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# Online resource for parents of children with congenital hand differences

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BOSTON UNIVERSITY  
SARGENT COLLEGE OF HEALTH AND REHABILITATION SCIENCES

Doctoral Project

**ONLINE RESOURCE FOR PARENTS OF CHILDREN  
WITH CONGENITAL HAND DIFFERENCES**

by

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Submitted in partial fulfillment of the  
requirements for the degree of  
Doctor of Occupational Therapy

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## **DEDICATION**

This work is dedicated to all who are living with a limb difference, and to all of the parents raising fierce children with limb differences. Through your strength, resilience, and kind hearts, you are building a more inclusive society where we can all be proud of and empowered by our bodies. Thank you.

## ACKNOWLEDGMENTS

Thank you for my mentor and surgeon Dr. Michelle James, for your continued support, guidance, and motivation throughout my life. Your work throughout your career has built a better world for all of us with limb differences, I am so grateful to work alongside you. Thank you Dr. Ellen Cohn for your academic mentorship and guidance.

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Thank you, mom, dad, and Madeline, for being my family support system, for believing in me, for having high expectations of me, and for encouraging me to follow my heart all over the world. You have taught me to be independent, resilient, and kind. Cannot wait for our next adventure!

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WITH CONGENITAL HAND DIFFERENCES**

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**ABSTRACT**

Children with congenital hand differences (CHD) are at risk for social isolation, increased anxiety, and lower rates of self-esteem when compared to their typically developing peers (Lumsdaine et al, 2016). Parents may feel guilt, isolation, and may have perceived lack of resources to best support their children (Ardon, Janssen, Hovius, Stam, & Selles, 2012; Murray, Kelley-Soderholm, & Murray, 2007). Goffman (1963) explained that society perceives that individuals with differences belong to the “other” category deprived of social privileges which are standard to those in the “in-group”. Many children with congenital hand differences and their parents have limited access to the resources to manage and cope effectively with the negative assumptions about their ability.. This doctoral capstone project describes the development of an online resource for parents of children with congenital hand differences. The content for the webpage is based on literature, clinical and personal experiences. The online resource was designed to increase feelings of support, community, and effective coping for parents of children with CHD.

Fourteen parents were recruited via social media platforms to review and evaluate the website. Responded evaluated the ease in website navigation, the usefulness of content, and rated their likelihood to use and to recommend the website. Parents on

average found the website easy to navigate, useful, and were likely to use, and recommend the website.

**Keywords:** Congenital hand differences, congenital limb differences, online resource, parent resource, parents

## TABLE OF CONTENTS

DEDICATION.....	iv
ACKNOWLEDGMENTS.....	v
ABSTRACT.....	vi
TABLE OF CONTENTS.....	viii
LIST OF FIGURES.....	xiii
LIST OF ABBREVIATIONS.....	xiv
CHAPTER ONE.....	1
Problem.....	1
Introduction to the Problem.....	1
Stigma.....	2
Description of the Explanatory Model of the Problem.....	5
Environmental factors.....	5
Personal factors.....	10
Problem.....	13
Proposed Solution.....	13
Stakeholders and Policy.....	15
Key stakeholders.....	15
Policy.....	17
Stakeholders and policy.....	18
CHAPTER TWO.....	19
Evaluation of the Evidence.....	21

Evaluative Summary .....	22
Child experience.....	22
Parent Experience.....	25
Coping. ....	26
CHAPTER THREE.....	31
Online Resources .....	31
Website Content.....	32
Resources.....	34
Faculty Mentor.....	34
Site Mentor.....	34
Digital Access. ....	35
Parents of and children/teens/ adults with CHD.....	35
Health Care Professionals.....	36
Budget.....	36
Activities.....	36
Literature Review.....	36
Interview Questionnaire.....	36
Website review.....	36
Outputs.....	36
Outcomes .....	36
Most accurate targeted information.....	36
Impact.....	37

Program Specifics .....	37
Detailed Weekly Activity Plan .....	39
Barriers/Challenges and Proposed Solution .....	40
Time management .....	40
Website formatting difficulties .....	40
Sustainability .....	40
CHAPTER FOUR.....	41
Evaluation Plan .....	41
Website Objectives.....	41
Outcome Cohort.....	42
Website Evaluation .....	42
Data Analysis .....	43
Limitations.....	45
CHAPTER FIVE .....	46
Target Audience.....	46
Primary Audience.....	46
Secondary Audience.....	46
Key Messages .....	47
Primary Audience.....	47
Secondary Audience.....	47
Sources/ Messengers .....	47
Primary Audience.....	47

Secondary Audience.....	48
Dissemination Activities .....	48
Special Education Opportunity.....	48
Submission for AOTA.....	48
Shrine Vine.....	49
Social Media.....	49
Flyer.....	49
Electronic communication.....	49
Presentation at Pediatric Hand Study Group (PHSG).....	50
Budget .....	50
Special Education Opportunity.....	50
Submission for AOTA.....	50
Shrine Vine.....	50
Social Media and flyers.....	50
Presentation at PHSG.....	51
Evaluation.....	51
Primary Evaluation.....	51
Secondary Evaluation.....	51
CHAPTER SIX.....	52
APPENDIX A: WEBSITE EVALUATION MEASURE.....	54
APPENDIX B: SPECIAL EDUCATION INVITATION.....	56
APPENDIX C: SHRINE VINE.....	57

APPENDIX D: ELECTRONIC FLYER.....	58
APPENDIX E: WEBSITE ADDRESS.....	59
APPENDIX F: EXECUTIVE SUMMARY.....	60
BIBLIOGRAPHY.....	63
CURRICULUM VITAE.....	69

## LIST OF FIGURES

Figure 1. Theoretical Model of the Problem .....	5
Figure 2. Proposed Logic Model .....	34
Figure 3. Sample landing page .....	38
Figure 4. Proposed Logic Model .....	41
Figure 5. Website Evaluation Graph.....	44

## **LIST OF ABBREVIATIONS**

AOTA	American Occupational Therapy Association
BU	Boston University
CHD	Congenital Hand Difference
HCP	Health Care Professionals
HRQoL	Health Related Quality of Life
OTD	Occupational Therapy Doctorate
PHSG	Pediatric Hand Study Group
SES	Social Economic Status
SHCNC	Shriners Hospitals for Children – Northern California
STI	Sexually Transmitted Infections

## **CHAPTER ONE**

### **Problem**

Children with congenital hand differences (CHD) may experience bullying and social isolation, but lack access to psychosocial and accommodation needs. However, there are few existing resources tailored to this population. For example, many psychosocial resources and recommended accommodations are designed for a general group of children with disabilities.

### **Introduction to the Problem**

Children, who have congenital hand differences (CHD), face the unique question “what happened to your hand?” Many children, parents, and medical professionals often do not know the reasoning behind the difference and are often are unaware of a difference until birth. Parents experience grief related to the loss of the children they imagined (Andrews, Williams, VandeCreek, & Allen, 2009; Angliss, 1981; De Jong, Reinders-Messelink, Janssen, Poelma, Van Wijk, & Van der Sluis, 2012; Murray, Kelley-Soderholm, & Murray, 2007). Numerous authors have reported that children who have physical appearance differences are limited in their social participation engagement. Physical and social barriers exclude children with disabilities (Knapp et al, 2013; Knight et al, 2013; Lumsdaine et al, 2016). Children with disabilities are at a high risk for social isolation, are more likely to have social anxiety in relation to peers , are less likely to engage in extracurricular programming (like Scouts and Brownies), are more likely to develop a negative self-body image, have emotional distress in relation to their disability, perceive stigma, and have feelings of treatment as “non-normal” (Knapp et al, 2013;

Knight et al, 2013; Lumsdaine et al, 2016). These feelings of separate, and “non-normal” prevent those with differences from engaging in social activities, an issue which falls within the scope of practice for Occupational Therapy as “social participation.” The American Occupational Therapy Association (AOTA) describes social participation “the interweaving of occupations to support desired engagement in community and family activities as well as those involving peers and friends” (AOTA, 2014, p. s21).

Children with CHD experience social, emotional, psychological and functional challenges. The chapter discusses the impact of these challenges on children’s school and social occupations.

### **Stigma**

Goffman’s Stigma Theory (1963) provides a useful theoretical lens to understand the experience of children with CHD. Individuals who have a physical disability may be perceived as “non-normals” by those without disabilities (Goffman, 1963). Children with a CHD may face difficulty integrating into the larger society, and their insecurities about their limb difference may be exposed and scrutinized. Society perceives individuals with differences as belonging to an “other” category, or “out-of-group”, deprived of social privileges which are standard to those in the “in-group”. For example, the “in-group” might assume that children with a CHD are “limited” in their ability to engage in various childhood occupations such as playing the violin. For a hypothetical example, a child with a CHD enters a music class where the music instructor and classmates have never interacted with someone who had a CHD. The child shows interest in the violin. The instructor and classmates assume that playing the violin requires the full use of two

“normal hands”. The instructor instead suggests that the child play the tambourine, the child agrees. This assumption stigmatizes the child with CHD and thus denies the child the opportunity to explore the violin and explore potential modifications or adaptations. The instructor’s assumption prevents the child from the agency to choose between the violin or the tambourine. The child feels the stigma, and by agreeing to the tambourine, the child denies himself/herself the opportunity to explore the instruments. The child internalizes these social limitations with the assumption such as “I cannot play the violin, I do not have two hands.”

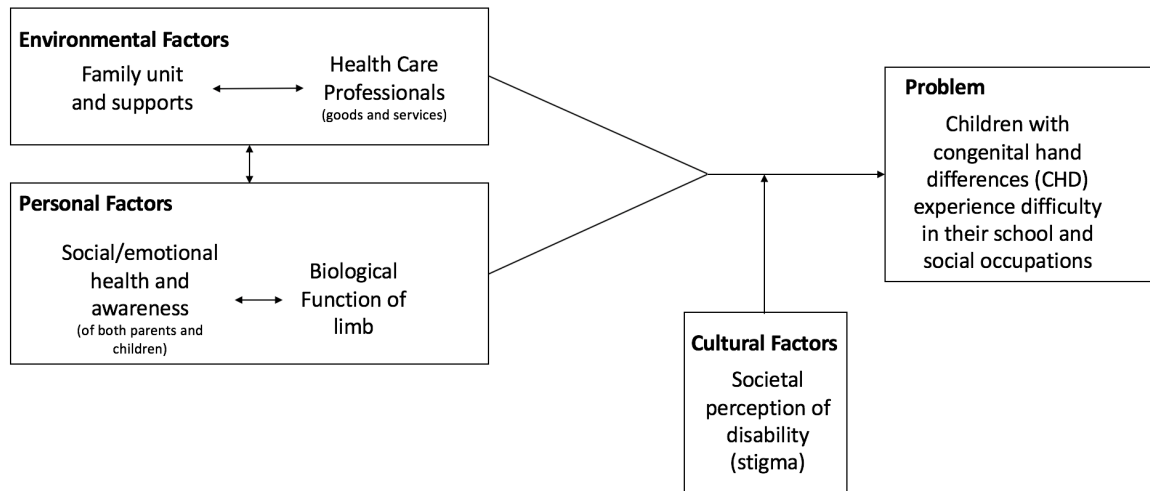
Society shuns individuals with physical differences making it difficult for them to identify and align with the “in-group”. They have difficulty aligning with the “out-of-group” because they may believe that they do not belong there and do not believe the social limitations apply to them (Goffman, 1963). Goffman (1963) proposed that individuals may choose to hide their differences in attempts to pass as “normal” to be in the “in-group”, or they may internalize the social and functional limitations projected on them and identify with the “out-of-group” (Goffman, 1963). He also suggested that individuals who identify with the “out-of-group” may spend a significant amount of time and effort attempting to reconcile their lack of alignment with either group (Goffman, 1963). This effort to align with a group results in a heightened self-awareness and a broader social awareness. This heightened awareness is a means of education to promote the social inclusion of “non-normal” groups (Goffman, 1963). In returning to the violin example, the child may develop the motivation to attempt the instrument, find adaptive strategies. The child’s ability to play the violin helps educate their peers that a child with

CHD, is in fact, capable of this occupational engagement with adaptive strategies. Educating others about the potential abilities of children with limb differences can be a complicated process, and children may not have the social and emotional awareness or coping skills to advocate for themselves effectively.

