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SOCIAL SUPPORT AND RARE DISEASE: INNOVATIONS IN HEALTH PROMOTION AND EDUCATION TECHNOLOGY

by

Bradden Joseph Blair

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STATEMENT OF THESIS APPROVAL

The thesis of Bradden Joseph Blair

has been approved by the following supervisory committee members:

Glenn E. Richardson , Chair **May 6, 2015**
Date Approved

Justine J. Reel , Member **May 6, 2015**
Date Approved

Debra L. Scammon , Member **May 6, 2015**
Date Approved

Quynh C. Nguyen , Member **May 6, 2015**

Nick A. Galli , Member **May 6, 2015**

and by **Les K. Chatelain** , Chair/Dean of

the Department/College/School of **Health Promotion and Education**

and by David B. Kieda, Dean of The Graduate School.

ABSTRACT

According to the Office of Rare Diseases Research (ORDR) at the National Institutes of Health (NIH), rare diseases affect more than 25 million Americans. The scarcity of information, poor prognosis, and lack of viable treatment options for many conditions causes significant anxiety for rare disease patients and their families. Increasingly, rare disease populations are going online to acquire the support necessary to cope with their health challenges. This dissertation builds upon earlier work by answering a question left largely unaddressed to date: what roles do social support and online support environments play for patients affected by rare disease?

This dissertation follows the three article format. In the first article, the author provides a review of important literature from three main areas of research; social support, online support groups/social media, and rare disease. The author also discusses implications of computerized health care services for the field of health promotion and education.

In the second article, the author reports the results of a recent study in which a conventional approach to qualitative content analysis was utilized to characterize the followers, focus, founders and formation of sarcoma related Facebook groups. Three different coding schemes for classifying online support groups were identified: group focus or orientation (person vs. population), founder treatment status (patient or nonpatient) and founder disease affiliation status (active treatment, survivor, in memoriam, or external organization). This study suggests that Facebook groups provide a mechanism not only for identifying disease specific groups, but also for facilitating connections between individuals with similar backgrounds or states of disease progression.

The third article reports the results of an additional qualitative study examining the online social support experiences of patients in active treatment for Osteosarcoma, a rare and aggressive form of cancer. Evidence of seven distinct types of social support were observed: appraisal, emotional, informational, spiritual, esteem, network and tangible. This study suggests that appraisal and spiritual support may play a bigger role in online support communities than has been previously suggested.

It is hoped that this dissertation will serve as a call to action for other researchers. Additional research is needed to adequately address and understand the needs of those affected by rare disease.

To my wife Jannette and my children, Paige, Evan, and Christian, for all of their love and support. I could not have done this without them. They are most important to me; much more important than any other earthly honor than can be bestowed.

I love you, team.

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this marathon journey without her support and am fortunate to be married to her. After reading and editing all of my papers, I am confident she knows as much about rare disease and social support as I do. Our own experience of receiving and coping with a rare disease diagnosis, also led to my focus on rare disease and social support for this dissertation. I hope this work will encourage additional research in this important area.

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CHAPTER 1

INTRODUCTION

This paper offers a fresh perspective on earlier work by answering a question left largely unaddressed to date: what roles do social support and online support environments play for patients affected by rare disease? Unlike members of other online communities who have the option of going elsewhere if they do not feel adequately supported online, rare disease patients often do not have that luxury. Given the rarity of some conditions and the unfamiliarity of providers and other healthcare workers with some conditions, an online support group may be the only source of information or support for patients with rare conditions. It is hoped that this dissertation will serve as a call to action for other researchers in the field of health promotion and education. Additional voices will be needed in order to adequately address the needs of an important and grossly underserved population; those affected by rare disease.

Article 1: Social Networking, Social Support, and Rare Disease: Implications for Health Promotion and Education

The purpose of the first article in this dissertation is 1) to review and expand upon prior work that has explored the relationship between social networking technology and social support, and 2) to discuss the implications of social networking technology and social support for rare disease patients and their families. The initial article also lays the groundwork for the subsequent articles in this study.

The first article begins with a brief overview of important literature from three main areas of research; social support, online support groups/social media, and rare disease. The articles that were selected for review have proven to be extremely valuable and have served to inform and guide the entire research process. The review of prior work helped the researcher not only to master important theoretical concepts, but also to identify important gaps within the existing literature. Indeed, many of the research questions that have been explored in this dissertation were developed during the formative stages of the initial article.

At the outset of this article, the researcher makes a case that rare disease is an important and necessary research area. Rare disease is not an insignificant health problem in the United States; it is a serious health concern. According to the Office of Rare Diseases Research at the National Institutes of Health, a rare disease is a condition that affects a small patient population; fewer than 200,000 people in the United States at any given time (see Rare Diseases Act of 2002). Although specific rare diseases affect a relatively small number of people, there are more than 6,800 rare diseases that collectively affect more than 25 million Americans.

The prevalence of rare disease conditions should make rare disease research a top priority for federal agencies, pharmaceutical companies, health researchers, and public health professionals. Unfortunately, such has not always been the case. Although the Rare Disease Act of 2002 together with the National Organization for Rare Disorders (NORD) and the Orphan Drug Act of 1983 helped to significantly increase legislative and financial support for rare disease research, education, and treatments over time, the research agenda that has emerged is largely unbalanced. The majority of research has centered on biomedical research. The author of this dissertation argues that the research agendas of the NIH, FDA, and other organizations should be expanded to also incorporate the behavioral and psychosocial concerns of rare disease patients and their families.

The dissertation author has also identified a related area that has been left largely untouched in the scientific literature: the intersection between social support and rare disease. Given the maturity of research in the area of social support, it was surprising to discover that this area has been ignored for so many years, especially by those in the field of health promotion and education. One of the primary challenges in the area of social support research, has been how best to define social support. As a multidisciplinary field, there has always been a great deal of controversy regarding how to operationalize the concept. The phrase social support has been used to describe many different aspects of social relationships, often without a clear "conceptual definition…or valid or reliable indicators of the concept" (Thoits, 1982, p. 146). Not surprisingly, this ambiguity has made it difficult to compare studies or generalize results.

Thoits (1982) proposed a solution to the definitional conundrum faced by many in the area of social support research: to only accept definitions of social support that can be operationalized. The impact of this recommendation on the current dissertation study cannot be overstated. This guideline was consistently followed while the coding frames for each of the subsequent articles were developed. This is one of the reasons why operational definitions for key concepts (e.g., patient status, disease affiliation status, functional components of social support, etc.) have been developed or included in every chapter of this dissertation. This recommendation also drove the researcher to utilize the four component model of social support developed by House (1981), to respond to some of the initial questions posed in this article. House (1981) defined social support as "an interpersonal transaction involving one or more of the following: (1) emotional concern (liking, love, empathy), (2) instrumental aid (goods and services), (3) information (about the environment), or (4) appraisal (information relevant to self-evaluation)" (p. 39). This is perhaps one of the best functional definitions of social support to date. It contains clearly defined constructs that are highly operational in nature.

In the first article, an important question is raised. What is the difference between a social network and a social support system? The focus of this dissertation is understanding the impact of social networking technologies on social support among rare disease patients. Research examining this question must go beyond merely evaluating whether or not rare disease patients are members of online social networks. Having access to a large number of connections does not necessarily mean that someone is socially engaged or supported. For example, if a rare disease patient joins an online support group, but never actually logs in to the group, it will be impossible for them to feel supported by the group. "The presence of actual social contact is required to provide any sense of support or lack of it" (Stephens, Alpass, Towers, & Stevenson, 2011). Therefore, research must also examine how groups differ, the types of support available via online support groups, and whether or not patients are taking advantage of such groups. These are just a handful of some of the many questions that are raised and addressed in the current dissertation study.

In addition to reviewing definitions of rare disease, social support, and social networks, some of the benefits and challenges of online support groups are also discussed. While not specific to rare disease, a number of studies have shown that participants in online support communities benefit from increased access to health information, feeling more confident in communicating with health care providers, feeling empowered and more engaged in medical decision making, experiencing greater levels of social support, and experiencing enhanced physical and mental health. There is also evidence to suggest that online support groups provide greater access to a diversity of perspectives and information on a health topic than can be found in face-to-face support groups. (Walther & Boyd, 2002; Wright, 2002). While there are many benefits of online support groups, such groups are also not without their challenges. Kevin Wright (2000) found that the single most frequently cited disadvantage of computer-mediated support groups was the absence of haptic communication, or communication by touch. This is not insignificant given the many studies that promote healing touch as a complementary therapy for stress and anxiety, pain, high blood pressure, depression, and other adverse health conditions (Anderson & Taylor, 2011; Wardell & Weymouth, 2004).

Finally, a detailed analysis of the implications of computerized health care services for the field of health promotion and education is presented. This analysis is positioned within the context of a three component framework developed by Patterson and colleagues. Patterson et al. (1997, p. 225) have suggested that computerized health care services are beneficial in three ways: (1) educating patients about health-related subjects, (2) bringing about changes in health behavior, and (3) providing social support, or assisting in obtaining social support. Although published over a decade ago, this framework also remains relevant for discussing some of the major implications of social networking technology, and social support for health promotion and education practice. The author of this dissertation makes one notable update to the framework: to include caregivers and clinicians alongside of patients in the discussion of the educational benefits of computerized health care services.

The first article provides strong support for the notion that online communities likely represent an important source of social support for rare disease patients and their families. It also suggests that different types of social support (informational, emotional, tangible, appraisal, and companionship) likely serve distinct, yet important, health promoting functions within these communities. Additional research is recommended to expand the current level of understanding regarding how online support networks function to enable socially supportive behaviors among rare disease patients.

Article 2: Social Media and Sarcoma: A Qualitative Content Analysis of Facebook Support Groups

In the second article, the focus is narrowed to one area of rare disease: sarcoma cancer. Sarcoma is a grouping of extremely rare forms of cancer that develop from tissues like bone or muscle (American Cancer Society, 2014). Of the approximately 1.6 million cases of cancer in the United States each year, fewer than 15,000 cases are sarcoma (American Cancer Society, 2013; Darling, 2007). The second article also concentrates on one type of online support community: Facebook groups. Although no specific estimate exists for the total number of rare disease groups on Facebook, many conditions have multiple groups, and a growing number of patients have multiple groups to choose from. The large number of registered users on Facebook has solved a challenge that was previously unsolvable by rare disease patients: how to locate others in a similarly rare situation. As of January 2015, there were over 1.39 billion registered Facebook users worldwide (Facebook Inc., 2015).

The aim of the second article is two-fold: 1) to increase scientific understanding of the role and influence of social media on the experience of support for sarcoma patients and their families, and 2) to characterize via content analysis the followers, focus, founders and formation of sarcoma related Facebook groups.

The research process begins by utilizing Facebook's built-in search engine and a predetermined list of sarcoma diagnoses to locate groups for study. In order to be included in the study sample, groups are required to contain a sarcoma diagnosis in the title, to be conducted primarily in the English language, and to be related only to human forms of sarcoma. These inclusion criteria result in the successful identification of 82 sarcoma support groups on Facebook. Total cumulative membership for the selected groups is calculated at nearly 26,000 members. Membership size for the selected groups is found to range from 2 to 2,715 members, and the average membership per group is 316 members. The top five disease classifications for the target groups are as follows: osteosarcoma (39%), leiomyosarcoma (18%), undifferentiated sarcoma (13%), rhabdomyosarcoma (10%) and synovial sarcoma (7%).

Once all of the potential study groups have been successfully identified, information extracted from group titles and descriptions is utilized to develop three coding schemes that guide content analysis: group focus (person vs. population), founder treatment status (patient or nonpatient) and founder disease affiliation status (active treatment, survivor, in memoriam, or external organization).

Content analysis of group titles, group descriptions and, for open groups, the initial posts of group founders, is used to classify all of the sampled groups and their founders. In cases where classification cannot be successfully completed using Facebook data, publicly available secondary data sources are used to confirm classification. Group classification reveals 40.2% of groups to be person-focused and 59.8% of groups to be population-focused. Nonpatient founders account for the greatest number of groups; 81.8% of person-focused groups and 75.5% of population-focused groups. The vast majority of person-focused groups (81.8%) have a founder associated with a patient in active treatment, while only 12.2% of population-focused groups have that affiliation. Population-focused group founders are found to be most likely to be affiliated with a cancer survivor (30.6%), a deceased patient (30.6%), or an external organization (26.5%). According to group founders, both intrinsic and extrinsic motivations contribute to the formation of online support groups. For patient founders in the study sample, being a survivor is the biggest motivation for group formation (56% of cases), but being in active treatment also accounts for a significant portion of group formations (>40%). Nonpatient founders are motivated by a variety of factors to form online sarcoma support groups. The primary motivations are: being connected to someone in active treatment (39%), keeping the memory of a loved one alive (30%), and promoting an external organization (20%).

As mentioned earlier, one of the objectives of the dissertation study is to increase scientific understanding of the role and influence of social media on the experience of social support for sarcoma patients and their families. The researcher seeks to describe and differentiate between groups in the study sample based on characteristics of group followers and founders, and on a group's focus and formation. This is an important first step in laying the groundwork for additional research regarding potential differences in socially supportive content between groups. If future studies demonstrate that different types of groups meet different support needs, such findings will reinforce the importance of properly matching individuals to groups. Joining a group that does not offer the type of support required by an individual will be counterproductive. The significance of any finding that differences exist in social support content between groups will also hinge on whether or not individuals participate in multiple groups. Although an individual group may not fully meet a member's support needs, those needs may be addressed in totality between all of the groups in which an individual participates. It is with this question in mind that the researcher sought (in article 2) to determine the amount of overlap in membership between groups. Analysis of group membership revealed that 87.8% of individuals accounted for in the study participated in only one of the groups in the sample. It is not yet known what this means, but this finding could prove important if future studies demonstrate clear separation in social support content between different types of groups.

The finding that groups can be classified as either patient-focused or population-focused has led the researcher to wonder whether there is a connection between tie strength and social support. Granovetter (1973), suggested that weak interpersonal ties play an important role in social circles, especially with respect to knowledge transfer and information dissemination. He saw weak ties, especially those that bridge disparate social structures, as helpful for enabling individuals to have access to ideas, influences or information that would otherwise be inaccessible.

In the second article, the researcher explores the impact of founders on group formation and function. Prior studies of organizational culture have shown that founders often have a lasting impact on the culture and behavior of their firms (Barringer, Jones, & Neubaum, 2005). Indeed, some of the world's most powerful social movements, and many industry leading brands have been built around an affection for charismatic leaders. Surprisingly, researchers have not yet examined the impact of group founders on online support groups. What impact do founders of online support groups have on group formation and processes? Recent studies suggest that this influence is significant. In 2014, Kraut and Fiore conducted a study in which they examined the role of founders in determining the fate of 472,231 Facebook groups. According the study, of the 100,000 new groups that are created on Facebook each day, "13% produce no content after the first day…and 57% have stopped all activity within three months of creation" (Kraut & Fiore, 2014).

In the present study, founders were observed playing an important role. Content analyses of publicly available information showed that group founders not only initiated group formation, but they also defined a group's focus and followers via carefully constructed group titles and descriptions. Founders also used introductory posts to establish group norms and expound the group's focus. Once established, group founders and administrators controlled group dynamics by managing group membership and privacy practices. Membership and privacy policies determined who could view, read, or post content within a group.

Although the second article has identified some important defining features of sarcoma groups, it does not address how these features relate to the different types of social support that are possible within such groups. Additional research is recommended to more fully investigate the relationship between group followers, group focus, group founders, group formation and social support. In the third article, the author begins to make these connections.

Article 3: A Qualitative Content Analysis of Social Support Messages Exchanged by Osteosarcoma Patients in Active Treatment on Facebook

The third article in the dissertation employs a directed approach to qualitative content analysis. According to Hsieh and Shannon (2005), directed content analysis is recommended when: a) existing theory or prior research exists about a phenomenon that is incomplete or would benefit from further description, and b) the researcher seeks to validate or extend conceptually a theoretical framework or theory (p. 1281). In the current study, the technique is utilized to describe the social support activities of patients currently affected by osteosarcoma, a rare and aggressive form of cancer, and the role that Facebook communities play in facilitating supportive interactions. The researcher also extends prior social support theory to the study of social support within an online support group for osteosarcoma patients. The study responds to the following important research questions: RQ1: What does the exchange of social support look like in an online support group dedicated to osteosarcoma? RQ2: How do patients in active treatment leverage online support groups to meet their needs for social support? These are important questions since “studies that examine the subjective experience of persons receiving and giving support are more likely to yield relevant information for understanding person-environment fit and for suggesting interventions" (Ell, 1984).

In article two, an investigation is conducted of the followers, focus, founders and formation of 82 sarcoma related Facebook groups. The most frequently encountered form of sarcoma represented in the sample groups is osteosarcoma (39% of groups). This fact strongly influenced the researcher's decision to select an osteosarcoma group for further examination in article three. Osteosarcoma is an extremely rare form of cancer that affects fewer than 800 new patients each year (American Cancer Society, 2015). Of the approximately 1.6 million cases of cancer in the United States each year, fewer than .0005% of cancer cases are diagnosed as osteosarcoma (American Cancer Society, 2013; Darling, 2007). The prognosis for osteosarcoma patients is affected by a variety of factors including: "primary tumor site, tumor size, presence of clinically detectable metastatic disease, surgical resectability and necrosis following induction or neoadjuvant chemotherapy" (National Cancer Institute, 2015). With effective diagnosis and treatment, the overall relative 5-year survival rate for osteosarcoma is estimated at between 41-55% (Damron, Ward, & Stewart, 2007; Dorfman & Czerniak, 1995). Given the complexities of osteosarcoma treatment and recovery, it is natural for patients to want to connect with as many supportive resources as possible. The rarity of osteosarcoma cases means that without organized support, it can be difficult for individuals to locate others affected by the disease. Faced with an uncertain diagnosis and future, many patients are turning to Facebook as their medium of choice for supportive exchange.

The first step in the research processes was to identify a study group and download a copy of all messages for the sample group. The selected group was founded in 2008 and has 627 members. Between September of 2008 and March of 2015, the group had nearly 4,500 messages exchanged between group members, 935 discussion posts and 3,521 replies. These numbers do not include nonnarrative responses, such as Facebook "likes." Since the researcher was interested in focusing only on patients in active treatment, the next step was to attempt to classify all messages based on the treatment status and disease affiliation status of the messages author at the time of posting. Independently classifying all 4,451 messages was important since the disease affiliation status of entry authors changes over time. For example, an osteosarcoma patient may move from active treatment to survivor status to relapse to death all within a very short period of time. If an individual had ever been an osteosarcoma patient, they were classified as a patient, regardless of their current disease affiliation status. The classification of message authors was accomplished using two of the categories that had been previously developed in article two for classifying group founders: treatment status (patient vs. nonpatient) and disease affiliation status (active treatment, survivor, in memoriam, and external organization).

Classification began by sorting messages in sequential order (newest to oldest) based on author name. After sorting, each message was independently coded by the researcher and a collaborator based on the treatment status and disease affiliation status of the author. Once classification was completed, interrater reliability was calculated based on each category. Cohen's Kappa ranged from .81 for disease affiliation status to .95 for treatment status. According to Jacob Cohen, this reveals almost perfect reliability between coders (see Cohen, 1960). Following the initial classification exercise, the researcher and collaborator met again to review all messages where differences in classification existed and come to perfect agreement on all entries where coding differed. The result was a highly differentiated dataset based on poster classification with which to begin analyzing social support content.

The next step in the research process was to separate out all messages by patients in active treatment. There were a total of 644 messages that met this criteria. This became the study sample. These messages were initiated by 32 unique individuals, and each of these users contributed an average of 20 messages while in active treatment status over the life of the group. The researcher then conducted a literature review to generate a list of the most common functional components of social support. There were six common components of social support identified from the literature search: informational, emotional, esteem, network, tangible and appraisal support.

Once the initial social support categories had been identified, the researcher next began to read through some of the group messages. It was observed that all of the initial messages were of one of two types: either support seeking or support giving. This was an important observation for two reasons: First, House (1981) noted that one of the key considerations in defining social support is understanding "who gives what to whom regarding which problems" (p. 22). He also argued that social support components are best understood as a matrix that incorporates both the functional definitions of support and the directionality of support between individuals. Second, work by Weiss (1974) and Cobb (1976) both suggest that social networks not only provide individuals with an opportunity to seek support, but that they also provide opportunities for individuals to provide nurturance or mothering (support) to others. These studies suggests that individuals benefit themselves by also being of help to others. Based on this information, a decision was made to modify the initial coding frame to encompass both giving and seeking behaviors for all six of the top social support dimensions identified in the scientific literature. This modification resulted in 12 social support codes.

Next, the researcher read all 644 messages and classified the content based on: 1) if the message contained evidence of socially supportive exchange, and 2) the appropriate coding classification for content determined to be supportive. Once all of the content had been coded into categories, steps were undertaken to confirm the accuracy of the coding process. Once the initial coding process had been completed, the research took steps to identify subcategories within each main category of social support. Operational definitions were created for each sub-category and content was coded accordingly. Once again, steps were taken to confirm the accuracy of the coding process. Finally, any content that could not be coded into existing categories were coded inductively into new sub-categories. The result of this effort was the creation of a new category of support (spiritual support) and three subcategories (prayer, helping thoughts, and well-wishing). In the end, the total number of codes created for the study was 56, 14 categories and 42 subcategories.

The results of the study were very interesting. For osteosarcoma patients in active treatment, nearly 80% of socially supportive messages involved giving support to others rather than seeking support for self. Approximately 85% of support giving messages involved only four types of social support: emotional (28%), appraisal (23%), informational (17%) and spiritual (17%). Posts having to do with seeking social support were predominantly focused on appraisal support (33%), informational support (30%) and tangible support (10%). Spiritual support also accounted for nearly 9% of messages where the poster was seeking support.

The results of this study clearly showed that a Facebook group can provide a rich environment where diverse types of social support are exchanged. In the study group, patients in active treatment not only utilized the group to seek or receive social support from others, but they also used the platform to provide support to others facing similar circumstances.

One of the most significant findings of the study concerned the importance and prevalence of spiritual support in an online environment. Although the need for a spiritual support category was not evident in the initial review of social support literature, the data strongly supported the creation of a separate category of spiritual support. A subsequent search of the literature revealed that spiritual support has been gaining prominence within the scientific literature in recent years. More articles containing the keyword “spiritual support” have been published in the last 5 years than had been published in the entire decade spanning the years 2000 to 2010.

In addition to spiritual support, there were two other social support dimensions that yielded surprising results: appraisal support and tangible support. Appraisal support was the most frequently encountered type of support in this study (25% of messages). Festinger’s (1954) social comparison theory suggests that “when an objective, non-social basis for evaluating one’s ability or opinion is…unavailable, people will…evaluate their opinions and abilities by comparing themselves with others.” Social comparison theory also suggests that, when possible, individuals prefer comparing themselves to others who are similar in terms of opinion or ability. If social comparison theory is correct, the high proportion of appraisal-related messages in the study group could be indicative of a systemic lack of objective information and health literacy among osteosarcoma patients. Another possible explanation for the high number of appraisal related posts, could be that appraisal support is highly correlated with other types of social support. Additional research should be performed to better understand this phenomenon and propose solutions if necessary.

Tangible support was the most infrequently expressed component of social support in messages posted to the study group by osteosarcoma patients in active treatment. This was surprising since tangible or instrumental support is the second most frequently researched component of social support in the literature to date. Osteosarcoma patients also have significant financial needs associated with their initial care ongoing treatment. Since most of the participants in the group have financial needs, group members may feel hesitant about asking one another to provide financial support.

Based on the finding of this research, there is no doubt that Facebook groups are revolutionizing the way those affected by rare disease connect with each other, and seek out and exchange health-related information and support. Facebook groups provide a mechanism not only for identifying disease specific groups, but also for facilitating connections between individuals with similar backgrounds or states of disease progression. While this study has identified some important defining features of sarcoma groups, it is not yet known how all of these features relate to the different types of social support that are possible within such groups. Additional research is recommended to more fully investigate the relationship between these many variables and social support.

References

American Cancer Society. (2013). Cancer Facts & Figures 2013. *Estimated Number* of New Cancer Cases and Deaths by Sex, US, 2013* (pp. 64): American Cancer Society.

American Cancer Society. (2014). What is a soft tissue sarcoma? Retrieved June 20, 2014, from http://www.cancer.org/cancer/sarcoma-adultsofttissuecancer/detailedguide/sarcoma-adult-soft-tissue-cancer-soft-tissue-sarcoma?docSelected=sarcoma-adult-soft-tissue-cancer-what-is-cancer

American Cancer Society. (2015). Osteosarcoma Overview: American Cancer Society [Brochure].

Anderson, J. G., & Taylor, A. G. (2011). Effects of healing touch in clinical practice: A systematic review of randomized clinical trials. *Journal of Holistic Nursing, 29*(3), 221-228.

Barringer, B. R., Jones, F. F., & Neubaum, D. O. (2005). A quantitative content analysis of the characteristics of rapid-growth firms and their founders. *Journal of Business Venturing, 20*(5), 663-687.

Cobb, S. (1976). Social support as a moderator of life stress. *Psychosomatic Medicine, 38*(5), 300-314.

Cohen, J. (1960). A coefficient of agreement for nominal scales. *Educational and Psychological Measurement, 20*(1), 37-46. doi: 10.1177/001316446002000104

Damron, T. A., Ward, W. G., & Stewart, A. (2007). Osteosarcoma, chondrosarcoma, and Ewing's sarcoma: National Cancer Data Base Report. *Clinical Orthopaedics and Related Research, 459*, 40-47.

Darling, J. (2007). A different view of sarcoma statistics. *ESUN, 4*(6).

Dorfman, H. D., & Czerniak, B. (1995). Bone cancers. *Cancer, 75*(S1), 203-210.

Ell, K. (1984). Social networks, social support, and health status: A review. *The Social Service Review*, 133-149.

Facebook Inc. (2015). Facebook Reports Fourth Quarter and Full Year 2014 Results. Retrieved April 20, 2015, from http://investor.fb.com/releasedetail.cfm?ReleaseID=893395

Festinger, L. (1954). A theory of social comparison processes. *Human Relations, 7*(2), 117-140.

