DEVELOPING SELF-MANAGEMENT SKILLS IN PERSONS WITH SPINA BIFIDA THROUGH mHealth APPLICATIONS: DESIGN AND CLINICAL EFFICACY

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The medical complexity of spina bifida results in equally complex self-care regimes that many persons with spina bifida (SB) struggle to effectively manage and maintain. Lack of consistent follow-through of self-care routines and health care recommendations, often results in the development of secondary conditions, impacting morbidity and mortality in this population. This dissertation provides a description of the development and initial clinical testing of an innovative mHealth system to help support adults with SB and improve self-management skills. The clinical needs and functional abilities of the SB population were integrated into a mHealh system called iMHere (Interactive Mobile Health and rehabilitation). The iMHere system consists of software including a suite of smartphone apps specifically designed for persons with SB, a clinician portal, and a communication system connecting the two.

The three primary phases of this study are 1) development, 2) usability testing and 3) clinical application. Due to the nature of development, feedback and improvements to the system continued throughout a year-long randomized control trial (N=26), conducted to determine the clinical efficacy of the system. Usability testing for this project was intensive and occurred in a step-wise manner. Detailed data was collected to revise and improve upon the design and functionality of the smartphone apps.

Results for the first six months of the clinical study are also shared comparing self-management outcomes for the intervention group using the iMHere system versus the control group receiving traditional care through the UPMC Adult SB clinic. Significance was found in looking at time x group (p = .006). In particular, improvement in self-management skills as assessed by the AMIS-II are noted in the intervention group participants from three months to six months. A moderate effect size of 0.46 was found in the association of group and time as calculated with change scores.

Limitations of the clinical study are discussed at length as well as potential future research opportunities including expanding usage of the iMHere system to other populations with chronic conditions and application of the intervention during the transition years (14-21 years).

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PREFACE

This study was made possible through funding from the Department of Education's, National Institute for Disability and Rehabilitation Research (NIDRR) with a grant for the Rehabilitation Engineering and Research Center on Telerehabilitation (RECR-TR) (Grant #: H133E090002). We were also fortunate to receive support from the Verizon Foundation for the overall project "Telerehabilitation Enhanced Wellness Program for Spina Bifida." Without funding from these sources, this project would not have been possible.

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1.0 INTRODUCTION

Spina bifida (SB) has been described as the most common permanently disabling birth defect, which affects over 166,000 Americans (SBAA, 2013). Occurring very early in development, SB is a form of neural tube defect (NTD). At only 18 days after conception, when the embryo is only about the length of a grain of rice, abnormal development of the spinal cord and surrounding structures occurs in the open forms SB (Sandler, 1997). Approximately 1,500 infants with SB are born in the United States annually (Parker, et al., 2006). Due to advances in medical care and interventions, 75% of children born who are born with SB are now expected to survive into adulthood and are living longer now than ever before (Bowman, McLone, Grant, Tomita & Ito, 2001).

While the UPMC Adult SB Clinic is a leader in providing care for persons residing in Pittsburgh and surrounding regions, this clinic is one of only four of its kind in the United States (Dicianno, et al., 2008). Our current health care system is unequipped to adequately provide services to adults with SB. Few SB clinics exist to provide care for a growing population of adults with SB who are outliving or no longer able to receive care and support from aging parents.

1.1 THE MEDICAL, FUNCTIONAL & PSYCHOSOCIAL IMPACT OF SB

Spina bifida, meaning “split spine” in Latin, is used to describe a group of NTDs resulting from an incomplete closure of the spinal column during prenatal development. Spina bifida and other NTDs occur very early in pregnancy, between the 14^{th} and 28^{th} day after conception. During embryonic development, an opening can occur in the formation of the neural tube over the affected area of the spinal cord. As a result, there is a short spine on each side of open spinal cord rather than a single one in the midline (Sandler, 1997). According to the Spina Bifida Association of America (SBAA), every day an average of eight infants are born in the United States affected by SB or a similar NTD.

SB is considered by many to be the most complex disability an individual can have that is compatible with life (SBAA, 2010). SB is a complex multisystem condition with orthopedic, urologic, gastrointestinal, neurological, musculoskeletal, immunological, and neuropsychological components that require life-long management (Dicianno, et al., 2008). Because SB involves so many bodily systems, this condition presents with an array of difficulties, which have varying degrees of functional impact from person-to- person.

There are several types of SB, with the three most commonly occurring types being: SB Occulta, SB Meningocele and SB Myelomeningocele [FIGURE 1]. These figures illustrate lateral cross sections of the lower back depicting the various types of SB impact the spinal cord and related structures.

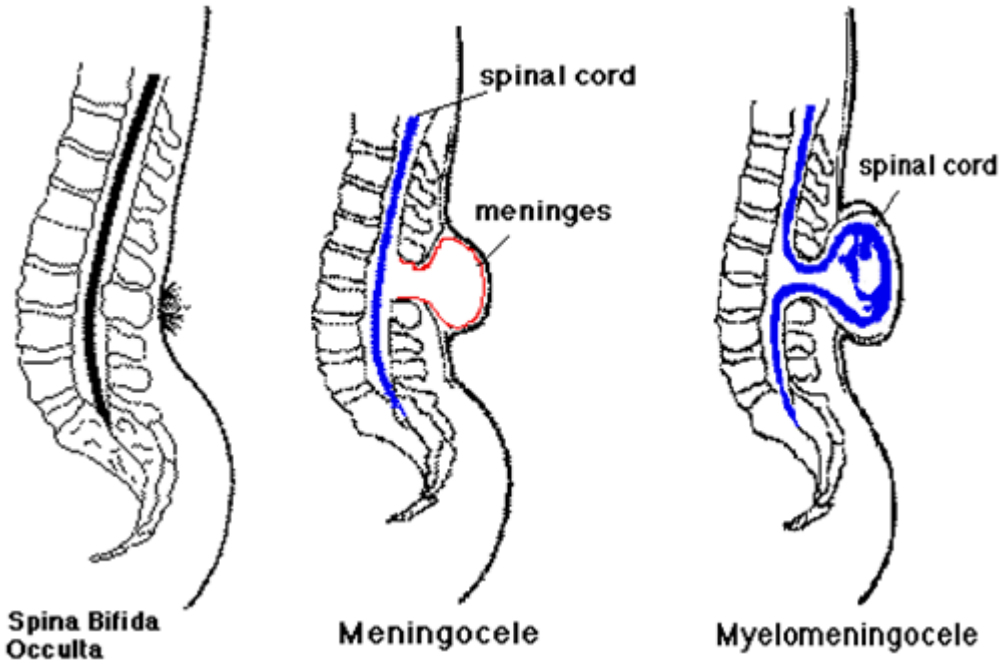


Figure 1. Types of Spina Bifida

Spina bifida occulta is a very mild, common form of SB usually resulting only in a slight deficiency in the formation of one of the vertebrae. "Occulta" means hidden, and the defect is often not at all visible externally. Visible signs, if any, are a dimple or small hair growth on the back and rarely is there any functional deficiency resulting. Persons with SB occulta have an opening in one or more of their vertebrae without any apparent damage to their spinal cord. Spina bifida occulta is usually discovered accidentally when the person has an x-ray or MRI for some other reason. The prevalence of occulta is not known, but it is probably the most common type of SB. It is has been estimated that as many as 5 - 40% of all Americans may have SB occulta, but because they experience little or no symptoms, very few ever know they have it (SBA, 2010).

Individuals whose functioning is most greatly impacted by SB generally have either: 1.) Meningocele (MC) – a cystic protrusion from the back with no neural elements contained in the sac and minimal neurologic deficits or 2.) Myelomeningocele (MMC) – defect with a cystic

protrusion in the posterior spinal elements with neural structures found within the hecal sac leading to major neurologic deficits (Sandler, 1997).

In persons with MC, the membrane that surrounds the spinal cord may enlarge, creating a lump or "cyst." This cyst is often invisible through the skin and causes no problems. If the spinal canal is cleft, or "bifid," the cyst may expand and come to the surface. In such cases, since the cyst does not enclose the spinal cord, the cord is not exposed. The cyst varies in size, but it can almost always be removed surgically if necessary. Myelomeningocele occurs in three to seven in 10,000 live births, with up to 50% of affected pregnancies terminated in some regions (IFSBH, 2008). Myelomeningocele is the most complex and severe form of SB. Usually involving neurological problems, the impact of MMC can be very serious or even fatal. A section of the spinal cord and the nerve roots that stem from the cord are exposed and visible on the outside of the body. Or, if there is a cyst, it encloses part of the cord and the nerve roots. Myelomenigocele has the greatest degree of negative impact on physical, cognitive and psychosocial functioning and is the most severe and medically complex form of SB. The impact of MMC upon these various areas of function is further described in the following sections.

1.1.1 The Medical Complexity of Myelomeningocele

The focus of this research study is on persons with a primary diagnosis of MC. Physical, sensory and cognitive deficits are often seen in persons with MC and hydrocephalus which may impact functional performance. As previously mentioned, SB impacts many systems of the body; therefore SB also has a diverse and significant impact across functional performance domains as well. The neurological deficits associated with SB, in particular, are directly related to mortality

and disability (Hunt & Oakeshott, 2003). The next sections discuss ways in which these deficits may present as functional impairments in everyday life for persons with SB.

1.1.2 Effect of Myelomeningocele on Physical Function

The orthopedic, urologic, gastrointestinal, neurological, musculoskeletal effects of MC often significantly impact physical function in this population (Sandler, 1997, Dicianno, Kurowski, et al, 2008). In particular, the neurological defects which occur during development of persons with MC, often results in paralysis leading to mobility impairments. Motor impairments tend to correspond with the individual's lesion level. The higher the level of the NTD lesion, the greater the physical deficits the person with SB will experience (Verhoef, et al 2007). The nerve roots innervate different muscle groups. For instance, if the lower thoracic region (T12) of the spinal cord has been impacted, the hip flexor muscles may not function properly. The hip flexors are important muscles for walking. As a result, a person with SB who has a T12 lesion may not be ambulatory. A person who does not have use of motor function in his or her lower extremities is described as having paraplegia. Persons with SB may have different levels of paraplegia. These levels are generally classified as: thoracic, high lumbar (L1 or L2), mid-lumbar (L3), low lumbar (L4 or L5) or sacral (S1) (Sandler, 1997). Due to limited motor function of the lower extremities, persons with MMC often require the use of crutches, braces, and/or wheelchairs for mobility.

Persons with MMC also commonly have a secondary diagnosis of hydrocephalus. Hydrocephalus is an accumulation of cerebrospinal fluid (CSF) which arises from an imbalance in the production and drainage of that fluid. "More than 85 % of children with SB also have hydrocephalus or develop this at a later stage" (IFSBH, 2008). Hydrocephalus can be medically

managed through surgical placement of shunts. Shunting is used to control the pressure by draining the CSF. However, shunting does not "cure" hydrocephalus. Most persons with hydrocephalus are likely to require several shunt revisions during their lifetime due to malfunctions. Shunt malfunctions can lead to significant morbidity, mortality, and sudden death (Tomlinson & Sugarman, 1995).

1.1.3 Cognitive Deficits in Myelomeningocele

As previously noted, SB is not just simply a physical disability, but a complex condition. Frequently, persons with MMC present with cognitive deficits. Shunted hydrocephalus is known to be associated with poorer outcomes in psychological health, function, and employment (Oi, 1996) compared to no history of hydrocephalus. Likewise, those with other types of SB besides MMC have fewer neurological and medical complications (Linder, 1982; Satar, 1997). The majority of individuals in the U.S. with non-occult forms of SB have MMC and hydrocephalus (Brinker, 1994; Kumar & Singhal, 2007), as does our clinic population. Each time the brain experiences the increase in pressure from hydrocephalus, damage can occur either temporarily or permanently, decreasing cognitive function. Studies have shown that presence of the associated pathology of hydrocephalus, rather than SB per se, has a negative effect on cognitive status (Barf et al., 2003; Iddon, Morgan, Loveday, Sahakian, & Pickard, 2004). Particularly those individuals with MMC and hydrocephalus who have undergone shunt revisions after the age of two years, "are associated with poor long term achievement as adults with SB" (Hunt, Oakeshott, & Kerry, 1999, p. 591). In absence of hydrocephalus, young adults with SB when compared to the general population showed relatively unimpaired cognitive functioning measured with standardized neuropsychological testing (Barf et. al., 2003; Iddon, et. al., 2004).

Performance on Intelligence Quotient (IQ) testing has also been correlated with ventricle size and the level of lesion or highest open vertebral arch at birth (Beeker, Scheers, Faboer & Tulleken, 2006; Bier, Moraels, Liebling, Geddes, & Kim, 1997). In persons with SB, specific higher order cognitive functions such as: executive function, processing speed and memory are often negatively impacted (Dise & Lohr, 1998, Iddon, et al., 1996). Overall, persons with SB and hydrocephalus, although often classified as physically disabled, also frequently present with significant cognitive challenges.

1.1.4 The Psychological Impact of Spina Bifia

Studies have shown increased incidence of depression in persons with SB in comparison to the general population. "Young people with SB are at a higher risk of depressed mood and lower self-worth, and are more likely to think about suicide" (Liptak, 2008). In a recent cross-cultural study which compared Norwegian adults with SB and adults with SB residing in Oregon, quality of life measures revealed similar findings between the two cultures that indicate significant concerns with regard to anxiety and depression. Forty-one percent of the Norwegian sample and 47% of the Oregon sample indicated that they suffered from depression. Likewise, 19% of the Norwegian subjects reported anxiety and 23.5% of the Oregon residents with SB reported difficulties with anxiety (Kalfoss & Merkens, 2006).

1.2 SELF-MANAGEMENT AND SPINA BIFIDA

Almost half of all Americans, more than 145 million people, are living with chronic conditions (Kung, Hoyert, Jiaquan & Murphy, 2008). Current evidence suggests that patients with chronic conditions who have effective self-management skills make better use of health care services and have enhanced self-care (Lorig, et al., 1999). The primary objective of developing self-management skills in persons with chronic conditions, such as SB, is to enable them to be actively involved in the center of their own health care in order to maximize quality of life. Lorig and Holman (2003) explain self-management involves the person through: *"… engaging in activities that protect and promote health, monitoring and managing symptoms and signs of illness, managing the impacts of illness on functioning, emotions and interpersonal relationships and adhering to treatment regimens"* (p.1). In providing services to enhance or increase self-management skills, health care professionals often work to enable individuals to: *"… make informed choices, to adapt new perspectives and generic skills that can be applied to new problems as they arise, to practice new health behaviors, and to maintain or regain emotional stability"* (Lorig, Mazonson & Holman, 1993, p. 438).

1.2.1 Impact of Cognitive Deficits on Self-Management Skills

Hayden, Davenport and Campbell (1979) were some of the first researchers to document the need for increased self-management skills in youths with SB. Despite the identification of this need, over thirty years later there still remains a limited understanding of the experience of self-management and strategies to develop these skills in persons with SB. The potential reasons for difficulty in following through with such a complex health regime are multiple for this

population. One of the most salient reasons is the persistence of cognitive deficits in the area of executive function for this population in relationship to the complexity and demands of what is required. Learning self-care requires the ability to understand treatments; to organize personal time; to remember appointments, treatments, and medications and their administration times; to remember potential complications and whom to call when problems arise; to be able to solve problems and make decisions about treatments. Without these abilities, independent of self-care tasks and maintenance of personal health care is unattainable.

Executive function is considered to be central to the completion of Activities of Daily Living (ADLs) particularly when these ADLs are expected to be initiated and completed at fixed time intervals throughout the day. Direct correlations have been found between executive dysfunction and inability to complete many of the necessary self-care tasks independently in those with MMC (Hunt, et al., 1999). Regardless of IQ, the standard measure of intelligence, more in depth neuropsychological testing has revealed that persons with SB often show evidence of significant impairment in the area of executive functioning (Dise & Lohr, 1998). Many persons with MC struggle to meet the demands of daily life and carry out ADLS independently. While the individual tasks of ADLs are fairly simple, self-care routines can be quite complex from a cognitive perspective involving the need to plan, initiate and execute numerous tasks at different times throughout the day. Medication routines, self-catheterization and daily skin checks are examples of common self-care tasks which are required of persons with SB and demand more complex cognitive processes. These complex cognitive processes include: "executive functions, including, but not limited to, organization, planning, attention, initiation, and pro-social skills that are often underdeveloped in children and adults with SB" (Calvery, M., 2008, p.10). Specifically, research has shown executive functioning deficits in persons with SB

in areas including: initiation, working memory, problem-solving and self-monitoring (Mahone, Zabel, Levey, Verda & Kinsman, 2002). Executive function deficits are not the same as "intelligence." In fact, individuals with MMC often present with strengths in verbal expression and facility, which are indicators of intelligence. Persons with MMC also tend to have extensive vocabularies and are very talkative. Often persons with MMC are typically strong academic performers as young children. As these individuals develop and age, an increased level of independence is anticipated developmentally in managing self-care, school work, and general activities of daily living. Unfortunately, as these persons mature, indicators of these executive function deficits, a lack of follow-through and the inability to develop or maintain organizational skills, are common functional barriers to successful transition to adult roles and the ability to independently maintain self-care routines without external support. Kafloss and Merkens (2006) discovered discouraging figures with respect to cognitive decline in their cross-culture comparison study of quality of life in adults with SB. Of the Norwegian participants in this study, 61% reported cognitive problems affecting daily activities and 31% experienced cognitive decline during the past six months. The Oregon sample of participants fared even worse with 65% reporting cognitive problems affecting everyday activities, and 33% reporting declining cognitive status (Kafloss & Merkens, 2006).

Self-management skills typically evolve from opportunities to engage in problem-solving activities, such as accessing resources (Hughes, 2004) or when one makes decisions about goals and treatment interventions (King, Shultz, Steel, Gilpin & Cathers, 1993). This evolution of self-management skills ideally begins in youth, within the protected home environment with the assistance of a parent or other caregiver. Reduced autonomy and decision-making skills have been observed in adolescents with SB (Monsen, 1992). Increasing knowledge of one's health

status is one method of intervention. The more comfortable persons are in their health knowledge and ability to carry out tasks the more likely they are to believe that engaging in such activities will result in desired outcomes (Nodhturft, et al, 2000).

1.2.2 Psychosocial Aspects of Self-Management

The following sections provide descriptions of several psychosocial issues commonly observed in persons with SB and other developmental disabilities. The literature reveals how each of these issues has an impact on self-managements skills in this population.

1.2.2.1 Self-Efficacy and Learned Helplessness

The Health Belief Model, developed to predict a person's compliance with preventive health behaviors (Hochbaum, 1956), is based on the theory that people weigh the perceived benefits of the behaviors against the perceived barriers when they are deciding whether to comply. Little time and attention is given by parents or professionals to discussing personal values of health care with young people disabled by SB, let alone helping them to identify and clarify their values. Lack of clear health care values contributes significantly to their failure to assume responsibility for healthy behaviors.

One study of self-care in chronic illness identified a direct relationship between self-concept and psychological status and the achievement of self-care (Connelly, 1993). This study also found that informing the person regarding the condition and treatment options was necessary if he or she were to maintain appropriate health care. The most unexpected finding was that a decrease in social support promoted an increase in self-care behaviors. This indicates that

the more helpful others are, the less likely it is the young person will assume total care for himself or herself. Hirot and Seligman's (1975) theory of learned helplessness and the literature on self-efficacy support these findings.

As a key founder in the concept of self-efficacy, Bandura (1977) developed social cognitive theory and defined self-efficacy as people's beliefs about their capabilities to produce designated levels of performance that exercise influence over events that affect their lives. Throughout his research, Bandura (1993) found that self-efficacy was the strongest predictor of behavior aimed at reaching a desired goal or outcome. Bandura identified four factors as influencing persons' levels of self-efficacy. The strongest factor is performance experience. Previous experience in obtaining or not obtaining desired outcomes most significantly influences self-efficacy. Obtained through observation of others, vicarious experiences follow, also having an impact on self-efficacy to a lesser extent than performance experience. The third factor is verbal persuasion. Verbal persuasion's effectiveness in bolstering self-efficacy is influenced by the level of expertise or trustworthiness of the person providing the persuasion. Verbal persuasion has the least influence on a person's level of self-efficacy when compared to performance and vicarious experiences. Finally, physiological and emotional states also influence self-efficacy. For instance, an athlete who is in pain or is fatigued may have less self-efficacy regarding his or her ability than if pain-free and well rested (Maddux, 2002).

Bandura's (1977) concept of self-efficacy was specific to particular situations. In other words, a person will possess certain self-efficacy beliefs given a specific physical environment, social context, and demand that are placed upon the individual. Sherer and colleagues (1982) modified Bandura's concept allowing for the generalization of self-efficacy from a situation and time specific scenario to a self-perception regarding one's ability to successfully function leading

to the achievement of defined goals (Magaletta & Oliver, 1999). Maddux (2002) succinctly and aptly explains self-efficacy as "the power of believing you can" in his chapter on self-efficacy within *The Handbook of Positive Psychology* (p. 336). Maddux further explains self-efficacy beliefs as what an individual believes can be accomplished with his or her skill set:

> *"Self-efficacy is not perceived skill; it is what I believe I can do with my skills under certain conditions. It is concerned not with my beliefs about my ability to perform specific and trivial motor acts but with my beliefs about my ability to coordinate and orchestrate skills and abilities in changing and challenging situations"* (Maddux, 2002, p. 278).

Conversely, learned helplessness is the perceived absence of control over the outcome of one's situation (Hirot & Seligman, 1975). Learned helplessness is essentially the opposite condition of a strong sense of self-efficacy. Importantly, Seligman's learned helplessness theory explains that hope and biological life are inextricably linked. Learned helplessness theory also connects persons who exhibit learned helplessness with clinical depression and other psychiatric conditions (Hirot & Seligman, 1975). For example, a pessimistic person tends to see negative situations as pervasive ("I can't do anything right"), personal ("It's my own fault") and permanent ("things will never change). Using a positive or strengths-based approach, it would make sense that learned helpless can potentially decrease through improvement of self-efficacy beliefs (Hirot & Seligman, 1975).

