IMPLEMENTING THE BIOPSYCHOSOCIAL APPROACH OF THE ICF-CY TO PREDICT SECONDARY CONDITIONS AND QUALITY OF LIFE IN INDIVIDUALS WITH SPINA BIFIDA

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ABSTRACT

FIKRIYE EDA KARACUL: Implementing The Biopsychosocial Approach Of The ICF-CY To Predict Secondary Conditions And Quality Of Life In Individuals With Spina Bifida
(Under the direction of Rune J. Simeonsson)

Spina Bifida (SB) is a complex neurogenetic disorder, diagnosed before or at birth that is caused by the defective closure of the backbone and the spinal cord (Antolovich &Wray, 2008). The condition is associated with many physical, or mental health conditions at varying severity levels. Additional health conditions that occur as a result of having a primary disabling condition are considered as secondary conditions.

The present study examined the nature of secondary conditions; the individual characteristics associated with their prediction and quality of life (QoL) and used ICF –CY (WHO, 2007) as a frame of reference to discuss their functioning. The findings suggested several significant small and medium strength relations among variables. After reaching a consensus between raters on linkage of the Secondary Conditions with ICF –CY codes, multiple hierarchical linear regression analyses were used to assess the ability of developmental and family characteristics to predict difficulty with body functions, activities participation and perceived QoL. The findings affirmed the overall predictor quality of the given characteristics, and direction of significant predictions, however there was variability of predictors for each criterion. Practical implications for school psychology are provided.

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LIST OF ABBREVIATIONS

Spina Bifida –SB

Secondary Conditions –SC

Survey of Family Characteristics –SFC

Quality of Life –QoL

World Health Organization –WHO

International Classification of Functioning –ICF

International Classification of Functioning for Children and Youth – ICF–CY

Body Functions – b

Body Structures –s

Activities and Participation – d

CHAPTER I –INTRODUCTION

Statement of the Problem

Spina Bifida (SB) is a complex neurogenetic disorder, diagnosed before or at birth that is caused by the defective closure of the backbone and the spinal cord (Antolovich &Wray, 2008). The condition is associated with many limitations at varying severity levels such as congenital malformations of the brain, hydrocephalus, Chiari II malformation, spinal curvatures or humps, neurogenic bladder, or muscle weaknesses. Additional physical or mental health conditions that occur as a result of having a primary disabling condition (spina bifida) are considered as secondary conditions. Recent research however has emphasized the need to measure secondary conditions and promote the quality of life of children with spina bifida.

Secondary conditions can be in the form of a variety of physiological, behavioral and psychological manifestations. At the physiological level, they may include primary health conditions such as pressure sores or scoliosis as well as mobility limitations, sleep disturbance, urologic infections, and pressure ulcers (Antolovich & Wray, 2008; Adriaansen, et al., 2012). It may be also expressed in eating disorders, self-injurious behaviors, learning disorders, and limitation in self-care. Of particular significance are behavioral and psychological, and cognitive manifestations related to participation, school integration, self-esteem, anxiety, and social isolation (Adriananansen, et al., 2012, Simeonsson, et al., 2002; Krahn, et al., 2009). Secondary conditions, and functional abilities greatly impact the individual’s and their caregiver’s quality of life through limitations in physical, academic, personal and social functioning, despite, limited research on this phenomenon in individuals with spina bifida (Guilcher et al., 2012; Simeonsson et al., 2002; Adriaansen, et al., 2012). Thus, understanding the relationships between psychological, social and physical functioning and environment would improve engagement of children with SB and their life fulfillment.

The purpose of this research is to investigate the nature of secondary conditions, the individual characteristics associated with their prediction and quality of life (QoL) and to use ICF-CY (WHO, 2007) as a frame of reference to discuss their functioning. Growing up with spina bifida can be challenging due to complex and demanding nature of the condition. The multifaceted nature of the physical and developmental problems of spina bifida makes it difficult to predict identification of the problems that are of secondary conditions. Therefore, a comprehensive holistic framework is needed for identifying key factors associated with functioning and development of children with SB. The biopsychosocial model of the ICF-CY provides a framework relating functioning characteristics at the body, person and societal domain, and the mediating role of the environment on such functioning (WHO ICF-CY, 2007).

There is minimal research that evaluates secondary conditions from a multidimensional perspective in individuals with SB. To date, much of the literature has focused on medical aspects of their activities and participation and functioning, and most known studies used spinal cord injury or orthopedic disability groups and included very few individuals with SB. Furthermore, there are no known studies that examine the underlying influences of secondary conditions and their dynamic relations by linking with ICF-CY. With the availability of the ICY-CY, there are growing efforts to define, not only the medical condition but also to describe the characteristics and well-being of the developing child and the influence of environment using a universal, standard language. Thus, this study attempts to measure the health and functioning of individuals with spina bifida using the ICF-CY as a frame of reference and items from secondary conditions questionnaire as a measurement tool. Linking the secondary conditions questionnaire to the ICF-CY provides information on the scope of the instrument; helps to estimate the characteristics of spina bifida with a standard language.

In sum, the nature of living with SB is not well researched; hence learning about the adjustment of the individuals with spina bifida and how they perceived the associated secondary deficits needs to be investigated. The current study describes and discusses health and functioning in individuals with spina bifida based on the results of the linkage between the secondary conditions and ICF-CY classification. It is expected that this study contributes to the description of risk and resilience factors to promote the quality of life and prevent secondary conditions in children and youth with spina bifida (Simeonsson & Leskinen, 1999).

Literature Review

Development of the Child with Spina Bifida

The development of children with SB is affected by primary as well as secondary conditions. Spina Bifida (SB) is a complex neural tube condition diagnosed before or at birth that is caused by the defective closure of the backbone and the spinal cord (Antolovich &Wray, 2008). It is the most common permanently disabling birth defect occurring in 1to 2 of every 1000 live births in the world and 0.3 to 0.5 of 1000 live births in U.S. (CDC, 2015). Over the years, the prognosis, survival rate, cognitive functioning and quality of life has changed with use of different treatments such as an implantable one-way shunt to hydrocephalus, use of clean intermittent catherization, prenatal surgical correction of myelomeningocele, promotion of folic acid consumption and improvements in prenatal testing (Zabel et al., 2011).

Spina bifida is classified into three types as “Spina bifida Occulta, Meningocele, and Myelomeningocele”. Spina Bifida Occulta is the “closed” form, Spina Bifida Meningocele, and Myelomeningocele are classified as “open” neural tube defects (CDC, 2015). Spina bifida occulta (hidden) is the mildest form caused by defective formation of backbones. It is the most common type, usually lacks physical sign or any complication so many individuals do not realize it by appearance. In some instances, the malformation may cause incomplete paralysis with urinary and bowel dysfunction and/or marked by malformations of fat, bone, or meninges (NINDS, 2015). In the spina bifida meningocele, spinal fluid and meninges protrude through an abnormal vertebral opening; the malformation contains no neural elements and may or may not be covered by a layer of skin with little or no nerve damage. Some individuals with meningocele may have few or no symptoms while others may experience such symptoms as paralysis with bladder and bowel dysfunction. Spina bifida myelomeningocele (SBM) is the most severe form, characterized by a lesion in which the spinal cord and nerves poke through the open part of the spine in a sack, that creates a barrier for the transmission of motor and sensory information at and below the lesion level. The severity of the spina bifida depends on the spinal level of the lesion (thoracic, lumbar, sacral) and shunting (Lomax-Bream et al., 2007) such that, higher lesions cause more significant nervous system damage than lower lesions (Rintoul et al., 2002). Hydrocephalus (fluid on the brain) as caused by Arnold-Chiari- II malformation (malformations of the hindbrain) is the most common neurological complication among children with SBM with 80% rate (Zabel, et al., 2011). Hydrocephalus has effects on the orthopedic, cognitive, behavioral and communication development (Tuminello, 2012).

Spina bifida does not have a single defining characteristic, and represents a complex multifactorial neurodevelopmental disorder. Some of the common sensory, motor or medical complications are difficulties with bladder and bowel control, ambulation difficulties, and skin infections due to pressure sores or burns, faecal and urinary incontinence, spasticity, contractures, deformity, scoliosis, epilepsy, constipation, renal impairment, hypertension, obesity, impairment of sexual function, chronic pain. In addition to the aforementioned conditions they experience cognitive impairments, communicational or psychosocial complications such as issues with school integration, socialization, psychological, social and family issues that affect their daily life (Antolovich & Wray, 2008; Brislin, 2008).

Secondary Conditions

Individuals with disabilities are at risk for “secondary conditions,” preventable health issues influenced directly or indirectly by the characteristics of their primary conditions (Adriaansen et al., 2012). Secondary conditions are physical, mental, and social disorders that "occur as a result of a primary condition and can either be a pathology, an impairment, a functional limitation or an additional disability" (Pope & Tarlov, 1991, p. 214). The primary deficit can only be a risk factor, however is not enough to account for the expression of the secondary condition (Simeonsson & Leskinen, 1999, p. 53). They are not specific to people with disabilities, but more prevalent in individuals with disabilities (Rimmer et al., 2011). The difference between personal and environmental interaction and developmental stages account for variations in the nature and extent of expressed secondary conditions (Simesonsson, McMillen & Huntington, 2002). Secondary conditions greatly impact the child's quality of life through limitations in physical, academic, personal and social functioning and even require frequent re-hospitalizations in some cases (Guilcher et al., 2012; Simeonsson et al., 2002).

It has been well documented that youth with spina bifida experience difficulty with executive functioning, cognitive and language skills that are important for managing social and academic performance (Lindquist et al., 2008). Their cognitive performance is "complex, dynamic, and at times unstable" (Zabel et al., 2011). Poor performance in cognitive abilities includes abstraction, sequencing, planning, problem solving, use of social inference, use of contextual language, visual-motor and spatial skills, mathematical performance, and attentional tasks (Lomax-Bream, et al., 2007). Executive functioning and social language difficulties in the early years cause school age issues and social problem solving difficulties that impact through adulthood (Landry et al., 2013). Learning difficulties in preschool children with SB has a strong association with slower cognitive growth (Lomax-Bream, et al., 2007). As an indicator of cognitive abilities, fine motor skills and self-perception, preschool children were asked to draw self-portraits. The spina bifida group drew less complicated figures, with fewer body parts and rated themselves as significantly different on physical and cognitive competence than their typically developing peers (Mobley et, al., 1996).

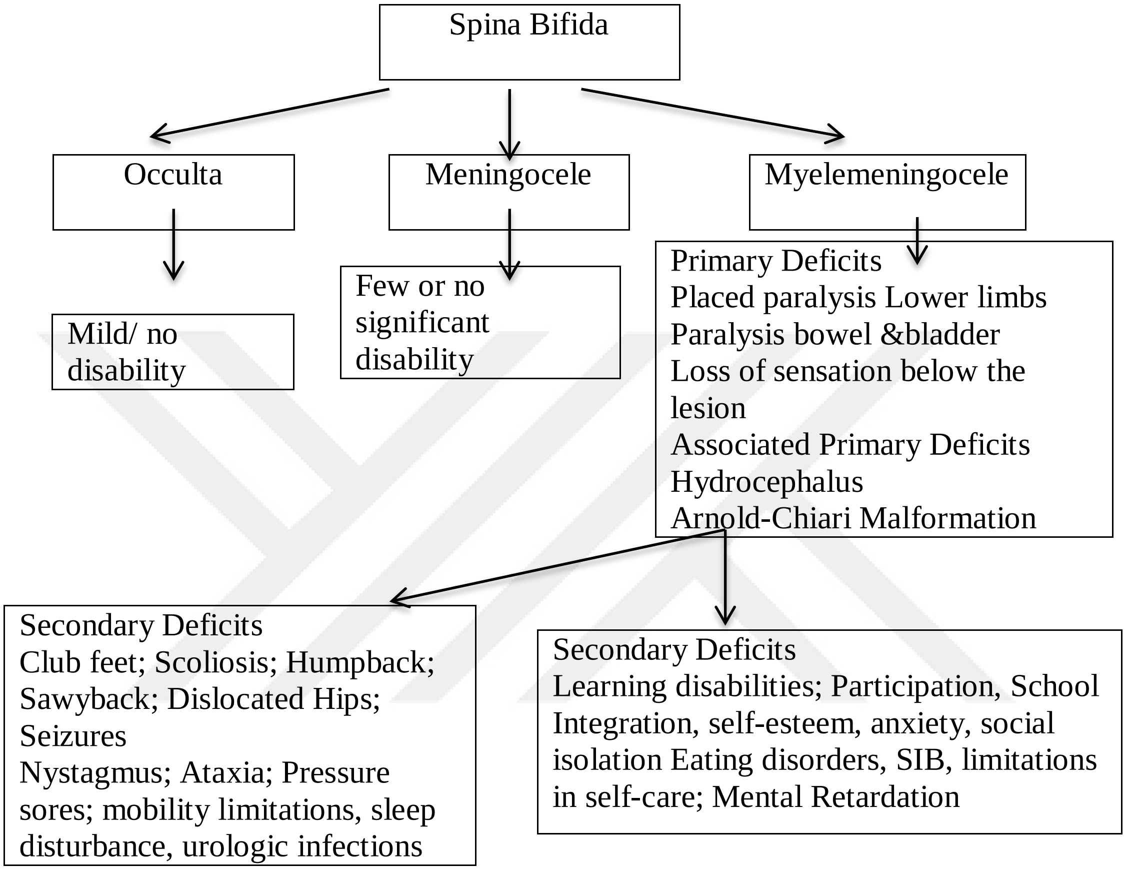


Figure 1. Disabilities associated with Spina bifida (Adopted and modified from Singh, D. K., 2003)

A common strength for children with SB frequently reported to be on verbal measures and rote memory abilities as compared to their performance IQ (Barnes, et al., 2014). Spina bifida presents mixed difficulties that can be presented in verbal or nonverbal abilities. In some individuals, the verbal deficits are often described as “cocktail party chatter syndrome”, in which the production of meaningful speech might be relatively reduced (Zabel et al., 2011).

Among academic skills, they are known to demonstrate better reading skills than math and about half of children with SB are diagnosed with a math disability (Barnes, et al., 2014).

Math related cognitive abilities are defined as phonological awareness, working memory and fine motor/ finger skills (English et al., 2009). Challenges on nonverbal measures (perceptual and motor tasks), reading comprehension, math and writing are exhibited as young as age three and continue through adulthood (Barnes et al., 2014). Children with spina bifida may not necessarily have skill deficits in all those areas, however their abilities may develop later than their TD peers during preschool years.

In a longitudinal study, Barnes, et al. (2014) tested the differences in the impact of math-related cognitive abilities on children with SB in comparison to TD peers, on their academic abilities and school related outcomes. They used cognitive abilities of children at 36 and 60 months of age and school related outcomes at 8.5 and 9.5 years of age. While visual spatial working memory effect was specific to mathematics, phonological awareness impacts both math and reading scores at 8.5 and 9.5 years of age. Also difficulties with staying on task, task initiation, time management, planning, organization, perception and orientation, motor control, visual perception are reported that is similar to children with ADHD (Janeslätt, et al., 2014; Burmeister, et al., 2005).

Children and youth with SB who have more difficulty on tasks assessing mental flexibility and switching as an indication of their executive functions, are predicted to have less autonomy, lower intrinsic motivation and higher parent involvement suggesting a relation between the three constructs (Tuminello et al., 2012). Social competence (social adjustment, social performance, and social skills) and executive function deficits also present increased risk for internalizing symptoms (Lennon et al., 2014). Positive, supportive social experiences with peers and parents are considered as a catalyzer for unpleasant health care rituals; however, lack of positive social support puts them at risk for depressive symptoms (Essner &Holmbeck, 2010).

Compared to TD adolescents, lack of positive peer and school contexts makes youth with SB more susceptible to internalizing disorders, particularly depression (Friedman et al., 2004; Essner & Holmbeck, 2010). Support, acceptance, and adaptive functioning in the family and school contexts is a stronger preventative condition for psychological risks (Holmbeck&Devine, 2010).

Their limitation in movement, communication, and challenges with self-care tasks becomes a barrier for developing positive social and environmental interactions. To function independently, individuals should be able to self-manage medical, physical and psychological consequences of spina bifida. Reducing the risk of secondary conditions while increasing self-efficacy would improve the likelihood that young people will function independently throughout their lifespan. Due to the impact of functional limitations, family related issues have higher impact during childhood years, but as they grow older, the areas of concern include peer relations followed (Stubberud et al., 2013).

Secondary conditions make it difficult to make and keep friends for youth with SB especially with TD peers. During adolescence, the experience of peer rejection would likely disturb other areas of functioning more severely. The individual would compare the self with the typically developing peers; they would feel a deep lack of worth, or confidence that is persistent through adulthood. Contrary to common research and beliefs, Zukerman et al. (2011) claim that children with SB are resilient in peer relations and by emerging adulthood they have similar quantity of peer friendships with TD peers. When the characteristics and quality of friendship in children with SB is compared to their TD peers and to youth with other chronic conditions, children with SB report the quality of proximity of their friends lower than that of other groups (Zukerman et al., 2011). They demonstrate the need of more social support and are more likely to identify “peers” as their best friends, despite limited amount of time spent together (Devine et al., 2012). Their feelings in a group of peers are different than when they are matched with a specific peer. In that case, youth with SB don't report feeling as close or as comfortable to others besides reporting lack of support (Devine et al., 2012). It is possible that lack of self-confidence and lower self-esteem would impact their perception of social environment (Brislin, 2008).

As individuals with spina bifida age their expectations and needs change and they are at higher risk for presenting lower self-esteem, confidence, body image, depression, poor social skills, isolation, exclusion, and poor relationships. It is well documented that these issues are presented as a reflection of their interaction with the environment and as secondary to their social difficulties. These emphasize the importance of participation in daily activities.

Quality of Life

Quality of Life is defined as "an individual's perception of their position in life in the context of the culture and value systems in which they live, and in relation to their goals, expectations, and concerns" (The WHOQOL Group, 1995, p.1405). Most individuals with myelomeningocele reported lower perceived Health Related Quality of Life (HRQoL) ratings when their participation levels were lower (Verhoef et al., 2007). Due to the complicated nature of the condition, there may be unique considerations to assess QoL (Sawin & Bellin, 2010).

Challenging expectations, being independent in mobility was found to be a higher predictor of QoL than other functional abilities such as being independent in self-care or being wheelchair dependent (Schoenmakers et al., 2005). Rendeli et al. (2005) used both parent and child reports to assess the health-related quality of life of children with SB, and reported continence problems as the main factor affecting QoL. Interestingly, higher disability predicted lower emotional problems, which is likely the result of how expectations change individual's mindset. An individual with less severe impairments may have a mental aspect of aiming to perform better, resulting in lower satisfaction of QoL. Despite both having similar physical impairments children and adolescents with SB reported lower “school and overall QoL” and participation in activities compared to those with spinal cord injury (Flanagan, Kelly, &Vogel, 2013). It is suggested that the presence of shunt may result in cognitive and executive functioning impairments, which have a chain effect on their school performance causing anxiety and self- consciousness, then their concentration (Flanagan, et al., 2013). Rofail et al. (2014) reviewed studies on HRQoL on individuals with SB, and reported the significance of low levels of QoL, and the need for a consistent communication intervention to support them. The qualitative part of this study also reflected the issues such as bullying, isolation, being hesitant in school activities.

The review of literature highlights the difficulties associated with Spina bifida and negative impact of those on their QoL reported by parents or the individuals. It is noteworthy that their perception had a key role on their QoL ratings regardless of who reported it. Studies also reflect the differences in parent and individual reports on which impairment affects their QoL. It is possible that parent characteristics would impact on their rating on the perception of the QoL of the individual with spina bifida. Despite the decrease in SB prevalence, they need to be understood and society should be more aware of their unmet medical and social needs to increase sensitivity and find better solutions for their behavioral and social challenges

Biopsychosocial Approach to Spina Bifida

Previous review demonstrates the complex nature of spina bifida and physical, social, psychological need for a comprehensive framework. Individuals with SB experience varying difficulties that is associated with activities and participation, body functions and structures and environmental factors as framed by International Classification of Functioning, Disability and Health: Children & Youth (ICF-CY). It is a widely-used framework for conceptualizing and measuring the nature and consequences of disability (WHO ICF-CY, 2007). It focuses on classification of the four aforementioned components that are specific to children and youth, whose age range from birth to 18 years old. ICF-CY is based on the "*biopsychosocial model*", which assesses children and youth functioning in the context of their stages of development and the environments in which they live and their interaction with each other (WHO ICF-CY, 2007). The "biopsychosocial model" presents the interactional role of biological, psychological, and social (socio-economical, socio-environmental, and cultural) factors, on human functioning.

Within this framework, *functioning* is explained as an "umbrella term that encompasses all body functions/structures, activities, and participation", whereas *disability* is defined as an "umbrella term for impairments, activity limitations and participation restrictions" (Gan et al., 2014, p. 25). ICY-CY is a function-focused model, and addresses the level of ability in functional areas, such as learning, mobility, communication, self-care, social relationships, and other similar characteristics. It encourages the development of interventions that targets at the development of individual' functioning in relation to their environment and personal conditions.

The ICF-CY functioning codes are grouped into *b- body functions* (i.e., physiological and psychological), *s- body structures* (i.e., anatomical parts such as organs, limbs and their components), *d- activities* (i.e., execution of a task or action), *and participation* is defined as "the involvement in life situations" such as learning, applying knowledge, self-care, communication, relationships or participation in school activities (WHO ICF-CY, 2007, p. 129). Contextual factors interact with the individual with a health condition and determine the level and extent of the individual’s functioning. *Environmental factors- e* is extrinsic to the individual (e.g. social support, relations, the attitudes of the society, the legal system). *Personal Factors* (e.g. gender, race, age, lifestyle, habits, coping styles) do not have categories within the ICF-CY, and open to the subjective view of the users (WHO ICF-CY, 2007).

