Comparing Patient Engagement and Patient Advocacy Activities and Measuring Preferred Roles in Medical Decision Making among Cystic Fibrosis Patients, Caregivers, Family Members and Patient Advocates

by

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Abstract

Background: Patient engagement has become a priority for healthcare organizations, yet remains inconsistently defined. Patient engagement has also become a priority for healthcare providers. Current discourse suggests that patients today desire more control over medical decision making than ever before, and that the proliferation of online health information is empowering patients to make autonomous medical decisions. Yet, prior research indicates that patients typically prefer to share decisions with their physician, raising questions about what patient engagement is and whether preferences have changed over time.

Methods: This study addresses multiple research questions using three phases of data collection. Phase one asked: how are ‘patient engagement’ and ‘patient advocacy’ defined and perceived by individuals who are involved in engagement and advocacy activities? This question was addressed by conducting semi-structured key informant interviews (N=11).

Phases two and three examined preferred roles in medical decision making among cystic fibrosis (CF) patients, caregivers, family members and patient advocates, and whether these roles were related to: trust in the physician, self-assessed health information literacy and demographics.

Phase two involved conducting a quantitative cross-sectional survey (N=165), which included three scales: the Problem-Solving Decision-Making Scale (PSDM), the Trust in Physician Scale (TPS), and the electronic Health Information Literacy Scale (eHEALS). T-tests and logistic regressions were computed to determine the relationships between variables. Phase three involved interviewing a subgroup of survey participants to help to explain survey results (N=19). Thematic analysis was used to identify common themes.

Results: Our findings suggest that patient engagement and advocacy have multiple meanings, that most survey participants preferred to share medical decisions with their physician (>80%), and that self-assessed health information literacy was associated with preferred roles; as eHealth literacy went down, so did desire for participation in making medical decisions. Follow-up interviews supported these results; participants mentioned how they assessed the credibility of online health information and how they preferred to direct questions to their physician.

Conclusion: This research highlights the need for clear language regarding patient engagement activities. This study also suggests that eHealth literacy may influence decision making preferences, and that preference for autonomous decision making should not be assumed.

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List of Abbreviations

CF	Cystic Fibrosis
DM	Decision Making
PS	Problem Solving
PSDM	Problem-Solving Decision-Making Scale
EHEALS	Electronic Health Information Literacy Scale
TPS	Trust in Physician Scale

CHAPTER 1

Background and Objectives

Engaging patients in decisions about their health and healthcare has emerged as a significant policy priority in recent years. Existing evidence suggests that patient engagement can improve quality of care, enhance patients' experiences and outcomes, and help to control healthcare costs (Carman et al., 2013). As a result, decision makers have sought to incorporate patients in a range of decision making processes, including policy decisions (i.e., at a macro-level), research and organizational activities (i.e., at a meso-level), and in decisions about their own medical care (i.e., at a micro-level). Despite these efforts, however, questions persist about what patient engagement is (i.e., how patient engagement ought to be defined and distinguished from other concepts and terms), what types of activities patients prefer to be involved in, and whether some patients desire less involvement in medical decisions over others.

An increasing volume of empirical literature and commentary have advised that patients, once characterized as 'passive bystanders', have come to desire more active control over medical decision making (O'Malley et al, 2011; Landro, 2017). This proposed shift, from passive bystander to 'active agent', has been supported by the fact that access to online health information is changing how physicians and patients interact, enabling increased information seeking and patient autonomy, and encouraging patients to self-diagnose their conditions using 'Dr. Google' (Pogacar, 2017; Lu, Xu & Wallace, 2017). However, these propositions assume that patients now consult the Internet as a substitute for professional medical advice, which may in fact not be the case.

Previous research conducted by Deber et al. (1996) distinguished between two aspects of preferred roles in medical decision making: Problem Solving (PS) and Decision Making (DM) tasks (measured by the PSDM scale). PS refers to decisions that are preference-independent; they require factual knowledge about a clinical situation and treatment options, whereas DM refers to preferences for outcomes and attitudes about treatment interventions. Three categories of preferred roles emerged from this work: *Passive* patients; who favour handing over PS and DM tasks to their physician, *Shared* patients; who favour handing over PS but sharing or keeping DM tasks, and *Autonomous* patients; who wish to exercise control over both dimensions.

Existing literature suggests that patients (mainly those with cancer) typically prefer sharing medical decisions and seldom desire to be autonomous (Chewning et al., 2012). However, it is conceivable that preferences have changed over time as health information has proliferated online, and that preferences might vary by health condition. In fact, little is known about the preferences of patients with rare diseases like cystic fibrosis (CF). In addition, few studies have considered the preferences of informal caregivers and family members who often support such patients and contribute to patient decision making. Yet, even those within a single disease community might have differing values and preferences. Experiences as a caregiver or family member may also influence one's preferences when interacting with a provider as a patient.

Beyond this, the academic literature includes little research on the decision making preferences of patient advocates (or those who formally advocate on behalf of patients with an organization). Identifying what roles patient advocates wish to assume in their own care, though, is important for uncovering if differences exist between the preferences of patients and those seeking to represent patient groups. Capturing potential differences is especially significant when one considers the responsibilities that advocates take on; particularly the task of identifying and representing patients' needs and preferences. One might also suppose that patient advocates, who are often very vocal and well-versed about the health system, are perhaps most likely to prefer an autonomous decision making role. Research is therefore needed to allow for a more nuanced understanding of patients', caregivers', family members' and patient advocates' preferences, rather than assuming that the distribution of preferences is similar across these groups. A survey was thus used in this dissertation to measure preferred roles in medical decision making among members within the CF community, and the extent to which trust in the physician, self-assessed health information literacy and demographic variables influence these preferences.

Self-assessed health information literacy (or eHealth literacy) involves measuring "a person's ability to assess their own skills at finding, evaluating, appraising, integrating, and applying what is gained in electronic environments towards solving a health problem" (Norman & Skinner, 2006, p. 2). It was hypothesized that participants with low eHealth literacy would be more likely to prefer a passive decision making role. In addition, because many studies have described how trust can facilitate patient-centered care and how distrust in physicians encourages patients to seek information from non-medical sources (including the internet and family and friends) (Wrede-Sach et al., 2013), trust was also measured in this study.

In this dissertation, a number of research questions are addressed through a mixed-methods research design that incorporated three phases of data collection: 1) key informant interviews, 2) a quantitative cross-sectional survey and 3) qualitative follow-up interviews of a subgroup of survey participants. Each phase was used to address different research questions within a broader patient engagement framework. Section 1.1 outlines these research questions and section 1.2 provides an overview of each data collection phase.

1.1 Research Questions

<u>Phase 1: Key informant interviews addressed the following research questions (1,2):</u>

1. How are ‘patient engagement’ and ‘patient advocacy’ defined and perceived by individuals who are involved in engagement and advocacy activities?
2. What are the perceived similarities and differences between engagement and advocacy activities?

<u>Phase 2: A cross-sectional survey addressed the following research questions (3-5):</u>

3. Within the Cystic Fibrosis (CF) community (patients, caregivers, family members and patient advocates), what are their: preferred roles in medical decision making, level of trust in physicians, and self-assessed health information literacy?
4. Are there differences in preferred roles in medical decision making, trust in the physician, and self-assessed health information literacy by participant category (i.e., between patients, caregivers, family members and patient advocates)?
5. Are preferred roles in medical decision making associated with: trust in the physician, self-assessed health information literacy, and demographics characteristics?
    a) What are the differences in trust and eHealth literacy scores between participants who preferred a passive, shared or autonomous role?

Based on these questions, it was hypothesized that:

A) Very few CF patients, caregivers, family members and patient advocates will have a preference for an autonomous decision making role despite the increased availability of health information on the Internet

B) Patient advocates will have a greater preference for a shared or autonomous decision making role than others in the CF community

C) Preference for a passive decision making role will be associated with lower self-assessed health information literacy

D) Preferences for a passive and shared decision making role will be associated with greater trust in the physician and lower education

Phase 3: Follow-interviews of a subgroup of survey participants addressed research topics (6-9):

6. Reasons for participants' preferred roles and factors that influence these preferences
7. Sources of health information participants use when considering decisions about their health, and the amount of trust they have in these sources of information
8. When and why participants use online health information
9. Factors that influence trust in their physician and the physician-patient relationship

1.2 Data Collection Phases

In phase one, key informant interviews were conducted with experts in the fields of patient engagement and patient advocacy. These interviews were used for the purpose of better understanding what 'patient engagement' and 'patient advocacy' mean (i.e., how these terms are being defined and interpreted). While these terms are regularly used in the grey and academic literature, they remain inconsistently defined and poorly understood. As a result, a more nuanced understanding of what these terms mean and how they might differ, both conceptually and operationally, was required. It was also hoped that interview responses would help clarify the perceived similarities and differences between engagement and advocacy activities. Phase one of this study was thus exploratory in intent and was pursued independently of phases two and three.

In phase two, an online cross-sectional survey was conducted of CF patients, caregivers and family members, as well as regional and provincial advocates who volunteer with Cystic Fibrosis Canada (CFC). This survey measured preferred roles in medical decision making (using the aforementioned PSDM), self-assessed health information literacy (using the electronic Health Information Literacy Scale [eHEALS]), trust in the physician (using the Trust in Physician scale [TPS]), and demographic variables. While previous research suggests that patients typically prefer to share medical decisions with their physicians, few researchers to date have explored the preferences of patients with rare diseases like CF. CF is a genetic disease that causes blockages in the lungs and other major organs and requires optimal medical management. Likewise, few studies have measured the preferences of caregivers, family members and patient advocates.

In phase three, follow-up telephone interviews were conducted with a subgroup of survey participants. These interviews were used to gather additional information about the research participants and helped to explain survey results.

1.3 Thesis Outline

This thesis consists of the following chapters:

Chapter 2 provides an overview of the relevant grey and academic literature. This includes providing a description of how patient engagement and advocacy have been conceptualized and defined, and outlining the literature on patients' preferred roles in medical decision making, self-assessed health information literacy, trust in physicians, and other factors which shape the physician-patient relationship. Chapter 2 begins with a broad discussion of patient engagement and advocacy, and then narrows in scope to address physician-patient interactions.

Chapter 3 provides a description of the methods used in this dissertation. As noted above, this study employed a mixed methods design that consisted of three phases of data collection. These descriptions include: objectives and research questions, study populations, and the data collection and analysis techniques used.

Chapter 4 presents themes that emerged from the eleven key informant interviews conducted in phase one that addressed research questions 1 and 2.

Chapter 5 describes the results of the cross-sectional survey administered to CF patients, caregivers, family members and patient advocates that addressed research questions 3-5. Data is presented on participants' demographic characteristics, medical decision making preferences, eHealth literacy, and trust in the physician. The relationships between these variables are also considered and presented.

Chapter 6 presents findings from the follow-up telephone interviews conducted in phase three that addressed research topics 6-9.

Chapter 7 discusses the findings of this dissertation and the implications of this work for physicians, policy makers and other health system stakeholders. Recommendations for future research and inquiry are also discussed.

CHAPTER 2

Concepts and Literature Review

This chapter provides an overview of the patient engagement and patient advocacy literature, and provides a description of the theoretical framework used to guide this dissertation. Literature on the physician-patient relationship and how this relationship is evolving is also discussed, as well as research on patients' preferred roles in medical decision making, trust in physicians, self-assessed health information literacy, and clinical and demographic factors which shape preferred roles. To facilitate this review, key online databases were searched for relevant content, including ProQuest, PubMed, Medline, and Google Scholar. Search terms used included: patient- and family-centered care, patient engagement, patient advocacy, preferred roles in medical decision making, treatment decision making, trust in physicians, theoretical frameworks, health information literacy and electronic health information literacy.

2.1 Patient Engagement

In Canada and internationally, engaging patients in decisions about their health and healthcare has emerged as a significant policy priority. Patient engagement is now widely recognized as being essential for improving patient safety and satisfaction, controlling healthcare costs, recognizing patient autonomy, and enhancing the management of chronic diseases (Barry & Edgman-Levitan, 2012; Bellows et al, 2015). Patient engagement has even been touted as being the "blockbuster drug of the century", being critical to a learning health system, and as empowering patients to take control of their health (Carman et al., 2013, Domecq et al., 2014). In many ways, this movement aligns closely with broader efforts to move away from paternalistic and physician-centric medicine (where physicians exercise control over healthcare tasks) towards a new paradigm of practice that seeks to empower patients. Whereas patients were once viewed as having limited knowledge and expertise, decision makers now recognize the value of engaging patients as a means to improving healthcare delivery (Lindblad et al., 2017).

Despite the optimism that has fueled this growing movement, however, what patient engagement means, when it is desirable, and what it ought to encompass remains unclear. Adding to this dilemma is the fact that patient engagement remains inconsistently defined in the literature, and is a term that is used synonymously with a number of other terms, including (but not limited to): patient advocacy, patient empowerment, citizen engagement, public participation, and shared decision making (Prey et al., 2014; Bellows et al., 2015; Gallivan et al., 2012). Closely related to engagement includes efforts to implement patient-centered care (or 'patient- and family-centered care') (Entwistle & Watt, 2013). Patient-centeredness is defined by the Institute of Medicine (IOM) (2001) as "providing care that is respectful of and responsive to individual patient preferences, needs and values".

The patient-centered movement places great emphasis on building strong physician-patient relationships, encourages providers to involve patients in decision making processes, and aims to improve patients' experiences and outcomes (Capko, 2014). Patient engagement is perhaps best regarded as falling under the umbrella of patient-centered care, although these terms are conceptually similar and also used interchangeably in the literature. The current state of patient engagement research and practice can thus be characterized as evolving and multi-directional. The author's own prior discussions with experts in the field have affirmed that disagreements persist about what patient engagement does and does not encompass, and that engagement is often perceived as being indistinguishable from other concepts and terms.

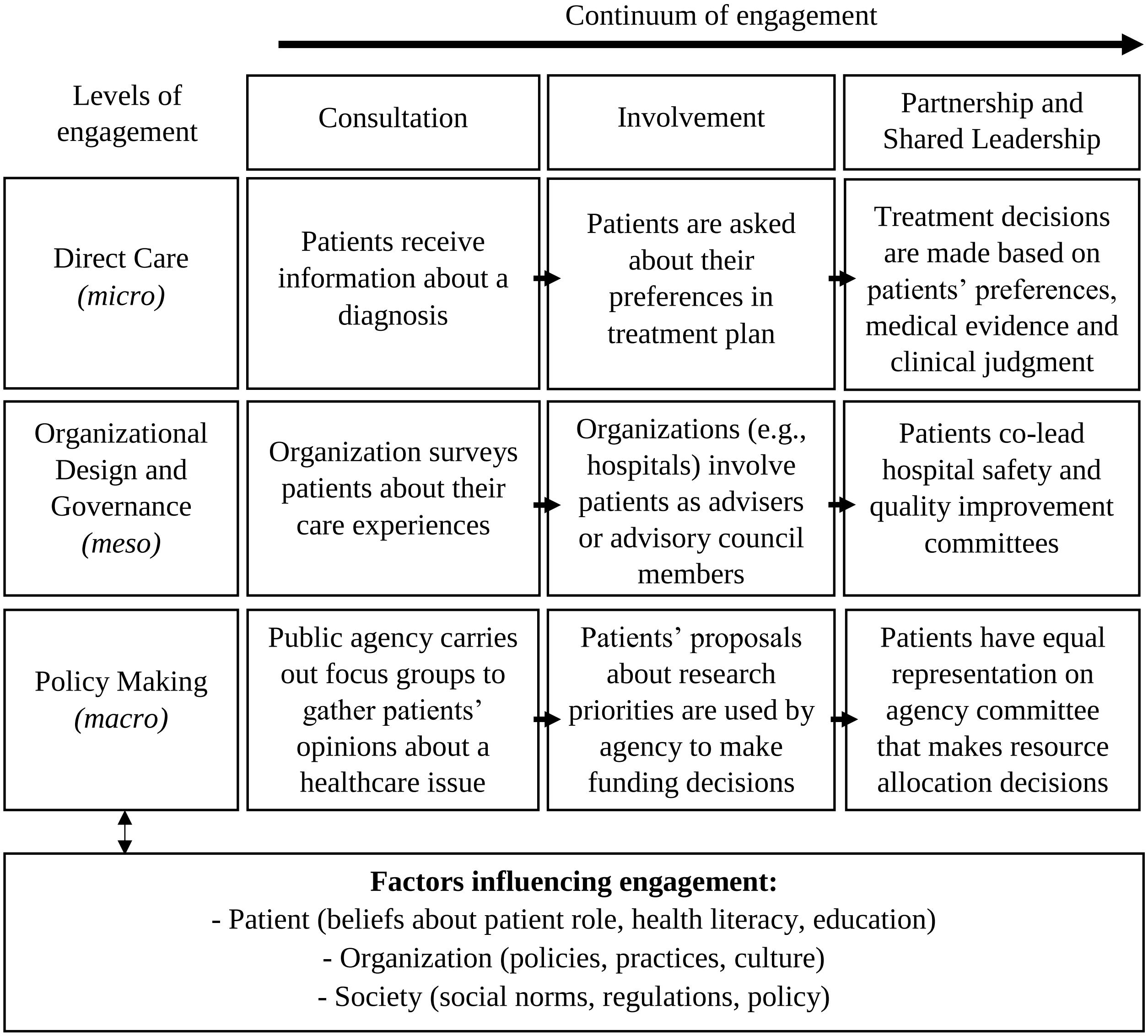
2.1.1 Types of Patient Engagement Activities

Accordingly, many scholars and agencies have sought to develop frameworks to help clarify when, how and where engagement takes place. These frameworks have employed a range of terms, with some being more specific to patients than others, but have all tended to emphasize that engagement has multiple meanings and occurs along a continuum. One framework that is regularly cited and expanded upon, for instance, originates from the International Association for Public Participation (IAP2) and their IAP2 spectrum. This spectrum was developed to help categorize the public's role in public participation processes and comprises five levels: inform, consult, involve, collaborate and empower (IAP2, 2007). In the first level, inform, decision makers convey information to the public about a problem and possible solutions. In contrast, empowerment occurs when the public is asked to make final decisions (e.g., via citizen juries). While this framework is typically equated with policy making, it has influenced the development of several other approaches to engaging patients, including Carman et al's. (2013) framework.

Carman et al's. (2013) patient engagement framework is amongst the clearest and most widely-consulted frameworks in North America and was used to guide this dissertation (see figure 2.1). This framework presents engagement as being influenced by multiple factors and as occurring at three levels: the direct care (micro) level, the organizational design and governance (meso) level, and the policy-making (macro) level. Each of these levels includes three stages: consultation, involvement, and partnership and shared leadership. At the direct care level, patients interact with their physician and may assume different levels of responsibility over their own care. At the organizational and governance level, patients may become involved in organizational decisions and help co-lead organizational committees. At the policy-making level, patients may be asked to participant in funding and/or resource allocation decisions.

This framework thus offers a useful way for thinking about the aspects of patient engagement. For example, phase 1 of our research sought to gain a better understanding of how patient engagement and patient advocacy are defined and operationalized by individuals who are involved in engagement and advocacy activities, which could occur at a meso and macro level. In contrast, phases 2 and 3 explored preferred roles in medical decision making and focused on the direct care or micro level.

Figure 2.1 – Carman et al's. (2013) Framework



Continuum of engagement →

Levels of engagement	Consultation	Involvement	Partnership and Shared Leadership
Direct Care *(micro)*	Patients receive information about a diagnosis →	Patients are asked about their preferences in treatment plan →	Treatment decisions are made based on patients' preferences, medical evidence and clinical judgment
Organizational Design and Governance *(meso)*	Organization surveys patients about their care experiences →	Organizations (e.g., hospitals) involve patients as advisers or advisory council members →	Patients co-lead hospital safety and quality improvement committees
Policy Making *(macro)*	Public agency carries out focus groups to gather patients' opinions about a healthcare issue →	Patients' proposals about research priorities are used by agency to make funding decisions →	Patients have equal representation on agency committee that makes resource allocation decisions

Factors influencing engagement:
- Patient (beliefs about patient role, health literacy, education)
- Organization (policies, practices, culture)
- Society (social norms, regulations, policy)

In accordance with Carman et al’s. (2013) framework, several examples can be found in the grey and academic literature which underscore the different ways that patient engagement has been defined, modeled and conceptualized across domains and levels of interaction. In her seminal work ‘Engaging Patients in Healthcare’, for instance, Coulter (2012) relates patient engagement to the physician-patient relationship (or the micro-level) and emphasizes the importance of shared decision making processes that “support active patient and public involvement in health and healthcare” (p. 87). Throughout much of the literature, shared decision making is described as being an ‘equal partnership’ between patients and physicians, and as being necessary when uncertainty exists about medical options (e.g., when there is no clear ‘right’ or ‘wrong’ treatment option) (Pomey et al., 2015). Physicians and patients must therefore work together to weigh the benefits and drawbacks of treatments until a consensus about an action is reached.

This ongoing dialogue differs considerably from other paradigms of medical practice, where decisions are made primarily by physicians (i.e., in a paternalistic model) or when decisions are made primarily by the patient (i.e., in an autonomous or consumerist model). Drawing attention to the quality of communication between patients and physicians, Coulter (2012) advises that most patients today wish to be involved in their own care and that information about a patient’s diagnosis, treatment options and potential outcomes is needed to facilitate engagement.

Research conducted by several scholars supports this position and recommends that there is considerable room for improving how patients and providers interact. In a survey of patients with various medical conditions, for example, Fagerlin et al. (2010) found that many patients did not receive information from their physicians about the risks and benefits of treatment options despite desiring to be informed. Likewise, Lee et al’s. (2011) study discovered that breast cancer patients treated with mastectomy had “low knowledge about breast reconstructive surgery” despite being heavily involved in treatment decisions (p. 6). These authors concluded that poor communication between patients and physicians likely resulted in this lack of knowledge, and proposed that interventions be implemented to support patient decision making (Lee et al., 2011). Patient engagement from this perspective, then, (i.e., at the patient-provider level), is primarily about strengthening the patient voice, accounting for patients’ informational needs and preferences, and ensuring healthcare providers work with (not just for) patients.

In contrast, Hibbard & Greene (2013) provide a much different characterization of patient engagement by suggesting that engagement is required to facilitate *patient activation*. According to these scholars, patient activation relates to a “patient’s ability to take independent actions to manage their own health and healthcare” (Hibbard & Greene, 2013, p. 207). Patient engagement interventions are designed to increase activation, such as training patients to seek preventative care, to self-manage their chronic diseases, and to engage in regular physical activity. The interplay between activation and engagement underscores how decisions and lifestyle behaviours can positively or negatively affect one’s health. Such actions can be captured using the Patient Activation Measure (PAM); a questionnaire which categorizes patients into one of four activation levels (Hibbard et al., 2004). Multiple studies have demonstrated that patients who score higher on PAM are significantly more likely to engage in healthy behaviours, to seek preventative care (e.g., immunizations), to search for health information, and to accumulate lower healthcare costs (Hibbard & Greene, 2013; Green & Hibbard, 2012; Mahoney, Stock & Tusler, 2007; Fowles et al., 2009).

Beyond engaging patients at a micro-level, researchers have also emphasized the need to engage patients in the planning and designing of comparative effectiveness research (CER), defined as comparing the “evidence on the effectiveness, benefits, and harms of different treatment options” (Agency for Healthcare Research and Quality [AHRQ], 2017). CER is said to be important for providing evidence-based information to patients, caregivers, providers and decision makers that can be used to facilitate informed decision making (Mullins, Abdulhalim & Lavallee, 2012). Whereas CER studies were once designed with little input from patients (typically referred to as ‘research subjects’), many researchers now recognize the value of engaging patients as ‘research partners’. It is also argued that engaging patients is necessary for reaching vulnerable populations (Las Nueces et al., 2012), promoting the democratic ideals of participation, accountability and transparency (Esmail, Moore & Rein, 2015), and satisfying the moral argument that patients, as the users of healthcare services, have a right to be involved in shaping research (Robinson, Newton & Dawson, 2012). The phrase ‘nothing about me without me’ encapsulates this position.

Patient engagement has additionally been described as “researchers doing research with patients, rather than for, at or to them” (Esmail, Moore & Rein, 2015, p. 133). Within the United States, this shift has been fueled (in part) by the creation of the Patient-Centered Outcomes Research Institute (or PCORI) in 2010 (Selby, Beal & Frank, 2012). Passed under the *Affordable Care Act*, this institute was designed to promote research that is guided by patients, caregivers and the broader healthcare community (PCORI, 2018). As the nation’s largest non-profit funder of CER, PCORI includes patients in the review of its research projects, encourages patients to identify research topics, and aims to produce research that is evidence-based (Sacristán et al., 2016).

A comparable approach to engaging patients in research can also be found in Canada, with the creation of the ‘Strategy for Patient Oriented Research’ (or SPOR) in 2012. Specifically, this initiative is a coalition of federal, provincial and territorial partners (comprising ‘research units’) that attempt to improve patient care through the levers of research (Canadian Institutes of Health Research [CIHR], 2017). SPOR units include patients, researchers, academic institutions, policy makers and other stakeholders that work together to address research priorities identified by patients (Légaré et al., 2017). Patient engagement is defined as occurring when patients “meaningfully collaborate in the governance, priority setting, and conduct of research, as well as in the summarizing, distributing and applying of resulting knowledge” (CIHR, 2017). Some of the ways that patients are involved in SPOR include: being part of research committees that design research projects, occupying positions on SPOR’s national steering committee (which oversees its implementation), and being more directly involved in research.