Many parents and professionals assume that young persons with developmental disabilities have significantly lower self-esteem and poorer self-concepts than their able-bodied peers. However, studies have found that there is no major difference in self-esteem between people with disabilities and their peers without disabilities (Arnold & Chapman, 1992; King, Shultz, Steel, Gilpin, & Cathers, 1993). Lowered self-efficacy beliefs may be hindering the achievement of personal health care behaviors in some young persons with SB. Other parallel

factors may also be contributing to this problem. Motivation and persistence in working toward a goal has been found to be lower in people with physical disabilities (King et al., 1993). Motivation is based upon the nature of the challenge, the experience of failure that sets the boundary of competence, and the reinforcement of efforts to succeed. A lack of persistence emerges when others are always there to provide assistance. Problems with motivation and persistence are common in the families of children with SB (King et al., 1993). Few parents allow a child with a developmental disability to experience failure. Many parents explain that there is so much their son or daughter cannot do because of the disability that letting the child fail at something else is not fair. The young person with SB, whose parent provides continuous support, does not experience competence by discovering personal boundaries or learning the consequences of inaction. Incompetence or helplessness learned in childhood contributes to the incompetent or helpless behavior of the adult (King et al., 1993).

Noncompliance can exasperate healthcare professionals, leaving them worrying about the effective outcome of their efforts and medical care. It also results in noncompliant persons being labeled as 'difficult' or 'troublesome'. It is suggested that professionals who label a patient as noncompliant are following convenient paternalistic principles rather than considering the impact of a prescribed regimen or a rehabilitation plan upon an individual. Autonomy and respect must be foremost in patient care. Furthermore, compliance does not necessarily indicate that professionals and individuals have developed a collaborative and understanding relationship. Noncompliance is described as a lack of recognition by the health care professional of the meaning of the regimen to the patient. Interventions will be most successful when the patient participates in the rehabilitation plan. Without acknowledgement of the individual as an equal partner, and listening to his or her narrative, care will be, at best, paternalistic (Moore, 1995).

While difficulties persons with SB experience in cognitive functioning play a part in the lack of follow through, there are yet instances when cueing or coaching is provided, yet some persons with SB resist this assistance. Further exploration of these persons' perception or attitudes toward receiving personal care assistance is necessary. Personal health care is a lifelong responsibility for all people. These important factors may impede a disabled person's assumption of responsibility for his or her personal health care:

- Poorly identified and developed health care values, and
- Low motivation and lack of experience in persistence behaviors.

Often the person's behavior may be misunderstood as "laziness" or seen as being "non-compliant," "uncooperative," "stubborn" or "unmotivated" even though they may eagerly engage in dialogue about interests and desires for independence (Calvery, 2008).

1.2.2.2 Influence of the Family System

The development of self-management skills can also be influenced strongly by the family environment as discovered in a retrospective study by Loomis, Lindesy, Javornisky and Monahan (1997). Over-protective parenting styles, commonly encountered in families with children who have SB, reduce opportunities for decision-making, which can hinder the development of self-management skills (Holmbeck, et al., 2002). A pattern of dependent behavior is often exhibited by adolescents with SB on their parents for self-care in areas that peers have developed independent skills such as bowel management and home responsibilities (i.e. chores) (Blum, et al, 1991). Likewise, Davis, Shurtleff, Walker, Seidel and Duguay (2006), discovered delays of two to five years in autonomy and self-care skills in teens with SB. Sawin, Bellin, Roux, Buran and Brei (2009) note: "enhanced self-management is critical to improve the

health status and quality of life" in persons with SB (p. 28). The SB population was selected for this study due to the need to establish effective self-management strategies as well as the nature and complexity of this diagnosis which creates an opportunity for the development of a unique intervention using telerehabilitation (TR) delivery methods for providing rehabilitation, monitoring of self-care tasks and on-going support. While, self-management strategies have been studied prospectively with other populations, this is the first study of its kind looking at the efficacy of a mHealth application as a clinical intervention to help develop or increase self-management skills in persons with SB.

1.2.2.3 Complexity of Self-Care Routines for Persons with Spina Bifida

Not only do persons with SB have difficulty following through with health care recommendations and maintaining self-care routines due to cognitive deficits, the complexity of the multi-system impact of their diagnosis also makes self-care routines complex. A multitude of activities must be completed every day, throughout the day for persons with SB to remain healthy and prevent the development of secondary conditions. The most common tasks or activities persons with SB need to engage in regularly include: medication management, self-catheterization routines, bowel management programs, monitoring of skin for signs of breakdown and/or wound care, nutrition management, maintaining adequate water intake, and engaging in physical activity as part of one's lifestyle despite physical limitations to ensure cardiovascular health and assist with weight management.

Medication Management:

Anyone who has ever been told to take some new medicine three or four times a day knows that it is not always easy to follow the prescribed regime on time consistently. Persons with SB are

frequently prescribed several medications for the management of incontinence (anticholinergics) or seizures, stool softeners, antidepressants, etc. In combination with the cognitive deficits frequently observed in persons with MMC, medication compliance is a significant challenge. A listing of the most commonly prescribed medications for persons with SB is included in Appendix A.

Clean Intermittent Catheterization (CIC):

About 95 percent of persons with MMC have a neurogenic bladder. This means they are unable to perceive the sensation of bladder fullness and lack the neurologic integrity to have coordinated contraction of the bladder muscle and opening of the bladder sphincter (Sandler, 1997). Many persons with MMC have uninhibited bladder contractions, which may be accompanied by high bladder pressures. Some persons may be able to empty their bladder partially by straining, but the emptying is incomplete. Even small amounts of residual urine in the bladder can lead to urinary tract infections (UTIs). The combined situation of high bladder pressure and infection can place the kidneys at risk. Simply emptying the bladder several times per day, CIC is performed by inserting a tube through the sphincter into the bladder or through an artificial stoma. This process serves to protect the kidneys and for many persons with SB when performed on a regular, routine schedule allows them to achieve urinary continence. Usually, CIC should occur five to six times during waking hours, which is every three to four hours throughout the day. Problems can be encountered during CIC which are indicators of medical issues such as infection or injury. Since many persons with SB lack sensation, they often do not experience the discomfort or pain that most persons who develop UTIs experience. However, limited volume, dark and/or foul-smelling urine are a few of the indicators of infection that can be quickly and easily detected during CIC. Noticeable amounts of blood in the urine can be a

sign of more serious UTI or kidney infection. Whereas, a small amount of blood may simply be caused by trauma to the urinary tract that occurs during catheterization. In addition to the appearance and smell of the urine, symptoms such as: dysuria, abdominal pain and/or fever, are indicators of possible infection. UTIs and kidney infections require prompt medical attention and the person will frequently need to follow up with a physician (Sandler, 1997).

Management of Neurogenic Bowel

Bowel continence is critical to medical issues (constipation, skin breakdown), mental health issues (self-esteem), and social issues (isolation). Persons with SB have a tendency toward constipation. The problem often begins in infancy and may never be properly addressed or treated. The lack of rectal sensation is the key factor in the fecal incontinence of people with SB. Persons with high spinal lesions have low internal sphincter pressure and rarely experience rectal sensation. Those persons with low spinal lesions have increased internal sphincter pressure and often have intact rectal sensation (Agnarsson, Warde, McCarthy, Clayden, & Evans, 1993a). Constipation, in both cases, can also be attributed to inadequate intake of fluids, limited physical activity, lack of fiber in the diet, or as a side effect of medication.

Constipation can lead to impaction with overflow incontinence or simply constant elimination of hard stool with movement. Treating the constipation aggressively is the first step in continence management. An effective bowel maintenance program needs to be completed at regular, consistent times as part of the person's routine. Forgetting to perform any portion of a bowel program on time or at regular intervals will make it ineffective, and incontinence may likely occur. People use a variety of different methods to maintain bowel continence. A bowel management program may consist of one or more of the following methods of intervention:

- Oral Medications (see Appendix A)

- Exercise and a healthy diet
- Timed toileting regardless of location
- Suppositories
- Digital stimulation
- Small and large volume enemas (SBAA, 2005, p. 23-24).

Management and Monitoring of Skin Integrity

Persons with SB have to be constantly vigilant for skin injury and breakdown. The neurologic problems associated with the lesion level of injury to the spinal cord result in loss of sensation typically to some portion of the lower body in persons with SB. Loss of feeling means that little or no discomfort is felt; therefore, there is no trigger to indicate a need to reposition oneself and reduce the pressure on a particular part of the body. Incontinence causes the skin to become even more susceptible to breakdown and infection. Furthermore, the lymphatic and circulatory systems of persons with SB often do not function as efficiently as they should (Mukherjee, 2007). These systems act to together to remove fluid and waste products. Without proper functioning of these systems, nutrients and oxygen are not received in adequate supply and edema will accumulate in the lower extremities (Dicken, et al, 1998). These combined issues mean that pressure ulcers can develop very quickly in persons with SB, but tend to heal very slowly (Sandler, 1997).

While there are many different types of wounds, pressure ulcers are the most common cause of skin injury resulting from remaining in one position such as prolonged sitting. This type of pressure ulcer generally begins as an area of redness just below the buttock, where the ischial tuberosity comes into closest proximity to the sitting surface. However, a pressure ulcer can occur anywhere on the body, not just person's sitting surface. As the underlying tissue is affected, there may be a "knot" of hard, swollen, or inflamed tissue under the skin. If the problem continues to progress, a blister may appear on the surface of the skin as a result of

damaged, dead tissue. Discharge of blood and pus may be apparent on clothing. A small red area or wound often is a sign of much more extensive damage to underlying tissues (Reddy, Gill & Rochon, 2006).

A retrospective study by Beierwaltes, et al., (2009) reveals 76% of persons with SB have had a dermal ulceration (48% sacral; 45% foot). Only 10% of these ulcers were caused by burns, the majority (90%) were caused by prolonged pressure. Since the majority of problems found in persons with SB related to skin integrity are classified as pressure or decubitus ulcers, we will focus on this type of wound for brevity sake. However, there are many other types of wounds which as well associated with nutrition, shear, temperature and a variety of other factors (Dicianno, et al., 2008). The incidence of pressure ulcers in this group of persons with SB was correlated with incontinence and lack of adherence to self-care programs (Beierwaltes, et al., 2009, Kinsman, et al, 1996) Treatment of pressure ulcers can include daily dressings and applications of topical gels. Relief from pressure is critical to allow for healing of the affected area. Special cushions and mattresses may be needed during the healing process and if the problem is recurrent, further assessment of pressure relief aids, equipment (i.e. wheelchairs and cushions) and lifestyle are warranted (Sandler, 1997). Daily activities to prevent the development of pressure ulcers are necessary components of self-care for persons with SB. The entire body should be inspected both morning and evening to detect any redness or changes in the skin. *The Health Guide for Adults Living with Spina Bifida* (SBAA, 2005) recommends: "Check skin for cuts, bruises, scratches, swelling, and red marks – especially inspect the buttocks and all sides of the feet and between toes" (p. 28). Assistance may be required from a care attendant or aide for bathing and other hygiene tasks, but the adult individual must be an active participant in the awareness of his or her own personal health. This includes awareness of skin integrity even if

the issues are located in areas that are insensate and not readily viewed by the person. Use of a long-handled mirror is encouraged for visual inspection of areas of the body such as this with or without the assistance of another person (SBAA, 2005).

Reducing the risk of a pressure ulcer developing is accomplished with vigilant efforts to maintain proper circulation to all insensate areas of the body. Pressure relief is recommended by changing the body's position at least every 20 minutes. Wheelchair push-ups can be performed to allow blood to flow normally, which is all the time that is needed. Persons who are unable to change their position by doing wheelchair pushups or leaning forward and side-to-side should have a power wheelchair that is fully equipped with tilt and recline functions that enable performance of pressure relief at the recommended time intervals. Otherwise, the person is continuously in need of assistance of others to perform pressure relief. Pressure relief must also occur when persons are in bed for extended periods of time unable to change positions. Persons may require a special bed or mattress to reduce pressure over insensate areas and bony prominences as previously described (SBAA, 2005).

1.3 DIFFICULTIES IN NAVIGATING THE HEALTH CARE SYSTEM

Over the past 30 years the quality of life and longevity has improved for persons with SB (Betz, 2004; Bowman, et al, 2001; Sawin, et al., 2010). Until the 1970's, when the concept of CIC was introduced, many persons with SB died from renal failure as young children (deJong, et al, 2008). Uncontrolled hydrocephalus was also another primary for morbidity in children with SB. With the advent of CSF shunting and CIC, this prognosis has been transformed and survival rates

have greatly increased (Davis, Daley & Shurtleff, 2005). We now have a new generation of young adults with SB who face age related tasks to become self-sufficient and plan for their futures. Once adulthood is reached, many persons with SB are left to navigate an adult healthcare system after having been served for their entire lives through a comprehensive and coordinated pediatric clinic (Rauen, et al, 2013). Historically, there has been little coordination across the multiple settings, providers and treatment interventions for persons with chronic conditions and disabilities. In addition, the clinical interventions for chronic diseases are often complicated (i.e. lymphedema therapy, wound care, etc.) making it difficult for patients to comply with treatment protocols. This is particularly true for adults with SB.

Effective medical care of persons with chronic conditions generally requires longer visits to the physician's office than is common in acute care due to the complexity of the condition. Moreover, in treating chronic illnesses, the same intervention, whether medical or behavioral, may differ in effectiveness depending on when in the course of the illness the intervention is recommended.

Fragmentation of medical care is a large risk for patients with chronic conditions, because secondary conditions or multiple chronic diseases coexist. Interventions may require input from multiple specialists that may not usually work together, and to be effective, they require close, careful coordination. As a consequence, patients with chronic conditions often fare poorly in the current acute-care model of care delivery. Persons with SB have shown some of the poorest outcomes even in comparison to other populations of persons with chronic conditions (Dicianno, et. al., *in press;* Dicianno & Wilson, 2010; Ouyang, et. al., 2007). This is most likely due to all of the reasons that have been described previously including: complexity of medical needs and self-

care regimes, cognitive deficits, poor self-management skills, lack of natural support, and a fragmented health care system (Exner, et. al.1993).

Dicianno and Wilson (2010) found secondary conditions are of significant concern to the health and quality of life for persons with SB (and many other disabilities / chronic conditions), leading to increased morbidity and mortality. Often these secondary conditions are preventable. Investing in proactive interventions to improve self-management skills and coordination of care can result in improved clinical outcomes as well as savings of healthcare dollars (Dicianno et al., *in press*). The following quote provided by a parent of an adult with SB provides a clear and compelling description of this problem and need:

> ***"The need for comprehensive, coordinated medical and social supervision for adults with spina bifida has never been greater. In order for this growing group of adults to experience quality of life and to contribute to society around them, they require assistance for the basic life-tasks most of us take for granted. A Wellness Program would contribute greatly to the enhancement of life of our adult population with spina bifida."*** ***- Howard Burrell (2006)***

Figure 2 provides an overview of the scope of the problem and proposed solution which will be further described in Chapter 2.

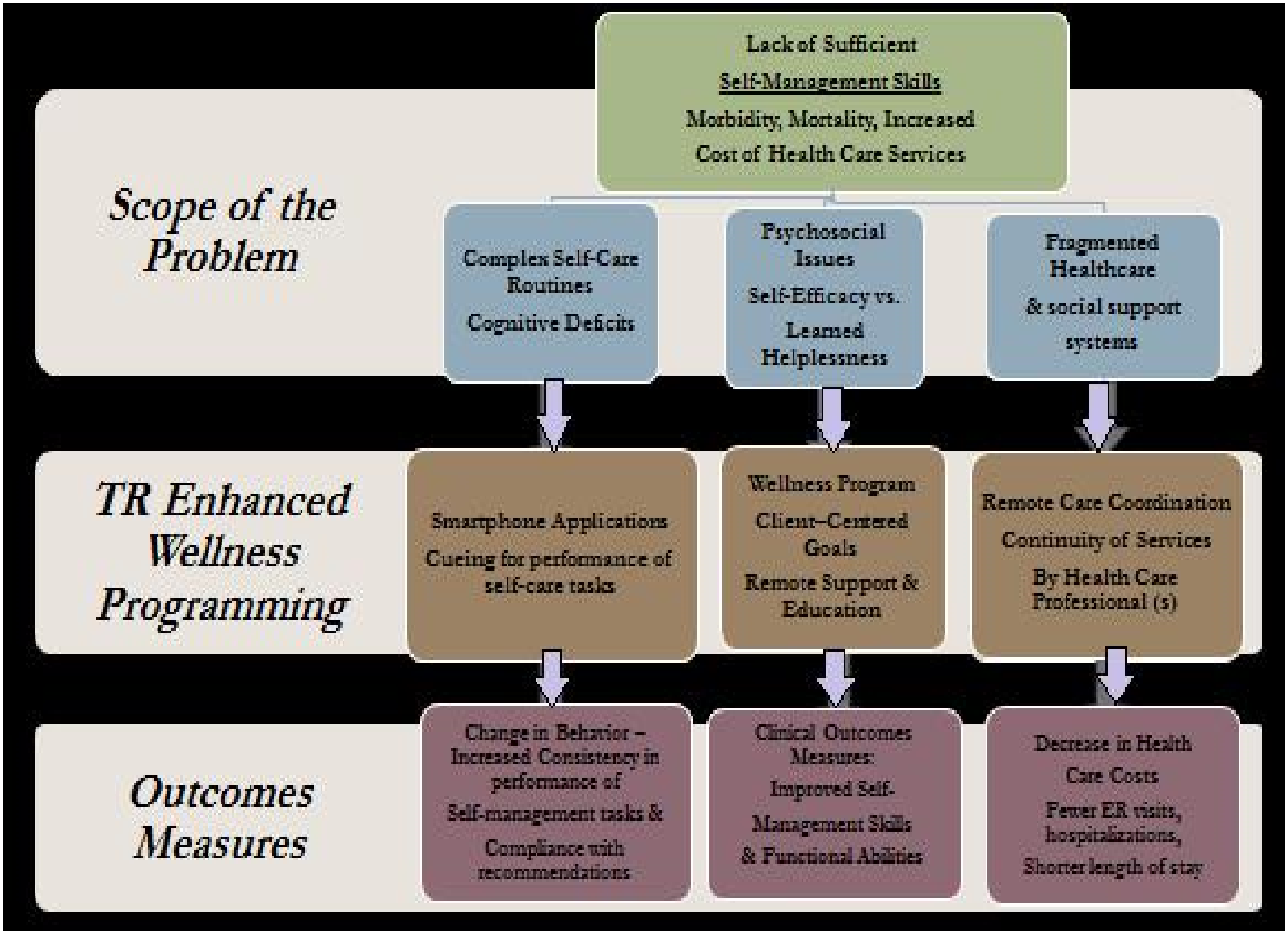


Figure 2. TR Model for Increasing Self-Management Skills for SB

2.0 DEVELOPING WELLNESS PROGRAMMING VIA AN INNOVATIVE MHEALTH SYSTEM FOR PERSONS WITH SPINA BIFIDA

Chapter two reviews the background related to the development of a wellness program and service provision of this programming for persons with SB. This chapter also discusses opportunity to further enhance and increase accessibility of these services by utilizing an innovative mHealth system, called iMHere (Interactive Mobile Health and Rehabilitation) to deliver the service. The iMHere system has two primary components which interact with one another to create a two-way communication system and share a variety of information between persons with SB and clinicians. One primary component is a suite of five smartphone applications: 1. MyMeds, 2. TeleCath, BMQ's, SkinCare and MoodTracker. The person with SB utilizes smartphone applications which allow them to communicate information about the performance of self-care routines often associated with potential problems these individuals may encounter leading to the development of secondary conditions. The other important component of the iMHere system is the clinician portal. The portal organizes all of the incoming data from the smartphones of the users with SB and displays this information in an easy to review dashboard. The dashboard allows the clinician to quickly visually scan the incoming information to quickly identify needs of specific individual among of a caseload of patients. This helps the clinician to prioritize the needs of persons on his caseload and assist those who require immediate attention. The clinician also uses the secure iMHere portal to send information back

to the person with SB using a secure messaging feature or can modify cues for self-care routines by changing the data in the apps remotely. In tandem, these two components (iMHere apps and clinician portal) provide a two-way system that can be tailored to each individual's needs and save the clinician time by automatically triaging the incoming data through the portal.

It is important to note the use of the iMHere system is not intended to replace traditional, in-person medical care. Nor is it to be used as an emergency response system. Instead, users are prompted to perform routine self-care tasks in a timely manner through the automated system. By performing these tasks as scheduled, the user is proactively reducing the possibility of developing secondary conditions. However, since it is not feasible to prevent every problem an individual may encounter, the system also provides non-emergency support and monitoring to help educate individuals regarding self-management of chronic conditions and accessing health care services as needed.

2.1 SUCCESS OF THE IN-PERSON SBAWP WELLNESS PROGRAM

With an appropriate level of support, persons with SB are fully capable of living healthy, productive lives while residing in the community; but, chronic health conditions often lead to disability and death in this population (Exner, Burgdöffer, Bohatyrewicz, Bomnüter & Wenck, 1993). The innovative mHealth system, described in detail later in this dissertation, was developed as an extension of a program that was first successful as an in-person pilot project to promote wellness in young adults with SB. In 2006, a three-year long pilot wellness program was developed by the Spina Bifida Association of Western Pennsylvania (SBAWP) funded by a $225K grant from the Highmark Foundation. The program employed registered nurses as

Wellness Coordinators (WCs) to proactively support young adults with SB in maintaining self-care routines, adopting healthy lifestyles and assisting with coordination of care. The program produced remarkably improved outcomes including fewer instances of UTIs and pressure ulcers and decreased incidence and duration of hospitalizations as compared with national data [See Table 1] (Ouyang, et. al, 2007; Dicianno, et al., *in press*):

Table 1. Outcomes of SBAWP Wellness Program

Outcomes of Wellness Program	SBAWP Survey		UPMC Adult SB Clinic population		Wellness Pilot Program	
# of subjects	142	%	31	%	31	%
skin breakdown episodes	41	28.9	11	35.5	3	9.7
# urinary tract infections	50	35.2			5	16.1
# hospitalizations	29	20.4	7	22.6	4	12.9
mean # days per hospital admission			11.4		5.7	

This program was staffed by two part-time registered nurses, who served as WCs. These WCs provided the participants of the program with coaching, education, support, direct care and monitoring services within the home and community settings. The direct care provided by these WCs was intended to be minimal, with other caregivers involved, so any tasks the person could not perform independently would be completed by routine personnel or family members. The original focus of this program was aimed toward developing self-awareness and self-

management skills for the young adults to be as independent as possible in managing their own care. Dicianno et al. *(in press*) describes the expanded in-person version of this program which is ongoing and now funded by the University of Pittsburgh Medical Center (UPMC) Health Plan for those patients covered by this managed care insurance program. The UPMC Center for Wellness is now delivering services for persons with SB and acquired spinal cord injury as a patient-centric Specialty Medical Home (SMH) model of care. The Institute for Healthcare Improvement's goals for healthcare reform coincide with this preventative approach to providing health care services (Beasley, 2009).