Every component of the hypothesized explanatory model (see figure 1) of the factors that create social isolation for children with limb differences incorporates Goffman's theoretical view of social stigma. This explanatory model of the problem proposes that children with a disability may not have the social and emotional awareness to understand or challenge the social structures that are placed on them, requiring parental support. The explanatory model includes the perspective of the family unit and children with CHDs who are navigating the implications of social stigma. The model suggests that social perceptions may influence the problems children with CHDs may confront in their school and social occupations.

Although Goffman's Social Stigma Theory is not applied directly to the experiences of children with CHD in the literature, the Social Stigma Theory has been applied to young women's likeliness for sexually transmitted infections (STI) testing. In US society, there is a tremendous stigma associated with STI. One study found that young women were less likely to seek testing for STI due to the associated stigma; women preferred to pass as "normal" (Balfe, Brugh, O'Connell, McGee, O'Donovan, & Vaughan, 2010). In another study of disclosure in the workplace among people with multiple sclerosis, individuals attempted to mask their multiple sclerosis and pass as "normal" in order to avoid the stigma associated with their disability (Vickers, 2017).

These studies suggest that social stigma does exist and that Goffman's Social Stigma Theory may be a useful theoretical lens for understanding the experiences of children with CHD.



**Figure 1. Theoretical Model of the Problem**

### **Description of the Explanatory Model of the Problem**

The visual model represents the relationship between environmental factors, including family units/supports and health care professionals, and the personal factors which include the children's social/emotional health awareness and the biological function of the limb. This model suggests that four components interact to contribute to problems that children with CHD experience.

#### *Environmental factors.*

Family unit/support. Parents and the parental support system (including extended family, friends, other parents of children with CHD or similar conditions) may have an impact on the parent's ability to support their children. Researchers have documented that when parents of children with CHD have extended supports and the ability to accept and

embrace their children's differences, they can better support their children (Bae, Canizares, Miller, Waters, & Goldfarb, 2018; Murray, Kelley-Soderholm, & Murray, 2007). When parents do not agree with each other about treatments or how to respond to their children's emotional comments such as "I wish I had two hands", they may experience marital discord and increased stress. This disagreement may impact their ability to effectively support their children (Angliss, 1981; Murray, Kelley-Soderholm, & Murray, 2007; Weir, Ephraim, & Mackenzie, 2010). Similarly, if parents are not able to support themselves by developing positive coping and self-acceptance, then they may be less likely to effectively support their children (Beresford, 1999; Murray, Kelley-Soderholm, & Murray, 2007). Parents can benefit from developing relationships with other parents of children with similar differences to create and broaden their support system (Murray, Kelley-Soderholm, & Murray, 2007). Parents may not know how to advocate for medical treatments or devices that may be helpful for their children if they do not have support. Parents may experience an economic burden as a result (or may not have the initial resources to access) treatment options and medical devices for their children (Weir, Ephraim, & Mackenzie, 2010).

Stigma. Parents may experience stigma. Often because, parents do not have limb differences themselves, they may have difficulty in imagining a life of happiness and success for their children. They may grieve the loss of the limb (Andrews, Williams, VandeCreek, & Allen, 2009; Angliss, 1981; De Jong, Reinders-Messelink, Janssen, Poelma, Van Wijk, & Van der Sluis, 2012; Murray, Kelley-Soderholm, & Murray, 2007). These emotional responses perpetuate the view that children with CHD have significant

limitations in their functional ability. That is, parents may reinforce the stigma by assuming that their children have limitations. Sheffler and colleagues (2009) conducted a study to understand how function and quality of life are perceived by children with CHD and their parents. The researchers found that parents consistently reported lower function than what the children self-reported. The researchers were unable to determine the causation for conflicting reports. However, they speculated a few reasons. First, they suggested that parents may have had a heightened awareness of the children's abilities and that the children inflated their engagement ability in the activity. Alternatively, the researchers suggested that the parents may have perceived their children as having a disability and therefore assumed their children are unable to fully engage in the activity (Sheffler, Hanley, Bagley, Molitor, & James, 2009).

The concept of perceived disability is congruent with Goffman's theory regarding the "in-group" perception of those in the "out-of-group" alignment. When parents are the "in-group" participants and make negative assumptions about their functional ability and quality of life for their children who are "out-of-group" participants (Bae, Canizares, Miller, Waters, & Goldfarb, 2018; Goodell, 2017; Murray, Kelley-Soderholm, & Murray, 2007). In 1981, Angliss wrote an article advocating for the early referral for children with CHD to specialized health care professionals. Although this article was written several decades ago, the language illustrates some of the stigmas that may still exist in society today. The language is negative and suggests catastrophic consequences such as a "crisis for the whole family", "guilt and distress over having a deformed baby", "the blight of a malformation, particularly where [the difference] is visible and hence embarrassing, is a

crushing blow to the parents and to everyone else who has shared in the event" (Angliss, 1981, p. 141-142). Although parents of children with CHD may not honestly believe these statements today, Angliss's writing suggests that these beliefs existed in 1981. In an article published almost 20 years later, Beresford (1999) reported "it is likely that family and friends are reluctant to handle the malformed hand for fear of the unknown" (p. 113). Beresford's more recent article provides evidence that this stigma was maintained for 20 years and suggests that this stigma exists today.

Health care professionals. CHD is an uncommon condition. Although the numbers vary across research studies, a recent study reported that there are 16 children born with CHD per 10,000 live births (Ardon, Janssen, Hovius, Stam, & Selles, 2012). Health care providers interact with the family and children in a range of settings, from family practice offices to acute care hospitals. Some health care professionals specialize in hand and limb differences; however, general health care providers may have little experience with this condition. Hands are not a priority in ultrasound imaging, and parents may not be aware of their children's limb difference until birth. CHD is not a life-threatening condition and is not heavily researched, providing an unclear prognosis for health care professionals. Moreover, many health care professionals may not have access to knowledge or resources to best support families with children who have CHD (Andrews, Williams, VandeCreek, & Allen, 2009).

Stigma. Health care providers may have a stigma towards children with CHD and may express perceived pity or reinforce the perception of limitations for the children. They may also project discomfort in interacting with the children. Both of which further

reinforce social norms that these children fall into the "out-of-group" social system.

Parental unit and supports and health care professionals. The explanatory model is drawn to reflect a bidirectional relationship between parental supports and health care providers. If parents have a robust support system, they are more likely to work with specialized health care professionals. They are also more likely to have access to accurate health information, resources, and other specialized professionals (e.g., prosthetists, hand therapists, and hand surgeons). If parents have a robust support system, and are unsatisfied with health information they receive, they are more likely to seek out different opinions from other health care professionals. When parents have access to accurate health information and resources/services provided by the health care professionals, they develop a stronger support system for their children.

More confident and accepting health care professionals may help parents accept their children with CHD and help to develop a more positive understanding of their children's functional abilities (Beresford, 1999). If health care professionals are uncomfortable in working with children with CHD, then this discomfort may impact parent's acceptance of their children's disability. This decreases their desire to seek out and work with health care professionals in the future. Awkward, uncomfortable, or harmful encounters with health care professionals may limit the possibility of parents receiving accurate health information and resources for their children, thus diminishing the parents' support network (Murray, Kelley-Soderholm, & Murray, 2007).

*Personal factors.*

Social and emotional health and awareness. The personal factors described in the explanatory model include both the children's and parent's understanding and perception of CHD, including personal factors such as emotional functioning, social functioning, school functioning, anxiety, and depression. Children with CHD typically score high on social and emotional health until they enter middle school and high school where some adolescents begin to experience higher rates of depression and anxiety and lower scores on social functioning (Ardon, Janssen, Hovius, Stam, & Selles, 2012).

Stigma. Stigma may influence the personal factors described in the explanatory model. For example, children may encounter the social consequences related to "out-of-group" experiences for the first time as they age into middle school. Children may not have developed the social and emotional awareness to reconcile these "out-of-group" experiences. Researchers reported that children with CHD in late elementary through high school ages had negative experiences when initially meeting people (Bradbury, 2006; Michielsen, Van Wijk, Keterlarr, 2010; Franzblau et al., 2016). In attempts to avoid negative experiences, some youth with CHD developed various methods to conceal their differences. For example, they hid their affected limbs in cosmetic prosthetic devices, in long sleeves, or in pockets in efforts to pass as "normal" (De Jong, Reinders-Messelink, Janssen, Poelma, Van Wijk, & Van der Sluis, 2012; Johansen, Dammann, Øinæs Andersen, & Andresen, 2016; Murray, Kelley-Soderholm, & Murray, 2007). Authors reported that those children 8-20 years in age, who concealed their limbs, reported higher self-acceptance than those who did not conceal their limbs (De Jong,

Reinders-Messelink, Janssen, Poelma, Van Wijk, & Van der Sluis, 2012). In the same study however, those who concealed their CHD reported less ability to cope with negative social experiences (De Jong, Reinders-Messelink, Janssen, Poelma, Van Wijk, & Van der Sluis, 2012). Conversely, those who did not conceal their arms reported in the study more effective coping, but lower self-acceptance when compared to their peers who engaged in concealment behaviors (De Jong, Reinders-Messelink, Janssen, Poelma, Van Wijk, & Van der Sluis, 2012). Ardon 2012 reports, children "do not have a negative self-sense until they are older and able to perceive stigma" (Ardon, Janssen, Hovius, Stam, & Selles, 2012, p. 356).