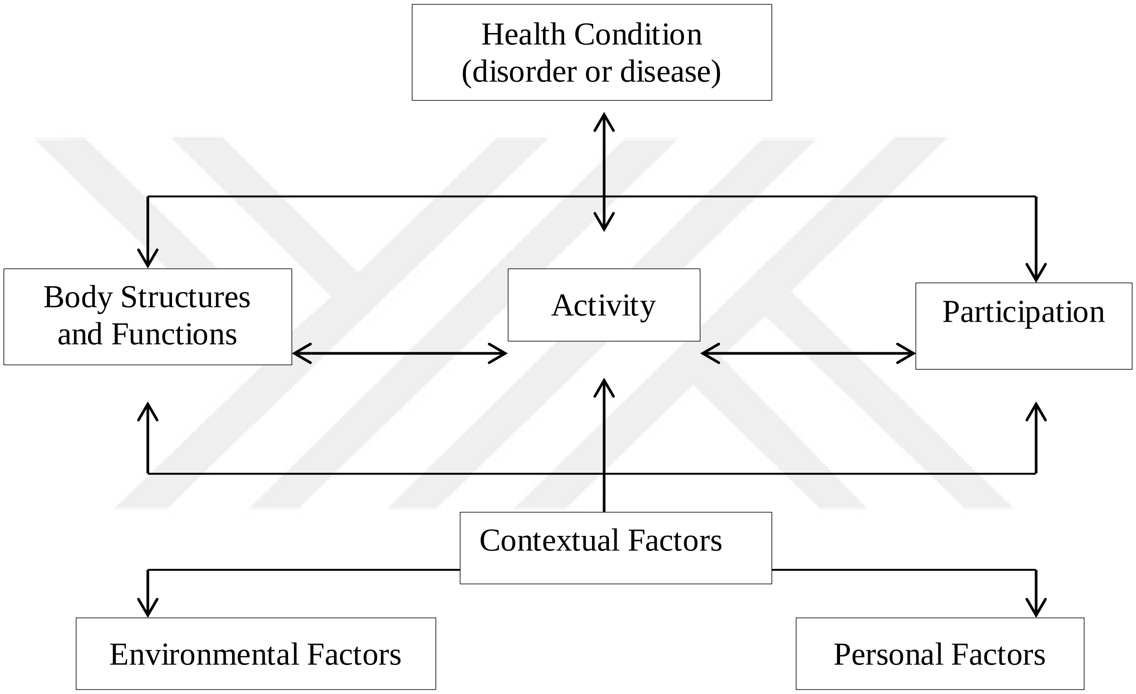


Figure 2. Interactions between the components of the ICF-CY Model

Each of the four domain is also divided into multiple chapters (1st level) than hierarchically listed categories (2nd level) such as (a) Mental functions; (b) Sensory functions and pain; (c) Voices and speech functions; etc. under the Body Functions domain. In level 3 and 4 the details of the categories gradually increase (Figure 4). Each of the components are quantified using a 0 to 4 scale that is defined as: 0- no problem; 1- mild problem (a problem is present less than 25% of the time); 2- moderate problem, (present less than 50% of the time); 3- severe problem, (present more than 50% of the time); and 4- complete problem, (present more than 95%

of the time).

ICF-CY has been increasing used as a standard for content comparison or examination of instruments, in order to select the most appropriate items or instruments or to report individuals functioning with a standard language for clinicians and researchers (Hwang et al., 2014; Krasuska et al., 2012; Sommer et al., 2015). Previous studies suggested the identification of reliable and valid instruments and mapping items based on linking rules such as the ones described by Cieza et al. (2004) (Ellignsen, K., 2011& Björck-Åkesson et al., 2010).

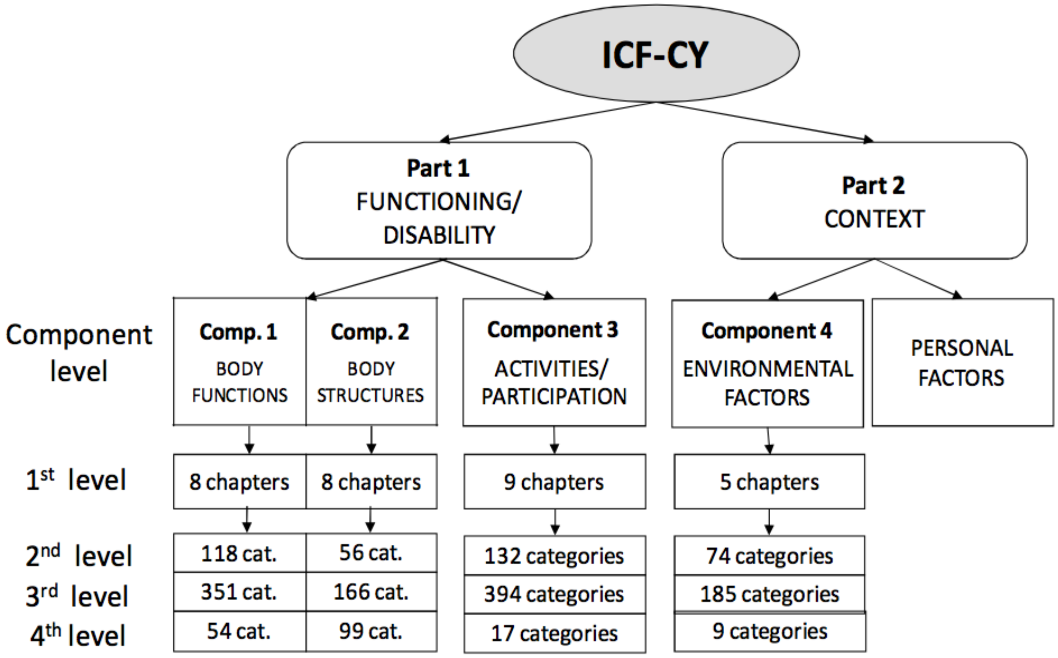


Figure 3. Hierarchically listed components of the ICF-CY (Pless &Granlund, 2011).

In the previous studies, linkage of ICF-CY codes is conducted on quality of life measures (Krasuska et al., 2012; Peterson et al., 2013; Sommer et al., 2015), environmental factors (Hwang et al., 2014), and on autism measures (Castro et al., 2013). Studies on linking the instruments with the ICF-CY claim to “raise awareness in clinicians and researchers about relevant domains from a well-being and functioning perspective” as well as making it easier to compare and better understand the domains of the instruments (Sommer et al., p. 439). Quantifying ICF-CY categories provide reliable and valid information about the functioning and well-being of the individual with health condition(s) (Krasuska, et al., 2012).

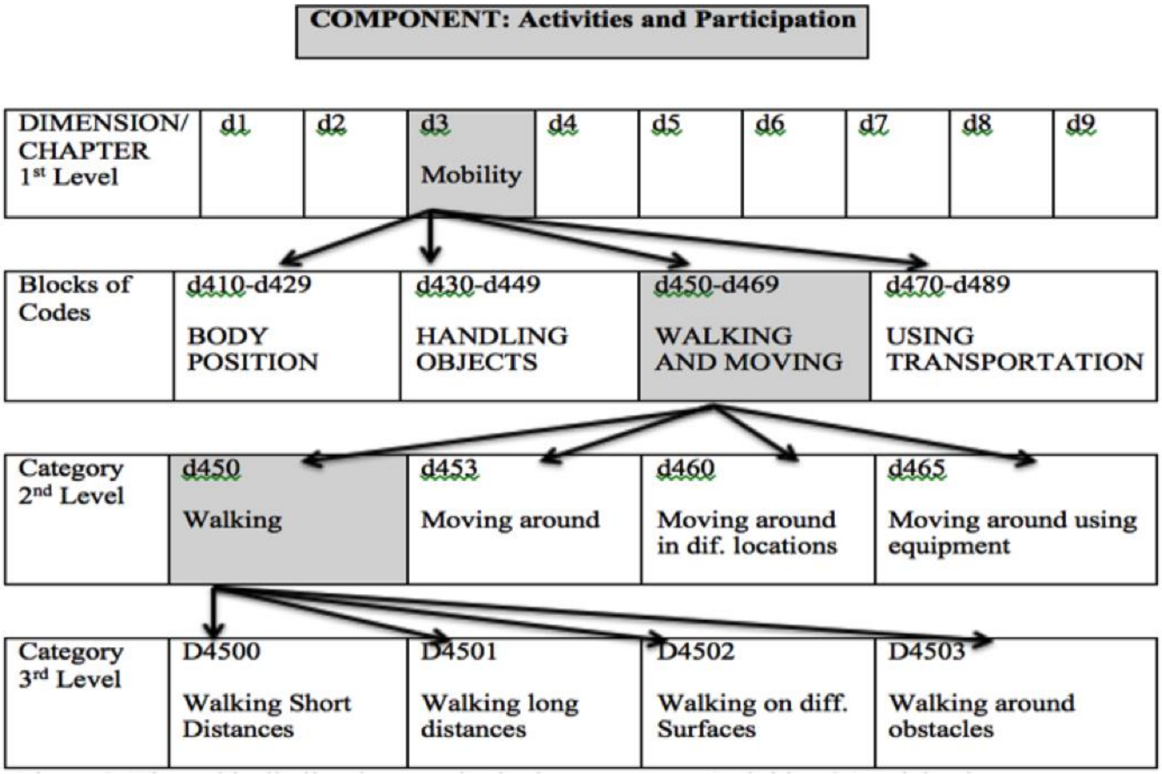


Figure 4. Activities and Participation Components

In the recent research ICF-CY is used as a framework to examine how quality of life measures are represented as well as providing a basis for a standard language for comparing and contrasting the nature and extent of the health conditions. Most studies used linking rules defined by Cieza et al. (2005), by first deriving meaningful concepts within the items, then linking those concepts with the ICF-CY by two raters. Castro et al. (2013), linked the ASD diagnostic measures the same way, to improve documentation of child functioning, inform interventions and identify functional characteristics of the tool. They also proved that linking existing measurements used for diagnosis, with the ICF-CY classification system could be possible. Through that study functional aspects of these tools were identified which in turn improved the documentation of child's functioning. Sommer et al. (2015), linked a health-related quality of life measure to the ICF-CY, which proved the scope and comparability of both the parent and child versions, provided a universal language and is expected to help to estimate the prevalence of the related disabilities. Krasuska et al. (2012) tried a different approach by applying linking process on a QoL measure and a disease specific module. Krasuska et al. (2012) followed the rules: "only items that were linked to one ICF/ICF-CY category was considered, and among the items confirming this requirement, the items that have been assigned to a particular ICF/ ICF-CY category with the higher inter-rater agreement were selected" (pg.78). As a result, health module presented content density, meaning more than one ICF-CY category was assigned to many items, and most important categories among the domains were recreation and leisure. The importance and need for considering the emotional aspect of functioning as well as the linking and validity studies on ICF-CY were emphasized.

A universal, standard language is needed to explain the health and well-being of individuals with spina bifida. The ICF-CY model is feasible and useful for research, clinical and policy purposes to develop effective interventions by taking all child and adolescent characteristics in consideration instead of focusing solely on the disability identifications. Each individual with or without a disability represent different strengths and limitations, hence having a complete picture of each individual with needs is of importance to improve their capacity and encourage independence in everyday activities. It also contributes to the description of risk and resilience factors for the identification and development of interventions to promote health and prevent secondary conditions in individuals with spina bifida.

This proposed study aimed to measure the health and functioning of children with spina bifida using the ICF-CY as a frame of reference and items from secondary conditions questionnaire as a measurement tool. Linking the secondary conditions questionnaire to the ICF-CY is provides information on the scope of the instrument; helps to estimate the characteristics of spina bifida. The implication of this study is not only to describe or classify the medical aspect of spina bifida, but also present how this can be applied to the secondary conditions. Thus, linking secondary conditions with ICF-CY would be an important contribution to understand these characteristics.

Participation: It is well established that participation has been linked with positive outcomes such as improved academic performance, better relationships and a more fulfilling life. However, individuals with myelomeningocele are reported to be hesitant to participate due to limitations of primary and secondary conditions. Given the difficulty of transition from childhood to adolescence, there are developmental differences in participation preferences of children and youth with SB in recreational, physical, social, skill-based, and self-improvement activities. In an assessment of age related differences, younger children participated more often in physical and skill-based activities than all other youth, and whereas older youth (12-18 years) participated less in recreational, physical, and skill-based activities Kelly et al. (2011). Other defining factors for improved participation were caregiver employment and shunt absence. Also more caregivers reported bladder and bowel needs as barriers to participation for children between the ages six to twelve.

Despite those findings, there is some disagreement among studies regarding the effect of secondary conditions on adolescents' participation in physical activities. Some studies claimed family support, athletic competence, and physical appearance correlated with physical activity, but not the level of ambulation (Buffard et al., 2009). The perception of restriction is independent from the level of severity, because each individual is in a different environment and their expectations increase with milder disability presentation (Barf et al., 2009). Some common areas of reported restriction were leisure activities, going to school or work and visiting family or friends, mostly caused by the lack of accessibility of buildings, or transportation and own emotional barriers, pain, or fatigue (Barf et al., 2009).

In a recent study, it was found that interventions that focus on environmental factors had significantly promising results for community involvement of children with physical disabilities (Law et al., 2015). In order to encourage participation of individuals with disabilities environmental factors such as familial (e.g. family functioning), physical (e.g. built environment, accessibility), physical task demands (i.e., balancing, getting on and off the bike), social (e.g. social and peer support), attitudinal (e.g. perceptions towards disability), institutional (e.g. policies and availability of programs), and cultural expectations (i.e., expectation to sit quietly during mass) are reported to be important (Law et al., 2006).

Contextual Factors: The outcomes of an interaction between spina bifida and environmental and personal factors form the concept of the disability. Contrary to peer relations, adolescents continue to have closer relationship with their parents. Parental overprotection is commonly mentioned as a meaningful reason of the behavioral and emotional dependence among children and youth with SB. Intrusive parents set lower expectations and get over concerned about their children’s abilities in tasks, causing lack of self-confidence, and self-reliability (McKernon et al., 2001). It is possible that executive function (EF) deficits among adolescents may be the reason of parent intrusiveness, or EF difficulties in children makes transitioning harder from being highly dependent on parents, to autonomous actions during adolescence (Tumineallo et al., 2012).

Mothers and fathers also differ in amount of parental fostering of independence between TD adolescents versus adolescents with spina bifida. Mother intrusiveness was consistent between both groups of adolescents; however, fathers were intrusive only to youth with SB, (Tuminello et al., 2012). Results on intrusiveness are consistent with the parental stress differences among mothers and fathers of preadolescents with SB (Friedman et al., 2004). There is considerable support for the assertion that parents of youth with chronic illnesses experience higher stress than parents of other groups (Gras et al., 2009). Even at lower levels, paternal stress nevertheless has a significant effect on child psychosocial adjustment along with marital functioning (Friedman et al., 2004). Despite greater levels of stress but families of children with a chronic condition were also highly resilient (Horton &Wallander, 2001). In order to develop resiliency, they use various coping methods that isolate the possibility of psychological maladaptation (Holmbeck et al., 2006; Gras et al., 2009). Among parents of children with SB, those coping methods include seeking social support, positive reappraisal as the most frequent style, whereas the internally focused thinking style, reflective and systematic thinking style and innovation-seeking were used the least (Gras et al., 2009).

The interplay of the factors effecting family stress and how families adapt and cope with those is well reflected in the Double ABCX Model of Stress and Adaptation by McCubbin and Patterson (1983). The Double ABCX Model describes the interplay between the pile-up of stressors (aA), such as a life event or transition impacting the whole family, the internal and external resources one has to address the stressor (bB), and the meaning of the stressor to the family (cC) and family adaptation (xX) (McCubbin and Patterson, 1983). When this model was used to examine the well-being and QoL of the families, behavior problems, and the severity as well as the support from the family and friends are found to be significantly related with the ability for adaptation and coping strategies (Sarria & Briosso, 2014). Several studies point that severity of the behavior and the deficits in social skills, or child-externalizing behaviors are more of a stressor than the disability itself (McStay, Trembath, and Dissanayake, 2014). Through the use of the same model perception towards stressors is reflected to be an identifying factor for child behaviors and (Saloviita, Italinna, & Leinonen, 2003).

There is not any methodologically valid, randomized, longitudinal and theory driven studies on the family functioning of children and youth with SB (Holmbeck et al., 2006). Most recent studies using this model focus on ASD, for example Paynter et al., (2013) found that the severity of the ASD behaviors, the support received from family and friends, their self-esteem, optimism, self-efficacy, social support, their perception of the stressor, family perception of the stressor and resilience in coping with the stressors were all linked with their adaption. Among those, social support was the most important resource to parents, however, families using active-avoidance style, is likely to increase the frequency or the severity of the challenging behavior, and this in turn increases family stress as well as building a barrier for reaching the social support (Paynter, Beamish, Davies, & Milford, 2013). Hope and social support contributes to the perceived stress by mothers as well as affecting each other. If a mother is satisfied with number of people supporting her as well as the quality of support, she presents higher hope that is defined as “the assessment of agency and pathways related to individuals’ goals” (Horton & Wallander, 2001; p.383). As families are satisfied with the social support, they had a better understanding of the disability and reported higher levels of benefit and meaning from the experience of having a child with disability that are all integral to coping with a child with a disability (Pakenham, Sofronoff, & Samios, 2004).

Being the parent of a child with a spina bifida has its own hardships, however there are no known studies on the family stress and coping of individuals with spina bifida based on Double ABCX Model that includes all stressor components in the same model of adaptation. Thus, the difficulties they face and the impact of those stressors needs to be investigated. It is also necessary to consider a reverse perspective, investigating the impact of family stressors on the child and adolescent with the disability. The studies up to date have not considered the impact of family stressors, (e.g. child problem behavior, severity, adaptive behavior, and pile-up of family characteristics) on the outcomes of the individual with disability. There is not yet sufficient information about the impact of family stress, or strength on the perceived quality of life of individual with the disabling condition. Furthermore, interventions to promote adaptive family functioning and better psychosocial outcomes are still a necessity with this population.

Limitations of Research on Spina Bifida

Historically, most of the research on SB has focused on the physical and neurocognitive domains, with less attention paid to the psychological and social domains of functioning (Holmbeck &Devine, 2010). Overall, studies reflect that children and youth with SB experience difficulties in developing peer relations due to restrictions in activities and participation, or deficits in social language. They are usually overly attached to their parents due to need of assistance in daily life. Similar issues have been the subjects of a few intervention studies. Outcomes of those studies suggest the need for variety of programs that can improve the psychosocial and academic outcomes for children, youth and their caregivers thus improving their participation in social life and enhance their quality of life.

There is minimal research for the development of independence and increased participation in children with SB (Friedman et al., 2004). Gender, parental support of autonomy achievement, and verbal abilities were only a few factors found that contributed to the differences in the autonomy trajectory of youth with spina bifida. In order to prevent secondary conditions or the contributing factors it is necessary to understand how they differ among variety of populations, besides developing assessment measures for secondary conditions (Simeonsson et al. 2002). More research is needed to find other possible factors contributing to autonomy development and also clarify the impact of specific family or social characteristics and how they mediate future outcomes of children with spina bifida (Friedman et al., 2004; Essner & Holmbeck, 2010).

Secondary conditions of the individuals with spina bifida plays a key role in their school, social, and family life, however there is no research that evaluates it from a multidimensional perspective. Based on biopsychosocial approach ICF-CY provides a framework to investigate various associated factors and characteristics. There is a need for research using a holistic framework and not only focusing on medical aspects of spina bifida but also use a standard language as defining the characteristics of those conditions. As recent research on ICF-CY suggests, providing a standard language for determining the nature and extent of spina bifida is needed (Sommer et al., 2015). A standard language would raise awareness and understanding across disciplines, and improve collaborative efforts.

Research Questions

1. What are the relationships among developmental characteristics, secondary conditions, and QoL of children & adults with Spina bifida?
2. Is there a difference in the distribution of secondary conditions within the domains of the ICF-CY model?
3. To what extent do developmental characteristics and family characteristics predict domains of the secondary conditions and QoL ratings in individuals with spina bifida?

   a. Do developmental (functional) characteristics of the individuals as measured by Abilities Index and Demographics Questionnaire significantly predict secondary condition domains and higher QoL ratings?

   b. Do developmental (psychosocial) characteristics of the individuals as measured by Harter's perceived competence scales significantly predict secondary conditions domains and QoL ratings over and above developmental characteristics?

   c. Do family characteristics (strength) of the individuals significantly predict the secondary conditions domains ratings and QoL ratings over and above developmental and psychosocial characteristics?

CHAPTER II –METHODOLOGY

To address research questions of this study a quantitative research design was used. Data for this study was drawn from the longitudinal research project named Spina Bifida: Surveillance of Secondary Conditions and Quality of Life (Simeonsson, McMillen, Huntington, 2002). Details about the participants, measures and data analysis are detailed below.

Participants

This study was conducted as a secondary data analysis on a de-identified data set of approximately 100 participants and their families. At the time of the original study participants were recruited through the University of North Carolina Hospitals' Physical Medicine and Rehabilitation Clinic, the Lenox Baker Children's Hospital Myelomeningocele Clinic at Duke University, and the North Carolina Spina Bifida Association. Data were collected from individuals, family members, or caregivers and/or teachers. The participants varied in levels of impairments and cognitive abilities. For this study, data for children, adolescents and adults were analyzed.