Granovetter, M. (1973). The strength of weak ties. *American Journal of Sociology*, 1360-1380.

House, J. S. (1981). *Work stress and social support*. Reading: Addison-Wesley.

Hsieh, H.-F., & Shannon, S. E. (2005). Three approaches to qualitative content analysis. *Qualitative Health Research, 15*(9), 1277-1288.

Kraut, R. E., & Fiore, A. T. (2014). The role of founders in building online groups. *Proceedings of the 17th ACM Conference on Computer Supported Cooperative Work & Social Computing*, 722-732.

National Cancer Institute. (2015). Osteosarcoma and malignant fibrous histiocytoma of bone treatment (PDQ®). Retrieved April 20, 2015, from http://www.cancer.gov/cancertopics/pdq/treatment/osteosarcoma/HealthProfessional

Patterson, T. L., Shaw, W., & Masys, D. (1997). Improving health through computer self-health programs: Theory and practice. In P. F. Brennan, S. Schneider & E. Tornquist (Eds.), *Computers in healthcare: Information networks for community health* (pp. 219-246). New York: Springer.

Rare Diseases Act of 2002, Pub. L. No. 107-280 [H.R. 4013] (2002).

Stephens, C., Alpass, F., Towers, A., & Stevenson, B. (2011). The effects of types of social networks, perceived social support, and loneliness on the health of older people accounting for the social context. *Journal of Aging and Health, 23*(6), 887-911.

Thoits, P. A. (1982). Conceptual, methodological, and theoretical problems in studying social support as a buffer against life stress. *J Health Soc Behav, 23*(2), 145-159.

Walther, J. B., & Boyd, S. (2002). Attraction to computer-mediated social support. *Communication Technology and Society: Audience Adoption and Uses, 153-188.*

Wardell, D. W., & Weymouth, K. F. (2004). Review of studies of healing touch. *Journal of Nursing Scholarship, 36*(2), 147-154.

Weiss, R. (1974). *The provision of social relationships in Zick Rubin (Ed) doing unto others* (pp. 17–26). Englewood Cliffs: Prentice Hall.

Wright, K. (2000). Perceptions of on-line support providers: An examination of perceived homophily, source credibility, communication and social support within on-line support groups. *Communication Quarterly, 48*(1), 44-59.

Wright, K. (2002). Social support within an on-line cancer community: An assessment of emotional support, perceptions of advantages and disadvantages, and motives for using the community from a communication perspective. *Journal of Applied Communication Research, 30*(3), 195-209.

CHAPTER 2

SOCIAL NETWORKING, SOCIAL SUPPORT, AND RARE DISEASE: IMPLICATIONS FOR HEALTH PROMOTION AND EDUCATION

Abstract

According to the Office of Rare Diseases Research (ORDR) at the National Institutes of Health (NIH), rare diseases affect more than 25 million Americans. The scarcity of information, poor prognosis, and lack of viable treatment options for many conditions causes significant anxiety for rare disease patients and their families. Increasingly, rare disease populations are going online to acquire the support necessary to cope with their health challenges. The purpose of this article is to review some of the recent literature that has explored the relationship between social networking technology and social support, and to discuss implications for improving health outcomes among rare disease patients.

Introduction

According to the U.S. Census Bureau (2013), 74.8% of all households have access to the Internet. Not surprisingly, the Internet has become an important source of health information for patients and families. Approximately 72% of Internet users report having searched online for health-related information within the past year, and a majority of those searches have been related to a specific disease condition (Fox & Duggan, 2013).

The preponderance of the Internet has also fueled the introduction and rapid growth of social networking platforms and related technologies. Since 2005, social networking participation among Internet users has skyrocketed from around 8% to over 73% (Rainie, 2013). As a result, social networking has also become an important source of health-related social support. Recent studies estimate that as many as 5-7% of Internet users participate in online support groups regardless of disease status (Chou, Hunt, Beckjord, Moser, & Hesse, 2009; Fox & Purcell, 2010)

Although the Internet and social media tools have benefitted the healthcare community generally, these implements have been particularly important for patients with rare conditions where often little information about diagnosis or treatment is available. Rare disease patients are increasingly using the Internet to connect to other patients, providers, and treatment resources. In the present article, we review some of the recent work that has explored the relationship between social networking technology and social support and discuss implications for improving health outcomes among patients with rare disease conditions.

Major Concepts and Definitions

Rare Disease

According to the Office of Rare Diseases Research at the National Institutes of Health a rare disease is a condition that affects a small patient population; fewer than 200,000 people in the United States at any given time (see Rare Diseases Act of 2002).

Some rare diseases, such as Huntington's disease, amyotrophic lateral sclerosis (ALS or Lou Gehrig's disease), Tourette syndrome, and cystic fibrosis have been highly publicized and are well known to the public (see Rare Diseases Act of 2002). However, other conditions such as Dercum's disease, Kabuki syndrome, Norrie disease, and rare cancers, such as Howel-Evans syndrome and leiomyosarcoma, are not as well known.

Although specific rare diseases affect a relatively small number of people, there are more than 6,800 rare diseases that collectively affect more than 25 million Americans, and that should make rare disease research a top priority for federal agencies, pharmaceutical companies, health researchers, and public health professionals. Unfortunately, such has not always been the case. Further, because there are so many rare diseases, it is often difficult for patients and disease advocates to attract research attention to a specific condition (Posada de la Paz & Groft, 2010, p. 7).

In the early 1980s, an organization called the National Organization for Rare Disorders (NORD) was formed to lobby for national legislation that would encourage the development of drugs for rare (or orphan) diseases. NORD's leadership recognized that drug manufacturers were largely ignoring the rare disease market. Pharmaceutical companies were unmotivated to direct research and development efforts towards finding cures for rare diseases given the niche market opportunity and comparatively lower profitability of such specialized interventions. As a result, NORD lobbied for and was instrumental in the passing of the Orphan Drug Act of 1983. The Orphan Drug Act created financial incentives for drug companies to conduct clinical research and develop orphan drugs for rare diseases. Prior to the Orphan Drug Act, only 38 orphan drugs had been developed (see Rare Diseases Act of 2002). Since then, over 450 orphan drugs have been approved and marketed in the United States and an additional 2,604 drugs are at various stages of development (Orphan Drug Designation Database, 2014).

In 2002, Congress amended the Public Health Service Act to statutorily establish an Office of Rare Diseases Research at the National Institutes of Health. This important legislation, known as the Rare Diseases Act of 2002, sought to further institutionalize rare disease research and to "increase the national investment in the development of diagnostics and treatments for patients with rare diseases and disorders" (see Rare Diseases Act of 2002).

Although the Rare Disease Act of 2002 together with NORD and the Orphan Drug Act of 1983 have helped to significantly increase legislative and financial support for rare disease research, education, and treatments, the research agenda has emerged as largely unbalanced. The majority of research has centered on biological research. Research devoted to psychosocial or behavioral interventions for rare disease patients deserves greater attention. Expanding the national research agenda to include psychosocial and behavioral approaches will significantly expand our understanding of how rare disease patients heal and cope with disease.

Social Support

As a multidisciplinary field, there has been a great deal of disagreement regarding how to define social support. The phrase social support has been used to describe many different aspects of social relationships, often without a clear "conceptual definition…or valid or reliable indicators of the concept" (Thoits, 1982, p. 146). The lack of conceptual clarity about what support is and how it functions has also made it difficult at times to compare studies or generalize results. Thoits (1982) argued that in order for a definition of social support to be meaningful, it must have clear implications for operationalization. Since the aim of the present article is to discuss practical implications of social support, we will adopt a similar standard, and favor definitions of social support that can be operationalized.

One of the most frequently cited definitions of social support is "information leading the subject to believe that he is cared for and loved, esteemed and valued, and a member of a network of communication and mutual obligation" (Cobb, 1976, p. 300). While this definition can be operationalized as Thoits suggests (see Thoits, 1982), and the sentiments articulated in the definition are likely to have meaning for rare disease patients, its scope is limited to emotional support. The implications of social support for rare disease patients go beyond emotional support. Recent studies suggest that in addition to emotional support, informational support, tangible support, appraisal support, and companionship also play an important role in patient well-being (Ahmad, Khan, & Shirazi, 2013; Coulson, Buchanan, & Aubeeluck, 2007; Franks, Cronan, & Oliver, 2004; Roscoe, Corsentino, Watkins, McCall, & Sanchez-Ramos, 2009).

Kaplan et al. (1977) classified studies of social support as being either one of two definitions. First, that support involves the gratification of a person's basic social needs (approval, esteem, succorance, etc.) through social interaction with others in the external environment. Second, that social support is often defined by "the relative presence or absence of psychosocial support resources from significant others" (p. 50). While this definition is much broader than the Cobb definition, it does not address informational needs that are not social in nature, such as questions about specific drugs or treatment options. Furthermore, rare disease patients form supportive relationships with many individuals who are not considered significant others (e.g. clinicians, clergy, etc.).

House (1981) defined social support as "an interpersonal transaction involving one or more of the following: (1) emotional concern (liking, love, empathy), (2) instrumental aid (goods and services), (3) information (about the environment), or (4) appraisal (information relevant to self-evaluation)" (p. 39). This is perhaps one of the best functional definitions of social support to date. It contains clearly defined constructs that are highly operational in nature. Many of the indicators listed in this definition have been widely validated and incorporated reliably into formal social support instruments (Heitzmann & Kaplan, 1988; Tardy, 1985; Uchino, 2004; Wills & Shinar, 2000). It is important to note that House's definition does not include companionship support, sometimes also referred to as belonging support, a construct that has been included in many other studies of social support (Rook, 1987; Wan, Jaccard, & Ramey, 1996; Wills, 1991).

Social Network: Supportive or Not?

A social network is "a unit of social structure composed of the individual's social ties and the ties among them" (Gottlieb & Bergen, 2010, p. 512). A social network provides social support only "to the degree that it provides one with technical and tangible assistance, emotional support, feelings of being cared about, self-esteem etc." (Stokes, 1983, p. 142). However, Gottlieb and Bergen (2010) observed that health and human services professionals often employ the term *support system* to describe both the function and structural content of social ties. Regrettably, the merging of these terms often creates confusion about the differences between a social network and social support. The risk of confusing these terms is that it oversimplifies the complexities of social support and networks. Solomon (1986, p. 240) noted that "social networks are not always supportive" and efforts on the part of support givers sometimes fail to meet the expectations of support recipients. Wellman further cautions,

> When we declare ahead of time that a set of ties constitutes a "support system," we assume in advance precisely that which we want to leave open for study. In order to study the conditions under which individuals do get support, we must allow for the possibility that many of their ties are not necessarily supportive (1981, p. 172).

From a research perspective, it is clear social networks do not always provide support. As an example, there has been a raft of studies regarding the prevalence of bullying behavior in online environments. While estimates of cyberbullying vary widely, one recent study estimated the percentage of school-age children experiencing threatening interactions online to be as high as 75% (Kowalski, Giumetti, Schroeder, & Lattanner, 2014).

Even in environments where support providers are attentive to the needs of support recipients, there is sometimes a mismatch between desired and received support (Dakof & Taylor, 1990; Dyregrov, Dyregrov, & Raundalen, 2008; Linden & VoderMaier, 2012; Peters-Golden, 1982; Reblin et al., 2014). When support does not materialize as expected, or when support is perceived to be inadequate, additional stress or victimization can occur (Coates, Wortman, & Abbey, 1979; Silver & Wortman, 1980; Solomon, 1986; Wortman & Dunkel-Schetter, 1979; Wortman & Dunkel-Schetter, 1987; Wortman & Lehman, 1985). Additionally, the potency of supportive efforts is likely to be diminished if not properly matched to the needs of the individual (Cohen & McKay, 1984; Vachon & Stylianos, 1988).

It is also noteworthy to mention that for most individuals, social networks are not unitary in nature. Individuals often engage simultaneously in a variety of social contexts (e.g., religious groups, community organizations, Facebook, Twitter, etc.). Having multiple networks can be health promoting since "different ties within a network provide different types of support…(and) an individual cannot rely on merely one or two others for all types of assistance" (Walker, Wasserman, & Wellman, 1993, p. 72)

Online Support

Benefits of Online Support

Online support groups have increased in popularity in recent years, as computer-mediated communication technologies have enabled social networks to expand online. A number of recent studies have shown online support community participation to provide significant health benefits. Group members have reported benefitting from increased access to health information, feeling more confident in communicating with health care providers, feeling empowered and more engaged in medical decision making, experiencing greater levels of social support, and experiencing enhanced physical and mental health (Bartlett & Coulson, 2011; Bell, 2007; Chung, 2014; Coulson & Knibb, 2007; Gustafson et al., 1999; Hoybye, Johansen, & Tjornhoj-Thomsen, 2005; P. K. H. Mo & Coulson, 2012; Shigaki et al., 2013; Sillence, Briggs, Harris, & Fishwick, 2007; Van Uden-Kraan, Drossaert, Taal, Lebrun, et al., 2008; Van Uden-Kraan, Drossaert, Taal, Shaw, et al., 2008).

Although online support does not obviate the need for face-to-face support, it does offer significant benefits, especially for rare disease patients. Some of these benefits include “24/7 availability, lack of geographical barriers, a greater degree of anonymity, and ability for people to carefully read and compose messages” (Oprescu, Campo, Lowe, Andsager, & Morcuende, 2013). There is also evidence to suggest that online support groups provide greater access to a diversity of perspectives and information on a health topic than can be found on face-to-face support groups (Walther & Boyd, 2002; Wright, 2002).

Challenges of Online Support

While there are many benefits of online support groups, such groups are also not without their challenges. Kevin Wright (2000) found that the single most frequently cited disadvantage of computer-mediated support groups was the absence of haptic communication, or communication by touch. This is not insignificant given the many studies that promote healing touch as a complementary therapy for stress and anxiety, pain, high blood pressure, depression, and other adverse health conditions (Anderson & Taylor, 2011; Wardell & Weymouth, 2004). Many of these are comorbid conditions in rare disease patients. Online communities also increase the risk of miscommunication that results from a lack of face-to-face visual and aural cues (Finfgeld, 2000; White & Dorman, 2001), the risk of off-topic or negative remarks (Wright, 2000), and the potential for misinformation to be indexed and widely distributed to patients (Hoch, Norris, Lester, & Marcus, 1999; Scanfeld, Scanfeld, & Larson, 2010; White & Dorman, 2001).

While social media technology has helped to increase social support connections within many health related communities, not all communities have benefitted equally from the advent of social media. Alexander et al. (2003) conducted a comparative case study in which they evaluated the communication practices of four different health-related online support groups. Their results showed that online support groups are not all created equal. Not only do groups vary in their communication styles, but they also vary with respect to how their members relate to each other and to their external environment. These findings have also been upheld in more recent studies of online support groups, especially in groups with a strong gender or ethnic identity (Im, Chee, Lim, & Liu, 2008; Phoenix K. H. Mo, Malik, & Coulson, 2009). For example, prostate cancer survivors utilize online support groups for information gathering, while breast cancer survivors view online support groups as a source of emotional support (Blank & Adams-Blodnieks, 2007; Blank, Schmidt, Vangsness, Monteiro, & Santagata, 2010; Gooden & Winefield, 2007).

Implications for Health Promotion and Education

Patterson et al. (1997, p. 225) suggested that computerized health care services are beneficial in three ways: (1) educating patients about health-related subjects, (2) bringing about changes in health behavior, and (3) providing social support or assisting in obtaining social support. Although published almost two decades ago, this framework remains relevant for also discussing some of the major implications of social networking technology, and social support for health promotion and education practice. In the present article, the author makes one notable update to the framework: the discussion of educational benefits of computerized health care services is extended beyond patients to also include caregivers and clinicians.

Educating People

Patients. Online networks play a critical role in educating rare disease patients. Some of the reasons that patients go online include “to find second opinions, seek support and experiential information from other patients, interpret symptoms, seek information about tests and treatments, help interpret consultations, identify questions for doctors, make anonymous private inquiries, and raise awareness” (Ziebland et al., 2004, p. 1). Patients may be especially motivated to go online for information when the cause of their disease and potential treatments are unknown. Walther and Boyd (2002) have suggested that online groups also afford convenience by allowing patients to immediately begin discussing sensitive topics without worrying about the social customs or taboos that often accompany face-to-face encounters. Given the influential role that online support groups play in information dissemination and consumption, additional research is warranted to better understand not only how patients interact and support each other, but also how to design effective online environments that support such interactions.

Some researchers and clinicians have expressed concerns that Internet resources and online support groups may offer misinformation, or reinforce maladaptive belief systems among patients (Barak, Boniel-Nissim, & Suler, 2008). Such misinformation may be counterproductive to the success of an online support group (P. K. Mo & Coulson, 2013). This concern is especially salient given the lack of information available for rare disease conditions. Lack of empirically-validated information not only makes it difficult for patients to verify information on their own, but also limits the ability of physicians and other health providers to adequately address questions from patients and caregivers during the medical encounter.

Many patients report that using the Internet to acquire information about their condition increases their health literacy (Bass et al., 2006). There is also evidence to suggest that social support can help to moderate the negative effects of low health literacy on health status via information transfer (Lee, Arozullah, & Cho, 2004). In a recent survey, 39-44% of Internet users reported having searched for health related information on behalf of another individual within the past year (Fox & Duggan, 2013). Sometimes patients depend on other members of their network to locate information about their condition because they “do not have access to the Internet, are not Internet savvy, or find they are too ill to search” (Kinnane & Milne, 2010, p. 1126). As the amount of clinically accurate and reliable information grows, the benefits for patients and their families will undoubtedly increase.

Caregivers. Although caregiving has been a topic of multidisciplinary research for many years, relatively few studies have focused specifically on the importance of online support groups for caregivers (Colvin, Chenoweth, Bold, & Harding, 2004; Klemm & Wheeler, 2005). Surprisingly, the growth in online support groups in recent years has done relatively little to boost interest in this area.

Schook et al. (2014) conducted one of the few studies examining differences between patients and caregivers, in terms of their online information gathering behaviors. The results of their study suggest that patients and caregivers seek out different kinds of information from online support groups. Patients reported being more interested in practical information about their current state of being than in finding out general information about their condition. Caregivers, on the other hand, were more interested in general information about a condition, as well as specifics on end of life planning and disease prognosis. These results suggest that a one-size-fits-all approach to educating patients and their families may not be desirable.

Clinicians. Clinicians working with rare conditions should be aware of both the informational and support needs of their patients. In addition to providing general information about a disease, 41% of patients strongly expect their healthcare provider to discuss social support systems with them (Price, Desmond, & Losh, 1991). The discussions posted on disease-specific online support networks represent an ideal opportunity for clinicians to learn directly from patients and their families about common challenges and support needs associated with specific conditions.

One of many challenges for clinicians while working with rare disease patients is a lack of personal understanding or training about rare diseases. Indeed, as many as 56.7% of primary care physicians and 40% of specialists report their training in rare diseases as being either neutral, ineffective, or very ineffective (Engel, Bagal, Broback, & Boice, 2013). Health providers often express concerns about the quality of information that patients find online and the impact such information may have on the provider-patient relationship. Yet, many providers lack sufficient knowledge about some conditions to adequately advise their patients on diagnosis or treatment options (Leonard, 2004). As a result, many patients are forced to become the experts on their own disease and assume the role of physician educator (Budych, Helms, & Schultz, 2012; Engel et al., 2013). Not surprisingly, many rare disease patients express dissatisfaction with how their diagnosis has been handled by their healthcare provider (Huyard, 2009; Schieppati, Henter, Daina, & Aperia, 2008).

Emerging research suggests that patients prefer online support groups that include active physician members because physician engagement increases perceptions of informational accuracy and reliability (Vennik, Adams, Faber, & Putters, 2014). Discussion postings from rare disease specialists also likely benefit the provider community by educating less knowledgeable physicians about specific conditions.

Changing Health Behavior

In the past decade, substantial literature has emerged to show the potential uses of the Internet for motivating individuals to change behavior, and adopt healthy lifestyles that reduce the risk of disease and enhance quality of life. Research has also shown that interpersonal technologies are the most effective at persuading individuals and effecting behavior change (Cassell, Jackson, & Cheuvront, 1998). Arguably, the influence of interpersonal technologies like social media on health behavior change, especially in rare disease patients, is an area of research that deserves greater attention.

One of the areas where social networking and social support has the potential to beneficially influence or change behavior in rare disease patients concerns lifestyle activities. There is a growing body of literature that recommends physical exercise as a means of improving physical, mental, and emotional outcomes in cancer patients (Courneya, 2001; Courneya & Friedenreich, 1999; Fairey, Courneya, Field, & Mackey, 2002; Rajarajeswaran & Vishnupriya, 2009; Wolin, Schwartz, Matthews, Courneya, & Schmitz, 2012). Substantial empirical support also exists for the positive influence of social support or of having an exercise buddy on motivation to exercise or compliance with a pre-established exercise regimen (Thoman, Sansone, & Pasupathi, 2007; Young, Gittelsohn, Charleston, Felix-Aaron, & Appel, 2001). This is especially true for patients that have recently experienced a major adverse health event (Damush, Plue, Bakas, Schmid, & Williams, 2007). That being the case, online support communities seem like an ideal environment for patients and caregivers to encourage each other to adopt lifestyle changes which may facilitate recovery, or promote healthy coping.

Lee et al. (2010) and Iverson et al. (2008) observed that higher levels of Internet use result in more active participation in medical decision making by patients. When patients participate in the medical decision making process, they are more likely to adhere to treatment protocols (DiMatteo, Haskard-Zolnierek, & Martin, 2012; Parchman, Zeber, & Palmer, 2010; Wilson et al., 2010). These studies suggest that social media participation could serve to motivate patients to be more engaged with their health care providers. As patients are engaged and compliant with treatment regimens, there will also likely be improved health outcomes.

Providing Support or Assisting in Obtaining Support

Informational support.

Patients with rare diseases share many frustrations. The rarity of some diseases can make it difficult to locate knowledgeable healthcare providers about a given condition, and to receive an accurate diagnosis. A 2013 survey of rare disease patients in the United States found that it takes, on average, 8 physicians, 2 to 3 misdiagnoses, and 7.6 years of waiting to receive a proper diagnosis (Shire Pharmaceuticals, 2013). The lack of information and resources associated with some conditions significantly adds to the stress and uncertainty felt by rare disease patients and their loved ones.

One of the first questions that patients and caregivers ask after a rare disease diagnosis is, “How many other people have this condition?” It can be difficult to find an answer to this question. Often, it is not until after stumbling onto an online support group for their condition that they become “convinced there are many others living with the same disease who could provide vital information about the presentation of the disease and how best to live with their condition” (Posada de la Paz & Groft, 2010, p. 5).

Informational support in the form of information exchange is one of the most frequently reported benefits of online support networks. Informational support takes many forms, but generally involves the exchange of factual information about a medical condition, or about what to expect in the future. Such support may include referrals to external sources of information (organizations, textbooks, or websites), as well as information based on the personal experiences of other group members (Coulson et al., 2007). Given the positive impact that informational support has on patient outcomes, knowledge resources for rare disease patients (e.g., rarediseases.org; rarediseases.info.nih.gov; eurordis.org; rarediseaseday.us; etc.) should continue to be expanded in the future.

Emotional support. Rare disease patients face a variety of psychosocial challenges. Due to the small numbers and geographic dispersion of patients with rare conditions, it is not uncommon for patients and their caregivers to experience feelings of isolation, loneliness, anxiety, and despair. Chronic conditions can be especially discouraging for support providers, since there are often few noticeable improvements in the support recipient's condition. Studies on burnout among caregivers have shown that efforts to provide support may diminish over time, as support providers tire or experience difficulty in meeting caregiving demands (Murphy, Christian, Caplin, & Young, 2007; Ybema, Kuijer, Hagedoorn, & Buunk, 2002). For such individuals, online support groups represent a vital link to others with similar health-related challenges (Leonard, 2004; Rimer et al., 2005; White & Dorman, 2001). In online support groups, rare disease patients and their loved ones can obtain "practical information and reassurance that they need not face illness or disability alone" (Lamberg, 1997, p. 1422).

Recent evidence suggests that support recipients are not the only ones to benefit from emotional support; support providers also benefit. Active participation in an online support group promotes closeness and trust between group members and helps to replace feelings of disillusionment with optimism; an important ingredient for psychosocial well-being and adaptive coping (Kim et al., 2012).

Instrumental aid/tangible support. Not surprisingly, tangible support is one of the least frequently reported sources of social support in online communities (Braithwaite, Waldron, & Finn, 1999; Coulson et al., 2007; Coursaris & Liu, 2009). However, tangible support does play a role, even in virtual communities. Examples of tangible support include donating money to support a patient's treatment, responding to a request to sign an online petition on behalf of a rare disease organization, joining an online donor registry, sending an e-mail or letter to another patient or caregiver with requested information, or agreeing to meet and transport another group member to an appointment.

Appraisal. Appraisal support involves the transmission of information to the recipient that is relevant to or instrumental for self-evaluation (House, 1981). The concept is similar to Festinger's (1954) social comparison theory that suggests individuals construct their reality by comparing themselves to others. Patients often compare themselves to others as a way of evaluating their own health status. Such comparisons serve to reassure, especially during times of heightened anxiety or uncertainty (Locock & Brown, 2010).

Research suggests that a patient's perceived need for appraisal support increases as information available from experts diminishes, or as uncertainty or anxiety about health status increases (Molleman, Pruyn, & Knippenberg, 1986). While appraisal support can increase quality of life for some patients, it can also be detrimental (Brakel, Dijkstra, Buunk, & Siero, 2012).

In 2004, a health data-sharing platform called PatientsLikeMe was launched by three MIT engineers with close personal and family ties to ALS, a rare neurogenerative disease. Social support research suggests that tools like PatientsLikeMe, which facilitate appraisal support, may be highly beneficial for health outcomes. Future research is recommended to understand the practical implications of online communities like PatientsLikeMe for appraisal support, as well as the connections between appraisal support and caregiver stress and wellbeing.