#### Social and emotional health and awareness and functional capacity of the limb.

The explanatory model (figure 1) is drawn to reflect a bidirectional relationship between the children's social and emotional health and awareness and biological functioning capacity of their limbs. Children who have higher scores of self-awareness and self-acceptance are less likely to engage in concealment behaviors when they are in the presence of peers who do not have CHD. Children who have higher self-esteem, do not engage in concealing practices, and feel confident and empowered, may be likely to engage in other novel activities. This mastery of experiences may encourage the children to continue seeking out these types of activities. On the contrary, if children experience stigma related to their CHD, they may be likely to use "disengagement coping mechanisms" such as concealment of their CHD (De Jong, Reinders-Messelink, Janssen, Poelma, Van Wijk, & Van der Sluis, 2012). This concealment decreases self-awareness and self-acceptance. If children are hiding their CHD in the presence of peers, then they

are limiting the functional capacity of their affected limbs. When children experience bullying or social isolation when attempting to engage in an activity, they may be less likely to engage in future experiences and more likely to conceal their limbs. Limb concealment limits functional capacity of their limbs. Conclusions drawn from research indicates that children with CHD are able to engage in their desired occupations effectively, however they struggle with social and emotional wellness (Ardon, Janssen, Hovius, Stam, & Selles, 2012; Johansen, Dammann, Øinæs Andersen, & Andresen, 2016; Mano, Fujiwara, & Haga, 2017; Sheffler, Hanley, Bagley, Molitor, & James, 2009).

Environmental factors and personal factors. The explanatory model depicts a bidirectional relationship between the environmental (family and health care supports) and personal factors (biological functioning and social and emotional health) relating to children with CHD. When parental supports and health care supports are strong, then children are more likely to have higher self-awareness and use the full functional capacity of their limbs (Bae, Canizares, Miller, Waters, & Goldfarb, 2018; De Jong, Reinders-Messelink, Janssen, Poelma, Van Wijk, & Van der Sluis, 2012). Conversely, when parental supports and health care supports are weak, children with CHD are more likely to have decreased self-awareness, and limited functioning of their limbs. Similarly, when children have decreased biological functioning and decreased social/ emotional health, then these limitations impact the environmental factors. This limits the children's overall engagement in necessary and desired occupations.

Cultural factors. In contemporary culture, the overall social perception of disability is negative. Society perceives those who have a disability as “non-normal” and

part of the “out-of-group” with social limitations. The proposed explanatory model suggests that stigma modifies the relationship between the environmental factors, the personal factors, and the problem. If social stigma is low, then, in theory, the problems experienced by the children will also be low. However, if social stigma is high, the stigma threatens the other factors in the model and increases the problems children experience when engaging in their school and social occupations.

### *Problem*

The explanatory model proposes that environmental factors and personal factors have a direct relationship with children with CHD experiencing difficulty in their school and social occupations. This model outlines the relationship between the environmental, personal, and the influence of cultural factors on children’s ability to effectively engage in their school and social occupations. Some children may experience less problems, and others may experience more problems. However, conclusions drawn from the literature note that children with CHD face social acceptance challenges throughout their life course.

### **Proposed Solution**

The purposed solution of this problems is the collaboration with Shriners Hospital for Children – Northern California (SHCNC) and the creation of an online resource for parents. This website provides parents with a community and supportive information as their children transition to school and engage in social occupations. The resources are included within the Shriners Hospital for Children – Northern California website to provide easy and reputable access to parents. The home page features a welcome section

and site overview with tabs to content in the following 4 domains: (a) parent experience, (b) child experience, (c) school resources and (d) additional resources. Each of these tabs include specific resources.

The online resource provides parents with medically accurate and applicable information. According to researchers, health information delivered via websites is effective for communicating health information (Sium, Giuliani, & Papakos, 2015). A majority of people in the United States have access to the internet, which is available at all times (Sium, Giuliani, & Papakos, 2015).

The information is a part of a reputable organization's website SHCNC, thus increasing the user's trust of accurate information (Sium, Giuliani, & Papakos, 2015). The top priorities for parents seeking information to support their children with CHD include parent experience, child experience, school resources, and additional resources, therefore this website is organized under these headings. The website contains a "Parent Experience Page" resource that synthesizes information from the literature about common parent experiences to decrease feelings of isolation. A "Child Experience Page" synthesizes information from the literature to help parents envision a future for their child. The third section of the website covers school resources. This section reviews available resources to support children with CHD in the school context, IEP, and 504s plans. The final section is a resource section with links to further information such as children's books, camps, organizations, and social platforms. When parents have a supportive network, they can more effectively support their children. This section aims to connect parents to other individuals with similar conditions.

All of the sections of the online resource integrate statements from children and parents with various viewpoints to reinforce the concept that the parents can choose what method of health care, communication, and intervention work best for families and their children. For example, some children enjoy using humor as a way to communicate the limb difference, whereas others avoid the questions, and others state what happened. The goal of this website is to provide accurate information in an accessible manner to empower parents to support their children.

The most significant barrier children face is stigma. This website is designed to support parents of children with CHD to support their children to address the stigma associated with CHD.

### **Stakeholders and Policy**

The capstone project operated within the Shriners Hospitals for Children – Northern California (SHCNC) system. Inherently stakeholders and policy are involved. This section briefly describes their roles.

*Key stakeholders.* Key stakeholders, relevant to this doctoral capstone project are the children themselves, parents, health care professionals (HCP), and the Shriners Hospitals for Children – Northern California (SHCNC) system. Children are the primary and ultimate recipients of care; however, they are young and unable to direct their caregivers how to best provide the social and emotional support necessary for their health and well-being. However, if parents have access to resources, then the children can develop adequate resiliency, self-awareness, self-confidence, and coping strategies. By supporting children in these ways, they may have lower rates of social exclusion,

depression, and anxiety and higher rates of social functioning. Before preparing an online resource for parents, it will be essential to gather input via interviews from the children and adults with CHD to ensure that the information provided on the website accurately reflects their experiences.

Parents are the more the direct recipient of the website as it will provide information to effectively support their children with CHD. Parental input is important and was planned to be collected via surveys, questionnaires, and interviews. This ensures the website addresses the specific areas in which parents are currently experiencing challenges, and accurate information is obtained and provided for them.

HCP will be both developing and using the online resources to communicate health care information to parents. HCP include the HCP who deliver children with CHD, as they may be with the parents when they initially discover the CHD. Other professionals include nursing, primary care physicians, physical therapists, occupational therapists, prosthetics and orthotic specialists, orthopedic surgeons, technicians, and aids. This resource can help challenge the stigmas HCP may have regarding the actual full capacity functioning of the limb and help provide parents with practical resources.

Shriners Hospitals for Children – Northern California provided funding for the development and implementation of the online resource. SHCNC is a reputable health care organization that focuses on research and teaching for non-routine and specialty conditions children face. Hospital administration staff need to ensure that information is accurate and does threaten the validity of their care and research. SHCNC is supportive as long as the content is accurate and beneficial for their clients, and they are most

interested if the content from the website leads to more referrals and therefore, more revenue.

*Policy.* Fortunately for families, insurance companies are billed for services, and the remaining balance is covered by funding through the SHCNC system, providing access for care for all clients. This model is currently functioning, however, as health care costs increase and insurance companies restrict the services provided, there may be more remaining costs threatening the sustainability of the SHCNC model.

Shriners Hospitals for Children – Northern California will implement the proposed program. SHCNC is located in Sacramento, California. It is essential to review Medi-Cal guidelines which guide the implementation of the federal Medicare and Medical programs. The most frequent problem for families of children with CHD is access for prosthetic and orthotic devices which, falls under 22 CR 51321 (g) Durable Medical Equipment (Reuters, 2018). The policy limits access to prosthetic devices that are a better fit and more functional for children with CHD. The policy requires children to trial the lowest cost devices. Often the lowest cost device is not the best option. However, before children can qualify for the best and most functional fitted device, they must try the least expensive device.

Children with CHD may qualify for Individualized Education Plans through the Individuals with Disabilities Education Act and section 504 through the Rehabilitation act of 1973 to best support their functioning in the school systems. These legislative policies provide guidelines for those seeking alternative ways of accessing the

curriculum. Children with CHD may qualify for services depending on their CHD's impact on their school success.

*Stakeholders and policy.* Parents' ability to pay for health care and services is an imperative factor influencing children's access to treatment. Parents are responsible for navigating the ever-changing policies and available services. In order to do so parents need to understand, seek, and qualify for services and supports. Additionally, children may greatly benefit from the goods and services provided by HCP. However, the number and length of treatment sessions, or the types of surgical interventions provided are dependent on medical insurance coverage and parents' resources to supplement costs. Reimbursement for services depends on the political climate of the time of service. Services supporting children with a non-life threatening condition, such as CHD, may be less likely to be covered if reimbursement tightens. As of now, SHCNC can cover the remaining costs for services provided regardless of family's ability to pay. However, this is not a sustainable model if health care costs continue to increase and reimbursement rates decrease. The hospital may need to resort to downsizing services and increasing fundraising efforts to continue operations.

## CHAPTER TWO

Due to the relatively uncommon birth frequency of CHD, parents feel there is a lack of support, resources, and interventions to cope with effectively and navigate the specific needs of children with congenital hand differences (Ardon, Janssen, Hovius, Stam, & Selles, 2012; Murray, Kelley-Soderholm, & Murray, 2007). As described in Chapter One, cultural factors impact the relationships between the environmental factors and the children's personal factors, which affects the child's engagement their social and school occupations.

In referencing to Goffman's Stigma Theory (1963), there is a social discrimination towards people with body differences and assumptions that these people are limited in their functional abilities. Researchers found that children with CHD had comparable scores on functional domains as their peers without CHD (Ardon, Jassen, Hovius, Stam, & Selles, 2012). This similar scoring suggests that children are functioning at the same level as their peers and not the assumed "limited functioning". Researchers noted however, as children with CHD approached 13-14 years old, they reported lower in the social functioning domains on the Health-Related Quality of Life Scale (HRQoL) (Ardon, Jassen, Hovius, Stam, & Selles, 2012). It can be inferred that children with CHD may be experiencing external social factors impacting their HRQoL (Ardon, Jassen, Hovius, Stam, & Selles, 2012).

Similarly, Goodell et al., 2007, suggested that parents' perception of their children's limited function contributed to lower parent scores on the ABILHAND- Kids assessment of children's hand functioning (Goodell et al., 2017). Stigma can cause an

increase in stress, as children are limited in their social identity and are devalued (Miller & Kaiser, 2001). This stress is unique to the individual that is stigmatized, as family and friends may not share the stigmatized experience.

Individuals experiencing stigma may struggle with psychological consequences, lack of control, low self-esteem/ self-concept, and depression (Miller & Kaiser, 2001). Therefore, the individual experiencing stress related to the experience of stigma will need to adopt various coping strategies to manage stress effectively. The importance of coping, for both parents and children, is repeatedly noted in the literature about the experience of childhood CHD (Andrews, Williams, VandeCreek, & Allen, 2009; Bae, Canizares, Miller, Waters, & Goldfarb, 2018; De Jong, Reinders-Messelink, Janssen, Poelma, Van Wijk, & Van der Sluis, 2012; Murray, Kelley-Soderholm, & Murray, 2007).

