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Philadelphia College of Osteopathic Medicine

Department of Psychology

A FEASIBILITY STUDY OF A LQTS-SPECIFIC PROBLEM-SOLVING
WORKSHOP: PARENTS' PROBLEM-SOLVING SKILLS, COPING, HOPE, AND
WORRY

By Elizabeth A. Phelps, M.S.

Submitted in Partial Fulfillment of the Requirements for the Degree of

Doctor of Psychology

April 2016

PHILADELPHIA COLLEGE OF OSTEOPATHIC MEDICINE
DEPARTMENT OF PSYCHOLOGY

Dissertation Approval

This is to certify that the thesis presented to us by Elizabeth A. Phelps on the 23rd day of November, 2015, in partial fulfillment of the requirements for the degree of Doctor of Psychology, has been examined and is acceptable in both scholarship and literary quality.

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Acknowledgements

Completion of this dissertation would not have been possible without the undying support, care, and guidance of many people. I would first like to thank my dissertation committee, Dr. Stephanie Felgoise, Dr. Susan Panichelli Mindel, and Dr. Victoria Vetter. It is only through their guidance, enthusiasm, encouragement, and support that this project has come to fruition. I would like to offer a very special thanks to Dr. Stephanie Felgoise, who has served as a dedicated mentor, offering her vision, time, encouragement, and direction in helping me to grow not only as a clinical psychologist, but also as a researcher and innovative thinker.

I would like to express my undying gratitude and appreciation to my co-author of this study, Katherine Corvi. If it were not for all of the many tireless days, nights, and weekends spent in the Psychology Department working together on this project and the other LQTS research, this project would still be a lingering idea. Your support, encouragement, and friendship has meant so much, and together we made it!

There are too many friends and family members that have been supportive, encouraging, patient, and loving to name them all in this section. I would like to especially thank my grandparents, Michael and Sandi Forese; my aunt, Michelle Dotson; my parents, Norb and Anita Waldron; and my brother, Vincent Waldron for their never-ending support as I follow my dreams and pursue higher education. A special note of thanks and appreciation to my mother for her ever critical eye as she has read over countless drafts of this dissertation and other scholarly work throughout my entire graduate career. I would also like to express my appreciation to my in-laws, Nina Phelps,

Eric D. Phelps, Janine Phelps, and Amy Phelps, for their continued support, encouragement, and inspiration.

I would like to thank my husband and best friend, Eric Phelps. I could not have done this without your never-ending support and encouragement to follow my dreams, never give up, and to stay strong no matter what challenges we faced together. I do not have the words to appropriately thank you for the love, support, and sacrifices you have made for me throughout this journey that we have been on together. I'm one step closer to having more degrees than you.

Lastly, I would like to thank all of the subjects who participated in this study and all of the other LQTS studies and have helped give personal meaning to this research.

Abstract

Long-QT Syndrome (LQTS) is an inherited cardiac condition that predisposes individuals to cardiac arrhythmias and is a potentially fatal disorder that affects approximately 1 in 2,000 people. The triggers are difficult to avoid and may cause children and their families to make major life changes to avoid scenarios that can precipitate cardiac events. Parents may become more aware of the risks and may be hypervigilant of their child's surroundings and exposure to potential triggers. Social problem-solving skills have been shown to enhance the ability to cope with both minor and major daily stressors and minimize psychological problems associated with physical health problems (Nezu & Nezu, 2012). This study was part of a larger study that included both children diagnosed with LQTS and their parents. Parents/caregivers participated in a workshop to learn problem-solving skills and coach their children to learn these skills. Results indicated that overall evaluations of the workshop by participants were positive (93.67% satisfaction rating), and the workshop was both feasible and effective. Participants demonstrated overall increases in adaptive problem-solving skills, decreases in maladaptive problem-solving skills, increases in coping abilities and hope, and decreases in worry, which were maintained at 1-month follow-up and 3-month follow-up. Parental inclusion is believed to have helped facilitate children's short-term gains on outcome measures. Further investigation of utilization of this workshop as a tool to help children and their parents better cope with LQTS-related stressors is needed.

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CHAPTER 1

Introduction

Statement of the problem.

Chronic illnesses have been defined as illnesses that last or must be predicted to last for at least one year, require ongoing medical attention, and limit the individual in activities of daily living. Some illnesses can linger for years or a lifetime. Chronic illnesses affect the physical, social, emotional, intellectual, vocational, or spiritual functioning of the diagnosed individual (Myers, 2011). It has been suggested that 30% of children under the age of 18 have a chronic illness (Weis, 2007), and approximately 6.5% of these children have a disabling chronic condition (Tak & McCubbin, 2002). Children are now surviving and living with chronic illnesses for long periods, which can place considerable burden on the family to care for the child.

Long QT Syndrome (LQTS) is an example of a chronic illness that affects children and their families. It is an inherited or mutated genetic defect within the muscle cell structure of the heart, specifically within the ion channels (Moss, 2009). The genetic mutation causes the ion channels to abnormally close or open, resulting in abnormal recharging of the heart; this causes instability of the electrical system of the heart leading to arrhythmias, abnormal heart rhythms in the heart (Goldenberg & Moss, 2008; Moss, 2009). In LQTS, the recharging or repolarization phase of the heart is prolonged resulting in a prolonged QT interval (Goldenberg & Moss, 2008; Moss, 2009). The prolonged QT interval predisposes individuals to a potentially fatal rapid heart rhythm

known as torsade de pointes (TdP) (Goldenberg & Moss, 2008), which can result in syncope and cardiac arrest (Ashworth, Levsky, Marley, & Kang, 2005).

LQTS is estimated to occur in 1 in 2000 individuals (Schwartz *et al.*, 2009).

LQTS is a familial, genetic disorder in 90% of cases, while the other 10% are without any familial history (Schwartz *et al.*, 2009). LQTS is a chronic condition with no obvious symptoms in 50% of cases until a serious incident occurs, such as syncope, TdP, sudden cardiac arrest (SCA), or sudden cardiac death (SCD) (Ashworth *et al.*, 2005). Cardiac events may be triggered by physical exertion (e.g., swimming, running), startle (e.g., alarm clock, school bell, ringing phone), emotions (e.g., fear, fright, anger, crying, stressful situations), or simply during sleep (Moss, 2009; Modell & Lehmann, 2006). These types of triggers are difficult to avoid, and may consequently cause children and their families to make major life changes in attempts to avoid scenarios that can precipitate cardiac events. As parents become more aware of the risks of SCD in their children diagnosed with LQTS, they may be hypervigilant of their children's surroundings (Gonzales, 2009). This hypervigilance can lead parents to feel more anxious about their ability to minimize their children's exposure to the potentially fatal triggers (Gonzales, 2009).

Parents' adaptation and adjustment may vary by chronic illness depending on its nature and course, their knowledge of the condition, and the suddenness of the onset (Dodgson *et al.*, 2000). As has been seen in parents of children diagnosed with other chronic diseases, the way a parent reacts to and handles the diagnosis of a chronic illness

can affect the parent, the child diagnosed, and the entire family. Parents of children with chronic illnesses experience increased emotional distress compared to parents of non-ill children, especially around the time of diagnosis (Barlow & Ellard, 2006). Long-term studies (i.e., at least 5 years post diagnosis) have indicated that parents experience levels of emotional disturbances similar to healthy controls, but continue to experience loneliness, uncertainty, fear of relapse, worry about children's future, and symptoms of post-traumatic stress (Grootenhuis & Last, 1997). Parents' ability to cope effectively with the diagnosis of their child with LQTS can depend on their perceived self-efficacy (Steffen, McKibbin, Zeiss, Gallagher-Thompson, & Bandura, 2002). The self-efficacy construct, in relation to caring for an individual who is diagnosed with a chronic illness, includes ability to self-care and obtain respite when needed, respond to care-related problems, and control upsetting thoughts and negative feelings activated by the caregiving activities that have been entrusted to the caregiver (Marquez-Gonzalez, Losada, Lopez, & Penacoba, 2009; Steffen *et al.*, 2002).

Mothers of children diagnosed with LQTS report that they experience problems coping with the diagnosis, fear and distress, guilt, sadness, loss, and challenges of attempting to keep their children as safe and healthy as possible (Gonzales, 2009). Children diagnosed with LQTS face daily challenges related to their diagnosis. These include fatigue, a medication regimen, restriction from activities with their friends, and feeling the need to explain possible effects from medical complications. Similarly, parents of children diagnosed with LQTS face daily challenges, such as to whom to

disclose the diagnosis, what activities to allow their children to participate in, and ensuring that their children are compliant with their medication regimen, to name a few. Research is needed to provide clinicians more options to assist parents of children diagnosed with LQTS in order to obtain skills to cope with the problems and challenges they face.

Psychosocial interventions.

The incorporation of a family member in psychosocial treatment for patients with chronic illnesses has been shown to positively impact health behaviors and emotional well-being of the patient and decrease symptomatology. Psychosocial treatment provides overall positive effects for the patient diagnosed with a chronic illness, and family members have also had an increase in empathy and supportiveness for the patients when included in the psychosocial treatment (Martire & Schulz, 2007).

One such psychosocial intervention that can be used to teach parents of children diagnosed with LQTS how to cope more effectively with the diagnosis and life challenges is Social Problem-Solving (SPS) Therapy. SPS is the process an individual takes to identify and carry out effective solutions to problems as they occur in natural environments (D'Zurilla & Nezu, 1982). SPS focuses on increasing the factors that produce adaptive functioning in real-life social environments (D'Zurilla, Nezu, & Maydeu-Olivares, 2004). Interventions that include SPS techniques attempt to teach participants to identify a problem, identify solutions to the particular problem, choose the best solution, and determine ways to implement the solution (D'Zurilla & Nezu, 1982).

Individuals who utilize constructive problem-solving skills and have positive problem orientation have better problem outcomes than individuals who have dysfunctional problem-solving skills and a negative problem orientation (D’Zurilla *et al.*, 2004). SPS focuses on attempting to help individuals learn to effectively and efficiently solve problems, believe in their own ability to solve problems, and feel less threatened when confronted with difficult problems.

Purpose of study.

LQTS is a chronic illness that affects the psychosocial well-being of the individual diagnosed with the chronic illness and also their family and caregivers. Parents of children diagnosed with a chronic illness face a number of psychosocial concerns. Helping parents to learn to cope with psychosocial concerns, decrease their worry, and increase their hope around their children’s chronic illness may be beneficial to the child, the parent, and the entire family. The present study evaluated the impact on the parents of a group intervention focused on SPS skills directed toward children diagnosed with LQTS, as part of a larger study. The inclusion of parents in a SPS intervention can help them to recognize that they are helping their child learn specific techniques to cope with their diagnosis of LQTS and make them more independent to positively handle LQTS related issues. In particular, this study evaluated whether the problem-solving workshop increased parents’ hope, coping skills, and SPS skills, and decreased parents’ worry about how their children will effectively handle LQTS related problems.

CHAPTER 2

Literature Review

Chronic illness.

Chronic illness affects the well-being of the individual. However, definitions have varied in the literature from “a chronic condition if that person’s condition had lasted or was expected to last 12 or more months and resulted in functional limitations and/or the need for ongoing medical care” (Hwang, Weller, Ireys, & Anderson, 2001, p. 288) to “a chronic disease or condition that has 1 or more of the following characteristics: is permanent; leaves residual disability; is caused by nonreversible pathological alteration; requires special training of the patient for rehabilitation; or may be expected to require a long period of supervision, observation, or care” (Bernstein *et al.*, 2003, p. 128). A majority of definitions add that the illness must last or be predicted to last for at least 1 year, require ongoing medical attention, and limit the individual’s activities of daily living. Chronic illnesses affect the physical, social, emotional, intellectual, vocational, or spiritual functioning of the diagnosed individual (Myers, 2011). Sometimes, there is no cure for a chronic illness.

Chronic illnesses that are most often diagnosed within the child and adolescent developmental periods include asthma, cerebral palsy, congenital heart disease, diabetes, and leukemia (Miceli, Rowland, & Whitman, 1999). The prevalence of childhood chronic illness varies from 0.22% to 44%, depending on the research design, type of chronic illness being investigated, and the definition of *chronic illness* that was used (van der Lee, Mokkink, Grootenhuis, Heymans, & Offringa, 2007). On average, it is reported

that about 30% of children under the age of 18 have a chronic illness (Weis, 2007), and approximately 6.5% of these children have a disabling chronic condition (Tak & McCubbin, 2002). Higher prevalence rates of chronic illness were reported for older children than younger children (van der Lee *et al.*, 2007).

A chronic illness may place considerable burden on the family in order to care for the child. Familial burdens to caring for a child diagnosed with a chronic illness include a variety of medical, developmental, social, emotional, and environmental needs and issues (Grey & Sullivan-Bolyai, 1999). Individuals diagnosed with a chronic illness are charged with managing the disease and its symptoms in the absence of a cure; these tasks become those of the parents when children are diagnosed with the chronic illness. The family also plays a pivotal role in the child's adaptation and adjustment to the illness (Knafl, Breitmayer, Gallo, & Zoeller, 1996).

Parents and chronic illness in children.

Caring for chronically ill children can be a source of significant stress for parents (Lewin *et al.*, 2005). Children's chronic illness is demanding for all family members, especially as it generally changes family life and requires parents to adapt to the medical needs of their children (Hentinen & Kyngras, 1998). Parents of children diagnosed with a chronic illness may experience a variety of feelings, including guilt, anxiety, shock, denial, confusion, anger, and depression (Hentinen & Kyngras, 1998; Melnyk, Feinstein, Moldenhouer, & Small, 2001). When children are diagnosed with a chronic illness, the parents must take on a long-term caregiver role in addition to the parenting role.

Caregiving has been described as an unexpected career, in which the caregiver moves

through a series of stages, each of which require adaptation and restructuring of responsibilities over time (Aneshensel, Pearlin, Mullan, Zarit, & Whitlatch, 1995).

Caregiving.

Caregiving for patients with chronic illnesses often has a negative impact on the caregiver's health, daily schedule, anxiety, and energy (Aranda & Hayman-White, 2001). As the needs of patients increase and there is an increased level of dependence upon caregivers, caregivers' ability to function is negatively affected (Given *et al.*, 1993). The caring role can have an impact on caregivers' life and social functioning. Compounding the challenges of providing regular care, caregivers without a reliable source of social support may feel abandoned by family members and friends who are not involved in the caring process and who do not understand the unique stress and changes in life with which the caregivers are dealing (Kurtz, Kurtz, Given, & Given, 2004). On the whole, caregivers report higher levels of psychological distress and lower levels of psychological well-being compared to the general population (Ross, Mosher, Ronis-Tobin, Hermele, & Ostroff, 2010). Depending on the type of illness, the onset of caregiving can occur suddenly. The sudden onset of a chronic illness may, therefore, cause heightened stress responses for caregivers (Sherwood *et al.*, 2008), which may also be compounded by the life-threatening nature of some chronic illnesses, such as LQTS.

Psychosocial factors.

Parents of children diagnosed with chronic illnesses take on an informal caregiving role. This role typically spans the course of the child's lives, from the time of diagnosis until children reach an age at which they can care for themselves (Raina *et al.*, 2004). As the informal caregivers, parents are responsible for providing the long-term

care that likely requires extraordinary physical, emotional, social, and financial resources (Lauver, 2008). They are also responsible for coordinating children's medical, educational, and developmental needs, while also balancing competing family demands (Silver, Westbrook, & Stein, 1998). Research indicates that lifelong informal caregiving for children with a chronic illness is associated with poor emotional and physical health of the caregivers. Long-term outcomes of informal caregiving have not been evaluated for parents of children diagnosed with LQTS.

Feelings of hopelessness and depression are very common among caregivers. Caregiver depression is the affective disturbance of the caregiver due to the stress of the caregiving process (Given *et al.*, 2004). Symptoms can include both the symptoms of clinical depression and increased symptoms of anticipatory grief and worry associated with patients' diagnosis, prognosis, and treatment often not seen in clinical depression (Haley, LaMonde, Han, Burton, & Schonwetter, 2003). Depression is more common when caregivers experience less self-efficacy or self-satisfaction in their roles as caregivers (Folkman & Moskowitz, 2000). The caregivers who do not feel competent in their caregiving abilities experience higher levels of depression (Nijboer, Tempelaar, Triemstra, van den Bos, & Sanderma, 2001). It has been predicted that between 32% and 50% of older adult caregivers experience depressive symptoms (Butler, Turner, Kaye, Ruffin, & Downey, 2005). The negative emotional aspects of caregiving can further decrease the quality of life of the caregiver and inadvertently affect the care the patient receives, although caregivers who report having better physical health often report having lower levels of depression (Haley *et al.*, 2003). Caregivers of children diagnosed with LQTS may experience many of these psychosocial issues and emotional

disturbances surrounding the caregiving of their children, due to the unpredictable nature of LQTS and the lack of an infrastructure of support available to parents whose children are diagnosed with LQTS.

