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Information Needs and Information Sources of Patients Diagnosed with Rare Cancers

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A dissertation submitted in partial fulfillment of the requirements for the degree of Doctor of Philosophy at Virginia Commonwealth University.

by

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I'd like to dedicate this work in memory of my grandmother, Grace Healy Jackson who always loved, encouraged, and believed in me.

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Abstract

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By Dana L. Ladd, Ph.D., MS, SLIS

A dissertation submitted in partial fulfillment of the requirements for the degree of Doctor of Philosophy at Virginia Commonwealth University

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BACKGROUND: Approximately 25% of all cancers diagnosed are considered rare. Patients may face many significant challenges including difficulty obtaining information about their rare conditions. Patients often have high information needs and may seek desired information from a variety of informational sources including healthcare providers, media, print, government and non-profit organizations in order to meet their needs. Accessing reliable consumer-level information can be challenging and often information needs are unmet. Dissatisfaction with health information provision can result in negative health-related outcomes and factors including decreased health-related quality of life.

METHOD: This cross-sectional design study used validated measures to assess the information needs, information sources, information satisfaction, and health-related quality of life of patients diagnosed with rare cancers (n=113). Adult patients at the VCU Health Massey Cancer Center who had been diagnosed with a rare cancer in the past 12 months were contacted via mail survey. Descriptive statistics were used to summarize patients' information need and information received levels. Unmet needs were analyzed using a two-sample T-test. Chi-square tests were used to analyze information needs and received by demographics for gender and race and logistic regression analysis was used for age. Descriptive statistics summarized information sources used and preferred. Finally, descriptive statistics were used to summarize information satisfaction. The relationship between information satisfaction and health-related quality of life was assessed using a two-sample T-test.

RESULTS: Study participants had high information needs, particularly for information about disease, medical tests, and treatment. Though patients also reported receiving information at high levels, 21 participants (18.9%) reported being unsatisfied with information provision. Unmet needs were found for information about disease and on the item level for information about causes of cancer, whether their cancer was under control, expected benefits of treatment, and financial, insurance, and work-related information. Although participants reported preferring information from their healthcare providers, they most commonly sought information from the Internet more than any other source.

CONCLUSION: By identifying patients' information needs and sources, this study fills an important gap in the information needs and sources literature of patients diagnosed with rare cancers. Identification of these needs allows healthcare providers to tailor information provision to more effectively meet patients' information needs.

Chapter I. Introduction

According to the National Cancer Institute, in 2016 over 1.6 million people will be diagnosed with cancer in the United States (National Cancer Institute, 2014). While there are few patients diagnosed with any one type of rare cancer, approximately 25% of all cancers are considered rare (Greenlee, Goodman, Lynch, Platz, & Havener, 2010).

Patients with rare cancers face many significant challenges in addition to the attendant complications of dealing with a potentially life-threatening and life-altering illness (Schaefer, 2012). Patients with rare cancers may have difficulty obtaining a correct and timely diagnosis; have few or no treatment options; and often lack access to specialists (Schaefer, 2012). In addition, patients with rare cancers often have difficulty accessing and obtaining consumer-level information about their types of cancers (Schaefer, 2012). Cancer-related information needs and sources of health information have been researched in patients diagnosed with common cancers; however, little is known about the unique health information needs (types of cancer information sought and desired) and the information sources used and preferred by patients diagnosed with rare cancers. The research found in the literature that examines the information needs and information sources of patients diagnosed with a rare disease is often limited to the study of parents of children with very rare genetic conditions.

A cancer diagnosis in general can have a profound impact on patients, and can result in a variety of health information needs throughout the cancer care continuum (Rutten, Arora, Bakos, Aziz, & Rowland, 2005). Many of these patients desire additional information about their diagnosis, treatment options, prognosis, coping, complementary and alternative medicine, relapse, legal and financial, and survivorship information.

Patients with cancer seek information from a variety of sources including healthcare providers, media, print, government and non-profit organizations, and others to fulfill their information needs. Most patients, when questioned, prefer to receive health information from their healthcare provider; however, many report that they actually sought and received information from a variety of other sources (Rutten et al., 2005; Chou, Liu, Post, & Hesse, 2011).

While patients desire and seek information regarding their cancers or rare diseases, often information needs are not met. Unsatisfied information needs can result in a variety of negative associated factors as compared to patients who report information satisfaction (Halkett, Kristjanson, Lobb, Little, & Shaw, 2012) Decreased trust in physician, decreased satisfaction with health care, and a low health-related quality of life can result from patients having unmet or unsatisfied information needs (Halkett et al., 2012). These factors are important to consider as they have been found to be associated with cancer patients' treatment adherence and overall survival (Halkett et al., 2012).

Using a modified version of Johnson's Comprehensive Model of Cancer Information Seeking (Johnson, 1997) as a guide, this cross-sectional survey study examines the unique information needs and information sources used and preferred by patients who have been diagnosed with rare cancers. Data were collected using validated and reliable measures via mail surveys of patients diagnosed with rare cancers at the Virginia Commonwealth University Massey Cancer Center. In addition, this study examines the association between information satisfaction and health-related quality of life. Overall, this study fills a gap in the rare cancer literature as it provides an understanding of the information needs and information sources of patients with rare cancers, an often overlooked and neglected patient population.

Chapter II. Literature Review

Definition of Rare Cancer

Before examining the information needs and information sources of patients diagnosed with cancers, it is important first to have an understanding of the definition of rare cancer and the various challenges faced by those diagnosed with rare cancers. Many different definitions of rare cancer have been developed by national and international rare disease organizations, but no single definition of a rare cancer has been agreed upon. Defining rare cancer is not easy and any definition of a rare cancer is considered artificial (RareCare, 2014). In general, in the United States, a cancer is considered to be rare if it affects fewer than 200,000 people. This is the definition of rare disease used by both the United States Orphan Drug Act and the National Institute of Health’s Office of Rare Diseases Research (Office of Rare Disease Research, 2014). The Orphan Drug Act states that “rare” may also be defined as a condition affecting more than 200,000 people but for which drug development will not be recouped from sales of the drug in the United States (Eslick, 2012).

When studying rare cancers, Eslick (2012) explains that it is necessary to understand the difference between incidence rate and prevalence rate. Some rare cancers with a low incidence rate and that are highly curable may have a high prevalence rate due to patients living longer with the cancer. Conversely, there are common cancers with very poor survival rates that make their disease prevalence low (Gatta, Capocaccia, Trama, & Martinez-Garcia, 2011). Therefore, defining a rare cancer using incidence rate rather than prevalence rate accounts for the varying life expectancy among patients with different types of rare cancers (RareCare, 2014).

Another consideration related to rare cancers is that they are not only rare histologic (primary cancer) variants, but also include: 1) subgroups that are difficult to study in common cancers; 2) common cancers or metastasis in uncommon sites; 3) common cancers in uncommon hosts; 4) cancers with unusual presenting symptoms, manifestations, or complications; and 5) cancers that rarely affect a particular age group or population. Delay in diagnosis or misdiagnoses can occur in these circumstances (Duffy, 2012).

For example, Miller (2010), describing rare subtypes of common cancers, provides the example of adenoid cystic carcinoma of the breast. While breast cancer is a common form of cancer in women, adenoid cystic breast cancer is a rare subtype of breast cancer, affecting only about one percent of those with breast cancer (Johns Hopkins Medicine, 2014). Examples of common cancers in rare sites include primary extra-ovarian tumors, inflammatory breast cancer, non-carcinoma breast cancers, ductal carcinoma in salivary glands, and non-cutaneous melanomas (Duffy, 2012; Greenlee et al., 2010).

Rare cancers can also be common cancers that present themselves in uncommon hosts such as breast cancer occurring in men (Eslick, 2012; Greenlee et al., 2010). Additionally, some literature also includes rare cancers that may have uncommon initial symptoms, making diagnosis difficult. Furthermore, cancers that do not often affect a particular age group, such as when a pediatric type of cancer occurs in an adult can be considered rare (Calhoun, 1998).

There are other factors that should be taken into account when studying rare cancers. It is important to know that some rare cancers do attract more media attention and obtain more funding and research than other rare cancers. An example of this includes malignant mesothelioma, a rare type of cancer that often affects the pleural tissue that lines the lungs, but can also affect tissue lining of the stomach, heart, and other organs. Many patients who are diagnosed with mesothelioma were occupationally exposed to asbestos. Mesothelioma has had considerable recent media exposure due to the many class-action, wrongful death lawsuits that have resulted from workplace exposure and subsequent mesothelioma diagnosis. Often, for these types of rare cancers, there is a greater awareness of the cancer and generally more information is available for both physicians and patients.

Individually, rare cancers affect only a very small percentage of people; however, collectively, rare cancers account for a large percentage of cancers. Miller (2010, p. 46) states, “Although individual rare diseases may be infrequently encountered, rare diseases in aggregate affect tens of millions of individuals in the US, Europe and worldwide.” Likewise, Greenlee et al. (2010) found that 25% of patients with cancers in the United States have a rare cancer. The research team defined “rare” as being fewer than 150 incident cases per one million per year. Incidentally, by defining rare cancers as less than 150 per one million, only 11 in the U.S are considered common: prostate, breast, lung/bronchus, colon, uterine, bladder, melanoma, rectal, ovarian, non-Hodgkin lymphoma, and kidney/renal pelvis cancer (Greenlee et al., 2010).

Rare cancers are also known as “orphan diseases” as they have been neglected by the medical and research community for many years (Eslick, 2012). Consequently, due to this long-standing neglect, patients with rare cancers encounter a lack of resources and organizational support compared to those diagnosed with more common types of cancers.

Rare Cancer Challenges

Gatta et al. (2011, p. 2494) state, “A major problem with rare cancers is that their overall burden on society has not been adequately estimated, although they are thought to constitute a major public health problem….Improving the quality of care for these cancers is a public health priority." In order to provide a brief overview of the significance of having a rare cancer, outlined below are some of the many challenges encountered by patients with rare cancers.

In addition to the difficulty of dealing with a cancer diagnosis and its attendant impact on the patient's health, patients diagnosed with rare cancers encounter many challenges due to the rarity of their conditions. While there are many rare cancers, few people have any one specific rare cancer. Consequently, rare cancers are often underfunded and understudied compared to other more common cancers ("Very Rare Cancers," 2001). Both epidemiological and natural history studies of rare cancers are very difficult to conduct due to lack of funding and few patients to participate in studies (Casali, Bruzzi, Bogaerts, & Blay, 2015). Also contributing to the difficulties of conducting rare cancer research is a lack of unique International Classification of Disease (ICD) codes for many very rare cancers (Ayme, Bellet, & Rath, 2015). For example, angiofollicular lymph node hyperplasia (Castleman Disease), a very rare lymphoma-like disease, lacks its own ICD code and is coded under a more general ICD code, "lymphadenopathy," along with other diseases, making research difficult (Castleman's Disease Collaborative Network, 2014). Additionally, there are fewer available cancer registries and tissue banks for rare cancer types than those for more common types of cancers (Schaefer, 2012).

Inadequate diagnostics and physicians' lack of knowledge of specific rare cancers (Schaefer, 2012) contribute to late or incorrect diagnoses of rare cancer (Feinberg, Law, Singh, & Wright, 2013; Jeffries & Clifford, 2009). Furthermore, patients may have difficulty finding and accessing clinical experts who have the expertise to diagnose the rare cancer. Patients with rare cancers may have to seek multiple medical opinions and travel distances to consult qualified experts for a complete diagnostic workup in order to obtain an accurate diagnosis. In a qualitative study by Jeffries and Clifford (2009) of women with cancer of the vulva (a rare cancer), women reported the frustration of delayed and incorrect diagnosis. Many women in the study expressed shock over a cancer diagnosis after being assured for years that they had a benign condition (Jeffries & Clifford, 2009). When physicians fail to find a medical diagnosis for the patients' symptoms, patients who repeatedly seek medical opinions for their undiagnosed symptoms may be diagnosed as having a psychosomatic or stress-related condition (Jeffries & Clifford, 2009).

Patients with rare cancers face additional challenges with regard to receiving timely and appropriate treatment. Delayed diagnosis, described above, may also result in treatment delays for the patient. Furthermore, appropriate therapies for the condition may not be available. There is often a lack of interest in drug development among pharmaceutical companies, with a limited/small potential market of patients resulting in difficulty recouping costs and few patients available to participate in existing clinical trials. These factors contribute to limited treatment options and often no U.S. Food and Drug Administration (FDA) approved therapies for many rare cancers.

Lack of research and physician knowledge; late and incorrect diagnosis; and insufficient treatments have a negative impact on patients. These factors contribute to the survival rate of patients with rare cancers being lower than the survival rate of patients with more common cancers (Eslick, 2012; Greenlee et al., 2010; Williams, 2011). As Greenlee et al. (2010, p.41) report, very rare cancers "can be rapidly fatal."

In addition to the challenges briefly outlined above, patients with rare cancers experience a lack of accessible and reliable information. Although they desire health information about their condition, finding authoritative information about rare cancers can be a daunting challenge (Walker, 2013).

Furthermore, there is a paucity of research in the literature regarding the health information needs and information sources of patients diagnosed with rare cancers. Even when broadening the scope of the search to include information needs of patients with any type of rare condition, there is very little written regarding this population. Most rare disease research has been conducted in Europe and focuses on the diagnostic challenges and information needs of parents of children who have rare genetic disorders (Bouwman, Teunissen, Wijburg, & Linthorst, 2010; Tozzi, Mingarelli, Agircola, Gonfiiantine, & Pandolfi, 2013). In a systematic review of research of cancer patients' information needs and sources for finding information, Rutten et al. (2005, p. 258) state, "Future research should address the limitations identified in past research by assessing the information needs and sources of large, representative, and diverse samples of cancer patients, including those with rare cancer diagnoses." Since this 2005 article, very little has been written to address the gap of patients' rare cancer information needs in the research literature.

General Cancer Patients' Information Needs and Information Sources

Because of the lack of data available on information needs and sources for patients with rare cancers, the following examines the types of information sought and sources used by those in the general (non-rare) cancer patient population.

Information needs. Cancer patients' information needs vary over the cancer care continuum (Halkett et al., 2012; Maddock, Lewis, Ahmad, & Sullivan, 2011; Matsuyama, Kuhn, Molisani, & Wilson-Genderson, 2013; Rutten et al., 2005). Cancer information needs are at their highest during the diagnosis and treatment phases. (Maddock et al, 2011; Matsuyama et al., 2013; Rutten et al., 2005). Cancer information needs reduce slightly following treatment, but remain high throughout the cancer care continuum. According to Matsuyama et al., 2013, p. 100).

"Information needs may be high because many patients have no experience with cancer at the time of diagnosis and want information to be able to understand and cope with their cancers. Information needs decrease over time indicating that patients are able to obtain and process needed information throughout treatment. However, since their needs for information remain high it would appear that new needs arise."

The quantity of information desired also varies from patients desiring no information about their cancers to those who want to know everything. In a study of cancer website users, researchers found respondents "wanted information on all aspects of cancer (Maddock et al., 2011)." In another study, most participants responding to a question about desired information stated that they wanted "everything, including bad news" for side effects, disease, and chance of getting worse (Matsuyama et al., 2013). In contrast, Balka, Krueger, Holmes, & Stephen's (2010) study of breast cancer patients' information needs found that some women did not want any additional information about their breast cancer.

Those patients who desire information have a variety of health information needs. Rutten et al. (2005) reported categories of informational needs of patients with cancers based upon a systematic review of the cancer information needs literature. The categories of cancer information needs outlined in more detail below are: cancer-specific information; treatment-related information; prognosis information; surveillance information and health information; coping and interpersonal/social information; financial/insurance/legal information; and medical system information.

Cancer-specific information: Cancer-specific information includes information on the type of cancer, etiology, physical effects, diagnosis, and symptoms of the patients' specific cancer (Rutten et al., 2005). A study by Shea-Budgell, Kosarus, Myhill, & Hagan (2014) about information needs of 411 patients with general cancers found that 43% reported searching for information on their specific cancer type. Women in a study of breast cancer information needs reported desiring breast cancer information to verify existing knowledge of their condition and to elaborate further on what they already know (Balka et al., 2010).

Treatment-related information: This category encompasses information about the patient's treatment, treatment options (Shea-Budgell et al., 2014), side effects, clinical trials (Maddock et al., 2011), and medication information. Shea-Budgell, et al. (2014) found that the majority of patients reported that treatment was the most important type of information. In addition to desiring information about specific treatment options mentioned by the physician, patients also desire information about all treatment options available for their conditions (Castleton, Fong, Wang-Gillam, Waqar, & Jeffe, 2010). Cancer patients report searching for information about treatment options such as chemotherapy, radiotherapy, and surgery. Patients also desire information about side effects of treatment (Castleton et al., 2010; Chou et al., 2011) and information about how to manage the side effect symptoms of treatment (Castleton et al., 2010; Tustin, 2010). A longitudinal study of the information needs of women receiving radiotherapy for breast cancer found that women desire information about treatment side effects and the impact of treatment on their overall health and life (Halkett et al., 2012). Beyond standard treatment options, cancer patients search for information about integrative and complementary and alternative (CAM) therapy options (Castleton et al., 2010; Rutten et al., 2005).

Prognosis information: Patients inquire about their prognosis, recovery, and long-term outcomes (Chou et al., 2011; Rutten et al., 2005; Shea-Budgell et al., 2014), with 58% of cancer patients reporting they sought information regarding prognosis for their particular types of cancer

(Castleton et al., 2010). In a study by Balka et al. (2010) using narrative to explore the information needs of breast cancer patients, many women reported seeking breast cancer health information for prognosis and long-term life expectancies. One respondent in the study stated, “I needed to know if I was going to die (Balka et al., 2010, p. 398).”

Surveillance information and health information: Patients with cancer also seek information about their physical and psychological health, prevention, and early detection (Rutten et al., 2005). Maddock et al. (2011) report that patients desire information about leading a healthy lifestyle, including diet and nutrition and engaging in physical activity. Along with health information, patients with cancer desire information about sexuality and physical appearance (Rutten et al., 2005). Patients in a study of cancer survivors reported needing information about sexual functioning and fertility (Kent, Arora, Rowland, Bellizzi, & Forsythe, 2012). Other types of health information included in this category are self-care, recovery, and post-treatment follow-up care (Rutten et al., 2005).

Coping information and interpersonal/social information: This category includes information about resources for emotional and spiritual support and how to cope with cancer (Castleton et al., 2010; Rutten et al., 2005). Although not reported as highly as other information needs, approximately 9% of general cancer patients need psychosocial information. Patients may seek information about support groups and desire to meet others diagnosed with their particular type of cancer (Rutten et al., 2005). Maddock et al. (2011) report patients seek counseling information and information about support groups that they can join. Rutten et al. (2005) report that patients need information regarding cancer’s effect on family, friends, and work and may seek information about how to talk to family members about their cancer.

Financial/insurance/legal information: Patients with cancer report financial, insurance, and legal information needs (Rutten et al., 2005). Patients may experience significant costs for treatment and care not covered by health insurance. Patients, particularly those with rare cancers may have to travel to consult disease-specific specialists and receive diagnostic tests and appropriate therapies. Due to illness, patients may also not be able to work or may miss time from work for medical appointments. This may cause a huge financial impact, and patients may need information about financial and insurance resources. Patients also report that they need legal information, which may include information about wills and advance directives (Maddock et al., 2011; Rutten et al., 2005).

Medical system information: This final category of information needs includes information about healthcare providers and specialists, experience and qualifications of physicians and healthcare providers, and information about health care systems (Rutten et al., 2005). Patients use information sources to assess the credentials of their oncologists and the hospitals from which they are receiving healthcare services (Tustin, 2010). Patients with rare cancers may search for cancer-specific specialists.

The above section describes the types of information that patients with cancer report needing and seeking to fill their information gaps. The section below outlines the different types of sources which patients want to find information about their cancer and from which they report receiving information.

Information Sources. Sources of cancer health information include healthcare providers; media (including the Internet); print and libraries; interpersonal; and cancer organizations (Balka et al., 2010; Kowalczyk & Draper, 2012; Roach, Lykins, Gochett, Brechting, & Graue, 2009; Rutten, 2005; Shea-Budgell et al., 2014; Tustin, 2010). Shea-Budgell et al. (2014) found that the majority of patients trusted their health care provider as a source of information compared to all other sources. However, while patients report that they prefer to receive information from their health care providers, they also seek and receive cancer-related information from a variety of sources (Chou et al., 2011; Rutten, 2005). Cancer information sources will be outlined and explained in detail below.

Health care providers: This category encompasses the healthcare team and includes: physicians (oncologists, general practitioners, surgeons, and radiologists), nurses, and other health care providers. Rutten's et al. (2005) secondary data analysis of Health Information National Trends Survey (HINTS) data found that 27.3% of patients reported receiving health information from a health care provider. A study by Shea-Budgell et al. (2014) showed that cancer patients rated their doctor or health professional as the most-trusted cancer information source over the Internet, family, friends, radio, newspaper, and television. The patients in the study responded that their preferred source of cancer information is meeting in person with a health care professional (84%).

Halkett's et al. (2012) study of breast cancer patients undergoing radiotherapy found that patients in reported that a consultation with a radiation therapist would be beneficial in order to receive additional information about radiotherapy. The authors of this study suggest that patients should be provided with additional information before their radiotherapy appointment.

Media: This category includes the Internet, television, videos, and radio. Rutten et al, (2005) found that 13.5% report the use of media and 37.1% of those reporting use of media report the use of the Internet to obtain cancer-related health information. A 2003 HINTS secondary data analysis found that 38% of cancer patients reported seeking information from the Internet, more than any other source (Roach et al., 2009). Another secondary data analysis of

HINTS 2007 data estimated percentages of respondents' first choices for accessing cancer health information and found that 25% responded that the Internet is their first choice (Kowalczyk & Draper, 2012). Patients reported searching for health information online and using the Internet to participate in online support groups, read health-related blogs, and email health care providers for information (Chou et al., 2011).

In contrast to high Internet use, Roach et al. (2009) found only 2% of patients with cancer reported using broadcast media (television, video) to search for health information. Likewise, in a study of breast cancer patients, Balka et al. (2009) found that patients rarely report watching videos to learn more about cancer.