Goffman (1963) proposed that individuals reconcile between the external social limitations placed on them by society and the self-perception of being "normal." Stigmatized people can come to terms with these two opposing constructs and bring stigmatizing situations into consciousness. The stigmatized individual realizes they do not have to follow the social limitations superimposed on them; they can break themselves free and develop self-confidence. With this newfound confidence and understanding, these individuals can then become advocates and teach society to redefine the expectation of "normal." This new definition of normal includes people with physical differences. Stigmatized individuals can teach society that people with disabilities are not subclass humans (Goffman, 1963). Effective coping mechanisms can assist individuals to reconcile between the two conflicting messages: the self-understanding of "normal" and

their perceived social limitations presuming them as “non-normal.”

The following literature review highlights coping and coping strategies parents and children employ when negotiating social perceptions and limitations.

### **Evaluation of the Evidence**

The evidence specifically related to children with CHD is low to moderate in quality. As of today, there is a lack of standard keywords to identify this particular subpopulation. Almost every study reviewed referred to limb differences by a different name. Keywords included "congenital hand deformity" "congenital limb difference (also including lower limb)" "congenital limb absence" and "below elbow deficiency."

Twelve studies were reviewed to develop the proposed explanatory model for this OTD capstone project. Among these 12 studies, none were randomized control trials. Two of the articles were expert opinions with an evidence level of 7 (on a 1–7 scale, with one being the highest level of evidence and seven being the lowest).

The review included five qualitative studies with an evidence level of 6, three cross-sectional qualitative studies with an evidence level of 4, and two studies that were non-randomized control trials with an evidence level of 3. There were no studies included with evidence levels of 1–2. Thus, most of the research reviewed had low evidence levels. Additionally, four studies included more than 100 participants, two studies included between 50–100 participants, and five studies included less than 50 participants. This further demonstrated the low levels of evidence available regarding CHD. Three studies occurred outside of the United States and had low generalizability for populations with CHD living in the United States.

All of the studies lacked social economic status (SES) data, lowering the generalizability to all SES groups. Some studies did not use valid and reliable outcome measures, but instead developed unique surveys and questionnaires. Studies that did use valid and reliable outcome questionnaires like the Vineland-II or Health Related Quality of Life measure (HRQoL) reported that the outcomes questionnaires may not be sensitive enough to capture the functional difficulties experienced by children with CHD.

The research tends to be mostly qualitative when focusing on social and emotional development for children with CHD. Because the lived experience of a child with a CHD is unique and subjective, randomized control trials in this subject matter is difficult. However, extrapolating from the available research studies, children with CHD are functionally able to do the activities they need to and want to do with adaptations. They perceive stigma and experience more challenges with self-acceptance and self-esteem in developing social relationships as they age. This cross-study agreement helps to strengthen the evidence reviewed. More research related to the experiences of children with CHD is indeed necessary.

### **Evaluative Summary**

*Child experience.* Multiple studies reported that children tend to score within normal ranges when compared to unaffected children in similar age groups on the following standard outcome measures: Piers- Harris Children's Self Concept subscale, Pediatric Quality of Life Scale, Child Depression Inventory, Culture Free Self-Esteem Inventory, State Trait Anxiety Inventory, Standard Intelligence Scales, Peds Quality of life Scale (Andersson, Gillberg, Fernell, Johansson, & Nachemson, 2011; Ardon,

Janssen, Steven, Hovius, Stam & Selles, 2012; Bond, Kent, Binney & Saleh, 1999). This research indicated that children with CHD are relatively functional with their limbs; therefore, interventions to enhance function is not a high need for children with CHD.

However, as children with CHD approached adolescence, they scored lower on social functioning domains on the Patient-Reported Outcomes Measurement Information System (PROMIS) and Health-Related Quality of Life (HRQoL) when compared to peers without CHD (Ardon, Janssen, Steven, Hovius, Stam & Selles, 2012; Bae, Canizares, Miller, Waters & Goldfarb, 2018; Gringas, Mongeau, Moreault, Dupuis, Herbert, & Corriveau, 1964). Based on these findings, the changes in social functioning as children transition from childhood to adolescence may have implications related to stigma. If these children experience stigma as they approach adolescence, they may need to develop effective coping strategies to manage the stigma they may experience. Ardon et al., (2012) reported that low scores in social functioning were indicators that children in the study encountered stigma and therefore, barriers to their social occupations (Ardon, Janssen, Steven, Hovius, Stam & Selles, 2012). These researchers also found that children with mild CHD (less noticeable: i.e., single-digit amputation) had lower scores on the emotional functioning domain of the HRQoL than children with more severe differences (more noticeable: i.e., below elbow amputation) (Ardon, Janssen, Steven, Hovius, Stam & Selles, 2012). This is important as children with less severe differences are far closer to “passing as normal” than those with more severe differences. Children with more severe differences have to face the reality of their condition more often and find acceptance with it as it is more difficult to hide than less severe conditions.

Applying Goffman's (1963) Stigma Theoretical framework, it can be inferred that children with CHD are experiencing an increase in perceived stigma as they approach adolescence. This increase in stigma impacts their ability to form social relationships. Similarly, those who have less severe differences may experience additional stigma and less coping as they are more likely to pass as "normal" when compared to children with more severe CHD (Ardon, Janssen, Steven, Hovius, Stam & Selles, 2012; Andersson, Gillberg, Fernell, Johansson, & Nachemson, 2011).

Exploring social relationships further, Franzblau et al. (2015) conducted semi-structured interviews with parents and children with CHD to evaluate stress associated with CHD. In the study, children reported stress due to social interactions, hand appearance, and emotional reactions. Children's stress was undetected by the parents 50% of the time (Franzblau, Chung, Carlozzi, Chin, Nellans, & Waljee; 2015). Kelly et al. (2016), reviewed hand function and appearance following reconstruction surgeries for children aged 6-17. 48% of the children reported they disliked or were bothered by their hand difference (disliking scarring, bone structure, and abnormal hair growth) (Kelley, Franzblau, Chung, Carlozzi & Waljee, 2016). The children focused on their perceived limitations during research interviews (Kelley, Franzblau, Chung, Carlozzi & Waljee, 2016). Inferring from the research, these experiences of stress and dislike for their limb difference suggest these children may be experiencing and internalizing the social limitations brought on by stigma.

Typically, children with limb differences participate in the activities they find meaningful by ways of various modifications and adaptations (Kelley, Franzblau, Chung,

Carlozzi & Waljee, 2016). Researchers found that younger children tended to require additional help in developing modifications whereas, older children/adolescents tended to report having their modifications set. Older children with CHD and their parents reported overall good functioning of the affected limb (Bond, Kent, Binney, & Saleh, 1999; Kelley, Franzblau, Chung, Carlozzi & Waljee, 2016; Michielsen, Van Wijk & Ketelaar, 2010). As children develop adaptive methods to engage in activities, they also develop adaptive coping mechanisms to address the perceived stigma of limitations. Based on semi structures interviews with parents and children, researchers identified with CHD eight coping strategies: humor, self-acceptance, avoidance, seeking external support, concealment, educating others, support programs and religion (Franzblau, Chung, Carlozzi, Chin, Nellans, & Waljee; 2015).

*Parent Experience.* Parents are often individuals who do not identify with the disability community and have lived within the “normal” group with learned stigmatizing views of individuals with a disability (Bradbury, 2007). Often parents will give birth to children with CHD and require a bereavement period as they grieve the loss of the imagined “perfect” baby and need to cope with the “deformed baby” (Bainbridge, 2009; Bradbury, 2007; Ho & Ulster, 2011; Kerr & McIntosh, 1999). Parents may have a period of turmoil and substantial emotional distress, shame, and guilt as they reconcile having children with a CHD. They may lack information, emotional support from health care professionals, and support from their community or extended family (Bainbridge, 2009; Bradbury, 2007; Kerr & McIntosh, 1999; Ho & Ulster, 2011). Parents may experience feelings of shock, sadness, anger, and denial, because they may speculate that the CHD

was a result of their previous actions or behaviors (Bainbridge, 2009). This strong emotional reaction suggests that the stigma (the belief that one with a disability is a subclass of society) is a significant barrier for parents as well as children (Bainbridge, 2009). This stigma can explain the challenges parents have in envisioning a future for their children and can explain feelings of isolation, concern for the future, and difficulty taking their child out for social outings (Kerr & McIntosh, 1999).

Parents experience stress related to making medical decisions for their children (Bradbury, 2007). This difficulty is a combination of multiple factors: the first being the challenge of including their child in the decision-making process. In interviews, parents of children with CHD reported anxieties when making decisions for younger children who were unable to contribute to the decisions as they were too young to participate in the decision-making process (Bradbury, 2007). Bradbury discussed the complexity of surgical decision making and recommended professional mental health therapy to support parents' emotional needs related to decision-making (Bradbury, 2007). Additionally, Bainbridge discussed the perspective of fathers in the decision-making process. Fathers often felt left out, as the medical team placed more emphasis on mothers and the children. Bainbridge suggested that fathers may be experiencing emotional distress, which may be a poor reflection on their "manhood" (Bainbridge, 2009). It is important to include fathers and children as much as possible and as appropriate for their developmental maturity. Having children with a congenital hand difference can be traumatic for the parents. Parents may need to develop coping mechanisms to address their own needs.

*Coping.* The coping mechanisms used to respond to the perceived stigma

experienced by children with CHD may be either helpful or detrimental.

Child Positive Coping. Children are very likely to adapt to their environment and utilize their limbs to engage in their activities. In semi-structured interviews, parents and children with CHD described the positive coping mechanisms their children use to address stigma. The coping strategies included using humor to explain their limb difference, self-acceptance, seeking external support (programs/ camps), educating others, support programs, and religion (Franzblau, Chung, Carlozzi, Chin, Nellans, & Waljee, 2015). Franzblau et al. (2015), suggested that the coping mechanisms such as preparing children with social scripts for explaining their CHD may improve self-esteem, and help children address or resolve feelings of inadequacy.

Ho and Ulster (2011) examined the effectiveness of Children with Obstetrical Brachial Plexus Day. The day included workshops and breakout sessions for teens and parents, lectures from health care professionals, meet and greets, and teen testimonials. All families rated the various components of the day as “good” or “excellent,” with workshops receiving very high ratings. All participants in the study reported they would attend the event in the future. Thus, we can infer the parents found the event valuable. The day also featured a workshop on bullying and teasing. This session provided the opportunity for teens to reflect on and share the challenges they experienced as well as their coping strategies. The workshop was designed to heighten their social awareness, facilitate acceptance of their differences, and promote better views themselves as equals (Ho & Ulster, 2011). Based on the framework of Goffman’s Stigma Theory (1963), these children and adolescents developed positive coping mechanisms to help combat the

shame associated with social limitations.