Instrumentation

A variety of measures were administered to the individuals with spina bifida and their primary caregivers, or family members in this study. The specific measures administered varied on the basis of the age of the individual, a standard battery of measures was given to all subjects regardless of the individual's age. The data analyzed in this study were drawn from the measures described below (see Table 1).

Demographic and Medical History Questionnaire. This questionnaire includes individual’s age, bladder and bowel methods, continence, and independence as well as mobility status to describe the demographic characteristics of the children in the study. Information was obtained from the subjects’ medical records and from parent or primary caregiver responses.

ABILITIES Index. The ABILITIES index (Simeonsson & Bailey, 1991) provides a profile of an individual's functional abilities across nine domains (ABILITIES), including Audition (i.e., hearing left and right ear), Behavior and social skills, Intellectual function, Limbs (i.e., right and left hand, arm, and leg), Intentional communication (i.e., understanding and communication with others), Tonicity (i.e., tightness and looseness), Integrity of physical health, Eyes (i.e., vision right and left eye), and Structural status. The ABILITIES Index rating scale consists of 19 items related to the nine domains. Each item is scored on a 6-point likert scale ranging from (0) normal ability to (5) profound lack of ability by primary caregivers or parents asked 40 teachers were to complete the index a second time, an average of 34 days after the first rating for the stability of ratings. The inter-rater agreement for 130 parent-teacher pairs was 68.5%; for 130 parent-specialist pairs was 65.5%; and for 125 teacher-specialist pairs was 67.8% (Bailey, et al., 1993).

Secondary Conditions Questionnaire: This questionnaire (Simeonsson, 1995) manifestation of secondary conditions by an individual with primary condition in the physical, academic, personal, and social domains. The scale captures the intensity of secondary condition problems on a 51 item, likert rating scale ranging from 0 (never a problem) to 5 (always a problem). Reliability estimates for the four subscales ranged from .76 to .87 using Cronbach’s alpha as an index of internal consistency (McMillen, 1997). In this study only the first item was utilized.

Quality of Life Rating. The Quality of Life Rating (Simeonsson, 1995) is part of the Secondary Conditions Questionnaire with the parent or primary caregiver providing ratings of the child’s quality of life on a 5 –point scale (1 –Poor; 2 –Fair; 3 –Good/Very Good; 4 –Excellent).

HARTER Perceived Competence Scales: There are four different Harter Scales targeting different age groups beginning with the Pre-K scale to the Adulthood scale. Each of these scales consists of many subscales, however for the purpose of this study only means of scales that reflect Physical Competence, Social Competence and Cognitive Competence subscale means are used.

- Pictorial Scale of Perceived Competence and Social Acceptance for Young Children (PSPCSC) (Harter, S. & Pike, R., 1984): This instrument is designed for children of Preschool and Kindergarten age. It taps four specific self-concept domains: Cognitive Competence, Peer Acceptance, Physical Competence, Maternal Acceptance. There are a total of 24 items, 6 for each subscale. Higher scores reflect a greater sense of competence or social acceptance. Alpha coefficients for the subscales ranged from .53 for Physical Competence in the Grades 1-2 sample to .83 for the Maternal Acceptance subscale in the Preschool-Kindergarten sample (Harter & Pike, 1984).
- HARTER’S Perceived Self-Competence Scale for Children (Harter, 1982): This 28 item instrument taps into three competence domains, Cognitive, Social and Physical as well as a subscale measuring general feelings of Self-worth. It is designed for children over age 8 and has also been adapted specifically for adolescents. The subscale reliabilities range from .75 to .86 (Harter, 1982).

- HARTER’S Perceive Self-Competence Scale for Teens (Harter, 1988): This instrument taps nine specific self-concept domains: Scholastic Competence, Athletic Competence, Social Competence, Physical Appearance, Job Competence, Romantic Appeal, Close Friendship and Behavioral Conduct. In addition, a separate, subscale, taps Global Self-Worth. There are a total of 45 items, five for each subscale. The same subscales with the children version of the scale are utilized for the purposes of this study. The internal Cronbach alpha internal consistency values ranged from .58 to .91 (Harter, 2012).
- HARTER’S Perceive Self-Competence Scale for Adults (Messer & Harter, 1986): This 50 item scale taps twelve specific domains: Intelligence, Job Competence, Athletic Competence, Physical Appearance, Sociability, Close Friendship, Intimate Relationships, Morality, Sense of Humor, Nurturance, Household Management, and Adequacy as a Provider. A thirteenth subscale taps Global Self-Worth with six items. The internal consistency reliabilities ranged from .63 to .92 (Messer &Harter, 2012).

The Survey of My Family Characteristics. In this instrument (Simeonsson, 1987), parents/primary caregivers report characteristic of their family on a five-point scale ranging from 0 (not at all like my family) to 4 (very much like my family) among 35 items. The scale represents family strength. Internal reliability of the survey was found to be .93 using Cronbach’s alpha as an index (McMillen, 1997).

Procedures

Following the submission of the study and approval by the IRB all the data was examined for missing values and cleaned. For the missing values, initially raw data files were checked and values were inputted when available. When values were available, pairwise deletion was used during the analysis procedures. The Statistical Package for the Social Sciences (SPSS) version 24.0 was used for the data analysis. Summary of the variables analyzed in the study are presented at Table 1.

Table 1. Summary of Variables

Variables	Measure	Scale Type	Subscales
Developmental Characteristics *(Demographic Ch.)*	Demographic and Medical History Questionnaire		Bladder control Bowel control Mobility status Gender Age
Developmental Characteristics *(Psychosocial Ch.)*	HARTERs perceived self-competence scale- Pre-K, Child, Teen, Adult versions	- Likert scale 1 to 4 - Individual ratings	Subscales: Cognitive Social Physical
Developmental Characteristics *(Functional Characteristics)*	ABILITIES index	- Likert scale 0 to 5 Normal to Profound - Parent ratings	-Audition (Hearing) -Behavior& Social skills -Intellectual Functioning -LIMBS -Intentional Communication -Tonicity -Integrity of Physical Health -Eyes (Vision) -Structural Status
Family Characteristics	Survey of My Family Characteristics	- Mean total score - Likert scale 0-4 - Parent ratings - 35 items	
Secondary Conditions	Secondary Conditions Scale	- Likert scale (0-5)	Subscales: Physical Academic Personal Social
Quality of Life	QoL item of the Secondary Conditions Scale	- Parent rating	Likert scale (1-4) 1 poor 2 fair 3 good/ very good 4 excellent

Research Design and Data Analysis

The datasets for each measure was initially on different .sav and excel files; therefore the initial step was to combine the files on a single dataset. In the data folder, there are many data files for each scale so the ones with the highest number of participants were chosen. First an excel sheet was formed with all the scales and participants which were then copied into an SPSS file. The Demographics file did not include the variables proposed for analysis so they were obtained (bladder and bowel control, mobility) from raw files at Frank Porter Graham Child Development Institute. Some family numbers had more than one entry for each questionnaire, given collection at different dates. Only the initial date entries were kept for each family number (participant) and the rest was not included. As there were more than one "family number" entries, only the cases with the initial dates are used. For example, there were entries for years 97, 99, and 00 for Family #608 so the variables for year 97 were used thereby limiting potential confusion. Also, when cases such as child#102 were not included they were found and added to the database. For the Harter Scales if data for Family# XX was used during Pre-K it wouldn't be used for further age/grade groups. This applied to each age/grade group from Pre-K to Adult. Data for the Survey of Family Characteristics was cleaned up based on the rules below for choosing the respondents:

1. Always use the respondent with the first contact number because focusing on who filled the given questionnaire is not important for the purpose of this study. Eventually at the time whoever was available or knew the child the best, filled the form.
2. If there are more than one, 1st respondent, use the 1st contact# then (e.g. Contact#1 Mother, Father, Teacher) mother is the preferred choice.

3. If there is no Contact#1 then use the Contact#2. In the case of more than one Contact #2, Mothers are the initial choice.

   Here is the order of whom to choose: 1 –Mother, 2 –Father, 3 –Self, 4 –Teacher.

- Analysis Plan for Research Question 1: What are the relationships among developmental characteristics, secondary conditions, and QoL of children & adults with Spina Bifida?

First, to understand the nature of the variables, descriptive statistics (mean, frequency distributions) were generated for nine subscale domain scores for ABILITIES index; for each demographic areas (age, gender, bladder and bowel independence, mobility); each subscale domain weighted mean scores for Harter's Perceived Competence Scales for (peer, cognitive, physical scales); each subscale ICF-domain scores of Secondary Conditions (a- Body Functions and Structures, b- Activities &Participation) and for the ratings on the Quality of Life item. Bivariate Correlation analysis were used to examine if there were any significant relationships among the variables.

- Analysis Plan for Research Question 2: Is there a difference in the distribution of secondary conditions within the domains of the ICF-CY model?

The correspondence of the items of the secondary conditions scale with the ICF-CY biopsychosocial model were examined by linking codes with the domains of the body functions and structures, activities, participation and contextual factors (environmental factors & personal factors). The linking was done by two professionals and rate of agreement was calculated before the linked items are analyzed for their distribution across the components of the ICF-CY.

- Analysis Plan for Research Question 3: To what extent do developmental (psychosocial and functional) characteristics and family characteristics predict domains of the secondary conditions and QoL in children and adults with Spina Bifida?

It was expected that developmental characteristics, and family characteristics would significantly predict secondary conditions domains and the quality of life. The primary predictor was expected to be developmental (functional) characteristics as measured by the ABILITIES index. Secondary conditions scale captured the perceived intensity of secondary condition problems on a likert rating scale ranging from 0 (never a problem) to 5 (always a problem).

Secondary Conditions Questionnaire items were regrouped in ICF domains of “body functions & structures” and “activities & participation” based on the two-rater agreement. The means of each domain was calculated for every participant. The prediction of these two domains by the three subdomains of Harter was calculated by using a regression model. After two-rater agreement there were 16 items for body functions and one item identified for body structures codes. For that reason, these were combined as Body Functions & Structures and were obtained means of 17 items for each individual. While calculating means for each Secondary Conditions ICF domain, the rule was to have at least half of the Secondary Conditions inventory items answered (e.g. mean body functions for individual number xx= pain+ weight+ bladder control+ bowel control+ skin breakdown+ latex energy)/6). The mean calculations were done based on how many items were answered. For Body Functions and Structures calculations, 17/2=8.5 a decision rule was set to be at least answers to 8 with no items missing. The same procedure applied for the 11 items identified with Activities & Participation domain codes. For Activities & Participation, 11/2=5.5, the decision was set to be at least answers to 5 items with a valid value. The prediction of these two domains by the three subdomains of Harter was calculated by using a regression model.

Functional characteristics are measured by the ABILITIES index and ABILITIES Index weighted scores are calculated based on guidelines (Simeonsson, 2014) about how to obtain weighted scores for each of the nine subindexes. The recommended weighted values are presented below. An aggregate score for the whole index was named Abilities Index Research Composite Score (ABIRCS) derived by multiplying the rating for each domain with the weight for that domain and summing the products for a total score. To calculate the ABIRCS, a scale of 0- normal to 5- extreme difference/ disability must be used.

ABIRCS= SUM (Audition left* 1.8; Audition right* 1.8; Behavior/ social* 1.4; Behavior/Inapprop.*1.7; Intelligence*2.0; Left Hand*1.5; Left Arm*1.4; Left Leg*1.6; Right Hand*1.5; Right Arm*1.4; Right Leg* 1.6; Receptive/ comm.*1.2; Expressive/ comm.*1.0; Tone/hyper*1.5; Tone/hypo*1.4; Health*1.5; Left Eye*1.7; Right Eye*1.7; Structure/form*1.3)

For this study, ABILITIES Index data was grouped into four indexes, because neither the use of each of the nine areas separately nor using Abilities Index Research Composite Score (ABIRCS) was seen as an effective way to capture individuals functioning.

- Index I- Intellectual & Communication= [Intellectual Functioning * 2.0 + Intentional Communication (Receptive Comm.*1.2+ Expressive Communication * 1.0)] / 3
- Index II- Behavior & Social Skills= [Behavior/ social* 1.4 + Behavior/Inappropriate *1.7] / 2
- Index III- Eyes & Audition= [Audition left* 1.8 + Audition right* 1.8 + Left Eye*1.7 + Right Eye*1.7]/4
- Index IV- Physical= [Left Hand*1.5+ Left Arm*1.4 + Left Leg*1.6+ Right Hand*1.5+Right Arm*1.4+ Right Leg* 1.6 +Tone/hyper*1.5+ Tone/hypo*1.4+ Physical Health*1.5 + Structural status/form*1.3] / 10

a. Do developmental (functional) characteristics of the individuals as measured by Abilities Index and Demographics Questionnaire significantly predict secondary condition domains and higher QoL ratings?

It is hypothesized that level of functional characteristics would predict the extent of secondary conditions. In order to test the hypothesis, multiple linear regression was conducted. Initially a check was made to examine whether the data meet the assumptions of multiple linear regression.

b. Do developmental (psychosocial) characteristics of the individuals as measured by Harter's perceived competence scales significantly predict secondary conditions domains and QoL ratings over and above developmental characteristics?

It is hypothesized that increased perceived competence significantly predicts lower scores on the domains of secondary condition and higher QoL ratings.

c. Do family characteristics (strength) of the individuals with SB significantly predict the secondary conditions domains ratings and QoL ratings over and above developmental and psychosocial characteristics?

It is hypothesized that higher scores on family characteristics will significantly predict lower scores on secondary condition domains and higher QoL ratings.

Survey of Family Characteristics (SFC) Means was calculated for each individual. SFC is composed of 35 items, so at least 17 items should have a value (not missing) to have the mean calculated. Calculation of the mean for each individual was based on the number of items with not missing values and with a valid entry. All individuals had at least 29 valid entries so none of the individuals were excluded. Multiple hierarchical linear regression analysis was used to predict domains of secondary conditions and QoL ratings. Before running regression analyses the variables were examined to meet assumptions, for the statistical approach. The predictor (independent) variables were child developmental (psychosocial and functional) characteristics measured by ABILITIES index and demographical questionnaires, HARTER subscales and Survey of My Family Characteristics. The mean scores were used for each scale. Dependent variables were Secondary Conditions domain mean scores and QoL ratings. The scale captures the intensity of secondary condition problems on a likert rating scale ranging from 0 (never a problem) to 5 (always a problem) and the QoL ratings range from 0 (poor) to 4 (excellent).

A hierarchical multiple regression was conducted by using the predictor variables in the following block order.

Block 1: Age, Gender, Bladder Independence, Bowel Independence, Mobility, Four Functional Ability Indexes

Block 2: Psychosocial Characteristics (Harter Perceived Cognitive/ Social/ Physical Competence Scales)

Block 3: Family Characteristics

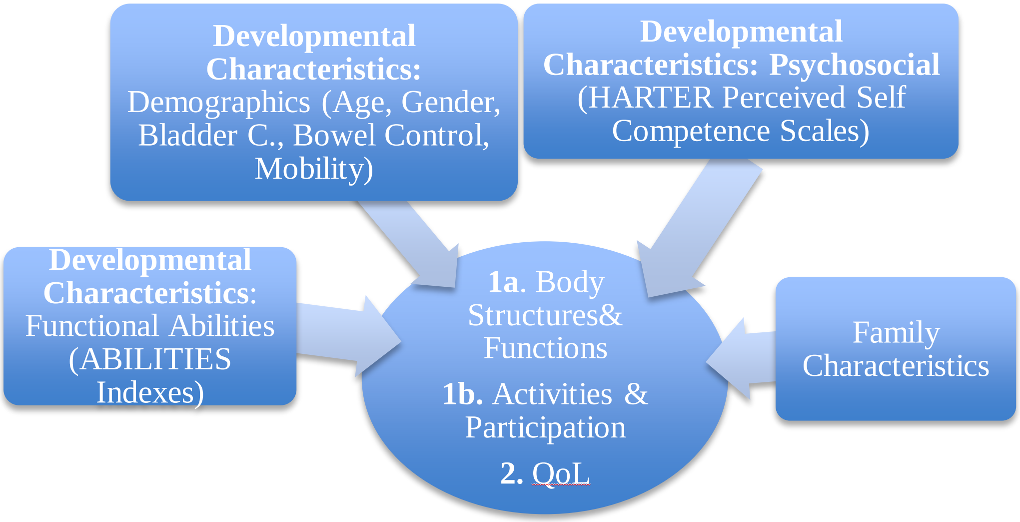


Figure 5. Regression Model for 3rd Question

CHAPTER III –RESULTS

The demographic characteristics of the study population and the mean scores of the fourteen variables are presented in Table I. Distribution of level of independence of bladder, bowel and mobility are presented in Tables 2 -4

Research Question 1

What are the relationships among developmental characteristics, secondary conditions, and QoL of children & adults with Spina bifida?

Pearson product moment correlation coefficients were calculated to examine the relationship between the functional abilities measured by ABILITIES index, perceived competence in physical, social and cognitive abilities as measured by Harter scales, Family Characteristics and ICF components of the secondary conditions (Body Functions, Activities & Participation), perceived Quality of Life, gender, age, bladder and bowel independence. The matrix of correlation coefficients is presented in Table 5. Note that the number of cases used for each correlation was determined on a "pairwise" basis. The Body Functions and Structures component of the Secondary Conditions Questionnaire such as how much of a problem the condition (pain, weight, bladder control, bowel control, skin breakdown, latex allergy, pressure sores, balance, strength, endurance, osteoporosis, breathing, sexuality, depression, lack of confidence, poor body image) was significantly positively correlated with age (r=.391) and physical functional abilities (r=.283), behavioral& social abilities (r=.276), and intellectual & communication abilities (r=.255) at the .05 level and with bladder independence (r=.206) at the

.01 level. The Body Functions Structures component was negatively correlated with perceived Quality of Life, Family Characteristics, and all three of the perceived self-competence areas (p<.05). The significance levels are all at the .05 except the correlation with Psychosocial – Social (p< .01), however the Pearson values were low ranging between r=.206 to r=.461. The strongest correlation with body functions was psychosocial- cognitive competence with a negative correlation of .461 indicating that lower values of the body functions variable was associated with higher values of the psychosocial cognitive variable. Knowing that psychosocial-cognitive competence is significantly associated with body functions, we might predict that it would be a statistically significant predictor variable in the regression model.

The Activities & Participation component of Secondary Conditions Questionnaire with items assessing how much of a problem or difficulty the condition presents significant positive relationship with functional abilities in the Behavioral and Social area and in the Intellectual & Communication area. It was found to have a strong negative relationship with perceived Quality of Life, Body Functions, perceived social competence and cognitive competence (p<.05).

A significant positive relationship was found between Perceived Quality of Life and perceived competence in cognitive, physical and social areas, and negative relationship with Functional abilities for the Behavioral & Social area with age (p<.05) and with bladder independence (p<.01). The strongest relationship with Quality of Life was the psychosocial-cognitive variable with a correlation coefficient of .376. Based on the significance level, psychosocial-cognitive was expected to be a statistically significant predictor in the regression model of Quality of Life. Family Characteristics was found to be significantly correlated with perceived competence in physical and social areas, and with intellectual& communicative functional abilities. Perceived competence in cognitive abilities was significantly correlated with perceived competence in social skills (p<.05) but negatively related with all areas of functional abilities, with bladder independence, and with age. Perceived competence in physical abilities was negatively related (p<.05) with Bladder Independence, Bowel Independence, Age, Functional abilities in all four-index areas, but positively correlated with perceived cognitive competence.

Within the areas of the Functional abilities, Intellectual and Communication was significantly related with the other areas of functional abilities. Functional physical abilities however were negatively related with Bowel Independence and Mobility variables. Significant relationships were also found for age with bladder independence, bowel independence and functional eye& audition abilities (p<.05).