While the SBAWP grant-funded Wellness program was very successful in reducing secondary conditions and decreasing healthcare costs, this program also had significant limitations in the number of persons which could be served over a small geographic region due to limited funding. Likewise, with an existing shortage of nursing staff in the United States (AACN, 2012), it did not seem feasible to consider expansion of the program to more persons in a wider geographic area by hiring more nursing staff to serve as WCs. Furthermore, persons with SB residing in outlying rural areas often have even fewer resources and could benefit even further from the program.

2.2 TELEREHABILITATION TO EXPAND AND IMPROVE WELLNESS PROGRAMMING

Telerehabilitation (TR) or "telehealth" is one means to efficiently and cost-effectively provide support to persons in rural areas. A remote system can employ the wireless technology of a smartphone and specially designed applications ("apps") for the most common needs of the SB

population in order to help support and maintain self-management skills. Increased support through a TR delivery method could potentially also decrease the burden of seeking assistance and healthcare services upon the consumer (person with MMC) and improve overall quality of life (McCue, Fairman & Pramuka, 2010). The smartphone was selected as the best device of delivery of remote support for multiple reasons. Smartphones are quickly growing in popularity in the United States due to their versatility. According to Neilson (2012), 56% of mobile phone users are now smartphone owners, and this growth is expected to continue. Software and support can be easily transported in the user's hand, whether he is at home, work, or at a community ball game. Portability is very important to achieve success in the clinical application of a system where self-care activities need to occur regardless of the location of the user. When emerging mobile communication and network technologies are integrated for use within healthcare systems, this is known as mobile health or mHealth (Istepanian, et al 2006). The American Telemedicine Association (2012) advises that mHealth is to be thought of as a tool through which telehealth or telemedicine can be practiced.

2.3 ASSISTIVE TECHNOLOGY AS COGNITIVE REHABILITATION, MONITORING AND SUPPORT

In addition to the potential positive outcomes of having persons follow through with necessary tasks to maintain their health, the potential exists to demonstrate a decrease in costs related to care. Freedman and colleagues (2006) were successful in demonstrating assistive technologies contribute to a decrease in dependence. In fact, use of assistive technology accounts for a 50% decrease in the number of people dependent on personal assistance in the elderly population

(Freedman et al., 2006). Likewise, Hoenig, Taylor and Sloans (2003) have found that that technological assistance may clearly substitute for at least a portion of the personal assistance required in coping with disability. Hoenig, et al (2003) describes: "People, who do not use equipment [assistive technology (AT)], report about four more hours of help per week compared with those who do use equipment" (p. 335). The potential for AT to reduce the need for personal assistance is apparent whether it is used some of all of the time (Hoenig, et. al., 2003).

While no clinical studies have been published that use smartphones or other hand-held electronic devices as cognitive orthoses for persons with SB, several studies have been published in which these devices are utilized to support persons with brain injuries for performance of everyday tasks. Like persons with SB, memory problems are one of the most frequently identified barriers to functional independence following a brain injury (Barnes, 1999). Cognitive deficits in areas such attention, executive function, working and prospective memory interfere with everyday function (Gentry, Wallace, Kvarford, & Bodisch-Lynch, 2008). There are two primary means to intervene in order to improve cognitive abilities in rehabilitation. Some interventions seek to remediate or change the person's skills and abilities. Other approaches seek to compensate for cognitive deficits by modifying the environment, task and or providing some form of assistive technology to improve the individual's function. Assistive technology comes in various forms including low technology (planners, calendars, sticky notes) and high technology devices (personal digital assistants, digital voice recorders, and paging systems). Over the past few decades, computers have become increasingly more powerful and smaller in size (Giles & Shore, 1989). A computer that once dominated an entire room can now function in a size small enough to fit in the palm of one's hand. In conjunction, these devices are becoming less expensive and more available to the general public as organizational tools for keeping track

of appointments, phone numbers, to-do lists and other important information previously memorized or maintained in a planner or calendar. Likewise, due to increased availability and use, persons are becoming increasingly tech-savvy and more open to using electronic devices in their everyday lives (Wade & Troy, 2001). Similar to persons with SB, persons with brain injuries often require some means to compensate for their cognitive deficits in order to perform everyday tasks (Barnes, 1999). As the use of hand-held electronic cognitive orthoses increases in rehabilitation settings, the programs and range of the technology are concurrently developing and improving (Scherer & Cushman, 2002). DePompei et al (2008) found that not only did students with TBI find using PDAs or smartphones improved their functional performance, but enjoyed the social independence from other persons it provided as well. "There was less need for 'nagging' to complete daily activities" …and… "They like a device reminding them rather than an individual telling them what to do all day long" (p. 493).

Gentry and colleagues (2008) conducted a study which examined the efficacy of personal digital assistants (PDAs) as cognitive aids in a sample of individuals with severe traumatic brain injury (TBI).. Two hypotheses were generated: "Eight weeks after the conclusion of training, participants will demonstrate: (1) significantly improved occupational performance of everyday life tasks and satisfaction with their performance, as measured on the Canadian Occupational Performance Measure (COPM) and (2) significantly improved participation in everyday life tasks as measured on the Craig Handicap Assessment and Rating Technique-Revised (CHART-R)" (Gentry et al., 2008, p. 20). This study uses a quasi-experimental (not randomized) pre – and- post assessment design with 23 participants. The intervention included three components: 1) Provision of a PDA, 2) Three to six 90-minute training visits provided by an occupational therapist (OT) in the home for no longer than a 30-day period. This training included one-on-one

verbal training, demonstration & instructional literature to meet individual learning styles and needs, and 3) use of the PDA for an eight week period. It is important to note, it is not just provision of the PDA's, but also the training in the use of PDA's that is the true intervention.

Outcomes of this study were measured using the COPM & CHART-R, self-assessment rating scales of occupational performance completed by the study participants and their family member or caregiver. The authors report statistically significant outcomes as mean change on the COPM & CHART-R scores as listed in [Table 2]:

Table 2. Outcomes of PDAs for person with TBI

Outcomes Measures	Mean – Pre	Mean -Post	Statistics
COPM			
Performance	2.86	7.28	t=11.36, p<0.001
Satisfaction	1.59	6.73	t=9.88, p<0.001
CHART-R			
Overall Improvement	74.7	85	F = 15.9, p<.0.001
Physical Independence	---	---	(data not reported)
Cognitive Independence	55.5	69.4	t = 5.85, p < 0.001
Mobility	87	95.7	t = 2.92, p < 0.001
Occupation	63.1	78.3	t = 3.18, p = 0.004
Social Integration	---	---	(data not reported)
Economic Self-Sufficiency	---	---	(data not reported)

Gentry's (2008) study demonstrates self-perceived improvement of functional independence in performing everyday life tasks as described by participants and their caregivers with the provision, training, and use of a PDA for an 8 week period by persons with severe TBI in a community-based setting.

PDAs and smartphones have many of the same features. However, the smartphone has the potential to serve as a cognitive orthoses in a more powerful way. Due to the multitude of

features, most individuals are unlikely to leave their smartphone behind; whereas, PDAs have been forgotten by individuals with cognitive disabilities. The device was not available in the necessary context to provide their intended support (Gentry, 2008). In addition, the development of specialized applications to assist in the management of self-care tasks will only serve to enhance the intervention. The Ubiquitous Smartphone and Developing "Apps"

Persons with SB often present with a variety of functional impairments including cognitive issues, fine motor and visual perceptual deficits as was introduced in Chapter 1. It is necessary to consider how these impairments impact the individuals' abilities to utilize smartphone applications. In exchange for the portability of this device, a trade-off occurs in that the small size of the screen limits visual display. This limitation creates a challenge to include targets that are large enough on the touch screen to accommodate the fine motor and visual perceptual difficulties which are common. Creating larger targets and text further limits the amount of information that can displayed on the small screen at any given instance. The need to navigate from screen-to-screen increases the number steps a person must perform and complexity of use from a cognitive perspective. All of these variables needed to be considered carefully in determining the functionality, design, visual layout and content available within each of the apps planned for development.

2.3.1 Developing Apps to Support Self-Care Routines

As previously discussed, self-management of SB varies according to the needs of the individual. The tasks and activities that many persons with SB need to perform on a consistent basis to maintain health and wellness include: medication management, bowel and bladder routines, skin care, exercise, nutrition and fluid intake, health care visits and mental health maintenance. Lack

of adherence to self-care routines and healthcare recommendations can quickly lead to a decline in health status and development of secondary conditions such as pressure ulcers and UTIs. To manage such complex self-care routines and change one's lifestyle is hard to do. In order to help this situation, there are various self-help tools that can be utilized, including AT.

Among AT devices, mobile phones can serve as an ideal platform to deliver self-management applications for the delivery of interventions to assist with the management of SB and prevention of secondary complications due to their ubiquitous characteristics and pervasiveness. Mobile cellular technology has been the most rapidly adopted technology with over 4.6 billion subscribers utilizing basic mobile services (i.e. mobile telephony and SMS) (ITU, 2009). Likewise, research conducted within our own geographic region specific to adults with SB shows that over 68% utilize cell phones (Dicianno, et al., 2009). Smartphones facilitate the potential adoption of new mobile applications. In addition, the rapidly evolving technologies used for mobile phones, namely smartphone applications "apps" increase the potential of mobile phones by allowing for the unique development and customization.

The home screen of the iMHere suite of apps is shown in Figure 3. Each individual app is accessed by an icon to represent the purpose of the corresponding self-care activity. The MyMeds app is highlighted in Figure 3. All of the apps include a scheduling component to program reminders to initiate performance of self-care activities. Five apps were prioritized based on availability of resources for development and the factors which most greatly impact

health outcomes in the SB population. The following section describes each app individually.

Figure 3. iMHere Home Screen

2.3.1.1 MyMeds App for Medication Management

The MyMeds app (Figure 4) provides cues to remind the user to take his medication at recommended times, tracks the number of doses and reminds the user to contact the pharmacy for refills. Generic and brand names are integrated into the app to make entry of one's medication regime easy. The user then also has a portable list of medications to share with clinicians which can be easily revised as prescriptions change. Having an accurate list of one's medications is a valuable resource for individuals who receive treatment from a variety of specialists who may not collaborate with one another. The list also enables the persons to describe the purpose of taking the medication. This information is valuable in helping persons to develop increased understanding of one's condition and how medications are used in the management of conditions. Figure 4 depicts what the user sees on the home screen of the smartphone when the MyMeds alarm is activated to alert the user to medication

Figure 4. MyMeds App Reminder

Additional apps were identified for future development and testing, but limited time and other resources prevented these apps from being included in this study. These include apps to manage appointments, exercise programs, nutrition and fluid intake, and ordering and maintaining self-care and health related supplies.

2.3.1.2 TeleCath App for Self-Catheterization

TeleCath allows the user to program the app to receive reminders to catheterize the bladder. Once the reminder is provided, the user can then easily indicate whether any problems were encountered when catheterizing (no or little output, bleeding, color differences, strong odor etc.). The information is automatically updated in real time to quickly alert the clinician of the difficulty the person is encountering when performing this self-care task as depicted in Figure 5. This app can also be set to ask users once per day about frequency of incontinence (Figure 6). Incontinence is a sign of an ineffective bladder management program. Tracking this information on a daily basis may help revise the plan of care including frequency of catheterization, fluid intake or prescription of medications frequently utilized in the urologic care of persons with SB.

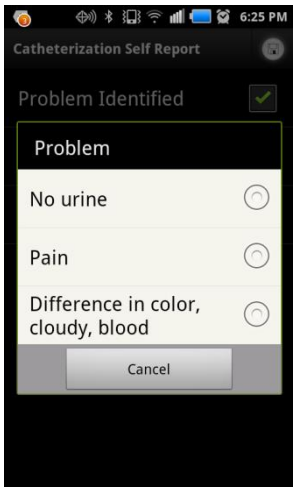


Figure 5. TeleCath App Screen to Report Problems

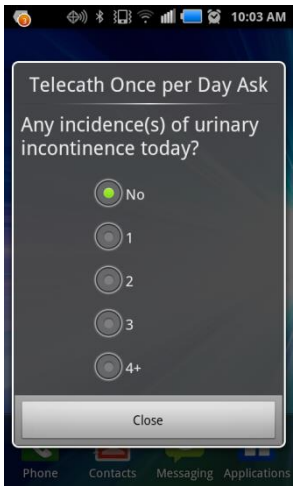


Figure 6. TeleCath App Screen to Report Frequency of Incontinence

2.3.1.3 BMQ App for Bowel Management

Much like TeleCath, the bowel movement cue, or **BMQ**, app provides reminders. However, these reminders can be set less frequently, such as every other day, or every third day, to reflect common bowel program schedules using enemas or medications. Figure 7 shows the screen utilized in setting alarms within the app, which can then serve as reminder to perform one's bowel management program. The save button is highlighted in this screenshot and is a common feature consistent within the design of all of the apps. This design consistency increases ease of use. Also, like TeleCath, BMQ allows the user to easily and quickly alert clinicians if problems are encountered while performing one's bowel management program.

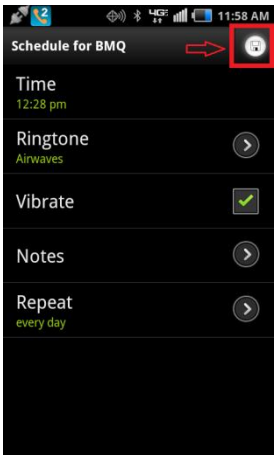


Figure 7. BMQ App

2.3.1.4 Mood Tracker App to Detect and Monitor Depression

The **Mood Tracker** app (shown in Figure 8) asks a series of 10 yes/no questions to help detect the user's level of depression. Questions included in this app are based on those developed as a depression screening by Mental Health America, formerly known as the National Mental Health Association (MHA, 2008). Mood Tracker can be set as frequently as daily, but is recommended for weekly or bi-weekly use similar to MHA's Depression Screening Quiz. Clinicians are then able to remotely monitor mood over time to determine changes and intervene if a user shows an increase in depressive symptoms as indicated through their responses to the questions. If a user indicates a high level of depressive symptoms 5/10 or greater (answers yes to five or more questions) or indicated presence of suicidal thoughts, the app is programmed to provide the user with the option to call 9-1-1 emergency response number or the mental health crisis as designated for his or her geographic region. Both phone numbers are programmed into the Mood Tracker app to provide this information automatically to the user upon completion of the questions.

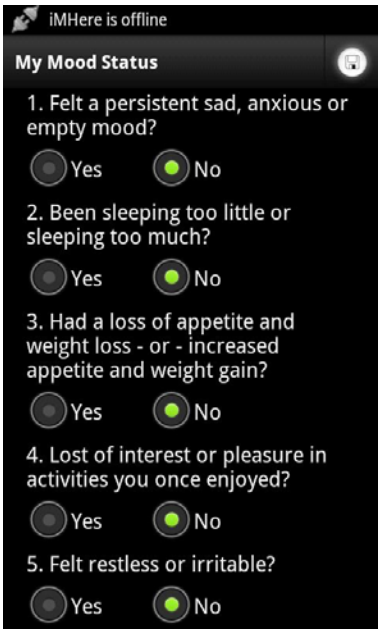


Figure 8. Mood Tracker App

2.3.1.5 SkinCare App for Prevention and Management

The SkinCare app is the most complex of the iMHere apps developed to date (Figure 9).

The SkinCare app has several screens and features: 1) Reminders can be set to cue users to perform daily skin checks as a preventative measure. 2) If skin breakdown or injury occurs, users can report it to the monitoring clinician using a combination of text feedback and also send a picture of the wound. 3) The wound is tracked over time to monitor progress in healing and/or provide instructions for how to best care for the wound. Also, see example of wound tracking in Figure 11, which depicts how information from the SkinCare app is displayed within the clinician portal for identification, monitoring and follow-up intervention.

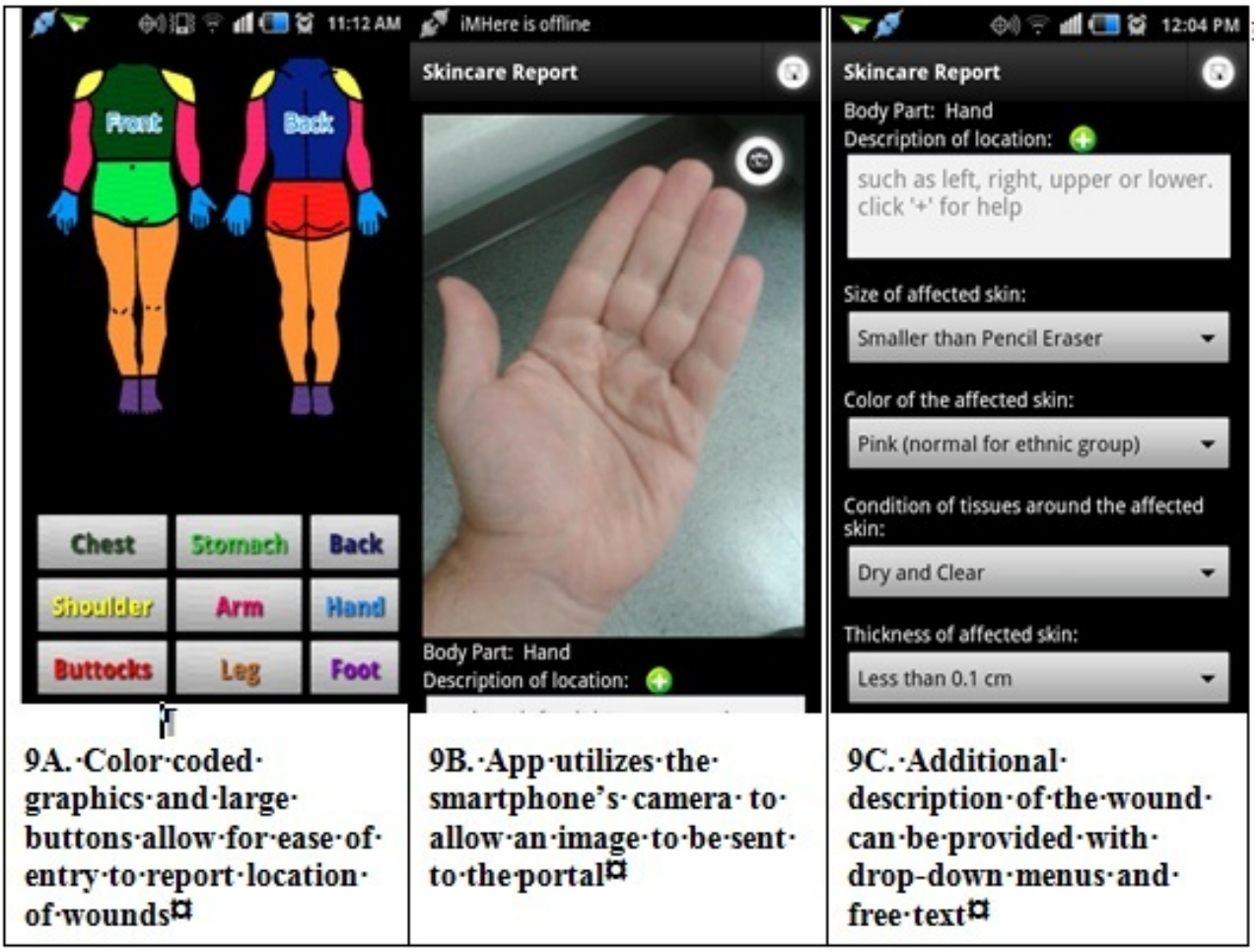


Figure 9. SkinCare App

Additional apps were identified for future development and testing, but limited time and other resources prevented these apps from being included in this study. These include apps to manage appointments, exercise programs, nutrition and fluid intake, and ordering and maintaining self-care and health related supplies. Persons are able to customize use of the system by using only the apps that are relevant to their individual needs. For instance, not all persons with SB complete a bowel program; therefore the BMQ app may not be utilized. Depression is a common problem for many persons with SB, but not every person will deem it

necessary to monitor mood if he or she has not encountered mental health problems. A person can use only one, a few or all of the apps for a customized remote wellness program.

2.4 THE CLINICIAN PORTAL: FEATURES & FUNCTIONS

The clinician portal is a web-based tool to monitor the activity of a caseload of patients currently using the iMHere app. The apps send monitoring data to, and receive self-care plans from, the web-based clinician portal. This portal includes a graphical dashboard for clinicians to monitor aggregate real-time data from all patients, and to identify patients in need of assistance. The clinician uses the portal to tailor treatment plans for each patient and can push the plan to the smartphone apps. It can be used to schedule drug regimens and maintenance plans, and to view any schedules the patient sets for themselves. Any problems the patient reports are communicated to the portal, and are flagged for clinicians' attention on the portal Dashboard. Patients may also create or modify self-care plans from their smartphone themselves, and these changes will be synchronized with the portal for the clinician's review. The iMHere Clinician Dashboard (Figure 10) provides a graphical overview of incoming information from the patients' apps. Clinicians can quickly scan the dashboard and determine how to prioritize contact with patients based on this information. The portal dashboard has been designed to allow the clinician to quickly review numerous patients at once, and to provide quick access to the areas of the portal that need attention. By clicking on attention flags on the dashboard, iMHere will direct the monitoring clinician or WC to the area that needs attention.