Child negative coping. As children are “discovered” as having a limb difference and experience the perceived social limitations of being in the “out-of-group”, they may develop negative coping mechanisms. If children are experiencing stigma in their school and social occupations, they may engage in concealing behaviors to avoid the stigma. (Franzblau, Chung, Carlozzi, Chin, Nellans, & Waljee; 2015). When children hide their hands, they then limit their functional engagement as they are not using their hands to maximum capacity. Children may experience decreased self-esteem and increased anxiety in social interactions and use disengagement coping mechanisms, such as concealment, ignoring questions, or not disclosing the CHD (Franzblau, Chung, Carlozzi, Chin, Nellans, & Waljee; 2015). Gringas and colleagues (1964) reported that children with CHD between the ages of 6-13 “invest, whether consciously or unconsciously, the deficient limb with all of their anxiety... the handicap is always a source more or less latent, of self-devaluation. Even if tendencies to overcompensate are developed and gratified, the disability is already a sign of inferiority” (pp. 117). These findings support the proposition that the perceived limitations felt by those with a physical disability impact their peer relationships. This is because children with CHD may perceive themselves as socially inferior.

Parent positive coping. It is unlikely that parents of children with CHD will meet each other by happenstance. Parents may have feelings of isolation and difficulty connecting about shared experiences with fellow parents who do not have children with CHD (Kerr & McIntosh, 1999). Kerr and McIntosh interviewed 63 parents on their

experiences of having children with congenital hand differences (Kerr & McIntosh, 1999). Researchers shared that many of these parents reported the benefit of meeting other parents. Such meetings supported the realization of shared experiences, provided a connection to those who understood, helped parents imagine the future by witnessing older children and adults with differences and hearing stories of success (Kerr & McIntosh, 1999). Researchers suggest that parents involved in community support groups were more likely to identify positive coping mechanisms than those who were not a part of community groups (Bradbury, 2007). Involvement in a community with peers who understand a shared experience helps to “normalize” the experience and support parents to envision a future, potentially mitigating parental stigma towards their children with CHD.

Parent negative coping. The strong emotional response of parents to their children with CHD varies. Some parents cope and accept their children rather quickly, while others require additional time or never develop effective coping strategies. When parents feel guilt or turmoil for their babies, these emotional reactions threaten the parent’s ability to bond with their babies (Bradbury, 2007; Bainbridge, 2009, Gringas, Mongeau, Moreault, Dupuis, Herbert, & Corriveau, 1964). Anxieties manifest in how parents interact with children. If parents are concerned for the children’s inability to complete a task, they may limit their children from attempting such a task. This action reinforces stigmatization of the children. The children may then believe these weaknesses and internalize these perceived social limitations (Bradbury, 2007; Gringas, Mongeau, Moreault, Dupuis, Herbert, & Corriveau, 1964). Parents may adopt concealment

behaviors for the CHD (covering with mittens or other devices) to promote the notion of “passing as normal.” This again reinforces anxiety, shame, and self-consciousness. It teaches the children to engage in concealment behaviors (Bradbury, 2007).

Similarly, when the parents’ do not process their emotional response, they are unable to discuss the limb difference with their younger children. By denying the opportunity for this conversation parents effectively teach children to perceive their CHD as a source of negativity and shame (Bradbury, 2007). Based on semi-structured interviews with parents of children with CHD, Kerr and McIntosh (1999) reported parental feelings of isolation, lack of resources, and difficulty envisioning a future for their children. For example, one parent shared, “The feelings I had were terrible. I just didn’t like the idea that everybody else’s baby had two hands,” and, “Initially I couldn’t see past this one-armed baby lying in the cot. I couldn’t see the future, I couldn’t see him in the future playing happily with [his sister]. I just couldn’t see past this missing arm” (Kerr & McIntosh, 1999, pp. 313-315). These responses reflect parents’ initial emotional reactions and highlight the need for coping. Gringas et al. (1964) theorized that unresolved emotional reactions to the limb difference negatively impact the children’s ability to cope. Thus, parental coping impacts the perceived stigma experienced by the children.

This content from this literature review was synthesized and disseminated via an online website for parents to provide resources for parents to support their children with CHD to most effectively engage in their school and social occupations.

### **CHAPTER THREE**

Research reported that parents of children with CHD feel stress associated with a lack of resources, lack of community, and concerns for their children's future (Bainbridge, 2007; Ho & Ustler, 2011; Franzblau et al., 2015). Parents are likely to experience negative social stigma related to disability (Bainbridge, 2007; Ho & Ustler, 2011). If parents experience difficulty coping with the CHD, there are potentially negative consequences for their children (Bradbury, 2009; Bainbridge, 2007; Gingras, Mongeau, Moreault, Dupuis, Hebert, & Corriveau, 1964). One solution to the lack of information and resources is the development of an online educational resource. Online resources are an effective mechanism to disseminate information, with higher rates of knowledge retention on the information presented when compared to traditional in-person training (Lioffi, Failo, Schoth, Williams, & Howard, 2018). Therefore, the proposed project is the development of an online resource. This resource provides content and addresses common needs to assist in parents in raising children with CHD.

#### **Online Resources**

The goal of this capstone project is the development of a user-friendly website for parents of children with CHD. A synthesis of the literature related to the needs of parents provides a guide for determining the website content. Content is developed from knowledge and themes gathered from the literature and lived experiences. Personal interviews provide a validation of parent experience and assist in the development of an envisioned future (Kerr & McIntosh, 1999). Information gathered from the interviews are synthesized into the website.

### Website Content

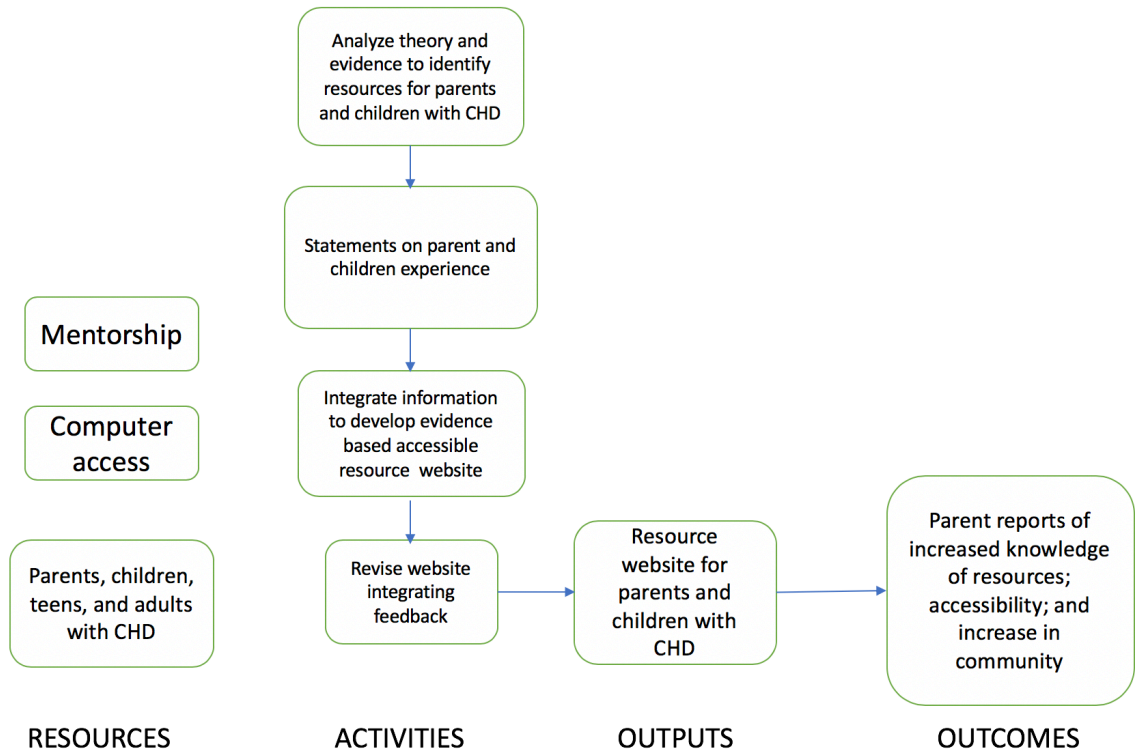
*Parent experience.* In response to parents feeling uninformed, isolated, and lacking emotional support (Bainbridge, 2009; Bradbury, 2007; Ho & Ulster, 2011; Kerr & McIntosh, 1999), the parent experience section features a description of common postpartum parent experiences, parent coping strategies, and stigma. Additionally, it reports parent reflections on these experiences. These resources are designed to decrease feelings of isolation, as parents will be able to read stories similar to their experience. Health care professionals can direct parents to this website for more information or confirmation of information received within the hospital setting.

*Child experience.* In response to children feeling self-conscious as they enter adolescence and parents' difficulty envisioning a future for their newborn child the online resource includes an "about me" section (Ardon, Janssen, Steven, Hovius, Stam & Selles, 2012; Bae, Canizares, Miller, Waters & Goldfarb, 2018; Gringas, Mongeau, Moreault, Dupuis, Herbert, & Corriveau, 1964). This section highlights the positive experiences of children, teens, and adults living with CHD who have overcome the challenges of living with CHD. Additionally, this section features suggestions for positive coping mechanisms for children, including using humor, self-acceptance, seeking support, educating others, and support programs (Franzblau, Chung, Carlozzi, Chin, Nellans, & Waljee; 2015). With the addition of this information, parents can better understand the child experience and better envision a future for their children, potentially decreasing perceived stigma associated with CHD. Additionally, children will be able to see other children like them, and older children/ teens/adults with CHD. This builds a broader

community and decreases perceived stigma.

*School resources.* The online website features resources to best support children in school to address the perceived lack of information for supporting children in this setting (Bainbridge, 2009; Bradbury, 2007; Ho & Ulster, 2011; Kerr & McIntosh, 1999). There is a section on the description of supports provided by 504, IDEA, and resources for communication of CHD with teachers and classrooms. This section features links to additional information and application materials for the state of California's public education system.

*Additional resources.* The website includes links to community organizations and resources that provide support for children with CHD to address the lack of information on resources (Bainbridge, 2009; Bradbury, 2007; Ho & Ulster, 2011; Kerr & McIntosh, 1999). Links includes national non-profit organizations to build community and pride around limb differences such as the Amputee Coalition, Lucky Fin Project, and Helpings Hands. There are links to events and outings that promote child-to-child and parent-to-parent meetings such as Camp Winning Hands, Camp No Limits, and Lucky Fin Project Picnic Day.



**Figure 2 Proposed Logic Model**

**Resources**

The following discusses the roles and actions of the resources, activities, outputs and outcomes from the logic model as seen in figure 2.

*Faculty Mentor.* The role of the faculty mentor was to provide resources and support for ongoing academic work to ensure academic integrity and guidance. The faculty mentor meet on a regular bases and reviewed the written materials for accuracy and clarity. The faculty mentor supported the development of the program.

*Site Mentor.* The role of the site mentor was to provide resources and support for scientific integrity and guidance on resources for this specific population. The site mentor reviewed written materials for accuracy and clarity while provided support for program

development.