Parents of children diagnosed with chronic illnesses face unique obstacles that are often different from those for parents of children not diagnosed with a chronic illness. Parents who are in the caregiving role for a chronically ill child may face problems as they learn to deal with their children's health and cope with the stress (Tew, Landreth, Joiner, & Solt, 2002). This may also be true of parents of children diagnosed with LQTS, although to date this not been researched. Parents report more parenting stress, less social support, increased strain on the role of parenting, and spending significantly more time with care-taking tasks compared to parents of healthy children. Parents of chronically ill children face a number of stressors at all stages of the chronic illness, including the time of diagnosis, the developmental transitions, continual stressors related to the ongoing health care needs of the children, and as the children experience illness exacerbations and hospitalizations (Melnik *et al.*, 2001). Parents of children who are diagnosed with a life-threatening illness, compared to a non-life-threatening illness, behave and adapt to the diagnosis differently (Chesler & Barbarin, 1987; Sterken, 1996). The constant fear of potential death tends to make it difficult for the family to function normally when children are diagnosed with life-threatening chronic illnesses (Sloper, 2000). The fear of potential death has been found to be extremely prevalent for families of children diagnosed with LQTS (Farnsworth, Fosyth, Haglund, & Ackerman, 2006; Gonzales, 2009; Rovinsky, 2010).

Lack of research on psychosocial interventions.

The incorporation of a family member to the psychosocial treatment for patients with chronic illnesses has been shown to positively impact the health behaviors and emotional well-being of the patient and decrease symptomatology (Martire & Schulz, 2007). Not only does psychosocial treatment provide overall positive effects for the patient diagnosed with a chronic illness, but the family members also had an increase in empathy and support for the patient (Martire & Schulz, 2007). Research has shown that rigorous, evidence-based interventions are lacking for families of children diagnosed with chronic illnesses (Anderson & Davis, 2011). Most of the studies that have been conducted with this population are descriptive studies, case studies, and reports of expert committees, or well-designed controlled studies, without randomization. These studies have also primarily focused on disease-specific interventions, none of which have been conducted on families who have children diagnosed with LQTS. While many of these studies report and describe relevant techniques for intervening with families of children diagnosed with a chronic illness, the interventions have not been empirically tested and are not considered evidence based (Anderson & Davis, 2011). This is unfortunate for a population in which the needs and stressors have been consistently identified, such as LQTS. Parents of children diagnosed with LQTS may be challenged to ensure their children understand their disease, including the need to carry an automated external defibrillator (AED), or that they take their medication every day. There is a notable scarcity of therapeutic interventions that focus on developing coping skills within the family (Anderson & Davis, 2011).

The parental caregiving role is an extremely important one, especially when the child has been diagnosed with a chronic illness. These parents often take on dual roles of parenting and informal caregiving for their children, which can cause psychological distress. Lifelong informal caregiving for children with a chronic illness is associated with poor emotional and physical health in the parents. The few well-designed controlled studies that exist have found that families of children diagnosed with a chronic illness could benefit from interventions, but research is lacking with randomized studies to test the effectiveness of these interventions.

Long QT Syndrome.

Long QT Syndrome (LQTS) is an inherited chronic cardiac condition, resulting from genetic mutations within the muscle cells of the heart, causing problems with the ion channels (Liu *et al.*, 2011; Moss, 2009). The abnormalities with the malfunction of the ion channels cause an inappropriate electrical charge to be generated. This results in the electrical system of the heart becoming erratic, generating an arrhythmia. With LQTS, the recharging or repolarization phase of the heart beat is prolonged resulting in prolongation of the QT interval on the electroencephalogram (EEG; Goldenberg & Moss, 2008; Moss, 2009; QTsyndrome.ch, 2006). As a result of the prolongation of the QT interval, a rapid heart rhythm known as torsade de pointes (TdP) can occur. TdP can result in syncope and cardiac arrest (Ashworth *et al.*, 2005). The actual prevalence of LQTS is unknown, but it is suspected to be a common cause of sudden and unexplained death in children and youth (QTsyndrome.ch, 2006).

LQTS is a somewhat rare clinical disorder, occurring in an estimated 1:2000 individuals (Schwartz *et al.*, 2009). It is estimated that 50,000 individual in the United

States are affected with LQTS, which results in 3,000 deaths annually (QTsyndrome.ch, 2006). It may be more common, but may be underdiagnosed or misdiagnosed in many cases. However, among the genetic cardiac arrhythmia syndromes, LQTS is considered to be one of the most common (Roden, 2008). LQTS is familial in 90% of the cases (Saenen & Vrints, 2008). Within families with the inherited form of LQTS, some individuals may carry more than a single gene mutation (Schwartz *et al.*, 2001).

There are hundreds of different mutations in 13 genes associated with LQTS that have been identified (Roden, 2008). Of these, eight of the genes code for specific ion channel (sodium, potassium, or calcium) mutations (Bokil, Baisden, Radford, & Summers, 2010). The most common mutations that cause LQTS result from potassium or sodium abnormalities, which result in the reduction of the action potential (Collins & van Hare, 2006). The flow of ions during the action potential affects the QT interval of the ECG. Certain variations of the sodium-channel mutations cause the sodium-potassium channels to malfunction, thereby resulting in a continued leakage or blockage of ions and a prolonged action potential (Goldenberg & Moss, 2008). Based on the genetic mutations, different subtypes indicate in which gene the mutation occurs (Collins & van Hare, 2006).

The inherited mutated genes indicate the particular type of LQTS with which an individual may be diagnosed (Saenen & Vrints, 2008). LQT1, LQT2, and LQT3 are the most commonly diagnosed forms of LQTS (Bokil *et al.*, 2010). The gene KCNQ1 (an encoded potassium channel mutation) is the gene responsible for LQT1 (Collins & van Hare, 2006). 30% to 35% of individuals diagnosed with LQTS are diagnosed with LQT1, and cardiac events for these individuals most often occur during exercise or

emotional arousal (Saenen & Vrints, 2008). KCHN2 (an encoded potassium channel mutation) is the gene responsible for LQT2 (Collins & van Hare, 2006). Thirty percent to thirty-five percent of individuals are diagnosed with LQT type 2. Cardiac events for individuals diagnosed with LQT2 most often occur due to an auditory stimulus, such as a loud noise or alarm (Saenen & Vrints, 2008) or emotional stress (Moss, 2009, QTSyndrome, ch, 2006). The gene SCN5A (an encoded sodium channel mutation) is responsible for LQT3 (Collins & van Hare, 2006). Only 5% to 10% of individuals with LQTS are diagnosed with LQT3. Arrhythmias and cardiac events associated with LQT3 most often occur during sleep.

LQTS manifests as a number of symptoms, which can include palpitations of the heart, syncope, seizures, SCA, and SCD. Individuals are usually diagnosed in childhood between the school-age years and young adulthood (Liu *et al.*, 2011), with the majority of individuals experiencing their first symptom prior to the age of 15 (Wedekind *et al.*, 2009). Symptoms do not occur every day in individuals diagnosed with LQTS. Symptoms often manifest during a triggering event (Wedekind *et al.*, 2009), which depends on the type of LQTS. Most of the time, individuals are diagnosed either because the individual was symptomatic or they had a family member who was symptomatic (Goldenberg & Moss, 2008; Liu *et al.*, 2011). Half of the time, SCA is the first symptom. Diagnosis of LQTS involves the evaluation of an electrocardiogram (ECG) by a cardiologist and a detailed family history (Vincent, 2003). Genetic testing is often involved in the diagnosis of LQTS, which can help to identify distinct types of LQTS gene mutations and genetic polymorphisms that may exist among individuals and families. The genetic testing can be extremely helpful in the overall treatment planning

for LQTS, as individuals diagnosed with different genetic variations of LQTS need different treatment plans (Shmimizu, 2005).

Treatment of LQTS can be very effective in managing the symptoms and preventing sudden death in patients diagnosed with LQTS. One of the main treatment options available for patients with LQTS is beta-blockers, which lower heart rates and blood pressure (Kubon *et al.*, 2011). A second modality used to treat abnormally fast heart rhythms is an implantable cardioverter-defibrillator (ICD; Collins & van Hare, 2006). This treatment is recommended for patients who experience recurrent syncope despite pharmacological treatments (Kwon *et al.*, 2012). ICDs have been shown to be highly effective for high-risk patients diagnosed with LQTS (Goldenberg & Moss, 2008). ICDs treat ventricular fibrillations (abnormal heart rhythms) from occurring through electrical defibrillation (Ganz, Olshansky, & Downey, 2012). The defibrillation is an electrical shock to the heart that corrects the abnormal heartbeats. The electrodes attached to the ICD and the heart record the heart rhythms. If the electrodes detect an abnormal heart rhythm, the ICD delivers a shock to the heart to restore the normal heartbeat (Ganz *et al.*, 2012).

Another treatment modality for LQTS is a pacemaker. Pacing is a treatment that can be used for infants and younger children instead of ICD implantation (Saenen & Vrints, 2008). Pacemakers are devices that are implanted under the skin and treat abnormal heart rhythms. They differ from ICDs because they are typically used to treat individuals with slow heartbeats. In the same way that the ICD works, pacemakers send electrical impulses the heart when they detect abnormal heart rhythms, causing the heart to beat at a normal rate. Pacemakers utilize pulse generators and leads that are connected

to the heart to determine how the heart is currently functioning and determine when to issue an impulse to the heart to maintain a normal heart rate and rhythm (Martin & Villalba, 2012). Pacemakers have been often used for the management of recurrent syncopal symptoms and cardiac arrest events, and have been shown to be an effective treatment modality for high-risk LQTS patients (Edlar *et al.*, 1987; Moss *et al.*, 1991).

Regardless of the type of treatment used to manage LQTS, most individuals diagnosed with LQTS have physical activity and recreational sport restrictions recommended by their cardiologists and/or electrophysiologists. As part of the Bethesda Conference on Eligibility Recommendations for Competitive Athletes with Cardiovascular Activities (2006), individuals with LQTS should be restricted from activities based on their presenting symptoms, previous history of LQTS-related events, family history, and presence of an ICD or pacemaker (Zipes *et al.*, 2005). For most types of LQTS, syncope, SCA, or SCD have been shown to be adrenergically mediated, and restrictions on athletic activities were previously recommended (Schwartz *et al.*, 2001). These restrictions were created out of concern for accelerated heartbeats and increased risk of LQTS-related events during these activities (Collins & van Hare, 2006). However, new recommendations have eased restrictions for some individuals, allowing those who are asymptomatic and those who are treatment compliant to participate in more physical activities than previously allowed (Ackerman, Zipes, Kovacs, & Maron, 2015). The literature indicates that swimming is a trigger for LQTS, especially for LQTS1, and should be avoided to some degree, at least competitively. Other types of restrictions are based on the exact genotype of LQTS, including things that could trigger a LQTS event (alarms, emotional stress, etc.; Roden, 2008). Individuals diagnosed with

LQTS are also often advised to avoid foods, over-the-counter medications, and prescription medications that may increase heart rate or interact with beta-blockers and to limit heat exposure, as these may place them at higher risk for a cardiac event (Shimizu, 2005).

Quality of life (QoL) in children diagnosed with LQTS was found to be lower than in children with other cardiac illnesses, including congenital complete heart block, bicuspid aortic valve, supraventricular tachycardia, and ventricular tachycardia (Czosek *et al.*, 2015). Children who had been diagnosed with LQTS and had a cardiac implantable device had lower QoL. Recent research has identified that expanding and lifting restrictions on activities for patients diagnosed with LQTS may be beneficial in increasing patients' QoL (Johnson & Ackerman, 2013; Lampert *et al.*, 2013).

Clinical presentations for individuals diagnosed with LQTS can vary from being asymptomatic during the entire course of an individual's life to sudden death as the first symptom or initial event. The variability and uncertainty causes great distress for those who are diagnosed with the condition and their family members. This drastic clinical spectrum of presentation most likely arouses fear and uncertainty for parents of children diagnosed with LQTS (Farnsworth *et al.*, 2006). It is often difficult for parents and children to avoid all of the specific triggers. In their qualitative study on parents' perceptions of LQTS, Farnsworth and colleagues (2006) described that parents are often fearful of their children's death, and many parents take actions to alleviate their fears. These actions include making lifestyle changes within the home and the community. Some parents provided their children with cell phones, used a baby monitor in the child's room at night to hear if they were still breathing, carried a defibrillator with their child,

and informed schools of the condition including putting a plan into place should an event occur at school. Parents also often engaged in health management behaviors that they taught their children, including listening to your body, understanding the seriousness of the illness, becoming informed of all medical information, and advocating for themselves (Farnsworth *et al.*, 2006).

Psychological health factors.

There are many different constructs that are evaluated when examining the role of cognitive appraisal in health-related behavior. These constructs can be evaluated in both patients diagnosed with a chronic illness, as well as their parents. Included among these factors are perceptions of worry, hope, and coping. These concepts have been widely researched as perceived to play an important role in the caregiving process for children diagnosed with a chronic condition. However, not all of these factors have been specifically examined in parents of children diagnosed with LQTS.

Worry.

Worry is a state of anxiety or uncertainty over a situation. Worrying can actually be a helpful tool in recognizing that action is needed to rectify a particular situation. However, worry can also be harmful when it inhibits a person from actually taking steps toward correcting a stressful, uncertain situation. Worry is the cognitive component of anxiety, the “thoughts and images that relate to possible negative or threatening outcomes” (Silverman, La Greca, & Wasserstein, 1995, p. 671). As a more cognitive process, the conceptual activity of worrying maintains the worry state in individuals (Borkovec & Inz, 1990).

When a child has been diagnosed with a chronic illness, many of these cognitive worries for the parents will focus on concerns related to the child's future well-being (Barlow & Ellard, 2006), sustained uncertainty surrounding the disease (Cohen, 1995), vulnerability of their child (Anthony, Gil, & Schanberg, 2003), susceptibility to illness (Spurrier *et al.*, 2000), and the day-to-day demands and long-term nature of the chronic illness (DeVet & Ireys, 1998). The amount of stress and worry that parents report when their child has been diagnosed with a chronic illness is quite variable (Hauenstein, 1990; Silver, Bauman, & Ireys, 1995). There are familiar features of worrying for families with children diagnosed with a potentially fatal illness (Garwick, Patterson, Meschke, Bennett, & Blum, 2002; Maclean, 1999; Woodgate & Degner, 2001).

Often for parents of children who have been diagnosed with a life-threatening illness, such as LQTS, worries are associated with not knowing or the uncertainty that of possible symptomatic episodes (Farnsworth *et al.*, 2006; Anderson, Oyen, Bjorvatn, & Gjengedal, 2005). Many of the worries of parents of LQTS patients include worry surrounding the condition, lack of knowledge of what LQTS is, attempting to understand why the condition occurred, lack of understanding about the cardiac events that had happened previously (either to the child, themselves, or another family member diagnosed with LQTS), uncertainty about events that *could* happen in the future, and decisions regarding their children's best treatment options (Anderson *et al.*, 2005; Farnsworth *et al.*, 2006). It has previously been noted that once parents of children diagnosed with LQTS become more knowledgeable and are introduced to ways of coping, the worry and uncertainty is less pervasive than when they did not have as much knowledge or coping abilities (Farnsworth *et al.*, 2006).

Hope.

Hope has been defined differently throughout the course of history. Typical dictionary definitions of the word *hope* emphasize a perception that something that is desired may happen. In the 1950s, 1960s, and 1970s, hope was often described as overall perceptions that an individual's goals can be met (Erickson, Post, & Paige, 1975; Gottschalk, 1974; Menninger, 1959). This conceptualization assumes that an individual's cognitions surrounding the goal-directed activities play an important role in the subsequent attainment of positive outcomes for the particular situation with which the individual is faced. However, these definitions lack explanation of how the goals are pursued and what impact that has on the overall hope of the individual toward attaining his or her goals. Over time, these conceptualizations have been expanded to further explain the different cognitive processes. Goal-directed thinking contains two interrelated components, *agency* and *pathway* (Snyder *et al.*, 1991). Agency is the sense of successful determination to meet goals in the past, present, and future. Pathway is the sense of being able to generate successful plans in order to meet the goals. The new definition of hope includes both the individual's "successful agency (goal-directed determination)" and "pathways (planning of ways to meet the goals)" (Snyder *et al.*, 1991, p. 571).

