“The World is Not All Rainbows and Butterflies:”
Facilitating Physical Activity and Quality of Life Among Children Living with
Cystic Fibrosis and Congenital Heart Disease – Toward a Conceptual Framework
and Parent - Mediated Behavioural Counselling Program

By

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A thesis submitted in conformity with the requirements for the degree of Doctor of Philosophy
Graduate Department of Exercise Sciences
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ABSTRACT

Although medical advances have improved prognosis for children with cystic fibrosis (CF) and congenital heart disease (CHD), these youth experience poor psycho-social health. Embedded within the Medical Research Council’s framework, the purpose of this Dissertation was to a) examine how CF and CHD children experience physical activity, b) explore parents’ perceptions toward their child’s activity, c) develop a theory of physical activity in childhood chronic diseases, d) develop a physical activity counselling program for youth with CF and their parents, and e) evaluate the program impact on quality of life and physical activity.

Study One explored how 14 CF youth experience physical activity. Active and inactive youth were characterized by different experiences, such as a sense of hope or despair. Given the burden of treatment and the fatal nature of the disease, youth negotiated temporal barriers to activity. Study Two explored perceptions toward activity among 29 CF and CHD parents. Parents discussed the benefits and barriers associated with physical activity for both child and self, and underscored the importance of role modeling.

By adopting a Grounded Theory approach, the theoretical constructs from Study One and Two were crystallized to develop a theory of physical activity in youth with CF and CHD. This theory was used to develop “CF Chatters:” A Six Week Physical Activity Counselling Program for Youth with CF and their Parents, and the intervention employed behavioural self regulation
skills. In Study Three, four case families in the CF clinic at the Hospital for Sick Children participated in CF Chatters. Improvements were noted in quality of life and physical activity, and participants described the program as convenient and relevant to their activity concerns. CF Chatters afforded therapeutic benefits to participants.

By employing an eclectic qualitative approach, this Doctoral Program has made theoretical and practical contributions toward our understanding of how physical activity is experienced among children living with CF/CHD. The findings support the use of behavioural counselling as an effective and feasible modality for enhancing quality of life and physical activity. This Dissertation calls on clinicians to attend to the activity needs of chronically ill Canadian youth.
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*When you are sorrowful, look again in your heart, and you shall see that in truth you are weeping for that which has been your delight* (Kahlil Gibran)

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“Philosophy begins in Wander” (Aristotle)

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“Curiosity, earnest research to learn the hidden laws of nature, gladness akin to rapture,
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interests were directed toward .... the secrets of the world” (Frankenstein, Mary Shelly)
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The world is hugged by the faithful arms of volunteers (Terri Guillemets)

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Chapter One: Introduction

“Jo took Beth down to the quiet place, where she could live much in the open air, and let the fresh sea breezes blow a little colour into her pale cheeks … so they were all in all to each other, and came and went, quite unconscious of the interest they excited in those around them – who watched with sympathetic eyes the strong sister and the feeble one, always together, as if they felt instinctively that a long separation was not far away (Alcott, p. 402).

The above passage is taken from Louisa May Alcott’s famous 19th century children’s novel, Little Women. In one poignant scene, young “dying Beth -” who suffers from rheumatic heart fever - weakly explains to her grieving sister Jo that she is not frightened of the dark road that lies ahead. Rather, she lives in fear of the rich and beautiful moments that she will miss with her family in their humble country home once she is gone. Beth’s passing away is quiet and tranquil. In the wake of her death, Beth’s family and friends continue to marvel at the simple pleasures, such as playing with a kitten or reading on her mother’s lap, that enriched her short life with much beauty. As time passes, Beth’s childlike goodness and generosity is immortalized. As though a faint but steady reverberating echo, the virtues of “Good Beth” are never forgotten by the bereaved March family who struggle for years to emulate her. Rather, they are comforted by the knowledge that Beth’s outdoor play adventures will no longer be interrupted by sudden “heart spells” that leave her weakened and bedridden for months.

Historically, sick children like Beth have invoked and engendered a range of complex cultural meanings and representations, such as fascination, fear, pity, hope, and courage. As suggested by Young, Dixon -Woods, Findlay, and Heney (2002), since they intensify notions of vulnerability and innocence, childhood illnesses may be considered as a concertized metaphor of childhood itself that magnifies psycho - social representations of what children are and the lives they should fulfill. Furthermore, childhood illnesses rudely shatter our sensibilities - that is, the accepted social order and the “natural progression of things” in which children grow up and make meaningful societal contributions. For this reason, there is a collective reluctance to talk of
or with chronically ill children, and, rather, we are compelled to uphold a mutual pretence that largely silences the experiences of children with chronic illnesses (Bluebond - Langener, 1978).

Furthermore, the broader childhood chronic illness literature has tended to overlook the physical activity experiences of ill children. Thus, by drawing attention to the physical activity experiences of ill children, my research bridges diverse academic disciplines such as psychology, pediatrics, behavioural medicine, childhood illnesses, and exercise sciences, in order to address a novel but neglected facet in our understanding of the chronically ill child. Drawing upon the creativity of qualitative research in this Doctoral program, I sought to understand and explore how youth with two chronic diseases, specifically cystic fibrosis (CF) and congenital heart disease (CHD), and their parents/caregivers understand, negotiate, and perceive physical activity. More importantly, informed by the voices of my participants, I sought to develop a theoretical model of physical activity in children and youth with CF and CHD. This Dissertation culminated in the development, implementation, and qualitative evaluation of a theoretically informed clinical behavioural intervention. Specifically, the impact of the program on participants’ quality of life and physical activity was explored. Before describing the program of work that was developed in this thesis, the rationale for studying physical activity and quality of life among children and youth with CF and CHD is first addressed below.

1.1 Physical Activity in the Chronically Ill Child: Rationale for the Program of Work

There are compelling and important reasons to study the physical activity experiences of children with CF and CHD. First, in the wake of a rising obesity epidemic, Canadian health officials, researchers, and clinicians have discussed the importance of regular physical activity participation for Canadian children and youth and identified the notable physical and psychosocial benefits (Tremblay & Willms, 2003). While “able-bodied” children and youths’ physical activity behaviours continue to occupy an important position on the Canadian research agenda (Tremblay & Willms, 2003), understanding and promoting physical activity among the ever
growing population of children and youth with chronic illnesses and disorders, is a glaring oversight. Since close to one in five Canadian children - or 20% - are living with a chronic health condition that requires a disproportionate amount of health care and social resources (Branstetter, Domian, Williams, Graff, & Piamjariyakul, 2008), attending to the physical activity needs of chronically ill children is, arguably, of critical importance to researchers and clinicians.

Second, various world governing bodies have advocated for children’s “right to play” and to access fun and enjoyable physical activity participation. This right is extended not only to healthy children, but, particularly, to those children who are the victims of oppressive and hostile social environments and experiences. Thus, arguably, the right to play and engage in recreation should be extended to chronically ill children too (Kidd & Donnelly, 2000).

Third, there are compelling practical and conceptual rationales for studying the physical activity experiences of children and youth living with CF and CHD specifically, and to contrast and compare their experiences. As a result of my previous, well established relationship with the CF and CHD clinic at the Hospital for Sick Children, these two clinical populations were readily available and accessible for me to study. Strong institutional support for my research and study facilitation within the clinic provided me with easy access to these two, highly coveted and policed clinical populations.

Furthermore, as discussed below, existing research and interventions have tended to focus on the physical activity experiences of children with physical disabilities, such as down syndrome or cerebral palsy. Pediatric exercise science and medicine for children with chronic illnesses such as cancer, CF, or CHD, is in its infancy and may reflect the relatively recent change in the prognosis of these once life threatening diseases. While there is an abundance of exercise training protocols for CF and CHD youth, there is an absence of qualitative research that has explored the broader role of physical activity in the lives of these youth, and little published information on the use of behavioural physical activity intervention approaches with these two
populations. This dissertation sought to address these lacunae in the CF and CHD physical activity research.

More importantly, CHD and CF greatly differ in terms of their etiology, treatment, and trajectory. Contrasting and comparing the similarities and differences in their physical activity experiences, was of personal interest to me. In particular, CHD can be considered as a chronic but manageable childhood illness. While these patients require ongoing medical monitoring for life, they are most often characterized by periods of health related stability post operatively, and the return of normalcy. In marked contrast, not unlike muscular dystrophy, CF is an ultimately fatal childhood illness that is characterized by progressive health losses and deterioration over the course of time. Its course and future outcome is thus uncertain. In selecting these two clinical populations, I was particularly interested in how the distinctly different etiology, treatment, and trajectory of CF and CHD may influence the physical activity narratives and experiences of patients and parents. In doing so, I was interested in illustrating some of the general and common physical activity experiences that characterize the broader group of children with chronic illnesses, with the hope of potentially transferring my results to other pediatric disease groups and contexts in the future, such as children with juvenile idiopathic arthritis. For instance, low self-efficacy, social exclusion, and fatigue are physical activity experiences that appear to be general to the broader group of children with chronic diseases (Culos Reed et al., 2002; Moola, Faulkner, Kirsh, & Kilburn, 2008). In contrast, the “social construction of risk” during physical activity (Moola, Fusco, & Kirsh, 2010), or temporal anxieties and dilemmas appear to be unique physical activity barriers that are specific to youth with CHD and CF respectively. Additionally, such a comparative approach allowed me to capture the unique and idiosyncratic nature of CF and CHD specifically. Thus, there were important practical and theoretical rationales for investigating the physical activity experiences of CF and CHD youth in particular.
Fourth, current research has documented the notable physiological benefits associated with physical activity participation for youth with a wide variety of chronic diseases, including CF and CHD. Furthermore, in light of the psycho-social health deficits that are experienced by this population, there is growing recognition of the role and importance of physical activity as a modality to enhance quality of life and psycho-social health in these youth (Culos Reed, 2002; Moons et al., 2005). While the quality of life enhancing benefits of physical activity is the subject of the review to follow, the extant quantitative and qualitative literature has confirmed the beneficial role of physical activity in the lives of youth with CF and CHD. In addition to well established physical benefits, physical activity offers a fun, non-pharmacological, and low resource pursuit for CF and CHD youth and promotes perceptions of normalization, skill building, mastery, and social inclusion among their same-age peers. As such, physical activity may enhance quality of life in children with CF and CHD and attenuate lingering psycho-social health problems (Culos-Reed, 2002; Goodwin & Staples, 2005). Given that psycho-social health is severely compromised in this population of sick children and they are more likely to suffer from depression, anxiety, reduced body image, and self-esteem in relation to healthy, able bodied peers, the potential psycho-social benefits of physical activity are particularly important.

Despite the numerous benefits of physical activity for children with CF and CHD, however, levels of participation remain low (Lunt, Briffa, Briffa, & Ramsay, 2003). Furthermore, studies indicate that these youth display reduced intensity of participation in relation to their healthy able bodied peers, and they do not reap the benefits of intense activity that are required to attain positive physiological outcomes. While the reasons for inactivity in youth with CF and CHD are not entirely known, a range of physical and psycho-social barriers appear to operate in tandem to reduce participation and restrict involvement. In addition to fatigue, pain, breathlessness, and coughing, for example, psycho-social barriers such as reduced physical activity self-efficacy, low value, low self-esteem, poor inclusion, liability concerns, and
parental overprotection may restrict participation and prevent youth from deriving the potential benefits of physical activity (Moola, McCrindle, & Longmuir, 2009).

Thus, while physical activity is a normalizing pursuit that is central to childhood itself (Taub & Greer, 2000), most youth with CF and CHD remain isolated and excluded from their peers in physical activity (Moola, Fusco, & Kirsh, 2011). Given that a range of complex barriers preclude these youth from deriving both the physical and psycho-social benefits of physical activity - and that they continue to be excluded from broader research initiatives for healthy youth - there is a compelling rationale for the development of theories and behavioural interventions that will help CF and CHD youth to circumvent these barriers and facilitate increased physical activity.

Thus, by drawing upon diverse fields, such as psychology, behavioural medicine, pediatrics, and exercise sciences, the purpose of this three-stage program of work was to draw upon the qualitative paradigm in order to develop a Grounded Theory of physical activity in children with CHD and CF. Guided by this conceptual framework, the study also sought to develop, implement, and evaluate a behavioural intervention with CF youth that will facilitate increased quality of life and physical activity participation. More specifically, the following research objectives and questions guided this Doctoral Dissertation:

1) What are the perceptions of youth with CF and CHD toward physical activity participation?
   What is the role of physical activity in their lives?

2) What are the perceptions toward physical activity in the parents of children with CF and CHD? What are the perceived benefits and barriers to participation? What role does physical activity have in their own lives? What interest do parents have in participating in a physical activity intervention with their child?

3) What are the central theoretical constructs that are relevant, important, and meaningful to the physical activity experiences of children and youth with CF and CHD?
4) How should a parent-mediated behavioural intervention be designed in order to facilitate physical activity for children with CF and CHD? What are the essential components of a parent-mediated physical activity intervention for those affected by CF and CHD?

5) What is the impact of a pilot, cognitive behavioural intervention on the quality of life and physical activity of parents and children affected by CF and CHD? How do children and parents feel after having participated in a parent-mediated physical activity intervention? How do different children and parents respond to the intervention? How may qualitative research be used as a way in which to evaluate the participant’s experience of the intervention?

6) What is the perceived feasibility and acceptability of a pilot behavioural intervention?

These important research questions guided this Dissertation and were explored throughout the program of work.

1.2 Definition of Terms

Particular terms were commonly referenced throughout this Dissertation. These terms are described below:

*Physical activity:* Physical activity is a broad and all encompassing term referring to any movement of the skeletal system that results in energy expenditure. While physical activity encompasses sport, exercise, and recreation, it also includes activities of daily living such as walking, gardening, and playing outdoors (Caspersen, Powell, & Christenson, 1985). World governing bodies have declared physical activity as an international children’s right.

*Quality of life:* As a result of the influx of injured soldiers returning home from abroad (Taylor, Gibon, & Franck, 2008), the term *quality of life* was first proposed after the Second World War. The need to attend to both wounded bodies and souls was well recognized and concerns for the wellness, activities, and occupations of the soldiers were paramount to their rehabilitation. However, the term was not indexed in academic forums until the 1970s (Taylor et
While the definition of quality of life is contested across multiple literatures, it can broadly be defined as one’s perception of physical, social, psychological, and emotional health within the context of culture, values, and personal goals, and therefore captures patients’ perceived wellness. Used interchangeably with other terms such as wellbeing, it broadly refers to perceived satisfaction and fulfillment in broad domains of health. Quality of life is multi-dimensional, thus emphasizing health as a holistic construct. Moreover, although there are attempts to measure quality of life, it is a subjective and personal construct and thus has a qualitative dimension (Waters, Maher, Salmon, Reddihough, & Boyd, 2005; Seid, Varni, & Jacobs, 2000; Matza, Sensen, Flood, Secnik & Leidy, 2004). In an effort to narrow the focus of quality of life interventions, the term health-related quality of life has been advanced. Eliminating non-health components, this term refers to one’s perception of the impact of disease and its treatment.

**Childhood chronic diseases:** Childhood chronic diseases refer to illnesses and diseases of childhood lasting longer than six months, such as CF, CHD, spina bifida, epilepsy, anorexia nervosa, attention deficit hyperactivity disorder or Acquired Immune Deficiency Syndrome (AIDS). Termed the acute to chronic disease era, advancements in medicine have resulted in an ever-growing population of children with chronic diseases (Newacheek & Taylor, 1992). While symptoms change and fluctuate over time, childhood chronic diseases significantly impact children’s daily functioning and wellbeing and are enduring in their nature and impact. Furthermore, unlike acute illnesses, childhood chronic illnesses require a disproportionate amount of resources, such as ongoing physician monitoring, home medical care, school assistance, and specialized physical activity. Some childhood chronic illnesses, such as CF or AIDS, are ultimately fatal (Newacheek & Taylor, 1992).

**Cystic fibrosis (CF):** CF is an ultimately fatal autosomal recessive genetic disease that affects 1/2500 live births. It is the most common fatal genetic disease among Caucasians. The
triumphs of science and medicine are particularly evident in the case of CF; while the majority of these patients died in childhood three decades ago, today, the mean life expectancy is approximately 30 years. While CF is a multi-system disease that affects the respiratory, digestive, and musculo-skeletal systems, the primary cause of death is respiratory failure. CF has no known cure, and, thus, treatment is oriented toward slowing the progression of the disease over time. The backbone of CF treatment is comprised of physical therapy, medicinal treatment, aerosol inhalers, and, in some cases, lung transplantation (Rosenstein & Cutting, 1998).

*Congenital heart disease (CHD):* CHD is characterized by structural anomalies of the heart that have functional implications (Mitchell, Korones, & Berendes, 1971). CHD affects 1/100 live births, and is therefore much more common than CF. CHDs vary tremendously in severity and functional impact, from mild to severe and debilitating. While CHDs may not be detected immediately, it is thought that they occur in utero during the process of heart development. Advances in surgery have dramatically altered the illness trajectory of CHDs. Approximately 15% of CHD patients require palliative care and are life limited. While this population may continue to display functional limitations and exercise intolerance, provided that routine medical care and cardiac management is administered in order to monitor the patient for signs of deteriorating health, the majority of post-operatively repaired patients have a normal life expectancy.

**1.3 Through the Looking Glass: A Note on Self-Reflexivity**

Prior to reviewing the extant literature, it is important to briefly discuss my personal interest in the topic of childhood chronic diseases and physical activity. Research endeavors are non-neutral and value laden. Rather, the researcher’s social position and location and own interests and preferences are brought to bear (Macbeth, 2001). Such experiences shape the research agenda and delimit the field of study, and the researcher’s personal experience, history, and encounters are reflected and refracted in and through the research. This brief self-reflexive
narrative will touch upon how my own experiences have stimulated my desire to study the physical activity experiences of chronically ill youth. In doing so, I illuminate sociologist C. Wright Mills’ (1959) declarative statement that the “personal is political,” and that we see our personal lives in the work that we do. Similarly, in the contemporary *Harry Potter children’s book series*, Gerstl-Pepin & Patrizio (2009) discuss Professor Dumbledore’s “Pensieve” as a metaphor for researcher self-reflexivity. The pensieve is a brewing cauldron that allows Professor Dumbledore to store, retrieve, re-live, and re-conceptualize past experiences and historical events. In teaching Harry about the pensieve, Professor Dumbledore imparts lessons about the value of self-reflexivity to any investigation.

I have devoted ten years of service to the Hospital for Sick Children as a volunteer and camp counsellor at a SickKids affiliated camping program for children with CHD. In doing so, I have been exposed to several clinical populations and inpatient units, including multi-organ transplant, dialysis, cardiology, infectious diseases, critical care, and bone marrow transplant. As an Undergraduate student in the Faculty of Physical Education and Health at the University of Toronto, in 2000, I was also pivotal to the establishment of a more enduring and sustainable University - Community volunteer partnership that “student-volunteers” still benefit from today. I acquired several skills and competencies through volunteerism, such as engaging in therapeutic play with sick children, assisting with medical procedures and infant care and feeding, engaging in leadership and teaching, and acquiring ethical mindedness.

Volunteers often have indelible memories and experiences which are described as “life altering,” and the impetus to greater self - reflexivity and introspection. A full account of the experiences and insights that I garnered as a volunteer at SickKids are beyond the scope of this Dissertation. I have observed, however, the various ways in which children and parents cope with serious childhood diseases and the multiplicity of grieving responses, which range from acceptance to chaos that they demonstrate. I have also noticed the isolation experienced by
diverse families for whom the Western medical model is largely unknown, and the tensions which often arise at the fragile and tenuous interface between religion, culture, and medicine. I have observed the sense of alienation and dislocation encountered by those families who are not from Toronto, and their struggle to establish some semblance of home in a large city that is unfamiliar to them. In shock, I have watched exhausted families turn against the advice of the clinical team and refuse further medical intervention in an attempt to preserve what remains of their child’s dignity and sanctity. I have listened to non-adherent and resistant patients’ dialogue about how medical non-compliance “frees them” to enjoy the little time they have left, heard children who are receiving palliative care question “where am I going?” spent time with neglected siblings who feel as though they are “lost in the shuffle,” and witnessed patients run away from the hospital on a “code white” upon receiving news that their illness will preclude them from engaging in a meaningful activity. When rapid deteriorations in patient health status occur, I have observed what Chapman et al. (2005) term, the struggle to facilitate “a good enough death.” I have talked with a broad range of allied health professionals - such as doctors, nurses, and social workers - who, in the wake of a child’s death, ask “did we fail her?” When I walk home from SickKids in the night time along the bustling and brightly lit University Avenue, I think about how, just beneath the famous Mother and Babe statue lies Toronto’s largest children’s graveyard.

More importantly, however, I have marveled at the incredible resilience and tenacity displayed by chronically ill youth who persist and endure through numerous surgeries, procedures, and hospitalizations and observed the great degree of care, compassion, and mutual understanding that chronically ill children display toward one another. With a sense of triumph, I have admired the vigour, vitality, and energy displayed by these youth when they engage in camp-based physical activities together in a way that does not mark them as disabled. Most notably, I have been surprised and humbled by the sophisticated and intelligent manner in which
chronically ill children become “medical experts,” and the high degree of independence, autonomy, and medical education they acquire in efforts to better manage their health. Lastly, I have been overwhelmed and perplexed on those rare occasions when the hospital itself emanates a sense of beauty and peace; the image of joyful faces in spite of painful experiences on Christmas morning at the hospital; children in wheelchairs playing under the arches of the oak trees on the front lawn; a single child reading a favourite book in some quiet corner of the hospital, momentarily oblivious to his surroundings and circumstances; and a parent bent in quiet and reflective prayer.

My experiences working at SickKids have exposed me to the vastness, richness, complexity, and nuances of the broad range of human emotions and ethical conundrums we face, in which I have vicariously experienced joy, horror, grief, confusion, sadness, learning, insight, and acceptance. It has exposed me to diverse cultures, nationalities, and peoples, who, despite vast differences in social location and position, are united in the common struggle to help their sick child get better. I was followed as a patient at the Hospital from 1993-2001 and was hospitalized on four occasions for several months. Although few would ever believe it of “well mannered and impeccably behaved Fiona,” in reflection, I was labeled as one of the most “non-adherent, rebellious, resistant, and recalcitrant” patients on the unit, one whose perceived wellbeing and quality of life far surpassed and exceeded objective indicators of illness. In reflection, I understand my experiences at SickKids as a struggle to articulate my voice, autonomy, health care expertise, and critique of the medical care; that by asking for what I perceived to be more ethical, humane, sensitive, and individualized care, my family and I confronted and challenged the unquestionable authority of the medical team. These experiences have fundamentally shaped my personal value and belief system, my philosophy, ethic, and interaction with children, how I have come to understand my relationship with the broader Toronto community, and the overwhelming sense of professional and personal obligation that I
have always had to intervene in lives of those that suffer. I feel compelled to publically advocate on behalf of chronically ill children, knowing that most people remain completely oblivious to - and ignorant of - their suffering. I want to draw attention to the “existential crises” (Westman, Bergenmar, & Andersson, 2010) of children and to communicate to the broader academic community that not all childhoods are characterized by the wonders of fun, innocent, and exploratory play. As I have learned, some childhoods are characterized by futile attempts to play and recreate with other children, or, rather by complete exclusion from childhood activity altogether. More importantly, my experiences as a volunteer and my personal and professional interest in physical activity and sport culminated in the desire to undertake a Dissertation in the field of childhood chronic diseases and physical activity. In this way, the self is always visible in the research we undertake (Macbeth, 2001), and I did not randomly stumble upon my research question. Rather, in the patients that I work with, I see shadows of a former self.

1.4 Summary

Despite the numerous physical and psycho-social benefits that physical activity may afford, children with CF and CHD remain inactive and excluded from broader physical activity initiatives for healthy children. They face numerous physical and psycho-social barriers to participation (Moola et al., 2009), and theories and methods for behavioural intervention development are required in order to circumvent these barriers and facilitate participation. This Dissertation drew upon a series of studies in order to address the question of physical activity in the lives of children with CF and CHD and their families and culminated in the development of a theoretical framework and behavioural intervention. By drawing attention to a population who are neglected in the research and centering their physical activity experiences, this chapter outlined the importance of considering how physical activity influences quality of life in chronically ill children and youth, and important research questions were stipulated. The following chapter will review the extant quantitative and qualitative literature on the physical
and psycho-social health status of children with chronic diseases and the role of physical activity in their lives. This will contextualize the problem my research addresses.
Chapter Two: Review of the Literature

In order to contextualize the role of physical activity in the lives of children and youth with CF and CHD, identify research limitations, and stipulate the rationale for the Doctoral Program of work, it was necessary to concisely review the extant quantitative and qualitative literature. A brief appraisal of this literature and mention of critical research issues will also be addressed. This chapter will draw upon the current research evidence in order to describe the diminished physical and psycho-social health status of children with CF and CHD. Following from this, this review will advance a critical discussion on the quality of life and psycho-social health benefits associated with physical activity.

Two important caveats are worth mentioning prior to reviewing the literature. First, since studies on the psycho-social benefits of physical activity in those with chronic diseases are scant, it is necessary to review exercise training interventions and make extrapolations to physical activity; this is a limitation associated with the review. Second, since physical activity studies for children with CF and CHD are emerging, it is necessary to draw upon studies conducted with other chronically ill youth. By introducing the reader to the problem of physical activity in children with chronic conditions and contextualizing the issue, this section explores the role of physical activity in their lives and examines evidence attesting to its physical and psycho-social benefits.

There is general consensus in the literature that amount and intensity of physical activity in children with CF and CHD is markedly reduced in relation to their healthy peers. For example, using accelerometry and self-report data, McCrindle et al. (2007) found that complex CHD patients who had undergone the Fontan procedure ¹ displayed levels of physical activity below

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¹ The Fontan procedure is a three stage palliative open heart surgery performed on children who cannot be offered a bi-ventricular repair (d’Udekem et al., 2007). Performed as a series of open heart surgeries under the age of five years, circulation is re-routed so that blood flow bypasses the dysfunctional ventricle and flows directly from the pulmonary
the 5th percentile for age and height. Sixty-two percent of these patients did not meet recommended levels of participation. Using a questionnaire in order to assess habitual physical activity in children with CHD, Lunt et al (2003) also reported that these youth display reduced intensity of physical activity. They are deprived of the cardio-vascular benefits which only participation in vigorous physical activities affords. Similarly, several studies have indicated that children with CF report reduced amount and intensity of physical activity. Using accelerometry and activity diaries, it was found that in relation to healthy controls, pubescent girls with worse pulmonary function displayed reduced amounts of physical activity (Selvadurai, Blimkie, Cooper, Mellis, & Van Asperen, 2004). These children are less engaged in structured activities; when they do participate, they prefer to be involved in unstructured play (Schneiderman -Walker et al, 2005). Illustrating the gendered dimension of physical inactivity, a longitudinal study which documented the habitual physical activity of CF youth, found that over half of the girls were inactive. Moreover, the steepest rate of lung function decline was observed in CF girls with the lowest amounts of activity (Schneiderman -Walker et al., 2005). In addition to diminished aerobic fitness and work capacity, children with CF display reduced intensity of physical activity in relation to their peers. Due to the increased oxygen cost associated with vigorous movements, children with CF may avoid these activities in order to reduce uncomfortable sensations such as breathlessness (Nixon et al., 2001). In light of the relationship between aerobic fitness and survival in children with CF, reduced engagement in vigorous activities may have important implications and hasten mortality (Nixon et al., 2001). Given this relationship, reduced amount and intensity of participation in CF is, arguably, a more pressing and immediate concern than for children with CHD. In this regard, the evidence suggests that youth with CF and CHD experience poor physical health and inactivity in relation to their peers.

artery to the lungs. As such, a single ventricle is responsible for both pulmonary and systemic circulation. These single ventricle or Fontan patients have reduced exercise capacity and life expectancy (d’Udekem et al., 2007).
Despite low physical activity frequency and intensity, however, several studies which utilize a variety of research designs and measurement tools, have documented the physical benefits associated with exercise training in youth with CHD and CF; physical training appears to attenuate diminished physical capacity and inactivity. While the benefits of physical training are well documented for both clinical populations, CF studies tend to be a more fertile area of inquiry. Research in exercise and CHD is still in its infancy.

Employing a home-based exercise program, Longmuir, Tremblay, and Goode (1990) noted improved exercise capacity in those Fontan patients who adhered to the intervention. Interestingly, the program assisted in normalizing levels of physical activity in youth with CHD, such that after the intervention, they were unremarkable from healthy peers. Moreover, benefits persisted for five years after the cessation of the intervention and similar findings have been reported elsewhere (Longmuir et al., 1990; Fredriksen et al., 2000). In a similar vein, there are numerous physical benefits associated with exercise training for CF youth across a range of interventions. While type of exercise differs, several authors report the numerous benefits associated with in-hospital supervised exercise training programs for CF youth. Improvements in FEV1\(^2\), peak aerobic capacity, BMI\(^3\), strength, maximal oxygen consumption, minute ventilation, total treadmill walking time, and anaerobic strength, were observed (Salvadurai, Blimkie, Meyers, Mellis, Cooper, & Van Aspersen, 2002; Turchetta, Salerno, Lucidi, Libera, Cutrer, & Bush, 2004; Klijn, Oudshoorn, van der Ent, van der Net, Kimpen, & Helders, 2004). Similarly, a 12 week home-based exercise training intervention was associated with improved maximal oxygen consumption and muscular strength in children with CF (Gulman, de Meer, Backel, Faber, Berger, & Helders, 1999). Similar findings were reported by Schneiderman-Walker et al (2000) who demonstrated a slower rate of lung function decline in those CF patients who participated in a three year home-based physical activity program. Finally, youth with CF

\(^2\) Forced expiratory volume in one second  
\(^3\) Body Mass Index
who participated in a four week physical activity, nutrition, and physical therapy camp displayed improved minute ventilation, peak work capacity, maximal voluntary ventilation, maximal aerobic capacity, oxygen pulse, and anaerobic threshold (Blau et al., 2002).

Although research designs and measurements vary, the extant literature indicates that children and youth with CHD and CF demonstrate reduced amount and intensity of physical activity and a diminished physical state. Moreover, a broad range of studies conducted in multiple settings indicate the beneficial impact of physical activity on several health indicators in these children and youth. While it is not the primary focus of this review, these youth are characterized by poor physical health and inactivity. Since improved physical health is a benefit associated with participation in physical activity, it may also be both an important secondary consequence of the intervention developed in this Doctoral program, an antecedent for continued participation, and an important line of inquiry for future research.

2.1 Quality of Life and Psycho-Social Health in Children and Youth with Chronic Illnesses

The extant quantitative and qualitative research indicates that quality of life and psycho-social health in children and youth with chronic conditions is poor. While the research is scant, it is evident that family members also encounter reduced psycho-social health and quality of life impairments. For example, Spurkland, Bjornstad, Lindberg, and Seem (1993) and Caset, Sykes, Craig, Power, and Mulholland (1996) employed a cross-sectional research design to explore psycho-social health and social, emotional, and academic functioning in children with severe heart disease. Youth with mild congenital heart lesions were included as comparison groups in both cases. Youth with severe CHD displayed high rates of clinical mental disorders. In addition, these youth displayed difficulties with social and emotional adjustment such as high levels of internalization and social withdrawal (Spurkland et al., 1993; Casey et al., 1996). In addition to impaired family functioning, these youth demonstrated severe learning and academic problems which warranted a diagnosis of a clinical learning disorder. In addition, children and youth with...
CHD have impaired health status and quality of life. Using a cross-sectional research design, Kamphuis et al. (2002) found that in relation to healthy peers, these youth report a diminished health status and poor quality of life. Deficits are in the physical domain such as reduced gross motor functioning and poor physical functioning. This indicates that the physical benefits associated with physical activity are critical, and underscores the importance of physical activity as a strategy to enhance quality of life.

Psycho-social health impairments in children with chronic diseases are not always overt and may not be detected using standardized clinical tools. Gupta, Mitchell, Giuffre, and Crawford (2001) found that in relation to healthy peers, children with CHD and asthma suffered from significantly greater covert and concealed specific fears. This included fear of injury and medical procedures and highlights the need to develop tools that are sensitive to the more subtle or hidden psycho-social pathologies in these youth. Interestingly, covert fears in youth with CHD were highly correlated with maternal anxiety. This demonstrates that mothers of youth with CHD who are particularly anxious and distressed, may further exacerbate youths’ specific fears. The children also displayed high levels of physiological anxiety (Gupta et al., 2001).

Quantitative studies have indicated that children and youth with chronic illnesses are likely to display poor quality of life and impaired psycho-social wellbeing when they are adults. In this regard, psycho-social health impairments linger and track into adulthood. For example, in a retrospective study of the Childhood Cancer Survivor Survey, Zebrack et al. (2002) found that as adults, childhood cancer survivors displayed high levels of depression and somatic distress in relation to their siblings. These lingering psycho-social problems were associated with reduced income, education, and employment as well as female gender and intensity of the treatment. Interestingly, a more severe and prolonged course of invasive chemotherapy was associated with greater psycho-social impairments thus illustrating the need to consider the mental health impact of treatment itself (Zebrack et al., 2002). Quantitative studies have also found that the parents of
youth with chronic diseases have impaired quality of life and psycho-social well-being. For example, in a prospective study of parental coping and adjustment to childhood cancer, mothers of children with cancer displayed high levels of anxiety and insomnia at the time of diagnosis in relation to fathers and healthy families. The level of family functioning deteriorated over the course of the child’s disease. With its child-centred focus, the pediatric system may be failing to meet the psycho-social needs of parents (Sawyer, Antonious, Toogood, Rice, & Baghurst, 2000).

Finally, reduced quality of life and impaired psycho-social functioning is a secondary outcome observed in studies that primarily seek to address the physical health status of children with chronic diseases. For example, De Jong et al. (1997) examined the physical and psychological components associated with quality of life in children with CF and results were compared to healthy peers. In addition to reduced exercise capacity and high rates of perceived dyspnoea, the participants displayed poor quality of life. Particular quality of life deficits included difficulty sleeping, resting, eating, engaging in home management, and physical activities (De Jong et al., 1997). In assessing habitual physical activity and health perceptions in youth with CHD, McCrindle et al. (2006) noted that these children have major deficits in quality of life areas, and, in relation to their peers, psycho-social health is worse in all domains.

While quantitative studies provide objective indicators of poor psychological health and quality of life in children and youth with chronic diseases and their families, they may foreclose the richness, complexity, and nuances through which poor psycho-social health is lived, experienced, and negotiated by these children and families. Studies that use qualitative inquiry as a lens for analysis have been extremely useful in elucidating some of the poor quality of life issues that quantitative studies eclipse. Drawing upon Leventhal's illness model that considers illness within the context of society and history, McMurray et al. (2001) sought to qualitatively describe perceptions of health and psycho-social wellbeing in children with CHD. These youth
found it difficult to cope with the disease and experiences such as hospitalization, surgery, medical procedures, and anaesthetics were taxing. Moreover, while they try to “forget about it,” these youth occasionally ponder “why me”? The children encounter limitations in physical education and report reduced stamina and endurance. While these youth are limited by others, they are also aware of the ways in which they limit themselves by acting with the utmost caution. Moreover, these youth are socially excluded from a variety of different activities and encounter bullying and stigma on account of their non-normative embodiment. While the children hope for a future that is free from medical intervention, they fear that ongoing stigma will prevent them from obtaining employment (McMurray et al., 2001). Similarly, qualitative literatures have illustrated that poor quality of life is experienced by CF patients over the course of the disease. In end stage CF, qualitative health literatures have “told stories” of how patients and clinicians struggle over the meaning of a “good enough death” and how to facilitate it in a manner which preserves sanctity and dignity (Chapman et al., 2005).

Berge et al (2007) used qualitative inquiry in order to illuminate the gendered nature of poor quality of life and psycho-social health in children with CF. Unlike CF boys, the girls require someone to be accountable to in order to engage in physical therapy and display a passive and dependent treatment orientation. They also fear that CF “marks” them as different and makes them stand out in relation to their peers; they feel excluded from activities such as dating and socializing. The girls are also at risk of developing severe mental health problems. As a result of the sadness of having to manage CF on their own, the girls shared their experiences of depression and suicidal ideations. Moreover, as they simultaneously navigate the nutritional demands associated with the disease with a broader cultural preoccupation with thinness, CF girls are at risk of developing anorexia nervosa, with devastating consequences sometimes. In contrast to the boys who integrate CF as an integral component of identity and “use it” as a teaching and learning opportunity for others, the girls come to a passive and reluctant sense of
acceptance through the use of psycho-therapeutic services. Influenced by parental overprotection that diminishes their social role, the mental health of CF girls is poor (Berge et al., 2007).

The qualitative literature base has illustrated that the parents of these youth also experience poor quality of life. Sparacino et al (1997) found that these parents navigate a range of psycho-social dilemmas in relation to their child’s poor health. Struggling to understand their child’s “normalcy,” they oscillate between perceptions of normal or not, and struggle to come to terms with the “shattered hope” of having a perfect infant. In light of the shame associated with CHD, parents struggle with knowing to whom this information may be safely entrusted. Since they do not know when or if their child will die, they are weighted down by a sense of uncertainty and are often reluctant to become attached to the ill child. Parents struggle with illness management; while they want to empower their child to have an independent role in disease management, they do not know how to safely transfer from the position of “manager to consultant.” Finally, in addition to social isolation in which parents become cognisant that their child lacks a same age peer group, parents express their concerns about the family impact of CHD. Indeed, problems such as marital strife, “lost and confused” siblings, and general family interruption and disruption are commonly reported (Sparacino et al., 1997). Glasscoe and Smith (2008) have also discussed the negative psycho-social impact of caring for a child with CF. In addition to experiencing stress and hopelessness, these parents experience significant temporal disjunctures in which there is a shattering of temporal integrity. In light of their child’s reduced lifespan, and the developmental effects of the illness on the child, parents’ sense of time and how it is passing with their child is significantly disrupted. Finally, other qualitative studies discuss the poor psycho-social health and reduced quality of life in the “shattered families” who assist in the management and care of chronic childhood conditions (Rempel & Harrison, 2007; Woodgate & Degner, 2003).
Based on a review of the extant literature base, there is consensus in both quantitative and qualitative health literatures that quality of life and psycho-social health is poor in children and youth affected by chronic diseases. The psycho-social impact of these diseases is not limited to children, and parents and other family members are also affected. The diminished psychological health in these children and youth must be attended to, through the incorporation of psycho-social health services (Duff, 2001). The following section briefly reviews current research evidence that makes the case for physical activity as a therapeutic modality to enhance quality of life and psycho-social wellbeing in children affected by chronic diseases such as CF and CHD.

2.2 Making the Case for Physical Activity: The Role of Physical Activity on Quality of Life in Children and Youth with CHD and CF

This section of the review will describe the role of physical activity in the lives of children and youth with chronic illnesses. The quality of life enhancing benefits of participation will be discussed as well as the psycho-social outcomes associated with participation. Critical research issues will be identified.

Most studies that address the psycho-social benefits associated with physical activity for children and youth with chronic illnesses occur in therapeutic camp settings. Therapeutic camps serve a wide range of purposes including opportunities for fun, normalization, disease education, skill building, and socialization. In addition, they serve as a forum for psycho-social research. As such, several studies have illustrated the psycho-social benefits associated with participation in therapeutic camps. For example, Moons et al. (2005) explored the impact of a multi-sports camp on perceptions toward health in youth with severe CHD; the Child Health Questionnaire (CHQ) was administered and a pre and post-test design was utilized. Camp participation resulted in enhanced perceptions of role functioning due to behavioural and emotional problems; general health, mental health, and health perceptions also improved. CHD youth have gross misconceptions regarding their physical abilities and encouragement and advocacy for physical
activity is required (Moons et al., 2005). Similar findings were reported by Briery and Rabian (1999) who examined the impact of a physical activity camp on attitudes toward the illness and levels of anxiety in children with multiple chronic diseases. A pre and post-test design was utilized and a battery of well-validated psychometric tools, such as the State Trait Anxiety Inventory for Children, was employed. Within the context of enhanced normalization, camp participation was associated with reduced anxiety levels and improved attitudes toward the illness. However, the psycho-social benefits were less notable for older, returning campers. These campers are more likely to be referred back to camp, and, as a result of having lived longer with a chronic health condition, they have a qualitatively different experience (Briery & Rabian, 1999).

Noting that camps are therapeutic landscapes that allow for a spatial and temporal removal from the stresses and strains associated with disease management, Kiernan, Gormley, and MacLachlan (2004) reported similar findings in a large camp study for children with multiple-chronic diseases and their siblings. Children from four different regions in Europe were included. Moreover, a battery of questionnaires was utilized and a pre and post-test repeated measures design was employed; the influence of age, type of illness, and geographic region on quality of life was considered. Camp participation was associated with reduced perceptions of physical symptoms and anxiety in the youngest campers. Enhanced self-esteem was observed in those campers from Central European regions only, thus illustrating the influence of social and cultural factors on self-esteem in children with chronic diseases. The authors call for the development of a comprehensive and multi-dimensional tool in order to assess the psycho-social impact of camp and note the non-linear nature of psychological change (Kiernan et al., 2004).

Group-based physical activity for children with CF is controversial (Blau et al., 2002). For example, due to the risk of cross-patient infection and acquiring potentially dangerous viruses and infections, the SickKids clinic recommends that CF patients remain greater than
three feet from one another (personal communication, CF clinic physiotherapist, 2009).

However, Blau et al (2002) noted several psycho-social benefits associated with participation in a four week sports and hiking camp for CF patients in the Swiss Alps. In addition to notable physical improvements, participation was associated with enhanced psycho-social wellbeing. This was influenced by positive peer pressure and social support for enhanced physical activity and nutrition (Blau et al., 2002). Finally, Hunter, Rosnove, Koontz, and Roberts (2006) note that while camps for children with chronic diseases serve multiple purposes, it is important to ascertain whether outcomes justify expenditures. Evidence attesting to the psychological benefits of camp informs critical decision making and ensures that measurements suit camp goals. In addition to enhanced disease knowledge and education, as well as improved blood glucose monitoring, diabetic children who participated in a one week mission based camp, displayed improved self-esteem, coping behaviours, and satisfaction (Hunter et al., 2006).

Qualitative literatures have discussed the benefits associated with physical activity for children with chronic diseases at camp. Goodwin and Staples (2005) suggest that children with disabilities feel a great deal of shame and have few positive role models. These authors suggest that developing a sense of self is contingent upon those identities which appear to be accessible. In addition to having fewer positive identities available to them, children with disabilities simultaneously navigate disabled and able-bodied identities. Seeking to explore the meaning of camp for children with disabilities, Goodwin and Staples (2005) found that camp participation in activities conjures up a sense of “not alone;” the children experience reduced disability isolation, appreciate being able to share individual biographies with one another, and a sense of “coming home.” Moreover, camp is associated with a sense of independence. There is positive peer pressure for disabled youth to undertake activities autonomously and children benefit from spending time away from their families. Finally, when engaging in activities at camp, the children acquire a sense of skill mastery and a chance to discover. They become cognizant that in
spite of the severity of the disability, they all, to some extent, have some level of ability (Goodwin & Staples, 2005). Other qualitative studies have illustrated that mastery experiences which confirm success in physical activity, engaging in non-traditional activities which limit the social and emotional demands that characterize mainstream sports, and positive reinforcement and social support for physical activity from significant others, may facilitate participation in children with complex CHD and enhance quality of life (Moola et al., 2008).

Data extraction was employed to illustrate that enhanced quality of life and psychosocial health are important secondary outcomes associated with participating in formal exercise training regimes for children with chronic diseases. For example, De Jong, Grevink, Roorda, & van der Schans (1994) conducted a home-based exercise training program for children with CF and evaluated the impact on physical performance and activities of daily living. In addition to improved oxygen consumption, lactate threshold, and work rate max, participation was associated with reduced ADL limitations; the ability of individuals to participate in the tasks of daily living is associated with enhanced quality of life. Salvadurai et al (2002) assessed the differences between an aerobic and resistance training program for children with CF. Improved quality of life was noted in the aerobic training group and the benefits persisted after the program was terminated (Salvadurai et al., 2002). Similarly, enhanced global self-worth and perceptions toward physical appearance, was an important psycho-social health outcome associated with participation in a 12-month home-based cycling program for children with CF (Gulman et al., 1999). Although formal measures were not used, Schneiderman-Walker et al. (2000) anecdotally reported that children with CF who participated in a clinical trial involving home-based physical activity felt better and reported favourable attitudes toward the program. Children with CF who participated in a 12 week clinical trial involving anaerobic training also experienced improved quality of life in the physical domain (Klijn et al., 2004).

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5 Activities of Daily Living
Finally, systematic reviews of literature have documented the quality of life enhancing benefits associated with physical activity for children and youth with chronic diseases. In their reviews on exercise for youth with CF, several authors note that exercise and physical activity may improve quality of life and psycho-social wellbeing in CF patients (Wilkes et al., 2009; Moran, 2008; Stevens & Williams, 2007). Systematic reviews of literature with other pediatric disease groups also underscore the psycho-social benefits of physical activity for youth with cancer. For example, Culos Reed (2002) indicates that physical activity plays a critical role in facilitating rehabilitation from pediatric cancers. In addition to improved self-esteem and quality of life, physical activity reduces perceived fatigue and pain. Moreover, improved physical competence mediates self-esteem in these youth and reduces anxiety and depression. Physical activity also enhances quality of life in children with cancer through its beneficial impact upon physical appearance. Indeed, by reducing gross motor functioning and increasing weight, cancer may have a detrimental impact on physical appearance. By attenuating these unpleasant cancer-related physical changes, physical activity improves quality of life (Culos Reed, 2002). Similar findings have been reported elsewhere. For example, Keats, Courneya, Danielsen, & Whitsett (1999) found that children who remain active during cancer treatment display improved quality of life and self-esteem. In addition to reducing disease related de-conditioning, the risk of acquiring secondary health complications associated with the disease, and improving physical functioning, physical activity improves quality of life in those with chronic conditions more generally (Painter, 2008). In this regard, although psycho-social health and quality of life does not feature centrally in interventions, it appears to afford important therapeutic benefits to youth with chronic diseases.

2.3 “The Things that Stand in my Way:” A Review of Barriers to Physical Activity Participation for Youth with CF and CHD
The above section indicated that there are numerous quality of life enhancing benefits associated with physical activity for children and youth with chronic diseases such as CF and CHD. However, despite these benefits, these populations encounter several barriers which thwart efforts to become more active. Parental overprotection or the vulnerable child syndrome, in which parents worry excessively about their child’s ill health and safety, may have a detrimental impact and cause these youth to doubt their physical abilities. Indeed, parental perceptions toward physical activity may override the objective severity of the cardiac diagnosis (Bar-Mor, Bar-Tal, Krulik, & Zeevi, 2000). Furthermore, clinician miscommunication regarding physical activity is a commonly reported barrier. Although these youth benefit when clinicians encourage physical activity (Schneiderman Walker et al., 2000), studies have indicated that many health professionals do not communicate clear physical activity guidelines or discuss the importance of physical activity. Moreover, termed the “vicious cycle of inactivity,” the disease itself may undermine participation. Symptoms such as fatigue, pain, dizziness, and breathlessness cause youth to avoid physical activity. In turn, physical inactivity merely serves to exacerbate CF and CHD symptoms thereby entrenching a broader pattern of physical inactivity (Schneiderman-Walker et al., 2002; Nixon et al., 2001). Other commonly reported barriers include social stigma and exclusion from “able bodied” peers; not being able to keep up increases perceptions of difference and non-normativity (Moola et al., 2008; McMurray et al., 2001). Particularly in the case of CHD, school-related barriers may undermine participation in physical education. Indeed, liability concerns, poor inclusion of children with chronic conditions into physical education classes, and misconceptions regarding the physical abilities of cardiac youth, impede participation in physical education. There are barriers to physical activity which affect those with chronic illnesses more broadly (Moola et al., 2009). For example, these populations are characterized by a changing course of illness and treatment, ongoing hospitalization and relapse, and numerous co-morbid conditions. These health-related instabilities make participation in
physical activity problematic (Painter, 2008). Moreover, people with chronic illnesses may not value physical activity or express interest in joining programs. In addition, given the small and heterogeneous nature of these populations, they are often difficult to access and recruit. When physical activity is not viewed as important and valuable by the clinical team more broadly, this may detract from the legitimacy of physical activity as a therapeutic modality for people with chronic illnesses (Painter, 2008). Finally, children with chronic diseases invariably find that there are community barriers to participation. Access to safe and enjoyable programs in the community that provide the adapted resources required to become active is limited (Goodwin & Staples, 2005).

Thus, while there are numerous quality of life enhancing benefits associated with physical activity for children with chronic illnesses, this needs to be balanced with a broader consideration of the barriers that impede participation. While being cognizant of these barriers then, camp-based studies, exercise training interventions, and reviews of literature, have documented the quality of life enhancing benefits of physical activity for children with chronic illnesses and improved psycho-social wellbeing.

Research Issues and Limitations: There are several outstanding research issues uncovered in this review that need to be addressed. Illustrating the close association between psyche and soma, mind and body, impaired quality of life in children and youth with chronic conditions is most often, in the physical domain. Since quality of life is a multi-dimensional construct, this suggests that the physical burden of the disease is most noxious and has the greatest impact on total quality of life. Since physical activity will enhance quality of life through improved physical functioning, all authors advocate for its incorporation into routine care.

Another research issue concerns the manner in which mental health diagnoses are made in children with chronic diseases and their families. When describing the psycho-social health status of these youth, diagnoses are made liberally and generally. Only in some cases are clinical
diagnostic tools, such as the DSM IV \(^6\), employed, and most often, general research labels are used. Clinical psychological disorders greatly differ from subclinical pathologies in terms of social and occupational impact and functioning. While qualified professionals should use clinical diagnostic criteria, researchers must report how diagnoses are made and the measurement tools employed. Not doing so impedes the accuracy of the diagnosis. Since psycho-social morbidity is high in these patient groups, it was important for me to accurately describe the psychological status of my participants.

There are also several limitations associated with the existing body of literature. Research on the psycho-social health benefits associated with physical activity for children with chronic diseases is in its infancy and still emerging. The majority of these studies address biomedical indicators, and the broader health benefits for these youth are not considered. Alternatively, psycho-social health is a *tangential or adjunctive issue* to studies that primarily seek to address medical health. This is a major limitation associated with the research base and more research is needed in order to discern the *psycho-social benefits of participation*. Moreover, there are limitations associated with camp-based studies that make claims to enhancing quality of life. For example, pre and post-test repeated measures designs are most often employed, and thus, it is difficult to discern how camp influences quality of life over time; more longitudinal and prospective studies are required. In addition, despite the numerous benefits of camp, the short duration (Epstein, Stinson, & Stevens, 2005), relative inaccessibility, and unrealistic setting impair the feasibility of such programs. Moreover, the review indicated that improved quality of life and psycho-social health may be a secondary outcome associated with exercise training interventions. However, few, if any, exercise training programs include quality of life outcomes.

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\(^6\) The Fourth Addition of the Diagnostic and Statistical Manual of Mental Disorders (DSM IV), is a manual for the classification and description of mental diseases. Different “axis’s” are used in order to indicate the clinical significance of the disorder and the manual is continually revised. Use of the DSM is “best practice”; patients should not be diagnosed as having a clinical disorder unless the DSM is used. Only licensed psychological clinicians can make diagnoses using the DSM (DSM- IV- TR, 2000).
In order to understand the broader influence of such programs on total health, these interventions should include quality of life measures.

2.4 Gaps in the Literature and the Rationale for Physical Activity Interventions in Children with CHD and CF

This review has garnered important insights and strategies that allow us to critically consider how to enable physical activity in children and youth with CF and CHD. While level and intensity of physical activity is low (McCrindle et al., 2007), they express interest in becoming more active. However, various barriers such as parental over-protection, low self-efficacy, peer exclusion, and fatigue may prevent them (Moola et al., 2009). Furthermore, qualitative literatures emphasize the importance of social support, mastery experiences, and enhanced self-efficacy as facilitators of physical activity (Goodwin & Staples, 2005; Moola et al., 2008). Although there are benefits associated with involvement in hospital-training programs, such research designs invariably encounter difficulties with sustainability. The review indicated that physical activity interventions should be specifically tailored to children’s local environments, and, thus, sensitivity to local contexts or ecological acuity must be displayed (Gannotti et al., 2007; Gulman et al., 1999). Children express positive attitudes toward physical activities in the home over which they have liberal choices, and this appears to offer sustainable benefits (Schneiderman-Walker et al., 2000).