Print: Books, magazines, newspapers, brochures, handouts, and libraries encompass print sources of health information. Rutten et al. (2005) report that 26.2% of patients use print materials for finding cancer health-related information. Additionally, patients also report using the library as a source of information about their cancers (Kowalzyk et al., 2012; Rutten et al., 2005).

Interpersonal: Sources of interpersonal communication include patients talking to friends, family, co-workers, clergy, and other patients. Cancer patients report using interpersonal communication (19%) as a source of health information (Maddock et al., 2011; Rutten et al., 2005). Of those who use interpersonal communication, 43% report communicating with friends and family, 31% report using support groups, 20% report seeking information from other patients, and 6% report that their clergy or church is a source of information (Rutten et al., 2005). Methods of interpersonal communication, for communicating with fellow patients, are through online, telephone, and in-person support groups (Rutten et al., 2005).

Organizations: According to Rutten et al. (2005), this category of information sources encompasses telephone information services, charitable and professional organizations, and health care organizations. In addition to raising awareness of general or specific cancers, organizations provide advocacy, lobbying, research, collaboration, funding, support, and education (Newlands, 2012). An example of a cancer organization is the American Cancer Society. Many of the organizations have online webpages that patients can use to access information and also telephone hotlines for information. Most cancer organizations also provide access to disease-specific online support groups (Ayme, Bellet, & Rath, 2008). These organizations may also publish journals with articles and information about cancer.

Information Satisfaction

The American Society of Clinical Oncology (ASCO) and the European Society for Medical Oncology (ESM) developed the Consensus Statement on Quality Care, a ten-point statement to ensure quality of cancer care. The first goal of the consensus statement is, "Access to information." (The American Society of Clinical Oncology, 2006). The access to information goal states, "Patients should receive adequate information about their illness, possible interventions, and the known benefits and risks of specific treatment options."

The above sections provided an overview of cancer patients' information needs by outlining their desired types of cancer health information and the information sources used. This section examines patients' met and unmet information needs and overall satisfaction with health information.

Accessibility to credible, accurate, and authoritative information is imperative for all patients, including those with rare diseases and cancers, so that they can make informed decisions about their health. The information that patients find often impacts health care decisions (Tozzi et al., 2013). Rutten's et al. (2005) review of the literature regarding cancer patients' information needs found that cancer information provision resulted in many benefits. These benefits included: an increased patient participation in decision making; increased treatment satisfaction; increased coping ability (Spring, 2014); reduced anxiety; and better communication with family.

Moreover, meeting information needs is important, as unmet needs are associated with negative health-related quality of life (Miyashita, Ohno, Kataoka, Tokunaga, Masuda, & Shien, 2015); high levels of anxiety and depression (Halkett et al., 2012); negative illness perceptions; and higher negative impact of cancer (Husson Mols, Oranje, Haak, & Nieuwlaat, 2014). General cancer patients report information needs impact their lives. Likewise, patients with rare diseases in a study by Huyard (2009) found their informational needs so important that they reported that it was the physician's moral obligation to provide them with authoritative, credible information. Rare disease patients in the study reported their overall health care experience would be improved if health care providers not only provided an adequate diagnosis, but also provided adequate information to the patient about the condition.

Meeting cancer patients' information needs is important; unfortunately, cancer patients often report that their needs were not adequately met (Rutten, Squiers & Hesse, 2006). In a study by O'Connor, Coates and O'Neill (2010, p. 275) of 40 patients with rectal cancer, patients reported having high levels of information needs; however, they reported that their needs were largely not met. Patients in the study reported receiving inadequate information about testing and "long-term physical, psychological and social sequelae of the disease." Likewise, Halkett's et al. (2012) study of breast cancer patients receiving radiotherapy found that many of the women's specific information needs regarding treatment effects were unmet or only partially met. Husson et al. (2014) found that nearly half of thyroid cancer survivors responded that they were, “not at all,” or “little satisfied” with the amount of information they received about thyroid cancer, and in particular were less satisfied with information they received about long-term effects, recurrence, aftercare, and current information about the disease.

Like patients diagnosed with cancer, patients with rare diseases, including rare cancers, often do not have their information needs met and are dissatisfied with the information they have received. Many patients with rare diseases report finding it difficult to obtain information about their rare conditions (Armstrong, Rochnia, Harries, Bundock, & Yorke, 2012; Budych, Helms, & Schultz, 2012; Feinberg et al., 2013; Huyard, 2009; Lim, Downs, Li, Bao, & Leonard, 2012; Mooney, Poland, Spalding, Scott, & Watts, 2013).

For example, Huyard (2009) conducted semi-structured interviews of 29 patients and 15 parents of patients diagnosed with rare diseases. The study found that patients with rare diseases expect not only to be well treated as patients, but to be listened to, taken seriously, and supported and informed according to need (Huyard, 2009). Patients who reported being dissatisfied with the diagnosis experience with their physician reported, among other reasons, “inadequate provision of information about the disease (Anderson, Elliott, & Zurynski, 2013, p. 4).”

In a rare cancer blog entry, Jan Geissler, Director of the European Cancer Patient Coalition states, “I believe that empowerment of patients is a pre-requisite for health, and access to high quality health information is fundamental to achieve this. [Patients] regard access to information as a fundamental right, as long as it meets stringent quality principles. Patients, especially those with a rare disease, are particularly isolated and vulnerable without effective access to information” (Geissler, 2010).

As shown above in examining information satisfaction and met and unmet needs of cancer patients and patients with rare diseases, meeting patients' information needs is important as unmet needs and low satisfaction with health information can be associated with many negative factors, including reduced satisfaction with care, trust in physician, and health-related quality of life (Husson Mols, & Van de Poll-Franse, 2011). These three factors are important in the overall health of cancer patients, as described below.

Patient satisfaction. Varying definitions of "patient satisfaction" exist due to the difficulty of defining the term "satisfaction" (Williams, Weinman & Dale, 1998). Bredart, Bottomley, Blazeby, Conroy, & Coens, (2005, p. 2121) define patient satisfaction as, "the extent to which an individual's health care experiences match his or her expectations." Kamo, Dandapain, Miksad, Houlihan, & Kaplan, (2010) expand this definition to encompass health care experiences increasing the possibility of the patient receiving favorable health outcomes. In general, patient satisfaction is used for measuring the quality of health care received by patients (Prakash, 2010).

The literature shows that patient satisfaction is important because it is related to patients' following of physician recommendations, cancer treatment adherence, and health status improvement (Bleich, Ozaltin, & Murray, 2009; Bredart et al., 2005). Bleich et al. (2009) found that in addition to patient satisfaction being related to increased treatment adherence, patients who are satisfied with their healthcare experience are generally more compliant and cooperative. Gupta (2012, p. 766) states, "…there has been a recent rise in awareness that patients' satisfaction with the quality of the services they receive at a healthcare institution can affect their treatment outcome." Results of Gupta's (2012) study of patients diagnosed with pancreatic cancer showed that patient satisfaction was an independent predictor of survival.

Patient trust in physician. While trust is important in any relationship, it is a particularly integral and important part of the patient-physician relationship (Anderson & Dedrick, 1990; Hillen, de Haes Hanneke, & Smets, 2011; Kaiser, Rauscher Jacobs, Strensk, Ferrans, & Warnecke, 2011; Pearson & Raeke, 2000). There are varying definitions of the concept of trust in a physician. Trust can be defined as the patient's set of beliefs about how their physician will perform in a certain way (Pearson & Raeke, 2000). Trust may also be defined more affectively using the patient's "reassuring feelings" or "feelings of confidence" in the physician as a basis of trust (Pearson & Raeke, 2000). Definitions also encompass qualities which patients expect their physician to possess and can include: competence, compassion, maintenance of privacy and confidentiality, reliability, dependability, and good communication skills (Pearson & Raeke, 2000). A study by Hillen et al. (2011) found that other components of physician trust among patients were perceived technical competence, honesty, and patient-centered behavior.

It is important for patients to have trust in their physicians. Pearson and Raeke (2000, p. 512) state, "Theoretically, patient trust should serve to reinforce the functioning of the clinical relationship as a health partnership, thereby increasing the probability of patient satisfaction, treatment adherence and improved health status, while decreasing the likelihood of leaving the physician's practice or withdrawing from a health plan." The literature shows that increased patient trust leads to greater treatment adherence (Pearson & Raeke, 2000; Kaiser et al., 2011; Hillen et al., 2011); patient satisfaction (Pearson & Raeke, 2000; Kaiser et al., 2011; Hillen et al., 2011); facilitated communication; medical decision making; and decreased patient fear (Hillen et al., 2011). In a study of patients with cancer, Hinnen, Pool, Holwersa, Sprangers, and Sanderman, (2014) found that low levels of trust in physician were associated with increased emotional distress and increased physical limitations. Kaiser et al. (2011) found that a trusting relationship with a regular provider facilitates trusting relationships with other health care providers on the care team.

Health-related quality of life. Health-related quality of life (HRQL) is the subjective perception of the patient's overall wellbeing, encompassing emotional and physical health and its impact on the patient's life (European Organization for Research and Treatment of Cancer, 2014). Traditionally, the outcomes of focus were on the narrow measures of mortality and morbidity (Centers for Disease Control and Prevention, 2011). Now, measures are expanded to include outcomes of HRQL in addition to morbidity and mortality. This reflects that more people are living longer with chronic illnesses, including cancers that may have limited treatments but not a cure (Institute of Health Economics 2008). Thus, with patients living longer with illnesses, it is essential that their physical, mental, and social lives are also improved.

Measuring HRQL is important because it assesses the positive and negative impacts of chronic disease and treatment on patients' overall wellbeing. Additionally, HRQL measures assess the current health status of the patient. This is vital due to the wide variability of patients' HRQL even if they have the same disease and are undergoing similar treatment (Institute of Health Economics, 2008).

HRQL is now considered critical to patient care and researchers are beginning to explore the relationship between HRQL and patients' overall survival (Lemonnier, Lewis, Ahmad, & Sullivan, 2014). In a study of non-small cell lung cancer patients, Efficace, Bottomley, Smith, Lianes, and Legrand (2006) found that self-reported HRQL provides independent prognostic information for survival. There was an increase in deaths for those who self-reported worse HRQL (Efficace et al., 2006).

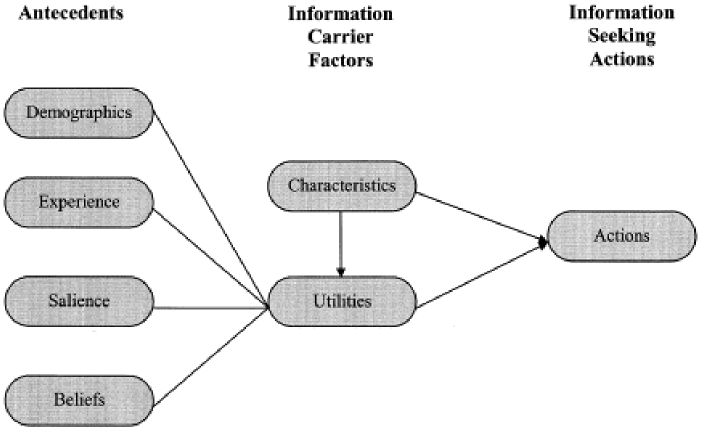
Since we know that information access and satisfaction of informational needs contribute to patients’ well-being, or can positively or adversely affect patients, we must begin to provide more/credible information for patients with rare cancers. The following section provides the conceptual framework used for this study that shows health information needs, sources, and information satisfaction in the context of health-related quality of life.

Theoretical Framework

This research study draws upon Johnson's Comprehensive Model of Cancer Information Seeking (CMIS) to conceptualize information needs and information seeking behavior of patients with rare cancers. First, an explanation of the CMIS will be presented, followed by the conceptual model that was used in this study.

Johnson's comprehensive model of information seeking. The CMIS posits that antecedents motivate patients to seek information and information carrier factors lead to information seeking actions. The CMIS model is shown in Figure 1 below and a brief overview of the model's constructs will follow.

Figure 1. Johnson's Comprehensive Model of Cancer Information Seeking



(Johnson, 1997)

Antecedents: The antecedents of CMIS are factors that motive a person to seek information and are comprised of the concepts of demographics, experience, salience and beliefs.

Demographic Factors: Research has shown that consumers' usage of sources of health related information varies by demographic factors (Johnson, 1997). For example, information searching behavior and information needs vary by age. Rutten (2006) examined informational needs of cancer patients who actively seek cancer information by socio-demographic status using

2003 HINTS survey data and found that younger aged cancer patients were more likely to seek health information than those aged sixty-five or older. Additionally, information seeking varies by gender. Females are more likely than males to search for cancer information (Rutten, 2005, 2006). Another example of demographic differences is educational level. Those with a college degree or reporting "some college" are also more likely to seek out cancer health related information than those who did not report any college experience (Rutten, 2006). Race/ethnicity also varies between those patients who search for cancer-related information online and those who do not (Castleton, 2011, Rutten, 2006).

Experience: The second antecedent in the CMIS model is experience. Direct experience predicts health information seeking. A person's own symptoms or experience with cancer may prompt him or her to seek health information. Examples include personally being screened or treated for cancer and having a friend or family member who is screened for cancer (Johnson, 1997).

Salience and beliefs: Salience and beliefs are motivational factors for seeking health information to fill a knowledge gap. Salience is the belief that health information will actually fill a knowledge gap. Beliefs encompass a person's belief that there is something that can be done to improve his or her medical condition. If a person believes that there is a medical therapy available that can improve his or her medical condition or that there is a test that can detect cancer, the person will seek health information (Johnson & Meischke, 1993).

Information Carrier Factors: First, the term "carrier" in the CMIS model is used to describe any informational source or information channel (i.e. print, video, physician). Information carrier factors are the second column of the CMIS model and are comprised of two concepts: characteristics and utilities.

Characteristics: This concept describes characteristics that health information seekers desire in the information sources they are using. For example, characteristics may include source credibility, editorial tone, or reading level of the information (Johnson, 1997)

Utilities: The second concept of information carrier factors is utility. Utility is related to the user's perceived usefulness of the information carrier. Characteristics, described above, make the information carrier useful. Describing utility, Johnson and Meischke, (1993, p. 349) ask, "… is the information contained in the medium important for the individual's purposes, relevant, and topical?" Utility of the information in CMIS leads to the concept of information seeking actions.

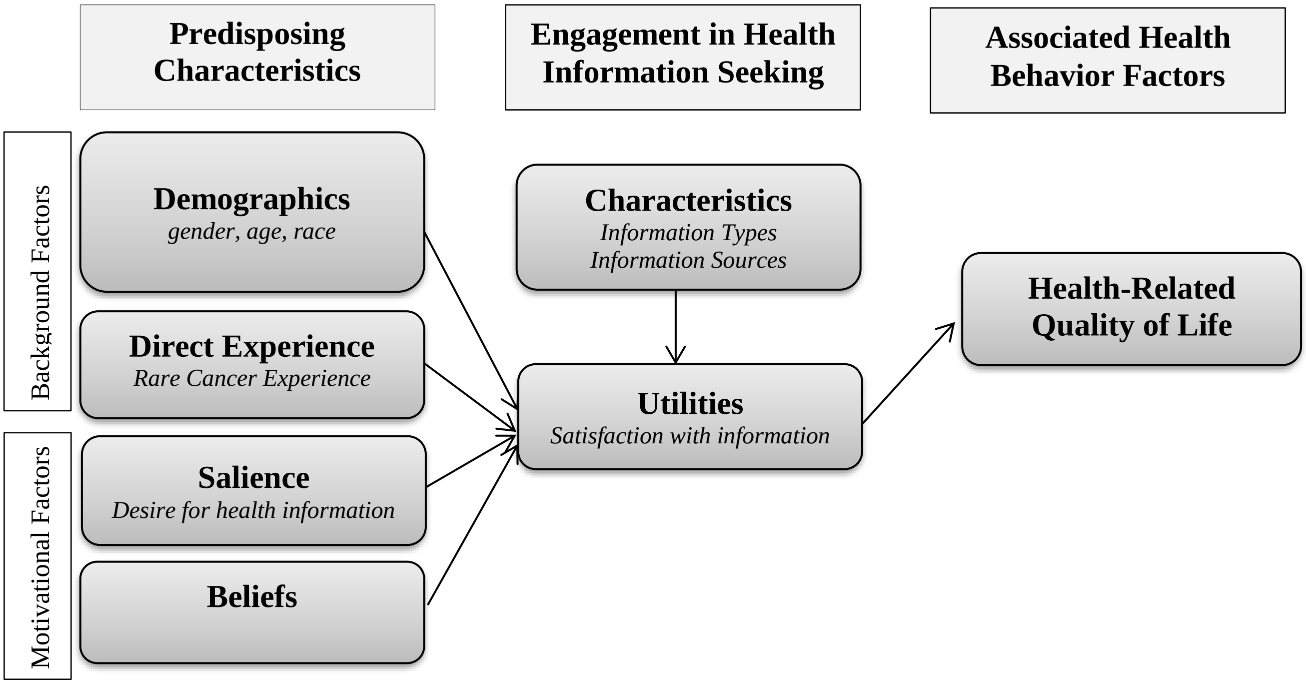
Information Seeking Actions: Antecedents and carrier factors predict information seeking actions. While the CMIS provides a framework for predicting information actions, it lacks a final outcome of patient health behavior. Anker, Reinhart and Feeley, (2011) expand the CMIS model by placing it in the context of health behaviors (see figure 2). This modification of the CMIS, integrating health behavior, is the basis for the conceptual model for this study (see figure 3) outlined in detail below.

Figure 2. Information Seeking in the Context of Health Behavior Outcomes

(Anker et al., 2011)

Conceptual model describing rare cancer patients' information seeking. Owing to a lack of theoretical models explaining information seeking in context of health, the conceptual model used for this study integrates Anker's et al. (2011) model into the CMIS to provide a conceptual framework that incorporates associated health behavior outcomes in addition to health information seeking. Figure 3 below demonstrates the model and a more detailed explanation of the model follows.

Figure 3. Conceptual Framework



In the conceptual framework used to guide this study, "predisposing characteristics" (called antecedents in the CMIS) are those characteristics that lead or motivate patients with rare cancers to seek information. These characteristics include demographics and direct experience. Information seeking varies by gender, age, and race. Direct experience can be defined as a patient's experience with rare cancers. Being screened, diagnosed, or treated for a rare cancer are all considered predictors to seeking information about rare cancers.

The motivational factors of the predisposing characteristics are salience and beliefs. Salience and beliefs provide the motivators to the background factors to actually engage in information seeking. If patients with rare cancers believe that there is information about their rare cancers (salience) and believe something can be done to improve their health, they will engage in information seeking. For example, if patients believe there is a treatment available for their rare cancers, they will seek out information about treatment.

These predisposing characteristics motivate or predict actual engagement in health information seeking (information carrier factors in CMIS). Health information seeking is defined as, “the purposive acquisition of information from selected information carriers (Johnson & Meischke, 1993, p.350).” Engagement in health information seeking is comprised of characteristics and utilities. Characteristics include information needs (types of information sought) and sources used.

The second concept of engagement in information health seeking is utilities. Utilities pertain to the usefulness of the information and answer the questions, “Was the patient satisfied with the information and did the patient receive information from his/her preferred information source?” Utilities, measured as satisfaction with information, leads to associated health behavior factors. In this study the associated health behavior factor being studied is health-related quality of life. For example, patients with information needs that are not met (unsatisfied information needs) may report a decreased health related quality of life as compared to patients whose information needs were satisfied.

Chapter III. Specific Aims and Research Questions

This research study examines the unique health information needs and sources of information of patients who have been diagnosed with rare cancers. Due to the lack of information on rare cancers for patients, this study also examines the relationship between rare cancer patients' satisfaction with health information, and their health-related quality of life.

The three specific aims of this study and their associated research questions are:

Specific Aim 1: Examine the information needs of patients who are diagnosed with a rare cancer.

RQ1: What types of cancer-related health information are desired by patients diagnosed with rare cancers?

RQ2: Do information needs vary by demographics: gender, age, and race?

Specific Aim 2: Examine the information sources used and preferred by patients with rare cancers.

RQ3: What sources do patients use to find information about their rare cancers?

RQ4: What sources do patients prefer when seeking information about their rare cancers?

Specific Aim 3: Examine patients with rare cancers' satisfaction with information and its association with health-related quality of life

RQ5: Are patients with rare cancers satisfied with the information they receive?

RQ6: Is information satisfaction related to health-related quality of life?

Based upon the literature review detailed above, it is hypothesized that rare cancer patients' unsatisfied information needs will have a negative association compared to rare cancer patients who report having a higher level of information satisfaction. For example, patients who report having unsatisfied information needs will report a lower perceived health-related quality of life as compared to patients who report a higher level of satisfied information needs.

Chapter IV. Research Design and Methods

Study Overview

This cross-sectional design study used mail surveys to assess the cancer information needs, information sources, and the relationship of health related quality of life to information satisfaction of patients diagnosed with rare cancers. Validated measures were used to collect data on cancer information needs, information sources, and source preferences. Additionally, validated measures were used to collect data on participants' satisfaction with information, as well as patient health-related quality of life. The overall purpose of this study is to describe rare cancer patients' information needs, information sources and source preferences, and to determine whether information satisfaction is associated with health-related quality of life.

Study Site

The study was conducted at the Virginia Commonwealth University Massey Cancer Center (MCC). The MCC is a National Cancer Institute (NCI) designated cancer center. In 2014, a total of 15,087 patients visited the MCC for consultations and treatments including chemotherapy, bone marrow transplantation, surgical procedures, and radiation therapy. In 2014, 5,814 new patients were seen comprising a patient population of 57% Caucasian, 38% African-American, and 5% other races.

List of Rare Cancers

The list of rare cancers used to identify eligible patients is derived from rare cancers included in the *Textbook of Uncommon Cancers* (2012) edited by Derek Raghavan MD, Medical Oncologist and President of the Levine Cancer Institute at Carolinas Health Care System. The textbook editorial board is comprised of six other oncology specialists, in addition to Dr.