Hope theory views hope as a goal-directed, positive motivational state that is based on the interactions derived from agency and pathway planning (Snyder, 1994, 2000). The theory behind hope can be divided into four categories: goals, pathway thoughts, agency thoughts, and barriers (Snyder, 2000). Goals, within the theory of hope, are the anchor that provides a direction and an endpoint for the hopeful thinking (Snyder,

2000). The pathway and agency thoughts are the particular routes taken to achieve the goal and the motivation to undertake the routes towards the goal, respectively. Barriers, on the other hand, are the things (both physical and mental) that can block or hinder attainment of a goal (Snyder, 2000). The attainment of goals has been found to be associated with positive emotions (Snyder *et al.*, 1996), while barriers, or the blockage of goals, has been found to be related to more negative emotions (Diener, 1984). However, this may not always be the case. Individuals who are rated as having high hope view barriers as challenges to overcome and utilize pathway thoughts to plan alternative routes when they encounter barriers to their goals, while also utilizing agency thoughts to keep the goal in sight and provide motivation to generative alternatives (Lopez, Snyder, & Pedrotti, 2004; Snyder, 1994).

Hope can be an influential factor in how individuals deal with problems. In terms of medical conditions, hope can be an important factor in how an individual diagnosed with the chronic condition and his or her caregiver interprets the illness. Hope theorists maintain that even among patients whose diagnosis and situation appear to be negative, individuals who embrace a high degree of hope are more interested in looking for ways to attain their goals, rather than feeling defeated over the diagnosis (Lopez *et al.*, 2004). It is especially important for caregivers to have high levels of hope to encourage the individual diagnosed with a chronic illness to continue “the fight”. For parental caregivers of children diagnosed with LQTS, it is likely that the presence of hope is a significant factor in determining parents’ problem-solving style and how they deal with LQTS-related problems that occur. Parents with high hope, including higher levels of agency thoughts and pathway thoughts, may be able to view problems they face related to

LQTS as an attainable challenge, rather than an obstacle, and will tend to find more efficient ways to solve the problems.

Coping.

In addition to worry and hope, clinical research has also focused on one's ability to cope. Coping is defined as "constantly changing cognitive and behavioral efforts to manage specific external and internal demands that are appraised as taxing or exceeding the resources of the person" (Lazarus & Folkman, 1984, p. 141). The functions of coping efforts are to regulate the emotional responses to the situation and to solve the problems of the situation (Lazarus & Folkman, 1984; Moos & Schaefer, 1986; Rutter, 1981). Coping does not represent a homogenous concept, but instead is a diffuse term that is used to describe a number of strategies that a person can engage in to help deal with the internal and external demands of a situation (Schwarzer & Schwarzer, 1996). Therefore, coping is often described as the specific strategies, tactics, responses, cognitions, or behaviors that an individual engages in when dealing with the situation. Coping is a combination of an individual's primary perception of the situation in combination with past experiences and acquired skills (Lazarus, 2000). This process is flexible and evolves over time, as the individual's life experiences alter his or her appraisal of situations (Lazarus, 2000). Coping can be an important mediator between illness and psychological well-being.

The manner of coping in which one engages has been linked to various health outcomes. The fact that the process of coping is dependent on how individuals appraise a situation means that the experience of the event is largely dependent upon how that individual thinks, rather than the particulars of the situation. Individuals may have more

control over their experiences than they may exercise in a given situation. Being able to cope effectively with a variety of difficulties in life is an important skill for people to obtain, and coping effectively may be a marker of psychological health. Maladaptive coping styles, on the other hand, may lead to breakdowns of psychological functioning and contribute to symptoms of stress, anxiety, and depression (Rippeto & Rogers, 1987).

There are a number of identifiable characteristics associated with coping with everyday life stressors, and there is a relationship between the ability to control the stressor and ability to cope effectively with the stressor (Aldwin & Yancura, 2004). The benefits of coping with daily stressors vary across time. Using avoidance to cope with a situation may initially improve psychological distress, but over time it may engender greater distress if a significant problem remains without resolution (Aldwin & Yancura, 2004). Coping with a chronic medical condition can be viewed as dealing with daily hassles or stressors, in contrast to a short, more traumatic stressor.

Research on coping with chronic illnesses, in general, has yielded various findings. Research conducted in the late 1960s reported evidence of maladaptive psychological functioning among persons diagnosed with childhood chronic illness (Cadman, Boyle, Szatmari, & Offord, 1987; Pless & Roghmann, 1971), although more recent findings have concluded that the impact of a childhood chronic illness can be significant (Barlow & Ellard, 2006; Sharpe & Rossiter, 2002). The majority of children diagnosed with a childhood chronic illness and their families respond adaptively and learn to adjust and cope well (Thompson & Raezer, 1998).

Barlow and Ellard (2006) suggest that parents and siblings of children diagnosed with a chronic illness may have psychosocial needs related to the presence of a chronic

medical condition within the family system. Most children diagnosed with a chronic illness did not have any clinical symptoms; they were mildly at risk for psychological distress (Barlow & Ellard, 2006). Parents of children diagnosed with a chronic illness often report more negative effects than their children regarding symptoms of distress (Sharpe & Rossiter, 2002). For example, families of children diagnosed with cystic fibrosis, another chronic childhood illness, were found to need to balance the extensive care of the child diagnosed with cystic fibrosis with personal investments in themselves as individuals, the family as a unit, and in increasing their overall understanding of the health care situation in order to cope effectively (McCubbin, Bowers, & Holaday, 1984). The evidence base needs to be extended regarding the psychosocial needs of families coping with various childhood chronic illnesses (Barlow & Ellard, 2006). Effective coping or being able to deal with the stressors related to the diagnosis of a chronic medical condition can help both the child and the parent to better deal with and resolve the stressors they confront due to the medical condition and achieve higher psychological functioning.

Parents of children diagnosed with LQTS have a number of factors related to the diagnosis with which to cope. Parents with higher hope and greater ability to cope and decreased worry may be able to effectively solve some problems and challenges with fewer psychological issues during the course of the chronic illness. Parents of children diagnosed with chronic illness who have lower self-efficacy, less hope, and fewer coping abilities may experience more challenges as they deal with the requirements of caring for a child diagnosed with a chronic illness, especially the needs of a child who has been diagnosed with a life-threatening illness such as LQTS.

Parents and LQTS.

There is very limited research on parenting a child who has been diagnosed with LQTS. The challenges and struggles that parents of children diagnosed with other chronic illnesses face can be helpful in drawing parallels to some of the same issues that parents of children with LQTS might encounter. Children diagnosed with LQTS are at risk for illness and life-threatening conditions; however, they are often not technically “sick”. Children diagnosed with LQTS are classified as having a chronic illness because of the life-long nature and implications of their condition. In general, distress for parents of chronically ill children may come from hospitalizations, pain, treatment, and restricted activity (Sallfors & Hallberg, 2003). On a daily basis, however, parents of children diagnosed with LQTS may contend with only a few of these stressors. Parents of children with LQTS must deal with the daily anxiety and worry of the potential for a cardiac event, including sudden death. Only a few qualitative studies have examined some of the psychosocial implications of parenting a child with LQTS.

Parents of children diagnosed with LQTS often face a number of other issues that are unique. LQTS has a broad spectrum of symptomatology, ranging from no symptoms at all to sudden death. Also, medical interventions, from long medical appointments to invasive and painful treatments (such as ICD implantation or ICD lead replacement), may be necessary and cause parents stress (Hendricks *et al.*, 2005). Due to the highly heritable nature of LQTS, some families may have multiple members in the family unit diagnosed with a chronic illness that is potentially life-threatening. This makes it extremely important to be able to identify and evaluate psychosocial interventions that will be able to ease the stress of parents of children diagnosed with LQTS.

Research on the psychosocial factors related to coping with the diagnosis of LQTS has found that parents experience psychological distress. In one study, 50% of parents who were told that their child tested positive for a LQTS mutation had high levels of clinical symptoms of distress (Hendricks *et al.*, 2005). After 18 months, 30% of the parents continued to indicate high levels of distress related to the diagnosis of LQTS. Parents also reported frustration with the limited knowledge of LQTS among health care practitioners (Farnsworth *et al.*, 2006). It was suggested at this time that increasing the amount of information available to parents and reports of clinical research findings would help to ease some of the distress (Hendricks *et al.*, 2005). Parents have also reported increased fears regarding their child's death more than of their own deaths after genetic confirmation of LQTS (Farnsworth *et al.*, 2006). Interventions that teach parents skills to increase their knowledge, solve their problems, and decrease their distress are needed within the LQTS population.

As mentioned previously, parents of chronically ill children face stressors at a number of different times (Melnyk *et al.*, 2001), and this is true also of mothers whose child has been diagnosed with LQTS. Gonzales (2009) conducted a qualitative study on mothers' reactions to and coping with the diagnosis of LQTS. The researcher identified different types of stressors at each time period. Mothers needed or were required to understand and comprehend the medical aspects of LQTS, a life-threatening illness, for their child. At the same time as understanding the medical aspects of the illness, mothers' were also contending with their own psychoemotional reactions to the diagnosis, which often included denial, disbelief, shock, and devastation. Over time, mothers began to cope with the new knowledge, which included emotions of fear,

distress, guilt, sadness, and loss, as well as engaging in behavioral activities to attempt to cope with and protect their children from the possibility of SCA or SCD (Gonzales, 2009).

As the mothers began to process their own emotions related to the diagnosis, they also were able to integrate the knowledge of the diagnosis of LQTS into part of their child's identity. This integration also included determining whether it was appropriate to tell other people outside of the immediate family about the diagnosis and confronting developmental issues that may be complicated by a life-threatening illness. At the same time, mothers always expressed concern about the child's future, especially as they noted being unable to monitor the child's immediate environment at all times. Mothers often expressed a sense of dread regarding the child's physical well-being and medical compliance when living independently, as the mother would not be able to have the control that she would have when the child is living with her (Gonzales, 2009). Mothers of children diagnosed with LQTS fared better when they utilized various coping skills and problem-solving abilities to deal with the diagnosis and treatment (Gonzales, 2009), suggesting that mothers (and all parents) of children diagnosed with LQTS may benefit from interventions that help to increase these skills. There has been a lack of research on the specific effects of a LQTS diagnosis on fathers and other family members.

Research regarding the psychosocial impact of LQTS is limited and warrants further investigation. LQTS is different from other more commonly studied childhood chronic illnesses in that the variation of symptoms has a considerable range, from asymptomatic to sudden death. A diagnosis of LQTS can have considerable effects on the parents of children who are diagnosed with this potentially life-threatening disease.

While research on the psychosocial effects of LQTS is beginning to emerge in the literature, there have not been any interventions published that have examined ways to help individuals diagnosed with LQTS and their parental caregivers learn to cope with the challenges of living with LQTS. One intervention that may be effective to help adolescents diagnosed with LQTS and their parental caregivers is Social Problem Solving.

Social problem solving.

There are many different types of interventions utilized by individuals diagnosed with a chronic illness and their families and caregivers to increase quality of life. Various types of psychosocial interventions have been shown to be effective to improve health and emotional well-being in individuals diagnosed with chronic illnesses and their families, including education, support groups, and cognitive-behavioral therapies (Martire & Shultz, 2007). At the current time, no formal interventions focus specifically on improving the health and emotional well-being of either individuals diagnosed with LQTS or their family members.

One specific modality of psychosocial treatment that may be effective for individuals diagnosed with LQTS and their family members is Social Problem Solving (SPS). Nezu *et al.* (1999) defined SPS as a metacognitive process by which individuals attempt to comprehend the nature of their problems in management of their daily lives while directing efforts at coping with difficult situations. SPS is demonstrated by the way in which individuals process information about the self, the environment, and the problems encountered during everyday life. These can range from problems resulting from the diagnosis of a chronic illness to the problems an adolescent faces while taking

over management of a chronic illness from his or her parents. SPS is a process by which the individual attempts to identify stressors in everyday living and attempts to implement effective and adaptive solutions (D’Zurilla & Nezu, 2007).

SPS is a process of solving problems within a natural environment situation (D’Zurilla & Nezu, 1982), with a focus on increasing factors that lead to adaptive functioning in real-life social environments (D’Zurilla *et al.*, 2004). There are a variety of problem-solving styles that have been demonstrated in individuals (D’Zurilla & Nezu, 2007). The particular type of problem-solving styles and the particular skills that an individual possesses will determine the adaptiveness and effectiveness of attempts to cope with the problem. Utilization of the wrong or an ineffective SPS style can lead to increased frustration, stress, negative mood, and maladaptive psychological and physical states (Nezu, Nezu, Freidman, Faddis, & Houts, 1998). On the other hand, individuals who have learned to draw on more adequate SPS skills are more likely to decrease and modify distress, anxiety, and other psychosocial consequences associated with the chronic conditions (Nezu, Nezu, Felgoise, McClure, & Houts, 2003). SPS plays a significant role in psychological adjustment and is an essential and important coping strategy to reduce or minimize psychological distress (D’Zurilla & Nezu, 1990).

SPS theory is a multidimensional construct composed of two different but related concepts, problem orientation and problem-solving style. Problem orientation refers to the set of stable cognitive-emotional schemas that reflect a person’s beliefs, appraisals, and feelings about problems in everyday life and ability to solve problems (D’Zurilla *et al.*, 2004). This is also believed to be an important motivational function in the application of SPS skills. D’Zurilla *et al.* (2004) indicate that there are two different

types of problem orientation, positive problem orientation (PPO) and negative problem orientation (NPO). PPO is a person's ability to assess a problem as a challenge, that is solvable, and that he or she has the ability to solve the problem. On the other hand, an individual who has a NPO is more likely to view problems as a threat to his or her well-being, doubt his or her ability to solve the problem, and become frustrated when faced with problems (D'Zurilla *et al.*, 2004).

Problem-solving styles refer to the cognitive and behavioral activities by which an individual attempts to understand problems and find effective solutions to cope with the problem (D'Zurilla *et al.*, 2004). There are three problem-solving styles: rational problem solving, impulsivity/carelessness style, and avoidance style. Rational problem solving is a constructive problem-solving style in which the individual has deliberate, systematic application of the problem solving skills. The impulsivity/carelessness style is a dysfunctional and ineffective problem-solving pattern, characterized by attempts to solve problems actively, but limited in the ability to generate solutions. Individuals with an impulsivity/carelessness style of problem solving tend to display impulsive, careless, and hurried attitudes and behaviors towards solving a problem. The avoidant style of problem-solving is also a dysfunctional and ineffective style of problem-solving, characterized by procrastination, inactivity, and withdrawal in the face of difficulties, or dependence upon someone else to deal and cope with the problem (D'Zurilla *et al.*, 2004).

The three major concepts within the SPS model are problem solving, problem, and solution. These concepts are very important in understanding the SPS model and for an individual to be able to distinguish between the concepts of *problem solving* and

solution implementation. Problem solving is the self-directed cognitive-behavioral process in which an individual attempts to identify or discover an effective solution for a specific problem (D’Zurilla *et al.*, 2004). Problem solving is an important and effortful approach in which individuals discover effective ways to cope with stressful problems they incur during everyday activities (D’Zurilla & Nezu, 2007). A problem is defined as “any life situation or task (present or anticipated) that demands a response for adaptive functioning but no effective response is immediately apparent or available to the person or people confronted with the situation because of the presence of one or more obstacles” (D’Zurilla *et al.*, 2004, p. 12). These demands can originate either from the environment or from within the person trying to solve the problem, and demands vary depending upon the particular type of problem at hand (D’Zurilla *et al.*, 2004). After assessing the problem, the solution is the situation-specific coping response or pattern that is the product of the problem-solving process. A solution is deemed to be effective when it achieves and meets the problem-solving goal while maximizing other positive consequences and minimizing negative consequences of the actions that were engaged in (D’Zurilla *et al.*, 2004).

Effective problem solving is postulated to be dependent on the effective application of rational problem-solving skills and on a positive orientation towards problem solving (problem orientation; D’Zurilla *et al.*, 2004). An individual who is an effective problem-solver is able to identify adaptive solutions, appears to have a positive problem orientation, and an effective application of problem-solving skills (Bell & D’Zurilla, 2009; Siu & Shek, 2005). On the other hand, individuals who are deemed to be an ineffective problem-solvers tend to avoid addressing problems, possess negative

attitude toward problem solving, have negative problem solving orientations, and experience stress, depression, and anxiety when faced with problems (Bell & D’Zurilla, 2009; Siu & Shek, 2005). In a study of middle-aged adults, it was found that both SPS skills and amount of perceived stress were involved in an individual’s sense of psychological well-being (Chang, D’Zurilla, & Sanna, 2009). SPS skills were found to partially mediate the link between stress and psychological well-being; specifically, negative problem-orientation was a unique predictor of particular dimensions of psychological well-being (Chang *et al.*, 2009). Health was rated significantly worse on days when SPS skills were decreased and negative mood prevailed for a group of college students who were prospectively evaluated on their experience of events, mood, and health on a daily basis (Baker, 2006). It was suggested that increasing individuals’ SPS skills may increase mood and they may evaluate problems that occur on a daily basis as less negative (Baker, 2006).

