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Health Concerns of Adolescents and Adults with Spina Bifida

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Abstract

The Transitional and Lifelong Care (TLC) program is a multidisciplinary clinical service providing comprehensive, coordinated care to adolescents and adults with spina bifida (SB). This study sought to identify the most common health concerns among this population at initial consultation and identify patient factors related to these concerns to ultimately improve health care delivery. A retrospective chart review was performed. Descriptive analyses were completed and associations between concerns and patient factors were determined using Spearman’s rho correlation coefficients. A multiple linear regression was performed to estimate the relationship between patient factors and total number of concerns reported. Ninety-four patient charts were reviewed (Mean age=29.04 ± 13.8 years). On average, patients reported 9 health concerns with care coordination being the most prevalent concern. Patient factors were not related to specific or total number of health concerns. Individualized, coordinated care and a medical home are warranted for adults with SB.

Keywords: spina bifida, health concerns, health care transition, youth, adolescent, young adult, adult, multidisciplinary health care, care coordination

Summary for Lay Audience

In recent decades, an increasing number of people with spina bifida (SB), a complex childhood-onset disability, are surviving well into adulthood, largely due to medical and technological advances in health care (Lorber, 2008; Mitchell et al., 2004; Shin et al., 2012). To ensure optimal health across the lifespan, adolescents and adults with SB require coordinated and planned health care transition services as they graduate from paediatric to adult health care (Binks, Barden, Burke, & Young, 2007; Fremion & Dosa, 2019). The Transitional and Lifelong Care (TLC) Program at Parkwood Institute in London, Ontario, is a program unique to Southwestern Ontario designed to fill this specific gap between paediatric and adult health care. The program provides ongoing multidisciplinary coordination of care and a 'medical home' for people with SB and other childhood-onset disabilities. However, like many other transitional models, the TLC program has not yet been evaluated or thoroughly studied (Bennett, Towns, & Steinbeck, 2005; Grant, Pan, & Grant, 2011; Mcdonagh, 2007). To improve the services provided by the TLC program and determine whether implementation of this model can improve the transition and standard of care received by adolescents and adults with SB, more needs to be known about the patients served by the program. The purpose of the present study was to identify the most common health care concerns or challenges that TLC patients with SB face and determine whether there are recognizable patient factors that are consistently related to these concerns. It is expected that this research will support future program evaluation and contribute to improvements in targeted health care delivery for TLC patients with SB, ultimately resulting in improved patient experience and outcomes.

Co-Authorship Statement

I would like to thank my co-authors, Dr. Laura Brunton and Dr. Caitlin Cassidy, who were instrumental in the conceptualization, design, and inception of this research, and who provided supervision, ongoing consultation, and assistance with statistical analyses and manuscript preparation. I would also like to thank Dr. Jacquelyn Marsh for her support with the statistical analyses and with the final manuscript.

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Chapter 1: Introduction and Background

Problem Statement

Where life expectancy was once poor, medical advances in the last several decades have resulted in an estimated 75% of people with spina bifida (SB), a complex neural tube disability originating in-utero, surviving well into adulthood (Bowman, McLone, Grant, Tomita, & Ito, 2001; Lorber, 2008; Mitchell et al., 2004; Shin et al., 2012). Because paediatric services typically cap between the ages of 18 and 21, adolescents with SB must face the challenge of transitioning to adult-oriented health care and navigating an ill-equipped health care environment throughout adulthood (Binks et al., 2007; Mennito & Clark, 2010).

The paediatric health care system typically houses specialized multidisciplinary clinics designed to address the needs of patients with complex health conditions (Binks et al., 2007; Liptak & Samra, 2010). In contrast, the adult health care system typically lacks coordinated, multidisciplinary care, and is frequently described as fragmented, resource-constrained, and lacking knowledgeable and interested health care professionals that can effectively address complex health care needs related to childhood conditions (Binks et al., 2007). Considering the complex nature of SB, it is unsurprising that the transition from paediatric to adult health care systems is associated with several negative outcomes, including difficulty accessing health and social services, higher rates of hospital admissions, and a decline in health status over time (Liptak et al., 2016; Okumura, Hersh, Hilton, & Lotstein, 2013; Young, Anselmo, Burke, Mccormick, & Mukherjee, 2014).

To ensure optimal health across the lifespan, adolescents and adults with SB require coordinated and planned health care transition at the individual level, as well as access to programming that addresses the common faults and lack of ongoing multidisciplinary care in the adult health care system (Binks et al., 2007; Fremion & Dosa, 2019). The Transitional and Lifelong Care (TLC) Program at Parkwood Institute in London, Ontario, is a unique program designed to address this specific gap. Unlike many other transition programs, the TLC program provides long-term coordination of care (or a “medical home”) for patients with SB, in addition to its transition services (st. Joseph’s Health Care London, n.d.). The TLC program provides access to knowledgeable clinicians and coordination of services, including assessment, intervention, and consultation, in one integrated and multidisciplinary care environment. Unfortunately, like many other transitional models, the TLC program has not yet been evaluated, despite the urgent need for formal assessment of transition services (Bennett et al., 2005; Grant et al., 2011; Mcdonagh, 2007). To improve the services provided by the TLC program and determine whether implementation of this model can improve the transition and standard of care received by adolescents and adults with SB in other settings, more needs to be known about the patients serviced by the program, including the most pressing challenges they face.

Literature Review

Spina Bifida

SB is one of the most common permanently disabling conditions in humans. (Mitchell et al., 2004). It is a non-progressive defect that results from incomplete closure of the neural tube during the first few weeks of embryonic development (Au, Ashley-Koch, & Northrup, 2010; Mitchell et al., 2004). Without normal closure of the neural tube, a variety of consequences may occur, including deficient development of the posterior elements of the vertebrae in a portion of the spinal column, allowing for the protrusion of the contents of the spinal canal (Fletcher & Brei, 2010). The most common form of SB is myelomeningocele, in which meninges and neural elements protrude through the spinal defect, necessitating neonatal surgical repair (Fletcher & Brei, 2010; Liptak & Dosa, 2010). The clinical spectrum of SB ranges from mild to severe neurological impairment, with severity determined by several factors, including the size and location of the spinal cord malformation, the degree of spinal cord or nerve involvement in the defect, and whether the opening in the spine is covered (Fletcher & Brei, 2010; Liptak & Dosa, 2010). The clinical consequences of SB are largely related to the degree of nervous system abnormalities resulting from the neural tube defect (Liptak & Dosa, 2010). Brain abnormalities, such as hydrocephalus and Chiari type II malformation, may occur and can be associated with learning disabilities, executive dysfunction, strabismus, precocious puberty, and/or epilepsy (Liptak & Dosa, 2010). The spinal cord malformation itself leads to a spectrum of sensory and motor impairments below the lesion level (located in the thoracic, lumbar, or sacral spine), and often to the development of neurogenic bowel and bladder dysfunction (Liptak & Dosa, 2010). The bony vertebral anomalies may lead directly to musculoskeletal consequences, including scoliosis and kyphosis (Liptak & Dosa, 2010; Liptak, Garver, & Dosa, 2013). In most cases of myelomeningocele, surgical closure of the defect is initiated shortly after birth (though recent advances have resulted in more frequent consideration of pre-natal fetal surgical repair (Rintoul et al., 2002)), and managing SB typically involves medical intervention throughout the lifespan (Mitchell et al., 2004). This may include continuous specialty care from urology, neurosurgery, orthopedics, and physical medicine and rehabilitation, as well as access to therapy services and support from psychology and assistive technology (Liptak & Dosa, 2010; Seeley & Lindeke, 2017).