Raghavan. The cancers included in the text were selected by oncologists in each cancer specialty area, with 198 cancer specialists contributing information. Each oncologist was responsible for identifying rare cancers in his/her area of specialty. All cancers, other than cancers identified in the pediatric cancer sections of this book were included in the rare cancer list used in this study.

Additionally, the compiled cancer list used for this study includes cancers from the Office of Rare Disease Research (ORDR) rare cancer list that were not included in the *Textbook of Uncommon Cancer*. Diseases on the ORDR list were included if they were an unusual cancer cell type. The following disease types on the ORDR list were excluded: those that are not specific cancer types; hereditary syndromes; syndromes that are not cancers but increase cancer risk; and precancerous conditions. Cancers were also excluded if they were exclusively pediatric cancers. Laurie Lyckholm, MD, Professor, Department of Hematology/Oncology, Holden Comprehensive Cancer Center, University of Iowa, assisted with the development of the rare cancer list that is included in this study and the inclusion/exclusion criteria for selecting cancers from the ORDR list. The list was comprised of approximately 500 different types of cancers that are considered rare.

This study was approved by the Virginia Commonwealth University (VCU) Institutional Review Board.

Study Sample

Inclusion criteria. Eligible participants (*N* = 113) were patients diagnosed with rare cancers of any type or stage being treated by a medical oncologist, radiation oncologist, or surgical oncologist at the MCC. Eligible patients must have been diagnosed within twelve months prior to being contacted for participation, aged 18 years or older, and fluent in the English language.

Exclusion criteria. Patients who did not have a diagnosis of a rare cancer were excluded due to this study specifically examining the information needs and information sources of those patients who have rare cancers. Patients who were diagnosed more than twelve months previously were not eligible because they may have satisfied a greater number of information needs given the length of time since diagnosis or may have forgotten their information needs, resulting in response bias. Patients younger than 18 years of age were also excluded from this study as this study seeks to examine the information needs and sources of adult patients, not that of children or the parents of children with cancer. Those who were not fluent in the English language were excluded since all measures used are written exclusively in the English language. Those patients known to be incarcerated currently or incarcerated at the time of diagnosis were also excluded from the study. Finally, patients were also excluded if they were deemed not appropriate for contact or participation by his/her oncologist.

Recruitment. A HIPAA partial waiver of authorization to use protected health information (PHI) was approved by the VCU IRB for purposes of identifying patients eligible to participate in the study.

To identify eligible participants, the researcher collaborated with Massey Data Analytics. The rare cancer list was used to match patients listed in the Massey Cancer Center cancer patient registry which is provided by the Massey Cancer Center Cancer Informatics Core and is updated on a six-month time delay from the patient's date of diagnosis. Each month, Massey Data Analytics emailed the researcher a password protected Excel spreadsheet containing a list of patients who were diagnosed with a rare cancer included in the rare cancer list and who met all eligibility requirements. Patients who were known to be deceased were excluded. Patients were determined to be deceased based upon VCUH hospital billing data, the Social Security Death

Index Data, and the cancer registry data vital status. Prisoners at the time of diagnosis and those currently incarcerated were also excluded. Prisoner status was determined by payer status listed as, “corrections,” and the latest available address in the billing data or registry data. The list of patients meeting eligibility criteria was also matched against prior lists sent to the researcher to exclude patients previously included in the rare cancer patient list. The MCC data analytics’ spreadsheet contained the following information: medical record number, patient name, diagnosis date, primary cancer site description, histology description, grouped histology description, behavior code (malignant), cancer stage, treatment status, and patient address.

In advance of mailing survey letters, the researcher contacted Massey Cancer Center oncologists and nurse practitioners via email and provided them details of the study. The oncologists and nurse practitioners were asked to contact the researcher if they had questions about the study, did not wish survey letters to be mailed to specific patients, or if they did not wish us to contact any of their patients with rare cancers.

This study used mail surveys to assess the information needs and information sources used by patients who have been diagnosed with rare cancers. The process for implementing the mail surveys followed the Tailored Design Method (Dillman, 2014) formerly known as the Total Design Method (Dillman, 1978). The Tailored Design Method uses concepts from social exchange theories and tailored design to motivate people to respond to written surveys. The Tailored Design Method promises a high response rate of at least 80% when following the specific detailed steps for conducting mail surveys (Dillman, 1978, Hoddinott S, Bass, 1986). By following this method, Hoddinott & Bass (1986) achieved a mailed survey return rate of 92.8%. The Tailored Design Method steps employed in this study are described below.

A timeline of mailings is found in Table 1 below. A preliminary introductory letter (Letter 1) was sent to all eligible patients introducing them to the study and providing a brief overview and contact information (phone number and email address) for them to opt out of participating in the study or to ask questions. Patients opting out were not sent any further letters regarding the study. Letter 1 also informed patients that they would be receiving a survey in the mail along with a written letter of request for participation in approximately one week. All patient and return addresses for the envelopes were hand-written and all letters and envelopes used VCU School of Medicine letterhead.

One week after mailing the introductory letter, an initial invitation letter/packet (Letter 2) was mailed to the patient. This contained a cover letter providing more details about the study, the survey booklet, a self-addressed stamped return envelope, and $2.00. The cover letter stated that by completing and returning the questionnaire, the patient was consenting to participate in the research study. The letter provided contact information, both telephone and email for the patient to ask additional questions about the study or to opt out of the study. The survey booklet contained all of the survey questions which were logically arranged in an easily understandable style (see appendix A). The survey booklet cover was tailored to the audience of patients diagnosed with rare cancers by using images to which they can relate. The survey was designed to be inviting to the patient, as well as easy to complete. The survey booklet contained a unique individual identification number at the bottom of each page linking to each patient. This identification number was used to avoid sending mailings after the thank you/reminder letter (letter 3) to those who had completed and returned the survey. A self-addressed stamped return envelope was included to lessen the burden on patients in returning the survey booklet. The return address was written on the final page of the survey booklet in case the participant lost the return envelope. The last item included in the packet was a token $2.00 as a “pre-thank you” incentive for completing and returning the survey that patients could keep regardless of whether or not they actually completed and returned the survey.

A week after mailing the initial invitation packet, a thank you/reminder letter (Letter 3) was mailed to everyone regardless of whether or not the patient had returned the survey unless they had opted out of participating in the study. This letter served to thank those who had responded and also reminded those who had not yet completed and returned the survey to do so.

Fourteen days following the thank you/reminder letter, a follow-up reminder packet (Letter 4) was sent to the patient if he/she had not responded by that time. This packet included a brief overview letter, an identical copy of the survey booklet with the patient’s unique identification number at the bottom of each page, and another self-addressed, stamped return envelope. This letter served as a reminder and also included another survey and return envelope in case the original survey had been lost or discarded.

A final reminder (Letter 5) was mailed four weeks after the follow-up reminder packet was sent to patients who had not yet returned their surveys. This letter served as a reminder to complete and return the survey and to let the patient know that we would not be contacting him/her further regarding the study. Applying the Tailored Design Method process, this letter was sent using priority mail. The purpose of a different mail delivery format is to pique the attention and interest of the recipient, emphasize the importance of the letter, and increase the likelihood that the survey would be completed and returned.

All patients deemed eligible to participate were sent letters 1-3 unless they had contacted us following Letter 1 requesting to opt out of further participation. Those not responding following Letter 3 were sent follow–up letters (Letters 4 and 5). Letters 1-3 also followed the timeline suggested by the Tailored Design Method. Due to a back-up in receiving mail at the university, completed surveys were delivered in large batches of ten or twenty, rather than individually as they arrived at the post office. These mail batches arrived in infrequent intervals, approximately every six to eight weeks. Due to this mail delivery constraint, and by strictly following the Tailored Design Method suggested time frame, some patients were sent Letter 4 (which contained an identical survey) when they had already completed and returned the first survey sent with Letter 2. This led to confusion and three patients returned both the first survey sent to them and the reminder survey. As a result, we adjusted the time frame for mailing Letters 4 and 5 based upon the return rates of surveys. Letter 4, which included the reminder survey, was not sent until after approximately 50% of the surveys sent following Letter 3 were received. The final letter, Letter 5 was sent approximately eight weeks following Letter 4 to allow time to receive surveys in the mail. This was intended to reduce participant confusion and ensure we no longer received duplicate surveys.

Table 1: Timeline of mailings following the tailored design method

Letter	Description	Mailing Timing (weeks)
Letter 1	Introductory Letter	1
Letter 2	Recruitment Letter and Survey	2
Letter 3	Thank You/Reminder Letter	3
Letter 4	Reminder Letter and Survey	5
Letter 5	Final Reminder Letter Priority Mail	9

Data received from the returned surveys were entered via key punch method into an Excel spreadsheet. Because three surveys were duplicates that arrived in batches (without a date postmark on the envelopes) it was difficult to determine which was the first survey the participant completed, so the researcher randomly selected one from each set of duplicates and entered the data received from the randomly selected surveys into the spreadsheet. The duplicate survey was stapled to the back of the survey used for data entry and was marked, “duplicate.” Upon completion of data collection and data entry, data were then imported into JMP (Version 12.0) statistical software for analysis.

Meeting recruitment goals. A plan was made for meeting recruitment goals. According to the MCC patient census records, a conservative estimate of 1030 patients (13.83%) were diagnosed with rare cancers listed on the Office of Rare Disease Research cancer list over a three-year time period, corresponding to an average of approximately 343 rare cancer patients annually eligible to participate in the study. Assuming a conservative estimate of a 50% participation rate, 172 patients annually were likely to participate in this study. This means that the needed sample size (N=100) for this study would easily be met. Attrition was not a consideration because the subjects were only surveyed at one-time point.

Sample Size and Power Analysis

Based upon the power analysis, a final sample size of 100 was used for this study. The power analysis calculations were based on research question number two, “Do information needs vary by demographics (gender, age, and race)?” since this research question requires the largest sample size to detect a significant effect. The sample size of 100 provides 97% or higher power to detect medium or moderate (range: .23 - .38) effect sizes. The power analysis type 1 error rate was .05.

Data Collection Procedures and Measurement

The mail survey session was anticipated to take approximately ten to fifteen minutes to complete. By completing and returning the survey, patients were informed they were consenting to participate in the study. Data were collected on: 1) socio-demographics; 2) cancer information needs; 3) patients’ received information and information satisfaction; 4) information sources used and preferred; and 5) health-related quality of life.

Variables and Measures

The following socio-demographic data were collected from the patient via survey: gender, age, race, type of medical insurance coverage, employment status, and income.

Description of Measures and Scoring (See appendix A for the survey containing measures and questions used in this study)

The European Organization for Research and Treatment of Cancer (EORTC) Quality of Life Questionnaire-Information scale (EORTC QLQ- (Information) INFO25).

Cancer Information Received, Information Needs, and Information Satisfaction were assessed using standardized and modified questions from the European Organization for Research and Treatment of Cancer (EORTC) Quality of Life Questionnaire-Information scale (EORTC QLQ-INFO25). The EORTC QLQ-INFO25 measure is valid and reliable. The instrument correlated with the EORTC Cancer Inpatient Satisfaction questionnaire (IN-PATSAT32) (r>.40), showing convergent validity and had a low correlation with the EORTC general cancer scale (EORTC QLQ-C30) showing divergent validity. Internal consistency (Cronbach’s alpha>0.90) of all items combined proved reliability of the measure (Arraras, Greimel, Sezer, Chie, & Bergenmar, 2010). The measure has been used in studies of patients with various types of cancers and in culturally diverse populations (Arraras et al., 2010; Husson et al., 2014).

Cancer Information Received: This 25 question instrument asks the stem question, “During your current disease or treatment, how much information have you received on…” followed by health information types such as “diagnosis,” “treatment,” and “procedures.” Question responses are measured on a four point Likert-type scale anchored by 1 (not at all) on one end and 4 (Very much) on the other.

In order to assess additional information received, four stem questions were added to this measure: 1.) The effects of treatment on fertility (the ability to have a baby); 2.) Other treatment options; 3.) Financial, insurance, and work-related resources; and 4.) Whether family members are at risk for cancer.

Cancer Information Needs: In addition to the above questions that assess information provision, the EORTC QLQ-INFO25 questions were modified to assess information needs. The questions in this instrument were modified to ask, “During your current disease or treatment did you/do you need information on…” Responses for these questions are on a four point Likert-type scale anchored by 1 (not at all) on one end and 4 (Very much) on the other.

Information Satisfaction: Information satisfaction was also assessed using the EORTC information received question, “Were you satisfied with the amount of information you received?” This question was modified to “Overall were you satisfied with the information you received?” to assess general information satisfaction. The original EORTC information satisfaction question was on a four-point Likert-scale ranging from “not at all” to “very much.” The response options for this survey were dichotomized to a “Yes” or “No” response for ease of response for participants and to facilitate analysis.

EORTC QLQ- INFO-25 Scoring:

Scoring EORTC QLQ-INFO25. Following directions from the manual, scores for both information needs and information received were linearly transformed into a 0-100 scale. First, a raw score, which is the mean of the component items was calculated for each scale: information about disease, information about medical tests, treatment, other services, different places of care, and things you can do to help yourself. The raw scores were then linearly transformed into a 0-100 scale. Higher scores indicate a higher information need and higher information received.

$Raw\ Score\ (RS)=(I_1+I_2\ldots I_n)/n$

Where I=component item (Likert-score) and n=the number of questions included in the scale

$Linear\ Transformed\ Score\ (S)=\{RS-1\}/range\}*100$

The range equals the maximum possible response value minus the minimum possible response value. Therefore, a 1-4 response scale's range is equal to 3 and the range for any yes/no (binary) response items is 1.

Following the EORTC QLQ-INFO25 manual, missing data were set to missing if more than half the data were missing from a particular participant for the particular scale being scored. Also, any single item was set to missing if not answered. If less than half the data for the scale were missing, then the missing data were imputed by simply ignoring the missing data and calculating the raw score by dividing by the number of questions in the scale that have a response. For example, if item 3 is missing then, RS=$(I_1+I_2+I_4)/3$.

Each individual information item was also transformed into a 100-point score. For each information item (individual questions), raw scores, (since they only consisted of one item) when converted using the formula above were essentially the Likert response score. The raw score was linearly transformed into a 100-point score using the same formula as above. Due to these being single questions only, data were unable to be imputed, so any missing items were set to missing in the database.

The Health Information National Trends Survey (HINTS). *Information Sources and Source Preferences* were ascertained using modified questions from the Health Information National Trends Survey (HINTS) developed by the United States National Cancer Institute. HINTS is a national survey that collects data regarding the use of cancer-related information in the United States. The HINTS survey has been administered in 2003, 2005, 2007, 2011 2012, and 2013. HINTS survey questions are derived from pre-existing national surveys and created by the HINTS program in 2001 (Nelson, 2004). Psychometric properties of the questions used in the HINTS survey are ensured through careful testing of each item (Health Information National Trends Survey [HINTS], 2014; http://hints.cancer.gov/faq.aspx). It is a standardized and accepted measure (Nelson, 2004). HINTS 2003 and 2005 were administered solely via telephone with questions being read to respondents and in 2007 HINTS was administered in two modes: via telephone and mail (Health Information National Trends Survey, 2014). The questions used in this study were taken from the HINTS “Cancer Communication” section and focus on cancer information sources.

One HINTS question included in the survey provides a list of sources and asks the participant to respond by indicating all the sources from which they sought information the first time they looked for information. This question was included in this study’s survey followed by two added questions (that are not in the HINTS survey) using the same sources listed in the HINTS question. These two modified questions ask what sources the patient has used to seek information; and the second modified question asks where the patient prefers to receive information. The following list of sources from the HINTS survey was included for patients to select: books; brochures and pamphlets; cancer organization; family; friend/co-worker; doctor or health care provider; Internet; library; magazines; newspapers; telephone information number; complementary, alternative, or unconventional practitioner; and other (specified).

HINTS scoring: Responses for this measure were reported as frequencies and percentages.

Health-related quality of Life

The Short-Form Health Survey (SF-12 v.1). *Health-Related Quality of Life* was assessed using the Short-Form Health Survey (SF-12 v.1) (License # QN035780) (Ware et al., 1996), a reduced instrument of the longer 36-item Short-Form Survey (SF-36) from the Medical Outcomes Study. The SF-12 measures two domains of quality of life: mental health and physical health. It correlates well with both the SF-36 physical component ($R^2 =0.911$) and the mental component ($R^2=0.918$). The 12-item measure is both valid and reliable (Cronbach's alpha>.80). The item response options are on a five-point Likert-scale ranging from 1 (excellent) to 5 (poor). The SF-12 has been used in multiple studies to assess health-related quality of life in cancer patients (Hamoen, De Rooij, Witjes, Barentsa, & Rovers, 2014; Neuner, Zokoe, McGinley, Pezzin, & Yen, 2014).

SF-12 v.1 Scoring:

Scoring for the SF-12v.1 was conducted using the client version 5.0 desktop scoring software provided by Optum (https://campaign.optum.com/optum-outcomes/what-we-do/health-surveys/sf-12v2-health-survey.html). The scoring software allows for missing score estimation and calculates both the physical component and mental health component scores of the SF-12 Health Survey. The software uses norm-based scoring, with the norm equaling 50. Higher scores indicate better health for both the physical health component and the mental health component, where lower scores indicate worse health for both physical and mental components.

Analysis Plan

Surveys and letters mailed and received are reported. The date mailed for each group of surveys sent is reported along with the number of letters sent for each mailing (letters 1-5). The number of surveys returned for each group is reported with frequencies and percentages. The number of patients reported by a family member as having died is reported for each group with percentages. Finally, the frequency and percentage of patients who requested to opt out of participating in the survey for each mailing group is reported. The participation rate is reported as frequency and percentage.

Descriptive analyses were used to characterize participant demographics: gender, age, race, employment status, insurance coverage status, and household income. Categorical variables (e.g., race, gender, employment status, insurance coverage and household income) are reported with frequency and percentages. Continuous variables: age, information needs scaled scores, information received scaled scores and health-related quality of life scores are reported with means and standard deviations.

JMP (Version 12.0) was used for all analyses. The significance level for all analyses is α=.05. Below are the research questions along with the statistical analyses that were used for each.

RQ1: *What cancer-related health information is sought by patients diagnosed with rare cancers?* Descriptive statistics were used to summarize the data regarding the health information that was reported needed and received by participants. Each cancer-related information type was listed with the corresponding percentages of information types reported that were needed by participants and the same was done for information reported received. The data are listed in two frequency distribution tables (one for information needs and another for information received) that included the type of information; number of respondents reporting each type; and percentage of respondents reporting each type of information for each response: “not at all, “a little,” “quite a bit,” and “very much.” The mean scaled scores for each subscale and item are also reported with standard deviations.

Unmet needs. The difference in information needs and information received (unmet needs) was scored by calculating scale scores for information needs and information received for each subscale: disease subscale, medical tests subscale, treatment subscale, other services subscale, and the individual items: different places of care, and things you can do to help yourself get well. The same was done for each of the individual information need and information received items. For each subscale and individual item, the average raw score and scaled score are reported. Additionally, each information need items and each information received items were linearly converted.

As previously described, larger mean scores indicate greater information needs or greater information received. To calculate unmet needs, the scaled score means of information received was subtracted from information needs scaled score means. A positive value indicates that information needs were greater than information received. A negative value indicates higher amount of information received than needed.

Information needs and received scales’ means were compared. A two-sample T-test was used to test the null hypothesis that the information needs and information received scales’ means are equal against a two-sided alternative hypothesis that the means are not equal. Data were assumed normal based on the Central Limit Theorem given that $n>30$. A Brown-Forsythe test was conducted to check for equal variances. If variances were not equal a Welch’s test was used. Information needs and received means for each scale are reported with the standard deviation. Differences between the means are reported with standard error, 95% confidence intervals, and p-value. In addition to comparing the means of the information needs and information received scales, means were also compared at the individual item level for all 23 information need items and information received items using the same analyses as outlined above for the information subscales.

RQ2: *Do information needs vary by demographics: gender, age, and race (Whites and non-Whites).*

Information needs. Information need levels were dichotomized into low needs and high needs. "Not at all" and "A little" were scored as 1 (low needs) where "Quite a bit" and "Very much" were scored as 2 (high needs). For the binary variables of gender (male and female) and race (White/non-White), a two sample Pearson chi-square test was used to compare the difference in the two groups' proportions of information needs. The null hypothesis that the proportion of no difference in level of need is tested against an alternative hypothesis that there is a difference in need level proportions. A Fisher's Exact test was used to assess differences if any group contained fewer than five subjects. For information need subscales and items, the number of subjects reporting high information needs and the number reporting low needs with a group total is reported. The proportion of those reporting high needs is reported with the proportion difference between the two groups (i.e. male high needs versus female high needs) and their associated 95% confidence intervals. The Chi-square test statistic, degree of freedom and p-value are also reported. If Fisher's Exact Test was used then no test statistic is reported, only the p-value.

Logistic regression was used to test for a relationship between the continuous variable age and level of information needs. Age was measured in whole years. Participants were indicated as having either high information needs or low information needs. Age was summarized for each information need level with sample sizes, means, standard deviations, and confidence intervals. Group high needs were summarized with frequencies and proportions. Parameter estimates, standard error, test statistics, p-values, and 95% confidence intervals are used to describe the change in odds of having high information needs due to a one-year increase in age. Data were assumed normal and each observation is independent. Linearity was assumed after the logit transformation.

Information received. Like information needs, information received was also dichotomized into "high amount of information received" and "low amount of information received" The same statistical analyses (chi-square test for the binary variables gender and race and logistic regression for the continuous variable age) were also used to assess if information received levels (high and low) vary by demographics: gender, age, and race (Whites/non-Whites). Pearson's chi-square was used to assess the null hypothesis that proportion of high levels of received information for males is equal to the proportion of high levels of received information for females against the alternative hypothesis that the two groups' proportions are different. The same analysis was conducted for proportions of received information for Whites and non-Whites. Logistic regression was used to test for a relationship between age and level of information received.

RQ3: *What sources do patients use to find information about their rare cancers?* Descriptive statistics were used to summarize the sources used by participants to find information about their rare cancers. The frequency and percentage of those who reported looking for information is described. For each type of source (physician, print, Internet, friends, etc.) corresponding frequencies and percentages of patients reporting use are reported. The data are listed in a frequency distribution table that includes: each source; number of respondents reporting using each source; and percentage of respondents reporting using each source.