The particular skills involved in the SPS model are: problem orientation, problem definition and formulation, generation of alternative solutions, decision-making, and solution implementation and verification (D’Zurilla *et al.*, 2002; D’Zurilla *et al.*, 2004). Problem definition and formulation skills include attempts to clarify and understand the problem by gathering specific and concrete facts about the problem, identifying demands and obstacles, and setting realistic goals. The second skill set in the SPS model is generation of alternatives, which includes focusing on the specific problem-solving goals and identifying as many potential solutions as possible. These solutions can include conventional and original solutions. In the decision-making step, individuals implementing the SPS model anticipate the positive and negative consequences of the

different solutions to the problem that were identified in the second step of the SPS process. After judging and comparing the consequences, the individual chooses the best, or potentially most effective solution. Finally, the last step of the SPS model is for the individual to implement the solution and to carefully monitor and evaluate the outcome of the chosen solution. The verification process occurs after attempting to implement the solution (D’Zurilla *et al.*, 2002; D’Zurilla *et al.*, 2004).

The model of Social Problem Solving described by D’Zurilla, Nezu, and Maydeu-Olivares (2004) includes two possible problem orientations (positive, negative) and three problem-solving styles (rational, impulsivity/carelessness, avoidant). The model predicts that effective outcomes of utilizing the model are from a combination of positive problem-solving orientation and rational problem-solving style. Individuals with these characteristics are more likely to have a combination of positive cognitions and behaviors that encourage active, persistent efforts. A combination of negative cognitive orientation and ineffective behavioral skills sets for solving a problem results in a lack of persistence, lack of effort, avoidance, or dependence on others to act (D’Zurilla *et al.*, 2004). Therefore, the steps of Social Problem Solving Therapy, as outlined above, have been found useful in helping those who are ineffective problem solvers learn to be more effective.

Effectiveness of Social Problem Solving Therapy.

Social Problem Solving interventions have been demonstrated to be effective in helping individuals in distress. Problem-solving ability served as a moderator between depressive symptoms and stressful life events in a study of college students (Nezu, 1986). Problem-solving dimensions were predictors of depression in individuals under high

levels of stress. In the study, those who were effective problems solvers reported significantly lower depression scores than ineffective problem solvers (Nezu, 1986). SPS ability has been found to be negatively related to stress in a sample of college students (D’Zurilla & Sheedy, 1991). Specifically, problem orientation (PPO or NPO) is a significantly stronger predictor of stress compared to the individual’s particular problem-solving skills (D’Zurilla & Sheedy, 1991).

SPS interventions have been commonly used with individuals who have been diagnosed with a chronic health condition (Hill-Briggs, 2003; Schmied & Tully, 2009) and their caregivers (Nezu *et al.*, 2003). SPS skills have been widely accepted as an aid for the self-management of diabetes at any developmental age (Bonnet, Gagnayre, & d’Ivernois, 1998; Paterson & Thorne, 2000). In a problem-solving intervention with 102 adults who were diagnosed with type 2 diabetes, the intervention revealed positive improvement in problem solving, diet, fat and caloric intake, weight reduction, and glucose testing frequency (Glasgow *et al.*, 1992). In studies of children diagnosed with diabetes, there have been mixed results of SPS interventions. While interventions have been generally effective in increasing SPS skills, there have been mixed results for increasing disease management behaviors, HbA_{1C} levels, personal control, diet, physical activity, and blood glucose levels. These are very similar to the disease management behaviors that individuals diagnosed with LQTS and their parents need to manage. In a study of 19 adolescents with type 1 diabetes, the problem-solving and behavior therapy intervention improved SPS skills, but did not produce significant changes for personal control in problem solving, blood glucose levels, and intake of carbohydrates or calories (Schlundt, Flannery, Davis, Kinzer, & Pichert, 1999). Another educational intervention

study, which included SPS skills for 49 children diagnosed with type 1 diabetes, increased problem-solving skills of the children, but did not show significant improvement in HbA_{1C} (Lucey & Wing, 1985). Similar problem-solving interventions may be useful for adolescents diagnosed with LQTS and their parents.

A third study of a diabetes education program for children diagnosed with type 1-diabetes and their parents found that parental problem solving skills increased as part of their participation in the educational program (Bloomfield *et al.*, 1990). There were no significant improvement in children's problem-solving skills, but the researchers did find decreases in HbA_{1C} levels and increased hypoglycemia of the children who participated in the study. There was no significant association between parents' problem-solving abilities and changes in the child's HbA_{1C} levels (Bloomfield *et al.*, 1990). Interventions have also aimed to utilize technology to model problem-solving barriers to disease self-management and psychosocial stressors, and these barriers were decreased through teaching SPS skills to the adolescents (Mulvaney *et al.*, 2011). SPS interventions have also been found to address self-esteem and social adjustment issues in adolescents who have been diagnosed with chronic health conditions. These preventative interventions were found to be helpful (Meijer, Sinnema, Bijstra, Mellenbergh, & Wolters, 2002). Interventions that address self-esteem and social adjustment issues that would benefit adolescents diagnosed with LQTS. Other SPS studies have been found to be effective in decreasing the reported levels of distress in both adult cancer patients and their caregivers, and these effects were still detected at 6-month and 1-year posttreatment follow-ups (Nezu *et al.*, 2003).

While parental caregiver problem-solving is involved in the care of children and adolescents diagnosed with a chronic illness, the child and adolescent also need to be able to make sufficient disease-related decisions and choices for situations encountered in daily life (Schlundt *et al.*, 1999; Thomas, Peters, & Goldstein, 1997). This gives the adolescent a sense of control and makes him or her part of the decision-making process in the treatment of their chronic illness. Previous studies have shown the effectiveness of including parents in interventions for children diagnosed with chronic illness (for example, Anderson & Davis, 2011; Goldbeck & Bakka, 2001; Lobato & Kao, 2002; Martire & Schulz, 2007). However, these interventions have not been specifically applied in a one-time social problem-solving workshop for children who have been diagnosed with LQTS and their parents. Utilizing other studies as a framework to examine parents' problem-solving style with their children's LQTS, a social problem-solving workshop may be effective in helping to teach parents effective problem-solving skills, to help increase their hope, coping skills, and self-efficacy, and to be able to help children learn to manage their LQTS in a more effective way.

CHAPTER 3

Hypotheses

Research Question.

Is it feasible to implement a one-time social problem-solving (SPS) workshop for children diagnosed with LQTS and their parents designed to increase problem-solving skills, hope, and coping strategies, and to decrease worry in the parents after participating in the workshop with their children?

LQTS is a chronic illness that affects the psychosocial well-being of the individual diagnosed with the chronic illness and also their family and caregivers. Parents of children diagnosed with a chronic illness face a number of psychosocial concerns. The present study will evaluate the impact on the parents of a group intervention focused on SPS skills directed toward children diagnosed with LQTS. The inclusion of parents in a SPS intervention can help parents to recognize that they are helping his/her child learn specific techniques to cope with their diagnosis of LQTS and in order to achieve greater independence to positively handle LQTS-related issues. This pilot study will examine if there are any long-term (3 month) gains for parents' social problem solving abilities, hope, and ability to cope with LQTS, and decreases in worry. The efficacy and effectiveness of the four-hour workshop will be perceived as useful to parents of children diagnosed with LQTS, as measured by the Satisfaction Survey that was created for this study.

H₁: There will be an increase in number of coping strategies from pretest to 1-month follow-up, and maintained at 3-month follow-up, following participation in a Social

Problem Solving Workshop as measured by the Coping Health Inventory for Parents (CHIP).

H₂: Participants will demonstrate an increase in total problem solving abilities as measured by the Social Problem-Solving Inventory-Revised (SPSI:R) Total Score, Positive Problem Orientation, and Rational Problem-Solving Skills subscales from pretest to 1-month follow-up and maintained at 3-month follow-up. Participants will also demonstrate a decrease on the Negative Problem Orientation, Avoidance, and Impulsive/Carelessness subscales of the SPSI:R from pretest to 1-month follow-up and maintained at 3-month follow-up.

H₃: Participants' degree of worry will decrease from pretest to 1-month follow-up and maintained at 3-month follow-up following participation in a Social Problem Solving Workshop as measured by the Worry Scale of the PedsQL Family Impact Module.

H₄: Participation in a Social Problem Solving Workshop will increase parents' ratings of hope from pretest to 1-month follow-up and maintained at 3-month follow-up, as measured by the Adult Hope Scale.

CHAPTER 4

Method

Overview.

The present study was part of a larger study evaluating the feasibility of a group-based Problem-Solving Therapy Workshop for children and adolescents diagnosed with Long QT Syndrome (LQTS) and their parents. The present study focused on the feasibility of including parents of adolescents diagnosed with LQTS in the workshop and investigating whether participation in the workshop was helpful to the parents. The focus of the workshop was to teach adolescents diagnosed with LQTS problem-solving skills to improve their ability to solve problems related to their LQTS diagnosis. Parents were included in the group as caregivers and participants who were able to encourage their adolescent to increase their use of the SPS skills. Parental participation in the group was utilized to help the adolescents learn new skills; increase the parent's hope, confidence related to solving LQTS-related problems, positively affect parents' coping style, and decrease parental worry. Only parental outcomes were measured for this particular study.

Design and design justification.

The study was a single-subject, within-group repeated measure design. Participants were assessed at 1- and 3-months post intervention to examine independent variable outcomes after completion of the workshop. The research goals to examine the feasibility of a group Social Problem Solving Workshop was to examine the effect of the workshop on parents' coping, worry and hope. The repeated measure design in this feasibility study provided the necessary conditions required to demonstrate whether Social Problem Solving Workshops are feasible with this population. The design began

with two scheduled groups, with four and two registered participants, respectively.

However, four participants dropped out prior to providing baseline data and participating in the workshop. Therefore, only one workshop was conducted, and all baseline and follow-up measures were obtained from the two parents who participated in this group.

Participants.

Participants were parents of children (ages 8-17) who had been diagnosed with LQTS by an electrophysiologist at least 6 months prior to participation in the workshop. Children and their parents were invited to participate in the workshop by sending letters of invitation from physicians to potential participants at the Children's Hospital of Philadelphia (CHOP), mailings to cardiac centers at hospitals in the Philadelphia area, and Internet announcements posted on LQTS-related sites such as Facebook groups, Twitter, Craigslist, and SADS. The current study recruited from hospitals that serve a diverse population of ethnic minorities. It was hoped that a total of 5 to 10 dyads (parents and adolescents) would be recruited for participation in the workshop. Workshops ranged in size based on interest; however, previous group research with individuals diagnosed with chronic illness and their parents has been completed with three to five adolescents and their respective parent assigned to each group (Ambrosino *et al.*, 2008). A total of six parents of children diagnosed with LQTS were registered for participation in the 4-hour workshops. Of the six recruited participants, four were not enrolled in the study due to inability to attend the 4-hour workshop. Inability to attend the workshop was due to a number of factors, including child's unwillingness to attend the workshop, scheduling conflicts, failure to attend scheduled workshop, and not responding to contact attempts. The participants, both Caucasian (ages 40 and 43; one male and one female),

were not diagnosed with LQTS. However, one spouse had been diagnosed with LQTS. One participant had been previously diagnosed with anxiety. Both participants had not been involved in other LQTS related support groups. Each of the children (ages 9 and 13; one male and one female) had been diagnosed with LQTS at least 6 months prior to participation. Both parental participants completed all baseline, post treatment, 1-month follow-up, and 3-month follow-up assessments.

Inclusion Criteria.

Parents were recruited for participation in the study if they met the following criteria: had a child/adolescent (ages 8-17) who had been diagnosed for at least 6 months with LQTS; capable of giving informed consent; read/write English, as indicated by their ability to read and sign the informed consent form; willing to give informed consent for both themselves and their child diagnosed with LQTS; and willing to participate in the four-hour workshop with their children. Parents were included regardless of whether or not they have a diagnosis of LQTS, and they must have been willing to provide personal information about themselves and their child.

Exclusion Criteria.

Parents were excluded if they were unable or unwilling to provide informed consent or were unable or unwilling to participate in the 4-hour workshop with their adolescent. Parents were excluded if their adolescent only had a probable diagnosis of LQTS or had been diagnosed with LQTS for less than 6 months.

Measures.***Personal Information Questionnaire (PIQ).***

A Personal Information Questionnaire (PIQ) was constructed to acquire descriptive information about the parent and child, including ages; medications; history of sudden cardiac arrest for parents and adolescents; presence of implantable devices; possession of automatic external defibrillator; number of siblings; other medical or psychological illnesses of parents, adolescents, and other family members; and other personal characteristics relevant to the description of the sample and study. Only parents completed the PIQ about the family members participating in the study.

Social Problem-Solving Inventory – Revised (SPSI-R:S).

The Social Problem-Solving Inventory – Revised, Short Form (SPSI-R:S) measures how individuals solve problems and evaluates different problem-solving styles (D’Zurilla *et al.*, 2002). The scale was developed based on a theory-based approach and is based on a conceptual model. The 25-item self-report scale measures ability to resolve problems in daily living. This measure helps to understand how a person typically resolves stressful situations and makes adequate decisions. In addition to a total score, the measure consists of five scales that measure two constructive dimensions (Positive Problem Orientation and Rational Problem Solving) and three dysfunctional dimensions (Negative Problem Orientation, Impulsivity/Carelessness Style, and Avoidance Style). Positive Problem Orientation (PPO) is described as a constructive, problem-solving cognitive set that involves appraising a problem as a challenge rather than a threat, a belief that problems are solvable, a belief in one’s ability to successfully solve problems, a belief that successful problem-solving takes time, effort and persistence, and a

commitment to solve problems with dispatch rather than avoidance. Negative problem Orientation (NPO) is the dysfunctional or inhibitive cognitive-emotional set that involves the tendency to view problems as a significant threat, doubt one's ability to successfully solve problems, and become upset or frustrated when confronted with problems. The Rational Problem Solving (RPO) subscale includes four specific tasks, which define a constructive problem-solving style: problem definition, generation of alternative solutions, decision-making, and solution implementation and verification.

The total social-problem solving score is comprised of a total of all of five scales, with higher scores (115 to 145) indicating above norm group average to extremely above norm group average, average scores (86 to 114), lower scores (71 to 85) indicating below norm group average, and (70 and below) very much below and extremely below norm group average (D'Zurilla *et al.*, 2002). The five scales assess positive and negative problem orientation dimensions and problem-solving styles, namely, rational problem solving, impulsivity/carelessness style, and avoidance style. The SPSI-R:S has demonstrated satisfactory levels of test-retest reliability ($r = 0.83$ to 0.90), and strong internal consistency ($\alpha = 0.79$ to 0.95) across all five scales (D'Zurilla *et al.*, 2002).

The Coping Health Inventory for Parents (CHIP).

The Coping Health Inventory for Parents (CHIP) is a measure of parent perceptions of behaviors used to manage family life when a child has a serious or chronic illness (McCubbin *et al.*, 1983). The 45-item questionnaire measures parental coping strategies, yielding three subscales that represent different positive coping styles: (a) Coping Style I: maintaining family integration, cooperation, and an optimistic definition of the situation; (b) Coping Style II: maintaining social support, self-esteem, and

psychological stability; and (c) Coping Style III: understanding the medical situation through communication with other parents and consultation with healthcare staff. Parents evaluate particular ways of coping, rating each as 3 (*extremely helpful*), 2 (*moderately helpful*), 1 (*minimally helpful*), 0 (*not helpful, chose not to use it, or not possible*). Items rated as *chose not to use it* or *not possible* are not added into the particular score for that subscale. The maximum possible score for Coping Style I is 54, the maximum possible score for Coping Style II is 51, and the maximum possible score for Coping Style III is 24. Raw scores are converted into normed scores, which differ for mothers and fathers. There are separate normed scores for parents of children with cardiac illnesses and diabetes. Higher scores on a particular subscale indicate increased utilization of the particular coping styles. The CHIP has satisfactory internal consistency for each of the three subscales ($\alpha = 0.79$, $\alpha = 0.79$, and $\alpha = 0.71$, respectively) (McCubbin, McCubbin, & Thompson, 1981).

PedsQL Family Impact Module.

The PedsQL Family Impact Module is a measure of the impact of pediatric chronic illness on the parents and the family, and is part of the PedsQL Measurement Model (Varni, Seid, & Rode, 1999). The PedsQL Family Impact Module is a 36-item self-report scale that measures parents' self-reported physical functioning (6 items), emotional functioning (5 items), social functioning (4 items), cognitive functioning (5 items), communication (3 items), and worry (5 items), along with parent-reported family daily activities (3 items) and family relationships (5 items). Parents are asked to indicate how much of a problem they have had with a particular statement, rating each as 4 (*almost always*), 3 (*often*), 2 (*sometimes*), 1 (*almost never*), and 0 (*never*). Scores are

transformed to a 0 to 100 scale. Higher scores indicate better functioning of the individual. Most of the scales on the PedsQL Family Impact Module approached or exceeded a Cronbach's alpha coefficient of 0.90 (range 0.82 to 0.97) for internal consistency reliability (Varni, Sherman, Burwinkle, Dickinson, & Dixon, 2004).