*Digital Access.* Computer access included the physical presence of a working computer with internet access, the ability to upload and download materials, access to data outcome measures, access to email capabilities, and access to journal articles. Computer access facilitated the research and ability to develop content for the website. The project needed access to the website development program. This platform access was used to develop content for the website, keeping in line with SHCNC branding and messaging. Hospital administrators reviewed website content for approval before public publishing. The computer station refers to the physical location for the computer station with internet access, the ability to save and access information on drives, this includes email access to download/ upload content with the understanding this does not authorize for remote access to protected health information. IT support referred to SHCNC IT department's assistance with problems as they arose concerning computer functioning, email, documents, website development access, and troubleshooting.

*Parents of and children/teens/ adults with CHD.* The project includes the perspectives of parents of and children with congenital limb differences. This perspective provides content for real parent, teen/ adult, and child experience to help develop the community. It was necessary to request access to contact information from SHCNC, whether it was through access in conjunction with clinic appointment, or at community networking events. Consent forms and photography release forms were needed to ensure those involved understand how content will be useful and authorize content to be shared in this platform.

*Health Care Professionals.* It was valuable to have a team of professionals to meet with as needed to communicate evidence and content for subsections of the website.

*Budget.* The budget included the payment for website access/ upkeep, Shriners Hospital administration manages this budget.

### **Activities**

*Literature Review.* Literature review refers to the multiple literature searches conducted and literature synthesized to develop evidence-based content on the website.

*Interview Questionnaire.* Interview questionnaires were developed for parents, teens, children, and adults to convey their experience. Content is uploaded into parent experience profiles and child/teen/adult experience profiles.

*Website review.* The website review referred to meeting with SHCNC administration to seek approval of the website and ensure messaging was consistent across all Shriners Hospital for Children systems.

*Outputs.* The output is an online resource for parents of children with CHD, including subsections of parent experience, child experience, school resources, and additional links.

### **Outcomes**

*Most accurate targeted information.* Parents can easily access accurate content and resources on the parent experience, child experience, school resources, and additional resources that targets members of the CHD community. Parents evaluated the website content on the premise of increased knowledge, increased community, and ease of website use.


### **Impact**









In providing an evidence-based and accurate informational website targeted at increasing positive coping for parents, parents will be more likely to cope with the CHD and in turn increase their children's ability to cope (Bainbridge, 2007). These higher rates of coping can decrease the feelings of perceived stigma, ultimately facilitating children engagement in school and social occupations.

### **Program Specifics**

The CHD website landing page is accessible via the "Hand, Arm, and Shoulder" resource page and the "Parents & Patients" tab. The Congenial Hand Differences landing page offers images of parents and children engaging in activities with the following subtabs: Parent Experience, Child Experience, School Resources, Additional Resources. Sample landing page is seen in figure 3.


CHOOSE LANGUAGE DONATE + CONTACT + REFER +

 **Shriners Hospitals for Children® Northern California**

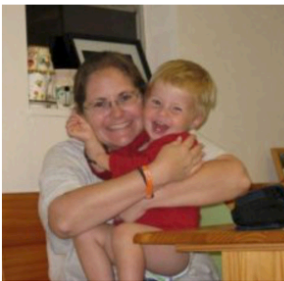
       

Cure Locations Education Research News & Events Stories Donate Patient Portal

## Congenital Upper Limb Differences




Paragraph outlining the definition of CoULD, difference from CoULD and amputation. Statistics on frequency of CoULD purpose for website page.




**Parent Experience**

Paragraph with brief overview of parent experience, with parent quote  
Paragraph with brief overview of parent experience, with parent quote




**Child Experience**

Paragraph with brief overview of child experience, with child quote



**School Resources**

Brief overview of IEP and 504, how they can help support children with limb



**Additional Links and Resources**

Description of links and resources to help build a stronger limb different

Figure 3 Sample landing page

## **Detailed Weekly Activity Plan**

### **Week 1-2**

- Orientation, networking with Health Care Professionals, begin shadowing/ observation of parents and families
- Meet with SHCNC website development team
- Begin website creation process, domain, login/passwords, determine layout and aesthetics consistent with SHCNC branding and messaging
- Weekly check-in with faculty and site mentors

### **Week 2-4**

Parent experience:

- Conduct Parent Experience Interviews
- Synthesize information from literature and interviews on parent experience
- Draft information for parent experience on the website
- Weekly check-in with faculty and site mentors

### **Week 5-7**

Child experience:

- Conduct child experience interviews
- Synthesize information from literature and interviews on child experience
- Draft information for child experience on the website
- Weekly check-in with faculty and site mentors

### **Week 8-9**

School resources:

- Review California policy for IDEA and 504 concerning services for CHD
- Draft information for school resources on the website
- Weekly check-in with faculty and site mentors

### **Week 10-12**

- Review and edit the draft version of the website
- Submit a draft website to parents for outcome measure feedback
- Analyze feedback, revise and make changes to the website
- Weekly check-in with faculty and site mentors

### **Week 13-14**

- Complete capstone report
- Complete capstone poster
- Complete capstone presentation
- Weekly check-in with faculty and site mentors

### **Barriers/Challenges and Proposed Solution**

Foreseen barriers included time management, website development difficulties, and weak evidence.

*Time management.* This project is robust, and ambitious in its conception; it was imperative to follow the week by week outline. Website content was planned to be completed and uploaded by the end of week 11 and beginning of week 12 order to allow for feedback and revisions before the completion of the capstone experience. In order to ensure an appropriate timeline, daily and weekly goals were developed in a reflection of the proposed weekly plan to ensure project components progressed at an expected rate. If meeting deadlines became challenging, project components were reviewed and revised with mentorship input.

*Website formatting difficulties.* Technology difficulties inevitability arise when formatting websites, whether it be layout challenges, uploading content challenges, inability to create consistency in font and sizing. In order to address this, contact with website developers and IT support can ensure that problems resolve efficiently. Publicly available tutorials and instructional videos, web forums can assist in problem solving.

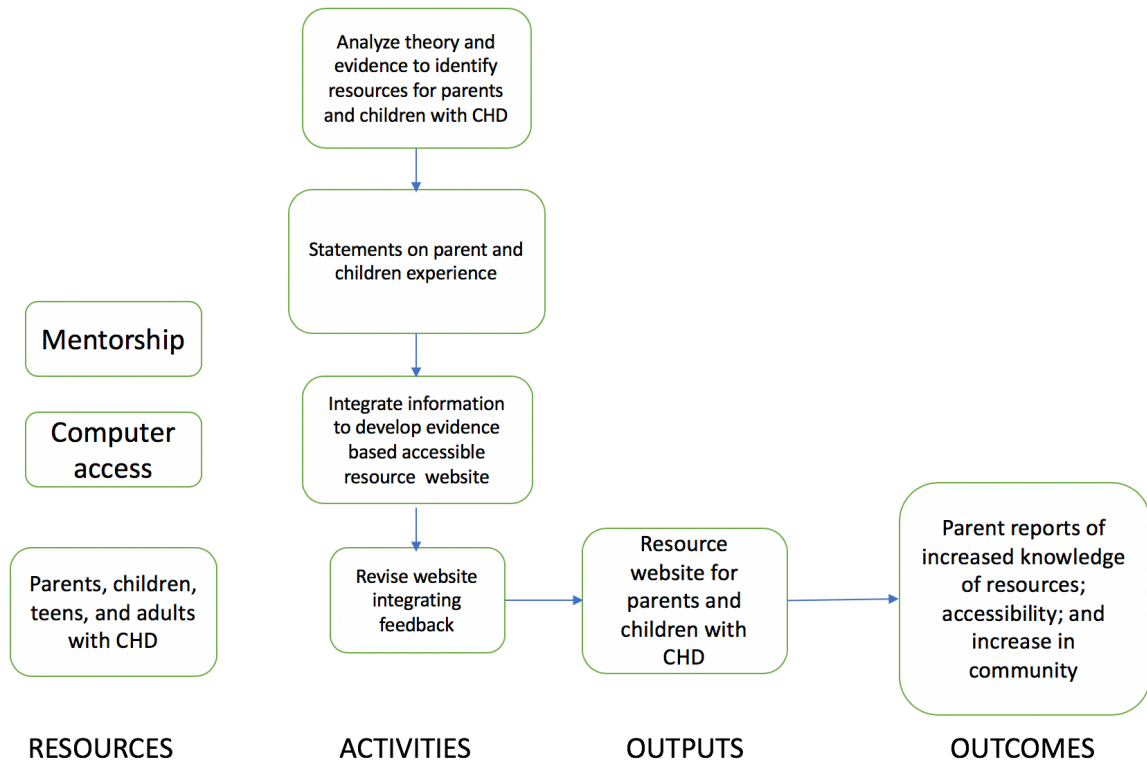
### **Sustainability**

The website is scheduled to be reviewed annually by members of the SHCNC Hand Team to update content, dates and links. The information included in these sections of the online resources will not address all of the resources that parents and children with CHD may be seeking. However, the four online resources will provide a foundation with the opportunity for future expansion. .

## CHAPTER FOUR

### Evaluation Plan

To determine program effectiveness, it is worth re-addressing the logic model. The output is an online resource for parents of children with congenital hand differences with the outcomes of accessibility, increased knowledge of resources, and increased feelings of community.



**Figure 4 Proposed Logic Model**

### Website Objectives

The original objectives of the website were to create an accessible website to increase parents' perception of knowledge about resources to support their children and to create a sense of community for parents. This objective became problematic to measure, these would have been very subjective self-reports that were difficult to

quantify, and much variation on question constructs between parents. Consequently, the outcome measure was redesigned to assess parent's perception of the usefulness of website content, ease of use, and likeliness to use and recommend the website. The outline of activities in the logic model supported these objectives. The supports and resources provided by mentors and the SHCNC setting facilitated the necessary activities to produce the website as an output. This output, in theory, provided the outcomes of providing a useful, easy to use website parents are likely to use and recommend.

### **Outcome Cohort**

Prominent limb different leaders on social medial platforms assisted with recruiting parents to evaluate the website. Parents were invited to review the website and complete an online questionnaire evaluating website features and content. Preferred respondents were parents of children with CHD aged newborn – 6 years old, as often this is the time of most uncertainty (Kerr & McIntosh, 1999). Parents were asked to review the website content before questionnaire completion. The goal was to recruit fifteen parents, with a 100% response rate.

### **Website Evaluation**

The outcome measure for this project was online survey questionnaire that was sent to parents along with the link to the CHD website. Questions were created in a 1-5 Likert scale, with one being the lowest rated and five being the highest rated. Each question had a not applicable option and additional comment boxes for parent feedback. The outcome measure featured two general questions for parent feedback and suggestions. These were excluded when scoring the program's effectiveness. The

comment boxes will not be required to complete and will not count towards the 100% completion rate. See Appendix A for the Website Evaluation Measure. The Website Evaluation focused on the Parent Experience, Child Experience, School Resources, and Resources pages were part of the outcome measure as these were the original pages focused on in the capstone projects. The About Us, Personal Experiences, Pediatric Hand Team and Landing Page, were not the focus of evaluation as they were added on after the initiation of the capstone project.