<u>Companionship or belonging.</u> When major life events like a rare disease diagnosis occur, support many not materialize as patients had expected. Sometimes, health care professionals or caregivers underestimate the support needs or support available to patients (Sharpe, Butow, Smith, McConnell, & Clarke, 2005). In other cases, family and friends may themselves feel threatened by the diagnosis, and being uncertain how to help, they withdraw (Dunkel-Schetter & Wortman, 1982; Weinberg, Uken, Schmale, &

Adamek, 1995; Wortman & Lehman, 1985). When this happens, patients often feel very alone. The companionship and sense of belonging afforded by online support groups can help to redress some of the sense of loss that patients feel when their needs are not being met in other social circles.

At times, there are also disincentives to participate in face-to-face support groups. Joachim and Acorn (2003) observed that for patients with scleroderma, a rare connective tissue disorder that often manifests with severe external symptoms, "...their major fear, a fear greater than dying from scleroderma, was being 'ugly' and looking bad to themselves and others" (p. 604). Davison, Pennebaker, and Dickerson (2000) further observed that patients with stigmatized illnesses, such as AIDS, alcoholism, breast cancer and prostate cancer, or illnesses that required home confinement due to debilitating symptoms or a compromised immune system, were as much as 250 times more likely to engage in online support groups than patients with nonstigmatizing diseases. These studies support the increased need for online communities, especially for patients where face-to-face support groups are not available or appropriate.

Conclusion

Although prior studies have examined the role of computer-mediated social support in healthcare, few studies have looked at online support participation through the lens of a rare disease patient. The present study suggests that online support communities may represent an important source of social support for rare disease patients and their families. It is also clear that different types of social support (informational, emotional, tangible, appraisal, and companionship) likely serve distinct, yet important health promoting functions within these communities. Additional research is recommended to expand understanding of how online networks function to enable supportive behavior in rare disease communities, and to recommend opportunities for improvement.

References

Ahmad, M., Khan, M. A., & Shirazi, M. (2013). Perception of Social Support by Cancer Patients. *International Journal of Psychology and Behavioral Sciences*, *3*(5), 114-122.

Alexander, S. C., Peterson, J. L., & Hollingshead, A. B. (2003). Help is at your keyboard: Support groups on the Internet. In L. Frey (Ed.), *Group communication in context: Studies of bona fide groups* (Vol. 2, pp. 309-334). Manwah: Erlbaum New York.

Anderson, J. G., & Taylor, A. G. (2011). Effects of healing touch in clinical practice: A systematic review of randomized clinical trials. *Journal of Holistic Nursing, 29*(3), 221-228.

Barak, A., Boniel-Nissim, M., & Suler, J. (2008). Fostering empowerment in online support groups. *Computers in Human Behavior, 24*(5), 1867-1883.

Bartlett, Y. K., & Coulson, N. S. (2011). An investigation into the empowerment effects of using online support groups and how this affects health professional/patient communication. *Patient Education and Counseling, 83*(1), 113-119.

Bass, S. B., Ruzek, S. B., Gordon, T. F., Fleisher, L., McKeown-Conn, N., & Moore, D. (2006). Relationship of Internet health information use with patient behavior and self-efficacy: Experiences of newly diagnosed cancer patients who contact the National Cancer Institute's Cancer Information Service. *Journal of Health Communication, 11*(2), 219-236.

Bell, V. (2007). Online information, extreme communities and internet therapy: Is the internet good for our mental health? *Journal of Mental Health, 16*(4), 445-457.

Blank, T. O., & Adams-Blodnieks, M. (2007). The who and the what of usage of two cancer online communities. *Computers in Human Behavior, 23*(3), 1249-1257. doi: http://dx.doi.org/10.1016/j.chb.2004.12.003

Blank, T. O., Schmidt, S. D., Vangsness, S. A., Monteiro, A. K., & Santagata, P. V. (2010). Differences among breast and prostate cancer online support groups. *Computers in Human Behavior, 26*(6), 1400-1404. doi: http://dx.doi.org/10.1016/j.chb.2010.04.016

Braithwaite, D. O., Waldron, V. R., & Finn, J. (1999). Communication of social support in computer-mediated groups for people with disabilities. *Health Communication, 11*(2), 123-151.

Brakel, T. M., Dijkstra, A., Buunk, A. P., & Siero, F. W. (2012). Impact of social comparison on cancer survivors' quality of life: An experimental field study. *Health Psychology, 31*(5), 660.

Budych, K., Helms, T. M., & Schultz, C. (2012). How do patients with rare diseases experience the medical encounter? Exploring role behavior and its impact on patient–physician interaction. *Health Policy, 105*(2), 154-164.

Cassell, M. M., Jackson, C., & Cheuvront, B. (1998). Health communication on the Internet: An effective channel for health behavior change? *Journal of Health Communication, 3*(1), 71-79.

Chou, W.-Y. S., Hunt, Y. M., Beckjord, E. B., Moser, R. P., & Hesse, B. W. (2009). Social media use in the United States: Implications for health communication. *Journal of Medical Internet Research, 11*(4).

Chung, J. E. (2014). Social networking in online support groups for health: How online social networking benefits patients. *Journal of Health Communication, 19*(6), 639-659.

Coates, D., Wortman, C. B., & Abbey, A. (1979). Reactions to victims. *New Approaches to Social Problems*, 21-52.

Cobb, S. (1976). Presidential Address-1976. Social support as a moderator of life stress. *Psychosom Med, 38*(5), 300-314.

Cohen, S., & McKay, G. (1984). Social support, stress, and the buffering hypothesis: A theoretical analysis. *Handbook of Psychology and Health, 4*, 253-267.

Colvin, J., Chenoweth, L., Bold, M., & Harding, C. (2004). Caregivers of older adults: Advantages and disadvantages of Internet-based social support*. *Family Relations, 53*(1), 49-57.

Coulson, N. S., Buchanan, H., & Aubeeluck, A. (2007). Social support in cyberspace: A content analysis of communication within a Huntington's disease online support group. *Patient Education and Counseling, 68*(2), 173-178.

Coulson, N. S., & Knibb, R. C. (2007). Coping with food allergy: Exploring the role of the online support group. *CyberPsychology & Behavior, 10*(1), 145-148.

Courneya, K. S. (2001). Exercise interventions during cancer treatment: Biopsychosocial outcomes. *Exercise and Sport Sciences Reviews, 29*(2), 60-64.

Courneya, K. S., & Friedenreich, C. M. (1999). Physical exercise and quality of life following cancer diagnosis: A literature review. *Annals of Behavioral Medicine, 21*(2), 171-179.

Coursaris, C. K., & Liu, M. (2009). An analysis of social support exchanges in online HIV/AIDS self-help groups. *Computers in Human Behavior, 25*(4), 911-918.

Dakof, G. A., & Taylor, S. E. (1990). Victims' perceptions of social support: What is helpful from whom? *Journal of Personality and Social Psychology, 58*(1), 80-89. doi: 10.1037/0022-3514.58.1.80

Damush, T. M., Plue, L., Bakas, T., Schmid, A., & Williams, L. S. (2007). Barriers and facilitators to exercise among stroke survivors. *Rehabilitation Nursing, 32*(6), 253-262.

Davison, K. P., Pennebaker, J. W., & Dickerson, S. S. (2000). Who talks? The social psychology of illness support groups. *American Psychologist, 55*(2), 205.

DiMatteo, M. R., Haskard-Zolnierek, K. B., & Martin, L. R. (2012). Improving patient adherence: A three-factor model to guide practice. *Health Psychology Review, 6*(1), 74-91.

Dunkel-Schetter, C., & Wortman, C. B. (1982). The interpersonal dynamics of cancer: Problems in social relationships and their impact on the patient. In H. S. Friedman & M. R. DiMatteo (Eds.), *Interpersonal issues in health care* (pp. 69-100). New York: Academic Press.

Dyregrov, A., Dyregrov, K., & Raundalen, M. (2008). *Effective grief and bereavement support: The role of family, friends, colleagues, schools and support professionals*. London: Jessica Kingsley Publishers.

Engel, P. A., Bagal, S. B., Broback, M., & Boice, N. (2013). Physician and patient perceptions regarding physician training in rare diseases: The need for stronger educational initiatives for physicians. *Journal of Rare Disorders, 1*(2), 1-15.

Fairey, A. S., Courneya, K. S., Field, C. J., & Mackey, J. R. (2002). Physical exercise and immune system function in cancer survivors. *Cancer, 94*(2), 539-551.

Festinger, L. (1954). A theory of social comparison processes. *Human Relations, 7*(2), 117-140.

Finfgeld, D. L. (2000). Therapeutic groups online: The good, the bad, and the unknown. *Issues in Mental Health Nursing, 21*(3), 241-255.

Fox, S., & Duggan, M. (2013). Health online 2013. Retrieved May 10, 2014, from http://www.pewinternet.org/files/old-media//Files/Reports/PIP_HealthOnline.pdf

Fox, S., & Purcell, K. (2010). Social media & health. Retrieved May 10, 2014 from http://www.pewinternet.org/2010/03/24/social-media-and-health/

Franks, H. M., Cronan, T. A., & Oliver, K. (2004). Social support in women with fibromyalgia: Is quality more important than quantity? *Journal of Community Psychology, 32*(4), 425-438.

Gooden, R. J., & Winefield, H. R. (2007). Breast and prostate cancer online discussion boards: A thematic snalysis of gender differences and similarities. *Journal of Health Psychology, 12*(1), 103-114.

Gottlieb, B. H., & Bergen, A. E. (2010). Social support concepts and measures. *Journal of Psychosomatic Research, 69*(5), 511-520.

Gustafson, D. H., Hawkins, R., Boberg, E., Pingree, S., Serlin, R. E., Graziano, F., & Chan, C. L. (1999). Impact of a patient-centered, computer-based health information/support system. *American Journal of Preventive Medicine, 16*(1), 1-9.

Heitzmann, C. A., & Kaplan, R. M. (1988). Assessment of methods for measuring social support. *Health Psychology, 7*(1), 75.

Hoch, D. B., Norris, D., Lester, J. E., & Marcus, A. D. (1999). Information exchange in an epilepsy forum on the World Wide Web. *Seizure, 8*(1), 30-34.

House, J. S. (1981). *Work stress and social support*. Reading: Addison-Wesley.

Hoybye, M. T., Johansen, C., & Tjornhoj-Thomsen, T. (2005). Online interaction. Effects of storytelling in an internet breast cancer support group. *Psycho-Oncology, 14*(3), 211-220.

Huyard, C. (2009). What, if anything, is specific about having a rare disorder? Patients' judgements on being ill and being rare. *Health Expectations, 12*(4), 361-370.

Im, E.-O., Chee, W., Lim, H.-J., & Liu, W.-M. (2008). An online forum exploring needs for help of patients with cancer: Gender and ethnic differences. *Oncology Nursing Forum, 35*(4), 653-660. doi: 10.1188/08.onf.653-660

Iverson, S. A., Howard, K. B., & Penney, B. K. (2008). Impact of internet use on health-related behaviors and the patient-physician relationship: a survey-based study and review. *JAOA: Journal of the American Osteopathic Association, 108*(12), 699-711.

Joachim, G., & Acorn, S. (2003). Life with a rare chronic disease: The scleroderma experience. *Journal of Advanced Nursing, 42*(6), 598-606.

Kaplan, B. H., Cassel, J. C., & Gore, S. (1977). Social support and health. *Medical Care*, 47-58.

Kim, E., Han, J. Y., Moon, T. J., Shaw, B., Shah, D. V., McTavish, F. M., & Gustafson, D. H. (2012). The process and effect of supportive message expression and reception in online breast cancer support groups. *Psycho-Oncology, 21*(5), 531-540.

Kinnane, N. A., & Milne, D. J. (2010). The role of the Internet in supporting and informing carers of people with cancer: A literature review. *Supportive Care in Cancer, 18*(9), 1123-1136.

Klemm, P., & Wheeler, E. (2005). Cancer caregivers online: Hope, emotional roller coaster, and physical/emotional/psychological responses. *Computers Informatics Nursing, 23*(1), 38-45.

Kowalski, R. M., Giumetti, G. W., Schroeder, A. N., & Lattanner, M. R. (2014). Bullying in the digital age: A critical review and meta-analysis of cyberbullying research among youth. *Psychological Bulletin*, *140*(4), 1073-1137.

Lamberg, L. (1997). Online support group helps patients live with, learn more about the rare skin cancer ctcl-mf. *JAMA, 277*(18), 1422-1423. doi: 10.1001/jama.1997.03540420016005

Lee, C. J., Gray, S. W., & Lewis, N. (2010). Internet use leads cancer patients to be active health care consumers. *Patient Education and Counseling, 81*, S63-S69.

Lee, S. D., Arozullah, A. M., & Cho, Y. I. (2004). Health literacy, social support, and health: A research agenda. *Social Science & Medicine, 58*(7), 1309-1321.

Leonard, H. (2004). How can the Internet help parents of children with rare neurologic disorders? *Journal of Child Neurology, 19*(11), 902-907.

Linden, W., & VoderMaier, A. (2012). Mismatch of desired versus perceived social support and associated levels of anxiety and depression in newly diagnosed cancer patients. *Supportive Care in Cancer, 20*(7), 1449-1456.

Locock, L., & Brown, J. B. (2010). ‘All in the same boat’? Patient and carer attitudes to peer support and social comparison in Motor Neurone Disease (MND). *Social Science & Medicine, 71*(8), 1498-1505.

Mo, P. K., & Coulson, N. S. (2013). Are online support groups always beneficial? A qualitative exploration of the empowering and disempowering processes of participation within HIV/AIDS-related online support groups. *International Journal of Nursing Studies*, *51*(7), 983-993.

Mo, P. K. H., & Coulson, N. S. (2012). Developing a model for online support group use, empowering processes and psychosocial outcomes for individuals living with HIV/AIDS. *Psychology & Health, 27*(4), 445-459.

Mo, P. K. H., Malik, S. H., & Coulson, N. S. (2009). Gender differences in computer-mediated communication: A systematic literature review of online health-related support groups. *Patient Education and Counseling, 75*(1), 16-24. doi: http://dx.doi.org/10.1016/j.pec.2008.08.029

Molleman, E., Pruyn, J., & Knippenberg, A. (1986). Social comparison processes among cancer patients. *British Journal of Social Psychology, 25*(1), 1-13.

Oprescu, F., Campo, S., Lowe, J., Andsager, J., & Morcuende, J. A. (2013). Online information exchanges for parents of children with a rare health condition: Key findings from an online support community. *Journal of Medical Internet Research, 15*(1).

Parchman, M. L., Zeber, J. E., & Palmer, R. F. (2010). Participatory decision making, patient activation, medication adherence, and intermediate clinical outcomes in type 2 diabetes: A STARNet study. *The Annals of Family Medicine, 8*(5), 410-417.

Patterson, T. L., Shaw, W., & Masys, D. (1997). Improving health through computer self-health programs: Theory and practice. In P. F. Brennan, S. Schneider & E. Tornquist (Eds.), *Computers in healthcare: Information networks for community health* (pp. 219-246). New York: Springer.

Peters-Golden, H. (1982). Breast cancer: Varied perceptions of social support in the illness experience. *Social Science & Medicine, 16*(4), 483-491.

Posada de la Paz, M., & Groft, S. C. (2010). *Rare diseases epidemiology* (Vol. 686). Dordrecht: Springer.

Price, J. H., Desmond, S. M., & Losh, D. P. (1991). Patients' expectations of the family physician in health promotion. *American Journal of Preventive Medicine*. 7(1), 33-39.

Rainie, L. (2013). Social Networking Fact Sheet. Retrieved May 10, 2014, from http://www.pewinternet.org/fact-sheets/social-networking-fact-sheet/

Rajarajeswaran, P., & Vishnupriya, R. (2009). Exercise in cancer. *Indian Journal of Medical and Paediatric Oncology: Official Journal of Indian Society of Medical & Paediatric Oncology, 30*(2), 61.

Rare Diseases Act of 2002, Pub. L. No. 107-280 [H.R. 4013] (2002).

Reblin, M., Cloyes, K. G., Carpenter, J., Berry, P. H., Clayton, M. F., & Ellington, L. (2014). Social support needs: Discordance between home hospice nurses and former family caregivers. *Palliative and Supportive Care*, 1-8.

Rimer, B. K., Lyons, E. J., Ribisl, K. M., Bowling, J. M., Golin, C. E., Forlenza, M. J., & Meier, A. (2005). How new subscribers use cancer-related online mailing lists. *Journal of Medical Internet Research, 7*(3).

Rook, K. S. (1987). Social support versus companionship: Effects on life stress, loneliness, and evaluations by others. *Journal of Personality and Social Psychology, 52*(6), 1132.

Roscoe, L. A., Corsentino, E., Watkins, S., McCall, M., & Sanchez-Ramos, J. (2009). Well-being of family caregivers of persons with late-stage Huntington's disease: Lessons in stress and coping. *Health Communication, 24*(3), 239-248.

Scanfeld, D., Scanfeld, V., & Larson, E. L. (2010). Dissemination of health information through social networks: Twitter and antibiotics. *American Journal of Infection Control, 38*(3), 182-188.

Schieppati, A., Henter, J.-I., Daina, E., & Aperia, A. (2008). Why rare diseases are an important medical and social issue. *The Lancet, 371*(9629), 2039-2041.

Schook, R. M., Linssen, C., Schramel, F. M., Festen, J., Lammers, E., Smit, E. F., . . . Westerman, M. J. (2014). Why do patients and caregivers seek answers from the Internet and online lung specialists? A qualitative study. *Journal of Medical Internet Research, 16*(2).

Sharpe, L., Butow, P., Smith, C., McConnell, D., & Clarke, S. (2005). The relationship between available support, unmet needs and caregiver burden in patients with advanced cancer and their carers. *Psycho‐Oncology, 14*(2), 102-114.

Shigaki, C. L., Smarr, K. L., Siva, C., Ge, B., Musser, D., & Johnson, R. (2013). RAHelp: An online intervention for individuals with rheumatoid arthritis. *Arthritis Care & Research*, *65*(10), 1573-1581.

Shire Pharmaceuticals. (2013). Rare Disease Impact Report. Retrieved on May 10, 2014, from http://shire-hgt.isebox.net/survey/results

Sillence, E., Briggs, P., Harris, P. R., & Fishwick, L. (2007). How do patients evaluate and make use of online health information? *Social Science & Medicine, 64*(9), 1853-1862.

Silver, R. L., & Wortman, C. B. (1980). Coping with undesirable life events. *Human Helplessness: Theory and Applications, 279*, 375.

Solomon, S. D. (1986). Mobilizing social support networks in times of disaster. In C. R. Figley (Ed.), *Trauma and its wake* (Vol. 2, pp. 232-263). New York: Brunner/Mazel.

Stokes, J. P. (1983). Predicting satisfaction with social support from social network structure. *American Journal of Community Psychology, 11*(2), 141-152.

Tardy, C. H. (1985). Social support measurement. *American Journal of Community Psychology, 13*(2), 187-202.

Thoits, P. A. (1982). Conceptual, methodological, and theoretical problems in studying social support as a buffer against life stress. *J Health Soc Behav, 23*(2), 145-159.

Thoman, D. B., Sansone, C., & Pasupathi, M. (2007). Talking about interest: Exploring the role of social interaction for regulating motivation and the interest experience. *Journal of Happiness Studies, 8*(3), 335-370.

U.S. Census Bureau. (2013). Computer & Internet Trends in America. Retrieved May 10, 2014, from www.census.gov/hhes/computer/files/2012/Computer_Use_Infographic_FINAL.pdf.

U.S. Food and Drug Administration (FDA). (2014). Orphan Drug Designation Database. Retrieved May 10, 2014, from www.accessdata.fda.gov/scripts/opdlisting/oopd/.

Uchino, B. N. (2004). The meaning and measurement of social support. In *Social support and physical health: Understanding the health consequences of relationships* (pp. 9-32). New Haven: Yale University Press.

Vachon, M. L. S., & Stylianos, S. K. (1988). The role of social support in bereavement. *Journal of Social Issues, 44*(3), 175-190.

Van Uden-Kraan, C. F., Drossaert, C. H. C., Taal, E., Lebrun, C. E. I., Drossaers-Bakker, K. W., Smit, W. M., . . . Van de Laar, M. (2008). Coping with somatic illnesses in online support groups: Do the feared disadvantages actually occur? *Computers in Human Behavior, 24*(2), 309-324.

Van Uden-Kraan, C. F., Drossaert, C. H. C., Taal, E., Shaw, B. R., Seydel, E. R., & Van de Laar, M. A. F. J. (2008). Empowering processes and outcomes of participation in online support groups for patients with breast cancer, arthritis, or fibromyalgia. *Qualitative Health Research, 18*(3), 405-417.

Vennik, F. D., Adams, S. A., Faber, M. J., & Putters, K. (2014). Expert and experiential knowledge in the same place: Patients' experiences with online communities connecting patients and health professionals. *Patient Education and Counseling, 95*(2), 265-270.

Walker, M. E., Wasserman, S., & Wellman, B. (1993). Statistical models for social support networks. *Sociological Methods & Research, 22*(1), 71-98.

Walther, J. B., & Boyd, S. (2002). Attraction to computer-mediated social support. *Communication Technology and Society: Audience Adoption and Uses, 153-188.*

Wan, C. K., Jaccard, J., & Ramey, S. L. (1996). The relationship between social support and life satisfaction as a function of family structure. *Journal of Marriage and the Family*, 502-513.

Wardell, D. W., & Weymouth, K. F. (2004). Review of studies of healing touch. *Journal of Nursing Scholarship, 36*(2), 147-154.

Weinberg, N., Uken, J. S., Schmale, J., & Adamek, M. (1996). Therapeutic factors: Their presence in a computer-mediated support group. *Social Work with Groups, 18*(4), 57-69.

Wellman, B. (1981). Applying network analysis to the study of support. In B. H. Gottlieb (Ed.), *Social networks and social support* (pp. 171-200). Beverly Hills: Sage Publications.

White, M., & Dorman, S. M. (2001). Receiving social support online: Implications for health education. *Health Education Research, 16*(6), 693-707.

Wills, T. A. (1991). Social support and interpersonal relationships. In M. S. Clark (Ed.), *Prosocial Behavior, Review of Personality and Social Psychology* (pp. 265-289). Thousand Oaks: Sage Publications, Inc.

Wills, T. A., & Shinar, O. (2000). Measuring perceived and received social support. In S. Cohen, L. G. Underwood & B. H. Gottlieb (Eds.), *Social support measurement and intervention: A guide for health and social scientists* (pp. 86-135). New York: Oxford University Press.

Wilson, S. R., Strub, P., Buist, A. S., Knowles, S. B., Lavori, P. W., Lapidus, J., . . . Group, B. O. o. A. T. S. (2010). Shared treatment decision making improves adherence and outcomes in poorly controlled asthma. *American Journal of Respiratory and Critical Care Medicine, 181*(6), 566.

Wolin, K. Y., Schwartz, A. L., Matthews, C. E., Courneya, K. S., & Schmitz, K. H. (2012). Implementing the exercise guidelines for cancer survivors. *The Journal of Supportive Oncology, 10*(5), 171.

Wortman, C. B., & Dunkel-Schetter, C. (1987). Conceptual and methodological issues in the study of social support. In A. Baum, S. Taylor, & J.E. Singer (Eds.), *Handbook of psychology and health, vol. 5* (pp. 63-108). Hillsdale: Erlbaum.

Wortman, C. B., & Dunkel-Schetter, C. (1979). Interpersonal relationships and cancer: A theoretical analysis. *Journal of Social Issues, 35*(1), 120-155.

Wortman, C. B., & Lehman, D. R. (1985). Reactions to victims of life crises: Support attempts that fail. *Social Support: Theory, Research and Applications*, 463-489.

Wright, K. (2000). Perceptions of on-line support providers: An examination of perceived homophily, source credibility, communication and social support within on-line support groups. *Communication Quarterly, 48*(1), 44-59.

Wright, K. (2002). Social support within an on-line cancer community: An assessment of emotional support, perceptions of advantages and disadvantages, and motives for using the community from a communication perspective. *Journal of Applied Communication Research, 30*(3), 195-209.

Young, D. R., Gittelsohn, J., Charleston, J., Felix-Aaron, K., & Appel, L. J. (2001). Motivations for exercise and weight loss among African-American women: Focus group results and their contribution towards program development. *Ethnicity and Health, 6*(3-4), 227-245.

Ziebland, S., Chapple, A., Dumelow, C., Evans, J., Prinjha, S., & Rozmovits, L. (2004). How the internet affects patients' experience of cancer: A qualitative study. *BMJ, 328*(7439), 564.

CHAPTER 3

SOCIAL MEDIA AND SARCOMA: A QUALITATIVE CONTENT ANALYSIS OF FACEBOOK SUPPORT GROUPS

Abstract

Background

According to the National Institutes of Health, as many as 1 in 13 Americans have been diagnosed with a rare disease condition ("FAQs About Rare Diseases", 2014). To date, relatively few research studies have examined the impact of the rapid expansion of social media technologies on the availability of support for rare disease patients and their families. Additional research is needed to understand the risks and benefits associated with social media use among those affected by rare disease.

Objective

The focus of the current study is on one area of rare disease: sarcoma. The aim of the current study is two-fold: 1) to increase scientific understanding of the role and influence of social media on the experience of support for sarcoma patients and their families, and 2) to characterize via content analysis the followers, focus, founders and formation of sarcoma related Facebook groups.

Methods

Facebook's built-in search engine and a predetermined list of sarcoma diagnoses were used to locate online groups for study. In order to be selected, groups were required to contain a sarcoma diagnosis in the title, to be conducted primarily in the English language, and to be related only to human forms of sarcoma. Using information extracted from group titles and descriptions, three coding schemes were developed to guide content analysis: group focus or orientation, founder treatment status, and founder disease affiliation status. Content analysis of group titles, group descriptions, and for open groups, the initial posts of group founders, was used to classify all of the sampled groups and their founders. In cases where classification could not be successfully completed using Facebook data, publicly available secondary data sources were used to determine classification. Using membership lists for all groups in the study sample, descriptive statistics were generated for each group and classification type. The amount of overlapping membership between groups was also calculated.