The Adult State Hope Scale.

The Adult State Hope Scale is a measure of an individual's level of hope as well as agency and pathway thoughts, and identifies dispositional hope in adults over the age of 15. Agency thoughts refer to goal-directed determination, while pathway thoughts refer to the planning of ways to meet the goals. The measure was created for several uses, including predicting outcomes among a sample and identifying individuals who need extra support and are low in hope (Snyder, 1995). The Adult State Hope Scale is a self-report questionnaire that contains 12 questions scored on an 8-point Likert scale. The scale creates three scores, a hope score that is created by summing the agency and pathway items and two separate scores that measure agency and pathways individually. The hope scale scores range from a minimum of 8 to a maximum of 64, while agency and pathway scores range from a minimum of 4 to a maximum of 32, with high scores reflecting higher levels of hope (Snyder *et al.*, 1991). Average scores on the Adult State Hope Scale are 48 (Lopez, Ciarlelli, Coffman, Stone & Wyatt, 2000), however, clinical samples have had significantly lower hope scores than undergraduate populations (Snyder *et al.*, 1991). The mean hope score for clinical samples was still toward the hopeful end of the response scale. The Adult State Hope Scale has good levels of reliability for overall hope, agency thoughts, and pathway thoughts ($\alpha = .74$ to $.84$; $.71$ to

.76; and .63 to .80, respectively). The test-retest reliability is .80 or above for period of up to 10 weeks in the student sampling populations (Snyder *et al.*, 1991).

Satisfaction Questionnaire.

Data on participant satisfaction and qualitative feedback was also collected as part of this feasibility study. A short feedback survey was developed for use at the end of the workshop. Participants were asked to rate how well they believe the session covered the designated objective and if the topics covered were useful and informative, using a 10-point Likert scale (1, *Not at all* to 10, *Definitely*). Each section (Workshop Information, Workshop Materials, Presenter Qualities, Topics of Workshop, Workshop Length/Location, Overall Evaluation of Workshop, and Parents Only) of the questionnaire was evaluated, and the workshop was deemed useful to parents, as indicated by an average score of 7 or higher on each section. Qualitative feedback on participants' most and least favorable components of the workshop was examined and evaluated for further determination of participant satisfaction of the overall workshop, as well as to determine if changes to the protocol were needed to make it more feasible for future studies. At the end of the workshop, each participant completed the satisfaction questionnaire to provide his or her opinion on the appropriateness and usefulness of the group intervention.

Workshop Evaluation Survey.

Data on group leader evaluations of the survey was collected as part of this feasibility study. A short survey form was developed for use at the end of the workshop. Group leaders were asked to evaluate the ease of following the protocol, time taken to complete each section of the protocol, usefulness of examples, and engagement of the

group using a 10-point Likert scale (1, *Not at all* to 10, *Definitely*). Group leaders also answered open-ended questions to further describe elements of the workshop that were effective and those that did not work.

Protocol Adherence Checklist.

A protocol Adherence Checklist was created for three independent raters to utilize while completing the integrity check of the protocol. The checklists evaluated specific group leader behaviors and completion of specific tasks as explained in the protocol.

Manuals.

The protocol for this feasibility study was created from a combination two manuals, but was adapted to incorporate material that is specifically related to the experiences of individuals diagnosed with LQTS. The protocol is primarily based on the problem-solving therapy described in *Helping Cancer Patients Cope* (Nezu *et al.*, 1998). The *I Can Problem Solve: An Interpersonal Cognitive Problem-Solving Program* (Shure, 2001) manual was utilized as a model to gauge age-appropriateness of examples and language within the protocol. Problem-solving skills training have been shown to be effective for helping other medical patients (Nezu *et al.*, 1998).

Helping Cancer Patients Cope: A Problem-Solving Approach.

The *Helping Cancer Patients Cope* manual outlines a 10-session Social Problem-Solving program. This particular program can be applied and adapted for individual, group, or family settings. The model of Social Problem-Solving outlined in this manual helps to diminish patients' sense of helplessness and despair, while instilling a sense of control and hope. Included are numerous case examples, transcripts from sessions, and

sample introductions that are a helpful adjunct to treatments for mental health professionals (Nezu *et al.*, 1998).

I Can Problem Solve: An Interpersonal Cognitive Problem-Solving Program.

This manual provides children with skills to think about how to solve problems at a young age to increase their chances of success and social competence in their future. An initial study of the *I Can Problem Solve* manual evaluated the interpersonal cognitive problem-solving training for children 10 years old. This study demonstrated that behavior change occurred, increasing prosocial behaviors and decreasing negative behaviors, thus increasing participants' concentration on task demands in the classroom and subsequently increased achievement in school (Shure, 2001).

Procedure.

All participants signed an informed consent form approved by the appropriate Institutional Review Board (IRB). The study was conducted at Philadelphia College of Osteopathic Medicine (PCOM). The investigators collected all pretest, posttest, and follow-up data on all participants.

Copies of materials created specifically for this study, including questionnaires, protocol, letters of invitation for participation, etc. can be requested directly from the author.

A number of different recruitment methods were utilized to attempt to reach as many interested people for possible participation in the 4-hour workshop. First, CHOP study staff identified individuals who met inclusion criteria for participation in the study. These individuals were sent a brochure and invitation letter by CHOP staff. Included in the mailing was a response card. Interested families returned the response card, e-mailed,

or called the investigators at PCOM denoting their potential interest in participation in the workshop. Second, cardiologists at local hospitals were contacted and sent study announcements to distribute in their practices. Third, study announcements were posted on several social media websites, including LQTS Facebook pages, Craigslist, SADS, and Twitter. Interested families were encouraged to call or e-mail the PCOM LQTS e-mail account, to express their interest. Families who responded were contacted with further information regarding the workshop. Those families who still expressed interest in participation in the workshop were scheduled for available workshop dates. Families registered for the workshop were contacted twice prior to the workshop, one week and two days in advance, by phone and e-mail to remind them of the date and time of the workshop and answer any last minute questions.

Families arrived at PCOM for the workshop on the designated day, with registration beginning at 9:00 am. The workshop ran for approximately 4 hours, with breaks and refreshments available for the participants. At the time of registration, both the parent and the child were present to read and sign the informed consent and assent forms for participation in the workshop. Parents signed two forms, one to consent for their participation in the study and a second one to consent for their child to participate in the study. The child signed the assent form. Once parents and children consented to participate in the workshop, they were given a folder of questionnaires to complete. Parents completed the PIQ, SPSI-R:S, CHIP, Adult State Hope Scale, and PedsQL Family Impact Module questionnaires. Questionnaires were returned to the investigators prior to the beginning of the workshop.

The 4-hour workshop was conducted jointly by two advanced clinical psychology doctoral candidates who were under the supervision of a licensed psychologist board certified in clinical psychology by the American Board of Professional Psychologists in Clinical Psychology. Children and their parents were introduced to and taught the steps of the Social Problem-Solving Model, which included Problem Orientation, Problem Definition and Formulation, Generation of Alternatives, Decision Making, Solution Implementation, and Solution Verification (Nezu, *et al.*, 1998). Vignettes of common LQTS-related problems were presented to the group for practice and implementation of the Social Problem-Solving Model. At this time, the children and parents were instructed to work together to discuss the vignettes and work through the presented problem utilizing all of the steps of the Social Problem-Solving model. Common discussion time followed each of the vignettes, which was led by the instructors. Areas in which individuals in the workshop seemed to be having more trouble with the Social Problem-Solving Model were explained and practiced further. Finally, each of the children, with the help of their parent, attempted to utilize the Social Problem-Solving model on a personal LQTS-related problem they were currently experiencing. The last half hour of the workshop was spent concluding and for both children and parents to fill out a post-workshop questionnaire and the Satisfaction Survey. Participants also completed a preference for contact form to gather contact information, which was used only for follow-ups. Snacks and beverages were available to participants at this time. Each family was given a list of resources and referral sites at the end of the workshop.

No participants disclosed experiencing emotional discomfort due to discussing topics related to problem-solving and LQTS during the workshop. Participants were

asked at the completion of the workshop if any of the study material made them uncomfortable or upset. If participants had experienced discomfort, they would have been provided with resources to talk to a qualified professional about their concerns and appropriate referrals would have been provided at that time. Additionally, a referral list was provided to all participants at the end of the workshop that included psychology referrals, online support groups, and informational websites. In addition, formal procedural guidelines were in place in the event of a participant experiencing extreme emotional discomfort.

Following completion, the supervising psychologist and two advanced doctoral candidates who conducted the research discussed the workshop. Three independent raters conducted the integrity checks utilizing the video-recorded session of the workshop. Raters were doctoral students who had been previously trained in Problem-Solving Therapy. The raters reviewed the workshop tapes, noting qualitative observations and following a checklist of the expected content included in the protocol, to verify whether techniques were or were not used during the workshop. They also determined the degree to which the group leaders adhered to the protocol manual. Each of the group leaders completed the Workshop Evaluation Survey, evaluating the ease of following the protocol, pacing of sessions, usefulness of examples, and engagement of the group.

One month following the workshop, each parent was contacted by the investigators for a 5 to 10-minute phone call. This phone call was utilized to check-in with the parents, evaluate effectiveness of the workshop, work through any current issues in implementing the problem-solving strategies that were taught at the workshop, and

given a small quiz on the steps of the Social Problem Solving model. At this time, parents were given an identification number and asked to complete the follow-up questionnaires via an online link to SurveyMonkey that was e-mailed from a PCOM LQTS e-mail account. The questionnaires completed at 1-month follow up were the SPSI-R:S, CHIP, PedsQL Family Impact Module, and the Adult State Hope Scale. If questionnaires were not completed within 1 week, participants were reminded via e-mail to complete the online questionnaires. After the 1-month follow-up was completed, participants were mailed a gift card for a nationwide retailer.

Three months following the workshop, each parent was contacted again by the investigator for a brief phone call and sent a link via e-mail to complete questionnaires. The questionnaires administered at 3-month follow-up were the SPSI-R:S, the Adult State Hope Scale, CHIP, and PedsQL Family Impact Module. All participants completed the 3-month follow-up and were mailed a gift card for a nationwide retailer.

CHAPTER 5

Results

Overview.

Two parents of children diagnosed with LQTS participated in the workshop, and data was examined and described within and between participants. The baseline assessments and workshop follow-up (1- and 3-month) assessments were analyzed to evaluate change in problem-solving abilities, coping strategies, worry, and hope. Feasibility and effectiveness of the pilot *Living Life With LQTS* Workshop were examined and described. Examination of the children's outcomes are reported elsewhere.

Social Problem-Solving Abilities.

The results of the study for social problem-solving abilities indicated an overall increase and maintenance of these skills from baseline to 1- and 3- month follow-ups for the parents who participated in the workshop. The total score on the Social Problem Solving Inventory – Revised, Short Form (SPSI-R:S); (D'Zurilla *et al.*, 2002) is a global indicator of a person's social problem-solving ability. Both participants scored in the average range for baseline total score ($M = 103, SD = 1$). Both participants had an increase in the total score for problem-solving abilities following the intervention, and had an average of 1 standard deviation increase in total score by the 3-month follow-up assessment (see Figure 1), which is considered a significant change. The total scores for problem-solving abilities at 3-month follow-up were significantly higher than the normative group (D'Zurilla *et al.*, 2002).

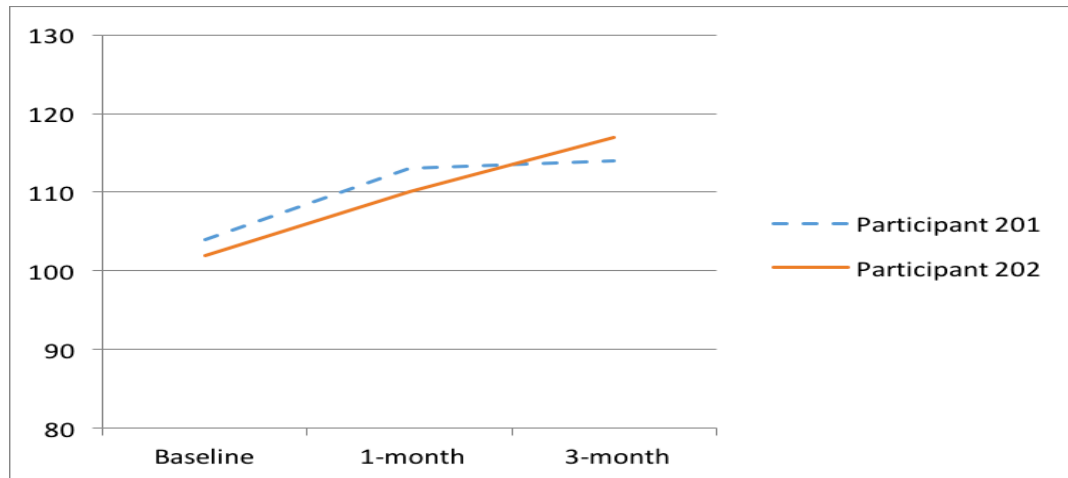


Figure 1. Change in Total Score of the SPSI-R:S.

The Positive Problem Orientation (PPO) scale of the SPSI-R:S measures an individual's constructive problem-solving abilities that involve the following: appraising a problem as a challenge rather than a threat; a belief that problems are solvable, a belief in one's ability to successfully solve problems, a belief that successful problem-solving takes time, effort, and persistence, and a commitment to solve problems with dispatch rather than avoidance. Participants in this study had a variety of change in PPO scores. At baseline, both participants had equal PPO scores ($M = 106$, $SD = 0$), considered average compared to the normative sample. This indicates that at the onset of the workshop, both participants were already able to appraise problems positively and had belief in their abilities to solve problems. One of the participants had an increase in PPO of over 1 standard deviation throughout the follow-up period, which is considered a statistically significant change in score. The other participant, however, had a slight decrease in PPO, although the scores were still average compared to the normative sample (see Figure 2); (D'Zurilla *et al.*, 2002).

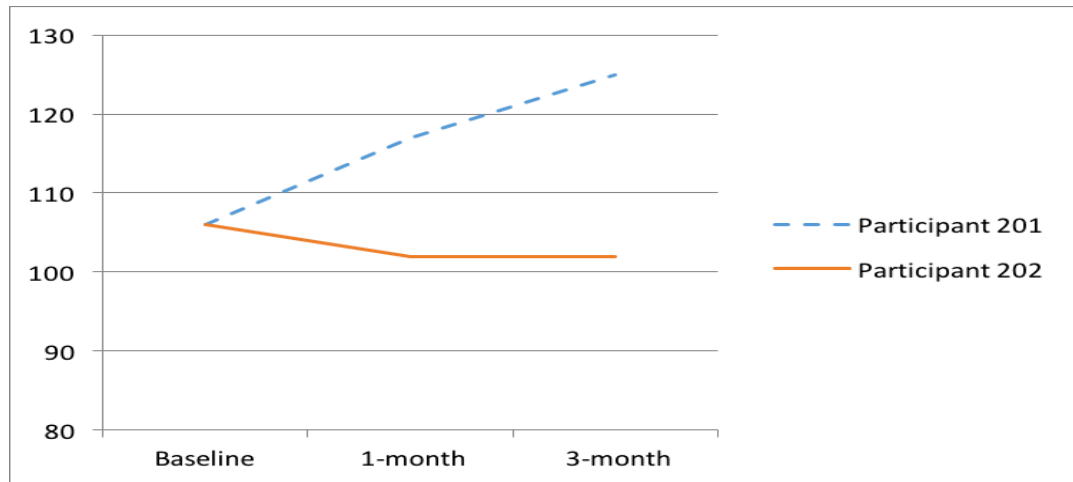


Figure 2. Change in PPO Scores on the SPSI-R:S.

The Negative Problem Orientation (NPO) scale measures dysfunctional or inhibited cognitive-emotional style, which involves the tendency to view a problem as a significant threat; to doubt one's ability to successfully solve problems, and to become upset or frustrated when confronted with problems. Participants in this study had a general decrease in NPO scores from baseline to follow-up periods. At baseline, participants in the study had average NPO scores ($M = 93$, $SD = 2$). One-month after participation in the workshop, participants were 1 standard deviation below the average of the normative sample ($M = 83$; $SD = 0$), a statistically significant difference, indicating that the participants did not view problems as a threat, believed in their ability to successfully solve problems, and did not have negative emotions when confronted with problems. At 3-months after the workshop, one participant continued to decrease in dysfunctional cognitive-emotive style of problem solving, while the other participant had a slight increase in NPO score, although the score was almost 1 standard deviation below the average of the normative group (see Figure 3); (D'Zurilla *et al.*, 2002).