However, this review indicated that there are significant knowledge gaps in relation to how we understand physical activity for chronically ill youth. For example, qualitative research is lacking, and little is known about how children with CF feel about physical activity and their perceived barriers. In addition, where parents are thought to be the critical gatekeepers to positive health behaviour change (Golan, 2006), there are currently no studies that examine parental perceptions toward physical activity for their ill youth. Along these lines, this review indicated that there are no studies that specifically examine the impact of physical activity on
quality of life in children and youth with CF and CHD; quality of life remains a neglected construct. More specifically, the literature is characterized by the absence of studies that have used the home as a setting for enabling physical activity and the overabundance of standardized exercise training programs that may not capture youths’ interests and motivations. Whether families may serve as the unit of analysis for enabling physical activity change, begs further investigation. Moreover, the literature is characterized by attempts to understand physical activity in single populations of chronically ill youth. While there are merits associated with this approach in terms of elucidating disease specific facilitators and barriers to participation, this limits a broader understanding of the general physical activity experiences of children with chronic diseases. For this reason, a general, cross childhood illness comparison is warranted. The literature is limited by the predominance of quantitative studies, and, thus, stories of how children with chronic conditions live and experience physical activity are lacking. Physical activity studies and interventions tend to be atheoretical, and we lack information about how to enable physical activity in these youth. Furthermore, despite the benefits of counselling approaches that allow researchers to tailor physical activity to the everyday lives, worlds, and dilemmas of patients, there is a lack of theoretically informed behavioural interventions that employ counselling as a method for intervention delivery to increase physical activity.

It is evident that there are critical lacunae and knowledge gaps in the existing literature on physical activity in children with CF and CHD. By employing a series of graded and sequential studies that allow for the accumulation of knowledge over time, this Doctoral Dissertation addressed these limitations and was developed within the context of these significant research absences. Specifically, this Dissertation attempted to fill these critical knowledge gaps by a) using qualitative inquiry as a lens to explore the perceptions, feelings, and attitudes toward physical activity in children with CF. Where little is known about how the parents of these youth feel about their own physical activity and their children’s, this Dissertation also b) explored
parental perceptions toward physical activity. Informed by the findings from Study One and Two, I also sought to c) develop a Grounded Theory of physical activity in youth with CF and CHD to guide and inform the development of a parent-mediated behavioural intervention designed to facilitate physical activity and quality of life. Finally, this Dissertation d) employed qualitative methods in order to explore how youth with chronic diseases and their parents perceive and appraise a physical activity program. The program of work was novel and built upon gaps and limitations extracted from the review of literature.

2.5 Summary: By describing the poor physical and psycho-social health status in these youth and the potential for physical activity to attenuate these deficits, this review has contextualized the problem of physical inactivity for children with chronic diseases. More importantly, this comprehensive and thorough review of the literature has drawn upon current quantitative and qualitative literatures in order to describe the state of inactivity and diminished quality of life and psycho-social health status of these youth (Spurkland et al., 1993). Moreover, it illustrated that the parents and families of these youth encounter diminished quality of life (Sawyer et al., 1999). This review appraised quantitative and qualitative health literatures that have documented the beneficial influence of physical activity on facilitating greater participation, quality of life and psycho-social health in chronically ill youth, thus making the case for physical activity interventions (Moons et al., 2005; Culos-Reed, 2002). Finally, the limitations of the review were noted, and this Doctoral program was contextualized within the existing research gaps. It is evident that physical activity is a therapeutic modality which may attenuate the quality of life and psycho-social health deficits encountered by children and youth with CF and CHD. This review has contextualized the problem of physical inactivity for these patients, illustrated the role that it may play in their lives, attended to knowledge gaps in the field, and outlined the rationale for physical activity interventions.
In the following chapter, a more detailed description of the physical activity program of work in this Dissertation will be outlined. In particular, the three, inter-related studies in this Dissertation will be discussed within the context of the Medical Research Council’s framework for complex behavioural intervention development (MRC), so that readers comprehend the structure of this multiple manuscript Dissertation.
Chapter Three: Overview of Doctoral Program of Work

This chapter will provide a broad overview of my Doctoral Program - toward a theory and behavioural intervention of physical activity in CF and CHD - and explain the structure of the Dissertation to the reader. Since the Dissertation culminated in the design of a behavioural intervention that aimed to facilitate increased physical activity participation and quality of life, it was embedded within the Medical Research Council’s (MRC’s) framework for complex behavioural intervention development and delivery. This framework is briefly described below.

The MRC Framework for Complex Behavioural Intervention Development

The MRC framework, depicted below, encourages researchers to adopt a four-stage process for the design and implementation of robust complex behavioural interventions. This approach recognizes that non-pharmacological interventions are subject to greater variation than drug trials, and, since they are comprised of various inter-connected parts and multiple components, it is critical that they be rigourously evaluated (Campbell et al., 2000; McEachan et al., 2008). Rather than developing evidence-informed interventions that are “context stripped” - that is, highly controlled but potentially irrelevant to the needs, interests, and contexts of participants’ lives - the MRC framework encourages researchers to develop programs that are grounded in and informed by potential user’s experiences and knowledge, and relevant theories. Furthermore, in their formative piece, Smith & Pell (2003) parody the researchers’ overwhelming insistence on conducting randomized control trials (RCTs) as the gold standard of research excellence. Since it lacks real world relevance and applicability, they facetiously write about the impossibility of ever conducting an RCT to assess whether parachute jumping results in a greater incidence of death! In this regard, studies conducted within the MRC framework also encourage researchers to think about whether an RCT should be developed in the given field.
Although there are slight semantic differences, the MRC framework is comprised of a four stage iterative process, in that new information discovered at the feasibility stage, for example, may require researchers to revisit and re-examine their evidence and theoretical base from previous phases. In general, the developmental (or identifying components stage) consists of identifying and developing the evidence and theoretical base that is necessary for the development of the intervention, and identifying and defining central components, such as reduced quality of life in youth with CF and CHD. To examine how the intervention works as well as potential barriers toward intervention development, this stage may also entail conducting qualitative research. This phase consists of critical developmental work (Campbell et al., 2000; McEachan et al., 2008).

The feasibility and piloting phase of the MRC framework is characterized by using the information obtained from the developmental phase in order to develop an optimal intervention. As in the case of my study, this stage may also entail pilot testing the intervention with patients in order to assess the feasibility and acceptability of the program. Thus, this exploratory trial allows the researcher to engage in necessary consultations and obtain feedback about the intervention itself (Campbell et al., 2000). Ideally, as in my program, outcome measures that will comprise the final trial should be included in the exploratory trial, such as quality of life and physical activity. Researchers are also encouraged at this stage to consider not only outcomes that are relevant to patients, but also those that could have relevance to health care providers or systems, such as the health economic measures (Campbell et al., 2000).

While the final two phases of the MRC framework were not addressed in this Dissertation and are the focus of future research, the evaluation phase consists of assessing the effectiveness of the intervention, identifying and understanding change processes, and assessing cost effectiveness. Furthermore, questions regarding sample size, inclusion and exclusion criteria, and methods of randomization, may continue to be addressed and modified at this stage.
Finally, the *implementation phase* of the MRC framework is concerned with the broader dissemination of the evidence produced into health care practice, as well as the ongoing surveillance, monitoring, and long term follow up of the study. At this stage, a change in health care practice – such as implementing physical activity counselling as a routine aspect of CF care - has hopefully been attained (Campbell et al., 2000; McEachan et al., 2008).

With its particular emphasis on encouraging researchers to conduct initial, preliminary and developmental work in order to devise interventions that are suitable and sensitive to participants’ lives, as well as theoretically informed and guided (McEachan et al., 2008), this Doctoral Dissertation was embedded within the developmental and feasibility phase of the MRC framework only. Indeed, research within other phases of the MRC framework - such as evaluation - are the focus of future research studies related to this Doctoral Dissertation. In working within the *developmental phase*, I sought to explore and address how users of potential interventions - youth with CF and their parents - feel about physical activity and their perceived interest in a physical activity intervention. In doing so, this initial developmental qualitative work allowed me to consult with “local communities of practice” in order to explore their physical activity experiences and needs and develop theories and interventions that are relevant and meaningful to their lives and grounded in their experiences. The studies in this Doctoral Dissertation that addressed Phase One and Two of the MRC framework - the developmental and feasibility phases - are described below.
Figure One: Doctoral Studies Embedded Within the MRC Framework

3.1 MRC Framework: Developmental Phase

Study One: “No Time to Play:” Perceptions Toward Physical Activity Participation in Children and Youth with CF

As a component of the developmental phase of the MRC framework (Campbell et al., 2000; McEachan et al., 2008), this qualitative research study sought to explore the perceptions, attitudes, beliefs, and feelings toward physical activity in 14 youth living with CF. Drawing upon qualitative inquiry as a lens for analysis, semi-structured in-depth interviews were conducted in the CF clinic at the Toronto Hospital for Sick Children. Specific research questions included:
1) What is the role of physical activity in the lives of children and youth with CF? 2) How active are youth with CF? 3) What are the barriers to physical activity that are encountered by these youth? 4) Are youth with CF interested in participating in a physical activity intervention?

A thematic analysis was employed in order to render findings. The findings from this study are discussed within the context of the existing literature base in Chapter Four and important implications and recommendations are proposed.

*Time Line: August-October, 2008*

*Study Two: Developing Physical Activity Interventions for Youth with Cystic Fibrosis and Congenital Heart Disease: Learning from their Parents*

As a second component of the *developmental phase* within the MRC framework and building upon the findings from Study One, Study Two of the Doctoral Program explored the perceptions of parents with chronically ill children toward physical activity. Twenty eight parents were recruited for this qualitative study from both the CF and CHD centres at the Toronto Hospital for Sick Children. Qualitative semi-structured in-depth interviews were employed in order to investigate the meaning of physical activity for youth with chronic diseases from the perspective of those most intimately involved in their health care and treatment.

Specific research questions included:

1) How do the parents of chronically ill children feel about their child’s physical activity?
2) Do the parents of these youth understand physical activity as important and valuable for their child? 3) How has the child’s chronic illness and hospitalization influenced the activity levels of these parents? 4) What are the barriers or concerns that parents have in relation to their child’s activity? 5) How interested are the parents of children with chronic illnesses in becoming more physically active with their child? 6) Does enhanced physical activity have health benefits for the parents of chronically ill youth?
All interviews were taped and transcribed verbatim. A qualitative thematic analysis was employed in order to garner general patterns across the parents’ data with particular attention to the similarities and differences between CF and CHD parents. Efforts were made to sample both mothers and fathers and similarities and differences between these two groups, were addressed. The findings from this study are interpreted within the literature base in Chapter Five, and what is already known about the struggles and triumphs of these pioneering parents who provide care in uncertain health contexts (Rempel & Harrison, 2007). Implications and recommendations were made. Importantly, Study Two identified the five case families who were interested in ongoing participation in the final study and thus attended to pragmatic concerns. Specifically, parents were questioned with respect to the perceived acceptability and feasibility of a parent-mediated physical activity intervention.

**Time Line, CF parent interviews:** October - December, 2009

**Time Line, CHD parent interviews:** January – February, 2010

**Identifying Relevant Theory:** The developmental phase of the MRC framework also encourages researchers to identify and develop relevant theories to guide and inform intervention development (Campbell et al., 2000; McEachan et al., 2008).

**Crystallization Phase - Toward a Grounded Theory of Physical Activity Participation in Children with CF and CHD and Their Parents**

This intermediate research phase - between Study Two and Three - consisted of employing the Grounded Theory qualitative research tradition in order to crystallize the central physical activity constructs from the children’s and parents’ data. As a preliminary form of theory building, this phase consisted of reading and re-reading the children’s and parents’ data in order to elicit the physical activity constructs which are most salient, important, and meaningful for them. The specific research questions which guided this phase included:
1) What are the general theoretical constructs that are most relevant, significant, salient, and meaningful to physical activity in youth with CF and CHD? 2) What are the general theoretical physical activity constructs that are most relevant, significant, salient, and meaningful to the parents of youth with CF and CHD? 3) Do these general theoretical constructs inform a preliminary conceptual framework in order to better understand and theorize the physical activity experiences of chronically ill youth? 4) Can these general theoretical principles be applied to physical activity intervention research for these families?

Grounded Theory was the qualitative method which informed the crystallization phase, including intense reading and immersion in the data as well as a critical analysis of the transcripts and findings. This allowed me to discern, distil, and crystallize the general principles and constructs which are most relevant to physical activity participation in children with CF and CHD. Examples of relevant constructs for this population included fatigue, pain, low value for physical activity, and the importance of social support. More importantly, by shaping and tailoring the physical activity intervention so as to “foreground” these constructs, my Grounded Theory influenced the general design and content of the physical activity intervention and provided a foundation for the delivery of the intervention.

**Time Line: March - April, 2010**

The developmental phase of the MRC framework also encourages researchers to identify other theoretical frameworks, such as *family systems theory, social ecological models, and the Movement Continuum Theory*, which are relevant to the development of their interventions, and, in this way, the field of potential theories that aid in intervention development, is quite eclectic. The following theoretical frameworks from the pre-existing literature also provided useful frameworks for guiding the development of my intervention and are briefly illustrated below.
i) *Family Systems Ecology* was originally informed by general systems theory and environmental science. A system is understood to be a set of complex elements in mutual interaction with one another (Cummings, 2002) and occurs on multiple scales of influence. Family systems theory is a framework for thinking about the systemic and bound nature of families, as systems. Not unlike environmental systems, family systems display adaptation, resilience, and homeostasis. However, when coping resources are taxed by chronic stressors such as illness, the system is characterized by dysfunction and chaos and risks weakening and collapse (Cummings, 2002; Crethar, Snow, & Carlson, 2004; Cowie, Quinn, Gunning, & Gunning, 1998).

The concept of wholeism and holistic family health is integral to family systems thinking. Phenomena in the family are not viewed in isolation, but rather, are understood within the context of total relationships, networks, and complex interactions (Crethar et al., 2004). As such, each member of the family is an individual system who interacts with other members in the larger, whole family system. The family itself consists of smaller parts or subsystems which are embedded within the larger family unit. For example, sibling and parental subsystems are embedded within the larger family system and are characterized by inter-relationships both within this sub-system as well as the whole system. In turn, the family itself is embedded within larger inter-linking systems, such as the school, work, religious, and recreational contexts. While each of these multiple and interlinking systems bear a disproportionate influence on the existence of the family, all are central to how emotions, behaviours, and cognitions are experienced (Crethar et al., 2004).

Moreover, family systems theorists consider the biological, social, psychological, spiritual, and cultural elements of family health. For example, while the physical health of the

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7 Systems theory is a paradigm that considers connections and inter-relationships among different components and requires transdisciplinary thinking (Leischow, 2006). It combines elements of chaos theory and complex adaptive systems. When applied to health behaviour change, central theoretical tenets of systems theory include: a) behaviour change is often a quantum event, rather than a linear one; b) behaviours can mimic chaotic processes which are sensitive to initial conditions and difficult to predict; c) behaviour change unfolds within the context of complex adaptive systems, in which there are multiple and inter-related component parts- interactions are non-linear, and the results of these interactions are greater than the sum of their parts (Resnicow & Page, 2008).
family is related to the state of the child’s condition, sociological health refers to the broader interface and support networks between the family, school, and health care system. Similarly, spiritual health refers to whether family members are able to find a sense of meaning in the child’s health condition (Cummings, 2002).

These theorists suggest that while the sick child and his/her associated behavioural difficulties - such as lack of physical activity - are often the presenting problem raised by families, these problems are not attributable to the individual child. Rather, they are indicative of broader, systemic problems in the family system. In order to ascertain what family patterns the child’s “symptom” is responding to, it is essential for exercise counsellors to decipher characteristics such as family functioning, boundaries, dynamics, and patterns of communication (Cowie et al., 1998; Crethar et al., 2004). It is thus important to bring attention to how behavioural problems displayed by the sick child may serve important broad purposes in the family and illustrate systemic family dysfunctions (Crethar et al., 2004). For example, family systems interventionists working with a family affected by childhood brain tumour recognized that “Michael’s” psychological distress and conflict in the parental sub-system, closely corresponded with his three-month visit to the pediatric oncologist at the hospital; the family was suppressing covert fears related to the cancer returning (Cowie et al., 1998). Interventionists noted that stress in the sibling sub-system was related to missing past family events and reunions, such as family picnics, and being responsible for their ill brother’s school work. Family interventionists try to alleviate individual symptoms by recalibrating how the family functions, interrelates, and communicates, and aims to facilitate more functional and healthy patterns of family relations. Interventionists sought to change family relations by encouraging members to talk about and share their feelings regarding Michael’s upcoming hospital appointments, alleviating siblings of the burden of being responsible for their brother’s school work, and mourning the loss of events and anniversaries that were missed as a result of his illness.
changes in the broader functioning of the family served to attenuate Michael’s psychological distress (Cowie et al., 1998).

Moreover, interventionists are cognizant that families are living and bound systems. While the family is characterized by particular patterns of relating, these will invariably change as the family evolves. When intervention work is complete, facilitators should be cognizant that the family will have to readjust to the loss of a valuable support system (Crethar et al., 2004).

Thus, family systems theory offers an important and valuable theoretical framework for understanding physical inactivity in CF/CHD children (Fiese, 2005; Berry, Sheehan, Heschel, Knafl, Melkus, & Grey, 2004). Indeed, physical inactivity may be conceptualized not as an individual behavioural problem, but rather, indicative of broader, systemic family functioning. In using this framework, it is possible to consider the family in its constitutive parts and its larger whole. In my intervention, efforts were made to observe the physical activity behavioural patterns, roles, modes, and channels of communication which characterize the family. Future research may seek to enable greater physical activity by recalibrating systemic physical activity behaviours in the family.

ii) Informed by Bronfenbrenner who proposed a framework for thinking about the inter-relationship between humans and their social settings and contexts, social ecological models also provide a valuable conceptual framework for thinking about physical activity in families affected by chronic childhood conditions. Social ecology recognizes that behavioural interventions should exceed the level of the individual (Elder et al., 2007), and that it is important to consider the multiple contexts that influence behaviour. This theory has been used previously in intervention research with chronically ill children. For example, individual coping and illness behaviours, such as depression, a sense of burden, warmth, and conflict are influenced by family and physician relationships (Bellin & Rice, 2009, Shapiro, Perez, & Warden, 1998).
These theorists propose that the individual is embedded within four nested structures that exert an influence on behaviour. These concentric nested structures consist of the micro-system, which refers to the individual’s immediate system, such as the family, work, and school environments (Shapiro et al., 1998). The meso-system refers to the systems of influence that are created in the inter-relationship between microsystems. For example, the inter-relationship between the family and school (microsystem) may create an influential mesosystem in and of itself. More broadly, the exosystem refers to the influence of those indirect and distal factors, such as neighbourhoods and governments. Lastly, the macrosystem considers the ideological patterns of the broader society, such as how the cultural environment regards physical activity.

Although all systems inter-relate with one another (Bellin & Rice, 2009; Shapiro et al., 1998), the system of greatest influence is most proximal to the individual. In turn, there is a hierarchy of influence such that each system itself is influenced and constrained by the system that it is embedded within (Shapiro et al., 1998). Moreover, each system of influence is composed of multiple physical, social, and cultural environments. For example, the microsystem of the family and school consist of both physical and social environments and physical activity interventionists may attempt to intervene upon both of these environments. By providing equipment to engage in physical activity and changing physical activity attitudes, beliefs, and norms in the family, the physical and social environments are intervened upon respectively (Elder et al., 2007; Bellin & Rice, 2009).

In addition to considering multiple domains of influence, social ecological models draw upon multiple theories and theoretically informed behavioural strategies. For example, principles of operant conditioning, such as behavioural reinforcement, are used to inform such interventions. Strategies such as providing positive reinforcement for physical activity, reducing positive reinforcement for sedentary behaviours, and reducing barriers toward physical activity, are utilized (Elder et al., 2007). Moreover, social cognitive theory has informed interventions
that employ a social ecological framework. Strategies such as increasing self-efficacy for physical activity, social modelling and observation of physical activity in the family, and vicariously learning physical activity behaviours, are utilized (Elder et al., 2007).

Social ecological models provide a conceptual framework for thinking about the ways in which family members affected by chronic illnesses are impacted by multiple systems of influence in a series of nested structures (Kitzmann & Beech, 2006; Sachkiki et al., 2002). During my intervention, this framework allowed me to consider how factors in the family environment as well as broader peer group, school, and community, will bear direct and indirect influences on physical activity. Although embedded within multiple contexts, each exerts a disproportionate influence with proximal microsystems, such as the family, bearing the greatest influence. In addition, this theoretical framework stipulates methods that may be effective in changing physical activity at multiple levels of influence.

iii) Finally, the Movement Continuum Theory offers an important theoretical framework for thinking about physical activity in families affected by childhood chronic diseases. Briefly, it is suggested that a grand theory of movement is required that relates to all aspects of a movement discipline and displays enduring characteristics over time (Cott & Finch, 1995). This theory offers a holistic framework and suggests that movement occurs on a continuum from the microscopic level to the person moving in the environment and society (Cott & Finch, 1995; Allen, 2007). For example, while some biophysically oriented researchers may be interested in movement parameters of patients with chronic diseases at tissue or organ system levels, other researchers who are more broadly interested in physical activity, can use this framework to think about the movement of people with chronic illnesses in society and the environment.

Moreover, the movement levels are influenced by physical, social, and psychological factors. For example, while genetic factors and the chronic illness itself affect levels of physical activity, motivation, personality, attitudes, and beliefs toward physical activity are also relevant
factors. In addition, environmental and contextual elements influence physical activity, such as the socio-economic status of the family, access to affordable and enjoyable physical activity opportunities, and cultural physical activity norms. In this way, movement is influenced by multiple determinants (Cott & Finch, 1995).

In addition, the movement levels are highly interdependent. For example, tissue level pathologies will invariably influence broader aspects of physical functioning (Allan, 2007; Cott & Finch, 1995). Moreover, an intervention that is delivered at a single movement level will exert effects upon all other levels. The movement continuum theorists also purport that each individual has a maximum achievable movement potential (MAMP), which denotes the finite nature and limits of movement. Moreover, individuals have a current movement capability (CMC) which refers to how much they are able to move. They are also characterized by a preferred movement capacity (PMC) that denotes the amount of movement that is desired (Allan, 2007; Cott & Finch, 1995). Interestingly, although the CMC and the PMC are usually the same, illness and disability often result in a discrepancy between these two constructs and the extent of the difference will vary based on the nature of the trauma in question. People with illnesses are usually referred to services when there is a presenting discrepancy between the preferred and the current movement capability and efforts are made to attenuate the movement deficit. In addition to increasing the CMC, preventing further deficits to the CMC, working towards the PMC, and closing the CMC and PMC gap, movement specialists should seek to uncover the physical, psychological, and social factors that are contributing toward the movement deficit and should attend to these determinants accordingly (Cott & Finch, 1995). For example, lack of parental support for physical activity or poor access to opportunities, may be factors which contribute toward the movement deficits of a child with a chronic illness.

In this regard, the Movement Continuum Theory offers a set of broad theoretical propositions and insights in order to understand physical activity in families affected by
childhood chronic diseases. By providing a holistic and contextual framework for thinking about different levels of movement in context (Allan, 2007; Cott & Finch, 1995), this theory was useful for my study. During my intervention, I considered how children move in their contexts, societies, and environments. Thus, the MCT provided a way in which to tailor the intervention to these specific levels of movement as well as to think about how the intervention may impact other levels of movement. As a contextual and holistic theory of movement, it was ideal for parent-mediated intervention research, where, arguably, factors in children’s social environments, such as peers, family members, and the school, either facilitate or undermine physical activity.

Summary of Theories

In addition to developing my own crystallized Grounded Theory from the child and parent data in Study One and Two - discussed in chapter Six - the developmental phase of the MRC framework also encourages researchers to identify theories in the existing literature that may be relevant and important for the development of an intervention. In this regard, family systems theory, social ecological models, and the movement continuum theory, also offered relevant theoretical frameworks for thinking about parent-mediated intervention research. These theories emphasize: the family as a system that is characterized by individually functioning parts as well as broader, systemic and complex relationships (Cummings, 2002); children with chronic diseases as embedded within nested spheres of influence and contexts (Kitzmann & Beech, 2006); and movement as a complex, holistic bio-psychosocial construct (Cott & Finch, 1995; Allan, 2007). While foundational elements and disciplines vary, all place emphasis on the importance of contexts and environments, as well as multiple levels of influence, in shaping behaviours such as physical activity. In tandem, these theoretical insights offered a broad and inter-disciplinary framework for thinking about physical activity in the families of children with chronic diseases, and, where relevant, were used as lens for interpreting the findings.
3.2 MRC Framework: Feasibility and Piloting Phase

Based on the developmental phase that identifies participants’ needs and relevant theory, the feasibility and piloting phase of the MRC framework consisted of developing and pilot testing a preliminary behavioural intervention (McEachen et al., 2008; Campbell et al., 2000). The purpose of this study was to assess the impact of the intervention on physical activity and quality of life, as well as to explore participants’ perceptions toward intervention feasibility and acceptability. This pilot work has ensured that future interventions are better tailored to participants’ lives before larger scale trials, such as RCTs, are conducted. Embedded within the feasibility and piloting phase of the MRC framework, Study Three of this Doctoral Dissertation consisted of a pilot intervention.

Study Three: “CF Chatters:” The Development and Feasibility of a Six - Week Theoretically Informed Physical Activity Counselling Program for Children and Youth with Cystic Fibrosis (CF) and Their Parents

Informed by my Grounded Theory, the final study in this sequential research program consisted of intervention development; a parent-mediated physical activity intervention was designed by the researcher based on the previous two studies and theory. Moreover, given the importance of quality of life to the overall health of chronically ill youth, this study examined how physical activity influences the quality of life of four case families. The primary research questions which guided this study included:

1) How should a parent-mediated physical activity intervention for children and youth with chronic illnesses and their families be designed? 2) What are the central components of a physical activity intervention for these families?

The specific research questions which guided the pilot testing of this intervention included:

1) What is the impact of a parent-mediated physical activity intervention on perceived quality of life in children and parents affected by CF? 2) How did the physical activity
intervention influence other aspects of psycho-social wellbeing and broader family functioning? 3) Did the physical activity intervention impact how parents and children feel about their health and illness? 4) How did different families enrolled in the intervention respond? 6) What were the barriers and limitations associated with this type of intervention research? 7) Is this type of intervention research acceptable and feasible for families?

Methods for the analysis of study three included qualitative case study research to investigate the impact of the intervention on physical activity and quality of life, and participants’ perceptions toward the feasibility and acceptability of the program. A self report physical activity tool was also utilized. The findings from the pilot study are reported in Chapter Six and are being used to develop a larger scale study at SickKids.

*Time Line: September - November, 2010*

In summary, this Dissertation consisted of a sequential series of three studies embedded within the *developmental and feasibility phases of the MRC framework* (McEachen et al., 2008). Knowledge garnered from each of these smaller, embedded studies was used to inform the subsequent stage of research. The series of studies in this research program allowed for the accumulation of knowledge over time.

The reader should note that individually, each study is contained and nested within a bound manuscript that is either in peer review or awaiting submission to a scholarly forum. Each manuscript consists of its own research questions, theoretical propositions, set of methods, and analysis, such that it explores one facet of the physical activity prism for chronically ill youth. However, collectively, the series of studies are inter-related. By building upon knowledge obtained from the previous study, it allows for a coherent and logical sequence and guides the reader toward a more complete, detailed, and thorough understanding of the role of physical activity in the lives of chronically ill children and youth and their parents. Thus, the studies may
be considered as both discrete entities as well as inter-related components. By developing a theoretically guided intervention, as well as evaluating the intervention, the broader vision of this research design was to establish an extensive research program in the field of physical activity for children with chronic illnesses.

The first study in the broader series, “No Time to Play,” is presented in the following chapter.
Chapter Four, Study One:

“No Time to Play:” Perceptions Toward Physical Activity Among Youth with Cystic Fibrosis (CF)

Physical therapy is time consuming…I guess that it helps your lung function. The bad thing about that, is like, it also takes away from some of your time - if you wanted to be active, you would not be able to be active, if you had to do your treatment.” (Zoe, age 12). The above quote exemplifies how one patient living with cystic fibrosis (CF) negotiates the experience of physical activity within the context of significant temporal losses. Despite the notable physical and psycho-social benefits of physical activity for patients with CF, the majority of these youth are inactive (Nixon et al., 2001). Furthermore, the extant literature is characterized by an absence of qualitative studies which help to illuminate why youth with CF are inactive. The study described below explores how one group of youth with CF experience and negotiate physical activity, and, in doing so, illustrates the numerous barriers and temporal constraints that limit their participation.

4.1 Review of the Literature

CF is a fatal, autosomal recessive genetic disease that affects 1/2500 North Americans (Lowton, 2004). Although the majority of people with CF died in early childhood several decades ago (Nixon et al., 2001), notable achievements in respiratory medicine have dramatically altered the illness course. While treatment remains palliative, novel therapies including medication, physical therapy, and nutritional supplementation, have reduced mortality with the mean life expectancy now being approximately 30 years (Williams, Mukhopadhyay, Dowell, & Coyle, 2007). Indeed, some authors have described CF’s coming of age and the need to re-conceptualize it not solely as a childhood illness, but as one that extends into adulthood (Jessup & Parkinson, 2010).
Despite the fact that patients with CF are living longer, they are still affected by significant morbidity (Moran & Bradley, 2010). For example, undertaking numerous, time consuming treatments on a daily basis or being hospitalized are particularly arduous for youth with CF and interfere with important social activities (Williams et al., 2007). In addition, youth with CF experience reduced psycho-social health, such as depression, anxiety, body image issues, behavioural disorders, concerns related to health in the future, and prolonged parental dependence (Berge et al., 2007). Described as “being shipwrecked” (Jessup & Parkinson, 2010), a pervasive sense of impending danger also invades the lives of many people with CF and may compromise psycho-social health. Furthermore, excessive coughing in public may cause significant social distress. Finally, young people with end stage CF may struggle with health providers over the meaning of a good enough death and how to facilitate it in a manner which preserves dignity (Chapman, Landy, Lyon, Haworth, & Bilton, 2005). Despite an improved prognosis, youth with CF experience compromised physical and psycho-social health (Berge et al., 2007). Interventions to increase quality of life and psycho-social health, including physical activity and exercise, have been advocated for.

4.2 The role of physical activity and exercise for youth with CF

Given that it may slow disease progression and contribute important psycho-social benefits, there is growing interest in the use of physical activity as an adjunctive treatment for CF (Moran & Bradley, 2010). Even for patients with severe disease, regular physical activity - defined as any bodily movement that is produced by the skeletal muscles which results in energy expenditure (Casperson et al., 1985) - is strongly encouraged and may be a normalizing pursuit that is central to childhood. For example, several studies undertaken in diverse research sites such as clinical, home, and camp settings have reported beneficial outcomes associated with physical activity and exercise for youth with CF (Stevens & Williams, 2007). The notable physical benefits include reduced rate of lung function decline, improved physical functioning,
enhanced muscular strength and endurance, increased aerobic and anaerobic capacity, and reduced mortality (Gruber et al., 2008; Klijn et al., 2004; Moorcroft, Morris, & Webb, 2004; Schneiderman - Walker et al., 2000; Turchetta et al., 2004). Physical activity may enhance quality of life and psycho-social well-being in youth with CF (Rose & Sandy, 1986). For example, the ability to engage in activities of daily living, well-being, body image, self-esteem, resilience, acceptance, social functioning, and perceptions of wellness, are secondary quality of life outcomes associated with physical activity for youth with CF (Enright, Chatham, Ionescu, Viswanath, & Shale, 2004; Selvadurai et al; 2002; Stanghelle, Hjeltnes, Bangstad, & Michaelson, 1998).

However, despite the notable physical and psycho-social benefits of physical activity, most youth with CF are less active than age-matched peers and activity levels diminish further during adolescence (Prasad & Cerny, 2002). Little is known about why youth with CF are inactive or how to facilitate increased activity. Potentially, the “vulnerable child syndrome” in which parents restrict youth from activity based on false perceptions of illness and adopt negative appraisals toward physical activity, are psycho-social barriers which deter participation (Wilkes et al., 2009). Unpleasant physical symptoms are also deterrents to physical activity participation (Swisher & Erikson, 2008). For example, termed the “vicious cycle of inactivity,” symptoms such as fatigue and breathlessness may lead to activity avoidance. In turn, physical inactivity only serves to exacerbate CF-related symptoms, thereby entrenching a broader pattern of inactivity (Nixon et al., 2001). Other youth with chronic illnesses also report that experiences such as bullying, social stigma, poor inclusion, liability concerns, and lack of access to physical activity opportunities, impede physical activity (Goodwin & Staples, 2005; Moola et al., 2009). More specifically, the inability to keep up with able-bodied peers may increase perceptions of non-normativity among youth with chronic illnesses (McMurray et al., 2001; Moola et al., 2008).
The inherently exploratory and explanatory nature of the qualitative paradigm may help to illuminate why youth with CF are inactive in spite of the notable benefits that physical activity may afford. However, thus far, qualitative investigations that explore youths’ perceptions and barriers toward physical activity, as well as the role of physical activity in their lives, remain a neglected area of research inquiry. Indeed, subsumed under literature that explores parents’ “adultist” perspectives, or, rather, the perspectives of other chronically ill children, the voices of youth with CF are absent in the existing literature. Uncovering such information is critical if we are to more fully grasp the perceived barriers to, and facilitators of, participation. Comprehending the potentially complex reasons why youth with CF are inactive and how to facilitate activity through the design of interventions that are more sensitive to their social lives and worlds, is a notable advantage which may be afforded by such a descriptive and exploratory patient-centered approach. Thus, the purpose of this qualitative study was to explore the role and meaning of physical activity in the lives of a group of youth with CF and to understand their perceptions and barriers toward physical activity.

4.3 Method

Conceptual Framework

In order to address the research question, concepts from the sociology of health and illness were drawn upon as a conceptual framework and lens for interpretation (Smith & Sparkes, 2008). As biographical disruption, the experience of illness may fragment the structures of everyday life, unsettling one’s sense of self and calling into question one’s role in the broader social world (Bury, 2005). More importantly, as wounded story tellers in a remission society, people with chronic illnesses may construct stories of their experiences (Frank, 1995). For example, culturally dominant narratives of restitution - that is, “feel-good” stories that are

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8 The “remission society” refers to a society in which it is assumed that people with illnesses can – and want to - recover from their illnesses and attain health. Healthism is the foundation upon which the remission society is premised.
pleasing to hear - emphasize triumph in the face of adversity and the success of individual survivors over illness. In contrast, stories of chaos emphasize pain and hardship, in which one’s life world is dominated by illness and the self is lost and all consumed (Frank, 1995). Furthermore, some scholars have explored how athletes narratively construct their experiences of illness within the context of sport (Sparkes, 2004). For example, the early stages of elite athlete Lance Armstrong’s battle with an aggressive form of cancer, was characterized by much pain and turmoil and the very integrity of the self was threatened. However, drawing upon dominant narratives of restitution, he triumphantly overcame his struggles and returned to sport unscathed (Sparkes, 2004). Thus, concepts from the sociology of health and illness provided a useful conceptual framework. In particular, in seeking to explore perceptions toward physical activity in youth with CF, I was alert to how youth may narratively construct stories of illness and its impact on physical activity.

Research Setting, Recruitment, and Participants

This study was approved by the Research Ethics Board at a metropolitan pediatric hospital in Canada, and the treatment of participants was in accordance with the ethical standards that are outlined by the American Psychological Association. The study occurred in the Cystic Fibrosis Clinic between July and September 2008 and was facilitated by a physiologist and nurse. Eligible participants were identified using the outpatient clinic registry, and I proceeded to contact participants, explain the study, and request participation. Fourteen youth between the ages of 11 and 17 participated (Table One). The mean age of participants was 14.2 years and there were an equal number of females and males. The degree of lung function decline and the age of diagnosis varied, and the participants had no other illnesses or disabilities. One youth was hospitalized at the time of the interview and another was receiving home intravenous therapy. However, the remaining participants were receiving routine medical follow-up in the outpatient CF clinic and were medically stable. Although the majority of the sample was Caucasian, one
participant self-identified as Black and the other as East Indian. Most participants were from
middle class families, with three participants from socio-economically deprived regions in the
Greater Toronto Area. It was our intent to conduct interviews with youth independently of
parents. However, three participants requested that their parents accompany them during the
interviews. Although this was an uncontrollable limitation of the study, the presence of parents
appeared to be comforting for youth and should be considered within the context of conducting
ethical research with chronically ill youth.

Table One: Participant Descriptions, Children’s Study

<table>
<thead>
<tr>
<th>Pseudonym</th>
<th>Age</th>
<th>Gender</th>
<th>Activity Category</th>
</tr>
</thead>
<tbody>
<tr>
<td>1. Rudy</td>
<td>12</td>
<td>F</td>
<td>Inactive</td>
</tr>
<tr>
<td>2. Faisal</td>
<td>16</td>
<td>M</td>
<td>Inactive</td>
</tr>
<tr>
<td>3 Danny</td>
<td>16</td>
<td>M</td>
<td>Inactive</td>
</tr>
<tr>
<td>4. McKayla</td>
<td>15</td>
<td>F</td>
<td>Active</td>
</tr>
<tr>
<td>5. Graydon</td>
<td>15</td>
<td>M</td>
<td>Active</td>
</tr>
<tr>
<td>6. Cassie</td>
<td>15</td>
<td>F</td>
<td>Active</td>
</tr>
<tr>
<td>7. Isabella</td>
<td>15</td>
<td>F</td>
<td>Inactive</td>
</tr>
<tr>
<td>8. Harriet</td>
<td>12</td>
<td>F</td>
<td>Active</td>
</tr>
<tr>
<td>9. Koby</td>
<td>11</td>
<td>M</td>
<td>Inactive</td>
</tr>
<tr>
<td>10. Gretchen</td>
<td>12</td>
<td>F</td>
<td>Inactive</td>
</tr>
<tr>
<td>11. Samantha</td>
<td>16</td>
<td>F</td>
<td>Inactive</td>
</tr>
<tr>
<td>12. Zoe</td>
<td>16</td>
<td>F</td>
<td>Active</td>
</tr>
<tr>
<td>13. Diana</td>
<td>15</td>
<td>F</td>
<td>Inactive</td>
</tr>
<tr>
<td>14. Celeste</td>
<td>14</td>
<td>F</td>
<td>Inactive</td>
</tr>
</tbody>
</table>

Qualitative Research Tradition: Thematic Analysis
Informed by the work of Boyatzis (1998) and Braun and Clarke (2006), thematic analysis (TA) was the qualitative research method that guided the development and analysis of this study. Before describing this method, it is important to discuss a few concepts regarding the use of TA.

Thematic analysis (TA) is a foundational qualitative method that is broadly used across a range of disciplines, such as psychology, sociology, marketing, and even science. It is a qualitative method that seeks to systematically identify, analyze, and report patterns and themes in the data and is thus a way of organizing, describing, and interpreting a qualitative data set. While TA is often used, it is a poorly demarcated qualitative method that has not garnered the same degree of attention as the other major qualitative research canons, such as Grounded Theory or phenomenology. Following Braun and Clarke’s (2006) suggestion that more information is required on the application and evaluation of TA, I would also suggest that the method requires further development regarding its history and epistemic foundations.

While there is some controversy regarding whether TA is a method in its own right (Braun & Clark, 2006), or, rather, a method that is comprised within other methods (Boyatzis, 1998), in my use of TA is this study, I define it as a flexible, independent qualitative method that is compatible with both realist – empirical as well as constructivist - interpretivist paradigms (Braun & Clarke, 2006). In this regard, so long as researchers make their ⁹ ontological and ¹⁰ epistemological assumptions explicit and transparent, it is a flexible method that can be employed in diverse ways. Thus, since I consider youth with CF to have multiple, rather than singular realities and experiences, and that the knowledge I produce about their lives and physical activity experiences is a historically and culturally situated interpretive act, my use of TA is this study is compatible with a constructivist ontology and an interpretivist epistemology.

In making my assumptions about reality and knowledge explicit and transparent - that I am

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 ⁹ Ontology refers to the nature of reality
 ¹⁰ Epistemology refers to the nature of knowledge (Weed, 2009). Much of the controversy between researchers is largely due to epistemic conflicts regarding the nature of reality and knowledge.
hoping to uncover a truth about youth with CF and their experiences of physical activity, rather than The Truth - I am hopefully circumventing one of the central problems that hampers the use of this method. I have made my constructivist and interpretivist views transparent, in which I am critical of the notion of unassailable, stable, and universal fixed and certain truths, and I do not assume linear relationships between language, meaning, and experience (Braun & Clarke, 2006).

Furthermore, TA is associated with both “bottom up” and “top down” approaches toward the description and analysis of data. Top down or theoretically driven approaches characterize how most research within TA is undertaken, in that a pre-existing theoretical framework drives and encodes the interpretation of the data. In contrast, inductive and emergent data driven approaches remain guided by and “close to the data,” and are not wedded to a particular theoretical framework. While this makes for greater analytical freedom, the invariably ambiguous nature of this analysis can overwhelm some qualitative researchers and lead them to avoid data driven techniques. While my study was theoretically sensitized by concepts from the Sociology of Health and Illness within the social constructivist paradigm, my analysis was not fixed to nor solely encapsulated within this theory, and my interpretation and analysis of the findings drew upon many theoretical frameworks. In this regard, the analytic approach that I adopted in this study can be characterized as bottom up or inductive.

Finally, TA is characterized by the identification and analysis of manifest or latent themes. Characterized by a description of reality, manifest themes aim to describe and reflect the reality of the participants to the reader. In contrast, the search for latent themes occurs at the interpretative level of analysis in which the analyst aims to interpret and analyze the meaning of participant’s narratives beyond the literal and semantic level, and to interpret their behaviours within the context of their life worlds. Since I made efforts to interpret and analyze CF youths’ commentary and to speculate and analyze their narratives, my thematic analysis was latent in nature and occurred at the interpretive level. Overall, TA is a flexible and diverse qualitative
method that is compatible with many research traditions. Characterized by pattern recognition and conceptual complexity, it equips researchers with the skills to analytically identify and interpret themes within qualitative data sets and corpus. In making my ontological and epistemic views transparent, I have made an effort to communicate the assumptions that guided my use of this method.

Data collection, data analysis, and trustworthiness

Semi-structured interviews served as the primary means of data collection and were 45 minutes to one hour in duration (See Appendix One). In order to explore the perceptions of youth with CF toward physical activity, I devised an interview guide (Creswell, 1994). Although the interview guide was not pilot tested prior to the study, it was informed by relevant literature and sensitizing concepts (Holt & Timminen, 2010) and approved for appropriateness by members of the research team. For example, commonly understood barriers to physical activity for youth with CF as well as theoretical concepts from the sociology of health and illness, assisted with the development of questions (Frank, 1995; Swisher & Erikson, 2008). Finally, I have worked with chronically ill children in pediatric facilities for over a decade and my clinical observations informed the interview guide. The interview questions were flexible in nature and unobtrusive and questions were guided by participants’ responses. For example, questions included: is physical activity important or not important for you? Can you explain whether or not CF impacts how active you want to be? Can you tell me what physical activity means to you? Furthermore, although participants were asked about what it is like to engage in physical activity with CF, the term physical activity was not defined by the researcher. Rather, youth were left to interpret the term on their own and participants’ tacit understanding of physical activity shaped their responses. Furthermore, although interviews probed youths’ physical activity experiences, participants conflated health, illness, and physical activity. Thus, as illustrated in the findings, youth explained physical activity in terms of living with CF.
The interviews occurred in appointment or patient rooms and were taped and transcribed verbatim. All participants gave informed and written consent to research participation, and, by continually asking children how the interview was going and if they wanted to proceed, informed consent was maintained throughout the interview process. Since competent children and youth are able to independently consent irrespective of age at this particular institute, parental consent was not sought. Finally, saturation refers to the point at which no novel data is emerging and redundancy in the data is obtained. Thus, data collection was terminated after 14 interviews (Guest, Bunce, & Johnson, 2006).

All interview data was audio-taped and transcribed verbatim, and, in line with my interpretive-constructivist assumptions, I had an active rather than passive role in the analysis of the data and make no claims to naive realism (Braun & Clarke, 2006). Firstly, in order to completely immerse myself within the data and to familiarize myself with it, I engaged in multiple in-depth readings of the data. Secondly, for the purposes of coding the raw data, or the smallest units that can produce meaningful insights related to the phenomena, the data was read again and coded. In this case, commonly occurring words and phrases across the data corpus—such as “time-” were coded. Furthermore, following Braun and Clarke (2006), I coded as much of the available data as possible and ensured that the coding process was not context stripped: in this regard, the coding process encapsulated the context and dialogue under which the discussion occurred. Thirdly, the coded data was collated and organized into higher order themes. This careful and recursive decision making process involved making important and critical judgments about which codes were similar and dissimilar, which codes could be grouped together, where particular codes “fit,” and the centrality of the overall theme to the broader story about the data. This iterative and recursive process involved continually moving back and forth between codes and themes and the relationship between them. Fourthly, through a process of revision and refinement, the themes were collapsed and refined. Additionally, in order to define the essence of
the theme and what it means in relation to other themes, the themes were carefully defined and named. In this regard, theme names adequately describe and explain what the theme is about and offer sufficient evidence - or data - to corroborate and describe it. This process was assisted by my supervisor who reviewed half of the transcripts. Through discussion, my supervisor and I shared and discussed interpretations in order to challenge the identified themes and their connections in a form of peer debriefing (Lincoln & Guba, 1985). Within and across the data, all of the themes are internally consistent as well as externally heterogeneous. In this regard, the data within each theme coheres together in a meaningful manner and there are identifiable differences between themes (Braun & Clarke, 2006; Boyatzis, 1998).

To provide the reader with an example of how higher order concepts, such as mastery experiences and parental support, were generated, I will turn to the concept of “narratives of despair.” Each of the transcripts was read individually. As I read the data, it was coded for what Maykut and Morehouse (1994) refer to as “meaning units” that provide data which illustrate and substantiate a concept. Although there is controversy regarding whether transcribed data should be coded by word, phrase, or paragraph, I coded the individual and group transcripts by phrase. Thus, while qualitative data analysis is always an interpretive exercise with the researcher both identifying and naming coded data, phrases such as “I feel helpless; I feel depressed; I don’t care about my health; I have no hope to give him; why has this happened to me?,” were given a numeric code on the transcript to denote that they all signified a common meaning – a sense of helplessness and despair as a result of CF. Although there are many ways in which to code, I worked with bold interface, highlighters, and cut and paste tables in order to identify, code, sort, and name categories such as “helplessness and despair” on my computer processing system’s word documents. At this preliminary stage of category development, I termed the category “helplessness and despair.” Similar “meaning units” were encapsulated into this initial category, and, through the process of constant comparison, different data were sorted and collated
elsewhere. In this regard, all of the data in the initial category of “helplessness and despair” alluded to and supported this broader, common psychosocial experience. After each individual transcript was coded for “helplessness and despair,” I proceeded to read, code, and sort across the broader corpus of data. Through a process of revision and refinement, I sought a higher level of conceptual analysis. For instance, since CF youth told stories to convey their sadness – thus reflecting a very specific way of articulating their distress – I used “narratives of despair” as the category name to reflect a higher level of abstraction and analysis. As described above, I then searched for the inter-relationships between narratives of despair and other categories, ensuring that these were supported empirically by the data. For instance, narratives of despair were related to whether youth felt positive or negative about physical activity. Finally, when presenting the data, I selected representative quotes from the category of “narratives of despair,” and strived to attain a balance between data presentation and interpretation. This data analysis process was also employed across study two, the crystallization phase, and study three.

Trustworthiness: Qualitative researchers are concerned with issues of trustworthiness. Trustworthiness refers to the truth - value of the findings or the degree to which the results accurately convey an authentic and believable account of the lives under study (Creswell, 1994). The measures put forth to increase trustworthiness in this study included conducting an audit trail which monitored all steps of the research process, maintaining a research journal in which the research process and emerging thematic analysis was documented, and engaging in discussions with the secondary investigator and the clinical nurse facilitator. Throughout the analysis, the interpretations were discussed with my supervisor as a critical friend to challenge the identified themes and their connections and to develop notions of authenticity, fidelity, and believability (Sparkes, 1998). More specifically, in order to discuss discrepancies and develop consensus concerning the main themes of the study, my supervisor also read and coded half of the transcripts (Spalding & Phillips, 2007).
The study drew upon the creativity of qualitative methods in order to explore the physical activity perceptions and experiences of youth with CF. The main themes, 1) positive perceptions toward physical activity, 2) negative perceptions toward physical activity, and 3) “no time to play,” are illustrated and explained below. Youth displayed either positive or negative perceptions toward activity and different experiences (subthemes), such as parental support or illness narratives, were associated with positive or negative activity appraisals. Finally, all participants negotiated physical activity within the context of reduced time. Regardless of youths’ physical activity perceptions, this final theme was central to how all participants interpreted the role of physical activity in their lives. In my presentation of the report below, I provide a story about the data within and across themes. In doing so, as an analyst who is both a cultural member and critical commentator, I offer a story that describes, explains, and interprets how youth with CF understand physical activity. See below for a conceptual illustration of the findings.

**Figure Two: Conceptual Illustration of Study One Findings**
Pseudonyms have been used in order to protect the anonymity of the participants.

4.4 Findings

“It Is Kind of Like My Sanctuary:” Positive Perceptions Toward Physical Activity

Five participants demonstrated positive perceptions toward physical activity and it was described as an enjoyable pursuit. These youth were regularly engaged in a variety of activities, such as walking the dog, walking to school, or participating in activities at local facilities for people with illnesses. More importantly, they were characterized by particular experiences which appeared to shape their positive activity perceptions. These experiences included a) parental support, b) mastery experiences, c) enjoyment, and d) a sense of hope.

a) Parental support: Advocates for physical activity. Youth who displayed positive physical activity perceptions stated that they receive a great deal of family support, and described their parents as advocates for physical activity. When these parents encountered obstacles, for example, they engaged in problem solving in order to seek out activities which their child could successfully participate in and they encouraged youth to be active. Moreover, youth suggested that their parents were knowledgeable about the benefits of physical activity. For example, Graydon describes his mother as an educated CF expert and states that:

My mom is always reading things about CF - Internet sites and books and all sorts of stuff. She knows a lot about it, and she knows all of the physical activities and sports that I need to do, to be healthy.

Similarly, by providing her with physical activity opportunities at an early age, McKayla’s family has positively influenced her physical activity perceptions and behaviours:

My mom and dad taught me to like sports when I was younger. I have always been that way; I grew up that way. I am used to it. I was never the person who was like, “let’s play Barbie’s.” I was the one who was like, “let’s play football,” when I was six. I was always active and with the guys. I was always like that.
Thus, perceived family support and encouragement was the most important experience for those youth who displayed positive perceptions toward activity.

b) Enabling success: Mastery experiences in physical activity. Participants who displayed positive perceptions toward physical activity, also had memorable mastery experiences which empowered them with an enduring sense of competence and achievement. Most often, mastery experiences were derived from participation in individual activities rather than team sports and youth commented that they were better at such non-competitive activities. For example, Zoe displays a positive attitude toward dance. In addition to enjoying the social atmosphere in the dance studio and the music, she derives a sense of competence through dance:

I have a lot of friends there, I guess. I really like music, so that allows me to be around it a lot. It (dance) is really exciting, there is a lot of anticipation leading up to it. I like working hard to achieve things.

Similarly, Rudy is cognizant of the activities in which she cannot excel due to CF-related fatigue and she accepts her physical activity limitations. However, she makes an effort to seek out activities which are personally achievable and likely to result in a sense of mastery:

As I said, track and field - it is harder to run straight for 1000 meters, or to jump as high as other people. But I guess I just try to find stuff that I could do, you know? Things that I can personally achieve.

Reflecting on the mastery experiences that he has derived in the sport of basketball, 11 year old Koby states that … “I feel good, because in basketball, I am really into basketball. If I have like a good game, I want to play more.”

Thus, youth who regarded physical activity positively were characterized by mastery experiences. Indeed, these memorable experiences allowed them to attain a sense of competence.

c) Enjoyment. An inherent sense of fun and pleasure during physical activity was another experience associated with positive physical activity perceptions and these youth looked forward
to being active. For example, Graydon expresses his preference for individual activities, and, unlike competitive team sports, he suggests they result in a sense of fun and pleasure:

I do not play soccer or hockey or any of those. I am just not interested in them - I do not have as much fun when I am playing soccer or hockey. I have more fun running, being in the gym, or biking.

Similarly, Rudy emphasizes that a sense of enjoyment and fun are more likely to facilitate activity. She explains that when youth are pressured to be physically active, rather than encouraged, physical activity becomes a burdensome task that resembles work:

I like being active in stuff that is fun - not hard work. Hard work is more tiring, and, just “ah, I have to do that now”? I would not mind doing something with my friends, because that is fun. But if I had to go run a kilometre all by myself, I would not enjoy that.

Deriving fun during physical activity and a sense that experiences were self authored rather than forced, were experiences associated with positive physical activity perceptions for active CF youth.

d) A sense of hope. Finally, youth who displayed positive perceptions toward physical activity adopted illness narratives that were characterized by hope and optimism. For them, physical activity was instrumental to the attainment of health and provided proof that CF had not conquered them. For example, Gretchen is resilient and hopeful. Although there are moments when she wishes that she did not have CF, she does not feel limited or regard herself as different from other youth. Gretchen uses physical activity as evidence of her abilities, and participation serves as a way in which to challenge the dominant stereotype that youth with disabilities are incapable of engaging in physical tasks. Since physical activity provides evidence that CF has not defeated her, she draws upon spiritual metaphors and describes it as sanctuary:

I do not want to be treated differently … so what, I have CF. One of my cousins, he went
easy on me in sports, and I got really mad. I was like “fine, I have a disability, but I still want competition, so how will I know if I improve or not”? I want to know that even though I have CF, I don’t want that to stop me. I want to be able to achieve … when I do sports, I will be like “oh, I am not letting this stop me. Even though this is happening, anybody can do sports.” It is kind of like, my sanctuary.