Other institutions have emphasized that patient engagement may extend beyond research. According to the US-based Institute for Patient- and Family-Centred Care (IPFCC) (2018), patient- and family-centered care (PFCC) is an “…approach to the planning, delivery, and evaluation of healthcare that is grounded in mutually beneficial partnerships.” PFCC can take many forms, such as involving patients and their families in quality improvement tasks (via hospital advisory committees), the education of healthcare providers, facility design, and policy improvement (IPFCC, 2018). The deliberate inclusion of families in this definition is based on the recognition that family members play a critical role in supporting patients in decision making processes both within and outside the health system.

Involving patients in policy decisions (at a macro level) is cited in the literature as being yet another form of patient engagement (Souliotis, 2016). Engagement at this level can occur when patients (or citizens) play a role in health system governance and inform resource allocation and distribution policies. Advocates of democratic participation have long contested that involving patients in policy decisions is essential for identifying and addressing population health needs.

2.2 Patient Advocacy

In addition to the patient engagement movement, patient advocates and advocacy organizations have also played a role in empowering the patient voice and advancing patient-centred care. Patient advocacy can take many forms, but broadly involves an individual, group or organization acting on behalf of a patient or population to improve their health or healthcare experiences (Gonzalez, 2018). Although the exact origins of patient advocacy remains somewhat contested, the term ‘advocate’ can be found in an ethical document produced in the 1920s by the American Nursing Association (ANA) which urged nurses to “…advocate for and strive to protect the health, safety and rights of patients” (Ronnebaum & Schmer, 2015, p. 238).

In many developed nations, patients first organized into advocacy groups in the 1940s and 50s in an effort to support people with chronic diseases. By the 1950s, for instance, non-profit cancer organizations were established in Canada, the UK and the US (Paul, 2008). These organizations brought together patients and cancer survivors who sought to educate the public about screening tests for diseases, solicit donations for cancer research and treatments, and lobby governments for funding. The 1960s and 70s were also characterized by extensive advocacy. For example, cancer advocates developed public awareness campaigns which contributed to changes in how breast cancer was viewed and treated by the medical profession (Ganz, 1995). Some patient groups also began urging that access to healthcare was a human right, and that patients should have input in decisions about their health (Aymé, Kole & Groft, 2008; Clarke, Pederson & Santalucia, 2015). In the 1990s, a significant victory was won in the US when the Food and Drug Administration (FDA) announced the creation of the ‘FDA Patient Representative’; an advisory body of patients that provides “input into the agency’s decision making processes associated with medical products for drugs, biologics, and medical devices” (Gonzalez, 2018; FDA, 2018).

Today, the term ‘patient advocate’ has come to have a variety of meanings and applications, and is often used interchangeably with the terms ‘patient representative’ and ‘patient liaison’ (Clarke, Pederson & Santalucia, 2015). In the legal, psychological, sociological and medical literature, advocates are described as playing many roles, representing diverse populations, and having various goals and aspirations (Ronnebaum & Schmer, 2015). Gilkey et al. (2008) offer a useful framework (described in section 2.2.1) which categorizes advocacy activities as occurring at four levels: the individual, interpersonal, organizational and policy level. This framework is similar to patient engagement models which distinguish between micro-, meso- and macro-level activities.

2.2.1 Types of Patient Advocacy Activities

At an *individual* level, access to information and patient literacy (or the ability for patients to understand and apply health information) are of particular concern to patient advocates, and efforts are taken to equip patients with the information they need to participate in decisions about their health (Gilkey et al., 2008). The Internet now plays a significant role in facilitating individual-level advocacy, as websites devoted to informing patients about healthcare services and programs, diseases and treatments, and other patients’ lived experiences allow patients to become more involved in their own care. These resources are especially critical for those living with rare diseases who benefit from finding support groups and engaging in dialogues to reduce social exclusion. For patients with cystic fibrosis who cannot easily have in-person interactions with other CF patients (due to the risk of acquiring an infection), learning about other patients' experiences and belonging to a virtual community can be very meaningful.

At an *interpersonal* level, patient advocacy occurs through interactions when a patient's family members and friends provide advice and emotional support and help patients with treatment regimens. Caregivers and family members are not formally involved in advocating for patients with an organization, but are rather 'informal advocates' who care for a loved one or significant other (Galiatsatos, Gurley & Hale, 2017). In many cases, however, caregivers and family members choose to coalesce into broader networks to raise awareness about patient needs, and find comfort in joining others that can relate to their challenges (Gilkey, Earp & French, 2008).

In the US, and increasingly in Canada, a number of private companies providing interpersonal advocacy support services have surfaced. These companies typically employ former health professionals (such as nurses) who attend medical appointments, visit a patient's home, track prescription medications, and offer a range of other services (Walkinshaw, 2011; Alliance of Professional Health Advocates, 2018). A number of certification programs seeking to legitimize and professionalize the patient advocacy role have also emerged (Patient Advocacy Certification Board, 2018). Collectively, these efforts, though not endorsed or paid for by governments, may help to challenge physician-centric models of practice, equip patients with resources and supports, and reinforce the notion that patients are critical stakeholders in their own care. At the same time, questions persist about whether certification programs might "exclude those who have gained their expertise through lived experience rather than formal advocacy training" (Gilkey, Earp & French, 2008, p. 581).

At an *organizational* level, advocacy transpires within the context of organizations, professional associations and other communities that are responsible for “establishing a tone or culture that can support or discourage patient-centeredness, patient safety and patient involvement” (Gilkey, Earp & French, 2008, p. 20). Within a hospital setting, for example, patient advocates may help to develop models of patient-centered care and patient safety, act as patient navigators, and help patients and their families grieve (Clarke, Pederson & Santalucia, 2015).

Lastly, at a *policy* level, advocacy organizations and patient advocates work to influence policy makers and health system actors. This can involve lobbying for greater patient input in decisions, representing the needs of vulnerable patient populations, and advocating for improved access to services. These advocates reach beyond individual patients and aim to represent a broader patient constituency. Advocacy organizations typically perform multiple functions, including educating the public and providing patient services, but also work to educate decision makers about the needs of patients. Such organizations have been particularly important for improving the lives of patients with rare diseases. Cystic Fibrosis Canada (CFC) is an example of a not-for-profit corporation that invests in CF research, innovation and care. CFC has contributed greatly to the CF community by successfully advocating for accessible drug coverage and newborn screening tests, educating providers about CF, and helping to improve clinical guidelines (CFC, 2018A).

These policy efforts, as well as the efforts of patient advocates at an individual, interpersonal and organizational level, underscore the important roles that advocates can play in raising awareness about patient needs, improving patient outcomes and experiences, and facilitating patient-centered care. Implicit in all forms of advocacy is the desire to give patients a greater voice. To this end, the goals of patient engagement and patient advocacy closely align, suggesting the need for inquiry that aims to better understand the similarities and differences between these concepts. Phase one of this study was pursued for this very reason; to understand how ‘engagement’ and ‘advocacy’ are being defined and interpreted by different stakeholders, and to identify perceived similarities and differences between engagement and advocacy activities.

2.3 The Physician-Patient Relationship

The next sections will focus on physician-patient interactions and medical decision making.

Throughout history, the physician-patient relationship has been the subject of intense scrutiny and debate in academic circles. From Plato’s early writings about medical doctrine, to more contemporary research about the role of the Internet in healthcare, questions have persisted about how to respect patient autonomy, how to balance patients’ preferences and values with evidence-based medicine, and how to ensure patients and physicians build strong relationships founded on trust (King, 1954; Pellegrini, 2017). In recent decades, researchers have sought to address these questions by contrasting three paradigms of medical practice: paternalism, consumerism, and shared decision making. Paternalism was once the standard of practice in Western biomedicine, whereas the latter two paradigms have gained traction in the past three or four decades. An examination of these paradigms and how they shape physician-patient interactions follows.

2.3.1 Paradigms of Medical Practice

Historically, the physician-patient relationship in most nations could be characterized as one based on a paternalistic approach to the practice of medicine. Paternalism, which contains the root word ‘paternal’ meaning ‘like a father’, can be conceived as an intervention in which “an action is carried out intentionally on behalf of another…with the purpose of providing benefit and avoiding harm" (Cody, 2003, p. 288). Since the emergence of Western biomedicine, physicians have been largely viewed as having specialized knowledge and training, the result of having spent years in medical school; this knowledge was used to diagnose, treat and make medical decisions on behalf of patients (Murgic et al., 2015). This obligation resembled that between a parent and their child, whereby the physician (the parent) had a responsibility to use his or her knowledge to benefit the patient (the child) since they lacked knowledge and expertise. The ethical principles of beneficence and nonmaleficence are closely linked to the practice of paternalism, as is deontological ethics (or the moral duty that physicians have to act in their patients' best interests) (Beauchamp & Childress, 2001). Paternalism has also been related to the 'principal-agent' theory, which posits that the patient (the principal) appoints a provider (the agent) to make decisions on their behalf (Laugesen, Hassanein & Yuan, 2015). This theory suggests that physicians have better health information at their disposal than patients (or asymmetrical information), and are therefore better positioned to make medical decisions.

More recently, however, paternalism has been challenged by an opposing worldview of medical practice; consumerism. Consumerism itself can be defined in many ways, but is based on the idea that patients or the 'consumers' of healthcare services, should make decisions about their health and healthcare (Latimer, Roscamp & Papanikitas, 2017). The right to exercise patient autonomy is a key underpinning of a consumerist paradigm. While this proposition is not new, especially as it relates to privatized health systems where services are paid for out-of-pocket (such as the US), consumerism has gained traction in several nations seeking to strengthen the patient voice. Within a consumerist paradigm, physician-patient interactions are likened to commercial transactions in which consumers bring their values and expectations to the table and expect to be heard (Hartzband & Groopman, 2009). Consumerism assumes that patients prefer to be involved in medical decision making, and that patients will take their concerns or ‘business’ elsewhere if they are not satisfied with their care (by changing providers) (Chin, 2002).

Consumerism is similar to patient engagement in that it emphasizes empowering patients in decision making processes, but moves beyond engagement by promoting patient autonomy and downplaying the role of physicians (Latimer, Roscamp & Papanikitas, 2017). Although outright calls for autonomy in Canada are limited, there have been calls to mandate autonomous decision making in nations around the world. In the UK, for instance, some libertarians have proposed that patients should make decisions to protect themselves from providers who cannot be trusted (with incidents of malpractice in the UK being used to justify this position) (Davies & Elwyn, 2008; Coulter, 1999). Consumerism is also endorsed as replacing “the supremacy of professional judgement of clinical need with a discourse of preference and choice” (Newman & Vidler, 2006, p. 199). This paradigm of medical practice therefore differs from paternalism on several fronts.

For example, whereas paternalism focuses on the obligation that physicians have to make decisions in their patients’ best interests, consumerism stresses that patient autonomy ought to be prioritized. Patients receive information from their physician not only to be informed about their diagnosis and treatment options, but also to make medical decisions. Consumerism likewise presumes that questioning a physician’s expertise is within a patient’s prerogatives, and that trusting providers ought not be automatic. In fact, while medical paternalism emphasizes trust, consumerism is best regarded as emphasizing accountability (Beisecker & Beisecker, 1993).

Consumerism has not gone without its criticisms however. One significant criticism is that a physician’s primary role in a consumerist relationship amounts to supplying information and to allowing patients to direct their own care (Rodriguez-Osorio & Dominguez-Cherit, 2008). This is especially problematic when patients desire an intervention that is medically inferior to other options (i.e., when a patient's choice of treatment conflicts with evidence-based medicine) (Newman & Vidler, 2006). Closely related to this argument is the belief that consumerism instills fear in physicians about the consequences of failing to support patient autonomy (Chin, 2002). Physicians might therefore supply information about a patient's treatment options while withholding their professional opinions. This paradigm likewise assumes that the knowledge gap between patients and physicians has narrowed in recent decades, so much so that patients can decipher medical jargon, determine which interventions align with their preferred outcomes, and make rational autonomous decisions. Of course, failure to do so could lead to patients making poor decisions that jeopardize health outcomes.

Given the potential for consumerism to be more harmful than helpful, what then has given rise to this contrasting paradigm of medical practice in recent decades? According to Zeckhauser & Sommers (2013), one factor that has enabled consumerism is the Internet and the role the Internet has played in empowering patients. Whereas physicians were once the sole gatekeepers of health information, effectively occupying a monopoly over medical knowledge, the Internet has now shifted health information into the public domain. As a result, patients can now go online to learn more about their symptoms, health conditions, medications and treatment options. Although patients are perhaps more informed about their health and healthcare than ever before, however, access to the Internet does not necessarily mean that patients can differentiate between credible and unreliable websites, nor does it imply that patients know how to apply online information (see section 2.5 for discussion about the internet).

Others have emphasized that ‘medical humanism’ is likewise influencing the patient experience (Hartzband & Groopman, 2009). Medical humanism is described as a movement that seeks to “understand the patient as a person [while] accounting for each patient’s values, goals and preferences with respect to clinical decisions” (p. 554). This signifies a transition away from ‘reductionism’; the view that biological processes can be best understood, diagnosed and treated as isolated parts. In the US, for instance, medical schools are increasingly teaching students to consider patients’ values, and routinely teach and assess cultural competence.

Efforts to enhance chronic disease management are also changing how physicians and patients interact (Stavropolou, 2012). Given that many chronic diseases can be managed through adherence to medications and lifestyle behaviours, physicians now recognize the importance of involving patients and their caregivers in medical decision making. To this end, neither paradigm, paternalism nor consumerism, may be preferred by patients and physicians.

A third paradigm of medical practice, characterized by shared or collaborative decision making, is perhaps most suitable for respecting patient autonomy while simultaneously leveraging physicians’ expertise. Within this paradigm, clinical encounters are akin to a ‘meeting of experts’ where patients and physicians make shared decisions. This occurs when a physician provides information about a medical problem, helps the patient consider the risks and benefits of treatment options, and then works with the patient to select a final treatment (Elwyn et al., 2010; Légaré et al., 2017). Shared decision making has been closely related to the concepts of ‘patients as partners’ and patient-centered care. These concepts assume that patients have experiential knowledge that can complement the scientific knowledge of medical providers (Elwyn et al., 2010). The relationship between patients and physicians is thus characterized by ongoing dialogue and a more equal distribution of power (Godolphin, 2009).

Shared decision making is also associated with greater trust in physicians, access to information and to a patient's emotional readiness (Charles, Gafni & Whelan, 2004). In many ways, shared decision making is the middle-ground between paternalism and consumerism; this model seeks to ensure that patients are informed and can contribute to medical decision making when preferred (rather than requiring patients or physicians to make unilateral decisions). Despite differing views about how patients should be engaged in their own care, then, it is clear that physician-patient interactions are being fashioned where patients are expected to be more active and physicians less paternalistic (Bury & Taylor, 2008).

2.4 Preferred Roles in Medical Decision Making

In accordance with this paradigm shift, researchers have sought to identify patients' preferences for involvement in their own care. Within the context of medical decision making, preferences have been discussed in relation to "…the potential benefits, harms and costs of [different] management options" (Yepes-Nuñez et al., 2017, p. 21). Many studies have explored preferred roles in medical decision making and the clinical and demographic factors which influence these preferences. Systematic reviews have also documented the tools used to measure preferred roles, and the challenges associated with comparing studies using different methodologies (Hubbard, Kidd & Donaghy, 2008; Yu, Enkh-Amgalan & Zorigt, 2017). In particular, prior research uses cross-sectional surveys, surveys pre and post physician visits, observations of physician-patient interactions, and structured and semi-structured interviews to examine the preferences of patients with various health conditions. To facilitate these studies, patients have been asked to consider a range of hypothetical scenarios, to consider their own medical conditions, to choose between gambles and assured outcomes, and to assign weights to the importance of different decisions (Eiring et al., 2015; Hubbard, Kidd & Donaghy, 2008).

Surveys administered have also consisted of a variety of decision making scales that categorize patients into three, five or seven decisional categories, with most surveys being administered at a single point in time. The most widely recognized scales include the ‘Control Preferences Scale’ (CPS) (Degner, Sloan & Venkatesh, 1997) and the ‘Problem-Solving Decision-Making Scale’ (PSDM) (Deber et al., 1996). These scales have been validated and tested on multiple patient populations, and are focused on in this review. The PSDM was used in this dissertation to explore the preferences of members within the CF community.

The CPS is a five-item tool which asks patients to sort decision making roles (A to E) from most to least preferred using five ‘cards’ (Degner, Sloan & Venkatesh, 1997). Each card consists of a statement and cartoon that illustrates varying levels of physician and patient involvement in decisions. Other versions of the CPS also exist, including those which measure perceived, actual and preferred roles (or concordance), with researchers assigning patients to one of three roles: an active, autonomous, or patient-controlled role; (where the patient makes medical decisions on their own), a shared or collaborative role; (where the patient and doctor make medical decisions together), or a passive, delegated or physician-controlled role; (where the patient leaves medical decision making entirely to their doctor) (De las Cuevas & Peñate, 2016).

Similarly, the PSDM comprises six questions and divides participation into two dimensions; Problem Solving (PS) and Decision Making (DM) (Deber et al., 1996) (see chapter 3 for details). PS refers to elements that require factual knowledge about diagnoses and treatment options, whereas DM incorporates preferences for outcomes and the risks and benefits associated with different medical interventions. Preferred roles are measured using two vignettes, one involving chest pain (a control vignette) and the other involving the patient’s current health condition. For each vignette, patients are asked to respond and to indicate their preference on each of the six items. Patients are then grouped based on their mean scores for both dimensions, where *passive* patients favour handing over PS and DM tasks to their physicians, *shared* patients favour handing over or sharing PS tasks but sharing or keeping DM tasks, and *autonomous* patients wish to be involved in both tasks. These roles relate to different paradigms of medical practice, where passiveness is akin to paternalism, sharing is akin to shared decision making, and autonomous decision making is akin to consumerism.

In accordance with increasing calls for greater patient engagement in decisions, current discourse suggests that patients today not only wish to be informed about their health, but also wish to exercise some control over medical decision making. Yet, considerable heterogeneity exists amongst patients' preferences, and predicting which patients will prefer a passive, shared or autonomous role is no simple task. The majority of the preference literature has also focused heavily on cancer patients (Chewning et al., 2012). This emphasis has allowed for extensive systematic reviews to be published on cancer patients' preferences in the past twenty years, but also underscores the limitations of existing research. This reality was particularly evident when searching for studies on the preferences of patients with rare and chronic diseases like CF. To our knowledge, no studies to date have explored the preferences of CF patients, underscoring the need for expanded inquiry and raising questions about whether patients with rare diseases, who often require specialized care and supervision, have similar or different preferences than those with cancers and more common diseases.

The literature generally advises that most patients prefer to share medical decisions with their physician, and that only a small minority of patients prefer to be autonomous. Of the 115 cancer studies reviewed by Chewning et al. (2012), for instance, 63% indicated that the majority of patients preferred a shared role. Similarly, in Deber al's. (2007) research, hospital outpatients with chronic diseases who completed the PSDM were found as desiring a shared role (78% for their current health condition and 65% for the chest pain vignette), with only 1% of patients wishing to be autonomous for both vignettes. Michaelis et al's. (2017) work also identified most patients with a mental health illness as preferring shared decision making.

Furthermore, the literature indicates that preferences are moderated by sociodemographic characteristics. In general, sex, age, and education have all been documented as significantly predicting preferences in multiple studies (Spooner et al., 2017; Hubbard, Kidd & Donaghy, 2008). In a survey of over 2700 adults under the age of 45, younger patients were found as preferring more active decision making than older patients (Levinson et al., 2005; Janz et al., 2004). Highly-educated breast cancer patients and multiple sclerosis (MS) patients have also been found as desiring greater involvement in decisions than their less-educated counterparts (Degner, Sloan & Venkatesh, 1997; Wallberg et al., 2000; Giordano et al., 2008).

Ethnicity and preferred spoken language may likewise shape how patients and physicians interact. It is possible that non-English speakers might have difficulties communicating and understanding information, which could in turn limit patients to a passive role. Nies et al's. (2017) research of breast cancer patients found ethnicity to be significantly related to preferences, with Malay women being about three times more likely to prefer autonomous decision making than patients of Indian descent.

Evidence about the associations between other variables and preferred roles, including income, marital status, employment status and health status is far less conclusive. For example, whereas some studies have documented an association between declining health status in cancer patients and role preference (Butow et al., 1997; Hinds et al., 2000), other studies have found no such associations (Bruera et al., 2002). On the other hand, the type of health condition has been described as being a significant predictor throughout much of the literature. Many systematic reviews have noted that patients with lung cancer and colorectal cancer tend to be more passive than other cancer patients (Schmidt et al., 2016; Damm, Vogel & Prenzler, 2012; Tariman et al., 2010). Brom et al. (2014) further observed that patients with life-threatening conditions favour greater input in decisions than individuals with less advanced diseases. This implies that the stage and severity of a health condition could influence preferences (e.g., the more serious the condition, the more patients might want to be involved in decision making tasks).

Research has also alluded to non-demographic factors which encourage or deter patients from participating in medical decisions, including: trust or distrust in physicians, the ability to acquire information, the type of decision being made, and prior experiences with treatment regimens (Maclean et al., 2012; Say, Murtagh & Thomson, 2006). Several studies have reported how distrust in physicians can encourage patients to seek information from non-medical sources, and how past experiences with physicians' decisions can inform attitudes towards medical providers (e.g., when a physician recommends a treatment that results in an undesired health outcome) (Wrede-Sach et al., 2013). Not surprisingly, patients who have less trust in their physician are more likely to prefer input in decision making (Kraetschmer et al. 2004; Skirbekk et al., 2011).

Patients may likewise prioritize and place varying degrees of value on different factors when considering their medical options. Belinchón et al's. (2016) systematic review describes how patients with psoriasis attach greater importance to treatment processes (such as treatment location and duration) than to potential outcomes (such as the probability of occurrence of adverse side effects) when selecting a final treatment. This finding implies that convenience could be an important determinant of whether a patient prefers one treatment over another.

There is also evidence to suggest that physicians fail to understand patients’ preferences for information, often perceive patients as being more involved in their care than they actually are, and regularly fail to engage in adequate communication (Nies et al., 2017; Kehl et al., 2015; Spooner et al., 2017; Tariman et al., 2010). In a cross-sectional survey of 562 prostate cancer survivors, about 20% of men who completed the CPS were found as desiring a passive role (Cuypers et al., 2016). However, these men reported being dissatisfied with the information they received from their doctors, and assessed information received as being less helpful than more active patients. Research on women undergoing fertility treatments in Canada reported similar findings, with woman indicating their preference to be involved in treatment decisions but lacking information from physicians to do so meaningfully (Stewart et al., 2001). Interestingly, prior studies indicate that patients achieve “greater satisfaction and less decisional regret when decisions are made in concordance with their values and preferences” (Spooner et al., 2017, p. 44). Indeed, physician-controlled decisions are linked to poorer physician-patient communication and quality of care, heightening the need for improved interactions (Anderson et al., 2009).

Still, it is important to acknowledge that desire for information does not necessarily mean that patients prefer to be involved in making medical decisions, nor does it imply that patients have a preference for autonomous decision making. In fact, Manson (2010) argues that information-seeking should be considered independent of participation in decisions for two reasons. First, humans have an intrinsic longing to know things; learning about how a medical procedure works, for instance, may have no impact on whether a patient decides to move forward with the procedure. Second, patients may desire information from their physician in order to achieve “psychological preparedness”. Patients may want to know about the risks and benefits of treatment options to avoid being surprised and to be prepared for what might happen to their bodies. In this case, a physician's responsibility is to provide information in order to respect the person experiencing the medical intervention; the person with the most at stake (Holzmueller, Wu & Pronovost, 2012). Following this logic, one might suppose that every patient, regardless of their preferred roles, should be informed about their treatment options.

A physician willing to engage in open communication may therefore help to instill confidence that the physician is trustworthy, competent, and willing to answer questions. In an investigation carried out by Alexander et al. (2012), patients with higher perceived quality of interpersonal exchanges with their physician (measured by physician behaviours), were significantly more likely to become involved in decisions than those with lower perceived quality. Several studies have also demonstrated how a physician's interpersonal skills can influence patient satisfaction, and how involving patients in decisions can reduce decisional conflict and improve medication adherence (Van Stam et al, 2018; Von Arx & Kjeer, 2014; Puschner et al., 2016).

Chan et al's. (2016) work further illustrates how failure to involve patients in decision making is associated with mental health consequences. In their study, women who preferred more active roles in in-vitro fertilization planning who did not receive it reported higher levels of depression and regret (Chan et al., 2016). On the other hand, patients in more vulnerable circumstances might be poorly positioned to make difficult and potentially life-threatening decisions, justifying their dependence on the skills and knowledge of medical experts (Jabbour et al., 2018).