Table 2. Descriptive Statistics of Study Variables

	Valid N	Range	Mean	Std. Deviation
Gender	106	1- 2	1.39	0.49
Age	105	5- 37	16.08	7.57
BladderIndependence	104	0- 5	3.37	2
BowelIndependence	103	0- 5	2.76	2.20
Mobility	104	0- 5	3.64	1.71
Functional- EyeAudition	109	1- 6	0.66	0.93
Functional- Physical	100	1- 6	2.02	1.123
Functional- BehaviorSocial	109	1- 6	0.55	1.13
Functional- Intellectual&Communication	110	1- 6	0.68	1.08
Psychosocial- Physical	110	1- 4	2.76	0.79
Psychosocial-Cognitive	107	1- 4	2.75	0.71
Psychosocial- Social	110	1- 4	2.99	0.72
Family Characteristics	106	0- 4	3.05	0.65
BodyFunctionsMean_SC	102	0- 5	0.99	0.43
ActivitiesParticipationMean_SC	88	0- 5	1.16	0.52
Quality of life	101	1- 4	3.23	0.67

Table 3. Distribution of levels of Bladder Independence of Study Population

Bladder Independence		Frequency	%	Valid %	Cumulative%
Valid	dependent	20	17.9	19.2	19.2
	takes interest	4	3.6	3.8	23.1
	assists	9	8.0	8.7	31.7
	needs supervision	9	8.0	8.7	40.4
	need reminding	9	8.0	8.7	49.0
	independent	53	47.3	51.0	100.0
	Total	104	92.9	100.0	
Missing	System	8	7.1		
Total		112	100.0		

Table 4. Distribution of levels of Bowel Independence of Study Population

Bowel Independence		Frequency	%	Valid %	Cumulative%
Valid	dependent	32	28.6	31.1	31.1
	takes interest	5	4.5	4.9	35.9
	assists	11	9.8	10.7	46.6
	needs supervision	7	6.3	6.8	53.4
	need reminding	4	3.6	3.9	57.3
	independent	44	39.3	42.7	100.0
	Total	103	92.0	100.0	
Missing	System	9	8.0		
Total		112	100.0		

Table 5. Distribution of levels of Mobility of Study Population

Mobility		Frequency	%	Valid %	Cumulative%
Valid	stroller, dependent wheelchair, no indep. mobility	6	5.4	5.8	5.8
	partial independent wheel chair	6	5.4	5.8	11.5
	independent wheelchair	26	23.2	25.0	36.5
	few steps, therapy ambulator	1	.9	1.0	37.5
	home ambutation	7	6.3	6.7	44.2
	community ambulation	58	51.8	55.8	100.0
	Total	104	92.9	100.0	
Missing	System	8	7.1		
Total		112	100.0		

Table 6. Distribution of levels of Gender of Study Population

Gender		Frequency	Percent	Valid Percent	Cumulative %
Valid	Female	65	58.0	61.3	61.3
	Male	41	36.6	38.7	100.0
	Total	106	94.6	100.0	
Missing	System	6	5.4		
Total		112	100.0		

Table 7. Distribution of levels of Age Ranges of Study Population

	Age Ranges	Frequency	Percent	Valid Percent	Cumulative %
Valid	PreK&Grade1-2	22	19.6	21.0	21.0
	ChildGrade3-8	19	17.0	18.1	39.0
	Adolescent	30	26.8	28.6	67.6
	Adult	34	30.4	32.4	100.0
	Total	105	93.8	100.0	
Missing	System	7	6.3		
Total		112	100.0		

The Figure 8 shows that both genders show progress (level1- takes interest) in Bladder Independence and up to adolescence they have a steady gain of independence up to level 5 of "needing reminders". From adolescence to adulthood they keep improving to gain independence though they may need less frequent reminders. Females tend to have a slightly slower increase from Pre-K to Elementary School ages, however they also appear to have more independence than males in adulthood.

Figure 9 shows that both genders are almost at same level (between levels 1 and 2 "takes interest to assists") up to age 8, then males experience a small decline up to level1 "bowel control" by supervision and improve to the level 5 of having "control with reminders" when they reach adulthood. Females appear to have bowel control by assistance up to adolescence followed by an increase reaching up to full independence. Furthermore, the overall frequencies (Table 3 and Table 4) indicate that 53% of the study population has bladder independence but 20% are dependent whereas 44 % have bowel independence, but 32% are bowel dependent. It is important to remember that having a study population at varying developmental stages would have an impact on the frequency results. Data from CDC's National Spina Bifida Patient registry of 3707 participants, age 5 years and older with impaired bladder function had a 35% rate of bladder independence, and 40 % with Bowel continence (CDC, 2016).

Males had home or community ambulation from age 5 to adolescence, however their mobility decreased to using "assisted ambulation and taking a few steps" in adulthood. Females were at the same level with males during adulthood, however their mobility was also at the same level up to age 8. Their mobility increases during elementary school years to adolescence with having a home ambutation (Figure 10).

ABIRCS score distribution (Figure 6), presents the increase in abilities for both genders, as individuals get older. The ABILITIES profile (Figure 6) of the study population reveals significantly higher mean scores for limbs (legs), followed by "muscle tone" and "structural status". Higher ABILITIES index score reflects greater severity of disability. As the lesion level increases in individuals with Spina Bifida, it causes a more profound disability with lower limbs the most commonly effected part.

The study population appears to have higher perception of Cognitive competence (Figure 11) up to age 8, then declines up to age 3, followed by an increase for males and decrease for females during adolescence. Both gender appear to have medium level cognitive perception during adulthood (age 18 and more). They also begin with a similar level of perceived social competence (Figure 13), females being slightly higher, however their level of perceived social competence tends to decline during adolescence. Females reach adulthood with a level of social competence similar to that in Pre-K. However, perceived social competence of males increases with age. Females experience a decrease in perceived physical competence with age, whereas males tend to have the same level of perceived physical competence at all age levels. Regardless of gender, the study population rated their perceived QoL as Very Good. Overall, all participants QoL decreased to Good as they age (Figure 14).