The survey was distributed via social media platforms and made it difficult for confirmation that only parents of children with congenital hand differences completed the survey. Therefore, a question was added to the outcome measure allowing for respondents to indicate if they were a parent of a child with CHD, a person with CHD, a health care provider, or other. Those who selected "parent of a child with CHD" were included in the data analysis.

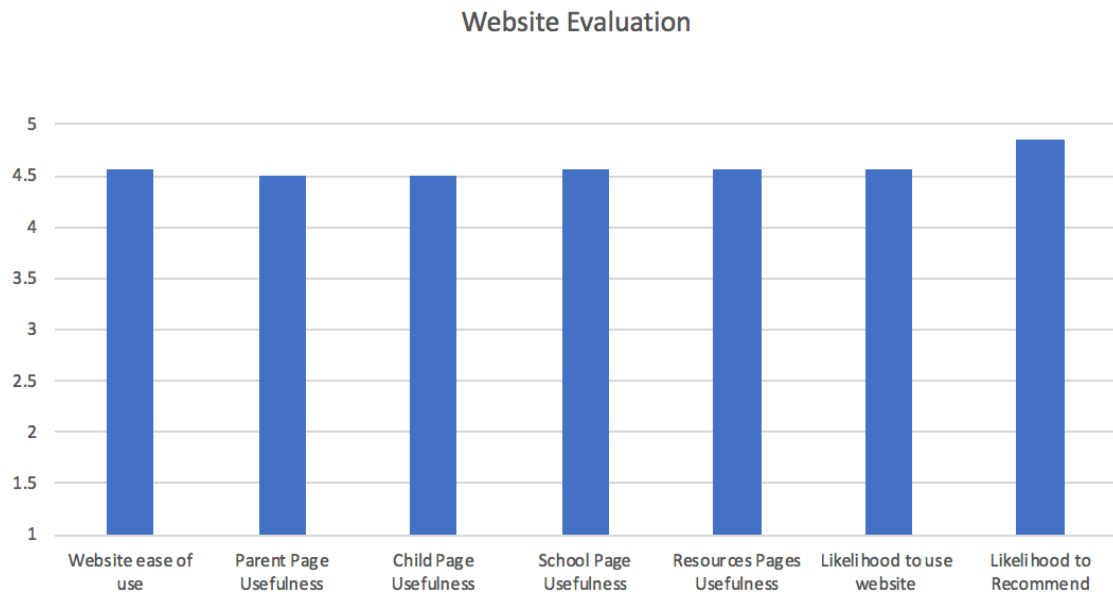
### **Data Analysis**

Social media was chosen as a recruitment platform as many parent communities exist on social platforms, and they are easy to access. Parents using social media were more likely to have older children, indicating they have grown through and experienced a variety of the concepts explained in the website. These parents were likely to provide insightful feedback, due to their experiences and the resources provided at the time of birth.

Twenty-three participants completed the survey. Fifteen were parents; five were people with congenital hand differences, three identified as "other." Persons with

congenital hand differences and "others" were excluded. One parent had a partially incomplete the survey and was excluded. The total responded parent number was fourteen.

Each response for each question was assigned a value. The value of 1 indicated lowest response (not easy, not useful, will not use, will not recommend) and 5 was assigned to the highest rated (very easy, very useful, will use, will recommend) — the average score of 4+ out of 5 determined website effectiveness. The following graph visually represents this data.



**Figure 5. Website Evaluation Graph n=14**

Each question received an average score of 4.5 or above for each section indicating that the website was considered easy to navigate and the content in the parent, child, school, and resources section was useful. Parents reported that they were likely to use the website and recommend the website to others.

Depending on the appropriateness of feedback, website edits were made, integrating feedback from comment boxes. Some suggestions were accommodated, such as parents' expansion of the Personal Experiences page with the inclusion of a downloadable template to facilitate submissions from parents and people with CHD. Other suggestions were not feasible to include and fell outside of the website scope and intent.. For example, parents suggested including a parent forum where parents can suggest resources and ask questions. This type of forum is better suited for parent groups and parent social media platforms. Therefore, this change was not made, however parent support group are referenced in the resources section on the website.

### **Limitations**

There are inherent limitations to the website evaluation process. First, 14 parents is a small sample size. A larger sample size could better represent the potential website user population. Second, the evaluation form is a self-report data which lacks the strength of objective measures. Thirdly, parents were recruited through social media platforms. It is likely that parents have a favorable bias due to a relationship with the website developer, or a relationship with the website developer's friends/ colleagues.

## CHAPTER FIVE

The Congenital Hand Difference website is designed specifically for parents of children with CHD. The content and resources within site can be beneficial for them and address common concerns when they are raising their children with CHD. It is advantageous to disseminate information about how to access the website to parents and health care professionals.

The short-term goal of the dissemination plan is to have an increase in viewing of the website over the next six months. This goal is measured in website hits, or how many times a website is visited. Data is collected via website analytics.

A long-term goal of this website is for the website to assist parents with transition of their children with CHD into school . This can be collected by a self-report from parents that they perceived the tools and resources assisted in the transition of their children into school.

### **Target Audience**

*Primary Audience.* The primary audience are parents of children with CHD. These are the individuals that will have the most impact from the website features and content.

*Secondary Audience.* The secondary audience are health care professionals (HCP). The content within the website can assist health care providers in understanding the parent and child experience of having a CHD. This understanding may help practitioners provide relevant care and connect parents to more useful resources. Secondly, HCP can connect parents to this resource website, providing supports for

parents.

Specific HCPs, are occupational therapists, as they have the distinct role of working with children and their families to engage in meaningful activities. Many of the topics included in the resource website can assist in their care delivery and connect parents to further resources.

### **Key Messages**

*Primary Audience.* The key message is that there is a resource page to support parents in raising children with congenital hand differences. The resource website contains the following topics:

- Addressing parent guilt and stigma
- Coping strategies
- Activity Adaptation
- Developmental Milestones
- Social and emotional anticipatory guidance
- Managing conversations with peers, new people, and teachers
- School resources such as 504/IEP plans
- Developing personal resumes and books
- Children's books and scientific articles on hand differences
- Stories from parents and people living with hand differences
- Roles of Pediatric Hand Team and questions to ask on a first visit

*Secondary Audience.* The key message for health care providers is that an easy to navigate and useful parent resource website exists to provide supports for children with congenital hand differences. The website includes the topics listed in the primary audience. HCP can additionally share this resource website with their patients as a form of support.

### **Sources/ Messengers**

*Primary Audience.* A messenger for the primary audience is Nicole Kelly. She is

a leader within the limb difference community. She has a congenital hand difference and won Miss Iowa in 2013. As a result of her Miss Iowa title, she is a speaker on diversity and inclusion in schools and corporations, and frequently speaks at events. She has studied disability rights and history and is an advocate. She has reviewed the website and agrees with the messaging. Nicole is well connected to share information about the website to reach a wider audience.

*Secondary Audience.* A messenger for the secondary audience is Dr. Michelle James a prominent leader in the pediatric hand surgery. She founded a professional organization, the Pediatric Hand Study Group (PHSG) that meets annually. Dr. James is well connected with other hand surgeons, both nationally and abroad. Dr. James plans to share the promotional flyer about the website with her colleagues encouraging them to review the resource and share it with their patients and their patient's families.

### **Dissemination Activities**

*Special Education Opportunity. Presentation to Health Care Professionals* SHCNC health care professionals attended a 45-minute presentation on the capstone project. The presentation included a review of the purpose, intent, and theory used to design the website along with sharing website design, and features. Participants were emailed the website link post-meeting. The hospital-wide invitation for this presentation is included in Appendix B.

*Submission for AOTA.* The capstone project was submitted for a poster presentation at the AOTA 2020 conference. Business cards with website address and QR codes will be distributed to occupational therapists attending the conference to help

facilitate visitor's remembrance of website post-conference.

*Shrine Vine.* Shrine Vine is a hospital-wide weekly newsletter. A story was drafted in partnership with Director of Public Relations at SHCNC. The article provided a recap of the presentation and provided hospital awareness for this resource website. The Shrine Vine article is included in Appendix C.

*Social Media.* The SHCNC Director of Public Relations worked with the graphic design team to create a condensed informational image with website hyperlink to share on SHCNC social media platforms such as Facebook and Instagram. Social media is used to provide a direct connection to the community, patients, patient families, and other health care professionals. This image can be shared and re-shared amongst the community member's personal and professional social media platforms.

*Flyer.* The hospital graphic artists developed an expanded promotional flyer to describe the website. This flyer was printed and is planned to be distributed by the SHCNC public relations office to patient families, health care professionals, and regional maternity wards in the fall. Parents of newborns with CHD will receive this flyer in their hospital welcome packet. SHCNC has a positive relationship with regional maternity wards and has disseminated this type of informational packet successfully in the past (with brachial plexus injury).

*Electronic communication.* The flyer drafted by the hospital graph artists has an online viewing option. The SHCNC public relations office plans to disseminate the flyer to the pediatric health care professionals, such as nurses, occupational therapists, physical therapists, surgeons, and social workers. These professionals are likely to work directly

with children with CHD and their families.

*Presentation at Pediatric Hand Study Group (PHSG).* The PHSG meets annually to share ongoing research, best practices, and discuss cases with national and international pediatric hand surgeons and occupational therapists. A presentation of this entire capstone project will be included in the 2020 annual meeting. While many health care professionals should have received electronic or paper flyers, the PHSG annual meeting provides a significant opportunity to present the website, and its features to a group of health care professionals working directly with children with CHD and their families.

### **Budget**

*Special Education Opportunity.* Dissemination opportunities occur monthly within the SHCNC hospital. Staffing, refreshments, space, and technology is well established and included within the SHCNC yearly budget.

*Submission for AOTA.* If accepted for the AOTA 2020 annual conference the registration fee is \$451 if purchased before March 13 for OT AOTA members. The anticipated cost is roughly \$1000, including public transportation in Boston, food and meals, renting a place to stay, and poster printing costs. Flights are complementary via as part of the benefits package of being an American Airlines employee.

*Shrine Vine.* Shrine Vine is a weekly newsletter that is already factored into hospital budgets for administrative and graphic artist duties.

*Social Media and flyers.* Social Media posting is within the scope of job duties of Public relations administration staff and hospital budget for administrative costs. Printed

flyers and mailings will cost roughly \$350 and are part of the SHCNC communications budget.

*Presentation at PHSG.* The registration fee for the PHSG is \$250. The total anticipated cost is \$800. This includes accommodations, meals, and local transportation. Flights are complementary via benefits package of being an American Airlines employee. Since the author of this capstone project was a American Airlines employee prior to enrolling in graduate school and has maintained her employment with American Airlines, this benefit continues. Potential scholarships may be available.

### **Evaluation**

*Primary Evaluation.* The website was developed on the WordPress platform. Wordpress allows for website creators to conveniently monitor traffic on to pages of the website. These numbers will be reviewed in six months. Disseminator efforts are considered effective if there has been an average increase in website hits over the six-month period.