Results

The study sample included 82 sarcoma support groups on Facebook. Total cumulative membership for the selected groups was calculated at nearly 26,000 members. Membership size for the selected groups ranged from 2 to 4,841 members, and the average membership per group was 316 members. One of the most significant findings of the current investigation was that groups could be classified according to three different coding schemes: group focus or orientation (person vs. population), founder treatment status (patient or nonpatient) and founder disease affiliation status (active treatment, survivor, in memoriam, or external organization). Group classification revealed 40.2% of groups to be person-focused and 59.8% of groups to be population-focused. Nonpatient founders accounted for the greatest number of groups; 81.8% of person-focused groups and 75.5% of population-focused groups. The vast majority of person-focused groups (81.8%) had a founder associated with a patient in active treatment, while only 12.2% of population-focused groups had that affiliation. Population-focused group founders were most likely to be affiliated with a cancer survivor (30.6%), a deceased patient (30.6%), or an external organization (26.5%). According to group founders, both intrinsic and extrinsic motivations contributed to the formation of online support groups. For patient founders in the study sample, being a survivor was the biggest motivation for group formation (56% of cases), but being in active treatment also accounted for a significant portion of group formations (>40%). Nonpatient founders were motivated by a variety of factors to form online sarcoma support groups. The primary motivations were: being connected to someone in active treatment (39%), keeping the memory of a loved one alive (30%), and promoting an external organization (20%).

Conclusion

Facebook groups are revolutionizing the way those affected by rare disease connect with each other, and seek out and exchange health-related information and support. Facebook groups provide a mechanism not only for identifying disease specific groups, but also for facilitating connections between individuals with similar backgrounds or states of disease progression. While this study has identified some important defining features of sarcoma groups, it is not yet known how these features relate to the different types of social support that are possible within such groups. Additional research is recommended to more fully investigate the relationship between group followers, group focus, group founders, group formation and social support.

Introduction

Rare disease is a serious health concern in the United States. Although specific conditions affect a relatively small number of people, fewer than 200,000, there are more than 6,800 rare diseases and counting (“FAQ About Rare Diseases”, 2014). The National Institutes of Health (NIH) estimates more than 25 million Americans and their families are affected (“FAQ About Rare Diseases”, 2014). Based on recent population estimates, that means that anywhere from 8-21% of all households in the United States have at least one person who has been diagnosed with a rare disease (U.S. Census Bureau, 2014). These figures have left many wondering what more can be done to support rare disease patients and their families.

In 2002, Congress amended the Public Health Service Act to statutorily establish an Office of Rare Diseases Research at the NIH. The amendment, which was part of the Rare Diseases Act of 2002, was hailed by rare disease advocates as an acknowledgement that not enough was being done to support the rare disease population in the United States. It was hoped that this legislation would “increase the national investment in the development of diagnostics and treatments for patients with rare diseases and disorders” (see Rare Diseases Act of 2002).

While these efforts have helped, there is still much more to be done. Because there are so many rare diseases, it is often difficult for patients and disease advocates to attract research attention to a specific condition (Posada de la Paz & Groft, 2010, p. 7). In 2014, the NIH estimates that it will spend approximately $142 per rare disease patient on orphan condition budget research ("Estimates of Funding for Various Research, Condition, and Disease Categories (RCDC)", 2014). This is a drop in the bucket compared to the nearly $23,000 per year that some estimate the average rare disease patient spends on treatment (Jolley, 2014). For some patients, treatments can run into the hundreds of thousands of dollars per year. The weight of this financial burden combined with the physical, mental, and emotional costs of illness can be overwhelming.

Without question, the road for most rare disease patients and their families is long and hard. Often, the journey begins with a constellation of symptoms that cannot be explained. Some patients wait years for a diagnosis, and for others, a definitive diagnosis never comes. Many rare disease patients and caregivers struggle to cope in isolation. The dearth of information and scarce resources associated with many diseases significantly adds to the stress of living with a rare health condition. Healthcare providers are expected to have all of the answers. Many providers admit that it is hard to be an expert in something they know nothing about (Paturel, 2012, p. 30).

In order to cope with the uncertainty of their condition, many patients are turning to social media for help. This should come as no surprise given that "73% of online adults [in the United States] use social networking sites" (Rainie, 2013). The average American is already spending up to 3 hours per day communicating via social media (Jacobsen & Forste, 2011; O'Brien, 2013, p. xx; Raacke & Bonds-Raacke, 2008). The large number of registered users on Facebook, over 1.23 billion worldwide (Facebook

Inc., 2014), and ability for users to form groups around specific topics or disease conditions, has been particularly helpful for those affected by rare diseases. Not only has Facebook made it easier to find and befriend others facing similar health conditions, but Facebook groups have also become an important conduit of supportive exchange.

The current study examines the role and influence of Facebook groups on the availability of social support within a specific segment of the rare disease population; those affected by sarcoma. Sarcoma is a grouping of extremely rare forms of cancer that develop from tissues like bone or muscle (American Cancer Society, 2014). Of the approximately 1.6 million cases of cancer in the United States each year, fewer than 15,000 cases are sarcoma (American Cancer Society, 2013; Darling, 2007). Data from existing online support groups was used to characterize the followers, focus, founders and formation of sarcoma related Facebook groups. It is hoped that the insights gleaned from this study will encourage additional research in this important area.

Methods

Sampling Procedure

Sample selection. The current study revolved around Facebook groups with an explicit emphasis on sarcoma cancer. To identify relevant groups, Facebook's built-in search engine and a predetermined list of sarcoma diagnoses were used to identify as many support groups related to sarcoma as possible. Groups were identified by typing a diagnosis into the Facebook search engine and then selecting "Find all groups named [diagnosis]" when prompted. See Table 3.1 for a list of included diagnostic keywords. In order to be selected, groups were required to contain a sarcoma diagnosis in the title, to be conducted primarily in the English language, and to be related only to human forms of sarcoma. Of the approximately 100 groups that were identified by the Facebook search engine, the chosen sampling criteria resulted in the successful identification and selection of 82 unique sarcoma-related support groups. The groups that were excluded from the study did not have any posts (dead groups), were foreign language groups, or were related to feline or canine forms of sarcoma.

Although there were likely many highly relevant groups that did not meet the selected search criteria, there were various reasons why the selected approach was adopted. First, there are an almost unlimited number of possible naming conventions on Facebook. Given the large number of groups on Facebook (over 600 million), there was no reliable way for the researcher to identify sarcoma groups without searching for groups with disease names in their titles. Were it possible to search the actual content of Facebook groups, additional groups might have been identifiable. However, Facebook's privacy policies restrict content indexing, especially for closed or secret groups (Facebook Inc., 2014). Second, searching by title ensured that all groups selected for study would be relevant to the activity of interest; to characterize via content analysis the followers, focus, founders, and formation of sarcoma support groups on Facebook.

<u>Data extraction.</u> The Facebook search engine produces a list of groups that match desired keywords. The search results include: a list of all of the groups whose names contain the target keyword, the privacy status for each group (open or closed), the number of members in each group, and an abbreviated version of the group description (if available). In the current study, the results list for each keyword searched was printed. Results lists ranged from one to nine pages in length. After printing, each page was cut

into strips so that every strip of paper contained only one group's worth of information. The group name, group description, privacy status, and member count for each group were also entered into a Microsoft Excel spreadsheet for future analysis. The membership lists for all 82 sarcoma support groups were downloaded and aggregated into a separate tab of the same Excel workbook for additional analysis. Unlike secret groups, membership lists for open and closed groups are visible to all Facebook users, regardless of the viewer's membership status. This capability is disclosed in Facebook's privacy policy, and accepted by all users as part of the Facebook terms of service (see Table 3.2).

Data Analysis

Data analysis began by selecting at random and analyzing the content of 20 of the paper strips created during the data extraction process. The purpose of this effort was to develop an initial coding scheme to guide the identification and classification of all 82 sarcoma support groups in the study sample. The initial content analysis focused only on the titles of the groups. As the researcher read through the group titles, it became readily apparent that there were two types of sarcoma support groups within the sample; groups where the primary focus was on a specific individual and his/her treatment (patient-focused groups), and groups where the focus was on many individuals affected by the same health condition (population-focused groups). This point of distinction was evident in the naming conventions of nearly every group. For example, patient-focused groups generally included a specific patient's name in the title (e.g., "Help for (Patient Name) with (Disease Name)") or had group titles that were written in the first person (e.g., "My Diagnosis of (Disease Name)"), whereas population-focused groups tended to adopt a more generic naming convention (e.g., “(Disease Name) Awareness”) without making reference to a specific patient. Using the identified naming conventions, the researcher classified and separated all of the groups into two piles: one for person-focused groups, and one for population-focused groups.

The next step was to verify the accuracy of the initial group focus classifications via manifest content analysis of group descriptions, and for open groups, the early postings of group founders. The full group descriptions available directly on Facebook were used for data verification purposes. Fortunately, 80% (66/82) of groups in the study sample included a narrative description. Where group descriptions were lacking, or where group focus was not apparent in a description, the first few posts in a group often contained sufficient information to confirm a group’s focus. When available and open to inspection, the researcher utilized this information on an as needed basis to confirm the focus.

In the 16 cases where no group description was given, only 6 of the groups were closed groups that precluded the viewing of group postings. Five of the 6 closed groups contained enough information in their titles that they could be clearly classified. In all five cases, other publicly available Facebook pages were located that also confirmed the focus of each of the groups. These sources were identified using Google searches of the group founders. The founder of the one closed group with both an ambiguous title and no description, had set up another publicly facing page on Facebook where she referenced her treatment journal. This confirmed her group was patient-focused. Using the methods described above, the accuracy of 100% of the initial group focus classifications was confirmed.

Following the classification of groups according to group focus, the next step was to reread all of the group titles and full descriptions, and for open groups, the initial postings of group founders to understand why groups had been established as either person- or population-focused. Insights were gleaned from manifest analysis of content contained within the groups. It was observed that group titles and group descriptions, where available, were presented as written by group founders, unless subsequently updated by other group administrators (a functional requirement imposed by Facebook). Group founders typically explained in the title, description, or an initial posting at the time the group was founded, the reasoning behind the group's formation. Based on whether or not text was written as a first-, second-, or third-person account, groups were identified as having a patient or nonpatient founder. Using this approach, 82 sarcoma support groups were classified as having either a patient or nonpatient founder.

In addition to the group focus and founder status, narrative themes related to the founder's relationship to, or experience with sarcoma were identified in the sample groups. An additional coding framework based on these themes was developed and utilized to further subcategorize the groups. This framework, referred to as the "disease affiliation schema," emerged from analysis of the manifest content contained in group titles, group descriptions, and the initial postings of group founders for open groups. The disease affiliation schema contains four main classifications of sarcoma support group founders:

- Active Treatment: the founder has either had sarcoma themselves or has formed the group on behalf of someone who is undergoing active treatment for sarcoma.

- Survivor: the founder has either survived sarcoma cancer themselves or has formed the group on behalf of someone who has survived sarcoma. To be classified as a survivor group founder, there must be references to someone being cancer free, in remission, having clear scans, or that there is no evidence of disease (NED).
- In Memoriam: the founder knows someone who has passed away from sarcoma and this was the motivation for starting the group.
- External Organization: the founder started the group on behalf of an organization that is somehow affiliated with sarcoma cancer (lobbying, fundraising, research, etc.).

Using the disease affiliation schema described above, founders of all of the support groups included in the study sample were classified. Of the 47 groups with open privacy settings, 92% (44/47) were categorized by disease affiliation using only the information contained in the group title, description, or initial posts. The remaining three groups were classified based on other publicly available data sources, such as obituaries connected to group founders. Of the 35 groups with closed privacy settings, 71% (25/35) were categorized using only group titles and descriptions. The remaining 10 groups were classified based on publicly available information, such as Facebook profile pages, news stories, or obituaries associated with group founders.

Once the coding process had been finalized for all 82 sample groups using the three schemas (group focus or orientation, founder treatment status, and founder disease affiliation status), the Microsoft Excel spreadsheet was updated with the group focus, founder treatment status, and founder disease affiliation status for each of the groups.

Using the combined information on the spreadsheet, from Facebook and the data analysis, descriptive statistics were computed for each of the groups, as well as for each of the study classifications (Table 3.3). The researcher also built a pivot table using the membership lists from each group to determine the amount of overlap in membership between all of the sampled groups.

Results

On June 17, 2014, a search of Facebook for sarcoma related groups was conducted. The search procedure and exclusion criteria mentioned earlier resulted in the identification of 82 online support groups for study. The diagnostic focus of the selected groups varied widely, but all groups were focused on sarcoma. The top five diagnostic keywords explicitly named in group titles were as follows: Ewing's sarcoma (21%), osteosarcoma (20%), leiomyosarcoma (17%), sarcoma (undifferentiated) (17%) and rhabdomyosarcoma (11%) (Table 3.4).

Group Followers

At the time of the study, total cumulative membership for the selected groups was calculated at nearly 26,000 members. Membership size for the selected groups ranged from 2 to 2,715 members and the average membership per group was 316 members. It is not known what the historical membership for these groups has been. However, the total historical number of group participants is undoubtedly much higher than 26,000. When an individual drops their membership in a Facebook group, they are no longer counted in the membership numbers, but their postings remain active, and are visible to other members of the group.

The amount of overlap in membership between groups was also examined. Of the nearly 26,000 members across all of the groups, more than 21,500 of these individuals were unique group participants. In fact, 88% (19,001/21,561) of all sarcoma support group participants in the sample participated in only one group. Of the 2,650 individuals who joined two or more support groups, 94% only joined groups of one orientation type (person-focused or population-focused). Of those individuals, 99.4% only joined population-focused groups.

Group Focus

Group classification revealed that population-focused groups were more prevalent than patient-focused groups within the study sample. As shown in Table 3.3, 59.8% (49/82) of the groups were population-focused, whereas 40.2% (33/82) of the groups were person-focused. The group focus coding scheme was found to be highly accurate. In 82% (28/34) of cases, groups that were initially classified as person-focused based on a person's name in the title, were subsequently confirmed to be person-focused. Of the six groups with a namesake in the title that ended up being population-focused, four of them were founded by nonpatients in memory of a patient, one was founded by a nonpatient associated with an external organization, and one was founded by a patient survivor.

Among groups initially classified as population-focused based on a generic naming format, 90% (43/48) of cases were later confirmed to be population-focused. Five person-focused groups adopted a generic naming format without naming a specific patient in their title. In two of the cases, group descriptions included lengthy definitions of the disease conditions. It appeared that those two founders saw information dissemination as an important objective of their groups. The rationale behind the generic naming convention of the other three groups remains unclear.

Group Founders

Treatment status. In addition to group focus, treatment status also proved to be an important attribute of the Facebook groups included in the study sample. Content analysis revealed that groups could be classified as either patient founded or nonpatient founded. Interestingly, data analysis also revealed that the vast majority of groups included in the study sample were founded by nonpatients. In total, nearly 82% (27/33) of patient-focused groups and approximately 76% (37/49) of population-focused groups were founded by nonpatients.

Disease affiliation. The disease affiliation status of group founders also emerged as an important distinguishing characteristic for the groups included in the study. Disease affiliation status was evident in 30% (25/82) of group titles, 55% (45/82) of group descriptions, and for those groups with open privacy settings, in 94% (44/47) of group postings. All but one of the open privacy groups showcased disease affiliation status in either the title, description, or postings. For the one open group that did not indicate disease affiliation status of the group founder on Facebook, disease affiliation status was confirmed via another public data source associated with the group founder (an obituary).

In groups with closed privacy settings, 69% (24/35) of groups revealed the disease affiliation status of their founders in either the group title or description. Group descriptions were more likely than group titles to indicate disease affiliation status. About half (17/35) of groups with closed privacy settings disclosed disease affiliation status in the group description, whereas only 43% (15/35) of groups disclosed disease affiliation status in the group's title. For all 11 closed privacy groups with no mention of disease affiliation status in either the group title or description, the disease affiliation status of the group founder was confirmed via a secondary public data source. Secondary data sources included online obituaries, news stories, or other internet resources.

Group Formation

According to the titles and descriptions of groups included in this study, both intrinsic and extrinsic motivations of founders contribute to the formation of online support groups. For patient founders in the study sample, being a survivor was the biggest motivation for group formation (56% of cases), but being in active treatment also accounted for a significant portion of group formations (>40%). Nonpatient founders were motivated by a variety of factors to form online sarcoma support groups. The primary motivations were: being connected to someone in active treatment (39%), keeping the memory of a loved one alive (30%), and promoting an external organization (20%).

Disease affiliation status was also helpful in characterizing the motivations behind forming patient-focused and population-focused support groups. Nearly 82% of all patient-focused groups were formed by someone who was either a patient in active treatment or someone associated with a patient in active treatment. The remaining patient-focused groups were either formed "in memoriam" by a family member or friend of a deceased patient (12%), or connected to a sarcoma survivor (6%). Population-focused groups were predominantly connected to a sarcoma survivor (31%), an external group or organization (27%), or were created "in memoriam" (31%). Only 12% of population-focused groups were associated with a patient in active treatment. Presumably, the low percentage of population-focused active treatment groups is explained by the high percentage of patient-focused active treatment groups. Founders appear to prefer forming patient-focused groups more than population-focused groups for patients undergoing active treatment.

Discussion

Group Followers

A key objective of the current study was to increase understanding of the role and influence of social media on the experience of support for sarcoma patients and their families. Groups in the study sample have been differentiated based on group followers, focus, founders and formation. This is an important first step in laying the groundwork for additional research regarding potential differences in socially supportive content between groups. If future studies demonstrate that different types of groups meet different support needs, such findings will reinforce the importance of properly matching individuals to groups. Joining a group that does not offer the type of support required by an individual will be counterproductive. If a group is found to be ineffective, it is not yet known whether or not an individual will change group affiliation, or if they will merely stop participating.

The significance of any finding that differences exist in social support content between groups will hinge on whether or not individuals participate in multiple groups. Although an individual group may not fully meet a member's support needs, those needs may be addressed in totality between all of the groups in which an individual participates. It was with this question in mind that the researcher sought to determine the amount of overlap in membership between groups included in the present study. Analysis of group membership revealed that 87.8% of individuals accounted for in the study participated in only one of the groups in the sample. It is not yet known what this means, but this finding could prove important if future studies demonstrate clear separation in social support content between different types of groups.

Group Focus

Analysis revealed that groups could be classified as either patient- or population-focused. This distinction seems particularly relevant to the study of social support within online rare disease communities. Buis and Whitten (2011) have noted, "although there have been several content analysis case studies of individual online support communities, to date, cross-community comparisons of social support content…[have been] limited" (p. 462). In the future, research exploring the relationship between group focus and social support should center around three distinct areas of inquiry: First, on the relationship between tie strength and group orientation. Second, on the relationship between tie strength and social support. Third, on the relationship between group orientation and social support. It is also quite possible that these relationships are interdependent on one another. For example, group focus could determine tie strength (who subsequently joins a group), and tie strength (connection between a group founder and patient) could determine group focus.

Tie strength and group focus. Notwithstanding the fact that there have not been any prior studies looking directly at the relationship between tie strength and group focus, some preliminary conclusions can be drawn based on prior research. Granovetter (1973) published one of the most influential and highly cited sociological papers of all time (Barabási & Frangos, 2002, p. 42). His paper, titled *The Strength of Weak Ties*, highlighted the important role that weak interpersonal ties play in social circles, especially with respect to knowledge transfer and information dissemination. Granovetter saw weak ties, especially those that bridge disparate social structures, as central to sociological understanding because they serve an integrative function and provide individuals with access to ideas, influences or information that would otherwise be inaccessible (p. 1370). It is upon this very premise that population-focused support groups are organized. Presumably, rare disease patients understand that keeping only to their strong tie connections will not adequately address their needs for support and information.

Unlike population-focused groups, person-focused groups are more likely to be comprised of strong tie connections than weak ties. Strong ties are broadly conceived as connections to family or kin, close friends, or significant others. These ties are characterized by an increased motivation to communicate, elevated emotional intensity, greater intimacy, increased reciprocity, a willingness to share personal information, and greater amounts and variety in the types of supporting resources exchanged (Granovetter, 1973; Haythornthwaite, 2002). Wellman (1990) has suggested that strong ties also exhibit three characteristics:

> (1) a sense of the relationship being intimate and special, with a voluntary investment in the tie and a desire for companionship with the tie partner; (2) an interest in being together as much as possible through interactions in multiple social contexts over a long period; and (3) a sense of mutuality in the relationship, with the partner's needs known and supported (p. 564).

Wellman's definition of strong ties is helpful for understanding why rare disease patients might find it difficult to establish strong tie connections in population-focused groups. For members of population-focused support groups, large geographic distances likely make it difficult to interact in other social contexts, or to provide a sense of intimacy or companionship to other group members. On the other hand, members of person-focused support groups are more likely to interact with other network members in multiple social contexts (both online and offline), to be well acquainted with the needs and personal histories of other group members, or to have an intimate or special relationship with another group member. Given the likelihood of person-focused groups being comprised of connections to family or kin, close friends, or significant others, it is also probable that members of person-focused groups will share similar values, attitudes, and life-styles (Adelman, Parks, & Albrecht, 1987; Lazarsfeld & Merton, 1954; McPherson, Smith-Lovin, & Cook, 2001). It is also likely "their social worlds will overlap – that they will have ties with the same third parties, a kind of transitivity" (Borgatti & Lopez-Kidwell, 2011, p. 40). In his paper *The Strength of Weak Ties*, Granovetter hypothesized that "the stronger the tie between A and B [two people], the larger the proportion of individuals in S [their combined network] to whom they will both be tied, that is connected by a weak or strong tie" (1973, p. 1362). Over the years, this hypothesis has been tested in a variety of experimental settings, and there does seem to be support for strong tie transitivity (Feld, 1997; Goyal, 2005).

Tie strength and social support. The transitivity present in strong tie networks can be counterproductive for those affected by rare disease whose primary need is informational support. The challenge with social overlap is that what one person knows, everyone knows, and there is very little unique information. Unless the illness affecting the focal individual in a group has a genetic component, it is unlikely that any strong tie connections will have prior information or experience related to the focal person's condition. Although less effective at providing informational support, strong tie networks have proven more effective at fostering other types of social support. In a paper comparing both weak and strong tie strength to support, Wellman and Wortley (1990) reported, "strong ties provide broader support than weaker active ties and contribute significantly more emotional aid, minor services, and companionship" (p. 566). The term "minor services" was used to refer to tangible aid, such as taking someone to a medical appointment or offering to tend children (Wellman & Hiscott, 1985, p. 210). Wellman and Wortley's "companionship" variable corresponds with the "network support" variable referenced in many other social support studies.

Unlike strong tie networks, weak tie networks are optimized to facilitate the exchange of informational support. Because weak ties have the potential to bridge otherwise isolated social structures, and because such structures contain diverse perspectives and experience, they are more likely to be purveyors of novel information (Granovetter, 1973). While there have been many concerns expressed about patients encountering misinformation online, it has rarely been discussed that for some conditions, there may not be other options. In the absence of other sources of information, those affected by rare diseases are often forced to rely on the internet to reach sufficient numbers of people to find answers to their health-related questions.

In addition to the structural benefits of weak tie networks, there are also pragmatic benefits. Weak ties are also helpful in reducing social cues that would otherwise inhibit support exchange. Davison, Pennebaker, and Dickerson (2000) conducted a study in which they compared face-to-face and online support group participation across a variety of health conditions. They observed that patients with stigmatized illnesses (e.g., AIDS, alcoholism, breast and prostate cancer) or illnesses that required home confinement due to debilitating symptoms or a compromised immune system, were as much as 250 times more likely to engage in online support groups than patients with nonstigmatizing illnesses. Online environments benefit patients who are hesitant to participate in face-to-face support groups because they allow participants to interact with one another without worrying about discrimination or conditions that might induce social stigma. Since weak-tie networks are comprised of distant relationships and "do not typically share an intimate relational history, they may be less likely to judge or feel judged by one another" (Wright, Rains, & Banas, 2010, p. 610). High levels of engagement are encouraged as participants "are able to be judged online only by their text-based communication, freed of the binding status associations inherent in face-to-face situations" (Haythornthwaite & Nielson, 2007, p. 169).

Given the potential for weak ties to transmit informational support, it is logical to wonder if other types of social support might also be transported by the same means. Although Granovetter's strength of weak ties (SWT) theory has been extensively applied to understanding informational support, it has not yet been thoroughly applied to other types of social support (e.g., emotional, tangible, esteem, or network). The potential for weak tie networks to transmit other types of support will likely be the subject of intense debate and rigorous scientific inquiry for years to come.

Recent work by Fowler and Christakis has shown that an emotion like happiness can be transmitted through a social network like a contagion (Christakis & Fowler, 2013; Fowler & Christakis, 2008). Similarly, negative emotions, such as loneliness, have also been shown to be amenable to diffusion via network processes (Cacioppo, Fowler, & Christakis, 2009). In theory, if an individual receives emotional support, there could be an increased desire on their part to extend emotional support to someone else; also like a contagion. However, the extent to which this happens, and the mechanisms by which this might operate, have not yet been addressed in the literature.

Unlike informational support, which is easily diffused throughout a network and transmitted via weak ties, emotional support is not easily portable. When an individual in a network receives emotional support, it is difficult for them to share that exact support with another network member further down the line. To be clear, it is not impossible to transmit emotional support, but it is much more difficult than information to mobilize. How researchers approach the discussion of transmitting emotional support via weak ties, and how they formulate research hypotheses, will likely center on theoretical or pragmatic lines. Granovetter argued that emotional intensity is one of the defining features of strong ties (Granovetter, 1973, p. 1361). In an empirical study of potential indicators of tie strength, Marsden and Campbell (1984) also found emotional intensity (or closeness) to be “the best indicator of tie strength among those available to us” (p.

497). While it may be true that emotional intensity engenders emotional support, it is also unlikely to be the sole correlate of emotional support. Low emotional intensity does not preclude weak ties from also initiating productive emotional support. Central to this argument is the observation that weak ties often coalesce under conditions in which network members require diverse types of support that may not otherwise be available from other sources. There is no "big bang" in support network formation. For example, population-focused online support groups come together because there is a common interest or goal that initiated them (e.g., a rare disease diagnosis).