Figure 6. ABILITIES Profile

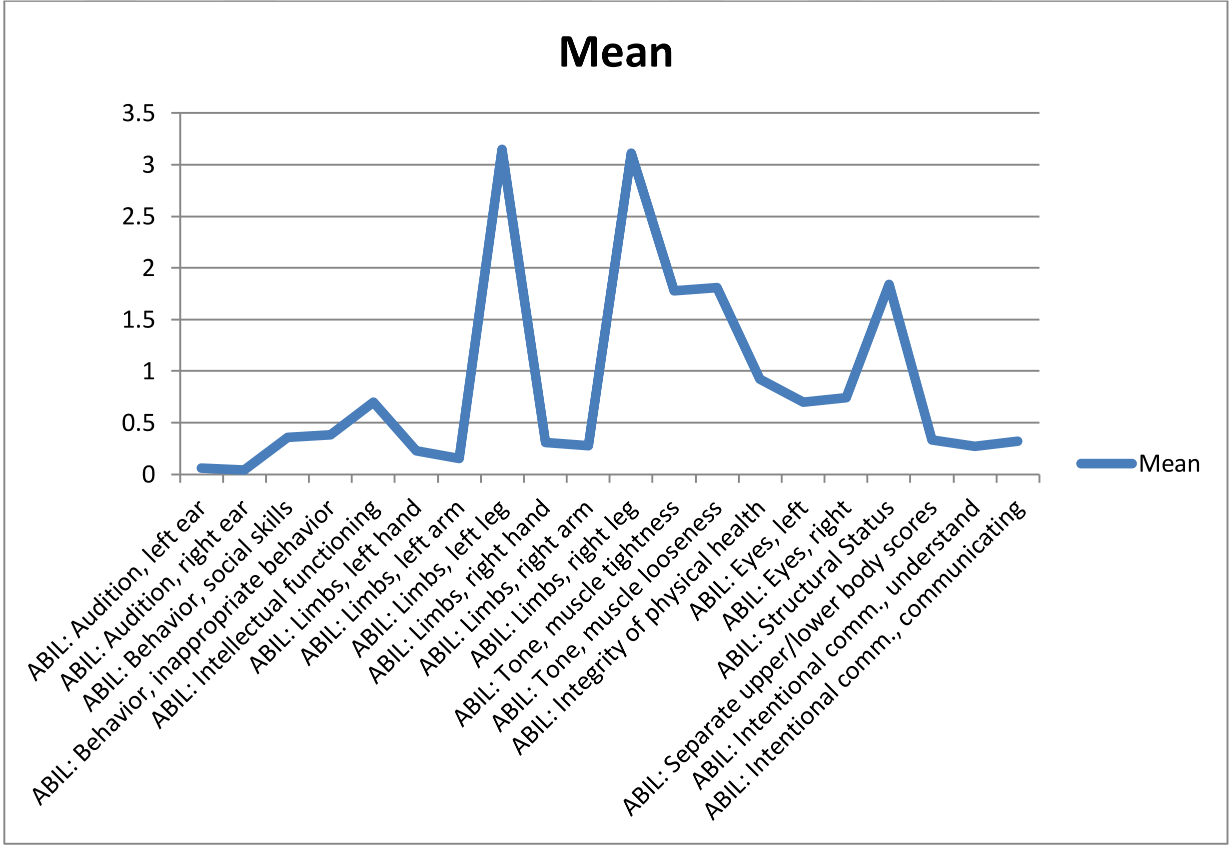


Figure 7. ABIRCS by Age and Gender

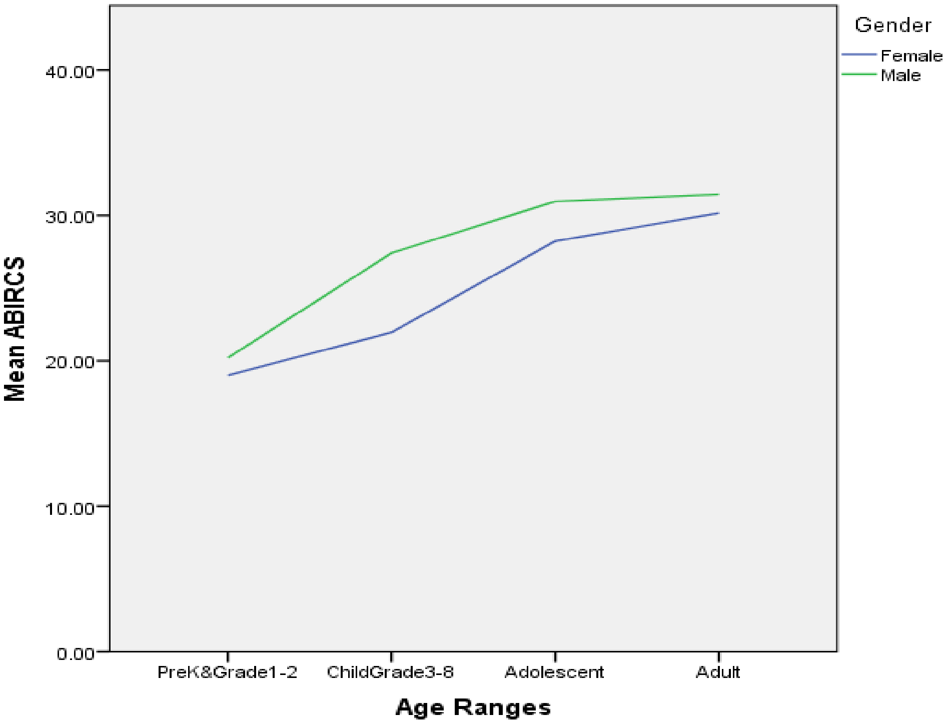


Figure 8. Bladder Independence by Age Ranges and Gender

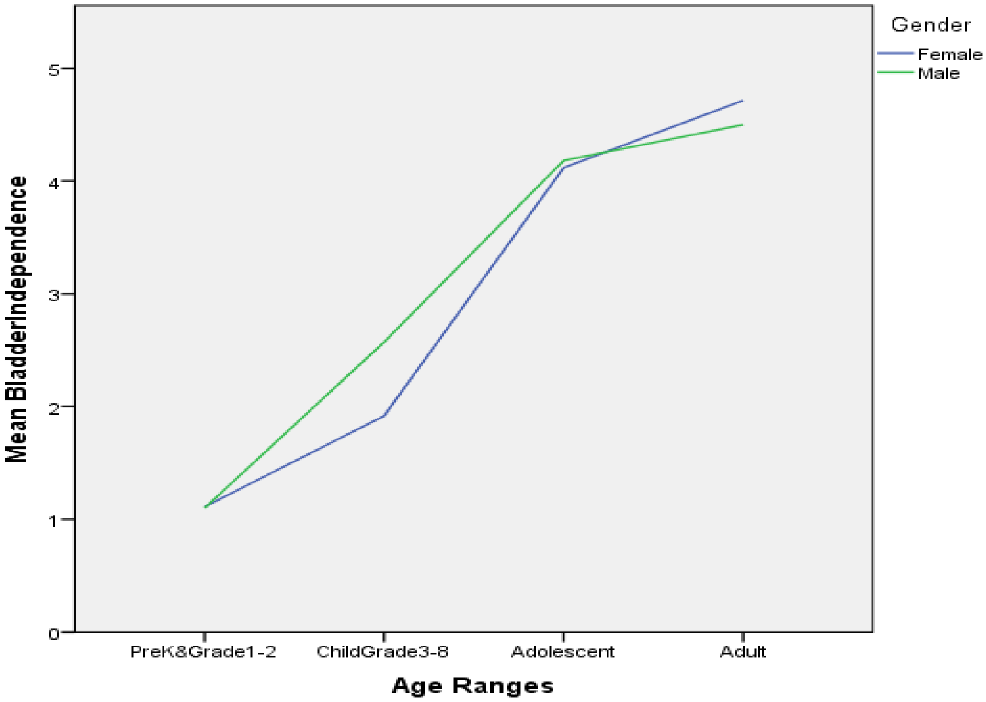


Figure 9. Bowel Independence by Age Ranges and Gender

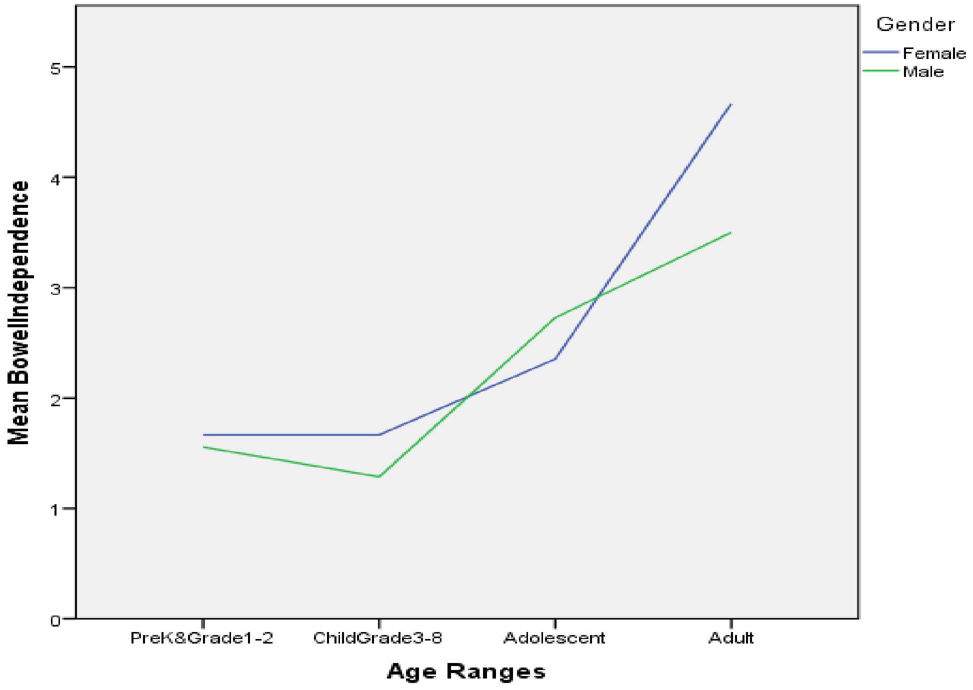


Figure 10. Mobility by Age Ranges and Gender

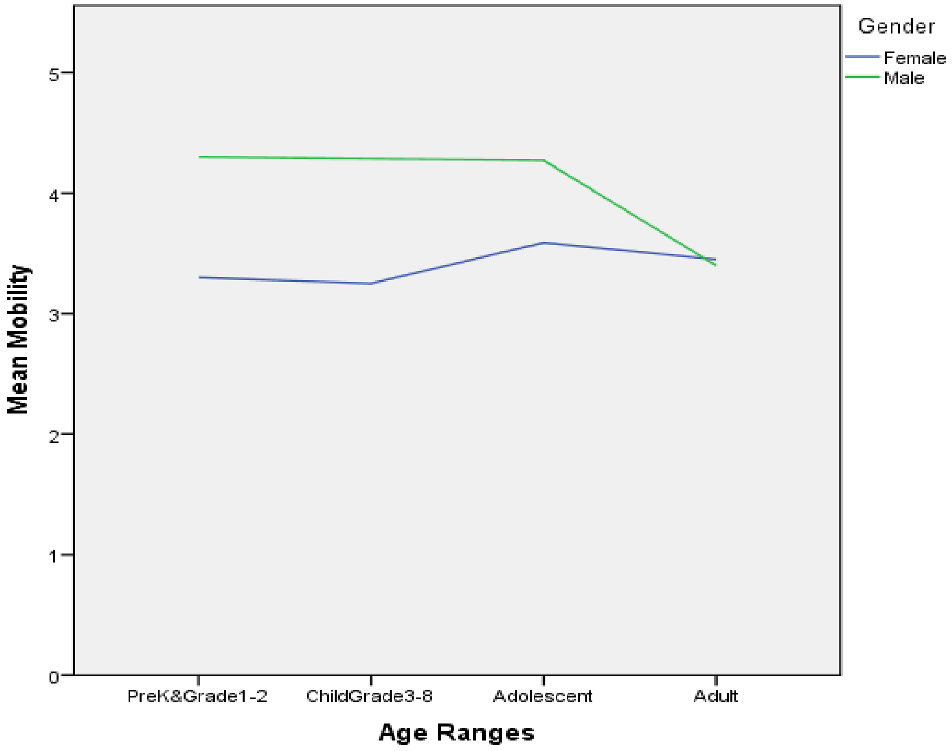


Figure 11. Psychosocial- Perceived Cognitive Competence by Age and Gender

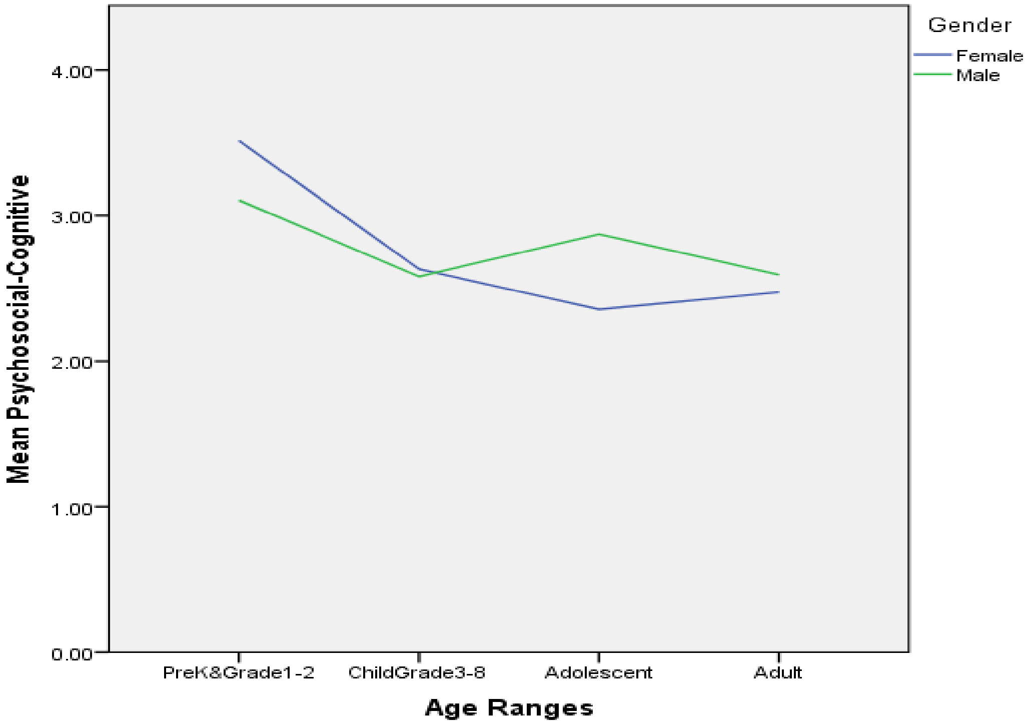


Figure 12. Psychosocial- Perceived Physical Competence by Age and Gender

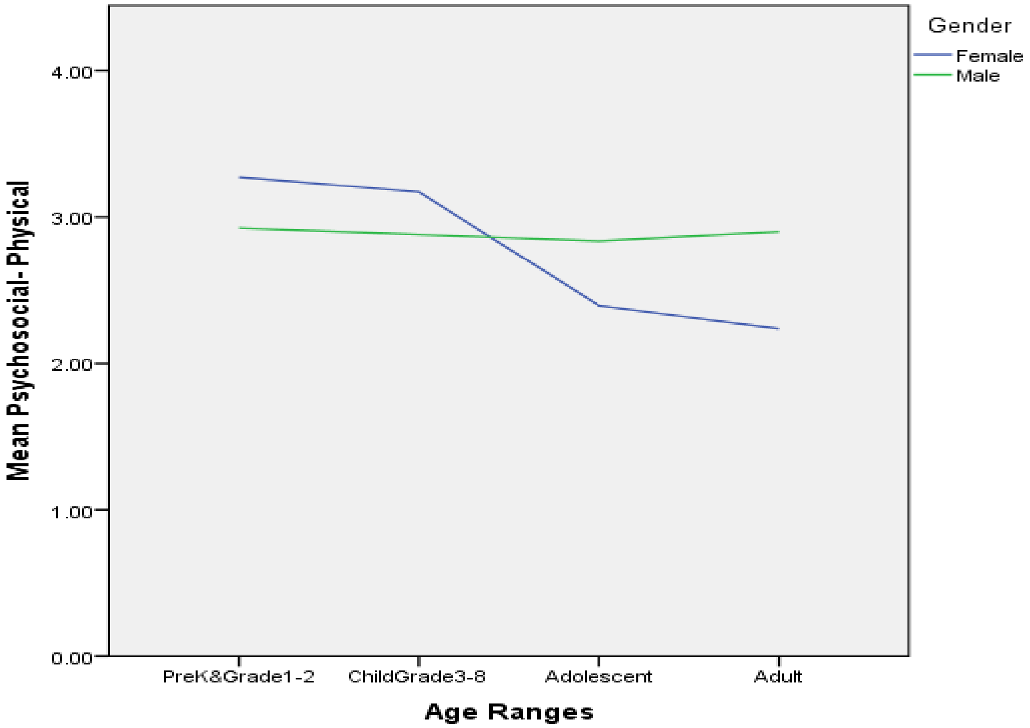


Figure 13. Psychosocial- Perceived Social Competence by Age and Gender

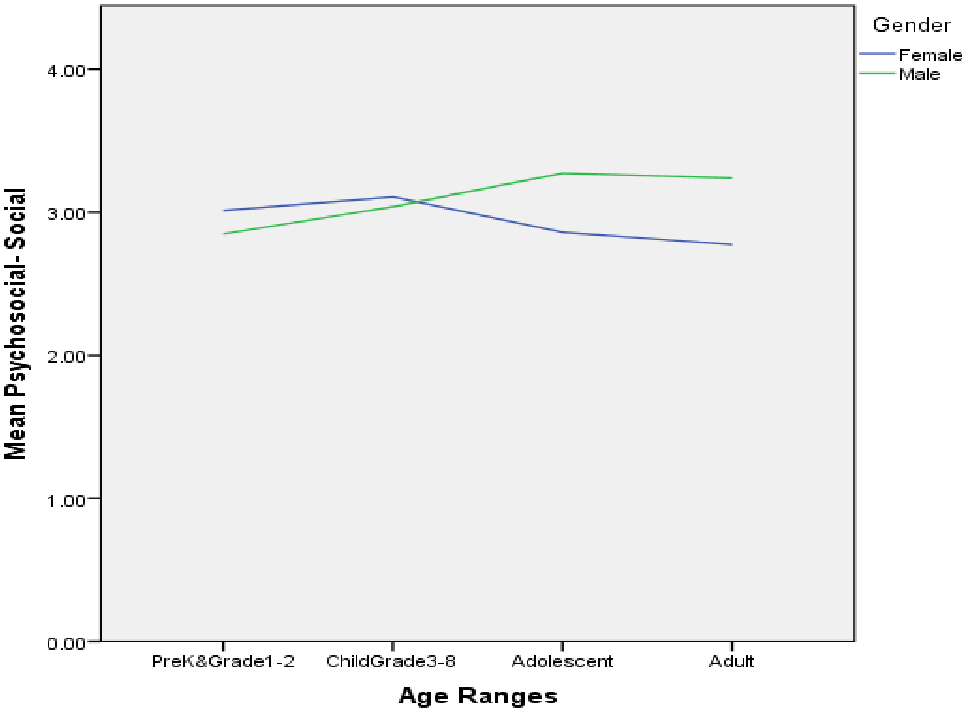


Figure 14. Perceived Quality of Life by Age and Gender

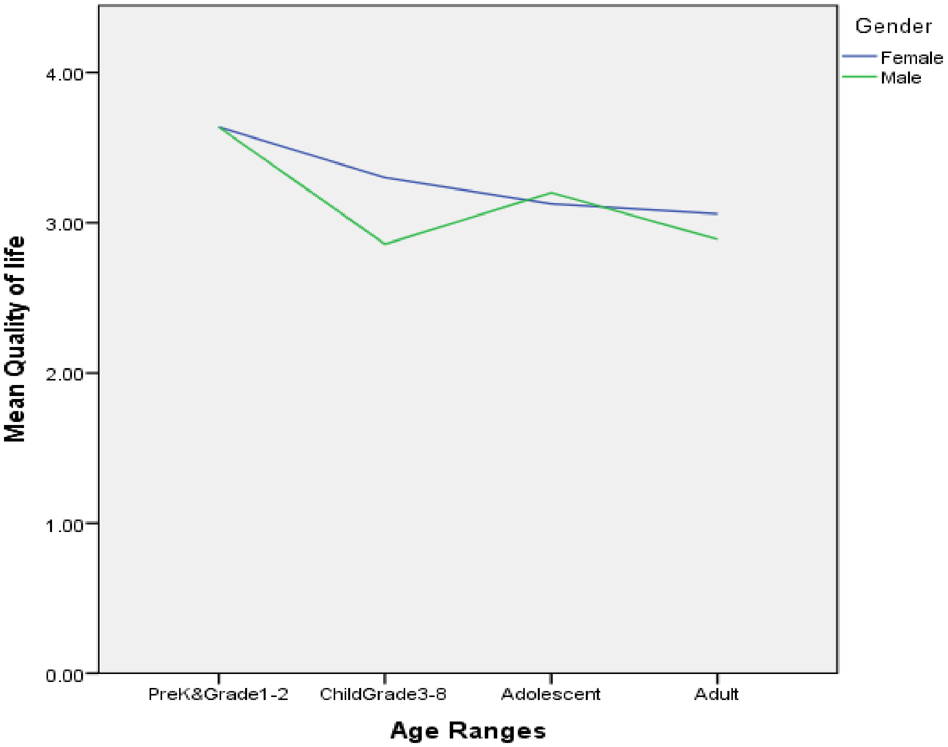


Table 8. Correlational Matrix of Study Variable

	Age	Bladder I.	Bowel I.	Mobility	Functional- Eye Audition	Functional- Physical	Functional- Behavior Social	Func.- Intellectual& Commun.	Psyc. Cognitive	Psych.- Physical	Psych.- Social	Family C.	Body Functions	Activities Participation	QoL
Age	1														
Bladder I.	.664**	1													
Bowel I.	.561**	.547**	1												
Mobility	-0.02	0.19	0.19	1											
Func.- Eye Audition	.208*	0.11	-0.03	0.04	1										
Func.- Physical	0.08	-0.12	-.208*	-.321**	0.12	1									
Func.- Behavior Social	0.10	0.07	0.01	-0.03	0.13	0.10	1								
Func.- Intellectual & Comm.	0.19	0.07	-0.05	-0.08	.317**	.290**	.485**	1							
Psyc.- Cognitive	-.315**	-.296**	-0.19	0.09	-.284**	-.227*	-.256**	-.403**	1						
Psyc.- Physical	-.339**	-.285**	-.216*	0.05	-.333**	-0.11	-.257**	-.287**	.397**	1					
Psych.- Social	-0.05	0.07	0.09	-0.04	0.01	-0.08	-0.19	-0.16	.292**	.439**	1				
Family C.	-.330**	-0.13	-0.06	0.06	-0.14	-0.19	-0.17	-.196*	0.18	.453**	.309**	1			
Body Func.	.391**	.206*	0.14	-0.15	0.18	.283**	.276**	.255**	-.461**	-.375**	-.220*	-.410**	1		
Activities Participat.	0.05	0.15	-0.12	-0.06	0.11	0.18	.368**	.494**	-.308**	-0.17	-.219*	-0.17	.393**	1	
QoL	-.326**	-.253*	-0.14	-0.03	-0.15	-0.17	-.271**	-0.14	.376**	.314**	.199*	0.20	-.398**	-.294**	1

**. Correlation is significant at the 0.05 level (2- tailed)

*. Correlation is significant at the 0.01 level (2- tailed)

Research Question 2

- Is there a difference in the distribution of secondary conditions within the domains of the ICF-CY model?

The correspondence of the items of the secondary conditions scale with the ICF-CY biopsychosocial model was examined by linking codes with the domains of the body functions and structures, activities, participation and environmental factors. Linking items from the secondary conditions questionnaire with the ICF-CY as a frame of reference provided a useful approach for the assessment of health and functioning in individuals with Spina Bifida. Linkage exercises serve as a basis for determining the nature and extent of impairments within a common cross-culturally consistent taxonomy (Sommer et al., 2015). Two professionals who had a good knowledge of the ICF-CY independently linked the Secondary Conditions Inventory Items with the codes. Then, rate of agreement was calculated before the linked items were analyzed for their distribution across the components of the ICF-CY.

Initially the first six steps in assigning ICF-CY classes and codes (page xix & xx) from the ICF-CY manual was used followed by the first seven rules defined by Cieza et al., 2016 study. A combination of steps as defined by the ICF-CY Manual (2007, pg. xix) and rules by Cieza et al. 2016 was used in the following order:

1. Acquire good knowledge of the conceptual and taxonomical fundamentals of the ICF, as well as of the chapters, domains and categories of the detailed classification, including definitions before starting to link meaningful concepts to the ICF (Cieza et al., 2016; pg.4).
2. Define the information available for coding and identify whether it relates to the domain

of Body Functions, Body Structures, Activities/Participation or Environmental Factors (ICF-CY Manual, 2007; pg. xix).

3. Identify the purpose of the information to be linked by answering the question “What is this piece of information about? or What is this item about?”. The answer to these questions will help to identify the main concept(s) most relevant to be linked to the ICF (Cieza et al., 2016; pg.4).
4. Identify any additional concepts contained in the piece of information in addition to the main concept(s) already identified in the previous step.
5. Identify and document the categorization of the response options
6. Locate the chapter (4-character code) within the appropriate domain that most closely corresponds to the information to be coded (ICF- CY Manual, 2007). / Link all meaningful concepts, the most relevant and additional ones, to the most precise ICF category (Cieza et al., 2016; pg.4).

   Health and health-related states should be recorded as specifically as possible, by assigning the most appropriate ICF category. For example, the most specific code for a person with night blindness is b21020 “Light sensitivity”. If, however, for some reason this level of detail cannot be applied, the corresponding “parent” code in the hierarchy can be used instead (in this case, b2102 Quality of vision, b210 Seeing functions, or b2 Sensory functions and pain) (ICF-CY Manual 2007, pg. 239).

7. Read the description of the 4-character code and attend to any notes related to the description (ICF- CY Manual, 2007; pg. xix).
8. Review any inclusion or exclusion notes that apply to the code and proceed accordingly (ICF- CY Manual, 2007; pg. xix).

9. Determine if the information to be coded is consistent with the 4-character level or if a more detailed description at the 5- or 6-character code should be examined (ICF- CY Manual, 2007; pg. xix).
10. Proceed to the level of code that most closely corresponds to the information to be coded. / Review the description and any inclusion or exclusion notes that apply to the code (ICF-CY Manual, 2007; pg. xix).
11. Use “other specified [8]” or “unspecified [9]” ICF categories as appropriate (Cieza et al., 2016; pg.4).

Based on the data obtained from the study population, the distribution and frequency of impairments in components of ICF-CY were calculated. Final codings of ICF components (b, s, d, e) were then quantified based on the 6-point likert type scale of secondary conditions questionnaire, followed by descriptive statistics. The percentage agreement between the health professionals in each of the instruments tested was calculated. In the 2005 and 2016 rules by Cieza neither “other specified” nor the “unspecified” categories of the ICF were used. Cieza claimed to “reduce ambiguity of the results of the linking process”.

In this study, the steps defined by the ICF manual were mainly used, therefore the “other specified” and “unspecified” categories were essentially not used as almost none of the Secondary Conditions items pinpoint a specific description. Furter, Cieza et al., 2005 used these linking rules for the results of intervention and outcome measures, but in this study, Secondary Conditions items provided a description of the extent of problems individuals’ experience, not the result of any measures. Different from previous studies, this study also addressed the issue raised by Cieza et al., 2005 “The information as to what extent an item refers to activity, to participation, or to both as well as the information as to whether an activity or participation from the perspective of capacity or of performance (pg. 217).

a. What is the rate of agreement between two raters?

According to the standards for linking measures to the ICF, linking is performed by the primary investigator and another professional who has acquired expert knowledge of the conceptual fundaments of the ICF-CY, as well as of the chapters, domains and categories of the detailed classification including their definitions. The linking versions of two raters were compared, discussed and were resolved any disagreements in the linkage.

For the rate of agreement is reflected in percentages. It was not possible to check the inter-rater reliability by calculating the kappa coefficient as it was proposed. In the table below, the coding are presented by 1st raters view, 2nd raters view, and the final code reached by consensus of the two raters.

The level of agreement between the two raters after initial coding was low (16%) so the raters discussed their reasoning for coding for each item to reach an agreement. For many items the disagreement was on the 3rd level of coding such as “b280- sensation of pain” vs “b289- sensation of pain, other specified and unspecified” or “b235- vestibular functions” vs “b2351- vestibular function of balance”. For that reason the level of agreement was calculated at the 2nd level of coding (e.g. b62, d17, d15, d77). The agreement increased up to 53% at the 2nd level. The percentage of rater agreement at the first level of domain was 75%.

There are 16 items representing body functions (b), which accounts for the %50 of all the items; only1 item representing body structures (s) accounting for the 3% of all items, 11 items representing activities and participation at the 34%, and 4 items were not coded.

Table 9. Secondary Conditions (SC) and ICF –CY Linkage

Item #	Secondary Conditions Items	Rater 1 Codes	Rater 2 Codes	Final Agreement	Agreement on Initial 2 step of the codes
	Physical	ICF	ICF Main codes		
1	Pain	b289 Sensation of pain, other specified and unspecified	b289 Sensation of pain, other specified and unspecified	b289	b28
2	Weight	b530 Weight maintenance functions	b530 Weight maintenance functions	b530	
3	Bladder Control	b6202 Urinary continence	b6202 Urinary continence	b6202	b620
4	Bowel Control	b5253 Faecal continence	b5253 Faecal continence	b5253	b525
5	Skin breakdown	b849 Functions of the skin, other specified and unspecified	b849 Functions of the skin, other specified and unspecified	b810	
6	Latex allergy	b849 Functions of the skin, other specified & unsp.	b849 Functions of the skin, other specified and unspecified	b810	
7	Pressure sores	b849 Functions of the skin, other specified and unsp.	b849 Functions of the skin, other specified and unspecified	b810	
8	Mobility	b7208 Mobility of bone functions, other specified	b7208 Mobility of bone functions, other specified	d450	
9	Scoliosis	s1200 Structure of spinal cord	s1200 Structure of spinal cord	s760	
10	Balance	b2351 Vestibular function of balance	b2351 Vestibular function of balance	b2351	b235
11	Strength	b4550 General Physical Endurance	b4550 General Physical Endurance	b730	
12	Endurance	b499 Functions of the cardiovascular, haematological, immunological and respiratory systems, unspecified	b499 Functions of the cardiovascular, haematological, immunological and respiratory systems, unspecified	b4550	

13	Osteoporosis	Health Condition	nc- Health Condition	b7700	
14	Breathing	b449 Functions of the respiratory system, other specified and unspecified	b449 Functions of the respiratory system, other specified and unspecified	b449	b44
15	Sexuality	b6409 Sexual functions, unspecified	b6409 Sexual functions, unspecified	b6409	b640
	Academic		ICF Main codes		
16	Math	b1729 Calculation functions, unspecified	b1729 Calculation functions, unspecified	d172	
17	Reading	d1669 Reading, unspecified	d1669 Reading, unspecified	d166	d166
18	Writing	d1709 Writing, unspecified	d1709 Writing, unspecified	d170	d170
19	Spelling	d1701 Using grammatical and mechanical conventions in written compositions	d1701 Using grammatical and mechanical conventions in written compositions	d1701	d170
20	Problem Solving	d1759 Solving problems, unspecified	d1759 Solving problems, unspecified	d175	d175
21	Learning/Attention	d159 Basic learning, other specified and unspecified / b140	d159 Basic learning, other specified and unspecified	d159/b1409	
	Personal		ICF Main codes		
22	Depression	b1528 Emotional functions, other specified	b1528 Emotional functions, other specified	b1528	b15
23	Low self-esteem	b1528 Emotional functions, other spe.	b1528 Emotional functions, other specif.	No code	
24	Lack of confidence	b1266- Confidence	b1266- Confidence	b1266	
25	Poor body image	b1801 Body image	b1801 Body image	b1801	
	Social		ICF Main codes		
26	Isolation	d799 Interpersonal interactions and relationships,	d799 Interpersonal interactions and relationships,	No code	

		unspecified	unspecified		
27	Exclusion	d799 Interpersonal interactions and relationships, unspecified	d799 Interpersonal interactions and relationships, unspecified	No code	
28	Unemployment	d8451-Maintaining a job	d8451-Maintaining a job	d8451	
29	Sexuality	d7702 Sexual relationships	d7702 Sexual relationships	d7702	
30	Economic Dependence	d8700 Personal economic resources	d8700 Personal economic resources	d870	d87
31	Family Stress	d2401 Handling stress	d2401 Handling stress	No code	
32	Lack of social skills	d7- interpersonal interactions and relationships	d7- interpersonal interactions and relationships	d729	

	b	s	d	e	pf	nc
#	16	1	11	0	0	4
	0.5	0.031	0.34			0.13

Both Raters Initially Agree on the Whole Code %	% of agreement only at the 2nd level	% of agreement at 2nd level	1st level of agreement
0.19	0.38	0.56	0.72

Research Question 3

To what extent do developmental characteristics (psychosocial-Harter and functional-ABILITIES & Demographics) and family characteristics predict domains of the Secondary Conditions and QoL in individuals with Spina bifida?

- **a.** Do developmental (functional) characteristics of the individuals as measured by Abilities Index and Demographics Questionnaire significantly predict secondary condition domains and higher QoL ratings?
- **b.** Do developmental (psychosocial) characteristics of the individuals as measured by Harter's perceived competence scales significantly predict secondary conditions domains and QoL ratings over and above developmental characteristics?
- **c.** Do family characteristics (strengths) of the individuals significantly predict the secondary conditions domains ratings and QoL ratings over and above developmental and psychosocial characteristics?

Predicting Body Functions & Structures

A multiple hierarchical linear regression was conducted to predict difficulty with Body Functions and Structures, based on developmental (demographics, functional abilities, perceived competence) and family characteristics. The functional abilities, age, gender, mobility, bowel and bladder control were used in the first block. Psychosocial characteristics as measured by the HARTER social, cognitive and physical perceived competence subscales were added in the second block, followed by the family strength as measured by Survey of Family Characteristics in the third block. Results are presented in the order of testing the blocks in the overall model for predicting secondary conditions (Body Functions & Structures, Activities & Participation).

In Block 1 the functional characteristics, age, gender, bowel and bladder control were entered in the regression analysis as predictors of the criterion “Body Functions &Structures”. The results of the regression analysis indicated that functional abilities, bladder control, bowel control, mobility, age and gender explained 27% of the variance and significantly predicted perceived difficulty level of living with secondary conditions that fell in the Body Functions & Structures component (R^2= .270, Adjusted R^2= .189, F (9, 81)= 3.335, p<.01). In this case, the adjusted R^2 value suggests that 19% of the variability of body functions is accounted for by the model, taking into account the number of predictor variables in the block (Table 10 & Table 11). The coefficients for each of the variables indicated the amount of change one could expect in body functions given a one-unit change in the value of that variable, given that all other variables in the model are held constant. The Coefficients table (Table 12) indicates that “Age” is a significant predictor such that an increase of .02 (Unstandardized B Coefficient= 0.02) would be expected in the body functions score is associated with one unit increase in Age, assuming that all other variables in the model were held constant (B=.020, Std. Error= .008; p <.05). This result indicates that as participants get older the perceived secondary condition difficulty level increases.

In Block 2, psychosocial characteristics were entered into the multiple regression model. Adding psychosocial characteristics as a predictor resulted in an R^2 change of .093. This means that there was an increase of about 9% in the proportion of variance explained by adding psychosocial characteristics (R^2 change= .093, F (3, 78)= 3.852, p < .05). The perceived competence in cognitive abilities (as measured by Harter scales) predicted significantly over and above the functional abilities, age and gender (Unstandardized B: -.181, β= -.30, Std. Error: .07;

p <.05). Age remained a significant predictor in the model as well (Unstandardized B: .019, Std. Error: .001; p< .05).

In Block 3 Family characteristics were entered into the multiple regression model. Adding family characteristics (as a measure of coping with stressors) resulted in an increase of .039 in the proportion of variance explained by family characteristics over and above the previous predictors. The family characteristics significantly predicted perceived difficulty level of living with secondary conditions that fell in the Body Functions & Structures over and above both psychosocial and functional abilities (R^2 change= .035, F (1, 77)= 4.52, p <.05, Table 10). The results indicated that one unit increase in family strength predicted .148 unit decrease in perceived difficulty of secondary conditions in the Body Functions area (B=.15, β= -.22, Std. Error= .007; p <.05).