The prevalence of SB differs by region and is influenced by race and ethnicity (Mitchell et al., 2004). Although the exact cause in each case of SB is often unknown, the potential causes are diverse and include both environmental and genetic factors (Liptak & Dosa, 2010; Mitchell et al., 2004). One critical such factor is folic acid supplementation during pregnancy (Mitchell et al., 2004). Adequate folic acid early in pregnancy (during early embryonic development) can reduce the risk of neural tube defects (Mitchell et al., 2004). In recent decades, in regions where there is mandatory legislation enforcing folic acid fortification in foods, there has been a vast (estimated 50% (Blencowe, Cousens, Modell, & Lawn, 2010)) reduction in the prevalence of neural tube defects, such as SB (Atta et al., 2016). Higher rates of prenatal screening and diagnosis of SB may have also contributed to this decline (Saavedra, MacLellan, & Gray, 2018). Even so, the lowest prevalence estimate for SB is 38.70 per 100 000 live births, stillbirths, and terminated pregnancies in North America (Atta et al., 2016).

Morbidity and Mortality in Adulthood

Although life expectancy for people with SB has increased, the population is at increased risk for excess morbidity and early mortality in adulthood (Liptak et al., 2013, 2016; Mitchell et al., 2004; Oakeshott, Hunt, Poulton, & Reid, 2009). Adults with SB continue to experience complex, multi-system disorders from childhood and adolescence throughout their lifespan (Liptak et al., 2013, 2016). Adults with SB experience co-morbid conditions and medical complexities that stem directly from their SB, as well as complications that may be indirectly related to SB, such as issues with infertility, poor mental health, obesity, and skin breakdown (Liptak et al., 2013, 2016). Moreover, adults with SB are at risk for health concerns and chronic health conditions common in the general population, such as hypertension, diabetes, and cancer (Liptak et al., 2013). The consequences of SB further extend beyond the physical body, such that specific learning disabilities or cognitive challenges and physical limitations that persist from childhood impact independence and educational, social, and employment opportunities in adulthood (Liptak et al., 2013, 2016). As such, a similar level of health care service and comprehensive support must exist throughout adulthood as in childhood and adolescence (Liptak et al., 2016). However, this is often not the case, and unsurprisingly, many adults with SB begin to experience a deterioration in their health soon after their discharge from paediatric health care services (Liptak et al., 2016; Okumura et al., 2013; Szymanski, Cain, Hardacker, & Misseri, 2017).

Current Standard of Care

In Canada and internationally, youth with SB typically receive specialized, multidisciplinary health care services, often in well-structured children's treatment centres or pediatric rehabilitation centres (Mennito & Clark, 2010; Mitchell et al., 2004; Sawyer et al., 1998). For instance, in the Southwest Local Health Integration Network (LHIN) of Ontario, children with special health care needs receive health care services through the Thames Valley Children's Centre. In these tertiary clinics, children and adolescents with SB receive assessment, diagnosis, and management for their condition from a team of diverse health care practitioners, most often including orthopaedics, urology, neurosurgery, nursing, social work, as well as occupational, speech, and physical therapy (Liptak & Dosa, 2010; Mitchell et al., 2004; Sawyer et al., 1998). The approach to care is family-centred, comprehensive, and holistic, considering the physical, emotional, social, and behavioural aspects of development ("Thames Valley Children's Centre," n.d.). However, transition to adult care is inevitable and typically occurs between 18 to 21 years of age once adolescents become ineligible for paediatric services (Binks et al., 2007; Mennito & Clark, 2010). Unfortunately, many people with SB struggle to find similarly equipped health care programs that can effectively address their complex needs after discharge from their children's treatment centre and throughout adulthood (Binks et al., 2007). Ideally, care for all people with SB should be delivered in a 'medical home', which (in principle) includes a primary physician and a team of health care practitioners, and provides care that is coordinated, continuous, comprehensive, accessible, patient- and family-centred, and holistic (American Academy of Family Physicians, American Academy of Pediatrics, American College of Physicians, & American Osteopathic Association, 2007; Thibadeau et al., 2020).

Patients, families, and a large body of scientific literature have noted many barriers inherent in the adult health care system – in Canada and internationally – that prevent successful transition and pose challenges to ongoing care (Bennett et al., 2005; Betz, 2004; Binks et al., 2007). Although the usual adult model of care may be appropriate for the general population, transition to adult care can be particularly difficult for those with complex disabilities who, in addition to managing their health care needs, face a lack of multidisciplinary care; fragmentation of services (separate appointments with different specialists across multiple settings must be booked and coordinated); scarce resources, poor coordination and communication between the adult and pediatric health care sectors and within the adult health care sector; lack of preparation or information for transition; and a paucity of health care professionals that are knowledgeable, properly trained, or interested in caring for adults with SB (Betz, 2004; Binks et al., 2007). These barriers can pose further challenges for those people with SB facing more significant cognitive or psychosocial challenges, or those who are heavily reliant on caregivers (Holmbeck, Kritikos, Stern, Ridosh, & Friedman, 2021). Difficulties in transition may also be explained by the significant cultural differences between paediatric and adult care services (Baltzer et al., 2007; Mennito & Clark, 2010; Rosen, 1995). Paediatric care is developmentally focused, multidisciplinary, and family-centred, where adult care tends to place a greater focus on independence, maturity, and disease management, necessitating deliberate guidance and additional support for those with SB (Baltzer et al., 2007; Fremion & Dosa, 2019; Mennito & Clark, 2010; Rosen, 1995).

Unsurprisingly, for many young people and adults with SB and their families, the transition experience is fraught with negative feelings (Levy, Wynd, & Carachi, 2014) and has even been likened to “falling off a cliff” (Stewart, Law, Rosenbaum, & Willms, 2009, p. 12). Several studies have identified several negative consequences of transitioning to adult care, including difficulty accessing health care services (Liptak et al., 2016), failure to seek medical attention or access age-appropriate services (Binks et al., 2007), challenges accessing funding or insurance (Liptak et al., 2016), higher use of emergency and inpatient health care (Liptak et al., 2013, 2016; Young et al., 2014), and a decline in health status over time (Okumura et al., 2013).

Transition Programming: A Call to Action

In a 2002 consensus statement on health care transitions for youth with special care needs, health care transition was defined as a dynamic process, continuing throughout the lifespan, that should ideally include the provision of planned, high-quality, individually-oriented, and developmentally appropriate services to facilitate uninterrupted movement from paediatric-oriented to adult-oriented health care systems (American Academy of Pediatrics, American Academy of Family Physicians, & American College of Physicians-American Society of Internal Medicine, 2002), Numerous other provincial, national, and international organizations, such as the Canadian Paediatric Society and the Provincial Council for Maternal and Child Health, have directed attention to the challenges associated with health care transition and the need to facilitate transition from paediatric to adult care and provide ongoing health and rehabilitation services for people with complex disabilities (American Academy of Pediatrics et al., 2002; Canadian Association of Paediatric Health Centres, 2016; Kaufman & Pinzon, 2007; Ontario Association of Community Care Access Centres, 2013; Provincial Council for Maternal and Child Health, n.d.). Guidelines, recommendations, and general health care goals published for best practice transition programming (American Academy of Pediatrics et al., 2002; Baltzer et al., 2007; Binks et al., 2007; Spina Bifida Association, n.d.) also advocate for a smooth transition to adult care, an enhanced ability to negotiate the adult health care system, optimized health, and achieving one’s maximum potential (Binks et al., 2007; Rosen, Blum, Britto, Sawyer, & Siegel, 2003).