Similarly, while Cassie acknowledges that her “lungs may not be great forever,” successful physical activity and sports participation is proof that CF cannot limit her, and she maintains positive perceptions toward physical activity:

I definitely do not think of myself differently because I have CF … I definitely do not lower my self esteem, because of that … the CF does not even bother me, and there is nothing physical that keeps me from doing anything. As long as I want to, there is nothing that is keeping me behind … I know that I can do it as long as I want to.

Victoria also demonstrates an enduring sense of hope and a positive attitude toward her health. Despite the fact that she was hospitalized as an inpatient at the time of her interview, she poignantly explains that CF has not detracted from her love for life:

Usually, I try to take that attitude, because what is the point - I am not going to keep drilling on the fact that I have CF because it is not really going to help with anything, and it is not going to make it go away. I would rather, you know, just love life, you know? … Obviously, it would be better if I did not have it (CF). You know, sometimes you wish that you did not have to go to the hospital at all, or whatever. But, you know, I guess that I have just kind of accepted it.

Thus, those youth who displayed positive perceptions toward physical activity were characterized by experiences of parental support, mastery, fun, and hopeful illness narratives.

“When I Am Really Sick, I Even Find Brushing My Teeth Difficult:” Negative Physical Activity Perceptions
Nine participants displayed negative perceptions toward physical activity. In addition, negative perceptions appeared to influence behaviour and these youth were physically inactive. Interestingly, although these youth were well educated about the benefits of participation, they were ambivalent regarding change and proposed logical explanations for inactivity. The experiences associated with negative physical activity perceptions included a) the detrimental impact of CF on physical activity, b) the belief that physical activity is not important, c) perceived lack of parental support for physical activity, and d) a sense of despair. Below, these experiences are discussed as sub-themes.

a) An “arduous burden” (Jessup & Parkinson, 2010): The detrimental impact of CF on physical activity. Unpleasant physical symptoms, such as breathlessness, coughing, and exhaustion, were the central experiences associated with negative physical activity perceptions. For example, Faisal explains that CF symptoms make it too difficult to be active and he believes that teachers and coaches would prefer to select youth who are stronger and better at sports:

   It (sports) uses all of my energy and it makes me feel tired, yes, it exhausts me … exercise makes me very tired. It makes me very drained, and makes me cough a lot. When I do exercise and I start to cough, I just continue coughing and coughing and coughing, and then I get a headache and a stomach ache.

Similarly, although she is aware of the benefits of physical activity for her health, Diana explains that CF symptoms make it too difficult to undertake even the basic activities of daily living, let alone physical activity:

   The physical therapist is just like “always stay active because it will increase your stamina.” But it is hard in the first place if you do not have enough stamina. Sometimes, when I am really sick, I even find brushing my teeth difficult.

As she recounts a recent experience playing soccer, Gretchen explains that unpleasant disease symptoms are frustrating because they compromise her ability to engage in sports:
It is kind of hard to breathe sometimes and you need to be able to breathe in order to play sports. That is one of the hard things … it is like “oh crap, I have to stop!” It is just that sometimes, it is really hard or that I will just pass out.

By making youth feel unwell and resulting in physical discomfort, the unpleasant symptoms associated with CF strongly influenced a negative physical activity orientation.

b) “What is the point?:” Low value ascribed to physical activity. Youth who displayed negative perceptions did not conceptualize physical activity as an important endeavor. Within the context of physical discomfort and social comparison, physical activity was not a valuable pursuit and they questioned the point of being active. For example, Diana states that:

I am not really good at sports. I know enough times from being sick and trying to run on the treadmill or something. I can’t. I feel really tired, so then I say, “if I am going to be tired, then why do it”? … having CF, I know that I should do it (activity) – it should be higher on the priority list. The more important things for me are like work and school … But physical activity is just something optional for me - I know that it should not be, but … because I know that it is hard, I do not want to make myself work hard.

Isabella concurs; the detrimental impact of CF makes her question whether she should engage in physical activity and whether it is a worthwhile pastime:

Sometimes, when I am running, I get more tired easily and faster than other people.

Sometimes that kind of makes me not want to always do sports … it is fun, and I want to, but I feel like it (CF) is kind of slowing me down. If it is slowing me down, then to keep doing the sport, it is not worth it.

Therefore, for the group of youth who were characterized by negative physical activity perceptions, the detrimental impact of CF made them question the point of being active.

c) Lack of parental support. Additionally, a perceived lack of family support and encouragement characterized the experiences of youth who regarded physical activity negatively. These youth
stated that parental involvement would facilitate increased physical activity and they expressed their desire to be physically active with their families. For example, Danny describes his family as lazy and expresses sadness when he remembers how active they were in the past. Danny expresses enthusiasm in becoming active with his family again:

   My family does have a big influence on me. My family is pretty lazy … we were all really active, but now, nobody does anything. Family is a big influence … if we all got active again, like if my dad bought bikes for all of us and was like ‘we are going to do a family bike ride once a week’ - I would be into that - that would be awesome.

Celeste also wishes that her family was more physically active together and she alludes to one of the important benefits of family-based or parent-mediated physical activity. Unlike other venues which may be characterized by competition and high performance expectations, family-based or parent-mediated physical activity is non-competitive and supportive …“if all of my family is going to go play one time in the week, going to play a sport, and there is no pressure, because it is just your family - I think that it definitely would benefit.”

   Although Koby’s parents are inactive, he suggests that parental support would facilitate physical activity … If my parents were more into physical activity, and we ran like two city blocks every few weeks or something, that would really help me.”

   Overall, youth with CF who relayed negative physical activity experiences were characterized by a perceived lack of family support for physical activity.

   d) A sense of despair. Finally, youth characterized by negative physical activity perceptions adopted narratives of despair and hopelessness in order to articulate their illness experiences. These narratives strongly shaped negative physical activity perceptions. For example, Faisal displays a less hopeful future orientation and his illness narrative emphasizes the ways in which CF accentuates difference; in relation to his peers, he feels thin and frail. He is excluded and ostracized and perceptions toward physical activity are framed through this
perspective. As he recounts his experiences of marginalization, he indicates that sports programs are designed for able-bodied youth:

I feel small, I feel skinny. I do not feel like I fit in with other kids. When I walk to school, I am the shortest kid—people ask me, “what grade are you in”? ... And I feel really sad, that they ask me that. Those are the two things that really keep me down ... and they think that I am bad and that I have some disease ... they talk rudely about me to themselves ... if it (sports programs) is for kids that are not sick—there is no point in going. It is all healthy kids, and they are active, and it is a place for them.

Diana also constructs a despairing illness narrative and her physical activity perceptions are refracted through this lens. Reduced physical functioning and observing that she is no longer able to do the same activities as she did in the past, serve as a painful reminder that her illness is progressing, that CF is “winning:”

I also know that I am not going to live as long as everybody else, so that is hard. I feel like it is out of my control, I feel helpless, how I used to be able to do it (physical activity), and now, I can’t. It is kind of depressing. It makes me think that it is a progressive disease, and it makes me think that it is getting worse ... it makes me worried.

For me, I do not want to get sicker.

The negative impact of CF, questioning the point of physical activity, lack of parental support, and despairing illness narratives characterized the experiences of these youth.

“No Time To Play” Finally, all participants expressed that due to the life limiting nature of CF as well as the time consuming and tedious nature of daily treatment, that they have less time available to them in relation to their peers and were “running out of time.” They alluded to reduced time in both a literal and symbolic sense. For example, although the mean life expectancy for patients with CF is 30 years, and many patients tentatively dream of such things as attaining a career or getting married (Jessup & Parkinson, 2010), participants understood
themselves as not living as long as their peers and questioned whether they would ever attain major developmental milestones. For this reason, reduced time had a metaphoric dimension. Similarly, due to the daily hassle of managing CF, such as medication-taking and physical therapy, youth discussed reduced time in a literal sense. Youth indicated that while necessary, daily treatments were time-consuming and hateful. Within the context of having to engage in such time-consuming treatments, there was a lack of available time for physical activity. Youth preferred to use this little, precious time for more important activities such as spending time with family and friends.

For example, as he describes his experiences at summer camp, Graydon’s narrative is punctuated with anger. By preventing him from engaging in camp activities with his peers, he explains how physical therapy robbed him of time:

I hate when it (physical therapy-PT) gets in the way of all of these things. At camp, it really gets in the way when you wake up and the bell goes and you have to go to breakfast. But I have to do PEP (breathing mask), and that holds me back. Or, it is night time and they are all going to go out somewhere. And maybe I have to go back to the cabin and get my PEP which is really far away. It is just annoying … that was the worst and I hated it so much … I know that I need to do physical activity, but it is just sometimes hard when things interfere, like medicine or PEP.

Gretchen also suggests that physical therapy is time consuming. She imagines that if it were shorter, she would have more time for activities such as outdoor play, exploration, or swimming:

The disadvantage of it, is that it takes a while and with extra-curricular activities, there is a time restraint. It would be cool (physical therapy), if it was a little bit shorter, so that I could be able to do some more stuff … sometimes, I would rather be outside playing-going for a swim or playing with my sister, or just going outside. But I am OK with it (PT), and I have been able to adjust. But it would be cool if it was shorter.
McKayla explains that the time consuming nature of PT detracts from time available for more enjoyable social pursuits, such as spending time with her friends or engaging in sports:

The bad things about PT is that I do not really have the time and it takes time. It just seems that I am always going in sports, or homework and that I never have time for PT. It is frustrating, because I want to be with my friends, but I have to do PT … I tell them (friends), and they are like “OK, call me when you are done “or whatever. Yah, it does not really matter, because I do it all the time - but sometimes, it gets me mad and I hate it.

Danny no longer adheres to PT treatments. Negative childhood associations, the fact that PT is a reminder of living with a fatal condition, and the time consuming nature of PT, are experiences which help explain his resistance and constitute a “logical explanation.” Danny worries that PT detracts from time available for physical activity:

I hate doing PT. When I was a kid, there was a round board and I would lie on it while my mom wacked my chest. Like a beating with a cupped hand - I hated it. Ever since I was little and she did that, and I knew that it was PT, I have despised it. It takes away from my time, and that is something that I do not like. When you have to sit down and do your mask for 30 minutes, and the PEP mask for 20, it drives the nail home that you do have a lung disease and sometimes, I just want to forget about it … as I am doing it- “wow, I have to do this, to prevent myself from dying.” It takes away from my time, for sports, friends, and family.

Similarly, while Cassie enjoys relatively good health at the moment, she understands that her grasp on health with CF is ultimately tenuous and fragile. She foresees that in the future, she will have to negotiate “physical activity time” within the context of her health status and that CF may ultimately compromise her ability to engage in physical activity:

I cannot see into the future, and I cannot tell how much CF is going to affect me.

Obviously, my lungs will not be amazing forever. I will try to do as much activity as I
can, to make sure that for a while, I can still do it. Of course, I know that in the future-hopefully, I will be lucky and I will be able to do activity for a while. Hopefully, many years and decades. For sure, in the future, it could prevent me from doing certain things, like physical stuff.

Youth with CF were aware of reduced life expectancy and how treatment detracted from available time. Physical activity occurred within the context of reduced time, and, in relation to the hassle of “hateful” CF treatment, there was less available time for physical activity. Youth wished to spend this time engaging in more important activities.

4.5 Discussion

This study addressed crucial limitations in the existing literature and has advanced our understanding of how youth with CF experience and perceive physical activity. The detrimental impact that CF had on the ability to be physically active, was one of the most significant experiences associated with negative physical activity perceptions. CF symptoms, such as coughing and breathlessness, were consistently reported as negative experiences and engaging in physical activity was not desirable if it was thought to exacerbate these unpleasant symptoms. Moreover, some youth alluded to the vicious cycle of inactivity that has been documented in the literature - or the “chicken or egg” dilemma that tends to characterize physical activity participation for youth with CF (Nixon et al., 2001; Selevdurai et al., 2004). Indeed, youth explained that breathlessness and coughing leads to activity avoidance; in turn, inactivity further exacerbates CF symptoms. The finding that disease symptoms are a significant barrier to physical activity for youth with CF and other chronic diseases is well documented in the literature (Moola et al., 2008; Swisher & Erikson, 2008). When youth feel unwell, they are unlikely to want to be physically active and disease symptoms lead to activity avoidance.

Furthermore, youth with CF adopted various illness narratives that appeared to influence physical activity perceptions (Sparkes, 2004). For example, some youth were resilient and
optimistic about CF and displayed a positive orientation toward physical activity. Drawing upon culturally dominant narratives of restitution, these youth believed that the attainment of health was possible and that they would not succumb to CF. Rather, they were convinced that CF-related obstacles would be overcome and that they would garner strength and insight through the process. Moreover, physical activity provided evidence that CF had not conquered them and was a testament to their will and resolve to fight. In contrast, other youth adopted chaos narratives in order to articulate their negative perceptions toward health and physical activity; and, in doing so, they conveyed a sense of desperation, futility, helplessness, and worry about the future. Recounting experiences of pain, hardship, and exclusion, these youth were deeply cognizant of their suffering and troubled by the terminal nature of CF. In turn, these despairing illness narratives influenced negative physical activity perceptions; it was an unpleasant reminder of CF. Alternatively, physical activity was regarded as an able-bodied site which emphasized exclusion.

As suggested by Smith and Sparkes (2008), these despairing chaos stories engendered fear and lacked a structured plot and feel-good overcoming narrative. For this reason, they invoked a sense of “narrative wreckage” and were difficult to hear. Finally, although “quest narratives” consider illness as a transformative process that profoundly changes the self, no participants adopted this narrative structure, and, rather, illness narratives appeared to be characterized by the restitution and chaos types only (Frank, 1995). Indeed, a quest or polyphonic illness narrative is characterized by a philosophy of the present and a fundamental change in values and beliefs, making it possible to both enjoy the present without investing in or depending on the future (Ezzy, 2000). While this may ultimately be a very helpful and positive way of storying CF, this narrative structure may evolve over the course of time, and did not appear to be accessible to youth with CF in this study. Other literatures have illuminated the despairing nature of the narratives that characterize other chronically ill youth and people with
disabilities (Smith & Sparkes, 2008; Yoos & McMullen, 1996), and, in this regard, the concept of illness narratives informed by the sociology of health and illness, lent interpretive insight into the findings.

Finally, this study provoked novel insights into how youth with CF experience physical activity within the context of reduced time. Indeed, although time was not a concept included in the research question or interview guide, it emerged as an important analytical point for further inquiry and was a concept that youth dialogued about and constantly grappled with. Within the context of living with a terminal illness, youth with CF spoke of time metaphorically and literally and illustrated their capacity to think in non-literal and abstract ways. Youth perceived their treatments as necessary but time consuming and unpleasant, and, as a result of such treatments, they understood themselves to have less time. Indeed, physical therapy was described as an arduous and at times hateful burden which results in less available time for physical activity. Moreover, youth preferred to repartition their limited precious time elsewhere and did not regard physical activity as a worthwhile time investment.

Other qualitative studies have illustrated the complex time tricks which children with CF negotiate and the ways in which their illnesses contribute toward a loss of temporal integrity (Glasscoe & Smith, 2008), or a shattering of one’s sense of time and how it passes. While time in a Newtonian sense is linear, chronic illness illustrates the iterative, cyclical, and fragmented nature of time for these youth. They are described as both diminutive and old souls at one and the same time. For example, having missed critical developmental milestones, they are always lagging behind. However, having experienced traumatic health encounters at tender ages, they are wise beyond their years and have mastered a complicated and technical medical jargon that is uncharacteristically childlike. By moving perceived time forward and backward and highlighting particular health-related time periods as salient, chronic illness may re-set the clock (Glasscoe & Smith, 2008). Similarly, by explaining the tentative and precarious nature of the future for
patients with CF in their phenomenological study of the body, space, time, and relationships, Jessup and Parkinson (2010) allude to the temporal dilemmas encountered by young people with CF and the salience of time to their lives. For example, at the time of diagnosis, the future is effectively cancelled and CF patients and their loved ones are confronted with the finality of the disease, the heart wrenching task of explaining death on to those that one was supposed to bestow life. However, as patients become aware of the medical advancements in CF care, they are permitted to tentatively dream about a provisional future. Thus, grappling with temporal issues is central to the experience of living with CF and finding the time for physical activity is, arguably constrained by youths’ weighty temporal dilemmas.

4.6 Recommendations

Youth with CF reported that disease symptoms restricted them from engaging in physical activity and they often felt too unwell to participate. While the benefits of physical activity for this population are well documented, health professionals must remain sensitive to the physical discomfort which de-trained youth experience during initial efforts to become active. Since perceptions of pain, fatigue, and breathlessness strongly influence non-participation, health professionals should recommend and promote physical activities which gradually increase fitness while minimizing physical discomfort, and remain sensitive to the burden of disease. Additionally, health professionals should be aware of the various illness narratives which youth construct in the process of making meaning as they come to terms with CF. In addition to being aware of how illness disrupts a sense of self and identity (Taylor & Field, 2003), it is important to understand that the narratives which youth construct to explain their illnesses strongly influence their physical activity perceptions. More importantly, since hopeful illness narratives appear to shape positive perceptions toward physical activity, health professionals should work with youth in order to develop more positive and functional ways of understanding and interpreting their illnesses. Given that youth with CF experience physical activity within the
context of reduced time, health professionals need to be aware of the complex way in which these youth bring meaning to the concept of time and display sensitivity to youths’ perceived time constraints. Health professionals should devise physical activity programs in a manner which is not perceived to be wasteful of time and the medicalization of physical activity should be avoided so that youth do not come to regard it as just another time consuming treatment.

Moreover, all participants underscored the importance of parental support for physical activity, a finding which is elaborated upon in the following chapter, Study Two. Parental encouragement was integral to physical activity for active youth with CF. In contrast, inactive youth lamented a lack of familial support for physical activity and wished that their parents were more active with them. Health professionals should thus consider parent-mediated intervention approaches, in which parents are actively engaged in youths’ physical activity (Fiese, 2005).

Finally, due to clinical concerns regarding the risk of cross contamination at the research site where this study was conducted, youth with CF are not advised to engage in physical activity together. From an adapted physical activity perspective, this is challenging. Youth with CF are arguably a more socially isolated group and typical approaches to forming physical activity social groups that allow youth to share illness narratives and engage in activity together, are not feasible. Indeed, the medical needs of this population must be carefully balanced with the provision of social support, and call on health professionals to devise novel physical activity approaches. Internet mediated physical activity which socially connects youth with CF to each other without compromising physical health, such as facebook or skype, are potential options.

4. 7 Limitations

Although youth with CF strongly supported the assertion that parental support is integral to physical activity through processes such as role modeling and facilitation (Prasad & Cerny, 2002), the actual level of parental physical activity knowledge and support cannot be ascertained from the available data and is a study limitation. More importantly, although lack of parental
support shaped negative perceptions toward physical activity for youth, the numerous barriers which these non-active and non-involved parents may encounter—such as a lack of knowledge about physical activity or socio-economic constraints—is unknown. Health professionals should resist the urge to blame parents for physical inactivity. Rather, it is important to consider the barriers which may prevent parents from being able to provide their children with safe and enjoyable opportunities. Clearly, further research is required in order to explore parental perceptions toward physical activity in youth with CF, and assisting those parents who are not able to support their children’s efforts to be active, should be a future priority for researchers and clinicians. The lack of objective indicators of disease severity was another study limitation. Without such information, it was not possible to ascertain whether disease severity influences perceptions toward physical activity.

4.8 Conclusion

The relationship between aerobic capacity and survival (Selevadurai et al., 2002) has sparked interest in the area of physical activity youth with CF. Despite the benefits however, youth with CF display low levels of physical activity (Nixon et al., 2001). This field is characterized by the absence of theoretically informed qualitative research which lends insight into why these youth are inactive, and this study sought to address these limitations. In conclusion, the participants displayed either positive or negative perceptions toward physical activity and all youth experienced physical activity within the context of reduced time. By illuminating how youth with CF perceive and experience physical activity, the findings call attention to how youths’ complex psycho-social realities, such as temporal dilemmas or illness narratives, may operate in tandem with reduced physiological health to constrain physical activity and contribute toward reduced participation. When planning physical activity for youth with CF, programs must consider their complex psycho-social realities and experiences. Doing so may contribute toward the development of more comprehensive and meaningful physical activity programs that are
sensitive and tailored to the social lives, worlds, and dilemmas of youth with CF.

In the findings discussed above, it was found that *parental support* is critical to physical activity participation among youth with CF. By examining how the parents of CF and CHD youth understand and negotiate physical activity for their child and themselves, the following study in this sequential series of manuscripts further explores, extends, and broadens this potential line of inquiry.
Chapter Five, Study Two:
Developing Physical Activity Interventions for Youth with Cystic Fibrosis and Congenital Heart Disease: Learning from their Parents

Yes, I think his scarring has affected him. Mind you, he is still swimming and he will throw off his top to go into the pool. I find that people ask me “what is wrong with him?” … A couple times, it has taken him aback. I am so used to it that I do not think of it. I will be like “what do you mean?” And they say that “he is all scarred” and I will be like, “oh”? I just say that “he is a miracle kid. He works on half a heart. What do you mean he has half a heart? …” It has affected us in some ways (CHD7).

5.1 Introduction: The above quote exemplifies one CHD parents’ perspective regarding her child’s physical activity. Indeed, despite the important instrumental and emotional support function of parents in the lives of children with chronic illnesses such as CF and CHD, little is known about their perceptions toward their own - and their child’s - physical activity. In the study described below, I chronicle and interpret how the parents of youth with CF and severe heart disease understand and interpret the role of physical activity in their own and their child’s life. In doing so, I discuss the numerous benefits of physical activity “for child and self,” and the importance of parental role modeling in facilitating participation. In addition to discussing the numerous barriers that prevent parents and children from being active, I explore the broader family context - that is characterized by a prevailing sense of chronic stress and complexity – in which CF and CHD families negotiate physical activity. Before doing so, a review of the literature is undertaken below.

5.2 Review of the Literature: Physical Activity for Youth with CF and CHD
Cystic fibrosis (CF) is an autosomal recessive genetic disease which is characterized by the accumulation of mucous secretions in the lungs and malnutrition. Treatment entails medicine,
physical therapy, and nutritional supplementation (Nixon et al., 2001). Because there is no cure, treatment is palliative rather than curative. In contrast, congenital heart disease (CHD) is characterized by structural defects of the heart which occur during heart development in utero and vary significantly from case to case along a continuum (Sparacino et al., 1997). The development of novel treatments has increased the median age of survival for CF patients to 31 years (Swisher & Erikson, 2008). Similarly, advances in surgical procedures have reduced mortality among patients with CHD such that approximately 85% of patients are expected to reach adulthood. Given the changing patterns of disease, CF and CHD can be considered chronic childhood illnesses that affect daily health and functioning.

Despite increased survival, these youth suffer from reduced quality of life and psychological health impairments, such as depression, anxiety, behavioural disorders, covert fears, poor quality of life, and diminished self-esteem (Berge et al., 2007; Moons et al., 2004; Spurkland et al., 1993). As a result of the psychological implications of chronic disease, there is interest in developing novel interventions that may improve quality of life.

Physical activity has been proposed as a therapeutic mechanism through which to enhance quality of life and diminish psychological impairments. Physical activity and exercise is associated with numerous health benefits for children and youth with CF and CHD. For example, for CF patients, participation is associated with improvements in pulmonary function, aerobic fitness, and muscular strength. Given the beneficial impact on VO2 peak, physical activity may be associated with reduced mortality and increased survival (Schneiderman-Walker et al., 2000; Van Doorn, 2010). For patients with CHD, participation in exercise and physical activity may improve maximum oxygen consumption, exercise capacity, and physical functioning (Longmuir et al., 1990; McCrindle et al., 2007).

In addition to the physical and functional benefits, physical activity is associated with enhanced psychological health and quality of life in these populations. For example, physical
activity improves role functioning due to behavioural and emotional problems, general health, mental health, and quality of life (Moons et al., 2005). Physical activity may also enhance perceptions of mastery and normalization. Youth with CF who engage in physical activity report that it is an enjoyable and preferred treatment modality, and it follows that perceptions of quality of life improve as a result of participation (Blau et al., 2002; Selvadurai et al., 2002). Indeed, as MacDonald and Greggans (2010) suggest, CF youth may desire to … “explore the boundaries of their physicality and have adventures - to do things that they would not normally do” (p. 2411).

However, despite the benefits associated with physical activity, patients consistently report low frequencies of participation and low intensities of exertion. For example, CF and CHD patients are less physically active than healthy children and do not participate in activities that are vigorous, thus depriving them of the benefits associated with intense activity (Lunt et al., 2003; McCrindle et al., 2007; Selvadurai et al., 2004). It appears that these youth encounter numerous barriers that occur at individual, parental, social, and structural levels. These barriers include the detrimental impact of disease symptoms (Moola et al., 2010; Swisher & Erikson, 2008), low self-efficacy for physical activity (Lunt et al., 2003; Moola et al., 2008), and the attribution of low value and importance toward physical activity (Lunt et al., 2003; Moola et al., 2008). Poor inclusion in physical education, bullying, teasing, and social stigma, fear of the legal ramifications if a cardiac event occurs on school property, and a lack of physical activity opportunities for ill youth in the community, are also significant barriers (Goodwin & Staples, 2005; Moola et al., 2010; Moola et al., 2009).

Of particular interest is the role of the parent or primary caregiver in encouraging and supporting their child (ren) to be physically active. In the context of children with chronic diseases, a pervasive psycho-social physical activity barrier is parental over protection. Termed the “vulnerable child syndrome,” the parents of youth with chronic diseases may worry excessively about their child’s health and their capacity to engage in physical activity (Wilkes et
al., 2009). Whether fears are justified by probable risks associated with physical activity or not, parents may worry that their sick child is too weak to engage in physical activity and feel that their child may become injured or ill if they participate. Perceiving their child to be weak and fragile, parents may unwittingly coddle them and prevent them from participating. Furthermore, parents may not hold the same standards for success as they do for their healthy, “able bodied” children and inadvertently restrict them from attaining their full potential. Illuminating the powerful impact of parental attitudes and beliefs on child physical activity, researchers have found that maternal attitudes toward physical activity for cardiac youth are a stronger predictor of participation than the severity of the cardiac lesion (Bar-Mor et al., 2000). It is likely that parents’ attitudes and beliefs about their child’s physical health affect the capacity and willingness of their children to participate in physical activity.

5.3 Engaging Parents: Considering Parents in the Physical Activity of Children and Youth with CF and CHD

Literature from the field of pediatric obesity has underscored the importance of engaging parents and eliciting their involvement when considering physical activity among children with chronic diseases. By either facilitating or hindering their child’s ability to participate, parents have been described as critical gatekeepers to health behaviour change (Golan, 2006). Indeed, parents provide opportunities for physical activity and are thus critical to enabling their child to be active. Parental attitudes and beliefs strongly influence youths’ physical activity behaviours, and, via processes such as role modeling and observational learning, children may adopt their parent’s physical activity practices. The familial context has a great influence on children’s physical activity. Processes such as dialoguing about physical activity in the home, how active the home is, the availability of physical activity equipment in the home, and the accessibility of the home to places in the neighbourhood to be active, are contextual influences that impact children’s ability to engage in physical activity. Moreover, although hospital-based
interventions ensure the high degree of supervision necessary for optimal compliance, such programs may lack ecological acuity and contextual relevance to participants’ social lives and worlds (Golan, 2006; Schneiderman-Walker, 2000). Interventions which involve parents are arguably more in tune with the nuances of participants’ local environments. Finally, given the genetic and behavioural similarities between members of the same family, intervening at the family level is likely to have benefits for all members (Nader, 1989).

Including parents in physical activity interventions not only has implications for the child’s health, but also for parents themselves. Because caring for a chronically ill child has been associated with poor physical and psychological health of the caregiver, engaging parents is critical. For example, in addition to lower immune functioning, higher rates of cardiovascular disease, as well as higher rates of mortality, caregivers are at a greater risk of developing psychological problems and disorders, such as depression, anxiety, insomnia, and symptoms of post traumatic stress (Barlow, Powell, Gilchrist, & Fotoadou, 2008; King & Brassington, 1997; Sawyer, Antonious, Toogood, Rice, & Baghurst, 2000). Caregiver distress may be particularly heightened during the initial shock of diagnosis but may gradually dissipate over time. However, the detrimental impact of the illness on family relations may escalate after diagnosis. Qualitative studies have revealed that parents experience their child’s illness as “life shattering, life altering, and self-obliterating” (Woodgate & Degner, 2003). Within the context of medicalized health care technologies, these parents are expected to demonstrate “constant vigilance” and perfect parenting, in which all of their time is devoted toward caring for their chronically ill child and properly monitoring their child’s treatment (Hafetz & Miller, 2010; Rempel & Harrison, 2007; Sparacino et al., 1997). Parents also struggle to come to terms with such questions as whether or not to disclose their child’s condition to others, what kind of expectations they should have of their child, as well as whether or not they should allow themselves to become attached to a child that will eventually die (Sparacino et al., 1997). Parents also struggle to understand whether their
child is normal or not and may feel resentment toward the child. As such, the evidence suggests that given the marked stressors associated with caring for a chronically ill child, the physical and psychological health of caregivers is affected.

Physical activity affords health benefits to caregivers. For example, engaging in physical activity may assist caregivers in attaining the appropriate levels of fitness that are required for the physical task associated with caregiving. Physical activity may afford caregivers with other important psychological benefits, such as reduced stress, distraction from caregiving, and improved quality of life (Farren et al., 2008; King & Brassington, 1997). However, physical activity has been tangential to studies that explore the experiences of parents who care for sick children. Although parents usually describe their fears associated with their child’s engagement in physical activity, or the difficulty associated with observing their child’s inability to keep pace with their peers (Sparacino et al., 1997), physical activity has not featured centrally as a topic for exploration in these studies.

In summary, physical activity levels of children with CF and CHD are less than optimal despite the important benefits that greater participation would confer. Existing research points to the important gate keeping role parents will play in supporting their children to access physical activity opportunities. Physical activity may also provide important psychological and physical benefits for parents themselves within the context of their caregiving demands. In order to develop behavioural interventions that are attractive, acceptable, and feasible for these families, it is essential to talk to parents and understand their experiences of caring for a child with CF or CHD. If interventions are not conceptualized keeping in mind the very real challenges and fears parents and their children face - and if they are not tailored to address common barriers and facilitators of physical activity - they may not be successful. Thus, the specific purpose of this study was to explore the perceived benefits and barriers of physical activity among parents of children with CF and CHD and identify implications for intervention development. Although
CHD and CF are both chronic childhood illnesses, CF is a progressive and life limiting condition with a high treatment burden (Berge et al., 2007). I was particularly interested in exploring the similarities and differences among CF and CHD parents. Identifying what is common across disease states may allow future theory and intervention development that can be applied across the spectrum of diseases while remaining sensitive to the unique needs associated with specific diseases.

5.4 Methods

Conceptual Framework

Family systems theory was the conceptual framework that guided this study (Murray, Kelley-Soderholm, & Murray, 2007). According to family systems theorists, the family might be considered as an open, dynamic, and ever changing system, in which each member’s actions bear an influence on all other members. Family system theorists aim to decipher the complex patterns of communication and roles that characterize families, and to recalibrate these patterns if particular subsystems are no longer functioning optimally. Family systems theorists are likely to consider behavioural problems - such as physical inactivity - not as a result of an individual’s failing, but, rather, as a response to the broader family context. Since family systems theory emphasizes the dynamic interaction between children and parents (Murray et al., 2007), as well as how children’s physical activity behaviours are reinforced by the family context, it was an important and relevant theoretical framework for this study.

Research Setting and Participants

This study was approved by the Research Ethics Board at the Hospital for Sick Children in Toronto, Canada and took place in the Cardio-pulmonary Exercise Lab and the Labatt Family Heart Centre. Twenty nine parents (16 parents from the CF clinic and 13 parents from the CHD centre) participated. Although all of the participants provided care to a child with CHD or CF, child medical history and disease severity varied. For instance, while some children had never
been hospitalized, others had been hospitalized on several occasions and had undergone numerous surgeries or procedures. Most parents had more than one child and provided care to only one sick child. However, three of the participants in the CF sample and one of the participants in the CHD sample were providing care to two children with chronic diseases.

Parents were between the ages of 35 and 55 years. The sample was gender balanced and ethno-culturally diverse. Although measures of parental health status were not obtained, most parents described themselves as healthy. A few parents, however, were dealing with health problems such as excess weight, diabetes, hypertension, or physical injury. In terms of employment, parents fulfilled a number of occupational roles. Some parents were professional white collar workers, such as nurses, teachers, or consultants. Other parents were engaged in blue collar jobs such as the skilled trades, sales, trucking, or farming. Additionally, a few parents were unemployed and receiving unemployment insurance. The Children’s Aid Society (CAS) was involved in one family, in which the level of socio-economic deprivation and ability to care for the sick child were judged to be inadequate. Some parents described themselves as “stay at home parents” and had made this decision in order to provide care for the sick child. While some participants were in long term married relationships, half of the sample was divorced, remarried, and/or single. Additionally, parents were from a range of urban and rural locations across Ontario and Quebec and access to physical activity opportunities varied.
### Table Two: Participant Descriptions, CF Parents

<table>
<thead>
<tr>
<th>Code</th>
<th>Birth Year</th>
<th>Gender</th>
<th>Occupation</th>
<th>Child age</th>
<th>Child Gender</th>
<th>Child Health Status</th>
<th>Hospitalization</th>
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</thead>
<tbody>
<tr>
<td>CF1</td>
<td>1964-1966</td>
<td>F/M</td>
<td>Nursing Web development</td>
<td>12</td>
<td>F</td>
<td>Good</td>
<td>Out</td>
</tr>
<tr>
<td>CF2</td>
<td>1959-1959</td>
<td>F/M</td>
<td>Analyst EI</td>
<td>13</td>
<td>M</td>
<td>Good</td>
<td>Out</td>
</tr>
<tr>
<td>CF3</td>
<td>1960</td>
<td>F</td>
<td>Caregiver</td>
<td>18</td>
<td>M</td>
<td>Fair-Poor</td>
<td>In</td>
</tr>
<tr>
<td>CF4</td>
<td>1964</td>
<td>F</td>
<td>Nurse</td>
<td>17</td>
<td>M</td>
<td>Good</td>
<td>Out</td>
</tr>
<tr>
<td>CF5</td>
<td>1965</td>
<td>F</td>
<td>Educational assistant</td>
<td>14</td>
<td>M</td>
<td>Good</td>
<td>In</td>
</tr>
<tr>
<td>CF6</td>
<td>1963</td>
<td>F</td>
<td>Cosmetic sales</td>
<td>17</td>
<td>M</td>
<td>Good</td>
<td>Out</td>
</tr>
<tr>
<td>CF7</td>
<td>1967</td>
<td>F</td>
<td>Tim Horton’s</td>
<td>9****</td>
<td>F</td>
<td>Good</td>
<td>Out</td>
</tr>
<tr>
<td>CF8</td>
<td>1972</td>
<td>M</td>
<td>Sales rep</td>
<td>7</td>
<td>F</td>
<td>Good</td>
<td>Out</td>
</tr>
<tr>
<td>CF9</td>
<td>1970</td>
<td>M</td>
<td>Consultant</td>
<td>11</td>
<td>F</td>
<td>Fair</td>
<td>In</td>
</tr>
<tr>
<td>CF10</td>
<td>1975</td>
<td>M</td>
<td>Consultant</td>
<td>13</td>
<td>F</td>
<td>Poor</td>
<td>Out – transplant list</td>
</tr>
<tr>
<td>CF11</td>
<td>1961</td>
<td>F</td>
<td>Caregiver</td>
<td>15</td>
<td>M</td>
<td>Good</td>
<td>Out</td>
</tr>
<tr>
<td>CF12</td>
<td>1963</td>
<td>F</td>
<td>Customer service</td>
<td>17</td>
<td>F</td>
<td>Poor</td>
<td>Out</td>
</tr>
<tr>
<td>CF13</td>
<td>1963</td>
<td>F</td>
<td>Nuclear energy worker</td>
<td>14</td>
<td>F</td>
<td>Good</td>
<td>Out</td>
</tr>
<tr>
<td>CF14</td>
<td>1961</td>
<td>M</td>
<td>Computers</td>
<td>12*****</td>
<td>F</td>
<td>Good</td>
<td>Out</td>
</tr>
</tbody>
</table>

*** Two children with CF

### Table Three: Participant Descriptions, CHD parents

<table>
<thead>
<tr>
<th>Code</th>
<th>Birth Year</th>
<th>Gender</th>
<th>Occupation</th>
<th>Child Diagnosis</th>
<th>Child Age</th>
<th>Child Gender</th>
</tr>
</thead>
<tbody>
<tr>
<td>CHD1</td>
<td>1970</td>
<td>M</td>
<td>Sales</td>
<td>TOF with absent PA</td>
<td>8</td>
<td>M</td>
</tr>
<tr>
<td>CHD2</td>
<td>1969</td>
<td>F</td>
<td>Banking</td>
<td>HLHS</td>
<td>9</td>
<td>F</td>
</tr>
<tr>
<td>CHD3</td>
<td>1963</td>
<td>M</td>
<td>Inspector</td>
<td>Aortic Atrésia; ICD implant</td>
<td>17</td>
<td>M</td>
</tr>
<tr>
<td>CHD4</td>
<td>1959</td>
<td>M</td>
<td>Electrical engineer</td>
<td>TOF</td>
<td>17</td>
<td>F</td>
</tr>
<tr>
<td>CHD5</td>
<td>1961</td>
<td>F</td>
<td>Finance manager</td>
<td>TGA</td>
<td>17</td>
<td>M</td>
</tr>
<tr>
<td>CHD6</td>
<td>1964</td>
<td>F</td>
<td>Teacher</td>
<td>TOF; Pulmonary Atrésia</td>
<td>13</td>
<td>M</td>
</tr>
<tr>
<td>CHD7</td>
<td>1961</td>
<td>F</td>
<td>Dental sales rep</td>
<td>Fontan</td>
<td>16</td>
<td>M</td>
</tr>
<tr>
<td>CHD8</td>
<td>1956</td>
<td>F</td>
<td>Caregiver</td>
<td>TOF</td>
<td>16</td>
<td>M</td>
</tr>
<tr>
<td>CHD9</td>
<td>1968</td>
<td>F</td>
<td>El</td>
<td>TOF</td>
<td>15</td>
<td>M</td>
</tr>
<tr>
<td>CHD10</td>
<td>1957</td>
<td>F</td>
<td>Jeweller</td>
<td>Truncus Arteriosus</td>
<td>18</td>
<td>F</td>
</tr>
<tr>
<td>CHD11</td>
<td>1957</td>
<td>F</td>
<td>Sales</td>
<td>CoA</td>
<td>16</td>
<td>M</td>
</tr>
<tr>
<td>CHD12</td>
<td>1966</td>
<td>F</td>
<td>Book keeping</td>
<td>Truncus Arteriosus; brain injury</td>
<td>17</td>
<td>F</td>
</tr>
<tr>
<td>CHD13</td>
<td>1975</td>
<td>M</td>
<td>Landscaping</td>
<td>TOF</td>
<td>15</td>
<td>F</td>
</tr>
</tbody>
</table>
Data Collection

In-depth semi-structured interviews were the primary means in which data was collected (Mason, 2002). Participants were phoned or approached directly in the clinic in order to request participation and informed consent was obtained. The interviews occurred in staff offices or clinic rooms and were 45 to 85 minutes in duration. Informed by the review of the literature, a general interview guide was developed. Although the interview guide was semi-structured in nature, participants’ responses drove further lines of inquiry. Interviews were audio-taped and transcribed (APPENDIX Two). The qualitative method which informed this study is described below.

Thematic Analysis

Informed by the work of Braun and Clarke (2006) and Boyatzis (1998), thematic analysis guided the development and analysis of this study and provided a useful method for understanding how the parents of youth with CF and CHD understand their child’s physical activity. Similar to Study One, the use of thematic analysis in this study was informed by a constructivist ontology and an interpretivist epistemology, and family systems theory provided a framework for thinking about the data. For a detailed and reproducible explanation of this qualitative method, and a more thorough explanation about my methodological decisions, please refer to the methodology outlined in Study One (Chapter Four).

Below, I provide a story about the data both within and across themes. In doing so, as an analyst who is both a cultural member and critical commentator, I offer a story that describes, explains, and interprets how the parents of youth with CF and CHD understand physical activity - both for their child and themselves and their perceptions of their child’s psychological response to physical activity. The findings are illustrated in Figure Three below. Codes have been used in order to protect the anonymity of the participants.
5. 5  
Findings  

*The Family System: Stress and Complexity*

In talking to parents about the benefits and barriers to physical activity, it was evident that such discussions could not occur without considering and appreciating the complexity, as well as high levels of stress, that characterized the experience of participants. The lives of most participants were characterized by separation, divorce, re-marriage, or even the death of a spouse. Although the reasons for parental separation were not explored in this study, in some cases, parents chose to disclose explanations to me. For example, in some cases, divorce was attributed to the chronic stress associated with caring for an ill child, and, more importantly, the inability for the couple to come to a consensus regarding how the ill child should be parented. Additionally, the ill child also played other roles in the marital relationship. For example, by revealing how one responds to stress, the ill child was thought to illuminate the spouses’ “true colours” and “authentic self,” and, interestingly, the parent was often grateful to the child. In other cases, parents felt that ending the relationship would add more stress to the life of a child.
who was already ill and they experienced guilt. This ensured that parents remained in an unhappy relationship for a lengthy period of time:

I just separated from Harriet’s dad. And that was me questioning, “is it OK to put myself first”? All along, it was like “no, she will be better off because we are in Toronto and close to SickKids.” But then I said, “but her disease is also crippling me. I know that I can be a better mother if I am happy.” That was such a huge thing. She has had a lot of changes in the past few years. I wanted to make things as easy as possible for her (CF14).

In this regard, marital failure tended to characterize the experiences of parents.

Parents also described feelings of stress, fragility, helplessness, grief, anxiety, and despair over their child’s health. For example, in some cases, participants repeatedly stated that they were at their “wits end” and “did not know what to do” about their ill child’s non-adherence to treatment. Expressing that they “worried about the child all the time” and did not know how to “reach” their child, it was evident that in some families, there was a dysfunctional relationship between the child and parent family subsystem. This resulted in grief, despair, and anxiety over their child’s health:

I would have to say that it has impacted our life for us as parents. It has constantly been on our mind. “When will he get sick? Will he die?” Because when you look at him, he looks healthy and he is “go, go, go” so it does not seem to be an issue. Him coming here on Saturday (hospital) has made a big impact even on his siblings ... He (husband) and I are constantly asking ourselves - “what would we do if- how would we prepare for”? That kind of stuff. Jessa (daughter) and I have talked about that too. “What would we do if? How sad would that be if?” (CFP5).

Similarly, Diana’s mother expresses sadness and fear regarding her child’s health. She lacks trust and does not believe that her daughter is adequately adhering to her life saving treatment protocol. This has led her to reluctantly snoop through her child’s garbage can at night.
in order to ensure that she has taken her pills. Her suspicion that her daughter has been medication skipping has been confirmed, resulting in sadness and confusion:

Stress is always there. It is like carrying this weight … it is not nice to say. It is the confidence. I do not have the confidence that she will follow through with all of it. I do not trust her with it and I know that she is not doing it (treatment). I go through her garbage, not through it, but I know with the Kleenex and I can tell about her doing her physiotherapy. She only did three. She only did three and she is supposed to do five. “Why not do six? Why not do an extra one”? You are just sitting there (CF12).

Another mother also expressed utter anger and frustration over her child’s inability to take care of his health and treatment, and is indicative of stress in the parent-child subsystem:

I have tried everything that I can with him, short of taking everything out of his room (to punish him). I do not know if I can do that. We will just be fighting all of the time. It just causes fighting and I do not want to do that with him (CHD8).

In this regard, parents experienced stress, anxiety, and grief over their child’s health and were often engaged in heated battles with their child over his/her adherence to treatment.

Finally, parents were celebratory of their child’s health professionals and grateful to staff for the excellent care they received at SickKids. Indeed, it was not uncommon for parents to express relief associated with receiving care from a world renowned children’s hospital. Parents knew that they were “in the best hands,” and vividly remembered “how bad things were” prior to their arrival at the “right place.” However, in light of the ongoing challenges they faced, parents were also reluctant and guarded when it came to receiving physical activity advice or programming for their children. They were not overly enthusiastic or motivated for their child to participate in a physical activity program and were ambivalent about intervention participation; not able to clearly identify what their children needed, they contemplated its effectiveness and merit and wondered whether it would really “work.” They frequently expressed that it would be
difficult for outsiders who lack a similar experience, to really understand what it is like to care for a chronically ill child … “I do not know if you would be able to give what we need” (CHD8).

Similarly, the father of two children with CF erects a barrier between those that parent sick children and those that do not, suggesting that years of education still do not equate to lived experience:

If you had not heard news about your child being given a death sentence, then you do not know how I feel. Sorry, you could have gone to school, but you do not know how I feel or cannot put yourself in my shoes (CF14).

Finally, demonstrating congruence with youths’ statements reported in Study One, temporal dilemmas and anxieties - especially among CF parents - characterized their caregiving experiences and parents discussed time in both an abstract and literal sense. For example, given the life threatening nature of the disease, they were aware that their children would be “here for less time,” and discussed time in an abstract sense. Parents explained how difficult it was to accept that their child would die before them. Furthermore, they discussed the strategies they employed - such as doing things now rather than later - "in order to circumvent the problem of lost time and obtain the most out of their available time:

A parent never wants to have a kid die before her and that is what she was upset about. That is why I was trying to tell her to spend as much time as you can, with her. And just think; every waking moment that you have, spend it with her. Even if you have both of them or one by themselves, spend that time with her. I had her quit her job and that is why I work 16 hours a day. So that she can spend more time with them (CF8).

Similarly, another parent states that:

We want to live for the moment while we can do these things. We have another girl in our town who is a few years older than our daughter who has been in the hospital
multiple times. If my daughter wants to take singing lessons now, I do not care how much it costs - she is going to take them (CF14).

Additionally, the time demands associated with their child’s treatment and ensuring that there was sufficient time to complete it, was a constant source of worry for parents. Parents regarded themselves as having too little time and were stressed by the time consuming nature of treatment. While treatment was the most important thing for parents, it impinged on their freedom, and they disliked their routinized lives that had come to be dominated by treatment:

When you want to go on day trips, it is like, “hurry up get it done, we want to get going.” You end up pushing him. “OK - now you have to eat something. We want it done before we go out for the day.” Sometimes, when you are gone all day, and you get home at 9:00 pm at night. We bring it with us to do in the car. It is always at the back of our mind. It never leaves. You are planning from the minute you get up as to how we will coordinate this mask and physiotherapy (CFP4).

Thus, the findings discussed below should be understood within the context of the “stress and complexity” which characterize these family systems and the sense of fragility and chaos that overshadows them.

The Benefits of Physical Activity

To child. While parents were ambivalent about the importance of a physical activity intervention in the broader context of their child’s health care needs, they did appreciate that physical activity could provide benefits both for their own health and their child. For example, when asked about the impact of physical activity for their child, parents discussed the potential physical benefits.

Interestingly, however, the parents of youth with CF were more knowledgeable about the disease specific benefits of physical activity and were able to discern clear links between engaging in physical activity and minimizing the detrimental impact of the disease. For example,
CF parents were generally aware that engaging in physical activity could improve their child’s lung function. Parents were able to draw upon complex medical terminology to explain, for example, that physical activity aids in dislodging mucous from the lungs:

Absolutely, specifically, I like her to be physically active because that seems to release the most mucous - so that is the most positive thing about it. It actually expropriates it more than anything, more that the physio (therapy). It is just important for health reasons - to keep her body active. To keep her functioning properly and to keep the muscles in tune with the body. Besides the fact that it helps the mucous, it also helps the body to grow and to develop (CF1).

In addition to being aware of the benefits of activity for CF, some parents used physical activity as a gauge or barometer to assess their child’s current health status, or how they were doing:

The more active he is, the better his lungs are doing, right? And that is how we know how he is doing, right? We know that if he is not able to keep up with the other kids, we can see that OK, he is not doing as well. So it not only helps him, but it helps us to gauge how he is doing, right? (CF2)

In contrast, although the parents of youth with CHD were aware of the importance of physical activity for their child, they were not as educated or knowledgeable as CF parents. They had more difficulty making clear links between physical activity and improved cardiac functioning … From what I understand, there was a concern about overworking cardiac kids. I think that there is a new way of thinking where it is important for them to be healthy. Therefore, exercise would be important (CHD2).

To self: Participants discussed the benefits associated with physical activity for their own physical health, such as weight loss, increased physical functioning, improved appearance, and enhanced energy. For example, in the following narrative, one CF mother discusses the numerous benefits associated with physical activity, such as reduced aging, stress, and weight:
Overall, you feel better when you are able to exercise. Right now, it is a weight loss thing as well as not feeling so old - it keeps you feeling better … I think that when you are in your 40’s I feel like you start to age quicker. I find that if I am not exercising, I feel lethargy in general, and general malaise. If I am out walking, I just feel better. My joints do not hurt as much. Definitely - it is a stress reliever. If you have had a crazy day at work, it just lets you shut down your mind and concentrate on the treadmill or the walk or whatever. And you sleep so much better for sure (CF13).

Furthermore, by enhancing their energy, parents were cognizant that physical activity readied them for the physical tasks associated with caregiving. Parents were aware that a lack of physical fitness could undermine their ability to adequately care for their child. Parents speculated that unhealthy and overweight parents would find it difficult and tedious to endure and persist through the long and arduous process of staying with their child in the hospital:

I think that it gives us more stamina. I think that an unhealthy person may not be able to be up at nights a lot and still function in the day. An unhealthy person, may not be able to stay in the hospital for seven days with their child. Yah - so I think that being healthy does help us help Emily. If he (unhealthy parent) was an overweight person, he might get tired during the day and not be able to sleep at night. When you say that it is a test of endurance; like you are in this little room for seven days (CF9).

Parents stated that there were several physical benefits associated with engaging in physical activity for their own health. Furthermore, by equipping them with energy and stamina, and improving physical functioning, parents felt that physical activity would prepare them for the physical tasks associated with caregiving.

**Distraction:** Additionally, for some participants, engaging in physical activity was thought to provide them with a temporary reprieve from constantly worrying about their sick child. Physical activity provided some parents with a temporary time out, in which they did not
have to constantly think about their child’s whereabouts and well-being. For example, Emily’s father eloquently explains how going to the gym provides him with the opportunity to “de-stress;” as sanctuary, it is a place in which he does not have to worry about his CF child:

Going to the gym! You go to the gym and just try and work out - while you are at the gym, you can know that she is at school, or with her mom or grandmother or whatever, and I can just concentrate on what I am doing here and just try and de-stress. And get my workout in. That helps (CF9).

Diana’s mother expresses similar sentiments:

With the yoga class, we would all talk about different things going on in our lives. So it was social. The benefits - you just felt more relaxed. Because it is stressful - my job is stressful and at home is stressful; always with worrying about Diana’s health and stuff like that. That does cause a lot of stress, for sure. So I understand the benefits (CF12).

Thus, from the perspective of parents, physical activity was associated with benefits for both child and self. In particular, parents commented upon the physical benefits associated with physical activity for their ill child. However, CF parents were better able to discern clear links between physical activity and diminished disease symptoms. Furthermore, parents talked about the benefits of physical activity for their own health. Caring for a chronically ill child was experienced as a chronic psychological burden, and, as sanctuary, physical activity provided some parents with a temporary reprieve from the stress of caregiving. More critically, parents who described themselves as physically active also described their children as physically active.

**Role Modeling**

From the perspective of parents, role modeling powerfully shaped youths’ physical activity behaviours, and, in general, active parents tended to have more active children. In contrast, inactive CF and CHD parents tended to have inactive children. Regardless of whether parents were active or not, they were cognizant of the impact that role modeling had on their
children’s attitudes towards physical activity as well as their behaviours; they knew that role modeling influences their child’s psychological response to physical activity. For example, parents who were regularly active were cognizant that they set a good example for their children and expressed that their own physical activity had in no way been “lost on their children.” In this regard, parents were aware that their positive physical activity behaviours and attitudes provided an important illustrative example to their children that physical activity is important … *When I see my parents-my dad, who could have lived a longer life if he has made healthier choices. Like, it is just so obvious. That is not lost on the kids either (CF6).*

Similarly, another father discusses his positive role modeling influence in the sport of hockey:

* I play hockey myself and that is what made him want to play hockey. I should do more activity, because I just sit at a desk job all day. I try and get out there and play with them. I try to be as active as I can. I would like to be more active for them (CHD1).