Group focus and social support. As mentioned earlier, the structural features of weak tie networks make them excellent conduits of novel informational exchange. This is likely one of the reasons why there are so many population-focused sarcoma support groups on Facebook. Online support groups represent an opportunity to reduce the uncertainty of diagnosis by exchanging information about treatment options and prognosis. The flow of informational support through population-focused online support groups has been supported in two prior studies of rare disease communities. Coulsen et al. (2007) found that informational support was the most frequently offered type of support within a Huntington's disease online support group (56% of cases). Similarly, a study of messages posted on a Primary Biliary Cirrhosis mailing list, revealed that biomedical information was the most frequent area of discussion (Lasker, Sogolow, & Sharim, 2005).

Although population-focused groups are optimized for information support, other types of supportive exchange are possible. Indeed, Coulson et al. (2007) found that 51.9% of posts to an online support group for Huntington's disease contained emotional support, 48.4% of posts contained network support, 21.7% contained esteem support, and 9.8% contained tangible assistance. Lasker et al. (2005) did not examine network, esteem, or tangible support, but they also observed frequent expressions of emotional support in a support group for individuals affected by rare disease.

An important next step for the scientific community will be to further explore the relationship between group focus and social support; especially in rare disease communities. Although there have been a handful of studies examining the prevalence of social support within population-focused groups, a review of the existing literature has not revealed any studies examining social support in the context of a person-focused rare disease community.

Group Founders

Prior studies of organizational culture have shown that founders often have a lasting impact on the culture and behavior of their firms (Barringer, Jones, & Neubaum, 2005). Indeed, some of the world's most powerful social movements, and many industry leading brands have been built around an affection for charismatic leaders. These observations have led this investigator to wonder what impacts founders of online support groups have on group formation and processes. Recent studies suggest that this influence is significant. In 2014, Kraut and Fiore conducted a study in which they examined the role of founders in determining the fate of 472,231 Facebook groups. According to the study, of the 100,000 new groups that are created on Facebook each day, "13% produce no content after the first day…and 57% have stopped all activity within three months of creation" (Kraut & Fiore, 2014).

In the present study, founders were also observed to play an important role. Content analyses of publicly available information showed that group founders not only initiated group formation, but they also defined a group's focus and followers via carefully constructed group titles and descriptions. Founders also used introductory posts to establish group norms and expound the group's focus. Once established, group founders and administrators controlled group dynamics by managing group membership and privacy practices. Membership and privacy policies determined who could view, read, or post content within a group.

Thoits (1986, 1995) and Cohen and McKay (1984) have argued that support is most productive when it comes from someone who is socially similar to the support recipient, and has faced similar circumstances. Likewise, social comparison theory (Festinger, 1954) suggests that support recipients turn to comparable peers because they are expected to be able to provide the most relevant information for coping with a given situation (Cohen & McKay, 1984, p. 257). Shared circumstances also promote empathetic understanding between support provider and support recipient. According to Heaney and Israel (2002), this understanding is "particularly relevant to the exchange of emotional support but also applies to instrumental and informational support" (p. 197). Given these prior assertions, it was encouraging that 56% of the patient founded groups in the present study sample were associated with a patient in active treatment, and 44% were associated with a sarcoma survivor. It is possible that these groups are affording opportunities for social comparison to those patients who need it most.

Unlike patient founded groups where survivorship accounted for nearly 56% of group formations, survivorship accounted for only 10.9% of group formations by nonpatient founders. The low number of survivor oriented groups founded by nonpatients suggests that many individuals do not see survivor support as a priority. There are several possible explanations for this finding. Studies on burnout among caregivers have shown that efforts to provide support may diminish over time as support providers tire or experience difficulty in meeting caregiving demands (Murphy, Christian, Caplin, & Young, 2007; Ybema, Kuijer, Hagedoorn, & Buunk, 2002). Culture also plays an important role in defining social support obligations and expectations. Cultural norms may dictate instances where support is withdrawn as individuals are expected or encouraged to recover on their own following a major life event. For example, in some cultures, males are expected to "be tough" or "buck up" in the face of stress, and showing emotion or distress is highly discouraged. Unfortunately, such cultural influences may obscure the level of stress that an individual may be experiencing and may inhibit help seeking behavior (Addis & Mahalik, 2003; Dunn & O'Brien, 2009; Vogel, Heimerdinger-Edwards, Hammer, & Hubbard, 2011).

Similar cultural effects have also been observed in studies of bereavement. In some cultures, individuals receive ample support immediately following a death in the family, but support diminishes rapidly as individuals are left to begin coping on their own (Kemp, 2005). Often, enacted support dissipates much too soon in relation to the needs of those in mourning (Dyregrov, Dyregrov, & Raundalen, 2008; Lehman, Ellard, & Wortman, 1986). Clearly, much more can be done to evangelize the benefits of social support for those who have recently experienced a traumatic life event. Many studies have shown social support to be predictive of better health related quality of life in cancer survivors (Ganz et al., 2002; Hipkins, Whitworth, Tarrier, & Jayson, 2004). Low support for survivors by nonpatients may also be the impetus for patients joining or forming cancer support groups on their own (Taylor, Falke, Shoptaw, & Lichtman, 1986).

Group Formation

An aim of the present study was to explore the impetus behind the formation of Facebook groups with a focus on sarcoma. According to Facebook, "…Facebook Groups are the place for small group communication and for people to share their common interests and express their opinion. Groups allow people to come together around a common cause, issue or activity to organize, express objectives, discuss issues, post photos and share related content" (Facebook, 2015).

It seems apparent that in order for a group to successfully coalesce around a common cause or issue, prospective members must understand the purpose or motivation behind a group's formation. In the present study, group founders indicated that both intrinsic and extrinsic motivations contributed to the formation of online support groups. For patient founders in the study sample, being a survivor was the biggest motivation for group formation (56% of cases), but being in active treatment also accounted for a significant portion of group formations (>40%). Nonpatient founders were motivated by a variety of factors to form online sarcoma support groups. The primary motivations were: being connected to someone in active treatment (39%), keeping the memory of a loved one alive (30%), and promoting an external organization (20%).

Group founders play an important role in initiating groups, as well as crafting group titles and descriptions that will attract the desired members. This is true of any group started on Facebook. Like a traffic signal, group titles and descriptions serve to help direct users towards groups that match their desired search criteria. For groups with closed privacy settings, unless a prospective member has been directly invited to participate or has prior knowledge of a group's formation, group titles and descriptions may contain the only visible clues to a group's purpose and function. Only 84% of group founders gave enough information in the title or description of their group for prospective members to be able to readily identify their group's purpose and mission. One of the clear messages of this study is that group founders can and should do a better job at crafting compelling descriptions for their groups. Doing so will greatly help them to better attract members to their group.

Limitations

There are several limitations of the present research. First, group selection was limited to groups containing a sarcoma diagnosis in the title, conducted primarily in the English language, and related only to human forms of sarcoma. These criteria likely resulted in the exclusion of many highly relevant sarcoma groups. For example, it is quite possible that support groups exist that reference "fighting cancer" in the title, but not sarcoma, even though the group's namesake is battling sarcoma. This may have caused the researcher to overlook important differences between groups. For instance, the generic keyword "cancer" is likely searched for more frequently on Facebook than keywords for rare forms of cancer like sarcoma. Founders employing a more generic nomenclature for their groups might experience greater visibility and success at attracting new members than those included in the current analysis. Similarly, by restricting the study to groups in the English language, important cultural differences between groups may have been overlooked.

Another limitation of the study was the inclusion of more than one type of sarcoma diagnosis in the group selection process. Table 3.4 shows the distribution of diagnostic conditions within the sample population. From the search results, it is clear that some conditions were more frequently represented in the study population than others. Disease specific factors, such as prevalence and prognosis, likely impacted not only the number of groups, but also the distribution of patient versus nonpatient founders between groups. The extent to which the distribution of diseases impacted the study findings was not examined.

Although one of the objectives of the current study was to characterize followers of sarcoma support groups, little analysis was completed beyond computing membership overlap and average membership per group. Treatment status and disease affiliation status were coded for group founders, but not for group followers. Given the sheer numbers of group members (almost 26,000) it would have been impractical to conduct a similar classification of every member of the sample groups. Nevertheless, a random sampling and classification of at least some of the group members might have yielded some additional interesting insights.

Opportunities for Future Research

Given the myriad benefits of social exchange between comparable peers, it was surprising to discover that 78% of groups in the study sample were founded by nonpatients. The impact of this phenomenon on the number of patients participating in online support groups warrants further investigation. Are support group members also predominantly nonpatients? The answer to this question will have important implications for understanding the role that online communities play in supporting those affected by rare disease. If so, this would greatly contribute to our understanding of how caregivers seek out and provide social support. The answer to this question will be an important opportunity for future research.

Although the present research helps to increase scientific understanding of the role and influence of social media on the experience of sarcoma support, it does not address differences in the types of support available between or within groups. Future studies are recommended to: 1) specifically examine the types of social support content within sarcoma support groups, and 2) examine differences in socially supportive content between groups and between different types of member within groups.

Conclusion

Facebook groups are revolutionizing the way those affected by rare disease connect with each other, and seek out and exchange health-related information and support. Facebook groups provide a mechanism for identifying disease specific groups, as well as facilitating connections between individuals with similar backgrounds or states of disease progression. While this study has identified some important defining features of sarcoma groups, it is not yet known how these features relate to the different types of social support that are possible within such groups. Additional research is recommended to more fully investigate the relationship between group followers, group focus, group founders, group formation and social support.

Table 3.1: Keyword Search Terms

Sarcoma Diagnoses		
Adenosarcoma	Ewing Sarcoma	Malignant Schwannoma
Alveolar Soft Part Sarcoma	Experimental Sarcoma	Mesodermal Mixed Tumor
Angiosarcoma	Extraskeletal Chondrosarcoma	Myeloid Sarcoma
Askin's Tumor	Fibrosarcoma	Myosarcoma
Chondrosarcoma	Gastrointestinal Stromal Tumor (GIST)	Myxosarcoma
Clear Cell Sarcoma	Hemangiopericytoma	Neurofibrosarcoma
Cystosarcoma	Hemangiosarcoma	Osteosarcoma
Cystosarcoma Phyllodes	Kaposi Sarcoma	Pleomorphic Sarcoma
Dermatofibrosarcoma	Leiomyosarcoma	Rhabdomyosarcoma
Dermatofibrosarcoma Protuberans	Liposarcoma	Sarcoma
Desmoid Tumor	Lymphangiosarcoma	Sarcoma Botryoides
Desmoplastic Small Round Cell Tumor	Malignant Fibrous Histiocytoma	Small Cell Sarcoma
Endometrial Stromal Tumor	Malignant Hemangioendothelioma	Synovial Sarcoma
Epithelioid Sarcoma	Malignant Peripheral Nerve Sheath Tumor (MPNST)	Undifferentiated Pleomorphic Sarcoma

Table 3.2: What Are the Privacy Options for Groups?

Facebook offers three privacy options for groups: Public, Closed and Secret. The table below shows who can join groups and what each privacy setting means.

	Public	**Closed**	**Secret**
Who can join?	Anyone can join or be added or invited by a member	Anyone can ask to join or be added or invited by a member	Anyone, but they have to be added or invited by a member
Who can see the group's name?	Anyone	Anyone	Current and former members
Who can see who's in the group?	Anyone	Anyone	Only current members
Who can see the group description?	Anyone	Anyone	Current and former members
Who can see the group tags?	Anyone	Anyone	Current and former members
Who can see what members post in the group?	Anyone	Only current members	Only current members
Who can find the group in search?	Anyone	Anyone	Current and former members
Who can see stories about the group on Facebook (like in News Feed and search)?	Anyone	Only current members	Only current members

Adapted from the following source: https://www.facebook.com/help/220336891328465

Table 3.3: Distribution of Groups

Groups & Followers	**Groups**	**%**	**Group Members**	**%**
Total	82	100%	25,927	100%
Avg. Size			316.18	
Group Focus				
Person	33	40.2%	5,234	20.2%
Population	49	59.8%	20,693	79.8%
Group Founders				
Treatment Status				
Patient	18	22.0%	6,202	23.9%
Nonpatient	64	78.0%	19,725	76.1%
Disease Affiliation Status				
Active Treatment	33	40.2%	3,878	15.0%
Survivor	17	20.7%	6,743	26.0%
In Memoriam	19	23.2%	6,016	23.2%
External Organization	13	15.9%	9,290	35.8%

Table 3.4: Distribution of Groups by Diagnostic Keyword

Diagnostic Keyword	**Number of Groups**	**Percent of Total**
Angiosarcoma	1	1.22%
Clear Cell Sarcoma	2	2.44%
Epithelioid Sarcoma	1	1.22%
Infantile Fibrosarcoma	1	1.22%
Leiomyosarcoma	15	18.29%
Liposarcoma	4	4.88%
Osteosarcoma	32	39.02%
Rhabdomyosarcoma	8	9.76%
Synovial Sarcoma	6	7.32%
Sarcoma (Undifferentiated)	11	13.41%
Cystosarcoma Phyllodes	1	1.22%
Total	**82**	**100%**

References

Addis, M. E., & Mahalik, J. R. (2003). Men, masculinity, and the contexts of help seeking. *American Psychologist, 58*(1), 5.

Adelman, M. B., Parks, M. R., & Albrecht, T. L. (1987). Beyond close relationships: Support in weak ties. *Communicating Social Support*, 126-147.

American Cancer Society. (2013). Cancer Facts & Figures 2013. *Estimated Number* of New Cancer Cases and Deaths by Sex, US, 2013* (pp. 64): American Cancer Society.

American Cancer Society. (2014). What is a soft tissue sarcoma? Retrieved June 20, 2014, from http://www.cancer.org/cancer/sarcoma-adultsofttissuecancer/detailedguide/sarcoma-adult-soft-tissue-cancer-soft-tissue-sarcoma?docSelected=sarcoma-adult-soft-tissue-cancer-what-is-cancer

Barabási, A. L., & Frangos, J. (2002). *Linked: The new science of networks science of networks*. New York: Basic Books.

Barringer, B. R., Jones, F. F., & Neubaum, D. O. (2005). A quantitative content analysis of the characteristics of rapid-growth firms and their founders. *Journal of Business Venturing, 20*(5), 663-687.

Borgatti, S. P., & Lopez-Kidwell, V. (2011). Network Analysis. In J. Scott & P. J. Carrington (Eds.), *The SAGE handbook of social network analysis*. Thousand Oaks: SAGE Publications.

Buis, L. R., & Whitten, P. (2011). Comparison of social support content within online communities for high-and low-survival-rate cancers. *Computers Informatics Nursing, 29*(8), 461-467.

Cacioppo, J. T., Fowler, J. H., & Christakis, N. A. (2009). Alone in the crowd: The structure and spread of loneliness in a large social network. *Journal of Personality and Social Psychology, 97*(6), 977-991. doi: 10.1037/a0016076

Christakis, N. A., & Fowler, J. H. (2013). Social contagion theory: Examining dynamic social networks and human behavior. *Statistics in Medicine, 32*(4), 556-577.

Cohen, S., & McKay, G. (1984). Social support, stress, and the buffering hypothesis: A theoretical analysis. *Handbook of Psychology and Health, 4*, 253-267.

Coulson, N. S., Buchanan, H., & Aubeeluck, A. (2007). Social support in cyberspace: A content analysis of communication within a Huntington's disease online support group. *Patient Education and Counseling, 68*(2), 173-178.

Coulson, N. S., & Knibb, R. C. (2007). Coping with food allergy: Exploring the role of the online support group. *CyberPsychology & Behavior, 10*(1), 145-148.

Darling, J. (2007). A different view of sarcoma statistics. *ESUN, 4*(6).

Davison, K. P., Pennebaker, J. W., & Dickerson, S. S. (2000). Who talks? The social psychology of illness support groups. *American Psychologist, 55*(2), 205.

Dunn, M. G., & O'Brien, K. M. (2009). Psychological health and meaning in life stress, social support, and religious coping in Latina/Latino immigrants. *Hispanic Journal of Behavioral Sciences, 31*(2), 204-227.

Dyregrov, A., Dyregrov, K., & Raundalen, M. (2008). *Effective grief and bereavement support: The role of family, friends, colleagues, schools and support professionals*. Philadelphia: Jessica Kingsley Publishers.

Facebook. (2014). *Wikipedia.* Retrieved January 29, 2014, from http://en.wikipedia.org/wiki/Facebook

Feld, S. L. (1997). Structural embeddedness and stability of interpersonal relations. *Social Networks, 19*(1), 91-95.

Festinger, L. (1954). A theory of social comparison processes. *Human Relations, 7*(2), 117-140.

Fowler, J. H., & Christakis, N. A. (2008). Dynamic spread of happiness in a large social network: Longitudinal analysis over 20 years in the Framingham Heart Study. *BMJ: British Medical Journal, 337.*

Ganz, P. A., Desmond, K. A., Leedham, B., Rowland, J. H., Meyerowitz, B. E., & Belin, T. R. (2002). Quality of life in long-term, disease-free survivors of breast cancer: A follow-up study. *Journal of the National Cancer Institute, 94*(1), 39-49.

Goyal, S. (2005). Strong and weak links. *Journal of the European Economic Association, 3*(2-3), 608-616.

Granovetter, M. (1973). The strength of weak ties. *American Journal of Sociology*, 1360-1380.

Haythornthwaite, C. (2002). Strong, weak, and latent ties and the impact of new media. *The Information Society, 18*(5), 385-401.

Haythornthwaite, C., & Nielson, A. L. (2007). Revisiting computer-mediated communication for work, community, and learning. In J. Gackenbach (Ed.), *Psychology and the Internet: Intrapersonal, interpersonal and transpersonal implications* (pp. 167-186). Burlington: Academic Press.

Heaney, C. A., & Israel, B. A. (2002). Social networks and social support. In K. Glanz, B. K. Rimer & K. Viswanath (Eds.), *Health behavior and health education: Theory, research, and practice* (Vol. 4, pp. 189-210). San Francisco: Josey-Bass.

Hicks, M. (2015). Facebook Tips: What's the Difference between a Facebook Page and Group? Retrieved March 1, 2015, from https://www.facebook.com/notes/facebook/facebook-tips-whats-the-difference-between-a-facebook-page-and-group/324706977130

Hipkins, J., Whitworth, M., Tarrier, N., & Jayson, G. (2004). Social support, anxiety and depression after chemotherapy for ovarian cancer: A prospective study. *British Journal of Health Psychology, 9*(Pt 4), 569-581. doi: 10.1348/1359107042304542

Jacobsen, W. C., & Forste, R. (2011). The wired generation: Academic and social outcomes of electronic media use among university students. *Cyberpsychology, Behavior, and Social Networking, 14*(5), 275-280.

Jolley, M. (2014). Rare Condition Health Management Solution. Retrieved June 7, 2014, from https://www.accordant.com/Apps/WebObjects/Accordant.woa/wa/HealthPlans

Kemp, C. (2005). Cultural issues in palliative care. *Seminars in Oncology Nursing, 21*(1), 44-52. doi: http://dx.doi.org/10.1053/j.soncn.2004.10.007

Kraut, R. E., & Fiore, A. T. (2014). The role of founders in building online groups. In *Proceedings of the 17th ACM Conference on Computer Supported Cooperative Work & Social Computing*. New York: ACM.

Lasker, J. N., Sogolow, E. D., & Sharim, R. R. (2005). The role of an online community for people with a rare disease: Content analysis of messages posted on a primary biliary cirrhosis mailinglist. *Journal of Medical Internet Research, 7*(1).

Lazarsfeld, P. F., & Merton, R. K. (1954). Friendship as a social process: A substantive and methodological analysis. In M. Berger (Ed.), *Freedom and control in modern society* (pp. 18-66): New York: Van Nostrand.

Lehman, D. R., Ellard, J. H., & Wortman, C. B. (1986). Social support for the bereaved: Recipients' and providers' perspectives on what is helpful. *Journal of Consulting and Clinical Psychology, 54*(4), 438-446. doi: 10.1037/0022-006x.54.4.438

Marsden, P. V., & Campbell, K. E. (1984). Measuring tie strength. *Social forces, 63*(2), 482-501.

McPherson, M., Smith-Lovin, L., & Cook, J. M. (2001). Birds of a feather: Homophily in social networks. *Annual Review of Sociology*, 415-444.

Murphy, N., Christian, B., Caplin, D., & Young, P. (2007). The health of caregivers for children with disabilities: Caregiver perspectives. *Child: Care, Health and Development, 33*(2), 180-187.

National Institutes of Health. (2014). *Estimates of Funding for Various Research, Condition, and Disease Categories (RCDC).* Retrieved June 7, 2014, from http://report.nih.gov/categorical_spending.aspx

National Institutes of Health. (2014). FAQ About Rare Diseases. Retrieved October 15, 2013, from http://www.ncats.nih.gov/about/faq/rare/rare-faq.html

O'Brien, P. C. (2013). Introduction: Social media history and use. In K. Langmia, T. Tyree, P. O'Brien & I. Sturgis (Eds.), *Social media: Pedagogy and practice* (pp. 250). Lanham: University Press of America.

Paturel, A. (2012). Too rare for research? *Neurology Now* (April/May), 29-33.

Posada de la Paz, M., & Groft, S. C. (2010). *Rare diseases epidemiology* (Vol. 686). Dordrecht: Springer.

Raacke, J., & Bonds-Raacke, J. (2008). MySpace and Facebook: Applying the uses and gratifications theory to exploring friend-networking sites. *CyberPsychology & Behavior, 11*(2), 169-174.

Rainie, L. (2013). Social Networking Fact Sheet. Retrieved May 10, 2014, from http://www.pewinternet.org/fact-sheets/social-networking-fact-sheet/

Rare Diseases Act of 2002, Pub. L. No. 107-280 [H.R. 4013] (2002).

Taylor, S. E., Falke, R. L., Shoptaw, S. J., & Lichtman, R. R. (1986). Social support, support groups, and the cancer patient. *J Consult Clin Psychol, 54*(5), 608-615.

Thoits, P. A. (1986). Social support as coping assistance. *Journal of Consulting and Clinical Psychology, 54*(4), 416.

Thoits, P. A. (1995). Stress, coping, and social support processes: Where are we? What next? *Journal of Health and Social Behavior*, 53-79.

U.S. Census Bureau. (2014). State & County Quick Facts. Retrieved June 7, 2014, from http://quickfacts.census.gov/qfd/states/00000.html

Vogel, D. L., Heimerdinger-Edwards, S. R., Hammer, J. H., & Hubbard, A. (2011). “Boys don't cry”: Examination of the links between endorsement of masculine norms, self-stigma, and help-seeking attitudes for men from diverse backgrounds. *Journal of Counseling Psychology, 58*(3), 368.

Wellman, B., & Hiscott, R. (1985). From social support to social network. In I. G. Sarason & B. R. Sarason (Eds.), *Social support: Theory, research and applications* (pp. 205-222). Hingham: Kluwer Boston, Inc.

Wellman, B., & Wortley, S. (1990). Different strokes from different folks: Community ties and social support. *American Journal of Sociology, 96*(3), 558.

Wright, K. B., Rains, S., & Banas, J. (2010). Weak-tie support network preference and perceived life stress among participants in health-related, computer-mediated support groups. *Journal of Computer-Mediated Communication, 15*(4), 606-624.

Ybema, J. F., Kuijer, R. G., Hagedoorn, M., & Buunk, B. P. (2002). Caregiver burnout among intimate partners of patients with a severe illness: An equity perspective. *Personal Relationships, 9*(1), 73-88.

CHAPTER 4

A QUALITATIVE CONTENT ANALYSIS OF SOCIAL SUPPORT MESSAGES EXCHANGED BY OSTEOSARCOMA PATIENTS IN ACTIVE TREATMENT ON FACEBOOK

Abstract

Facebook groups are becoming an important medium of social exchange for rare disease patients. On Facebook, patients meet others with whom they have an opportunity to regularly interact and cultivate socially supportive relationships. Although the scientific literature is replete with research in the area of social support, few studies have examined the role of Facebook groups on the provision of social support. Even fewer have examined social support in the context of rare disease. The aim of the present study was to characterize, via a directed approach to qualitative content analysis, the social support experiences of patients currently affected by osteosarcoma, a rare and aggressive form of cancer. This research extends prior social support theory to the study of social support within an online support group for osteosarcoma patients. Evidence of seven distinct types of support were found within the study group: appraisal, emotional, informational, spiritual, esteem, network and tangible. All of these support types are found elsewhere in the literature. The results of this study suggest that appraisal and

spiritual support may play a bigger role in online support communities than had previously been indicated.

Introduction

Facebook and Social Support

By most estimates, Facebook is the largest and most successful social networking platform in the world. As of January 2015, Facebook had over 1.39 billion registered users worldwide (Facebook Inc., 2015). This represents the single largest congregation of individuals anywhere in the world. To put this in perspective, if Facebook were a country, it would be more populous than the largest country on earth: China (1.37 Billion as of April 2015). Never before has there been such an opportune time to undertake research in the area of social media enabled social support.

The growth of Facebook, and worldwide adoption of social media has opened the door to new areas of research, especially around social media and social support systems. For example, social media technologies are enabling patients to connect with their healthcare providers and others in ways that were previously unimaginable (Bacigalupe, 2011; Hawn, 2009). As a result, not only do patients have access to larger social support networks, a significant health benefit, but they are also able to use social media for effective inbound and outbound information dissemination activities. Importantly, these technologies also allow researchers to directly measure and quantify the contribution of social media enabled social support towards the achievement of positive health outcomes in individuals and communities.

While Facebook has greatly simplified the process for identifying and connecting to disease specific groups, some disease communities have benefitted more than others. Social media and social networking sites have been particularly helpful for communities where there are disincentives to participate in face-to-face support groups (e.g., due to stigmatizing illnesses) or where there are otherwise few opportunities to connect in person. For example, Davison, Pennebaker, and Dickerson (2000) found that patients with stigmatizing illnesses (e.g., AIDS, alcoholism, etc.) were as much as 250 times more likely to engage in online support groups than their peers with non-stigmatizing illnesses.

Rare disease communities are also increasingly using social media to connect members to other patients, providers, and treatment resources. According to the National Institutes of Health (NIH), a rare disease is a condition that affects fewer than 200,000 people in the United States at any given time. While specific rare diseases affect a relatively small number of people, there are more than 6,800 rare diseases that collectively affect more than 25 million Americans, and that has made funding rare disease research a strategic priority for both the NIH and the Food and Drug Administration (Dunoyer, 2011; Hampton, 2006; Montoya, 2011; Rados, 2003; Seoane-Vazquez, Rodriguez-Monguio, Szeinbach, & Visaria, 2008). The rarity of these conditions makes it difficult for individuals to receive an accurate diagnosis, to locate healthcare providers knowledgeable about a given condition, and to connect with other patients and families with the same health condition. For such patients, online support groups may be the only way to organize, and meet others with similar health-related challenges (Rimer et al., 2005; White & Dorman, 2001).