Table 10. Model Summary for Prediction of Body Functions & Structures

					Change Statistics				
Model	R	R^2	Adjusted R^2	Std. Error of the Estimate	R^2 Change	F Change	df1	df2	Sig. F Change
1	$.520^{a}$	.270	.189	.38852	.270	3.335	9	81	.002
2	$.603^{b}$	.364	.266	.36974	.093	3.852	3	78	.013
3	$.632^{c}$	.399	.298	.36166	.035	4.525	1	77	.037

a. Predictors: (Constant), (Functional- Intellectual& Communication, Functional- Eye Audition, Functional- Physical, Functional- Behavior Social), Bladder Independence, Bowel Independence, Mobility, Gender, Age

b. Predictors: (Constant), Predictors in Model 1+ (Psychosocial- Social, Psychosocial- Cognitive, Psychosocial- Physical)

c. Predictors: (Constant), Predictors in Model 2+ Family Characteristics

Table 11. ANOVA Model for Prediction of Body Functions & Structures

Model		Sum of Squares	df	Mean Square	F	Sig.
1	Regression	4.531	9	.503	3.335	.002
	Residual	12.227	81	.151		
	Total	16.758	90			
2	Regression	6.095	12	.508	3.715	.000
	Residual	10.663	78	.137		
	Total	16.758	90			
3	Regression	6.686	13	.514	3.932	.000
	Residual	10.072	77	.131		
	Total	16.758	90			

Dependent Variable: BodyFunctionsMean_SC

a. Predictors: (Constant), (Functional- Intellectual& Communication, Functional- Eye Audition, Functional- Physical, Functional- Behavior Social), Bladder Independence, Bowel Independence, Gender, Age, Mobility

b. Predictors: (Constant), Predictors in Model 1+ (Psychosocial- Social, Psychosocial- Cognitive, Psychosocial- Physical)

c. Predictors: (Constant), Predictors in Model 2+ Family Characteristics

Table 12. Regression Coefficients for Body Functions & Structures

	Model 1			Model 2			Model 3		
	B	Std. Error	β	B	Std. Error	β	B	Std. Error	β
(Constant)	0.53	0.20		1.40	0.32		1.78	0.36	
Gender	-0.01	0.09	-0.01	0.03	0.08	0.03	0.03	0.08	0.03
Age	0.02	0.08	0.34*	0.02	0.01	0.33*	0.01	0.01	0.25
BladderIndependence	-0.01	0.03	-0.01	-0.02	0.03	-0.10	-0.02	0.03	-0.07
BowelIndependence	0.02	0.03	0.01	-0.01	0.03	-0.05	0.00	0.03	-0.02
Mobility	-0.02	0.03	-0.07	-0.01	0.03	-0.04	-0.01	0.03	-0.04
Functional- EyeAudition	0.03	0.05	0.07	-0.01	0.05	-0.02	-0.01	0.05	-0.01
Functional- Physical	0.08	0.04	0.20	0.05	0.04	0.14	0.05	0.04	0.12
Functional- BehaviorSocial	0.08	0.04	0.21	0.06	0.04	0.16	0.06	0.04	0.15
Functional- Intellectual&Communication	0.00	0.05	0.01	-0.03	0.05	-0.08	-0.03	0.05	-0.08
Psychosocial-Cognitive				-0.18	0.07	-0.30*	-0.19	0.07	-0.31*
Psychosocial- Physical				-0.09	0.07	-0.16	-0.04	0.07	-0.08
Psychosocial- Social				-0.03	0.07	-0.01	0.01	0.07	0.02
Family Characteristics							-0.15	0.07	-0.22*

**p* < .05. ***p* < .01.

Dependent Variable: Body Functions Mean_ SC

a. Model 1 Predictors: (Constant), Functional- Intellectual& Communication, Functional- Eye Audition, Functional- Physical, Functional- Behavior Social, Bladder Independence, Bowel Independence, Mobility, Gender, Age
b. Model 2 Predictors: (Constant), Predictors in Model 1+ (Psychosocial- Social, Psychosocial-Cognitive, Psychosocial- Physical)
c. Model 3 Predictors: (Constant), Predictors in Model 2+ Family Characteristics

Predicting Activities and Participation

A multiple hierarchical linear regression was conducted to assess the ability of developmental (demographics, functional abilities, perceived competence), psychosocial, and family characteristics to predict perceived difficulties related to Activities And Participation. The order of the blocks was the same as the previous regression model for predicting secondary conditions. Functional abilities, bladder independence, bowel independence, mobility, age and gender were entered in Block 1, explained 36% of the variance (R^2= .362) significantly predicting difficulties with the Activities And Participation component (R^2= .362, Adjusted R^2= .278, F (9, 68)= 4.43, p<.01). The model accounted for 28% of the variability in activities and participation, taking into account the number of predictor variables in the model (Adjusted R^2: .278) (Table 13 & 14).

The Coefficients for the first block indicate that “Bladder Independence” and “Intellectual & Intentional Communication” functioning were significant predictors of Activities & Participation with Bladder Independence beta value being 0.32 (p <.05) and for Intellectual & Intentional Communication abilities beta being 0.43 (p<.01). These results indicated that every unit increase in Bladder independence is associated with .08 unit increase in the Activities and Participation mean score. Every unit increase in Intellectual & Communication abilities mean score is associated with a .21 unit increase in Activities and Participation mean score, assuming that all other variables in the model are held constant (B: .21, Std. Error: .06; p <.05, Table 13).

In Block 2 Psychosocial characteristics were added into the multiple regression model. This block was designed to test if adding psychosocial characteristics added any additional benefit to the overall prediction model. Adding psychosocial characteristics in Block 2 resulted in an R square change of .035, reflecting a non-significant increase in the proportion of variance explained (F (3,65) =1.43, (p>.05)). Bladder independence, and Intellectual & Communication abilities, continued to be significant predictors at the first block, however psychosocial competence did not contribute to the model. Gender also became a significant predictor variable (p<.05, B=.23, Beta=.21).

In Block 3 Family characteristics were added into the multiple regression model. Adding family characteristics (as a measure of coping with stressors) over functional and psychosocial characteristics also did not significantly increase the predictability of the model above the variables included in Block 1. An R square change of 0.06 meant almost no increase in the proportion of variance explained by adding family characteristics, the change in F (1,64) = .75 was not significant (p> .05). The effect of significant predictor variables in Block 1 continued to have similar level of effect, with the addition that gender also became a significant predictor variable (p<.05, B=.215). With every unit increase in gender, Activities & Participation difficulty mean was expected to increase by .22 units. Gender was coded as Female: 1, and Male: 2, so Males are expected to experience higher levels of difficulty in the area of Activities and Participation. Family Characteristics scale could be considered to represent a positive resilience factor, it was interesting that having a positive family support system did not contribute into individual’s experiences of secondary conditions. This may indicate that the difficulty living with secondary conditions is very personal despite surrounding support systems.

Table 13. Model Summary for Prediction of Activities and Participation

					Change Statistics				
Model	R	R^2	Adjusted R^2	Std. Error of the Estimate	R^2 Change	F Change	df1	df2	Sig. F Change
1	.602	.362	.278	.44	.362	4.43	9	68	.000
2	.634	.401	.291	.44	.039	1.41	3	65	.248
3	.638	.407	.286	.44	.005	.57	1	64	.451

Table 14. ANOVA Model for Prediction of Activities and Participation

Model		Sum of Squares	df	Mean Square	F	Sig.
1	Regression	7.466	9	0.83	4.295	.000
	Residual	13.135	68	0.193		
	Total	20.601	77			
2	Regression	8.268	12	0.689	3.632	.000
	Residual	12.333	65	0.19		
	Total	20.601	77			
3	Regression	8.378	13	0.644	3.374	.001
	Residual	12.223	64	0.191		
	Total	20.601	77			

Dependent Variable: Activities ParticipationMean_SC

a. Predictors: (Constant), Functional- Intellectual&Communication, Functional- EyeAudition, Functional- Physical, Functional- BehaviorSocial, BladderIndependence, BowelIndependence, Gender, Age, Mobility

b. Predictors: (Constant), Predictors in Model 1+ (Psychosocial- Social, Psychosocial- Cognitive, Psychosocial- Physical)

c. Predictors: (Constant), Predictors in Model 2+ Family Characteristics

Table 15. Regression Coefficients for Activities and Participation

	Model 1			Model 2			Model 3		
	B	Std. Error	β	B	Std. Error	β	B	Std. Error	β
(Constant)	0.74	0.25		1.22	0.42		1.39	0.48	
Age	-0.01	0.01	-0.13	-0.01	0.01	-0.19	-0.02	0.01	-0.22
Gender	0.19	0.11	0.18	0.23	0.11	0.21*	0.23	0.11	0.21*
BladderIndependence	0.08	0.04	0.32*	0.09	0.04	0.36*	0.10	0.04	0.37*
BowelIndependence	-0.04	0.03	-0.16	-0.03	0.03	-0.12	-0.03	0.03	-0.11
Mobility	-0.02	0.03	-0.07	-0.03	0.03	-0.10	-0.03	0.03	-0.11
Functional- EyeAudition	-0.03	0.06	-0.05	-0.01	0.06	-0.02	-0.01	0.06	-0.01
Functional- Physical	0.01	0.05	0.03	0.01	0.05	0.02	0.00	0.05	0.01
Functional- BehaviorSocial	0.07	0.05	0.15	0.06	0.05	0.12	0.05	0.05	0.12
Functional- Intellectual&Communication	0.21	0.06	0.43*	0.19	0.06	0.40*	0.19	0.06	0.40*
Psychosocial-Cognitive				-0.06	0.09	-0.09	-0.07	0.09	-0.09
Psychosocial- Physical				0.05	0.08	0.08	0.08	0.09	0.11
Psychosocial- Social				-0.14	0.09	-0.20	-0.14	0.09	-0.19
Family Characteristics							-0.07	0.09	-0.09

**p* < .05. ***p* < .01.
Dependent Variable: Activities Participation Mean_ SC

a. Model 1 Predictors: (Constant), (Constant), Functional- Intellectual&Communication, Functional- EyeAudition, Functional- Physical, Functional- BehaviorSocial, BladderIndependence, BowelIndependence, Mobility, Gender, Age

b. Model 2 Predictors: (Constant), Predictors in Model 1+ (Psychosocial- Social, Psychosocial-Cognitive, Psychosocial- Physical)

c. Model 3 Predictors: (Constant), Predictors in Model 2+ Family Characteristics

Predicting Quality of Life

A multiple hierarchical linear regression was conducted to predict perceived Quality of Life, based on psychosocial, functional and family characteristics as well as secondary conditions (Activities & Participation component). In addition to the predictor variables in the previous regression analyses of secondary conditions the Activities & Participation ICF component of secondary conditions was added as a mediator in the model as a 4th block.

Based on Anova results, the first block of predictors did not yield an overall effect, but adding in the perceived competence in social, physical and cognitive areas at the second block resulted in the overall effect to be significant (p<.05, Table 17). Adding Family Characteristics in the third block indicated that it was a significant predictor of QoL (p<.05). Adding the Activities and Participation with the fourth block resulted in the overall model being significant (p<.05), indicating its strong prediction value. The results of the hierarchical regression indicated that adding other set of predictors over and above the existing ones did not have any significant changes in the model.

Table 16. Model Summary for Prediction of Quality of Life

					Change Statistics				
Model	R	R^2	Adjusted R^2	Std. Error of Estimate	R^2 Change	F Change	df1	df2	Sig. F Change
1	0.45	0.20	0.10	0.64	0.20	1.92	9	68	0.06
2	0.53	0.28	0.14	0.63	0.08	2.24	3	65	0.09
3	0.53	0.28	0.13	0.63	0.00	0.00	1	64	0.97
4	0.56	0.31	0.16	0.62	0.04	3.21	1	63	0.08

a. Predictors: (Constant), Functional- (Intellectual& Communication, Eye Audition, Physical, Behavior Social), Bladder Independence, Bowel Independence, Mobility, Gender, Age

b. Predictors: (Constant), Predictors in Model 1+ (Psychosocial- Social, Psychosocial- Cognitive, Psychosocial- Physical)

c. Predictors: (Constant), Predictors in Model 2+ Family Characteristics

d. Predictors: (Constant), Predictors in Model 3+ Activities Participation Mean_SC

Table 17. ANOVA Model for Prediction of QoL

Model		Sum of Squares	df	Mean Square	F	Sig.
1	Regression	7.14	9.00	0.79	1.92	0.06
	Residual	28.10	68.00	0.41		
	Total	35.24	77.00			
2	Regression	9.77	12.00	0.81	2.08	0.03*
	Residual	25.47	65.00	0.39		
	Total	35.24	77.00			
3	Regression	9.77	13.00	0.75	1.89	0.05*
	Residual	25.47	64.00	0.40		
	Total	35.24	77.00			
4	Regression	11.01	14.00	0.79	2.04	0.03*
	Residual	24.23	63.00	0.39		
	Total	35.24	77.00			

Dependent Variable: Quality of life

a. Predictors: (Constant), Functional- Intellectual&Communication, Functional- EyeAudition, Functional- Physical, Functional- BehaviorSocial, BladderIndependence, BowelIndependence, Gender, Age

b. Predictors: (Constant), Predictors in Model 1+ (Psychosocial- Social, Psychosocial-Cognitive, Psychosocial- Physical)

c. Predictors: (Constant), Predictors in Model 2+ Family Characteristics

d. Predictors: (Constant), Predictors in Model 3+ ActivitiesParticipationMean_SC

Table 18. Regression Coefficients for Quality of Life

	Model 1			Model 2			Model 3			Model 4		
	B	Std. Error	β	B	Std. Error	β	B	Std. Error	β	B	Std. Error	β
(Constant)	4.15	0.36		2.92	0.60		2.93	0.69		3.38	0.72	
Age	-0.02	0.02	-0.26	-0.02	0.02	-0.25	-0.02	0.02	-0.25	-0.03	0.02	-0.31
Gender	-0.06	0.16	-0.05	-0.11	0.15	-0.08	-0.11	0.16	-0.08	-0.04	0.16	-0.03
BladderIndependence	-0.03	0.05	-0.09	-0.01	0.06	-0.02	-0.01	0.06	-0.02	0.02	0.06	0.07
BowelIndependence	0.01	0.05	0.03	0.03	0.05	0.08	0.03	0.05	0.08	0.02	0.05	0.06
Mobility	-0.03	0.05	-0.07	-0.04	0.05	-0.10	-0.04	0.05	-0.10	-0.05	0.05	-0.13
Functional- EyeAudition	-0.04	0.09	-0.06	0.01	0.09	0.01	0.01	0.09	0.01	0.01	0.09	0.01
Functional- Physical	-0.11	0.07	-0.18	-0.08	0.07	-0.13	-0.08	0.07	-0.13	-0.07	0.07	-0.12
Functional- BehaviorSocial	-0.16	0.07	**-0.27***	-0.13	0.07	-0.22	-0.13	0.07	-0.22	-0.11	0.07	-0.19
Functional- Intellect.&Com.	0.07	0.09	0.11	0.12	0.09	0.19	0.12	0.09	0.19	0.18	0.09	0.28
Psychosocial-Cognitive				0.25	0.13	0.26	0.25	0.13	0.26	0.23	0.13	0.24
Psychosocial- Physical				0.11	0.12	0.13	0.12	0.13	0.13	0.14	0.13	0.16
Psychosocial- Social				0.02	0.13	0.02	0.02	0.13	0.02	-0.03	0.13	-0.03
Family Characteristics							-0.01	0.13	-0.01	-0.03	0.13	-0.03
ActivitiesParticipationMean										-0.32	0.18	-0.24

**p* < .05. ***p* < .01.

Dependent Variable: Quality of life

a. Predictors: ((Constant), Functional- Intellectual&Communication, Functional- EyeAudition, Functional- Physical, Functional- BehaviorSocial, BladderIndependence, BowelIndependence, Mobility, Gender, Age

b. Predictors: (Constant), Predictors in Model 1+ (Psychosocial- Social, Psychosocial-Cognitive, Psychosocial- Physical)

c. Predictors: (Constant), Predictors in Model 2+ Family Characteristics

d. Predictors: (Constant), Predictors in Model 3+ ActivitiesParticipationMean_SC

CHAPTER IV –DISCUSSION

The present study examined the nature of secondary conditions and quality of life of children, adolescents and adults with Spina bifida and to identify individual characteristics associated with their prediction and (QoL) using the ICF –CY (WHO, 2007) as frame of reference for analyses. Spina bifida is a complex neurogenetic disorder that can limit functioning of individuals, their activities and participation and quality of life. The core disability diagnosis is also a risk factor for secondary conditions that can be seen in individuals without the disability or with other disability diagnoses. Some of the secondary conditions individuals with Spina bifida experience include deficits in executive functioning, cognitive abilities, learning, social and language skills, as well as anxiety, depression, poor self-perception, or physical difficulties such as mobility. As any other secondary conditions, they are likely to co–occur. In this context, the biopsychosocial model of the ICF-CY (WHO, 2007) is used as a comprehensive holistic framework to group the secondary conditions and identify key factors that may be associated with the functioning and development of individuals with SB.

The first research question focused on the nature of the variables and relationships among demographic characteristics (age, gender, bladder and bowel independence, mobility), functional characteristics (4 Indexes derived from ABILITIES index), psychosocial characteristics (perceived social, physical and cognitive competence), secondary conditions (1-Body functions & structures, 2-Activities &Participation), and quality of life of individuals with Spina bifida. Overall, there were several significant relationships among variables ranging from r=.199 to r=.672 (p<.05).

Majority of the significant correlation coefficients were of small and medium strength at the p<.05 level. Expected significant relationships among age, bladder independence and bowel independence were at large strength (r >.50). The relatively small sample size may account for the fact that the relationship among variables was not high.

The second research question focused on the difference in the distribution of secondary conditions within the domains of the ICF-CY model and the rate of agreement between the two raters based on the rules at ICF-CY coding manual and steps used by Cieza et al. (2005, 2016). The items of the secondary conditions questionnaire are composed of single word descriptions such as “pain”, “math”, “and unemployment” making it difficult to link with a specific ICF –CY code. Most items could be classified with “other specified” or “unspecified” categories which Cieza et al. (2005) initially encouraged in order to avoid ambiguity of the results of the linking process. The lack of clarity of terms resulted in initial low rate of inter-rater agreement, which increased, from 19 % to 56% after reaching the decision to use 2nd level of coding (e.g. b62, d17, d77). In the latest rules again suggested by Cieza et al. (2016) this is enhanced by assigning the concept “not definable” with addition of general health (nd-gh), physical health (nd-ph), mental health (nd-mh), disability (nd-dis), functioning (nd-func), development (nd-dev). These steps carry the purpose of further clarification, however they also counteract with the rules set up at the ICF –CY manual, so not used in this study. Final model summarizing secondary conditions related to ICF-CY codes was composed of 16 items representing body functions (b), 1 item representing body structures (s), and 11 items characterizing activities and participation (p). The results of the linking process also reflect the fact that the majority of the secondary conditions are related with body functions and opens the way to improve the scale structure by wording it in detail. For example, the item “pain” on a scale of “never a problem to always a problem” may mean as “I have high tolerance of pain so it is usually not a problem” or “I do not experience much pain so it is usually not a problem”.

With regard to the third research question, the present study affirmed that demographic characteristics and functional abilities were significant predictors of the perceived level of problem/ difficulty with Secondary Conditions and Quality of Life in overall models. A multiple hierarchical linear regression was sequentially used to test the role of developmental (demographics, functional abilities, perceived competence) and family characteristics in predicting difficulty with Secondary Conditions and perceived Quality of Life. The functional abilities, age, gender, mobility, bowel and bladder control were entered at Block 1. Psychosocial characteristics as measured by the HARTER social, cognitive and physical self-perceived acceptance subscales are added in the Block 2, followed by adding the Family Characteristics in Block 3.

The strength of variables to predict the Body Functions component of the Secondary Conditions was assessed with Block 1 functional variables, revealing only age as a significant predictor of secondary conditions at the body functions component. For Block 2, entry of perceived physical, social and cognitive competence increased the total variance explained by the model, however only perceived cognitive competence significantly contributed to the model. Participants with higher perceived competence would thus have a lower level of perceived difficulty level with secondary conditions (e.g. bladder control, skin breakdown, breathing, etc.) It may also be that the more an individual believes in his/her cognitive abilities, the less difficulty/problem is reported in body functions such as bladder control, skin breakdown, and breathing. This is not likely to mean that perceived competence in cognitive abilities is a direct predictor of body function difficulty level. The individuals with higher cognitive competence may be focusing more of their attention in other areas or may come up with better ways of dealing with the difficulty or may simply not perceive those difficulties as problematic due to their positive focus in cognitive abilities. For Block 3, adding in Family Characteristics over and above the demographics, functional abilities and perceived competence variables maintained the significance of the overall model. Positive family perceptions and/or strong family connections were meaningful predictors of lower problem/ difficulty in Secondary Conditions-Body functions component. It should be noted that parents filled Survey of Family Characteristics. Their perception of the family characteristics is likely to be different than the children, adolescents or young adults' perception of the family characteristics and may not be accurately reflecting on their experiences with family and their secondary conditions. Positive statements of the survey would represent family strength, however using more surveys or contribution of the children, adolescents and young adults would likely provide a more complete perspective. Despite the statistical significance, parents having a positive viewpoint of the family may not be a good predictor of the individuals' perception of how much of a problem/ difficulty is their secondary condition.

The analysis preceded with another multiple hierarchical regression in which the role of predictor variables to predict Activity and Participation component of the Secondary Conditions was assessed. At the individual variable level, higher bladder control and lower level of functioning in Intellectual & Communication abilities were significant predictors for the activities and participation component. It was interesting to find that higher control of bladder was associated with a decrease in the perceived difficulty with secondary conditions Activities and Participation component. This was an unusual finding with no apparent documentation found in the literature.

The last question, the role of functional, psychosocial, and family variables ability to predict perceived Quality of Life of individuals with SB was assessed. Results of the hierarchical multiple regression with all four blocks entered indicated a significant overall model but an individual variable, Behavior-Social Skills was only significant when entered in Block 1. Then, higher mean scores in Behavior& Social skills functional abilities associated with more profound disability significantly predicted lower perceived Quality of Life. Adding in the psychosocial characteristics (perceived competence scales), family characteristics or the Activities and Participation on all the previous Blocks, did not have any significant impact on the model.

Limitations

Several limitations need to be considered with regard to the present study. First, the sample size for the multiple hierarchical regression analyses was relatively small which would affect the generalizability of the study (Pallack, 2010). Tabachnick and Fidell (2007, p. 123) suggest the sample size to be larger than eight times the number of independent variables plus fifty (N> 50+8m). Based on this formula, the sample size of this study should be larger than 140. The participant range of this study was between 88 to 110 individuals, resulting from using pairwise deletion.

The large age range of the participants (up to age 37) is another limitation. It is difficult to draw conclusion from the data, due to the wide range of developmental characteristics of individuals and their experiences. Thus, while younger individuals are just learning how to live with a disabling condition, the nature and extent of their secondary conditions and QoL may be different from older individuals who likely to develop skills to accommodate their needs.