RQ4: *What sources do patients prefer when seeking information about their rare cancers?* Descriptive statistics were used to summarize the data regarding preferred informational sources. For those who report searching for information, each type of source (physician, print, Internet, friends, etc.) is listed with the corresponding frequencies and percentages of source preferences reported by participants. The data are listed in a frequency distribution table that includes: source type; number of respondents reporting the source as preferred; and percentage of respondents reporting the source as preferred.

RQ5: *Are patients with rare cancers satisfied with the information they receive?* Descriptive statistics were used to summarize the data regarding participant satisfaction for information they received. Frequencies and percentages for each category (“Yes” and “No”) for the questions, “overall were you satisfied,” “did you desire additional information, and “did you desire less information” are reported. Perceived helpfulness, searching confidence, amount of effort to find information, frustration level, difficulty of information, concern about quality, and source trust are all reported on a Likert-type scale. Frequencies and percentages for each response category are reported. For all questions, frequencies and percentages are listed in a distribution table with number of respondents for each category and percentages of patients reporting each.

Information satisfaction/dissatisfaction differences by gender, race and age are also reported. A chi-square test was used to analyze differences in proportions of information dissatisfaction between males and females and to analyze differences between White and non-White participants. The frequency of reporting satisfaction and dissatisfaction are reported for each group with the group totals, group portions reporting dissatisfaction, proportion differences and 95% confidence intervals, Chi-square test statistic, degrees of freedom and the p-value. Logistic regression was used to assess the relationship of age and information satisfaction.

RQ6: *Is information satisfaction related to perceived health-related quality of life?* A t-test was used to test the association between information satisfaction and perceived health-related quality of life. A two-sample T-test was used to test the null hypothesis that the perceived health-related quality of life mean scores of patients reporting information satisfaction and those reporting dissatisfaction with information are equal. This was tested against a two-sided alternative hypothesis that the mean health-related quality of life scores is not equal between the two groups. Mean comparisons were conducted for both the physical health component scores and the mental health component scores of the SF-12 Health Related Quality of Life Measure.

Data are assumed normal using the Central Limit Theorem: $n>30$. A Brown-Forsythe test was conducted to check for equal variances. If variances were not equal a Welch's test was used. Mean scores and standard deviations for the physical component of health-related quality of life and the mental health component are reported for both the information satisfied group and the dissatisfied group. Differences between the means are reported with standard error, 95% confidence intervals, and p-value.

Chapter V. Results

Survey Mailings

Data collection began in October 2015 and was completed in May 2016. Table 2 below contains a timeline of letters and surveys sent and received. A total of 199 patients were identified as having a rare cancer and were asked to participate in the study via mailed survey. Letter 1 was mailed to a total of 199 patients identified as having a rare cancer. Letters 2 and 3 were mailed to all patients unless the patient or patient's doctor contacted us opting out of the study or if a patient's spouse contacted us informing us of the patient's death. Letter 2 was sent to 197 patients and Letter 3 was sent to 195 patients. Those not returning the completed survey and who had not opted out were then mailed follow-up letters. Letter 4 was mailed to 68 patients and of those not responding, 48 were sent the final Letter 5.

Of the 199 surveys mailed to patients, 113 were returned resulting in a 56.7% overall response rate. Six patients were reported as being deceased (3.0%), three surveys were returned as undeliverable (1.5%), and six patients opted out of participation (3.0%). Of the resulting 184 eligible patients, 61.4% participated in the study.

Table 2. Letters and surveys sent and received

Group #/Date of First Mailing	Letter1 # Sent	Letter2 # Sent	Letter3 #Sent	Letter4 #Sent	Letter5 #Sent	Surveys Received Frequency, (%)	Patients Deceased	Returned as Undeliverable	Requested to opt out of study	Participation Rate Frequency (%)
1 (10/25/15)	85	85	85	45	32	46 (54.1%)	3	3	2	46/77 (59.7%)
2 (11/12/15)	12	12	12	8	8	4 (33.3%)	0	0	0	4/12 (33.3%)
3 (11/27/15)	13	13	13	6	3	9 (69.2%)	1	0	0	9/12 (75%)
4 (12/2/15)	17	17	17	9	5	11 (64.7%)	0	0	1	11/16 (68.7%)
5 (1/13/16)	15	14	14	-	-	8 (53.3%)	1	0	1	8/13 (61.5%)
6 (2/13/16)	19	18	18	-	-	10 (52.6%)	0	0	1	10/18 (55.5%)
7 (2/29/16)	25	25	24	-	-	17 (68.0%)	0	0	1	17/24 (70.8%)
8 (3/23/16)	13	13	12	-	-	8 (61.5%)	1	0	0	8/12 (66.6%)
TOTAL	199	197	195	68	48	113 (56.7%)	6 (3.0%)	3 (1.5%)	6 (3.0%)	113/184 61.4%

Demographics

Socio-demographics of the sample are reported in table 3. Fifty-one respondents were male (45.1%) and 62 were female (54.9%). The mean age of the sample was 59.0 years, (SD=15.90). Of those responding, 69.9% self-identified as White/Caucasian, 25.7% as Black/African American, and 4.2% other races. Less than 2% identified as Hispanic or Latino. The majority of the sample reported being retired (44.1%) and thirty-one participants (27.9%) reported working full-time. Twenty-one respondents (18.6%) reported a mean annual household income of greater than $100,000. Most participants reported having employee sponsored health insurance (40.7%) followed closely by Medicare coverage (32.7%). Appendix C contains tables listing participants' cancer types, sites, and stages by demographics.

Table 3. Demographics

n=113	Mean (SD)
Age, Years	59.0 (15.90)
	Frequency (%)
Gender	
Female	62 (54.9)
Male	51 (45.1)
Race	
Black or African American	29 (25.7)
White or Caucasian	79 (69.9)
Asian	1 (.89)
Native American or Alaska Native	0 (0)
Native Hawaiian/Pacific Islander	0 (0)
Multi-Racial	2 (1.8)
Other	2 (1.8)
Hispanic or Latino Origin	
Yes	2 (1.8)
Employment Status (n=110)	
Full-time paid	31 (27.9)
Part-time paid	11 (9.9)
Retired	49 (44.1)
Unemployed	13 (11.7)
Homemaker	6 (5.4)
Medical Insurance Coverage (n=112)	
None	3 (2.7)
Private insurance	14 (12.4)
Employer sponsored insurance	46 (40.7)
Self-pay	0 (0)
Medicare	37 (32.7)
State-sponsored insurance	1 (0.9)
Virginia Coordinated Care	1 (0.9)
Medicaid	7 (6.2)
Military	1 (0.9)
Not sure	1 (0.9)
Other	1 (0.9)
Total Household Income (n= 108)	
Less than $5,000	4 (3.5)
$5,001-$10,000	8 (7.1)
$10,001-15,000	6 (5.3)
$15,001-$20,000	5 (4.4)
$20,001-$25,000	2 (1.8)
$25,001-$30,000	6 (5.3)
$30,001-$40,000	5 (4.4)
$40,001-$50,000	14 (12.4)
$50,001-$60,000	9 (8.0)
$60,001-$70,000	12 (10.6)
$70,001-$80,000	9 (8.0)
$80,001-$90,000	2 (1.8)
$90,001-$100,000	5 (4.4)
Greater than $100,000	21 (18.6)

Information Needs

Below are the mean score results for each information need for the four information need subscales with scores for each of the items comprising the scale. Table 4 shows each of the subscale scores' means and standard deviations and each of the 23 items included in the information needs scale. Information need items are reported with respondent frequency and percentage reporting needs on a Likert scale from 1 (not at all) to 4 (very much). The Likert scale for each subscale and each item was converted into a 100-point scale called the scaled score. Scores for each are continuous ranging from 0 indicating low need to 100 indicating high information need.

Information about disease. The mean information need score for the subscale, "information about disease" was found to be high at 74.62 (SD=23.8). Four items comprise this subscale with the following mean level of information need: "diagnosis about your cancer" (M=79.0 SD=28.8); "the extent of your cancer" (M=72.4, SD=33.1); "the possible causes of your cancer" (M=64.8, SD=34.8); and "whether the cancer is under control" (M=81.8, SD=28.8).

Information about medical tests. Participants had a high mean information need score for the subscale, "information about medical tests" (M=69.7, SD=28.9). Three information need items comprise this subscale with the following mean level of information need: "the purpose of medical tests" (M=68.2, SD=33.0); "the procedures of the medical tests" (M=65.8, SD= 33.3); and "the results of the medical tests" (M=75.5, SD= 28.4).

Information about treatment. The "information about treatment" subscale was also found to be high with a mean score of 64.8 (SD=26.31). This subscale included six items. The mean information need scores for the items were: "treatment modality" (M=76.6, SD=32.0); "expected benefit of treatment" (M=80.0, SD=28.9); "possible side effects of treatment"

(M=75.7, SD=31.4); “expected effects of treatment on symptoms” (M=74.6, SD=34.2); “effects of treatment on social and family life” (M=51.4, SD=36.5); and “effects of treatment on sexual activity” (M=30.0, SD=37.8).

Information about other services. The reported mean need score for the “information about other services” subscale was 33.1 (SD=29.3), which was comprised of four items. These items included: “additional help outside the home” (M=30.6, SD=34.9); “rehabilitation services” (M=27.9, SD=35.7); “aspects of managing illness at home” (M=47.4 SD=36.9); and “possible professional psychological support” (M=26.0, SD=33.4).

Single items. There were two items that were single items in the information needs scale. “Different places of care” scored a mean level of information need of 40.4 (SD=38.2) and the other single item, “things you can do to help yourself get well” had a mean level of information need of 56.5 (SD=35.1).

Added items. Four items were added to the existing information needs measure. These included information need for: “the effects of treatment on fertility” (M=13.0, SD= 29.5); “other treatment options” (M=53.3, SD=40.4); “financial/insurance resources” (M=48.2, SD=39.3); and “whether family members are at risk” (M= 49.2, SD=40.5).

Table 4: Information needs

	Not at All	A Little	Quite a Bit	Very Much	Scaled Score (0 low – 100 high) Mean (SD)
			Frequency (%)		
Disease Subscale					74.6 (28.3)
The diagnosis of your cancer (n=111)	4 (3.6)	16 (14.4)	26 (23.4)	65 (58.6)	79.0 (28.8)
The extent of your cancer (n=110)	9 (8.2)	19 (17.3)	26 (23.6)	56 (50.9)	72.4 (33.1)
The possible causes of your cancer (n=107)	10 (9.3)	31 (29.0)	21 (19.6)	45 (42.1)	64.8 (34.8)
Whether the cancer is under control (n=110)	5 (4.5)	12 (10.9)	21 (19.1)	72 (65.5)	81.8 (28.8)
Medical Test Subscale					69.7 (28.9)
The purpose of any medical tests you have had (n=110)	10 (9.1)	21 (19.1)	33 (30.0)	46 (41.8)	68.2 (33.0)
The procedures of the medical tests (n=110)	9 (8.2)	29 (26.3)	28 (25.5)	44 (40.0)	65.8 (33.3)
The results of the medical tests you have received (n=110)	3 (2.7)	20 (18.2)	32 (29.1)	55 (50.0)	75.5 (28.4)
Treatment Subscale					64,8 (26.3)
The medical treatment (chemotherapy, radiotherapy, surgery, or other modality) (n=111)	9 (8.1)	12 (10.8)	27 (24.3)	63 (56.8)	76.6 (32.0)
The expected benefit of treatment (n=111)	7 (6.3)	7 (6.3)	32 (28.8)	65 (58.6)	80.0 (28.9)
The possible side effects of the treatment (n=111)	10 (9.0)	8 (7.2)	35 (31.5)	58 (52.3)	75.7 (31.4)
The expected effects of the treatment on cancer symptoms (n=109)	13 (11.9)	8 (7.3)	28 (25.7)	60 (55.0)	74.6 (34.2)
The effects of the treatment on social and family life (n=111)	24 (21.6)	31 (27.9)	28 (25.2)	28 (25.2)	51.4 (36.5)
The effects of treatment on sexual activity (n=108)	57 (52.8)	23 (21.3)	10 (9.3)	18 (16.7)	30.0 (37.8)
Other Services Subscale					33.1 (29.3)
Additional help outside the hospital (n=111)	51 (45.9)	32 (28.8)	14 (12.6)	14 (12.6)	30.6 (34.9)
Rehabilitation services (n=110)	58 (52.7)	27 (24.5)	10 (9.1)	15 (13.6)	27.9 (35.7)
Aspects of managing your illness at home (n=111)	29 (26.1)	31 (27.9)	26 (23.4)	25 (22.5)	47.4 (36.9)
Possible professional psychological support (n=108)	59 (54.6)	25 (23.1)	14 (12.9)	10 (9.3)	26.0 (33.4)
Single Items					
Different places of care (n=109)	40 (36.7)	28 (25.7)	19 (17.4)	22 (20.2)	40.4 (38.2)
Things that you can do to help yourself get well (n=111)	16 (14.4)	35 (31.5)	27 (24.3)	33 (29.7)	56.5 (35.1)
Added Items					
The effects of treatment on fertility (n=108)	87 (80.6)	8 (7.4)	5 (4.6)	8 (7.4)	13.0 (29.5)
Other treatment options (n=109)	30 (27.5)	21 (19.3)	21 (19.3)	37 (33.9)	53.3 (40.4)
Financial, insurance, work-related or legal resources (n=110)	32 (29.1)	27 (24.5)	21 (19.1)	30 (27.3)	48.2 (39.3)
Whether family members at risk for developing cancer (n=109)	34 (31.2)	21 (19.3)	22 (20.2)	32 (29.4)	49.2 (40.5)

Information Received

In addition to information needs, participants were also asked to report the amount of information they received for each informational item for which they reported information needs. Below are the mean score results for information received for the four information received subscales along with scores for each of the items comprising the scale. Table 5 shows each of the 23 informational received items included in the scale along with respondent frequency and percentage reporting level of information received on a Likert scale from 1 (not at all) to 4 (very much). The Likert scale for each subscale and each item was converted into a 100-point scale called the scaled score. Table 5 also contains the mean scaled scores and standard deviations for each information received subscale and contains the scaled score and standard deviation for each information received items. Scores for the subscales and individual items are continuous ranging from 0 (low level of information received) to a score of 100 (high level of information received).

Information about disease. The mean information received score for the subscale, “information about disease” was high at 62.1 (SD=26.9). Four items comprise this scale with the following mean information received scores. The mean level of information received for the item “diagnosis about your cancer” was 73.6 (SD=27.4); “the extent of your cancer” was 65.2 (SD=35.1); “the possible causes of your cancer” was 42.9 (SD=37.0); and “whether the cancer is under control” was 66.0 (SD=32.8).

Information about medical tests. Participants’ mean information received score for the subscale, “information about medical tests” was also high at 72.8 (SD=25.4). Three information received items comprised this subscale with the following mean level of information need: “the purpose of medical tests" (M=71.2, SD=28.7); "the procedures of the medical tests" (M=71.5, SD=27.0); and "the results of the medical tests" (M=75.8 SD=27.1).

Information about treatment. The mean received score for "information about treatment" subscale was 59.9 (SD=27.3). This subscale included six items. The mean information received score for "treatment modality" (M=76.1, SD=28.4); "expected benefit of treatment" (M=71.5, SD=30.9); "possible side effects of treatment" (M=69.4, SD=34.0); "expected effects of treatment on symptoms" (M=65.5, SD=34.8); "effects of treatment on social and family life" (M=47.5, SD=36.2); and "effects of treatment on sexual activity" (M=27.5, SD=34.7).

Information about other services. The mean for the information received subscale, "information about other services" was low with a mean reported score of 32.36 (SD=26.892). Four items comprised this subscale of information received. These items included: "additional help outside the home" (M=27.6, SD=33.1); "rehabilitation services" (M=28.2, SD=35.0); "aspects of managing illness at home" (M=50.5, SD=34.2); and "possible professional psychological support" (M=23.6, SD=29.4).

Single items. There were two items that were single items in the information received scale. "Different places of care" mean received score was 32.4 (SD=35.1) and the other item, "things you can do to help yourself get well" was found to have a mean score of information received of 56.2 (SD=35.9).

Added items. Four items were added to the measure. This included information received for: "the effects of treatment on fertility" (M=18.2, SD=31.6); "Other treatment options" (M=38.1, (SD=36.1); "financial/insurance resources" (M=35.8, SD=35.8); and "whether family members are at risk" was (M=32.4, SD=36.9).

Table 5: Information received

	Not at All	A Little	Quite a Bit	Very Much	Scaled Score Mean (SD)
	Frequency (%)				
Disease Subscale					62.1 (26.9)
The diagnosis of your cancer (n=111)	4 (3.6)	16 (14.1)	44 (39.4)	47 (43.3)	73.6 (27.4)
The extent of your cancer (n=110)	15 (13.6)	18 (16.4)	34 (30.9)	43 (39.1)	65.2 (35.1)
The possible causes of your cancer (n=108)	33 (30.6)	33 (30.6)	20 (18.5)	22 (20.4)	42.9 (37.03)
Whether the cancer is under control (n=104)	10 (9.6)	21 (20.2)	34 (32.7)	39 (37.5)	66.1 (32.8)
Medical Test Subscale					72.8 (25.4)
The purpose of any medical tests you have had (n=110)	5 (4.5)	19 (17.3)	42 (38.2)	44 (40.0)	71.2 (28.7)
The procedures of the medical tests (n=110)	1 (.90)	26 (23.6)	39 (35.5)	44 (40.0)	71.5 (27.0)
The results of the medical tests you have received (n=110)	2 (1.8)	19 (17.3)	36 (32.7)	53 (48.1)	75.8 (27.1)
Medical Treatment Subscale					59.9 (27.3)
The medical treatment (chemotherapy, radiotherapy, surgery, or other modality) (n=109)	5 (4.6)	13 (11.9)	37 (33.9)	54 (49.5)	76.1 (28.4)
The expected benefit of treatment (n=110)	8 (7.3)	16 (14.5)	38 (34.5)	48 (43.6)	71.5 (30.9)
The possible side effects of the treatment (n=111)	12 (10.8)	17 (15.3)	32 (28.8)	50 (45.0)	69.4 (34.0)
The expected effects of the treatment on cancer symptoms (n=112)	14 (12.6)	20 (18.0)	33 (29.7)	44 (39.6)	65.5 (34.8)
The effects of the treatment on social and family life (n=108)	27 (25.0)	31 (28.7)	27 (25.0)	23 (21.3)	47.5 (36.2)
The effects of treatment on sexual activity (n=108)	57 (52.8)	25 (23.1)	14 (13.0)	12 (11.1)	27.5 (34.7)
Other Services Subscale					32.3 (26.8)
Additional help outside the hospital (n=110)	53 (48.2)	35 (31.8)	10 (9.1)	12 (10.9)	27.6 (33.1)
Rehabilitation services (n=110)	56 (50.9)	29 (26.4)	11 (10.0)	14 (12.7)	28.2 (35.0)
Aspects of managing your illness at home (n=111)	21 (18.9)	35 (31.5)	32 (28.8)	23 (20.7)	50.5 (34.2)
Possible professional psychological support (n=110)	55 (50.0)	40 (36.4)	7 (6.4)	8 (7.3)	23.6 (29.4)
Single Items					
Different places of care (n=112)	50 (44.6)	28 (25.0)	21 (18.8)	13 (11.6)	32.4 (35.1)
Things that you can do to help yourself get well (n=111)	18 (16.2)	33 (29.7)	26 (23.4)	34 (30.6)	56.2 (36.0)
Added Items					
The effects of treatment on fertility (n=106)	73 (68.9)	17 (16.0)	7 (6.6)	9 (8.5)	18.2 (31.6)
Other treatment options (n=106)	37 (34.9)	35 (33.0)	16 (15.1)	18 (17.0)	38.1 (36.1)
Financial, insurance, work-related or legal resources (n=108)	40 (37.0)	38 (35.2)	12 (11.1)	18 (16.7)	35.8 (35.8)
Whether family members at risk for developing cancer (n=106)	49 (46.2)	28 (26.4)	12 (11.3)	17 (16.0)	32.4 (36.9)

Unmet Information Needs

Unmet information needs were assessed for information subscales and each of the 23 individual items.

Information subscales. Unmet information needs (differences in information needs and information received) were calculated by computing scale scores for information needs and information received for each of the information needs and information received subscales (information disease, medical tests, treatment, other services) and the two single items (different places of care, and things you can do to help yourself get well). Greater mean scores indicate greater amounts of information needs or greater amounts of information received. The scaled score means of information received was subtracted from information needs for each information subscale and for each individual information item. A positive value indicates that information needs were greater than information received. A negative value indicates a higher amount of information received than was needed.

Patients reported a high level of information needs for information about disease with a mean scale score of 74.6, however, reported an information received mean scale score of only 62.1, resulting in a significant difference of 12.4 (p=0.0003). No other information sub-scales were significantly different between information needs and information received.

Unmet information needs for individual items. In addition to differences in information subscale scores between information needs and information received, differences in all 23 individual items of information needs and received were assessed for differences. There were significant differences in information needs among two information about disease subscale items where patients reported higher need than information received. Patients reported high mean need (64.8) for information about possible causes of their cancer, but only reported information received at a mean of (42.9), resulting in a significant difference of 21.9 (p<.0001). Patients also reported an unmet need for information about "whether their cancer is under control" with a difference of 15.79 (p=0.0306) and the added item, "whether family members are at risk for developing cancer" with a mean difference of 16.8 (p=0.0016).

Unmet needs were also reported in one treatment subscale item with patients needing more information than received about "the expected benefit of treatment" (difference=8.3, p=0.0388). Patients desired more information than received about treatment options (difference=15.28, p=0.0037) an added treatment-related item. Another added item, "information about financial, insurance, work-related or legal resources" also had a significant difference of 12.38 (p=0.0158).

Information Need Differences by Demographics

Chi-square and logistic regression analyses were conducted to assess the demographic differences in level (high need versus low need) for each of the information need sub-scales and all 23 information need items. There were differences found in information needs by gender, race, and age.