While in person patient support groups have existed for many years for some of the most prevalent health conditions, patients with rare diseases have lacked the benefit of such interactions. The rarity and geographic dispersion of rare disease patients has made it difficult for such patients to similarly organize. With the advent of the internet, online support groups have proliferated and, as a result, many more patients and disease conditions (including rare diseases) are being served by virtual social support communities. Thanks to services like Facebook, rare disease patients and their loved ones can now obtain “practical information and reassurance that they need not face illness or disability alone” (Lamberg, 1997).

The large number of registered users on Facebook, over 1.39 billion worldwide (Facebook Inc., 2015), and ability for users to form groups around specific topics or disease conditions, has been particularly helpful for those affected by rare diseases. In 2010, there were more than 620 million groups of different types on Facebook (O'Neill, 2014). A more recent estimate is not available due to changes in Facebook’s privacy policies, but a current estimate would likely be much higher. Some estimates suggest that as many as 100,000 new groups are created each day (Kraut & Fiore, 2014). A small number of these groups are dedicated to rare disease. Although not all rare disease conditions have readily identifiable support groups, many do, and a growing number of patients have multiple support groups to choose from.

The growing number of Facebook groups for rare disease patients raises questions like, “Who is using Facebook groups, and what types of support are being exchanged on these groups?” Prior research has suggested that fundamental differences exist between users of online support groups, based on treatment and disease affiliation status. However, it is not yet known how social support differs between these groups. The researcher has undertaken this study as an initial step towards answering this question.

One of the aims of the current study was to describe the support needs of patients currently affected by osteosarcoma, a rare form of cancer, and the role that Facebook communities play in facilitating supportive interactions by these individuals. The study has also responded to the following two questions: 1) What does the exchange of social support look like in an online support group dedicated to osteosarcoma?, and 2) How do patients in active treatment in an online support group leverage the platform to meet their social support goals? These are important questions since "studies that examine the subjective experience of persons receiving and giving support are more likely to yield relevant information for understanding person-environment fit and for suggesting interventions" (Ell, 1984).

Osteosarcoma

Osteosarcoma is an extremely rare form of cancer that affects fewer than 800 new patients each year (American Cancer Society, 2015). To put this in perspective, of the approximately 1.6 million cases of cancer in the United States each year, fewer than .0005% of cancer cases are diagnosed as osteosarcoma (American Cancer Society, 2013; Darling, 2007). The majority of osteosarcoma cases occur in children and adolescents, but a handful of studies have also found an elevated incidence of osteosarcoma among the elderly (Dorfman & Czerniak, 1995; Geller & Gorlick, 2010; Mirabello, Troisi, & Savage, 2009; Savage & Mirabello, 2011). The estimated incidence of osteosarcoma is 2.4 cases/million/year in children, 7.6-8.2 cases/million/year in adolescents, and 1.5-4.5 cases/million/year in persons over 60 years of age (Kager et al., 2010; Savage & Mirabello, 2011). Although there are a variety of different types of primary bone malignancy, all of them rare, osteosarcoma is the most common. Other less common malignancies of the bone include chrondrosarcoma, Ewing's sarcoma, chordoma and malignant fibrous histocytoma.

The prognosis for osteosarcoma patients is affected by a variety of factors including: "primary tumor site, tumor size, presence of clinically detectable metastatic disease, surgical resectability and necrosis following induction or neoadjuvant chemotherapy" (National Cancer Institute, 2015). Amputation is generally only required in 10-20% of cases, but skilled surgical resection is necessary in all cases to ensure a successful treatment outcome (Picci, 2007). With effective diagnosis and treatment, the overall relative 5-year survival rate for osteosarcoma is estimated at between 41-55% (Damron, Ward, & Stewart, 2007; Dorfman & Czerniak, 1995). Age specific 5-year survival rates are "60% for those younger than 30 years, 50% for those aged 30 to 49 years, and 30% for those aged 50 years or older" (Damron et al., 2007). Survival rates have increased dramatically over the past 24 years, and the increases have no doubt been the result of the development of effective multimodal therapies to combat high grade tumors (National Cancer Institute, 2015; Picci, 2007).

Given the complexities of osteosarcoma treatment and recovery, it is natural for patients to want to connect with as many supportive resources as possible. The rarity of osteosarcoma cases means that without organized support, it can be difficult for individuals to locate others affected by the disease. Faced with an uncertain diagnosis and future, many patients are turning to Facebook as their medium of choice for supportive exchange. To date, few studies have been conducted to understand the nature and types of social support exchanged within these groups.

Study Aim

The aim of the present study was to examine the social support experiences of patients currently affected by osteosarcoma, a rare and aggressive form of cancer, and the role that Facebook groups play in facilitating supportive interactions. Until now, research in this area has been very limited, and the scientific community would benefit greatly from further description. The current research extends prior social support theory to the study of social support within an online support group for osteosarcoma patients.

Methods

The current study employed a directed approach to qualitative content analysis. According to Hsieh and Shannon (2005), directed content analysis is recommended when: 1) existing theory or prior research exists about a phenomenon that is incomplete or would benefit from further description, and 2) the researcher seeks to validate or extend conceptually a theoretical framework or theory (p. 1281).

Participants

Background. In June of 2014, an examination was conducted to better understand the organization and membership of sarcoma support groups on Facebook. This was part of a larger investigation into the role and influence of social media on the social support experiences of sarcoma patients and their families. Facebook’s built-in search engine and a predetermined list of sarcoma diagnoses were used to locate relevant groups for study. Groups were identified by typing a diagnosis into the Facebook search engine and then selecting “Find all groups named [diagnosis]” when prompted. In order to be selected, groups were required to contain a sarcoma diagnosis in the title, to be conducted primarily in the English language, and to be related only to human forms of sarcoma. This search procedure resulted in the successful identification of 82 sarcoma-related support groups. Membership size for the selected groups ranged from 2 to 4,841 members and the average membership per group was 317 members. Total cumulative membership for the selected groups was nearly 26,000 members. The top five categories of sarcoma represented by the selected groups were: osteosarcoma (39%), leiomyosarcoma (18%), undifferentiated sarcoma (13%), rhabdomyosarcoma (10%) and synovial sarcoma (7%).

Using information extracted from group titles and descriptions, three group classification schemes were developed: group orientation (patient vs. population), founder treatment status (patient vs. nonpatient), and founder disease affiliation status (active treatment, survivor, in memoriam, external organization). These coding schemes were used to classify all 82 of the sampled groups and their founders. Group classification revealed 40.2% of groups to be person-focused and 59.8% of groups to be population-focused. Nonpatient founders accounted for the greatest number of groups: 81.8% of person-focused groups and 75.5% of population-focused groups. The vast majority of person-focused groups (81.8%) had a founder associated with a patient in active treatment, while only 12.2% of population-focused groups had that affiliation. Population-focused group founders were most likely to be affiliated with a cancer survivor (30.6%), a deceased patient (30.6%), or an external organization (26.5%). According to group founders, both intrinsic and extrinsic motivations contributed to the formation of online support groups. For patient founders in the study sample, being a survivor was the biggest motivation for group formation (56% of cases), but being in active treatment also accounted for a significant portion of group formations (>40%). Nonpatient founders were motivated by a variety of factors to form online sarcoma support groups. The primary motivations were: being connected to someone in active treatment (39%), keeping the memory of a loved one alive (30%), and promoting an external organization (20%).

Group selection criteria. In the present study, one of the publicly available osteosarcoma support groups from the prior study was selected for additional examination. The selected group was chosen based on its privacy settings (public group), the total number of messages posted within the group (robust, yet reasonable for the current analysis) and its focus on osteosarcoma, the most frequently encountered form of sarcoma identified in the prior groups. The target group was founded in 2008 and has 627 members. Between September of 2008 and March of 2015, the group had nearly 4,500 messages exchanged between group members, 935 discussion posts and 3,521 replies. These numbers do not include nonnarrative responses, such as Facebook "likes."

Data extraction. Once a group had been selected for study, the next step was to extract the message content for analysis. Since the group was an "open" group, the researcher was able to extract the content for analysis without joining the group. The researcher utilized functions built into a web browser to capture and create a local archival copy of the group. Data mining techniques were then utilized to extract site content to a spreadsheet file that was utilized, both for the initial data classification processes, and for uploading into a qualitative research software package that was utilized to facilitate content analysis and the reporting of results.

Sampling method. In this investigation, a purposeful homogeneous sampling method was used to identify and examine the messages posted by patients in active treatment, for evidence of socially supportive exchange. The goal of the chosen sampling method was not to generalize to a population, but to select "information-rich cases" that would allow the researcher to maximize understanding of the phenomenon of interest; online social support behaviors by osteosarcoma patients in active treatment.

According to Patton, "the logic and power of purposeful sampling lies in selecting information-rich cases for study in depth" (Patton, 1990, p. 169). In order to be able to identify and select relevant entries for study, the researcher first undertook efforts to classify all 4,451 messages in the group based on the treatment status (patient vs. nonpatient) and disease affiliation status of message authors. Independently classifying all 4,451 messages was important since the disease affiliation status of entry authors changes over time. For example, an osteosarcoma patient may move from active treatment to survivor status to relapse to death all within a very short period of time. If an individual had ever been an osteosarcoma patient, they were classified as a patient, regardless of their current disease affiliation status. Definitions of disease affiliation status were developed in an earlier study. The same definitions were utilized to classify message authors in the current study:

- Active Treatment: An individual has either had osteosarcoma themselves, or is connected to someone who is undergoing active treatment for osteosarcoma.
- Survivor: An individual has either survived osteosarcoma themselves, or is connected to someone who has survived osteosarcoma. To be classified as a

survivor, there must be references to being cancer free, in remission, having clear scans, or that there is no evidence of disease (NED).

- In Memoriam: An individual is connected to someone who has passed away from osteosarcoma and this was the motivation for starting the group.
- External Organization: An individual is posting on behalf of an organization that is somehow affiliated with osteosarcoma cancer (lobbying, fundraising, research, etc.).

Classification began by sorting messages in sequential order (newest to oldest) based on author name. After sorting, each message was independently coded by the researcher and a collaborator based on the treatment status and disease affiliation status of the author. Once classification was completed, reliability was calculated based on treatment and disease affiliation status. The analysis revealed "almost perfect reliability" between coders (Cohen, 1960). Cohen's kappa ranged from .81 for disease affiliation status to .95 for treatment status. Following the initial classification exercise, the researcher and collaborator met again to review all messages where differences in classification existed and come to perfect agreement on all entries where coding differed. The result was a highly differentiated dataset based on author classification with which to begin analyzing social support content. Table 4.1 contains the recorded frequencies for each classification of author.

Content Analysis

Unit of analysis. The unit of analysis consisted of a single message (post or reply) contained within the target group, and determined by the researcher and collaborator to be directly posted by an osteosarcoma patient in active treatment. There were a total of 644 messages that met this criteria. These messages were initiated by 32 unique individuals, and each of these users contributed an average of 20 messages while in active treatment status over the life of the group. Separate posts, whether initial or subsequent to a thread of discussion, were readily distinguishable by the date of post, and picture and name of the posting member.

Coding frame. The aim of the present study was to develop an understanding of social support needs and behaviors from the perspective of an osteosarcoma patient in active treatment. Cutrona and Russell (1990) note that theorists have proposed a variety of models to explain the functional components of social support. In 1990, they provided a brief comparison of five component models of social support (Carolyn E. Cutrona & Russell, 1990). In the present study, this earlier work was extended by comparing social support components from sixteen prominent research articles. Table 4.2 contains a comparison of the social support components from these articles. The initial coding frame for the current study was developed based on the synthesis of these articles. Social support categories were determined by analyzing prior models and identifying overlapping concepts. The following six concepts were determined to be most prevalent; informational support, emotional support, esteem support, network support, tangible support, and appraisal support. For the purposes of this study the following definitions were adopted for each of these concepts:

Cutrona and Russell (1990, p. 322)

- Emotional Support: "…the ability to turn to others for comfort and security during times of stress, leading the person to feel that he or she is cared for by others."

- Informational Support: "…providing the individual with advice or guidance concerning possible solutions to a problem."
- Tangible [Support]: "...concrete instrumental assistance, in which a person in a stressful situation is given the necessary resources (e.g., financial assistance, physical help with tasks) to cope with the stressful event."
- Esteem Support: "…the bolstering of a person's sense of competence or self-esteem by other people."
- Network Support: "a person's feeling part of a group whose members have common interests and concerns."

  House (1981, p. 25)
- Appraisal Support: "Like informational support, involves only transmissions of information, rather than the affect involved in emotional support or the aid involved in instrumental support. However, the information involved in appraisal support is relevant to self-evaluation… (a.k.a.) social comparison."

Main analysis. Once the initial coding frame had been finalized for the study, the researcher began by reading some of the messages posted by patients in active treatment. The first thing the researcher noticed was that messages appeared to be of two different types; giving support and seeking support. This was an important observation for two reasons: First, House (1981) noted that one of the key considerations in defining social support is understanding "who gives what to whom regarding which problems" (p. 22). He also argued that social support components are best understood as a matrix that incorporates both the functional definitions of support and the directionality of support between individuals. Second, work by Weiss (1974) and Cobb (1976) both suggest that social networks not only provide individuals with an opportunity to seek support, but that they also provide opportunities for individuals to provide nurturance or mothering (support) to others. These studies suggest that individuals benefit themselves by also being of help to others. Based on this information, the researcher decided to modify the initial coding frame to encompass both giving and seeking behaviors for all six of the top social support dimensions identified in the scientific literature. This modification resulted in 12 social support codes. The researcher also added an "unclassified content" category for tagging any apparent social support content which would not fit into any of the predetermined categories.

The next step was to read all 644 messages and classify the content within messages. Messages were analyzed to determine: 1) if the message contained evidence of socially supportive exchange, and 2) the appropriate coding classification for content determined to be supportive. If a post contained language that was relevant to multiple categories of support, it was considered acceptable for the content within a post to be coded separately into each of the relevant categories. Any text that appeared supportive but did not meet the definition of an existing category was coded as "unclassified" and set aside for subsequent analysis. Once all of the data in the study sample had been coded, the researcher then took steps to verify the accuracy of the coding process. The researcher reread all of the coded content by category to confirm that content had been coded in accordance with the pre-established category definitions. If any content had been misclassified, it was recoded into the appropriate category.

Once the initial coding process had been completed, the researcher next sought to identify subthemes or subcategories within each of the main categories of social support. This was an inductive process that involved reading all of the posts within a category and looking for commonalities between posts. As subthemes were identified, giving and seeking codes for each of these themes were created within each of the main categories. In total, 18 unique subthemes were identified and 36 subcategories were created (one of each for giving and seeking).

Once the researcher felt confident that all of the potential subthemes had been identified, the content within each category was analyzed and separated into the new subcategories. Fortunately, there were not any data that could not be reclassified from categories into subcategories. This was a strong indicator for the researcher that all of the relevant subthemes within categories had been successfully identified. Once all of the data had been coded into subcategories, the researcher once again reread all of the coded data to confirm the accuracy of the coding process. It is important to note that there were some subcategories that had data in either giving or seeking, but not both. It is not known whether this finding is significant or not. Future studies should examine whether such data exists within the same group for other classifications of users (other than patients in active treatment) or if such data exists in other larger osteosarcoma groups.

Finally, data within the unclassified content bucket were coded into themes. Three themes were identified in the data that were not encompassed under any of the other social support themes: prayer, well-wishing, and helping thoughts. An additional functional category of social support was added to encompass these three themes: spiritual support.

Table 4.3 contains a listing of all of the identified categories and subcategories utilized in the study. The total number of codes created for the study was 56. This total includes giving and seeking codes for each of the identified categories and subcategories.

This study was determined by the University of Utah Institutional Review Board (IRB) to be exempt from requiring review because all information utilized in the study was available in the public domain.

Results

Table 4.4 contains descriptive statistics for the 14 categories and 42 subcategories of social support within the data set. For osteosarcoma patients in active treatment, nearly 80% of socially supportive messages involved giving support to others rather than seeking support for self. Approximately 85% of support giving messages involved only four types of social support: emotional (28%), appraisal (23%), informational (17%) and spiritual (17%). Posts having to do with seeking social support were predominantly focused on appraisal support (33%), informational support (30%) and tangible support (10%). Spiritual support also accounted for a little more than 8% of messages where the author was seeking support. **Note:** Contained within this article are many direct quotes from the study group. Some of these quotes contain erroneous spellings or grammatical mistakes. All materials have been transcribed exactly as they were found in the source text.

Emotional Support

Emotional support was the second most common form of socially supportive message, accounting for nearly 24% of social support messages. Emotional support was determined to be comprised of five main categories: cheering (84.4% giving/15.6% seeking), empathy (97.4% giving/2.6% seeking), love and affection (97.3% giving/2.7% seeking), condolences or sympathy (100%giving), and expressing concern (100% giving). Patients were nearly 14 times more likely to give emotional support to another than to seek emotional support for themselves.

Cheering. Messages coded as giving cheering were generally congratulatory in nature and expressed shared excitement for desirable test results or good news. Some examples of these types of postings include the following:

"So good to hear the positive results!!"

"…that is fantastic news to hear! Good for you guys"

On the other hand, messages coded as seeking or encouraging cheering were frequently expressed as statements about progress towards a personal goal or treatment milestone. Many of these posts culminated in an exclamation point which seemed to not only express the strong emotions of the poster, but also to invite others to engage in cheering behavior. For example, "I had my post-op scan Christmas Eve and it came back 100% clear...no more cancer!!!" generated the following response from another group member: "I am also elated that you have had such success!!" Similarly, "We received good news! The bone scan was clear!" resulted in the following response: "THANKS BE TO GOD!!!"

Empathy. Messages where empathy was given generally manifested the ability of the poster to understand what another group member was feeling. For example, one poster expressed guilt at not being as available as they would like for their children during chemotherapy. In response, another user posted the following message:

> "That's the feeling. The guilt of not being there. They seem bigger every time I come home. I'm missing 17 days in the hospital for every 35 with the intense schedule my doc has me on."

Empathy seeking behavior was expressed as asking if anyone else knew how an individual felt about a situation or stressor. For example, one user posted the following:

> "first chemo after surgery, awful! just 3 more times and the chemo part is over...i think the hardest part will be finding my new normal. anyone else feel that way?"

Love and affection. This subcategory was used to classify messages containing expressions of caring between group members. Examples of group members giving love and affection to another include the following:

> "I love you too honey."
>
> "Hugs!!"
>
> "Sending love to (name withheld) and your family!"

The support group also provided a medium for members to seek love and affection from others. For example, "You can send hugs and well wishes on here too."

Condolences or sympathy. Messages coded as offering condolences or sympathy contained expressions of sorrow or regret for something that either had happened, or was currently happening to another group member. For example, members giving condolences or sympathy to other members expressed the following:

> "im ao sorry to hear she relapsed"
>
> "I'm so sorry...all I can say is cancer sucks!"
>
> "I am very sorry to hear of your child's diagnosis. I do not wish this on anyone at

all, ever."

There were not any messages posted by patients in active treatment that sought condolences or expressions of sympathy from other members.

Expressions of concern. Messages in which members gave expressions of concern most often contained an inquiry from one individual about the state of being or health of another individual. Some of the examples of members expressing concern or interest in the health of another include the following:

"How are you feeling today?"

"How are you feeling over all?"

Although there were several instances of patients expressing concern for other group members, no examples of patients actively seeking expressions of concern from other members were found.

Informational Support

Informational support was subcategorized into three areas: referral (92% giving/8% seeking), suggestion or instruction (73.1% giving/26.9% seeking), and awareness (53.6% giving/46.4% seeking). Messages in these categories accounted for nearly 20% of all social support messages posted to the sample group. Study participants were also 2.2 times as likely to give informational support as to seek informational support.

Referral. Messages coded as a referral contained recommendations for individuals to seek input from an outside expert, or source of information that would enlighten patients regarding their health status. The purpose of the referral was to encourage the support recipient to consult with, or take advantage of, the recommended resource prior to taking additional action. Unlike network referrals, the primary purpose of these messages was to connect recipients to information, rather than to expand their support network. Examples of referral giving messages include the following:

> “I'd def call yours and ask what sort of follow ups you need to have. Heck call your surgeon, or ask the doc who runs rounds in the nursing home his opinion”.
>
> “Get with your oncologist to see if this is normal and see what their recommendations are.”
>
> “The livestrong foundation will send you one for free, it has alot of great tools in it.”

Referral seeking behavior was also identified. In one instance, an individual looking for a research group posted the following inquiry:

> “(name withheld) is this one of the biggest foundations for osteosarcoma research? I am trying to get more involved and want to help out and give back to osteosarcoma research but don't know where to start”

Suggestion or instruction. Messages coded as a suggestion or instruction contained information or recommendations about health treatments or diets, or suggested steps for relieving symptoms or discomfort related to osteosarcoma care. Such messages were posted by their authors for the purpose of helping either the support giver or the seeker to contemplate a course of action related to disease management. Suggestion or instruction giving messages were initiated either in response to a request for suggestions (direct solicitation) or were unsolicited. For example, one patient posted the following information in response to a question about diet, exercise and cancer treatment:

> “Just be careful what you eat. I don't know if I can give any other advice for stopping the hunger besides drinking a lot of water all day to help curb the feeling of being hungry. Get with your oncologist to see if this is normal and see what their recommendations are.”

Another patient posted the following suggestion with respect to treating mucositis, a painful inflammation of the mucous membranes lining the digestive track.

> "You'll need soft bristle toothbrushes for him and NO mouthwash with alcohol in it. Biotene brand is great! It will help with dry mouth too."

Suggestion seeking messages were very similar to suggestion giving messages. The primary difference between giving and seeking messages was the directionality of the information request (seeker to giver). Two of the suggestion- or instruction-seeking messages include the following:

> "Has anyone every tried anything other than Chemo that seemed pretty sufficient? Just curiosity Chemo is kicking my butt"

> "I'm on coumadin, so my diet is restricted from green veggies etc. which bums me out because juicing and natural cleansing was what I was planning on. If you know of a way around all that awesome!"

<u>Awareness.</u> Content coded as awareness focused contained information designed to increase the general level of consciousness or mindfulness of a message recipient regarding an idea or issue experienced by other group members. Such messages did not contain an explicit call to action, but helped to increase the health literacy of message recipients with respect to osteosarcoma treatment, services and considerations. Some examples of awareness giving messages include the following:

> "FYI. An email hoax about cancer is going around…"

> "The burning was from the steroid they give you, decadron probably"

> "The drugs can also change taste buds and give a metallic taste in your mouth."

Awareness seeking messages were similar to awareness giving messages. However, the content of awareness seeking messages was generally formatted as a question that implied an interest on the part of the information seeker in understanding or knowing more about an idea or issue experienced by other group members. For example, posts included the following questions:

> "Has anyone used the chemo pill Pazopanib? What can you tell me about your experience?"

> "Anyone heard of trying a spirit healer? My kid actually brought one home today, lol. Sounds different to me, but I won't say no to anything...."

Tangible Support

Tangible support accounted for only 3.8% of all social support messages posted by osteosarcoma patients in active treatment within the study group. These patients were also only slightly more likely to seek tangible support (55% of cases) than to provide tangible support to others. Tangible support was subcategorized into two areas: financial (46.7% giving/53.3% seeking) and time or activity (42.9% giving/57.1% seeking).

Financial. Messages coded as financial support contained requests for, or offers of, material assistance. The following posts exemplify giving financial support:

> "I have alot of this tube feeding left over from my surgery.. if you or anyone you know in need of this please contact me…Its for anyone who has an Ng tube or G tube ...the feedings for it…They are expensive and I dont want to just throw them away"

> "BTW if you decide to make more bracelets I will purchase one:)"

The target group also contained instances of patients seeking financial support. Here are two examples:

> "Click here to support Living expenses"

> "I dont know if this is tacky or not, but my family started a fundraiser online for me. I think its fantastic, I am just not sure how to 'advertise' it? I am posting it here, but no pressure."

Time or activity. Messages coded as time or activity support did not contain requests or offers of direct financial support, but rather contained requests for, or offers of, help with specific tasks or actions that would benefit another. In the following giving example, a patient offers to share a cancer booklet they have written with others affected by cancer:

> "Hi I just wanted to share my new cancer booklet I compiled for anyone affected by cancer…"

Patients also asked others to give time, or engage in activities related to osteosarcoma. Here is an example:

> "Please can people endorse my blog by going on my blog and going on link for the @wegohealth award to give me a chance of winning for blogging about Osteosarcoma so more people are award as its so rare!"

Appraisal Support

Appraisal support was the most frequent form of social support content observed in messages posted by patients in active treatment. Approximately 25% of all messages coded contained expressions of appraisal support. Appraisal support content was sub-categorized into two areas: validation (82.2% giving/ 17.8% seeking) and social comparison (70.3% giving/29.7% seeking). Patients in the study sample were 2.7 times more likely to offer appraisal support than to seek appraisal support from others.

Validation. Validation support was most often expressed as agreement with another person's perspective or point of view on a stressful situation. For example, group members often validated one another's thoughts or concerns about coping with bone cancer. Examples of validation giving messages include the following:

> "I agree with you…This is awful."

> "no i don't think it's strange to wait until you get your reports back to celebrate! i

> think that's the right thing to do...you went through chemo hell for a much longer period of time, what's a couple more weeks to wait and be absolutely certain that it's gone...then you won't have any worries in the back of your mind like if you were to start celebrating now"

Patients also utilized the group to seek validation from other members. Some examples include the following statements:

> "I'm actually worried I won't receive chemotherapy. From what I understand it is usually given both as a treatment for existing (detected) cells, as well as undetectable (precaution) if the doctor just says no chemo, and I have undetected cells / metesases... That worries me the most. Am I just being crazy?"

> "Am I being strange by refusing to celebrate until I hear pathology reports and finish chemo."