Figure 10. The iMHere Clinician Dashboard

The portal maintains a persistent connection with all registered patients' devices currently running iMHere. The patients whose devices are powered on and currently connected will be indicated on the Patient Roster with a green checkmark. The Patient Roster can be used to bring up the Client Details Box which has the advantage of allowing you to navigate one patient at a time. The clinician portal provides a dashboard using symbols to make it easy to visually review a caseload of patients' incoming information. The symbols help the clinician to quickly differentiate which patients do and do not require attention as well as the degree of urgency associated with their needs (See Figures 11, 12 & 13).

Figure 11. Does Not Require Attention Symbol

Figure 12. May Need Attention Symbol

Figure 13. Needs Immediate Attention Symbol

Figure 14. iMHere Clinician Potral View of SkinCare App Entry

3.0 USABILITY TESTING (PHASE I)

The purpose of this usability study is to evaluate potential strengths and weaknesses of the system, and identify additional features which may improve the usability of the smartphone applications and the overall iMHere mHealth system. The Phase I Usability testing component of this project has focused specifically on users of the applications (patients) diagnosed with SB. Apps that can be customized have been created for this population and individuals' needs specifically to serve as self-management tools for persons with SB.

3.1 RATIONALE FOR USABILITY TESTING

In order to be an effective intervention, participants will need to utilize the apps and adopt the use of this technology as part of their daily routine. Usability is a key factor in adoption of technology as is originally described in Davis' Technology Acceptance Model (TAM). The TAM describes the adoption and prolonged use of an information system and can be explained by two beliefs: p*erceived usefulness and perceived ease of use* (Davis, 1989, Davis et.al., 1989). Perceived ease of use is defined as "the degree to which a person believes that using a particular system would enhance his or her…performance" (Davis, 1989, p. 320). Davis (1989) defines perceived ease of use as "the degree to which a person believes that using a particular system would be free of effort" (p. 320). Development of usable applications is crucial for both clinical

and consumer entities to accept smartphone apps as a method of remote intervention and support. The purpose of this usability study is to evaluate potential weaknesses of the system, and identify additional features which may improve the usability of the smartphone applications and the overall system. At this time we will focus specifically on users of the applications (patients) diagnosed with SB. Usability testing of the overall system by clinicians will occur at a later date as part of the overall project.

Even with the rapid growth of smartphone technologies, there are certain limitations in physical specifications of mobile devices, such as limited size of display or physical user interaction. In the design process of self-help applications, it is necessary to make the following compatible by taking the situations of use into account; the necessary functions to support the self-management activities, operability, ease of understanding, giving effective feedback to keep on motivating the users, and unobtrusiveness in daily life. In order to achieve this, the project focuses on the following aspects considering accessibility with regard to physical dexterity, perception and cognition:

• Visual interfaces and application structure for enhancing ease of operation and effective and motivating interaction.

• Auditory and tactile interfaces including their operability itself and their effect in usability by use of them with visual interface.

• Automatic tailoring, context awareness and self-configuration of the user interface depending on user's cognitive level, functional abilities severity and preferences.

The methods will follow design principles of human-computer interaction, with an emphasis on involving real users in the design process.

3.2 RESEARCH METHODS: USABILITY TESTING

3.2.1 Recruitment Activities

Approval was obtained through the University of Pittsburgh's Institutional Review Board (IRB) prior to the initiation of any recruitment activities. Recruitment for the usability testing component of this study occurred primarily through the Adult Community Services Programs of SBAWP. Fliers were posted and two informational group sessions were delivered to persons at SBAWP's residential programs. Persons who indicated interest in participating in the usability component of the project received full explanation of the study in person at the informational sessions or later via telephone utilizing the script found in Appendix A. An appointment was then scheduled to meet with the potential participant at their place of residence. Utilizing a natural environment is especially important to understanding the cognitive demands of using the apps within a potentially distracting setting (Johnson, 1998). Meeting with the participants to complete the data collection at their residence also aided in the recruitment process. Access to transportation can be very difficult to arrange or expensive to obtain for persons who are wheelchair users and live outside of the bus lines. During the initial individual appointment the Co-Investigator met with each potential subject at his or her home and obtained informed consent after again reviewing all aspects of the study including explanation of the inclusion criteria for participation in the usability testing study. In order to participate, the person must:

- be an adult between the ages of 18-40 years
- have a primary diagnosis of spina bifida (type: Myelomeningocele) as diagnosed by a physiatrist
- have a history of shunted hydrocephalus confirmed by a physiatrist

- reside in the community (not in a residential facility which provides support; e.g. a group home, personal care home or nursing home)
- be able to use a smartphone device after receiving brief instructions to make phone calls, send text messages, access the internet and use apps to find information

Participants completed the Adapted MacArthur Competence Tool (Appendix B) to ensure that they are able to fully understand the study and are able to provide informed consent (Appelbaum & Grisso, 2001). Potential participants must score a minimum of eight out of ten to qualify for informed consent. Signatures were obtained on the consent form . The next step was to determine whether subjects met the inclusion and exclusion criteria and subsequent performance of the screening tasks. The last bullet point listed above refers to our screening procedure. Subjects were asked to utilize the Motorola Droid ™ smartphone device to: 1) place a telephone call, 2) compose and send a specific text message, 3) determine the weather for the following day using a weather app, 4) report movie show times using the internet. These tasks were intended to aide in determining whether or not participants will have the ability to access and successfully utilize the iMHere apps developed for the study. The rationale was that if subjects were able to perform all of these tasks successfully, they should also have sufficient fine motor skills, visual perceptual abilities, and cognitive function in order to be able to successfully utilize the specially designed smartphone apps of the iMHere system.

3.2.2 Usability Testing Set-Up, Setting & Equipment

Since the Smartphone apps were to be utilized in natural environments, not a laboratory setting, usability testing also occurred in natural settings in which the participants live, work, learn and engage in recreational activities. Appointments were scheduled to meet with the potential

participants in their homes. Co-Investigators met with participants at their place of residence. Utilizing a natural environment was especially important to understanding the cognitive demands of using the apps within potentially distracting settings. The in-person testing occurred at the participants' residence. Going to the participants to complete the data collection also aided in the recruitment process since access to transportation can be very difficult to arrange or expensive to obtain for persons who are wheelchair users and live outside of the bus lines.

During this initial visit, a background survey was completed with each of the usability study participants to gather information related to demographics (age, gender) functional abilities (motor skills and sensory functions) and technical experience. Also at this first visit, subjects were instructed in the use of basic smartphone features and given the smartphone device to become comfortable and acclimated to aspects that are new to them during the first week. The following week usability testing of the apps began. The duration of participation in the usability testing component of this study is approximately four weeks. Each week a new app was introduced and the participant to utilize it in managing the related daily life task(s) over the next week. Each of the apps was utilized by three participants for at least one week in duration. The following graphical representation (Figure 15) illustrates the step-wise process in which usability testing of the iMHere apps occurred with Phase I participants over a five week duration:

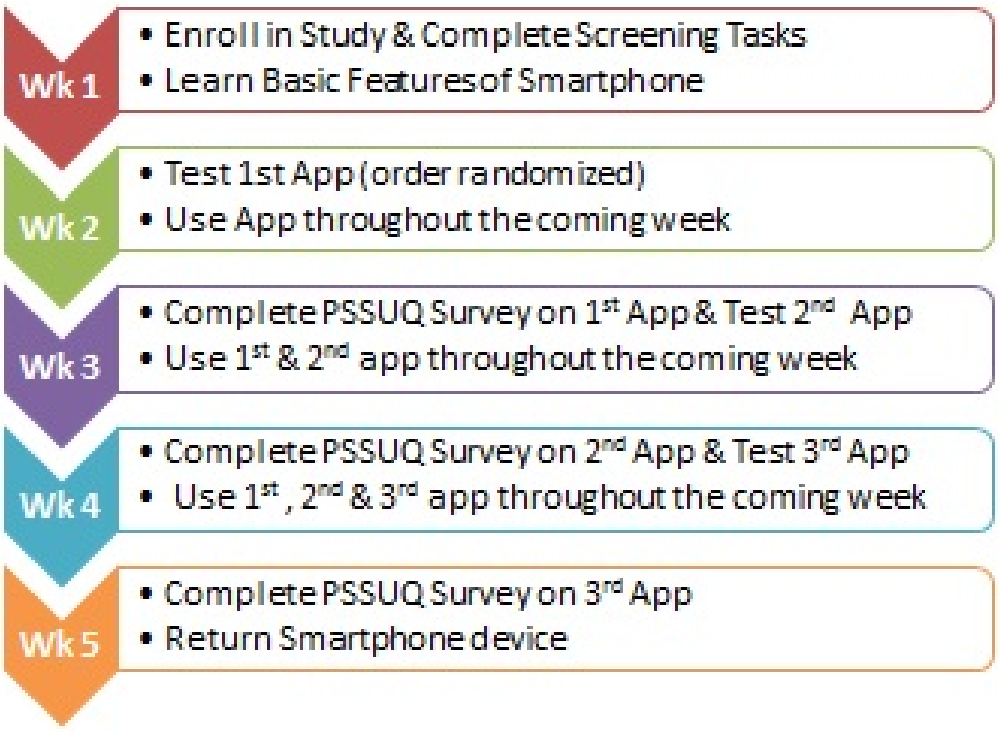


Figure 15.Activities & Timeline of Usability Testing

Laboratory evaluations do not simulate the context where the smartphone will be used (Johnson, 1998). A laboratory environment lacks the desired ecological validity a user would encounter in natural settings such as interruptions, movement, noise, etc. (Tamminen, Oulasvirta, Toiskallio, & Kankainen, 2004). Because the apps would be utilized in natural environments, not a laboratory setting, usability also occurred in natural settings in which the participants live, work, learn and engage in recreational activities. Utilizing a natural environment was especially important to understanding the cognitive demands of using the apps within potentially distracting settings. The in-person testing was completed in the participants' homes or other location of their selection. Going to the participants to complete the data collection also aided in the recruitment process since access to transportation can be very difficult to arrange or expensive to obtain for persons who are wheelchair users and live outside of the bus lines. During the initial meeting,

the participants were introduced to the first app and will perform relevant tasks to determine their ability to access and enter information using the smartphone app.

Data for two objective measures will be collected during this usability testing:

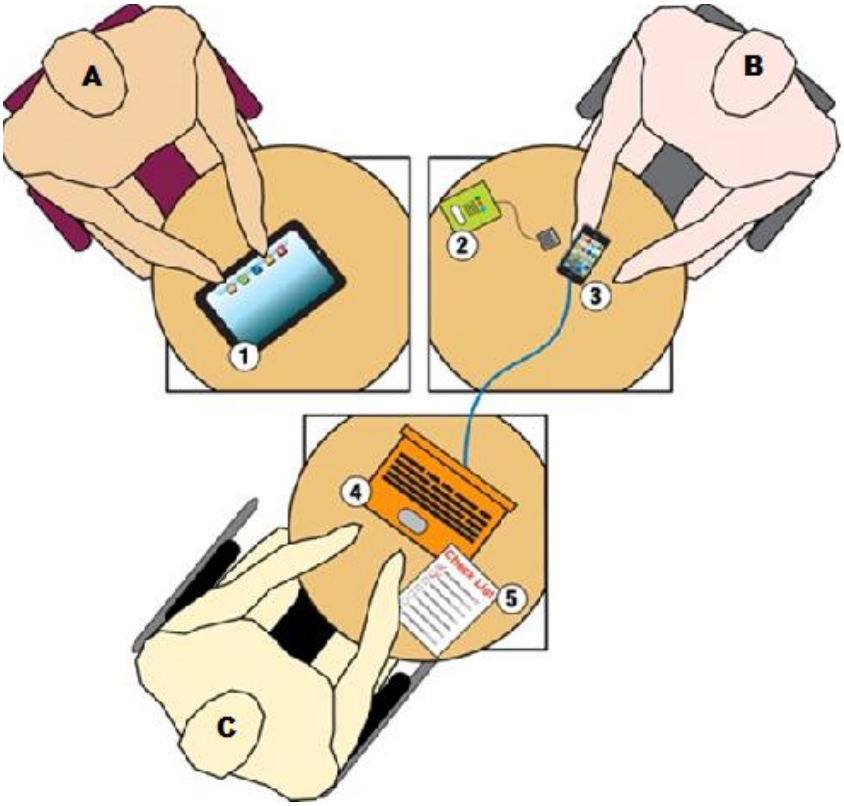
- **Completion of Tasks** – user is able to input or retrieve information from the apps accurately and verbalizing when they are "done"

- **Erroneous Actions** – Defined as an action that does not get the user closer to their goal of completing the task

At the time this usability study was initiated in June 2011, there were quite a few models of smartphones to choose from and there are many more available today. The selected device utilized in the usability testing phase of the study was the Motorola Droid due to its combined features for input through both the touch screen and pull-out keyboard (Figure 16).

Figure 16. Motorola Droid ™

Screencast for Android© is a free application available from the Google Play store which provides the facility to capture the android screen and created video of the screenshots as tasks are performed (Google, 2011). The Screencast software was created as a means to record and to provide video by recording the Android screen. In this usability study, it allowed for recording of the use of the apps while they were being initially tested. Display of the Android smartphone screen simultaneously on a laptop screen was also possible. This display enabled the research assistant to easily view the subject's smartphone screen while the testing was occurring. Figure 17 depicts the physical set-up of the persons involved and equipment utilized during usability testing of the apps.



Persons & Roles

A. Researcher - instructs subject in use of app, demonstrating step-wise actions as needed using tablet

B. Usability Testing Subject

C. Research Assistant - Monitors the subject's activities in using the app using Screencast software to view activities, listen to subject's verbal feedback and cues from Researcher to record errors made while using the app in real-time

Equipment

1. Electronic Tablet
2. Audio Tape Recorder
3. Smartphone
4. PC (laptop) running Screencast software
5. Log to record accuracy of actions completed while using app

Figure 17. Physical Set-Up of Usability Testing of Apps

Subjective self-report measures will be gathered using the *IBM Post-Study Usability Questionnaire* (PSSUQ) (Lewis, 1993) [See Appendix C]. The PSSUQ is primarily a close-ended questionnaire that has been found to be both a valid and reliable instrument for usability testing. The scale has been adapted to meet the needs of this usability study, but continues to use a seven-point Likert scale format with numbers correlating with higher levels of satisfaction. Three main categories of subjective are analyzed through the PSSUQ:

1) **System Usefulness** – belief that the system will improve the user's performance

2) **Interface Quality –** relates to the perception of the user interface layout

3) **Information Quality –** belief to which a system provides information that is helpful to complete the given tasks

Open-ended questions were also used to record users' opinions about the best and worst aspects of the system. (Scotch et al., 2007, p. 81-82). Aspects integral to understanding the likelihood that persons with SB will utilize these apps to support them in completing self-care tasks include: learnability, efficiency, ease of use, memorability and user satisfaction (Ryan & Gonsalves, 2005). [See Table 3]

Table 3. Outcomes Measures of Usability Testing (Phase I)

Usability Attributes Measured	Description	Data Collection Technique
OBJECTIVE MEASURES		
Performance *(Collect during Initial Training & 1 week post use)*	Time taken to complete task measured in seconds at the sub-task level of each app	Observed time for task completion in using the app. Note if time gradually decreases, stays the same or fluctuates.
Number of Errors *(Collect during Initial Training & 1 week post use)*	Errors committed by the user *	Observation of the number of errors performed by the individual as directly observed by the investigators
SUBJECTIVE MEASURES		
Learnability *(Initial Training & 1 week post use)*	How the user interacted with the application based on training received	Post experience questionnaires completed by the participants (these created from IBM PSSUQ) Ranked by the user on a Likert scale 0-7 Memorability will also be able to be detected by the difference in the amount of time from week to week and change in the number of errors
Efficiency *(1 week post use)*	How effective the application was in achieving the task	
Ease of Use *(1 week post use)*	How intuitive the application is to utilize	
Memorability *(1 week post use)*	How well after the user leaves a program and, when he or she returns to it, remembers how to do things in it	
User Satisfaction *(1 week post use)*	How pleasant the user's experience was in using the application	

(Adapted from: Ryan & Gonsalves (2005). *The Effect of Context and Application Type on Mobile Usability: An Empirical Study*. Australian Computer Society, Inc. 28th Australian Computer Science Conference.)

A think-aloud protocol will be used for this study. Virzi, Source and Herber (1993) found this approach as generally more sensitive to other methods, uncovering a broader array of problems in the user interface. A think aloud approach requires the participants to verbalize their thoughts as they interact with the system and attempt to complete the designed tasks. Using think aloud approach enables researchers to discover "what" and "why." The information provided determines "what" the participants are doing in relation to the system interaction and "why" they are interacting in this manner (Nielsen, 1993). Audio tape recordings captured this information, were transcribed and reviewed following the usability testing sessions. This data provided a

mechanism to determine if the user interface matches the natural way of thinking and acting as the process highlights features to be improved (Virzi, et al., 1993).

The time for task completion is an important variable to measure. In order for use of the apps to be integrated into daily life routines, it is important use is minimally intrusive. One way to ensure this to ensure usage requires only minimal time. This data provided an objective performance measure, which we would expect to decrease over time. The participant utilized the device and iMHere throughout the following week. At the end of this first week, the subject provided feedback (Likert scale and qualitative survey) on the experience of utilizing the app. In addition, during this second session (week two) another app was added. The participant performed tasks relevant to this second app for use throughout the week. At the end of this second week, the person will again provide feedback (Likert scale and qualitative survey) on the experience of utilizing these two apps in tandem with one another in completing the relevant self-care tasks. During the third session the final app for this round of testing will be reviewed. The participant will perform tasks relevant to this third app use the app throughout the week to complete the round of testing. During this final week of use, the participant will be utilizing three apps in tandem with one another. The fourth visit to the participant at the end of week four, when final was gathered and the smartphone was retrieved from the participant.

3.3 USABILITY TESTING RESULTS

A total of eight subjects participated in the usability testing. One subject dropped out of the study due to changes in her availability to participate after the first two meetings. All six apps were tested by a minimum of three subjects who met the inclusion criteria. During the initial

phase of usability testing, the apps were not yet linked to the iMHere portal. Additional testing was needed once apps have been revised and are linked to the portal. However, this additional tested did not result in much change to the user interface of the apps. Instead this later testing focused on difficulties encountered in getting the data from the apps to sync with the iMHere clinician portal. Two additional rounds usability testing were completed after the initial testing of six apps prior to initiation of the clinical phase of the study.

3.3.1 Usability Testing Subject Demographics Summary

A total of 8 subjects (n=8) participated in the first phase of usability testing. Prior to enrolling in the study, subjects passed a participant screening process to verify they were between of 18-40 years of age, did not have a diagnosis of mental retardation or serious mental illness. Subjects demonstrated sensory, cognitive and sensory ability to utilize an off-the-shelf, Motorola ® Droid, smartphone device. Each subject successfully completed a series of three tasks:

1) Utilize the smartphone to place a phone call to the researcher's phone and save contact information in device
2) Download "The Weather Channel" app, input settings for his/her region, read weather report aloud including current temperature and forecast for rain
3) Access the internet, use search function to find local movie times, read aloud latest show of the evening for testing date

None of the subjects reported or demonstrated sensory (vision, hearing) or motor deficits which would interfere with operation of the smartphone device. All of the subjects also had shunted hydrocephalus, with one subject reporting a recent shunt revision. The subject who

dropped out was also already an owner of the same smartphone device, the Motorola Droid ™, being utilized in the study. A summary of the demographic data collected on each of the usability testing subjects is included in Table 4.

Table 4: Usability Testing - Subject Demographics

SUBJECT DEMOGRAPHICS	SUBJECT 1	SUBJECT 2	SUBJECT 3	SUBJECT 4	SUBJECT 5	SUBJECT 6	SUBJECT 7	SUBJECT 8
Gender:	Male	Female	Female	Male	Male	Male	Female	Female
Age in years:	24	30	23	22	33	35	32	29
Race:	Caucasian	Hispanic	Asian	Caucasian	Caucasian	Caucasian	Caucasian	Caucasian
Marital Status	Single	Single	Single	Single	Single	Single	Single	Single
Level of Education	HS Diploma	HS Diploma + Some Trade/Tech	Currently enrolled in college	HS Diploma	Associates Degree	Some college	Some college	Some college
Special Education Services	yes	yes	no	yes	yes	no	yes	yes
Level of Lesion	Unknown	L1 -L2	L1 - S2	Unknown	Lumbar	L4 - L5	Sacral	Unknown
Hours of Personal Assistance / week	35	14	20	28	25	15.5	27.5	24
Employment Status / Place of Employment	Part-time / Seasonal	Unemployed	Part-time	Part-Time	Part-Time	Part-Time	Unemployed	Part-Time
Currently a student?	No	No	Part-time college	No	No	No	No	No
Vision Deficits	yes, for distance, but no problems up close (near-sighted)	wears glasses, vision sometimes blurry since shunt revision	no vision problems	Wears glasses, fully correct vision	no vision problems	Wears glasses, fully correct vision	Wears glasses, fully correct vision	Wears glasses, fully correct vision
Fine Motor Deficits	no	no - not known	yes, but WFL	no	no	no	no	no
Cell Phone User?	yes	yes	yes	yes	yes	yes	yes	yes
Smart Phone User?	no	no	yes - has own Droid	no	no	yes - a little	no	yes
Familiar with apps	no	no	yes	no	no	no	yes - iPod Touch	yes

Subject three dropped out of the study after only two meetings. The subject's schedule and availability changed following new employment opportunities, in addition to her college studies.

3.3.2 Results of iMHere Apps Usability Testing

Recruitment, screening and testing all occurred within the home environment of the subjects. These subjects are already participating in an in-person Wellness Program provided by SBAWP. The subjects' experience with the concept of participating in a preventative wellness program also enabled them to better understand the concept of using proactive measures to avoid and manage health problems.

Results of the adapted PSSUQ are summarized in Table 5 below. The PSSUQ has been adapted (see Appendix D) to meet the needs of this usability study, but continues to use a 7-point Likert scale format with numbers correlating with higher levels of satisfaction.