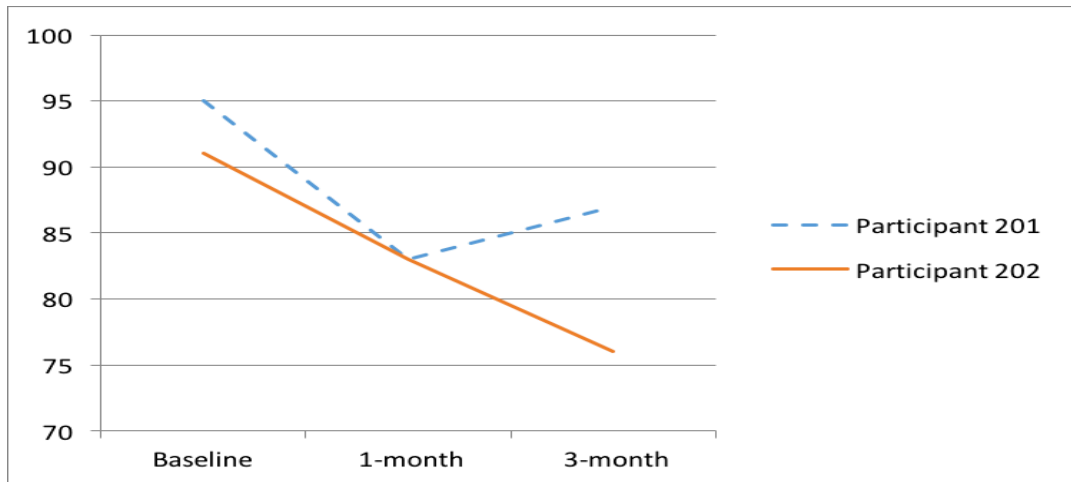


Figure 3. Change in NPO Scores on the SPSI-R:S.

The Rational Problem-Solving Style (RPS) scale measures the rational, deliberate, and systematic application of effective problem-solving strategies and techniques.

Participants had a general increase in RPS scores from baseline through the 3-month follow-up assessment, indicating retention and utilization of problem-solving skills. At baseline, the participants scored in the average range ($M = 96.3$, $SD = 3.5$) in comparison to the normative sample. By 3-months following the workshop, participants had increased their RPS ($M = 112$, $SD = 2$) to nearly 1 standard deviation above the mean in comparison to the normative sample (see Figure 4); (D’Zurilla *et al.*, 2002).

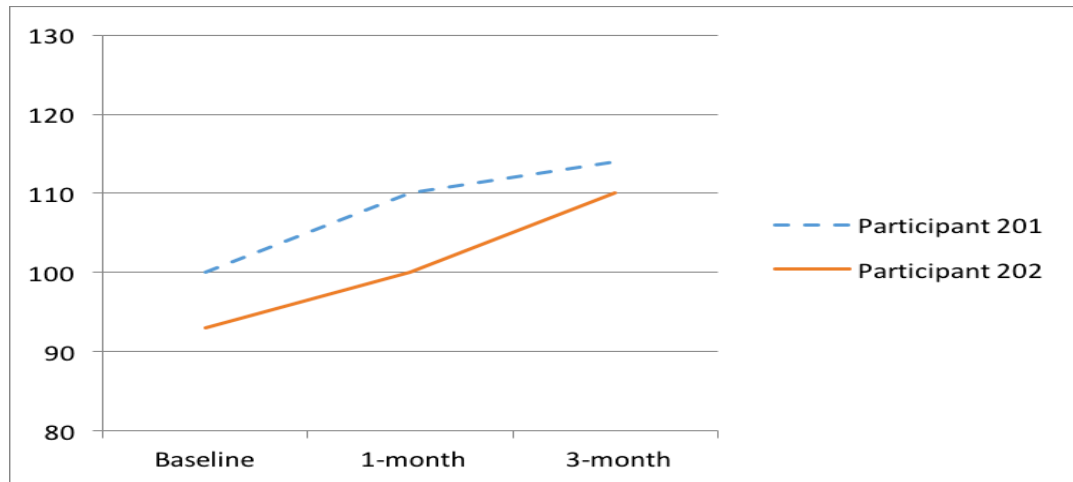


Figure 4. Change in RPS Scores on the SPSI-R:S.

The Impulsive/Carelessness Style (ICS) scale measures narrow, impulsive, and careless attempts at applying problem-solving skills. Higher scores on this index indicate individuals who are unable to generate solution alternatives, are impulsive and try the first idea that comes to mind, and quickly, carelessly, and unsystematically scan alternatives and consequences. Participants in the research study had an overall decrease in ICS scores, indicating that they became less impulsive and careless in attempts at problem solving. At baseline, participants scored in the average range in comparison to the normative group ($M = 99$, $SD = 6$). By the 3-month follow-up, participants scored in the lower limits of the average range in comparison to the normative group ($M = 89$, $SD = 9$); (see Figure 5); (D’Zurilla *et al.*, 2002).

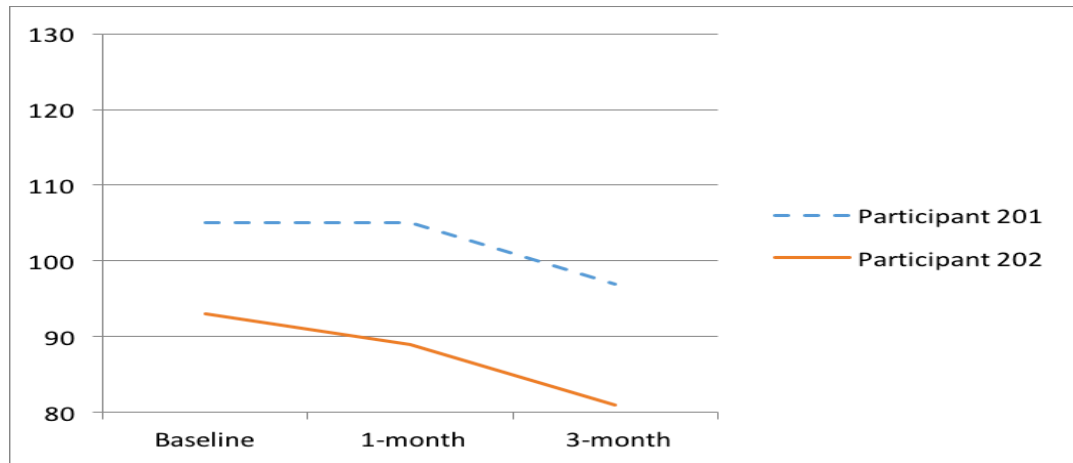


Figure 5. Change in ICS Scores on SPSI-R.

The Avoidant Style (AS) scale measures other deficient problem-solving patterns, which include procrastination, passivity, inaction, or dependency when faced with problems. Persons with higher scores on this scale are likely to avoid problems, put off solving problems for as long as possible, wait for problems to solve themselves, and/or attempt to shift responsibility of problem solving to others. Participants in the study scored in the average range in comparison to the normative sample ($M = 97$, $SD = 5$), indicating that the participants tended to not procrastinate or avoid solving problems. At 1-month following participation in the workshop, both participants had improvements in avoidance style problem-solving, indicating that they were not avoiding solving problems or shifting responsibility to others ($M = 90$, $SD = 2$). However, at 3-months following the workshop, one participant continued to show decreases in avoidant problem-solving styles, while one participant increased in avoidant problem-solving style. However, the score was still in the average range in comparison to the normative sample and would not negatively impact the individual's overall problem-solving style (see Figure 6); (D'Zurilla *et al.*, 2002).

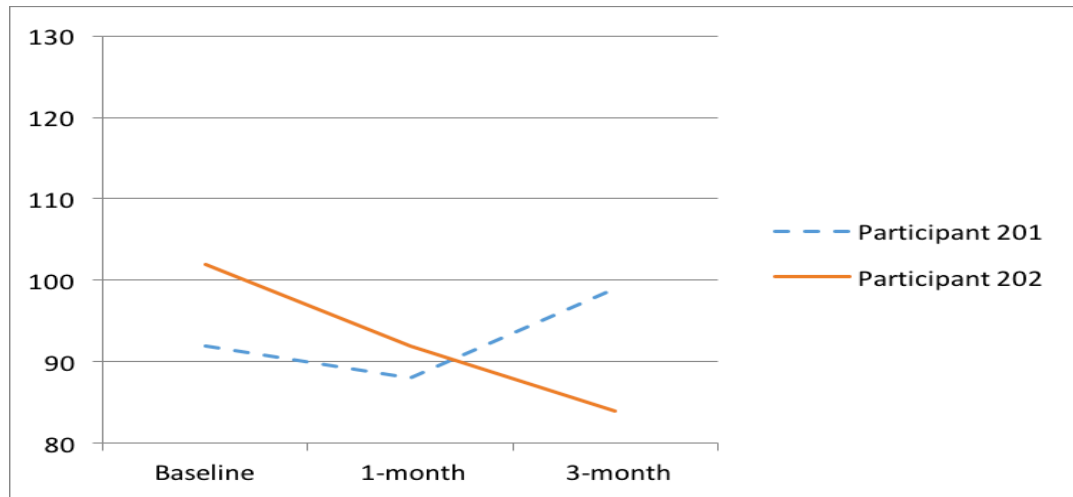


Figure 6. Change in AS Scores on the SPSI-R:S.

Coping Strategies.

Coping strategies were measured by the Coping Health Inventory for Parents (CHIP), which is a 45-item, self-report scale that asks parents to indicate if they utilize certain coping strategies and the extent to which each coping behavior is helpful when caring for a child who has been diagnosed with a chronic illness. Strategies are grouped in to three subscales of coping patterns, (a) Family Integration, Cooperation and an Optimistic Definition of the Situation; (b) Maintaining Social Support, Self-Esteem, and Psychological Stability; and (c) Understanding the Health Care Situation Through Communication with Other Parents and Consultation with the Health Care Team. Participants scores were compared to normative groups for children with cardiac illness based on gender (McCubbin, 1996).

Coping Pattern I (Maintaining Family Integration) identified a variety of ways in which the parent participants engaged in behaviors that focused on strengthening family life and relationships, and the parents' outlook on life with a chronically ill child. Each

parent coped with family integration in different ways after participation in the workshop. Participant 201 had scores slightly below those of the normative group at baseline and throughout the follow-up assessments. This participant remained consistent in reports of coping behaviors that he/she found to be *Extremely Helpful* over the course of the study, potentially indicating that this coping pattern was already strong for this participant and was not changed due to participation in the workshop. Participant 202 had scores significantly higher than those of the normative group at baseline and 3-month follow-up, but was only slightly higher than the norms at the 1-month follow-up (see Figure 7). Similarly, this participant remained relatively stable in the use of family integration coping behaviors, with changes in total score indicating a change in finding a behavior *Moderately Helpful* rather than *Extremely Helpful* (McCubbin, 1996).

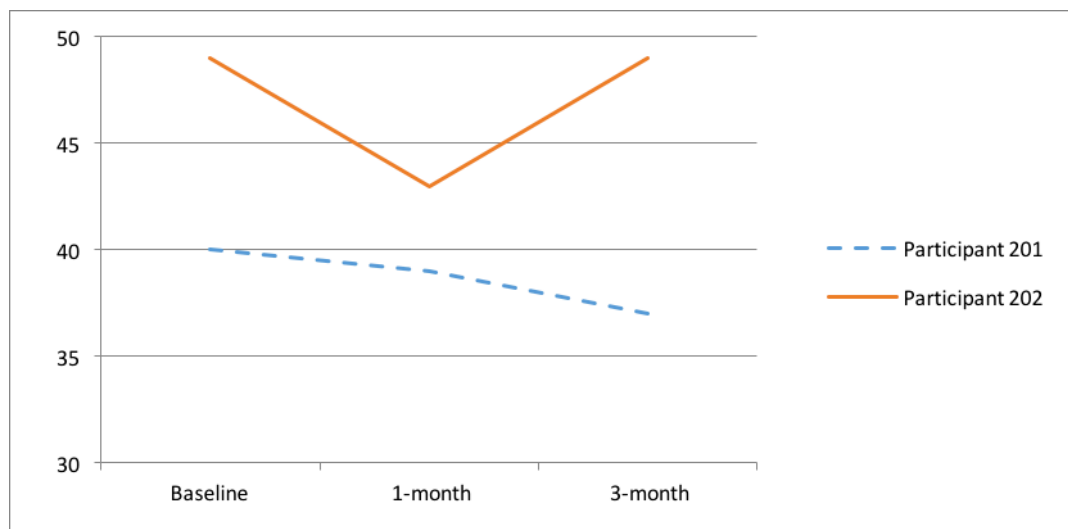


Figure 7. Change in Coping Pattern I on the CHIP.

Coping Pattern II (Maintaining Social Support) identified a variety of ways that parents participated in behaviors that focus on their efforts to develop relationships with

others, engage in activities that enhance feelings of individual identity and self-worth, and manage psychological tensions and pressures. Each parent engaged in different behaviors to cope with maintaining social support after participation in the workshop. Both participants had overall decreases in total coping scores after participation in the workshop (see Figure 8). Participant 201 had scores below the norm at baseline and throughout the follow-up assessments. Overall, this participant reported a number of maintaining social support coping behaviors, such as allowing myself to become angry, talking to a professional about how I feel, and being able to get away from the home care tasks and responsibilities for some relief as either *Chose not to use it* or *Not possible*. Interestingly, this participant reported at baseline choosing not to be more self-reliant and independent, but at 1- and 3-month follow-ups reported finding this skill to be *moderately/extremely helpful*. Participant 202 had significantly higher scores in maintaining social support compared to the normative group at baseline and following participation in the workshop. Differences in this participant's scores throughout the time points seemed to be due to slight changes in finding the specific coping behaviors *Moderately Helpful* vs. *Extremely Helpful* (McCubbin, 1996).

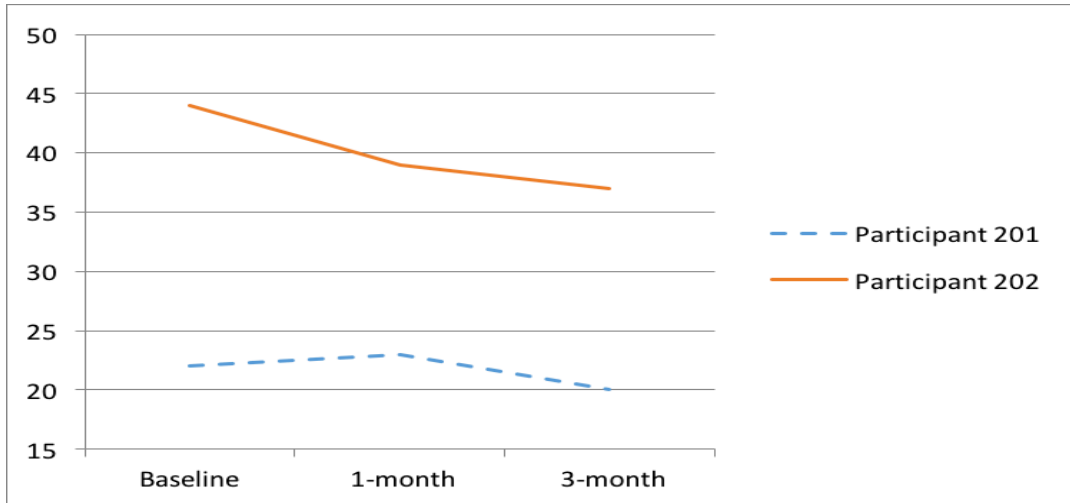


Figure 8. Change in Coping Pattern II on the CHIP.

Coping Pattern III (Understanding the Medical Situation) identifies behaviors directed at the parents' relationships with health care professionals and other parents of chronically ill children, such as increasing knowledge and understanding of the illness and mastering treatment and medical regimens. Participants had opposite changes in their engagement in behaviors to cope with understanding the medical situation, although both participants had scores above the normative group of parents of children with cardiac illness at all measured time points (see Figure 9). Both participants reported slight changes in their determination of how helpful each medically-related coping behavior was to them over the course of the study, with the majority of change occurring due to changes between *Moderately Helpful* and *Extremely Helpful* ratings of each coping behavior (McCubbin, 1996).