In contrast, inactive parents explained that laziness, lack of time, or complacency prevented them from engaging in physical activity. Parents did not deny their poor physical activity behaviours and recognized the implications that lack of activity could have for overall health. These parents often chastised themselves. They acknowledged that they were not positive influences for their children and that their children likely mimicked their lack of physical activity:

* Her father and I are not the best example either. We will finish dinner and say that we are full and then have a cup of tea and a few cookies and sit down for the night. You know what I mean? (CHD12).

Similarly, another CF mother states that:

* I am not a good example at all. I am not. The only motivation that I have is her health and I know that it should not be that way. But, I will do forms of activity with her. I do not
enjoy it. I enjoy it while I am doing it but it is the thought of getting there and doing it. I do not like it (CF12).

Thus, from the perspective of participants, the physical activity attitudes and behaviours of parents strongly influence their child’s orientation toward physical activity. Active CF and CHD parents described having active children. By ensuring that they provided their children with opportunities to be active and encouraging them, they were also gatekeepers toward health behaviour change. In contrast, inactive parents tended to have inactive children and were less likely to seek out opportunities for their children or to make physical activity a priority.

**Barriers to Physical Activity**

Despite the benefits associated with physical activity, parents suggested that numerous barriers prevent them and their child from engaging in physical activity. These barriers - to self and to child - are briefly discussed below.

**To self:** While parents were well aware of the physical and psycho-social benefits associated with engaging in physical activity for their own personal health, particular barriers prevented them from being active. Parents described common physical activity barriers that are not unlike those that prevent participation among the parents of healthy youth, such as laziness, lack of motivation, lack of time, or a general sense of complacency. However, they also reported more disease specific barriers to physical activity. In particular, parents reported a tremendous sense of guilt associated with taking time off for the self to engage in physical activity. The concept of self care was not evident to parents, and, rather, they foresaw that they would feel guilty about the temporary disruption in the caregiving routine that engaging in physical activity for themselves would require:

Yes, guilt about activity is the case. I would definitely say that. I am in a relationship and that is what is hampering it. He always says that I am doing too much for the kids and that I do not take time for myself. That is the way that it is. I think it is who I am. It is
easier now, as the boys are getting older. Even leaving them at 12, 12, and 14 (Two, 12 year old twins). Leaving them in the evening for activity - I just cannot do it. I just do not feel right about doing it. It has created issues (CHD6).

Similarly, in the following narrative, Harriet’s mother demonstrates her slowly evolving understanding of the importance of physical activity self care:

I am getting better at saying, “it is OK to put myself first.” But then I said, “but her disease is also crippling me. Harriet will always be right beside me. I know that it is important for me to be healthy and that it is important for her to be healthy (CF13)

Furthermore, some participants explained that parenting a child with a chronic illness meant changing life roles, responsibilities, and priorities. Within the context of caring for an ill child, physical activity and health were no longer a priority for parents:

This is what is most important (caregiving). I know that physically, I should be more active, but it is only the circumstances and I know that it is temporary. Through the last three years, I should have been more active, but given the circumstance, I do not know how I would have been more physical. For me I do not think so. No. Caring is the thing that is my only priority (CF10).

Similarly, the father of two children with CF states that:

I am in a new role now. Activity is not a priority. I am fortunate because I am healthy. Maybe I take it for granted, I am not sure. They are the focus, with both of them having fatal illnesses. I chose to do this (CF14).

In this regard, parents reported “self barriers” and described that an ongoing and lingering sense of guilt, as well as changing life roles and responsibilities, prevented them from taking “time off” from their caregiving duties to engage in physical activity.

To Child: From the perspective of parents, their children also encountered several barriers in the effort to be physically active and they commented upon their child’s negative psychological
response to physical activity. Unfortunately, these disease related barriers, including disease burden, low self-efficacy, and scars and size, made it more challenging for their children to engage in PA.

The burden of disease

From the perspective of parents, the detrimental impact of the disease - and unpleasant symptoms - was reported as the primary barrier to physical activity for their children. The nature of these negative disease symptoms varied slightly. For example, for children with CF, breathlessness, fatigue, and coughing were the primary deterrents. In contrast, fatigue, dizziness, and breathlessness were experienced as debilitating for youth with CHD:

He has had so many surgeries. He has so much done to him. He has had three open heart surgeries and many caths (catheters). The last few years, he has had two valves put in, a bovine and a pig valve. He has had an ICD put in. Because he is coarc (Coarctation of Aorta) now, he has always had poor circulation and no ability to get his heart rate up to get working to do stuff. If he starts to do something and he is as gray as that - and people go, “oh my God, sit down, what is wrong?” He sits down and he stops. The sweat pours. He will come back from the store, you can ring it out and it is soaked (CHD3).

Similarly, another parent discusses the burden of disease in CF by stating that:

She cannot walk very far. Running - out of the question. She was at school when there was physical education, and if there was running, she was the timer. She found another way to be involved in it ... That is how it did affect her ... I know that if I say “OK, let’s go to the school and we will walk there.” She is not going to be able to walk over there. At two minutes, she is stopping, and we can see that her lips are turning blue a little bit (CF10).

For children with severe disease, the detrimental impact of disease symptoms was so intrusive that it was thought to entirely change the children’s activities and motivations and
contribute toward an entirely new set of interests. For example, Crystal has been hospitalized repeatedly for close to two years, and, as she awaits a double lung transplant, she is receiving oxygen therapy and total parental nutrition through a $^{11}$G tube. Crystal has a great deal of difficulty walking; she becomes frustrated when her peers will not wait for her and she always struggles to keep up with them. Crystal’s father is convinced that the severity of her symptoms has contributed toward re-shaping her interests and detracted from any potential that she could have had in physical activity. For example, rather than engaging in physical activity, Crystal has become a very artistic child - interested in drawing and painting - and also spends a great deal of time watching television or engaged in other solitary pursuits:

We knew that she was fragile and could not play as hard as the other friends. During two summers ago, we registered her. She played maybe two games a summer. She liked it, but if she does not feel well, then no. I have registered her for a karate course; I say “I will go with you.” She could not follow the group and it was hard. At school, she was playing kin ball. But at the game, she was only in the group, but not really playing. Many different sports, but she was never really-she is more artistic than sporty. We wanted her to do activity-but she was more turned onto drawing, painting. I do not know if she would have been as artistic as she is right now. I think maybe the CF influenced her interests (CF10).

Low self-efficacy for physical activity

Related to the burden of the disease, low self efficacy was a common barrier for these parents’ children, contributing toward a negative psychological response toward physical activity. While parents did not use this word to describe their child’s perception, negative experiences in physical education, sport, or exercise often led children to feel “bad about themselves.” The

$^{11}$ Some children have medical conditions that prevent them from being able to take adequate nutrition orally. A gastrostomy tube (also called a G-tube) is a tube inserted through the abdomen that delivers nutrition directly to the stomach. It is used to ensure adequate nutrition, growth, and development.
inability to keep up with their peers led children to be excluded; this invariably left children feeling frustrated and they came to doubt their capabilities. Ultimately, these negative experiences led children to develop a more persistent belief in their inability to do sports and usually led them to drop out from the activity. Thus, self doubt, low-self efficacy, and feelings of inadequacy characterized how children with CF and CHD felt about physical activity … *She does not feel well and she does not have the energy—and then she does not really want to.* It makes sense. *If you are not feeling well, how are you going to exert yourself? (CF12).*

When you hold them back and hold them back, they are like - “mom I do not want to go anymore.” They lose their interest and want to drop out. Yes. And then there is a group of kids that are sporty and kids that are not (CHD9).

Parents also expressed their dissatisfaction with standards-based physical education or physical activity, which demands that normative criteria be attained before progressing to the next level. For unskilled and ill children who have difficulty meeting such criteria, this invariably led to failure and dropout. For example, Jaime’s mother reflects on the negative experience which her daughter encountered during community swimming classes. She states that because of her medical condition, Jaime will “never be able to keep up with her peers” or to meet the standards based curriculum. Indeed, this experience has led Jaime to feel defeated and to drop out of swimming. Jaime’s mother urges teachers to better modify their programs so that children like her may be able to participate too:

Swimming lessons are crazy - But Jaime, until she was maybe ten or so, she reached a plateau … they would not move her up. She could swim forever on her back and could not do front crawl or breast stroke. They did not move her up because she could not physically do the four lengths. And I did say to them that “you need to-not give her the badge, or whatever, but move her up, so that she can learn different strokes, learn CPR.
You still need to teach them.” Yes-some children will never be able to meet the criteria that are set (CHD9).

Crystal’s father expresses similar sentiments. Although at one time Crystal enjoyed walking with her friends outdoors, her debilitating CF symptoms make it too difficult to keep up with them. Although she asks her peers to wait for her, the group frequently forgets after a few minutes; they resume their faster walking pace which invariably leaves Crystal feeling frustrated, angry, and defeated. Ultimately, Crystal gave up walking with her friends. Although she always waves through her bedroom window when they are walking outside, she no longer joins them and prefers to stay indoors where she can do quiet activities:

At first she was angry - she said “I ask them, and they do not listen.” After a while, she just; when they are coming home, she is happy. They play in her room or watch movies. If it is outside, she does not go. At first, when it happened, she was angry when she has come back home. When she sees them outside, she does not care. She is playing on the computer (CF10).

Aaron’s parents also emphasize how low self-efficacy contributes toward disinterest in active pursuits. Furthermore, as their son approaches mid adolescence, they have noticed a progressive worsening of his self efficacy for physical activity:

He seems to – if he does not think that he is good at a sport, he is not as interested in doing it. Soccer sign up is on Monday, and I am saying, “you should sign up.” He says, “well I am not very good at soccer,” and I am saying “that does not matter - the important thing is that you have a good time and that you get out there.” And it is like “let me think about it.” I will be really upset if he does not take soccer, you know. He seems less interested in things, where he knows that he is not as good as the rest of the team and that is a source of worry for you as he approaches adolescence (CF2).
Low self efficacy - or lacking confidence in one’s ability to successfully undertake physical activity - characterized the experiences of children with CF and CHD in physical activity.

*Scars and size:* From the perspective of parents, youth with CHD and CF are also plagued by body image concerns which detract from physical activity. ‘Scars and size’ refer to the disease specific body image concerns that are related to the pathogenesis and treatment of CHD and CF respectively. These body image concerns taint their experiences in physical activity and lead them to avoid engaging in certain activities altogether.

For example, CHD parents stated that the numerous surgical scars which their children have as a result of open heart surgery, often left them feeling weird, abnormal, or different. In addition to being worried about the appearance of their scars, children feared that scars would elicit undue attention and lead to unwanted questions from others. Children were generally uncomfortable disclosing their condition to others, and, unfortunately, surgical scars were thought to prompt unwanted disclosure in social settings. Body image sensitivity to scars varied across the group, but, in general, such anxieties were exacerbated in physical activity and physical education settings when the body was more exposed. Children took strides to avoid others seeing their scars when changing in the change room. They purposely avoided activities in which it was difficult to conceal scars, such as swimming … *She has never wanted people to know what is wrong with her. Unfortunately, it comes out, because of her mechanical valve, she ticks loudly and people have caught onto this ... Yes, scars are tough (CHD9).*

I think that body image would have been with the scars. He did ask the doctors if that was ever going to go away. It was all red and it did bother him a little. When he went swimming, he was worried about it (CHD8).

In contrast, given the nutritional complications associated with CF, many patients are underweight and have a small stature. While adequate nutrition is critical to the proper management of CF, parents stated that their children often struggle to consume a heavy,
calorically rich diet as well as the numerous dietary enzymes which are required to adequately digest. Although thinness and short stature were interpreted differently between boys and girls - indeed, given the current cultural emphasis on slenderness, some girls sadly commented upon how CF allowed them to fit in with the in crowd and look like their peers-in general, small stature and thinness was thought to be detrimental in physical activity settings. For example, some children felt that their small size signified a young age; they were worried that they did not appear the same as their peers and would be infantilized. Some parents felt that their CF child was too delicate and fragile to participate in certain activities. Because their child was small and thin, they worried that participation would result in injury … *I think with her being thin ... She does say stuff like that. There is that. She does say - about her ribs. “You can see my ribs and I do not want to wear a two piece bathing suit.”* (CF12)

The only body image thing - he is so desperate to gain weight, but then again, that could be a teen boy thing and all of his friends are the same way, who do not have CF … That is the only thing really. And now he is the shortest of his friends and has not really grown very much in the past couple of years. We don’t care how tall he is as long as he is healthy - but he does care (CF6).

5.6 Discussion

This qualitative investigation sought to explore the perceived benefits and barriers of physical activity among parents of children with CF and CHD and identify implications for intervention development. Experiences of physical activity occurred within a broader family system that is characterized by stress, complexity, and fragility. Indeed, these families were characterized by divorce and perceptions of helplessness and stress. In line with a family systems perspective (Murray et al., 2007), this context of stress and complexity tended to taint and overshadow parents’ physical activity experiences, making them unlike - and more fragile than -
other families. Many parents described a dysfunctional child-parent sub-system characterized by heated battles and feuds over whether their adolescent was adhering to treatment, or not.

Although CF parents were better able to discern links between engaging in physical activity and reducing the detrimental impact of the disease, both CF and CHD parents were aware of the physical benefits associated with physical activity for their child. Parents discussed the benefits of physical activity for their own health. Specifically, it enhanced parental physical health, increased the physical functioning that is necessary for caregiving, and provided parents with a temporary reprieve from caring for an ill child. However, despite the benefits, parents discussed the barriers which prevented both them and their child from being able to engage in physical activity, such as disease symptoms, poor self-efficacy, and the disease-specific body image concerns of scars and size. Role modeling appeared to be a significant social process which facilitated physical activity. These findings have significant implications for the development of behavioural physical activity interventions targeting children with CF and CHD.

Such intervention development will need to be sensitive to the complexity associated with these family systems. Parents characterized their experiences as being one of stress, marital failure, and ongoing feelings of grief, despair, and anxiety. Rates of marital discord were also particularly high. The literature lacks consensus regarding whether the parents of youth with chronic diseases are at greater risk of marital failure; studies report contradictory results and marital discord may not be the most accurate indicator of family stress (Sabbeth & Leventhal, 1984). None the less, marital stress and failure was high in this study, and should alert researchers to family complexity and systemic malfunction. Other researchers have documented the tremendous stress associated with caring for a chronically ill child. Indeed, parents often describe their experiences as life shattering and life altering, requiring them to question their competence as good parents (Rempel & Harrison, 2007; Woodgate & Degner, 2003).
Skepticism, ambivalence, and reluctance to participate in research and interventions, and the belief that others do not understand me, may taint CHD and CF family’s receptiveness toward physical activity promotion messages and interventions. Since parents are skeptical of others’ capacity to understand what it is really like to parent and care for a chronically ill child, it is important to display empathy and understanding. If parents do not regard the exercise counsellor as an empathetic and credible source, it is likely to taint their receptiveness toward physical activity messages and compromise the uptake of programs. Methods of intervention delivery should incorporate elements of peer delivery, in which the delivery of physical activity messages include members of the target audience - that is, parents and children with chronic diseases.

Complex temporal dilemmas also characterized the experiences of CF parents. This is a unique finding that is not significant for CHD parents, suggesting that post operative repair, the impact of CHD is less intrusive and parents do not regard their child as life limited. Indeed, the demanding schedule associated with CF treatment was described as time consuming and left parents feeling too exhausted to engage in physical activity. The time consuming burden of disease has been reported elsewhere in the literature (Hafetz & Miller, 2010; Schneiderman-Walker et al., 2005) and accords with the children’s findings from Study One. CF parents also discussed time in a more abstract sense and expressed difficulty in coming to terms with their child’s reduced life span. Thus, physical activity interventions must be tailored to suit the busy treatment schedules of CF youth and parents and it is important that programs not be perceived as wasteful of time. Encouraging children and parents to engage in brief bouts of physical activity - such as walking to school - as well as devising novel ways to incorporate physical activity into treatment, may be a particularly useful recommendation.

In comparison to CF parents, CHD parents appeared less knowledgeable about the benefits of physical activity for their child in terms of their condition. They were not able to
establish relationships between physical activity participation and disease outcomes. The reasons for this are speculative. For example, lack of knowledge could be due to greater disease complexity and variability in comparison to CF, or poor staff communication about the benefits of physical activity. Different illnesses conjure up different cultural interpretations (Barnes, Mercer, & Shakespeare, 1997), and the heart may connote particular social imagery. There may be greater worry and fear about physical activity participation among the parents of children with heart disease, leading them to inappropriately overprotect and “coddle” their child. Historic and current literature suggests that perceptions of frailty, vulnerability, and risk have negatively influenced physical activity in youth with heart disease and served to exclude them from participation (Bar-Mor et al., 2000; Moola et al., 2010). For example, in a 1967 seminal piece, Bergman and Stamm (1967) proposed that the very notion that children with heart disease are more fragile and at risk, is a disease in and of itself. Termed the “vulnerable child syndrome” (Wilkes et al., 2009) or “cardiac non-disease” (Bergman & Stamm, 1967), such perceptions thwart youth with CHD of opportunities to be “normal.” This may serve to further “cripple” cardiac children and deprive them of opportunities to participate in physical activity with their same age peers. Most asymptomatic children with surgically repaired CHD can and should be encouraged to participate in safe and enjoyable physical activity (Moola et al., 2009), and continued efforts are required to dispel myths regarding their supposed frailty and vulnerability.

From the perspective of parents, unpleasant disease symptoms, such as fatigue and breathlessness, prevent children with CF and CHD from engaging in physical activity and are barriers to participation. This has been reported elsewhere and is well documented in the literature (Moola et al., 2008; Swisher & Erikson, 2008). For example, in a qualitative study undertaken with CF youth, it was found that breathlessness and coughing were experienced as formidable barriers to physical activity (Swisher & Erikson, 2008). Another study reported similar findings with CHD youth (Sparacino et al., 1997), and other researchers have found that
children with heart disease negotiate physical activity within the context of chronic and
prevailing fatigue (Moola et al., 2008). Understanding these barriers and designing physical
activity programs in such a way so as to circumvent them, is critical. The need to optimize
physical benefits should be balanced with ensuring children’s comfort during activity. It is likely
that children will not adhere to activities that exacerbate symptoms. Activities which minimize
physical discomfort, or which gradually expose children to increasing exercise stress over time,
are recommended.

Parents also reported that their children display poor self-efficacy for physical activity
and this aligns closely with the literature. For example, several studies have reported that youth
with chronic diseases experience feelings of inadequacy and low self confidence in physical
tasks. Self efficacy appears to be critical to the physical activity experiences of youth with
chronic diseases and may override the severity of the disease (Bar Mor et al., 2000). In addition
to physiological status, vicarious learning, and social modeling, experiences of mastery, success,
and competence in any given task is critical to establishing self-efficacy (Bandura, 1997). Thus,
experiences of exclusion, feeling demoralized, and being ostracized from physical activity
invariably lead children with CF and CHD to doubt their abilities, feel defeated, and terminate
their involvement in physical activity. It is critical for exercise counsellors to develop programs
which are self-efficacy enhancing and include elements which promote mastery.

Additionally, this study has bettered our understanding of disease specific barriers to
physical activity which affect youth with CHD and CF particularly. Although problems with
surgical scar sites for CHD youth have been documented previously (Sparacino et al., 1997), as
well as small stature in CF youth (Berge et al., 2007), the ways in which scars and size impede
physical activity has not been extensively discussed in the literature. In particular, due to the fact
that it led to unwanted social attention and perceptions of abnormality, children with CHD
avoided undertaking those physical activities that revealed cardiac scars. Furthermore, small
stature and thinness was experienced as a physical activity barrier for youth with CF. In addition to leading others to think that they are younger than they actually are, it made children feel vulnerable in physical activities where greater size and mass were necessary. Thus, it is important to be aware of the specific body image concerns that are experienced by youth with CF and CHD and how these may taint perceptions of activity, shape psychological responses to activity, and lead to activity avoidance. Exercise counsellors should circumvent such sensitivities by offering an encouraging physical activity environment. They should also encourage children with CF to participate in physical activities where small stature is not experienced as a disadvantage.

While parents were aware of the benefits of physical activity for their own personal health, a sense of pressing guilt for taking time off from caregiving duties to engage in physical activity was reported as a barrier. The concept of self-care was difficult for parents to comprehend and they had difficulty understanding that enhanced self-care may improve their capacity to engage in caregiving duties. A sense of ongoing obligation and duty or - constant vigilance (Rempel & Harrison, 2007) - has also been reported among other caregivers. In light of the psycho-social and physical benefits that physical activity may afford to caregivers (King & Brassington, 1997), pediatric caregivers - such as the parents of youth with CF and CHD - should be educated about the importance of self care and encouraged to engage in physical activity for themselves.

As a form of observational learning, role modeling is a concept that has been advanced by those schooled in social-cognitive psychological theories, such as Bandura (1997). According to this hypothesis, one of the primary ways by which children learn about the world and develop new behaviours, is by observing and mimicking others. How parents behave - their exercise and eating practices - are thought to powerfully influence their children’s learning and uptake of new information. For children with chronic diseases, parental role modeling of physical activity...
behaviours may be more critical for children with chronic diseases given the range of barriers these children face to be active. Parents were cognizant of the impact that their behaviours had on their children and active parents tended to have active children. Thus, parental role modeling powerfully influences child physical activity in these populations. As such, efforts must be made to engage parents in physical activity interventions for CF and CHD youth. Interventions that involve parents should be considered as a potential design – to benefit both child and parent. For example, family based intervention research is a potentially useful approach to facilitating physical activity participation in this population.

5.7 Conclusion

By opening up the psychological and social worlds of parents, this is the first theoretically informed qualitative study which has examined how parents and caregivers of CF and CHD youth understand physical activity, for both themselves and their child. Overall, this study provides insight into the physical activity barriers that are encountered by CF and CHD youth and their parents, as well as directions for future physical activity interventions. The information from this study is essential for developing interventions that are acceptable, feasible, and attractive for this relatively neglected and poorly understood group of youth and parents affected by CF and CHD.

Building upon the findings and concepts discussed in the previous two studies, such as “reduced time, role modeling, and disease burden,” the following intervention chapter in this sequential series of studies, builds upon these important lines of inquiry. More specifically, the intervention that was developed and subsequently pilot tested with a group of CF patients and parents, was designed with the view to considering and incorporating the findings and concepts discussed above.
Chapter Six, Study Three

“CF Chatters:” The Development and Feasibility of a Six-Week Theoretically Informed Physical Activity Counselling Program for Children and Youth with Cystic Fibrosis (CF) and Their Parents

6.1 Introduction

Yes, the program will be helpful for other kids with CF in the clinic. They will feel better after the program. They may go from feeling good to maybe down, to maybe good again and think that “this is good. It helps” ... It was valuable time - I would rather be here than doing something silly (Chase, age 17).

The above quote illustrates one CF patients’ perceptions toward a novel, physical activity counselling program entitled CF Chatters. Informed by my disease specific Grounded Theory of physical activity in CF, in this chapter, I describe the development of a six week physical activity counselling program for youth with CF and their parents, known as CF Chatters. I will also explore participants’ perceptions toward the program and its influence on physical activity and quality of life. Before describing CF Chatters, I will contextualize the study by providing a brief review of the literature on the efficacy of physical activity and exercise interventions for the CF population, and, in doing so, draw attention to critical intervention gaps that further the rationale for a program like CF Chatters.

6.2 Review of the Literature: Physical Activity and Exercise Interventions for Youth with CF

Physical activity is conceptualized as any bodily movement produced by the skeletal muscles that result in energy expenditure (Caspersen et al., 1985). In contrast, exercise training refers to planned, structured, and repetitive bodily movement generally with the objective of health enhancement (Bouchard & Shephard, 1994). Despite the benefits of employing habitual physical
activity (HPA) as a broader approach to physical activity among children (Schneiderman-Walker et al., 2000), the literature is characterized by an overabundance of exercise training approaches. For this reason, the rationale for physical activity posited in this study is based on an inadequate body of knowledge that is heavily exercise based, and, for this reason, I have extrapolated the findings from exercise training protocols to physical activity in CF. This is elaborated upon below.

Given that it facilitates the expectoration of mucous from the lungs, physical activity and exercise may mediate survival in children and youth with CF. Although definitive statements cannot be made, some researchers suggest that physical activity is a useful adjunctive treatment and should be incorporated into a model of disease in CF (Stevens & Williams, 2007). Despite the important benefits of physical activity, and evidence that it may slow disease progression over time, children and youth with CF display reduced intensity of physical activity in relation to healthy peers (Nixon et al., 2001) and deficient aerobic and anaerobic capacity (Stevens & Williams, 2007). Thus, interest in physical activity and exercise interventions for CF patients is a burgeoning area of inquiry. A range of different intervention modalities - which vary in terms of the type of activity, the program duration, the number of participants, the research setting, and the outcome measurements employed, have been conducted. Despite the great variation in intervention designs, all point to the notable benefits associated with physical activity for the CF population.

For example, reviews of the literature have documented the notable benefits associated with exercise training for children and youth with CF and some researchers advocate for the inclusion of physical activity as a variable that is related to health status in this population (Decramer & Gosselink, 2006). For example, Stevens and Williams (2007) purport that although young patients with CF display exercise intolerance, reduced muscle size and strength, and deficient aerobic and anaerobic capacity, novel exercise training models have been developed
that afford patients with important functional and psychological benefits. Since exercise testing offers an objective and integrated assessment of the metabolic, respiratory, musculoskeletal, and cardiovascular status of the patient, it is a tool with much prognostic value that can assist physicians in making important clinical judgments and decisions. Similarly, although the variation in exercise training programs limits the development of generic guidelines, a recent Cochrane review suggests that exercise affords important physical benefits to patients with CF and it should not be discouraged. In addition to negatively influencing overall health and body image, inactivity in patients with CF may worsen lung disease and compromise activity of daily living participation (Moran, 2008). Additionally, although patients with CF face numerous barriers to participation, evidence obtained from aerobic, anaerobic, and strength training programs support the beneficial role of physical activity and exercise on health and quality of life in patients with CF (Wilkes et al., 2009). In addition to improved pulmonary function and nutrition, these reviews of the literature conclude that physical activity may influence other physiological and psychological parameters, such as reduced risk of osteoporosis and diminished anxiety and depression (Moran, 2008; Rose & Sandy, 1986; Goodhill, 2005; Moorcroft et al., 2004).

Furthermore, four recent randomized control trials illustrate the important physical and functional benefits associated with exercise training for children and youth with CF. In a hospital-based clinical trial by Klijn et al (2004), 20 children and youth with CF between the ages of nine and 18 years were randomized to either a 12 week, bi-weekly anaerobic training group or a control group. Notable increases in aerobic performance, anaerobic performance, and quality of life were observed in the training group. Similarly, in a hospital-based control trial by Selvadurai et al. (2002), 66 youth with CF were randomized to a resistance training program, an aerobic training program, or a control group. The mean age was 13.2, 13.1, and 13.2 years respectively, and, with a mean Forced Expiratory Volume in one second (FEV 1) of 56.8%, the group was
relatively ill. The aerobic training group demonstrated significant improvements in peak aerobic capacity, activity levels, and quality of life. In contrast, youth in the resistance training group demonstrated improved weight gain, leg strength, and lung function (Selevadurai et al., 2002). In light of these different benefits, the authors advocate for a combined physical activity approach that incorporates both training modalities. While hospital settings allow for a high degree of control and optimal participant supervision, it is important to ensure that youth with CF are equipped with the skills and abilities to engage in physical activity in their local environments and communities (Schneiderman - Walker et al., 2000). In their home-based study, 65 CF patients between the ages of seven to 19 were randomized to a three-year physical activity program. Patients in the training condition were encouraged to engage in 20 minutes of large muscle group physical activity at least three days /week, and emphasis was placed on selecting enjoyable activities. The rate of pulmonary function decline was less notable in the exercise intervention group (Schneiderman-Walker et al., 2000), and, these telling findings point to the potential role of exercise in the attenuation of disease progression. Suggesting that physical activity improves prognosis and enhances the ability of patients with CF to manage the physical tasks associated with daily living, Orenstein et al (2004) conducted a year long, home-based semi-supervised exercise training study. Sixty seven patients with CF between the ages of eight and 18 were randomized to either a resistance or aerobic training group. Participants were encouraged to engage in three sessions of physical activity/week. Although participants in the resistance training group reported greater increases in strength, all youth demonstrated improvements in physical work capacity (Orienstein et al., 2004).

Beyond this evidence from RCTs, quasi experimental studies also demonstrate the notable benefits associated with exercise training for youth with CF. For example, utilizing a hospital-based intervention with rolling enrolment, Gruber et al. (2008) enrolled 286 children.

Patients with CF tend to be malnourished and underweight. Maintaining a healthy weight is critical to overall health and disease outcomes.
and youth with CF to a four-six week program that consisted of 45 minute, mixed activity
sessions five days/week. The program resulted in improved pulmonary function and increased
performance on a multiple-component fitness test. As such, the authors underscore the
importance of devising training interventions that incorporate multiple domains of fitness
(Gruber et al., 2008). Gulman et al (1999) studied the response of 14 youth with CF to a 12-
month, home-based cycling program and patients engaged in five cycling sessions /week.
Notable improvements in muscular strength and maximal oxygen consumption were observed.
Since youth with CF did not find the program enjoyable or fun, the perceived acceptability of the
program was low. However, psycho-social benefits were observed in the area of perceived
competence, feelings about physical appearance, and general self-worth (Gulmans et al., 2008).
De Jong et al. (1994) studied the response of ten, relatively ill patients with CF to a three month
home-based cycling intervention that consisted of 15 minute daily cycling sessions. Program
participation resulted in improved maximal exercise capacity, maximal oxygen uptake, and
oxygen pulse, and reduced activity of daily living (ADL) limitation. Utilizing a novel setting,
Blau et al (2002) studied the response of 13 Israeli patients with CF to a four-week training
program at a camp in the Swiss Alps. The participants engaged in daily mountain climbing and a
range of different indoor activities. Improvements were noted in exercise tolerance, peak work
capacity, maximal oxygen uptake, minute ventilation, and anaerobic threshold. Although such
intervention designs are not feasible given cross contamination concerns, participants also
reported improved nutrition and perceptions of well-being (Blau et al., 2002). In a landmark
longitudinal study, Schneiderman-Walker et al (2005) studied the relationship between forced
expiratory volume (FEV1) and physical activity participation among 109 patients with CF over
the course of three years. Using the Habitual Activity Estimation Scale (HAES), participants
were divided into four activity quartiles that generally corresponded to the patients’ activity
status. Schneiderman -Walker et al (2005) found that girls with CF in the least active physical
activity quartiles demonstrated a steeper decline in FEV 1. These compelling findings suggest that lung function decline occurs at a faster rate among the least active patients (Schneiderman-Walker, 2005), and that adolescent girls are particularly at risk.

6.3 Research Limitations

The literature reviewed above has been critical to furthering evidence that supports the important role of physical activity in the lives of patients with CF - and the notable physiological benefits. However, because current physical activity and exercise programs in the CF population are characterized by varied research designs, exercise modalities, outcome measurements, program durations, disease severities, and participant ages, it is difficult to propose conclusive recommendations for a physical activity dose. I have also identified several research limitations related to intervention design and research methodology. Firstly, there is an absence of theoretically informed studies that draw upon existing theories and models of health behaviour change to design and implement programs. Methods and strategies for increasing physical activity among CF youth are not theoretically informed. The atheoretical nature of studies and interventions may increase the likelihood that studies do not address the central theoretical constructs that are relevant to physical activity behaviour change in youth with CF. Furthermore, in the absence of relevant health behaviour change theories and models, researchers lack a conceptual framework for the interpretation and analysis of intervention findings. Secondly, perceptions of choice and autonomy are important theoretical constructs in the health behaviour change literature and are associated with positive psychological health (Ryan & Deci, 2000). However, despite the recommendation by Wilkes et al (2009) that youth with CF engage in physical activities that are fun, enjoyable, and sensitive to their perceived barriers and local contexts and environments, with the exception of Schneiderman - Walker et al (2000), current interventions prescribe standard exercise approaches that may not capture participants’ interests, motivations, or contexts. Indeed, Schneiderman -Walker et al (2000) suggest that while exercise
training in clinical settings results in notable and important physiological benefits that furthers the evidence for exercise in this population, the resource intensive nature of this approach is plagued by numerous limitations. In addition to lacking intuitive appeal for children and failing to capture children’s actual activity patterns, the sustainability of exercise training may be compromised once the intervention is terminated (Gulmans et al., 2008). Rather, Habitual Physical Activity (HPA) - such as childhood play - may more accurately capture the nature of how CF patients move in their local contexts, environments, and homes and provide children with a more realistic, enjoyable, and manageable approach to activity.

Thirdly, with the exception of Swisher & Erickson (2008) who employed a qualitative research design in order to explore the perceptions of youth with CF toward physical activity and exercise, the literature is characterized by an absence of qualitative research studies that have explored how youth with CF experience physical activity. Swisher and Erickson (2008) found that although youth with CF identify numerous benefits associated with being active, unpleasant disease symptoms and a lack of interest in physical activity, are barriers. In their review of the literature, Wilkes et al (2009) suggest that it is critical to pursue Swisher and Erickson’s (2008) qualitative lines of inquiry in order to understand why most patients are inactive in spite of the benefits that activity would afford. Although I have embarked on these tasks in Study One and Two, this remains a neglected question that is arguably best addressed through the inherently exploratory and explanatory nature of the qualitative paradigm. Specifically, more information is required in order to elucidate youths’ perceptions toward activity and their thoughts about potential behavioural interventions.

Fourth, only a few interventions include direct measures that assess the influence of physical activity on psycho-social variables - such as perceptions of wellness or quality of life. In most cases, psycho-social health remains tangential to exercise intervention research in CF. Indeed, despite the beneficial role of physical activity in enhancing psychological wellbeing
among CF patients (Stevens, Oades, Armstrong, & Williams, 2010), no studies have included psycho-social health as an outcome or a component of the broader research question. Given that patients with CF may have compromised psycho-social health, such as depression, anxiety, or perceptions of futility (Berge et al., 2007), not addressing the influence of exercise and physical activity on psycho-social health parameters is a notable oversight.

Fifth, the development of behavioural self regulation skills are required in order to independently regulate physical activity behaviours and to adhere to physical activity over the course of time. However, to my knowledge, no studies include a behavioural skills development component that will equip participants with the knowledge and skills necessary in order to sustain participation in their local environments. Sixth, despite the benefits of counselling approaches that allow researchers to explore participants’ resistance to behaviour change and tailor physical activity in a meaningful way to the everyday lives of patients (Whitlock, Orleans, Pender, & Allan, 2002), thus far, the literature has not utilized counselling as a modality for behaviour change. Finally, there are no published studies in the area of physical activity for youth with CF that have engaged parents and caregivers in behaviour change attempts, and interventions have largely been delivered at the individual level without considering the broader family context. Given that parents heavily influence youths’ physical activity (Golan, 2006), this is a particularly glaring oversight.

In summary, most youth with CF are inactive despite the numerous benefits that physical activity would afford, and, arguably, behavioural interventions are required in order to facilitate activity. Furthermore, despite great variation in intervention design, this review of literature revealed that there is an absence of theoretically informed behavioural interventions and an overabundance of studies that utilize standardized exercise prescription approaches. Psycho-social health in CF is not included as a direct intervention outcome. Although I have embarked upon this important line of inquiry, there is a lack of qualitative research and an absence of
studies that include a behavioural skills regulation component. Counselling has not been employed as a method for intervention delivery and parents have not been engaged as potential gatekeepers to health behaviour change. Thus, the development of the CF Chatters program described below aimed to address these critical limitations. In light of these limitations, my intervention is designed by keeping in mind 1) the need to use theory, 2) encourage habitual physical activity rather than exercise, 3) ensure the use of the qualitative paradigm, 4) explore the impact of physical activity on psycho-social measures of health, and 5) exploit the facilitative function of parents. In addition to describing the development of a theoretically informed behavioural intervention, the purpose of this study was to draw upon the qualitative paradigm in order to explore participants’ perceptions toward CF Chatters. This study also sought to examine the impact of the program on physical activity and quality of life and participants’ perceptions toward intervention feasibility and acceptability. The method that guided the development of this theoretically informed intervention is described below.

6.4 Methods

Development of the CF Chatters Program: Theoretical Framework

The CF Chatters program is a six-week parent mediated cognitive-behavioural intervention. CF Chatters was informed by my own initial, qualitative research undertaken with 14 CF youth and 28 parents (N=42) in Study One and Two, and my relevant Grounded Theory. There is great variation in the duration of physical activity programs for youth with CF and consensus has not been established in the literature (Moran, 2008). While study durations range from one month to three years, I selected a six week time frame. This program duration has been reported previously in the CF and exercise literature (Gruber et al., 2008). It allowed me to avoid overburdening a population that already experiences significant time constraints. At the same time, it was an ideal time frame in which to intensively deliver the intervention content, allow
participants to practice their newly acquired self-regulation skills at home, and potentially observe short term increases in physical activity and quality of life.

Developing pilot studies on the basis of developmental research broadly accords with the Medical Research Council’s (MRC’s) framework for complex behavioural interventions that was outlined in Chapter Three (McEachan et al., 2008). In order to crystallize the data and to develop a substantive theory that captures the central theoretical constructs that are relevant to their physical activity experiences, a Grounded Theory analysis was performed based on the findings from Study One and Two. This Grounded Theory was then used to guide the development of my intervention. A brief description of Grounded Theory - or the crystallization phase in this Dissertation - is briefly outlined.

*Grounded Theory: Qualitative Research Tradition*

Grounded Theory is a popular and “sexy” qualitative research tradition that continues to engender controversial and heated debates regarding its proper use. While Grounded Theory is differently defined and conceptualized, it is best considered as a total qualitative research design that encapsulates both methodology and method. In this regard, it is both a way of conceptualizing how knowledge about the social world should be produced, as well as a *set of tools* for the collection of data about the social world, and enables researchers to develop theories about poorly understood social experiences and human phenomena. There are different variants of Grounded Theory. However, in line with my use of thematic analysis in Study One and Two, I adopted Charmaz’s version of *constructivist Grounded Theory* for conceptualizing and analyzing my data. In making my *constructivist ontology and interpretivist epistemology* transparent, I understand that my theory does not reflect true or accurate accounts that are simply “excavated from buried data that are waiting to be found.” In contrast, my Grounded Theory is an interpretive exercise; I consider my theory to be culturally and historically situated, offering co -
created interpretations about participant’s physical activity experiences, rather than true facts. Thus, I concur with Charmaz when she states that … data do not provide a window on reality. Rather, the discovered reality arises from the interactive process and its temporal, cultural, and structural contexts” (Charmaz, in Weed, 2009).

Weed (2009) and Holt and Timminen (2010) have recently contributed toward the scholarly dialogue on Grounded Theory in a manner that furthers the debate in fruitful and productive ways. In addition to encouraging Grounded Theory researchers to take responsibility for their ontological and epistemological assumptions and methodological decisions, they offer a set of eight guiding principles for planning and evaluating Grounded Theory studies. Seeking to avoid methodological policing, they suggest that these eight criteria should be considered as guidelines rather than hard and fast rules. However, adhering to these principles of “full fat” Grounded Theory may ensure that the theory is both internally consistent at the micro-level as well as externally relevant to other bodies of knowledge at the macro-level. I will briefly discuss my Grounded Theory of physical activity in CF and CHD in light of these eight criteria (Weed, 2010).

The process for creating my Grounded Theory entailed conducting a detailed analysis both within and across the multiple data sets from Study One and Study Two. In this regard, I worked with the original transcripts from 1) the CF children’s data set, 2) the CF parents data set, as well as 3) the CHD parents data set for a total of three data sets and 43 transcripts. Firstly, my collection and analysis of data in Study One and Two was an iterative and recursive process. Thus, data analysis was not a separate research activity, and, rather, my emerging analysis continued to drive data collection such that the two processes occurred simultaneously. Secondly, I employed theoretical sampling throughout the investigative process. I sampled newly emerging areas of inquiry – such as parental guilt for physical activity – to ensure that these emergent ideas were “fleshed out” in further interviews with children and parents. Third,
theoretical sensitivity - or the ability to remain open minded and free of preconceived hypothesis or bias - guided my analytical endeavor. Although I have extensive knowledge about the area of physical activity and childhood chronic disease, I endeavored to open myself to new lines of unexpected inquiry and constantly reflected upon my preconceived notions in order to adopt a theoretically sensitive approach. Fourth, while I did not adhere to Glaser and Strauss’s (1967) three way coding process, I systematically coded the transcribed data and ensured that I used written memos throughout the process to guide my systematic and analytic interpretation of the codes. This detailed coding process occurred both within and across data sets from Study One and Two, and entailed identifying, naming, and coding “chunks of data” or meaning units that are the smallest units which encompass meaning and conceptual clarity with respect to the research question. In this regard, my coding process was detailed and rigorous. Fifth, the constant comparative method guided my identification and sorting of data into “like and unlike” categories, and, although this is an interpretive activity, I ensured that there was conceptual consistency between categories and conceptual distinctions between them. Sixth, in both the children’s and parents study, the collection of fresh data failed to render new theoretical insights or to expand my concepts. In this regard, the data was theoretically saturated. Seventh, while different criterion are used to ensure the micro - level internal consistency and quality of a qualitative analysis, the principles of fit, work, relevance, and modifiability guided my judgments about research quality. In this regard, the resulting theory accurately “fits” and describes the data. Furthermore, the theory offers explanations to problems that are observed within the research context, such as parental stress, and reflects the concerns that are relevant to the participants. Finally, the theory is modifiable and open to further extensions and insights that may be offered in the future about the data. Finally, the development of a substantive theory of physical activity in youth with CF, CHD, and their parents (Figure Four) that is relevant to this substantive area was the final result of my analytic activity, a criterion which many Grounded
Theory researchers often fall short of. Although not stipulated by Weed (2009), I also tested my Grounded Theory by using it to inform a behavioural intervention, and the application of my Grounded Theory is arguably, a strength.

In summary, the resulting crystallized or Grounded Theory that is described in detail below was generated through a Grounded Theory analysis of the transcribed data from Study One and Two and offers an understanding of how a social group - children and parents affected by CF and CHD - experience physical activity. Furthermore, my resulting Grounded Theory and crystallization adheres to the eight guidelines recommended for “full fat” Grounded Theory (Weed, 2009), suggesting that I have not selectively chosen certain components of the method, while disregarding others. Readers should thus place confidence in the findings. Informed by social constructivism, it offers a historically and socially situated interpretation of how physical activity is experienced by these children and parents. My Grounded Theory or crystallization phase is illustrated and explained below within the context of the existing literature.
Figure Four: A Grounded Theory of Physical Activity in Youth with CF, CHD, and their Parents and Caregivers

“Living on the Edge of Health and Illness:” Family Fragility, Uncertainty, and Chaos

Disease Burden

Negative Activity Perceptions
- Illness Narrative: Despair
- Parental Support: Lacking
- Low Value

Positive Activity Perceptions
- Illness Narrative: Hope
- Parental Support

Low Self Efficacy

“Crystallization:” Toward a Grounded Theory of Physical Activity in Youth with CF, CHD, and their Parents and Caregivers
For CF and CHD youth and parents, *the burden of disease and treatment* was the most significant barrier toward physical activity. Because physical activity made youth feel chronically unwell, symptoms such as coughing or breathlessness led to activity avoidance. The negative impact of disease symptoms on the ability to be physically active has been reported in other studies involving youth with CF (Swisher & Erikson, 2008), and youth with other chronic diseases more broadly (Moola et al., 2008). This suggests that interventions must be designed in a way that remains sensitive to youths’ real and perceived illness symptoms. Youth who chronically feel unwell are not likely to want to be active.

Furthermore, youth and parents displayed *low value toward physical activity* and did not define it as an important pursuit. Rather, they demonstrated a gradual shift in priorities. Within the context of feeling unwell and lacking experiences that confirmed feelings of success in physical activity - youth did not define it as an important pursuit. This is a novel theoretical construct that has not been discussed in previous CF literature. However, other youth with chronic diseases display a progressive repartitioning of their attention in which they gradually come to define other activities as “more important” and divert their attention away from physical activity (Lunt et al., 2003; Moola et al., 2008). Additionally, parents stated that *low self-efficacy for physical activity* - or lacking experiences of success - were barriers that led their children to avoid physical activity. The environments that children undertook physical activity in, such as physical education settings or community physical activity groups, were often not self-efficacy supportive and were characterized by experiences of exclusion and hostility. Low self-efficacy for physical activity is well documented (Lunt et al., 2003; Moola et al., 2008), and appears to be a generic experience that characterizes youth with chronic diseases more broadly.

CF patients and their parents were generally well educated about the health benefits of physical activity, and, in comparison to youth with CHD and their caregivers, their knowledge was superior. However, participants were generally uninformed about the specific relationship
between physical activity participation and health outcomes in CF. Furthermore, unlike nutrition or nursing, there is no standard method of physical activity message delivery in the CF clinic. For this reason, knowledge and education about the importance of physical activity for CF is an important theoretical construct. In order to ensure that patients are informed and aware of how their current physical activity behaviours influence future health outcomes in CF, physical activity knowledge is an important theoretical construct to address.

Additionally, parental involvement in physical activity was central for all youth. For CF and CHD youth that were active, parents were critical to the provision of tangible and emotional support. In contrast, inactive CF youth lacked parental support for activity. They wished that their parents were more involved and expressed an interest in being active with their parents. Additionally, parents were aware of the importance of role modeling. Regardless of whether they described themselves as good or bad role models, in all cases, parents were aware that children role model and mimic parental physical activity behaviours. No studies have discussed the importance of parental involvement and support in CF youths’ physical activity. However, there are strong theoretical arguments - from the field of pediatric obesity - for the inclusion of parents in behavioural interventions that target youths’ physical activity and health. Given that parents provide instrumental and emotional support to engage in physical activity, by, for example, verbally encouraging their children or providing monetary assistance, they can be considered as critical “gate keepers” (Golan, 2006) to health behaviour change. Thus, parents either support or hinder health behaviours. Additionally, social cognitive learning theories underscore parents’ important role modeling function. Indeed, through social processes such as observational learning, children mimic their parents’ behaviours. Moreover, social ecological theorists increasingly support the move away from individual models of health behaviour change and suggest that multiple environmental determinants - such as parents, peers, schools, or the broader culture - exert an influence on children’s health behaviours (Elder et al., 2007). In this regard,
individual behaviours are not context stripped, and, rather are strongly influenced by the behaviours unfolding within the broader familial environment and social milieu. Finally, families are likely to be genetically and behaviourally similar. In addition to the clustering of physiological indicators, such as high cholesterol or high blood pressure, physical activity behaviours are likely to be similar among family members (Nader et al., 1996). In addition to the notable health benefits reported from studies that include and involve parents in youths’ physical activity, there is strong theoretical support for parents’ role modeling and supportive function, and this appears to be a relevant theoretical construct for CF and CHD youth and their families.

Youths’ illness narratives - and general outlook and perspective toward CF and CHD (Frank, 1995) - strongly shaped their perceptions toward physical activity. Resilient youth generally felt hopeful and optimistic in spite of CF/CHD and demonstrated a positive outlook toward physical activity. In contrast, other youth reported chronic despair, a sense of hopelessness, and a sense of futility and were more likely to be uninterested in physical activity. Clearly, youths’ perceptions toward their health and illness will have important implications for readiness to participate in physical activity research investigations, and this is discussed further.

In this regard, physical activity behaviours were a component of youths’ broader understanding of their own health and illness. Previous research has not discussed the inter-relationship between patients’ illness narratives and their orientation toward physical activity. However, several authors have drawn attention to the multiple illness narratives and stories that people with illnesses construct (Frank, 1995), and the importance of such narratives in making meaning out of their experiences.

Finally, the concept of “Living on the Edge of Health and Illness:” Family Fragility, Uncertainty and Chaos refers to the broader familial context and backdrop that characterizes these families. Indeed, these families are characterized by a fragile and precarious family structure. Living with uncertainty and a sense of foreboding, their grasp on health is tenuous and
they endeavor to prevent the encroachment and invasion of illness into their everyday lives. Specifically, parents reported high rates of marital failure as a result of the chronic stress of caring for an ill child and there were often dysfunctional communicative pathways between family sub-systems, such as “child - parent, sibling - sibling, or parent - parent.” High levels of stress among families caring for a child with CF are well documented in the literature, as well as patients and families affected by chronic diseases more broadly (Eiser, Zoritch, Hiller, Havermans, & Billig, 1995). Furthermore, child – non adherence to treatment was also reported as extremely stressful for parents as well as a lack of trust in health care providers and the sense that “others do not understand me.” It is critical to understand that the “Living on the Edge of Health and Illness” permeates and influences perceptions toward physical activity among CHD and CF youth and parents, underscoring that physical activity experiences are inseparable from health. Arguably, it is this sense of fragility, uncertainty, and chaos – characterized by stress, lack of trust, and marital conflict - that demarcates CHD and CF families as distinctly different from healthy able bodied families, adding a layer of complexity. That families affected by childhood chronic disease live with uncertainty, is well documented in the literature (Rempel & Harrison, 2007; Woodgate & Degner 2003). Family fragility, uncertainty, and chaos is not intrinsic to the family structure itself, and, rather, appears to be a consequence of caring for an ill child as well as other psycho-social and institutional barriers. However, it is critical to understand the sense of fragility and chaos that overwhelms these families and influences their global physical activity perceptions, as well as the precarious health/illness balance that parents and children negotiate.

Additionally, it is important to take note of an important caveat with respect to my Grounded Theory. My Grounded Theory offers a descriptive and interpretive tool to understand how physical activity is experienced by youth with CF and CHD as well as their parents and caregivers. The theoretical constructs in the model thus reflect the general physical activity
experiences of both the CF and CHD population, and has generally applicability to both groups. This is a strength of the model.

However, the behavioural intervention described below includes a disease-specific barrier that is specific to the CF population and not to the CHD group, that of temporal dilemmas. Participants with CF only understood CF to be a life-limiting disease that robs children of life time. Physical therapy or medicinal treatment, which may be undertaken multiple times a day, was defined as a time consuming and demanding practice that dominated youths’ time. Thus, youth with CF stated that they have insufficient time available to them in relation to their healthy, able-bodied peers. With “no time to play,” they did not feel that physical activity was a worthwhile time investment and they repartitioned their limited time elsewhere, to more important and valuable endeavors. The ways in which youth with CF negotiate physical activity within the context of significant temporal losses, appears to be a novel theoretical construct that has not been addressed previously. However, other studies have discussed CF youths’ complex time tricks and how the illness shatters a sense of temporal integrity (Glasscoe & Smith, 2008). Thus, in the intervention described below, this disease specific physical activity experience—which is likely common to those that live with life shortening pediatric diseases and/or involve a time consuming and arduous treatment burden such as muscular dystrophy—has been incorporated into the physical activity program.

In summary, the above theoretical constructs from my Grounded Theory and crystallization phase offer a conceptual illustration of how youth with CF and CHD and their parents understand and perceive physical activity (Figure Four), and was used to develop the CF Chatters parent-mediated behavioural counselling program described below.

Using Theory to Develop Interventions
Accordingly, the CF Chatters program described below was informed by my Grounded Theory and the central theoretical constructs that were distilled and crystallized from the qualitative data sets in Study One and Two (McEachan et al., 2008).

The CF Chatters manual was comprised of an introductory section that explained the program to the participants and discussed the program requirements. The three main chapters in the CF Chatters intervention and manual consisted of: 1) Knowledge and Awareness about Physical Activity in CF and Understanding my Values; 2) Planning to Engage in Physical Activity and Understanding my Environment, as well as; 3) My Barriers Toward Physical Activity and Dealing with Relapse. The three intervention chapters in the manual corresponded to the three program contact visits at the hospital. Each chapter included a series of interactive and engaging workbook activities that I developed. Participants were encouraged to read the chapter and to complete the workbook activities as homework between sessions. During each 90 minute counselling session, the chapter activities served as a way in which to prompt discussion between the researcher and the participant about the intervention content. Efforts were made by the researcher to adhere to a structured conversation that closely followed the content in the manual. However, when participants spontaneously engaged in new discussions that were not a component of the manual, such as cross-contamination issues in CF or academic accommodations for students with illnesses and disabilities at University, I discussed these concerns with the participants and temporarily veered away from the structured content. In keeping with the client - centred and participant driven nature of counselling and the emphasis of the qualitative paradigm on engaging participants in conversations that are meaningful and relevant to them (Mason, 2002), I regarded these conversations as important and relevant to the lives of CF patients and their families. Given that the CF Chatters Manual requires intensive training and workshop attendance to employ with patients – as well as appropriate clinical and
institutional support – it is not included in the appendix. Readers should contact the author of this dissertation to obtain further information about the Chatters manual.