Social comparison. Patients often provided or requested detailed information about one another's disease symptoms, progression or prognosis. The primary motivation for these requests seemed to be that of social comparison. Group participants regularly compared themselves to others, or held themselves out as a point of comparison to others facing similar stages of disease progression or treatment options. Many such messages were posted in response to an individual expressing uncertainty about their treatment or disease outcomes. Such reciprocal exchanges seemed to serve two purposes: 1) to reassure support recipients by providing a basis for comparing oneself to others in similar situations, and 2) to satisfy an innate drive on the part of group members to provide nurturance and to support others in similar circumstances. The following example of social comparison giving was posted to the osteosarcoma support group by the mom of a patient in active treatment:

> "We just found out my step son has 3 tumors on his right lung and they don't think there is no more options for him because everything they have done for his osteosarcoma has made a tumor pop up he lost his leg n half of his left lung from it...we aren't sure how to handle this can someone help us please...thank you"

In this post, a mother asks the group for help in understanding “how to handle” recent diagnostic news for her step-son. It is important to note that she asks for help, but does not directly ask for social comparison. In response to her request, a patient in active treatment not only suggests ways of handling the cancer diagnosis, but also self-discloses her own treatment status as a way of modeling beneficial coping behavior. Here is the posted response to the mother’s request for help:

> “Have you talked to different Dr's or went to different hospitals? Don't give up...don't ever give up until that's absolutely the last option. I'm battling this disease as well and I refuse to let death be my answer. Sometimes I feel really down and i hopeless but I always pick myself up and i keep fighting!!”

In another example, a grandfather shares, “My 4 year old grandson was just diagnosed with Osteo Sarcoma of the right upper arm. He began Chemo yesterday. So far there has been no mets to his lungs and we are hoping that because we caught this early he has a good chance. Your thoughts and suggestions are welcome.”

Once again, the response comes from a patient in active treatment who attempts to provide comfort by emphasizing that someone else is also going through the same thing. She responds, “I also have Osteosarcoma of my upper right arm for 17 months now..I've had 7 rounds of inpatient and outpatient chemo..6 weeks of radiation..2 surgeries and one coming up in January!”

Although the responder does outline the steps they have taken as part of their treatment, the response seems intended more as a “me too” message, rather than as providing insight into treatment options.

In addition to social comparison giving messages, there were also social comparison seeking messages in the target group. In this example, a patient author posts a question related to a prior resection surgery, and wonders if anyone else has experienced similar circumstances. She states,

> "For some reason in my initial resection surgery they left my patella in, and I'm wondering how many know of Dr.'s doing this or just replacing everything in the joint. I ask because I apparently have pain because my patella is rubbing against the metal...as well as I fractured it a little. Sighs. Still waiting to see what the Dr. says. They found the fracture with a bone scan....said they think it looks like a fracture...only i didn't hit my knee,...so maybe its something else. Something worse."

A patient in inactive treatment responds with the following personal information for comparison:

> "Mine was left in last year when I had my total knee and partial femur/tibia replacement. I was not able to start physical therapy for about six months. Because my muscles had so weakened, my patella shifted during PT to the side of my joint implant and severely wore down. I had surgery to have the patella removed in April. He did not replace it, so I no longer have one."

In the final and most explicit example of all, a patient in active treatment asks overtly for others to volunteer themselves as persons of reference to whom comparisons can be made:

> "Hey everyone I was recently diagnosed with osteosarcoma I was wondering if anyone would like to talk about what's going on and vent and compare whats going on with us...I have osteosarcoma in my left femur with metastatic lung noduals...message me if you want to chat I'd really love to meet people going through this same thing!!!"

Network Support

In the present study, network support messages accounted for only 6.2% of all messages coded. However, for the messages that were coded, participants were 2.4 times more likely to give than to receive network support. The following three subthemes of network support were identified: connection and friendship (62.5% giving/37.5%

seeking), unity (100% giving), and network referral (71.4% giving/28.6% seeking).

Connection and friendship. Content coded as being in the connection and friendship category contained language that either reinforced an existing connection between group members, or contained an invitation to establish a new supportive relationship. Examples of network support giving include the following:

"you ever wanna talk I'm all ears!"

"Feel free to contact me or friend me on here and ask questions anytime."

"You can add me add a friend or talk to me on here if you wish."

"if he ever wants to talk or has questions let me know! I'd more than happy to!!"

Patients in active treatment sought out new connections and friendship via the online support group. Patients posted the following messages:

> "I have osteosarcoma in my left femur with metastatic lung noduals...message me if you want to chat I'd really love to meet people going through this same thing!!!"

> "Hey all! 28 year old mom of 2. Diagnosed with patriarchs on 9/11/12. 9/25/12 started the 4 rounds of the harshest chemo (cisplatin and adrianmycn). finally had surgery on 1/7/12 to remove the one localized tumor in my right knee via limb sparing surgery. I found this group just looking around on Facebook. I am hoping to find some good tips and possibly friendship/support."

Unity. Content coded as unity support contained reminders from one group member to another of the strength that comes in numbers. Patients posting unity messages seemed to understand that a coherent online support group contributes to the successful coping of its members. Some examples of unity giving messages include the following:

"we will all fight this together"

"Just know that whatever the outcome...you are among family here."

"we are all routing for you!!"

While there were a number of unity giving posts, there were not any posts where patients in active treatment requested messages of unity from other members.

Network referral. Messages in this category contained tips and suggestions for expanding one's existing social support network. For example, group members often referred one another to additional support groups related to osteosarcoma. Group members also utilized the network to request content "shares" that would expand the reach of their individual posts or personal stories beyond the reach of their own individual networks. Some examples of network referral giving are as follows:

> "Have you joined sarcoma alliance? If not please do, they have thousands of current and former patients that offer much needed support and great info!"
>
> "Gofundme.com has been great for us!"

An example of referral seeking behavior includes the following:

> "Hi everyone Please like and share this page with your friends"

Esteem Support

Like network support, esteem support accounted for only a little more than 6% of all social support content posted to the study group by patients in active treatment. Although relatively infrequent, when it did occur, esteem support was very strongly biased in favor of support giving. Study participants were 15 times more likely to give esteem support to others than to seek esteem support for themselves. Esteem support messages were subcategorized into three areas: affirmation (91.7% giving/8.3% seeking), inspiration (90.9% giving/9.1% seeking), and compliment (100% giving).

Affirmation. Message content coded under the affirmation category contained statements by one user regarding the competence or effectiveness of another individual or group as providers of social support. The implied purpose of such messages appeared to be to increase the self-confidence and esteem of the intended message recipients. For example, patients in active treatment made the following affirmational statements to other group members:

> "You are definetely a hero. I'm battling cancer now and I have a son. You have been an inspiration for me."
>
> "I'm naturally a very positive thinking and optimistic person but Mary you give me even better outlook so thanks!"
>
> "great group with awesome support!"

Patients in active treatment also sought affirmation from other members. For example, one user posted, "Can you let us know if something worked that we suggested?" The response to this post will have two benefits for the group: 1) to enhance the credibility of the advice given and the social stature of the support provider, and 2) to share the results of following the given advice with others who may benefit from taking similar action.

Inspiration. Group members often expressed confidence or optimism on behalf of other group members, empowering or encouraging other members to achieve positive results in their treatment. Some examples of patients in active treatment giving inspiration to other members include the following:

> "I too am 31 with 2 boys 10 and 4 and it's not fair but I continue to fight for them as you will!!"
>
> "Fight, fight, fight and you'll be alright!"

One example of a patient in active treatment seeking inspiration was identified.

This category is represented by the following post:

> "Im going through a hard time emotionally. I would love to hear some survival stories."

Compliment. Group members posted expressions of praise, admiration or respect for one another. The following examples of these behaviors were observed in the sample group:

> "Everyone here is awesome."
>
> "Shes beautiful:)"
>
> "you are a peach."

There were not any examples of group members seeking compliments from other members.

Spiritual Support

In the present study, a little more than 15% of all social support content contained some form of spiritual support. Spiritual support was classified into three categories: prayer (75% giving/25% seeking), helping thoughts (80% giving/20% seeking), and well-wishing (95.9% giving/4.1% seeking). In cases where spiritual support content was observed, message authors were eight times more likely to offer spiritual support to others than to seek spiritual support for themselves from others.

Prayer. Of the three subthemes of spiritual support in the sample group, prayer was the most common. Prayer messages included both offers to pray for others and requests for prayers. Prayer requests included nonspecific requests (pray for me) as well as requests to pray for specific outcomes. Some examples of prayer giving messages include the following:

"You are in my prayers"

"my prayers are with you and your family!"

"praying for all of you"

Examples of prayer seeking messages include the following:

"keep me in your prayers... im nervous"

"shoot I'd love some prayers and good vibes."

"Please pray for God to give me the strength I need to fight this disease and pray that He gives me peace, hope, and comfort."

"Please pray for my journey to continue on this positive path."

Helping thoughts. There was a commonly expressed belief among group members that having one member think about another member would bring positive health benefits to the person being thought of. Any messages exhibiting such beliefs were categorized as "helping thoughts." In some messages, the word thought seemed to be used interchangeably with prayer. In other cases, it was obvious that the message author clearly distinguished between prayer and thoughts as separate sources of support (e.g., individuals asking for thoughts AND prayers). Some examples of patients in active treatment giving helping thoughts to others include the following:

"I will keep u in my thoughts"

"Thinking about you"

"In my thoughts!!"

"I'm praying for you and your son. I know it doesn't seem like much, but every positive thought out there counts for something."

"Brave kid my thoughts and prayers go out to him that he gets through it all just fine."

Group members also sought out helping thoughts from other members, especially during periods of heightened stress or anxiety. Two examples of members seeking helping thoughts are as follows:

> "missing my beau and babies hard right now. Please think of us. xo"

> "My first chemo treatment starts tomorrow..2-3 days in the hospital. Please keep me and my family in your thoughts. I'm pretty scared!"

Well-wishing. Like prayer messages, well-wishing messages often contained language about invoking a higher power to intercede on behalf of a patient undergoing cancer treatment (e.g., seeking God's blessings). The difference here is that the message author is prospectively stating a desired outcome from an external source rather than expressly describing prayer or asking for help. Well-wishing messages which did not include references to Deity, generally contained expressions of luck or good fortune instead. Whether well-wishing expressions credited Deity or luck, they always invoked an external source of strength for help. For this reason, well-wishing messages were placed in the same category as prayer and helping thoughts; spiritual support. Examples of well-wishing giving messages include the following:

> "may God give you peace and comfort"

> "good luck to u!"

> "Best wishes for your family and son."

> "God bless good luck"

> "God bless you"

Examples of well-wishing seeking include the following:

> "And I guess wish me luck."

> "Wish me luck!!"

Discussion

The present study addressed two research questions. First, how can the exchange of social support in an online support group dedicated to osteosarcoma be characterized? Second, how do patients in an online support group who are in active treatment leverage the platform to meet their social support goals?

The results of this study clearly showed that a Facebook group can provide a rich environment where diverse types of social support are exchanged. In the study group, patients in active treatment not only utilized the group to seek or receive social support from others, but they also used the platform to provide support to others facing similar circumstances. In fact, nearly 80% of all socially supportive messages posted to the group were related to support giving rather than support seeking behaviors.

One could conclude from these results that that the study participants were inherently more altruistic than self-interested. Although this could be true, such a conclusion would be incredulous without further empirical investigation. There are also other factors which more easily explain the phenomenon. For example, the number of support seeking posts in the group is inherently limited by individual decisions to seek support. There is no “big bang” in support seeking behavior. Such behaviors are only initiated when an individual encounters or perceives a need for support. Although it is possible for someone to encourage another member to post a support seeking message, this is difficult to do in informal relationships, where one member may not be intimately aware of the support needs of another member.

At the same time, there are many stimuli within the group that have the potential to initiate support giving behaviors. Each support seeking message that is posted to the group has the potential to generate multiple responses; there is no theoretical limit to the number of replies possible per posting. In the study group, the average number of replies per posting was 7.24 with an SD of 7.53 messages. The minimum number of replies per post was 1 and the maximum number of replies per post was 49. The large number of replies per post could certainly have impacted the proportion of support giving to support seeking behaviors.

Although it would be unwise to place undue emphasis on the proportion of support giving versus support seeking behavior, the researcher is not suggesting that support giving behavior is irrelevant to osteosarcoma patients, or that this dimension is immaterial to the present research. In fact, quite the opposite is true. First, there is a symbiotic relationship between support giving and support seeking behavior. One cannot exist without the other. Having access to a large number of connections also does not necessarily mean that someone is socially engaged or supported. For example, if an osteosarcoma patient joins an online support group, but never actually logs in to the group, it will be impossible for them to feel supported by the group. “The presence of actual social contact is required to provide any sense of support or lack of it” (Stephens, Alpass, Towers, & Stevenson, 2011).

Second, Weiss (1974) and others have argued that personal relationships provide “opportunities for nurturance,” and these opportunities, can in and of themselves, be health promoting. According to Weiss, nurturing relationships provide “meaning to an individual’s life and to sustain commitment to goals in a wide variety of activities” (1974, p. 23). Rare disease research has confirmed this finding, and has demonstrated that many patients find personal meaning in feeling needed, or that they can help or inspire others with similar conditions (Olsson Ozanne, Graneheim, Persson, & Strang, 2012).

One of the findings of the present study concerned the importance and prevalence of spiritual support in the online support environment. As mentioned earlier, spiritual support was the fourth most frequently encountered form of social support in the study group, comprising approximately 15% of all social support content identified by the researcher. Spiritual support was most often expressed as requests for prayer, or offers to pray on behalf of another group member. While some theorists (e.g., Cutrona & Suhr, 1990) have proposed that prayer be coded as emotional support, it was clear that most of these patients saw prayer as doing much more than supporting emotional health. Prayer was perceived by these individuals as communicating with, or drawing upon an external source of strength, and seeking assistance from that external source.

Group members also expressed beliefs that positive or helping thoughts from one member could result in positive health outcomes for another member. In some cases, the phrase “in my thoughts” appeared to be used synonymously with “in my prayers,” but in other cases users clearly distinguished between these concepts (e.g., by keeping others in thoughts AND prayers). These messages also strongly supported separating spiritual support from emotional support. The meaning of these messages was clear; participants viewed positive thoughts as an external force for good that had the power to help beyond altering the emotional state of a support recipient. The positive thoughts resided within the support provider and somehow the energy from those thoughts would benefit and sustain the support recipient.

Since the need for a spiritual support category did not emerge until after the study had begun, the researcher had not thought to include the keyword phrase “spiritual support" in the initial literature search and review. This also why spiritual support was not included in Table 4.2. It was surprising to discover that spiritual support was absent from all of the early writings and many of the well-known literature reviews on social support, especially since the concepts of positive spiritual energy and wellness are not foreign to researchers in the health fields. Similar concepts are found in the naturalistic Qi-based medicinal philosophies of east Asian cultures. For example, Qi Gong practitioners believe it is possible to "project one's internal Qi towards another body" (Eisenberg & Wright, 1995, p. 211). Nevertheless, not one of the initial frameworks of social support proposed by theorists contained a spiritual support component.

One reason why some theorists may not consider spiritual support to be a separate component of social support is because of the potential for intercorrelations between spiritual and emotional support. For example, Peacock, Wong and Reker (1993) found a statistically significant correlation ($r = .16$, $p<.05$) between emotion-focused and spiritual coping schemas. It should be noted, however, that correlations have also been found between other functional components of social support, and this does not necessarily mean that the concepts are singular in nature. For example, Sarason et al. (1987) conducted a study of correlations between social support measures, and found highly significant correlations between a variety of social support measures, including emotional support and tangible assistance ($r=.44$, $p<.001$, two-tailed). The question of whether emotional support and spiritual support are separate components merits additional analysis and investigation. If, at a functional level, patients perceive these components to be distinct, then perhaps the concepts could benefit from further definitional clarity. Definitional slurring between spiritual and emotional support could partially account for observed correlations. Findings from the current study suggest that spiritual support is more likely a multidimensional, rather than a unitary concept. Future investigations should examine, in greater depth, the relationship between each of the subcomponents of spiritual and emotional support.

A quick search of the literature reveals that researchers are beginning to aggressively examine the nature and influence of spiritual support (see Table 4.5). A Google Scholar search reveals that more articles containing the keyword "spiritual support" have been published in the last 5 years than had been published in the entire decade spanning the years 2000 to 2010. This is also true of every major functional component of social support, except emotional support. There are also more articles about spiritual support than about some of the other more traditional social support concepts. Table 4.5 contains a recap of the growth in published articles by keyword for each of the major functional components of social support.

The emergence of spiritual support as a research discipline has followed a path similar to that of many of the other social support constructs. Early work has focused primarily on establishing the underlying theory and definition of the construct. Later work has focused on developing measurement instruments. A variety of definitions have been put forth for spiritual support and these definitions will be of use to future researchers seeking to examine the role of spiritual support in online rare disease communities (see Ai, Peterson, & Huang, 2005; Conrad, 1985; Krause, Ellison, Shaw, Marcum, & Boardman, 2001; Kuuppelomaki, 2001; Maton, 1989; Stiles, 1994) . Of all of these definitions, the definition put forth by Krause et al. (2001) is most closely aligned with the current research. Krause et al. (2001) have defined spiritual support as individuals "help[ing] people maintain and deepen their faith, as well as apply their religious beliefs in daily life." In the present study, individuals often extended offers of prayer, well-wishing, or helping thoughts to others or sought such actions for themselves. These efforts served in different ways to maintain and deepen the faith and personal convictions of both support givers and believers with respect to divinity, luck, external intervention and health.

In spite of the many references to God in group messages, the researcher was careful not to restrict the grouping of "spiritual support" content to theistic or non-naturalistic posts, or to posts exemplifying the belief system of any one group of people. Researchers have argued that any definition of spiritual support must encompass diverse cultures and belief systems (Baldacchino & Draper, 2001; Paley, 2008; Tuncay, 2007). Limiting definitions of spiritual support to participants from only one form of religion or belief system, would overlook the important but different ways in which diverse groups express their needs for support from others in similar circumstances. The need for a broader definition of spiritual support is reinforced by the following post from an active treatment patient in the study population:

> "Ok. I don't want to sound disrespectful or ungrateful, or offensive. I have a serious question. ALOT of people have told me that now I need to find god, pray, and so on....Now that I have cancer.....Now that..well whatever...the point is...is this normal..How do I get them to stop. While I appreciate their beliefs and prayers, its not mine. I appreciate the support. How do I get them to stop?" (March 2013)
>
> "I believe in him...I'm just not talking to him...haven't for many years. And just because of this challenge...I'm not going to suddenly start. Thank you for responding and understanding. I'm sure some people might think thats horrible of me...but honestly...I'm not sorry." (March 2013)

Definitional flexibility is necessary for achieving a complete understanding of the social support experiences of patients who may exhibit multiple spiritual orientations, or whose belief systems change over time. For example, the following messages were posted to the group by the same individual that posted the prior statements about not wanting prayer support.

> "Anyone heard of trying a spirit healer? My kid actually brought one home today, lol. Sounds different to me, but I won't say no to anything...." (June 2013)
>
> "I'm praying for you and your son. I know it doesn't seem like much, but every positive thought out there counts for something. And remember...you can always message group members for support or just to chat." (August 2013)

Krause et al. (2001) have suggested that individuals providing spiritual support help others "apply their religious beliefs in daily life." This can be accomplished no matter what a person's religious orientation might be. Those seeking spiritual support will also expect others to help them to apply the tenets of their faith toward their treatment and recovery from illness.

In addition to spiritual support, there were two other social support dimensions that yielded surprising results: appraisal support and tangible support. As shown in Table 4.5, appraisal support is one of the least researched dimensions of social support. Only esteem support has received less attention in the scientific literature, and yet, appraisal support was the most frequently encountered type of support in this study (25% of messages).

There are at least two possible explanations for this phenomenon. First, the Facebook group examined in this study provides a unique opportunity for osteosarcoma patients to seek out information, and compare themselves to others in similar circumstances. Presumably, some of the information posted to the group about common challenges and support needs associated with the disease cannot be found anywhere else, even from a patient's own physician. Although not specific to osteosarcoma, recent studies have supported this notion in the scientific literature. For example, a study examining the level of comfort of clinicians at counseling with rare disease patients found that many medical professionals lack personal understanding or training in the area of rare diseases. As many as 56.7% of primary care physicians and 40% of specialists report their training in rare diseases as being either neutral, ineffective, or very ineffective (Engel, Bagal, Broback, & Boice, 2013). The lack of objective knowledge and understanding about these conditions makes it difficult for providers to adequately advise their patients on diagnosis or treatment options (Leonard, 2004). Festinger's (1954) social comparison theory suggests that "when an objective, non-social basis for evaluating one's ability or opinion is…unavailable, people will…evaluate their opinions and abilities by comparing themselves with others." Social comparison theory also suggests that, when possible, individuals prefer comparing themselves to others who are similar in terms of opinion or ability. If social comparison theory is correct, the high proportion of appraisal-related messages in the study group could be related to a systemic lack of objective information and health literacy among osteosarcoma patients. Additional research should be performed to evaluate this concern, and propose solutions.

Another possible explanation for the high number of appraisal related posts, could be that appraisal support (like spiritual support) is highly correlated with other types of social support. Sarason et al. (1987) have presented evidence to suggest this may be the case. Correlations between appraisal support and belonging support (r=.66, p<.001,two-tailed for men/r=.72, p<.001, two-tailed for women), appraisal support and tangible support (r=.42, p<.001, for men/r=.62, p<.001, for women), and appraisal support and self-esteem (*r*=.52, *p*<.001, for men/*r*=.73, *p*<.001, for women) have all been found to be highly significant.

Thoits (1986, 1995) and Cohen and McKay (1984) have argued that support is most effective when it comes from someone who is socially similar to the support recipient, and has faced similar circumstances. When the support provider is similar to the support recipient, the potency of the support provider as a point of social comparison is enhanced. Shared circumstances also promote empathetic understanding between support provider and support recipient. According to Heaney and Israel (2002), this understanding is “particularly relevant to the exchange of emotional support but also applies to instrumental and informational support” (p. 197). Since emotional and information support were the second and third most frequently expressed forms of social support in the group, this may help to explain the high occurrence of appraisal related discussions.

Finally, tangible support was the most infrequently expressed component of social support in messages posted to the study group by osteosarcoma patients in active treatment. This was surprising, since tangible or instrumental support is the second most frequently researched component of social support in the literature to date (see Table 4.5). Osteosarcoma patients also have significant financial needs associated with their initial care and ongoing treatment. Coulson et al. (2007) found similar results in a study of a message board for Huntington’s disease; 51.9% of posts contained emotional support, 48.4% of posts contained network support, 21.7% contained esteem support, and 9.8% contained tangible assistance.

The following post suggests that this phenomenon may also be related to social comparison. Since most of the participants in the group have financial needs, group members may feel hesitant about asking one another to provide financial support.

> "I dont know if this is tacky or not, but my family started a fundraiser online for me…I am posting it here, but no pressure."

This does raise an important question. If online support groups are the primary medium of supportive exchange for rare disease patients, and patients are uncomfortable posting online about their need for financial support, where else do they turn? This is another area that warrants further investigation.

Conclusion

The present study demonstrates that patients in active treatment can derive many benefits from participating in osteosarcoma-oriented Facebook groups. Facebook groups provide an ideal medium for patients to exchange diverse types of social support, and patients seem comfortable both giving and receiving support to one another online. Given the many benefits of Facebook groups for osteosarcoma patients, further research is warranted to understand how to better improve the reach and implications of such groups, not only for patients in active treatment, but also for other types of Facebook users (e.g., survivors, caregivers, etc.). Additional research is also recommended to better understand the role that spirituality, social comparison, and tangible support play in osteosarcoma and other rare disease populations. Such research will facilitate better matching between different types of patients and available support communities. Health benefits in underserved populations will also be increased as study findings are disseminated to clinicians, patient advocates, caregivers, and community organizers who seek to create more effective online support environments, and direct patients to relevant support groups.