Information need scale differences by gender. For the subscale, "information about disease" there were gender differences in information needs. Ninety-two percent of males reported high needs, whereas 76.6% of females reported high needs. This resulted in a significant proportion difference of 15.4 (p=0.0378). There was also a significant difference in gender information needs in the subscale of "information about medical tests." Males (84.3%) reported higher need than females (63.3%). This difference in proportions (20.9) was found to be significant (χ^2=6.153, DF=1, p=0.0131). The only other significant difference in information needs by gender was "information needs about treatment." Males (86.2%) reported high needs and only 66.6% of females reported high needs resulting in a significant difference of 19.6 (χ^2=5.758, DF=1, P=0.0164).

Information need item differences by gender. Gender differences in information need were seen for the "extent of your cancer (item in the information about disease subscale);" "purpose of medical tests (item in the information about medical tests subscale)" "expected effects of treatment on cancer symptoms (item in the treatment subscale);" and "different places of care (single item)." Forty-three males reported high information needs (86.0%) for the extent of cancer where 39 females reported high needs (65.0%), with a proportion difference of 21 (95% CI [0.04, 0.35]). Differences in need between males and females were found to be significant (χ^2=6.334 df=1, p=0.0118). Males (82.4%) and females (62.7%) reported high information needs for the purpose of medical tests, with males reporting a higher proportion difference of 19.6 (95% CI [0.02, 0.34]) in need which was found to be significant (χ^2=5.214 df=1, p=0.0224).

Males had significantly higher levels of need for other items, with 90% of males reporting high needs for the "expected effects of treatment on cancer symptoms" and 72% of females reporting high needs for the same item (χ^2=5.099, df=1, p-value=0.0239). There was a 21.4 difference in proportions of high needs for "different places of care," with males having a significantly higher level of need (χ^2=5.313, df=1, p-value=0.0212).

Information need scale differences by race: None of the information scales showed a significant difference in race (Whites and non-Whites), although there were race differences between Whites and non-Whites, with Whites reporting less information need than non-Whites for specific items. For Whites, the mean level of information need for "aspects of managing your illness at home" was 38.4 and for non-Whites 60.6, with a significant proportion difference of

22.2 (χ^2=4.594 df=1, p=0.0321). For Whites the mean level of need for "effects of treatment on sexual activity" was 19.7 and for non-Whites 43.7, resulting in a significant proportion difference of 24.0 (χ^2=24.013 df=1, p=0.00101).

Information need scale differences by age: Older age proved significant for higher levels of information need of the subscale, "information about disease." The mean age for high needs for this subscale was 60.7 (SD=14.9, 95% CI [57.63, 63.76]) whereas the mean age for low needs was 49.4 (SD=18.3, 95% CI [40.27, 58.50]). For every one-unit increase in age, the odds of having high needs for "information about disease" increase by 0.04 (χ^2=7.00, DF=1, p=0.0082).

Information need item differences by age: Age proved significant for high information needs for information for two items in the information about disease subscale: "cancer extent;" and "whether the cancer is under control." The mean age of high needs (82.0%) was 60.7 years and for low needs (28.0%) the mean age was 52.9 (SD=14.0, 95% CI [57.64, 63.84]. A one-unit increase in age increases the odds of having high information needs for "the extent of cancer" by 1.03 (χ^2=4.85, df=1, p=0.0277). The mean age of patients reporting high levels of information need for "whether the cancer is under control" was 60.3 and the mean age reporting low needs is 49.6. The odds of having high information need for this item increase by 1.04 for each unit increase in age (χ^2=6.04 df=1, p=0.0124).

There were also age differences in information needs in three items included in the "information about medical tests" subscale: the "purpose of the medical tests;" "the procedures of the medical tests," and "the results of medical tests." The mean age reporting high needs for the purpose of medical tests was 60.7 and the mean age reporting low needs was 53.4. The odds of having higher needs for the purpose of medical tests was 1.02 greater for each one-unit increase in age. (χ^2=4.16, p=0.0413). The mean age of patients reporting high levels of information need for “procedures of the medical tests” was 61.1 and the mean age reporting low needs was 54.4. The odds of having high information received for this item increase by 1.026 for each unit increase in age (χ^2=4.19 df=1, p=0.0405). The mean age of patients reporting high levels of information need for “results of medical tests” was 61.2 and the mean age reporting low needs is 49.9. The odds of having high information need for this item increase by 1.04 for each unit increase in age (χ^2=8.20 df=1, p=0.0042).

High information needs also significantly increased with age for the item, “the expected benefit of treatment” which is an item in the treatment subscale. The mean age of patients reporting high levels of information need for “expected benefit of treatment” was 60.3 and the mean age reporting low needs is 48.6. The odds of having high information need for this item increase by 1.045 for each unit increase in age (χ^2=6.15 df=1, p=0.0132).

Information needs significantly decreased with age for “the effects of treatment on sexual activity (an item included in the treatment subscale),” and “the effects of treatment on fertility.” The mean age of patients reporting high levels of information need for “the effects of treatment on sexual activity” was 53.3 and the mean age reporting low needs is 60.3. The odds of having high information need for this item decrease by 0.972 for each unit increase in age (χ^2=4.01 df=1, p=0.0451). The mean age of patients reporting high levels of information need for “effects of treatment on fertility” was 40.8 and the mean age reporting low needs is 61.1. The odds of having high information need for this item decrease by 0.91 for each unit increase in age (χ^2=14.22 df=1, p=0.0002).

Information received differences by demographics. Information received levels (high/low) were also assessed for each of the information received subscales and all 23 information received items to assess if information received varied by demographics. Chi-square and logistic regression analyses showed that information received items varied by the demographics of gender, race, and age.

Information received scale differences by gender: There were no differences in any sub-scales by gender, although there were significant item differences in the level of information received about the "medical treatment modality (an item in the information about treatment subscale)" between males and females. Ninety-two percent of males reported receiving high levels of information received about "medical treatment" as compared to only 76.3% of females reported high levels, resulting in a significant difference of 15.3 in proportions between the two groups using Fisher's Exact Test (p=0.0376). There were no other significant differences by item in this subscale.

Information received scale differences by race: There were no differences in any of the information received sub-scales by race, however, there were differences in an individual item. There were differences between information received about "the extent of your cancer" (an item in the information about disease subscale) between Whites and non-Whites with Whites reporting receiving more information than non-Whites. Fifty-six percent of non-White participants reported receiving high levels of information about "the extent of their cancer" as compared to 76.3% of Whites. This difference of 20.4 in proportions was a significant difference in information received between the two groups (χ^2=4.670, DF=1, p=0.0307). There were no other significant differences by item in this subscale.

Information received scales by age: There was a significant difference in age for the information received scale, "information about other services." The mean age for having high level of information received for the scale was 51.6 (SD=15.6, 95% CI [45.08, 58.03]) and the mean age for low level of information received was 61.1 (SD=15.4, 95% CI [57.77, 64.38]). For every one-unit increase in age, information received for information about other services decreases by 0.03 (χ^2=6.52, DF=1, p=0.0106).

Information received item differences by age: Information received levels varied by age for three items. There were lower levels of information received for "effects of treatment on sexual activity (an item in the information about treatment subscale)" and also the "effects of treatment on fertility (an added single item)." Information received for both of those items decrease with older age. The mean age reporting high received for the "effects of treatment on sexual activity" is 49.2 and the mean age reporting low received is 61.7. The odds of having received high levels of information about this item decreased by 0.95 for each unit increase in age (χ^2=10.68 DF=1, p=0.0011). Likewise, the odds decrease by 0.95 for each unit increase in age for the information received about "the effects of treatment on fertility (χ^2=7.33, df=1, p=0.0).

Levels of information received also differed by age for the "other services" subscale item, "additional help outside the home." The mean age for high levels received for this item was 60.7 years and for low levels received the mean age was 51.5. Being an older age was significant for higher levels of information received for this item. For each one-unit increase in age, the odds of receiving higher levels of information about additional help outside the home increased by 1.04 (χ^2=5.52, df=1, p=0.0188)

Information Sources

Sources sought. Participants were asked to select all sources where they seek health information from a list of 12 various sources. Source types to choose from included: books, brochures, cancer organizations, friends, doctor, Internet, library, magazines, newspapers, telephone information number, and complementary medicine sources. Participants were also asked to specify whether they sought information from any other sources and, if so, they were prompted to list them. Out of 112 participants responding to the question, nearly all (93.8%) reported searching for health information from any source. The most sought source for information was the Internet (n=89, 80.9%), followed by doctor or health care provider (74.5%). Over a third of participants (n=43) reported using cancer organizations (39.1%) as a source for finding information and seeking information and 36.3% reported receiving information from brochures and pamphlets. The least reported sources of information for health information were complementary and alternative medicine (3.6%) and telephone information hotlines, with only two participants (1.8%) reporting using this channel to seek information. A complete list of all sources participants used to search and the frequency of reported use is found in Table 6.

Sources preferred. Participants were also asked to select one preferred choice of information from the same list of sources from which they sought information. The majority of the sample (n=91) reported preferring their “Doctor or health care provider (80.5%)” as a source for information, followed by Internet (23.0%) and cancer organizations (17.7%). The least preferred sources for health information were magazines, newspaper, telephone information numbers, library, and friends/co-workers, with less than 2% of participants preferring each of these. No participants reported preferring complementary alternative, or unconventional practitioners as a source of information. Participants’ health information source preference frequencies and percentages are found in Table 6.

Table 6: Sources preferred and used for information

	Preferred Source (n=113)	Sources Used (n=112)
Sources	Frequency (%)	Frequency (%)
Doctor or health care provider	91 (80.5)	82 (74.5)
Internet	26 (23.0)	89 (80.9)
Cancer organization	20 (17.7)	43 (39.1)
Brochures, pamphlets, etc.	9 (8.0)	40 (36.3)
Books	5 (4.4)	17 (15.5)
Family	3 (2.7)	35 (31.8)
Friend/Co-Worker	2 (1.8)	28 (25.5)
Magazines	2 (1.8)	16 (14.5)
Other	2 (1.8)	1 (.90)
Library	1 (.89)	10 (9.1)
Newspapers	1 (.89)	8 (7.3)
Telephone information number	1 (.89)	2 (1.8)
Complementary, alternative, or unconventional practitioner	0 (0.0)	4 (3.6)

Information Satisfaction

In addition to participants' information satisfaction, questions assessed perceived helpfulness of the information they received about their rare cancer. Two questions assessed whether participants desired additional information and an open-ended question asked what additional information they desired. Questions also assessed the participants' self-confidence finding health information about their condition and their perceived level of difficulty in finding and receiving the information to fill their information needs. Finally, participants' trust in the health information sources was also assessed.

Overall information satisfaction. Participants' general information satisfaction was assessed with the question, "Overall, were you satisfied with the information you received about your rare cancer?" with a yes/no response. While the majority of participants (81.1%) reported being satisfied with the information they received about their rare cancer, 21 participants (18.9%) reported dissatisfaction. Even though the majority reported being satisfied with the information they received, 46 participants (43.0%) reported they desired additional information about their conditions, while only 4 participants (4.3%) desired receiving less information. Information satisfaction data can be found in Table 7 below.

Table 7: Overall information satisfaction

	Yes N (%)	No N (%)
In general, were you satisfied with the information? N=111	90 (81.7)	21 (18.9)
Did you desire additional information? N=107	46 (43.0)	61 (57.0)
Did you desire less information? N=92	4 (4.3)	88 (95.7)

Information satisfaction by demographics. Information satisfaction by demographics can be found in Table 8 below. Information satisfaction was not found to be significantly different between race (White/non-White) and was not found to be significantly different due to age. There was, however, a significant difference between the proportion of males and females who reported dissatisfaction with the information they received. Ten percent of males were dissatisfied with information received, whereas 25.8% of females reported dissatisfaction. The difference in these proportions of 15.6 was found to be significantly different (χ^2=4.343, df=1, P=0.0372).

Table 8: Information satisfaction differences by demographics

Demographic		Information Satisfaction Satisfied	Dissatisfied	n	Proportion reporting dissatisfaction	Proportion difference (95% CI)	Chi-Square DF=1	P-value
Gender						-15.60	4.343	*0.0372**
	Male	44	5	49	10.2	[-0.28, -0.00]		
	Female	46	16	62	25.8			
Race								
	White	64	13	77	16.8	6.66	0.679	0.4099
	Non-White	26	8	34	25.5	[-0.09, 0.23]		
Age			N	mean age (SD)				
		Satisfied	90	54.5 (16.3)		95% CI [55.02, 61.88]		
		Dissatisfied	21	61.3 (14.3)		95% CI [54.81, 67.85]		

Parameter Estimates

Term	Estimate	Std. Error	ChiSquare	Prob>ChiSq	Odds Ratio	95% CI
Intercept	-2.1617716	0.994	4.73	0.0296	1.01186	[0.981, 0.988]
Age	0.01179059	0.015	0.55	0.4502		

Perceived helpfulness of information. Participants' perceived helpfulness of the information they received was also assessed using one question, "How helpful was the information?" Response categories were on a four-point Likert scale anchored with "not at all" on one end and "very much" on the other. The majority of the participants (81.5%) responded with "quite a bit" or "very much" to the question, while only 19 participants reported that the information they received was not at all helpful (0.9%) or only "a little' helpful (17.5%). Perceived helpfulness of information can be found in Table 9 below.

Table 9: Perceived helpfulness of information

	Not at All N (%)	A Little N (%)	Quite a Bit N (%)	Very Much N (%)
How helpful was the information? N=103	1 (0.97)	18 (17.5)	37 (35.9)	47 (45.6)

Searching confidence. Participants' self-confidence in their ability to obtain information was assessed with the question, "Overall, how confident are you that you could get advice or information about health or medical topics if you needed it?" Responses were on a five-point Likert scale anchored on one end with "completely confident" and on the other end, "not confident at all" and can be found in Table 10 below. Overall, the majority of the sample reported being confident in finding information about their rare cancer. Thirty-two participants (28.6%) reported being completely confident and 43 participants (38.4%) reported being very confident in their ability to obtain health information. Thirty participants (26.8%) reported being only somewhat confident, five participants reported being only a little confident (5.4%), and one participant (0.9%) reported being, "not at all" confident in their ability to find health information.

Table 10: Searching confidence

	Completely N (%)	Very N (%)	Somewhat N (%)	A Little N (%0	Not at All N (%)
How confident are you that you could get advice or information? (n=112)	32 (28.6)	43 (38.4)	30 (26.8)	6 (5.4)	1 (.90)

Amount of effort to find information. The amount of effort perceived to find information was assessed by having participants respond to how much they agree with, "It took a lot of effort to get the information you needed." Responses were on a four-point Likert scale anchored on one end with "strongly agree" and "strongly disagree" on the other end. Although the majority of the participants (67.0%) reported having confidence in their ability to obtain information, 47 participants (44.0%) responded they "strongly agree (17.8%)" or "somewhat agree (26.2%)" it took a lot of effort to obtain the information they needed.

Level of frustration in finding information. Nearly half (44.4%) of the participants also reported a level of frustration finding information. Participants were asked to respond on a four point Likert scale from "strongly agree" to "strongly disagree" to the question "You felt frustrated during your search for information." Twenty-one participants reported they "strongly agree (19.4%)" and 27 participants reported they "somewhat agree (25.0%)" they were frustrated during their search for the information.

Difficulty understanding information found. Participants reported on a four point Likert-type scale from strongly agree to strongly disagree to the question, "The information found was hard to understand" to ascertain participants' difficulty understanding the information they received. Although not a majority, nine participants responded they "strongly agreed (8.3%)" and 34 "somewhat agreed (31.5%)" that the information found was difficult to understand.

Quality of Information. Participants were asked to report their level of concern about the quality of information received with the stem question, "You were concerned about the quality of the information." Question response format was on a four point Likert scale anchored by "strongly agree" on one end and "strongly disagree" on the other end. Responses were mostly evenly distributed with 27 participants responding, “strongly agree (25.0%)” and 28 participants responding, “somewhat agree (25.9%).” About half of the respondents reported, “somewhat disagree (25.0%)” or “strongly disagree (24.1%).” Search effort, frustration with finding information, understanding of information, and information quality can be found in Table 11 below.

Table 11: Search effort, frustration, understanding, and information quality

	Strongly Agree N (%)	Somewhat Agree N (%)	Somewhat Disagree N (%)	Strongly Disagree N (%)
It took a lot of effort to get the information you needed (n=107)	19 (17.8)	28 (26.2)	36 (33.6)	24 (22.4)
You felt frustrated during your search for the information (n=108)	21 (19.4)	27 (25.0)	25 (23.1)	35 (32.4)
The information found was hard to understand (n=108)	9 (8.3)	34 (31.5)	41 (40.0)	24 (22.2)
You were concerned about the quality of the information (n=108)	27 (25.0)	28 (25.9)	27 (25.0)	26 (24.1)

Source Trust. As reported above, approximately 50% of participants were concerned about the quality of the information they received. Participants were also asked to rate their trust in sources. Sources they were asked to rate included: doctor, family or friends, newspaper or magazines, radio, Internet, television, government health agencies, charitable organizations, and religious organizations and leaders. Participants’ reported trust in sources can be found in Table 12 below. Responses were on a four point Likert-scale anchored by “not at all” on one end and “a lot” on the other end. More than any other source, participants reported trusting their doctor. Eighteen participants (15.9%) reported trusting their doctor “some” and 93 participants (82.3%) reported they trust their doctor as a source of information “a lot.” Following trust of doctors was participants’ trust in the Internet, with 58 participants reporting “some” (53.2%) trust and 20 reporting, “a lot” (18.4%) of trust. Other reported trusted sources of information are government health agencies and charitable organizations, with 66 participants reporting “some” (44.5%) or “a lot” (15.5%) of trust for government agencies and 57 participants reporting “some” (44.5%) or “a lot” (7.2%) of trust for charitable organizations. The least trusted sources reported by the sample were radio, with 91 participants reporting “not at all” (46.2%) or “a little” (40%) trust and television, with 44 participants (40.7%) responding “a little” and 36 participants (33.3%) reporting “not at all.”

Table 12: Trust in information sources

Frequency (%)	Not at All	A Little	Some	A Lot
Doctor (n=113)	0 (0.0)	2 (1.8)	18 (15.9)	93 (82.3)
Internet (n=109)	6 (5.5)	25 (22.9)	58 (53.2)	20 (18.4)
Government health agencies (n=110)	17 (15.5)	27 (24.5)	49 (44.5)	17 (15.5)
Family or Friends (n=110)	16 (14.5)	37 (33.6)	48 (43.6)	9 (8.2)
Charitable organizations (n=110)	23 (20.9)	30 (27.2)	49 (44.5)	8 (7.2)
Religious organizations and leaders (n=108)	38 (35.2)	35 (32.4)	30 (27.8)	5 (4.6)
Television (n=108)	36 (33.3)	44 (40.7)	27 (25.0)	1 (.92)
Newspaper or magazines (n=110)	20 (18.2)	49 (44.5)	41 (37.2)	0 (0.0)
Radio (n=108)	50 (46.2)	41 (40.0)	17 (15.7)	0 (0.0)

Health-Related Quality of Life

Health-related quality of life was assessed using the SF-12 and was scored using scoring software. Scores are norm-based with a norm mean of 50 (SD=10.0) and range from 0 to 100. Higher scores indicate better health for both the physical health component and the mental health component, where lower scores indicate worse health for both physical and mental components. The overall health-related quality of life physical health mean component score for the sample was M=40.0 (SD=11.6) and the mean mental health component score for the sample was M=49.6 (SD=9.7). Health-related quality of life scores and information satisfaction can be found in Table 13 below.

Information Satisfaction and Health-Related Quality of Life

Two-sided t-tests were used to compare health-related quality of life (physical health component and mental health component) scores between the information satisfied group and the information dissatisfied group. The mean physical health component score for the information satisfied group was M=39.8 (SD=11.7) and the mean physical health component for the information dissatisfied group was M=41.0 (SD=10.9). A difference of -1.2 (SE=2.8, 95% CI [-6.93, 4.48] between the two groups was shown not to be significant (t_{106}=0.42485, p=0.6718). Likewise, the 1.2 difference (SE=2.4, 95% CI [-3.51, 6.10] between the information satisfied and information dissatisfied groups' mental component score was found to not be significant (t_{106}=0.5335, p=0.5948).

Table 13: Information satisfaction and health-related quality of life

	All Participants HRQL Score (SD) n=111	Information Satisfied Group HRQL Score (SD) n=88	Information Dissatisfied Group HRQL Score (SD) n=20	Group Difference SE [95% CI]	t (DF)	p-value
Physical Component Scale (0-100) (PCS)	40.091 (11.645)	39.894 (11.776)	41.067 (10.901)	-1.223 2.879 [-6.932, 4.485]	-0.42485 (106)	0.6718
Mental Component Scale (0-100) (MCS)	49.69 (9.704)	49.949 (9.700)	48.655 (10.171)	1.293 2.424 [-3.513, 6.100]	0.5335 (106)	0.5948

Chapter VI. Discussion

Information Needs and Information Received

This study aimed to fill an important gap in the literature by identifying the types of cancer-related health information that patients diagnosed with rare cancers need in order to improve information provision to this often over looked population. In addition to identifying the needs of patients with rare cancers, this study also explored the levels of health information patients reported receiving. Owing to noted discrepancies in reported information needs and information received, gaps were subsequently examined to identify unmet needs in this population in order to gain further insight into ways to meet the information needs of patients with rare cancers.

Information needs. Participants in this study had high needs for most subscales and items and very high needs were reported for subscales “information about disease,” “information about medical tests,” and “information about treatment.” Specifically, participants had very high needs for the individual items, “whether the cancer is under control,” and “the expected benefit of treatment.” These categories of information, particularly “information about disease” and “information about treatment” are cancer-specific types of information, which are more difficult to find and obtain than other subscale information such as “information about other services.” This is particularly true for rare cancers where there may be very little consumer-level cancer-specific information available. These high reported needs demonstrate the importance for information provision of cancer specific information to patients with rare cancers. While most needs were high, participants in this study reported lower need for “information about other services” and single subscale item, “different places of care.” These reported lower needs may be a reflection of where patients are in their health care journey and only a small sub-set of patients may need access to other services or other places of care. While these needs are reported as low, health-care providers should still be cognizant of patients who may need access to this type of information.