Hence, it would appear that the common thread between most studies is that patients desire to be informed about their treatment options but not for the purpose of making autonomous decisions. Nevertheless, it is difficult to predict what a patient's preferred role might be at any given time. This is especially true since current decision making tools differ, since patients' preferences might vary over time, and since little is known about the decision making preferences of those with rare and chronic diseases (making it difficult to generalize findings from one population to another). It is not surprising, then, that physicians have a difficult time identifying patient preferences. As a result, researchers have advocated for the use of decision aids to help improve physician-patient interactions (Holzmueller, Wu & Pronovost, 2012; Stacey, Légaré & Lewis, 2017; McAlpine et al., 2018). This is precisely where interpersonal support groups comprising caregivers and family members might likewise be of benefit; where close others support patients by contributing to patient decision making.

2.4.1 Caregivers and Family Members

Beyond assigning attention to the decision making preferences of patients, the literature offers insights about the roles that caregivers and family members play in shaping patients' healthcare experiences. Patients rely heavily on their caregivers and family members for emotional and practical support, and often depend on significant others to attend physician visits, to assist in managing chronic diseases, and to participate in a range of medical decisions (Pardon et al., 2010). As a result, caregivers and family members are often called '*informal advocates*'; unpaid individuals who advocate on behalf of patients by providing assistance with health-related needs and limitations (Galiatsatos, Gurley & Hale, 2017). In this dissertation, caregivers were defined as those (e.g., family members and significant others) who provide informal care to patients on regular basis, whereas family members were defined as those who are not actively involved in caregiving tasks but may still contribute to patient decision making (e.g., a distant parent or sibling) (see section 3.4.4 for further clarification).

Research indicates that active family involvement in physician visits is related to improved patient satisfaction, greater understanding of medical information, and to greater patient input in decision making (Wolff et al., 2015). Rini et al. (2011) investigated patients' perceptions of the roles that family members and close others played in deciding whether or not to undergo a colectomy. Regression analyses showed that patients with inflammatory bowel disease (IBD) were more inclined to have a colectomy when others persuaded them, and had lower decisional conflict when decisions were made with close others. Hobbs et al. (2015) also discovered language and ethnicity to be strongly associated with family involvement, with non-English speaking patients relying heavily on family members when making decisions.

Despite the benefits of having an interpersonal support network, however, much remains to be learned about whether the preferences of these stakeholders differ from patients. For example, it is possible that patients, caregivers and family members within a particular disease community might have differing values and preferred roles in medical decision making (Shin et al., 2013). This could create challenges for those seeking to develop clinical practice guidelines and for physicians seeking to practice patient- and family-centered care. It is also possible that experiences as a caregiver may inform one's personal preferences when interacting with a medical provider as a patient (i.e., through 'dual role' experiences). Measuring the preferences of patients, caregivers and family members was thus a major focus of this dissertation.

2.5 The Role of the Internet

In recent years, the amount of health information available online has increased considerably. It is estimated that over 54% of the global population (or more than 4 billion people worldwide), and over 85% of Canadians can now access the Internet (Internet Society, 2018; Statistics

Canada, 2019). While access to health information was once controlled and managed by the medical profession, health information is now readily available online. Patients can now use the Internet to acquire online medical advice, to learn about their symptoms and treatments, and to connect with others through virtual communities (Lu, Xu & Wallace, 2017). Caregivers and family members may also search websites to learn about a loved one's diagnosis and to gather information that can be used to support them. In a survey of 705 adults, 56% of respondents reported searching the Internet for health information on behalf of a relative or friend (Sadasivam et al., 2013).

Several studies have described why and how patients search for online health information. In particular, scholars have measured Internet-use by asking if patients have looked for material about a health condition within a given time frame, or have asked patients to rate the importance of information sources when making medical decisions (Couper et al., 2010). Demographic factors, as well as experiences with using the Internet have likewise been related to information seeking behaviours (Fiksdal, et al., 2014). Women are more likely to search for online health information than men, and Internet-users tend to be younger and more highly educated than non-Internet users altogether (Atkinson, Saperstein & Pleis, 2009; Wangberg et al., 2008).

With regards to medical decision making, frequent Internet users have additionally been found as being more likely to ask about treatment options (Hall, Bernhardt & Dodd, 2015; Levinson, Kao & Thisted, 2005; Holzmueller, Wu & Pronovost, 2012). In one study, 94% of pregnant women reported using the Internet to supplement information received from a doctor (Lagan, Sinclair & Kernohan, 2010). In another study, 75% of chronic disease patients reported using online health information when making treatment decisions (Fox, 2007).

Proponents of the Internet argue that patients today are equipped to make medical decisions, and that patients can use the Internet to hold physicians to account (e.g., by using online information to raise questions and concerns about diagnoses and treatment recommendations). Others suggest that the benefits of the Internet depend on the quality and credibility of information accessed. Those seeking to self-diagnose themselves using ‘Dr. Google’ may be unable to differentiate between low and high quality information and may have difficulties sifting through medical jargon. While some patients might access information from credible sources, others may view content from websites which are unreliable (e.g., when patients visit websites based on the order they appear in their search engine) (Peterson, Aslani & Williams, 2003). Indeed, research has shown that search engines can steer Internet-users toward medical interventions “that offer no benefit and that affect people’s decisions about the efficacy of medical treatments” (Pogacar et al., 2017, p. 8; White & Hassan, 2014).

Given the potential for patients to make unsafe medical decisions, then, Mathieu (2010) argues that physicians should direct patients to websites that are trusted by the medical profession. In a survey of adults’ navigation support preferences, medical providers were reported as being the most preferred mode of support for helping patients locate web-based information (Lee et al., 2017). As McMullan (2006) proposes, physicians can react to online information in three possible ways: 1) physicians can feel threatened and respond defensively, 2) physicians can work collaboratively with patients to review information, or 3) physicians can steer patients to reliable websites. Given that the Internet is becoming increasingly accessed and relied upon by patients and caregivers, it is these latter two responses (both of which emphasize effective communication) which could help to cultivate improved physician-patient relationships.

2.6 Health Literacy and eHealth Literacy

The need for physicians and patients to work together is heightened even further when one considers that patients may have difficulties understanding health information. Health information literacy (referred to as ‘health literacy’) is another important factor that could influence patients’ preferred roles in medical decision making. Health literacy is defined as the “degree to which individuals have the capacity to obtain, process, and understand basic health information to make appropriate health decisions” (Institute of Medicine, 2004). In contrast to electronic health literacy (discussed below), health literacy focuses on the information patients acquire from all sources, primarily during physician-patient interactions.

To date, a wealth of tools have been developed to measure literacy skills, including the newest vital sign (NVS), the test of functional health literacy in adults (TOFHLA), the rapid estimate of adult literacy in medicine (REALM), and other scales (Mackert et al., 2014). While these tools vary in their evaluation approach, they typically involve capturing survey responses on a Likert scale and categorizing health literacy as being either low/limited or high/adequate (Monkman et al., 2017). In a systematic review conducted by Berkman et al. (2011), limited health literacy was associated with “more hospitalizations, greater use of emergency services, lower receipt of mammography screening and influenza vaccine, poorer ability to demonstrate [appropriate medication use], poorer ability to interpret labels and health messages, and poorer overall health status and higher mortality rates” (p. 97). In contrast, higher health literacy is associated with better information gathering from the Internet and better self-management of chronic diseases (Czaja et al., 2013; Neter & Brainin, 2012). These findings are especially significant since 6 in 10 adults in Canada are not health literate (Government of Canada, 2018).

Research also suggests that the way physicians communicate may be perceived differently by patients with varying literacy levels, and that low health literacy may limit patient involvement in decision making (Ishikawa et al., 2009; Barton et al., 2014). In a cross-sectional survey of diabetes patients, DeWalt and colleagues (2007) found that those with low general literacy (below a grade 6 level) had less desire to participate in medical decisions. Patients with low health literacy are also more likely to ask for clarification, less likely to ask physicians' questions, and less likely to use medical terminology during physician visits (Katz et al., 2007).

Beyond this, the concept of electronic health information literacy (or eHealth literacy) has additionally gained traction as health information has proliferated online. According to Norman & Skinner (2006), eHealth literacy is "the ability to seek out, find, evaluate and appraise, integrate, and apply what is gained in electronic environments towards solving a health problem" (p. 2). The eHEALS, which combines six component literacies and includes eight questions, is one of the most widely used scales for eHealth literacy assessment (Monkman et al., 2017). Rather than measuring literacy objectively, the eHEALS asks respondents to assess their own skills at finding and using online sources of information (thus measuring 'perceived' or 'self-assessed' health information literacy). This measure was used in this dissertation for two reasons.

Firstly, those within the CF community depend heavily on the Internet and virtual communities for information about diagnoses, treatments and research. As mentioned previously, CF patients are frequent Internet-users and may have higher eHealth literacy than other patient groups. Secondly, it was hypothesized that lower eHealth literacy will be associated with a greater preference for a passive decision making role. Indeed, Goggins et al's. (2014) study of over 1200 cardiovascular patients found that patients with higher eHealth literacy desired greater involvement in PS and DM tasks than passive patients.

It is equally necessary to consider the information needs of caregivers and family members. Research indicates that patients and their caregivers search WebMD for information about treatments and health habits, and when physicians have not addressed their questions (Kanthawala et al., 2016). The literature also suggests that people overestimate their eHealth literacy skills, and that eHealth literacy is lower among older adults and those with limited education and computer skills (Canadian Council on Learning, 2008; Jensen et al., 2010; Choi & DiNitto, 2013). Knapp et al's. (2011) survey of parents whose children had special care needs found parents as experiencing difficulties differentiating between low and high quality websites. While patients often rely upon close others for support, then, those with low eHealth literacy might be limited in their ability to assist patients in decision making. In a European quality of life survey, caregivers who provided the most care were discovered to be between the ages of 50 to 64 (Efthymiou et al., 2017).

Recognizing the need to improve health literacy and eHealth literacy, researchers and decision makers have sought to implement programs designed to help patients gather, understand and apply information. Jacobs et al's. (2016) systematic review identified the potential for computer-based applications to improve health outcomes, health-related behaviours, and literacy scores. Simplifying medication labels has likewise been found to improve health literacy (Yin et al., 2008). While these initiatives have proven to be beneficial, however, few studies have explored how eHealth literacy might impact medical decision making preferences. The eHEALS was thus employed in this dissertation to contribute to this limited body of knowledge. Accounting for this type of literacy is especially pertinent since the consequences for using low quality, misleading online information can be significant.

2.7 Trust in Physicians

Trust in physicians (in all clinical settings) is further noted in the literature as shaping clinical interactions and decision making preferences. Within the physician-patient relationship, trust is best thought of as “the acceptance of a vulnerable situation in which the ‘truster’ (the patient) believes the ‘trustee’ (the physician) will care for their interest” (Hall et al., 2001, p. 615). Ideally, trust in the physician-patient relationship is developed over time as a physician exhibits their technical expertise and interpersonal skills. In emergency or dire circumstances, however, the decision to trust a physician might arise out of necessity rather than a voluntary act of compliance or based on prior interactions. At the same time, building trust is necessary for providing physicians with the latitude to make decisions for those who are vulnerable and dependent upon medical experts to remedy their health problems (Skirbekk et al., 2011). It is precisely this outlook that allowed paternalism to flourish as the dominant paradigm of medical practice in Canada for centuries.

The physician-patient relationship is also unique as it relates to the potential benefits and harms of clinical interventions. Given that patients are the ones receiving treatments and services, patients stand to gain and lose the most from choosing to trust (or not trust) in their physicians’ diagnosis and treatment recommendations (Hall et al., 2001). Closely linked to this reality is the emotional component of trust that can result in exaggerated expectations and feelings of anger and betrayal when expectations are not met. Such emotions might arise when a physician causes harm, or when patients feel unheard or challenged. Croker et al’s. (2013) research highlights well the associations between trust and interpersonal aspects of the physician-patient relationship; in their study, patients showed greater trust and confidence in their physicians when their problems were ‘taken seriously’. Still, the amount of trust required of patients might differ depending on the severity of the health condition. Not surprisingly, cancer patients typically report higher levels of trust in their physician than primary care patients (Hillen et al., 2011; Hall et al., 2002).

Furthermore, the development of a trusting relationship is facilitated by a range of institutional and personal factors, such as patient-centred approaches to the delivery of healthcare services, the way in which physicians communicate, and socio-demographic factors (Pearson & Raeke, 2000; Croker et al., 2013). The existence of trust is also associated with improved self-reported health outcomes, medication adherence, patient safety in primary care settings, patient recovery, and patient satisfaction (Skirbekk et al., 2011; Krot & Rudawska, 2016; Ricci-Cabello et al., 2016; Russell, 2005; LoCurto & Berg, 2016). Research further suggests that trust influences how patients respond when physicians make mistakes; with patients with higher levels of trust being more understanding of errors (White, Cohrs, & Göritz, 2011).

Given the importance of trust, a number of tools have been developed to measure this construct. The Trust in Physician Scale (TPS) developed by Anderson & Dedrick (1990) is perhaps the most widely consulted of these tools. The TPS has been used to investigate primary care patients, patients with malaria, rheumatic diseases and others health conditions, and was used in this dissertation (see section 3.4.4 for details). This scale comprises 11 items, where respondents' answers are collected on a five-point Likert scale. Scores are then utilized to categorize patients as having low, moderate, high or blind/perfect trust in their physician. Studies using the TPS have found that lower trust is associated with higher education, older age and poorer health outcomes (Marcinowicz et al., 2017; Freburger et al., 2003).

Existing research indicates that patients' willingness to participate in medical decisions is related to trust in physicians. Interviews of patients with type 2 diabetes, for instance, led researchers to conclude that those who trusted their physicians to act in their best interest were significantly more likely to pursue a shared decision making role (Montori, Gafni & Charles, 2006). Moreover, Kraetschmer et al. (2004) examined the relationship between trust and preferred roles in over 600 patients with breast cancer and prostate cancer (who completed the PSDM and TPS). As hypothesized by these scholars, the majority of 'autonomous patients' had low trust in their physician whereas most 'shared patients' had high trust.

As it relates to the CF community, it is possible that CF patients, caregivers, family members and patient advocates may have varying degrees of trust when compared to other patients and populations. Learning more about what conditions produce trust might therefore be used to improve clinical interactions, especially as efforts are taken to replace paternalism with a more patient-centric model of care. In addition, the relationships between trust and eHealth literacy have yet to be sufficiently explored in the literature, let alone amongst those from a rare disease community. This dissertation aims to contribute to this existing research.

CHAPTER 3

Methods

This dissertation employed a mixed methods research design that incorporated three phases of data collection: 1) qualitative key informant interviews, 2) a quantitative cross-sectional survey, and 3) qualitative follow-up interviews of a subgroup of survey participants (see figure 3.1). This chapter discusses the methods of each phase, including: objectives and research questions, study populations, and data collection and analysis techniques.

3.1 Research Design Overview

According to Creswell & Plano Clark (2011), mixed methods designs can be broadly characterized as being convergent, explanatory, exploratory, embedded, transformative or multiphase. To facilitate this study's objectives, elements from both a multiphase and explanatory design were leveraged. In a multiphase design, researchers use a pragmatic orientation to examine a topic of interest through an iteration of phases or studies. Each phase addresses different research questions that advance an overall objective. This design has several variants, but was once called the sandwich design since researchers would alternate "quantitative and qualitative methods across three phases" (Creswell & Plano Clark, 2011, p. 100).

In an explanatory design, researchers alternate between a 'post-positivist' and theoretical orientation to collect quantitative and qualitative data strands (with researchers using different assumptions for each strand) (Creswell & Plano Clark, 2011). To begin, quantitative data is collected to answer quantitative research questions and to test hypotheses. The data is then statistically analyzed, and analyses are used to identify which results warrant further explanation.

Qualitative research questions are subsequently developed and answered, and the researcher discusses to what extent the qualitative strand helped to explain quantitative results. Phases 2 and 3 of this study aligned closely with this methodology (i.e., a survey paired with interviews).

Figure 3.1 – Research Design

Phase 1: Key informant interviews (Qualitative)	**Phase 2:** Cross-sectional survey (Quantitative)	**Phase 3:** Follow-up interviews of survey participants (Qualitative)
- Semi-structured telephone interviews conducted with those involved in patient engagement and/or patient advocacy activities - Data used to better understand perceived similarities and differences between terms and activities	- Online survey conducted to investigate CF patients', caregivers', family members', and patient advocates' preferred roles in medical decision making - Data analyzed to determine which results warranted further explanation	- Qualitative interview questions developed to help to explain survey results - A subgroup of survey participants were interviewed - Data analyzed to identify common themes

3.2 Ethical Considerations

The entirety of this study received ethics approval from the University of Toronto (see appendix 4 for approval letter). The risks associated with this research were minimal. All participation was voluntary and consented to, and measures were taken to safeguard and de-identify the data.

3.3 Phase 1: Key Informant Interviews

3.3.1 Objectives and Research Questions

As noted in section 1.1, key informant interviews were conducted for the purpose of better understanding what 'patient engagement' and 'patient advocacy' mean (i.e., how these terms are being defined and interpreted). As evidenced by the plethora of literature on these topics, these terms continue to have multiple meanings, applications and similarities despite being commonly used by healthcare stakeholders. As a result, a more nuanced understanding of what these terms mean and how they might differ was required. This clarification was perceived as being important for two reasons. Firstly, it was necessary to gain a greater understanding of how ‘patient engagement’ and ‘patient advocacy’ are being defined in different settings. Secondly, it was hoped that interview responses would help clarify the perceived similarities and differences between terms, including how activities might differ depending on where they occur, who is involved, and for what purpose. Phase 1 of this dissertation was thus pursued independently of phases two and three; it considered micro-, meso- and macro-level contexts and addressed research questions 1 and 2:

1. How are ‘patient engagement’ and ‘patient advocacy’ defined and perceived by individuals who are involved in engagement and advocacy activities?
2. What are the perceived similarities and differences between engagement and advocacy activities?

3.3.2 Study Population and Data Collection

Telephone semi-structured interviews were conducted with key informants who were selected for their expertise in the fields of patient engagement and/or patient advocacy. This was achieved by leveraging existing partnerships with those who were knowledgeable about patient engagement or patient advocacy activities, some of whom had contributed to this research during its developmental stages. These partners were also asked if they were aware of others who could provide useful insights, and if they would be willing to forward them the study invitation. A snowball sampling technique was therefore employed until thematic saturation was reached. To be included in this phase, participants were expected to be English-speaking, to be residing in Canada, and to be currently involved in engagement and/or advocacy activities as part of a health organization or agency (e.g., a hospital, patient network, research group etc.).

Potential respondents were emailed with an invitation to participate and an attached consent form outlining the details of the study. Eleven out of twelve people emailed agreed to participate (for a 92% response rate). All informants were encouraged to contact the researcher with questions, and were asked to return the consent form prior to being interviewed. An interview guide was also developed, and notes were taken during the interview process. All interviews were audio recorded (lasting from 20 to 40 minutes), transferred to an encrypted hard drive, and transcribed by the researcher in Microsoft Word. Questions about definitions and roles used to describe and categorize engagement and advocacy activities, and how to distinguish between terms made up the majority of the interview content. To ensure clarity, these questions were piloted beforehand with three partners.

In total, 11 people were interviewed (counting those in the pilot study), including 6 informants from Ontario, 2 from Alberta, 1 from Nova Scotia, 1 from British Columbia and 1 from Quebec. Key informants included: four patient and family advisors/council members who volunteer on an organizational or hospital committee/board, one person who oversees a patient engagement department, one person who manages a company that specializes in patient- and family-centered care, one person who works with a patient and family network, a leader of a provincial SPOR unit, one person with a patient advisory and advocacy background, and two people with an extensive background in disease-specific patient advocacy.

3.3.3 Data Analysis

Interview transcripts were analyzed using thematic analysis, which is a method "for identifying, analyzing, and reporting patterns within qualitative data" (Braun & Clarke, 2006, p. 78). While researchers can employ various methodologies to identify themes, this study employed a simple qualitative descriptive approach (which assumes a "unidirectional relationship between meaning and experience") (Sandelowski, 2002) and followed Braun & Clarke's (2006) procedures. To begin, all transcripts were printed and reviewed several times. The researcher then highlighted relevant content and created initial codes within the margins of each transcript. After reviewing and coding each transcript separately, all transcripts were reviewed again (in one sitting) to assess the overall sense of consistency in coding. Once the data was coded and organized based on similarities and differences in content, the codes were re-examined to consider how themes fit together. For some interview questions, the frequencies of responses were tabulated.

To minimize the likelihood of misinterpreting the data, another PhD student also reviewed four transcripts separately to identify codes and themes. Once this student was finished, a meeting was arranged where the researcher and reviewer presented their interpretation of the data. After a discussion, the researcher elected to modify some of his codes until an agreement about themes was reached. Finally, headings which captured the essence of each theme were developed, and quotes were selected for reporting. All quotes were linked to a participant number (i.e., Participant 1 = P1). Given that most participants worked with multiple organizations and preferred not to be called a 'patient' or an 'advocate' (for reasons discussed in chapter 4), the term 'informant' was used during reporting. The following interview guide was used:

3.3.4 Interview Guide for Key Informant Interviews

Patient Engagement

1. What does the term ‘patient engagement’ mean to you? How would you define patient engagement?

2. Has the term patient engagement been defined and used differently by others in your organization and field?

- a) If so, can you please provide examples of these different perspectives?
- b) Do you agree or disagree with any of these perspectives?
- c) Is there one particular definition that you agree with or would tend to refer to?

3. What are the patient engagement activities that individuals in your organization engage in?

- a) Are there formal roles/titles that have been assigned to people based on these activities?
- b) What are the terms used to refer to these different roles and activities?
- c) Is there a consensus amongst your colleagues about these different roles?

4. What are the patient engagement activities that you currently engage in?

- a) Do you have a particular title?

Patient Advocacy

5. What does the term ‘patient advocacy’ mean to you? How would you define patient advocacy?

6. Has the term patient advocacy been defined and used differently by others in your organization and field?

- a) If so, can you please provide examples of these different perspectives?

b) Do you agree or disagree with any of these perspectives?

c) Is there one particular definition that you agree with or would tend to refer to?

Distinctions

7. Why do you think patient engagement and advocacy continue to have multiple meanings?

a) Is there any overlap or similarities between these terms?

8. It has been suggested that engagement and advocacy activities can be distinguished as taking place at a micro-level, a meso-level or a macro-level. Do you agree with this suggestion? Can you provide examples of how activities might vary across these different levels?

3.4 Phase 2: Cross-Sectional Survey

3.4.1 Objectives and Research Questions

In phase 2, an online cross-sectional survey was conducted to explore the medical decision making preferences of patients with cystic fibrosis (CF), regional and provincial advocates representing CF patients, and caregivers and family members of CF patients. This was made possible by working closely with Cystic Fibrosis Canada (CFC) to acquire access to these populations. While much of the current literature proposes that the Internet is changing how physicians and patients interact, and that healthcare users now desire greater input in their own care, much remains to be learned about the relationships between trust, eHealth literacy and preferred roles in medical decision making. This is especially the case for those affected by CF; a rare and hereditary chronic disease that is difficult to manage. To our knowledge, no study to date has investigated the decision making preferences of CF patients.

Adding to this reality is the fact that limited research exists on the decision making preferences of caregivers, family members and patient advocates. Accounting for the roles that these stakeholders wish to assume in their own care, though, is important for uncovering if differences exist between the preferences of patients and those seeking to represent patients' interests. Measuring preferences might also be used to enhance advocacy efforts and to ensure patient engagement models reflect what patients want and need. A survey was thus used to explore the relationships between: preferred roles in medical decision making, trust in the physician, self-assessed health information literacy, and demographics. Phase 2 addressed research questions 3-5;

3. Within the CF community (patients, caregivers, family members and patient advocates), what are their: preferred roles in medical decision making, level of trust in physicians, and self-assessed health information literacy?
4. Are there differences in preferred roles in medical decision making, trust in the physician, and self-assessed health information literacy by participant category (i.e., between patients, caregivers, family members and patient advocates)?
5. Are preferred roles in medical decision making associated with: trust in the physician, self-assessed health information literacy, and demographics characteristics?
    b) What are the differences in trust and eHealth literacy scores between participants who preferred a passive, shared or autonomous role?

Based on these questions, it was hypothesized that:

A) Very few CF patients, caregivers, family members and patient advocates will have a preference for an autonomous decision making role despite the increased availability of health information on the Internet

B) Patient advocates will have a greater preference for a shared and autonomous decision making role than others in the CF community

C) Preference for a passive decision making role will be associated with lower self-assessed health information literacy

D) Preferences for a passive and shared decision making role will be associated with greater trust in the physician and lower education

3.4.2 Study Population

Phases two and three of this research were made possible through a partnership developed with Cystic Fibrosis Canada (CFC). CFC is a national charitable organization that is committed to finding a cure for cystic fibrosis (CF) and improving the care that CF patients receive. According to CFC's (2018A) website, CF is a serious genetic disease that affects more than 4100 Canadians (nearly 60% of which are adults), with one in every 3600 children being born with this rare disease. Around 70,000 people worldwide have CF (Cystic Fibrosis Foundation, 2018).

CF causes various effects on the body that differ in severity from one person to another, but typically affects the lungs and digestive system (CFC, 2018B). As a result of the production of abnormally thick mucus that blocks tubes, ducts and passageways in these major organs, and results in ongoing respiratory infections and a decline in lung function over time, most people with CF die at a young age and have a life expectancy of 52 (CFC, 2018B). Ongoing infections paired with other CF-related complications makes it difficult for patients to fight off common illnesses, leading to lengthier recovery times, and heightening the need for specialized care and appropriate medical management (Quittner et al., 2014). While recent advances in treatment have improved quality of life, CF remains one of the most difficult chronic diseases to manage.