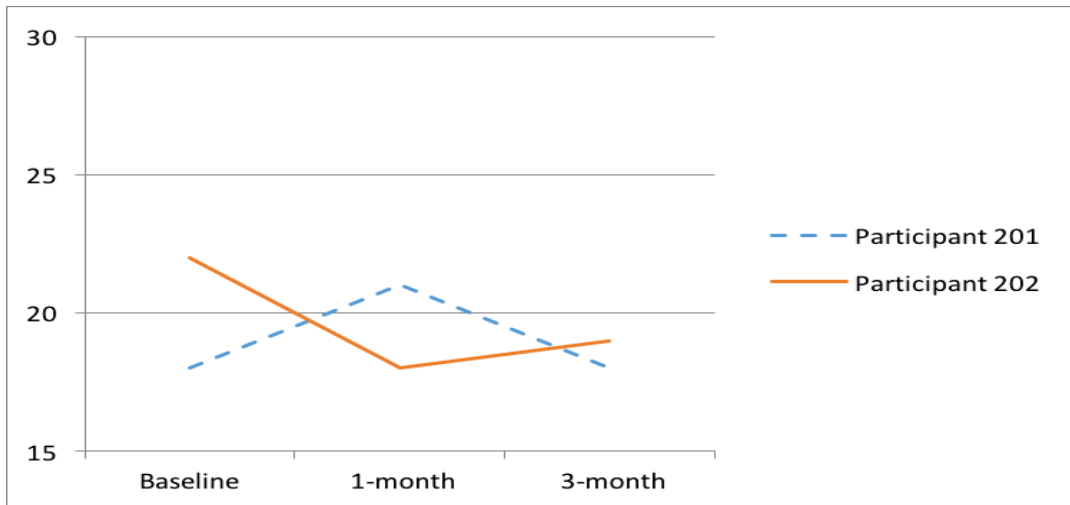


Figure 9. Change in Coping Pattern III on the CHIP.

Worry.

The Worry subscale of the PedsQL Family Impact Module (Varni *et al.*, 1999) measures parents' problems with worrying, including worry about a child's treatment and side effects, others' reactions to the child's condition, effect of the illness on the rest of the family, and the child's future. Higher scores indicate lower levels of worry and more adaptive functioning to the chronic illness, or LQTS, for the population utilized in this research study. Participants in this study had overall decreases in worry following participation in the workshop. Both participants remained relatively stable in their scores of worry between baseline and 1-month follow-up. However, one participant (202) had a significant decrease in worry score between the 1-month and 3-month time points. The other participant also had a decrease in worry scores between 1-month and 3-month time points (see Figure 10).

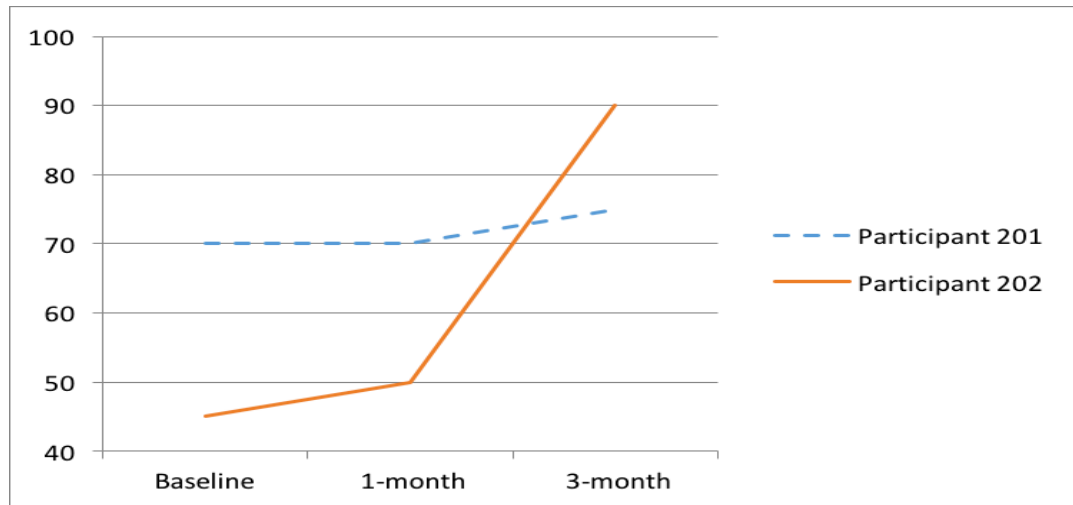


Figure 10. Change in Worry Score on the PedsQL.

Hope.

The Adult State Hope Scale measures an individual's level of hope in a self-report questionnaire that contains 12 questions scored on an 8-point Likert scale, identifying dispositional hope in individuals. Hope is defined as the perceived capacity to devise pathways towards a goal and obtain motivation towards completing those goals (Snyder, 2002). Agency thoughts refer to goal-directed determination, while pathway thoughts refer to the planning of ways to meet the goals. The total hope score is obtained by summing agency and pathway scores.

Agency scores reflect goal directed energy. Participants in this study were divided in their scores of agency. Participant 201 recorded the maximum score for agency at both baseline and 1-month follow-up time points. However, this participant reported a slight decrease in agency over the course of the 1-month to 3-month time frame. Participant 202 had a general increase in agency scores throughout participation

in the research study, resulting in better goal-directed energy after participation in the problem-solving workshop.

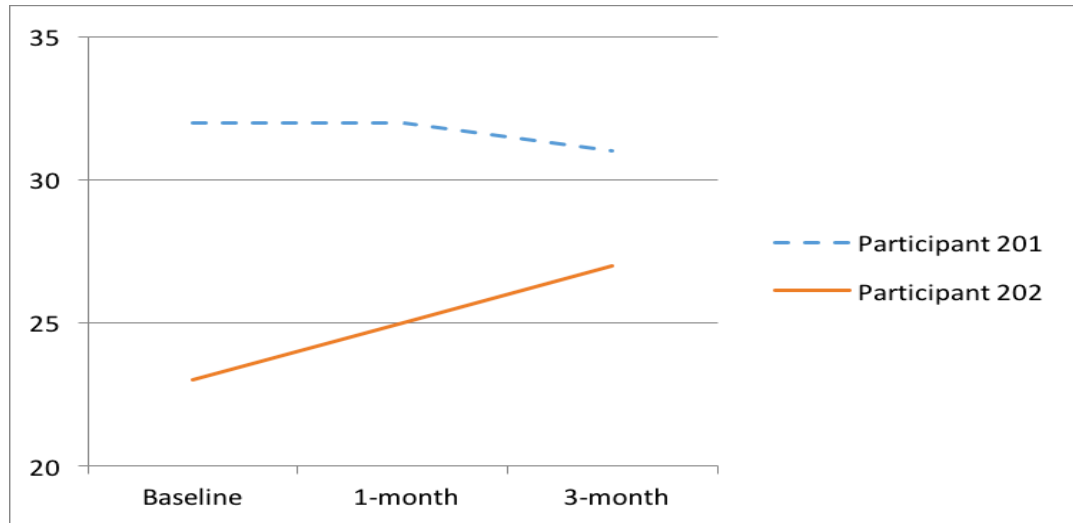


Figure 11. Change in Agency Score on the Adult State Hope Scale.

Pathway scores reflect ability to plan towards accomplishing goals. Participants in the study differed in their scores for pathway. Participant 201 recorded the maximum score for pathway at both baseline and 1-month follow-up time points. However, this participant also reported a slight decrease in pathway over the course of the 1-month to 3-month time frame. Participant 202 had an increase in pathway scores from baseline to 1-month follow-up and then was able to maintain this increase in ability to plan towards accomplishing goals at the 3-month follow-up measurement (see Figure 12).

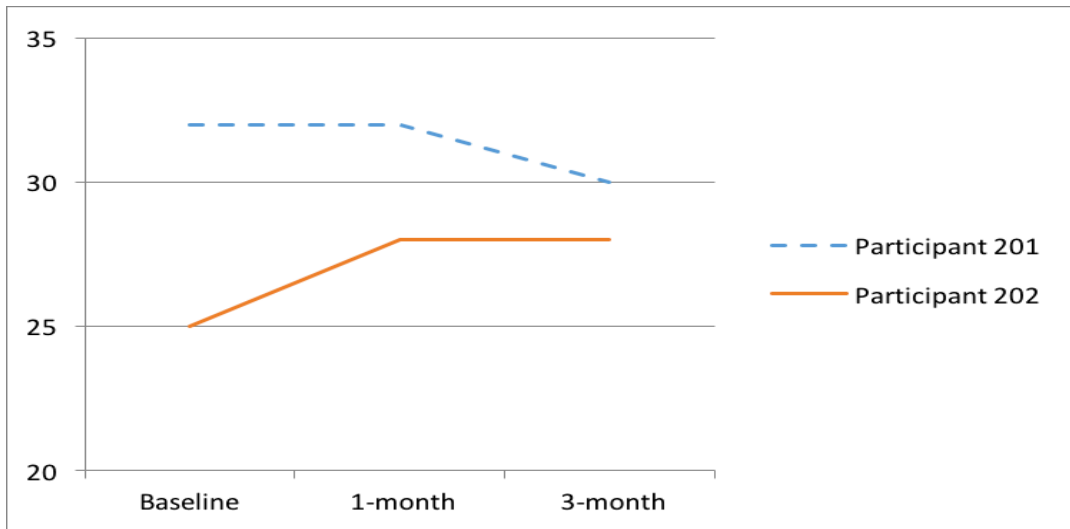


Figure 12. Change in Pathway Score on the Adult State Hope Scale.

Total score on the Adult State Hope Scale indicates a combination of both goal-directed energy and ability to plan for engaging in goal-directed behavior. Both participants were at or above the average ($X = 48$) of the normative group at all measured time points. Participant 201 exhibited very high levels of hope, and had a small decrease in this during the course of the study. Participant 202, however, had a general increase in hope throughout the course of this study, with a large increase at the 1-month follow-up in comparison to the increase from the 1-month to 3-month follow-up time points (see Figure 13).

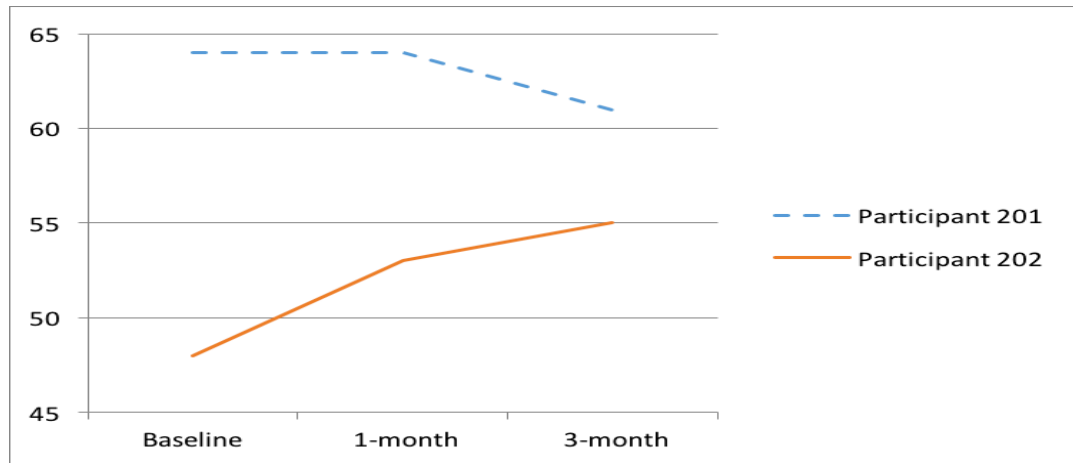


Figure 13. Change in Total Score on the Adult State Hope Scale.

Integrity Checks.

Interrater reliability ratings $\kappa = 0.986$ indicated very good agreement between the raters of the extent to which the protocol was implemented as intended. All of the raters identified that each of the particular steps of the Social Problem Solving model was explained in detail and that utilization of the worksheets and handouts further facilitated the participants' learning of the model. One rater indicated that identification of "what makes a particular situation a problem" was not discussed, and noted that the group leaders did not summarize the problem in the terms given by the child before moving on to the goal-setting portion of the manual. Two of the raters noted that problem orientation was discussed at a later time during the workshop than was established in the manual. One rater questioned whether the children understood some of the terms that were utilized in the manual, such as Quality of Life and assumptions. Qualitatively, it was noted that there was considerable participation from the parents, especially at the beginning of the workshop, which may have overshadowed some of the children's participation or internalization of the concepts being presented in the workshop.

Leader evaluations of overall satisfaction with the workshop of $\kappa = 0.969$, indicating very good agreement between the leaders. Specifically, the leaders had mutual agreement that the topics covered during the workshop were definitely useful and informative to the participants, the workshop was well planned and executed, incorporated participants' LQTS specific problems in the workshop materials, and that participants were actively engaged in the workshop. Furthermore, leaders believed that participants acquired specific strategies to use when faced with LQTS-related problems, and learned how to define problems, investigate the details of a problem, generate new alternatives to solving problems, evaluate the pros and cons of a solution alternative to the problem, and implement a solution and evaluate the outcome. There was slight disagreement between the leaders in regards to the acceptability of the length of the workshop, whether too much information was presented to the participants, and how well the workshop was paced for the participants.

Feasibility and effectiveness.

The feasibility and effectiveness of the *Living Life With LQTS* Workshop was measured by the Satisfaction Questionnaire. Results indicated that overall evaluations of the workshop by participants were positive and that the workshop was both feasible and effective. Both participants rated the question "All and all, I am glad that I attended the workshop" as a 10 (*Definitely*). Parents reported that they enjoyed being with people who understand LQTS issues, and being in a very comfortable setting is what they liked most about the workshop. One participant mentioned that the length of the workshop and the drive to the location were too long. Both parents noted that they would recommend this workshop to others. The overall satisfaction rating with the workshop was 93.67%.

Parents reported a 95% satisfaction rating for the Workshop Information. This suggests that the parents found the topics covered during the workshop to be useful and informative and that enough information was presented during the workshop. Parental participants reported that 25% or 50% of the material presented during the workshop was new to them.

Participants reported a 100% satisfaction rating for the Workshop Materials. This indicates that participants found the handouts useful and easy to read, felt that the workshop was well planned, and were able to understand the material that was presented during the workshop.

Parents also reported on the Presenter Qualities, indicating a 98.8% satisfaction rating. These results suggest that the parents felt respected, believed that the presenters understood the material they were teaching, the material was presented in an user-friendly manner, and found the workshop to be interesting.

Of the topics covered in the workshop, the parents indicated that they were 91.9% satisfied with the topics. Specifically, parents reported that they believed they now had some specific strategies to use when faced with LQTS-related problems. There was a discrepancy in parental reported increase in understanding of how to deal with LQTS-related problems effectively, with one parent reporting only a 5 (*somewhat*) increase and another parent reporting a 10 (*Definitely*) increase. This may be due to the differences in parents' abilities to deal with the LQTS-related problems prior to their participation in the workshop. Also, the parents reported that they learned how thoughts and feelings affect their understand of problems, to define problems, to investigate the details of problems, to generate new alternative to solving problems, to look for the pros and cons of each

alternative to solving a problem, and to implement a solution and evaluate how well it worked. These results suggest that, at the end of the workshop, the parents increased their knowledge of problem-solving skills through participation in the workshop.

Participants reported an 85% satisfaction rating for the workshop Length/Location. Parents reported average ratings of the location and setting (7/8 of 10) and length (8 of 10). One of the participants reported that they believed that the workshop should be 3 hours maximum and suggested that participants fill out questionnaires prior to coming to the workshop so as not to use workshop time. However, all participants reported 10s (*Definitely*) of feeling comfortable about asking questions and giving opinions during the course of the workshop.

Parents reported a 91.3% satisfaction rating for the Parents Only section of the Satisfaction Questionnaire. This indicates that parents overall believed that it was beneficial for their child to participate. Also, parents reported positively that as a result of the workshop, they felt that their child was better able to handle problems or decisions related to LQTS, were less worried about their child's ability to handle life with LQTS, and they were more hopeful about their child's ability to cope with their LQTS. These results support the increases in problem-solving skills, decreases in overall worry, and increases in hope of the parent participants following participation in the problem-solving workshop.

CHAPTER 6

Discussion

Summary of Findings.

This study presents the initial findings of a pilot project aimed at assessing the feasibility and acceptability of a problem-solving workshop for children diagnosed with LQTS and their parents. The primary purpose of the *Living Life with LQTS* Workshop was to teach children diagnosed with LQTS and their parents' effective skills in problem solving. The workshop afforded children and their parents time to apply these new skills of problem solving to both hypothetical and actual LQTS-related problems. The findings in this study suggest that a one-time problem-solving workshop may be feasible and effective for parents of children diagnosed with LQTS. Parental participants reported an overall satisfaction rating of 93.67% following participation in the workshop. They provided positive feedback on the content, instructional materials, group leaders, and skills taught during the course of the workshop. The parents also reported that the information presented during the workshop was useful to their children.