More specifically, each workbook activity in the CF Chatters manual addressed one or more of the above theoretical constructs from my Grounded Theory, and, in this regard, the intervention activities were theoretically based. For example, workbook activities such as the “Wheel of Life” or “Where am I Going?,” were designed in order to specifically address participants’ values and whether or not physical activity could be incorporated into their broader value system. Additionally, intervention activities such as “Falling off the Wagon,” were designed to address the burden of disease and treatment. In this regard, each workbook activity in the CF Chatters manual addressed one or more of the central theoretical constructs from my Grounded Theory. A table of the CF Chatters manual activities and the corresponding theoretical construct(s) that it addressed from my Grounded Theory is provided below.
Table Four: CF Chatters Workbook Activities and Theoretical Constructs

<table>
<thead>
<tr>
<th>CF Chatters Session</th>
<th>Work Book Activities</th>
<th>Theoretical Construct</th>
</tr>
</thead>
</table>
| One                 | -What does PA mean to me?  
|                     | -How does PA make me feel?  
|                     | -What are some examples of PAs that I could engage in?  
|                     | -What are some examples of PAs that I would really enjoy engaging in?  
|                     | -PA and the lungs  
|                     | -What is CF?  
|                     | -How does PA affect CF?  
|                     | -The benefits of PA for CF  
|                     | -The benefits of PA for overall health  
|                     | -Other physical benefits of PA  
|                     | -Psycho-social benefits of PA  
|                     | -PA is safe  
|                     | What do I value in life?  
|                     | Where did I come from and where am I going?  
|                     | What are the advantages and disadvantages of becoming physically active?  
|                     | What does time mean to me? How do I want to use my time?  
|                     | -Is treatment time consuming?  
|                     | -Do I have enough time for PA?  
|                     | -Planning PA  
|                     | -PA calendar activity  
|                     | -PA action plan  
|                     | -PA coping plan  
|                     | -My neighbourhood and community  
|                     | -My PA environments  
|                     | -How can I better engage in PA in the environment?  
|                     | -Google earth map activity  
|                     | -How do you feel when you are ill?  
|                     | -What happens to your PA when you are ill?  
|                     | -Staying active during illness  
|                     | -Falling off the wagon  
|                     | -What are high risk PA situations?  
|                     | -PA relapse prevention plan  
|                     | -Obtaining social support  
|                     | -My PA barriers  
|                     | -Developing strategies to deal with barriers  
|                     | -The wheel of life  
|                     | -Caregiver health  
|                     | Physical Activity 
|                     | Knowledge 
|                     | Value Toward Physical Activity 
|                     | Temporal Loses 
|                     | Understanding Physical Activity Environments 
|                     | Disease Burden 
|                     | Low self efficacy for physical activity 
|                     | Parental support and role modeling 
|                     | Stress |

Secondly, one or more behavioural self regulation skills and strategies were employed in order to address each of the theoretical constructs in the workbook activities. Here, I broadly drew from behavioural self regulation skills theory - a school of thought that has advanced our understanding of how regulatory skills are necessary in order to link positive intentions to subsequent behaviours (Lewis, Marcus, Pate, & Dunn, 2002; Kahn et al., 2002; Rhodes, Naylor,
& McKay, 2010; Rhodes & Pfaeffli, 2009). Reviews of the literature on mediators of behavioral interventions purport that self regulatory constructs are consistent and predictable agents of behaviour change. Self regulatory skills training does not automatically ensure that intentions are translated into behaviour and may, for example, overlook the role of the environment in influencing behaviour change (Lewis et al., 2002; Rhodes and Pfaeffli, 2009). Although much more information is required in order to understand behaviour change in healthy populations, thus far, a self regulatory approach appears to be effective in promoting physical activity.

More research is required on the application of behavioural self regulation skills training to populations affected by chronic illnesses. However, in this study, I drew upon well documented, evidence-based behavioural self regulation skills for incorporation into the CF Chatters program, such as planning, self-monitoring, and contingency strategies. Respectively, these involve developing and describing the specifics of a behavioural plan, monitoring ones’ action toward the plan, as well as developing alternative, contingency plans should action toward the desired goal not occur.

In most cases, I drew upon evidence-based behaviour change strategies from the literature in order to address the theoretical construct in the workbook activity, such as goal setting, recording, planning and scheduling, monitoring, and psycho-education (Beech et al., 2003). For example, the workbook activity entitled “my barriers toward physical activity,” addressed “the burden of disease and treatment,” from my Grounded Theory. In turn, the behaviour change strategy of “behavioural costs and rewards” - or the pros and cons of changing - was employed in order to address this theoretical construct. Similarly, the workbook activity “understanding my time” was designed in order to address participants’ temporal constraints and perceived lack of time for physical activity. In turn, the behaviour change strategy of planning and scheduling was employed in order to address this theoretical construct from my grounded theory. Thus, a behaviour change strategy – outlined below –
was employed to address each of the theoretical constructs in the intervention workbook activities.

**Table Five: CF Chatters Workbook Activities, Theoretical Constructs, and Behaviour Change Strategies**

<table>
<thead>
<tr>
<th>CF Chatters Session</th>
<th>Work Book Activities</th>
<th>Theoretical Construct</th>
<th>Behaviour Change Strategy or Process</th>
</tr>
</thead>
</table>
| One                 | -What does PA mean to me?  
-How does PA make me feel?  
-What are some examples of PA’s that I could engage in?  
-What are some examples of PA’s that I would really enjoy engaging in?  
-PA and the lungs  
-What is CF?  
-How does PA affect CF?  
-The benefits of PA for CF  
-The benefits of PA for overall health  
-Other physical benefits of PA  
-Psycho-social benefits of PA  
-PA is safe | **Physical Activity Knowledge** | -Psycho-Education  
-Cognitive behavioural skills training (CBT)  
-Goal setting |
| One                 | What do I value in life?  
Where did I come from and where am I going?  
What are the advantages and disadvantages of becoming active? | **Value Toward Physical Activity** | -Psycho-Education  
-CBT  
-Behavioural cost and reward  
-Goal setting |
| Two                 | -What does time mean to me? How do I want to use my time?  
-Is treatment time consuming?  
-Do I have enough time for PA?  
-Planning PA  
-PA Calendar activity  
-PA action plan  
-PA coping plan | **Temporal Losses** | -Planning and Scheduling  
-Goal setting  
-Monitoring and recording  
-Psycho-education  
-CBT |
| Two                 | -My neighbourhood and community  
-My PA environments  
-How can I better engage in PA in the environment?  
-Google earth map activity | **Understanding Physical Activity Environments** | -Psycho-Education  
-CBT  
-Mapping exercises |
| Three               | -How do you feel when you are ill?  
-What happens to your PA when you are ill?  
-What happens to your PA when you are ill?  
-Staying active during illness  
-Falling off the wagon  
-What are high risk PA situations?  
-PA relapse prevention plan  
-Obtaining social support  
-My PA barriers  
-Developing strategies to deal with barriers | **Disease Burden** | Goal setting  
Behavioural Cost and Reward  
Psycho Education  
CBT |
| All sessions        | -The wheel of life  
-Caregiver health | -Low self efficacy for physical activity  
-Parental support and role modelling  
-Stress | -Psycho-Education and CBT |
Additionally, the three theories identified in the developmental phase of the MRC framework in Chapter Three, broadly informed the development of the intervention too. While specific content was not designed to address these theories, these conceptual models broadly informed my thinking and delivery of the intervention. For instance, family systems theory allowed me to consider physical inactivity not as a personal problem but perhaps indicative of broader family functioning (Cummings, 2002). It also directed my attention to how sub-systems within the family operate. Furthermore, the social ecological models sensitized me to thinking about the multiple environments that children and parents are embedded within when they seek to engage in physical activity (Elder et al., 2007). Finally, the movement continuum theory drew my attention to participants’ current, preferred, and maximal physical activity capacity and how to reduce discrepancies between these constructs. It also reminded me of the broad, body-society conceptualization of movement (Cott & Finch, 1995).

Psycho-education (PE) and elements of the cognitive behavioural psychological tradition (CBT) were also employed in this study. While these traditions are more commonly used for patients with mental health conditions, psycho-education entails providing patients with information, support, coping, and problem solving skills, and is particularly effective in reducing remission and relapse (Chow et al., 2010). While the formalized delivery of CBT entails a four-stage process, the process does include identifying, modifying, and challenging the illogical thoughts that guide behavior (Spangler, 2010). PE and CBT were critical to the delivery of some of the intervention content and are also illustrated in Table Two. For example, workbook activities such as “is physical activity safe,” required me to discuss participants’ and parents’ underlying beliefs about the safety of physical activity for CF. In some cases, parents felt that physical activity in CF could be unsafe, reflecting errors in reasoning and conceptualizations about physical activity. This required me to discuss the “cognitive error” with participants and then explain what the research evidence states about the safety of physical activity for medically
stable and asymptomatic patients with CF. Similarly, in the “understanding my environment” workbook activity, I often had to educate parents and participants about how broader cultural, school, and policy environments influence their physical activity. In this regard, elements of the PE and CBT tradition were employed in this study, especially if participants demonstrated cognitive errors related to health and physical activity.

In summary, CF Chatters was informed by developmental work and qualitative evidence from Study One and Two, by people who are living with CF and their caregivers. As a concept map and Grounded Theory, these theoretical constructs may also be conceptualized as mediators of physical activity in children and parents with chronic diseases. The constructs tap into the physical activity experiences of youth with CF and their parents and have provided a useful theoretical model to guide the development of CF Chatters. Each workbook activity was designed based on concepts from my theory and a behaviour change strategy was employed to facilitate change.

Research Setting and Participants

This pilot study occurred in the CF Clinic at the Hospital for Sick Children (SickKids) in Toronto, Canada. SickKids is a world renowned pediatric facility that specializes in the care of children with acute and chronic illnesses and disabilities. Given the ethical issues involved in counselling sick children and their caregivers, it was important to ensure the safety of my participants. Firstly, the counselling sessions were approved by the Research Ethics Board at the Hospital for Sick Children, in which it was deemed that the slight emotional risks to participants were of no greater magnitude than that which they would have encountered in their everyday lives. Furthermore, as outlined in the REB application, steps were documented in order to assist me with the management of any emotional risks that may have arisen during the investigation, such as reporting adverse psychological outcomes to the clinical team. Furthermore, in order to ensure the safety of the participants and caregivers, the counselling program was carefully
monitored by two nurses, the exercise physiologist, and an overseeing physician, and, in this regard, I had a great deal of institutional support and oversight of the program. Although no significant problems arose during the sessions that indicated participant distress, I regularly reported participants’ progress to these clinicians and was in constant dialogue and conversation with them. Furthermore, my qualifications – as a Doctoral Student in Health and Exercise Psychology – were made transparent, and participants were aware that I was not a clinical psychologist.

In addition to these institutional measures which ensured participant safety and protection from harm, I took actions during the program to ensure their comfort. Participants were continually reassured that participation in the program was voluntary and that they were free to skip over or terminate any discussions that aroused discomfort or upset. Finally, children’s participation was closely monitored by their parents and caregivers. Parents were directly involved in the sessions with two of the participants, and engaged in the content with their child. Chase’s father accompanied him to all of the sessions and quietly read a book in the corner during session two and three. Although Layla’s father was distantly involved, I continued to make efforts to report her progress to her caregiver. In this regard, parental support and consent for CF Chatters was maintained throughout the course of the investigation.

It is important to note that although there is always an inherent risk associated with participation in research, literature on the history of pediatric research suggests that the greatest risk afforded through research to sick children comes from Phase One pharmacological trials. Indeed, this type of research – which tracks drug mechanisms - is often undertaken with the most ill patients who have not benefited from other interventions; it is often falsely equated with treatment and is pursued as a desperate “last chance” for children and parents, and often imbued with false hopes (Oberman & Frader, 2003). While the risk through research is inherent, according to the REB matrices of risk, the risk afforded through the counselling sessions was
low, no greater than the risks associated with everyday life, and of an emotive nature. In addition to taking all foreseeable institutional and social measures to ensure my participants’ safety and security during the program, I had well developed and immediate plans – to report adverse psychological outcomes to overseeing staff and refer to psychological services – should events have arisen. Most importantly, as discussed below, the participants in the program underscored the importance of psychology and discussing psycho-social health in the care of cystic fibrosis, and were celebratory of psychological counselling.

A total of six participants were enrolled in the CF Chatters program. More specifically, four children and youth with CF participated as well as two parents/caregivers; two parent-child dyads participated and the remaining two youth opted to participate without a parent. The child and youth participants ranged in age from 11 to 17; the mean was 14. Although FEV1 values ranged from 70 to 87%, functional status and activity of daily living participation varied. Three girls and one boy participated. One male and one female parent participated and the mean age of the parent was 44 years. Participants were from low to mid socio-economic status brackets. Finally, five of the six participants were enrolled in the previous study and only one participant was new to the study. The participants’ characteristics are described below. An indicator of disease severity is included in this table (FEV 1), and is the measure of health status that corresponds to the time in which the study was undertaken.
Table Six: Participant Descriptions, CF Patients

<table>
<thead>
<tr>
<th>Name</th>
<th>Age</th>
<th>Disease Severity: FEV 1</th>
<th>Gender</th>
<th>Hospitalized Previously</th>
<th>Participated with or without parent</th>
<th>Region</th>
</tr>
</thead>
<tbody>
<tr>
<td>Chase</td>
<td>17</td>
<td>85%</td>
<td>M</td>
<td>Yes</td>
<td>Without parent</td>
<td>Toronto</td>
</tr>
<tr>
<td>Layla</td>
<td>16</td>
<td>85%</td>
<td>F</td>
<td>Yes</td>
<td>Without parent</td>
<td>North Toronto</td>
</tr>
<tr>
<td>Emily</td>
<td>11</td>
<td>71%</td>
<td>F</td>
<td>Yes</td>
<td>With parent</td>
<td>Whitby</td>
</tr>
<tr>
<td>Zoe</td>
<td>12</td>
<td>87%</td>
<td>F</td>
<td>No</td>
<td>With parent</td>
<td>Georgetown</td>
</tr>
</tbody>
</table>

Table Seven: Participant Descriptions, CF Parents

<table>
<thead>
<tr>
<th>Name</th>
<th>Age</th>
<th>Child</th>
<th>Gender</th>
<th>Occupation</th>
<th>Martial Status</th>
<th>NO. of Children</th>
<th>Region</th>
</tr>
</thead>
<tbody>
<tr>
<td>Erik</td>
<td>35</td>
<td>Emily</td>
<td>M</td>
<td>Consultant</td>
<td>Married</td>
<td>2</td>
<td>Whitby</td>
</tr>
<tr>
<td>Gretchen</td>
<td>53</td>
<td>Zoe</td>
<td>F</td>
<td>Consultant</td>
<td>Remarried</td>
<td>2 (also has CF)</td>
<td>Georgetown</td>
</tr>
</tbody>
</table>

Overview of CF Chatters Program

The CF Chatters program is a theoretically informed parent-mediated cognitive behavioural program that relies upon counselling and behavioural skills regulation training as methods of intervention delivery. Individual counselling was employed with the two youth that participated independently of their parents, and small group based counselling was employed with the remaining two parent-child dyads. The intervention includes a manual that outlines the program content. As a) an individual who has a great deal of experience working in the field of pediatrics, b) has a high degree of Post Graduate training and education in psychology, and c) possesses a
great deal of empathy and awareness, I considered myself as an exercise counsellor. In addition to helping participants to identify and reduce barriers to physical activity and “rolling with resistance” (Loughlan & Mutrie, 1995), in my capacity as an exercise counsellor, I was well equipped to deal with the tremendous complexity associated with these families. Patients affected by chronic disease are likely to live with uncertainty and experience good “days and bad days” that are characterized by disease exacerbation (Charmaz, 1993). For this reason, in my capacity as an exercise counsellor, I was also well equipped and prepared to deal with patient disease exacerbation throughout the program, and, so long as youth remained medically stable, I encouraged adherence to the program even during periods of illness.

The intervention occurred over the course of six weeks in the CF clinic at SickKids. With the exception of one parent-child dyad, the meetings occurred after clinic hours in the late afternoon or evening. There were four meeting points in the CF Chatters program as well as multiple reminder phone calls throughout the two week intervals. At Time One (T1), the researcher met with the participants for half an hour and explained the CF Chatters program. Informed consent was obtained at T1 and the Habitual Activity Estimation Scale (HAES) was administered to the participants, as well as the CF Chatters Parent and Child manual. Session one of the CF Chatters program occurred two weeks after the initial meeting at Time Two (T2). The purpose of this 90 minute session was to increase participants’ knowledge and awareness of physical activity and discuss the value of physical activity in participants’ lives. Session one concluded with setting a physical activity goal. Session two of the CF Chatters program occurred two weeks later at Time Three (T3). The purpose of this 90-minute session was to address participants’ temporal anxieties by increasing their capacity to plan and schedule physical activity. Session two also aimed to increase participants’ ecological acuity or their ability to better access physical activity opportunities in the environment. This session concluded with physical activity goal setting. The final meeting was held two weeks later at Time Four (T4).
This 90 - minute session addressed participants’ barriers toward physical activity and aimed to equip them with the skills necessary in order to better manage CF relapses and deteriorating health. The program concluded on this day. The HAES physical activity tool was administered again and all participants engaged in an exit qualitative interview. A schematic representation of the program is depicted below. Each participant or child-parent dyad engaged in approximately seven counselling hours and the entire study involved 28 to 30 counselling hours.

Figure Five: Overview of the CF Chatters Intervention

**Qualitative Research Tradition: Case Study Research and the Stories we Tell**

Objective quantitative data is necessary in order to make certain evidence - based claims about the efficacy of the CF Chatters program and is the focus of future studies. However, it is necessary to draw upon the qualitative paradigm as a forum of inquiry that can help us to better understand how participants perceive and appraise behavioural interventions (Stake, 2000; Flyvberg, 2004). Garnering such information at the initial or developmental phase of intervention development is critical in order to ensure that future programs are relevant and sensitive to participants’ social lives and dilemmas, and is supported by the MRC framework (McEachan, 2008).
The qualitative research tradition that guided the development of this study was case study research. Case study research, which employs multiple methods, can be described as the in-depth study and investigation of a few lives in context (Stake, 2000). Although case study research has a long history in medicine, it has been adopted by a range of diverse fields such as marketing, psychology, and law. While case research was initially grounded in a positivist epistemology, researchers are increasingly employing the case study in ways that take account of the socially and historically situated nature of social life and the culturally contingent production of knowledge. Central to case research is the foundational assumption that valuable and important knowledge can be derived from the study of the particular and the individual (Stake, 2000). While case studies cannot be generalized to a larger group or population, important and novel information can be derived from the case study. The case study carefully treads the line between the general and the specific, or the generic and the particular (Stake, 2000). Indeed, as suggested by Woods, case studies “emphasize the context bound uniqueness of participants’ stories while common themes reveal the junctures in their shared experiences” (1992, p. 96). Through case contrast and comparison, then, it can thus act as a useful forum in which to confirm existing understandings and insights about CF and physical activity, as well as generate novel insights which capture the particularities of participants’ lives. Researchers are advised to conduct multiple case studies that allow for simultaneous comparisons between many cases. Finally, the case study is a particularly valuable form of intervention research in that it illuminates participants’ individual uptake and appraisal of a program (Stake, 2000; Faulkner & Sparkes, 1999; Flyvbjerg, 2004). By “invite (ing) the reader into a vicarious experience (however brief) of the life or lives being described” (Sparkes, 2000, p. 33), case studies always provide a provocative story and rich description of a few lives in context and are a particularly useful way of understanding an individual’s experience of an intervention. Case study research is composed of multiple methods and these are briefly described below.
Semi structured interviews were employed in order to explore participants’ experiences of the CF Chatters program. Although a semi-structured interview guide was developed by the researcher in order to understand their experience of the intervention, the participants’ responses drove further lines of inquiry and the interview proceeded in terms of a recursive conversation. Thus, the semi-structured interview provided a useful lens in order to understand participants’ experiences of CF and physical activity and their perceptions of the program. These interviews were digitally taped and transcribed verbatim (Kvale, 1996; Mason, 2002) (See APPENDIX Three for the CF Chatters Interview Schedule).

Field notes: A detailed field note journal was used throughout the study in order to record, describe, and characterize participants’ responses to the CF Chatter’s program, institutional observations, personal reflections, and novel insights about the program. A few hundred pages of field work data was collected over the course of the CF Chatters program and observations were recorded before, during, and after each session. The field work data further corroborated the interviews and provided an excellent forum in which to understand the unique facets of participants’ lives as well as their response to the program (Faulkner & Sparkes, 1999).

Habitual Activity Estimation Scale (HAES): Finally, the HAES was employed as a component of the broader case research. I selected the HAES tool as a self report measure of physical activity in order to understand whether or not the program was associated with increases in physical activity (Wells et al., 2008). The HAES is a reliable and valid tool that was developed by pediatric researchers in order to assess level of habitual physical activity in children with chronic diseases. Initially derived from work with childhood survivors of cancer, the HAES has been validated for use with the CF population. The HAES is an easy and convenient tool that can be completed in ten minutes. It estimates the percentage of time that children and parents in this case, spend being a) non active b) somewhat inactive c) somewhat active or d) active. The HAES is divided into time quartiles which relatively approximate children’s daily time routines and
schedules, such as activities before breakfast or at school (Wells et al., 2008). Data is collected based on one typical, regular, or average weekday or weekend day. The HAES is oriented toward a broad conceptualization of physical activity, rather than exercise and was thus well suited toward a type of program such as CF Chatters. Accelerometer data is needed in order to provide objective information about physical activity and is the focus of future interventions. However, the purpose of this study was to explore participants’ perceptions toward the CF Chatters program through the qualitative paradigm, and, as a researcher, I sought to avoid burdening participants with too many time consuming data collection methods. For this reason, I selected the HAES as an efficient and informative measure of physical activity (Wells et al., 2008; See APPENDIX Four).

Case Study Data Analysis: There are multiple ways in which case study research can be analyzed. For example, case studies can be analyzed in a chronological fashion which describes an individuals’ story over the course of time (Stake, 2000). Case studies can also be analyzed by theoretical postulates, in which a theoretical framework, such as social cognitive theory or family systems theory, provides a framework for thinking about and analyzing the case. Alternatively, case studies can be conducted in a descriptive fashion in which the in-depth story of a particular life is storied (Stake, 2000).

In order to analyze the case studies, first, a thematic analysis of the transcribed qualitative interview data was generated (Braun & Clarke, 2006). These interview question responses, such as the program impact on physical activity and quality of life, were coded, named, and grouped into broader themes. Novel responses that were not a component of the interview guide, such as how the program facilitated “tough talks” or the psychological and therapeutic benefits of the program, were also named, coded, and grouped into themes. This thematic analysis of the transcribed data allowed me to understand participants’ overall perceptions toward the CF Chatters intervention (Braun & Clarke, 2006).
Secondly, multiple analytical techniques - such as analysis by way of description and theoretical postulate - were employed in this study and novel information was garnered by contrasting the different CF cases (Stake, 2000). For example, my case studies were analyzed within my *Grounded Theory of Physical Activity in CF*, and I sought to create “story profiles” of each participant. Then, the field work and transcribed data were combined as multiple data sources in order to compile case stories, and provided the broader framework for how the cases were analyzed and organized. For example, field work notes and the transcribed interview data were drawn upon in order to compile a description of the participants’ history and background, personal story living with CF, and response to the CF Chatters program.

In summary, case study research - involving interviews, field notes, and the HAES tool - were employed in order to evaluate how six patients and parents living with CF experience physical activity and their uptake of the counselling program. The findings, including extracts from the interviews and field notes, are provided below.

### 6.5 Findings

*Aggregate Overview*

Eight participants enrolled in the CF Chatters program. However, after the first session, one parent-child dyad (two participants) dropped out due to a CF related exacerbation, and, thus, there was an 80% retention rate. The remaining six participants (two parent-child dyads and two adolescent participants) demonstrated 100% attendance in all sessions. Across the case studies, all participants reported that the intervention was easy, convenient, interesting, enjoyable, and relevant to their physical activity concerns. This suggests that overall, the intervention was acceptable and satisfying. CF staff reported that the program was timely, efficient, and likely to be delivered to other patients. It did not disrupt clinic flow or excessively burden the clinic, suggesting that the intervention was feasible.
Across the four case studies, the qualitative findings indicate that the program increased physical activity behaviour and quality of life in the physical domain for the majority of the participants. The program also enhanced knowledge about the importance of physical activity for CF, as well as perspective and insight about “the self.” The program resulted in improved caregiver health and facilitated “tough talks” that would have been difficult to discuss in the absence of a trained exercise counsellor. The program was reported to be easy, convenient, and relevant to participants’ physical activity concerns and offered important therapeutic benefits. In this regard, the broader study findings across all of the case studies point to the overall benefit of the program to participants’ lives and their positive perception toward - and appraisal of - the program.

The pre and post-test HAES data indicate that for the majority of the participants, the CF Chatters program resulted in increased physical activity - on a typical, average, or regular weekday - over the course of the six-week intervention. Despite reporting uncertainty over whether or not their physical activity behaviour changed in their qualitative interviews, the HAES data indicate that for two of the six participants (one parent-child dyad), physical activity did not increase over the course of the intervention. However, the pre and post-test HAES data from the remaining four participants (one parent child dyad and two youth) indicate significant increases in percentage physical activity/day, total hours of daily physical activity (the sum total of the “somewhat active” and “active” categories), as well as percentage daily activity in the “most active” category. Indeed, in order to ensure that physiological benefits are derived from physical activity, activity occurring in “vigourous or most active categories,” is required. Therefore, across the participants, the CF Chatters intervention was successful in increasing physical activity among 67% of the participants. A more detailed explanation of the CF Chatters HAES data is provided below.
**Table Eight: HAES Data**
1) Percentage Change in Daily PA 2) Change in Total Hours of Daily PA 3) Change in Percentage and Total Hours of “Active” Category PA

*Child Participants*

<table>
<thead>
<tr>
<th>Name</th>
<th>Age</th>
<th>Pre to Post Test Change, % Total Daily Activity</th>
<th>Pre to Post Test Change in Hours of Daily PA</th>
<th>Pre to Post Test Change, Active Category</th>
<th>Outcome</th>
</tr>
</thead>
<tbody>
<tr>
<td>Layla</td>
<td>16</td>
<td>7.9%</td>
<td>1.89 hours</td>
<td>3.3 % 0.8 hours</td>
<td>Increase daily PA</td>
</tr>
<tr>
<td>Chase</td>
<td>17</td>
<td>6.6%</td>
<td>1.58 hours</td>
<td>1.9% 0.46 hours</td>
<td>Increase daily PA</td>
</tr>
<tr>
<td>Emily</td>
<td>11</td>
<td>18.5%</td>
<td>4.44 hours</td>
<td>24.7% 5.93 hours</td>
<td>Increase daily PA</td>
</tr>
<tr>
<td>Zoe</td>
<td>12</td>
<td>7.6%</td>
<td>1.83 hours</td>
<td>1.1% 0.25 hours</td>
<td>Decrease daily PA</td>
</tr>
</tbody>
</table>

**Table Nine: Adult Participants**

<table>
<thead>
<tr>
<th>Name</th>
<th>Age</th>
<th>Pre to Post Test Change in % Total Daily Activity</th>
<th>Pre to Post Test Change in Hours of Daily PA</th>
<th>Pre to Post Test Change, Active Category</th>
<th>Outcome</th>
</tr>
</thead>
<tbody>
<tr>
<td>Erik (Child, Emily)</td>
<td>35</td>
<td>21.6%</td>
<td>5.17</td>
<td>14.2% 3.49 hours</td>
<td>Increase daily PA</td>
</tr>
<tr>
<td>Gretchen (Child, Zoe)</td>
<td>53</td>
<td>6.2%</td>
<td>1.48</td>
<td>No change</td>
<td>Decrease daily PA</td>
</tr>
</tbody>
</table>

Since case studies provide a rich and provocative story that straddles the line between collective experience and the unique particularities of participants’ lives (Woods, 1992), below, I explore, discuss, and interpret the individual case stories and the response of the participants toward the CF Chatters program. Each case study follows a similar narrative structure to discuss a) the history and context of the participants, b) the participants’ health and illness story, as well as c) their response to the intervention.
Chase: “I Want To See the World Through Another Person’s Perspective”

Context and background: Chase is an intelligent and thoughtful 17-year-old boy with CF who was experiencing a respiratory infection at the time of the intervention. His FEV1 is 85%. Although Chase coughed several times throughout the sessions, and required time to regulate his breathing, he has the outward appearance of a healthy boy. Chase is from a two-parent middle class family and resides in a comfortable urban home. His father is a consultant and his mother is a cosmetic specialist. Chase has a younger brother who does not have CF.

Chase’s story: During the program, Chase explained that his perspective toward his illness had recently undergone a radical change. He recounted negative stories of himself as a child and spoke of obtaining a “new lease on life.” With a grimace and cringe, Chase conjured up an unpleasant image of himself as a child in his mind’s eye - one who was uninformed, unhealthy, and lazy, lacked awareness into health-related matters, and passively followed the rules outlined by his parents without question. Chase explained that as a child, he did not know what CF “really was” and simply “did what his parents told him to” without understanding the rationale for or consequences of treatment. With a near visible disdain and repugnance for the boy that he once was, Chase recounted memories of a child who lacked a sense of passion and commitment. For example, the “former Chase” was one who enjoyed physical activity, but who only undertook it at specialized centres for children with disabilities. The “former” Chase did not want to go away to college or University for fear of the health-related problems, and, instead, envisioned himself living at home with his parents where he did not have to expose himself to unnecessary risks. The “former” Chase had a small social network and was largely isolated from his peers. The “former” Chase expressed disdain for the distressing bodily changes that the medication prednisone - used to treat his CF - had caused.

Chase attributed his personal metamorphosis to a 60 day “River Trip” that he undertook at a Northern Ontario residential summer camp. On the River Trip, Chase’s social network
expanded to include socially conscious and interesting new friends who saw past CF and accepted him as he was. The physically demanding nature of the trip - and the endurance required to paddle the river on a daily basis - allowed Chase to develop a new found respect for his body, and, more importantly, a desire to take better care of himself through nutrition and physical activity. Furthermore, camp provided Chase with the opportunity to meet other youth with disabilities, such as autism. Chase has a contradictory relationship with his own disease. However, meeting others with disabilities at camp has led him to be fascinated by various illnesses and fostered an overwhelming desire to help others who are ill.

Thus, the changed boy who participated in the CF Chatters intervention was insightful and reflective and possessed a strong health orientation. Although he dislikes much of the high school curriculum which “really does not teach you anything useful,” Chase diligently pursues his studies and is focused upon attaining a meaningful career in the future. Chase has taken strides to educate himself outside of the constraints of the high school academic curriculum and is an avid reader of philosophy. For example, he is particularly interested in the history of time and frequently urged me throughout the program to share my current book list with him. Although Chase expressed a desire to “see the world from another person’s perspective,” he is unsure of how to go about doing so. To start, Chase will be spending a year abroad with a colleague from school working at “Larche” in France - that is, an internationally renowned community for the intellectually disabled that has provoked the attention of scholars and health professionals alike. Moreover, Chase’s social network has - and continues to - expand, and he expressed an interest forming new social relationships. Shyly and somewhat awkwardly, he also described the importance of a new romantic relationship in his life and how important her interests, such as Celtic dance, are to him. Thus, despite the daily hassle of treatment, it appears that Chase has learned how to balance his competing health needs with his social life and enjoys an ever expanding and stimulating social world. Additionally, Chase’s complete commitment to
the CF Chatters program was evidence of his resolution to ensure good physical health and to slow the progression of CF over time.

Finally, Chase expressed an almost insatiable desire to know more about the lives of other children with CF and was interested in details, such as how ill other children are in relation to him. Whenever any mention was made of other patients during our sessions, Chase was immediately captivated and hungered to know more. He was also fascinated by the volunteer work that I do with other sick children. Unfortunately, the medical restrictions that prevent children with CF from interacting with one another are particularly burdensome for Chase. Chase skeptically questioned clinical recommendations that CF youth not congregate with one another and illuminated a complicated ethical dilemma, in which the medical risks of congregating-cross contamination-are in tension with the social risks of congregating-isolation. The inability to satisfy Chase’s desire to know more about other CF patients and to be unable to abate his hunger for this knowledge was an ethical conundrum which plagued me throughout the program:

I learned a little about other kids with CF through the program. I wish that I could see more of them and to relate my CF to theirs … I want to know their fitness plans and their lifestyles. I do not know them. Maybe they have something that I do not know about. And I have no way to learn about that. You will learn about all that stuff, but I cannot know that stuff … Because there is no one that I can really relate to. When I am having a conversation, there is no one that I can talk to, unless it is asthma or another disease …

But, I don’t know, it would be nice to talk with someone that has CF as well.

CF Chatters Program Response: According to the pre and post-test HAES data, Chase demonstrated significant improvements in percentage daily physical activity (6.6 %), total hours of daily physical activity (1.58 hours), and percentage daily activity in the most active category (1.9%) on a typical, average, and regular weekday, over the course of the intervention.

Furthermore, according to his qualitative report, Chase greatly benefited from the program in the
area of physical activity behaviour change, quality of life, and learning and insight about himself.

“Not being able to keep up with my peers” was distressing for Chase, and, for this reason, he tailored his physical activity goals during the program in order to facilitate increased endurance and running ability. Chase was not interested in pursuing a diverse range of activities, and, rather, his goals were very specific and centred upon running. Specifically, Chase challenged himself to run more on the treadmill at his school and community gym; he sought to increase the frequency of running from zero to four times/week and the duration from 15 to 20 minutes / session. He also set physical activity goals that allowed him to experiment with novel running activities and tasks, such as treadmill running, running outdoors, and parcour or “urban play” running - an activity that entails using the shortest possible distance to run from point A to point B. Goal setting was the most successful behavioural regulation skill for Chase, and, by setting physical activity goals on a regular basis, Chase increased his level of physical activity and engaged in behaviour change:

This program, when I learned that there was a goal that I wanted to reach-I always knew that I wanted to get more endurance, but I did not think about it much. But this program, made me think well “what is your goal?” And once I did that-how long, where, when-it was easier to do and I took action and did it. Otherwise, I would not have done it.

For Chase, engaging in more physical activity was associated with a slight increase in the physical domain of quality of life. Specifically, since improved endurance reduced Chase’s exposure to unpleasant and distressing social situations where he is not able to “keep up” with others, the program improved his quality of life:

My quality of life altered a little. With the running-wanting to run more and then actually changing to run more-that is only going to help. So, my quality of life did raise a little.

_Fiona:_ What part of your quality of life did it help?
It helped the physical part of quality of life, with endurance and being able to run more. Endurance comes up all of the time. It does not have to be running. It could be who knows- whatever kind of activity. Endurance and stamina are important in CF … I can avoid situations that bring me down, because I am not able to do something. I will be able to avoid those things with better endurance and have a better quality of life.

Chase reported that he acquired insight and knowledge about himself as a result of participation in the program. While he had to some extent always known certain things about himself, more focused and structured discussions, around issues such as values and beliefs, allowed him to better identify his motivations, drives, intentions, and barriers. Indeed, it appeared that enhanced “knowledge about the self” was the greatest benefit afforded to Chase through the program. Chase was adamant and suggested that the discussion-counselling format utilized in the program was critical to enhanced self knowledge:

Yes, I learned things about myself, by doing things like this -“the life wheel” (activity) - I saw things that I had structured but had not realized … I have always known about my barriers but I have never thought about them as much as I have in the program. I have always known that CF is a barrier, but, until we talked about it on this page, I did not know just how much it was … I have someone to talk to and talk with you. To have someone here with me to talk about this … You definitely need a counsellor for this.

Lastly, Chase reported that the program was easy to do and convenient and he described it as a worthwhile time investment. However, unlike the other participants who unquestionably accepted the program as “great,” Chase was unafraid to offer polite commentary and critique that would enhance the program. For example, since good nutrition is necessary to engage in physical activity, Chase suggested that the program should also include content on nutrition:
Well, I feel that in order to fully and to efficiently exercise, you need to eat healthy too. Without that, you will get healthier but not to the same extent as you could. Nutrition should be thrown into one of the chapters.

**Summary:** Chase is an exemplar of a young person who is living well with CF and who changed his physical activity behavior, quality of life, and knowledge about himself by participating in the program. More specifically, Chase’s response to the program can be interpreted within the context of my existing Grounded Theory, as well as the other theoretical frameworks in this study. In addition to enhanced self awareness, Chase reported increases in knowledge about the importance of physical activity for CF, as well as enhanced strategizing about how to deal with the burden of disease and treatment. Chase also reinforced the importance of physical activity and health to his broader value system. Chase broadened his understanding of the role of CF in his life and displayed an improved disease outlook and perspective. He emphasized the importance of living well with CF for as long as possible and undertaking health behaviours today that will influence his future health outcomes. Furthermore, in line with family systems theory and the social ecological model, although Chase’s father did not directly participate in the program, he was very supportive of Chase’s involvement, facilitated his participation, and remained informed about Chase’s physical activity progress (Cummings, 2002; Elder et al., 2007). As such, theoretically, the program appeared to alter patterns of physical activity communication in the child-parent sub-system or Chase’s immediate micro-system. Furthermore, according to the Movement Continuum theory, Chase’s desire to challenge himself and improve his stamina through more vigorous running activities, arguably attenuated the gap between his CMC and PMC (Cott & Finch, 1995).

Chase has largely overcome his early struggles to have an active and informed role in his health, and the avoidant approach that he once adopted to deal with the burden of CF. Chase is a motivated, intelligent, inquisitive, caring, and socially conscious boy who is devoted to attaining
optimal personal health with CF. Chase desires to take care of himself despite knowledge of the harsh health-related implications of CF. He shows no indication of helplessness or futility, and, engaging in treatment, physical activity, and good nutrition appear to be worthwhile time investments that enhance perceptions of control and autonomy over his health. While he foresees that he may need “treatment reminders,” Chase expressed no fear of transition to the adult CF centre. Chase also displayed concern for the broader social world around him - and its complexities - especially to help those that suffer. At the end of the program, the therapeutic benefit to Chase was evident and he expressed sadness at the loss of an important social relationship. Based on a telephone assessment, two weeks after the termination of the program, Chase continues to adhere to regular physical activity goals.

Layla: “I Never Tell Others What I Really Think”

*Context and background:* Layla is a 16-year old girl with CF and her FEV1 is 85%. During the study, Layla was experiencing prolonged and excessive fatigue and lethargy of unknown etiology, and reduced energy not relieved by sleep or rest. Layla is thin, pale, and fragile in appearance and her countenance can generally be described as one of sadness and despondency. She has great difficulty making eye contact with staff members and her voice lacks confidence and enthusiasm. Layla was the only participant in the study who is a visible minority. Layla is from a two parent, low socio-economic status suburban family. She resides in a housing complex with her six brothers and sisters and her parents who emigrated to Canada from Afghanistan a few years ago. Layla’s parents do not work and she “has no idea how they make their money.” With the exception of her youngest brother who also has CF, Layla’s siblings are healthy. Layla’s family adheres to the principles of fundamentalist Islam. Although Layla at times expressed disagreement with the strict religious sanctions in her home, her relationship to her religious faith is contradictory. She wears the hijab and suggested that she enjoys attending
the mosque and other religious gatherings with her family. Indeed, for Layla, religion is a “guide which keeps me on track and explains things.”

_Layla’s story:_ Within the clinic, Layla is the quintessential “negative,” “high risk,” or difficult patient. Known as a “trickster,” clinical staff strongly suspect that Layla fails to adhere to her life saving treatment protocol and regularly produces medical evidence to indicate her propensity to tell falsehoods. She rarely tells the truth about how she is feeling or whether she is taking her medication and the staff suggest that she “lies to your face.” Layla has also mastered the art of the “silent treatment” and often does not answer questions, or, rather, answers with one or two syllable responses. Furthermore, Layla lacks initiative and independence in matters related to her health. Rather than contacting the clinic when she is feeling unwell, for example, she waits until she is very ill and relies on parents and staff, rather than herself, to intervene. Thus, Layla has often encountered largely preventable health deteriorations associated with the poor monitoring of CF. Layla’s poor adherence to her clinical treatment practices extend to multiple health behaviours and she is physically inactive and very underweight. Theoretically, from Layla’s inactivity and poor state of health, I could infer that it is important to prevent further losses to her CMC (Cott & Finch, 1995).

In addition to poor personal health, Layla has a complex social life and world. With little command of the English language, and very strict parenting sanctions, such as not allowing their children to attend social gatherings, school parties, or mixed gender activities, Layla’s relationship with her parents is characterized by conflict. Informed by family systems theory (Cummings, 2002), there are multiple problematic communicative pathways in the family system itself, but, most particularly, within the _child and parent sub-system._ While Layla’s parents did not participate in the intervention, I could infer that there were also problems in the _parent-parent subsystem,_ characterized by a dominating father and largely passive and uninvolved maternal figure. In addition to her illness, Layla appeared to be somewhat trapped in a
complicated cultural clash between two opposing world views, and her parents strict sanctions around social activities saddened and angered her. Indeed, I was quite taken aback one evening when Layla’s father entered our discussion room at the end of a session. Her body language and facial expression changed dramatically in his presence; she appeared visibly upset and immediately lapsed into silence. Furthermore, given that several of her siblings are very young, she has few points for identification with them. Layla has good relationships with two older sisters and two same aged girl friends at school. Thus, Layla’s social network is small, and, interestingly, she does not derive social support from adults.

Furthermore, Layla spends between five and six hours doing homework in the evening and often goes to sleep between 2:00 and 3:00 am. However, her ability to concentrate at school for prolonged periods of time is severely compromised and her academic performance is mediocre at best. Indeed, I was quite astounded by the sight of Layla’s weekly schedule. With little time for play or relaxation, her scholastic efforts do not allow her to attain desired academic outcomes. I felt particularly dismayed, when, on top of this grueling homework schedule, Layla grudgingly admitted that CF often leaves her stricken with fatigue and bed ridden at home. Additionally, Layla’s scholastic aptitude for the future and career related ambitions are poorly conceptualized. Although she stated that she wants a “job and a home one day,” she is worried that CF will thwart her attainment of these goals and she lacks passion and interest. Layla unenthusiastically stated throughout the program that she “may apply to college.” Although I repeatedly encouraged her to complete the necessary applications and tried to foster broader discussions about the benefits of education for women, I am not convinced that Layla will ever attend college.

*CF Chatters Program Response:* Despite her general negative world view and resistance toward talking to adults about her health, Layla tolerated the program well and derived benefits. According to the pre and post-test HAES data, Layla demonstrated significant improvements in
percentage daily physical activity (7.9%), total hours of daily activity (1.89 hours, and percentage daily activity in the most active category (3.3%), on a typical, average, or regular weekday—over the course of the intervention. Furthermore, although it required multiple phone calls home, as well as a great deal of verbal encouragement, Layla attended all of her appointments in the program, attained all of her weekly physical activity goals, mastered the program content, and demonstrated the ability to continue with the program after getting sick. Furthermore, when Layla learned to trust me—and understood that I was interested in why—rather than whether—she was doing treatment and physical activity, she eloquently and honestly articulated her complex health dilemmas and demonstrated insight.

Since Layla was completely inactive prior to the program, she set physical activity goals that would facilitate reduced sedentary time, improved participation in activities of daily living as well as mild to moderate physical activities. For example, Layla challenged herself to walk to school and to play basketball after school everyday, an activity that she enjoys. Layla only slightly increased the duration and intensity of her goals over the course of the program, and, rather, her goal setting was reflective of the desire to avoid inactivity and to maintain some level of activity. While Layla is given to falsehoods, I repeatedly emphasized throughout the program that she was in charge and that she did not have to shape her responses in order to suit what she thought I wanted to hear. I explained that before increased activity was possible, it may be important to understand why she has difficulty engaging in physical activity. Receiving consistent reinforcement and encouragement to set realistic physical activity goals that were meaningful for her, I think, reduced Layla’s propensity to lie and acquiesce to researchers. In terms of self-regulatory skills and processes, psycho-education and CBT were most effective for Layla. She appeared to be deeply engrossed and engaged by discussions that addressed the potential benefits of taking better care of her health.
More specifically, for Layla, the program resulted in increases in physical activity, quality of life, and learning about the self. For example, although her physical activity increased only slightly, Layla described a slight shift in thinking. When she is “lying around at home,” she is more likely to think about whether she should be active:

My activity, it did change. Now, whenever I am sitting around and doing nothing, I am starting to think that “maybe I could go outside and run around or play basketball or soccer.” It gets you active and it also helps your body … Yes, now I sort of try to make time for activity. To do something, even if it just a little, during the day.

Similarly, Layla’s subjective definition of quality of life appeared to have cognitive components. By fostering a greater understanding of what self care means and why it is important, she explained that the program allowed her to contemplate what she needs to do to live a better life, and slightly increased her quality of life:

Um, my quality of life feels much better. I do not really feel lazy any more and you feel healthy. Well, the program has made me think more about myself, and to think about what I need to do to stay healthy and to have a good life.

Finally, Layla reported increases in her knowledge and understanding as a result of participation in the program. Specifically, her awareness of the important role of physical activity for CF increased as well as her knowledge about herself:

I learned that—you have to stay on schedule and stuff. It is not good to leave things until they get worse. It is good to stay on top or things … I learned that exercise is really important for a lot of different things. Like physically and mentally too … yes, like the questions in the book make you think about things and the kind of person that you are. Layla also found the program to be easy and convenient and suggested that there were no major limitations in the program content, method, or delivery.
Summary: Layla is an intelligent but profoundly sad girl who rejects health and begs researchers and clinicians to further study the experiences of those who do not adhere to treatment. Characterized by a prolonged sense of sadness and resignation regarding her CF, Layla does not always “see the point of taking care of herself” if the end result of CF is always death. I further elaborate on “difficult patients” like Layla in the discussion section of this paper. Layla made significant cognitive and behavioural gains in the area of physical activity, quality of life, and self knowledge that exceeded those of the other patients in the program. For example, using my Grounded Theory, Layla’s *knowledge and awareness* about the importance of physical activity for CF dramatically increased. While physical activity has not been incorporated into Layla’s broader value system, program participation resulted in her thinking and reflecting about her *values* and whether taking care of her health and physical activity could become a part of her value system. Layla also discussed strategies and tactics that would allow her to minimize the *burden of disease and treatment*, such as engaging in household chores or other ambulatory activities - rather than lying in bed - when she is feeling unwell. While Layla’s *disease outlook and perspective* remained negative, program participation allowed her to dialogue and discuss her sense of futility, powerlessness, and hopelessness. While Layla did not change her outlook toward her illness during the program, she was receptive and listened attentively to how and why a change in attitude could help her. Furthermore, according to insights from the social ecological model (Elder et al., 2007), it did appear that Layla has multiple, hostile physical activity environments that are not conducive to increasing physical activity. Her immediate family micro-system, for example, was characterized by a lack of support and encouragement for physical activity. Furthermore, more distal macro-systems, such as the neighbourhood and the community, were largely inaccessible to her.

I believe that Layla’s important gains in the program were temporary and attributed to the high level of intervention assistance and emotional support. Unless Layla acquires a strong social
support network and learns to cognitively and behaviourally re-appraise CF such that she adheres to treatment, physical activity, and nutrition, she will continue to be a high risk patient who suffers tremendously with CF. By way of a telephone assessment, two weeks after the termination of the program, Layla is no longer physically active and it is difficult to have extended verbal conversations with her on the phone or in clinic. Layla is a patient who I continue to worry about.

*Emily: Opening Doors Between Reality and Fantasy*

*Context and Background:* Emily is an inquisitive, precocious, and imaginative eleven-year old child with CF who was experiencing a pulmonary exacerbation at the time of the study. Her FEV1 is 71%. Although Emily was diagnosed and treated for a co-morbid anxiety disorder during her last hospital admission, her anxiety appears to have abated. During the program, she regularly discussed the various strategies that she has employed in order to overcome her anxiety. Although Emily is very small, slender, and almost whimsical and fairy like in appearance - and appears to be far younger than eleven years of age - she is an animated and happy child who frequently laughs.

Emily is from a two-parent middle class family that resides in a comfortable suburban home about 1.5 hours west of the city. Emily’s father is a consultant and her mother is a stay at home parent who assumed the role of Emily’s primary caregiver ten years ago. Although her mother did not participate in the program, she is highly educated about CF and has played a critical role in educating her daughter about the disease.

*Emily’s Story:* Despite having undergone eight previous hospitalizations – and having worse lung function than the other youth in the study - Emily makes an effort to care for and attend to her physical health. Engaging in treatment, physical activity, and eating well-with a particular emphasis on trying to gain weight-is of critical importance. From a developmental perspective, Emily has impressive knowledge about CF and its implications. For example, she
can explain what CF is and articulate the difference between asthma airways and CF airways. Furthermore, she knows that children with CF may be hospitalized, require transplants, or even die—but that whether these events happen, depends upon the individual case. Despite the harsh reality of CF, however, treatment and self-care occupy an unquestioned place in her life. She has a strong pro-health orientation that is largely cultivated and assisted by her parents.

Emily enjoys a rich and imaginative life world. When she talks, she easily and seamlessly traverses between reality and fantasy, and her fantasy life and magical world continues to enrich her daily routine existence. She is an avid reader with a well developed vocabulary that far surpasses her eleven years. Much like Zoe, one of the other participants in the study, I was incredibly impressed by the breadth of reading that Emily has done and her profound ability to imagine new places and realities. Furthermore, Emily displayed impressive insight not only about medical health, but also her psychological and social worlds. For example, she described the detrimental impact of being criticized by others and how she has learned “not to care about those people” and to say “Whatever! Who cares”! She also frequently expressed that things which exert pressure on her, such as competition or high expectations, can be particularly psychologically damaging. Like Chase, she is socially conscious and aware, and displays empathy, for example, for the children of Haiti or the earth that “people are destroying.”

Although Emily is still recovering from her year long struggle with CF related anxiety—and her sense of self remains fragile but developing - she is engaged in a range of childhood activities which she enjoys. For example, Emily participates in Girl Guides as well as ballet dancing, and, during the course of the study, geared her physical activity goals such that she could better prepare for a dance recital. Emily also enjoys a pleasant school and social life and explained that she “enjoys her life because she is doing the things that she wants to do.” Although she frequently has to miss school due to CF-related appointments or sick days, her scholastic performance is good and she is interested and engaged in the lessons. At the age of 11
years, she has definitive career interests and expressed her desire to become a school teacher in the future. Emily stated that she has a great deal of social support, and the CF Chatters program underscored the importance of Emily’s family and friends to her life. Thus, although Emily is a medically fragile and ill child who has had past psychological struggles, she is deeply committed to her treatment and is living as well as she can with CF. Theoretically, both of Emily’s parent-child subsystems appear to function optimally, and, although she clings excessively to her mother during periods of hospitalization, as would be expected of someone her age, she does appear to have very healthy relationships with both of her parents. Furthermore, the fun and exciting interests that Emily pursues, such as dance, figure skating, and travel, are indicative of micro and macro systems that appear to facilitate optimal health and physical activity in spite of CF (Cummings, 2002).

Erik is Emily’s father and the parent who participated in the program. Although he is an intelligent man, he is generally reserved and quiet and engaged in discussion only when prompted. At 35 years of age, Erik is satisfied both personally and professionally in his life. He is not seeking career advancements, and, rather, explained that his primary future goal is to see “Emily and her brother achieve their ambitions.” Like his wife, Carroll, Erik is completely devoted toward caregiving for his daughter and the best possible management of CF. He is very educated about the disease and its treatment and is generally interested in CF research—or finding out new things that can help Emily. Theoretically, like Emily, Erik’s child and parent subsystems as well as spouse-spouse sub-system, all appear to function optimally, and the range of activities and interests that are available to him in the environment to pursue physical activity are indicative of contexts that are health and physical activity supportive (Cummings, 2002).

Interestingly, both Erik and Emily display a shared disdain for spontaneity and lack of structure. Recounting negative past experiences, they explained how not “being in control of their time” contributed toward a worsening of CF symptoms. Erik and Emily explained that
without schedule and routine, life with CF “feels chaotic and crazy.” As such, Erik, Emily, and their family have a very structured and routine existence characterized by doing the exact same thing at the exact same time, a pattern that was somehow incongruous with Emily’s rich and timeless fantasy life. Erik and Emily did not express the desire to live more spontaneous lives. Rather, they enjoy being in control of their time and it appears to facilitate better management of CF and to increase perceptions of control and autonomy.

Finally, both Erik and Emily displayed the capacity to easily dialogue about CF and talking about the disease is central to family conversations. They generally have realistic perceptions of the disease, neither over emphasizing its negative aspects by incorporating illness as master status (Bury, 1991) - nor denying the role of CF in their lives. For example, in the narrative below, Emily articulated the difference between a diagnosis of CF at birth, or, rather, in childhood. In doing so, she illustrated the difference between congenital and acquired disabilities and the ease with which she can speak about her illness:

Yah, because if you find out when you are older-like in school- or three or something. Anything over being a toddler. You have not grown up with CF, so you do not really know how to deal with it. It is harder for the parents then too. “We just found out about this-what do we do”? You have not had it since you were a baby. Now, if someone asks me why I get out of breath, I can say, “yes, it is because I have CF”. Whereas, it could be like “I don’t know! I just found out about it!!” Or, it could be like “what is everyone thinking?” What do I do! Rather than if you found out about when you are little, you do not worry about that stuff. You grow up with CF and do not worry about that stuff.