The scales used in the study may also represent a limitation. Perceived Quality of Life was measured with response to a single item. Using a comprehensive scale may have revealed different results.

The Secondary conditions scale originally defined the three components of Physical, Academic, and Personal conditions. With this study, the items are linked with ICF –CY codes and then grouped into two components named 1- Body functions& Structures and 2- Activities& Participation based on coding by professionals. Using an empirical approach, such as factor analysis may have yielded different groups with different items, which in turn would influence the regression results. Also, a factor analysis of the secondary conditions items could reveal components different than both the original scale and the ICF-CY based scale. A similar limitation is pertaining to the use of the ABILITIES scale in which a total weighted score is available to summarize the results of assessment. In this study, an alternate approach to differentiating components would provide a more detailed perspective than the use of a total score, by dividing the scale into four components named Eye & Audition, Physical, Behavior & Social Skills, Intellectual Communication. Prior to deciding on these four indexes, factor analysis was tried resulting in different number of factor structures (3, 4, or 5 factors), but none of them provided indexes that were plausible. The division of the index into four groups based logic was thus used in this study by it may not be the most effective way to characterize the functional abilities on child and adults with Spina bifida.

Implications for School Psychology

In educational settings children and youth with Spina bifida are considered under the Individuals with Disabilities Education Act (IDEA) category of Orthopedic Impairment which includes many physical disabilities such as birth defects, cerebral palsy, amputations, fractures, burns, or physical impairments caused by disease. The IDEA category of Orthopedic Impairment may be underestimating the possible secondary conditions such as learning difficulties, or social-emotional challenges that co-occur with medical and physical condition. Considering the biopsychosocial approach, school staff especially the school psychologist and teachers should be well informed about the condition, modifications, accommodations, and interventions to help children with spina bifida navigate their educational environment and succeed.

School psychologists are the mental health and special education experts in school setting, so they are actively involved in the preparation of Individualized Education Plans and

504 Plans, coordinate with school staff and parents to apply evidence based interventions. As school psychologists, we can provide trainings for teachers and other school staff informing them of the etiology of spina bifida, what it is like to live with the condition, accommodations, its effect on learning and social and emotional well-being and evidence based interventions.

Besides training of school staff, school psychologists should also train students with spina bifida on their condition, strengths, and weaknesses and be able to self- advocate. With the right accommodations and support systems, students with spina bifida can be successful, have a rewarding educational and social life just like any other student. Reviews of the literature report positive interventions aimed to increase participation in domestic life including psychosocial and academic interventions focused on self-management, self- advocacy, goal attainment, and management. For example, Lindsay et al. (2014) investigated self-management interventions for children and youth with physical congenital or acquired physical disabilities between the years 1980 to 2012. With the rule of having a comparison group and at least one quantifiable health-related outcome only two intervention studies were found for the individuals between the ages of 8-18 with SB. The results indicated that self-management interventions are effective ways of improving health related outcomes and self-management. Steward et al. (2011), used a six month long (25 sessions) computer mediated peer support intervention for twenty-two adolescents with SB and cerebral palsy to provide them with a wider social network and connect them with peer sharing similar issues. It was also aimed to overcome their self-concept and isolation issues by psycho-educational sessions on “living independently, health concerns, bullying, making friends, career planning, traveling, sports, and building relationships”. The only significant improvement was on sense of community based on quantitative measures however; during interviews participants reported increased social network, social acceptance and self-confidence. They also mentioned the need for improved social networking with typically developing peers. A similar online intervention was also used by Stewart et al. (2011), in which both peer mentors and mentees reported positive reactions to the sessions. Stubberud et al. (2013) described a mindful therapy approach for adults with spina bifida myelomeningocele (SBM), using Goal Management Training for executive functions. The training used seven modules that included a) orientation to the program and defining absentmindedness, and slip-ups, b) stop the automatic pilot, c) working memory and present mindedness, d) teaching state goal, e) completing goals and making decisions, f) handling overwhelming tasks, and g) recognizing to stop to monitor output/ error correction. The participants' attention control significantly improved and they performed better on real-life multitasking situations.

Spina bifida not only affects the individual but also the caregivers and people surrounding the individual with the condition. It is a hard task to take care of an individual with congenital disorder. It creates stress as well as confusion on how to approach various situations. Parents of children with SB find it challenging to talk about puberty as their adolescents face challenges associated with Spina bifida. Through Spina bifida association Greenley et al. (2006), provided an individualized family self-management, hospital-based intervention that was conducted in two 60–90 min sessions over the span of three months. Key components of the program involved providing education for families about Spina bifida, teaching problem solving skills, homework and goal setting. Interventions to promote adaptive family functioning and better psychosocial outcomes are a continuing priority with this population focusing on emotion regulation, stress management, parenting skills to balance their protectiveness versus child's need for autonomy. In addition, interventions need to focus on family cohesion for higher paternal involvement, and supportive care programs focusing on the families' resilience and coping strategies as targets of interventions for families of individuals with SB. Despite the scarcity of literature on evidence-based interventions with families of individuals with spina bifida, results of the available studies are encouraging of parent involvement in intervention programs. The most effective program by O’Mahar et al. (2010) included 90-minute psycho-educational trainings for five days for youth with SB in grades 6 and 7. Topics included “knowledge on SB, sharing responsibility, mastery of self-care tasks, setting social goals, and self-concept”, through dynamic camp based intervention, using role plays, discussions, memory diary, verbal rehearsal, and problem solving approach. Holbein et al. (2013) replicated and extended O’Mahar’s et al. (2010) intervention, by using a new and larger sample of youth and adults with spina bifida who participated in a modified camp-based intervention targeting independence and social skills. They had 119 campers between the ages of 7 to 41 years of age and their parents were included at pre-and post intervention for progress monitoring purposes. The intervention aimed on goal attainment, management of health-related self-care, and independence through choosing a goal to work on through1 hour daily psycho-educational workshops and counselor monitoring. This study, with participants in three age groups (7-12, 13-18, 18+ years) was found effective for most campers in reaching their goals however; income status unexpectedly affected the results, with skill enhancement of low SES campers. In both studies, it was concluded that youth with SB did not spend enough time with their peers and that there was a need to include typically developing peers in those interventions.

There are a few evidence-based interventions that can be applied in school settings, and future studies should consider using a mindful therapy approach for children and adolescents as well. Furthermore, it is necessary to consider the effect of growth mindset and resilience factors on the educational and social outcomes of students with congenital disorders.

Conclusion

This study aimed to contribute to the literature to increase awareness of the impact of Spina bifida on the daily experiences and lives of children and adults with the condition. Such awareness and knowledge may further contribute to policy and practice innovations to creative interventions for accommodating their needs. Although there is a decline in the prevalence of SB in North America and Western Europe, there are still about 1,500 babies are born each year with the condition (CDC, 2016). It will continue to be a common birth defect due to the lack of knowledge on dietary prevention factors, and unawareness of folic acid supplement intake (Fletcher &Brei, 2010). Thus, there is a need to continue scientific research on the impact of Spina bifida on the development and functioning of children with this condition as well as the creation of intervention and support programs.

The findings of this study reinforce the use of the ICF-CY framework to document the nature of secondary conditions and providing a more comprehensive perspective of the impact of the condition on the lives of individuals with Spina bifida. Webb (2010) pointed the need for further studies on Spina bifida that would include younger and older age participants considering the importance of quality of life throughout lifespan. Future research would benefit from use of a perceived quality of life measure that takes into account the comprehensive experiences of individuals and their caregivers/ families. The outcome of such research could identify interventions for all age groups to address coping with functional difficulties and improve quality of life. There is a need for further research on how persons with spina bifida transition from childhood through adulthood. Through such research, t it may be possible to find and apply effective methods for transitions across the life span based on their developmental level and perception of difficulties. Future studies should include longitudinal studies of the developmental, psychological, and functional growth experiences of children, adolescents and adults with spina bifida. The use of multiple measures to assess their developmental, psychological and functioning characteristics linked to the ICF –CY can create stronger and unified findings to advance interventions and support.

APPENDIX

1. Secondary Conditions Questionnaire
2. Quality of Life item
3. ABILITIES Index
4. HARTER’s Perceived Competence Scales (Pre-K, Child, Teen, Adult)
    - Pictorial Scale of Perceived Competence and Social Acceptance for Young Children
    - HARTER’S Perceived Self-Competence Scale for Children
    - HARTER’S Perceive Self-Competence Scale for Teens
    - HARTER’S Perceive Self-Competence Scale for Adults
5. Survey of My Family Characteristics

1. Secondary Conditions Questionnaire

Physical	0 Never a problem	1 Usually not a problem	2 Sometimes not a problem	3 Sometimes a problem	4 Usually a problem	5 Always a problem
Pain						
Weight						
Bladder control						
Bowel control						
Skin breakdown						
Latex Allergy						
Pressure Sores						
Mobility						
Scoliosis						
Balance						
Strength						
Endurance						
Osteoporosis						
Breathing						
Sexuality						
Other						

Academic	0 Always Easy	1 Usually Easy	2 Sometimes Easy	3 Sometimes Difficult	4 Usually Difficult	5 Always Difficult
Math						
Reading						
Writing						
Spelling						
Problem Solving						
Learning						
Attention						
Other						

Personal	0 Always absent	1 Usually absent	2 Sometimes Absent	3 Sometimes Present	4 Usually present	5 Always present
Depression						
Low self esteem						
Lack of self						
Poor body image						
Other						

Secondary Conditions Questionnaire (continued)

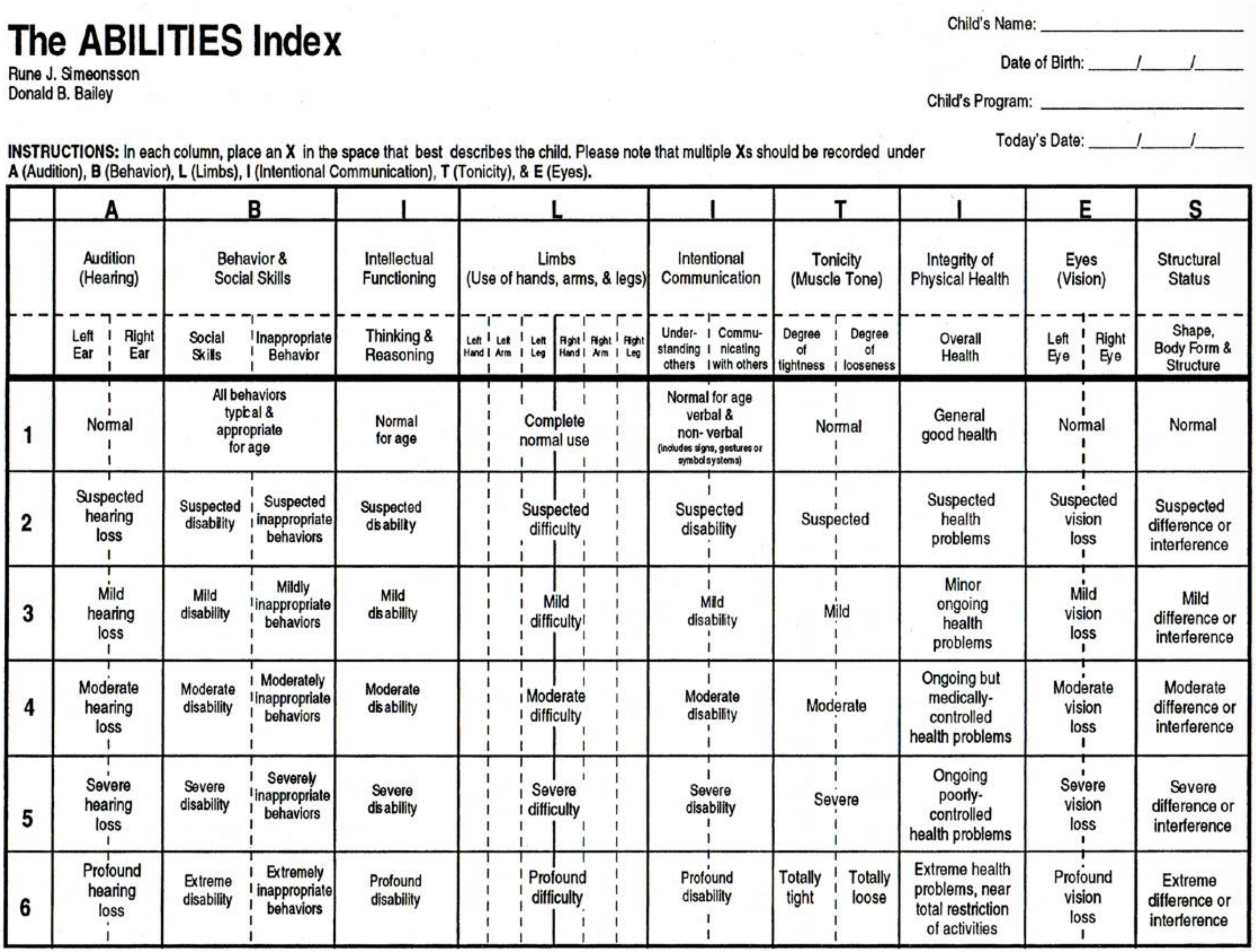
Social	0 Always absent	1 Usually absent	2 Sometimes Absent	3 Sometimes Present	4 Usually present	5 Always present
Isolation						
Exclusion						
Unemployment						
Sexuality						
Economic						
Family stress						
Lack of social						
Other						

2. Perceived Quality of Life Item

5. How would you describe your child’s Quality of Life, overall?

1	2	3	4
Poor	Fair	Good /Very Good	Excellent

3. The ABILITIES Index



The ABILITIES Index

Rune J. Simeonsson
Donald B. Bailey

Child's Name: ____________

Date of Birth: ____/____/____

Child's Program: ____________

Today's Date: ____/____/____

INSTRUCTIONS: In each column, place an **X** in the space that best describes the child. Please note that multiple **X**s should be recorded under **A** (Audition), **B** (Behavior), **L** (Limbs), **I** (Intentional Communication), **T** (Tonicity), & **E** (Eyes).

	A		**B**		**I**	**L**						**I**		**T**		**I**	**E**		**S**
	Audition (Hearing)		Behavior & Social Skills		Intellectual Functioning	Limbs (Use of hands, arms, & legs)						Intentional Communication		Tonicity (Muscle Tone)		Integrity of Physical Health	Eyes (Vision)		Structural Status
	Left Ear	Right Ear	Social Skills	Inappropriate Behavior	Thinking & Reasoning	Left Hand	Left Arm	Left Leg	Right Hand	Right Arm	Right Leg	Understanding others	Communicating with others	Degree of tightness	Degree of looseness	Overall Health	Left Eye	Right Eye	Shape, Body Form & Structure
1	Normal		All behaviors typical & appropriate for age		Normal for age	Complete normal use						Normal for age verbal & non- verbal (includes signs, gestures or symbol systems)		Normal		General good health	Normal		Normal
2	Suspected hearing loss		Suspected disability	Suspected inappropriate behaviors	Suspected disability	Suspected difficulty						Suspected disability		Suspected		Suspected health problems	Suspected vision loss		Suspected difference or interference
3	Mild hearing loss		Mild disability	Mildly inappropriate behaviors	Mild disability	Mild difficulty						Mild disability		Mild		Minor ongoing health problems	Mild vision loss		Mild difference or interference
4	Moderate hearing loss		Moderate disability	Moderately inappropriate behaviors	Moderate disability	Moderate difficulty						Moderate disability		Moderate		Ongoing but medically-controlled health problems	Moderate vision loss		Moderate difference or interference
5	Severe hearing loss		Severe disability	Severely inappropriate behaviors	Severe disability	Severe difficulty						Severe disability		Severe		Ongoing poorly-controlled health problems	Severe vision loss		Severe difference or interference
6	Profound hearing loss		Extreme disability	Extremely inappropriate behaviors	Profound disability	Profound difficulty						Profound disability		Totally tight	Totally loose	Extreme health problems, near total restriction of activities	Profound vision loss		Extreme difference or interference

4. HARTER'S Perceived Self-Competence Scales

Scored as ➔	Really True For me 1	Sort of True For me 2 BUT:	Sort of True For me 3	Really True For me 4
Subscales Used in this study	Pre-K & Grade 1-2	Child (Grade 3-8)	Teen (Age 13-18)	Adult (Age 20-60)
Peer	Peer 2 Has lots of friends 6 Stays overnight at friends 10 Has friends to play with 14 Has friends on playground 18 Gets asked to play by others 22 Eats dinner at friends'	Social 2. Hard/easy to make friends 8. A lot/fewer friends 14. Like more/have enough friends 20. Do things with others/alone 26. Wish more age-peers liked them 32. Popular/not very popular	Social 2 Find it hard to make friend 11 Have a lot of friends 20 Very hard to like 29 Popular with others their age 38 Are socially accepted	Sociability 2 Feel enjoyable to be with 14 Uncomfortable with new people 27 At ease with other people 39 Not very sociable
Physical Appearance	Physical Appearance 3 Good at swinging 7 Good at climbing 11 Can tie shoes 15 Good at skipping 19 Good at running 23 Good at hopping	Physical Appearance 4. Happy/or not with own looks 10. Happy/or not with height & wt 16. Wish their body was different 22. Wish physical appearance diff 28. Wish face or hair looked diff 34. Think are good-looking/not so	Physical Appearance 4 Not happy with way one looks 13 Wish their body were different 22 Wish physical appearance diff. 31 Think they are good looking 40 Really like their looks	Physical Appearance 6 Happy with way they look 19 Thinks not very good-looking 31 Like one's physical appearance 44 Unsatisfied with face or hair
Cognitive	Cognitive 1 Good at puzzles 5 Gets stars on papers 9 Knows names of colors 13 Good at counting 17 Knows alphabet 21 Knows first letter of name	Scholastic 1. Feel good at school work 7. Just as smart kids own age 13. Slow/quick doing school work 19. Forget/remember what learned 25. Do very well at classwork 31. Trouble figuring out answers	Scholastic 1. Feel just as smart as others 10 Slow in finishing school work 19 Do very well at classwork 28 Have trouble figuring answers 37 Are pretty intelligent	Intelligence 12 Feel stupid if don't understand 24 Feel they are intelligent 37 Not very intellectually capable 49 Feel just as smart as others

5. Survey of My Family Characteristics

Rune J. Simeonsson, 1987

1 My family has a sense of humor	0=Not at all like my family 1=Little like my family 2=Somewhat like my family 3=Like my family 4=Very much like my family
2 I am able to help my child learn	As Above
3 Family works to solves problems	As Above
4 Family members talk w/ each other	As Above
5 I feel good about my family	As Above
6 I can make a difference in things	As Above
7 I draw strength from religion	As Above
8 Family is caring & loving	As Above
9 I am can care for my children	As Above
10 Family able to settles differences	As Above
11 I know how to relax or unwind	As Above
12 Family appreciates eachother	As Above
13 Fam does stuff together outdoors	As Above
14 Fam. finds support in religion	As Above
15 I can handle my children behav.	As Above
16 Family supports each other	As Above
17 Family can organize to do work	As Above
18 Family members trusts eachother	As Above
19 I have positive view of life	As Above
20 Cultural important to family	As Above
21 Family talks things out	As Above
22 We have friends who care	As Above
23 Family spends time together	As Above
24 I know how children grow	As Above
25 Family knows how to get service	As Above
27 Family member accept each other	As Above
28 I have religious faith	As Above
29 Family eats meals together	As Above
30 I face & solve problems	As Above
31 We count on our relatives	As Above
32 Family see positives in crisis	As Above
33 Family has $ to do things	As Above
34 Often spends holidays together	As Above
35 Family has nicknames children	As Above

REFERENCES

Adriaansen, J., J., E., van Asbeck, F., W., A., Lindeman, E., van der Woude, L., H., V., de Groot, S., & Post, M., W., M. (2012). Secondary health conditions in persons with a spinal cord injury for at least 10 years: Design of a comprehensive long-term cross-sectional study. *Disability and Rehabilitation*, *35*, 1–7. doi:10.3109/09638288.2012.712196

Antolovich, G., C. & Wray, A., C. (2008). Habilitation of Children and Young Adults with Spina Bifida In F. M. Ozek, M., Cinalli, G., Maixner, W., J. (Ed.), *Spina Bifida Management and Outcome* (pp. 341-348). Milan: Springer. doi: 10.1007/978-88-470-0651-5_28

Bailey, D. B., Simeonsson, R. J., Buysse, V., & Smith, T. (1993). Reliability of an index of child characteristics. *Developmental Medicine and Child Neurology*. *35*, 806- 815.

Barf, H. A, Post, M. W. M., Verhoef, M., Jennekens-Schinkel, A, Gooskens, R., H., J., M., & Prevo, A., J., H. (2009). Restrictions in social participation of young adults with spina bifida. *Disability and Rehabilitation*, *31*(11), 921–927. doi:10.1080/09638280802358282

Barnes, M., A., Raghubar, K., P., English, L., Williams, J., M., Taylor, H., & Landry, S. (2014). Child longitudinal mediators of achievement in mathematics and reading in typical and atypical development, *Journal of Experimental Psychology*, *119*, 1–16.

Björck-Åkesson E., Wilder J., Granlund M., Pless M., Simeonsson R., Adolfsson M., Almqvist L., Augustine L., Klang N., Lillvist A. (2010). The International Classification of Functioning, Disability and Health and the version for children and youth as a tool in child habilitation/early childhood intervention--feasibility and usefulness as a common language and frame of reference for practice. *Disability and Rehabilitation*, *32*, 125–138.