Those with CF must also receive their care in specialized CF clinics that offer multidisciplinary services (with teams consisting of a primary physician, nurses, respiratory specialists and other health professionals) (Gravelle et al., 2015). There are currently 42 of these clinics located in hospitals throughout Canada (CFC, 2018A). In the majority of provinces, children receive their care in a pediatric clinic and then transition to an adult clinic after turning 18 (Gravelle et al., 2015). In less-populated regions, however, both children and adults receive their care in the same CF clinic, allowing for patients transitioning from childhood to adulthood to be cared for by many of the same care providers (Gravelle et al., 2012; Gravelle et al., 2015). Most patients are assessed in a clinic every three to four months, with an increased frequency of appointments occurring for younger children and for those with more advanced diseases. For the purposes of this study, however, only CF patients above the age of 18 were asked to participate.

Furthermore, caregivers and family members may play a critical role in helping patients manage their health conditions, may attend physician visits, and are sometimes tasked with carrying out daily activities on behalf of their loved one (Gravelle et al., 2015). It was recommended by members of CFC’s research team that caregivers and family members be included in this study but be categorized separately. ‘Caregivers’ were defined as participants who provide informal unpaid care for a person with CF (e.g., family members and significant others). In contrast, a ‘family member’ or ‘non-caregiver’ included participants who did not provide care for a person with CF (e.g., individuals affiliated with CFC who, despite not living with a CF patient, may still contribute to patient decision making). In some instances, caregivers and family members may also advocate for patients by volunteering their time to raise awareness about CF.

Since its establishment in 1960, CFC has relied heavily on such volunteers to develop its strong advocacy presence and to influence policy makers (CFC, 2018A). As it stands, CFC has more than 100 regional and provincial advocates who perform numerous tasks, including (but not limited to): building relationships with government officials in order to increase support for CF programs and services, monitoring CF-related issues at a provincial and regional level, organizing advocacy events, and using social media platforms to raise awareness about CF. In general, those concerned with improving the lives of CF patients tend to be vocal and well-versed about the challenges that come with having this rare disease, and play a critical role in representing patients' needs and preferences.

Hence, upon working closely with members from CFC's research team, including the manager of government relations and advocacy, it was agreed upon that patients, caregivers, family members and patient advocates who are affiliated with CFC would be asked to complete the survey. According to a primary contact, most people become aware of CFC by word of mouth or after a family member has received a diagnosis. Individuals can then choose to be added to one of CFC's email lists for the purpose of receiving information about CF. It was these individuals, those who were interested in learning more about CF treatments and research (namely, Internet users), adults, English-speakers, and those living in Canada who were asked to participate. The sampling frame also included family members and caregivers of patients who were under 18.

3.4.3 Data Collection

Given the distribution of participants across Canada, it was determined that an online survey was the most feasible method for collecting data. Using Qualtrics survey software, an online survey was created and piloted with members from CFC's research team. Once the survey was finalized, an anonymous survey link was generated which allowed the link to be copied and pasted into an email format. Using an anonymous link ensured that participants completing the survey could not be identified unless they agreed to be contacted for a follow-up interview (i.e., unless participants provided their email address at the end of the survey). The researcher then worked with CFC to draft an email, which included a description of the study and the survey link, and to distribute this email to three email lists (see appendix 2). The first list consisted of 111 patients, caregivers and family members. The other two lists included 40 family members and caregivers, and 171 patient advocates. Members from CFC's research team sent out survey invitations and reminders to participate on the researcher's behalf. In total, 328 people were contacted, with 165 people responding (for a 51% response rate).

3.4.4 Survey Content

Upon clicking the survey link, participants were greeted with a consent page which outlined the details of the study (see appendix 1 for the consent form). The researcher's contact information was also provided, and a section was included describing the researcher's desire to conduct a follow-up interview. Each participant was asked to consent prior to beginning the survey by checking off a box which stated: "yes, I am above 18 and would like to participate in this study". After consenting, participants were forwarded to the online survey which consisted of four parts: 1) demographic questions, 2) questions used to measure preferred roles in medical decision making (the PSDM), 3) questions used to measure self-assessed health information literacy (the eHEALS), and 4) questions used to measure trust in the physician (the TPS). At the end of the survey, participants were asked if they were interested in a follow-up interview. Those who were interested were asked to provide their email address to allow the researcher to contact them.

Demographic Questions

The first part of the survey included eight questions which were designed to gather demographic information. Since CF patients, caregivers, family members and patient advocates were all sent the same survey link, question one sought to differentiate between who was completing the survey (referred to hereafter as the ‘category descriptor question’). This question was developed with the help of CFC. It was recommended that multiple response options be provided and that caregivers and family members be kept separate rather than grouping them together (e.g., since some family members might not assist CF patients on a regular basis but still provide input in patient decision making). Participants were thus asked to identify themselves by selecting one or more of the following descriptors:

- I am a person with cystic fibrosis (CF)
- I am a caregiver of a person with CF who is under the age of 18
- I am a caregiver of a person with CF who is 18 or above
- I am a family member of a person with CF, but I am not a caregiver
- I am an advocate that volunteers with Cystic Fibrosis Canada who has CF
- I am an advocate that volunteers with Cystic Fibrosis Canada who does not have CF

Participants were also asked to indicate their sex (Q2), year of birth (Q3), country of origin (Q4), preferred spoken language (Q5), highest level of education (Q6), employment status (Q7) and current health status (Q8). These questions can be found in appendix two.

Preferred Roles in Medical Decision Making (PSDM)

Preferred roles in medical decision making were measured using the PSDM (Deber et al., 1996). The PSDM has been tested and validated on multiple populations and continues to be used today. This scale comprises six questions and divides participation into two dimensions, *Problem Solving* (which is measured using questions 1 to 4 of the scale), and *Decision Making* (which is measured using questions 5 and 6). These questions were asked for two vignettes, one involving the participant's health condition and the other involving chest pain (allowing for comparison with other studies). The current health condition vignette read: "Please select the box which best describes how you feel about each of the below statements related to making decisions about cystic fibrosis (CF). If you do not have CF, please provide the name of your current health condition or a condition which you had in the past". The chest pain vignette read: "Suppose you had mild chest pains for three days and decided that you should visit your doctor about this".

At the end of this vignette, participants were asked if they had experience with this clinical situation and to select all that applied from the following list: 'I have had personal experience with it', 'I know of family members or close friends who have experienced it', 'I have read or heard about it', and 'I do not know much about it' (Deber et al., 1996).

Problem Solving (PS) questions included:

1. Who should determine (diagnose) what the likely causes of your symptoms are?
2. Who should determine what the treatment options are?
3. Who should determine what the risks and benefits for each treatment option are?
4. Who should determine how likely each of these risks and benefits are to happen?

Decision Making (DM) questions included:

5. Given the risks and benefits of these possible treatments, who should decide how acceptable those risks and benefits are for you?
6. Given all the information about risks and benefits of the possible treatments, who should decide which treatment option should be selected?

For each vignette, participants were asked to respond and to rate their responses on a five-point Likert scale (where answers included; 1 = ‘doctor alone’; 2 = ‘mostly the doctor’; 3 = ‘doctor and you equally’; 4 = ‘mostly you’; and 5 = ‘you alone’). To determine preferred roles, mean scores were computed separately for PS and DM tasks for each participant and placed into one of three categories: Hand over (mean score < 3), Share (mean score between 3 and 3.99), or Keep (mean score ≥ 4) (Deber et al., 1996).

Once PS and DM scores were classified, participants were categorized as being either passive, shared or autonomous (as illustrated in table 3.1). ***Passive*** participants wished to hand over PS and DM tasks to their physician, ***Shared*** participants wished to hand over PS tasks but share or keep DM tasks, and ***Autonomous*** participants wished to exercise some control over both dimensions (see table 5.1). Prior research conducted by Kraetschmer et al. (2004) suggested that it would be theoretically implausible for individuals to desire control over PS tasks but not DM tasks; indeed no participants were found as falling into that category.

Table 3.1 – PSDM classification categories

Responsibility for DM	Responsibility for PS		
	Hand Over (< 3)	Share (3 - 3.99)	Keep (≥ 4)
Hand Over (< 3)	Passive	Theoretically implausible	
Share (3 - 3.99)	Shared (Leaning Passive)	Shared (Equally)	Autonomous (Leaning Shared)
Keep (≥ 4)	Shared (Divide and Share)	Shared (Leaning Autonomous)	Autonomous (Consumerist)

<u>Self-Assessed Health Information Literacy (eHEALS)</u>

It was hypothesized that preference for a passive decision making role will be associated with lower self-assessed health information literacy. Self-assessed health information literacy was measured using Norman & Skinner's (2006) eHealth Literacy Scale or eHEALS. The eHEALS is an 8-item scale that was designed to measure perceived skills "at using information technology to acquire the knowledge needed to find, evaluate, and apply electronic health information when making healthcare decisions" (Norman & Skinner, 2006, p. 2). Responses are collected on a five-point Likert scale with answers ranging from 1 = 'strongly disagree' to 5 = 'strongly agree'. The eHEALS is scored out of 40, and participants are categorized as having low, moderate or high eHealth literacy; with scores ≤ 20 indicating ***low eHealth literacy***, scores between 21 and 30 indicating ***moderate eHealth literacy***, and scores ≥ 31 indicating ***high eHealth literacy***. The eight items are:

1. I know what health resources are available on the Internet
2. I know where to find helpful health resources on the Internet
3. I know how to find helpful health resources on the Internet
4. I know how to use the Internet to answer my health questions
5. I know how to use the health information I find on the Internet to help me
6. I have the skills I need to evaluate the health resources I find on the Internet
7. I can tell high-quality from low-quality health resources on the Internet
8. I feel confident in using information from the Internet to make health decisions

Trust in the Physician (TPS)

The fourth section of the survey measured trust in the physician by using Anderson & Dedrick's (1990) Trust in Physician Scale (TPS). The TPS assesses three dimensions, "physician dependability, confidence in physician knowledge and skills, and confidentiality and reliability of information received from the physician" (Anderson & Dedrick, 1990, p. 1092). This scale has been validated by numerous scholars (exhibiting strong internal reliability) and continues to be heavily used in research (Marcinowicz et al., 2017). It comprises 11 items where respondents' answers are collected on a five-point Likert scale (with answers ranging from 1 = 'strongly agree' to 5 = 'strongly disagree'). Mean scores are computed to allow participants to be grouped as having either ***low trust*** (mean score < 3), ***moderate trust*** (score averaging from 3 to 3.99), or ***high trust*** (mean score ≥ 4). To avoid response bias, questions 5, 7 and 11 are reversed before scoring. The TPS consists of the following items:

1. I doubt that my doctor really cares about me as a person.
2. My doctor is usually considerate of my needs and puts them first.

3. I trust my doctor so much I always try to follow his/her advice.
4. If my doctor tells me something is so, then it must be true.
5. I sometimes distrust my doctor's opinions and would like a second one.
6. I trust my doctor's judgments about my medical care.
7. I feel my doctor does not do everything he/she should about my medical care.
8. I trust my doctor to put my medical needs above all other considerations when treating my medical problems.
9. My doctor is well qualified to manage (diagnose and treat or make an appropriate referral for) medical problems like mine.
10. I trust my doctor to tell me if a mistake was made about my treatment.
11. I sometimes worry that my doctor may not keep the information we discuss totally private.

3.4.5 Data Coding and Missing Values

All survey results were collected through Qualtrics software and directly exported into SPSS (version 23). The data was then checked for errors by generating frequency tables for all survey items. Each participant was also assigned a unique identification number, and questions with missing values were addressed in several ways. For example, participants who did not provide their age were assigned missing age values (n=4). In the event that a single non-demographic response was missing in the data (e.g., when participants answered 7 out of 8 questions in the eHEALS), the researcher would calculate a mean score from the questions answered and input this score into the missing response. This did not affect whether participants were categorized as having low, moderate or high literacy. Overall, less than 5% of the dataset had missing values.

In addition, a handful of variables were collapsed into fewer categories. While participants could identify themselves by selecting multiple categorical descriptors (in question 1), participants were recatergorized into four groups: CF patients, caregivers (which included all caregivers), family members, and patient advocates (which included regional and provincial advocates with and without CF). The decision to reduce the number of groups was two-fold. First, some of the initial groups lacked sufficient numbers needed to conduct statistical tests. For instance, only 2 participants identified themselves as being an advocate who had CF. Secondly, despite having formal advocacy roles with CFC, many advocates elected to identify themselves as caregivers, or as being both an advocate and a caregiver (n= 9). This was identified by comparing the number of participants emailed (171 regional and provincial advocates and 70 to 80 caregivers), with the number of participants who identified as advocates (n=24) and caregivers (n=78). This was also confirmed by a contact from CFC who suggested that this finding was a common occurrence since “most advocates view themselves as being a caregiver first and foremost” and “that some caregivers eventually become advocates who volunteer with CFC”. The researcher was thus required to determine how to best categorize participants who selected both the patient advocate and caregiver categories.

As a result, it was decided that participants who identified with both roles would be coded as an ‘advocate’ since these participants formally volunteered with CFC. This ensured that results would not be double-counted for the same person, but also meant that some participants who self-identified as a caregiver were indeed advocates (who could not be identified and moved into the ‘advocate’ category due to the anonymous survey link). This limitation was mitigated by the fact that most participants had similar scores and preferences.

3.4.6 Data Analysis

Survey data was analyzed using a number of descriptive and inferential statistics, including Pearson and Spearman's correlations, cross-tabulations, chi-square and Fisher's exact tests, t-tests, and binary logistic regressions (see chapter 5 for results). The analysis process began by conducting univariate statistics in SPSS; the distribution and frequencies of demographic variables were generated and reported, as well as scores, means and standard deviations for the PSDM, eHEALS and TPS. Both dimensions of the PSDM were further examined to determine the proportion of participants who preferred passive, shared or autonomous roles in PS and DM tasks (for both the current health condition and chest pain vignettes).

Cross-tabulations were then computed to examine the frequencies of responses by participant category. Chi-square and Fisher's exact tests were used to compute measures of association between categorical variables. The Pearson chi-square statistic was reported for 2x2 contingency tables and Fisher's exact tests was reported for larger contingency tables and when assumptions were violated (i.e., when more than 20% of cells had expected frequencies of less than 5). Independent samples t-tests were also computed to explore the differences in mean trust scores and eHealth literacy scores between passive and shared participants. Cohen's d was reported to indicate the effect size, with Cohen's d < 0.2 indicating a small effect, Cohen's d around 0.5 indicating a medium effect, and Cohen's d > 0.8 indicating a large effect.

Finally, binary logistic regression models were generated to determine whether trust and eHealth literacy were significant predictors of preferred roles in medical decision making for both vignettes. This was made possible since no participants were found as preferring an autonomous role, meaning the dependent variable could be coded as dichotomous (i.e., passive or shared).

These regressions included the following features: degrees of freedom (df) (which considers the number of independently variable factors affecting a given system), the Wald statistic (which measures the relationship within or between data items), Nagelkerke R^2 square and Cox & Snell R^2 (which considers the strength of the relationship between variables), Exp(B) (which indicates the odds ratio) and standard error (Field, 2009). Prior to conducting these regressions, several assumptions were also considered (e.g., the inclusion of a dichotomous dependent variable, etc.).

3.5 Phase 3: Follow-up Interviews of Survey Participants

3.5.1 Objectives and Research Questions

In phase 3, follow-up interviews were conducted with a subgroup of patients, caregivers, family members and patient advocates who had completed the online survey. These interviews were used to gather additional information about research participants and to help to explain survey results. For example, upon conducting statistical tests, it was determined that most participants preferred a shared decision making role, that the majority of participants had high or moderate trust in their physician, and that self-assessed health information literacy was a significant predictor of preferred roles. These results were then used by the researcher to develop interview questions which provided insights about research topics 6-9:

6) Reasons for participants’ preferred roles and factors that influence these preferences

7) Sources of health information participants use when considering decisions about their health, and the amount of trust they have in these sources of information

8) When and why participants use online health information

9) Factors that influence trust in their physician and the physician-patient relationship

3.5.2 Study Population and Data Collection

Telephone semi-structured interviews were conducted with survey participants. This was accomplished by emailing individuals who had provided their email address at the end of the survey (in an optional response box) and by arranging a time to be interviewed. In total, 64 people were willing to participate in an interview, including 17 patients, 35 caregivers, 6 family members and 6 patient advocates. The goal was to capture responses from a reasonable number of participants until thematic saturation was reached. To determine which individuals would be interviewed, then, the researcher alphabetized email addresses in each group and selected addresses beginning from the letter ‘A’ and worked his way down the list. Those who were initially selected were then contacted and given time to reply to the researcher’s email. When individuals did not reply or no longer wanted to be interviewed (n=6), the researcher proceeded down the list. In total, 19 interviews were conducted with 7 CF patients, 6 caregivers, 3 family members and 3 patient advocates.

At the beginning of each interview, the researcher explained how responses would be used, asked participants if they had any questions or concerns, and acquired verbal consent to record the conversation. A semi-structured interview guide was developed to facilitate discussions, several prompts were developed, and the researcher took notes during each interview to aid his final analysis. All interviews were audio recorded (lasting from 25 to 40 minutes), transferred to an encrypted hard drive, and transcribed by the researcher in Microsoft Word.

3.5.3 Data Analysis

Interview transcripts were analyzed using thematic analysis (identical to the process conducted in phase 1) (described in section 3.3.1). All transcripts were reviewed several times, the data was organized based on similarities and differences in content, codes were re-examined to consider how themes fit together, and another PhD student reviewed five transcripts to identify themes. Once a consensus about common themes was reached, quotes were selected for reporting. The qualitative findings were reported separately in chapter 6 and are described in relation to the quantitative results in chapter 7. The following interview guide was used:

3.5.4 Interview Guide for Follow-up Interviews

1. Medical decision making can include any number of decisions that affect a person's health. These can include diagnosing a person's health condition based on their symptoms, determining treatment options, assessing the risks and benefits of treatment options, deciding which treatment(s) to pursue, and so on.

a) What are the types of decisions you *prefer* to be involved in?
b) What are the types of decisions you *do not* prefer to be involved in?
c) Does your preferred level of involvement depend on how serious the health problem is?
d) When are you most likely to leave medical decision making entirely to your doctor?

2. What sources of information do you turn to when you have questions about your health?

a) How much do you trust these sources of information?
b) What are the benefits of using these sources of information?

c) What are the drawbacks of using these sources of information?

d) When are you mostly likely to use the Internet (i.e., for what purpose)?

e) How much do you trust the information you receive from your doctor when compared to other sources of information (such as the Internet?)

f) Does the amount of trust you have in your doctor influence how much you wish to be involved in decisions about your health? If so, how?

3. What are the benefits of involving patients in medical decision making? What are the drawbacks of involving patients in medical decision making?

a) When do you think patients should *be less* involved in making medical decisions?

b) When do you think patients should *be more* involved in making medical decisions?

The next chapter presents findings from phase 1.

CHAPTER 4

Findings from Phase 1: Key Informant Interviews

This chapter presents themes that emerged from the key informant interviews conducted in phase one. As noted in section 3.3, eleven key informants (2 men and 9 women) who had expertise in the fields of patient engagement and/or patient advocacy were interviewed from across Canada, including: six people from Ontario, two from Alberta, one person from Nova Scotia, one person from British Columbia and one person from Quebec. These included: four patient and family advisors/council members who volunteer on an organizational or hospital committee/board, one person who oversees a patient engagement department, one person who manages a company that specializes in patient- and family-centered care, one person who works with a patient and family network, a leader of a provincial SPOR unit, one person with a patient advisory and advocacy background, and two people with a background in disease-specific patient advocacy.

A total of eight themes were identified: 1A) multiple definitions and activities, 2A) differences in preferred terminology, 3A) multiple roles and titles, 4A) patients as valuable contributors, 5A) patient advocates as promoting a specific agenda, 6A) attached connotations, 7A) individual and interpersonal advocacy, and 8A) multiple factors driving definitional differences.

4.1 Patient Engagement

4.1.1 - Theme 1A: Multiple definitions and activities

When informants were asked to define patient engagement, each informant described patient engagement as comprising several activities that can occur at different levels of the health system. Some participants provided specific examples of engagement activities, emphasizing that their past experiences had informed their perspective, whereas others articulated a much broader definition. In many cases, participants used the term ‘spectrum’ or ‘continuum’ to emphasize that engagement varies depending on where it occurs, who is involved, and how much involvement takes place. The majority of informants also equated patient engagement to meso-level activities, such as including patients in research activities and on hospital advisory boards. A lesser number of informants discussed engaging patients in their own care. Two quotes help capture this theme:

> *“Patient engagement can run the whole spectrum, from asking someone to complete a survey to having patients seated on different governing levels and positions within an organization, all the way up to the co-design and implementation of any initiative that has to do with research or clinical care” (P9).*

> *“…when we are talking about planning services or designing a healthcare system, we need to engage patients if we are going to be able to design services that are for people. Of course, we could also talk about engaging patients in their own care” (P8).*

Six informants discussed definitions or frameworks which had informed their understanding of patient engagement. Two informants alluded to Carman et al’s. (2013) framework, two others referred to the IAP2 spectrum, and two others referred to the Institute for Patient- and Family-Centred Care’s definition. Each person also pointed out that defining patient engagement is a difficult task, and that this term has multiple meanings, applications and is used interchangeably with a number of other terms.

> *“…what I do understand is that there are as many different nuances along the continuum as there are people. It is different things for different people” (P4).*

> *"There are certainly a lot of different terms that float around. You can have client-centered care, you can have person-centered care, you can have patient- and family-centered care, you can have patient partnership. These terms may or may not mean the same thing depending on the organization and the context they are being used in" (P9).*

When asked if patient engagement had been defined differently by others in their organization and field, informants discussed a range of activities that are associated with the term.

> *"…it's decision making, it's being a partner, being involved in research..." (P4).*

> *"…patient engagement can apply to research. I also hear it a lot in regards to quality improvement, I hear it a lot in regards to innovation and in regards to policy and system planning. I would say that you are hearing it more and more in regards to anything that is going to affect the patient experience" (P2).*

4.1.2 - Theme 2A: Differences in preferred terminology

Patient engagement was further described as being a term that is confusing and not preferred by informants.

> *"…the whole engagement thing is a relatively new term, and to be honest, I am not sure it's my favourite. To me it implies that someone is engaging the patient, which is different than collaboration where you sit down and talk about shared goals and…how we are going to work together. Patient engagement implies that there is someone with more power and influence, and if they decide to, they will try to engage patients" (P5).*

> *"A bunch of us don't particularly like the term, but it's the term de jour so we stick with it because it's what everybody seems to understand" (P1).*

Many informants went as far as to suggest that the term 'patient engagement' should be replaced altogether, and that replacing this term could help clarify patient expectations. Two informants suggested referring to patients as 'collaborators' and four participants recommended referring to patients as 'partners'. Other informants did not like the term 'partners', were hesitant to propose new terms since "they are frequently changing and evolving" (P3), and expressed that terminological differences were inevitable yet inconsequential so long as patients could provide input in decisions (to the extent they desired to be involved). Three informants said:

> *"Partnership would be better because it clearly defines what it is" (P11).*

> *"In research we are trying to use the terminology of 'patient partners'. I think there is consensus that if you say patient partners, then there is an understanding of that…" (P2).*

> *"I don't prefer any of these words like involvement, partnership and engagement. I think you have to define these terms and I don't think there is any standardization. This whole patient revolution is in different stages in different places and is rapidly changing" (P5).*

4.1.3 - Theme 3A: Multiple roles and titles

When asked about whether formal roles or titles had been assigned to patients based on their engagement activities, informants discussed a plethora of terms that were used by organizations to characterize their tasks. These roles and titles included: patient advisors, patient and family advisors, patient members, patient representatives, patient partners, patient collaborators, and patient leaders. It was made clear that these roles and titles are not standardized, and that depending on the context or organization, patients could be assigned several other names.

> *"It's not standardized, everybody you talk to will give you a different definition because I can say well, I'm a patient advisor and that's what I'm called right now…, but some[one] else can say, oh that's not what a patient advisor means at all" (P7).*

> *"…some people might refer to committee positions as patient representatives, but others might refer to them as patient members. One of our committees calls them public members even though they operate more like patient members in representing the views of patient groups" (P6).*

Of the several names mentioned, 'patient advisor' and 'patient partner' were the most commonly cited as being used across Canada (by informants from Ontario, Alberta and British Columbia). Patient advisors were described as having less decision making capacity than patient partners. One informant said:

> *"I volunteer as a patient advisor with a healthcare organization and I bring a patient and family perspective to that organization. I would say that in our discussions around our names, we have always come back to advisor because when it comes right down to it, the final decision is not ours. We feel that our role is to bring the patient perspective and to advise the organization. In another role that I am in, I am a patient partner, and in that role, I am far more actively doing the work" (P10).*

4.1.4 - Theme 4A: Patients as valuable contributors

All informants discussed the need to engage patients in a meaningful way and emphasized that patients are valuable contributors to the health system. Patient expertise was primarily discussed at a micro-level (i.e., in relation to patients' knowledge about their own health).