In response to this call to action, transition programs have been and continue to emerge both internationally and within Canada (Binks et al., 2007). However, many gaps still exist in health care transition practice and research. Although organizations and experts in the field have put forth general principles and agreed on general features of effective and appropriate transition programming, there is currently no standardization of care (Grant et al., 2011; Mcdonagh, 2007). While many programs may have similar underlying themes or may be based on previous models or programs, each program’s components and elements often differ and vary in the degree to which they follow expert recommendations (Grant et al., 2011). Numerous experts and organizations have also previously placed a heavy emphasis on the simple transfer of care from paediatric to adult care, rather than on comprehensive, long-term programming that extends beyond the transition period (Betz et al., 2018). Many treatment centres, hospitals, and academic settings in Canada have developed transition programs that aim to prepare adolescents and adults for life outside of the paediatric health care system (Grant et al., 2011). However, these services primarily target health behaviour at the individual level, through developing patient skills in self-management and capacity for navigating health care services, and neglect to address the major faults and lack of infrastructure (including ongoing, comprehensive programming) within the adult health care system and the challenges faced by individuals who lack the cognitive or physical capacity to achieve self-management (Binks et al., 2007; Holmbeck et al., 2021). Unfortunately, many adults with SB lack a medical home, and poor transition outcomes are still common (Liptak et al., 2016; Okumura et al., 2013; Szymanski et al., 2017; Young et al., 2014).

Moreover, as transition to adult health care is a relatively new field and science, limited evidence exists to support specific transition practices (Betz et al., 2018; Fremion & Dosa, 2019). Historically, guidelines and recommendations have been primarily developed through expert consensus rather than rigorous evidence from well-designed studies (Betz et al., 2018; Fremion & Dosa, 2019). Previous model evaluations have been associated with significant limitations in design and methodology (Betz et al., 2018). Existing studies have also lacked information on the services or models themselves despite the urgent need for model replication and program development (Betz et al., 2018). As such, a large body of research advocates for robust program and model evaluation related to transition and lifelong care (Binks et al., 2007; Grant et al., 2011; Stewart, 2009).

TLC Program

The TLC program was developed in 2014 at the Parkwood Institute site of St. Joseph's Health Care in London, Ontario. Prior to 2014, there was no such transition program in the Southwest LHIN to serve people with SB after discharge from the local children's treatment centre (the Thames Valley Children Centre) or surrounding areas. Since its inception, the standard of care for this population has been redefined. The program was created in line with best-practice recommendations around transition to adult care (Cooley et al., 2011) and strives to address the diverse needs of adolescents and adults with chronic childhood-onset disabilities, including SB, cerebral palsy, Rett syndrome, and other developmental disabilities. In particular, the program services a significant patient population of adolescents and adults with SB. The program is unique to Southwestern Ontario and unlike existing transition programs in Canada. It functions both as a transition service and an ongoing clinical program that provides support on a regular and as-needed basis for patients with SB (and other childhood-onset disabilities) throughout their lifespan. It provides a ‘medical home’ for people with SB, with access to knowledgeable clinicians and coordination of services in one comprehensive care setting. The program was developed and designed by a team representing Parkwood Institute and the Thames Valley Children Centre, using the existing outpatient facilities located at Parkwood Institute. It has provided services to over 500 patients with childhood-onset disabilities and expects to grow as government estimates show that over 315,000 people aged 15 years and older were living in Canada with a developmental disability in 2017 (Morris, Fawcett, Brisebois, & Hughes, 2018), with Ontario having one of the highest developmental disability prevalence rates in the country (1.1%) (Berrigan, Scott, & Zwicker, 2020).

The TLC program houses a multidisciplinary team of health care practitioners, including a physiatrist, nurse practitioner, social worker, physiotherapist, occupational therapist, speech-language pathologist, registered dietitian, and rehabilitation therapist. In line with paediatric services offered to children with complex health care needs, this diverse team provides a more holistic approach to health care, considering physical and psychosocial well-being. See Table 1 for specific program services.

Table 1

TLC Services Offered in Initial Consultation and Follow-Up Visits

Initial Consultation	Follow-Up Visits
Transitional (“overlap”) clinics at Thames Valley Children’s Centre for adolescents nearing transition	Outpatient clinic visits at Parkwood Institute for adult (“post-transition”) patients
Access to interdisciplinary services, including (but not limited to) physiotherapy, occupational therapy, speech and language therapy assessment and treatment, and dietitian consultation and support	System navigation Telephone/telehealth support for patients and community partners (e.g., Family Physicians, home-care providers) Access to interdisciplinary services, including (but not limited to) physiotherapy, occupational therapy, speech and language therapy assessment and treatment, and dietitian consultation and support

Study Purpose

At this time, little research supports the effectiveness of programming currently being offered by the TLC (Betz et al., 2018), and as such, the program requires evaluation. However, as the TLC program is a relatively new service, prior to a program evaluation, more needs to be known about the populations currently being served to appropriately prioritize foci of future research, plan for continuing education of health care professionals, and allocate health care resources both within the TLC and Canadian and international health care systems. Therefore, the purpose of the present study was to identify the most common health care concerns affecting TLC patients with SB and determine whether there are recognizable patient factors that are consistently related to these pressing concerns. This work aligns with the Institute for Health Improvement’s (IHI) Triple Aim Framework, which suggests that health care system reform must: 1) improve the experience of care: 2) improve the health of populations; and 3) reduce per capita costs of health care, where the accomplishment of the aims requires a focus on a defined population and an organization or an ‘integrator’ that can coordinate health care services

(Berwick, Nolan, & Whittington, 2019). The objectives of this study were to (1) identify the most pressing initial clinical health care concerns among TLC patients with SB; (2) determine whether specific patient factors, such as age, sex, and neurological level, are associated with the concerns; and (3) determine whether patient factors (sex, age, neurological level, type of SB, and ambulatory status) can predict total number of health concerns. Initial patient consultation information was of interest to understand initial concerns at presentation to the TLC clinic, including the number and types of unmet healthcare needs among new TLC patients.

It was hypothesized that patients will present with multiple health concerns spanning multiple clinical areas, and that the most frequently identified patient concerns will relate to common conditions specific to SB, such as neurogenic bowel and bladder, as well as common secondary consequences of SB, such as osteoporosis and pressure ulcers. Specifically, it was hypothesized that sex, age, and neurological level would correlate with specific health concerns. Additionally, that total number of health concerns will correlate with:

1) neurological level, since neurological level of impairment typically predicts the extent of sensory and motor loss in people with SB (Liptak & Dosa, 2010) and has been associated with the presence of secondary impairments (Verhoef et al., 2004);

2) sex, as previous research has shown that women demonstrate greater healthcare-seeking behaviour for health concerns compared to men (Thompson et al., 2016).

3) age, due to the physical and psychosocial consequences of SB that extend beyond childhood, as well as the added risk of secondary health conditions later in adulthood (Liptak et al., 2013, 2016; Oakeshott et al., 2009);

4) ambulatory status, as wheelchair use has been associated with consequences such as pressure ulcers (Kim et al., 2015), and several other factors, including shunt status and history of hip or knee contracture surgery, have been (inversely) associated with ambulation status (B. E. Dicianno et al., 2015); and

5) type of SB, since several subgroups of SB exist, where myelomeningocele is the most common and most severe (Fletcher & Brei, 2010).

It is expected that identifying this population’s major health care challenges will help to improve the quality of services provided within the TLC program, target health care delivery, and in line with the first and second aims of the IHI Triple Aim Framework, improve the experience of care and improve the health of patients with SB within the program (Berwick et al., 2019).