Table 5. Summary of Modified PSSUQ Usability Testing Results

Apps Tested	**Round 1**	**Round 2**
MyMeds	2.0-4.3	1.0 (all scored 1.0)
TeleCath	1.0-2.3	1.0 (all scored 1.0)
BMQ	1.0-2.7	1.0 (all scored 1.0)
Mood	2.3-3.0	1.0 -3.33
SkinCare	1.3-3.0	1.0 – 3.0
Nutrition	1.3-2.0	Not tested

Table 6. Results of MyMeds App Usability Testing

DATE SURVEY COMPLETED⇳ ⇳ SURVEY QUESTIONS	PSSUQ AVG	Subject 1 6/9/2011	Subject 3 6/16/2011	Subject 4 6/16/2011	Subjects' Narrative Comments (Direct Quotations) *(# corresponds to subject)*
1) Overall, I am satisfied with how easy it is to use this smartphone app.	4.333	2	6	5	3) I liked it, but the information won't stay in the phone 4) only worked part of the week
2) It was simple to use this app	2.333	2	1	4	3) It was great for reminders. Would be good for someone with a busy schedule - they could have the reminder at work to take care of themselves better 4) Only when it worked
3) I could quickly utilize this app as part of my daily routine.	2.333	1	1	5	3) Most definitely, love it.
4) I was able to easily enter information using this app.	3.333	3	1	6	3) Yes, I was
5) I felt comfortable using this app.	2	1	1	4	3) I would recommend it for anyone
6) It was easy to learn to use this app.	3	3	skipped	3	3) OH YEAH (: !!
7) Whenever I made a mistake using the app, I could recover easily and quickly.	3	2	skipped	4	3) Yes cuz I was able to ask ?'s to the person who taught me how to use the app
8) It was easy to revise or change information I entered into the app.	3	1	5	skipped	3) Yup, whenever the information would stay on the phone
9) The organization of information in the app was clear.	3.667	2	4	5	3) The only thing I didn't like was having to set it for 5:00, 5:01, 5:02, etc. That would make it confusing and you had to turn off the alarm each time. It kinda gets annoying" "It would be nice for it to just be set for one time."
10) I liked using the interface of the app.	3	1	4	4	3) I did but the information would not stay in so that was a little frustrating
11) This app has all the functions and capabilities I expect it to have.	3.667	1	7	3	3) half/half "The reason I say half/half is that it has the capability, but the information does not stay in there."
12) Overall, I am satisfied with this app.	2	1	skipped	3	3) Yes, I am as long as the information stays in the phone

Table 7. Results of TeleCath App Usability Testing

DATE SURVEY COMPLETED→		Subject 1	Subject 2	Subject 4	Subjects' Narrative Comments (Direct Quotations)
↓ SURVEY QUESTIONS	PSSUQ AVG	6/2/2011	6/9/2011	6/25/2011	*(# corresponds to subject)*
1) Overall, I am satisfied with how easy it is to use this smartphone app.	1.667	1	3	1	3) Apps and the phone are so easy to use after ur taught a couple times. "I learn best 1:1 and if you teach me 1-2 times, I can get it - especially when it comes to technology."
2) It was simple to use this app	1	1	1	1	
3) I could quickly utilize this app as part of my daily routine.	1.667	1	3	1	3) "Simple is good - you don't want to be playing with your phone at work or school."
4) I was able to easily enter information using this app.	1.667	1	3	1	1) "Sometimes I cathed earlier, sometimes later" The subject did not actually cath when he received the alert from the alarm.
5) I felt comfortable using this app.	1	1	1	1	
6) It was easy to learn to use this app.	2.333	1	5	1	
7) Whenever I made a mistake using the app, I could recover easily and quickly.	2.333	1	5	1	3) It was OK cuz the person who taught me had to reteach me how to fix certain thing on the certain App."
8) It was easy to revise or change information I entered into the app.	1.667	1	3	1	3) After you learn how to use it it's easy
9) The organization of information in the app was clear.	2	1	4	1	3) It was greatly worded
10) I liked using the interface of the app.	1	1	1	1	3) I thought it was great
11) This app has all the functions and capabilities I expect it to have.	1	1	1	1	3) Yes, the information was great
12) Overall, I am satisfied with this app.	1	1	1	1	3) Absolutely Yes

Table 8. Results of BMQ App Usability Testing

DATE SURVEY COMPLETED⇨	PSSUQ	Subject 1	Subject 3	Subject 4	Subjects' Narrative Comments (Direct Quotations)
⇩SURVEY QUESTIONS	AVG	6/9/2011	6/16/2011	6/16/2011	*(# corresponds to subject)*
1) Overall, I am satisfied with how easy it is to use this smartphone app.	1.667	1	3	1	1) All the alarms going off kinda drove me nuts, but overall I had no problems. 1) I did not use BMQs - needs to be worked on
2) It was simple to use this app	2	1	4	1	1) I turned off the phone when I was at work because of the alarms
3) I could quickly utilize this app as part of my daily routine.	2.667	1	6	1	3) "The screen is simple - all you have to do is push 1 or 2 buttons. Not too distracting, you don't have to fuss with it. Just put the information in you need and you're done."
4) I was able to easily enter information using this app.	1.667	1	3	1	
5) I felt comfortable using this app.	2.667	1	6	1	
6) It was easy to learn to use this app.	2	1	4	1	3) It was easy but I learn better if something is explained more than once
7) Whenever I made a mistake using the app, I could recover easily and quickly.	2	1	4	1	3) Yes, I could but with some verbal instructions
8) It was easy to revise or change information I entered into the app.	1	1	1	1	3) Yes, it actually was fun because I was able to see other apps
9) The organization of information in the app was clear.	1.333	1	2	1	3) Researcher Note: This subject's bowel program was performed at a different time of day every time. She also has "in-person" reminders because staff help her with her bowel program and they determine when it is performed.
10) I liked using the interface of the app.	1	1	1	1	
11) This app has all the functions and capabilities I expect it to have.	2.667	1	6	1	
12) Overall, I am satisfied with this app.	3	1	7	1	3) Cuz I don't use it often

Table 9. Results of SkinCare App Usability Testing

DATE SURVEY COMPLETED⇨		Subject 1	Subject 3	Subject 4	Subjects' Narrative Comments (Direct Quotations)
⇩ SURVEY QUESTIONS	PSSUQ AVG	9/20/2011	9/15/2011	9/13/2011	*(# corresponds to subject)*
1) Overall, I am satisfied with how easy it is to use this smartphone app.	2.667	6	1	1	5) "App was not easy to use - taking pictures was hard. Would be easier to just use the button on the top as with camera for phone"
2) It was simple to use this app	2.667	5	1	2	Researcher Note: Neither Subjects 5 or 6 recorded any information using this app. Subject 7 had to be re-trained after the first week.
3) I could quickly utilize this app as part of my daily routine.	3	6	1	2	6) Have a "save" and "retake" button for pictures
4) I was able to easily enter information using this app.	2.667	6	1	1	
5) I felt comfortable using this app.	2.667	5	1	2	
6) It was easy to learn to use this app.	3	6	1	2	
7) Whenever I made a mistake using the app, I could recover easily and quickly.	3	6	1	2	
8) It was easy to revise or change information I entered into the app.	2.667	5	1	2	
9) The organization of information in the app was clear.	2.333	4	1	2	
10) I liked using the interface of the app.	3.333	6	1	3	
11) This app has all the functions and capabilities I expect it to have.	3	5	1	3	
12) Overall, I am satisfied with this app.	3.333	6	1	3	

Table 10. Results of Mood App Usability Testing

DATE SURVEY COMPLETED⇨		Subject 5	Subject 6	Subject 7	Subjects' Narrative Comments (Direct Quotations)
⇩SURVEY QUESTIONS	PSSUQ AVG	9/6/2011	9/22/2011	9/13/2011	*(# corresponds to subject)*
1) Overall, I am satisfied with how easy it is to use this smartphone app.	3	4	1	4	
2) It was simple to use this app	2	3	1	2	
3) I could quickly utilize this app as part of my daily routine.	1.667	2	1	2	
4) I was able to easily enter information using this app.	2	2	3	1	
5) I felt comfortable using this app.	1.667	3	1	1	
6) It was easy to learn to use this app.	1.333	2	1	1	
7) Whenever I made a mistake using the app, I could recover easily and quickly.	2	4	1	1	
8) It was easy to revise or change information I entered into the app.	1.333	2	1	1	
9) The organization of information in the app was clear.	1.667	3	1	1	
10) I liked using the interface of the app.	2	3	1	2	
11) This app has all the functions and capabilities I expect it to have.	2.333	2	1	4	7) It would not let me set the alarm for every day easily
12) Overall, I am satisfied with this app.	2	2	1	3	7) Subject reports that once per day feature of this app keeps reverting back to monthly cue

Table 11. Results of Nutrition App Usability Testing

DATE SURVEY COMPLETED⇨		Subject 5	Subject 6	Subject 7	Subjects' Narrative Comments (Direct Quotations)
⇩ SURVEY QUESTIONS	PSSUQ AVG	9/13/2011	9/8/2011	9/20/2011	*(# corresponds to subject)*
1) Overall, I am satisfied with how easy it is to use this smartphone app.	1.333	2	1	1	** None of the subjects provided written narrative comment for this app*
2) It was simple to use this app	1.333	2	1	1	
3) I could quickly utilize this app as part of my daily routine.	2	4	1	1	
4) I was able to easily enter information using this app.	1.667	2	1	2	
5) I felt comfortable using this app.	1.667	3	1	1	
6) It was easy to learn to use this app.	1.333	2	1	1	
7) Whenever I made a mistake using the app, I could recover easily and quickly.	2	4	1	1	
8) It was easy to revise or change information I entered into the app.	1.333	2	1	1	
9) The organization of information in the app was clear.	1.333	2	1	1	
10) I liked using the interface of the app.	2	3	1	2	
11) This app has all the functions and capabilities I expect it to have.	1.667	2	1	2	
12) Overall, I am satisfied with this app.	1.667	2	1	2	

3.4 IMPROVEMENTS IN DESIGN OF APPLICATIONS "APPS"

A few overall recommendations were provided to development team for revisions across the apps:

- Check all apps closely for spelling & grammar errors, several of these already noted and corrected
- Increase width of scrollbar on right side of screen to make this more obvious and/or ensure that text is not hidden when initial screen is displayed. Otherwise persons do not realize additional information is available by scrolling down and will exit and/or save incomplete data.

3.4.1 MyMeds App Modifications

MyMeds initially had lowest scores of all six apps and was the first app developed as part of the iMHere system. Significant revisions were needed for the MyMeds app to be successfully utilized by persons with SB. The way in which medications are entered into the app currently resulted in a separate alarm function for every medication. A more convenient way for persons to utilize the app would be to enter all of the medications included in a person's regime. The MyMeds app could then be programmed to allow for the number of daily reminders to be received at specific times (i.e. Morning 8:00 AM, Afternoon 12:30 PM, Evening 6:30 PM, Bedtime 10:00 PM). In conjunction with each time interval the appropriate medication(s) can

then be added from that person's medication regime list. This is particularly important for persons who take many different medications throughout the day. This changes the structure of the app and prevents the need for a separate alarm for every single medication, while still allowing the recording and tracking of each medication prescribed and taken by the patient. Improvements include: When the person accesses the app after receiving the reminder and they respond to the alarm, they can see a list of medications to take. A check box is included next to each medication to indicate they have taken this dose. If a person does not or cannot take their medication a place to indicate the reason why can be included and this information is then immediately reported back to the WC. Other minor changes to the MyMeds app include:

- Frequency of medications - Add ability to take medication less frequently than on a daily basis. For instance: only on certain days of the week, once per month, etc.
- Ensure all possible methods of intake of medications (inhaler was missing as an option for one subject).

3.4.2 Modifications of TeleCath & BMQ Apps

No major changes were needed for the TeleCath app. This app is simplistic and easy for persons to utilize. For the BMQ app, changes were made with scheduling options was necessary. The previous design of this app only allowed for daily reminders. Persons who complete bowel program regimes typically do not perform this self-care task on a daily basis. Persons are more likely to perform this task every other or every third day. A person may also need to vary the time of day they perform their bowel management program if it interferes with other activities or

they are reliant on caregiver to assist them with performing this task in some way. Otherwise simplistic design of the BMQ app was well-received similarly to TeleCath.

3.4.3 SkinCare App Modifications

Revisions were necessary to simplify the SkinCare app. The Green "+" button was not readily noticed by the usability testing subjects. If not drawn to the attention of the participant, the individual would simply skip utilizing this feature completely. The photo of wound function also required several revisions. The on-screen taking picture button is difficult for persons to use. This is especially true if the area of the body the person is attempting to photograph is on a more difficult to reach area of their body. Instead of using an on-screen button to take the picture, persons could more easily utilize the physical control button on the device that is already built in to perform this function. None of the subjects successfully recorded information using this app the first week it was demonstrated to them. One subject successfully utilized this app after being re-trained the following week. One subject was scheduled to test this app the final week; therefore, he did not have a chance to re-test it. The final subject did not indicate any difficulty in using this app, but no data was stored in the device to show that he had successfully utilized it.

3.4.4 MoodTracker App

No major changes needed for the Mood app. One subject did note difficulty in receiving the reminder to report on a daily basis; however, the frequency of this app's use clinically will likely be once per week at most.

3.4.5 Modifications for Nutrition App

As it currently is designed, the Nutrition app does not provide intended clinical function for wellness promotion. The purpose was intended to remind persons to avoid or increase certain foods/fluids in their diet. For instance, too much cheese and other dairy products can cause problems with bowel function for many persons with SB. Persons who have wounds need to make certain that they are eating foods high in protein to promote healing of their skin. Person often drink too much caffeine and not enough water. The app functions adequately from a technical perspective, but it only allows report of what has been ingested. Persons are also confused by the start and end time. Instead, persons should be able to receive reminders to ingest or avoid certain foods and app should be able to track progress. Example – Person needs to drink 6-8 glasses of water per day. Reminders received and person inputs # of glasses, at end of day can see if goal was achieved.

3.5 LIMITATIONS OF USABILITY TESTING AND DISCUSSION

These methods of usability testing of the apps allowed for the provision of specific feedback on each individual smartphone app by at least three subjects for three apps each round of usability testing. This is very important due to the fact that each person's wellness program will be customized to meet their individual needs. The apps utilized by one person with SB will vary from another, while some may be the same. If all of the apps were tested together as one package, we would not know if the strengths of one app are making up for the weaknesses in another. Ideally, we would want to randomize the order for all of the apps tested. This is a

limitation of this usability testing design. Unfortunately, time constraints did not allow for all of the apps to be developed before randomizing the order of implementation of usability testing. However, randomizing three apps per group will help to distribute the efficiency effect rather than testing all of the apps simultaneously or implementing and testing the apps in the same order.

Due to limited resources, it was determined the Nutrition app would not be further developed at this time. The other five apps would be integrated with the iMHere portal in preparation for additional usability testing and use in the clinical phase of the project as described in Chapter 4.

4.0 CLINICAL STUDY INVESTIGATING DEVELOPMENT OF SELF-MANAGEMENT SKILLS VIA THE IMHERE MHEALTH SYSYEM

4.1 OVERVIEW & HYPOTHESES

The aim of this study is to determine if delivery of wellness services through the mHealth system, iMHere, will result in improve of self-management skills in persons with SB. The outcomes of the intervention group receiving mHealth wellness services to enhance traditional care were compared to a control group receiving traditional care alone. After six months of participating in the TR Enhanced Wellness Program for SB, participants in the intervention group will demonstrate:

H_O (1) a self-perceived improvement in functional performance and/or satisfaction ratings on individualized goals as indicated by the COPM

H_O (2) significantly improved self-management scores as measured on the AMIS-II (Total Score and Condition Management Subscale)

in comparison to a control group receiving standard care alone.

A randomized controlled trial (RCT) design was implemented to test these hypotheses. The following sections provide a detailed account of the methods, analysis and results obtained from this study in relationship to outcomes specific to self-management skills. As previously mentioned this dissertation is part of an overall study which will look at a variety of outcomes

measures related to clinical impact, cost efficiency, quality of care and patient satisfaction at the end the year-long clinical trial. Some of the additional assessments intended to capture these outcomes are utilized to briefly describe the subjects, but are not analyzed in detail at this early phase of the overall study. Self-management skills are a foundational component in developing and maintaining wellness (Lorig & Holman, 2003). Therefore, it has been anticipated that change will occur in developing improved self-management skills through our mHealth intervention in order to impact the other outcomes over the longer duration of the overall study.

The story board, depicted in Figure 18, provides a visual summary of the overall study including: the steps taken for recruitment, enrollment, randomization, collection of outcomes measures as described in the subsequent sections of this chapter.

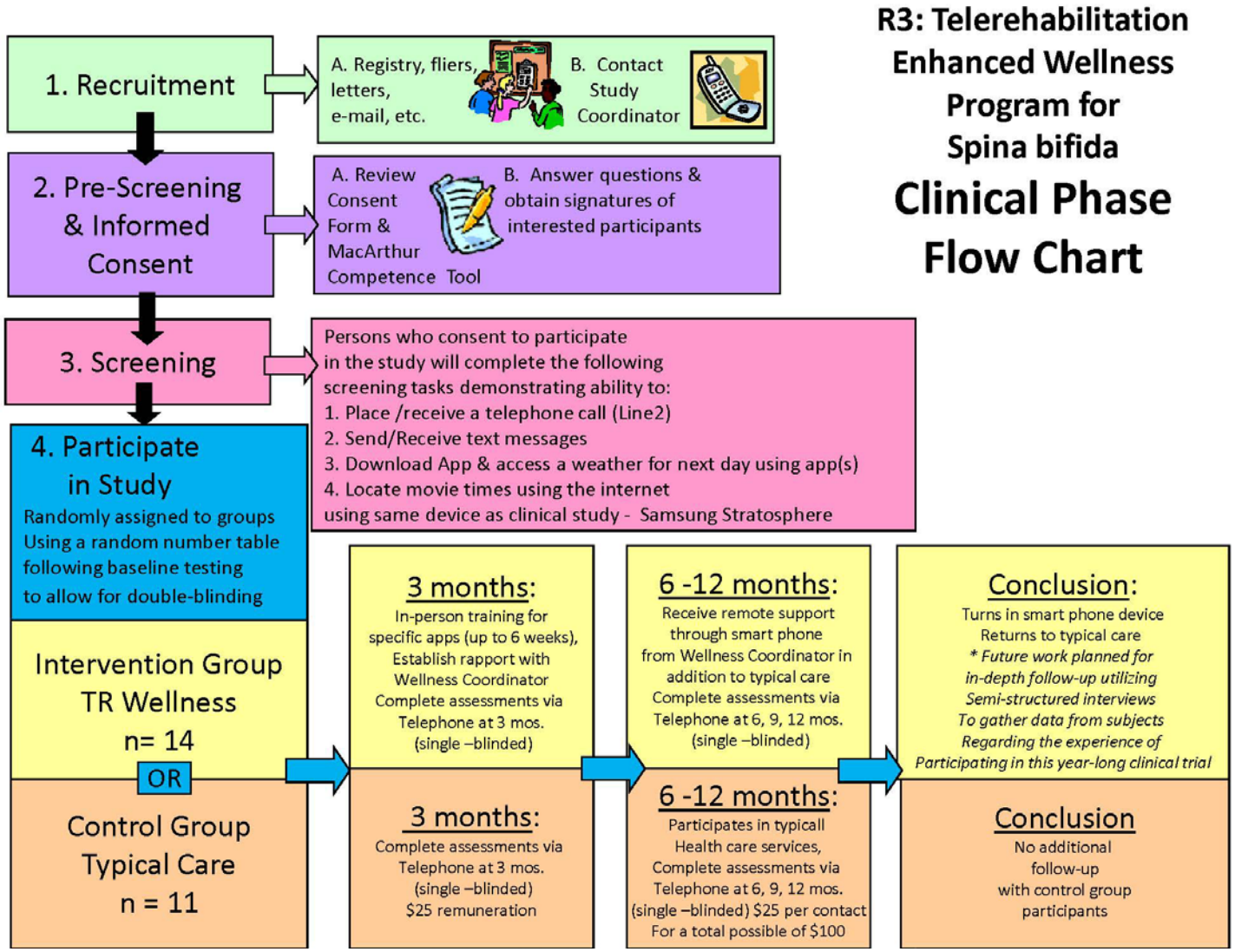


Figure 17. Story Board for Clinical Phase of Study

4.2 METHODS

4.2.1 Participant Recruitment & Enrollment

Approval was obtained from the University of Pittsburgh's IRB prior to the initiation of any recruitment activities Subjects were recruited primarily through the UPMC Adult SB Clinic. A research registry is maintained through the Department of Physical Medicine and Rehabilitation at the University of Pittsburgh providing contact information and supporting documentation of patients interested and willing to participate in research studies. Post cards and fliers were mailed and/or distributed to patients through this research registry or in-person at the clinic. A copy of the image used for these recruitment materials is included in Appendix B. All participants enrolled the study are under the care of the Principal Investigator, Brad Dicianno, MD, who is also the Director providing clinical oversight to the UPMC Adult SB Clinic.

4.2.1.1 Informed Consent Process

Voluntary, written informed consent for participation was obtained from all of the participants before the initiation of any research activities. Following the initial contact, either in-person or via telephone, individual appointments were scheduled to meet with these recruits at their home residence. Once the recruiter arrived at the potential participant's home in-depth information was provided regarding the nature of the study including its purpose and significance, as well as the data that will be collected. If the participant was still interested in participating, written, informed consent will then be obtained. The consent form was constructed using simple

sentences with basic vocabulary (Flesch-Kincaid Grade Level 7.7), pictures, and large print (Flesch, 1944). The recruiter also read the form aloud to all potential participants, pointing out primary aspects of the study, before obtaining their signature. In addition, the recruiter provided additional explanations and examples of potential scenarios that may be encountered while participating in the study and answered any questions about the study or participation prior to obtaining written consent. This dialog helped to ensure that potential participants who have limited literacy skills were able to fully understand the information being presented to them. The consent form is included here as example for future researchers who may wish to conduct similar studies. Considerable time and effort was placed on developing a consent form which provided the participants with a clear and understandable description of activities involved in participating in this research study. When the devices and costs of the service plan is provided by the institution, an addendum such as the one included here, removes potential liability for the institution in the event of improper usage by the participants. The addendum section of the consent form provides protection for the institution and consequences for removal from the study should the participant utilize the device for any unlawful purposes.