> ***"…****the reason to involve patients is because they are experts of their own health in a system that is often characterized by professional advantage" (P5).*

> *"…patients have expertise the same way that researchers and health professionals have expertise. Patients are experts of their own chronic conditions" (P8).*

Informants additionally mentioned how their personal experiences, which was often a negative experience as a patient or caregiver, prompted them to become involved in engagement or advocacy activities. Although personal experiences varied from one person to another, informants believed that their efforts could help others affected by an illness or disease. Three quotes help illustrate this theme:

> *"I think that the people who get involved are people who really want to make a difference so they are very committed to bringing their perspective" (P10).*

> *"Patient harm is common, [and] out of it, there are people who find the need to ensure that others don't experience harm. Patients make these decisions as a rebound for bad experiences…, they want to improve [care] for others. As long as people feel like they are making an impact, they will look to continue to contribute" (P5).*

> *"I think that people who get involved in sitting on a patient committee or advisory council or working on research projects…, I think the main reason they do it is for altruism. They want to make a difference for other people. They believe they have some kind of experience or knowledge based on their use of the healthcare system or living with a certain condition that can be of value" (P8).*

Two informants also pointed out that those who participate in engagement activities are not compensated for their time, meaning that each person is a volunteer. One person passionately expressed how this made her feel undervalued. She said: *"I do a lot of work for an [agency] and they are looking for my expertise so I should be an expert around the table and valued equally with the physicians and everyone else there. The difference is that everyone [is] being paid for their time. As a consultant, no one is paying me to sit there and pay for my parking and pay for my time so obviously they are not valuing me as an equal partner around the table*" *(P11).*

4.2 Patient Advocacy

4.2.1 - Theme 5A: Patient advocates as promoting a specific agenda

Unlike 'patient engagement', most informants described patient advocates as working for disease-specific advocacy organizations, being politically-motivated, and promoting a specific agenda or course of action(s) at a macro or policy level.

> *"Advocacy implies that there is a particular point of view that you believe is under-adopted that needs adoption. I tend to think of advocates at the policy level because if you have, you know, disease-based organizations, they do research, they develop policy papers, position papers, and they advocate for the people who have that disease" (P1).*

> *"I think for advocacy, I would have to have a particular issue that I was trying to put forward and that would be the main reason that I was involved…, because I wanted to advance my ideas" (P3).*

Informants also distinguished between patient engagement and patient advocacy activities by contrasting the objectives and intentions of patients and patient advocates. Those involved in engagement activities were described as being more impartial and receptive to an open dialogue, and less likely to be confrontational than advocates. In contrast, patient advocates were described as being difficult to work with and unwilling to consider different viewpoints. Two informants alluded to their personal interactions with advocates as being "unpleasant" or "frustrating at times", especially when advocates would "dominate discussions". Conversely, two informants with advocacy backgrounds provided more positive descriptions, with one person emphasizing the critical roles that advocates play "in representing vulnerable populations and fighting for improved health". To this end, those involved in patient engagement activities were more pessimistic when discussing advocacy activities than advocates themselves.

> *"I think advocacy is more about trying to represent patient interests to the best of one's ability, but in patient engagement…, we want their perspective to be as a partner in the healthcare system more broadly. We don't want [people] to be advocating in the strong sense, we want them to be providing a more impartial view" (P6).*

> *"An advisor technically does not push something because their goal is to work collaboratively… so you either push your position or you work collaboratively, but you can't do both at the same time. You will have people who are advocates for a particular disease, for instance, so it's a valuable role but it's quite a different role" (P1).*

4.2.2 - Theme 6A: Attached connotations

The term 'patient advocate' was further described as having attached connotations. Many informants believed that advocates were likely to be negatively perceived by others, that pharmaceutical companies had co-opted advocacy agencies, and that the meaning of the term 'advocacy' had changed over time (from positive to negative). Even one informant who had previously held advocacy positions shared this perspective (P4). In total, six out of the eleven participants spoke of attached connotations.

> *"It's not a term that is used in a positive way by people who are in the system" (P8).*

> *"I think what's happened with advocacy is that its credibility has been completely eroded by the funding from pharma. So when you say you are an advocate, yeah, for who? I just don't trust most people" (P4).*

Four informants also expressed that they would never call themselves an 'advocate' in fear of being mischaracterized or looked upon negatively by their peers.

> *"Once you say you're an advocate, it immediately puts people on the other side as looking at you as an adversary. We don't want to call ourselves advocates because advocates can be annoying and never give up on their position. If I said I was an advocate, people would be less receptive [and] I'm trying to look unthreatening" (P3).*

> *"...some of my colleagues sometimes refer to themselves as advocates as oppose to advisors, I virtually never do because in my experience out there, people sometimes feel that patient advocates have a position and a point of view that they are pushing. It's not a label that works well in all contexts" (P1).*

4.2.3 - Theme 7A: Individual and interpersonal advocacy

In addition to patient advocates being described as macro-level actors who promote a specific agenda, some participants discussed a different form of advocacy. More specifically, informants believed that advocacy can occur on a more personal level, whereby individuals seek to acquire the best possible care for themselves or for a loved one. A clear distinction was therefore made between advocacy at a macro-level that intends to benefit a broader population, and advocacy at a micro-level that intends to benefit a single person or family member.

> *"…to me, there is a world of a difference between people who are involved in their own care, and people who are active in the system in order to benefit others. The language is confusing but the activities are distinct" (P5).*

> *"Well, I think [you want] to make sure that the person you are a caregiver for is getting the best possible care. But there are caregivers who have become involved in trying to improve the health system and make change in healthcare as well…, but I think you separate those two roles" (P3).*

4.2.4 - Theme 8A: Multiple factors driving definitional differences

When informants were questioned about why they thought patient engagement and patient advocacy continue to have multiple meanings and applications, a number of different opinions were provided. Almost all informants had difficulty answering this question, with many suggesting that they were unaware why so many definitions existed. Yet, there was a general consensus that multiple factors have contributed to this issue, including different contexts, people's experiences, and the fact that patient engagement practices are still evolving in Canada.

> *"It probably just reflects the different experiences and knowledge that people bring with them when they [become] involved in activities. From a space of advocacy, for example, they might be coming from outside the health system so they bring that vocabulary and those concepts with them" (P6).*

> *"I think it [depends] on the context and organization, depending on what the need is. But more than anything, I think it's probably an evolution of what engagement can be and what it is. As we learn, we grow to understand that there are nuances" (P10).*

The final interview question sought to determine the usefulness of categorizing patient engagement and patient advocacy activities as occurring at different levels of interaction (i.e., at a micro, meso and macro level). The majority of informants found these distinctions to be helpful, but were quick to point out grouping activities can be difficult and that these categories might need to be modified depending on patients' activities.

> *"I think you have to make a distinction between something that benefits the individual and something that benefits the system. I think it can happen at all three levels, but the distinction isn't more about what level but who is the beneficiary" (P5).*

> *"I'm not sure that I would say micro, meso and macro, but if we look at the micro and say that's the individual level, then yes. At the meso level, I would say that that has to do with organizations, and at the macro level you are working more systemically at the policy level. So I think that you can define it in these ways, I would just caution that trying to put [activities] into tidy boxes means that there are things that don't fit" (P1).*

4.3 Summary of Interview Findings

Interviews of patient engagement and patient advocacy experts addressed research questions 1 and 2 of this dissertation and revealed eight themes. Patient engagement was primarily discussed as occurring at a meso-level, was inconsistently defined by key informants, and was mentioned as taking multiple forms (with engagement activities, roles and titles ranging from one context to another). In contrast, patient advocacy was commonly defined as occurring at a macro-level (e.g., through disease-specific advocacy organizations), and at a micro-level (e.g., when seeking the best possible care for a loved one).

Informants distinguished between patient engagement and patient advocacy activities by emphasizing perceived similarities and differences. Those involved in engagement activities were perceived as being impartial and willing to listen and to compromise, whereas patient advocates were perceived as being difficult to work with and as promoting a specific agenda. However, both engagement and advocacy were discussed as occurring at a micro-level and as being important for improving patients’ lives and experiences.

CHAPTER 5

Phase 2: Survey Results

In total, 165 people responded to the online survey, including: 46 CF patients, 78 caregivers, 17 family members and 24 regional and provincial advocates. This chapter is organized into two parts. Firstly, data is presented on participants' demographic characteristics, including: sex, age, year of birth, country of origin, preferred spoken language, education level, employment status, health status, and health condition. Next, data is presented on participants' preferred roles in medical decision making (PSDM) for both vignettes (chest pain and current health condition), self-assessed health information literacy (eHEALS), and trust in the physician (TPS). The relationships between these variables and several hypotheses are analyzed. Select tables and statistics are also provided to break down the data by participant category.

5.1 Descriptive Statistics

5.1.1 Participant Category

As noted in section 3.4.3, a total of 328 people were emailed the online survey, with 165 people responding (for a 51% response rate) (shown in Table 5.1). A descriptor question was used to distinguish between who was completing the survey. To reiterate, all participants were affiliated with CFC, were above 18, and were categorized into four groups: CF patients, caregivers (which included all caregivers), family members, and patient advocates (which included all regional and provincial advocates). Fourteen respondents held multiple roles, 9 of which identified as being both a caregiver and an advocate (discussed in section 3.4.5).

Table 5.1 – Distribution of Participants by Participant Category

Participant Category	Total (total percent)
CF patient	46 (27.9%)
Caregiver of a person with CF who is <18	65 (39.3%)
Caregiver of a person with CF who is ≥18	13 (7.9%)
Family member	17 (10.3%)
Advocate who has CF	2 (1.2%)
Advocate who does not have CF	22 (13.3)
Total	165 (100%)

5.1.2 Demographic Characteristics

Demographic characteristics are presented in Tables 5.2-5.6. The sample consisted of 45 males (27%) and 120 females (73%). The mean age of participants was 43.8 years, and the standard deviation (SD) was 14.3 years. Almost all CF patients were below the age of 45 (91.3%), which was expected given the life expectancy of those with CF. Four participants did not specify their year of birth, resulting in four missing age values.

All participants identified ‘English’ as their preferred spoken language and 93.3% of participants specified Canada as their country of origin. The remaining participants were originally from Armenia, Bangladesh, Bulgaria, Iran, Netherlands, the United Kingdom (n=3), and the United States (n=3). Education was measured using eight categories, but was reduced to five categories since all participants had completed some high school (see appendix 2). Overall, the sample was well-educated, with about 90% of participants having had completed some university, college, professional or graduate school. About two-thirds of participants were either self-employed or employed full time or part time (66.8%).

When asked to rate their current health status, 63% of participants rated their health as being 'excellent' or 'very good'. The sample consisted of 46 CF patients (27.9%), 42 participants who had a different health condition (25.4%), and 77 participants who did not specify their health condition (46.7%). However, decision making preferences were similar across these groups.

Table 5.2 – Distribution of Participants by Sex

Sex	CF Patient	Caregiver	Family Member	Advocate	Participant Total (total percent)
Male	15 (32.6%)	16 (20.5%)	7 (41.2%)	7 (29.2%)	45 (27%)
Female	31 (67.4%)	62 (79.5%)	10 (58.8%)	17 (70.8%)	120 (73%)
Total	46 (100%)	78 (100%)	17 (100%)	24 (100%)	165 (100%)

Table 5.3 – Distribution of Participants by Age

Age	CF Patient	Caregiver	Family Member	Advocate	Participant Total (total percent)
Missing	0 (0%)	3 (3.8%)	0 (0%)	1 (4.2%)	4 (2.4%)
18-25	10 (21.7%)	1 (1.3%)	0 (0%)	2 (8.3%)	13 (7.9%)
26-35	18 (39.1%)	20 (25.6%)	0 (0%)	1 (4.2%)	39 (23.7%)
36-45	14 (30.4%)	24 (30.8%)	1 (5.9%)	5 (20.8%)	44 (26.7%)
46-55	3 (6.5%)	24 (30.8%)	1 (5.9%)	2 (8.3%)	30 (18.1%)
56-65	0 (0%)	6 (7.7%)	10 (58.8%)	5 (20.8%)	21 (12.7%)
65+	1 (2.2%)	0 (0%)	5 (29.4%)	8 (33.3%)	14 (8.5%)
Total	46 (100%)	78 (100%)	17 (100%)	24 (100%)	165 (100%)

Table 5.4 – Distribution of Participants by Level of Education

Level of Education	CF Patient	Caregiver	Family Member	Advocate	Participant Total (total percent
Some high school or completed high school	8 (17.4%)	6 (7.7%)	2 (11.8%)	1 (4.2%)	17 (10.3%)
Some post high school (university, college or trade school)	11 (23.9%)	15 (19.2%)	4 (23.5%)	2 (8.3%)	32 (19.4%)
Completed university or college	21 (45.7%)	39 (50%)	1 (5.9%)	10 (41.7%)	71 (43%)
Some professional or graduate school	2 (4.3%)	4 (5.1%)	1 (5.9%)	0 (0%)	7 (4.2%)
Completed professional or graduate school	4 (8.7%)	14 (18%)	9 (52.9%)	11 (45.8%)	38 (23%)
Total	46 (100%)	78 (100%)	17 (100%)	24 (100%)	165 (100%)

Table 5.5 – Distribution of Participants by Employment Status

Employment Status	CF Patient	Caregiver	Family Member	Advocate	Participant Total (total percent
Employed full time (40+ hours/week)	15 (32.6%)	35 (44.9%)	4 (23.5%)	6 (25%)	60 (36.4%)
Employed part time (up to 39 hours/week)	9 (19.6%)	18 (23%)	2 (11.8%)	5 (20.8%)	34 (20.7%)
Not employed	2 (4.3%)	2 (2.6%)	1 (5.9%)	1 (4.2%)	6 (3.6%)
Retired	1 (2.2%)	1 (1.3%)	4 (23.5%)	8 (33.3%)	14 (8.5%)
Homemaker	1 (2.2%)	14 (17.9%)	3 (17.7%)	0 (0%)	18 (10.9%)
Student	7 (15.2%)	0 (0%)	0 (0%)	1 (4.2%)	8 (4.8%)
Self-employed	4 (8.7%)	7 (9%)	3 (17.6%)	2 (8.3%)	16 (9.7%)
Unable to work or temporarily sick	7 (15.2%)	1 (1.3%)	0 (0%)	1 (4.2%)	9 (5.4%)
Total	46 (100%)	78 (100%)	17 (100%)	24 (100%)	165 (100%)

*In Ontario, many employers now consider full time employment as comprising 30+ hours/week

Table 5.6 – Distribution of Participants by Current Health Status

Current Health Status	CF Patient	Caregiver	Family Member	Advocate	Participant Total (total percent
Excellent	3 (6.5%)	23 (29.5%)	7 (41.2%)	10 (41.7%)	43 (26.1%)
Very Good	13 (28.2%)	31 (39.7%)	6 (35.3%)	11 (45.8%)	61 (37%)
Good	14 (30.4%)	20 (25.6%)	3 (17.6%)	2 (8.3%)	39 (23.6%)
Fair	15 (32.7%)	3 (3.9%)	1 (5.9%)	1 (4.2%)	20 (12.1%)
Poor	1 (2.2%)	1 (1.3%)	0 (0%)	0 (0%)	2 (1.2%)
Total	46 (100%)	78 (100%)	17 (100%)	24 (100%)	165 (100%)

Current Health Condition

Those who completed the online survey were asked to specify their current health condition. It total, 46 participants (27.9%) had CF, 42 participants (25.5%) specified having a different health condition (most of which were chronic conditions), and 77 participants (46.6%) did not mention having a health condition (likely because they were in good health). Other health conditions mentioned included: asthma (n=5), coronary heart disease (n=4), respiratory problems (n=3), diabetes (n=3), pregnancy-related problems (n=3), thyroid problems (n=3), blastomycosis (n=2), depression (n=2), hypertension (n=2), intestinal issues (n=2), migraines (n=2), ulcerative colitis (n=2), allergies (n=1), osteoarthritis (n=1), prostate cancer (n=1), a brain tumour (n=1), kidney disease (n=1), epilepsy (n=1), obesity (n=1), gall bladder disease (n=1), and a hernia (n=1).

Correlations

Bivariate correlations were conducted to determine whether demographic variables were correlated. Given that the data consisted primarily of categorical variables, Spearman correlations were computed and are presented below (in table 5.7).

Table 5.7 – Correlation Matrix

	Participant category	Sex	Age	Level of education	Employment status	Current health status
Participant category						
Correlation coefficient	1					
Significance						
Sex						
Correlation coefficient	.003	1				
Significance	.972					
Age						
Correlation coefficient	.531**	-.044	1			
Significance	.000	.570				
Level of education						
Correlation coefficient	.293**	-.181*	.301**	1		
Significance	.000	.020	.000			
Employment status						
Correlation coefficient	-.025	.113	-.038	-.243**	1	
Significance	.751	.147	.631	.002		
Current health status						
Correlation coefficient	-.416**	-.051	-.072	-.133	.124	1
Significance	.000	.518	.360	.088	.112	

(Note: **= $p< 0.05$, *= $p< 0.001$)

5. 2 Preferred Roles in Medical Decision Making

Statistics on preferred roles in medical decision making were generated in SPSS. As noted in section 3.4.4, participants were categorized based on their PSDM scores for the current health condition and chest pain vignette. For both vignettes, ≈80% of participants desired a shared role.

5.2.1 Chest Pain Vignette

Table 5.8 presents preferred roles for the chest pain vignette. Over 90% of participants preferred to hand over PS tasks to a physician. This finding was consistent with prior research (Deber et al., 1996; Thakkar, 2018). In contrast, over 70% of participants indicated their preference to assume some control over DM tasks, such as determining the acceptability of treatment options and selecting a final treatment. More than 25% of the sample wished to *keep* DM (n=44). A sub-analysis revealed a weak correlation between DM scores and trust in the physician (r = -.300, p < 0.01). Participants who wished to keep control over DM tasks were also highly educated, with 16/44 participants having had completed professional or graduate school (or 42% of the sample).

After calculating mean scores for PS and DM tasks and placing participants into their preferred role categories, 81.2% of participants were identified as preferring a shared decision making role. This role was subdivided into ‘shared leaning passive’ (49.7%), ‘shared equally’ (4.9%), ‘divide and share’ (24.2%), and ‘shared leaning autonomous’ (2.4%). The remaining participants were classified as passive (18.8%), indicating their desire to hand over PS and DM to their physician. Not a single participant preferred to be autonomous (0%), and the distribution of preferences was similar across groups. Participant category was not related to preferred roles (for both vignettes).

Table 5.8 – Distribution of Preferences for the Chest Pain Vignette

Responsibility for DM	Responsibility for PS		
	Hand Over (< 3)	Share (3 - 3.99)	Keep (≥ 4)
Hand Over (< 3)	Passive n=31 (18.8%)	Theoretically implausible n=0 (0%)	
Share (3 - 3.99)	Shared (Leaning Passive) n=82 (49.7%)	Shared (Equally) n=8 (4.9%)	Autonomous (Leaning Shared) n=0 (0%)
Keep (≥ 4)	Shared (Divide and Share) n=40 (24.2%)	Shared (Leaning Autonomous) n=4 (2.4%)	Autonomous (Consumerist) n=0 (0%)

Table 5.9 – Distribution of Preferences for the Chest Pain Vignette by Participant Category

Preferred Roles	CF Patient	Caregiver	Family Member	Advocate	Participant Total (total percent)
Passive	9 (19.6%)	15 (19.2%)	2 (11.8%)	5 (20.8%)	31 (18.8%)
Shared	37 (80.4%)	63 (80.8%)	15 (88.2%)	19 (79.2%)	134 (81.2%)
Total	46 (100%)	78 (100%)	17 (100%)	24 (100%)	165 (100%)

To control for prior information about this vignette, participants were asked about how much experience they had with the chest pain situation. On balance, 47.3% (n=78) of participants indicated that they had 'personal experience with this vignette', 35.8% (n=59) indicated that they had 'family members or close friends who had experienced it', 12.1% (n=20) indicated that they had 'read or heard about it', and 12.7% (n=21) indicated that they 'did not know much about it'. The relationship between personal experience and preferred role was not statistically significant. However, there was a relationship between those who did not know much about the chest pain scenario and preferred role (Fisher's exact test; p = 0.03), with those who were unfamiliar with this situation being more likely to prefer a passive role. This finding was similar to previous research conducted by Kraetschmer et al. (2004).

5.2.2 Current Health Condition Vignette

Similarly, for the current health condition vignette, the majority of participants indicated that PS should be decided 'mostly by the doctor' or by 'the doctor and participant equally' (see table 5.10). Alike the chest pain vignette, not a single participant wished to assume control over PS tasks, and a large proportion of participants preferred to be involved in DM tasks. Nearly the same number of participants wished to *keep* DM (n=45 or 27.3%), although 8 more participants fell under the 'shared leaning autonomous' category for the current health condition vignette.

In total, 81.8% of participants preferred a shared role, 18.8% preferred to be passive, and 0% preferred to be autonomous. The type of health condition was not associated with preferred roles. This finding was almost identical to the distribution of preferences for the chest pain vignette. Given that CF is a serious disease, it is possible that members of the CF community could relate closely to the chest pain scenario; resulting in similarities in preferences between vignettes.

Furthermore, it was hypothesized (in hypothesis B) that patient advocates would be more likely to prefer a shared or autonomous decision making role than others participants. However, no participants preferred to be autonomous, meaning this hypothesis could not be tested. It was also observed that CF patients and advocates had similar preferences, and statistical tests revealed no significant relationship between preferred roles and participant category (chi-square; p = .702).

Table 5.10 – Distribution of Preferences for the Current Health Condition Vignette

Responsibility for DM	Responsibility for PS		
	Hand Over (< 3)	Share (3 - 3.99)	Keep (≥ 4)
Hand Over (< 3)	Passive n=30 (18.2%)	Theoretically Implausible n=0 (0%)	
Share (3 - 3.99)	Shared (Leaning Passive) n=81 (49.1%)	Shared (Equally) n=9 (5.4%)	Autonomous (Leaning Shared) n=0 (0%)
Keep (≥ 4)	Shared (Divide and Share) n=33 (20%)	Shared (Leaning Autonomous) n=12 (7.3%)	Autonomous (Consumerist) n=0 (0%)

Table 5.11 - Distribution of Preferences for the Health Condition Vignette by Participant Category

Preferred Roles	CF Patient	Caregiver	Family Member	Advocate	Participant Total (total percent)
Passive	6 (13%)	15 (19.2%)	4 (23.6%)	5 (20.8%)	30 (18.2%)
Shared	40 (87%)	63 (80.8%)	13 (76.4%)	19 (79.2%)	135 (81.8%)
Total	46 (100%)	78 (100%)	17 (100%)	24 (100%)	165 (100%)

5.3 Self-Assessed Health Information Literacy

Self-assessed health information literacy (hereafter eHealth literacy) was measured using the eHEALS and was scored out of 40 (with scores ≤ 20 indicating low eHealth literacy, scores between 21 and 30 indicating moderate eHealth literacy, and scores ≥ 31 indicating high eHealth literacy). The mean eHealth literacy score was 28.5, with a SD of 6.65. Total scores ranged from a minimum of 8 to a maximum of 40 (with seven people scoring perfect on the eHEALS). Nearly 90% of participants had moderate or high eHealth literacy, and only 10.9% had low eHealth literacy (none of which were family members) (see table 5.12).

EHealth literacy categories were correlated with preferred roles for both the chest pain vignette (Spearman's r = .221, p < .001) and current health condition vignette (Spearman's r = .250, p < .001). Trust scores and eHealth literacy scores (presented in section 5.21) were also correlated; as eHealth literacy went down, trust in the physician increased (Pearson R^2 = -.160, p = .041). While highest level of education was not significantly related to eHealth literacy, approximately half of those who completed some professional or graduate school had high literacy scores (see table 5.19).

Table 5.12 – Distribution of eHealth Literacy by Participant Category

eHealth Literacy categories	CF Patient	Caregiver	Family Member	Advocate	Participant Total (total percent)
Low literacy	3 (6.5%)	10 (12.8%)	0 (0%)	5 (20.8%)	18 (10.9%)
Moderate literacy	24 (52.2%)	36 (46.2%)	8 (47%)	10 (41.7%)	78 (47.3%)
High literacy	19 (41.3%)	32 (41%)	9 (53%)	9 (37.5%)	69 (41.8%)
Total	46 (100%)	78 (100%)	17 (100%)	24 (100%)	165 (100%)

5.4 Trust in the Physician

The Trust in Physician Scale (TPS) was used to measure and categorize trust (where participants were grouped as having either low trust (mean score < 3), moderate trust (score averaging from 3 to 3.99), or high trust (mean score ≥ 4)). The TPS comprised eleven items, for a minimum score of 11 and a maximum score of 55. The mean raw trust score was 41.2 and the SD was 6.7. Two participants did not complete the TPS and were excluded from analyses. More than 55% of the sample had moderate trust in their physician. Current health status was positively related to trust (Fisher’s exact test; p = .038), although trust was not significantly related to preferred roles.