Chapter 3: Methodology

Study Design

This research is guided by the Institute for Health Improvement's (IHI) Triple Aim Framework (Berwick et al., 2019). Ethics approval was initiated through the Lawson Health Research Institute and approved by the Health Sciences Research Ethics Board at Western University (Appendix A). This study used an observational design involving a retrospective medical chart review of initial TLC patient consultation encounters between 2014 (time of the program's inception) and December 2017.

Data Collection

See Table 2 for extracted data elements. All data extraction was conducted electronically using a data extraction tool created in the REDCap research database platform (Appendix B). The physiatrist within the TLC program developed the tool for both retrospective and prospective studies based on the clinic initial assessment form (where the same form allows for data extraction for both study designs). It is reflective of data gathered for patient medical charts collected within clinical consultations, and of characteristics and issues relevant to people with SB. A pilot test of the data collection tool was performed, where the tool was trialed, and data from approximately ten patient medical charts was extracted, by two members of the research team. This process ensured that the extractors were able to reliably use the form and extract patient data in the same manner. The pilot test also provided an opportunity to adjust the tool when there were discrepancies between extractors.

In this study, neurological level is defined as the lowest level at which sensory or motor function was preserved in a patient, where function was fully intact above that particular level. As SB lesions are typically not discrete, with some blurring of normal and abnormal function in the zone of the anatomical spinal cord defect (Rintoul et al., 2002), neurological levels were broadly grouped as thoracic, upper lumbar, lower lumbar, and sacral. In the event that patients had asymmetrical neurological presentations on the right and left side, the patient was classified as the higher (i.e., worse) of the two neurological levels. Presenting concerns included any health care or social matter that the patient or caregiver felt required the attention of the physician or of another TLC team member, or any issue that the physician felt needed attention at the time of presentation to the TLC program. Comorbid conditions that were controlled or stable and not leading to any active concerns (i.e., were not raised as pressing concerns by the patient/caregiver and/or the physician at the initial clinical consult) were not identified as presenting concerns for this study. Presenting concerns (found within the dictated chart notes of the initial clinical consult, in the concluding treatment plan/list of final recommendations) were summarized and extracted in a list format alongside the collection of other data elements using the REDCap data extraction tool. Patients were included in the study if they presented to the TLC clinic between 2014 and 2017 and had a diagnosis of SB. There were no restrictions regarding age or other patient characteristics.

Table 2

Patient Factors and Concerns Extracted

Data Elements Extracted
Age (in years, at the time of initial consult)
Date of birth
Sex
Individual reporting concerns (communication status)
Self, other, unknown, or not reported
Type of SB
Myelomeningocele, meningocele, occulta, other (normal exam), or not reported
Neurological level
Thoracic, high lumbar, low lumbar, sacral, unknown, or not reported
Ambulatory status (according to the Hoffer Classification of Ambulation for patients with spina bifida or other diagnoses (Bartonek, Saraste, & Knutson, 1999))
Community ambulator, household ambulator, or non-ambulator
Surgical history
Hip Status
Epilepsy history
Current medications and active non-medical treatment
Physical exam results
Hip flexion contracture, knee flexion contracture, plantar flexion contracture, or scoliosis
Presenting issues or concerns at initial consult

Note. Although “communication” and “hip status” were initially included on the data collection instrument, the information related to these two categories was omitted due to inconsistent reporting within patient medical charts.

Note. “Sex” was used to describe male/female patients (rather than “gender”) as it is unclear if patients were explicitly asked about their preferred gender (gender as socially constructed) at their initial consultation. Within the TLC program’s patient medical charts, only patients’ male/female designation from their patient hospital chart (obtained from TLC program registration) was available.

Data Analysis

Data Transformation

All presenting concerns (for each patient) were exported from the REDCap database. From there, each unique health concern was inductively identified from the raw data. With the information extracted from patient medical charts, each patient was coded as either having or not having each identified health concern. Individual health concerns were then grouped into broader concern categories to report the most common concerns broadly across the cohort and assist with statistical analyses. Within each concern category, patients were coded as having no health concerns, at least one health concern, or two or more concerns.

Objective 1: identify the most common clinical health care concerns

Descriptive statistics were used to summarize the characteristics of the sample using frequencies, percentages, mean, standard deviation, median, range, minimum, and maximum as appropriate. Concern categories and the individual health concerns within each category were reported as frequencies and percentages. To determine the most common concerns, only the proportion of patients with at least one concern within each concern category was considered. The “most common” concerns were defined as all health concerns that affected over 25% of the sample. This percentage was not based on any pre-defined criteria but was thought to be a large enough proportion of patients to warrant clinical consideration of a concern area.

Objective 2: determine whether specific patient factors are associated with the concerns

Spearman correlation coefficients were used to identify associations between the most common health concerns and patient-related factors of age, sex, and neurological level. Spearman correlation coefficients were used due to the level of the data (ordinal), where Pearson correlation coefficients are more appropriate for interval-level data. A Bonferroni correction factor was applied to adjust for multiple comparisons. The Bonferroni correction is more precise and reduces the chance of a type I error, where there is an increased risk of type I error from conducting multiple statistical tests. Multiple methods for controlling family-wise error rate were considered, and Bonferroni was chosen as it was the most simple yet conservative approach.

Objective 3: determine whether specific patient factors can predict total number of health concerns

Total number of concerns were reported as mean, median, standard deviation, and range. Patient factors (sex, age, neurological level, type of SB, and ambulatory status) were associated with total number of concerns using Spearman correlation coefficients. Total concerns were considered ordinal data (and thus, the choice of Spearman correlation coefficients) due to the inability to attribute a meaningful interpretation or claim that the difference between one concern and another is the same, although the data was rankable. A Bonferroni correction factor was applied to adjust for multiple comparisons. Finally, a backward, stepwise multiple linear regression analysis was performed where number of concerns was the dependent variable, and sex, age, neurological level, type of SB, and ambulatory status were independent variables. A backward stepwise regression model was chosen due to the exploratory nature of this study. To this study's knowledge, limited research exists on which to base the order of entry of the predictors into the model regarding their importance in predicting total number of health concerns (for hierarchical regression), and limited theoretical underpinnings also exist for forced variable entry. The backward method was selected (as opposed to the forward) to minimize suppressor effects (which occur when a specific variable only has a significant effect when another variable is held constant) (Portney & Watkins, 2008). In stepwise regression, the order in which the predictors are entered into the model is based on mathematical criteria (Portney & Watkins, 2008). In the backward stepwise method, all of the predictors are entered into the model simultaneously. The contribution of each predictor is then evaluated in a stepwise fashion (using the significance (p-value) of its t-test) (Portney & Watkins, 2008). If a predictor does not make a statistically significant contribution to the model, it is removed, and the contribution of the remaining predictors is reassessed (Portney & Watkins, 2008). Assumptions for the regression model were tested, including assumptions of additivity and linearity, independent errors (or independence of observations), homoscedasticity, normally distributed errors (or normality of residuals), variable types, multicollinearity, and non-zero variance.

Chapter 4: Results

Patient Characteristics

A total of 94 patients met the inclusion criteria for the study (Table 3). The mean age of the patients included in this study was 29.04 years *(SD*= 13.8). Age was not normally distributed, the median was 26 years, the mode was 19 years, and few patients were over the age of 60, establishing that most patients were within younger age cohorts. Patients were predominantly female (70%) and had myelomeningocele (77%). Neurological level was relatively evenly distributed across the sample, where most patients had a low lumbar lesion (37.2%) or thoracic level lesion (26.6%). In terms of ambulation, 43% of patients were community ambulators, and 50% were non-ambulators. The remainder only ambulated within their household. The most common surgical procedures were in neurosurgery (75%), including ventriculoperitoneal (VP) shunt procedure(s), tethered cord release, and Chiari decompression. A significant portion of patients had also previously had an orthopaedic (56%) or a bowel or bladder surgery (20%). Specifically, the most common surgical procedures experienced by patients were VP shunt procedure(s) (62.8%) and tethered cord release (21.3%). Fifteen percent of patients had a history of epilepsy. The most commonly used medications were bowel/GI agents (23%), Vitamin D supplements (19%), psychotropic agents (12%), and pain agents (11%). Although this information was not explicitly collected, a large proportion of patients (81%) were taking other regular medications and 29% were taking other medications on an as-needed basis (different than the ‘other’ medication grouping found within Table 3) that were not found on the data extraction instrument. Finally, 27% of patients had at least one hip flexion contracture, 36% had at least one knee flexion contracture, and 53% had scoliosis.