Although the outcomes are preliminary, they suggest that there may be important benefits of teaching parents problem-solving skills and helping parents to coach their children in learning skills to deal with LQTS-related problems. The preliminary results also offer promising indications about the potential effectiveness of the intervention. These findings support the overall research goal of this pilot study to determine if it was feasible to implement a one-time social problem-solving (SPS) workshop for children diagnosed with LQTS and their parents designed to increase problem-solving skills, hope, and coping strategies, and to decrease worry in the parents after participating in the

workshop with their child. Results garnered from participation in the one-time problem-solving workshop should be interpreted with caution, given the small sample size.

Possible factors that may have influenced the results include a possible selection bias for individuals who support research, motivation to find psychosocial supports for their child diagnosed with a chronic illness, and camaraderie with other parents who face the same challenges in raising a child with a life-threatening chronic illness.

Possible treatment effects were explored in this study by assessing changes in problem-solving skills, coping behaviors, worry, and hope in the parents of children diagnosed with LQTS. The changes in problem-solving skills indicated an overall increase in adaptive problem-solving skills and decrease in maladaptive problem-solving skills. Study outcomes indicate that parents decreased avoidant strategies for problem solving, decreased impulsive and careless attempts at problem solving, and increased utilization of the rational problem-solving skills taught during the workshop. Most of the previous research has indicated that teaching problem-solving skills to individuals diagnosed with chronic illnesses has shown disease-related improvements (Bonnet, Gagnayre, & d'Ivernois, 1998; Hill-Briggs, 2003; Paterson & Thorne, 2000; Schmied & Tully, 2009). Interventions that have targeted parents of children diagnosed with chronic illness have also indicated that being educated on problem-solving skills can reduce disease-related distress (Sahler *et al.*, 2002). One of the only previous studies that taught both children diagnosed with a chronic illness and their parents problem-solving skills also found that parental problem-solving skills increased due to their participation in the study (Bloomfield *et al.*, 1990), which was similar to the results of the current research study. Of note, the current research study did not specifically tailor the teaching of

problem-solving skills to the parents. Skills were taught to the children diagnosed with LQTS, and parents were utilized as coaches during the workshop, to aid in the child's ability to learn problem-solving skills. This finding suggests significant opportunities to teach problem-solving skills to diverse populations and using modalities (i.e., individual, group, combined parent-child group) to further the research on utilizing problem-solving therapy as a therapeutic intervention for individuals diagnosed with chronic illnesses and their family members. Participants strongly believed that the workshop was beneficial for themselves and their children, especially in relation to their child's ability to better handle problems or decisions related to LQTS, handle life with a chronic illness, and cope with LQTS.

Changes in coping behaviors of parents trended toward continued use of current coping strategies or increases in currently utilized coping behaviors. Each of the participants in the study utilized different methods of coping. Scores on the three subscales of the CHIP (McCubbin, 1996) were significantly different for the two participants in the study. Participant 201 indicated lower than normed scores on each of the coping behaviors throughout the study. However, it was noted that this participant remained consistent in reports of utilizing coping behaviors deemed to be *Extremely Helpful*, suggesting that this participant might have already been engaging in successful individualized coping behaviors prior to participation in this study. Participant 202 had overall slightly higher than the normed scores for each of the subscales throughout the course of the study. However, changes in this participant's scores varied more than those of Participant 201, due to changes from finding certain coping behaviors to previously have been *Moderately Helpful* at baseline to *Extremely Helpful* at follow-up time points.

It is believed that this study may not have been sufficiently powered to detect changes in coping behaviors over time following participation in a workshop to increase problem-solving skills. Also, another potential reason for lack of change would be characteristics of the participants who decided to participate in the study already had adequate coping behaviors in place prior to participation in the study and therefore, coping behaviors were not affected by participation in the workshop.

As hypothesized, parental worry decreased following participation in the problem-solving workshop. Many of the worries that are seen in parents of LQTS patients include worry surrounding the condition, lack of knowledge of what LQTS is, attempting to understand why the diagnosis occurred, lack of understanding about the cardiac events that had happened previously (either to the child, themselves, or another family member diagnosed with LQTS), uncertainty about events that could happen in the future, and decisions regarding their children's best treatment options (Anderson *et al.*, 2005; Farnsworth *et al.*, 2006). For the parents in the current study, worry seemed to center on the uncertainty about events that could happen in the future. By providing parents and children diagnosed with LQTS the skills necessary to engage in effective problem solving, it decreased the worry of the parents about the future and whether their children could handle LQTS-specific situations in an effective manner. It is important to note, however, that the instrument used to measure worry was not LQTS-specific, but rather related to worry in chronic illnesses. Therefore, further research is needed to determine if decreases were in disease-related worry or worry in general.

The changes seen in scores for hope were in general above the norm. The score for participants in the current study were both above those of the normative group prior to

participating in the problem-solving workshop. Both participant's scores remained above those of the normative group throughout their participation in the research study, with one participant having a slight decrease in hope, while the other participant had an increase in hope. Hope theory views hope as a goal-directed, positive motivational state that is based on the interactions derived from agency and pathway planning (Snyder, 1994, 2000). Parents with high hope, including higher scores on the agency thoughts and pathway thoughts, may be able to view problems they face related to LQTS as an attainable challenge, rather than an obstacle, and may tend to find more efficient ways to solve the problems. Hope is likely a motivating factor for engaging in effective problem-solving skills, especially for individuals diagnosed with chronic illnesses and their caregivers.

Clinical impressions.

A clinical anecdote discussed during the workshop is offered to demonstrate the feasibility and effectiveness of the problem solving workshop for children with LQTS and their parents. As part of the effort to provide participants with opportunities to meet others with LQTS and discuss their life experiences, the group leaders provided the participants with moments in which to share their personal experiences. One child shared his experience of a time when he was required to wear a Holter monitor for a period of time while he was in school. When he presented to class, the teacher misinterpreted the Holter monitor as a bomb and sent the child to the school principal. The child and parent described this situation as frightening, embarrassing, and humiliating. After describing the experience, the other child in the group validated the feelings of the child who wore the Holter monitor, and the parents were able to discuss their common reactions to the

child's experience. This further emphasizes qualitative data that the group was deemed to be effective by the participants.

Recruitment issues.

The original goal of this pilot study was to include five to ten dyads of adolescents diagnosed with LQTS and their parents. After the first mailing of invitations to participate from an electrophysiologist, a few participants responded. The investigators experienced difficulty scheduling the first group of adolescents (aged 13 to 17 years); therefore, they decided to decrease the age of participants to 9 years-old. After attempting to contact these individuals, a group that would have included six children and four adults was formed. Unfortunately, one family dropped out due to the child's fear of being different, and a second family with three children did not come to their scheduled workshop, despite confirmatory phone and e-mail contact. The group was therefore conducted with two child and parent dyads.

Further recruitment was attempted via a second mailing to the electrophysiologist's patients, announcements of the workshop at tristate area hospitals and clinics, postings on SAD Websites, LQTS-related Facebook pages, Craigslist, and through contact with other local cardiac organizations. This increased recruitment effort yielded minimal increase in responses. The investigators therefore explored actual interest in the topics of the workshop. The following announcement was posted on the PCOM LQTS Research Team Facebook page in order to gain a better understanding of the limited response to the workshop.

As many of you have seen our posts, we've been offering studies including a free workshop for children with LQTS offered in the Philadelphia area,

however, we have not received high responses. We are trying to brainstorm other ways to reach those who may be interested or understand why our workshop may not be of interest. We greatly appreciate all of your help and feedback! (Post by study investigators on Facebook).

Feedback indicated that individuals were interested in participating in the workshop, but barriers to participation included time, money, child's age, energy to travel, location (living outside of the tristate area), and fear of feeling different. Unfortunately, many of these concerns were unable to be addressed or modified to increase recruitment.

Facebook users provided recommendations for alterations in the protocol to reach more individuals, which included podcasts, online workshops, Google to distribute handouts of materials, and providing significant transportation help (i.e., paying for plane tickets).

Of the barriers to participation that were identified, it was determined that it was feasible to modify the child's age in the current protocol, which was believed would identify more participants for the workshop. The requirement for age of child participants was lowered to 8 years old, in the hopes of identifying enough participants to conduct a second workshop. A second workshop group was formed with two children (ages 8 and 9) and their parents who were interested in participating. Unfortunately, due to unforeseen scheduling circumstances for the participants, this workshop was not held. Recruitment was discontinued at that time. These barriers to participation and suggestions from Facebook users are important ways in which this protocol for a one-time problem-solving workshop can be further enhanced for future studies.

Feasibility and effectiveness.

Concerns may be raised about the feasibility and effectiveness of the workshop, given the small number of participants who were able to attend. The definition of *feasible* is “capable of being done or carried out” (feasible, 2015). Therefore, as the workshop was able to be conducted in a group format with multiple participants, the workshop can be defined as feasible. The definition of *effective* is “producing a decided, decisive, or desired effect” (effective, 2015). Therefore, due to the changes noted in participant scores, the workshop can be defined as effective. However, the effectiveness of the workshop is not generalizable to the greater population of parents of children who have been diagnosed with LQTS given the small sample size utilized in this pilot study. The limitations of the study need to be taken into consideration when examining the results of the study and further research is needed in order to determine the exact effects of the workshop on children diagnosed with LQTS and their parental caregivers.

Limitations.

As detailed above, one of the major study limitations is the small sample size. The small sample size limits the generalizability of these findings to the larger population of children diagnosed with LQTS and their parents, therefore limiting external validity. However, the participants recruited for participation in the problem-solving workshop had life-threatening medical conditions and related psychosocial stressors. Future considerations for use of this protocol with children diagnosed with LQTS and their parents should consider other methods of delivery.

Another limitation is that the treatment was not tailored to the parents who were involved in the study. Parental problem-solving skills were measured and parents were

included in the workshop; however, the workshop was directed toward the children and the issues that they face due to their LQTS diagnosis. The issues that the parents face, such as having a child diagnosed with a life-threatening illness, such as LQTS, were not discussed in the context of the workshop. Nevertheless, this is consistent with other psychosocial interventions that have focused the content of the intervention on the issues of the patient diagnosed with the chronic condition rather than on the parent's or caregiver's concerns (Bloomfield *et al.*, 1990; Nezu *et al.*, 2003; Scholten, *et al.*, 2013). If a parents-only workshop were provided, topics might include issues that parents face raising a child diagnosed with LQTS (e.g., sex, drugs, caffeine intake, dating), problem-solving skills for parents related to their child's LQTS, and managing spousal differences in raising a child. A parents-only workshop may influence the amount of change seen on study-specific variables, such as problem-solving abilities, hope, worry, and coping skills.

A third limitation is that the results of this pilot study may not be generalizable to other populations or parents of children who have been diagnosed with other potentially life-threatening chronic illnesses or for the general population. The role of SPS skills may be very different for individuals diagnosed with other chronic conditions compared to children diagnosed with LQTS and their parents. Furthermore, there may have been participants included in the study who are already effective problem-solvers; therefore, change scores for problem-solving skills, coping, hope, and worry would not have been as great as for ineffective problem-solvers. One-time psychosocial interventions have been found to be feasible for populations including women diagnosed with gynecological cancer discussing issues and concerns of the cancer diagnosis (Powell *et al.*, 2008),

reappraising married couples' conflict (Finkel, Slotter, Lunchies, Walton, & Gross, 2012), and psychological debriefing following trauma (Rose, Bisson, & Wessely, 2003), although there were not any identified studies for children diagnosed with a chronic illness and their caregivers.

Another limitation is that problem-solving interventions have not been empirically tested for individuals diagnosed with LQTS, including empirically supported evidence for the types of LQTS-related examples that were used in the workshop. There are no recommended examples in the literature that must be addressed in problem-solving interventions with children diagnosed with LQTS. Therefore, this may limit the generalizability of the results, given that the LQTS-related examples were the primary means of teaching problem-solving skills to the children and their parents.

The combination of parents and children in the same group may be a limitation of the current study, as it may limit learning of the problem-solving skills for both parents and children due to parent-child relationship variables that are extraneous to the goals of the current study. One intervention that included both parents and adolescents in the same behavioral group intervention to treat pain associated with inflammatory bowel disease also included a communication and limit-setting component, which seemed to influence the parent-child relationship and indirectly benefited the adolescents' ability to utilize the problem-solving skills (Hayutin, Blount, Lewis, Simons, & McCormick, 2009). The current pilot study did not include a communication and limit-setting component due to time constraints, and it is unknown how having both parents and children in the intervention group may have affected learning of the problem-solving skills.

Unlike other problem-solving interventions in the literature, this one-time intervention format did not allow participants to practice and further develop their problem-solving skills, which may be another limitation of the current pilot study. Participants did not have the ability to take time to practice the skills and then return back to the group or to the group leaders to work through problems with implementing the problem-solving skills, as was available in all of the other problem-solving interventions currently in the literature.

There was likely a selection bias for those participants who enrolled in the pilot study. Parents and children who responded to the letter/brochure to participate in the workshop may have significantly differed from those who did not choose to respond to the invitation for participation. The researchers were unable to determine how exactly the participants who responded to the invitation differed from those who did not respond.

Although there are indications that the intervention was feasible and effective, the inconsistent effects of other measured variables (problem-solving abilities, worry, hope, coping) may limit the conclusions. However, the noted changes in score for the participants suggest that the intervention provided during the workshop was informative, worthwhile, and helpful to the participants. The variability in follow-up scores indicates that future problem-solving workshops and interventions may need to include a component of reinforcement of the skills. For parents, this could include guidelines on how to reinforce use of problem-solving skills of their children, and perhaps, may include telephonic coaching at 1- and 3-months post workshop.

Conclusion.

Outcomes of this study have important implications for the LQTS population, given that there are currently no other psychosocial treatments available for families affected by LQTS. There are a number of qualitative studies that have examined some of the psychological issues that parents of children diagnosed with LQTS face, such as frustration, fear, distress, guilt, sadness, loss, and problems coping with the diagnosis (Farnsworth *et al.*, 2006; Gonzales, 2009; Hendricks *et al.*, 2005; Rovinsky, 2010). Determining the efficacy and feasibility of a psychosocial intervention aimed directly at some of the specific LQTS related problems that parents face helps add to the available psychosocial literature for this population. Problem-solving skills are important in the management of LQTS, due to the daily needs of patients and their families to ensure that they minimize the likelihood of a cardiac event, such as managing water intake, avoiding swimming pools or medications that may prolong the QT interval, and carrying an AED, just to name a few. Teaching children to manage these behaviors on their own may allow parents to worry less about the child's actions that could potentially initiate clinical manifestations. Family members, especially parents, play invaluable roles in the child's adjustment to a chronic illness, but are also affected themselves by the patient's condition, activity restriction, and need for emotional support or physical assistance (Martire, 2005).

Future research should continue to examine the feasibility and effectiveness of social problem-solving treatment with a larger sample of children diagnosed with LQTS and their parents. The addition of guidelines for helping parents' reinforcement of problem-solving skills with their child once the workshop has been completed or the use

of telephonic coaching for parents at scheduled follow-up points may also be beneficial for the parental participants in the workshop. Furthermore, research should also continue to explore the relationship between social problem-solving skills, coping, hope, and worry in parents of children diagnosed with LQTS. Not all of these variables have been specifically evaluated and investigated for parents of children diagnosed with LQTS, and understanding the relationship between these variables will help to enhance the future outcomes of one-time social problem-solving workshops with this population. Finally, further research should examine the direct benefits of learning social problem-solving skills for distressed parents of children who have been diagnosed with LQTS. Evaluating the effects of a workshop with a distressed population will enhance the generalizability and outcomes of a social problem-solving workshop with children diagnosed with LQTS and their parental caregivers.

Despite the limitations, the *Living Life with LQTS Workshop* appears to be a promising addition to care for children with LQTS and their parents. To date, this workshop is the first intervention study designed specifically to address problem-solving skills for children and adolescents with LQTS and to include parents in the workshop to help teach and coach the skills to their children. The participants in this study were taught social problem-solving skills to help manage everyday stressors related to their LQTS. The workshop appeared to be a feasible and effective psychosocial intervention for this population, although further research is needed.

The limited psychosocial interventions related to the specific problems that individuals diagnosed with LQTS encounter, combined with research that has shown that patients with LQTS face psychosocial issues similar to those faced by patients with other

cardiac diseases, make the results of this feasible intervention an important contribution for physicians and medical professionals working with children with LQTS (Czosek *et al.*, 2015; Waldron, Felgoise, Tress, Lawrence, & Vetter, 2013). The results of this study provide physicians and medical professionals with opportunities to refer patients and their parents to mental health providers who offer problem-solving treatment as part of collaborative care. Overall, this study demonstrated the feasibility and efficacy of a problem-solving workshop for children diagnosed with LQTS and their parents.

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