An addendum was added to the consent form to further protect the institution from potential liability issues (APPENDIX D). While subjects, had been informed verbally the system was not intended to replace traditional medical care, it was important they did not attempt to utilize the iMHere system to seek care or advice in the event of an emergency situation. An additional addendum (APPENDIX E) was signed by all participants assigned to the intervention group. It was reiterated throughout the course of the consent process and during the study, participation is completely voluntary and based on participants' interest in offering their

perspectives and opinions to guide clinical programming through the innovative iMHere mHealth system. Participants have the right to withdraw from the study at any time.

4.2.1.2 Screening Procedures and Equipment

Due to the nature of the activities involved a screening procedure was conducted following informed consent to participate in the study. The inclusion criteria were reviewed with the subject through a series of questions and tasks performed utilizing a the same model of smartphone device, the Samsung Stratosphere™ a Galaxy S phone (Figure 19) that will be utilized in the year-long study by the participant if he or she should be randomly assigned to the intervention group.

Figure 18. Samsung Stratosphere ™

Verizon Wireless© is the carrier Samsung Stratosphere ™. The Stratosphere smartphone was designed to maximize the speed of Verizon's 4G LTE network (LTE is a trademark of ETSI). This smartphone device has a large four-inch wide video graphics array (WVGA) super active-matrix organic light-emitting diode (AMOLED ™) touchscreen. This device was specifically selected for use in this study due to its fast connection and processing speed, in comparison to

the original Motorola Droid ™, used in Phase I (Usability Testing) of the study. In addition, when the clinical phase of this study began the Samsung Stratosphere was the only 4G LTE compatible devices provided through Verizon Wireless©, which still included the slide-out QWERTY keyboard. QWERTY is the most common modern keyboard layout and we anticipated subjects in the intervention group for this clinical phase of the project may rely heavily on the pull-out keyboard for any text input that is more than a few words since this was our observation during the usability testing phase of the project. The Samsung Stratosphere ™ runs on the Android 2.3 Gingerbread Operating System (OS). This open-source software is continually being upgraded. Several newer versions have since been released with the very latest version being Android 4.1 JELLY BEAN OS, just released March 21, 2013 (Android Inc., 2013). Additional information regarding the specifications of the Samsung Stratosphere ™ can be located on Samsung's © website (Samsung, 2013).

In Chapter 3, the Motorola Droid ™, was described as the smartphone device utilized in the usability testing phase of our project. While this newer model of smartphone device was selected for the clinical testing phase of this project, it did not negatively impact the usability of the apps for our subjects. In fact, the slightly larger screen display, and raised buttons on the slide-out QWERTY keyboard more likely further enhanced the ease of access from a physical (fine motor) and visual perspective.

The inclusion and exclusion criteria were very similar for Phases I & II of this project. The only difference is that for Phase II, persons who are covered under the UPMC Health Insurance Plan (2013) are also excluded in participating. During the development (Phase I) of the project, UPMC Health Plan, insurance provider, agreed to fund an in-person version of the program (Dicianno, et. al., *in press*). Since these individuals were now eligible for in-person

services, it was determine the best way to prevent any overlap of the remote (TR) and in-person wellness services was to exclude persons covered by the UPMC Health plan from participating in Phase II of the project. The following table provides a list of the inclusion and exclusion criteria specified for the clinical phase of the study (Table 12):

Table 12. Phase II Clinical Study Inclusion & Exclusion Criteria

Inclusion Criteria	Exclusion Criteria
• Primary diagnosis of Myelomeningocele (MMC) SB confirmed by physiatrist • History of shunted hydrocephalus confirmed by physiatrist • Age 18 to 40 years • Must be able to use a smartphone • Reside within the community (not within a residential facility that provides support; e.g. group home, personal care home or nursing home) • Location of residence within 100 miles of the City of Pittsburgh, Pennsylvania	• Formal diagnosis of Mental Retardation • Primary diagnosis of other types of SB: meningocele, lipomeningocele, lipoma, or spina bifida occulta. • Persons with a concomitant diagnosis of severe and persistent psychiatric illness (schizophrenia, bipolar disorder), active suicidal, homicidal ideation, and/or drug/alcohol addiction • Enrollment in UPMC Center for Wellness (In-Person Wellness Program for SB)

Staggered enrollment of subjects into the study began December 2011 and continued through the end of September 2012. While we had targeted 30 subjects for enrollment to account for attrition over the duration of the year-long study, recruitment of subjects who met all of our inclusion criteria to create a somewhat heterogeneous group of subjects proved to be challenging. Initially we sought to enroll subjects who resided only within Allegheny County, Pennsylvania, in attempt to limit travel time for enrollment, training and on-going in-person technical assistance by our research assistants. We later expanded the geographic region for recruitment to an approximately 100-mile radius of the city of Pittsburgh, Pennsylvania.

A short series of questions and observations made to determine whether or not the subjects met the criteria described in Table 13. Subjects performed several tasks as part of the screening process in order to enable the research recruiter to determine his or her ability to

effectively utilize the smartphone device. All potential recruits were briefly instructed and asked to use the smartphone device to complete the following tasks:

1) place a telephone call us, using the Line2 app and service
2) access and utilize text messaging
3) download and access information from a commonly used weather application
4) utilize the internet browser to look up a local movie theater's website and determine a current movie time

These screening tasks were almost identical to the screening tasks required to participate in Phase I of the study. The Line2 app was integrated into the project for Phase II to minimize costs. Line2 is a service which allows the user to place and receive call without cellular voice service. Instead, Line 2 uses the data plan on the device and can place and receive calls to wireless or landline phone numbers over Wi-Fi or the 4G data plan. This reduced the expense for the study considerably as we were able to eliminate the need to pay for cellular voice service on the plans and only needed unlimited data plans for phone calls and text messaging with the use of Line2 (Toktumi, 2012). Observation while the potential subject is completing these tasks significantly aided in determining whether or not participants would have the ability to successfully access and utilize the smartphone apps developed as part of the iMHere mHealth system. The Screening form utilized in this process is provided in Appendix A.

4.2.1.3 Collection of Demographic and Other Related Data

If the recruit successfully completed all of the tasks as part of the screening procedures identified above, he or she was enrolled as a subject into the clinical phase of the study. At this time, detailed demographic data was obtained through self-report as well as reviewing medical records through the UPMC Adult SB clinic. Many persons with SB are unaware of their own medical

histories. Reviewing medical records through the clinic helped to serve as verification of reported information related to clinical history such as a confirmed diagnosis of MMC, specific lesion level, and shunted hydrocephalus. The medical record also provided a source to obtain this information when the subject was unaware. The form utilized to collect and record demographic information in this process is included in Appendix B.

After completing the enrollment process, baseline data was obtained via telephone interview by a separate clinician/researcher, who remained blinded to the assignment of groups for the duration of the study. These data collected at baseline and on an on-going quarterly (every three months) basis included the following standardized assessments:

- Canadian Occupational Performance Measure (COPM) (Law, et al, 1990)
- Adolescent Self-Management and Independence Scale II (AMIS-II) (Austin, 1993)
- Craig Handicap Assessment Reporting Technique – Short Form (CHART-SF) (Mellick, 2000)
- Beck Depression Inventory (BDI-II) (Beck, 1996)
- World Health Organization Quality of Life Brief Instrument (WHOQOL-BREF) (WHO, 1998)

These telephone interviews lasted approximately 60 – 90 minutes for each subject at the five time intervals. At baseline the study was double-blinded. Both the research investigator administering the assessments and the subject were unaware of group assignment.

Following collection of baseline data, participants were randomized into intervention (TR – TeleRehabilitation-Enhanced Wellness Program) and Control (TC - Typical Care) groups. A Microsoft Excel © (Microsoft, 2010) file was utilized to generate odd (TC- control) and even (TR - intervention) values from a number table for random assignment to a group. Subjects were

then informed by the Co-investigator, who also serves as the WC for the study, whether they had been randomly assigned to the intervention or control group. The research activities involved as relevant for that subject's assigned group were again reviewed via telephone.

The subjects assigned to the TC group were advised to continue to go about typical daily routines and access to traditional health care services. In three months and later at six months, these subjects would hear again from the researcher, blinded to their group assignment. They would again complete the standardized assessments as outcomes. Subjects in the TC group received remuneration of $25 per follow-up telephone call completed with the blinded investigator who is an Occupational Therapist (OT). It was stressed that during these follow-up telephone calls, they should not discuss which group they had been assigned. The research administering the assessment via telephone, provided updates to the co-investigator aware of subject assignment to provide remuneration to the subjects in the control group.

Those subjects who were assigned to the intervention group learned they would soon be contacted by research assistants, who were undergraduate students, are serving in this role on the project. These research assistants travelled to the subjects' homes to provide a smartphone device and training in the use of the basic smartphone features during their initial visit. The usage of the smartphone along with an unlimited data plan, calling, text and picture messaging was the only incentive provided to the subjects in the TR group. Subjects were informed they were permitted to use the device for any personal activities (i.e. calling friends, family, on-line shopping, gaming, etc.) they chose, with the only exception being illegal activities as outlined in the addendum of the consent form.

The ability for subjects to utilize the smartphone device for activities outside of the research project was purposeful. The more the subjects utilized the smartphones, the more

meaningful the device would become in the subjects' daily routines. Because the subjects were provided opportunity to participate in the study for a full year, even those who already maintained a service plan through a wireless provider typically chose to end the contract and benefit from the free service and device provided through the study. The cost of the service plans was a significant portion of the overall budget for this research study. However, it was considered critical to ensure all subjects had the same access to wireless regardless of their ability to afford the cost of the device or service plan. Many persons with SB are of lower socioeconomic status and would otherwise be unable to afford this additional expense in order to participate in the study. Moreover, our survey conducted at the 2011 National Annual SB Conference (Fairman, et al, *in press*) revealed concern about individuals' abilities to remain aware of usage limitations and the potential to create exorbitant bills for overage charges of wireless services. To eliminate potential overage charges, it was determined that unlimited usage overall would provide the greatest freedom for our subjects in the TR group while also ensuring a predictable cost of service provision over the duration of the year-long study.

Staggered enrollment was planned beginning December 2011 and continuing through the end of September 2012. While we had targeted 30 subjects for enrollment to account for attrition over the duration of the year-long study, recruitment of subjects who met all of our inclusion criteria to create a somewhat heterogeneous group of subjects proved to be challenging. Initially we sought to enroll subjects who resided only within Allegheny County, Pennsylvania, in attempt to limit travel time for enrollment, training and on-going in-persons technical assistance. We later expanded the geographic region for recruitment to an approximately 100-mile radius of the city of Pittsburgh, Pennsylvania.

4.2.2 Training & Technical Assistance

The Wellness Coordinator (WC), who is co-investigator on this study, is a licensed OT with extensive experience in working with the SB population. The WC remotely discussed with intervention group participants the types of self-care activities they typically engage in to maintain health and wellness. This allowed for the development of a client-centered wellness plan and goals in conjunction with the results of the COPM, in instances where subjects had identified goals. A subject may have utilized only one or two of the apps appropriate for his or her specific needs. Many of the subjects in the TR group made use of all five of the smart phone (developed in Phase I – see Chapter 2) applications specific to his/her unique needs. Individual, in-person meetings occurred between the research assistants and the subjects to instruct participants in use of the device and relevant apps. Length of time for training varied by subject according to the number of apps utilized from three to six weeks. Following this development and training phase, persons in the TR group continued to receive remote wellness support for the remainder of their enrollment in the study. Those subjects who experienced technical difficulties were provided in-person support by the research assistants as necessary. Common reasons for in-person tech support visits included: replacement of broken devices, upgrades to smartphone applications, difficulties with maintaining charge due to faulty or worn out batteries and problems maintaining wireless connections. Wellness services with any clinical guidance were provided entirely remotely either through the iMHere mHealth system, telephone calls and basic text messaging. No in person contact occurred with the WC other than during the initial visit to obtain informed consent, since the WC was also the recruiter for this study. Weekly team meetings occurred between the undergraduate students providing technical assistance to the subjects and the WC to ensure all potential problems with detected and addressed quickly.

4.2.3 Provision of Wellness Programming through mHealth

The TR Enhanced Wellness Program utilizes a similar targeted case management approach like that of the successful SBAWP Wellness Program (Dicianno, et al, *in press*) described in Chapter 2. Wellness interventions in both of these programs are based on an enhanced health care management model. This model utilizes a proactive and holistic approach to health care by identifying the issues a client faces, and then designing an individualized plan of care to ensure that appropriate care and supportive services are delivered. This model of wellness programming seeks to meet the need for a comprehensive approach to addressing the needs of young adults with the medically complex diagnosis of spina bifida. The WC works collaboratively with health care professionals (i.e. physicians, physical therapists, nurse practitioners, insurance providers). In this instance, Dr. Dicianno, as the Director of the UPMC Adult Spina Bifida clinic, serves as the primary clinical liaison. However, it should also be noted the WC for this study is a licensed OT with over ten years of clinical experience and has extensively worked with the SB population. Clinical collaboration typically occurred on a weekly basis through a conference call to discuss subjects' clinical needs. The WC provided daily monitoring of the iMHere portal of incoming information from the subjects' apps. It is important to distinguish use of the iMHere system for wellness programming is not just remote monitoring, but rather a program in which the WC intervenes and encourage participants to change their behavior and become more responsible in carrying out their own health and wellness activities. In addition, the WC can help the individuals in the TR group to access services they may otherwise not receive.

4.2.4 Outcomes Measures

Two standardized, reliable assessment tools have been selected to measure the impact of TR-Enhanced Wellness programming upon increasing self-management skills in young adults with SB. These measures, the COPM © and AMIS-II, are described in detail below:

4.2.4.1 Canadian Occupational Performance Measure (COPM) ©

Designed as an outcome measure, the COPM is an individualized, client-centered, outcome measure designed for use by occupational therapists. The measure is designed to detect change in a client's self-perception of occupational performance over time. The COPM is appropriate to be utilized across developmental stages and a wide variety of disability groups. It has also been used to assess the effectiveness of various treatment interventions including assistive technology (Petty et al., 1999; Reid et al., 1999; McColl et al., 2000, Gentry, et al., 2008). The COPM is a standardized assessment tool which uses a semi-structured interview format of delivery and a structured scoring method. Change scores between assessment and reassessment using the COPM are the most meaningful scores derived from this standardized tool. The COPM is appropriate to administer as an initial assessment. The COPM will aide in developing a positive rapport while setting the tone of the therapeutic relationship. The issues that the person feels are priorities are identified through the COPM assessment process, letting the individual know you will be working as partners to achieve their goals in relationship to areas they identify occupational performance issues including self-care, productivity, and leisure domains of function.

The COPM is fairly easy to administer and takes approximately 20-40 minutes in most instances. The individual is interviewed and asked to provide an account of their typical day.

The person is asked to consider which activities they encounter throughout the day are difficult to perform. The top five occupational performance problems according to the level of importance of the individual are then identified and rated (1=lowest level of performance, 10=highest level of performance). The person then rates their current level of satisfaction with their performance on a scale of 1-10 (1=least degree of satisfaction, 10=greatest degree of satisfaction). Scoring of the COPM is also very easy to calculate. Performance and satisfaction scores are totaled and then divided by the number of problems. The person is then reassessed and the change in performance and satisfaction scores determines the outcome of the intervention Ninety-eight papers have been published which have positively supported the clinical utility, validity and responsiveness of the COPM within a variety of treatment settings and clinical populations (Law, et al, 1990; Law, et al, 1998). The COPM has been implemented with the SB population in a study that looked specifically at the perceived benefits of word prediction software (Tam et al, 2002).

4.2.4.2 Adolescent Self-Management Independence Scale Version II

The Adolescent Self-Management Independence Scale Version II (AMIS-II) is a reliable tool used to assess clinically relevant self-management skills in teens and young adults with SB. Psychometric properties of the AMIS-II have been evaluated using a sample of 135 parent-child dyads from four different spina bifida clinics. The age range of the participants was 12-25 years. While participants in this study extend up to age 40 years, questions were revised and worded appropriately for the developmental age-level. The AMIS-II is comprised of 17 questions demonstrating construct validity and reliability (Buran, et al, 2006). The assessment's items are rated using a 7-point response category (1 = total assistance to 7 = no assistance). The AMIS-II requires rating by a health care provider, but can be administered fairly quickly. This assessment

tool provides data that allows for the measurement of skills which are critical for successful self-management skills as an adult with SB. The AMIS-II is administered by interview and measures the level of independence across multiple domains, including personal care skills such as taking medications and toileting, and community living skills such as making and keeping appointments and refilling prescriptions. The AMIS-II has been utilized in several transition research studies for persons with SB. For instance, Waring (2009) conducted a study using a convenience sample of 26 young adults and 13 parents attending a transition program in Wisconsin found that most of these young adults (ages 18-35 years, mean age 23 years) with spina bifida, required supervision in managing their personal health needs and community living skills. Likewise, Bellin and colleagues (2011) utilized the AMIS-II in their study to increase the knowledge base around how gender, lesion level, self- management, community integration and quality of life (QOL) are interrelated in young adults with MM. These researchers found a need for young adults with MM to receive ongoing support in the performance of ADLs and condition-specific self-care tasks utilizing the AMIS-II assessment (Bellin, et al, 2011).

4.2.5 Statistical Analyses

To reduce the probability of obtaining a Type II error, an "a priori" power analysis was completed using the G*Power software program, version 3.1.3 to determine sample size (Faul, et al, 2007). A moderate effect size of 0.30 was applied based on preliminary data gathered through an in-person SBAWP Wellness program for persons with SB. The outcomes data from this pilot study suggest the potential for a 30-50% reduction in the incidence of secondary conditions (i.e. pressure ulcers and urinary tract infections) in comparison to a similar population of persons with SB not receiving wellness coordination services (Dicianno, et al, *in*

press). Using a one-way analysis of variance approach for the repeated measures design with five data collection points, a total n of 22 subjects yields a power of 95%. It was our aim to over-recruit a total of 30 subjects to account for subject attrition during the year-long study. All subsequent statistical analysis was conducted using the IBM SPSS version 21.0 software program (IBM, 2012) or Microsoft Excel (Microsoft, 2010).

Descriptive analyses were used to examine demographics, prior experience with technology and baseline functioning though the administration of the: AMIS-II, BDI, CHART-SF, COPM, and WHOQOL-BREF. All demographic data were examined in order to determine if the four basic assumptions of parametric tests had been met. These assumptions in this initial phase of the analysis include: 1) normality and 2) homogeneity of the data, 3) all data has been measured at least at the interval level, and 4) each subject's data is independent from the data of other subjects enrolled in the study.

Histograms and boxplots were created using SPSS to visually examine the distribution of the data, using a bar graph format. In addition the skewness and kurtosis of the variables were examined for the entire sample (N=25). Data were considered normally distributed if skewness and kurtosis values fell between -1.0 and 1.0 and data appeared to be normally distributed on plots. Values which fall outside the range of -1.0 to 1.0 do not meet the assumption of normality (Wright, 2002).

In three instances the information obtained during the telephone follow-up interviews for the AMIS-II assessment were coded as not applicable at the time of the three month follow-up. For the AMIS, this was not an appropriate way to handle this data when scoring the assessment. The assessor had mistakenly coded these individual items incorrectly, but this represents a very small amount of the data that was collected. In these three instances, the subjects' scores at

baseline were utilized in what is known as a last observation carried forward (LOCF) approach to managing missing data (Portney & Watkins, 2009). This was the most conservative estimate of how the person could have been doing at this point in the study, given that scores were expected to improve or remain the same.

The main analysis of the outcome measures for intervention versus control group was carried out as a 3 × 2 mixed design ANOVA. Three measured time points of the outcomes variables include: 1) baseline, 2) three months, and 3) six months the two levels of the independent variable (intervention and control groups). The main analysis reported for change in self-managements skills is reflected by the AMIS-II assessment alone, since the COPM did not yield any useable data for analysis. At baseline, analyses to determine if the assumptions of a mixed design ANOVA had been met were performed for the AMIS-II outcomes measure. The assumption of normality and compound symmetry were violated for the AMIS with a few outliers identified from this small group of subjects. Transformations of the AMIS data including log, square root and reciprocal transformations, were attempted and unsuccessful in achieving normality.

Bootstrapping is most useful as an alternative to parametric estimates when the assumptions of those methods are in doubt (as in the case of regression models with heteroscedastic residuals fit to small samples), or where parametric inference is impossible or requires very complicated formulas for the calculation of standard errors (as in the case of computing confidence intervals for the median, quartiles, and other percentiles) (Azzalini & Hall, 2000, Boos & Brownie, 1989, Effron, 1994). Therefore, bootstrap p-values were computed rather than making other adjustments in order to correct for the violation of

assumptions. Correcting for the violations of assumptions could have greatly complicated the interpretation of the results, especially with this small group of subjects.

Effect size was calculated to determine the strength and practical clinical significance of the association found between time and group for the AMIS-II measure. Since the TR and TC groups did not have the same number of subjects the pooled standard deviation (*sd*) was calculated to show the level of spread of the AMIS-II scores. Cohen's d was utilized to calculate the effect size by subtracting the mean of the TC group from the mean of the experimental group and dividing by the pooled *sd* (Portney & Watkins, 2009).

4.3 RESULTS

Informed consent was obtained from 27 subjects by our targeted end date of September 2013. The consort diagram (Figure 20) provides a summary of subject enrollment, randomization, drop-outs and continued participation through the first six months of the overall study.