Brislin, D., C. (2008). Reaching for Independence: Counseling Implications for Youth With Spina Bifida. *Journal of Counseling & Development*, *86*(1), 34–38. doi:10.1002/j.1556-6678.2008.tb00623.x

Buffard, L., M., van den Berg-Emons, R., J., G., van Meeteren, J., Stam, H., J., & Roebroeck, M. E. (2009). Lifestyle, participation, and health-related quality of life in adolescents and young adults with myelomeningocele. *Developmental Medicine and Child Neurology*, *51*(11), 886–894. doi:10.1111/j.1469-8749.2009.03293.x

Burmeister, R., Hannay, H. J., Copeland, K., Fletcher, J. M., Boudousquie, A., & Dennis, M. (2005). Attention problems and executive functions in children with spina bifida and hydrocephalus. *Child Neuropsychology*, *11*(3), 265–283. doi:10.1080/092970490911324

Castro, S., Ferreira, T., Dababnah, S., & Pinto, A. I. (2013). Linking autism measures with the ICF-CY: Functionality beyond the borders of diagnosis and interrater agreement issues. *Developmental Neurorehabilitation*, *16*(5), 321–31. doi:10.3109/17518423.2012.733438

Centers for Disease Control and Prevention, National Center on Birth Defects and Developmental Disabilities (2016, Oct. 17). *Spina Bifida Basics.* Retrieved from https://www.cdc.gov/ncbddd/spinabifida/facts.html

Cieza, A., Stucki,G., Weigl, M., Disler, P., Jackel, W., & van der Linden, S. (2004). ICF Core Sets for low back pain. *Journal of Rehabilitation Medicine, 44*, 69-74.

Cieza, A., Geyh, S., Chatterji, S., Kostanjsek, N., Ustun, B., & Stucki, G. (2005). ICF Linking rules: An update based on lessons learned. *Journal of Rehabilitation Medicine, 37*, 212-218.

Cieza, A., Fayed, N., Bickenbach, J., & Prodinger, B. (2016). Refinements of the ICF Linking Rules to strengthen their potential for establishing comparability of health information. *Disability and Rehabilitation*, *8288*, 1–10. http://doi.org/10.3109/09638288.2016.1145258

Devine, K., A, Holmbeck, G., N., Gayes, L., & Purnell, J., Q. (2012). Friendships of children and adolescents with spina bifida: social adjustment, social performance, and social skills. *Journal of Pediatric Psychology*, *37*(2), 220–31. doi:10.1093/jpepsy/jsr075

Ellignsen, K. (2011). *Deriving Developmental Code Sets from The International Classification of Functioning, Disability and Health, for Children and Youth (ICF-CY).* Retrieved from UNC electronic theses and dissertations collection.

English, L., H., Barnes, M., A, Taylor, H., B., & Landry, S., H. (2009). Mathematical development in spina bifida. *Developmental Disabilities Research Reviews*, *15*(1), 28–34. doi:10.1002/ddrr.48

Essner, B., S., & Holmbeck, G., N., (2010). The impact of family, peer, and school contexts on depressive symptoms in adolescents with spina bifida. *Rehabilitation Psychology*, *55*(4), 340–50. doi:10.1037/a0021664

Flanagan, A., Kelly, E. H., & Vogel, L. C. (2013). Psychosocial outcomes of children and adolescents with early-onset spinal cord injury and those with spina bifida. *Pediatric Physical Therapy*, *25*(4), 452–9. doi:10.1097/PEP.0b013e3182a5d35c

Fletcher, J. M., & Brei, T. J. (2010). Introduction: Spina Bifida—A Multidisciplinary Perspective. *NIH Public Access*, *16*(1), 1–5. http://doi.org/10.1002/ddrr.101.

Friedman, D., Holmbeck, G. N., Jandasek, B., Zukerman, J., & Abad, M. (2004). Parent functioning in families of preadolescents with spina bifida: Longitudinal implications for child adjustment. *Journal of Family Psychology, 18*(4), 609–619. doi:10.1037/0893-3200.18.4.609

Gan, S., M., Tung, L., C., Yeh, C., H., Chang, H., Y., & Wang, C., H. (2014). The ICF-CY-based structural equation model of factors associated with participation in children with autism. *Dev Neurorehabil*, *17*(1), 24–33. doi:10.3109/17518423.2013.835357

Gras, R. M. L., Berná, J. C., Lopez, P., S. (2009). Thinking Styles and Coping When Caring for a Child with Severe Spina Bifida, *J Dev Phys Disabil 21:*169–183.

Guilcher, S., J., T., Craven, B., C., Lemieux-Charles, L., Casciaro, T., McColl, M., A., & Jaglal, S., B. (2012). Secondary health conditions and spinal cord injury: An uphill battle in the journey of care. *Disability and Rehabilitation*, *35*, 1–13. doi:10.3109/09638288.2012.721048

Greenley, R. N., Coakley, R. M., Holmbeck, G. N., Jandasek, B., & Wills, K. (2006). Condition-related knowledge among children with spina bifida: Longitudinal changes and predictors. Journal of Pediatric Psychology, 31, 828–839

Harter, S. (1982). The Perceived Competence Scale for Children. *Child Development*, *53*(1), 87-97.

Harter, S. (1985). The Self-Perception Profile for Children: Revision of the Perceived Competence Scale for Children. Denver, CO: University of Denver.

Harter, S. (1988). Manual for the Adolescent Self-Perception Profile. Denver, CO. Retrieved from https://portfolio.du.edu/SusanHarter/page/44210.

Harter, S., & Pike, R. (1984). The Pictorial Scale of Perceived Competence and Social Acceptance for Young Children. *Child Development*, *55*(6), 1969-1982. doi:10.2307/1129772

Harter, S. (2012). Self-Perception Profile For Adolescents: Manual And Questionnaires. Retrieved from https://portfolio.du.edu/SusanHarter/page/44210

Holmbeck, G., N., & Devine, K., A. (2010). Psychosocial and family functioning in spina bifida. *Developmental Disabilities Research Reviews*, *16*(1), 40–6. doi:10.1002/ddrr.90

Holmbeck, G., N., Greenley, R., N., Coaklet, R., M., Greco, J. & Hagstrom, J. (2006). Family Functioning in Children and Adolescents with Spina Bifida : An Evidence-based Review of Research and Interventions, *27*(3):249-77

Holbein, C. E., Murray, C. B., Psihogios, A. M., Wasserman, R. M., Essner, B. S., O'Hara, L. K., & Holmbeck, G. N. (2013). A camp-based psychosocial intervention to promote independence and social function in individuals with spina bifida: moderators of treatment effectiveness. *Journal of Pediatric Psychology*, *38*(4), 412–24. doi:10.1093/jpepsy/jst003

Horton, T. V., & Wallander, J. L. (2001). Hope and social support as resilience factors against psychological distress of mothers who care for children with chronic physical conditions. *Rehabilitation Psychology*, *46*(4), 382–399. doi:10.1037//0090-5550.46.4.382

Hwang, A. -W., Liao, H. -F., Granlund, M., Simeonsson, R. J., Kang, L.-J., & Pan, Y.-L. (2014). Linkage of ICF-CY codes with environmental factors in studies of developmental outcomes of infants and toddlers with or at risk for motor delays. *Disability and Rehabilitation*, *36*(2), 89–104. doi:10.3109/09638288.2013.777805

Janeslätt, G., Kottorp, A., & Granlund, M. (2014). Evaluating intervention using time aids in children with disabilities. *Scandinavian Journal of Occupational Therapy*, *21*(3), 181–90. doi:10.3109/11038128.2013.870225

Kelly, E., H., Altiok, H., Gorzkowski, J., A., Abrams, J., R., & Vogel, L., C. (2011). How does participation of youth with spina bifida vary by age? *Clinical Orthopaedics and Related Research*, *469*(5), 1236–1245. doi:10.1007/s11999-010-1693-x

Krasuska, M., Riva, S., Fava, L., von Mackensen, S., & Bullinger, M. (2012). Linking Quality-of-Life Measures Using the International Classification of Functioning, Disability and Health and the International Classification of Functioning, Disability and Health–Children and Youth Version in Chronic Health Conditions. *American Journal of Physical Medicine & Rehabilitation*, *91*(2), S74–S83. doi:10.1097/PHM.0b013e31823d4f35

Landry, S. H., Taylor, H. B., Swank, P. R., Barnes, M., & Juranek, J. (2013). Longitudinal mediators of social problem solving in spina bifida and typical development. *Rehabilitation Psychology*, *58*(2), 196–205. doi:10.1037/a0032500

Lars, W. (1995). Harter's Self Perception Profile for Adolescents: Reliability, Validity and Evaluation of the Question Format. *Journal of Personal Assessment*, 65(1), 100-116.

Law, M., King, G., King, S., Kertoy, M., Hurley, P., Rosenbaum, P. et al. (2006). Patterns of participation in recreational and leisure activities among children with complex physical disabilities. *Developmental Medicine & Child Neurology, 48,* 337–342.

Law, M., Anaby, D., Imms, C., Teplicky, R., & Turner, L. (2015). Improving the participation of youth with physical disabilities in community activities: An interrupted time series design. *Australian Occupational Therapy Journal*, *62(2):* 105-15. doi:10.1111/1440-1630.12177

Lennon, J., M., Klages, K., L., Amaro, C., M., Murray, C., B., & Holmbeck, G., N. (2014). Longitudinal Study of Neuropsychological Functioning and Internalizing Symptoms in Youth With Spina Bifida: Social Competence as a Mediator, 1–13.

Lindquist, B., Persson, E., K., Uvebrant, P., & Carlsson, G. (2008). Learning, memory and executive functions in children with hydrocephalus. *Acta Paediatrica), 97*(5), 596–601. doi:10.1111/j.1651-2227.2008.00747.x

Lindsay, S., Kingsnorth, S., Mcdougall, C., & Keating, H. (2014). A systematic review of self-management interventions for children and youth with physical disabilities. *Disability and Rehabilitation*, *36*(4), 276–88. doi:10.3109/09638288.2013.785605

Lomax-Bream, L., E., Barnes, M., Copeland, K., Taylor, H., B., & Landry, S., H. (2007). The Impact of Spina Bifida on Development Across the First 3 Years. *Developmental Neuropsychology*, *31*(1), 1–20. doi:10.1207/s15326942dn3101_1

Kelly, E., H., Altiok, H., Gorzkowski, J., A., Abrams, J., R., & Vogel, L., C. (2011). How does participation of youth with spina bifida vary by age? Clinical Orthopaedics and Related Research, 469(5), 1236–1245. doi:10.1007/s11999-010-1693-x

Krahn, G. L., Suzuki, R., & Horner-Johnson, W. (2009). Self-rated health in persons with spinal cord injury: Relationship of secondary conditions, function and health status. *Quality of Life Research*, *18*(5), 575–584. doi:10.1007/s11136-009-9477-z

McCubbin, H. I., & Patterson, J. M. (1983). The family stress process: The Double ABCX model of adjustment and adaptation. In H. I. McCubbin, M. B. Sussman, & J. M. Patterson (Eds.), Social stress and the family: Advances and developments in family stress theory and research (pp. 7–37). New York: Haworth.

McKernon, W., L., Holmbeck, G., N., Colder, C., R., Hommeyer, J., S., Shapera, W., & Westhoven, V. (2001). Longitudinal Study of Observed and Perceived Family Influences on Problem-Focused Coping Behaviors of Preadolescents With Spina Bifida, *Journal of Pediatric Psychology*, *26(1)*, 41–54.

McMillen, J., S., (1997). Child and family characteristics as predictors of child's quality of life (Doctoral dissertation). Retrieved from UNC Chapel Libraries online database.

McStay, R. L., Trembath, D., & Dissanayake, C. (2014). Stress and Family Quality of Life in Parents of Children with Autism Spectrum Disorder: Parent Gender and the Double ABCX Model. *Journal of Autism and Developmental Disorders*, 3101–3118. doi:10.1007/s10803-014-2178-7

Messer B., & Harter. S., (1986). The Self-Perception Profile For Adults: Manual And Questionnaires. Denver, CO. Retrieved from https://portfolio.du.edu/SusanHarter/page/44210

Messer B., & Harter. S., (2012). The Self-Perception Profile For Adults: Manual And Questionnaires 2012 Revision of 1986 manual. Retrieved from https://portfolio.du.edu/SusanHarter/page/44210

Mobley, C., E., Harless, L., S., & Miller, K., L. (1996). Self-perceptions of preschool children with spina bifida. *Journal of Pediatric Nursing*, *11*(4), 217–24. doi:10.1016/S0882-5963(96)80094-7

National Institute of Neurological Disorders and Stroke (NINDS, 2015). Spina Bifida. Retrieved from http://www.ninds.nih.gov/disorders/spina_bifida/spina_bifida.htm

O'Mahar, K., Holmbeck, G. N., Jandasek, B., & Zukerman, J. (2010). A camp-based intervention targeting independence among individuals with spina bifida. *Journal of Pediatric Psychology*, *35*(8), 848–56. doi:10.1093/jpepsy/jsp125

Pakenham, K. I., Sofronoff, K., & Samios, C. (2004). Finding meaning in parenting a child with Asperger syndrome: Correlates of sense making and benefit finding. *Research in Developmental Disabilities*, *25*(3), 245–264. doi:10.1016/j.ridd.2003.06.003

Pallant, J. (2010). *SPSS survival manual : a step by step guide to data analysis using SPSS* (4th ed.). Maidenhead, Berkshire, England: McGraw Hill.

Paynter, J., Riley, E., Beamish, W., Davies, M., & Milford, T. (2013). The double ABCX model of family adaptation in families of a child with an autism spectrum disorder attending an Australian early intervention service. *Research in Autism Spectrum Disorders*, *7*(10), 1183–1195. doi:10.1016/j.rasd.2013.07.006

Petersson, C., Simeonsson, R. J., Enskar, K., & Huus, K. (2013). Comparing children's self-report instruments for health-related quality of life using the International Classification of Functioning, Disability and Health for Children and Youth (ICF-CY). *Health and Quality of Life Outcomes*, *11*, 75. doi:10.1186/1477-7525-11-75

Pope, A. M., & Tarlov, A. R. (Eds.). (1991). Disability in America: Toward a national agenda for prevention. Washington, DC: National Academy Press.

Rendeli, C., Ausili, E., Tabacco, F., Caliandro, P., Aprile, I., Tonali, P., … Padua, L. (2005). Assessment of health status in children with spina bifida. *Spinal Cord : The Official Journal of the International Medical Society of Paraplegia*, *43*(4), 230–235. doi:10.1038/sj.sc.3101707

Rimmer, J., H., Chen, M-De, Hsieh, K. (2011). A Conceptual Model for Identifying, Preventing, and Managing Secondary Conditions in People With Disabilities. *Physical Ther., 91:1728-1739, doi:10.2522/ptj.20100410.*

Rintoul, N. E., Sutton, L. N., Hubbard, A. M., Cohen, B., Melchionni, J., Pasquariello, P. S., & Adzick, N. S. (2002). A new look at myelomeningoceles: Function level, vertebral level, shunting, and the implications for fetal development. *Pediatrics, 109,* 409-414.

Rofail, D., Maguire, L., Kissner, M., Colligs, A., & Abetz-Webb, L. (2014). Health-related quality of life is compromised in individuals with spina bifida: results from qualitative and quantitative studies. *European Journal of Obstetrics & Gynecology and Reproductive Biology*, *181*, 214–222. doi:10.1016/j.ejogrb.2014.07.048

Saloviita, T., Itälinna, M., & Leinonen, E. (2003). Explaining the parental stress of fathers and mothers caring for a child with intellectual disability: A Double ABCX model. *Journal of Intellectual Disability Research*, *47*(4-5), 300–312. doi:10.1046/j.1365-2788.2003.00492.x

Sarriá, E., & Brioso, A. (2014). Family quality of life and psychological well-being in parents of children with autism spectrum disorders: A double ABCX model. *Journal of Intellectual Disability Research*, *58*(may), 442–458. doi:10.1111/jir.12042

Sawin, K. J., & Bellin, M. H. (2010). Quality of life in individuals with spina bifida: A research update. *Developmental Disabilities Research Reviews*, *16*(1), 47–59. doi:10.1002/ddrr.96

Schoenmakers, M. A. G. C., Uiterwaal, C. S. P. M., Gulmans, V. A. M., Gooskens, R. H. J. M., & Helders, P. J. M. (2005). Determinants of functional independence and quality of life in children with spina bifida. *Clinical Rehabilitation*, *19*(6), 677–685. doi:10.1191/0269215505cr865oa

Simeonsson, R. J. (1987). Survey of My Family Characteristics. Chapel Hill, NC: Frank Porter Graham Child Development Center.

Simeonsson, R. J., & Bailey, D. B. (1991). The ABILITIES Index. Retrieved from http://fpg.unc.edu/node/365

Simeonsson R., J., Bailey, D., B., Scandlin, D., Huntington, G., S., Roth, M. (1999). Disability, Health, Secondary Conditions and Quality of Life: Emerging Issues in Public Health. In: Simeonsson, R., J., McDevitt L., N., editors. Issues in Disability and Health: The Role of Secondary Conditions and Quality of Life. Chapel Hill: University of North Carolina Press, pp. 239–55.

Simeonsson R., J., & Leskinen, M. (1999). Disability, Secondary Conditions and Quality of Life: Conceptual Issues. In: Simeonsson, R., J., McDevitt L., N., editors. Issues in Disability and Health: The Role of Secondary Conditions and Quality of Life. Chapel Hill: University of North Carolina Press, pp. 52-72.

Simeonsson, R., J., McMillen, J., S., & Huntington, G., S. (2002). Secondary conditions in children with disabilities: spina bifida as a case example. *Mental Retardation and Developmental Disabilities Research Reviews*, *8*(3), 198–205. doi:10.1002/mrdd.10038

Singh, D. K., 2003. Families of Children with Spina Bifida: A Review. *Journal of Developmental & Physical Disabilities. 15*(1), 37-55. doi: 10.1023/A:1021452220291

Sommer, R., Bullinger, M., Rohenkohl, A., Quitmann, J., & Brütt, A. L. (2015). Linking a short-stature specific health-related quality of life measure (QoLISSY) to the International Classification of Functioning – Children and Youth (ICF-CY). *Disability and Rehabilitation*, *37*(5), 439–446. doi:10.3109/09638288.2014.923528

National Center on Birth Defects and Developmental Disabilities, Centers for Disease Control and Prevention (2016, October 17). Spina bifida basics. Retrieved from https://virtuallab.unc.edu/vpn/index.html

Stewart, M., Barnfather, A., Magill-Evans, J., Ray, L., & Letourneau, N. (2011). Brief report: an online support intervention: perceptions of adolescents with physical disabilities. *Journal of Adolescence*, *34*(4), 795–800. doi:10.1016/j.adolescence.2010.04.007

Stubberud, J., Langenbahn, D., Levine, B., Stanghelle, J., & Schanke, A., K. (2013). Goal management training of executive functions in patients with spina bifida: a randomized controlled trial. *Journal of the International Neuropsychological Society : JINS*, *19*(6), 672–85. doi:10.1017/S1355617713000209

Tabachnick, B. G., Fidell, L. (2007). *Using multivariate statistics* (5th ed.). Boston: Pearson/Allyn & Bacon.

Tuminello, E. R., Holmbeck, G. N., & Olson, R. (2012). Executive functions in adolescents with spina bifida: relations with autonomy development and parental intrusiveness. *Child Neuropsychology : A Journal on Normal and Abnormal Development in Childhood and Adolescence*, *18*(2), 105–24. http://doi.org/10.1080/09297049.2011.590470

The North Carolina Office on Disability and Health (2003). Growing up with Spina Bifida: What we have learned. Retrieved from http://fpg.unc.edu/sites/fpg.unc.edu/files/resources/reports-and-policy-briefs/NCODH_SpinaBifida.pdf

The WHOQOL Group (1995a). The World Health Organization Quality of Life assessment (WHOQOL): Position paper from the World Health Organization. Soc. Sci. Med., 41, 1403.

Tuminello, E., R., Holmbeck, G., N., & Olson, R. (2012). Executive functions in adolescents with spina bifida: relations with autonomy development and parental intrusiveness. *Child Neuropsychology: A Journal on Normal and Abnormal Development in Childhood and Adolescence*, *18*(2), 105–24. doi:10.1080/09297049.2011.590470

Verhoef, M., Post ., M., W., Barf, H., A., van Asbeck F., W, Gooskens R., H., Prevo, A., J., (2007). Perceived health in young adults with spina bifida. *Developmental Medicine & Child Neurology; 49*: 192–97.

Zabel, T. A., Jacobson, L. a, Zachik, C., Levey, E., Kinsman, S., & Mahone, E. M. (2011). Parent- and self-ratings of executive functions in adolescents and young adults with spina bifida. *The Clinical Neuropsychologist*, *25*(6), 926–41. doi:10.1080/13854046.2011.586002

Zukerman, J., M., Devine, K., A, & Holmbeck, G., N. (2011). Adolescent predictors of emerging adulthood milestones in youth with spina bifida. *Journal of Pediatric Psychology*, *36*(3), 265–76. doi:10.1093/jpepsy/jsq075

Webb, T. S. (2010), Optimizing health care for adults with spina bifida. Dev Disabil Res Revs, 16: 76–81. doi:10.1002/ddrr.99

World Health Organization (2007). International classification of functioning, disability and health: children & youth version: ICF-CY. Retrieved from *apps.who.int/iris/bitstream/10665/43737/1/9789241547321_eng.pdf*