Table 5.13 – Distribution of Trust by Participant Category

Trust categories	CF Patient	Caregiver	Family Member	Advocate	Participant Total (total percent)
Low trust	4 (8.7%)	7 (9.2%)	1 (5.9%)	3 (12.5%)	15 (9.2%)
Moderate trust	24 (52.2%)	40 (52.6%)	12 (70.6%)	14 (58.3%)	90 (55.2%)
High trust	18 (39.1%)	29 (38.2%)	4 (23.5%)	7 (29.2%)	58 (35.6%)
Total	46 (100%)	76 (100%)	17 (100%)	24 (100%)	163 (100%)

5.5 Comparing eHealth Literacy and Trust Scores between Passive/Shared Participants

As noted in sections 5.2-5.3, participants were identified as preferring passive or shared decision making roles for both vignettes. As a result, further analyses were generated to determine whether differences by role preference could be found in eHealth literacy and trust scores. Since no participants preferred to be autonomous, this category was removed from subsequent analyses. The next section presents the results of two independent sample t-tests used to compare means between groups (i.e., ‘passive’ and ‘shared’ participants).

Chest Pain

For the chest paint vignette, participants who preferred a shared decision making role scored higher on the eHEALS than those who preferred a passive role. This difference was statistically significant (t= -2.80, p= 0.006), and the effect size was medium (Cohen’s d= -0.560). The effect size was calculated manually for each t-test (using an effect size calculator). Similarly, passive participants had a significantly higher level of trust in their physician than shared participants (t= 2.86, p= 0.05, Cohen’s d = 0.587).

Table 5.14 – Chest Pain Vignette: eHealth Literacy by Participants’ Preferred Roles

	Categories	N	Mean	Std. Deviation	Std. Error Mean
eHealth Literacy	Passive	31	25.516	6.4698	1.1620
	Shared	134	29.154	6.5239	.5636

Equal variances assumed	F	Sig.	t	df	Sig. (2-tailed)	Mean Diff.	Std. Error Diff.	95% CI (Lower, Upper)
	.269	.605	-2.80	163	.006	-3.64	1.298	-6.201, -1.074

Table 5.15 – Chest Pain Vignette: Trust by Participants' Preferred Roles

	Categories	N	Mean	Std. Deviation	Std. Error Mean
Trust	Passive	30	44.2667	6.24739	1.14061
	Shared	133	40.4827	6.62425	.57440

Equal variances assumed	F	Sig.	t	df	Sig. (2-tailed)	Mean Diff.	Std. Error Diff.	95% CI (Lower, Upper)
	.093	.761	2.86	161	.005	3.79	1.33	1.166, 6.402

Current Health Condition

For the current health condition vignette, participants who preferred a shared decision making role had higher eHealth literacy than those who preferred a passive role. This difference was statistically significant (t= -2.89, p= 0.004), and the effect size was medium (Cohen's d= -0.553). In addition, there was a statistically significant difference in trust scores between passive and shared participants (t= 2.16, p= 0.032). Passive participants scored about five points higher on average on the TPS (Cohen's d= 0.448). Given the significance of these findings, binary logistic regressions were computed to determine predictors of preferred roles (presented in section 5.7).

Table 5.16 – Current Health Condition Vignette: eHealth Literacy by Participants' Preferred Roles

	Categories	N	Mean	Std. Deviation	Std. Error Mean
eHealth Literacy	Passive	30	25.367	7.3601	1.3438
	Shared	135	29.160	6.3041	.5426

Equal variances assumed	F	Sig.	t	df	Sig. (2-tailed)	Mean Diff.	Std. Error Diff.	95% CI (Lower, Upper)
	.704	.403	-2.89	163	.004	-3.79	1.313	-6.386, -1.200

Table 5.17 – Current Health Condition Vignette: Trust by Participants' Preferred Roles

	Categories	N	Mean	Std. Deviation	Std. Error Mean
Trust	Passive	29	43.5862	6.36125	1.18125
	Shared	134	40.6582	6.68135	.57718

Equal variances assumed	F	Sig.	t	df	Sig. (2-tailed)	Mean Diff.	Std. Error Diff.	95% CI (Lower, Upper)
	.033	.856	2.16	161	.032	2.93	1.356	.2748, 5.608

5.6 Select Demographic Comparisons

Several demographic variables were also analyzed to determine whether they were related to preferred roles, trust and eHealth literacy. While scores and responses differed across groups, most of these differences were not statistically significant. For example, only current health status and age were significantly related to trust, and no demographic variables were related to eHealth literacy and preferred roles. In general, trust scores decreased and eHealth literacy scores increased as level of education went up. Select demographic tables are presented below.

Table 5.18 – eHealth Literacy and Trust Scores by Education

Highest level of education	Self-assessed health information literacy (Mean ± SD)	Trust in physician (Mean ± SD)
Some high school	29.3 ± 11.6	48.7 ± 6.6
Completed high school	29 ± 6.0	40.5 ± 5.2
Some post high school (university, college or trade school)	28.6 ± 6.6	42.2 ± 7.9
Completed university or college	27.6 ± 6.9	41.5 ± 6.5
Some professional or graduate school	31.4 ± 4.0	39 ± 4.9
Completed professional or graduate school	29.1 ± 6.6	39.8 ± 6.6

Table 5.19 – eHealth Literacy and Trust Scores by Participant Sex

Sex	Self-assessed health information literacy (Mean ± SD)	Trust in physician (Mean ± SD)
Male	28 ± 6.4	40.7 ± 6.7
Female	28.6 ± 6.8	41.3 ± 6.7

Table 5.20 – Chest Pain Vignette: Education and Preferred Roles

	Preferred Roles		
Highest level of education	Passive	Shared	Total (total percent)
Some high school	2 (6.5%)	1 (0.8%)	3 (1.8%)
Completed high school	1 (3.2%)	13 (9.7%)	14 (8.5%)
Some post high school (university, college or trade school)	8 (25.8%)	24 (17.9%)	32 (19.4%)
Completed university or college	15 (48.4%)	56 (41.8%)	71 (43%)
Some professional or graduate school	0 (0%)	7 (5.2%)	7 (4.2%)
Completed professional or graduate school	5 (16.1%)	33 (24.6%)	38 (23%)
Total	31 (100%)	134 (100%)	165 (100%)

Table 5.21 – Current Health Condition Vignette: Education and Preferred Roles

	Preferred Roles		
Highest level of education	Passive	Shared	Total (total percent)
Some high school	1 (3.3%)	2 (1.5%)	3 (1.8%)
Completed high school	0 (0%)	14 (10.3%)	14 (8.5%)
Some post high school (university, college or trade school)	7 (23.3%)	25 (18.5%)	32 (19.4%)
Completed university or college	16 (53.3%)	55 (40.8%)	71 (43%)
Some professional or graduate school	0 (0%)	7 (5.2%)	7 (4.2%)
Completed professional or graduate school	6 (20%)	32 (23.7%)	38 (23%)
Total	30 (100%)	135 (100%)	165 (100%)

5.7 Binary Logistic Regression

Binary logistic regressions were conducted to determine whether eHealth literacy and trust were significant predictors of preferred roles in medical decision making. Since no participants were found as preferring an autonomous role, the dependent variable was coded as dichotomous (i.e., passive or shared). Before carrying out these regressions, several assumptions were considered. For example, the dependent variable was recoded, and a weak correlation was identified between trust and eHealth literacy (indicating that multicollinearity was not an issue). Two regressions are presented below; one for the chest pain vignette and one for the current health condition vignette.

5.7.1 Chest Pain Vignette

To begin, reference categories were selected in SPSS. 'High eHealth literacy' was selected as the reference category for self-assessed health information literacy, and 'high trust' was selected as the reference category for trust in the physician. A 'shared role' was selected as the reference category for medical decision making. The same categories were used for both vignettes.

Table 5.22 – Binary Logistic Regression: Reference Categories

		Frequency	Parameter coding	
			(1)	(2)
eHealth Literacy	Low literacy	18	1.000	.000
	Moderate literacy	77	.000	1.000
	High literacy	68	.000	.000
Trust in the Physician	Low trust	15	1.000	.000
	Moderate trust	90	.000	1.000
	High trust	58	.000	.000

The omnibus tests of model coefficients (presented in table 5.22) demonstrated the model of interest with the inclusion of predictors (by comparing the predictor/full model against a constant model). A non-significant finding was identified (Chi square = 12.690, p = .080), indicating a non-significant improvement in the model with the addition of predictors.

Table 5.23 – Omnibus Tests of Model Coefficients

Step	Chi-square	Degree of freedom	Significance
1	12.690	7	.080

R square values were found as ranging from 0.075 to 0.122. Given that these indices were far away from 1, it was concluded that the full model was limited in making predictions about preferred roles. This was supported by a classification table generated in SPSS which calculated the overall percentage of cases that are correctly classified by the model with predictors. The original percentage of 81.6% did not increase from the null model to the full model.

Table 5.24 – Model Summary

Step	-2 Log likelihood	Cox & Snell R Square	Nagelkerke R Square
1	141.968	0.075	0.122

Of the two variables entered in the regression, only eHealth literacy was a significant predictor of preferred roles in medical decision making (as shown in table 5.24). Specifically, participants with *moderate* eHealth literacy were .232 times (or about 23%) less likely to prefer a shared role than participants with *high* eHealth literacy (indicated by Exp(B)). Put differently, one would expect participants with higher eHealth literacy to prefer greater involvement in medical decision making than less literate participants. This finding aligned with the results of our samples t-tests.

Table 5.25 – Predictors of Preferred Roles for the Chest Pain Vignette

	B	S.E.	Wald	df	Sig.	Exp(B)
Trust in the Physician			1.447	2	.485	
Low trust	1.137	1.107	1.055	1	.304	3.116
Moderate trust	.361	.432	.696	1	.404	1.434
eHealth Literacy			7.458	2	.024	
Low literacy	-1.076	.752	2.048	1	.152	.341
Moderate literacy	-1.462	.536	7.449	1	**.006**	**.232**
Constant	2.015	.747	7.275	1	.007	7.504

Summary of Findings: Chest Pain Vignette

A binary logistic regression analysis was conducted to determine whether eHealth literacy and trust in the physician were significant predictors of preferred roles in medical decision making for the chest pain vignette. A test of the full/predictor model compared with a model without predictors was not statistically significant (p= .080). The predictive power of the model was also relatively weak with Cox and Snell's R square= 0.075 and Nagelkerke's R square= 0.122. Only eHealth literacy was significantly related to preferred roles. Specifically, those with moderate eHealth literacy were less likely to prefer a shared role than participants with high eHealth literacy (Exp(B) = 0.232, p = .006). While it was hypothesized that preference for a shared and passive decision making role will be associated with greater trust in the physician, our analysis did not confirm a relationship; we fail to reject the null hypothesis.

5.7.2 Current Health Condition Vignette

For the current health condition vignette, the same reference categories were utilized. However, unlike the chest pain vignette, the omnibus tests of model coefficients approached statistical significance and indicated a marginal improvement in the model with the addition of predictors (Chi square = 13.908, p = .053).

Table 5.26 – Omnibus Tests of Model Coefficients

Step	Chi-square	Degree of freedom	Significance
1	13.980	7	.053

R square values ranged from 0.082 to 0.135 (table 5.26). Given that these indices were far away from 1, it was concluded that the full model was limited in making predictions about preferred roles for the current health condition vignette. This was supported by a classification table which calculated the percentage of cases that are correctly classified by the addition of predictors. The original percentage of 82.2% did not increase from the null model to the full model.

Table 5.27 – Model Summary

Step	-2 Log likelihood	Cox & Snell R Square	Nagelkerke R Square
1	138.730	0.082	0.135

Similar to the chest pain vignette, only eHealth literacy was found to be a significant predictor of preferred roles in medical decision making (table 5.27). Specifically, those with low eHealth literacy were .171 times (or 17.1%) less likely to prefer a shared role than participants with high eHealth literacy. Likewise, those with moderate eHealth literacy were .270 times (or 27%) less likely to prefer a shared role than participants with high eHealth literacy.

Table 5.28 – Predictors of Preferred Roles for the Current Health Condition Vignette

	B	S.E.	Wald	df	Sig.	Exp(B)
Trust in the Physician			1.890	2	.389	
Low trust	-1.123	1.115	1.014	1	.314	3.074
Moderate trust	-.501	.442	1.283	1	.257	1.651
eHealth Literacy			7.503	2	.023	
Low literacy	-1.768	.712	6.170	1	**.013**	**.171**
Moderate literacy	-1.310	.547	5.733	1	**.017**	**.270**
Constant	-2.043	.759	7.249	1	.007	7.717

Summary of Findings: Current Health Condition

A binary logistic regression analysis was conducted to determine predictors of preferred roles in medical decision making for the current health condition vignette. Two predictor variables were included in the model. A test of the full model approached statistical significance (p= .053), although the predictive power of this model was relatively weak with Cox and Snell's R square = 0.082 and Nagelkerke's R square = 0.135.

Only eHealth literacy was significantly related to preferred roles. Specifically, those with low and moderate eHealth literacy were less likely to prefer shared decision making than participants with high eHealth literacy ($\text{Exp}(B)_{\text{low}}$ = .171, p= .013 and ($\text{Exp}(B)_{\text{moderate}}$ = .270, p= .017. Alike the chest pain vignette, preference for a shared and passive decision making role was not associated with greater trust in the physician. We therefore fail to reject the null hypothesis.

5.8 Summary of Survey Results

A quantitative survey of CF patients, caregivers, family members and patient advocates revealed that not a single participant preferred to be autonomous for both the chest pain and current health condition vignettes. Rather, about 80% of participants preferred sharing medical decisions with their physician. Most participants also had moderate to high eHealth literacy and trust.

It was hypothesized that patient advocates would be more likely to prefer shared or autonomous decision making roles than others participants. However, this hypothesis could not be tested. In addition, there were no significant differences between preferred roles for the current health condition and chest pain vignettes, and trust in the physician was not related to preferred roles. Only eHealth literacy was found to be a significant predictor.

The next chapter will discuss findings from the follow-up interviews conducted with a subgroup of survey participants. Chapter 7 will discuss the implications of survey and interview results.

CHAPTER 6

Findings from Phase 3: Follow-up Interviews

This chapter presents findings from the follow-up interviews conducted in phase 3. Follow-up interviews were conducted with 19 survey participants, including 7 CF patients, 6 caregivers, 3 family members and 3 patient advocates. A total of six themes were identified, including: 1B) participants' preferred roles and factors that influence them, 2B) benefits and drawbacks of involving patients in medical decision making, 3B) sources of health information used by participants, 4B) how participants assess online health information, 5B) factors that influence trust in medical providers, and 6B) prior caregiving experiences as influencing preferred roles.

6.1 - Theme 1B: Participants' preferred roles and factors that influence them

The online survey conducted in phase 2 found that no participants preferred an autonomous decision making role. Instead, most participants preferred to share medical decisions with their physician. This meant that participants wished to be involved in DM tasks (i.e., determining the acceptability of treatment options and selecting a final treatment), but wished to hand over or share PS tasks. This finding was supported by the qualitative follow-up interviews. For example, one caregiver said: "*I prefer to be given information about a diagnosis, condition and treatment options, and then I like to be involved in making a treatment decision based on the risks and benefits that are valuable to me*" *(Caregiver 3).* The majority of respondents also discussed their preference to leave diagnosing up to a medical provider. A lack of medical knowledge and expertise was commonly cited as deterring patients from attempting to diagnose their own health conditions. Two participants said:

> *“I prefer to be involved mainly in decisions around treatments, because I usually don’t feel qualified to diagnose. I [may] have questions around the diagnosis, but I usually don’t feel qualified to determine the diagnosis” (Family member 1).*

> *“I think patients don’t necessarily have the training or insights to be able to make and interpret the appropriate diagnosis” (Patient 5).*

When participants were asked about whether the seriousness of a health problem would shape their preferred level of involvement in medical decision making, several different responses were provided. Although some individuals discussed being more likely to leave decision making to a doctor or healthcare team for more serious problems and conditions, others talked about desiring greater input in their own care. Three participants also mentioned their willingness to hand over decision making tasks for routine interventions (i.e., decisions that involve minimal risk). Despite these differing responses, however, almost every participant underscored the need for medical providers to share medical information and to ensure that patients’ questions and concerns are being addressed (i.e., for physicians to communicate clearly and effectively when a serious health problem arises). One patient said: *“the more serious the health problem is, the more information I want. I want to be informed about what the doctor is thinking and how different courses of action will affect my health” (Patient 2).*

Furthermore, participants described when patients should be less involved in making medical decisions. A number of examples were provided for when patients should assume a passive role, including: when patients are indecisive, when patients have difficulties understanding medical information, when English is not a patient’s first language, and when patients lack competence. One caregiver suggested: “*For people who are very young or who have different cognitive* *abilities or challenges, I don't think it's fair to expect them to make decisions in those periods of time where their capacity is limited"* (Caregiver 6). Yet, participants also stressed that patients and their families should be involved in decisions when preferred, and that providers must always be willing to communicate information.

6.2 - Theme 2B: Benefits and drawbacks of involving patients in medical decision making

Participants mentioned several benefits and drawbacks of involving patients in medical decision making. Benefits included: empowering patients, allowing patients to voice their values, questions and concerns, improving trust between patients and medical providers, improving treatment and medication adherence, and allowing physicians to narrow down health problems faster. In many ways, these benefits resembled those discussed by key informants in phase one (with regards to the benefits of patient engagement).

> *"…I really think that it helps to establish trust and those long-term therapeutic relationships. I think patients are more likely to follow recommendations if they are involved in the process and if they get a chance to raise questions and concerns and to discuss alternative options. I think they are a lot more likely to actually follow through on the treatment" (Patient 5).*

> *"I think that involvement can probably have an impact on the reliability of someone taking their medications. I think the more you feel you are involved, the easier it is to cope with a more difficult situation" (Patient 2).*

> *"…the most important benefit is that the patients' values and priorities can be taken into account. Although clinicians have a better understanding of what the treatment options*

> *are, what the risks and benefits are and what the prognosis is, the individual has more information about what they value most and what is important to them"* (Caregiver 6).

In addition to mentioning the benefits of involving patients in medical decisions, a number of drawbacks were also talked about. Specific drawbacks comprised: burdening and overwhelming those in vulnerable circumstances, providing patients with too much decision making autonomy (to the point where patients make suboptimal decisions that jeopardize health outcomes), and asking patients who are indecisive to contribute to decisions. Two participants mentioned how some patients prefer for decisions to be made on their behalf.

> *"You can find yourself somewhat overwhelmed at times…, and that can just build anxiety. I think this is the reason why some patients would prefer for a doctor to make final decisions for them"* (Patient 1).

> *"If you are an indecisive person and you worry a lot, it might be better for someone to make decisions on your behalf like a spouse or a parent or your physician. Some people are just really indecisive. If you give them two options, they are going to spend time that maybe they do not have"* (Caregiver 1).

6.3 - Theme 3B: Sources of health information used by participants

The majority of survey participants were found as having moderate or high eHealth literacy. Follow-up interviews were therefore used to determine what sources of information participants turn to when they have questions about their health. Two main sources were mentioned: in-person and online. In-person sources included a range of healthcare providers (primarily physicians and nurses), and caregivers, family members and friends. Online sources included websites (with Google being the most frequently mentioned search engine), medical journals, articles, YouTube, and virtual communities. Of these sources of information, websites were the most commonly used by interview participants (mentioned by 14 of 19 people). Participants discussed using multiple websites to learn about health conditions, and five people discussed searching PubMed for studies about CF. CFC's website was additionally described as providing reliable information about the latest advancements in CF research and care (by four participants).

Three participants also talked about using Facebook or online support groups to learn about other patients' healthcare experiences. One patient mentioned: "*I have a lot of friends in the CF community, and there is a webpage where we post on and see who has experienced a [similar] problem or symptom. We are not there to diagnose anyone, but are there to provide counsel and to talk about medical issues. It's very helpful" (Patient 1).* Another patient advocate emphasized the importance of virtual communities, saying: "*It's a really frustrating thing that people who are going through something so life-consuming cannot be around to support each other, so the Internet is a valuable tool for people with CF" (Patient Advocate 1).*

In addition, participants provided many reasons for consulting the Internet, such as to: learn about a diagnosis, health condition and treatment options, acquire general health information, learn about drug reactions and medication side effects, identify treatment alternatives, and to search for information about a family member's health condition. Interestingly, not a single participant discussed using the Internet to diagnose themselves. Moreover, six individuals talked about consulting the Internet when experiencing new symptoms, mainly to determine the seriousness of their symptoms and whether or not to contact a medical/healthcare provider.

> *"If I'm waiting to see the doctor and I have symptoms that are unclear, I might be googling to try to understand what [the] symptoms [are]. I also use the Internet to understand the implications of a diagnosis and what it means, as well as different treatment options for a specific diagnosis, side effects of medications and potential complications" (Family member 1).*

> *"I don't consult the Internet for diagnoses, but if I don't know what's happening, I use the Internet to gauge how concerned I should be" (Caregiver 2).*

> *"…if I have a new symptom or if there is a different drug that they have given me, I will also go online… just to do a bit more research to see what information I can dig up. A lot of times, I will take that information with me and then go and talk about it with my physician" (Patient 7).*

Furthermore, as noted above, in-person sources of information used by participants included healthcare providers, family members and close friends. Most participants mentioned how they preferred to direct questions to their physician, and that their physician or healthcare team was their primary source of information. In contrast, family members and close friends were mostly looked to for general advice and support. As one CF patient said: *"I usually check with my doctor and healthcare professionals first because my CF clinic has a physician, physiotherapist, dietician and nurse; you know there is a whole conglomerate of people right there that you can ask questions to…" (Patient 4).*

6.4 - Theme 4B: Assessing online health information

Beyond talking about the sources of information participants turn to when they have questions about their health, about half of respondents also discussed how they assess the credibility and trustworthiness of online sources of information. In particular, participants mentioned looking for research links on a website, considering whether bias viewpoints were presented by an author or organization, and researching the publisher of an article (i.e., their credentials and affiliations).

> *"I tend to trust the links at the bottom of an article more than the actual article itself. I would be wary about any preconceived biases or biased viewpoints depending on where the information is coming from" (Patient advocate 1).*
>
> *"…for me, anything on the Internet that doesn't have sources cited is something that you should not really trust" (Patient 3).*
>
> *"I think it depends on the website. I think if I was second-guessing the website, I would probably google the actual publisher to learn more about who they are…" (Caregiver 3).*

The Internet was further described as offering several benefits. Many participants referred to the Internet as being accessible, providing a variety of data and information, and allowing patients and providers to share their experiences and knowledge. One participant said: *"…the Internet just expands my knowledge, gives me points of reference, and allows me to ask questions, to investigate further and to gather insights about treatments"* (Caregiver 1). However, online websites were likewise noted as being potentially misleading and as creating unnecessary anxiety by suggesting that patients are in fact sicker than they actually are. Two participants said:

> *"There is a ton of information that can mislead you, so I try to avoid looking online unless it's by a source that I know I can trust" (Patient 6).*

> *"...if you don't know how to navigate the* Internet *with a grain of salt, it can quickly become quick sand. You can easily go from having a mild headache to thinking that you are dying. I think people see what they read and think its iron clad regardless of the source (...). There is nothing that beats that one*-on-one *personal connection with a physician..." (Caregiver 5).*

6.5 - Theme 5B: Factors that influence trust in medical providers

Participants also talked about the importance of building trust in the doctor-patient relationship and described trust in medical providers as being influenced by several factors, such as: a physician's willingness to communicate information and to answer questions, how long a patient has known their provider, and prior outcomes and experiences related to a physician's decisions.

> *"I have been with the same doctor since I was 18 and he has never led me wrong. I think my level of trust comes from the fact that my doctor has always taken really good care of me. I just really feel that he has my best interest in mind..." (Patient 3).*

> *"When a provider is willing to communicate with you openly, it really helps to trust them more" (Family member 2).*

The amount of trust that patients have in their physician or medical provider was also mentioned as influencing their preferred level of involvement in medical decision making. Lower trust was related to patients being more actively involved in making decisions and changing their providers, whereas higher trust was discussed in relation to patients being more passive.

> *"I think if I was going to someone that I didn't trust, I would pull away from that person and be seeking help elsewhere. I would probably try to take things more into my own hands" (Patient advocate 2).*

> *"If it's a steady relationship where trust has been built, then I think there is definitely more flexibility in being able to trust your physician to make decisions for you…without you having to really look at the situation and say, I still need you to prove yourself. So absolutely, the level of trust [I have] in a physician would affect how much I want to be involved in decisions" (Caregiver 2).*

> *"…I want to be involved in any case, I think it's probably just an easier discussion when I have a high level of trust. I mean to be clear, if I didn't trust the provider, I would try to switch providers" (Family member 3).*

Three participants talked about how trust in medical providers might vary for different members of a care team. For instance, one patient said: *"…because CF is dealt with in a team approach, I can have different levels of trust for different people. So for example, a dietician might go for maternity leave and another might come on…, and I might have a different level of trust for that person based on how much they seem to know and understand" (Caregiver 4).* While this study measured trust in the physician, this finding suggests the need to consider how trust might vary for members of a multidisciplinary team.

6.6 - Theme 6B: Prior caregiving experiences as influencing preferred roles

The final theme that was identified pertained specifically to caregivers and family members. Six participants discussed how their past experiences in the health system, particularly when caring for and supporting a loved one, influenced their preferred level of involvement in their own care. Four participants discussed how a family member's negative experiences, poor outcomes, and poor interactions with physicians urged them to adopt a more active role in medical decision making (both for themselves and for their loved one).