Table 3

Demographic Characteristics

Patient Characteristic	*n*(94)	
	n	%
Sex		
Male	28	29.8
Female	66	70.2
SB Type		
Myelomeningocele	72	76.6
Occulta	7	7.4
Other	13	13.8
Not Reported	2	2.1
Neurological Level		
Thoracic	25	26.6
High Lumbar (L1-L3)	19	20.2
Low Lumbar (L4-L5)	35	37.2
Sacral	10	10.6
Normal Exam	5	5.3
Ambulatory Status		
Community Ambulator	40	42.6
Household Ambulator	7	7.4
Non-Ambulator	47	50
Independent with transfers	31	66
Assistance with transfers	2	4.3
Dependent with transfers	14	29.8
Surgical History		
Neurosurgery	70	74.5
Bowel or Bladder	19	20.2
Orthopaedic	53	56.4
Other Non-Orthopaedic	90	95.7
Epilepsy History		
Yes	14	14.9
No	77	81.9
Unclear	2	2.1
Unknown	1	1.1
Medications		
Antiepileptic	7	7.4
Psychotropic	11	11.7
Tone	4	4.3
Pain	10	10.6
Bowel/GI	22	23.4
Other	31	32.9
None	9	9.6

Patient Characteristic	*n*(94)	
	n	%
Physical Exam Results		
Hip Flexion Contracture	25	26.6
Knee Flexion Contracture	34	36.2
Plantar Flexion Contracture	0	0
Scoliosis	50	53.2
None	8	8.5
Not Reported	25	26.6

Note. The “Other” subcategory within “SB Type” represented those with lipomyelomeningocele, dermal sinus tract, occulta with diastematomyelia, or an unknown SB subtype.

Note. The “Normal Exam” subcategory within “Neurological Level” represented patients who had an atypical neurological presentation.

Note. The “Other Non-Orthopaedic” subcategory within “Surgical History” represented other surgical procedures that were not explicitly collected on the data extraction instrument.

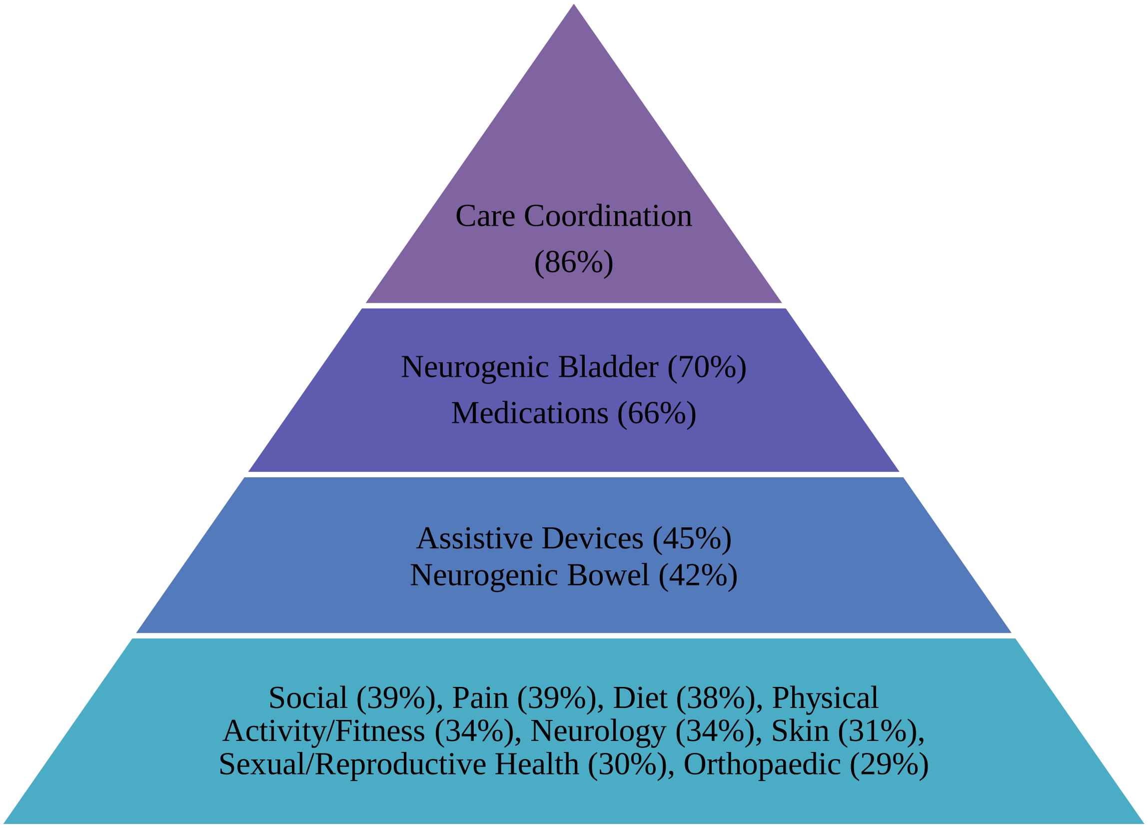
Note. “Other” medications included calcium, vitamin D, iron, melatonin, Trazodone, Zopiclone, and Scopolamine.

Top Concerns

One hundred individual patient concerns and 18 concern categories were identified (Table 4). Thirteen health concern categories were most common, found in Figure 1.

Figure 1

Most Common Concerns (In Order)



The most common health concern was care coordination, with 86% of patients requiring coordination relating to one or more health issue(s). Unpacking this further, the majority of patients required physiotherapy services (35%), most often to address physical activity concerns, and also often to address pain, weight loss, wheelchair, and gait concerns. The medical team also referred 15% of patients to recreational therapy, 14% to dietetics, 14% to social work, and 14% to occupational therapy. Secondly, patients commonly had at least one concern regarding neurogenic bladder (70%), and/or neurogenic bowel (42%). Bladder or kidney concerns related to monitoring bladder/renal function with urinalysis, blood pressure monitoring, renal and bladder ultrasound and/or urodynamics; bladder management (including catheterization, incontinence, hygiene, bladder irrigation, bladder emptying, and consideration of need for surgical intervention); bladder education; and concerns about upper or lower urinary tract infections. Concerns related to neurogenic bowel included constipation, establishing bowel routines, addressing incontinence or changes in bowel patterns, and hygiene.

More than half of the patients (66%) had medication concerns. Medication concerns included requiring or considering new prescriptions; restarting medications; needing to switch or stop medications or alter their dosage; needing to refill existing medications; and concerns with medication compliance. Patient medication concerns most often related to the management of pain, seizures, bladder or bowel routines, spasticity, hypertension, and ADHD. Several initial clinical encounters also included discussions about supplementation, including vitamin D and calcium for bone health, and magnesium, fibre, or probiotics for bowel management. Concerns about assistive devices were reported by 45% of patients and mostly involved orthotics or braces (25%) and wheelchair or seating (19%). Concerns about existing devices, device assessments, and new device options or prescriptions were often discussed.