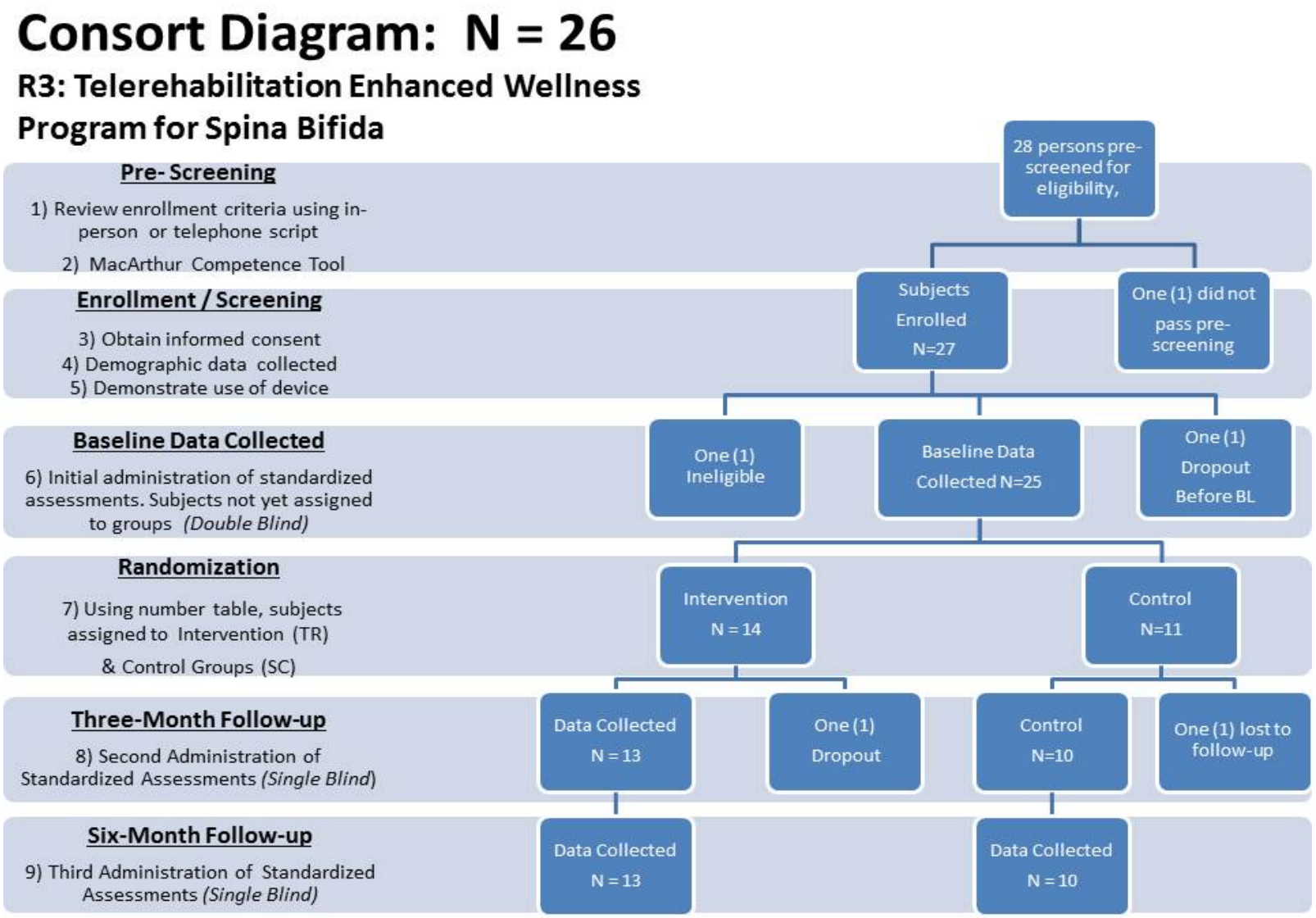


Figure 19. Consort Diagram Clinical Phase

As indicated in the consort diagram, four subjects are no longer participating in the study for the following reasons:

1. Upon review of medical records, it was determined that one subject did not actually have a diagnosis of SB
2. A second subject was no longer interested in the study by the time she was contacted to complete the baseline assessment measures via telephone
3. One subject declined further participation in the study due to the fact that wireless service was so poor where he resided and worked that he could not effectively utilize the smartphone
4. Thus far, only one subject has been lost to follow-up after baseline data was collected

A total of 23 subjects have completed the first six months of the study for analysis at three time points. Subjects were enrolled in the states of Pennsylvania, Ohio and West Virginia, with some of the farthest subjects residing in central PA (see Figure 21).

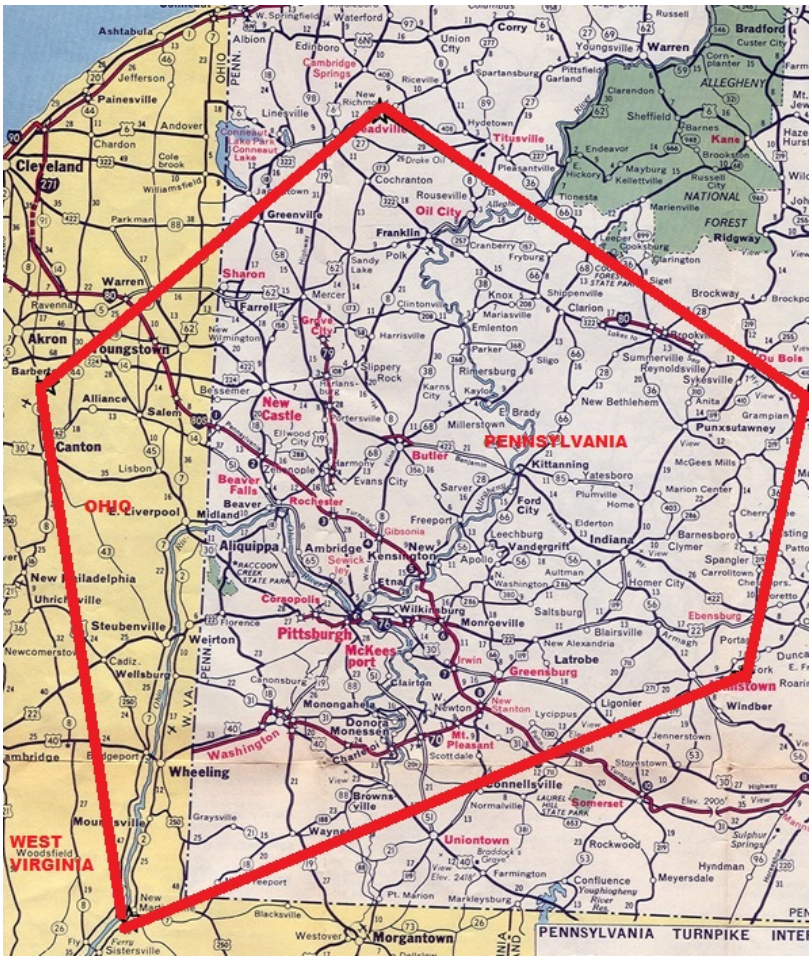


Figure 20. Geographic Reach of Clinical Phase

Demographic characteristics of the study population are listed in Table 12.

Table 12. Phase II Clinical Intervention – Demographic Variables

Demographic Variables		Intervention TR, n = 14	Control TC, n = 11	Statistical Test p value
Mean Age in Years		30.7 (SD 4.999)	29.9 (SD 6.564)	Mann-Whitney 0.936
Gender	Male Female	9 (64.3 %) 5 (35.7%)	5 (45.5) 6 (54.5%)	Chi-Square 0.346
Ethnic Origin Caucasian Hispanic		13 (92.9%) 1(7.1%)	11 (100%) 0	Fisher's Exact 1.000
Marital Status	Single Married	13 (85.7 %) 1 (7.1%)	11(100%) 0	Fisher's Exact 1.000
Highest Level of Education Achieved	High School Diploma (only) Higher Education or Training (College, Trade, Tech)	3 (21.4%) 11 (78.6%)	3 (27.3%) 8 (72.7%)	Fisher's Exact 1.000
Received Special Education Services		12 (85.7%)	9 (81.8%)	Fisher's Exact 1.000
Lesion Level By Group	L2 & Above L3 – L5	9 (64.3%) 5 (35.7%)	7 (63.6%) 4 (36.4%)	Fisher's Exact 1.000
Assistive Device used for Mobility	Ambulates w/ or w/o Assistive Device Non-Ambulatory uses WC or other PMD	2 (14.3%) 12 (85.7%)	3 (27.3%) 8 (72.7%)	Fisher's Exact 0.623
Utilizes Paid Personal Assistance		5 (35.7%)	4 (36.4%)	Fisher's Exact 1.000
Living Situation	Resides Alone Resides with others	1 (7.1%) 12 (85.7%)	4 (36.4%) 5 (45.5%)	Fisher's Exact 0.350
Employment Status*	Unemployed Employed (FT or PT)	11 (78.6%) 3 (21.4%)	2 (18.2%) 7 (63.6%)	Fisher's Exact 0.005*
Describes Self as "Tech-Savvy"		13 (92.9%)	10 (90.9%)	Fisher's Exact 0.697
Has previous experience using smartphones		9 (64.3%)	6 (54.5%)	Fisher's Exact 0.648
Has previous experience with "apps"		8 (57.1%)	7 (63.6%)	Fisher's Exact 1.000
Is a smoker		(7.1%)	0 (0%)	Fisher's Exact 0.560

The functional abilities of the SB population vary considerably. This variability has the potential to impact the outcomes of this study; therefore, we attempted to utilize a stratified sampling

approach. It is more appropriate to classify patients with SB into these three main functional motor levels rather than using specific vertebral level. Lesion level has been grouped according to three functional categories as identified by Verhoef, et al (2006). While gathering demographic data, questions were also posed to gain an understanding of the participants' previous exposure and use of technology. Subjects in the TR and TC groups share similar experiences. All individuals in both groups received formal computer training while in high school and/or as part of an advanced educational program (i.e. trade school or college). The majority of participants described themselves as "Tech Savvy," meaning they view themselves as knowledgeable with regard to technology.

4.3.1 DESCRIPTIVE ANALYSIS OF GROUPS - BASELINE DATA

A summary of the participants' scores from standard assessments administered at baseline can be found in Table 13. The purpose of presenting the data in this manner is to provide a general sense of the sample's functional status as a group for comparison to other populations of persons with SB and related disabilities.

Table 13**.** Description of Subjects by Outcomes Measures

Outcomes Measures at Baseline Only	**TC Group (n= 10)**		**TR Group (n =13)**		**p-values**
AMIS – II	Mean	101.30	Mean	89.92	Mann-Whitney
Total Independence Score	SD	21.14	SD	21.44	0.107
	Mean	43.60	Mean	38.92	t-Test
Condition Mgt. Subscale	SD	7.69	SD	9.29	0.350
CHART-SF	Mean	93.60	Mean	77.85	Mann-Whitney
Physical	SD	10.01	SD	30.62	0.244
	Mean	73.80	Mean	71.54	Mann-Whitney
Cognitive	SD	25.07	SD	34.98	0.647
	Mean	82.00	Mean	66.69	Mann-Whitney
Mobility (N=25)	SD	13.70	SD	22.22	0.134
	Mean	58.13	Mean	38.56	Mann-Whitney
Occupational	SD	36.90	SD	34.43	0.317
	Mean	92.35	Mean	81.69*	Mann-Whitney
*Social	SD	18.40	SD	18.40	0.434
	Mean	68.04	Mean	54.88**	Mann-Whitney
**Economic	SD	37.47	SD	31.59	0.201
WHOQOL (N=25)	Mean	13.89	Mean	13.80	t-Test
Domain 1- Physical	SD	1.59	SD	1.63	0.270
	Mean	14.73	Mean	14.87	t-Test
Domain 2- Psychological	SD	1.58	SD	1.81	0.384
	Mean	16.00	Mean	16.00	t-Test
Domain 3- Social	SD	2.40	SD	2.93	0.528
	Mean	16.75	Mean	17.15	t-Test
Domain 4 - Environmental	SD	1.87	SD	2.13	0.662
BDI (N=25)	Mean	3.90	Mean	6.46	t-Test
Total Score	SD	5.38	SD	6.37	0.290

*Two subjects did not feel comfortable discussing sexuality to complete this domain
**Two subjects did not wish to discuss information related to their financial situation

The WHOQOL domains, BDI, and AMIS-II Condition Management Subscale were normally distributed variables and therefore t-tests were conducted to ensure the groups were not significantly different at baseline. The AMIS Total Independence and CHART-SF sub-domain scores did not meet the assumptions for normality and the Mann Whitney U was utilized. While employment status was found to vary significantly (as reported in Table 13) when comparing subjects of the TR and TC groups, it is not necessary to

incorporate this as a confounding variable in future analysis since we used the CHART-SF as a more robust measure of occupational status, and the groups were statistically similar with respect to this measure. In particular, the CHART-SF Occupation Subdomain seeks to measure function with regard to occupation as a broader construct than employment status alone. Occupation does not merely refer to one's job or paid vocational activities, but overall engagement in meaningful, purposeful activity of any type. So the argument here is that while many persons in the intervention group were unemployed, this does not mean they were less engaged in occupations than the intervention group.

4.3.2 MAIN ANALYSIS: OUTCOMES OF THE COPM & AMIS-II

Unfortunately, the COPM did not yield any useable data for analysis. Many subjects in both the intervention and control groups could not identify goals at baseline or in subsequent follow-up interviews. The primary focus on self-management skills has been effectively captured by the AMIS-II assessment.

As previously described in the methods section, bootstrapping was applied due to the fact that normality could not be achieved with application of transformations to the baseline data. Similar results found between the bootstrap p-values and ANOVA p-values. This similarity suggests the mixed ANOVA results are robust even with the violations of the test assumptions as indicated in Table 14.

Table 14. AMIS II Total Independence Score Results

	Boot *p*	**Mixed ANOVA *p***
Time	.308	.323
Time x Group	.208	.006
Group	.375	.366

Significance was found in looking at Time X Group with *p* value of .006. Table 15 provides the mean change difference for the AMIS-II Total Independence Score from baseline to three months (Change 1) and three months to 6 months. The greatest change occurred in the intervention group from three months through six months. In addition, the intervention group gradually improved their scores over time. However, the control group improved slightly from baseline to three months and then declined in their functional performance as assessed by the AMIS-II from three months to six months, for a lower score than had originally been obtained at baseline.

Table 15. AMIS-II Total Independence Change Scores

Group	Change 1	Change 2	Overall Change
Intervention	0.83	6.92	7.75
Control	1.82	-3.82	-2.00
Group	**Baseline**	**3 mos**	**6 mos**
Intervention	89.17	90.00	96.92
Control	101.09	102.91	99.09

These change score were also utilized to calculate the effect size of time and group. A moderate effect size of 0.46 was found in the association of group and time.

Figure 22 provides a visual, graphical image of the results provided in Table 15.

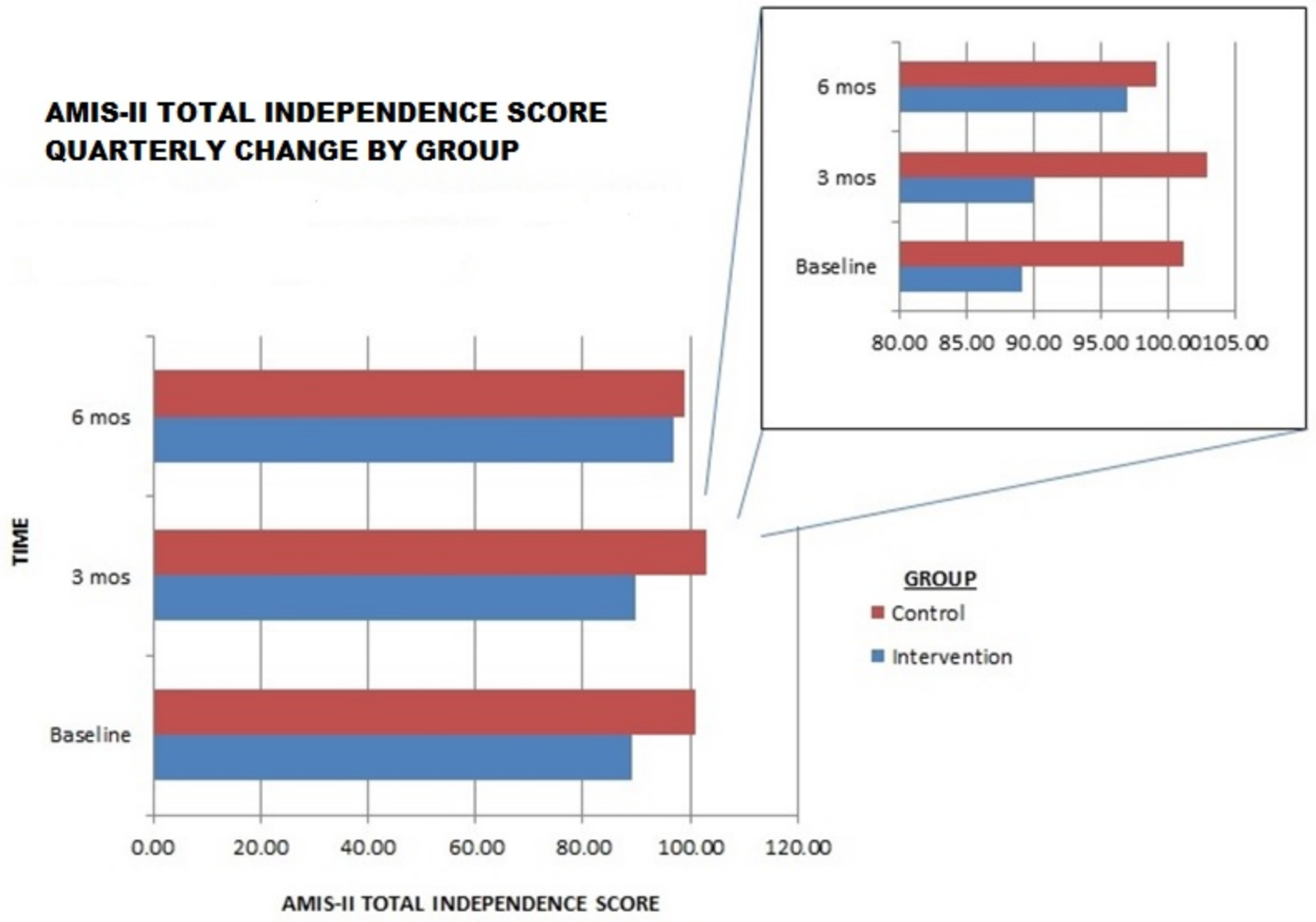


Figure 21. AMIS-II Total Independence Score Quarterly Change Scores by Group

4.4 INTERPRETATION AND DISCUSSION

These results suggest the mHealth Wellness intervention was a successful method for improving self-management skills as assessed through the AMIS-II. In particular subjects in the intervention group exhibited the majority of improvement during the three to six month time interval. Considering that subjects were involved in training in the use of the apps anywhere from three – seven weeks, it was anticipated change in self-management skills may require the longer duration of time. Participants in the study will continue to be followed. AMIS-II assessment scores, in addition to the other outcomes measures previously described, will be obtained again at nine and twelve months. This additional time will help to determine if this improvement in function will continue or if subjects have already reached a maximum degree of improvement with use of the system.

There are a variety of limitations must be taken into consideration with this small study. As baseline testing was being completed, we found that many of the subjects lacked sufficient insight into personal health needs and were unable to identify any potential areas for improvement in self-management skills. The researcher, who conducted the telephone based interviews to gather the data from our battery of standardized assessments, noted that the COPM was typically administered as the last assessment at the end of an hour-long or longer phone call. Due to fatigue, some subjects may have been reluctant to continue the conversation any longer than necessary at this point. This researcher also noted that a number of the participants lacked

the insight necessary to establish goals. As a result, the COPM is not included in the reported findings of this analysis.

This lack of insight is present despite numerous difficulties the subjects are encountering in relationship to their health and well-being. The model developed by Stuifbergen et al (2000), suggests changes in self-efficacy often precede changes in health-promotion behavior and quality of life. Without sufficient knowledge and self-efficacy persons cannot identify the need, strategies or their ability to change and influence their own health status. We anticipate being able to develop increased insight through participation in the intervention and expect persons to gradually gain these skills throughout the duration of the year-long intervention.

Only the AMIS-II scores were available for analysis to determine if the intervention had any impact on self-management skills with our study population. In some subjects, there may have been a ceiling effect using the AMIS-II standardized assessment t if they had high scores in several domains at baseline. In this case, the AMIS-II would not reflect any further positive change in these individuals. The assessment may not have accurately represented these subjects' full functional abilities and/or change over the time they were enrolled in the study. While this is a potential limitation in this study, the AMIS-II was the most appropriate tool to measure self-management skills in the population. If the COPM had been successfully incorporated for all of the subjects, this would have allowed for a more individualized means to look at each subject's change in function over time as well as satisfaction ratings with regard to their current level of satisfaction in self-identified goal areas (Law, et al, 1990).

In our attempts to control for extraneous factors by using a fairly homogenous sample, we were able to improve internal validity in the design of this study. However, the cost is that our sample size is relatively small and not representative of the overall population of persons

with SB. There were many potential subject traits we felt may interfere with the independent variable (i.e. age, lesion level, cognitive status, etc.). Therefore we developed very specific inclusion and exclusion criteria to control for and eliminate potential confounding factors. We were able to create a fairly homogeneous group in doing so, but the impact of SB is still incredibly diverse. We recognize this limitation and expect utilizing a mHealth intervention such as this would be effective only for a portion of the SB population. In addition, our research group did not provide much ethnic diversity. With the exception of one participant in the intervention group who is of Hispanic descent, all other subjects described themselves as Caucasian. The resulting ethnic distribution of the participants is fairly representative of the geographic area where we recruited our subjects. However, our study population is not representative of the overall population of persons with SB in the United States. The National Birth Defects Prevention Network, a workgroup of the CDC, reports women of Hispanic decent currently have the highest rate of having a child affected by spina bifida (Boulet et al, 2008).

There was likely a degree of sampling bias due to self-selection by only persons who felt comfortable with using the smartphone technology. While both groups self-selected and were equally comfortable with using the smartphone device, the overall study sample is not representative of a population of persons with SB. Many persons do not view themselves as "tech-savvy" or may be much less comfortable using a smartphone or have little interest in technology as was identified in our recent survey at the 2011 SBA Annual Conference (Fairman, et al, *in press*).