> *"I have had a great deal of interaction with the medical system because my oldest son has CF. I guess it really taught me to be a little more discriminating in terms of quality of care, because I have seen some really good physicians and I have seen some not so good ones, with outcomes that have been quite impactful. So I am much more mindful of details and attentive when interacting with physicians" (Family member 3).*

> *"…my kids were diagnosed with CF when they were 2 and 4 years old. Back then, I would have answered these questions completely differently and put 100% trust in my doctor, which I did, until they misdiagnosed them and put them in jeopardy. It really caused me to speak up more, both for them and for myself" (Caregiver 3).*

> *"I guess obviously my opinions are coloured by my experiences with my daughter. I learned that sometimes the decisions doctors make are really poor and it made me want to be more involved" (Caregiver 4).*

CHAPTER 7

Discussion, Implications and Future Research

This chapter discusses the findings of this dissertation, the contributions and significance of each phase of research, and the implications of this work for different stakeholders. Recommendations for future research and inquiry are also provided.

7.1 Research Questions 1 and 2 - Key Informant Interviews

7.1.1 Patient Engagement

Interviews of patient engagement and patient advocacy experts addressed research questions 1 and 2, and provided insights about the various ways patient engagement and advocacy are defined. While these terms are regularly used in discussions about improving patients' healthcare experiences, they continue to have different meanings for different people and vary depending on where they occur (i.e., at an individual, organizational, research, or policy-level) (Aymé, Kole & Groft, 2008; Carman et al., 2013). Even key informants who participate in engagement or advocacy activities provided varying definitions, perspectives and opinions. This was especially the case with regards to patient engagement, as most informants defined engagement while conflating other concepts and terms, and while drawing on their own personal experiences within the health system. An analysis of interview transcripts thus revealed clear similarities with the existing patient engagement literature, which is described by Bellows et al. (2015) as consisting of blurry lines between concepts and lacking consistent messaging.

The majority of informants also equated patient engagement to meso- and macro-level activities (such as including patients on hospital advisory boards and in research activities), while less informants discussed engaging patients at a micro-level (such as involving patients in decisions about their own care). Not surprisingly, though, participants themselves acknowledged that patient engagement occurs along a continuum, varies from one context to another, and is an evolving practice that has only recently gained traction in Canada. This latter point may help to explain (in part) why roles and titles used to characterize patient engagement activities differ across organizations. To this end, the use of terminology meant to describe patients and their responsibilities should not be overlooked or trivialized. In fact, it was precisely differences in preferred terminologies that evoked passionate responses from key informants. For example, whereas some informants preferred to be called ‘patient partners’, others preferred to be called ‘patient advisors’ or ‘patient collaborators’. This finding was consistent with prior research (Gallivan et al., 2012), and likely reflects a broader concern with ensuring patients are being valued and meaningfully involved in decisions. The theme of ‘patients as valuable contributors’ seems to support this assertion.

It is also worth mentioning that efforts to involve patients in meso- and macro-level decisions reflects a relatively new approach to health system governance in Canada, and addresses a prolonged history of absence of the patient voice in positions of authority. Hence, to view patients as experts is to move towards a more patient-centered system and to recognize the value that healthcare users bring to the table. It is the opinion of this author that this recognition alone, namely, the belief that patients should be involved in organizational and research activities, could help to improve the quality of care that patients receive (i.e., by drawing attention to the challenges that patients face).

At a basic level, then, those seeking to leverage patient expertise must carefully consider their objectives, be transparent about what is expected of patients, and convey their intentions to relevant stakeholders (Bellows et al., 2015). This involves asking a number of questions during planning processes, including: how should patients be engaged, what type of engagement is required, and how can patients be empowered in decision making. Tending to these questions will not only help to account for patients' perspectives, but could also be used to achieve the meaningful engagement which all patients long for. This is especially critical given the altruistic intentions of those who become involved in engagement activities; many of which are unpaid volunteers. In contrast, "misunderstanding and misinterpretation of expectations and goals poses a barrier to achieving successful [engagement]" (Gallivan et al., 2012, p. 4).

Prior research also indicates that patient engagement is understood differently by patients, providers and administrative leaders, adding yet another layer of complexity and heightening the need for clear goals and expectations (Bellows et al., 2015). Given that patient engagement research and practice are evolving and inconsistently defined, reaching a consensus about when, where, and how patients should be involved in decision making is challenging. To this end, patient engagement experiences must be studied and shared, both within and across healthcare settings, for the purpose of learning about the supports, information, and resources that patients need to meaningfully contribute to decisions. The development of patient feedback mechanisms (via quantitative and qualitative measures) could additionally be used to systematize and standardize best practices.

7.1.2 Patient Advocacy

Unlike with the term ‘patient engagement’, patient advocacy was commonly defined by key informants. Advocates were described as working primarily for disease-specific advocacy organizations and as promoting a specific agenda or course of action(s). Yet, a distinction was made between advocacy that occurs at a macro-level that intends to benefit a broader population, and advocacy that occurs at a micro-level that intends to benefit a single person (i.e., individual and interpersonal advocacy). This distinction aligned with Gilkey et al’s. (2008) research, which describes advocacy as occurring at an individual, interpersonal, organizational or policy level (in section 2.2). At an interpersonal level, for instance, advocacy ensues when a patient’s family members provide advice, attend physician visits, or help patients with treatment regimens. Caregivers and family members are not formally involved in advocating for patients with an organization, but are rather ‘informal advocates’. At a macro or policy level, advocacy groups seek to educate policy makers about the needs of patient populations.

Beyond determining how patient advocacy was defined, phase one was also used to clarify the perceived similarities and differences between engagement and advocacy activities. Participants distinguished between activities by underscoring three perceived differences. Firstly, advocates were primarily discussed as functioning at a macro-level. On the other hand, patient engagement was mainly discussed as occurring at a meso or organizational-level. This suggests that the strategies used to promote and advance patient interests, as well as the decision makers that patients seek to influence, differ when engagement and advocacy activities take place.

Secondly, patient advocates were described as promoting a specific agenda. Some informants expressed how this made advocates difficult to work with and confrontational at times. In contrast, patients involved in engagement activities were perceived as being more impartial and more interested in working collaboratively with different stakeholders. This implies that the goals of engagement and advocacy activities vary, and that people's preferences and experiences may prompt them to become involved in some tasks over others. Future research should consider whether those who become involved in engagement and advocacy activities differ (e.g., demographically and in terms of their healthcare experiences).

Thirdly, the term 'patient advocacy' was mentioned as having a negative connotation. The fact that some participants opposed to being called an advocate in fear of appearing adversarial suggests that 'advocacy' is not a neutral term. On the other hand, the term 'patient engagement' was not negatively perceived by informants (although some informants favoured the term 'partnership' over engagement). This finding stresses the importance of terminology, specifically within the context of hindering or facilitating teamwork and shared decision making.

Beyond these differences, a few similarities were also identified between patient engagement and advocacy activities. For example, both engagement and advocacy were discussed as occurring at a micro-level when individuals seek to acquire care for themselves or for a loved one. Moreover, at a meso- and macro-level, those who participate in engagement or advocacy activities are tasked with representing the interests of a broader population (be it through a patient advisory role or when advocating for a specific disease community), must aim to identify patients' needs and preferences, and must carefully consider whether patients are being accurately represented in order to maximize their contributions.

7.2 Research Questions 3, 4 and 5 - Survey and Interview Findings

7.2.1 Preferred Roles in Medical Decision Making

A quantitative cross-sectional survey of CF patients, caregivers, family members and patient advocates was conducted to measure preferred roles in medical decision making. Qualitative follow-up interviews were also conducted with the goal of identifying themes which could help to explain survey results. It was hypothesized that very few participants in this study, despite being frequent Internet users and despite having reason to be heavily involved in their own care, would prefer an autonomous decision making role. Survey results aligned with this hypothesis; not a single participant preferred to be autonomous for both the chest pain and current health condition vignettes. Rather, about 80% of participants preferred sharing medical decisions for both vignettes. This finding is not surprising given that the sample was fairly well-educated and that most participants had moderate to high trust in their physician and eHealth literacy. This finding is also consistent with previous research on cancer patients and heterogeneous patient groups (Chewning et al., 2012; Michaelis et al., 2017).

Furthermore, it is likely that members of the CF community could relate closely to the chest pain scenario (perhaps due to the seriousness of CF), which could explain similarities in preferences between vignettes. About 47% of participants indicated that they had personal experience with the chest pain scenario. Nevertheless, our results suggest that participants see a clear division of labour between problem solving (PS) and decision making (DM) responsibility. In particular, over a quarter of participants preferred to keep control of DM tasks for both vignettes (i.e., to ‘divide and share’ or to ‘share [while] leaning autonomous’). A lesser proportion of participants (<20%) wished to hand over PS and DM tasks to their physician (or preferred a passive role).

Follow-up interviews supported this result; participants mentioned their desire to be informed and to be involved in making medical decisions. As one caregiver said: "*I prefer to be given information about a diagnosis, condition and treatment options, and then I like to be involved in making a treatment decision based on the risks and benefits that are valuable to me*". This finding aligned with prior studies, although a larger proportion of participants wished to keep control of DM tasks in this study than in previous research (Deber et al., 2007; Thakkar, 2018).

It was further hypothesized that patient advocates would be more likely to prefer shared or autonomous decision making roles than others participants. However, given that preferences were similar across groups, that participant category was not significantly related to preferred roles, and that not a single participant preferred to be autonomous, the null hypothesis could not be tested. In addition, this study did not find significant differences between preferred roles for the current health condition and chest pain vignettes, and trust in the physician and demographic variables were not related to preferred roles (e.g., level of education). This differed from other investigations which found demographic variables as significantly influencing preferences (Spooner et al., 2017). One possible reason for this difference is that factors related to medical decision making could depend heavily on the sample being investigated. Here, we examined members from the CF community (a sample which consisted of little variability), while other scholars have primarily studied cancer patients and outpatients (Goggins et al., 2014; Chewning et al., 2012). Yet, eHealth literacy was found to be a significant predictor of preferred roles in medical decision making (see section 7.2.3).

Interview findings underscored how prior experiences as a caregiver or family member may influence preferences. For instance, a caregiver may choose to become more involved in their own care after experiencing their loved one being mistreated or misdiagnosed. This finding was important since most of the existing literature has focused on how patients make decisions within the context of people close to them (Rini et al., 2011). In this study, however, participants discussed how the opposite was true as well; experiences with patients had an influence on caregivers' and family members' own decision making preferences.

Nevertheless, participants stressed that patients and their families should be informed about their care at all times, and that their physician or healthcare team was their preferred source of health information. Several benefits of involving patients in medical decision making were likewise mentioned, such as allowing patients to voice their values, questions and concerns and improving medication and treatment adherence. Participants also mentioned the potential drawbacks of involving patients in decisions, including overwhelming those in vulnerable circumstances and providing patients with too much decision making autonomy (to the point where patients make suboptimal decisions that jeopardize their health outcomes). It is precisely these factors which are cited in the literature as justifying medical paternalism (Cody, 2003).

In many ways, then, these findings challenge the belief that the Internet is being used as a substitute for professional medical advice and that consumer autonomy is the new preferred norm. To the contrary, our findings recommend that shared decision making remains the most suitable middle-ground between paternalism and consumerism.

7.2.2 Trust in the Physician

The Trust in Physician Scale (TPS) was used to measure trust in this study. Less than 10% of participants had low trust in their physician, and most participants had moderate trust (55%) or high trust (36%). While trust was not significantly associated with preferred roles in medical decision making, (likely because trust was relatively high overall), interview participants did mention how trust could influence their preferences. Specifically, those with greater trust in their physician would be more willing to hand over decisions, whereas those with lower trust would assume a greater role in their own care (as mentioned in theme 5B).

Moreover, trust was described as being influenced by a physician’s communication skills, willingness to answer questions, and by prior experiences and outcomes related to a provider’s decisions. These findings were not surprising. Prior research suggests that distrust in physicians encourages patients to seek information from non-medical sources (such as the Internet), and that past experiences with physician decisions can inform attitudes towards medical providers (e.g., when a physician recommends a treatment that results in an undesired health outcome) (Wrede-Sach et al., 2013). Trust was further discussed as being important for improving medication and treatment adherence. This finding is important, especially for patients with chronic diseases like CF who require ongoing treatment and supervision. In addition, literature underscores the importance of a physician’s transparency and communication skills for building strong physician-patient relationships (Alexander et al., 2012). While some scholars have speculated that trust in physicians is declining, with the Internet playing a significant role in this decline, our study, though focused on a single rare disease community, implies that this is not the case.

7.2.3 EHealth Literacy and Sources of Health Information

The majority of survey participants had moderate to high self-assessed health information literacy; eHealth literacy was found to be a significant predictor of preferred roles in medical decision making for both vignettes. As eHealth literacy decreased, preference for a shared decision making role decreased also (or put differently, one would expect patients with higher eHealth literacy to prefer greater involvement in decisions). This finding aligned with our initial hypothesis and indicates that members of the CF community are confident in their ability to find, evaluate and apply online information. This finding is plausible given that this population is spread out across Canada and relies heavily on websites and virtual communities (e.g., to learn about other patients' healthcare experiences). It is conceivable, then, that these individuals would gravitate toward shared decision making precisely because they understand the limitations of their own knowledge, yet desire to be involved in decision making processes (particularly, in DM tasks). Despite being frequent Internet-users, however, no participants preferred autonomous decision making. On the basis of this study and others, decision makers should be aware that eHealth literacy may influence preferences and that those with lower eHealth literacy may desire less involvement in medical decision making. However, this does not forego the need for physicians and medical providers to inform such patients. In fact, Durand et al's. (2014) research indicates that shared decision making interventions can significantly improve health outcomes for vulnerable and marginalized populations.

Furthermore, follow-up interviews revealed the sources of information participants turn to when they have questions about their health, and how participants assess online health information. Websites were typically used to search for information about a range of health-related topics, although participants preferred to direct questions to their physician when possible. These findings imply that the Internet is being used for informational purposes and not necessarily as tool for replacing medical judgment as most models of consumerism would imply.

7.2.4 Limitations

This research consists of five limitations. Firstly, a non-probability convenience sampling strategy was used to acquire access to survey participants. One disadvantage of this approach is the problem of selection bias (since those without access to the Internet could not participate). The sample also consisted of those who were relatively young. While this presents a limitation, it should be noted that participants were affiliated with CFC, highly-educated, and were perhaps most likely to prefer an autonomous decision making role. Yet, even these participants did not desire to be autonomous, implying that those missed might not want to be autonomous either.

Secondly, using a single cross-sectional survey limits the ability to make longitudinal inferences about whether preferences are changing over time. It is possible that some CF patients will evolve in their preferences as their health deteriorates and as the decisions they face become increasingly more meaningful for their quality of life. Similarly, it is likely that those who have known their physician for a longer period of time will have higher trust scores. Yet, no questions were incorporated in the survey about the duration of the physician-patient relationship. The researcher also overlooked how trust might vary for different providers (i.e., how those receiving care from a multidisciplinary team may have greater trust in some team members than others).

Thirdly, there was an uneven distribution of patients, caregivers, family members and patient advocates who participated in the online survey, and only 46 participants with CF. The sample size may therefore lack sufficient power to draw statistically significant conclusions and to make generalizations (e.g., the logistic regressions). Despite this limitation, however, it is important to acknowledge that the data was collected from a rare disease population and that CF patients under 18 were excluded from participating (justifying the inclusion of caregivers in this study).

Fourthly, a number of patient advocates identified themselves as being caregivers on the survey. This limited the ability to provide accurate numbers about the distribution of participants, but was mitigated by the fact that most participants preferred shared decision making roles and had moderate to high trust and eHealth literacy. While ‘participant category’ was not significantly associated with differences in scores and preferences, it is possible that those occupying multiple roles might formulate their preferences based on their various experiences. Indeed, participants alluded to how their negative experiences as a caregiver or family member encouraged them to become more involved in their own care.

Finally, this study occurred in Canada where there is coverage for medically necessary hospital and physician services. If this study had occurred in nations where services are paid for out-of-pocket, it is possible that patients would be less likely to trust their physician and more likely to prefer autonomous decision making. In fact, Huang et al’s. (2018) research proposes that “people living in healthcare-commodified countries are significantly less likely to trust doctors than people living in countries [where healthcare is publically-funded]” (p.6). The commodification of healthcare through consumerism, it is argued, might therefore incentivize patients to view physicians as market actors, resulting in a desire for greater patient control over decision making.

7.2.5 Implications for the Physician-Patient Relationship

Phases two and three of this research have implications for several stakeholders. With regards to the physician-patient relationship, our findings suggest that patients do not prefer autonomous decision making and that current discourse about patients preferred roles should be revisited. The majority of participants were found as preferring a shared role, yet about a quarter of participants preferred to be keep control of DM tasks for both vignettes. This proportion was larger than in previous studies, and indicates that patients may wish to be informed about their diagnosis and treatment options in order to make final treatment decisions. As such, it would appear that neither paternalism (where physician autonomy is prioritized) nor consumerism (where patient autonomy is prioritized) are suitable in the current era of shared decision making.

Beyond this, knowledge of patients' preferences is relevant for informing the development of clinical practice guidelines and for informing providers about what resources are needed to ensure patients' needs and preferences are being met (Yepes-Nuñez et al., 2017). Providers would additionally benefit from designing their approach to engaging patients by considering different eHealth literacy levels and the relationships between eHealth literacy and decision making. The literature recommends that these approaches cannot be generalized and that each individual patient may have different capabilities. According to Hawley et al. (2007), one strategy to identity preferences is to "develop a feedback loop where physicians inquire about patient's preferred roles at the beginning of clinical encounters and then assess level of satisfaction at the end" (p. 394).

It is important to acknowledge that involving patients in medical decision making does not mean that physicians must agree to every patient request. Rather, patients and their interpersonal support networks carry their own values and expertise, and should therefore be acknowledged during clinical encounters. This requires patients to be informed about their diagnosis, treatment options and the risks and benefits of different treatments. At the same time, physicians should supply information and advice based on their own judgement and medical expertise. Doing so could help to establish trust and two-way communication; both of which are essential for fostering good physician-patient relationships. This is consistent with the concept of patient-centered care, where patients and physicians work together as 'equal partners'.

7.2.6 Policy Implications

Policy makers and medical schools should consider whether physicians are equipped to provide patient-centered care. If shared decision making is intended to become the new paradigm of medical practice, then providers must be trained in its use (O’Malley et al., 2011). One way to facilitate this training is through medical schools and education programs which can provide physicians with the skills needed to communicate effectively (Davies & Elwyn, 2008). Decision aids could additionally be used to benefit patients seeking to participate in medical decisions. Such aids can supply balanced information about the possible outcomes of treatment options and can help patients consider what matters most to them (Holzmueller, Wu & Pronovost, 2012).

The fact that eHealth literacy was a significant predictor of preferred roles is also important for organizations like CFC who aim to produce resources and disseminate information about CF. This finding highlights the necessity for such agencies to produce materials and information that is accessible and evidence-based, and to equip patients to distinguish between low and high quality information. Given that those affected by CF are likely to consult the Internet to acquire health information, efforts must be taken to ensure that patients, caregivers and family members can understand and critically appraise the information they receive.

Lastly, researchers should explore the potential consequences related to promoting consumerism and patient autonomy. It is the opinion of this author that perpetuating a discourse that patients now desire greater autonomy in their own care, as a result of the Internet, will lead to more harm than good (e.g., patients making poor decisions based on unreliable online information). While some patients will be interested in playing a more active role in medical decision making than others, further exploration is needed about how to solicit patients’ preferences appropriately.

7.3 Conclusions and Future Research

Findings from phase one underscore the important roles that patients can play in improving other patients' health and healthcare experiences, but also underscores the need for clear terminology and future research (e.g., about best practices). This is especially the case with regards to patient engagement. To move forward on this front, some scholars have developed frameworks that distinguish between different levels of patient engagement (e.g., Carman et al., 2013). While some informants found these frameworks to be helpful, others were quick to point out that grouping activities into tidy boxes can be problematic. This research sheds light on how a common term used throughout the literature is still very much ambiguous; in short, patient engagement has a different meaning for different people. With regards to patient advocacy, many of the above challenges appear to be non-existent since a consensus about what advocacy means appears to exist. This research thus offers some conceptual clarity by describing the perceived similarities and differences between engagement and advocacy activities.

With regards to phases 2 and 3, to our knowledge, little research to date has examined the medical decision making preferences of those in rare disease communities like CF, let alone the preferences of caregivers, family members and patient advocates. Instead, most research has attempted to generalize findings from cancer patients and outpatients. This research found that most survey participants preferred to share medical decisions, and that eHealth literacy was related to preferred roles. This study might therefore be used to enhance patient engagement models and practices to better suit the eHealth literacy levels and preferences of patients.

Qualitative interviews helped to support and explain our survey findings by revealing themes that influence physician-patient relationships, the sources of information participants turn to when they have questions about their health, and how participants assess online health information. Many participants mentioned their preference to acquire information from their physician, and trust in the physician was found to be relatively high in this study.

Increased awareness about the factors that might hinder or facilitate patient-centered care is needed. More longitudinal research is also needed about how patient preferences in medical decision making might change over time, whether differences exist between preferred and actual roles (and if so, what might be done to bridge this gap), and about what interventions can be implemented in order to improve eHealth literacy. Researchers in general should consider eHealth literacy as a determinant of a range of health-related behaviours. For example, little is known about how eHealth literacy might impact trust in physicians and how online sources (e.g., websites) can be improved to allow healthcare-users to make better use of online information.

It is difficult to predict what a patient's preferred role might be at any given time. This is especially true since current decision making tools differ, since patient preferences might change over time, and since preferences might vary by health condition. It is not surprising, then, that physicians have a difficult time identifying preferences and that some researchers propose that preferences are not generalized. For these reasons, physicians would do well to assess each patient individually and to ask patients' questions about what information they require, what concerns they might have, and whether they prefer to be involved in medical decision making.

Appendices

Appendix Outline:

- *Appendix 1:* Phase 1 – Key Informant Interviews
- *Appendix 2:* Phase 2 – Quantitative Online Survey
- *Appendix 3:* Phase 3 – Qualitative Follow-up Interviews
- *Appendix 4:* Ethics Approval Letter
- *Appendix 5:* Letter of Support from Cystic Fibrosis Canada

*****Please note that the ethics protocol was amended and reapproved at a later date by the Office of Research Ethics at the University of Toronto. An amendment was pursued to reflect several changes made to the initial study protocol. Changes included (but were not limited to): renaming the thesis and expanding the study population to include CF caregivers and family members.

Appendix 1: Phase 1 – Key Informant Interviews

A) Informed Consent

<u>Study Title:</u> Preferences for Involvement in Medical Decision Making: Comparing Patients and Patient Advocates

<u>Overview:</u>

There is a growing need to identify what roles patients and patient advocates wish to play in making decisions about their own medical care. As part of Daniel Saliba's doctoral thesis, he will be circulating an online survey for patients with cystic fibrosis (CF), as well as patient advocates representing CF patients to complete. This survey will employ scales that will be used to measure preferred roles in medical decision making, trust in physicians and self-assessed health information literacy. Recognizing that 'patient engagement' and 'advocacy' can have different meanings, and that advocates often engage in various activities, his study will begin by attempting to clarify these terms and by distinguishing between who is completing the survey.

Purpose and Participation

Given your expertise and knowledge about patient engagement and advocacy, you are being asked to participate in a 20 to 30 minute interview. This interview may be conducted in-person or by phone depending on your preference. More specifically, this will involve interviewing you about the different ways that organizations and individuals advocate for patients, and the various patient engagement activities that individuals engage in.

The goal is to help remedy the lack of clarity that currently exists in the literature pertaining to how ‘patient engagement’ and ‘patient advocacy’ have been defined. If possible, interview results will then be used to develop a typology (that is, a way to categorize the engagement and advocacy activities that people engage in).

Interviews will be audio recorded and transferred to an encrypted hard drive within 24 hours of being taped. The results will also be de-identified (not linked to you) unless you would prefer otherwise. Once the data is gathered and analyzed, a question will be incorporated at the beginning of the survey that will ask participants to choose which statement (from a prespecified list based on the results) best describes their activities. This will provide a framework that will guide the content and wording of the survey, and allow for a more nuanced understanding of how preferred roles in medical decision making might vary across a continuum (i.e., based on people’s activities). As such, your contribution would be greatly appreciated. There are no risks or benefits associated with taking part in interviews.

You may also choose to withdraw your responses at any time. You can ask to have your responses removed by emailing the researcher or calling him directly. The researcher will then shred all interview transcripts. No reason for choosing to withdraw is needed.

*If you require more information, please contact Daniel Saliba using the information below.

Statement of Consent: I have read the above information and understand how my contribution will be used. I consent to take participate in an interview.

Signature__________________ Date __________________

B) Interview Guide for Key Informant Interviews

Patient Engagement

1. What does the term ‘patient engagement’ mean to you? How would you define patient engagement?

2. Has the term patient engagement been defined and used differently by others in your organization and field?

a) If so, can you please provide examples of these different perspectives?
b) Do you agree or disagree with any of these perspectives?
c) Is there one particular definition that you agree with or would tend to refer to?