Social concerns were prevalent (39%) in this population. The most common social concerns involved finances or funding (15%) and social participation (15%). Social participation concerns related to community involvement, social opportunities, leisure activities, support groups, and specific social programming. Other less common social concerns were in school or education (4%) and future care or living arrangements (4%). In addition, 39% of patients had pain concerns, most commonly back pain (19%), lower extremity pain (14%), and required pain management (14%). Diet concerns were also prevalent, specifically related to food/nutrition (21%) or weight management (13%). A total of 34% of patients had a physical activity or fitness concern. In this area, patients were prescribed specific exercise or physiotherapy programs, stretching or strengthening and range of motion exercises, and/or had concerns about physical activity, exercise, fitness, or sports participation. Furthermore, 34% of patients had concerns about neurologic function, most involving spasticity (12%) and symptomatic tethered cord syndrome (7%). Skin health concerns affected 31% of the sample and included issues around skin breakdown/redness, a need for plastic surgery, and preventative strategies to maintain skin health. Sexual/reproductive health and family planning concerns were reported by 30% of patients, with family planning being the most common (20%), representing concerns centred around birth control, folate/folic acid supplementation, and SB prevention. Finally, 29% of patients experienced orthopaedic concerns, and 24% had other (miscellaneous) concerns that could not be sorted into any of the existing categories. Some concern areas that were less prevalent, but notable, were mental health, bone health, and functional mobility concerns. The fewest number of concerns were in the environment and bone health categories.

Table 4

Health Concern Categories and Individual Concerns

Concern Category & Individual Concerns	Summary	*n*	%
Functional Mobility			
Improve Ambulation		2	2.1
Maintain/Improve Functional Mobility		6	6.4
Maintain/Improve Range of Motion		4	4.3
Functional Decline		6	6.4
Gait		5	5.3
Falls/Fall Prevention		3	3.2
Functional Mobility Other		2	2.1
	No concerns	73	77.7
	One concern	16	17
	Two Concerns	5	5.3
Assistive Devices			
Orthotics and Braces		23	24.5
Wheelchair/Seating		18	19.1
Gait Aids		6	6.4
	No concerns	52	55.3
	One concern	37	39.4
	Two Concerns	5	5.3
Miscellaneous			
Swallowing		5	5.3
Respiratory		2	2.1
Endocrinopathies		2	2.1
Inpatient Stay (Care Coordination)		2	2.1
Twitching/Cramping		2	2.1
Driving		8	8.5
Miscellaneous Other		7	7.4
	No concerns	71	75.5
	One concern	19	20.2
	Two Concerns	4	4.3
Medications			
		62	66
Physical Activity/Fitness			
		32	34

Concern Category & Individual Concerns	Summary	*n*	%
Neurogenic Bladder			
Current Bladder/Kidney Status/Function		49	52.1
Monitoring Bladder/Kidney Health		49	52.1
	No concerns	28	29.8
	One concern	34	36.2
	Two Concerns	32	34
Bone Health			
Bone Health/Fractures		6	6.4
Osteoporosis		6	6.4
Osteoporosis/Bone Health Treatment		11	11.7
	No concerns	79	84
	One concern	7	7.4
	Two Concerns	8	8.5
Neurogenic Bowel			
Neurogenic Bowel		38	40.4
Specific Device Concerns		2	2.1
	No concerns	55	58.5
	One concern	38	40.4
	Two Concerns	1	1.1
Care Coordination			
Physiotherapy		33	35.1
Occupational Therapy		13	13.8
Rehabilitation Therapy		8	8.5
Recreational Therapy		14	14.9
Orthotics		7	7.4
Dietetics		13	13.8
Specialist		8	8.5
Family Physician Nurse Practitioner		6	6.4
Nephrology		4	4.3
Gynaecology		2	2.1
Urology		6	6.4
Social Work		13	13.8
Speech Language Pathology		6	6.4
Optometry		2	2.1
Family Physician Search		3	3.2
Transition/Ongoing Care		11	11.7
Seating		11	11.7
Wound Clinic		7	7.4
Care Coordination Other		4	4.3
	No concerns	13	13.8
	One concern	34	36.2
	Two Concerns	47	50

Concern Category & Individual Concerns	Summary	*n*	%
Skin Health			
Edema Management		4	4.3
Pressure Management		10	10.6
Wound/Skin Management		20	21.3
Edema/Swelling/Lymphedema		4	4.3
	No concerns	65	69.1
	One concern	21	22.3
	Two Concerns	8	8.5
Environment			
Home Accessibility		10	10.6
Workplace Accommodations		2	2.1
	No concerns	82	87.2
	One concern	12	12.8
	Two Concerns	0	0
Diet			
Food/Nutrition		20	21.3
Weight Management		12	12.8
Supplements		4	4.3
Dietary Management of Bowels		11	11.7
	No concerns	58	61.7
	One concern	25	26.6
	Two Concerns	11	11.7
Sexual/Reproductive Health & Family Planning			
Women's Health Issues		4	4.3
Family Planning		19	20.2
Pregnancy/Childbirth/Childbearing		5	5.3
Sexual Health		2	2.1
Fertility		6	6.4
Menstruation/Menorrhagia		2	2.1
Sexual Function		4	4.3
	No concerns	66	70.2
	One concern	16	17
	Two Concerns	12	12.8

Concern Category & Individual Concerns	Summary	*n*	%
Pain			
Lower Extremity Pain		13	13.8
Back Pain		18	19.1
Upper Extremity Pain		5	5.3
Musculoskeletal Pain		2	2.1
Pain Management		13	13.8
Headaches		3	3.2
Bursitis		2	2.1
Pain Sensitization		2	2.1
Pain Other		3	3.2
	No concerns	57	60.6
	One concern	18	19.1
	Two Concerns	19	20.2
Orthopaedic			
Joint Management		8	8.5
Joint Stability/Instability		3	3.2
Leg Length Discrepancy		3	3.2
Contractures/Flexion Contractures		3	3.2
Foot Concerns		4	4.3
Spinal Curvature		10	10.6
	No concerns	67	71.3
	One concern	23	24.5
	Two Concerns	4	4.3
Mental Health			
Affective Disorders		14	14.9
Coping		2	2.1
Behaviour		3	3.2
Sleep		3	3.2
Substance Use Cessation		3	3.2
Mental Health Other		2	2.1
	No concerns	72	76.6
	One concern	17	18.1
	Two Concerns	5	5.3

Concern Category & Individual Concerns	Summary	*n*	%
Neurology			
Neurological Monitoring		2	2.1
Numbness/Sensory Loss		2	2.1
Vision		3	3.2
Articulation/Speech Clarity		2	2.1
Seizures		5	5.3
Shunt Function		4	4.3
Spasticity		11	11.7
Symptomatic Tethered Cord		7	7.4
Hand Paresthesias/Carpal Tunnel		2	2.1
Neurology Other		2	2.1
	No concerns	62	66
	One concern	25	26.6
	Two Concerns	7	7.4
Social			
Return to School		4	4.3
Social Support/Participation		14	14.9
Independence		5	5.3
Employment/Volunteering		6	6.4
Future Care/Living Planning		4	4.3
Advocacy		2	2.1
Transportation		3	3.2
Financial/Funding		14	14.9
Social Other		3	3.2
	No concerns	57	60.6
	One concern	23	24.5
	Two Concerns	14	14.9

Note. The concerns labelled "__ Other" within each category included individual concerns indicated by only one patient that were therefore grouped together.