Table 4.1: Number and Percentage of Entries Based on User Classification

Classification	**Number of Postings**	**% of Total Postings**
Nonpatient	2120	47.63
Active Treatment	686	15.41
External Organization	112	2.52
In Memoriam	590	13.26
Survivor	724	16.27
Unknown	8	0.18
Patient	2213	49.72
Active Treatment	644	14.47
External Organization	2	0.04
Survivor	1562	35.09
Unknown	5	0.11
Unknown	118	2.65
External Organization	10	.22
Unknown	108	2.43
Grand Total	4451	

Table 4.2: Comparison of Functional Components of Social Support

Articles	**Emotional Support**	**Informational Support**	**Tangible Support**	**Esteem Support**	**Network Support**	**Appraisal Support**	**Active Support /Nurturance**	**Motivational Support**	**Status Support**
Weiss (1974)	Attachment	The obtaining of guidance	A sense of reliable alliance	Reassurance of Worth	Social Integration		Opportunity for Nurturance		
Caplan (1974)	Help to master emotional burdens	Cognitive guidance	Sharing of tasks. Money, materials, tools and skills.						
Cobb (1979)	Emotional Support	Instrumental Support	Material Support	Esteem Support	Network Support		Active Support /Mothering		
Kahn (1979)	Affect		Aid	Affirmation					

Table 4.2: Continued

Articles	Emotional Support	Informational Support	Tangible Support	Esteem Support	Network Support	Appraisal Support	Active Support /Nurturance	Motivational Support	Status Support
Hirsch (1980)	Emotional Support	Cognitive Guidance	Tangible Assistance	Social Reinforcement	Socializing				
House (1981)	Emotional Support	Informational Support	Instrumental Support			Appraisal Support			
Schaefer, Coyne, & Lazarus (1981)	Emotional Support	Information Support	Tangible Support						
Lin, Dean, & Ensel (1981)	Expressive Support		Instrumental Support						

Table 4.2: Continued

Articles	**Emotional Support**	**Informational Support**	**Tangible Support**	**Esteem Support**	**Network Support**	**Appraisal Support**	**Active Support /Nurturance**	**Motivational Support**	**Status Support**
Vaux (1982)	Emotional Support	Advice/Guidance	Practical Assistance, Financial Assistance		Socializing				
Cohen & Hoberman (1983)		Appraisal Support	Tangible	Self-Esteem	Belonging				
Cohen & McKay (1984)	Emotional Support		Tangible Support			Appraisal Support			
Wills (1985)	See Esteem Support	Informational Support	Instrumental Support	Esteem Support	Social Companionship			Motivational Support	Status Support

Table 4.2: Continued

Articles	**Emotional Support**	**Informational Support**	**Tangible Support**	**Esteem Support**	**Network Support**	**Appraisal Support**	**Active Support /Nurturance**	**Motivational Support**	**Status Support**
Barrera (1986)					Social Embeddedness				
Cutrona & Russell (1990)	Emotional Support	Informational Support	Tangible Aid	Esteem Support	Network Support				
Cutrona & Suhr (1992)	Emotional Support	Informational Support	Tangible Assistance	Esteem Support	Network Support				
Langford, Bowsher, Maloney & Lillis (1997)	Emotional Support	Informational Support	Instrumental Support			Appraisal Support			

Table 4.3: Categories and Subcategories of Social Support for Content Analysis

Support Type	**Definition**
Emotional Support	"…the ability to turn to others for comfort and security during times of stress, leading the person to feel that he or she is cared for by others" (Cutrona & Russell, 1990).
Cheering	Congratulatory messages. Shared excitement for desirable test results or good news.
Empathy	Communication that indicates an ability for one person to understand another person is feeling.
Love & Affection	Expressions of caring between two or more individuals.
Condolences or Sympathy	Statements of sorrow or regret for something that either has happened or is currently happening to an individual.
Expressions of Concern	Inquiries from one individual about the state of being or health of another individual.
Informational Support	"…providing the individual with advice or guidance concerning possible solutions to a problem" (Cutrona & Russell, 1990).
Referral	Recommendations for individuals to seek input from an outside expert or source of information. The purpose of the referral is to encourage additional consultation prior to taking further action. Unlike network referrals, the primary purpose is to connect recipients to information rather than to expand one's own support network.
Suggestion or Instruction	Information or recommendations about health treatments or diets, or suggested steps for relieving symptoms or discomfort related to care.
Awareness	Information designed to increase the general level of consciousness or mindfulness regarding an idea or issue (no specific call to action).
Tangible Support	"...concrete instrumental assistance, in which a person in a stressful situation is given the necessary resources (e.g., financial assistance, physical help with tasks) to cope with the stressful event" (Cutrona & Russell, 1990).
Time or Activity	Requests for or offers of help with specific tasks or actions that would benefit another person.
Financial	Requests for or offers of material assistance

Table 4.3: Continued

Support Type	**Definition**
Appraisal Support	"Like informational support, involves only transmissions of information, rather than the affect involved in emotional support or the aid involved in instrumental support. However, the information involved in appraisal support is relevant to self-evaluation… (a.k.a.) social comparison" (House, 1981).
Validation	Expressing agreement with another person's perspective or point of view on a stressful situation.
Social Comparison	Exchanging detailed information with another about their disease symptoms, progression or prognosis for the purposes of making comparisons.
Network Support	"a person's feeling part of a group whose members have common interests and concerns" (Cutrona & Russell, 1990).
Connection & Friendship	Either reinforcing an existing social connection or extending an invitation to establish a new relationship.
Unity	Communicating shared concerns and togetherness.
Network Referral	Tips and suggestions for expanding one's existing social support network.
Esteem Support	"…the bolstering of a person's sense of competence or self-esteem by other people" (Cutrona & Russell, 1990).
Compliment	Messages that uplift by expressing praise, admiration or respect.
Inspiration	Expressions of confidence or optimism on behalf of another.
Affirmation	Statements regarding the competence or effectiveness of an individual or group as a provider of social support.
Spiritual Support	"…help[ing] people maintain and deepen their faith, as well as apply their religious beliefs in daily life" (Krause, 2001).
Well-Wishing	Pronouncing future blessings, luck, or good fortune on another.
Prayer	Communicating with or petitioning a source of higher strength or Deity.
Helping Thoughts	Initiating positive thoughts or sending positive energy on behalf of another.

Table 4.4: Descriptive Statistics Based on Social Support Classification: Frequency of Supportive Exchanges by Category

Support Classification	N	% of Total	Mean	Median	SD	Range
Giving Support	601	79.50%	85.86	102	57.12	13-166
Emotional (cheering, empathy, love and affection, condolences or sympathy, expressions of concern)	166	27.62%	33.20	36.00	15.90	10-54
Informational (referral, suggestion or instruction, awareness)	102	16.97%	34.00	30.00	13.45	23-49
Tangible (time or activity, financial)	13	2.16%	6.50	6.50	0.71	6-7
Appraisal (validation, social comparison)	139	23.13%	69.50	69.50	45.96	37-102
Network (connection and friendship, unity, network referral)	33	5.49%	11.00	8.00	7.94	5-20
Esteem (compliment, inspiration, affirmation)	45	7.49%	15.00	13.00	6.24	10-22
Spiritual (prayer, well-wishing, helping thoughts)	103	17.14%	34.33	24.00	32.75	8-71
Seeking Support	155	20.50%	22.14	14	18.61	2-51
Emotional (cheering, empathy, love and affection, condolences or sympathy, expressions of concern)	12	8.50%	2.40	1.00	4.28	0-10
Informational (referral, suggestion or instruction, awareness)	46	30.07%	15.33	18.00	12.22	2-26
Tangible (time or activity, financial)	16	10.46%	8.00	8.00	0.00	8-8
Appraisal (validation, social comparison)	51	33.33%	25.50	25.50	24.75	8-43
Network (connection and friendship, unity, network referral)	14	9.15%	4.67	2.00	6.43	0-12
Esteem (compliment, inspiration, affirmation)	3	1.31%	1.00	1.00	1.00	0-2
Spiritual (prayer, well-wishing, helping thoughts)	13	8.50%	4.33	3.00	3.21	2-8
Grand Total	756					

Note: $\bar{x}$, M, SD, and ranges are based on the number of social support exchanges per subcategory

Table 4.5: Published Articles by Year for Each of the Major Social Support Constructs

Google Scholar Keyword Search	<1970	1970-1979	1980-1989	1990-1999	2000-2009	2010-Present
“Social Support”	1,570	4,070	30,500	180,000	1,070,000	206,000
“Emotional Support” and “Social Support”	63	70	3,250	11,000	28,800	18,100
*“Instrumental Support” and “Social Support”	7	6	508	2,130	7,210	8,380
*“Tangible Support” and “Social Support”	3	9	214	866	2,580	2,890
“Informational Support” and “Social Support”	7	4	248	1,210	4,020	4,920
“Spiritual Support” and “Social Support”	4	5	93	589	3,000	3,410
“Network Support” and “Social Support”	1	21	421	769	2,190	2,270
“Appraisal Support” and “Social Support”	2	2	129	379	1,120	1,290
“Esteem Support” and “Social Support”	1	6	126	444	1,050	1,010

Source: Google Scholar (April 18, 20015)

*Instrumental support and tangible support are often used interchangeably.

References

Ai, A. L., Tice, T. N., Peterson, C., & Huang, B. (2005). Prayers, spiritual support, and positive attitudes in coping with the September 11 national crisis. *Journal of Personality, 73*(3), 763-792.

American Cancer Society. (2013). Cancer Facts & Figures 2013. *Estimated Number* of New Cancer Cases and Deaths by Sex, US, 2013* (pp. 64): American Cancer Society.

American Cancer Society. (2015). Osteosarcoma overview. Retrieved April 20, 2015, from http://www.cancer.org/acs/groups/cid/documents/webcontent/003069-pdf.pdf

Bacigalupe, G. (2011). Is there a role for social technologies in collaborative healthcare? *Families, Systems, & Health, 29*(1), 1.

Baldacchino, D., & Draper, P. (2001). Spiritual coping strategies: A review of the nursing research literature. *Journal of Advanced Nursing, 34*(6), 833-841.

Barrera, M. (1986). Distinctions between social support concepts, measures, and models. *American Journal of Community Psychology, 14*(4), 413-445.

Caplan, G. (1974). *Support systems and community mental health: Lectures on concept development*. New York: Behavioral Publications.

Cobb, S. (1976). Social support as a moderator of life stress. *Psychosomatic Medicine, 38*(5), 300-314.

Cobb, S. (1979). Social support and health through the life course. In M. W. Riley (Ed.), *Aging from birth to death: Interdisciplinary perspectives* (pp. 93-106). Boulder: Westview Press.

Cohen, J. (1960). A coefficient of agreement for nominal scales. *Educational and Psychological Measurement, 20*(1), 37-46. doi: 10.1177/001316446002000104

Cohen, S., & Hoberman, H. M. (1983). Positive events and social supports as buffers of life change stress. *Journal of Applied Social Psychology, 13*(2), 99-125.

Cohen, S., & McKay, G. (1984). Social support, stress, and the buffering hypothesis: A theoretical analysis. *Handbook of Psychology and Health, 4*, 253-267.

Conrad, N. L. (1985). Spiritual support for the dying. *The Nursing clinics of North America, 20*(2), 415-426.

Coulson, N. S., Buchanan, H., & Aubeeluck, A. (2007). Social support in cyberspace: A content analysis of communication within a Huntington's disease online support group. *Patient Education and Counseling, 68*(2), 173-178.

Cutrona, C. E., & Russell, D. W. (1990). Type of social support and specific stress: Toward a theory of optimal matching. In B. R. Sarason, I. G. Sarason & G. R. Pierce (Eds.), *Social Support: An Interactional View* (pp. 319-366). New York: Wiley-Interscience.

Cutrona, C. E., & Suhr, J. A. (1992). Controllability of stressful events and satisfaction with spouse support behaviors. *Communication Research, 19*(2), 154-174.

Damron, T. A., Ward, W. G., & Stewart, A. (2007). Osteosarcoma, chondrosarcoma, and Ewing's sarcoma: National Cancer Data Base Report. *Clinical Orthopaedics and Related Research, 459*, 40-47.

Darling, J. (2007). A different view of sarcoma statistics. *ESUN, 4*(6).

Davison, K. P., Pennebaker, J. W., & Dickerson, S. S. (2000). Who talks? The social psychology of illness support groups. *American Psychologist, 55*(2), 205.

Dorfman, H. D., & Czerniak, B. (1995). Bone cancers. *Cancer, 75*(S1), 203-210.

Dunoyer, M. (2011). Accelerating access to treatments for rare diseases. *Nature Reviews Drug Discovery, 10*(7), 475-476.

Eisenberg, D., & Wright, T. L. (1995). *Encounters with Qi: Exploring Chinese medicine*. New York: WW Norton & Company.

Ell, K. (1984). Social networks, social support, and health status: A review. *The Social Service Review*, 133-149.

Engel, P. A., Bagal, S. B., Broback, M., & Boice, N. (2013). Physician and patient perceptions regarding physician training in rare diseases: The need for stronger educational initiatives for physicians. *Journal of Rare Disorders, 1*(2), 1-15.

Facebook Inc. (2015). Facebook Reports Fourth Quarter and Full Year 2014 Results. Retrieved April 20, 2015, from http://investor.fb.com/releasedetail.cfm?ReleaseID=893395

Festinger, L. (1954). A theory of social comparison processes. *Human Relations, 7*(2), 117-140.

Geller, D. S., & Gorlick, R. (2010). Osteosarcoma: A review of diagnosis, management, and treatment strategies. *Clin Adv Hematol Oncol, 8*(10), 705-718.

Hampton, T. (2006). Rare disease research gets boost. *JAMA: The Journal of the American Medical Association, 295*(24), 2836-2838.

Hawn, C. (2009). Take two aspirin and tweet me in the morning: How Twitter, Facebook, and other social media are reshaping health care. *Health Affairs, 28*(2), 361-368. doi: 10.1377/hlthaff.28.2.361

Heaney, C. A., & Israel, B. A. (2002). Social networks and social support. In K. Glanz, B. K. Rimer & K. Viswanath (Eds.), *Health behavior and health education: Theory, research, and practice* (Vol. 4, pp. 189-210). San Francisco: Josey-Bass.

Hirsch, B. J. (1980). Natural support systems and coping with major life changes. *American Journal of Community Psychology, 8*(2), 159-172.

House, J. S. (1981). *Work stress and social support*. Reading: Addison-Wesley.

Hsieh, H.-F., & Shannon, S. E. (2005). Three approaches to qualitative content analysis. *Qualitative Health Research, 15*(9), 1277-1288.

Kager, L., Zoubek, A., Dominkus, M., Lang, S., Bodmer, N., Jundt, G., . . . Bielack, S. (2010). Osteosarcoma in very young children. *Cancer, 116*(22), 5316-5324.

Kahn, R. L. (1979). Aging and Social Support. In M. W. Riley (Ed.), *Aging from birth to death: Interdisciplinary perspectives* (pp. 77-91). Boulder, CO: Westview Press.

Krause, N., Ellison, C. G., Shaw, B. A., Marcum, J. P., & Boardman, J. D. (2001). Church-based social support and religious coping. *Journal for the Scientific Study of Religion, 40*(4), 637-656.

Kraut, R. E., & Fiore, A. T. (2014). The role of founders in building online groups. *Proceedings of the 17th ACM conference on Computer supported cooperative work & social computing*, 722-732.

Kuuppelomäki, M. (2001). Spiritual support for terminally ill patients: Nursing staff assessments. *Journal of Clinical Nursing, 10*(5), 660-670.

Lamberg, L. (1997). Online support group helps patients live with, learn more about the rare skin cancer ctcl-mf. *JAMA, 277*(18), 1422-1423. doi: 10.1001/jama.1997.03540420016005

Langford, C. P. H., Bowsher, J., Maloney, J. P., & Lillis, P. P. (1997). Social support: A conceptual analysis. *Journal of Advanced Nursing, 25*(1), 95-100.

Leonard, H. (2004). How can the Internet help parents of children with rare neurologic disorders? *Journal of Child Neurology, 19*(11), 902-907.

Lin, N., Dean, A., & Ensel, W. M. (1981). Social support scales: A methodological note. *Schizophrenia Bulletin, 7*(1), 73-89.

Maton, K. I. (1989). The stress-buffering role of spiritual support: Cross-sectional and prospective investigations. *Journal for the Scientific Study of Religion*, 310-323.

Mirabello, L., Troisi, R. J., & Savage, S. A. (2009). International osteosarcoma incidence patterns in children and adolescents, middle ages and elderly persons. *International Journal of Cancer, 125*(1), 229-234.

Montoya, I. D. (2011). Rare diseases: Challenges and resources. *Journal of Nursing Education and Practice, 1*(1), p1.

National Cancer Institute. (2015). Osteosarcoma and Malignant Fibrous Histiocytoma of Bone Treatment (PDQ®). Retrieved April 20, 2015, from http://www.cancer.gov/cancertopics/pdq/treatment/osteosarcoma/HealthProfessional

O'Neill, N. (2014). Google Now Indexes 620 Million Facebook Groups. Retrieved June 7, 2014, from http://allfacebook.com/google-now-indexes-620-million-facebook-groups_b10520

Olsson Ozanne, A., Graneheim, U. H., Persson, L., & Strang, S. (2012). Factors that facilitate and hinder the manageability of living with amyotrophic lateral sclerosis in both patients and next of kin. *Journal of Clinical Nursing, 21*(9-10), 1364-1373.

Paley, J. (2008). Spirituality and nursing: A reductionist approach. *Nursing Philosophy, 9*(1), 3-18.

Patton, M. Q. (1990). *Qualitative research & evaluation methods*. Newbury Park, CA: Sage Publications, Inc.

Peacock, E. J., Wong, P. T., & Reker, G. T. (1993). Relations between appraisals and coping schemas: Support for the congruence model. *Canadian Journal of Behavioural Science/Revue canadienne des sciences du comportement, 25*(1), 64.

Picci, P. (2007). Osteosarcoma (osteogenic sarcoma). *Orphanet J Rare Dis, 2*(6).

Rados, C. (2003). Orphan products: Hope for people with rare diseases. *FDA consumer, 37*(6), 10.

Rimer, B. K., Lyons, E. J., Ribisl, K. M., Bowling, J. M., Golin, C. E., Forlenza, M. J., & Meier, A. (2005). How new subscribers use cancer-related online mailing lists. *Journal of Medical Internet Research, 7*(3).

Sarason, B. R., Shearin, E. N., Pierce, G. R., & Sarason, I. G. (1987). Interrelations of social support measures: Theoretical and practical implications. *Journal of Personality and Social Psychology, 52*(4), 813.

Savage, S. A., & Mirabello, L. (2011). Using epidemiology and genomics to understand osteosarcoma etiology. *Sarcoma, 2011*.

Schaefer, C., Coyne, J. C., & Lazarus, R. S. (1981). The health-related functions of social support. *Journal of Behavioral Medicine, 4*(4), 381-406.

Seoane-Vazquez, E., Rodriguez-Monguio, R., Szeinbach, S. L., & Visaria, J. (2008). Incentives for orphan drug research and development in the United States. *Orphanet Journal of Rare Diseases, 3*, 33.

Stephens, C., Alpass, F., Towers, A., & Stevenson, B. (2011). The effects of types of social networks, perceived social support, and loneliness on the health of older people accounting for the social context. *Journal of Aging and Health, 23*(6), 887-911.

Stiles, M. K. (1994). The shining stranger: Application of the phenomenological method in the investigation of the nurse family spiritual relationship. *Cancer Nursing, 17*(1), 18-26.

Thoits, P. A. (1986). Social support as coping assistance. *Journal of Consulting and Clinical Psychology, 54*(4), 416.

Thoits, P. A. (1995). Stress, coping, and social support processes: Where are we? What next? *Journal of Health and Social Behavior*, 53-79.

Tuncay, T. (2007). Spirituality in coping with HIV/AIDS. *HIV & AIDS Review, 6*(3), 10-15.

Vaux, A., Brownell, A., & Hill, P. (1982). Life Stress and Social Support in the Family System. Public Policy Research Organization, University of California,working paper #158.

Weiss, R. (1974). The provision of social relationships. In Zick Rubin (Ed), *Doing Unto Others* (pp. 17–26). Englewood Cliffs: Prentice Hall.

White, M., & Dorman, S. M. (2001). Receiving social support online: Implications for health education. *Health Education Research, 16*(6), 693-707.

Wills, T. A. (1985). Supportive functions of interpersonal relationships. In S. Cohen & S. L. Syme (Eds.), *Social support and health* (pp. 61-82). Orlando: Academic Press, Inc.

CHAPTER 5

CONCLUSION

The current dissertation study has helped to illuminate the role that online communities play in facilitating social support for those affected by rare disease. Although prior studies have examined the role of computer-mediated social support in healthcare, few studies have looked at online support participation through the lens of a rare disease patient. This study has generated many insights which will serve to facilitate a better understanding of this important and underserved population. It is hoped that these findings will encourage additional research in this important area.

The results of this study suggest that online support communities represent an important source of social support for rare disease patients and their families. Rare disease has many different constituents. In the first article, the researcher utilized the Patterson et al. (1997) framework, and work by other authors to discuss how computerized healthcare services, such as online support groups, have the potential to benefit not only patients, but also caregivers and clinicians. Research has suggested that different types of users utilize online support groups in different ways. For example, rare disease patients utilize online support groups for a variety of purposes, including seeking out information, support, treatments, providers, etc. Caregivers, on the other hand, are more likely to seek out general disease information, or information about end of life planning and disease prognosis. For clinicians, online communities represent an opportunity to share expertise, as well as to increase their knowledge and understanding of specific rare disease conditions. Clinicians benefit not only from reading the posts of patients, but also the opinions of other more experienced medical professionals.

The author of this dissertation has taken several steps to deepen the current understanding of how social support differs among individuals affected by rare disease, who also participate in online support groups. First, the researcher developed three different classification schemes for distinguishing between users of online support groups: group focus or orientation (person vs. population), treatment status (patient or nonpatient) and disease affiliation status (active treatment, survivor, in memoriam, or external organization). These classification schemes were found to be highly useful for classifying both the founders and participants of online support groups. Although an interrater reliability analysis was not computed for group focus, Cohen's kappa ranged from .81 for disease affiliation status to .95 for treatment status.

Second, the researcher began to analyze and identify the specific types of support exchanged within an online support group for those affected by rare disease. Although this dissertation has focused on only one group of participants, patients in active treatment, this study has laid the foundation for future studies involving other user populations (e.g., survivors, caregivers and clinicians). The results of this study clearly demonstrate that a Facebook group can provide a rich environment where diverse types of social support are exchanged.

In the study group, patients in active treatment not only utilized the group to seek or receive social support from others, but they also used the platform to provide support to others facing similar circumstances. The researcher found that several different types of social support were exchanged by patients in active treatment. The distribution of messages posted by patients in active treatment, according to each type of social support, was as follows: appraisal support (25.1%), emotional support (23.5%), informational support (19.6%), spiritual support (15.3%), network support (6.2%), esteem support (6.3%) and tangible support (3.8%). All of these support types were found to serve distinct, yet important health promoting functions.

Duncan (1989) has argued that more health educators should be familiar with content analysis. This study has demonstrated that content analysis can be useful for examining the content of online communities and for identifying objectives for health promotion and education research.

Limitations

There were several limitations of this research. First, group selection was limited to groups containing a sarcoma diagnosis in the title, conducted primarily in the English language, and related only to human forms of sarcoma. These criteria likely resulted in the exclusion of many highly relevant sarcoma groups. For example, it is quite possible that support groups exist that reference “fighting cancer” in the title, but not sarcoma, even though the group’s namesake is battling sarcoma. The prescribed search criteria would have included such groups in the study sample. While this restriction did not impede the researcher’s ability to acquire a sufficient number of groups for analysis, it may have caused the researcher to overlook important differences between groups. For instance, the generic keyword “cancer” is likely searched for more frequently on Facebook than keywords for rare forms of cancer, like sarcoma. Founders employing a more generic nomenclature for their groups might experience greater visibility and success at attracting new members than those included in the current analysis. Similarly, by restricting the study to groups in the English language, the researcher may have overlooked important cultural differences between groups.

Another limitation of the study concerns the distribution of patient versus nonpatient founders between groups. From the search results, it was clear that some conditions were more frequently represented in the study population than others. Disease specific factors, such as prevalence and prognosis, may have impacted not only the number of groups, but also the distribution of patient versus nonpatient founders between groups. The extent to which the distribution of diseases impacted the study findings was not examined.

Finally, the examination of specific types of support was only conducted using a single online support group. Although single group analyses are acceptable within the realm of qualitative research, additional studies are recommended to determine whether the patterns of communication observed in this study are also found in other rare disease oriented online support groups.

Opportunities for Future Research

The researcher was surprised to discover that appraisal support was the most prevalent type of social support in the study group. Festinger’s (1954) social comparison theory suggests that “when an objective, non-social basis for evaluating one’s ability or opinion is…unavailable, people will…evaluate their opinions and abilities by comparing themselves with others." Social comparison theory also suggests that, when possible, individuals prefer comparing themselves to others who are similar in terms of opinion or ability. If social comparison theory is correct, the high proportion of appraisal-related messages in the study group might suggest that there is a systemic lack of objective information and health literacy among the osteosarcoma patients in the study group. In the future, additional research should be performed to evaluate the prevalence of the concern and, if necessary, to propose solutions.

Given the myriad benefits of social exchange between comparable peers, it was surprising to discover that 78% of groups in the study sample were founded by nonpatients. The researcher wonders what impact this phenomenon might be having on the number of patients participating in online support groups. Are support group members also predominantly nonpatients? The answer to this question will have important implications for understanding the role that online communities play in supporting those affected by rare disease. If so, this would greatly contribute to our understanding of how caregivers seek out and provide social support. The answer to this question will be an important opportunity for future research.

There is evidence to suggest that self-appraisal may have also accounted for the low prevalence of expressions of tangible support within the group. For example, some patients expressed discomfort about seeking financial support from others while knowing that others likely faced similar financial challenges. This is unfortunate, especially since it will be difficult for patients to obtain financial support without expressing a need for it. This also raises an important question. If online support groups are the primary medium of supportive exchange for rare disease patients, and if they are uncomfortable posting online about their need for financial support, where else do they turn? Future studies will need to address this question.

Spiritual support was also found to play an important role in the sample group. In this dissertation study, a little more than 15% of all social support content identified by the researcher contained some form of spiritual support. Spiritual support was most often expressed as requests for prayer, or offers to pray on behalf of another group member. While some theorists (e.g., Cutrona & Suhr, 1990) have proposed that prayer be coded as emotional support, it was clear in this study that most patients saw prayer as doing much more than supporting emotional health. Prayer was perceived by these individuals as communicating with, or drawing upon an external source of strength, and seeking assistance from that external source. Additional research is recommended to determine to what extent spiritual support exists in other rare disease communities.

Although the present research has helped to increase understanding of the role and influence of social support for osteosarcoma patients in active treatment, it does not address differences in the types of support exchanged by members of other groups. Future studies are recommended to: 1) specifically examine the types of social support content within other sarcoma support groups, and 2) examine differences in socially supportive content between groups and between different types of members within groups. An investigation is also recommended to understand the impact of group focus (patient versus population) on social support. Although a group focus classification schema (patient versus population) was developed in this study, the significance of this schema for social support was not addressed.

Finally, additional research is recommended to better understand the role that spirituality, social comparison, and tangible support play in osteosarcoma and other rare disease populations. Such research will facilitate better matching between different types of patients and available support communities. Health benefits in underserved populations will also be increased as study findings are disseminated to clinicians, patient advocates, caregivers, and community organizers who seek to create more effective online support environments, and direct patients to relevant support groups.

References

Cutrona, C. E., & Suhr, J. A. (1992). Controllability of stressful events and satisfaction with spouse support behaviors. *Communication Research, 19*(2), 154-174.

Duncan, D. F. (1989). Content analysis in health education research: An introduction to purposes and methods. *Health Education, 20*(7), 27-31.

Festinger, L. (1954). A theory of social comparison processes. *Human Relations, 7*(2), 117-140.

Patterson, T. L., Shaw, W., & Masys, D. (1997). Improving health through computer self-health programs: Theory and practice. In P. F. Brennan, S. Schneider & E. Tornquist (Eds.), *Computers in Healthcare: Information networks for community health* (pp. 219-246). New York: Springer.