3. What are the patient engagement activities that individuals in your organization engage in?

a) Are there formal roles/titles that have been assigned to people based on these activities?
b) What are the terms used to refer to these different roles and activities?
c) Is there a consensus amongst your colleagues about these different roles?

4. What are the patient engagement activities that you currently engage in?

a) Do you have a particular title?

Patient Advocacy

5. What does the term ‘patient advocacy’ mean to you? How would you define patient advocacy?

6. Has the term patient advocacy been defined and used differently by others in your organization and field?

a) If so, can you please provide examples of these different perspectives?
b) Do you agree or disagree with any of these perspectives?
c) Is there one particular definition that you agree with or would tend to refer to?

Distinctions

7. Why do you think patient engagement and advocacy continue to have multiple meanings?

a) Is there any overlap or similarities between these terms?

8. It has been suggested that engagement and advocacy activities can be distinguished as taking place at a micro-level, a meso-level or a macro-level. Do you agree with this suggestion? Can you provide examples of how activities might vary across these different levels?

Appendix 2: Quantitative Online Survey

Project Overview:

There is a growing need to identify what roles patients and patient advocates wish to play in making decisions about their own medical care. Rather than assuming what these preferences are, this study aims to capture your preferred roles in medical decision making, and the extent to which trust in providers and online information shapes these roles. This study will be used as part of Daniel Saliba's PhD thesis. It is hoped that your responses will be used to improve patient engagement efforts and the care that people with cystic fibrosis receive. There are no right or wrong answers to the questions being asked, I am simply interested in your opinion. Please consider sharing your valuable opinions.

Voluntary Participation:

Survey: You are invited to participate in a survey that will take about 10 to 15 minutes to complete. Your participation in this survey is voluntary and can be stopped at any time. You may also skip questions that you do not wish to answer. All data collected will not be shared with your doctors or nurses.

At the end of the survey, a question will appear which asks if you interested in taking part in a follow-up telephone interview. You may choose the “yes” or “no” box option. If are willing to be interviewed, you will be asked to leave your email address so that the researcher can contact you in the future.

Follow-up Interview: To better understand your decision making preferences, we would like to conduct a follow-up telephone interview with individuals who have completed the survey. This interview will ask questions about the information you rely upon when choosing to make medical decisions. Interviews will be scheduled at a time that works for you and will take about 20 to 30 minutes to complete. Combining survey responses with this interview will provide a better picture of your preferences.

Risks and Benefits:

Risks: Those who agree to be interviewed will provide their email address. These emails will not be shared with anyone, and will only be used by the researcher to contact you. In addition, all data collected will only be used for the purposes of this study. As such, there are no risks associated with participating.

Benefits: The results of this study will be shared with the wider academic and CF community, as well as with Cystic Fibrosis Canada (CFC). The main goal of this research is to help to improve the quality of care that people with CF receive. Learning about your decision making preferences will help to achieve this important goal.
Sharing the results with CF agencies will also be vital for developing courses of action that can be used to improve patients' experiences. The researcher will be working closely with CFC to decide how the results of this research can be used in a meaningful way.

Confidentiality:

Those who complete the survey but do not wish to be interviewed will remain anonymous, meaning their responses cannot be removed. If you provide your email information in order to be interviewed, you will be able to withdraw your survey data. You can ask to have your responses removed by emailing the researcher or calling him directly. No reason for choosing to withdraw your data is needed. All survey and interview data will be protected in many ways. This will include using a password to restrict access to the online survey database and using a computer to analyze data that is password protected. Audio recordings of interviews will also be moved to an encrypted hard drive within 24 hours of being taped (a technology that protects information). Any survey and interview results reported in future papers will not be linked to any person.

Access to Information and Publication of Results:

Data collected in support of this study will be kept for 5 years after it is completed. Access to the final results and reports produced will be made available in many ways. CFC will help to share results on their website, social media platforms, and will help to send out emails to participants. Presentations may also be held (such as webinars and or at conferences) in the future. Even if you choose not to participate in this study, you may email the researcher to learn about the results.

Summary:

This consent page explains the study you are being asked to participate in. *If you require more information, please contact Daniel Saliba using the information below. You may also contact Kim Steele who is helping to forward this study. You can also call the Research Oversight and Compliance Office at ethics.review@utoronto.ca or 416-946-3273 if you have any questions about your rights as a participant.

Daniel Saliba
416-951-7900
Daniel.saliba@mail.utoronto.ca
Institute of Health Policy, Management and Evaluation

Kim Steele
1-800-378-2233 (extension #297)
ksteele@cysticfibrosis.ca
Cystic Fibrosis Canada

Would you like to participate in this study?

○ Yes, I am above 18 and would like to participate in this study (please click the arrow button below)

○ No, I am not interested in participating in this study (please click the arrow button below)

1. Please select the box or boxes which best describe you:

○ I am a person with cystic fibrosis (CF)
○ I am a caregiver of a person with CF who is under the age of 18
○ I am a caregiver of a person with CF who is 18 or above
○ I am a family member of a person with CF, but I am not a caregiver
○ I am an advocate that volunteers with Cystic Fibrosis Canada who has CF
○ I am an advocate that volunteers with Cystic Fibrosis Canada who does not have CF

2. Please indicate your sex

○ Male
○ Female
○ Other (please specify) ____

3. What year were you born in?

____

4. What country were you born in?

____

5. What is your preferred spoken language?

____

6. What is the highest level of education you have received? (Select one box)

○ Some elementary school (up to grade 8)
○ Completed elementary school
○ Some high school
○ Completed high school
○ Some post high school (university, college or trade school)
○ Completed university or college
○ Some professional or graduate school
○ Completed professional or graduate school

7. What is your employment status at present? (Select one box)

- ○ Employed full time (40 hours or more per week)
- ○ Employed part time (up to 39 hours per week)
- ○ Not employed
- ○ Retired
- ○ Homemaker
- ○ Student
- ○ Self-employed
- ○ Unable to work or temporarily sick
- ○ Other, please specify ____

8. Overall, how would you rate your current health? (Select one box)

- ○ Excellent
- ○ Very good
- ○ Good
- ○ Fair
- ○ Poor

9. Please select the box which best describes how you feel about each of the below statements related to making decisions about cystic fibrosis. *If you do not have CF, please provide the name of your current health condition or a condition which you had in the past.

____

A. Who should determine (diagnose) what the likely causes of your symptoms are?

- ○ Doctor alone (1)
- ○ Mostly the doctor (2)
- ○ Doctor and you equally (3)
- ○ Mostly you (4)
- ○ You alone (5)

B. Who should determine what the treatment options are?

○ Doctor alone (1)
○ Mostly the doctor (2)
○ Doctor and you equally (3)
○ Mostly you (4)
○ You alone (5)

C. Who should determine what the risks and benefits for each treatment option are?

○ Doctor Alone (1)
○ Mostly the doctor (2)
○ Doctor and you equally (3)
○ Mostly you (4)
○ You alone (5)

D. Who should determine how likely each of these risks and benefits are to happen?

○ Doctor alone (1)
○ Mostly the doctor (2)
○ Doctor and you equally (3)
○ Mostly you (4)
○ You alone (5)

E. Given the risks and benefits of these possible treatments, who should decide how acceptable those risks and benefits are for you?

○ Doctor alone (1)
○ Mostly the doctor (2)
○ Doctor and you equally (3)
○ Mostly you (4)
○ You alone (5)

F. Given all the information about risks and benefits of the possible treatments, who should decide which treatment option should be selected?

- Doctor alone (1)
- Mostly the doctor (2)
- Doctor and you equally (3)
- Mostly you (4)
- You alone (5)

Q15. Please select the box which best describes how you feel about the statements below assuming you had the medical condition which is described.

"Suppose you had mild chest pains for three days and decided that you should visit your doctor about this".

A. Who should determine (diagnose) what the likely causes of your symptoms are?

- Doctor alone (1)
- Mostly the doctor (2)
- Doctor and you equally (3)
- Mostly you (4)
- You alone (5)

B. Who should determine what the treatment options are?

- Doctor alone (1)
- Mostly the doctor (2)
- Doctor and you equally (3)
- Mostly you (4)
- You alone (5)

C. Who should determine what the risks and benefits for each treatment option are?

- Doctor alone (1)
- Mostly the doctor (2)
- Doctor and you equally (3)
- Mostly you (4)
- You alone (5)

D. Who should determine how likely each of these risks and benefits are to happen?

- Doctor alone (1)
- Mostly the doctor (2)
- Doctor and you equally (3)
- Mostly you (4)
- You alone (5)

E. Given the risks and benefits of these possible treatments, who should decide how acceptable those risks and benefits are for you?

- Doctor alone (1)
- Mostly the doctor (2)
- Doctor and you equally (3)
- Mostly you (4)
- You alone (5)

F. Given all the information about risks and benefits of the possible treatments, who should decide which treatment option should be selected?

- Doctor alone (1)
- Mostly the doctor (2)
- Doctor and you equally (3)
- Mostly you (4)
- You alone (5)

G. How much experience have you had with the clinical situation described in the above scenario? (Please select all that apply)

- I have had personal experience with it. (1)
- I know of family members or close friends who have experienced it. (2)
- I have read/heard about it. (3)
- I do not know much about it. (4)

22. Please select the box which best expresses your opinion about the following statements.

A. I know how to find helpful health resources on the Internet.

- Strongly disagree (1)
- Disagree (2)
- Neither agree nor disagree (3)
- Agree (4)
- Strongly agree (5)

B. I know how to use the Internet to answer my health questions.

- Strongly disagree (1)
- Disagree (2)
- Neither agree nor disagree (3)
- Agree (4)
- Strongly agree (5)

C. I know what health resources are available on the Internet.

- Strongly disagree (1)
- Disagree (2)
- Neither agree nor disagree (3)
- Agree (4)
- Strongly agree (5)

D. I know where to find helpful health resources on the Internet.

- ○ Strongly disagree (1)
- ○ Disagree (2)
- ○ Neither agree nor disagree (3)
- ○ Agree (4)
- ○ Strongly agree (5)

E. I know how to use the health information I find on the Internet to help me.

- ○ Strongly disagree (1)
- ○ Disagree (2)
- ○ Neither agree nor disagree (3)
- ○ Agree (4)
- ○ Strongly agree (5)

F. I have the skills I need to evaluate the health resources I find on the Internet.

- ○ Strongly disagree (1)
- ○ Disagree (2)
- ○ Neither agree nor disagree (3)
- ○ Agree (4)
- ○ Strongly agree (5)

G. I can tell high quality from low quality health resources on the Internet.

- ○ Strongly disagree (1)
- ○ Disagree (2)
- ○ Neither agree nor disagree (3)
- ○ Agree (4)
- ○ Strongly agree (5)

H. I feel confident in using information from the Internet to make health decisions.

- o Strongly disagree (1)
- o Disagree (2)
- o Neither agree nor disagree (3)
- o Agree (4)
- o Strongly agree (5)

30. Please select the box which best expresses your opinion about each of the following statements.

A. My doctor is usually considerate of my needs and puts them first.

- o Strongly disagree (1)
- o Disagree (2)
- o Neither agree nor disagree (3)
- o Agree (4)
- o Strongly agree (5)

B. I doubt that my doctor really cares about me as a person.

- o Strongly disagree (1)
- o Disagree (2)
- o Neither agree nor disagree (3)
- o Agree (4)
- o Strongly agree (5)

C. I trust my doctor so much I always try to follow his/her advice.

- o Strongly disagree (1)
- o Disagree (2)
- o Neither agree nor disagree (3)
- o Agree (4)
- o Strongly agree (5)

D. If my doctor tells me something is so, then it must be true.

- Strongly disagree (1)
- Disagree (2)
- Neither agree nor disagree (3)
- Agree (4)
- Strongly agree (5)

E. I sometimes distrust my doctor’s opinion, and would like a second one.

- Strongly disagree (1)
- Disagree (2)
- Neither agree nor disagree (3)
- Agree (4)
- Strongly agree (5)

F. I trust my doctor’s opinion about my medical care.

- Strongly disagree (1)
- Disagree (2)
- Neither agree nor disagree (3)
- Agree (4)
- Strongly agree (5)

G. I feel my doctor does not do everything he/she should for my medical care.

- Strongly disagree (1)
- Disagree (2)
- Neither agree nor disagree (3)
- Agree (4)
- Strongly agree (5)

H. I trust my doctor to put my medical needs above all other considerations when treating my medical problems.

- Strongly disagree (1)
- Disagree (2)
- Neither agree nor disagree (3)
- Agree (4)
- Strongly agree (5)

I. My doctor is a real expert in taking care of medical problems like mine.

- Strongly disagree (1)
- Disagree (2)
- Neither agree nor disagree (3)
- Agree (4)
- Strongly agree (5)

J. I trust my doctor to tell me if a mistake was made about my treatment.

- Strongly disagree (1)
- Disagree (2)
- Neither agree nor disagree (3)
- Agree (4)
- Strongly agree (5)

K. I sometimes worry that my doctor may not keep the information we discuss totally private.

- Strongly disagree (1)
- Disagree (2)
- Neither agree nor disagree (3)
- Agree (4)
- Strongly agree (5)

Thank you for taking the time to complete this survey!

Would you be interested in a follow-up telephone interview? This interview should take 20 to 30 minutes. If yes, please select "yes" and provide your email address to allow the researcher to contact you.

○ Yes, I am willing to be interviewed.
____
○ No, I am not willing to be interviewed.

If you are not interested in being interviewed, but would still like to receive the study results, please select "yes" and provide your email below.

○ Yes, I am interested in receiving the study results.
____
○ No, I am not interested in receiving the study results.

Appendix 3: Qualitative Follow-up Interviews

1. Medical decision making can include any number of decisions that affect a person's health. These can include diagnosing a person's health condition based on their symptoms, determining treatment options, assessing the risks and benefits of treatment options, deciding which treatment(s) to pursue, and so on.

a) What are the types of decisions you *prefer* to be involved in?
b) What are the types of decisions you *do not* prefer to be involved in?
c) Does your preferred level of involvement depend on how serious the health problem is?
d) When are you most likely to leave decision making entirely to your doctor?

2. What sources of information do you turn to when you have questions about your health?

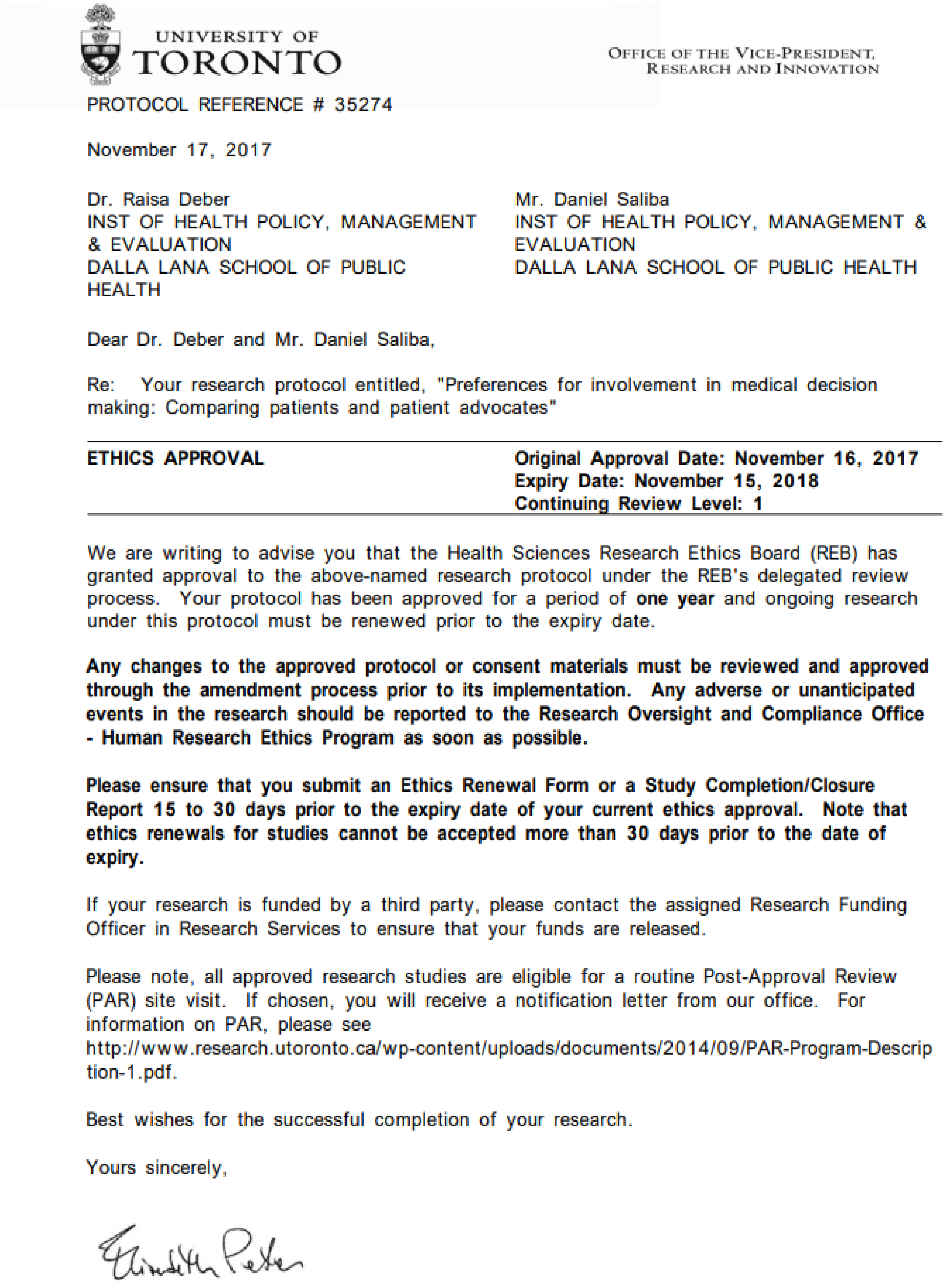
a) How much do you trust these sources of information?
b) What are the benefits of using these different sources of information?
c) What are the drawbacks of using these different sources of information?
d) When are you mostly likely to use the Internet (i.e., for what purpose)?
e) How much do you trust the information you receive from your doctor when compared to other sources of information (such as the Internet?)
f) Does the amount of trust you have in your doctor influence how much you wish to be involved in decisions about your health? If so, how?

3. What are the benefits of involving patients in medical decision making? What are the drawbacks of involving patients in medical decision making?

a) When do you think patients should *be less* involved in making medical decisions?
b) When do you think patients should *be more* involved in making medical decisions?

Appendix 4: Ethics Approval Letter

*Please note that the ethics protocol was amended and reapproved at a later date.



UNIVERSITY OF
TORONTO

OFFICE OF THE VICE-PRESIDENT,
RESEARCH AND INNOVATION

PROTOCOL REFERENCE # 35274

November 17, 2017

Dr. Raisa Deber
INST OF HEALTH POLICY, MANAGEMENT & EVALUATION
DALLA LANA SCHOOL OF PUBLIC HEALTH

Mr. Daniel Saliba
INST OF HEALTH POLICY, MANAGEMENT & EVALUATION
DALLA LANA SCHOOL OF PUBLIC HEALTH

Dear Dr. Deber and Mr. Daniel Saliba,

Re: Your research protocol entitled, "Preferences for involvement in medical decision making: Comparing patients and patient advocates"

ETHICS APPROVAL

Original Approval Date: November 16, 2017
Expiry Date: November 15, 2018
Continuing Review Level: 1

We are writing to advise you that the Health Sciences Research Ethics Board (REB) has granted approval to the above-named research protocol under the REB's delegated review process. Your protocol has been approved for a period of **one year** and ongoing research under this protocol must be renewed prior to the expiry date.

Any changes to the approved protocol or consent materials must be reviewed and approved through the amendment process prior to its implementation. Any adverse or unanticipated events in the research should be reported to the Research Oversight and Compliance Office - Human Research Ethics Program as soon as possible.

Please ensure that you submit an Ethics Renewal Form or a Study Completion/Closure Report 15 to 30 days prior to the expiry date of your current ethics approval. Note that ethics renewals for studies cannot be accepted more than 30 days prior to the date of expiry.

If your research is funded by a third party, please contact the assigned Research Funding Officer in Research Services to ensure that your funds are released.

Please note, all approved research studies are eligible for a routine Post-Approval Review (PAR) site visit. If chosen, you will receive a notification letter from our office. For information on PAR, please see http://www.research.utoronto.ca/wp-content/uploads/documents/2014/09/PAR-Program-Description-1.pdf.

Best wishes for the successful completion of your research.

Yours sincerely,

Elizabeth Peter

Appendix 5: Letter of Support from CFC

Cystic Fibrosis Canada

2323 Yonge Street, Suite 800
Toronto, Ontario, M4P 2C9
Phone: 1 800-378-2233
www.cysticfibrosis.ca

September 8, 2017

To whom it may concern:

This letter is in support of the doctoral work being undertaken by Daniel Saliba at the University of Toronto in the Institute of Health Policy, Management and Evaluation (IHPME). This work will take place under the supervision of Professor Raisa Deber and will focus on the roles that cystic fibrosis (CF) patients and patient advocates play, and wish to play, in their own medical decision making.

Cystic fibrosis is the most common fatal genetic disease affecting Canadian children and young adults. At present, there is no cure. CF causes various effects on the body, but mainly affects the digestive system and lungs. The degree of CF severity differs from person to person, however, the persistence and ongoing infection in the lungs, with progressive loss of lung function, will eventually lead to death in the majority of people with CF. Other typical complications caused by cystic fibrosis are: difficulty digesting fats and proteins; malnutrition and vitamin deficiencies because of inability to absorb nutrients; CF-related diabetes; sinus infections.

Approximately one in every 3,600 children born in Canada has CF, making CF a rare – or orphan - disease. More than 4,100 Canadian children, adolescents, and adults with cystic fibrosis attend specialized CF clinics. Medical management promotes significantly improved survival outcomes of what historically was a fatal chronic disease of children and young adults. Canadian CF patients attended over 15,500 CF-specific outpatient clinic visits in 2014; cumulatively they spent almost 25,000 days in the hospital. To put things in perspective, that adds up to 68.5 years.

Canadians with CF live 10 years longer than their U.S. counterparts, likely due to the high standard of clinical care and access to medicines; however, this gap could close if Canadians with CF can't access disease-modifying therapies through public drug plans like Americans with CF can. Ages of individuals living with cystic fibrosis today range from birth to almost 80 years old. In 2015, the median age of survival in Canada was 52.1 years; still, of the 47 patients who died in 2015, half were under 29.7 years of age.

Cystic Fibrosis Canada is a national charitable not-for-profit corporation established in 1960, and is one of the world's top three charitable organizations committed to finding a cure for CF. As an internationally recognized leader in funding innovation and clinical care, we invest more in life-saving CF research and care than any other non-governmental agency in Canada. We are committed to helping people with CF to live healthy and well.

Since its establishment, Cystic Fibrosis Canada has invested more than $235 million in leading research, innovation and care. As a result, Canadians with cystic fibrosis have one of the highest median survival rates in the world. As we work toward new treatments and ultimately a cure for this devastating disease, we also work to improve the services and supports people with CF need to live healthy and well.

In 2017, Cystic Fibrosis Canada is investing $3.16 million dollars in Ontario to support CF clinical care, transplants and research.

Cystic Fibrosis Canada

2323 Yonge Street, Suite 800
Toronto, Ontario, M4P 2C9
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www.cysticfibrosis.ca

One of the goals of Daniel's work is to determine whether or not advocacy initiatives can be improved to ensure that CF patients' and caregivers' preferences are being accurately articulated to the broader medical and public policy community. We believe that the experiences of Canadians with CF and their families have had in working with clinicians at the 42 specialty CF clinics across Canada to manage their health will go a long way in helping Daniel meet this objective.

To achieve his research objectives, it is our understanding that Daniel intends to conduct surveys and interviews of people with CF and their caregivers, as well as individuals who represent and/or support CF patients who are involved in a range of patient engagement activities.

According to the research brief, Daniel will conduct online surveys to determine and compare the roles that CF patients and patient advocates wish to assume in their own medical decision making, and how these roles might vary with: trust in providers, self-assessed health information literacy, and demographics. His work may also involve follow-up interviews of survey respondents for the purposes of explaining survey results, and to provide insights into the sources of information that people rely upon when considering courses of action pertaining to CF.

Cystic Fibrosis Canada is a proud supporter of this important work and we are pleased to help Daniel and Raisa ensure that this research is meaningful and has a positive impact on Canadians living with CF and their loved ones. To that end, Cystic Fibrosis Canada has agreed to promote participation in the study by assisting with the development of a direct email list and to send a study invitation to those who are on said list. We will also help determine expected response rates based on the size of this list. Cystic Fibrosis Canada will also be involved in the piloting of surveys and will work with Daniel to ensure that communications with the study participants is compliant with the Canadian Anti-Spam Legislation (CASL), and is both transparent and ongoing.

In closing, Cystic Fibrosis Canada is pleased to partner with Dr. Deber and Mr. Saliba on this initiative. Should you have any questions about our partnership, please contact Kim Steele, Manager, Government Relations and Advocacy at ksteele@cysticfibrosis.ca or by phone at 416-485-9149 ext. 297.

Sincerely,

Norma Beauchamp
President & CEO

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