Associations

A Bonferroni correction factor of (0.05/39) resulted in an adjusted significance level of 0.001. All Spearman's rho correlations between the demographic factors of age, sex, and neurological level and the top 13 concerns were weak and non-significant ($p>0.001$). Although none of the associations reached clinical significance with the adjusted alpha level, some were more strongly correlated and significant prior to correcting for multiple comparisons (Table 5).

Table 5

Correlations Between Most Common Health Concerns and Specific Grouping Factors

Health Concern (Category)	Sex		Age		Neurological Level	
	r_s	p	r_s	p	r_s	p
Pain	-.24	.02	—	—	.24	.022
Diet	—	—	.26	.012	—	—
Sexual/Reproductive Health and Family Planning	—	—	-.27	.009	—	—
Skin Health	—	—	—	—	-.23	.027
Physical Activity/Fitness	—	—	—	—	.33	.001

Note. All correlations were not statistically significant at the 0.001 level, but were significant at the 0.05 level (2-tailed), *n*=94, df= 92

Note. Only correlations that reached significance prior to the application of the Bonferroni correction factor were presented to account for associations that may exist in larger sample sizes with greater power and to reduce the need for a large number of simultaneous statistical tests in future studies.

Total Concerns and Predictors of Total Number of Concerns

The median number of concerns reported at initial consultation was 9.0 (*M*= 9.2, *SD*= 3.9), where the range was 21 (1 to 22). A Bonferroni correction factor of (0.05/5) resulted in an adjusted significance level of 0.01. None of the variables (age, sex, neurological level, type of SB, or ambulatory status) were significantly correlated with total number of concerns (Table 6).

Table 6

Correlations Between Specific Grouping Factors and Total Number of Health Concerns

Patient Factor	r_s	p
Age	.228	.027
Sex	.087	.402
Neurological Level	-.138	.184
Type of SB	-.134	.198
Ambulatory Status	.094	.365

Note. Significance was set at the 0.01 level (2-tailed), n=94

Assumptions of homoscedasticity, variable types, multicollinearity, non-zero variance, independent errors (Durbin-Watson value= 1.84), and normally distributed errors were met. However, none of the relationships between patient factors (independent variables) and total concerns (dependent variable) were linear. In the original regression analysis, age, sex, neurological level, type of SB, or ambulatory status did not explain a significant proportion of variance in total concerns (R^2= .051, F(5, 88)= .939, p= .460) (see Table 7 for results of individual predictors). Due to the violation of the linearity assumption, the dependent variable (total concerns) was log-transformed, and a log-level regression was conducted using the same backward stepwise regression method. The log-level regression resulted in one significant model (R^2= .066, F(2, 91)= 3.2, p= .045) with age and neurological level predicting total number of concerns. However, despite being statistically significant, the model containing age and neurological level explained only 5% of the variance in total concerns (adjusted R^2= .045), which is unlikely to be clinically significant. Furthermore, the individual variables in the model (age and neurological level) were not significant predictors of total number of concerns (see Table 8 for results of individual predictors). A post hoc power analysis of the full (R^2= .076) model containing 5 independent variables, and nested significant model containing age and neurological level revealed a power of 0.039 (n= 94).

Table 7

Summary of Regression Analysis for Variables Predicting Total Number of Health Concerns

Patient Factor (Independent Variable)	*B*	*SE B*	β	*t(5)*	*p*	95% CI	
						LB	*UB*
(Constant)	9.41	1.58	—	5.94	—	6.26	12.55
Age	.034	.03	.120	1.15	.253	-.025	-.093
Sex	.719	.9	.084	.801	.425	-1.063	2.501
Neurological Level	-.713	.49	-.208	-1.47	.145	-1.677	.250
Type of SB	.001	.003	.055	.518	.605	-.004	.007
Ambulatory Status	-.268	.385	-.099	-.695	.489	-1.033	.5

Table 8

Summary of Log-Level Regression Analysis for Age and Neurological Level Predicting Total Number of Health Concerns

Patient Factor (Independent Variable)	*B*	*SE B*	β	*t(2)*	*p*	95% CI	
						LB	*UB*
(Constant)	.890	.065	—	13.759	.000	.762	1.019
Age	.003	.002	.161	1.587	.116	-.001	.006
Neurological Level	-.041	.021	-.197	-1.940	.055	-.082	.001

Chapter 5: Discussion

Top Concerns

In line with the literature (Lidal, Lundberg Larsen, & Hoff, 2019; Liptak et al., 2013, 2016), TLC patients with SB continue to face multiple conditions associated with SB, affecting multiple organ systems, and secondary complications during and after the transition to adult health care. On average, patients had a total of 9 health care concerns and as many as 22 at their initial consultation. The results of this study provide evidence that many health concerns persist from childhood and that new health concerns may arise over the life course, necessitating continued, lifelong care, in addition to the support provided in the initial health care transition.

Care coordination services and multidisciplinary health care teams are rarely available or accessible to adults with SB (Binks et al., 2007). Young et al. (2009) found that adolescents with SB were more likely to receive health care services from a greater variety of health professionals than adults with SB (Young et al., 2006). Many studies have also noted a decline in contact with health and social services after discharge from paediatric care (Binks et al., 2007; Liptak et al., 2016; Okumura et al., 2013). This decline perhaps points (at least partially) to the importance of care coordination and an adult-centred medical home in facilitating access to the appropriate care professionals in the adult years. Care coordination was the most common concern reported by TLC patients with SB, with almost all patients (86%) reporting at least one need in this area. This finding further highlights the importance of a service like the TLC program and access to multidisciplinary care in a single setting. It underscores the need to remove the burden of coordinating care from individuals and their caregivers and emphasizes the need for continued access to multiple health care professionals, including allied health, in the management of SB throughout adulthood. Just at the initial clinical encounter, patients were referred to several health care professions, including physiotherapy, recreational therapy, dietetics, occupational therapy, and social work, as well as other specialties regarding assistive device concerns and wound or skin management. Although it is expected that the need for multiple specialist referrals will decline in follow-up consultations, half of the patients in this study were referred to two or more specialty services, highlighting the immense need for care coordination and multidisciplinary care in this population. Of note, many referrals were to services available within the TLC program, where there are knowledgeable clinicians interested in caring for people with complex health care needs. Elsewhere, people with SB often have difficulty navigating the health care system and accessing competent health care providers who have are interested or knowledgeable in SB (Binks et al., 2007). The volume of care coordination concerns suggests that the TLC program fills a major gap in the health care system by providing coordinated, multidisciplinary care in a single setting.

It is well documented that youth and adults with SB are at substantial risk for bowel and bladder dysfunctions (Liptak & Dosa, 2010; Mitchell et al., 2004). In their initial consultation, a high proportion of patients were experiencing neurogenic bowel and bladder concerns. Other studies have also found that bowel concerns, including fecal incontinence, and bladder concerns, such as incomplete bladder emptying, urinary tract infections, and urinary incontinence, are significant issues for adults with SB (Betz et al., 2018; Lidal et al., 2019; Liptak et al., 2013). The present study further identified a specific need for bladder and kidney monitoring, which is particularly important as renal failure secondary to neurogenic bladder dysfunction continues to be a frequent and sudden cause of death in people with SB (Liptak et al., 2013; Oakeshott et al., 2009). The various methods of managing and monitoring bowel and bladder function include surgical interventions, oral medication, and other specific procedures (Liptak et al., 2013; Mitchell et al., 2004). These interventions, and any others, should be continuously discussed and incorporated in TLC consultations and follow-up appointments to address neurogenic bowel and bladder concerns. Unfortunately, due to the retrospective nature of this study, it is unclear whether these neurogenic bladder and bowel concerns were concerns of the health care professional or reported by the patient/caregiver. Future studies should delineate bladder and bowel concerns identified by the patient/caregiver(s) and the health care provider to better address bladder and bowel needs.