Similar to participants with rare cancers in this study, studies of general cancer patients show that cancer-specific information is reported as a high need. General cancer patients report desiring to learn more about their specific cancer type including etiology, physical effects, diagnosis, and symptoms (Rutten et al., 2005). Likewise, general cancer patients also report high need for treatment-related information including specific cancer treatment and treatment options (Maddock et al., 2011) similar to the high levels of need found in patients this study of patients with rare cancers, emphasizing the continued importance of information provision to patients with general and rare cancers.

Information received. Participants reported high levels of information received for the subscale "information about medical tests," and moderately high levels of information received for "information about disease," and "information about treatment." This moderate to high level of information received reported demonstrates that healthcare providers are delivering high levels of information and most likely directing patients to other sources of information about their rare cancer. This also demonstrates that this level of information provision should be maintained.

Information needs/received by demographics. This study showed that information needs and information received varied by demographics, specifically by gender, race and age. There were differences in information needs and information received between males and females, Whites and non-Whites, and by age. These gender, age, and race differences in information needs can inform healthcare providers and information professionals so they can tailor information for specific demographic groups based upon greater identified need. These demographic differences can also help inform website development when creating web portals of information about rare diseases so specific demographic groups can be targeted and provided with potentially needed types of health information.

Gender. One surprising finding from this study is that men reported having significantly higher levels of information need for the scales: information about “disease,” “medical tests,” and “treatment,” and also many information items including the “extent of cancer,” “the purpose of medical tests,” “expected effects of treatment on symptoms,” and “different places of care.” This number of differences demonstrates that in some circumstances males have higher information needs. This finding was so surprising that the researcher verified that the coding of gender of participants had not been inadvertently switched anywhere throughout the process.

The research literature of the information needs of general cancer patients demonstrates information needs vary by gender, with females reporting higher levels of information need than men (Rutten, 2005, 2006). Females also report needing general cancer site information and psychological support at a higher level than men, whereas men report higher treatment information needs (Rutten, 2006). The findings of this study conflict with gender differences of information need found in studies of general cancer patients.

Not only do females report higher levels of information need for cancer information, research studies show that females are more likely to have higher information need in other non-cancer areas of health (Tong, Raynor, & Asiani, 2014; Ek, 2013; and Manierre, 2015). Very few studies have been conducted to assess the reasons gender based discrepancies exist in health information needs. Few theories exist that may explain gender differences but one theory is gendered perceived risk of illness (Manierre, 2015). If men do not perceive a high risk or threat from their condition, they may not be motivated to seek information. Men diagnosed with rare cancers, as in the case of this study, may have a higher perceived risk due to the fact that their cancer is considered rare. One concept in Johnson's Comprehensive Model of Cancer Information Seeking is perceived risk and increased perceived risk is a motivational factor of searching for information.

For healthcare information provision, this finding of higher information need among males is important. Healthcare providers and information professionals may need to be cognizant that with rare cancers, males may desire more information. Given the fact that this finding of greater information needs for males is very different from other research on general cancers, more research on the information needs of patients with rare cancers should be investigated. Interestingly, although men in this study reported higher information needs, females actually reported a higher level of dissatisfaction with information provision than did males in this study which should also be further explored in future studies.

Men reported higher need for many information items, however, there was only one item where males received higher levels of information. Specifically, males reported receiving more information about "medical treatment" than females.

Race. This study found that Whites and non-Whites in this population largely needed and received the same levels of information for most information items, however, there are differences in needs for two information need items and one received item. Non-Whites have greater information needs for "aspects of managing your illness at home" and "effects of treatment on sexual activity." Whites reported receiving more information than non-Whites for only one item, "the extent of your cancer."

Likewise, a study by Asare, Peppone, Roscoe, et al (2016) of patients diagnosed with general cancers found there were information need differences between Whites and Blacks. Blacks reported needing significantly more information about cancer tests, follow-up, healthy living, stress management, and handling stigma.

Being cognizant of these differences of information need by race can help healthcare professionals be more proactive in providing information. The information need differences between race found in this study allow providers to target specific racial groups with tailored health information that will meet their identified information needs.

Age. The research literature also shows there are age differences in information needs, with older individuals reporting higher needs for information about treatment and younger individuals most likely to report higher needs for information about general cancer (Rutten, 2006). Rutten (2006) examined informational needs of cancer patients who actively seek cancer information by socio-demographic status using 2003 HINTS survey data and found that younger cancer patients were more likely to seek health information than those who are sixty-five or older.

This study also found significant age differences in information need items. The likelihood of reporting high needs for, "whether the cancer is under control, "the procedures of the medical tests," "the results of the medical tests," and "expected benefit of treatment" increased with age. This study found that younger age was significant for higher needs about "effects of treatment on sexual activity," and "the effects of treatment on sexual activity." Also, younger participants in the study reported receiving this information at higher levels than older participants. The likelihood of reporting high needs for the subscale "information about other services" and the "information about other services" subscale item, "additional help outside the home” increased with age. Being aware of information differences in needs and received for patients with rare cancers can help providers tailor information for younger and older patients so that needs can be met for all age groups.

Participants in this study of all demographics (gender, race, and age) report high levels of need and received information, however, a minority of the participants report being dissatisfied with the information they received. This prompted the researcher to explore where there were unmet needs. Identifying unmet needs provides additional information where information provision should be increased.

Unmet Information Needs

Although analyzing unmet information needs was not part of the research aims of this study, the findings are intriguing. The unmet needs found in this study provide clarification regarding how provision of information to patients with rare cancers should be improved. Examining the unmet needs found in this study (information need scores greater than information received scores) for the information sub scales, the largest and statistically significant unmet need was found in “information about the disease.” This unmet information need is not surprising in the population of patients diagnosed with rare cancers. The literature shows that patients with rare diseases and general cancers often express specific disease information as an important need, but report it is often unmet. For example, Anderson et al. (2013) reported that patients with rare diseases were dissatisfied with their diagnosis experience. One of the reasons they reported dissatisfaction was that they did not receive adequate information about their specific rare disease. A study conducted by Shea-Budgell, Kosarus, Myhill, & Hagan (2014) about information needs of patients with general cancers found that 43% reported searching for information on their specific cancer type.

All other information needs and received scale comparisons were not shown to be statistically significant for unmet needs. This may be due to the fact that more information is available about each of these information types because most would be the same types of information given to patients diagnosed with general cancers. For example, information about different places of care, other services, and things you can do to help yourself get well may be easier to obtain because they are general and not disease-specific informational items.

Disease information items: Participants in this study of patients with rare cancers reported unmet needs in two particular disease information items. Patients reported high information needs for etiology (possible causes of their cancer) and whether the cancer is under control. Given that little is known about many rare cancers, cancer etiology may not be known or may be very difficult information to locate. For example, one rare cancer, Multicentric Castleman Disease has no known cause. While there are many recent theories as to the etiology of the disease, currently it is unknown. Patients or their healthcare providers would have to search the current medical literature to assess the current theories of the etiology of Castleman Disease and for any other rare cancers where etiology is unknown.

Treatment information items: Two treatment need items were shown to be unmet in this study: “other treatment options (an added item),” and “the expected benefit of treatment.” Treatment information has been found to be very important among patients diagnosed with general cancers. Treatment information was reported as the most important type of information, more than any other information need in a study by Shea-Budgell, et al. (2014).

In addition to desiring information about specific treatment options offered by their own physician, patients also desire information about all treatment options available for their conditions (Castleton, Fong, Wang-Gillam, Waqar, & Jeffe, 2010). Many rare cancers have few treatment options available. This may be one reason why patients may experience difficulty finding treatment information about specific rare diseases. Likewise, there may be a lack of knowledge regarding treatment options among many oncologists who do not see many patients with any particular type of rare cancer, especially very rare cancers. Treatment option information may also be difficult to find if treatments are very new and may only be found in the latest medical literature.

Likewise, the "expected benefit of treatment" may not be fully known, particularly for many rare cancers. These cancers often do not have specific United States Food and Drug Administration (FDA) approved treatments and off-label treatments may be used to treat a rare cancer. For example, oncologists may treat a rare cancer with a drug that is approved for use in another related type of cancer, basing treatment options on a few case studies found in the research literature. There is little research showing expected outcomes of treatment with these drugs for the particular rare cancer because no randomized controlled trials have been conducted for many rare cancers. Benefits may be largely unknown resulting in patients having unmet needs about the benefits of treatment.

Financial, insurance, work-related, and legal resources information item: The only other statistically significant item where an unmet information need was found was the added item regarding financial, insurance, work-related, or legal resources information needs. Interestingly, this was the only item of information need found to be significantly unmet that would be considered not applicable to a specific cancer. For example, this type of information can be answered with general informational resources. Though reported needs for this item were at a moderate level, information received was reported as very low compared to need.

Financial, insurance, work-related, and legal resources information may be particularly important for the rare/genetic disease population due to many significant barriers throughout their healthcare journey (Schaefer, 2012) which may lead to greater financial burden, insurance, and work-related issues. These patients may require more tests to diagnose their condition, need to travel long-distances to an oncologist who specializes in their condition, or face battles with insurance companies over treatments that are not covered because they are considered experimental or off-label for a rare cancer. This may result in greater financial burden and loss of time away from work if, for example, patients have to take off more time to travel for tests and to see specialists and have to pay more money out of pocket. Additionally, patients living with rare diseases/cancers experience work-place and health insurance discrimination (Williams, 2014).

This unmet information need is an important consideration as it can be addressed. For example, patients can be referred to social workers or free legal assistance centers that can address issues related to finances, insurances, and work. Public and consumer health libraries can also direct patients to information about financial, insurance, legal, and work-related resources as there are many online resources that address these information needs. By recognizing this as an information need in this population, an emphasis on disseminating this information should be made a priority to ensure patients with rare cancers have easy access to financial, insurance, legal, and work-related information resources.

Information Sources

Source preference and trust: While the majority of participants (80.9%) reported *seeking* information from the Internet, only 23% reported this as a source they *preferred*. Participants reported a preference for receiving information from their healthcare provider over all other sources; however, in contrast to their stated preference, reported using the Internet over all other sources. While sources used least were also reported as preferred least, patients may not be receiving sufficient information or they may perceive they are not receiving a sufficient amount of information from their preferred source, their healthcare provider. This may be difficult to ascertain given some patients may believe there is more information to access than is available and some may desire to know everything about their rare cancer.

Furthermore, participants reported they trust their healthcare providers more than any other source including the Internet. The responses from this study demonstrate that patients are seeking information from the Internet, which is not their preferred source of information but is also not as trusted as health care providers a source for information about their rare cancers.

This discrepancy in source preference and trust is concerning; however, it is not unexpected. Responses of the patients in this study are similar with regards to source preference and trust of sources in studies of patients diagnosed with general cancers. Those with general cancers report trusting their healthcare providers more than any other source, but also report predominantly using the Internet. Studies of general cancer patients show that patients trust their doctor or healthcare provider as the most trusted source of all sources including the Internet, family friends, radio, newspaper, or television (Shea-Budgell et al 2014; Halkett et al. 2012). A secondary data analysis of HINTS survey data found that 38% of cancer patients reported seeking information from the Internet, more than any other source (Roach et al., 2009).

A study that aimed to answer why patients use the Internet as a source of information over their healthcare providers found that the healthcare provider’s empathy and amount of time spent with the patient affect whether patients use the Internet substantially more as a source for finding health information (Tustin, 2010). Owing to the complexities of many rare diseases, including rare cancers, physicians may not have the time to adequately address all information needs that patients with rare cancers may have. If patients perceive that their information needs have not been adequately addressed, patients then may seek the Internet to fill their unmet needs.

Also, due to a lack of specialist physicians for many different rare cancers, particularly those that are very rare, there may be few physicians with knowledge of the specific rare cancer. (Spring, 2014b). Healthcare providers may have to search the literature to obtain an adequate level of information but for many rare cancers may also experience difficulty finding information about the condition due to the paucity of information. They may also not have the expert searching skills to find information that may be available and may also need to turn to other sources, such as physicians, government rare disease organizations, or a health sciences librarian. With very rare cancers, there simply may not be much at all known about them to find any substantive information in the consumer health or even the medical literature.

Issues with patients searching the Internet. There are negative implications for patients not receiving information from their preferred source and then searching for information from the Internet. Studies show that patients make medical decisions based upon information they find on the Internet. Patients have been found to decline medical advice from qualified healthcare professionals, instead following the advice of information they found online. Patients report making decisions about their own or a loved one’s health based upon information they found online (Weaver, 2009; Fox, 2002). One study found that 11% of patients refused to follow the advice or discontinued their physician’s recommended treatment based upon information found online (Weaver, 2009). In another study, 68% of patients seeking online information reported the Internet had some impact on healthcare decisions and 16% reported the Internet had a major impact on healthcare decisions (Fox, 2002).

Reliable information provision for patients with rare cancers is crucial, particularly since some of these patients are utilizing Internet health websites and may potentially make decisions about their health based on this uncertain information. It is imperative that those with rare cancers be provided with access to health information portals containing high quality, accurate and credible sources of information. Patients who search the Web via online search engines may be unaware how to evaluate critically health information sources for accuracy and reliability and may thus be making decisions based on unreliable or inaccurate information.

Information Satisfaction

Though the majority of participants reported being satisfied with the information they received about their cancer, they reported frustration with finding and comprehending it. This may reflect the lack of consumer health information available online about many rare diseases including rare cancers. There are few authoritative consumer-level rare disease/rare cancer websites available and are difficult to locate without sophisticated searching skills. Patients may be overlooking these and other reliable sources of additional information about their cancers. For example, most participants in the study did not prefer or report using libraries, print resources, cancer organizations, videos, or interpersonal (communication with other patients) as sources of information. Patients may benefit from using the help of trained health science librarians to access consumer level websites about rare conditions. Most information available by doing a simple Google search of a rare disease are links to medical journal articles aimed at healthcare providers. These articles are written at a very high reading level, containing complicated medical jargon, which makes it very difficult for the lay-person to understand. This may have reflected the high level of reported difficulty understanding information among patients in the study.

Furthermore, as discussed earlier, information from the Internet may contain inaccurate, biased, or even nefarious health information. The possible dangers of navigating the Internet can be ameliorated through the use of expert search services. For example, a consumer health cancer library that provides information about complicated medical issues, current therapies, and rare cancer types and subtypes reports benefits of their expert search service's impact to its users (Volk, 2007). Volk (2007) found 96.2% of users of the service reported "some or all" information that they received through the expert searching service was not provided to them through any other informational sources (Volk, 2007) demonstrating the importance and benefit of expert searching for rare cancer information.

In addition to overlooking libraries as a potential source of finding information, few reported using cancer organizations as a source for information. Cancer organizations can be an excellent source of finding additional information about specific rare cancers. Many cancer organizations contain general cancer information but also contain information about specific cancers or can provide links to or contact information for disease-specific information and can also often provide disease specific brochures and pamphlets which are particularly helpful for those patients who do not prefer using the Internet. These organizations not only provide disease information but also often provide users with support group information and places to connect with other patients who share similar medical issues providing a source for interpersonal information about rare cancers. Patients with rare cancers can meet, communicate with, and learn from others who are experiencing similar medical issues.

Like general cancers, patients with rare cancers do not report using videos as a source of information. The low reported usage of video may be due to the lack of authoritative consumer-oriented online cancer videos and difficulty obtaining DVD cancer videos. Even if located, DVDs can often be cost prohibitive for patients. Videos are often an important resource for low-literacy patients and having access to consumer level authoritative videos may be beneficial to those patients who have lower literacy or who prefer learning through audio-visual resources.

Despite participants reporting unmet needs, feeling that it took a lot of effort to find information, and feeling that the information was difficult to understand, the majority of the participants in this study of patients with rare cancers reported information satisfaction. The number who reported being unsatisfied with information is roughly equal to other research of satisfaction with general cancer information. For example, Davies et al. (2008) report that 16% of patients diagnosed with breast or prostate cancer report that they were dissatisfied with the information they received about their cancers. A recent study showed that 34% of prostate cancer patients were dissatisfied with the information they received about their cancer, which was at a slightly greater level of dissatisfaction than what was found in this study of rare cancer patients (18.9%) (Lamers, Cuypers, Hunson, de Vries, & Kil, et. al, 2015). Even though these results are similar to patients diagnosed with general cancers, it was expected that the satisfaction with information would have been less in the rare cancer population than those diagnosed with more general types of cancers due to decreased information availability and difficulty finding information. One hypothesis about this is that participants in this study may have reported satisfaction because they were satisfied with the care they received by their oncologist and healthcare team. Due to the satisfaction of participants at the Massey Cancer Center, it may be beneficial to conduct a study of patients at other institutions to ascertain if patients with rare cancers are generally satisfied with information provision or whether information satisfaction may be an anomaly due to the fact that patients at the Massey Cancer Center are receiving adequate levels of health information from their healthcare providers.

Information satisfaction by demographics. Though men in this study reported higher information needs as discussed above, females actually reported a higher level of dissatisfaction with information provision than did males. This is also an important finding in this study as it shows that even though only 18.9% of the participants in this study are dissatisfied with the information they have received, the majority of those dissatisfied with information are females. Further research is needed to explore reasons why patients are dissatisfied with information, particularly females, so that information dissatisfaction can be addressed in the rare cancer patient population.

Health-Related Quality of Life

The research literature shows that unmet needs and low satisfaction with health information can be associated with negative health outcomes including reduced satisfaction with care, trust in physician, and health-related quality of life (Husson Mols, & Van de Poll-Franse, 2011). A recent study of prostate cancer patients focusing on information provision and quality of life found that approximately 33% of the patients in the study were dissatisfied with health information provision and those patients had a significantly worse health-related quality of life than those who were satisfied with information received (Lamers et al, 2015).

Surprisingly, this study of patients with rare cancers conflicts with findings in the literature and showed no significant difference between health-related quality of life between those patients who reported information satisfaction and those reporting information dissatisfaction. One possible reason may be due to the fact that only 18.9% of the participants in this study reported information dissatisfaction which is a low percentage of those who are dissatisfied. Lamers et. al (2015) study was a larger sample size including 999 patients with a response rate that produced a sample size of 697 participants. Of those participating, 222 (32%)

participants reported being dissatisfied with the information they received about prostate cancer. Perhaps information satisfaction and health-related quality of life should be examined in a subsequent study of information needs in a larger sample size of patients to identify if there is a correlation between information satisfaction and health-related quality of life among patients diagnosed with rare cancers. Another hypothesis of the reason for the discrepancy between this study and other studies regarding the relationship between information satisfaction and health-related quality of life is that given the six-month time delay in obtaining eligible patients for this study, patients with lower health-related quality of life may have been deceased by the time they received the survey or may have been too ill to respond. It is possible the responders were those who had a higher health-related quality of life compared to non-responders.

Study Impact

Traditionally, the rare disease/rare cancer community has been ignored and there is a general lack of knowledge about many individual rare cancers, resulting in many challenges for patients including the challenge of finding information about their condition. This long-standing neglect by the research community of the rare disease population has led to the growing trend of patient engagement in their own care and a sense of patient empowerment in the rare disease/rare cancer patient community (Ayme, Kole, & Groft, 2008). The World Health Organization defines empowerment as, “a process through which people gain greater control over decisions and actions affecting their health (1998).” On an individual level, through learning everything they can about their particular disease, patients with rare diseases become patient experts about their specific conditions (Ayme, Kole, & Groft, 2008; Spring, 2014a&b).

Patient empowerment also expands the concept of the engaged individual patient level to the greater rare disease community level. The web has created the opportunity for establishing rare disease/cancer online organizations and support groups where people with similar diseases can meet and discuss their shared experiences. These organizations also present opportunities to build collaborative relationships with physicians to create online information portals that contain patient-level information that address the information needs of the rare cancer community.

Overall, the identified information needs and unmet needs identified in this study show the importance of information provision, particularly disease-specific and treatment information to patients with rare cancers. Information provision and access to reliable information is important for this population. In an effort to provide additional information patients can be encouraged to use other reliable sources to find additional information about their rare cancer if they are interested in more information.

One easy and cost-effective way of promoting additional sources is via information prescriptions. The National Institutes of Health and the National Library of Medicine sponsor the Information Rx project (www.informationrx.org). Information Rx provides free customizable prescription pads that can be used to direct patients to rare disease information portals, government agencies, directories for rare disease organizations and nearby libraries. Healthcare providers are encouraged to write the name and stage of the cancer on the prescription so patients have the correct cancer name/spelling when searching for information. This also lessens the burden of the patient so that he/she does not have to remember the name of the cancer or how to spell it when searching online or when seeking information from other information sources.

Because patients report using the Internet as a source of information over all other sources, patients should be directed by healthcare providers to websites that contain authoritative and accurate health information. Information prescriptions can also be used to provide the patient with pathfinders to reliable online resources. As described above, a Google search may yield results containing information that is written at a very high reading level, and may even contain bias or untrue information and patients may not critically evaluate informational sources before using them to make health care decisions. While there is little information about rare cancers online, there are several portals of health information available that contain reliable information about many rare cancers. Patients should be directed to these portals.

Given the findings of this study that patients with rare cancers report needing information about their diagnosis and treatment over all other information types, providing patients with this information is important. Future efforts of rare disease organizations should focus on providing patients with basic information about rare cancers, with an emphasis on diagnosis information and treatment. The National Institute of Health's Genetic and Rare Disease Information Center (https://rarediseases.info.nih.gov/GARD/) provides a list of rare diseases including a section with links to many rare cancers. The site provides very limited information about many rare cancers; however, this may be an excellent starting point for developing more consumer health information content about rare cancers with a focus on diagnosis and treatment information.

Study Limitations

Study limitations are outlined below with efforts used to ameliorate their effects on the study. Additionally, recommendations regarding how this study could be improved are included.

Nomenclature. A few participants reacted negatively to the word "rare" that was listed on the survey and in all letters informing the patients that the study explored the cancer information needs of patients diagnosed with rare cancers. These individuals were unaware that their cancer is considered rare or thought "rare" is an actual type of cancer. Using the word "uncommon" or the phrase "not commonly occurring" may have been a better choice of terminology and would not have elicited a negative response from patients. Most patients, however, were keenly aware they had been diagnosed with a rare cancer and expressed their appreciation that this study focused on patients with rare cancers.