**CF chatters program child and parent response:** According to the pre and post-test HAES data, Emily demonstrated significant improvements in *percentage daily physical activity, total hours of daily activity, and percentage daily activity in the most active category, for an average, typical, or regular day* - over the course of the intervention. Emily demonstrated an
18.5% increase in daily physical activity, corresponding to 4.44 more hours of daily physical activity and 24.7% more activity in the most active activity category.

According to the qualitative data, while both Emily and Erik enjoyed the program, they derived different benefits. Although Emily’s HAES data indicate that her activity increased, in her qualitative report, she explained that she often feels tired and that she is already doing the maximum amount of physical activity that she can … *I think that my activity stayed the same, because, I was already doing the highest level of physical activity that I could. Maybe, I can do a different kind of dance, but I think that I would be tired. I do not think I could do more.*

For this reason, her goals centred upon maintaining or slightly increasing her physical activity, as well as attending and completing her daily physical activities. For example, during the program, Emily set physical activity goals such as attending 100% of her dance classes each week, rather than skipping class. Emily also challenged herself to participate completely in all of her physical education classes.

While Emily’s quality of life remained unchanged throughout the program, she reported increases in knowledge about the self. For example, she credited the program for providing her with more information and knowledge, both about the importance of physical activity for physical and mental health, and about herself:

*I always knew this, but I learned that physical activity helps your psychological - and your thinking and stuff. And, I learned that I never knew that it could help that much, but it can ... I kind of - when I was doing the “wheel of life” (workbook activity) ... I noticed a lot more, of how big the family part is, and how small other parts are. Because, I really had to think about that, to think about it.*

Erik also reported clear increases in physical activity behaviours. According to the HAES data, he demonstrated a 21.6% increase in daily physical activity, corresponding to 5.71 more hours of daily physical activity, and 14.2% more activity in the most active activity category, on a typical,
average, or regular weekday. Erik’s physical activity goals were narrow and specific and he
displayed no interest in experimenting with novel activities. Specifically, Erik was frustrated
with himself for resuming an inactive and sedentary lifestyle after Emily’s most recent
hospitalization and he expressed a strong desire to regularly attend the gym as he used to. Erik
used the program as an opportunity to resume his gym-based physical activity routine. Each
week, he challenged himself to increase the frequency of his gym attendance and the intensity of
his physical activity sessions. Erik progressed from no physical activity to three, 45-60 minute
physical activity sessions, involving running and weight training per week. Specifically, the
program allowed him to “get back on the physical activity bandwagon,” and equipped him with
the behavioural skills required to overcome his physical activity barriers:

It definitely increased. It (program) got me back on track. For a few months there, I had
not been going to the gym. Writing down those goals at the end of the first session, where
I said that I would go six times. I wrote it down and I had to do it. I only went five times,
but I did do some stuff at home too. So my activity definitely increased and it got me
back on track to where my PA should be. From a personal standpoint, it was great.

Furthermore, at the end of the program, Erik was pleased that he had become more active and
was noticing the transference of physical activity behaviour change to other areas of his life, such
as nutrition.

Erik reported no change in quality of life or learning about the self. However, he
appreciated that the program provided him with the opportunity to have discussions in advance
about CF relapses and how they influence his-and his daughter’s-physical activity. In particular,
the chapter on *relapse prevention* allowed Erik to think about Emily’s potential hospitalizations
in the future and devise strategies in order to better deal with these situations. Rather than a
difficult discussion, he saw this as necessary “advance planning:”
I think that it was important to talk about, so, again, you can plan it out ahead of time. Get used to thinking about it, ahead of time. “Like, Emily is going into the hospital. What can I do? We can walk up the stairs, right. We can walk around the hospital or outside.”

Both Emily and Erik reported that the program was easy and convenient and had no program criticism. As useful feedback, Emily suggested that it was important for future CF Chatters participants to be able to talk about CF without fear and to feel comfortable talking to strangers. She thus offered critical information with respect to participant readiness for the program:

I think that it depends on how much they want to talk about CF. And maybe, if we are talking about all of this physical activity, they are thinking “ohhh, I do not do all this and that must make me bad.” Or, when they do the survey (HAES), they think that “oh, I really do all that?” Or, it could be like “I am shy and I do not like talk to new people, like nurses and stuff.” It depends on how they feel. It could be scary.

Summary: Emily is a precocious and imaginative child with CF who displayed improvements in disease and self knowledge as a result of participation in the program. Despite her frail medical status and her slowly recovering self esteem, she is living as well as she can within the context of CF and has a good quality of life. She has well developed coping strategies and a great deal of familial support. Erik is a highly educated caregiver who is completely devoted toward his daughter’s health. Although his daughter’s illness is stressful, he is not overwhelmed by his caregiving tasks and is in control of CF. He derived notable benefits in the area of physical activity behaviour change as well as planning for relapse, from participating in the program. With respect to self regulatory skills, this highly structured father-daughter dyad - who both enjoy structuring time - were, not surprisingly, most successful at skills such as planning for physical activity.

In terms of my Grounded Theory, Erik and Emily were aware of the importance of physical activity for CF; physical activity was already a component of their broader value
system. Emily and Erik increased their ability to deal with the burden of disease and its treatment, by for example, developing a realistic relapse prevention plan for times of illness and hospitalization. While Erik was already a good health and physical activity role model for Emily, the program reinforced knowledge about the importance of physical activity role modeling and parental support. Emily displayed slight improvements in her disease outlook and perspective and there were notable reductions in her anxiety. By continuing to learn about CF and that “not everything bad will happen to me,” Emily enhanced her resilience, confidence, and sense of hope and displayed a more robust and less helpless response to CF.

Furthermore, according to the MCT, both Emily and Erik demonstrated improvement. In her qualitative interview, Emily stated that she was happy to maintain her current level of physical activity and was worried that more physical activity would make her feel too tired, indicating that her CMC and PMC were the same. In spite of this, there was a discrepancy between her interview and self report questionnaire. While the reasons for this discrepancy are not entirely known, Emily’s stable and unchanging state of health throughout the CF Chatters program may have allowed her to unknowingly undertake more activity than usual without setbacks or relapses. Emily demonstrated progress toward achieving her MAMP and perhaps demonstrated a shift in what is preferred. Similarly, Erik was dissatisfied with his CMC at the start of the program. Increasing his physical activity throughout the program reduced the gap between his CMC and PMC, and, at the end of the program, he was pleased with his level of activity (Cott & Finch, 1996).

Using a phone assessment, two weeks after the termination of the program, Emily and Erik are still active.

Zoe: “I Like Being Different - It Lets Me Do Things and Think Things That Other Kids Can’t:”

Background and context: Zoe is a quirky, expressive, and precocious twelve year old with CF and her FEV1 is 87%. Zoe enjoys a rich and imaginative life world and often spends
several minutes lost in thought. Zoe is very artistic and enjoys acting, singing, and writing. Although Zoe has good pulmonary function and diligently adheres to her treatment protocol like Chase and Emily, she is not inclined toward physical activity and runs the risk of continuing to be a sedentary adolescent with CF. Theoretically, despite her relatively good pulmonary function, Zoe’s CMC and PMC are low and the same, due to a general lack of awareness about the benefits of physical activity (Cott & Finch, 1995). Zoe’s relatively good health means that she has not had to encounter difficult experiences, such as hospitalization or intravenous treatments. Unlike Emily and Layla, she also has the external appearance of health and maintains a healthy weight. Zoe is from a middle class suburban family that resides one hour north of the city. Zoe and her brother, who also has CF, both live with their mother and father. Zoe’s parents are professionals; her mother is a consultant and her father is in the field of computers.

*Zoe's story:* Zoe loves her family and described them as “anchors” who support her. However, her school social relationships are complex and she has frequently been the victim of peer bullying. These negative experiences at school, such as being considered as a freak because of CF, have left her feeling ostracized and excluded and contributed toward low self esteem:

> In my mind, I thought that people thought that I was a freak. I really think that they think that. So, my self esteem was really low when I was littler. And it has built up more. But, my self esteem is still not where I wish that it was.

In spite of the bullying that she has endured, Zoe is starting to understand the advantages of avoiding social mores and conventions. She articulated the benefits of being different and avoiding the futile struggle to fit in:

> Sometimes, when you are different, you are willing to accept things that other kids cannot accept and to do things that other kids cannot do. Unlike other kids who follow the crowd to be accepted. Some kids do not want to be accepted and want to stand out.
It is clear that Zoe’s relationship to social conventions is complex and contradictory. She expressed the desire to fit in with her friends and be popular, while, at the same time, underscoring the desire to be different. I found myself wondering about Zoe’s future developmental trajectory at high school and how she will resolve the “sameness - difference” dilemma. Although Zoe’s social world is more complex and difficult than Chase’s and Emily’s - and she is constantly avoiding the “mean kids -” she too enjoys a range of social and extra-curricular activities that make for an enjoyable childhood. For example, Zoe is actively engaged in school plays and musicals and also sings in a church choir. Although Zoe does not perform as well in school as Chase or Emily, she too has well defined career pursuits. Also an avid reader, she wants to become an author of children’s fiction. Theoretically, while both parent-child sub-systems and sibling-sibling subsystems appear to function optimally for Zoe, her more immediate mesosystem at school does not appear to facilitate optimal health. Although Zoe does not always access them, the more distal environments in her neighbourhood and community are health and physical activity supportive (Cummings, 2002; Elder et al., 2007).

Gretchen’s story: Gretchen is Zoe’s mother and the primary caregiver who participated in the program. Gretchen is re-married; her and her second husband are raising two children with CF. Gretchen is an eloquent, soft spoken, sophisticated, principled, and intelligent woman who is very much, an old soul. While she cannot reach the imaginative and fantasy heights that her daughter can, she has immense appreciation for it. Theoretically, Gretchen appears to have optimally functioning child-parent sub-systems with both CF children. From what I could infer, Gretchen and her husband greatly differ in their interests and values, with Gretchen assuming the lion’s share of the caregiving burden. Thus, there could be some strife in the partner-partner subsystem. Furthermore, as a middle class family that resides north of the GTA, Gretchen and her family have immediate micro and mesosystems that are health and physical activity supportive. Although Gretchen and her family rarely use these environments to recreate in, as I
learned throughout the program, trail systems, parks, and rivers comprise their immediate environment (Cummings, 2002; Elder et al., 2007).

At the age of 53, Gretchen is the quintessential overworked mother who puts in long hours at the office but is also completely devoted to her children. While she is not in a state of ill health, Gretchen is overweight and does not have time to make physical activity a priority. Theoretically, Gretchen’s CMC is low. She is aware that she is unhealthy and states that if she had the time, she would want to be more active, and work toward her PMC (Cott & Finch, 1995). Furthermore, Gretchen is a strict disciplinarian and pragmatist who is very concerned about ensuring her children’s proper conduct and moral development. Furthermore, Gretchen and her husband both know that although they have two healthy kids with CF, that their grasp on health is tenuous. They are concerned that their children participate in as many activities as possible while they are still relatively healthy with CF:

Once we found out that as long as they are well, that we can treat them as anyone else. I mean it has caused us to do things in our lives at a different point than we would have otherwise. For example, we went to Disney World when Zoe was seven and Rick was three. We said that “they are well now, and we do not know how long they will be well for, so let’s go now.” So there are things we have done in our lives, because we wanted to make sure that they had the opportunity to do those things when there were not health issues.

In particular, ensuring that they treat their CF children as normally as possible and equip them with the skills to engage in independent health management, is of great importance to Gretchen and her husband:

You cannot stop the kids from being exposed to germs. If you let them out the door, that will happen. They have to be able to deal with the world as it is, and to deal with their
association to the world. If you are constantly hiding them away from having that exposure—when you are not there to provide that support for them, they are going to be lost. They need to be independent.

*CF chatters program child and parent response:* According to the pre and post-test HAES data, both Gretchen and Zoe displayed decreases in percentage daily physical activity and total hours of daily activity over the course of the intervention. Gretchen demonstrated a 6.2% decrease in daily physical activity, corresponding to 1.48 fewer hours of daily physical activity and no change in percentage activity in the most active category, *on a typical, average, or regular weekday*. Zoe demonstrated a 7.6% decrease in daily physical activity, corresponding to 1.83 fewer hours of daily physical activity, and 1.1% less activity in the most active category *on a typical, average, or regular weekday*. The reasons as to why Gretchen and Zoe’s activity decreased over the course of the intervention, is not entirely known. However, as illustrated by their somewhat uncertain and ambivalent qualitative responses regarding whether or not the program resulted in increased physical activity, the HAES report and the qualitative findings are somewhat concordant.

Even though the program did not result in significant increases in physical activity, Gretchen and Zoe set important physical activity goals together. Since Gretchen and Zoe have both previously been inactive, their goals centred upon attaining some physical activity, avoiding inactive states, and staying committed to their goals. Furthermore, in order to motivate one another as well as spend important mother-daughter time, Gretchen and Zoe set physical activity goals together. For example, Gretchen and Zoe challenged themselves to attain goals such as running around the block for 15 minutes two times a week with each other, walking the dog, raking the leaves for a few hours, as well as doing a fitness video together at home. For this mother-daughter dyad who were relatively uneducated about the benefits of physical activity for CF, psycho-education and CBT, were, by far, the most important behaviour change processes.
Despite displaying difficulty with physical activity behaviour change, important precursors to behaviour change, such as knowledge, awareness, and commitment to being active, increased. Gretchen and Zoe realized that physical activity had not been a priority in the past and underscored the importance of changing their perspectives. Thus, for both mom and daughter, the program appeared to enhance knowledge, attitude, and perspective about the importance of physical activity:

It reinforced more what I know needs to happen. The fact that during the six weeks, we usually agreed to do two new activities together. And, each time, we usually only managed to do one of those things that we agreed to. That showed me that the attention to activity and the commitment to it, was not as high as it should have been. Again, it is easier to come up with the excuse that “we are busy”, but that is a part of life. But, the fact that we at least did one of the two, at least showed that there was a certain level of commitment. But, if we had done everything, that would have shown that there was total commitment (Gretchen).

Furthermore, both mother and daughter reported slight increases in quality of life in the physical domain as a result of program participation … *Mine went up a small bit, but not very much … Well, I felt that I was getting more physical activity. So, I felt that my life was getting better. My quality of life increased (Zoe).*

Gretchen’s quality of life response was similar to Zoe’s and she reported slight increases in the physical domain. Furthermore, Gretchen illustrated the cognitive components of quality of life. She explained how the program content and manual enhanced quality of life by facilitating a new attitude and perspective about the importance of health and physical activity:

I would say that there was a slight increase in quality of life. What I got more out of it, was the change in attitude and the knowledge that there was-that we needed to have a different perspective-that is what I will come away with.
Finally, the therapeutic benefit of the program to both mother and daughter in making “tough talks” possible was particularly striking especially for Gretchen. Gretchen continuously underscored the important role that the program played in helping her to dialogue about potentially distressing topics that would have been difficult to have with her daughter in the absence of an exercise counsellor:

Well, I think that it did give me a much better insight and perspective into Zoe. We do not have these conversations, about these kinds of issues and these areas of concerns, etc. As a result, I would never have had this kind of conversation with Zoe and not had the chance to get her input and her feedback on these kinds of issues. She has impressed me incredibly over these weeks, with her insight-into herself and into her disease, and into the world around her. How she fits in. She has been very articulate and able to express herself. I have been very, very impressed with her in this time period. As a parent, we tend not to have the time to have these conversations with our children. We tend to be focused on our day to day existence. We do not have the time to have an in-depth conversation and we do not have the time to spend time together, with somebody leading the discussion. It has been excellent from that perspective.

Similarly, Zoe discussed the benefits of the program in facilitating “tough talks” with her mother. She explained that she would not know how to go about discussing such topics, such as the psychological ramifications of the disease, with her mother in the absence of an exercise counsellor:

I found it easier to talk about all of this. If mom asked me this, I would not really know what to say. Or how to say it for that matter. I would not know what to say … It would be hard, because I would not be able to understand what she was asking. Until then, I would not know how to say it at all.
Both mother and daughter also emphasized the importance of psychology in the treatment of CF and enjoyed discussing the psychological and social ramifications associated with CF. The lack of emphasis on psychological well-being in CF was noted as a primary barrier to clinical care:

> We get familiar with the medical staff. We know the RP (Respiratory Therapist), we know the nutritionist, we know the nurses, and we know the doctors. But, we do not know the psychological side of things. That is because we only see psychology unless we ask for that. If you do not have a significant issue (mental health issue), then, you will just coast along, as though it is just a part of your life. This is a significant issue within our lives. Not just from the kids perspective, but from our perspective, as well.

Zoe and Gretchen found the program easy and convenient and reported no criticisms.

**Summary:** Zoe and Gretchen are a high functioning mother-daughter dyad who, unlike the other youth, have not had to deal with particular stressors such as inpatient hospitalizations or prolonged pulmonary exacerbation. Although Zoe continues to endure unpleasant bullying at school that has taken a toll on her self-esteem, she is an upbeat and talkative child who is interested and motivated to engage in a range of different activities. Zoe and Gretchen are grateful to be dealing with minor pulmonary difficulties, but understand their grasp on health to be tenuous. For Zoe and Gretchen, the program facilitated enhanced quality of life and an attitudinal shift toward better understanding the importance of physical activity in CF. Furthermore, the program was therapeutic as it made it possible to facilitate discussions about difficult topics, and, by facilitating these necessary dialogues between Gretchen and Zoe, there were improvements in this particular child-parent subsystem. In terms of my theoretical model, Zoe and Gretchen increased their *knowledge* about physical activity and seriously re-conceptualized their values to include physical activity as a priority. While CF is currently not a significant barrier, Gretchen and Zoe increased their ability to *identify their barriers*-particularly
psychological ones—and to develop strategies to deal with such barriers. The program reinforced the importance of *parental role modeling* of physical activity for Gretchen. The program facilitated slight shifts in Zoe’s *outlook and perspective toward her illness*. Since it allowed her to talk with her daughter about distressing topics in a controlled and sensitive clinical setting, the program reduced Gretchen’s *stress* related to her child’s health.

Theoretically, Zoe and Gretchen’s CMC decreased over the course of the program. Zoe and Gretchen’s low level of physical activity appeared to be unrelated to CF, and, rather, maybe attributed to a lack of awareness about the importance of physical activity, as well as interest and motivation. Zoe and Gretchen appeared to display the more general pattern of inactivity that is characteristic even of healthy children and adults in the broader Canadian population, in which lack of time, motivation, commitment, and interest are barriers to participation (Tappe, Duda, & Eharwald, 1999), and there appears to be an inexplicable disconnect or gap between their intentions and their behaviours. Regular physical activity is still not a habitual activity for their family, and, although they are a high functioning and relatively well family, I suspect that they will still require regular physical activity reminders and more information on the optimal dose of physical activity that is required for health benefits. Two weeks after the termination of the program, Zoe and Gretchen are still adhering to regular physical activity goals.

In summary, the case findings indicate that the program was well received by CF patients and parents and was associated with increased physical activity behaviour, quality of life in the physical domain, and enhanced knowledge about the self. Improved caregiver health, as well as facilitating “tough talks,” were also notable results. My Grounded Theory provided a useful rubric for understanding the findings. By contrasting and comparing the cases to reveal new insights and discussing the cases within the existing literature, an analysis of the findings is undertaken below. In particular, the case study analysis reveals that the 1) *burden of disease* and
2) *participant age* differentiate how CF patients and parents interpret physical activity and health and influence their response to the program.

### 6.6 Discussion

Overall, the CF Chatters program was effective in facilitating enhanced physical activity participation and quality of life among most of the participants. The *retention rate* in the study was 80% and a CF-related exacerbation was the cause of program drop out. This suggests that although most participants adhered to the intervention, illness, relapse, and hospitalization may compromise the participation of some CF youth and their parents in behavioural interventions. Moreover, all participants demonstrated a *100% attendance rate* and completed all of the workbook activities and the program content. This suggests that although regular phone calls were required in order to encourage participation, participants were seriously committed to the CF Chatters program. Participants described the program as easy, convenient, enjoyable, and relevant to their physical activity concerns. Furthermore, the staff members who facilitated the delivery of the intervention suggested that from a clinical perspective, the program was not burdensome or disruptive to clinic flow. For this reason, the intervention was feasible.

Broadly informed by behavioural self regulation theory (Rhodes et al., 2010), behavioural self regulation skills training appeared to be an effective way in which to translate participants’ intentions into behaviour change. While other factors, such as parental support or access to physical activity also appeared to influence participants ability to change their behaviours, in general, behavioural self regulation skills training was particularly useful in promoting physical activity among CF patients and participants. By far, goal setting and planning the specifics of a behavioural plan to be more active were the most successful and well used strategies employed by the participants. Participants appeared to enjoy creating the “goal setting and planning card” at the end of each session, in which they clearly stipulated the specifics of their goal and plan for the week. Participants reported that they referred to their goal card throughout the week and
returned to each, subsequent session with their goal card in hand. Additionally, the behavioural self regulation skill of contingency planning, in which participant’s stipulated alternative arrangements that they could do if their physical activity goal could not be realized, was also very useful. All participants tailored contingency planning to illness exacerbations and relapses, and appeared quite astounded that they could in fact create alternative plans for physical activity if illness compromised their original intentions. Thus, this skill was useful for both the development of contingency plans, but also for communicating to the participants that they are less helpless than they think during periods of illness. Finally, “behavioural costs and rewards,” in which participants stipulated the pros and cons of changing their behaviour, also appeared to a useful behavioural self regulation skill. In addition to facilitating behaviour change, it was useful for the participants to discuss the perceived advantages and disadvantages of changing their behavior. In this regard, drawing from a broader theory, behavioural self regulation skills training (Rhodes et al., 2010) appeared to have applicability and relevance to this population of sick children and facilitated behaviour change. Thus, in summary, the CF Chatters program was effective in facilitating changes in quality of life and physical activity behaviour and should be considered as feasible and acceptable for the participants.

Furthermore according to the HAES data and the qualitative exit interviews, the intervention was successful in increasing physical activity participation among the majority of the participants. In addition to participants’ increases in physical activity in the qualitative exit interview, the HAES data demonstrated increases in 1) percentage daily physical activity, 2) total hours of daily physical activity, as well as 3) percentage daily physical activity in the most active category. While avoiding sedentary time and increasing physical activity in small doses may be particularly important for populations affected by chronic illness and disability, moderate to vigorous activity (MVPA) appear to be the most important in order to derive physiological benefits. Indeed, Yankaskas, Marshall, Sufian, Simon, and Rodman (2004) suggest that
“appropriate vigorous physical exercise enhances cardiovascular fitness, increases functional capacity, and improves quality of life (p. 2),” supporting the notion that a particular intensity of activity is necessary to garner benefits. The version of the HAES used in my study does not have a measure of vigorous or intense activity, which is an inherent limitation of the study tool. Thus, while percentage activity increased for the participants in the most active category, judgments cannot be made with respect to whether the intervention is associated with physiological benefits that may influence disease outcomes. Furthermore, the findings indicate that my Grounded Theory derived from Study One and Two is a relevant and important conceptual framework for understanding physical activity in youth with CF. For example, while the CF population appears to be well educated about the benefits of physical activity, prior to the program, not all participants were aware of the specific relationship between physical activity and the progression of CF over time. Furthermore, while participants were aware that staying active is important, Layla, Gretchen, and Zoe lacked knowledge about the optimal duration, intensity, and dose of physical activity that they should engage in to reap health benefits, as well as how physical activity influences overall health. Thus, with respect to my theoretical framework, the study appears to have facilitated enhanced knowledge about the importance of physical activity for patients with CF. Closely related to this, as the participants gained knowledge and awareness about the relationship between activity and the progression of CF, they displayed a slight shift in the value and importance that they ascribe toward physical activity. Enhanced value was most notable for Layla, Gretchen, and Zoe; while Layla was generally uninterested in engaging in any self-care activities prior to the program, Gretchen and Zoe were ignorant about the benefits of physical activity. As such, the program was associated with value shifts in which greater importance was ascribed toward living an active life with CF.

Self-efficacy was another theoretical construct from my Grounded Theory that was relevant to participants. While participants did not display notable increases in their physical
activity self-efficacy, the program made them more aware of how different factors, such as the broader social context and the type of activity, influence physical activity self-efficacy. For example, Chase engaged in an important discussion about the contextual nature of his physical activity self-efficacy. While he is plagued by pressing feelings of self-doubt during swimming, a physical activity setting that worsens his symptoms and makes “my lungs stop working,” he feels relatively confident when lifting weights at the gym. In this regard, the program allowed participants to view physical activity self-efficacy not as a static and unchanging construct, but, rather, as one that changes based on the context, the social situation, and the nature of the physical task (Bandura, 1997).

After discussing the intervention with Chase and Layla at the assessment phase (T1), they decided that they wanted to engage in the program independently of their parents. While Chase’s father stayed abreast of his son’s progress in the program, Layla’s parents remained uninvolved. This limits my ability to discuss parental support and role modeling as theoretical constructs within the context of my study. However, for the two participating child-parent dyads, there were clear increases in parents’ understanding of the importance of role modeling positive physical activity behaviours to their children. For Erik, who was already engaged in good parental role modeling, the program reinforced the importance of this construct to him. In contrast, Gretchen made significant strides in her ability to understand the significance of parental role modeling for Zoe’s physical activity and she demonstrated important cognitive shifts.

A lengthy discussion about the “burden of disease and its treatment and illness narratives,” is reported below as one of the central case differences that distinguished the participants from one another. Indeed, according to case study research, the burden of disease and illness narratives, were experiences that straddled the line and juncture between general experiences and the unique particularities of participants’ lives (Woods, 1992). However, all participants identified disease burden as a central physical activity barrier that detracted from
being able to participate. The program allowed the participants to identify exactly how CF symptoms compromise physical activity. Chase, Layla, and Gretchen, in particular, appeared to enjoy the discussion-based nature of the program and the opportunity to dialogue about their experiences with a supportive exercise counsellor in a sensitive setting. In this regard, it appears that the CF Chatters program may have attenuated participants’ level of stress. The program also enhanced their ability to devise strategies that would allow them to cope with illness and to remain as active as possible during times of relapse. Arguably, by enhancing CF youths’ ability to manage and deal with relapses and set backs, the program equipped participants with greater resilience and agency in terms of taking an active role in their health and physical activity, and facilitated an improved outlook toward CF. Finally, participants did not discuss their temporal constraints at length in the study. While they touched upon the time consuming nature of treatment and the life limiting nature of CF, they demonstrated a “just get through it” approach to physical therapy. However, the participants stated that the program was a worthwhile time investment and suggested that a longer program would have been more beneficial. This somewhat contradictory finding is incongruent with my theoretical tenant of negotiating physical activity within the context of reduced time. While “no time to play” requires further investigation, it may be that youth with CF and their parents are more selective, careful, thoughtful, and judicious about their time use. If this is the case, then CF Chatters met their more strict criteria for worthwhile time investments. In this regard, my Grounded Theory allowed me to devise a behavioural intervention that was relevant to participants’ lives. In addition to demonstrating the utility of my theory for understanding CF youths’ physical activity experiences, it offered a useful lens for the interpretation of the findings.

It appears that other theoretical frameworks that were identified in the developmental phase of the MRC framework, such as family systems theory, the social ecological model, as well as the movement continuum theory were relevant to the interpretation of the findings and
were useful theoretical frameworks that greatly informed my thinking during the delivery of the intervention. Specifically, in addition to reminding me to think of physical inactivity not as a individual issue, but, rather, as a familial one, family systems theory provided a way of characterizing how family members communicate with one another, whether particular family sub-systems are functioning optimally or not, and how to influence the broader families’ communication about physical activity (Cummings, 2002). The social ecological model sensitized me to the multiple environments that children and parent’s physical activity unfold in, providing me with an opportunity to characterize these environments as health and physical activity supportive, or, rather, hostile (Elder, 2007). For example, while Chase, Emily, and Zoe all had access to environments that would facilitate physical activity, Layla’s access to environments was heavily restricted and her social world was, subsequently, quite narrow. One work book activity, entitled “Understanding my Environment,” required the participants to both draw and discuss their broader environment and context and the physical activity opportunities available to them in their environments. While Chase, Zoe, and Emily’s environments were large and expansive, Layla’s visual environment – and the narrative that she shared about it – was strikingly narrow from a social ecological model perspective. Finally, the MCT was a very useful theoretical framework for generally being able to describe participants CMC, PMC, and MAMP and whether or not they were interested in closing gaps between these three constructs (Cott & Finch, 1995). In this regard, while I drew much more heavily upon my own Grounded Theory from Study One and Two in the design of my intervention, family systems theory, the social ecological model, and the movement continuum theory broadly informed my thinking during the intervention and allowed me to interpret the findings. Future researchers should consider these models as relevant and applicable to physical activity intervention research with chronically ill populations.
The Burden of Disease: In addition to the above discussion that outlines the relevance of my Grounded Theory to participants’ physical activity experiences, it is important to discuss and analyze the case studies in terms of their similarities and differences. Indeed, the burden of disease and its treatment and illness narratives, were the most important experiences that differentiated the participants. At the critical juncture between group and individual experiences (Woods, 1992), these case differences offer novel information about the CF population. Furthermore, age was a relevant case difference that differentiated the participants. The burden of disease and illness narratives are discussed below, with a particular focus on recommendations for future intervention research.

The burden of CF and treatment was the primary experience which differentiated the participants and revealed new insights about CF and physical activity. There were striking differences in how CF was experienced by the participants and their ability to cognitively reframe their experiences. Despite negative experiences and “hard knocks,” Chase, Emily, and Zoe displayed an enduring sense of hope, optimism, and resilience that dampened the influence of CF. In all cases, these youth had developed useful coping mechanisms that helped them to better manage CF, such as “just talking about it” or rather, taking better control of their time. Furthermore, Chase, Emily, and Zoe all pursued meaningful and fulfilling activities which equipped them with a sense of mastery and belonging - such as attending summer camp or acting lessons. Chase, Emily, and Zoe did not perceive CF in binary, “black or white” terms. The benefits of slowing the progression of the disease were evident and they wanted to attain the best possible health with CF. Although treatment was sometimes experienced as time consuming, and they wished they were doing other things, it occupied a central role in Chase’s, Emily’s and Zoe’s lives. Supported by their parents, adherence to treatment was non-negotiable and simply something that they had to do. Finally, although Chase, Emily, and Zoe started from different physical activity base lines, with Chase and Emily undertaking more activity than Zoe, engaging
in physical activity made sense to all three youth. Indeed, when the children learned that physical activity was important for CF, they were all interested in becoming more active through the program. Thus, Chase, Emily, and Zoe had both the psychological resources-cognitive appraisal of CF and coping mechanisms-as well as social support-activities and family-to equip them with a degree of optimism and resilience. This sense of optimism attenuated the burden of CF, making all three youth more interested in attaining a full life with CF that included physical activity.

In marked contrast, Layla’s case narrative illuminated the difference between children and youth who are living well with CF and those in which the burden of disease is significant. Layla’s experience with CF is one characterized by chronic pain, suffering, and turmoil and shows no signs of abating. Psychically, Layla conceptualizes CF in black and white terms-either sick or well. She does not see the point of treating a disease that is inherited and has no cure, despite evidence that treatment will dampen the trajectory and magnitude of disease progression. Chase, Emily, and Zoe adopt a more transitional way of thinking about CF in which there are periods of relative illness and relative wellness and they understand the benefits of living well with CF. On the other hand, Layla appears to have resigned herself to the fact that she will ultimately lose her battle with CF. The burden of disease for Layla is never relieved, and, while she identified new ways of coping during the study - such as phoning the doctor earlier when she was sick - up until then, she did not have a single coping mechanism in her arsenal against CF unlike the other youth. Rather, Layla demonstrated strong maladaptive coping, such as becoming silent or skipping treatment.

While it is not clear whether the appraisal of illness and the consequent burden of disease is a psychic or social process, Layla’s social environment is hostile and did little, if anything, to alleviate her burden of disease. Her experiences unfolded against the harsh backdrop of multiple and additive barriers - such as living in a multiple child, low income family in which there is no financial aid to enroll Layla in activities that she is interested in, parents who are not educated
about CF, and - more devastatingly - not a single source of social support from adults. Chase, Emily, and Zoe’s reduced burden of disease was facilitated by social support from parents who provided both tangible and emotional support, as well as finding meaningful activities outside of CF that provide vehicles for escapism and perceptions of mastery. The harsh socio-economic reality in Layla’s family meant that she has not had the opportunity to develop new interests or identify and pursue talents.

With extensive support, Layla demonstrated—however temporary—the ability to share her feelings and thoughts and to change her physical activity. She has the capacity to form supportive social relationships with trusting adults and to gain insight into why physical activity is beneficial for her health. However, unlike Chase, Emily, and Zoe who have the skills and resources to continue being active, Layla’s program response was temporary and she relapsed into inactivity after the program despite receiving monetary assistance to enroll in a physical activity of her choice.

Thus, Layla’s story illuminates the difference between patients who are living well with CF and those in which the burden of disease is overwhelming. While definitive statements cannot be made from case research (Stake, 2000), Layla’s story serves as a template for understanding those patients with CF who not only fail to adhere to physical activity, but also to a range of other health behaviours such as treatment or good nutrition. In many ways, Chase, Emily, and Zoe’s narratives, contrasted and compared against Layla’s, points to the bifurcation of the CF trajectory, or road; those that will continue to take the actions that will lead to living well with CF and those that will deteriorate. From this, we should consider physical activity not as a singular behaviour, but, rather, an activity that CF youth adhere to or not, within the broader context of how they interpret their overall health and illness. Patients like Layla, who feel a profound sense of futility and helplessness, are not likely to see the point of being active at all –
suggesting that until they receive psychological counselling, they are unlikely to derive long term benefits from a program like CF Chatters.

The striking case difference aligns well with my conceptual model derived from Study One and Two, in which the “burden of disease and treatment” and “illness narratives” were central to CF youths’ perceptions toward activity. Indeed, when youth feel unwell because of CF and report feelings of helplessness and despair, they are not likely to want to be physically active. Similar findings have been reported elsewhere. For example, by making children feel unwell, Swisher & Erikson (2008) found that the negative impact of disease symptoms were a primary deterrent to physical activity for youth with CF. Similarly, Moola et al. (2008) found that within the context of prevailing fatigue and a sense of social exclusion, that the point of being active was often not evident for youth with severe heart disease.

The case findings reported here on the burden of disease require researchers and clinicians to help patients to develop cognitive and behavioural coping strategies that will facilitate the cognitive re-appraisal of CF. Before sustained physical activity behaviour change is possible, it is necessary to change youths’ outlook toward their health such that being active is recognized as a benefit. More critically, the findings illuminate how supportive or hostile social environments and contexts, or the social determinants of health, either facilitate or impede health and physical activity and shape how patients interpret and appraise CF. Clinicians should help CF youth to develop more supportive social environments, including enlisting friends and family that are health and physical activity facilitating (Raphael, 2006).

In terms of the recommendations and implications for exercise counsellors from this case finding, unfortunately, it is unlikely that helpless and disengaged patients like Layla will derive long-term benefits from a program such as CF Chatters. Far more intensive individual and family counselling may be necessary in order to ensure that patients like Layla have the psychological and social resources necessary in order to derive sustainable benefits from the program. For this
reason, exercise counsellors may wish to initially screen participants for psycho-social health status prior to the intervention. Amotivated and helpless patients should not be engaged until they have received necessary counselling and assistance. This initial assessment of psycho-social readiness to participate may allow CF Chatters exercise counsellors to identify and refer such patients to counselling prior to readiness for the CF Chatters program again.

Before discussing the next case difference, it is important to note that case differences on the basis of disease burden taught me a great deal about resistance to treatment among difficult patients like Layla. By suspending my judgment, the CF Chatters program allowed me to better comprehend the complex worlds of difficult patients, and, I think, further a more complex understanding of non-adherence to treatment in CF. For example, gaining knowledge over time that CF is a fatal genetic disease has led Layla to develop a profound sense of helplessness and powerlessness, and, in her perspective, fully justifies “slacking off or skipping treatment.” The chronicity and incurability of CF- and its intractable nature - is particularly insidious for Layla, and, she frequently expressed that if CF had a cure, she would take the actions necessary to ensure her complete recovery. Layla conceptualizes CF in black and white terms; the benefits of “slowing the disease down” through treatment are not directly evident to her-if not ridiculous- and rather, she desires complete freedom from disease. Layla feels victimized, both by the CF that makes her “different, sickly, and abnormal,” as well as the various adults in her life-parents and doctors-who tell her to undertake treatments that she does not believe are in her best interest. Layla is not able to cognitively reappraise CF. Instead, she displays an illness narrative (Frank, 1995) that is characterized by sadness and resignation and the pain and loss that has characterized her childhood and adolescence shows no signs of abating:

After a while, treatment gets very irritating. It is like “I have to do this, for the rest of my life? Screw it! Who cares! I am not going to do it anymore.” You know that you have to do it all the time. You think of it and you say, “it is OK if I miss it once or twice. Because
I will have to do it for the rest of my life anyways” … if there was something that would get rid of CF, I would do it all the time! (laughing!). It is not like that. But the thing is with this CF - it is “like whatever, I have to do it for the rest of my life anyways” … when I started to grow up in grade eight and nine - when you start to learn more about CF. When I learned that it is actually inherited, you cannot get rid of it - there is no cure. That is when I started to slack off and to not care as much (Layla).

Furthermore, the program made me contemplate Layla’s treatment “secrets and lies -” not as an indication of being a bad patient - but, rather, as illustrative of those patients who are not provided with the opportunity to tell the truth, within a medical field that unquestionably subscribes to the inherent good of medical treatment. The clinic is frustrated by Layla’s inability to tell the truth, and, similarly, Layla feels guilty and bad when she lies to the doctor. As an embodied child actor with a self reflexive capacity (Mayall, 1998), Layla knows that her treatment dishonesty leaves both doctor and patient feeling lousy and is ultimately in no one’s best interest:

It does make you feel bad when you do tell the doctor that you do take the treatment. Sometimes, I will tell them that I have missed it once or twice. But, like, afterwards, you know how important it is and you know that they know too. When you do not do it, you feel bad.

By way of recommendations, arguably, clinicians and researchers need to facilitate a comfortable and non-judgmental dialogue that makes Layla’s “unsayable” illness narrative (Smith & Sparkes, 2008), an intelligent patient that does not see the point of treating a fatal, genetic illness, articulable. In this way, learning about “difficult patients” like Layla, and their treatment struggles, was an important caveat to explore that allowed me to more fully comprehend case differences according to disease burden.
Participant Age: Additionally, there were important case similarities and differences with respect to participant age. Although Emily and Zoe are at different stages of pubertal development, at 11 and 12 years, their biological age generally corresponds to the pre-adolescent or “tween” stage of cognitive development. In contrast, at the age of 16 and 17 years of age, Layla and Chase can be characterized as mid to late adolescents. According to Piaget’s stages of cognitive development, both Chase and Layla demonstrated the capacity for formal operational thought and abstract reasoning and cognition (Inhelder & Piaget, 1958). Thus, the four case units generally capture the beginning, middle, and end of the pre-adolescent to adolescent continuum.

The first age-related case difference was the ability of the younger participants to dialogue more openly and honestly about CF. While Chase and Layla eventually discussed their illnesses with me, in general, it was a more difficult and distressing conversation to have. Layla and Chase also did not elaborate upon the terminal nature of CF, and CF dialogues were a more sensitive and emotive topic for them. It was clear that Layla and Chase waited to ensure my trust before sharing their thoughts about CF. In contrast, Emily and Zoe discussed CF with relative comfort and ease. CF was central to how they understood themselves and they spoke of their illnesses in an unabashed manner. For Emily and Zoe, CF was more “self than other.” In contrast, although Chase had learned to accept CF and Layla vehemently resisted it, they both distanced CF from themselves and CF was more alien, more “other than self.”

The reasons as to why age appeared to influence CF youths’ ability to talk about CF are not entirely clear. However, literature and theory suggest that illness disrupts and fragments one’s sense of identity and their role, place, and purpose within the broader social world (Bury, 1991). Furthermore, illness often involves narrative reconstruction and significant identity repair work, in order to mend a fragmented identity in which former and present selves are called into question (Bury, 1991). Within this context, developmental psychology literature also underscores the significant struggle that the developing adolescent faces in the effort to secure a sense of
selfhood that is distinct from that of the parent. Indeed, the formation of a sense of self is the central task of adolescence, and, the developing adolescent wagers into broader questions regarding the self (Inhelder & Piaget, 1958). Thus, arguably, chronically ill adolescents, like Chase and Layla, face multiple barriers to identity work and their ability to reconcile whether CF is a part of the self or not, is arguably more complicated than for younger patients. The combined influence of adolescence as a temporal period as well as the experience of chronic illness, may account for why the younger and older case patients were positioned differently in their ability to discuss CF.

The younger case patients’ health and physical activity remained strongly under the influence of their parents decisions. For example, Emily and Zoe’s definitive pro-health orientation, and their adherence to treatment as well as physical activity, were an outgrowth and extension of their parents’ insistence on health. Emily and Zoe’s caregivers-Gretchen and Erik—communicated to their children that non adherence to treatment or lack of physical activity—was non-negotiable and their children articulated similar responses and behaviours. In contrast, Layla and Chase’s health and physical activity behaviours were not under as much parental influence. Although Chase’s parents facilitated physical activity, by, for example, paying for him to attend camp or driving him to sporting venues and facilities, his decision to be physically active and to adhere to treatment, was clearly his own; Chase has independent health and physical activity behaviours. Similarly, although Layla displayed a range of maladaptive behaviours, such as non adherence to treatment, nutrition, and physical activity, her health behaviours and decisions also appeared to be under less parental influence. Interestingly, when Layla and Chase were discussing parental involvement, they both chastised their parents for often communicating incorrect or erroneous information and explained that it was far easier to attend clinic without their parents. Thus, although Chase’s parents played an indirect facilitator role in his health and physical activity behaviours, and Layla’s parents appeared to have no role, parental involvement
for the older, adolescent case patients were less direct and involved. Their health behaviours and decisions were their own. In contrast, Emily and Zoe engaged in healthful behaviours largely because their parents exercised direct control and provided no other option.

The finding that physical activity and health behaviours are under less parental control as children age makes intuitive sense. As the developing child ages, the influence of the parent shrinks; the impact of the social, peer group grows (Krosnick & Judd, 1982) and parents come to occupy a facilitative, rather than directive role in their child’s treatment. Indeed, Krosnick and Judd explain the adolescents’ gradual shift away from parents by suggesting that “all theories of adolescence agree that this is the time when the child must cut and run, when he must disentangle himself from the family and become his own man – emotionally, behaviourally, in his values and controls” (1982, p. 359). As the developing child matures and separates from the parent to develop an independent identity, they are arguably better able to differentiate between their own values and those of their parents. This finding provides important information for exercise counsellors who are seeking to work with families. Physical activity interventions that are targeted at younger children should enlist the parent directly and parents should have an active role in overseeing their children’s behaviour change efforts alongside the child.

Interventions targeting older adolescents should involve the parent indirectly in a way which more distantly facilitates, supports, and encourages physical activity.

Case differences on the basis of participant age also offer important information for physical activity counsellors. Specifically, the age of the participant may change the delivery of the CF Chatters program. Younger patients between the ages of 10 and 12 years whose physical activity behaviours are still under the direct influence of their parents, should participate with a caregiver. This will ensure that the child has the emotional and tangible support from their parent to engage in physical activity. Based on the results from my study, the developing child is more likely to regard the presence of the parent as comforting and parental involvement is not likely to
threaten their sense of autonomy. In contrast, adolescents with CF are not likely to want to engage in the program with a parent and are in the process of developing their own physical activity values and beliefs. Based on the case findings, CF youth are critical of their parents’ understanding of the disease and often accuse them of communicating incorrect and erroneous information to health professionals. As well, their developing sense of autonomy is likely to be threatened by the presence of a parent, and, while developmental and chronological age are not always concordant, adolescents may be more comfortable engaging in the program with a familiar and trusted staff member. For adolescent patients with CF, it is advisable to indirectly engage the parent in a facilitator role, such as making the parent aware of how their child is progressing in the program as well as requesting that the parent drive their child to the appointment. Thus, parents should have a more background and peripheral role. Thus, the age of the participant will have a direct impact on how the intervention is delivered and the developing CF adolescents’ wish for autonomy and independence should be respected.

Finally, the beneficial impact of the CF Chatters program on caregiver physical activity and quality of life is a novel finding. In addition to supporting the inclusion of parents and caregivers into CF youths’ physical activity interventions, and extending the family based intervention research to the area of CF, this novel finding points to the need to better consider caregiver health among the parents of youth with CF. In addition to the benefits for their children, the two caregivers reported increases in physical activity, knowledge about the self, as well as quality of life. Indeed, it is well documented that the parents of youth with CF encounter high levels of stress related to their child’s illness and high caregiver burden (Eiser et al., 1995). Furthermore, no previous studies have described whether physical activity may reduce caregiver burden among CF parents. Indeed, caregiver literature from other fields, such as female caregivers of patients with dementia, have demonstrated that physical activity readies caregivers for the physical tasks associated with care provision and improves psychological wellbeing.
(Farren et al., 2008). The important physical and psychosocial benefits reported by the parents in this study not only supports the inclusion of parents in physical activity interventions, but points to the need to further research caregiver health in CF and whether physical activity can reduce caregiver burden (King & Brassington, 1997). The finding related to caregiver health among CF parents has relevance for future intervention development. Researchers and clinicians who are interested in caregiver health should consider physical activity as a modality to enhance the health and wellbeing of parents and caregivers who provide care to youth with CF. Such research and intervention work will make a novel and notable contribution toward our understanding of caregiver health and physical activity.

In summary, the CF Chatters program was perceived to be favourable and resulted in increased physical activity, quality of life, caregiver-health, and self-knowledge. The intervention was effective, feasible, and acceptable. Furthermore, the findings from the intervention make sense within the context of my Grounded Theory derived from Study One and Two, suggesting that the theory is a robust tool that requires further development and research. An analysis and discussion of the case studies revealed important differences in how the burden of disease and participant age influence health, physical activity, and the response of participants to the program. According to case research, disease burden and age illuminate the differences between general and particular experiences of physical activity in CF youth (Stake, 2000). In doing so, new insights about the similarities and differences between CF youth have been raised with particular recommendations for future intervention research. This extends and supports the existing literature and offers researchers important information with respect to health behaviour change in this population.

6.7 Limitations and Future Directions

The CF Chatters pilot intervention that was described in this chapter was an effective, feasible, acceptable, and enjoyable program that enhanced physical activity behaviour, quality of life, and
knowledge about physical activity and the self in a supportive setting that provided therapeutic benefits. More importantly, my theoretically guided behavioural counselling program makes sense, and is relevant to the social lives and dilemmas of the participants. While there were strengths associated with the approach that I adopted in my study in terms of describing participants’ response to the program, there were also limitations.

Specifically, with its intent focus on generating rich descriptions about participants’ health and physical activity stories (Stake, 2000), qualitative case research is particularly useful for generating provocative narratives. At the juncture between individual and generic experiences (Woods, 1992), new information was garnered in the multi-case study approach in this study. For example, unpleasant CF symptoms were consistently reported as a barrier toward physical activity for youth with CF and is well documented in the existing literature, suggesting that disease burden is a common experience to all youth with CF. In contrast, at the juncture and point between Layla’s narrative, the other three children, and the literature, “narratives of despair” appear to be unique to Layla in particular, drawing attention to a novel aspect of the CF experience that researchers may not have known previously and should explore.

However, while the multiple case study approach allowed me to generate new information about CF, the small sample size and reliance on self report data, impacts upon the generalizability of the findings, in the classic definition of the term that underpins most quantitative studies. Given that self report data may lack accuracy, this limits generalizability and confidence in whether the intervention changed physical activity. Therefore, in the classic sense of generalizability, the sample cannot be said to be representative of all youth with CF and the findings, particularly concerning any intervention effects, may not be statistically generalized from the sample to a population. However, in discussing the rigour of qualitative research, Peter Conrad (1990) proposes a new way of conceptualizing generalizability and suggests that we require different criteria and viewpoints in order to make sense of generalizability in qualitative
research. Conrad (1990) suggests that the generalizability of the concepts and ideas from qualitative research – to other settings and contexts – should comprise how generalizability is considered in qualitative research. Drawing on concepts such as “felt stigma” in epilepsy or “deviance disavowal” in polio, for instance, Conrad encourages us to consider not whether the findings apply to a broader population of epilepsy and polio sufferers, but rather, what these concepts and constructs help to illuminate and teach us about other illnesses in other settings. In this regard, generalizability of the concept requires researchers to think about whether the concepts that they have derived have relevance and value to other settings and contexts, helping to explain and understand the experiences of other participants in other settings. Although other researchers have coined terms such as case generalizability, transferability, or reader transferability to describe generalizability of the concept, all encourage the researcher to think about whether the concept has relevance to other pediatric chronic disease groups (Polit & Beck, 2010).

Since generalizability of the concept and transferability is a collaborative process, I have created in-depth, thick, and detailed descriptions for readers who will make judgments about whether inferences and extrapolations can be made from my findings to another group of participants, and the consumers of my research will have to decide whether my findings apply to their research settings as well. Transferability by the readers and consumers of my research – or generalizability of concept - will be enhanced by considering proximal similarity - or where the research setting and context in my study is located on a gradient of similarity in relation to other research settings and contexts. The findings from my study are most transferable to research settings that have the greatest similarity in terms of people, time, and context (Polit & Beck, 2010), and the consumers of my research will have to self reflexively consider whether the results make sense to new research contexts.
Guided by the MRC framework, the development of a CF Chatters feasibility trial is the next recommended step in the future. Such a feasibility trial should be characterized by a well powered sample, objective measures of physical activity, and the inclusion of a usual care condition that does not receive the CF Chatters intervention. A feasibility trail will allow researchers to further develop and stipulate the self regulatory skills needed to address theoretical constructs, develop their own outcome measures to measure novel and emerging constructs from the study, and tease out mediators of physical activity behaviour change. Generating such evidence will allow researchers to assess whether a definitive RCT should be conducted, comment upon the feasibility of a larger scale trial, distinguish whether changes in physical activity behaviour can be attributed to the intervention, and further the rationale for employing physical activity behavioural counselling programs with CF youth.

6. 8 Conclusion

Despite the notable benefits of physical activity and exercise for children and youth with CF, the literature is characterized by the absence of theoretically based interventions that utilize a counselling format which engages parents, as well as qualitative methods and psycho-social health indicators as methods and outcomes associated with intervention delivery. This study explored participants’ perceptions toward a novel, theoretically informed pilot program entitled CF Chatters, and the positive impact that it had on their quality of life, their understanding and knowledge about CF and the self, and their level of physical activity. In addition to demonstrating that my Grounded Theory is a robust tool that both guides intervention work and acts as a lens for interpretation, the results from my study are promising. The findings beg researchers and clinicians to consider similar intervention designs with the CF population that engage parents both directly and indirectly in the delivery. I hope that this is just the beginning of novel intervention formats that are tailored to the everyday lives and dilemmas of CF patients
and caregivers. In the final concluding chapter below, the findings from all three studies in this sequential Doctoral Program of Work are synthesized, analyzed, and concluded.
Chapter Seven: Conclusion

Reflections on the Dissertation Process

7.1 Summary of Doctoral Dissertation

Embedded within the MRC framework for complex behavioural intervention development (McEachan et al., 2008), the purpose of this dissertation was to 1) explore how physical activity is experienced in the lives of children with CF and CHD and their parents, 2) to develop a conceptual framework of physical activity in children with chronic diseases, 3) to develop and pilot test a theoretically informed behavioural intervention, and 4) to employ qualitative research to explore the impact of the program on participants quality of life and physical activity. More specifically, this study drew upon qualitative research traditions, such as Grounded Theory and thematic analysis (Braun & Clarke, 2006; Glaser & Strauss, 1967), in order to explore how youth living with CF and CHD and their parents experience and perceive physical activity, and their potential interest, if any, in behavioural interventions.

By thoroughly reading and immersing myself in the transcribed interview data from Study One and Two, a preliminary Grounded Theory of Physical Activity in CF and CHD was developed that illuminates the main theoretical constructs that are relevant to physical activity participation in these youth and caregivers. This critical developmental work within the MRC framework, equipped me with the knowledge and skills necessary in order to develop and pilot test a behavioural intervention. Guided by my Grounded Theory as well as other relevant theories from the literature - such as family systems theory, social ecological theory, and self regulation theory (Cummings, 2002; Elder, et al., 2007; Rhodes et al, 2010) - the feasibility and piloting phase of the MRC framework was also characterized by developing and pilot testing a behavioural intervention called “CF Chatters” with four case families. In addition to important findings from Study One and Two, as well as the developmental theoretical work, the results
from the pilot intervention suggest that “CF Chatters” was feasible, acceptable, interesting, enjoyable, and relevant to participants’ lives. Furthermore, for the majority of the participants, the intervention increased perceptions of quality of life, physical activity behaviour, and knowledge about the importance of physical activity for CF and “the self.” The intervention offered important therapeutic benefits to participants and facilitated “tough talks” that would have been difficult to have in the absence of an exercise counsellor. For a detailed summary of the results from the studies in this Dissertation, please see below.