*Secondary Evaluation.* Dissemination efforts will be evaluated by the self-report within 12 months from parents who perceived this website to be useful for the transition of their child with CHD into school. Self-reports will be collected from conversations with parents in the clinic setting at SHCNC.

## CHAPTER SIX

The Congenital Hand Difference website has the potential for long lasting positive impact on a broad audience. Parents, children, and people with limb differences can access this resource when convenient for them and for as many times as they need. This resource will help provide resources for families to choose what is best for their child and their family.

Developing a resource website such as this marks innovation in the medical communities. Topics covered and addressed here help build a deeper understanding of a child; they are not merely their diagnosis. The resource website takes into consideration who the child is and the environmental context in which the child exists. An occupational therapy perspective was used to highlight the personal and environmental factors that may influence the life experiences of a child with hand differences. Information about occupational therapy is included in the website and may enhance the understanding of the distinct role of OT for children and their families. Occupational therapists are trained to assess the barriers for activity limitations and identify strengths and supports to facilitate interventions for people to engage in their meaningful occupations. Occupational therapists have the skills for activity analysis and effective communication with their clients. These skills and perspective create an impactful comprehensive resource for client populations in need for this support.

Educating others about the occupation, health and well-being is a type of intervention provided by occupational therapists. The website developed for this capstone project provides educational resources to help people feel better prepared to manage their

health condition and engage in the activities they find meaningful. The website helps give voice to potentially everyday experiences and helps to families prepare for medical appointments. The creation of this website may inspire other occupational therapists to build similar resources for their client populations.

Future incentives would be to develop future parent or client resources for a range of conditions to support people to live meaningful lives.

**APPENDIX A: WEBSITE EVALUATION MEASURE**

1. Please rate the CHD website layout, design, and features.
  - a. Very Difficult
  - b. Difficult
  - c. Neutral
  - d. Easy
  - e. Very Easy
  - f. N/AComments:
2. Please rate the information of the Parent Experience Page.
  - a. Not Useful
  - b. Somewhat Useful
  - c. Neutral
  - d. Useful
  - e. Very Useful
  - f. N/AComments:
3. Please rate the information of the Child Experience Page.
  - a. Not Useful
  - b. Somewhat Useful
  - c. Neutral
  - d. Useful
  - e. Very Useful
  - f. N/AComments:
4. Please rate the information of the School Resources Page.
  - a. Not Useful
  - b. Somewhat Useful
  - c. Neutral
  - d. Useful
  - e. Very Useful
  - f. N/AComments:
5. Please rate the information of the Resources page.
  - a. Not Useful
  - b. Somewhat Useful
  - c. Neutral
  - d. Useful
  - e. Very Useful
  - f. N/AComments:

6. Please rate the likelihood that you will use the CHD website as a resource
  - a. I will not use this website as a resource
  - b. I might not use this website as a resource
  - c. I may or may not use this website as resource
  - d. I might use this website as a resource
  - e. I will use this website as a resourceComments:
  
7. Please rate the likelihood that you recommend this website to a family with a new baby who has a congenital hand difference?
  - a. I will not recommend this website.
  - b. I might not recommend this website.
  - c. I may or may not recommend this website.
  - d. I might recommend website.
  - e. I will recommend website.Comments:
  
8. Elective feedback: What is your overall feedback of the website? What are the areas of improvement? What did you find most helpful?
  
  
9. Elective feedback: What additional information should be included in future revisions of the website?
  
  
10. I am a:
  - a. Parent of a child with a CHD
  - b. A person with CHD
  - c. Health Care Professional
  - d. Other

For questions 1-7, A=1 and E=5.

**APPENDIX B: SPECIAL EDUCATION INVITATION**

*You, too, can have*

***A Hand in Progress...***

You're invited to attend a special presentation by Sarah Tuberty. Sarah worked with our Pediatric Hand team this summer to complete her capstone project for her Doctorate of Occupational Therapy Degree from Boston University. As a Shriners Hospital alum and former patient of Dr. Michelle James, Sarah has developed an online resource for parents of children with congenital hand differences. She'll be sharing her project with us.

Thursday, August 15  
7th Floor Boardroom  
12 Noon – 12:45 PM



Refreshments will be served. Feel free to bring your lunch. Thank you.

## APPENDIX C: SHRINE VINE

### PLEASE POST

August 16, 2019



*The ShrineVine is a newsletter for the staff at Shriners Hospitals for Children — Northern California. Its purpose is to celebrate accomplishment and strengthen the community of care through shared news and information.*

### A Hand in Success • OI Meeting • Lunchtime Lecture • Positive Feedback

**A Hand in Success:** A new online resource will soon be available to parents of children with congenital hand differences. **Sarah Tuberty**, a former patient of **Michelle James, M.D.**, worked with the hand team to develop the site this summer as part of her capstone project for her Doctorate of Occupational Therapy from Boston University. Sarah introduced the site to the hand team and other hospital staff at a lunchtime talk in the boardroom on Thursday. Topics featured include coping strategies, developmental milestones, school resources, patient and parent testimonials, recommended



reading, and even questions to ask your doctor. Among the 20 attendees providing feedback were several members of the hand team including **Janice Conroy**, **Sally Martens**, **Claire Manske, M.D.**, **Relton McCarroll, M.D.**, and Dr. James. Several other interested staff members were there to provide comment and support. The feedback provided allows Sarah to update the online resource. Sarah said, *“I have had the pleasure of volunteering at Camp Winning Hands. In all my conversations with parents, they express a need for more resources and support. I felt that providing an avenue that connects parents to resources was a wonderful role for me.”*

## APPENDIX D: ELECTRONIC FLYER



### A New Online Resource for parents of children with **Congenital Hand Difference (CHD)**

Congenital hand differences occur when a child is born with arms and or hands that look a little different than expected. Often parents feel lack of support and resources for raising their children. This website is here to change that!



**Visit:**  
[shrinerschildrens.org/HandDifferences](http://shrinerschildrens.org/HandDifferences)

#### Topics featured include:

- Addressing parent guilt and stigma
- Coping strategies
- Activity Adaptation
- Developmental Milestones
- Social and emotional anticipatory guidance
- Managing conversations with peers, new people, and teachers
- School resources such as 504/IEP plans
- Developing personal resumes and books
- Children's books and scientific articles on hand differences
- Stories from parents and people living with hand differences
- Roles of Pediatric Hand Team and questions to ask on your visit

Working in partnership with Michelle James, M.D., chief of orthopaedics, Sarah Tuberty developed the new web page as a capstone project for her Doctorate of Occupational Therapy. Born with Congenital Hand Difference (CHD), Sarah was referred to Shriners Hospital for care at a very young age and is a former patient of Dr. James.



***"I have had the pleasure of volunteering at Camp Winning Hands for 10 years. In all my conversations with parents, they express a need for more resources and support. I felt that providing an avenue that connects parents to resources was a wonderful role for me. By facilitating these connections, we can build a stronger community where children and parents feel they belong."***

– Sarah Tuberty, Occupational Therapist living with CHD

***"This website is a substantial blend of community resources, recommended reading, scientific literature, and personal experiences. The result is a site filled with information that specifically addresses common parent concerns and anxieties. The information helps parents who visit the site feel supported, part of a community and able to imagine a beautiful future for their family."***

– Michelle James, M.D., Pediatric Hand Surgeon & Chief of Orthopaedics



**Shriners Hospitals**  
for Children®— Northern California

#### For more information:

Parents with questions, comments or concerns regarding Congenital Hand Difference (CHD) are encouraged to contact the Northern California hand team. Please email us at [ncl.chd@shrinenet.org](mailto:ncl.chd@shrinenet.org)

**APPENDIX E: WEBSITE ADDRESS**

<http://shrinerschildrens.org/handdifferences/>



**APPENDIX F: EXECUTIVE SUMMARY**

Online Resource for Parents of Children with Congenital Hand Differences

Shriners Hospital of Northern California

Sarah Tuberty

Ellen S. Cohn, ScD, OT, OTR, FAOTA

Michelle James, MD

**Problem statement**

Children with congenital hand differences (CHD) may experience bullying and social isolation, but lack access to psychosocial and accommodation needs. However, there are few existing resources tailored to this population (example: many resources are for a general group of children with disabilities). Therefore, with the implementation of an online parent resource, parents can be better equipped to assist their children's engagement in social and school occupations.

**Problem Background**

Children with congenital hand differences (CHD) are at risk for social isolation, increased anxiety, and lower rates of self-esteem when compared to their typically developing peers (Lumsdaine et al, 2016). Parents may a sense of isolation or guilt that they caused the abnormality. In addition, they may be searching for resources to best support them in their parenting (Ardon, Janssen, Hovius, Stam, & Selles, 2012; Murray, Kelley-Soderholm, & Murray, 2007). When societal expectations of how to behave or look a particular way are imposed on people, individuals with differences may be viewed as belonging to an "other" category deprived of social privileges which are standard to

those in the “in-group” (Goffman, 1963). Many children and parents lack access to the resources to effectively manage and cope with negative assumption of their ability.

### **Program Overview**

This doctoral capstone project is based on a synthesis of the research literature, clinical and personal experiences. An online parent resource page was developed to provide parents with resources to increase feelings of support, community, and to develop effective coping for parents of children with congenital hand differences. The website includes resources in the following domains:

<b><u>Landing Page</u></b>	<b><u>Parent Experience</u></b>	<b><u>Child Experience</u></b>	<b><u>School Resources</u></b>
Welcome to website	Imagined Future	Developmental Milestones	School Requirements
Brief introduction to CHD	Greif and Coping	Activity Adaptation	School Services
	Stigma	Stigma	Talking to Your Child’s
<b><u>Pediatric Hand Team</u></b>	Making Medical	Social and Emotional	Teacher
Hand Surgeons	Decisions	Anticipatory Guidance	About Me
Geneticists		Coping	Personal Resume
Anesthesiologists	<b><u>Experience Profiles</u></b>	Managing Conversations	Navigating
Advanced Practice	Child	Social Scripts and Creating	Conversations with in
Providers	Teen	a Story	the classroom
Nursing Physical and	Adults		
Occupational Therapists	Parents	<b><u>Additional Resources</u></b>	<b><u>About Us</u></b>
Prosthetists		Organizations	Sarah Tuberty, OT/s
Care Management		Camps	Dr. Michelle James, MD
Child Life Specialists		Children’s Literature	
Transition Care		Scientific Literature	
		Media	

### **Outcomes**

Parents were asked to rate the ease and usefulness of the website and whether they were likely to use and recommend the website as a resource. Parents reported that they found the website easy to navigate with useful content and that they would use and recommend the website to new parents of children with CHD.

### **Recommendations**

Online resource websites are beneficial at communicating health information. It was recommended to add additional parent profiles, additional pictures of people with CHD engaging in tasks, and a section on how parents can apply for grants or scholarships to help cover health care costs and equipment.

Hospitals should consider building similar resource websites for various diagnosis in order to best support parents.

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**CURRICULUM VITAE**