Six-month embargo on cancer registry list. The Massey Cancer Center cancer registry was used to identify patients eligible for participation in the study. The registry list, however, is on a six-month time delay meaning that patients are not listed in the registry until six months' post diagnosis. As a result of this delay in identifying eligible patients, several patients died prior to or after receiving the letter. Six patients were reported as having died by their spouse or other family member. It is also possible that many patients who did not respond to the survey were gravely ill or had recently died given the time from diagnosis to survey contact.

Generalizability of the study. This study may or may not be generalizable to other settings; however, since this study takes place at an academic safety net medical center, it may be representative of other similar safety net academic medical centers and generalizable to those centers.

Absence of level of education data. Originally, the researcher intended to collect data on cancer health literacy using the CHLT-6, however the CHLT-6 had not been validated for use in written surveys. The CHLT-6 was removed from the survey and the question regarding highest level of education was inadvertently left out in error. Education level is acknowledged to be an important factor in health information needs and should be addressed in future studies of patients with rare cancers' information needs.

Method limitations. This study used a cross-sectional design. While there are many advantages to using a cross sectional design such as ease of study design, relatively low cost, and no concerns about the possibility of attrition, there are some disadvantages. One important limitation is that cross-sectional studies allow only an examination of one-time point. Results may have varied if the patient had received the survey at a different time-point in their healthcare journey. Also, because we examined patients that were diagnosed within the past year, different patients were at varying stages in their healthcare journey. For example, some patients may have finished treatment, whereas others were still undergoing treatment. This is especially important regarding questions related to health-related quality of life. Patients who had finished treatment and were in remission may have felt better, thus, experiencing a higher health-related quality of life than patients who were still undergoing chemotherapy or some other treatment.

Additionally, cross-sectional design studies do not allow for making causal inferences so this study only examined relationships between variables. As such this study did not attempt to make any type of causal inference between any of the variables included in this study.

Survey instruments. This study utilized the SF-12v.1 to measure patients' health-related quality of life. At the time of study conception there were many versions of this instrument available online. Some contained different response options for the question about physical or emotional health interfering with social activities. This researcher obtained the survey initially through the organization that developed the measure for the Patient Reported Outcomes study. Subsequently, the rights to the SF12 were obtained by Optum and this researcher obtained approval from Optum to use the measure. Optum, does not recognize a previous response option "A good bit of the time" and no longer allows for scoring of this response option with their scoring software. Additionally, in order to ensure validity of the measure, Optum does not allow for any formatting changes of the measure. Because this measure was included in a survey booklet with other measures (following the Dillman Tailored Design Method), formatting of the SF-12 was changed to align with the formatting of the survey booklet to ensure patient ease in following instructions and completing questions. A brief description of the measure was added before the survey questions, questions were renumbered to align with sequencing of all questions in the survey booklet and minor changes were added to wording to allow for patients to follow instructions of the questionnaire easily. Optum made an exception to their formatting rules since the content remained the same and licensed it to Virginia Commonwealth University for this study. (License # QN035780).

Additionally, minor changes were made to the EORTC questionnaire. The EORTC instrument is a reliable and validated instrument that measures information received. In addition to measuring information received, questions about different types of information found in the EORTC information received measure were also reworded to measure information needs. This allowed us to 1.) measure information needs since there were no other measures that adequately measured various types of information needs that we desired to measure in this study, and 2.) allowed for a direct comparison of information needs and received information. Because the modified EORTC questions used to measure information needs in this study have not been validated to measure needs, a limitation exists in using this to measure information needs.

Mailed survey limitations. There are benefits and limitations to using written mailed surveys. First, mailed survey are only received by patients with a permanent mailing address, resulting in possible selection bias. Additionally, there is no way of ensuring that only the actual patient completes the survey. The participant’s spouse, parent, or adult child may have completed and returned the survey for the patient without our knowledge. Also, without assistance and guidance which is possible with in-person or telephone surveys, patients may skip questions, check multiple boxes, not follow the survey directions, or accidentally skip large sections of the survey. Missing data is a limitation of mailed surveys since participants may either accidentally skip sections or may decide not to answer certain questions.

Measurement error is another consideration in mailed surveys (Dillman, 1991). Participants may intentionally or unintentionally be unable to provide accurate information in response to the survey. Also, unlike having the questions read to them in a telephone or in-person survey, mailed surveys require a high level of literacy to read and complete. Low literacy may result in measurement error if the patient is unable to understand the questions.

Even though there were limitations of the mailed survey there are also many advantages to using a mailed survey method. The promised 80% response rate of the tailored design method of mailed surveys is a benefit to using mailed surveys. This study achieved only a 53.2% response rate; however, this population was comprised of patients who have been diagnosed with rare cancers that have high associated morbidity and mortality rates. Lack of response may be due the result of some participants being too ill to participate. Because there was a six-month delay receiving the patient list from the cancer registry, patients were six to twelve months' post-diagnosis and their health status could have been adversely compromised by the time the surveys were mailed. As noted, six (3.0%) patients' family members called to inform us that the patient had recently passed away.

Importantly, mailed surveys are also much faster for participants to complete and are less burdensome than other methods such as telephone surveys, an advantage when working with patients who may be ill. Participants can begin and complete the surveys on their own time and pace. Although a potential limitation for the study, an advantage of mailed surveys to participants is that those who do not wish to participate do not have the additional burden of declining participation directly and can simply not return the survey.

Receiving mail. Patients were asked to return the mailed surveys in a self-addressed stamped envelope containing the researcher's mailing address at the university. Because the mail schedule was unpredictable, receiving in batches every six to eight weeks following the tailored design method frequency of mailing letters was difficult. The mailing schedule for the fourth letter containing an identical survey, in case the original was lost, was supposed to be sent two weeks following the third letter. Participants who had previously completed and returned the survey were also sent a second survey due to the mail delay. A small number of participants (n=3) completed the second identical survey and returned it, thus resulting in multiple surveys received from the same participants. For duplicates, one survey from each participant was randomly selected for inclusion in the study. To correct for potential duplications and to lessen the burden on participants so they did not feel obligated to complete two identical surveys, the researcher delayed sending the fourth letter until after approximately 50% of that groups' surveys had been returned. The mail delivery issues noted above may have affected the overall response rate. Had mail delivery been timely, the study may have achieved a higher response rate as demonstrated by other studies using the Dillman method. In future studies, a dedicated postal box should be used to ensure mail is received in a timely manner.

Study Strengths

Overall, it is recognized the limitations outlined above exist, however, they are outweighed by the many strengths and benefits of this study.

Fills a needed gap in the literature. Most importantly, this is the first study to examine the information needs and sources of the rare cancer patient population. The results from this study contribute to the rare cancer information needs and sources literature, filling an important gap in the literature.

Ameliorates gap in knowledge. The information from this study about information needs and sources in this traditionally overlooked population is beneficial so that patients' information needs can be addressed and needs can be met. The results from this study provide healthcare providers with important information regarding needs which are most important to patients with rare cancers and identifies the areas where there are unmet needs. With these unmet needs identified, pertinent resources can be made available to patients.

Study location. This study was conducted at a large academic medical center, which is a safety net hospital serving a diverse population. This provided the opportunity for a diverse population of patients to participate in the study.

Validated measures. Another strength of the study is that validated measures were used to assess information needs/received, information sources, and health-related quality of life.

Rare cancer list. There are few lists available of cancers that are considered rare to use as a guide to identify which cancers are considered rare. This study uses cancers listed in the *Textbook of Uncommon Cancers* and selected cancers from the Office of Rare Disease Research (ORDR) rare cancer list. The researcher consulted with a medical oncologist and the editor of the Textbook *of Uncommon Cancers* to create the list. The resulting list is a very thorough delineation of cancers that are considered rare in the United States.

Future Studies

This study filled a much needed gap in the study of information needs and sources of patients with rare cancers, however, there is much more to learn regarding the informational needs and sources of this population. While the cancers used to identify eligible rare cancers in this study were all considered rare, a next step may be to examine the differences between the needs of those in this study who had the rarest cancers seen at the study site compared to those whose rare cancers are more commonly seen there. Based upon the results, it may also be beneficial to conduct a similar study with exclusively rarer cancers that are not commonly seen at the Massey

Cancer Center. Due to information satisfaction at the Massey Cancer Center, another next study may need to be conducted at other sites to assess information needs of this population and ascertain health information satisfaction in the rare cancer population.

Additionally, a comparison of information needs of patients with rare cancers with those diagnosed with general/common cancers would also be important to ascertain the differences in information needs, sources, and information satisfaction between those diagnosed with general cancers and those diagnosed with rare cancers.

This study also did not examine where patients were on the cancer care continuum since these patients were at different stages along the healthcare continuum. It would be beneficial to conduct a longitudinal study of the information needs and sources of patients with rare cancers throughout the cancer care continuum to ascertain the needs of patients throughout their cancer care journey.

A future project based upon the results of the information needs of patients would be the development of a government-sponsored rare cancer information portal. As discussed earlier, a rare cancer information portal aimed specifically at meeting the information needs of patients with rare cancers would be especially beneficial for providing reliable and accurate information to patients and fill a much needed information gap. This resource would benefit patients who want to learn more about their cancers. Additionally, it would contribute to the building of information services for the rare cancer community and would enable and empower patients by providing them information to become patient experts about their rare condition.

VII. Conclusion

This cross-sectional study examined the information needs and sources used by patients who have been diagnosed with rare cancers. As detailed above, patients with rare cancers experience many challenges during their health care journey, including difficulty finding authoritative information to satisfy their information needs.

As described above, the rare cancer population has traditionally been ignored by the research community and very little has been written describing the unique information needs and sources of patients with rare cancers. Most of the literature regarding information needs focuses on patients with general cancers (non-rare) or on the parents of children with rare genetic diseases. Additionally, most of the existing studies published on the information needs of rare diseases are small qualitative studies of patients in European countries.

Overall, this study fills a critical gap in the rare cancer literature by identifying patients' information needs and sources. This study demonstrates that there are many unmet needs, particularly in the provision of disease-specific information, treatment, and treatment options. The patients in this study reported they are not receiving information from their preferred source, healthcare providers, and are turning to the Internet for information even though they do not report high levels of trust for this source. Even though the majority of patients in the study were satisfied overall with the information they received, nearly 20% were not satisfied. Almost all patients reported desiring more information. Additionally, many reported that it took effort to find, were frustrated with their searches, found that the information was difficult to understand,

and were concerned about the quality. Even though there are government organizations created to address the informational needs of patients with rare cancers, these results show that much more needs to be for this traditionally underserved population.

Perhaps, with a better understanding of rare cancer patients’ information needs and sources of information, improvements can be made in the provision of authoritative information at the health care level and development of improved online information portals. Improved information provision may, in turn, improve rare cancer patients’ efforts to become empowered patients and enable them to become patient experts about their particular rare cancers. Effective information provision can lead to increased met needs and satisfaction which may also have a positive effect on patient outcomes such as improved health-related quality of life.

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Appendix A

Survey

Health Information Needs of Patients with Rare Cancers

A study of the information needs and information sources of patients diagnosed with rare cancers

Department of Social and Behavioral Health
Virginia Commonwealth University

Thank you for taking the time to complete this survey about your health information needs. We appreciate your help.

Your responses are voluntary and will be kept confidential. By completing and returning the survey you are consenting to participate in this research study.

If you have any questions about this study, please contact Dana Ladd by telephone at 804-628-2429 or by email at dlladd@vcu.edu.

Health Information Needs

These are some questions about the different types of information you may have ***needed*** about your cancer or treatment. For each type of information, check the box to show how much information you ***needed***.

Q1**. During your current disease or treatment, did you need information on…**

	Not at all	**A Little**	**Quite a Bit**	**Very Much**
1. The diagnosis of your cancer?	☐	☐	☐	☐
2. The extent (spread) of your cancer?	☐	☐	☐	☐
3. The possible causes of your cancer?	☐	☐	☐	☐
4. Whether the cancer is under control?	☐	☐	☐	☐
5.Whether family members are also at risk of developing cancer?	☐	☐	☐	☐
6. The purpose of any medical tests you have had or may undergo?	☐	☐	☐	☐
7. The procedures of the medical tests?	☐	☐	☐	☐
8. The results of the medical tests you have already received?	☐	☐	☐	☐
9. The medical treatment (chemotherapy, radiotherapy, surgery or other modality)?	☐	☐	☐	☐
10. The expected benefit of the treatment?	☐	☐	☐	☐
11. The possible side-effects of the treatment?	☐	☐	☐	☐
12. The expected effects of the treatment on cancer symptoms?	☐	☐	☐	☐
13. The effects of the treatment on social and family life?	☐	☐	☐	☐
14. The effects of the treatment on sexual activity?	☐	☐	☐	☐
15. The effects of treatment on fertility (the ability to have a baby)?	☐	☐	☐	☐
16. Other treatment options?	☐	☐	☐	☐
17. Financial, insurance, work-related or legal resources?	☐	☐	☐	☐
18. Additional help outside the hospital (e.g. help with daily activities, self-help groups)?	☐	☐	☐	☐
19. Rehabilitation services (e.g. physiotherapy, occupational therapy)?	☐	☐	☐	☐
20. Aspects of managing your illness at home?	☐	☐	☐	☐
21. Possible professional psychological support?	☐	☐	☐	☐
22. Different places of care (hospitals/outpatient services/home)?	☐	☐	☐	☐
23. Things that you can do to help yourself get well (rest, contact with others…)	☐	☐	☐	☐

Health Information Received

These are some questions about the different types of information you may have ***received*** about your cancer or treatment. For each type of information, check the box to show how much information you have ***received***.

Q2. During your current disease or treatment, how much information have you *received* on…

	Not at all	A Little	Quite a Bit	Very Much
1. The diagnosis of your cancer?	☐	☐	☐	☐
2. The extent (spread) of your cancer?	☐	☐	☐	☐
3. The possible causes of your cancer?	☐	☐	☐	☐
4. Whether the disease is under control?	☐	☐	☐	☐
5. Whether family members are also at risk of developing cancer?	☐	☐	☐	☐
6. The purpose of any medical tests you have had or may undergo?	☐	☐	☐	☐
7. The procedures of the medical tests?	☐	☐	☐	☐
8. The results of the medical tests you have already received?	☐	☐	☐	☐
9. The medical treatment (chemotherapy, radiotherapy, surgery or other modality)?	☐	☐	☐	☐
10. The expected benefit of the treatment?	☐	☐	☐	☐
11. The possible side-effects of the treatment?	☐	☐	☐	☐
12. The expected effects of the treatment on cancer symptoms?	☐	☐	☐	☐
13. The effects of the treatment on social and family life?	☐	☐	☐	☐
14. The effects of the treatment on sexual activity?	☐	☐	☐	☐
15. The effects of treatment on fertility (the ability to have a baby)?	☐	☐	☐	☐
16. Other treatment options?	☐	☐	☐	☐
17. Financial, insurance, work-related or legal resources?	☐	☐	☐	☐
18. Additional help outside the hospital (e.g. help with daily activities, self-help groups)?	☐	☐	☐	☐
19. Rehabilitation services (e.g. physiotherapy, occupational therapy)?	☐	☐	☐	☐

Q2. (continued) During your current disease or treatment, how much information have you received on…	**Not at all**	**A Little**	**Quite a Bit**	**Very Much**
20. Aspects of managing your illness at home?	☐	☐	☐	☐
21. Possible professional psychological support?	☐	☐	☐	☐
22. Different places of care (hospitals/outpatient services/home)?	☐	☐	☐	☐
23. Things that you can do to help yourself get well (rest, contact with others…)	☐	☐	☐	☐
24. Have you received written information?	☐ **Yes**	☐ **No**		
25. Have you received information on CD or tape/video?	☐ **Yes**	☐ **No**		
26. Overall, were you satisfied with the information you received?	☐ **Yes**	☐ **No**		
27. Do you wish to receive more information?	☐ **Yes**	☐ **No**	**If yes go to question 27a** **If no go to question 28**	
27a. If yes, please specify on which topics?				
28. Do you wish you had received less information?	☐ **Yes**	☐ **No**	**If yes go to question 28a** **If no go to question 29**	
28a. If yes, please specify on which topics?				
	Not at all	**A Little**	**Quite a Bit**	**Very Much**
29. Overall has the information you have received been helpful?	☐	☐	☐	☐

Where you Look for Health Information

These questions are about where you look for health information about cancer. For each, check the box for your answers about where you look for information.

Q3. Have you ever looked for information about health or medical topics from any source?

☐ Yes **[If yes, go to question 3A]**

☐ No **[If no, go to question, go to question 4]**

Q3A. The most recent time you looked for information about health or medical topics, where did you go *first*? <u>Choose one</u>

a. Books

b. Brochures, pamphlets, etc.

c. Cancer organization

d. Family

e. Friend/Co-Worker

f. Doctor or health care provider

g. Internet

h. Library

i. Magazines

j. Newspapers

k. Telephone information number

l. Complementary, alternative, or unconventional practitioner

m. Other-Specify- ☐

Q3B. When you look for information about health or medical topics, where did you go? **Choose all that apply**

a. Books

b. Brochures, pamphlets, etc.

c. Cancer organization

d. Family

e. Friend/Co-Worker

f. Doctor or health care provider

g. Internet

h. Library

i. Magazines

j. Newspapers

k. Telephone information number

l. Complementary, alternative, or unconventional practitioner

m. Other-Specify- ☐

Q4. From where do you prefer to receive information about your cancer? <u>Choose one</u>

a. Books

b. Brochures, pamphlets, etc.

c. Cancer organization

d. Family

e. Friend/Co-Worker

f. Doctor or health care provider

g. Internet

h. Library

i. Magazines

j. Newspapers

k. Telephone information number

l. Complementary, alternative, or unconventional practitioner

m. Other-Specify-

Q5. Based on the results of your most recent search for information about health or medical topics, how much do you agree or disagree with <u>each</u> of the following statements?

		Strongly Agree	Somewhat Agree	Somewhat Disagree	Strongly Disagree
a.	It took a lot of effort to get the information you needed.	☐	☐	☐	☐
b.	You felt frustrated during your search for the information	☐	☐	☐	☐
c.	You were concerned about the quality of the information	☐	☐	☐	☐
d.	The information found was hard to understand	☐	☐	☐	☐

Q6. Overall, how confident are you that you could get advice or information about health or medical topics if you needed it?

1. Completely confident
2. Very Confident
3. Somewhat confident
4. A little confident
5. Not confident at all

Q7. In general, how much would you trust information about health or medical topics from each of the following?

	Not at all	**A Little**	**Some**	**A Lot**
a. A doctor	☐	☐	☐	☐
b. Family or friends	☐	☐	☐	☐
c. Newspapers or magazines	☐	☐	☐	☐
d. Radio	☐	☐	☐	☐
e. Internet	☐	☐	☐	☐
f. Television	☐	☐	☐	☐
g. Government health agencies	☐	☐	☐	☐
h. Charitable organizations	☐	☐	☐	☐
i. Religious organizations and leaders	☐	☐	☐	☐

Your Health-Related Quality of Life

These questions ask for your views about your health. These questions are about how you feel and how well you are able to do your usual activities. Check the box for each question that describes how you feel.

Q8. In general, would you say your health is:

☐ Excellent
☐ Very Good
☐ Good
☐ Fair
☐ Poor

Q9. The following two questions are about activities you might do during a typical day.
Does your health limit you in moderate activities, such as moving a table, pushing a vacuum cleaner, bowling, or playing golf?

☐ Yes, Limited A Lot
☐ Yes, Limited A Little
☐ No, Not Limited At All

Does your health limit you in climbing several flights of stairs?

☐ Yes, Limited A Lot
☐ Yes, Limited A Little
☐ No, Not Limited At All

Q10. During the past 4 weeks have you had any of the following problems with your work or other regular activities as a result of your *physical health* and . . .

Accomplished less than you would like:

- ☐ Yes
- ☐ No

Were limited in the kind of work or other activities:

- ☐ Yes
- ☐ No

Q11. During the past 4 weeks, were you limited in the kind of work you do or other regular activities as a result of any *emotional problems* (such as feeling depressed or anxious) and…

Accomplished less than you would like:

- ☐ Yes
- ☐ No

Didn’t do work or other activities as carefully as usual:

- ☐ Yes
- ☐ No

Q12. During the past 4 weeks, how much did pain interfere with your normal work (including both work outside the home and housework)?

- ☐ Not At All
- ☐ A Little Bit
- ☐ Moderately
- ☐ Quite A Bit
- ☐ Extremely

Q13. The next three questions are about how you feel and how things have been during the past four weeks. For each question, please give the one answer that comes closest to the way you have been feeling. How much of the time during the past 4 weeks –

Have you felt calm and peaceful?

- ☐ All of the Time
- ☐ Most of the Time
- ☐ A Good Bit of the Time
- ☐ Some of the Time
- ☐ A Little of the Time
- ☐ None of the Time

Did you have a lot of energy?

- ☐ All of the Time
- ☐ Most of the Time
- ☐ A Good Bit of the Time
- ☐ Some of the Time
- ☐ A Little of the Time
- ☐ None of the Time

Have you felt downhearted and blue?

- ☐ All of the Time
- ☐ Most of the Time
- ☐ A Good Bit of the Time
- ☐ Some of the Time
- ☐ A Little of the Time
- ☐ None of the Time

Q14. During the past 4 weeks, how much of the time has your physical health or emotional problems interfered with your social activities (like visiting with friends, relatives, etc.)?

- ☐ All of the Time
- ☐ Most of the Time
- ☐ A Good Bit of the Time
- ☐ Some of the Time
- ☐ A Little of the Time
- ☐ None of the Time

Questions About You

Q15. What is your gender?

- ☐ Male
- ☐ Female

Q16. What is your age? ☐

Q17. Are you Hispanic or Latino?

- ☐ Hispanic
- ☐ Not Hispanic

Q18. What is your race?