**Table Ten: Doctoral Dissertation Summary of Results**

<table>
<thead>
<tr>
<th>Chapter</th>
<th>Primary Focus and Aims Addressed</th>
<th>Participants</th>
<th>Data Collection /Analysis</th>
<th>Key Findings</th>
</tr>
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<tbody>
<tr>
<td><em>Four</em></td>
<td>Perceptions of youth with CF toward physical activity</td>
<td>15 children</td>
<td>Interviews; thematic analysis</td>
<td>Youth with CF display positive or negative perceptions toward physical activity. <em>Parental support, fun, mastery experiences, and a sense of hope,</em> are experiences associated with positive perceptions. <em>Lack of parental support, disease burden, a sense of despair, and low value toward activity,</em> are associated with negative perceptions. Since youth with CF engage in time consuming treatments and understand themselves to be life limited, all youth negotiate physical activity within the context of reduced time.</td>
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<tr>
<td></td>
<td></td>
<td>and youth with CF</td>
<td></td>
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</tr>
<tr>
<td></td>
<td></td>
<td>28 parents of youth with CF and CHD (14 CF parents; 14 CHD parents)</td>
<td>Interviews; thematic analysis</td>
<td><em>Parents of youth with CF are more educated</em> and knowledgeable about the benefits of physical activity and were able to discern clear links between physical activity participation and improved disease outcomes. Parents were aware of the <em>benefits</em> of physical activity for both “child and self.” For example, physical activity provided parents with a distraction from the chronic stress of care giving. Despite the benefits, parents reported that there were many <em>barriers</em> associated with physical activity for both “child and self”. Disease burden and low self-efficacy were reported as barriers to PA for their children. Lack of time and a sense of guilt were parental physical activity barriers. Parents were aware of the importance of <em>role modeling</em> to facilitate physical activity. Parents’ physical activity should be understood within the broader family context of ongoing stress and complexity.</td>
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Directions for future research, the lessons learned from this dissertation, the limitations of this project, and a “self-reflexive story,” are now presented.

7.2 Major Contributions of This Work and Future Recommendations

This Doctoral Program of Work makes five major contributions to the field of Pediatrics, Psychology, Physical Activity, and Chronic Disease.

Theoretical contributions have been made from this Doctoral Dissertation. According to qualitative methodologists, theories are not the sole preserve of science. Within the qualitative tradition, a theory is defined as a set of inter-related ideas and concepts that facilitate a better understanding of particular phenomena. Theories also direct and inform the inquiry process in the social world or other naturalistic settings and can be developed in situ from the data, or rather, from pre-existing theories (Sandelowski, 1993). In this regard, theories sensitize researchers and clinicians to particular areas, and facilitate a better understanding of particular
problems or phenomena. Not unlike scientific theories, theories are both closed and bound, as well as open and dynamic to change and development over time. In particular, while substantive theories facilitate a better understanding of specific problems - for example, how CHD youth experience physical activity or how families respond to mental illness (Moola et al., 2008; Rose, Mallinson, & Walton – Moss, 2002) - formal theories are further developed and refined and have broader applicability to a wider range of phenomena, such as how chronically ill and disabled youth more broadly experience their health and physical activity (Carter & Little, 2007).

The development of a substantive Grounded Theory of physical activity in children with CF, CHD, and their parents is one of the most important outcomes associated with my investigation. Currently, the literature is atheoretical and the existing research is characterized by the absence of theories that pertain to childhood chronic diseases and physical activity. Interventions in CF and CHD are either atheoretical, or, rather, characterized by the application of “able bodied theories” that may not be appropriate for use with sick children - to these populations. The failure of exercise counsellors to employ a theoretically grounded approach may lead to the development of interventions that eclipse and overlook the critical physical activity experiences, perceptions, and needs of these populations, and may compromise the relevance of the intervention to participants’ lives.

Although the conceptual framework developed in this study requires further theoretical development, this task was beyond the scope of this Dissertation. However, my Grounded Theory of Physical Activity in Youth with CF and CHD and their Parents derived from Study One and Two, is a major theoretical contribution. It will provide future researchers with a framework in which to understand and interpret study findings, and a guiding rubric in which to develop tailored interventions that are more specific and relevant to participants’ needs, interests, and motivations. In particular, theoretical constructs derived in this study - such as “no time to play” or the negative influence of “despairing illness narratives” on physical activity - are novel,
disease specific theoretical constructs. These constructs may help exercise counsellors to understand why youth with CF are inactive and facilitate the development of strategies to overcome or re-conceptualize these perceived barriers.

Additionally, for qualitative researchers, the validity of theoretical frameworks is judged according to a different set of criterion than quantitative researchers. In particular, the following questions guide judgments about the validity of qualitative findings and theoretical frameworks (Sousa & Hendriks, 2006). 1) Does the concept in question adequately express and fit the pattern in the data that it purports to denote, and, in this regard, does the theory reflect and fit the data? Termed “validity,” theoretical fit ensures that the theory fits the data and is not forced, much like Cinderella’s step sister’s attempted to force - fit their feet into too-small slippers (Sandelowski, 1993). 2) Does the theory deal with what is truly important and relevant to those in the substantive area, and deal with the central concerns of the participants? 3) Through further development and evolution, can the theory be modified, such that the plausibility, credibility, and applicability of the theory increase? Finally, does the integrated and conceptually plausible grounded hypothesis properly account for how the main concerns of the participants are continually resolved, demonstrating workability? In this regard, according to the criterion set by qualitative methodologists, my Grounded Theory of physical activity in CF and CHD is valid, in the sense of offering a trustworthy, authentic, and believable account of the lives under study. Since the theory demonstrates fit, workability, modifiability, and relevance, trust can be placed in the theory (Sousa & Hendriks, 2006). In terms of future directions and recommendations, my theory should be further developed and modified. In order to generate a different kind of research evidence that is characteristic of quantitative forms of inquiry, however – such as the strength of the association between the theoretical constructs and the physical activity behaviour of youth - theoretical modeling and testing with a larger group of youth in diverse settings is required, and it should be subjected to experimental testing (Wilson, 1978).
The second contribution was the development of *CF Chatters*, a cognitive-behavioural parent mediated intervention that employed counselling to enhance physical activity and quality of life. CF Chatters proved to be a feasible and acceptable program that participants enjoyed and was relevant to their lives. The program was also successful in enhancing quality of life and physical activity behaviours. To my knowledge, this is the first physical activity behavioural counselling program for youth with CF and their parents that is grounded in their experiences and perceived physical activity needs. Since the pre-existing literature is characterized by the absence of behavioural self regulation skills programs to facilitate physical activity, this addressed a central limitation and gap in the existing literature. CF Chatters requires further development and testing in order to generate more evidence on its utility. However, it is my hope that I have provided researchers and clinicians with a basic, useful template in which to further develop and test the efficacy of a theoretically informed and contextually relevant behavioural counselling physical activity program for youth with CF that can, at a later date, be modified and adapted for other pediatric disease groups.

With respect to future directions for the CF Chatters program, it is necessary to discuss a few issues. The MRC framework allows researchers to make decisions about whether or not a randomized control trial should be conducted. Indeed, since RCT’s may be context stripped or not feasible, the MRC notes that there are disadvantages associated with RCT approaches and encourages researchers to use the early phases of the framework to make judgments about the potential for an RCT (Campbell, 2008). The participants in my study reported that the program was easy, enjoyable, and feasible and there are no major ethical issues or hazards associated with the program. For this reason, so long as a “wait list control group” approach is employed, thus ensuring that all participants receive the benefits of the program, it can tentatively be concluded that an RCT of the CF Chatters program can be safely conducted according to the MRC framework. Such an approach - including objective measures of physical activity, a
representative and well powered sample of CF youth, as well as a control condition that does not receive the CF Chatters program - is an important future direction that will generate the kind of research evidence that is needed to make institutional changes to clinical care within the current context of evidence - based medicine. For example, a definitive RCT will allow researchers to assess whether or not differences in physical activity and quality of life can be attributed to the CF Chatters intervention received by the experimental group, thereby furthering the rationale for the implementation of CF Chatters as a routine aspect of clinical care.

However, as a qualitative researcher who has personally witnessed the many health related struggles and dilemmas that these children and parents encounter, I am personally interested in investigating patients’ individual physical activity and illness stories, am keenly aware of the detrimental aspects associated with overly medicalizing sick children, and question whether physical activity should be the main outcome measure in intervention work with life limited children. I am interested in maximizing the quality of life of sick children - which may or may not be correlated with objective increases in physical activity – as well as the preservation of patient autonomy and dignity. Therefore, although I support the development of an RCT approach of the CF Chatters program for other researchers and would work as an interdisciplinary team member, I would prefer not to be involved in the measurement and assessment aspects of such a program given that it compromises some of my deeply held personal beliefs about children with chronic diseases.

It is evident that the epistemic differences between the quantitative and qualitative paradigms – and the type of very different research evidence that each produces – bear out in a program such as CF Chatters. This is a debate that I am conscious of and will continue to dialogue about in the future. Since an RCT of the CF Chatters program may fundamentally change the participant centred and exploratory nature of the intervention, there are clear disadvantages. However, in the current context that values evidence based medicine, it is not
likely that the CF Chatters program will be funded and implemented without evidence from an RCT.

Thirdly, although parents of youth with CF and CHD were not the direct focus of this Doctoral Program of work, this work has identified and drawn attention to a neglected group of caregivers, who, as a result of their child’s illness, experience significant personal, social, emotional, and financial strains and losses (Woodgate & Degner, 2007). My findings suggest that, as a result of the stress associated with caregiving for a chronically ill child, parents are more likely to experience anxiety and regularly question whether their child is “normal or not.” Parents also draw upon different “narratives of etiology” in order to explain their child’s condition to others and engage in “boundary play” in order to gauge and assess how ill their child is in relation to others (Moola, 2011, in review – See Appendix Five). As illustrated in Study Two, although parents enjoy physical activity and see it as a necessary reprieve from caregiving, they have difficulty understanding the concept of self care and regularly experience a pressing sense of guilt when engaged in physical activity. For this reason, findings related to poor caregiver health are a novel contribution from this study, and the utility of physical activity as a modality to enhance caregiver health among these parents is evident. Thus far, the benefits of physical activity for caregivers are mainly isolated to literature on elderly caregivers (King, & Brassington, 1997). In terms of future recommendations, the results from this study are potentially useful for caregiver and physical activity scholars who may wish to examine the stress encountered by pediatric caregivers and the utility of physical activity as a method in which to attenuate caregiver distress.

The focus upon children living with both CF and CHD and their caregivers was another notable contribution from this program of work. Most previous studies have worked with and engaged only one illness population. While this has notable benefits in terms of understanding the specific concerns of one group of ill children, the cross-childhood illness comparison
conducted in this study was particularly useful to my understanding of physical activity among children with chronic diseases more broadly. For example, the findings indicate that while CF parents are more educated about the benefits of physical activity, CHD parents cannot discern links between disease outcomes and physical activity participation. Similarly, although youth with CF are not concerned with “issues of risk” during activity like CHD patients are, their temporal constraints and negotiations are unique and have not been documented in other pediatric disease groups. In this regard, my work with both CF and CHD youth may make a notable contribution toward understanding the general and disease specific barriers toward physical activity among different youth with chronic diseases. In terms of future recommendations, a more generic formal theory of physical activity in children with chronic diseases more broadly - such Cystic Fibrosis, Congenital Heart Disease, Juvenile Idiopathic Arthritis (JIA), and Acute Lymphoblastic Leukemia (ALL) - should be developed which broadly encompasses their overlapping physical activity experiences. For example, despite notable differences, it has been found that youth with CF, CHD, and ALL report poor physical activity self-efficacy (Culos-Reed et al., 2002; Moola et al., 2008), suggesting that this construct may be incorporated into a generic theory of physical activity for ill children. In contrast, “no time to play,” “risk during physical activity,” as well as severe body image concerns resulting from chemotherapy, appear to characterize the different, disease specific experiences of youth with CF, CHD, and ALL respectively. Thus, future research should entail developing a broad and general formal theory of physical activity in childhood chronic diseases, which can then be tailored to specific disease groups.

Quantitative studies continue to dominate research in the area of physical activity for children with chronic diseases. Where qualitative research has been employed to understand health and illness interpretations, the physical activity experiences of chronically ill youth have remained tangential; the qualitative paradigm has been used instead to understand patients’
health and illness experiences. The use of the qualitative paradigm - including Grounded Theory, thematic analysis, and case study traditions - to understand physical activity, was a major, notable contribution of this work (Holloway, 2005; Mason, 2002). Indeed, it illuminated some of the individual and contextual issues and barriers, such as lack of parental support or illness narratives that prevent researchers from being able to translate and implement their evidence.

The qualitative inquiry undertaken in this study illuminated hidden or poorly understood areas in the experience of physical activity for CF and CHD youth. In terms of future directions and recommendations, the qualitative paradigm should feature as a central intervention component, both as an adjunctive method to quantitative inquiry, as well as a valuable forum for investigation in itself. Drawing upon the qualitative paradigm will ensure that interventions are tailored to participants’ local environments, needs, interests, desires, and motivations and continue to bring novel and poorly understood health and physical activity experiences to researcher’s attention (Mason, 2002). Arguably, mixed method designs that use objective measures while still engaging with the unique particularities of participants’ lives, offer an ideal approach in which to provide evidence informed, but contextually relevant, behavioural interventions.

Finally, the use of the iterative, stage-like MRC process to develop the CF Chatters program was another strength and contribution from this program of work. Where it is recognized that non pharmacological trials, such as developing physical activity interventions for chronically ill youth, are characterized by much complexity and many variables and components, it is challenging to design robust interventions that will have long term sustainable benefits. Furthermore, in suggesting that “… complex interventions may work best if they are tailored to local contexts rather than completely standardized,” Craig (2008) emphasizes that encapsulating the needs, interests, and contexts of participants may be an inherent benefit of this approach. Arguably, the preliminary developmental and piloting work conducted in this Program of Work
will ensure the development of future interventions that are evidenced informed, theoretically based, tested in many settings, and relevant to participants lives. The MRC states that:

“best practice is to develop interventions systematically, using the best available evidence and appropriate theory - and then to test them using a carefully phased approach that starts with a series of pilot studies targeted at each of the key uncertainties in the design, and moving onto an exploratory and then definitive evaluation” (Craig, 2008).

In adhering to these recommendations, arguably, the CF Chatters program has furthered best practice in complex intervention development. In summary, this Doctoral Program of Work has made major contributions and inroads into our understanding of physical activity in youth with CF and CHD, including, most importantly, a novel theory and behavioural intervention.

7.3 Lessons Learned Along the Way

There were important lessons learned and garnered from this Doctoral Program of Work, that provide researchers and clinicians with important information for future intervention development. Social isolation and the desire to participate in physical activity with other CF youth is, by far, participants’ most commonly identified physical activity need. Indeed, youth with CF are eager to engage in physical activity with others that are “just like me.” Given that CF is a very rare disease that affects 1/2500 youth, they explained that group-based physical activity with other CF youth would not only facilitate enjoyable and fun participation, but also equip them with an opportunity to tell and share their illness narratives and stories so that they can better strategize with others about how to manage CF. As recently as six years ago, youth with CF were permitted to engage in physical activities together, such as CF camp, and other scholars have emphasized the importance of reducing social isolation in CF (Blau et al., 2002; MacDonald & Greggans, 2010).

However, given the risk of cross contamination, clinicians do not recommend that youth with CF engage in social interaction with one another (personal communication, CF clinic
physiotherapist, SickKids, 2009). This uncontrollable medical issue compromises the ability of
the exercise counsellor to create physical activity programs that will address youths’ desire for
social support and detracts from one of the most important benefits of physical activity for
people with chronic diseases. This is not an issue that other populations, such as youth with CHD
or ALL, have to contend with and adds a layer of complexity for the exercise counsellor that is
seeking to develop programs. Indeed, I was quite taken aback when two, adult participants in the
parents study stated that they were happy to have two CF children, rather than one! They
explained that having multiple CF children circumvented the total loneliness and isolation
characteristic of families with only one CF child. Exercise counsellors should use “e-health” and
other online resources to connect youth with CF together in physical activity interventions, as the
only other feasible option (Ball & Lillis, 2010). However, despite the potential to exploit exciting
and novel internet resources such as facebook or skype, all youth with CF stated that meeting in
person was far preferable to online chatting. This was an important lesson learned from this
Dissertation and has illuminated a potential bioethical issue, in which the health benefits of
restricting patients from interacting with another, appear to have significant social costs.

As noted in the review of the literature in Chapter Two, children with chronic diseases
report reduced quality of life. Additionally, although quality of life is the sum total of physical,
social, emotional, psychological, and spiritual health, quality of life deficits are most often
reported in the physical domain. In my study, the majority of the participants reported improved
quality of life in the physical domain after participation in the CF Chatters program, suggesting
that the results from my study accord with previous literature. This important research issue once
again illuminates that poor physical health is particularly burdensome for patients with chronic
diseases, exerting the most significant impact on quality of life. The potential for physical
activity to improve participants’ physical functioning and well-being is evident. The finding that
physical activity appears to offer benefits in the physical component of quality of life for patients
with chronic diseases, should continue to further the rationale for continued physical activity promotion in this population.

As illustrated by Layla’s rather despairing and hopeless illness narrative, this study also allowed me to further study “recalcitrant and resistant” patients who are difficult to engage in research studies. Although there are semantic differences in the literature, such as “pre-contemplative, low motivation, or medical futility,” such resistant patients have been described (Lopez, Yager, & Feinstein, 2010). Although resistance to health and treatment has been described in populations such as people with mental health conditions, generally, less is known about treatment resistance in CF. While Layla acquired information and knowledge through the CF Chatters program and improved her physical activity and quality of life, in general, she demonstrated a sense of profound hopelessness and futility; she often questioned the point of being active and displayed disregard for other such self care activities, such as healthy eating. In addition to her negative and sad appraisal of CF, Layla was characterized by other experiences such as lack of parental support, poor career aspirations, lack of independence, and social deprivation and marginalization. Layla is a patient who does not adhere to “healthiest” assumptions about the inherent goodness and value of health, and requires health professionals to better consider those patients that reject health and treatment entirely. Thus, Layla was instructive not only in demonstrating why it is difficult to engage poorly motivated parents in research studies, but, more importantly, illustrating how such patients subscribe to different notions and values of health. This is an important lesson learned and will be instructive for CF researchers and clinicians who are attempting to engage difficult patients in treatment. It is likely that intensive social and psychological support is necessary to change underlying conceptualizations toward health before readiness to participate in interventions is possible.

Although it is unrelated to physical activity, “difficult transitions” was another issue and lesson learned from this Dissertation. Across numerous studies at SickKids, “difficulty with
transition” emerged as a salient theme time and time again. Youth with CF and CHD in my studies foresee that they will have great difficulty with the process of “transitioning” from child to adult clinical and treatment health care settings. Numerous barriers, such as parental overprotection, child fear, poor communication between child and adult physicians, and differences between child and adult “treatment cultures,” make it difficult for these patients to adequately transition (Kirk, 2008). The transitional difficulties that chronically ill children experience when moving to adult centres, is well documented in the literature (Kirk, 2008). However, demonstrating the transitional concerns of youth with CF and CHD - and how their transitional process occurs within the context of fears about the future, limited life time, and significant discrepancies between chronological and developmental age - were novel contributions from this thesis. A manuscript entitled “Down the rabbit hole”: Enhancing the transition process for young patients with cystic fibrosis and congenital heart disease by re-imagining the future and time” will be published with a co-author in the journal Child: Care, Health, and Development – in press.

Finally, the cumulative impact of the time consuming nature of daily physical therapy treatments as well as the knowledge that CF is a life-limiting condition, contribute toward participants’ belief that they are “running out of time.” An important finding from this study was that youth with CF and their parents negotiate physical activity within the context of perceived temporal constraints. They are more judicious and careful with their time use and only incorporate interests and activities that are thought to be worthwhile time investments, eliminating extraneous or unnecessary temporal activities. As a group, they are more structured and less spontaneous, and, although they are “masters of their time,” my intervention appeared to change participants’ beliefs about the temporal value of physical activity. All of the participants found CF Chatters to be a worthwhile time investment.
Like space, time is a broad concept that has been debated by philosophers and scientists alike for centuries, and, like religion or culture, it is a meta-construct that has generated much debate (Heidegger, 1992). For example, while scientists tend to be concerned with the objective quantification of time, other scholars urge us to consider the cyclical and iterative nature of time, begging us to consider *Kairos* rather than *Chronos*; while the latter refers to “passing time,” the former refers to “meaningful time” (Sandelowski, 1993). My study adds to Charmaz’s (1993) ground breaking work on how illness and disability fundamentally changes our sense of time and how it passes. Although the “time and physical activity findings” were very useful to my thesis, the ways in which CF patients negotiate and embody time is an important research issue that deserves further inquiry and investigation.

Thus, insights about social isolation, difficult patients, the transitional concerns of youth with chronic diseases, time in CF, as well as the impact of physical activity on physical quality of life, were novel and important lessons learned in this Dissertation that may contribute more broadly to general pediatric literature.

7.4 Limitations

There were several limitations associated with this Doctoral Program of Work. These limitations are pragmatic as well as methodological and conceptual in nature. One of the central research objectives associated with this Dissertation was to work toward the development and implementation of a physical activity intervention for youth with CF and CHD. Since it would allow for a *cross-childhood illness comparison* to illustrate the physical activity and health similarities and differences between these two clinical groups, it was thought to be a novel contribution from this Dissertation. Patients with CHD participated in Study One, Study Two, and the Grounded Theory - Crystallization Phase of the Dissertation. However, in the final stages of this program of work, pragmatic and practical concerns prevented the implementation of a similar intervention for CHD youth. Factors such as an already overburdened CHD clinic, a lack
of nurse facilitation, and the effort to conduct a manageable and timely thesis, prevented me from being able to deliver a similar “CHD Chatters” intervention to youth with CHD and their parents. Furthermore, although it is dyadic and informational in nature, there is a similar physical activity intervention currently being undertaken in the CHD clinic. In this regard, although the CHD population contributed toward Study One, Study Two, and the Grounded Theory crystallization phase, they did not participate in the intervention itself. However, the insights gained from CHD parents and youth in the qualitative components of the study were very informative in terms of understanding how this population experiences physical activity as well as their perceived intervention interests. In the future, a disease-specific “CHD Chatters -” with both general and disease specific content - should be delivered and implemented with the CHD population too, as well as other clinical populations. This remains the focus of future interventions.

With the assistance of my Doctoral supervisor, I experimented with a wide range of qualitative research traditions, such as Grounded Theory, thematic analysis, as well as case study research. This decision was based on the desire to expand my methodological repertoire. Following Ussher (1999), I was eager to explore the potentially fruitful dialogue and theoretical and methodological insights that may result from the cross fertilization of a variety of different qualitative techniques. Seeking methodological pluralism and eclecticism\(^\text{13}\) (Ussher, 1999), I was eager to discover potentially thought provoking insights by addressing the research question from a variety of different methodological perspectives. As suggested by Ussher (1999), such a diverse methodological approach may produce a more rich and detailed account of the particular phenomena that is under question, in this case, how physical activity is experienced by

\(^{13}\) Although in the past methodological pluralism referred to a diverse and eclectic approach adopted between quantitative and qualitative research, the use of this term here refers to diversity and eclecticism within the qualitative paradigm itself. For example, the use of thematic analysis, Grounded Theory, and case study research within the same dissertation– all variants of qualitative research – may be considered as a methodologically pluralistic approach.
chronically ill youth and their parents. Furthermore, Carter and Little (2007) also support the notion of methodological pluralism, suggesting that so long as the researcher demonstrates congruence between epistemology-methodology and method, and that these decisions are communicated and justified, a multiple qualitative methodological approach may facilitate the production of unbound knowledge that “knows no barriers (2007).”

However, while I have certainly enhanced and diversified my ability to conceptualize and employ diverse qualitative methodologies - and this is a strength - it is important to note that there are epistemic tensions between them. Indeed, some qualitative researchers would suggest that the methodologies employed within each study are not entirely congruent with one another across the broader Dissertation (Denzin & Lincoln, 2003), or that they are incommensurable (Carter & Little, 2007). For example, Grounded Theory falls within the “social constructivist” epistemological “camp” suggesting that the production of knowledge is culturally and historically situated, as well as produced at the interface between the researcher and the participant. In this regard, it has relativistic and constructivist interpretations of reality. In contrast, thematic analysis is a foundational qualitative method. So long as qualitative researchers make their assumptions and epistemic positions evident, it is a flexible and diverse method that is compatible with both realist and constructivist paradigms (Braun & Clarke, 2006). Finally, while qualitative case study research requires further ontological and epistemic development, it is a “post positivist” methodology, and, in relation to Grounded Theory, it is inherently more conservative in its epistemological assumptions. For example, while case study researchers acknowledge the social construction of reality, empirical methods are still required for the derivation of knowledge and they are less likely to deconstruct notions of “true” knowledge. As such, there are ontological and epistemological differences in how qualitative case research, Grounded Theory, and thematic analysis conceptualize the production of
knowledge. Some qualitative methodologists may critique this approach and suggest that the methodologies are incommensurable (Carter & Little, 2007; Denzin & Lincoln, 2003).

In addition to selecting qualitative traditions that best suit the research question at hand (Ussher, 1999), as well as greatly enhancing my methodological aptitude, I have dealt with these methodological conundrums. In particular, following Carter and Little (2007), I have made my epistemic position within each of the three studies, transparent. More importantly, I have ensured that each study is internally consistent, demonstrating congruence and compatibility between epistemic insights, methodological decisions, and the selection of methods (Carter & Little, 2007). Following Carter and Little (2007), it is my hope that methodological pluralism encounters less resistance in the future, so that the “vicious cycles of contempt between opposing positions” – and parochialism - can be avoided.

There were also limitations with respect to the delivery of the intervention. For example, CF Chatters was designed as a “parent-mediated” physical activity counselling program for youth with CF as well as their caregivers. Indeed, the literature reports that the parents of chronically ill youth experience compromised physical and psycho-social health. By facilitating their children’s health behaviours, parents are gatekeepers to health behaviour change. For these reasons, the inclusion of parents in my intervention was well justified in the literature. However, once the participants were enrolled, the youth-adolescent participants (age 16 and 17) opted out of participating with their parent and expressed their desire to participate with the researcher independently. Thus, of the six participants, two parent-child dyads (four participants) engaged in CF Chatters together, and, the remaining two adolescents participated in the program independently. While this was an unexpected limitation associated with the study, the desire of the older, adolescent patients to engage in the program alone was very informative for me as a researcher and exercise counsellor and accords with theoretical insights about child and adolescent development, in which the adolescents’ burgeoning sense of autonomy leads them to
disengage from their parents. Thus, based on my findings, it appears that parents are more likely to be involved in the intervention if the participating patient is a child. In contrast, parents are more likely to adopt a distant and facilitative role if the participating patient is an adolescent. It is desirable to provide patients with the choice of participating with a parent, and adolescents’ desire for autonomy and reluctance to participate with their parents, must be respected.

Finally, qualitative research allows for the generation of rich “thick” descriptions about participants’ lives, allowing researchers to story experiences and produce provocative and interesting interpretive narratives (Stake, 2002). For marginalized populations - such as sick children - the inherently exploratory and participant driven nature of the qualitative inquiry may equip patients with a sense of participant centredness, agency, and control. However, based on quantitative criterion for generalizability – a well powered sample that is representative of the population under study - it is important to note that the samples selected for qualitative investigations and the subsequent findings, may not be generalized to larger groups of children with CF and CHD beyond the study population itself (Stake, 2002). However, as described in the previous chapter, when we adopt qualitative criterion, the findings may afford generalizability of the concept and transferability, in which important concepts and ideas from the study, such as low self efficacy or reduced time, can be transferred to other settings and contexts, helping to illuminate how people in other settings and situations resolve their dilemmas (Conrad, 1990). In this regard, concepts derived in my study may display case generalizability since they are relevant to other participants in other settings. Furthermore, the findings and descriptions produced in case study research allow us not only to better interpret and describe participants’ lives, worlds, and dilemmas, but may sensitize us to broader health and physical activity issues that the larger group of children with chronic diseases experience. As such, the qualitative paradigm employed in this study, has, arguably, helped us to learn more about physical activity and health among children with CF and CHD. To generate a different kind of “objective”
research evidence - studies powered by larger sample sizes as well as objective measures, are required. In summary, there were pragmatic as well as conceptual limitations associated with this study. Before concluding this Doctoral Dissertation, I will discuss the self reflexive insights that I acquired throughout the process.

7.5 Self-reflexivity and the “Penseive” (Gerstl-Pepin & Patrizio, 2009)

It is important to return, for a moment, to Professor Dumbledore’s penseive that was discussed in the introduction to this Dissertation - that is, a deep, stone made brewing cauldron of stored memories and stories that scholars (or wizards!) repeatedly return to in order to better understand themselves, and work toward the resolution of present problems. Although the boy Harry Potter remained skeptical and unconvinced about the virtues of the penseive, under Professor Dumbledore’s tutelage, he learned about the value of attending to past selves in order to interpret events today. In his subsequent childhood adventures, he was all the more wiser for it.

In Gerstl-Pepin and Patrizio’s (2009) article, they draw upon Dumbledore’s penseive or brewing cauldron, as an analogy to the concept of self-reflexivity, arguing that it is critical to qualitative inquiry. Indeed, a self-reflexive stance allows the qualitative researcher to understand that their investigations are never value-free and non-neutral. Rather, we bring past histories and selves to all of our research inquiries, and, we can see and locate ourselves in research. Thus, Professor Dumbledore’s brewing cauldron of stored memories - his penseive - reminds young investigators of the importance of re-remembering past events, how the self is always embedded in research, and the ways in which we are always changed through our research investigations. Below, my self reflexive musings and thoughts related to this Doctoral Program of Work - what brews in my penseive - are briefly discussed in “Lilly’s Story.”

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14 Professor Dumbledore is the “Master” at Hogwarts Academy of Witchcraft and Wizardry. Professor Dumbledore serves as mentor and teacher to the young protagonist, Harry Potter. It is thought that in the future, the Harry Potter books will garner much fame for illustrating the dilemmas of the post modern child. The “penseive” is used as a metaphor by Gerstl-Pepin and Patrizio (2009) to discuss the importance of research reflexivity.
I was introduced to Lilly at the beginning of Study Two of my Doctoral Dissertation. After two and a half years of being cared for as an inpatient in another, out of province children’s hospital, Lilly and her father arrived in Toronto. Described as their “new home,” Toronto’s SickKids was where Lilly and her father would begin the long and tedious process of waiting for double lung transplant surgery and their battle to get “new lungs on time.” Although Lilly was ambulatory when I met her, she was receiving oxygen therapy 24 hours a day. Despite her weakened state, Lilly was exuberant and excited. She enthusiastically dialogued about her upcoming physical therapy sessions that she would undertake with her father and expressed excitement over the possibility of receiving new lungs. That a patient could be as excited as Lilly over an upcoming transplant surgery made me marvel at the fortitude and resilience of the human spirit. Although she was several hours away from home and separated from her family, Lilly and her father embraced SickKids and described the sense of safety and security that had enveloped them since arriving at the “world’s best hospital.”

The severity of Lilly’s illness and her difficulty with English excluded her from participating in the children’s study. However, her English speaking father, Francis, participated in Study Two. Twelve year old Lilly and I took an immediate liking to each other and were “kindred spirits” (Montgomery, 1908). With an angelic face and large, hopeful eyes, I was truly amazed by Lilly’s resilience and excitement, and her ability to conceptualize her upcoming lung transplant, not as a setback or limitation, but, rather, as a new and fighting chance.

Lilly had not attended school for nearly two years. Since she was vulnerable to infections, she was isolated from her brothers and sisters. Francis explained that the debilitating effects of CF over the past two years had led to prolonged periods of bed rest and resulted in the development of a new set of non-active childhood activities and interests, such as drawing and painting. Furthermore, Francis took an extended leave from his job in order to “wait for lungs”
with his daughter in Toronto, and described how, in his mid thirties, he was largely isolated from his friends. Additionally, Lilly’s illness caused conflict between her biological mother and father, and had led to the ultimate termination of their marriage. Francis was Lilly’s primary caregiver.

In spite of these numerous and repeated stressors, the resilience and robustness of this father-daughter dyad astounded me, and, more importantly, the enduring sense of love and tenderness that Lilly garnered from everyone who interacted with her. Lilly remained everyone’s “sweetheart” and left an indelible and lingering impression on family members, friends, staff members, and even strangers. Lilly was more soul than body, and “wore” hope and optimism as though it was a magnificent garb. I joined in Lilly and Francis’ joy and found myself “wishing for lungs” right alongside this beautiful but ailing child.

*Lilly’s fall:* As Lilly’s illness rapidly progressed, unfortunately, the late spring and summer of 2010 was characterized by a rather rapid decent. Within a few weeks of arriving in Toronto to wait for lung transplantation, Lilly and Francis moved out from their Toronto apartment and were admitted to the hospital. Lilly never left the hospital again. A series of serious health-related complications left her in the inpatient unit for the entire summer.

When I arrived home from a trip in the fall, the clinical team notified me that Lilly had been admitted to the critical care unit (CCU). A nurse notified me that Lilly had been intubated and was attached to the ECMO machine (Extracorporeal transmembrane oxygenator) in order to provide life support. She kindly informed me that although it was “scary” to see Lilly, that it was my choice to visit her and that her family would very much appreciate my visits. Smith and Sparkes (2004) discuss the importance of narrating illness both for those with illnesses, and those that bear witness to their experiences. It was at this particular juncture in my construction of Lilly’s illness when my storying began to reflect elements of “narrative incoherence and the collapse of meaning” (Charon, 2006). My belief in Lilly’s ability to recover and attain health was called into question, and, from this point onward, the events in Lilly’s health and illness
story resembled elements of chaos (Frank, 1995). Characterized by rapid change, I no longer knew what was happening to Lilly, or how to explain these events to myself. The linearity of her story collapsed for me as her story teller.

I observed other changes in Lilly’s care that served as ominous signifiers of the events to come. Her biological mother and grandmother arrived in Toronto for an extended stay in order to better support Francis in his care giving duties. Additionally, I was somewhat confused by the ever growing discrepancy between Francis’s perspective toward his daughters health, and the response of the clinical team. Although the clinical team continued to describe the situation as “really bad,” Francis generally remained optimistic and hopeful, and simply stated that he and Lilly were “still waiting for lungs.” .... “Waiting for lungs”... indeed, Westman et al (2010) has described how prolonged waiting in a health care setting often contributes toward “existential dread” in which the integrity of the self is threatened (Westman et al., 2010). I wondered what it was like for Francis to wait for such a prolonged period of time, whether his patience was tested, and how time was passing.

_Bodies that freak (Garland - Thomson, 1997):_ Despite having worked for years at SickKids, I was shocked, stunned, and taken aback when I visited Lilly in her new unit, and my sensory system was affronted. Her physical appearance had dramatically altered from the bright and talkative girl I had met just a few short months ago, who, despite her frailty, was as exuberant and exited as any other “healthy” child. Although I regularly visited Lilly to stand by her bed or to hold her hand, she was heavily medicated and her tongue was too swollen to speak. Lilly’s hair had changed colour and she was so thin that she appeared to be only seven or eight years old. It was nearly impossible to communicate with Lilly. Given the heavy administration of medication, it was only possible to interact with her in non-verbal ways for ten minutes at most. During these brief interactions, Lilly communicated with me through her eyes only and made futile attempts to talk or to smile. I wondered if Lilly’s drab and cold hospital environment in the
CCU mimicked her internal state. A single photograph of Lilly’s smiling “former self” was attached to one of the CCU machines at the foot of her bed, and occasionally, Lilly’s eyes moved and rested on this picture. For me, this picture only served to magnify the ever growing discrepancy between Lilly Now and Lilly Yesterday.

Admittedly, I found the site of Lilly’s ECMO machine to be particularly horrific and I wondered whether the treatment was doing more harm than good. The large, invasive machine appeared to sit heavily upon Lilly’s emaciated body. While logically understanding that the machine was providing life supportive functions to Lilly, it appeared to be an almost violent procedure, contributing toward medicine looming large (Foucault, 1979) in Lilly’s life. I watched through tears as the machine repeatedly shook her body.

October and November were particularly difficult months for Lilly and her family, who were confronted by repeated health deteriorations and complications that prevented lung transplantation. At this time, I often found myself feeling very angry with Canada’s “opt in” approach to multi-organ transplant donation and began to consider this important health and social issue.

My final interaction with Lilly was very difficult. While the CCU nurses encouraged me to talk with her, Lilly’s tiny body appeared to reduce the content and depth of my speech to infantile interactions, and I found myself talking to an 12 year old girl as though she were only two. The difference between conscious and non-verbal, semi conscious patients was profound for me as a researcher. Although I intently read Lilly’s eyes through a sort of evolving “vision speech,” I found myself not able to discern whether Lilly was pining for life, asking to be let go, or, rather, some other choice that was clearly not evident to the adults attending to her. I often felt confused. Having worked in various capacities at the hospital for ten years, I knew that Lilly was gravely ill. And, yet, I fought against Lilly’s possible death. Instead, I found myself, like her dad, “hoping for lungs.”
Lilly was not even conscious when she turned 13 years old in the CCU. In the late fall, in addition to her failed lungs, Lilly entered cardiac failure. With the dilemma of both not being able to transport Lilly in such a severely compromised medical state, as well as the fact that SickKids does not perform heart-lung transplant surgeries, the clinical team explained to Francis that they could not-and would never be able to-do Lilly’s surgery. With this knowledge, Francis explained to Lilly that she would be joining her grandmother in heaven. In many ways, Francis’s communication to Lilly was a perfect, loving, and utterly intuitive explanation by a father to his daughter. However, following Bluebond Langener (1978), I found myself sitting with uncomfortable questions regarding the nature of what dying children know: did Lilly have all the information she needed? While still heavily medicated in order to allow for a painless passing, Lilly’s care was withdrawn and her life support machines turned off. In late fall, a nurse notified me that Lilly had passed away immediately after the withdrawal of the machine, indicating the failure of her respiratory system to do any work on its own.

There was a near palpable sense of despair that hung drearily over the CF clinic that week. The nurses sadly explained that many members of the clinical team felt that they had failed Lilly and her father who had unconditionally and without doubt placed so much confidence and hope in them. The clinical discourse shifted (Foucault, 1979) from medical to religious, and, instead of dialoguing about Lilly’s treatment plan, the clinicians reminisced about what an “amazing and inspiring” family they were, one they would never forget. Despite its brevity, Lilly and her father touched me profoundly and they are a father-daughter dyad whom I will never forget. Francis and I continue to stay in touch; Francis tells me that Lilly continues to give him courage every day.

Lilly and my dose of self-reflexivity: In my self reflexive pensieve, Lilly’s story-and my relationship to it-is relevant and pertinent and forms the bulk of the self reflexive insights that I learned throughout the course of this Doctoral Dissertation. It provides a forum in which to
understand the numerous ethical constraints and moral dilemmas that plague researchers and clinicians who work with this vulnerable population of ill youth.

The ethical dilemmas are particularly striking in Lilly’s case, begging researchers to consider broader questions such as what dying children know. However, ethical considerations of less gravity and magnitude still figured prominently in my interactions with many patients across the entire Doctoral Program of Work. For instance, it provoked me to ponder questions such as “what is the researcher to do when the child conveys “logical reasons” for why they do not adhere to treatment?” What is the role of the researcher when a participant’s health status rapidly deteriorates? What is the researcher to do when the participant discloses confidentially that they have received neglectful or improper health care? What is the researcher to do when social exclusion, pain, and fatigue clearly make physical activity more harmful than good? In more minor instances, what is the researcher to do when a young research participant is having difficulty differentiating between research and clinical care? What is the researcher to do when an intervention that is affording therapeutic benefit to the patient, is terminated because the study period ended? Does the researcher have an ongoing obligation to the patient, or not?

I dealt with all of these situations in a manner that accords with hospital-university ethics, moral framework that are based on liberal humanist notions of morality. However, none of the situations were easily rectified and all have left me with an ongoing and lingering sense of moral dis-ease and angst (Knifed, Goyal, & Bernstein, 2010). As a part of my researcher reflexivity, questions of ethics and morality were central to this investigation and directly implicated me in the ethical lives of my participants.

As witness, as observer, as story teller: More poignantly, I draw on the writing of Bluebond Langener (1978) to explain my interpretation of what it is like to conduct research with chronically ill children. Bluebond - Langener (1978) worked with children with cancer in the late 1970’s, at a time when nearly all of the participants in her ethnographic study died. I was
born in 1980, when she conducted her ground breaking and formative anthropological study on “what dying children know,” and, in this way, her work preceded mine by three decades. Three of the 50 children that I have worked with have died over the course of my Masters and Doctoral Dissertations. However, like Bluebond - Langener (1978), I fell to working with this population in my late teens and early twenties. Unlike Bluebond, I was hospitalized extensively as a child, and, thus, my research at SickKids is born out of more than chance (Bluebond-Langener, 1978). However, this investigation has exposed me to darker, more painful and sinister aspects of life that I would have remained ignorant of had I not undertaken this Dissertation. The private lives of chronically ill children (Bluebond-Langener, 1978) - and the harsh experiences they are often exposed to - are a facet of social life that I am both privileged and frightened of having observed. I am grateful for how quickly parents and children at SickKids have embraced me and the close nature of the interaction that characterizes the enduring bond between us. And, on most days, I am happy to be a part of their process, story, and rehabilitation, and the opportunity to be involved vicariously in their health related victories and triumphs.

However, at times, and at a young age, I am sometimes frightened by the intensity of what I have witnessed. More pressingly, while I am usually convinced that my work has bettered the health and physical activity of patients, when a child deteriorates or dies, the researcher is provoked to ask “did I do everything I could?” Did I do more harm than good?” Was there a point to this inquiry? Where are these children, now?” Who has benefited? What is the value of my work? “What does it mean to suffer?” Are those that do not suffer currently, obligated to intervene in the lives that do”? Is this work too hard? Should I research something else?” These are questions that I will think about always. At best, I know that in my work with sick children, I have tried to be a dedicated and honest witness. I would like to think that I am an inquisitive observer “on the side lines,” listening, observing, and storying—but not necessarily intervening upon-the events occurring around me. The researcher can never represent the Other’s voice, and
only ever offer up an always partial interpretation. However, through publications, conference activity, teaching, and more importantly—my daily banters and “rants” to friends and family who so graciously listen—I have done my best to interpret and communicate what the private health and physical activity lives of sick children are like, to a largely unknowing public.

In my self-reflexive penseive, I see that my experiences working at the Hospital for Sick Children are rife with contradiction. I feel inspired, hopeful, and touched by these children and grateful for having acquired a body of knowledge that I can share with others. At the same time, by exposing me to the harsh realities of childhood chronic diseases, my work is sometimes painful and frightening and exhausting. I understand why health professionals wear a “coat of armor” that conceals them from the emotional tumult of their work that they would encounter if they took off their “masks.” I can identify the discourses that clinicians often draw upon—medicalized language and jargon—to talk away deeply human issues such as the suffering or death of a wonderful child. Although it makes for more difficulty, however, I am happy and grateful that I feel each child’s story as intensely, richly, and closely as I do, and that each narrative reverberates.

And, at all times, this work is thought provoking and intense, begging me to consider broader existential questions that are larger than myself and my immediate environment, and I am more thoughtful as a result. In this regard, Lilly’s story illuminates just some of the self-reflexive insights that I have gathered in my penseive (Gerstl-Pepin & Patrizio, 2009), underscoring, once again, that the self is always embedded in, and transformed by, the research activities that we undertake.

7.6 Conclusion

This Doctoral Dissertation has allowed me to make major forays and inroads into our understanding of physical activity in two neglected populations of chronically ill youth. It has illuminated and revealed the numerous barriers that constrain CF and CHD youths’ physical
activity. Furthermore, it has facilitated the development of a conceptual framework in order to better understand CF and CHD youths’ health and physical activity experiences and laid the ground work for future behavioural interventions that will facilitate greater physical activity and quality of life in these populations. As adjunct and complimentary issues, this Dissertation has also drawn attention to novel health issues and dilemmas encountered by chronically ill children and their caregivers, such as “losing time,” “fear of transition,” “the difficult patient,” “the benefits of physical activity for caregivers” and the “social versus medical bioethical dilemma in CF.” All studies in this Doctoral Dissertation will contribute toward the academic and scholarly literature in diverse fields, such as psychology, adapted physical activity, pediatrics, sociology, childhood studies, disability studies, and philosophy, and may one day – hopefully - result in changes to clinical practice. At the juncture between childhood, psychology, physical activity, and illness, at best, I hope that my work will push these bodies of knowledge forward, and, more importantly, place the physical activity needs of a previously neglected group of chronically ill Canadian children on researchers’ and clinicians’ agendas.

It is with much excitement and triumph, utter disbelief, a sense of nostalgia and gratitude, and-admittedly-much fatigue, that I bring my Doctoral Dissertation Adventure to a bitter-sweet close. After three years, I have more questions than answers and a desire to listen more than I speak. In doing so, I draw attention to a recent article on how a young woman with anorexia nervosa negotiates physical activity. In their article, Zanker & Gard (2008) suggest that:

The idea that physical activity means different things to different people hardly needs to be reiterated. And yet, it is surely part of the ongoing role of scholarship to map and probe the evolving meanings of physical activity and peoples experiences of it … perhaps most important of all is the need to offer narratives that run counter to dominant ways of thinking about physical activity … narrow understandings can in turn, delegitimize the experiences of some people, sanction dangerous or unethical practices in physical

Working with chronically ill children is emotionally taxing—if not exhausting. Before one can inquire into this fascinating but highly coveted and policed clinical group of children, there are numerous institutional barriers to circumvent. However, the physical activity *counter narratives* that I had the privilege of listening to were fascinating, intriguing, and informative. Following Gard and Wright (2008), as physical activity scholars who seek to know how and why people move - or rather, why movement is alienating and unpleasant for some - it is critical to attend and to *listen to* (Smith & Sparkes, 2008) the physical activity experiences of sick children and youth, and bear witness to physical activity counter-narratives that run contrary to what we know and assume. In articulating that “*life is not all rainbows and butterflies,*” – the title of this Dissertation - my participants drew upon poignant metaphors and counter narratives to describe the weighty health and illness dilemmas that make their childhoods and physical activity pursuits particularly challenging and burdensome. For this reason, in addition to the physical activity stories that I have listened to, and the theory and intervention that I have advanced, it is my hope that this Dissertation calls upon and compels clinicians and researchers alike to attend to the physical activity needs of this neglected population of chronically ill youth. With so much to teach us - *about physical activity, health, and the human condition itself* - it is my hope that the stories of youth with CF and CHD are heard too.
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APPENDIX ONE

Children’s Interview Schedule

General
1) Can you explain how physically active you are at the moment? How many days of the week do you engage in physical activity and for how many minutes?

2) On a scale of one to ten, how hard would it be for you to become more physically active? Why?

3) In your opinion, what are the good things about becoming more active? What will becoming more active allow you to do? In your opinion, what are the bad things about becoming more active? (outcome expectations)

4) What would need to change in order for you to become more active in physical activity?

Facilitators
1) Can you explain the things that would help you to become more active in physical activity? Who would need to help you? What would they need to do?

2) If your school and community offered different types of physical activities and sports programs, would this help you to become more active? Why or why not?

3) What would the CF clinic need to do and say in order to help you to become more physically active?

4) If your parents were more active in physical activity and sport, would it help you to be more active? Why or why not?

Barriers
1) Can you explain the things which may stand in your way of becoming as active as you want to be?
2) Do you think that your parents ever worry about you when you do physical activity and sports? If so, why do you think that they worry? Does their worry impact you? How does it make you feel?

3) If physical activity was important for your friends, would it be important for you too? (social norms)

*Health and physical activity values*

1) How do you feel when you are doing physical activity? Is it fun for you or not?

2) Does physical activity make you feel better or worse about yourself?

3) Do you think that it is important to be involved in physical activity? Why or why not? If not, can you explain the activities that are more important than doing sports?

*Disease specific*

1) If you knew that physical activity could help your CF, would you want to do more than you are at the moment? Why or why not?

2) In your opinion, does your CF affect how active that you are able to be? If so, can you explain?

3) When you get a respiratory infection or have to come to the hospital, does it affect how active you want to be? If so, how?

4) What are the easy things and the hard things about becoming active again after you have been sick?

*Health*

1) What are your feelings about doing physical therapy for your CF? What are the good things about treatment and what are the bad things about treatment?

2) How do you feel about taking nutritional supplements for your CF? What are the good things about taking nutritional supplements and what are the bad things?
3) Do you think that CF could affect you in the future? How active do you want to be in the future?
APPENDIX Two

Parents Interview Schedule

Parental Physical Activity Attitudes and Beliefs

1) Do you think that physical activity and exercise is important for your child? Why or why not?

2) Do you think that physical activity is important for your child’s CHD/CF? Why or why not?

How did you obtain this information?

3) If you knew that physical activity was important for your child’s health, would you be interested in encouraging him/her to become more active?

4) Do you think that physical activity is important for your own health? Why or why not?

5) If you knew that physical activity was important for your own health, would you be interested in becoming more active?

6) What are your physical activity beliefs and attitudes? For example, do you think that it is dangerous and risky for your child to be active? Do you think that it is safe and normal for your child to be active?

7) Is physical activity and exercise a priority for you? Why or why not? Can you explain the things which are more or less important than physical activity?

Parental Physical Activity Behaviours

1) Can you describe how physically active your child is at the moment? For example, can you describe the amount and type of activities that your child participated in last week?

2) Can you describe how active your family generally is at the moment? For example, can you explain the amount and type of activity that you engaged in with your family last week?

3) Can you describe how active you are at the moment? For example, can you describe the amount and type of activity that you participated in last week?
4) Please reflect for a moment to the time when your child was first diagnosed with CHD/CF. Can you explain whether your family’s physical activity has changed as a result of your child’s illness? Can you explain if your activity has changed as a result of your child’s illness? How?

5) Do you have other children? How active are your other children? Do you think that your other children have an impact on your sick child’s physical activity? If so, how?

6) How “active” is your home? For example, can you explain if you talk about physical activity and sports in your home, do activities together, seek out and learn about new activities, and own sports equipment to play with?

*Parental Physical Activity Barriers*

1) In your opinion, are there things that make it difficult or challenging for your child to become more physically active? For example, does the illness itself affect how active your child can be? Do the types of activities offered in gym class at school affect how active your child can be? Do your child’s friends and peers impact how physically active they can be? Do the types of activities offered in your community affect how active your child can be?

2) Do you affect or influence your child’s physical activity? If so, how?

3) Has there ever been a time when you felt too worried, scared, or anxious to allow our child to be physically active? If so, can you explain?

4) Do you trust your child’s judgement and ability to know when they are tired during physical activity and sport? If so, can you explain?

5) Do you trust your child's judgment and ability to know when they are tired during physical activity and sport? Please explain

6) Are there any things which make it difficult or challenging for you to become more active? For example, does taking care of our child affect how active you can be? Does work or family life affect how active you can be?
Parental Physical Activity Facilitators

1) What would make it easier for your child to become more active? For example, what would your child require in order to become more active?

2) What would make it easier for your family to become more active? For example, what does your family require in order to become more active?

3) What would make it easier for you to become more active? For example, what do you need to do to become more active?

4) What needs to change in your life right now in order for your family to become more active? Are you interested in changing in order to become more active?

5) Do you help your child to be active? If so, please explain how you help your child to be active. Are there things which you could do differently in order to help your child to become more active?

Physical Activity Intervention Interest

1) Are you interested in becoming more physically active with your child? Would your family be able to participate in a program in order to help you to become more active? Please explain.

2) If a program was offered to help you and your family become more active, what would it include? For example, please describe an ideal physical activity program to help your family to become more active.

3) Do you think the staff at the Hospital can help you and your child to become more active? If so, what kind of help do you need from the hospital in order to help you and your child to become more active?

4) Would physical activity counselling, education, and goal setting help your family to become more active? Why or why not?

5) What would make it difficult to participate in a physical activity program with your family? For example, do you have enough time? Are you interested?
6) May we contact you at a later date for participation in a physical activity program?
APPENDIX Three

CF Chatters Interview Schedule

Program Elements

1) In your opinion, what were the good things and the bad things about the “CF Chatters” physical activity counselling program? What did you like about the program? What did you not like about the program?

2) What do you think about the “CF Chatters” manual? Was it useful for you, or not?

3) What was it like to have a personal physical activity counsellor? Was it helpful or not helpful to have a personal physical activity counsellor?

4) In comparison to your regular medical appointment with the doctors and nurses in the clinic, how did it feel to participate in the CF Chatter program? (did the CF Chatters program feel the same or different than other appointments in the CF clinic?)

5) Was the “CF Chatters” program offered on the best day and time for you? Was it convenient for your schedule or not?