Others may not be able to utilize a smartphone device due to physical, sensory or cognitive limitations. Our inclusion criteria required that subjects have physical dexterity (fine motors skills) and little or no sensory deficits (vision or hearing) which would interfere with use of the

smartphone device. While most subjects in our study had some degree of cognitive disability, exclusion criteria prevented anyone with intellectual disability severe enough to qualify for a formal diagnosis of mental retardation (IQ < 60). There were persons excluded from the study who expressed interest in participating either because of an MR diagnosis or they lacked the cognitive skills necessary to successfully perform the screening tasks.

Another potential limitation of our study involved the incentives or perceived incentives for participation. For individuals participating in the intervention group, a free smartphone with a full unlimited service plan for the duration of an entire year was extremely motivating. Some individuals in the control group expressed disappointment they had been randomly assigned to the TC group where the incentive could reach $100 over the duration of the year-long study. It was recognized by some subjects that the value of the smartphone and service plan far exceeded this with a monetary value of approximately $1200. Such a strong incentive may have encouraged continued participation in the intervention even if the individual was dissatisfied with the services or no longer had an interest in participating. However, it should also be noted the majority of individuals had a cell phone or even a smartphone with a full service plan prior to participating in the study. Some participants in the control group also expressed their genuine interest in participating in the wellness program with hopes they may be able to receive the service through the clinic in the future. Several of the subjects in the control group expressed disappointment upon learning they had not been assigned to the intervention group, despite the remuneration incentive. Others were glad they were assigned to the control group, because they already owned a smartphone and were more interested in receiving the monetary incentive. None of the subjects dropped out of the study upon learning which group they had been assigned.

Another concern may be that it is not actually the use of the iMHere service that resulted in the improvement in scores for the intervention group. Instead it is the smartphone device and the increased access to information and communication options that has improved self-management scores. Commercially available smartphone devices have a number organization tools and features already built into them. In addition, there are literally tens of thousands of apps that can be downloaded to these devices for use. While not all of these apps have the potential to improve self-management skills, some certainly could be utilized in this way. The subjects were able to utilize any aspect of the device in an unlimited manner. It is possible that they used other features on the phone which were actually the cause of their improved functioning. It is also possible subjects may have utilized other apps and features on their study-issued smartphone that further enhanced their functioning, many of the participants of the control group also had access to their own personal smartphone device prior to participating and throughout the duration of the study. At baseline subjects were asked if they had previous experience in using smartphones or apps. Of the overall group (N=23), 69.6% had previously utilized a smartphone and apps in the past. Alternatively, if budget limitations had not been an issue and we had been able to successfully recruit more persons for the study, the control group could have also received smartphone devices. Also, if budget restrictions and more subjects had been recruited, a third intervention group could have been created in which persons were issued the devices and trained to use the smartphones' basic features as well as some commercially available smartphone apps intended to improve health and well-being as a possible "placebo" intervention. Analysis would have included comparison of the outcomes measures across the three groups and over time. However, cost was a concern for this study particularly related to the expense of the service plans for unlimited data, text messaging and calling.

While we were careful to plan for no further in-person contact by the WC, we were unable to provide services without some degree of in-person support for training and technical assistance. As previously described, undergraduate students provided the initial individualized training in the use of the apps at the subject's home. This training period lasted for three weeks (0ne visit per week) to six weeks. However, if subjects required additional support for difficulties with the device itself or other technical problems the assigned support person would follow-up as needed. Attempts were initially made to assist the person remotely. In some instances, remote support could not remedy the problem and additional visits were made. A potential argument could be that it was not the intervention that improved the individuals' scores but the increased social contact and support provided through the training and technical assistance support staff. Persons in the control group did not receive this extra contact. In fact, there was at least one instance where one of the intervention group participants seemed to fabricate technical problems in order to receive additional in-person visits for technical support and really just seemed to crave the opportunity for social interaction during the visit. Once this issue of "technical malingering" was identified by the research team, we developed strategies to decrease it. Additional attempts to have the individual trouble-shoot problems on her own were directed by the WC. The clinician portal has been developed in order that the clinician can program and correct problems remotely as well as view any changes made to the apps by the user through the smartphone. This subject's use of the apps was reviewed and it was identified that she was deleting information and then indicating her smartphone was faulty. The WC identified these activities with the subject and the behavior ceased. Providing all support remotely would have been ideal, but simply not realistic given the many various scenarios and technical problems that were encountered. In-person visits were minimized to the greatest extent

possible and were focused only on training and technical assistance for use of the smartphone device and apps. Clinical support by the WC only occurred remotely, which was the best way we could try and control for this potential limitation in the study design.

Despite the randomization of subjects into groups, in the end more persons were assigned to the intervention group than the control group due to the small sample size and attrition. The study experienced a loss one subject in each group after randomization. Two additional subjects were lost prior to completion of baseline testing and randomization. Used on-protocol or completer analysis was utilized because further data were not available on the subjects lost to attrition and voluntary drop out. Portney and Watkins (2009) note the on-protocol approach tends to favorably bias the results of a treatment effect. This is due to the fact that those persons who experience positive results from an intervention are more likely to stay involved than a person who does not. However, since we had one person lost from each group this remains less of a concern.

The single-blind design of this study is a strength increasing rigor of the results, especially given the nature of the outcomes measures utilized. The researcher collecting the data from the standardized assessments remained blinded to the assignment of subjects to groups for the entire duration of data collection. The subjects themselves were aware of their group assignment following baseline testing. A double-blind study would have been impossible to carry-out. However, the previous scenario described with two levels of intervention where the 1) subjects use commercially available apps, or 2) subjects use iMHere system, could have allowed for a double-blind study to be carried out.

The results reported in this dissertation only include baseline, three month and six month time intervals. The length of the overall study is twice this duration of time. It is possible that

increase in skills or perception of improved skills is only temporary. We will continue to follow these subjects and analyze the AMIS-II as an outcomes measure again at nine months and twelve months.

Finally, as with most any research study there is a potential for the Hawthorne effect to influence the results. In addition to the fact they knew their responses were being studied, they may have responded favorably in attempt to support the concept of the intervention. Many of the subjects were excited about the potential for this innovative system to become a successful intervention.

4.5 SUMMARY

While there have been quite a few limitations identified in this study, the process of implementing such an extensive clinical trial with the innovative iMHere system, has provided an incredible amount valuable information in itself. This study is the first of its kind in implementing a mHealth intervention within the community setting to promote self-management skills across such a wide variety of self-care activities. The limitations of the study must be carefully considered before attempting to develop clinical programs and interventions. Additional research is necessary with a few of the many potential opportunities for future research activities are discussed briefly in Chapter 5.

5.0 FUTURE RESEARCH

5.1 OVERALL STUDY R3: TELEREHABILITATION ENHANCED WELLNESS PROGRAM FOR SPINA BIFIDA

Goals of this research project are improve health outcomes for persons with SB via Telerehabilitation (TR). Aims of TR- Enhanced Wellness program include improving the access, efficiency and overall effectiveness of care. Our hypotheses is that in comparison to the control group (typical care) the intervention group (TR-Enhanced Wellness Services) will demonstrate: 1) improved health and rehabilitation outcomes, and 2) decreased time and costs related to supporting persons with Spina Bifida in managing health and wellness needs.

On-going collection during the enrollment in the study also includes data related to medical complications and health care utilization. In addition to the COPM and AMIS-II standardized assessments, the other outcomes measures utilized in the overall study are listed below. Our hypothesis is that, in comparison to the control group, persons participating in the intervention group will demonstrate improved:

<u>Functional outcomes</u> as assessed by:

- Craig Handicap Assessment Reporting Technique – Short Form (CHART-SF) (Mellick, 2000)
- Beck Depression Inventory (BDI-II) (Beck, 1996)

Quality of Life, Satisfaction with health care services, as assessed by:

- World Health Organization Quality of Life Brief Instrument (WHOQOL-BREF) (WHO, 1998)
- Patient Assessment of Chronic Illness Care (PACIC) (Glasgow, 2005)

Medical Outcomes, as assessed by between group comparison of:

- Duration & frequency of urinary tract infections and wounds
- Incontinence episodes
- Missed medications, appointments & testing
- Number of emergency room visits
- Number & length of acute hospital stays

In addition, we plan to calculate an estimated cost-benefit analysis of medical care based on typical charges for services. Much of this information can be automatically captured and tracked using the iMHere portal through use of the Smartphone apps for those subjects enrolled in the intervention group.

5.2 FOLLOW-UP SEMI-STRUCTURED INTERVIEW REGARDING EXPERIENCE OF R3 INTERVENTION GROUP PARTICIPANTS

Following completion of the year-long, it will be valuable to seek follow-up with participants to the intervention regarding their experiences to gather feedback for improvements to the system and the programming. Subjects' usage of the system was varied, with some extensively using all five of the apps throughout the day and others intermittently using only two of the apps. Due to the fact there is only a small group of persons in this intervention group, it is not likely we would

be able to attention much data through seeking quantitative feedback alone. Qualitative data would provide richer content and can be gathered through semi-structured interviews. The nature of qualitative research is inductive. Therefore, this type of research does not usually have a conceptual framework or hypothesis to be tested (Morse, J.M. & Field, P.A., 1995).

This study will utilize a phenomenological approach. Phenomenology as an approach to qualitative research has two primary implications: knowing what people experience and how they interpret the world (Patton, 2002). In order to gain this understanding, methods must allow the researcher to understand what the person experiences as directly as possible. The goal of phenomenological method is to provide an accurate description of the phenomena being studied. The use of well designed, in-depth individual interviews with the R3 intervention group participants will help to capture the essence of their experiences and perspectives. This insight will allow for decisions to be made regarding future improvements to the iMHere system as well as the remote wellness.

Separate IRB approval will need to be obtained prior to conducting the interview including informed consent of these research participants on a new consent form specific to these research activities. Voluntary and written informed consent for participation in this study will be obtained from all of the participants prior to the initiation of any research activities. Once the recruiter arrives at the potential participant's home in-depth information will be provided regarding the nature of the study including its purpose and significance, as well as the data that will be collected. If the participant is still interested in participating, written, informed consent will then be obtained. A small financial incentive (i.e. $25 debit card) could be issued in

exchange for the participants' time spent in the interview, which would last approximately one hour. This data should be gathered as subjects from the intervention exit the study near their one year anniversary date. Ideally, the interview will occur in the individual's home or other private, quiet environment. The researcher will seek permission to audio record the interview. Narrative data will include transcripts of all interviews as well as any field notes relative to direct observations from the natural settings where the individual interviews will take place, in the participants' homes

The aim is to provide a phenomenological understanding of the participants' experiences through an interactive, collaborative story (Padilla, 2003). Categories and themes derived from the data will be used to generate a one page summation and/or graphic display of the participants' experiences. This one page summation and/or graphic display will be shared with each participant individually and the participants' reactions and commentary to the summation will be audio taped, transcribed and analyzed. A constant comparative methodology will be employed throughout the data collection and data analysis cycle (Strauss & Corbin, 1998). Concept formation will begin using a process of open coding. Initial codes will be generated by analyzing what and how participants respond to guided reflections and interview questions in an attempt to understand how the participants were interpreting the events that they discussed. Concepts in the data will be further developed through the processes of axial coding, the reduction of categories, and theoretical sampling. As the coding process continues, central ideas will be refined as concepts, and the properties and dimensions of these concepts will be identified in such a way that categories of concepts will be delineated, and the range of properties of any given category can be specified and grouped together (Lincoln & Guba, 1985).

Reducing the number of categories is a process that will continue throughout the data analysis. Initial analyses of the data will be completed by hand. The primary investigator will conduct this initial analysis and finds this method to be most productive. In each successive coding session a clean printout of an interview will be used and hand coding will continue. Memos will be generated throughout this iterative coding process and recorded in electronic format. During the concept development phase of analysis, and after several iterations of hand coding, an initial coding matrix will be entered into the latest version of ATLas.ti© (Scientific Software Development, 2013) computer software. ATLas.ti© is computer software that qualitative researchers have employed to manage, access, and evaluate data (Muhr, T., 1997). This software supports the coding process and the handling, processing, and organization of the qualitative data, and will allow for comparisons within and between cases.

Trustworthiness of this study will occur by multiple means. Triangulation will occur by data source and analysis. Multiple sources of data include: chart reviews for basic demographic data, data base of health-related outcomes, and individual interviews / in-depth conversations with young adults with SB. Finally, this project will utilize member checking to ensure categories and themes defined in the data resonate with authenticity to the participants who provided the data.

5.3 APPLICATION TO YOUNGER POPULATIONS (TEENS) WITH INTERGRATION OF NATURAL SUPPORT SYSTEMS

Opportunity also exists to apply this intervention to a younger population of teens with SB. The early teen years are a developmentally appropriate time in one's life to be seeking separation from one's parents and gain full independence with regard to self-management. As previously discussed in Chapter 1, many teens fail to develop the self-management skills necessary to independently manage medical and self-care routines (Holmbeck, et al., 2002; Davis, et al. 2006; Sawin et.al. 2009; Bellin, et al, 2011). A recent article by Rauen et al (2013) emphasizes a need for successful pediatric to adult healthcare transition programs.

Incorporating mechanisms for caregiver and family involvement within the iMHere system is another means to expand application of the innovative iMHere system to a broader population.

5.4 APPLICATION TO OTHER CHRONIC CONDITIONS

Likewise, another logical step for expanding usage of this system is to consider other populations of persons with chronic conditions who may benefit from receiving wellness services with iMHere as the vehicle for service delivery. Potential populations which have significant medical needs, a variety of self-care tasks to perform and difficulty following through with these regimes are ideal candidates beyond the SB population.

5.5 USABILITY TESTING OF THE PORTAL FROM THE CLINICIANS' PERSPECTIVE

While we have sought extensive feedback from persons with SB regarding their experiences in utilizing the smartphone apps, we have not tested the usability of the portal from clinicians' perspectives. In order for clinicians to effectively utilize the iMHere system as part of their existing workflow this additional clinician usability testing will need to be conducted. The iMHere system has the potential to not only improve the patients' quality of care and adherence to recommendations, but also increase efficiency of healthcare personnel, but likely requires a number of revisions and improvements to the clinician portal to begin to develop into a viable commercial product.

5.6 GAMIFICATION

Gamification is ""The integration of the mechanics that make games fun and absorbing into non-game platforms and experiences in order to improve engagement and participation" (Deterding et al. 2011a, p.10). After time, utilizing any system can become less engaging. Gamification may allow for on-going novelty and reinforcement for use of the system and completion of self-care tasks to maintain health and wellness. This may appeal to all ages of individuals; however, it may have increased appeal to a younger population who grew up with on-line gaming as a recreational and social outlet. Behavioral changes can be elicited through gamification of tasks related to maintaining health and wellness using principles of partial reinforcement. Gamification is a very new area of research and development when it comes to

APPENDIX A.

COMMONLY PRESCRIBED MEDICATIONS FOR PERSONS WITH SPINA BIFIDA

Purpose of Rx	Brand / Trade Name	Generic Name
Urologic Care / Bladder Management	Detrol	tolterodone
	Detrol LA	tolterodone XR
	Ditropan	oxybutynin
	Ditropan	oxybutynin
	Ditropan XL	oxybutynin XR
	Toviaz	fesoterodine
	Vesicare	solifenacin
	Enablex	darifenacin
	Sanctura XR	tropsium XR
	Sanctura	tropsium
Bowel Management: Laxatives, Stool Softeners, Fiber supplements	Colace	bisacodyl
	MiraLax	senna
	Kristalose	docusate sodium
	Fibercon	lactulose polycarbophil

Purpose of Rx	Brand / Trade Name	Generic Name
Psychiatric: Depression, Anxiety, Mood Stabilizers	Paxil Paxil CR Zoloft Wellbutrin Wellbutrin XL Wellbutrin Wellbutrin SR Lexapro Celexa Effexor Effexor XR	paroxetine paroxetine XR Sertraline bupropion HCl bupropion HCl XR bupropion HCl bupropion HCl XR escitalopram citalopram venlafaxine venlafaxine XR
Antibiotics – Infection	Bactrim Cipro Augmentin Keflex Levaquin Moxatag Amoxil	trimethoprim sulfamethoxazole ciprofloxacin amoxicillin/clavulanate cephalexin levofloxacin amoxicillin

Purpose of Rx	Brand / Trade Name	Generic Name
Pain Relief	Ultram	tramadol
	Motrin	ibuprofen
	Aleve	naprosyn sodium
	Advil	ibuprofen
	Oxycontin	oxycodone extended release
	n/a	oxycodone
	Percocet	acetaminophen/oxycodone
	Vicodin	acetaminophen/hydrocodone
	Tylenol	acetaminophen
	OxyIR	oxycodone immediate release
	OxyFast	oxycodone
	Neurontin	gabapentin
	Lyrica	pregabalin
	Elavil	amitriptyline

APPENDIX B.

ADAPTED MACARTHUR COMPETENCE TOOL

Participant Initials: _____ Interviewer: _____ Date: _____

Please rate the participant's answers to questions in the below 5 areas using the following scale:

0 = inadequate understanding 1 = partial understanding 2 = adequate understanding

1. Purpose of the Project Rating: 0 1 2

   What is the purpose of the research project that I described to you?

2. Activities involved in Participation Rating: 0 1 2

   How many study visits are you asked to participate in?
   How long will your participation in this research be if you decide to stay until the end?

3. Benefits of participation Rating: 0 1 2

   In what way may you benefit by volunteering to participate in this study?
   What might health care workers learn if people decide to be in this research project?

4. Risks/Discomforts of participation Rating: 0 1 2

   Can you tell me about the possible risks of participating in this project?

5. Ability to withdraw Rating: 0 1 2

   What will you do if you decide that you no longer want to participate in this study?

   Total Score: _____ (Total score must be 8 or higher to participate in research.)

APPENDIX C.

MODIFIED IBM POST-STUDY QUESTIONNAIRE (PSSUQ)

Usability Evaluation Form (Modified PSSUQ) **Subject: _____**

Smartphone Application ("App"):_____ **Date: _____**

1. Overall, I am satisfied with how easy it is to use this smartphone app.

	1	2	3	4	5	6	7	
Strongly Agree	□	□	□	□	□	□	□	Strongly Disagree

□ Check for not-applicable

Additional Comments:

2. It was simple to use this app

	1	2	3	4	5	6	7	
Strongly Agree	□	□	□	□	□	□	□	Strongly Disagree

□ Check for not-applicable

Additional Comments:

3. I could quickly utilize this app as part of my daily routine.

	1	2	3	4	5	6	7	
Strongly Agree	□	□	□	□	□	□	□	Strongly Disagree

□ Check for not-applicable

Additional Comments:

Subject: 1
Date:
App tested:

4. I was able to easily enter information using this app.

	1	2	3	4	5	6	7	
Strongly Agree	□	□	□	□	□	□	□	Strongly Disagree

□ Check for not-applicable

Additional Comments:

5. I felt comfortable using this app.

	1	2	3	4	5	6	7	
Strongly Agree	□	□	□	□	□	□	□	Strongly Disagree

□ Check for not-applicable

Additional Comments:

6. It was easy to learn to use this app.

	1	2	3	4	5	6	7	
Strongly Agree	□	□	□	□	□	□	□	Strongly Disagree

□ Check for not-applicable

Additional Comments:

7. Whenever I made a mistake using the app, I could recover easily and quickly.

	1	2	3	4	5	6	7	
Strongly Agree	☐	☐	☐	☐	☐	☐	☐	Strongly Disagree

☐ Check for not-applicable

Additional Comments:

8. It was easy to revise or change information I entered into the app.

	1	2	3	4	5	6	7	
Strongly Agree	☐	☐	☐	☐	☐	☐	☐	Strongly Disagree

☐ Check for not-applicable

Additional Comments:

9. The organization of information in the app was clear.

	1	2	3	4	5	6	7	
Strongly Agree	☐	☐	☐	☐	☐	☐	☐	Strongly Disagree

☐ Check for not-applicable

Additional Comments:

10. I liked using the interface of the app.

	1	2	3	4	5	6	7	
Strongly Agree	□	□	□	□	□	□	□	Strongly Disagree

□ Check for not-applicable

Additional Comments:

11. This app has all the functions and capabilities I expect it to have.

	1	2	3	4	5	6	7	
Strongly Agree	□	□	□	□	□	□	□	Strongly Disagree

□ Check for not-applicable

Additional Comments:

12. Overall, I am satisfied with this app.

	1	2	3	4	5	6	7	
Strongly Agree	□	□	□	□	□	□	□	Strongly Disagree

□ Check for not-applicable

Additional Comments:

Other comments:

Subject: 4
Date:
App tested:

APPENDIX D.

ADDENDUM 1: PHASE-II CONSENT FORM

Smartphone device not to be utilized for unlawful purposes

If I agree to take part in this research study, can I be removed from the study without my consent?

It is possible that you may be removed from the research study by the researchers if, for example, your status would change to prevent you from meeting the inclusion/exclusion criteria for the study. For persons enrolled in the intervention group, inappropriate use of the smartphone device and service package (voice, text/picture messaging, and internet access) provided to you may result in removal from the study. This includes, but is not limited to:

- Intentionally or recklessly abusing or misusing the smartphone and service to cause damage or system interruptions.
- Lending the smartphone device to any other person for use. Use of the device and service is limited only to the person who is enrolled in the study.
- Using electronic media to harass or threaten other persons, or to display, design, copy, store, draw, print or publish obscene language or graphics
- Using the device and related service to gain or attempt to gain unauthorized access to computing resources i.e. "computer hacking."
- Intercepting or attempting to intercept or otherwise monitor any communications not explicitly intended for him or her without authorization.
- Making, distributing and/or using unauthorized duplicates of copyrighted material, including software applications, proprietary data, and information technology resources. This includes sharing of entertainment files (e.g. music, movies, video games) files in violation of copyright laws.

_____ _____

Participant's Signature Printed Name of Participant

Date: _____

APPENDIX E.

ADDENDUM 2: PHASE-II CONSENT FORM

NON-EMERGENCY MEDICAL NEEDS

If I am enrolled in the intervention group using the smartphone and apps to communicate health and wellness needs, I understand that it does not replace the need for urgent or emergency care. The system does not replace traditional medical care. In the event of an urgent or emergency health situation, I would need to seek care from community resources such as emergency room treatment, crisis phone lines, or calling 9-1-1.

_____ _____

Participant's Signature Printed Name of Participant

Date: _____

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