- ☐ Black or African American
- ☐ White or Caucasian
- ☐ Asian
- ☐ Native American or Alaska native
- ☐ Native Hawaiian or Other Pacific Islander
- ☐ Multi-racial
- ☐ Other

Q19. What kind of medical insurance coverage do you have?

☐ None
☐ Private insurance
☐ Employer sponsored insurance
☐ Self-pay
☐ Medicare
☐ State-sponsored Indigent Care Program
☐ Virginia Coordinated Care Program
☐ Medicaid
☐ Military
☐ Not sure
☐ Other ___________________________

Q20. Which of the following best describes your employment status?

☐ Full-time paid
☐ Part-time paid
☐ Retired
☐ Unemployed
☐ Homemaker

Q21. Which category represents your combined total household income during the last 12 months? This includes money from jobs, net income from business, or farm or rent, pensions, dividends, interest, social security payments, disability and any other money income received by everyone in your household.

☐ Less than $5,000
☐ $5,001 - $10,000
☐ $10,001 – $15,000
☐ $15,001 - $20,000
☐ $20,001 - $25,000
☐ $25,001 - $30,000
☐ $30,001- $40,000
☐ $40,001 - $50,000
☐ $50,001 - $60,000
☐ $60,001 - $70,000
☐ $70,001 - $80,000
☐ $80,001 - $90,000
☐ $90,001 - $100,000
☐ Greater than $100,000

Thank You!

Comments:

Please return your survey in the enclosed envelope

or use the following address:

Dana Ladd
c/o Robin Matsuyama Ph.D
Box 980149
Department of Social and Behavioral Health
Virginia Commonwealth University
Richmond, VA 23298-0149

Appendix B

Letters

Research Participant Introductory Letter

<Date>

<Address Line 1>
<Address Line 2>
Address Line 3>

Dear <*patient name*>:

I am writing to ask for your help with an important research study I am conducting at the Virginia Commonwealth University Massey Cancer Center. The purpose of this research study is to find out more about the health information needs and health information sources of patients who have been diagnosed with cancers that are considered rare. In the next few days you will receive a request to participate in this project by answering a survey about your health information needs and the sources of information you use to find health information about cancer. I hope this study will contribute to improving patients' access to reliable information about rare cancers.

We would like to do everything we can to make it easy and enjoyable for you to participate in the study. I am writing in advance because many people like to know ahead of time that they will be asked to fill out a questionnaire. This research can only be successful with the generous help of people like you.

To say thanks, you will receive a small token of appreciation with the request to participate. I hope you will take 10-15 minutes of your time to help us.

Sincerely,

Dana L. Ladd, MS SLIS
Department of Social and Behavioral Health
Virginia Commonwealth University

Research Participant Initial Invitation Letter

<Date>

<Address Line 1>
<Address Line 2>
<Address Line 3>

Dear <*patient name*>:

I am inviting you to participate in a research study I am conducting at the Virginia Commonwealth University Massey Cancer Center. You are being contacted because you have been diagnosed with a cancer that is considered rare and your physician agreed this was a study for which you are potentially eligible. Your participation will not interfere with any care or treatment you may be receiving at this time. The purpose of this research study is to find out more about the health information needs and health information sources of patients who have been diagnosed with rare cancers.

You are being asked to complete a written survey about your health information needs. The questions take about 15 minutes to complete. Your responses are voluntary and will be kept confidential. By completing and returning the survey you are consenting to participate in this research study. If you have any questions about this study, please call Dana Ladd at 804-628-2429 or email at dlladd@vcu.edu. Data are being collected only for research purposes. All personal identifying information will be kept in password-protected files on a secure server. Your data will be identified by ID numbers, not names, and stored separately from research data in locked files in the researcher's office. Although the study team has put in safeguard to protect your information, there is always a potential risk of loss of confidentiality. We will not tell anyone the answers you give us and we will not use your name when we are reporting on the study. This study has been reviewed and approved by the Virginia Commonwealth University Institutional Review Board, and if you have any questions about your rights as a participant in this study, you may contact them by telephone at 804-827-2157.

You may not receive any direct benefit from this study, but by taking a few minutes to share your thoughts about your health information needs, you will help us in improving access to reliable health information about rare cancers. If you wish to participate in this study, please complete the enclosed survey and mail back to us in the enclosed addressed stamped envelope. A small token of appreciation is enclosed as a way of saying thank you.

Your alternative is to not participate in the study. Your care at Massey Cancer Center will not be affected by whether or not you participate in the study. If you do not wish to participate in the study, you may opt out of receiving any future letters by emailing dlladd@vcu.edu or calling Dana Ladd at 628-2429.

I truly appreciate your time and help with the study.

Sincerely,
Dana L. Ladd, MS SLIS
Department of Social and Behavioral Health
Virginia Commonwealth University

Research Participant Reminder Letter

<Date>

<Address Line 1>
<Address Line 2>
Address Line 3>

Dear *<patient name>*:

Last week a questionnaire was mailed to you asking you to participate in a study about the health information needs of patients diagnosed with cancers that are considered rare.

If you have already completed and returned the questionnaire, please accept our sincere thanks. If not, please consider completing and returning the survey as soon as possible. We are especially grateful for your help with this important study. If you did not receive the questionnaire, if you have any questions about this study, or if you do not want to receive any future letters about this study, please call Dana Ladd at 804-628-2429 or email at dlladd@vcu.edu.

Sincerely,

Dana L. Ladd, MS SLIS
Department of Social and Behavioral Health
Virginia Commonwealth University

Research Participant Information (Follow-up) Letter

<Date>

<Address Line 1>
<Address Line 2>
Address Line 3>

Dear *<patient name>*:

In <insert date> we sent you a letter requesting that you complete a questionnaire about your cancer health information needs. To the best of our knowledge, it has not yet been returned.

We are writing again because of the importance that your feedback can provide to our study about patients’ health information needs. We hope that you will fill out and return the enclosed questionnaire as soon as possible.

As mentioned before the questions take about 15 minutes to complete. Your responses are voluntary and will be kept confidential. If you have any questions about this study, please call Dana Ladd at 804-628-2429 or email at dlladd@vcu.edu. This study has been reviewed and approved by the Virginia Commonwealth University Institutional Review Board, and if you have any questions about your rights as a participant in this study, you may contact them by telephone at 804-827-2157. By completing and returning this survey, you are consenting to participate in this research study.

Thank you for your time and help with this research study.

Sincerely,

Dana L. Ladd, MS, SLIS
Department of Social and Behavioral Health
Virginia Commonwealth University

Research Participant Information Final Letter

<Date>

<Address Line 1>
<Address Line 2>
Address Line 3>

Dear <*patient name*>:

I am writing one last time to thank you for participating in the Health Information Needs study. The responses we have received have been invaluable. If you have not yet had a chance to send us your responses, you are welcome to do so and we would still appreciate your help completing the survey. We will continue to accept questionnaires until <date> at which point we will move to the next stage of the study.

If you have any remaining questions about this survey of cancer health information needs, I will be happy to help and can be reached by telephone at 804-628-2429 or by email at dlladd@vcu.edu.

I truly appreciate your time and help with the study.

Sincerely,

Dana L. Ladd, MS, SLIS
Department of Social and Behavioral Health
Virginia Commonwealth University

Appendix C

Cancer Types, Sites, and Stage by Demographics

Table 14: Cancer histology group and behavior descriptions of study participants by demographics

Histology Group Description Histology Behavior Description	Primary Cancer Site	Derived Stage (AJCC V7)	Derived Stage Grouped	Gender M=Male F=Female	Age	Race W=White NW=Non-White
Acinar Cell Carcinoma						
Acinar cell carcinoma	Parotid gland	III	III	F	63	NW
Adenocarcinoma with Metaplasia						
Metaplastic Carcinoma	Breast	IIIB	III	F	63	W
Metaplastic Carcinoma	Breast	IIIA	III	F	49	NW
Adenocarcinoma, NOS						
Adenocarcinoma, NOS	Endocervix	IIIB	III	F	33	W
Adenocarcinoma, NOS	Ileum	IV	IV	F	67	W
Adenocarcinoma, NOS	Duodenum	IV	IV	M	73	W
Adenoid Cystic & Cribriform CA.						
Adenoid Cystic Carcinoma	Cheek mucosa	I	I	F	65	W
Adenoid Cystic Carcinoma	Overlapping lesion breast	IIA	II	F	72	NW
Astrocytoma, NOS						
Astrocytoma, anaplastic	Frontal lobe	NA	NA	M	58	W
Astrocytoma, anaplastic	Parietal lobe	NA	NA	F	35	NW
Carcinoid Tumor, Malignant						
Carcinoid Tumor, Malignant	Rectum, NOS	I	I	M	76	NW
Carcinoid Tumor, Malignant	Rectum, NOS	UNK	UNK	F	62	NW
Carcinoid Tumor, Malignant	Ileum	IIIB	III	M	62	W
Carcinoid Tumor, Malignant	Ovary	IA	I	F	66	W
Carcinoid Tumor, Malignant	Ileum	IIIA	III	F	68	NW
Carcinoid Tumor, Malignant	Ileum	IIA	II	F	69	W
Carcinoid Tumor, Malignant	Small Intestine	IIIB	III	F	61	W
Neuroendocrine carcinoma	Gallbladder	NA	NA	F	55	NW
Neuroendocrine carcinoma	Ovary	IV	IV	F	26	W
Merkel cell carcinoma	Skin-upper limb	IIB	II	F	74	NW
Merkel cell carcinoma	Skin-scalp/neck	UNK	UNK	M	68	W
Goblet Cell Carcinoma	Appendix	IIA	II	F	59	W
Carcinoma, NOS						
Carcinoma, NOS	Parotid Gland	IVC	IV	M	60	W
Carcinoma, Undiff., NOS						
Pleomorphic carcinoma	Parotid Gland	IVC	IV	M	72	W
Carcinosarcoma, NOS						
Carcinosarcoma, NOS	Endometrium	IIIA	III	F	78	NW
Cholangiocarcinoma						
Cholangiocarcinoma	Biliary Tract, NOS	NA	NA	M	42	W
Cholangiocarcinoma	Intrahepatic bile duct	II	II	M	73	W
Chronic Myeloproflierative Dis.						
Myelosclerosis w/myeloid metaplasia	Bone Marrow	NA	NA	F	56	W
Essential thrombocythemia	Bone Marrow	NA	NA	F	41	W
Essential thrombocythemia	Bone Marrow	NA	NA	M	68	NW
Clear Cell Adenocrcinoma, NOS						
Clear Cell Adenocarcinoma, NOS	Cervix uteri	IB1	I	F	39	W
Clear cell adenocarcinoma, NOS	Upper lobe, lung	IB	I	M	71	W
Renal Cell Carcinoma, Chromophobe type	Kidney	I	I	F	68	W
Cystadenocarcinoma, NOS						
Serous cystadenocarcinoma,NOS	Fallopian Tube	IA	I	F	55	W
Endometrioid Adenocarcinoma						
Endometrioid carcinoma	Cervix Uteri	IVB	IV	F	67	NW
Endometrioid carcinoma	Ovary	IIIA	III	F	74	W
Erythroid Leukemia						
Acute myeloid leukemia, M6	Bone Marrow	NA	NA	F	53	W

Ewing Sarcoma						
Ewing Sarcoma	Long Bones/upper limb	IIA	II	M	31	NW
Ewing Sarcoma	Pelvic Bones	IVA	IV	M	20	W
Fibromatous Neoplasms						
Fibrosarcoma	Connective, subcutaneous	III	III	M	77	W
Solitary Fibrous tumor, malignant	Connective, subcutaneous	IIB	IIB	F	40	W
Fibromyxosarcoma	Connective, subcutaneous	III	III	M	77	W
Fibromyxosarcoma		IIA	II	F	69	W
Fibromyxosarcoma		IIB	II	M	79	W
Fibromyxosarcoma		III	III	M	53	NW
Fibrous Histiocytoma, Mal.						
Dermatofibrosarcoma, NOS	Skin of trunk	NA	NA	F	35	NW
Dermatofibrosarcoma, NOS	Vulva, NOS	NA	NA	F	63	NW
Follic. & Marginal Lymh, NOS						
Follicular lymphoma, NOS	Small intestine, NOS	IIEA	II	M	65	W
Marginal zone B-cell lymphoma	Ethmoid sinus	IIEA	II	M	73	W
Glioblastoma, NOS						
Gliosarcoma	Temporal Lobe	NA	NA	M	62	W
Glioblastoma, NOS	Frontal Lobe	NA	NA	M	83	W
Glioblastoma, NOS	Parietal Lobe	NA	NA	M	47	W
Glioblastoma, NOS	Parietal Lobe	NA	NA	M	78	NW
Glioblastoma, NOS	Temporal Lobe	NA	NA	M	53	W
Hemangioendothelioma						
Epitheloid Hemangioendothelioma	Liver	NA	NA	F	60	W
Immunoproliferative Diseases						
Waldenstrom macroglobulinemia	Blood	NA	NA	M	72	W
Kaposi Sarcoma						
Kaposi Sarcoma	Skin, NOS	NA	NA	M	41	NW
Liposarcoma Neoplasms						
Myxoid Liposarcoma	Connective, subcutaneous	IB	I	F	67	W
Lymphoepithelial Carcinoma						
Basaloid squamous cell carcinoma	Base of tongue, NOS	IVA	IV	M	50	W
Basaloid squamous cell carcinoma	Larynx, NOS	IVA	IV	M	58	W
Malignant Lymphoma, NOS						
Primary Cutaneous Follicle Centre Lymphoma	Skin of scalp and neck	IE	I	F	74	NW
ML, Large B-Cell, Diffuse						
Burkitt Lymphoma, NOS	Breast NOS	IEA	I	F	36	NW
ML, Large B-cell, diffuse	Connective, subcutaneous	IEA	I	F	35	W
ML, Large B-cell, diffuse	Connective	IIEA	II	M	93	NW
ML, Large B-cell diffuse	Short bones/lower limb	IEA	I	M	60	W
ML, Small B-Cell Lymphocytic						
Mantle cell lymphoma	Lymph nodes/multiple regions	IVB	IV	M	70	W
Mantle cell lymphoma	Palate, NOS	IEA	I	F	46	W
Mucinous Adenocarcinoma						
Mucinous Adenocarcinoma	Lower lobe, lung	IB	I	M	63	W
Mucoepidermoid Carcinoma						
Mucoepidermoid Carcinoma	Parotid Gland	I	I	F	50	W
Myeloid Leukemia, NOS						
Chronic Myeloid Leukemia	Bone Marrow	NA	NA	M	71	W

Chronic Myeloid Leukemia	Bone Marrow	NA	NA	M	44	W
Myomatous Neoplasms						
Leiomyosarcoma, NOS	Connective, subcutaneous	IIB	II	F	42	NW
Leiomyosarcoma, NOS	Connective, subcutaneous Mandible	IIA	II	F	65	W
Leiomyosarcoma, NOS	Connective,	IIA	II	F	36	W
Leiomyosarcoma, NOS	subcutaneous	III	III	F	59	W
Neurofibrosarcoma						
Malignant peripheral nerve sheath tumor	Peripheral nerves and autonomic	IA	I	M	44	W
Nevi & Melanomas						
Malignant Melanoma	Partotid gland	NA	NA	M	61	W
Nonencapsul. Sclerosing Ca.						
Nonencapsulating sclerosing carcinoma	Thyroid gland	I	I	F	19	W
Oligodendroglioma, NOS						
Oligodendroglioma, NOS	Frontal Lobe	NA	NA	F	46	W
Oligodendroglioma, anaplastic	Frontal Lobe	NA	NA	M	33	W
Osteosarcoma, NOS						
Osteosarcoma, NOS	Bones of skull and face	IA	I	M	84	NW
Chondroblastic osteosarcoma	Vertebral column	IVB	IV	M	19	W
Other Leukemias						
Hairy cell leukemia	Bone marrow	NA	NA	M	77	W
Hairy cell leukemia	Bone marrow	NA	NA	M	41	W
Other Myeloid Leukemias						
Chronic myelogenous leukemia, BCR/ABL pos	Bone marrow	NA	NA	F	77	NW
Chronic myelogenous leukemia, BCR/ABL, pos	Bone marrow	NA	NA	F	56	W
Other Spec. Non-Hodgkin Lymphoma						
NK/T-cell lymphoma, nasal and nasal type	Nasal cavity	IEA	I	F	64	W
Anaplastic large cell lymphoma, T-cell and	Ethmoid sinus	IVA	IV	F	86	W
Oxyphilic Adenocarcinoma						
Oxyphilic adenocarcinoma	Thyroid gland	II	II	F	56	NW
Paget Disease, Extramammary						
Paget disease, extramammary	Skin of trunk	Unk	Unk	M	59	W
Paget disease, extramammary	Skin of trunk	II	II	M	65	W
Papillary & Follicular Adenoca.						
Mixed medullary-papillary carcinoma	Thyroid Gland	IVA	IV	M	79	W
Papillary Carcinoma, NOS						
Verrucous carcinoma, NOS	Ventral surface of tongue	I	I	F	69	W
Papillary squamous cell carcinoma	Pharynx, NOS	NA	NA	M	36	W
Papillary Cystadenoca., NOS						
Intraductal papillary-mucinous carcinoma	Head of pancreas	IA	I	F	64	NW
Phylllodes Tumor, Mal.						
Phyllodes tumor, malignant	Breast, NOS	IIA	II	F	91	NW
Polycythemia Vera						
Polycythemia vera	Bone marrow	NA	NA	M	69	W
Prolymph/Precurs Leukemia						
Prolymphocytic leukemia, T-cell type	Bone marrow	NA	NA	M	68	W
	Bone marrow	NA	NA	M	59	NW

Prolymphocytic leukemia, T-cell type						
Refractory Anemia						
Refractory anemia with excess blasts	Bone marrow	NA	NA	F	58	W
Refractory anemia with excess blasts	Bone marrow	NA	NA	F	59	NW
Refractory anemia with excess blasts	Bone marrow	NA	NA	M	78	NW
Sarcoma, NOS						
Giant cell sarcoma	Connective, subcutaneous	III	III	F	68	NW
Squamous Cell Carcinoma						
Sq. cell carcinoma, keratinizing, NOS	Vulva	IVA	IV	F	53	W
Sq. cell carcinoma, keratinizing, NOS	Overlapping lesion nasopl	III	III	F	41	W
Sq. cell carcinoma, keratinizing, NOS	Ovary	IIB	II	F	55	NW
Sq. cell carcinoma, keratinizing NOS	Vulva, NOS	IB	I	F	49	W
Sq. cell carcinoma, spindle cell	Vagina	III	III	F	63	NW
Sq. cell carcinoma spindle cell	Glottis	I	I	M	74	W
Sq. cell carcinoma, lg. cell, non-ker	Nasopharynx, NOS	III	III	M	23	W
Stromal Sarcoma						
Adenosarcoma	Endometrium	IA	I	F	58	W
Sweat Gland Adenocarcinoma						
Apocrine adenocarcinoma	Upper outer quadrant of bre	IA	I	F	58	NW
Thymoma, Malignant						
Thymic carcinoma, NOS	Thymus	NA	NA	F	73	W

Table 15: Histology group descriptions by gender and race

Histology Group Description 54 Types of Rare Cancers	Gender		Race	
	Male	Female	White	Non-White
Acinar Cell Carcinoma	0	1	0	1
Adenocarcinoma with Metaplasia	0	2	1	1
Adenocarcinoma, NOS	1	2	3	0
Adenoid Cystic & Cribriform CA.	0	2	1	1
Astrocytoma, NOS	1	1	1	1
Carcinoid Tumor, Malignant	3	9	7	5
Carcinoma, NOS	1	0	1	0
Carcinoma, Undiff., NOS	1	0	1	0
Carcinosarcoma, NOS	0	1	0	1
Cholangiocarcinoma	2	0	2	0
Chronic Myeloproflierative Dis.	1	2	2	1
Clear Cell Adenocrcinoma, NOS	1	2	3	0
Cystadenocarcinoma, NOS	0	1	1	0
Endometrioid Adenocarcinoma	0	2	1	1
Erythroid Leukemia	0	1	1	0
Ewing Sarcoma	2	0	1	1
Fibromatous Neoplasms	4	2	5	1
Fibrous Histiocytoma, Mal.	0	2	0	2
Follic. & Marginal Lymh, NOS	2	0	2	0
Glioblastoma, NOS	5	0	4	1
Hemangioendothelioma	0	1	1	0
Immunoproliferative Diseases	1	0	1	0
Kaposi Sarcoma	1	0	0	1

Liposarcoma Neoplasms	0	1	1	0
Lymphoepithelial Carcinoma	2	0	2	0
Malignant Lymphoma, NOS	0	1	0	1
ML, Large B-Cell, Diffuse	2	2	2	2
ML, Small B-Cell Lymphocytic	1	1	2	0
Mucinous Adenocarcinoma	1	0	1	0
Mucoepidermoid Carcinoma	0	1	1	0
Myeloid Leukemia, NOS	2	0	2	0
Myomatous Neoplasms	0	4	3	1
Neurofibrosarcoma	1	0	1	0
Nevi & Melanomas	1	0	1	0
Nonencapsul. Sclerosing Ca.	0	1	1	0
Oligodendroglioma, NOS	1	1	2	0
Osteosarcoma, NOS	2	0	1	1
Other Leukemias	2	0	2	0
Other Myeloid Leukemias	0	2	1	1
Other Spec. Non-Hodgkin Lymphoma	0	2	2	0
Oxyphilic Adenocarcinoma	0	1	0	1
Paget Disease, Extramammary	2	0	2	0
Papillary & Follicular Adenoca.	1	0	1	0
Papillary Carcinoma, NOS	1	1	2	0
Papillary Cystadenoca., NOS	0	1	0	1
Phylllodes Tumor, Mal.	0	1	0	1
Polycythemia Vera	1	0	1	0
Prolymph/Precurs Leukemia	2	0	1	1
Refractory Anemia	1	2	1	2
Sarcoma, NOS	0	1	0	1
Squamous Cell Carcinoma	2	5	5	2
Stromal Sarcoma	0	1	1	0
Sweat Gland Adenocarcinoma	0	1	0	1
Thymoma, Malignant	0	1	1	0
TOTAL:	**51**	**62**	**79**	**34**