6) How could the “CF Chatters” program be improved and enhanced?

7) Was it helpful or not helpful to receive a physical activity start up fee for the “CF Chatters” program?

8) Was it helpful or not helpful to receive weekly physical activity reminder calls for the “CF Chatters” program?

9) Was working with a parent in the “CF Chatters” program helpful or not helpful?

10) Did you use any of the information in your daily life?

Efficacy

1) What did you learn from participating in the “CF Chatters” program?
2) Did your knowledge about physical activity change or not change as a result of participating in the program?

3) Did the content of the “CF Chatters” program cover everything that you needed to know? (knowledge, planning, environment, barriers, illness)? If not, what should be added to the content in order to make it better?

3) Did your involvement in physical activity change or not change as a result of participating in the “CF Chatters” program?

4) Did your quality of life (wellness/happiness/how you feel about you) change or not change as a result of participating in the “CF Chatters” program?

5) Did your knowledge and understanding about CF change or not change as a result of participating in the “CF Chatters” program?

6) Did your ability to plan and schedule physical activity change or not change as a result of participating in the program?

7) Did your ability to deal with CF related relapses/illness barriers change or not change as a result of participating in the program?

8) Did the “CF Chatters” program help or not help with the management of CF?

9) Did the “CF Chatters” program make any difference to the quality of the care that you have received at Sickkids, or not?

*Future*

1) Do you think that you will continue or not continue to do physical activity in the future?

2) Do you think that the “CF Chatters” program should be delivered to other CF kids, or not?
THE HAES (HABITUAL ACTIVITY ESTIMATION SCALE)

This questionnaire will ask you questions about your daily activities. Please read all of the instructions carefully and answer each question as truthfully as you can.

Name: ____________________________________________

Date: ____________________________

INSTRUCTIONS (please read!)

Please recall the activities of one typical weekday (choose from Tuesday, Wednesday or Thursday) and one typical Saturday within the past 2 weeks. For each given time period, please estimate the percentage of time that you spent in each of 4 different activity levels. For each of the time periods, the total time spent in all activity levels must add up to 100%.

The different activity levels are described below:

<table>
<thead>
<tr>
<th>ACTIVITY LEVEL DESCRIPTIONS</th>
</tr>
</thead>
<tbody>
<tr>
<td>These descriptions give you examples of activities that are typical of each activity level. You should refer back to these descriptions as often as you need when completing your estimates.</td>
</tr>
<tr>
<td>a) <strong>inactive</strong> – lying down, sleeping, resting, napping</td>
</tr>
<tr>
<td>b) <strong>somewhat inactive</strong> – sitting, reading, watching television, playing video games, time in front of the computer, playing games or activities which are mostly done sitting down</td>
</tr>
<tr>
<td>c) <strong>somewhat active</strong> – walking, shopping, light household chores</td>
</tr>
<tr>
<td>d) <strong>very active</strong> – running, jumping, skipping, bicycling, skating, swimming, games that require lots of movement and make you breathe/sweat hard</td>
</tr>
</tbody>
</table>
Following is a sample of a completed time period:

<table>
<thead>
<tr>
<th>Activity Level</th>
<th>Percentage</th>
</tr>
</thead>
<tbody>
<tr>
<td>a) inactive</td>
<td>5%</td>
</tr>
<tr>
<td>b) somewhat inactive</td>
<td>60%</td>
</tr>
<tr>
<td>c) somewhat active</td>
<td>25%</td>
</tr>
<tr>
<td>d) very active</td>
<td>10%</td>
</tr>
<tr>
<td>TOTAL</td>
<td>100%</td>
</tr>
</tbody>
</table>
**WEEKDAY ACTIVITY**

For *one typical weekday in the past 2 weeks*, (choose from one of Tuesday, Wednesday or Thursday), please estimate the percentage of time that you spent in each activity level.

1. **After getting out of bed until starting breakfast:**
   - a) inactive ______ %
   - b) somewhat inactive ______ %
   - c) somewhat active ______ %
   - d) very active ______ %
   
   **TOTAL** 100%

2. **After finishing breakfast until starting lunch:**
   - a) inactive ______ %
   - b) somewhat inactive ______ %
   - c) somewhat active ______ %
   - d) very active ______ %
   
   **TOTAL** 100%

3. **After finishing lunch until starting supper:**
   - a) inactive ______ %
   - b) somewhat inactive ______ %
   - c) somewhat active ______ %
   - d) very active ______ %
   
   **TOTAL** 100%

4. **After finishing supper until bedtime:**
   - a) inactive ______ %
   - b) somewhat inactive ______ %
   - c) somewhat active ______ %
   - d) very active ______ %
   
   **TOTAL** 100%
For the typical weekday that you are referring to, please answer the following questions as accurately as possible in the spaces provided.

5. At what time did you get out of bed in the morning? ______

6. At what time did you start eating breakfast? ______

7. How long did you spend eating breakfast? ______ minutes

8. At what time did you start eating lunch? ______

9. How long did you spend eating lunch? ______ minutes

10. At what time did you start eating supper? ______

11. How long did you spend eating supper? ______ minutes

12. At what time did you go to bed that evening? ______

13. For the typical weekday that this questionnaire has asked you about, please rate your overall level of activity (please circle one response only):
   a) very inactive
   b) inactive
   c) somewhat inactive
   d) somewhat active
   e) active
   f) very active

14. Is this “typical” Tuesday, Wednesday or Thursday that you described in this questionnaire (please circle one response only):
   a) a lot like most weekdays
   b) a little bit like most weekdays
   c) a little bit different from most weekdays
   d) a lot different from most weekdays
SATURDAY ACTIVITY
For one typical Saturday in the past 2 weeks, please estimate the percentage of time that you spent in each activity level.

15. After getting out of bed until starting breakfast:
   a) inactive ______ %
   b) somewhat inactive ______ %
   c) somewhat active ______ %
   d) very active ______ %
   TOTAL 100%

16. After finishing breakfast until starting lunch:
   a) inactive ______ %
   b) somewhat inactive ______ %
   c) somewhat active ______ %
   d) very active ______ %
   TOTAL 100%

17. After finishing lunch until starting supper:
   a) inactive ______ %
   b) somewhat inactive ______ %
   c) somewhat active ______ %
   d) very active ______ %
   TOTAL 100%

18. After finishing supper until bedtime:
   a) inactive ______ %
   b) somewhat inactive ______ %
   c) somewhat active ______ %
   d) very active ______ %
   TOTAL 100%

For the typical Saturday that you are referring to, please answer the following questions as accurately as possible in the spaces provided.
19. At what time did you get out of bed in the morning? ______
20. At what time did you start eating breakfast? ______
21. How long did you spend eating breakfast? ______ minutes
22. At what time did you start eating lunch? ______
23. How long did you spend eating lunch? ______ minutes
24. At what time did you start eating supper? ______
25. How long did you spend eating supper? ______ minutes
26. At what time did you go to bed that evening? ______

27. For the typical Saturday that this questionnaire has asked you about, please rate your overall level of activity (please circle one response only):
   a) very inactive
   b) inactive
   c) somewhat inactive
   d) somewhat active
   e) active
   f) very active

28. Is the “typical” Saturday that you described in this questionnaire (please circle one response only):
   a) a lot like most Saturdays
   b) a little bit like most Saturdays
   c) a little bit different from most Saturdays
   d) a lot different from most Saturdays

29. If you have any comments about your activity patterns that you think are important, please mention them on the back of this page. Thank-you.
“This is the Best Fatal Illness you can Have”: Exploring and Comparing the Parenting Experiences of Youth With Cystic Fibrosis and Congenital Heart Disease

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Bios

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Abstract

Despite the important instrumental and emotional role that parents play in the lives of children with cystic fibrosis (CF) and congenital heart disease (CHD), qualitative researchers have not examined the similarities and differences between caregivers’ experiences. Informed by thematic analysis, in this qualitative study, I explored what it is like to care for a child with a chronic illness from the perspective of CF and CHD parent’s at a children’s hospital in Canada. Pediatric caregiver stress was qualitatively different between CF and CHD parents, and temporal dilemmas were unique sources of stress for CF parents only. To alleviate stress, all parents drew on a three way, inter-related process to comprehend their child’s illness and acquire perspective.

By opening up the social worlds of parents, I have illuminated important similarities and differences in the caregiving experience of CF and CHD parents, and offer novel contributions to the literature.

Keywords

Parents; perceptions; children; chronic disease; cystic fibrosis; congenital heart disease; qualitative inquiry
Childhood chronic diseases, such as congenital heart disease (CHD) and cystic fibrosis (CF), are illnesses that persist for greater than six months and affect children’s daily functioning. Given advances in health care over the past several decades, however, more children with illnesses that were once characterized as life threatening are now reaching adulthood. Termed the acute to chronic disease era, approximately 20 - 29% of North American children are living with chronic health conditions (Branstetter, Domian, Williams, Graff, & Piamjariyakul, 2008). This begs clinicians to better attend to the needs of this new group of children who are characterized by complex medical and psycho - social needs.

Although quantitative and qualitative researchers have studied the impact of caring for a chronically ill child, to my knowledge, there are no qualitative studies that contrast and compare the experience of parents that care for children with CF and CHD. In addition to describing both the commonalities and context specific idiosyncratic differences in their care giving experiences, and developing descriptive theoretical frameworks, such comparative qualitative literature is required to identify groups that experience greater psycho - social morbidity and appropriately direct health services. Given the strong relationship between parental and child health outcomes, understanding CF and CH parents’ experiences might help clinicians to assist parents in better supporting their children. Thus, by opening up the social world of parents, the purpose of this study was to explore what it is like to parent a child with CF or CHD and to compare these experiences. Before describing this study, a review of the literature is undertaken.

**Review of the Literature: Parenting a Medically Fragile Child**

Because it magnifies notions of dependence and vulnerability, childhood illnesses have been described as a condensed symbol or concretized metaphor of childhood itself (Young, Dixon -Woods, Findlay, & Heney, 2002). The notion that (a) children are “natural innocents,” (b) that illness in childhood is rare, (c) that illness threatens the futurity of children, or our cultural investment in who children are, (d) that we associate illness with death and (e) that it is
not the natural order of things, intensifies notions of childhood innocence. Childhood illnesses invoke a sense of catastrophe in the cultural imagination (Young et al., 2002).

A review of the quantitative and qualitative research evidence revealed five themes that characterize the experience of parents that care for children with chronic diseases. These include (a) the negative physical and psycho-social health implications of caring for a child with a chronic health condition, (b) the ways in which childhood diseases threaten the integrity of parenthood, (c) the systemic barriers that parents encounter in efforts to better care for their child (d) parents desire to normalize their child’s life, and (e) the sense of hope that results from caring for a medically complex child. These research themes and limitations are discussed below.

In relation to the parents of healthy youth, Ingersky, Shaw, Gray and Janicke (2010) found that the parents of youth with chronic illnesses report higher rates of psycho-social distress and impairment. Specifically, Ingerski et al (2010) examined the prevalence of subclinical post traumatic stress symptomology among children and parents affected by multi organ transplant, HIV, and sickle cell anaemia, as well as the correlates associated with post traumatic stress. All three parent groups reported higher levels of post traumatic stress in relation to healthy parents, and parents of youth awaiting organ transplantation reported the highest rates of psychological morbidity. Illustrating the important relationship between parental health and child health outcomes, parents’ post traumatic stress symptomology was associated with greater perceived child symptoms. Other recent quantitative studies have also reported high rates of psycho-somatic stress among the parents of children with chronic diseases (Cabizuca, Portella – Marquest, Mendlowicz, Coutinho, & Figueira, 2008). For example, in a prospective study of parental adjustment to childhood cancer, mothers displayed high levels of anxiety and insomnia in relation to fathers and healthy families (Sawyer, Antonious, Toogood, Rice, & Baghurst, 2000). Similarly, fathers of children with asthma report excessive worry, anxiety, and uncertainty related to caregiving, and learn to live with a sense of chronic concern (Cashin, Small, &
Foster et al (2001) explain that given the uncertainty associated with CF in the future and the intrusive nature of treatment into one’s daily life, the parents of youth with CF experience a great deal of stress. Described as an insidious disease, parents recall how periods of relative health in the life of the CF child falsely lulled them into a deceptive sense of security, and they fear the progressive nature of the disease (Foster et al., 2001). For parents who care for children with severe heart disease, the constant vigilance required to oversee their child’s complex home medical technology is also described as stressful (Rempel & Harrison, 2007).

In addition to poor physical and psycho-social health, these parents also encounter systemic barriers. For example, it is stressful for parents to interact with unsupportive health professionals who do not listen to their concerns and treat their child as a diagnosis, rather than a unique individual (Kratz, 2009). Parents report that there are school related barriers associated with caring for a child with a chronic illness. School policies related to their child’s medication use are experienced as restrictive, and parents often break school policies to ensure that their child has access to medication in a timely fashion (2008).

Researchers also suggest that parenting a child with a chronic illness might threaten parental integrity and identity. In a qualitative study that examined the experience of caring for a child with cancer, Young et al (2002) found that cancer disrupts both the mothers’ and child’s biography. These mothers are embedded within dominant constructions of motherhood which emphasize that good mothers subsume their own needs under those of the child. Within the context of caring for a child with cancer, however, mothers protect their precarious biographies and construct new identities. Much like other parents who find the experience of caring for a child with cancer to be life shattering (Woodgate & Degner, 2003), Hodkinson and Lester (2002) found that the mothers of children with CF encounter a threatened identity in which important aspects of their former selves, such as spending time with friends or attaining more education, are lost. Similarly, Grob et al (2008) discuss the compromised parental integrity associated with
caring for a newborn with CF. Although genetic newborn screening should be heralded as an important medical breakthrough, the disease is at the forefront of the initial parenting experience and marks the end of a normal relationship with the infant (Grob, 2008). Although parents eventually find ways to powerfully connect with their child, the newborn period is characterized by threatened parental integrity.

The parents of youth with chronic illnesses express concerns related to their child’s normalcy. In a recent qualitative study, Earle, Clarke, Eiser, & Sheppard (2006) found that while the mothers of children with Acute Lymphoblastic Leukemia (ALL) strive to normalize their child’s life, the search for normalcy is potentially damaging if it continues to elude parents. Rather than a state, mothers describe normalcy as a transient process that is characterized by peaks and valleys, and something that is only realized once it is threatened. Mothers only experience a sense of normalcy in between clinic visits to the hospital when they temporarily forget about ALL (Earle et al., 2006). Hill, Higgins, Dempster & McCarthy (2009) also found that striving for normalcy is a central concern for fathers that care for children with ALL. In the post treatment maintenance phase, fathers actively minimize their child’s difference and encourage their child to partake in pre-cancer activities. Fathers often encounter resistance to normalcy from women spouses who are more likely to adopt an overprotective stance even during their child’s remission (Hill et al., 2009). Similarly, Anderson (1981) found that although the parents of chronically ill youth semantically label their child as normal and engage in normalizing practices that diminish difference, they insist that their child engage in non-normative behaviour, such as isolating themselves from other children to avoid infection, without considering such practices as incongruent with their understanding of normalcy.

Despite poor parental health, threatened parenthood, and negotiating their child’s normalcy, however, current studies have explored the positive aspects associated with caring for a child with a chronic health condition. In a qualitative study that explored the experience of
fathers that care for children with Juvenile Rheumatoid Arthritis, fathers described their child’s illness as a “catalyst” for a more meaningful, rich, and engaging interaction with their child (MacNeill, 2007). In addition to broadening a narrowly defined gender role to include space for caregiving, fathering a child with a chronic health condition was associated with a more intimate and loving interaction. In a recent qualitative study, Samson et al (2009) examined the “fabric of hope” for parents of children with Duchene’s Muscular Dystrophy (DMD). Despite their initial despair, as time passes, parents’ perceptions toward their child change and they come to recognize their child’s unique individuality regardless of illness. Parents experience a sense of serenity and despite their child’s significantly reduced life expectancy, the child is described as the fabric of hope itself. Kratz et al (2009) also report that there are positive experiences associated with caring for a child with a chronic illness, such as gaining appreciation for the things that really matter and personal growth.

From this review of literature, it is evident that the parents of youth with CF and CHD as well as other chronic illnesses, experience reduced psychosocial and physical health related to caregiving. In addition, they encounter systemic barriers to care giving, experience threatened parental integrity, and question whether their child is normal or not. However, recent literature has documented the positive and transformative aspects associated with caring for a child with a chronic health condition, and, rejecting the dominant assumption that caregiving belongs to the sole preserve of motherhood, also illuminated the perspective of fathers.

While this literature has been critical to furthering a better understanding of caregiving, there are three limitations. Specifically, while CF and CHD caregivers’ experiences have been studied separately (Grob, 2008; Rempel & Harrison, 2007), the research is characterized by the absence of studies that contrast and compare their experiences. It should be noted that CF and CHD are different childhood diseases that greatly differ in terms of trajectory and treatment. Given the unique differences between these two conditions, exploring and contrasting their
parenting experiences was thought to be a novel and intriguing line of investigation, that, given several years of volunteer work with these two populations, also sparked a great deal of personal interest. This type of investigation allowed me to compare the similarities and differences between those parents that care for children with chronic but manageable diseases, such as CHD, as well as those that care for children with fatal illnesses such as CF that are characterized by gradual health losses over time - and how these very different illness trajectories might potentially influence their storied experiences. Such comparative research may facilitate the development of theories that better describe the commonalities between CF and CHD parents. The development of explanatory frameworks might also illustrate the context specific idiosyncrasies that characterize the unique and particular experiences of CF and CHD parents.

Furthermore Ingersky et al (2010) suggest that such comparative research is required to identify, develop, and deliver specific psycho-social health services to those children and caregivers who have greater psycho-social morbidity. Furthermore, parents’ attitudes toward their child’s illness influence child health outcomes, such as adjustment to illness and quality of life (Hill et al., 2009). For this reason, it is of great importance to explore how the parents of children with a variety of chronic diseases construct their child’s health. Gathering such information might allow professionals to educate parents on the relationship between parental and child health outcomes, so as to better support their child’s social and emotional adjustment to illness. The need to capture the similarities and differences between CF and CHD parents’ experiences, identify groups with greater psycho-social morbidity, and better support children with CF and CHD, were powerful rationales for this research. Thus, in this study, I sought to address the following research questions: What is it like to parent a child with CF? What is it like to parent a child with CHD? Are there similarities and differences in the parenting experiences between CF and CHD parents? The methodology which guided this study is outlined below.

**Methodology: Research Setting and Participants**
This study was approved by the Research Ethics Board at a metropolitan children’s hospital in Toronto, Canada and took place in the CF and CHD outpatient clinics. The original study sought to explore parental perceptions toward physical activity among CF and CHD parents. The results reported in this article relate to parents’ emergent discussions about their child’s illness. Recruitment for the study was facilitated by two nurses in the CF and CHD clinics and a purposeful sampling strategy was employed. After the nurses identified eligible participants for me using the patient data base, I called parents to request participation and arranged a convenient interview time. During the interviews, some participants mentioned that I appeared familiar. However, I had no formal previous research or clinical role with any of the participants and they were unknown to me.

Twenty eight parents (14 parents from the CF clinic and 14 parents from the CHD centre) participated. Although all participants provided care to a child with CHD or CF between the ages of 10 and 18 years, medical history and disease severity varied. Although some children had never been hospitalized, others had been hospitalized on several occasions and had undergone numerous procedures. At the time of the study, three participants’ children were hospitalized, and one participants’ child was receiving oxygen therapy and was currently on the lung transplant list. Most parents had more than one child and provided care to only one sick child. However, three of the participants in the CF sample and one of the participants in the CHD sample were providing care to two children with chronic diseases.

Parents were between the ages of 35 and 55 years and the sample was gender balanced. Objective indicators of race and culture were not employed during this study and it was unclear as to how culture might have influenced parents’ perceptions toward their child’s health. However, while the majority of participants could be characterized as White, during the interviews, some participants self identified as African Canadian, Spanish, Middle Eastern, or East Indian. Although most parents described themselves as healthy, a few parents were dealing
with health problems such as excess weight, diabetes, or physical injury. In terms of employment, parents fulfilled a number of occupational roles. Some parents were professional white collar workers, such as nurses, teachers, and engineers. Other parents were engaged in blue collar jobs such as trucking or farming. Additionally, a few parents were unemployed and receiving unemployment insurance benefits. Finally, some participants described themselves as stay at home parents. While it was not the case for all of the stay at home parents, most had made this decision to provide care for the sick child. Although most participants were in long term married relationships, some participants were divorced, remarried, or single. Parents were from a range of urban and rural locations across Ontario and Quebec. The conceptual framework which informed this study is described below.

Conceptual Framework

This study was informed by the formative work of Arthur Frank (1995). The theoretical concept of the illness narrative from the Sociology of Health and Illness provided a conceptual lens to interpret the data. Narratives, that is, the stories that people with illnesses and their loved ones construct, are central to human existence and to a better understanding of the clinical and medical world. The availability of an explanatory framework or narrative structure by which to explain their experiences is of great importance to those with illnesses or those touched by it (Ezzy, 2000). When life is disrupted by illness and the integrity of the self is threatened, narrative allows for the reconstruction of the self (Whitehead, 2006). Illness narratives are not just personal, and, although they are told within the context of one’s personal history, they speak to social and political dimensions associated with contemporary social life (Ezzy, 2000). As suggested by Nettleton (2005) who states that …“the story reveals as much about the culturally available discourses as it does about the minutiae of a teller’s life” (p. 49), it is evident that in listening to an individual’s illness narrative, we learn much about the discursive cultural resources that patients and caregivers have available to them to interpret their experiences, and
Frank (1995) proposed that there is a typology of three illness narrative types which characterize how people with illnesses and those that care for them interpret their experiences. As the culturally preferred story line, the restitution narrative is characterized by a departure from health when one discovers that they are ill. Adopting Parson’s classic sick role in which the ill person is temporarily abdicated from normal responsibilities, the patient engages in a brief period of convalescence. The restitution narrative concludes with the patients’ eventual triumph over illness and their return to the land of health. In contrast, the chaos narrative lacks a clear start and finish, and, rather, the patient or caregiver remains totally consumed by illness. The chaos narrative is characteristic of chronic and incurable illnesses, or those conditions that lack a clear diagnosis and treatment plan, such as chronic fatigue syndrome. The chaos narrative is characterized by feelings of terror and loss of control, and the sense that the body has been completely engulfed by illness. The stories of bodies in chaos are never resolved and patients brace themselves for continued health losses. Given that it rouses terror and forces us to contemplate our own invincibility and ontological insecurity, the horror associated with the chaos narrative renders it culturally unsayable (Smith & Sparkes, 2005; Nettleton et al., 2005). Finally, the quest illness narrative is more than one’s departure from health to illness and back to health again. Rather, body and self are profoundly transformed through illness, and illness is seized as a tool for introspection and self-reflexivity (Whitehead, 2006). Values are re-prioritized through illness, and, in some cases, illness acts as the impetus for social change or the discovery of destiny (Whitehead, 2006). Although there are weaknesses associated with using narrative as a way to understand illness, by, for example, foreclosing a discussion on the elusory nature of embodiment, narrative is a useful tool for understanding how parents make sense of illness in a world of health (Radley, 1997). The qualitative method that guided this study is described below.
Qualitative Method and Analysis: Thematic Analysis

Informed by the work of Braun and Clarke (2006), thematic analysis was the qualitative research method that guided the development and analysis of this study. Thematic analysis is a foundational qualitative method that seeks to identify, analyze, and report patterns and themes in the data; it is thus a way of organizing, describing, and interpreting a qualitative data set. While thematic analysis is a flexible and diverse qualitative method that is compatible with realist and constructivist paradigms, it has not garnered the same degree of attention as the other major research canons. Furthermore, Braun and Clarke (2006) suggest that both “bottom up” or “top down” approaches might be adopted in a thematic analysis. By remaining close to the data, not attempting to fit it into pre-existing frameworks, and drawing on multiple theories to interpret my results, my analysis was data driven and can be characterized as a bottom up or inductive approach. Moreover, I did not simply aim to reflect and describe parents’ experiences at the literal or semantic level. Rather, at the latent level, I made efforts to contextualize and interpret their narratives within their life worlds (Braun and Clarke, 2006).

Data Collection and Analysis

In employing thematic analysis as the method in this study, I conducted semi-structured interviews (Mason, 2002). Informed by the conceptual framework and the review of the literature, a general interview guide was developed. The interviews occurred in clinic rooms and were 45 to 85 minutes long. Although the interview guide was semi-structured in nature, participants’ responses drove further lines of inquiry.

To facilitate the analysis of data, all interview data was audio-taped and transcribed verbatim as the study was occurring. I also recorded detailed field notes during the data collection process and potential analytical insights. Following Braun and Clarke (2006), first, to completely immerse myself within the data and to familiarize myself with it, I engaged in multiple in-depth readings. Second, for the purposes of coding the raw data, or the smallest
units that can produce meaningful insights related to the phenomena, the data was read again and coded. In this case, commonly occurring words and phrases across the data corpus - such as “stress, burden, or exhaustion -” were coded. Furthermore, I coded as much of the available data as possible, and ensured that the coding process was not context stripped by encapsulated the context under which the discussion occurred. Third, the coded data was collated and organized into higher order themes. This careful and recursive decision making process involved making important judgments about (a) which codes were similar and dissimilar, (b) which codes could be grouped together, and (c) where particular codes “fit.” This iterative process involved continually moving back and forth between codes and themes and the relationship between them. Fourth, through a process of revision, the themes were collapsed and refined. Fifth, to define the essence of the theme and what it means in relation to other themes, the themes were carefully defined and named. Theme names adequately describe and explain what the theme is about and offer sufficient evidence, or data, to corroborate and describe it. This process was assisted by my Doctoral Supervisor in a form of peer debriefing. He reviewed half of the transcripts and dialogued with me about my interpretations to challenge the identified themes and their connections (Lincoln & Guba, 2000). Finally, in my final presentation of the report below, I provide a story about the data both within and across themes. In doing so, as an analyst who is both a cultural member and critical commentator (Braun & Clarke, 2006), I offer a story which describes and interprets how the parents of youth with CF and CHD understand their child’s illness. Pseudonyms have been used to protect the anonymity of the participants.

**Results: Overview**

Three main themes are reported in this study. First, while a *sense of stress* was ever present for all parents, CHD and CF parents narrated stress in different ways. Second, the *temporal losses* associated with the time consuming nature of treatment and the life limiting nature of the disease, was a specific source of stress for CF parents only. Third and finally, both
CF and CHD parents dealt with the chronic stress of caregiving by *coming to terms with their child’s illness and learning to put things into perspective*. The first theme is described below.

**The Stress of Parenting a Child with a Chronic Illness: CHD and CF Parents’ Narratives**

When explaining what it is like to parent a child with CF or CHD, all parents described their child’s condition as stressful. Stress was a chronic burden that was always “in the back of one’s mind.” The nature of parent’s stress varied and included such worries as the deterioration of the child’s health, whether their child properly adhered to treatment, and the negative impact of illness on scholastic performance. Caring for a sick child was associated with detrimental health consequences for parents, and while participants did not disclose clinical mental health conditions to me, they reported negative feelings such as depression and anxiety. Some parents reported engaging in self harming behaviours, such as consuming alcohol excessively.

However, there were important differences in the way that CF and CHD parents narrated their experience of caregiver stress. For CHD parents, stress was located in the past, in history and memory. Some parents had lived with an undiagnosed cardiac baby – or “blue baby-” for months or even years. When parents approached clinicians regarding their infant’s poor health, they were sometimes questioned sceptically or told that they were simply over anxious. Although clinician scepticism and blame was stressful, an intuitive sense that their child was not well - not eating, sleeping, or behaving like a normal baby - motivated them to seek out second opinions and to have their claims legitimized. Although stress remained high for CHD parents throughout this process, arriving at the hospital and receiving a diagnosis which confirmed their initial beliefs, was associated with relief. Stress for CHD parents remained high during the process of surgical repair to correct their child’s heart defect, and, in some cases, parents had to endure their child receiving multiple stressful surgical procedures.

However, the experience of stress for CHD parents rapidly dissipated in the post surgical repair period, and, despite parents’ initial fears about open heart surgery, the functional benefits
of surgery to their child were immediate and rapid. For example, observing that their once “blue baby” now had a “normal pink complexion” resulted in great relief for parents. Similarly, observing their child gradually resume normal feeding behaviours and gain weight signified to parents that their child was beginning to recover. Therefore, while CHD parents narrated stressful experiences in the pre-surgical period, such as obtaining a diagnosis that legitimized their concerns, the experience of stress rapidly diminished in the post surgical repair period when their child began to recover. For example, in the narrative below, the father of a child with CHD explains that it was stressful to observe his child’s failure to thrive in the pre-surgical period.

When his child began to thrive in the post surgical repair period, however, he experienced relief:

   Yes, at the beginning it was stressful, he was only seven months old. He was like 11 lbs and friends of ours that had babies at the same time, you see them getting big and strong. That gave us worries, then. After his surgery, he just caught up to everyone else (Gregory).

The father of an adolescent patient with CHD eloquently narrates the stress and fear associated with being on “survival mode” in the pre-surgical period when his daughter was gravely ill:

   It was a big stress at the beginning. At the beginning, the first year and half until she had an operation. She was about four weeks old ... She did not develop as a normal kid and was behind, until she had another operation, when she was about a year old. That was difficult. We could not feed her and keeping her on the right track. Then everything settled. She became better and the problems went away. Just making it, just surviving. She had a few operations and it did not seem that she would pull through or make it. It was scary (Charles, father)

In contrast, CF parents narrated their experience of caregiver stress in qualitatively different ways. Not unlike the parents of youth with CHD, the initial diagnostic period was
characterized by shock and sadness, and, as Jerry describes below, learning about the life
limiting nature of the disease was particularly devastating and stressful for him and his wife:

She (wife) was upset by that. She had learned about the lifespan. That is what really upset
her, that she (daughter) was going to die before her. A parent never wants to have a kid
die before her and that is what we were upset about (father).

Parents explained that as they became more educated about CF and ensured that
treatment become a routine component of everyday family life, over the course of time, CF
entered a period of relative normalcy and mundaneness. However, unlike CHD parents, CF
parents narrated a qualitatively different experience of stress that was chronic and never
resolved. In particular, it was frightening for parents to observe their child’s non-adherence to
treatment during the typical rebellious phase that characterizes adolescence, and they worried
that their child was intentionally compromising their health. Parents projected their stress into the
future and it had a protracted and futuristic dimension. Knowing that a time would come when
their child’s health deteriorated - and that their child would eventually succumb to CF – was a
chronic and unresolved concern encountered by CF parents. Despite how normal their child was
at the moment, they worried about their child’s deterioration in the future and vehemently fought
to delay the onset of that time. In the following narrative, for example, the mother of a
hospitalized adolescent with CF explains that her entire family constantly worries about Evan’s
potential deterioration and death in the future. Grace explains how Evan’s current hospitalization
has shattered any semblance of normalcy and reminds her about the finality of CF:

I would have to say that it has impacted our life for us as parents. It has constantly been
on our mind. “When will he get sick? Will he die?” That has played huge on Rick
(husband) and I. Sean and Jason (other sons) - I think they think about it, sometimes, but
not in any impact. Because when you look at him (CF child), he looks healthy and he is
“go, go, go” so it does not seem to be an issue. Him coming here on Saturday (hospital)
has made a big impact even on his siblings ... He (husband) and I are constantly asking ourselves - “what would we do if - how would we prepare for”? ... Jessa (daughter) and I have talked about that too. “What would we do if? How sad would that be if?” (Grace).

Mary also explains that it is stressful to think about her son’s health deterioration in the future. She feels a sense of helplessness in knowing that there is nothing that she can do except encourage him to abide by the doctor’s orders:

You feel usefulness and helpless ... And then they talk about there being no cure for CF-sometimes I think we worry about that too and him getting sicker in the future ...You try to find answers but there is really nothing that I can give him. Just that, you know, we have to do what the doctor says and follow the rules and keep on going (mother).

Thus, CF and CHD parents have qualitatively different narrations of caregiving stress. For CHD parents, stress was largely located in the past and dissipated post surgically. In contrast, for CF parents, stress was unresolved and had a future orientation in which they worried about their child’s deteriorating health and death. In the following theme, a unique source of stress experienced only by the parents of youth with CF is described: stolen time.

“Stolen Time:” The Temporal Dilemmas of CF Parents

The parents of youth with CF narrated significant temporal stress related to caring for a child with a chronic illness. Time was thought to be stolen by CF and parents’ narratives about time had both literal and metaphoric dimensions. For example, parents described the tedious and time consuming nature of engaging in daily physical therapy treatment with their child. Treatment detracted from parents’ spontaneity and freedom and time was experienced in highly structured ways. Furthermore, the time consuming nature of physical therapy treatment required parents to conceptualize their daily time differently. Ensuring that there was adequate time to complete treatment with their child was of great importance to parents, weighed heavily on them,
and dictated their use of time. All other activities, such as engaging in recreation or socializing, were secondary to ensuring that their child completes treatment on time.

Despite the importance of treatment in the lives of parents, during the interviews, parents self reflexively considered how treatment had come to dominate their time. Although it was of utmost importance to parents and was a non-negotiable facet of CF care, the effort required to structure and plan treatment was stressful. Parents were cognisant of the daily toll that treatment had on their and their child’s use of time. For example, in the following narrative, Sandy expresses stress and frustration as she describes how the time consuming nature of CF treatment, and the dietary requirements, interfere with important family time:

When you want to go on day trips, it is like, “hurry up get it done, we want to get going.” You end up pushing him (child with CF). “OK - now you have to eat something.” We want it done (therapy) before we go out for the day.” Sometimes, when you are gone all day, and you get home at 9:00 pm at night. We bring it with us to do in the car. It is always at the back of our minds. It never leaves. You are planning from the minute you get up as to how we will coordinate this mask and physiotherapy (Sandy, mother).

Similarly, the father of a ten-year old child with CF who was hospitalized at the time of the study explains the high degree of planning required to ensure that his daughter completes her treatment and its impact on the entire family’s routine. Because CF treatment is performed prior to school, it requires his daughter to wake early and interferes with her sleep:

The one thing that Emily (daughter with CF) jokes about is that “you wake me up so early for school and my friends wake up 20 minutes before the bell rings.” With Emily, we have to get up; she has to have her physiotherapy done in the morning, and she has to have it done at night. The answer to that is that we see the effect in time. We need to plan our day. We need to wake up earlier than other families. This includes Sharon (wife) and myself ... We need to plan to wake up early to fit in this physiotherapy. We need to
plan it in at night time to make sure that we fit it in before bed, along with homework.

The answer is time. We have to plan the day out. Other families can fly by the seat of their pants and do this and that (Erik).

Additionally, when discussing the interaction between their child’s illness and time, CF parents described time in a more metaphorical and abstract sense. Parents were cognisant of their child’s reduced life span and this was experienced as stressful. Within the context of reduced life time, parents described wanting to get the most out of life as possible in the time they have. Specifically, some parents described a temporal re-organization in which they took strides to do certain activities now, such as spending as much time with their child as possible or signing up for a particular set of lessons, that, had they had a healthy child, they would have done later.

For example, learning about reduced life expectancy was experienced as particularly stressful for the father of a child with CF. This husband-wife dyad chose to parent their child in ways that accord with dominant gender ideologies, and, thus, his wife is the primary caregiver for their child with CF. To compensate for reduced life time, Billy encourages his wife to spend as much time with their child as possible:

A parent never wants to have a kid die before her and that is what she was upset about. That is why I was trying to tell her to spend as much time as you can, with her. And just think; every waking moment that you have, spend it with her. Even if you have both of them or one by themselves, spend that time with her. I had her quit her job and that is why I work 16 hours a day. So that she can spend more time with them (father).

Similarly, within the context of reduced life time, the father of two children with CF described the importance of living for the moment. In the event that CF compromises the ability to engage in certain enjoyable activities in the future, Joe ensures that his children have the opportunity to pursue their interests immediately:

We want to live for the moment while we can do these things. We have another girl in
our town who is a few years older than our daughter who has been in the hospital multiple times. If my daughter wants to take singing lessons now, I do not care how much it costs - she is going to take them (father).

Stolen time was a unique source of stress for the parents of youth with CF only and did not characterize the experience of CHD parents. Furthermore, some CF parents compensated for reduced life time by ensuring that they live for the moment. In the final theme, the process by which CF and CHD parents alleviated the stress associated with caregiving is explained. Specifically, parents alleviated stress by developing a better understanding of their child’s condition and learning to put things into perspective.

Learning to Put Things Into Perspective: CF and CHD Parents’ Constructions of their Child’s Illness

Parents alleviated stress by coming to an understanding of their child’s illness and learning to put things into perspective. Specifically, they drew on three inter-related processes to understand their predicament and these are described as sub-themes below.

“The more disabled other:” There are those that are worse off than my child. Parents came to an understanding of their child’s illness by comparing their son or daughter against those children who were thought to be more ill. Although parents were cognisant of their stress, at all times, they were aware of how much worse things could be. When contrasting and comparing their child with a more severely disabled “other,” it appeared that parents had constructed their own tacit hierarchy of illness, that is, their own understanding of what constitutes a really sick child. For CHD parents, children with CF were thought to be more ill than their child. This spontaneous comparison made by CHD parents may be because of the fact that youth with CF and CHD receive care in adjacent clinics, and are often exposed to one another.

For both CF and CHD parents, the proverbial image of the “dying child with cancer” served as a powerful representation of just how sick a child may become. Parents’ understanding
that things could be so much worse occurred through observations of the social and spatial milieu around them. For example, parents interacted with other families whose child had received more prolonged treatment, profound disruptions to family life, or even the deterioration and death of the child. Below, the father of two children with CF states that in relation to other illnesses, CF is manageable. Joe’s narrative is premised on the assumption that caring for a child with CF at home is, somehow, less stressful than receiving chemotherapy or using a wheelchair:

This is the best fatal illness that you can have. Many of the elements of the illness are up to you. We can do them (treatment) at home. We do not have to come in for chemotherapy or radiation. We are not in a wheelchair. The treatments take a long while, but again, the fact that the only difference for my kids is that they are having enzymes with lunch. It is not much at all (father).

Parents contemplated how difficult it must be to parent a more chronically ill child. Despite displaying empathy for these other parents, interacting with more severely disabled children in the hospital setting dampened parents’ stress and allowed them to narrate their experience in a more positive light. Although seeing other sick children did not make parents feel better, it allowed them to put things into perspective and to develop an understanding that their situation was bearable after all. When one walks through SickKids, it is very common to see children with a wide variety of illnesses, as well as the various stigma symbols that mark their young bodies as different, such as wheel chairs, intravenous poles, or naso–gastric feeding tubes. During the interview, the mother of a child with CF explains that walking through SickKids serves as a stark reminder of others’ more extreme suffering in relation to one’s own child, and her narrative takes the form of a plea to the unknowing public:

“You take a walk through the hospital. Take one look. Even when your child has the worst diagnosis with the heart or cancer, you look at those kids; there are those worse then you.” It just makes - it does not make you feel better but it puts it in perspective, it
does. You do not dwell on anything and you are just thankful (Angela).

Comparing their child with a more severely disabled other was the first process by which parents learned to put things into perspective.

“Walk a mile in my shoes:” “Healthy” families just don’t get it. The healthy normal child served as a point for comparison for CF and CHD parents, and was a part of the process of coming to terms with their child’s condition. For instance, since they lacked the same experiences, participants frequently explained that healthy parents will never be able to grasp the difficulty associated with parenting a chronically ill child. With a sense of annoyance or resentment, parents like Erik trivialized their relatively minor health concerns, such as broken bones or colds:

We tell people about Emily (daughter) and how we are in hospital and how we have been in the hospital seven times. They will tell you a story of how once their child’s fever went up to 101 degrees and they freaked out. Right? How you felt that one time in your ten year olds life is a weekly to daily occurrence for us, in our life. If Emily is wheezing and coughing, at anytime, it can go downhill ... Even if she is fine today, next week she could be in the hospital and that is at all times. So we are always living on that edge. At anytime, she could be in hospital. It is definitely more stressful. Other families with CF-ers will. Families with completely healthy kids who have never been in the hospital and have had a fever once, I do not think they quite understand (father).

Parents consistently emphasized that it was difficult to really comprehend what it is like to parent a chronically ill child if one lacked personal experience. Stating that “others do not understand,” they drew stark divisions between parenting a healthy and a sick child. Parents felt a degree of connection with the parents of other ill children and were detached from the parents of healthy children. For example, the father of two children with CF poignantly states that:

If you had not heard news about your child being given a death sentence, then you do not know how I feel. Sorry, you could have gone to school, but you do not know how I feel
Recognizing that their child was not like healthy children, and that healthy families would not be able to understand their experiences, was part of the process by which parents learned to bring legitimacy and gravity to their experiences and to put things into perspective. “My child’s pain is greater than my own.” Recognizing that their child bears the greatest burden was the final process that parents drew on to better comprehend their child’s illness. Parents were cognisant that since their child’s body is inflicted by illness – and not their own – they experience greater suffering. Parents put their own pain, fear, and stress into perspective by reminding themselves that their child hurts more than they do. For example, the mother of a child with CF states that “what I feel is nothing compared to what it puts on him.” (Rose)

Similarly, although Shelly expressed stress related to her child’s non adherence to treatment, as the interview progressed, her anger dissipated and she became reflective. Witnessing her son’s ongoing struggle with CHD has put things into perspective, and made her realize the relatively trivial nature of her own health issues in relation to her son’s CHD:

You know, they make you strong, watching what they go through. I do not even care. I mean, I do care about myself. But they really do make you stronger, watching them go through something like that. If you have watched your kid go through that, you can go through anything. When you get told that you are sick, it will not hit you as hard, when you have watched our kids grow up that way, yes (mother).

Thus, the parents of youth with CF and CHD bring meaning toward their child’s illness and learn to take perspective by drawing on three inter-related processes. Furthermore, CF and CHD parents construct their child’s illness in the same way. By comparing their child over and against a more disabled other, parents are able to dampen their fears and to put things into perspective. By recognizing that parents of healthy children will not ever be able to understand
their experiences, parents draw stark divisions between sick and healthy children, and give voice and legitimacy to the grave nature of their parenting experiences. Although parents acknowledge that their child’s illness has resulted in personal turmoil, parents humble themselves and find courage in the recognition they do not bear the greatest burden of suffering. The three findings from this study, that is, caregiving stress, stolen time in CF, as well as learning to take perspective, are analysed below within the context of the literature.

**Analysis and Discussion**

The conceptual insights which informed this study lent much interpretive value to the analysis. Drawing on Frank’s (1995) typology of illness, there appeared to be qualitative differences in the way that CHD and CF parents narrated their stressful parenting experiences. Although stress related to parenting a child with CHD is ever present and requires managing a chronic childhood illness, parents were elated that they had pulled through a difficult experience and survived, and they recalled that their most stressful experiences were a thing of the past. Thus, CHD parents were characterized by a frightening departure from a state of health at the time of diagnosis. Adopting a sick role when hospitalized, children and parents experienced a prolonged convalescence which was trying at times and tested survival. However, the post-surgical repair period marked their child’s return to relative health; they had triumphed over CHD and it was a thing of the past. Drawing on Franks’ typology of illness (1995), it appears that CHD parents adopted a protracted restitution narrative that is marked by a departure from health, a socially appropriate occupation of the sick role, and a triumphant return to health.

In contrast, the parents of youth with CF displayed different stress narratives that contained elements of both chaos and quest typologies. The diagnosis of CF was met with initial shock, remorse, and a sense of chaos. As parents educated themselves about CF and routinized treatment as an everyday part of life, CF entered a period of normalcy and stability. While it was not characteristic of most participants, a few parents storied quest narratives. They dialogued
about how the onset of CF was a life-altering learning moment that had led them to engage in introspection; they learned to re-prioritize values and weigh up what was really important. However, CF parents knew that their grasp on health was precarious and fragile at best; chaos was projected forward and involved taking stock of the fact that their child’s health would deteriorate in the future (Whitehead, 2006). In this regard, CF parents were never afforded with an opportunity to triumph over illness and their stress narratives contained elements of suffering and pain in the shadowy future. As clinicians and researchers, it is important that we be aware of the very different illness narratives that the parents of youth with CHD and CF take up in the process of storying their stressful caregiving experiences (Frank, 1995). For CF parents who live with chronic unresolved stories and a protracted sense of chaos, it might be important to facilitate the development of more hopeful ways of storying their experiences.

CF parents also described significant temporal stress that was absent from the narratives of CHD parents. For CF parents, time appeared to be “stolen” in both a literal and abstract sense. The time consuming nature of treatment was onerous and stressful for parents and required so much scheduling that it detracted from spontaneous opportunities and a sense of freedom. Furthermore, parents alluded to time in a more abstract sense. They were aware of their child’s reduced life span and this was distressing. While it was not characteristic of all parents, some parents described wanting to get the most out of life in the time that they have. In contrast, the parents of youth with CHD did not dialogue about stolen time. Although this requires further research, it appears that post surgical repair, the treatment burden for CHD is experienced as minor and parents generally do not consider their child to be life limited.

While CF parents’ narrations about stolen or lost time are a unique contribution from this study, other authors have discussed how CF changes perceptions toward the passage of time. Qualitative studies by Swisher and Erikson (2008), Berge, Patteron, Goetz, and Milla (2007), Foster et al (2001) and Glasscoe and Smith (2008) emphasize the time consuming and intrusive
nature of treatment for families affected by CF. In addition to being a daily reminder of living
with a fatal condition, the never ending cycle of treatment is extremely stressful for parents.
Parents constantly worry about the consequences associated with non-adherence to treatment and
often neglect their other healthy children (Foster, 2001). Additionally, time collapses for CF
parents. Rather than experiencing a linear series of events in their child’s life, the temporal
experience for parents is punctuated by illness related emergencies in the child’s life (Glasscoe &
Smith, 2008). Thus, although the management of other childhood diseases may also be perceived
as time consuming, temporal losses appear to be a unique feature that distinguishes CF and CHD
caregivers (Jessup & Parkinson, 2010; Tluczek, Chevalier, McKenhnie, & Lynam, 2010).

The theoretical base aids in better understanding the concept of stolen time for CF
parents. Gibson, Zizelsberger & McKeever (2009) suggest that “…etched into the social fabric
of Western societies is a conception of time that encourages individuals to exert control over its
passing, to be future oriented, enjoy free time, and not waste time.” (p. 554). Gibson et al (2009)
explain how time is a resource for control and conflict, and, in late modern capitalist states, time
is conceptualised as an objective entity which must be seized, controlled, and quantified.
Informed by Sir Isaac Newton, linear time describes our current objective consideration of time.
As a late capitalist modernist construct associated with progress, time is thought to be an
objective entity which stands independently of social life and the meanings which we ascribe to
it. Linear time emphasizes the progress of productive rationale actors toward the attainment of
outcomes on time. Furthermore, the dominant time culture, which is engrained in childhood and
embedded within the cultural imagination, has profound implications for how we understand
childhood development. Indeed, children quickly learn about the importance of attaining taken
for granted temporal roles, such as graduating or gaining employment. Furthermore, we are all
measured against the expectations associated with a linear time course. Linear time has no place
for idle activities, and, thus, activities which do not contribute toward instrumental outcomes,
such as rest or self care, are thought to be wasteful of time.

The dominant linear time culture might be particularly damaging for those, such as “futureless” children or people with certain life limiting conditions, who might not achieve temporal milestones on time. In a qualitative analysis of people with muscular dystrophy, for example, Gibson et al (2009) suggest that for those with life limiting conditions in which the future is uncertain, modernist conceptualizations of time might be oppressive and lead to a profound sense of powerlessness. Always measured against normative standards of time, people with disabilities are often found lacking. Drawing on the work of Bourdieu who proposes that we take stock of the subjective experience of time, Gibson et al (2009) propose a radical re-conceptualization of time. In addition to dismantling the social and temporal discourses that shape the existence of people with disabilities, she proposes that we address the inequalities of times by celebrating the cycles and processes that occur in the shadow time of the present. Similarly, in his discussion of those living with HIV/AIDS, Ezzy (2000) discusses a philosophy of living in the present. Engaging with present time and not investing in or depending on the future, characterizes such a philosophy. Informed by Aristotle’s politics, those that live with a philosophy of the present are agentic subjects who act on their environments, yet embrace the finite nature of human life and forces that exist outside of our control. They can look to the future without a dependence on it to enjoy today.

Although we all vary in our capacity to resist the dominant time culture (Gibson et al., 2009), most CF parents spoke of a loss of time and this roused stress and sadness. Thus, it appears that living with a philosophy of the present is not characteristic of how these parents cope with their child’s illness or how they conceptualize time. As we all are, it appears that the parents of youth with CF are embedded within dominant conceptualizations of time. They are aware of, and hold their child against, the temporal norms associated with modernist linear time. In addition to taking stock of the temporal dilemmas that are unique to CF parents, professionals
might empower these parents to work toward developing a philosophy of the present. It is important to note that developing a philosophy of the present might not be a successful strategy for all CF parents, and might be influenced by parents’ position along their child’s illness trajectory. Indeed, there are likely many ways in which these parents come to terms with and story their child’s experience. However, developing such a philosophy that is characterized by a radical re-conceptualization of time, in which one (a) enjoys the present (b) plans for the future without depending on it or colonizing it and (c) acknowledges uncertainty and finality, might promote more positive ways of coping for some parents, and more meaningful ways of enjoying time with their child. We must also take stock of the ways in which illness changes one’s sense of time (Glasscoe & Smith, 2008). Indeed, as health care advances continue to increase survival in CF, to prolong time and sustain life in a dominant linear sense, we can also expect that the narratives of CF patients and parents will change and begin to reflect the dominant time culture relationships that currently characterize healthy populations.

The three stage process by which CF and CHD parents learned to put things into perspective is a novel contribution from this study. Indeed, it alleviated parental stress and facilitating a sense of meaning and understanding. Moreover, CF and CHD parents both appeared to employ the same, three stage perspective taking process. While this appears to be a novel finding that has not been reported previously, particular aspects of this process accord with some previous literature. For example, the fathers of children with ALL often report frustration related to the minor health issues that parents with healthy children complain about. They tend to trivialize these experiences, and, like the participants in my study, state that others will never be able to understand their experiences (Hill et al., 2009). Similarly, recognizing that “things could be so much worse” is one of the strategies that the parents of youth with illnesses draw on to normalize their situation. Additionally, Upward Theory (Festinger, 1954), in which people appraise their experiences by engaging in social comparison with others, also supports my
results. As well, while it is beyond the scope of this study, other scholars might wish to apply Said’s (1979) original and adapted theoretical work on Otherness to further analyse how ones’ illness identity may only come into existence in and through its Other. Further to this, the experience of those parents whose child is “Othered” within the context of treatment for childhood chronic illnesses, such as children with cancer, should be sensitively explored.

Described as forgotten parents (Hill et al., 2009), dominant gender ideologies exclude fathers from caring for children with illnesses, and incorrectly assume their more instrumental and less emotional role. In contrast, my study adds to the growing body of literature on fathering children with chronic illnesses, and is a notable contribution. Like McNeill (2007), my study rejects the problematic assumption that fathers are uninvolved in caregiving. My study involved dialogues with fathers who wanted to occupy a central caregiving role in their child’s life in both emotional and instrumental ways. Fathers rarely employed dominant gendered discourses to conceal their caregiving impact, and expressed a strong desire to care give. Future researchers might wish to examine how gender influences caregiving among CF and CHD parents.

Conclusion

Because the ability of parents to adapt to and cope with their child’s chronic illness is central to child health outcomes (Hill et al., 2009), it is important to explore the meaning of parenting a child with a chronic health condition. By cleaving open the parenting experiences of CF and CHD parents, this theoretically informed study sought to address critical research limitations, and revealed novel and important similarities and differences in how the parents of youth with CF and CHD experience and narrate caregiving. Although stress was present for both groups, CF parents narrated protracted future worries that, in some cases, illustrated an unresolved chaos narrative. Stolen time was a unique source of stress for CF parents, and they grieved the loss of life time and spontaneous uses of free time. By drawing on a three way, “Sick Other - Healthy Child – Self” process that allowed them to put things into perspective, CF and
CHD parents dealt with caregiver stress in similar ways. The findings add to the literature on caring for children with chronic illnesses. Ingersky et al (2010) suggest that comparative research is useful for identifying high risk groups so that appropriate services are delivered. From this, it can tentatively be concluded that since psycho-social morbidity is greater among CF parents, more services are required. By opening up their private worlds, this study has not only illustrated the parenting dilemmas encountered by CF and CHD caregivers, but illuminated the subtleties, nuances, and differences, between them.
References


APPENDIX Six

List of Publications and Conferences

Published


In press


Moola, F. J. “This is the best fatal illness you can have”: Contrasting and comparing the parenting experiences of youth with cystic fibrosis and congenital heart disease. *Qualitative Health Research.*


*Conferences*


**Moola, F. (2008).** Towards a sociology of physical activity in congenital heart disease (CHD): The social determinants of physical activity and health in children and youth with CHD. *Canadian Student Health Research Forum, Canadian Institute for Health Research (CIHR),* The University of Manitoba, Canada.