Two-thirds of patients had concerns regarding medications, suggesting that the TLC program plays a role in recommending or prescribing pharmacological treatment and managing current medications and medication regimens. People with SB often have multi-system disorders treated pharmacologically (B. E. Dicianno et al., 2008). Similar to the findings of Lidal et al. (2019), the present study found that only 10% of study patients were not taking any medications. Additionally, the mean age of the participants in the study by Lidal et al. (2019) was 58 years, suggesting some stability of medication use usage throughout adulthood and the importance of continued attention to medication management over time.

Half of the patients in this sample were non-ambulatory, with only 11% having a sacral neurological level of impairment, pointing at a large proportion of patients with significant limitations in motor function (Liptak & Dosa, 2010). Therefore, it is unsurprising that concerns relating to assistive devices, most notably orthotics and braces, were prevalent. Assistive devices (such as orthotics, braces, wheelchairs, and gait aids) can improve mobility and ambulation in adults with SB, where preservation of mobility is an important determinant of functioning and quality of life (B. E. Dicianno et al., 2008). However, many adult practitioners have limited medical training and experience with chronic childhood-onset disabilities (Binks et al., 2007). It is likely that many lack expertise in recommending or prescribing assistive devices and identifying and meeting the general ambulatory and mobility needs of people with SB. The assistive technology needs of people with SB are complicated by features such as scoliosis and short limb length, necessitating care by multidisciplinary teams experienced in prescribing assistive devices (B. E. Dicianno et al., 2008). The TLC program compensates for this gap by providing patients with a medical home, thereby mitigating the challenges patients with SB have in accessing services that address their specific assistive device needs within the typical adult care system. TLC programming must continue to address concerns with existing patient devices and prescribe or advocate for appropriate devices that support mobility, functioning, and participation (B. E. Dicianno et al., 2008). Physiatrists should also aim to appropriately preserve the physical function and mobility of ambulatory patients who are ambulatory and those who use wheelchairs through specific rehabilitative programming (B. E. Dicianno et al., 2008).

Social well-being is vital to overall health (Helliwell & Putnam, 2004). Many people with SB experience brain abnormalities that present cognitive and physical challenges that may affect independence, education, employment, and/or other social outcomes (Liptak & Dosa, 2010; Roach et al., 2011). Research suggests that such consequences may be mitigated or well-managed in a multidisciplinary care setting (Roach et al., 2011). The prevalence of social concerns reported by TLC patients suggests that it was challenging to access social services, including social work and recreational therapy, for concerns related to funding and socialization prior to the inception of the TLC clinic.

In a recent study conducted by Lidal et al. (2019), a cohort of older adults (mean age 58) with SB reported that their most notable health concern was pain. Although pain was not the most common concern in this study, it affected 40% of patients, with most reporting pain in the lower extremities or the back, in line with the findings in Lidal et al. (2019). The younger average age in this study may explain why pain was reported less frequently, as in the literature, pain is more common among older adults with SB (Werhagen, Hultling, & Borg, 2010). A significant portion of patients in the current study also had two or more pain concerns, suggesting that pain in one area coincides with pain in another body region. The findings from this study further support the literature that adults with SB are likely to experience pain and have a higher prevalence of pain than the general population (Peterson, Haapala, Kamdar, Lin, & Hurvitz, 2021). For this reason, and because pain can affect quality of life and point to underlying treatable conditions (Liptak & Dosa, 2010), specific attention to pain in health care interactions is warranted (Alriksson-Schmidt, Josenby, Lindquist, & Westbom, 2018; Lidal et al., 2019; Liptak & Dosa, 2010).

Diet is also an important concern to consider in the clinical encounter as a high proportion of older adults with SB are overweight or obese and have hypertension (Lidal et al., 2019; Liptak et al., 2016). The likelihood of being overweight or obese in adults with SB also increases with age (Oakeshott, Poulton, Hunt, & Reid, 2019) and links to a decline in mobility or ambulatory functioning (Lidal et al., 2019; Oakeshott et al., 2019). In the current study, weight management specifically was only a concern for 13% of patients. However, food and nutrition concerns were reasonably significant (with 21% of patients reporting it as a concern) and were often discussed in the context of other concerns, such as bone health and bowel management, for which diet is also important (Mcpherson, Chen, O'neil, & Vanderbom, 2020). Combined with physical activity and fitness programming, proper diet, weight management, and recognition of nutrition problems should continue to be central in TLC programming to ensure optimal health and to aid in the prevention of secondary conditions (Mcpherson et al., 2020; Oliveira, Já, & Marques, 2014). There may be opportunities to run group programming related to dietary needs across multiple populations served by the TLC program as a cost-effective method of addressing these patient concerns. Future research should also include the prospective collection of information regarding any changes in ambulatory functioning related to weight as these patients age to better understand this association.

Another of the most commonly discussed concerns in the initial clinical encounter was physical activity/fitness (34%). Physical activity or exercise is particularly important in youth and adults with SB due to their increased risk of obesity, pain, hypertension, and decline in mobility later in life (Lidal et al., 2019). A narrative review of the literature found that adults with SB are more likely to be inactive, have decreased aerobic capacity, lower daily physical activity, and higher levels of obesity compared to other groups of people (Crytzer, Dicianno, & Kapoor, 2013). Bloeman et al. (2019) found that children and adolescents who use a manual wheelchair are more sedentary and less physically active than their peers with typical development, increasing the risk for secondary health conditions (Bloemen et al., 2019). At the initial encounter with the TLC program, physical activity was often prescribed for concerns such as pain, weight management, range of motion, and bone health. Physical activity concerns were often co-reported with physiotherapy concerns as patients were prescribed physical activity or specific exercises (such as home programing, stretching, and strengthening) in the physiotherapy setting (within the medical home) when concerns around physical activity were discussed. Exercise training in people with SB can improve elements of fitness (Oliveira et al., 2014). Therefore, it may be beneficial to consider physical activity as a preventative strategy or to encourage general physical activity for all TLC patients to promote optimal health and wellbeing. Similar to addressing dietary concerns, a group physical activity promotion class or series may be a cost-effective method of addressing the physical activity concerns of a high proportion of TLC patients.

While a modest number of patients had neurological concerns upon presentation to the TLC clinic, each concern was most often limited to a small number of patients and concerned spasticity and symptomatic tethered cord. Patients in this cohort commonly had a past surgery to place a VP shunt (63%), which is comparable to the results of the National Spina Bifida Patient Registry clinical data profile by Sawin et al. (2015), where 66% of participants had a VP shunt placement (Sawin et al., 2015). However, even though shunts frequently fail or become infected and often require revision (Liptak & Dosa, 2010), very few patients in this study had shunt concerns (4%) at their initial appointment. The literature suggests that acute hydrocephalus or shunt malfunction is not commonly recognized and is a common cause of death among people with SB (Oakeshott et al., 2009). Shunted hydrocephalus is also associated with hospitalizations and acute outpatient visits in people with SB (Liptak et al., 2016; Young et al., 2014). As clinicians within the TLC program have greater competence and exposure to adolescents and adults with SB and can recognize the signs and symptoms of shunt deterioration, the program positions itself appropriately to watch for signs of neurologic deterioration and continue to adequately monitor those with hydrocephalus and a VP shunt (Liptak & Dosa, 2010). In providing a medical home for people with SB, the TLC program creates a first line of defence in the prevention of morbidity and mortality related to hydrocephalus and VP shunt in adulthood. As such, the program may also help to reduce preventable shunt complications causing hospitalization and emergency visits, which are both costly to the health care system and riskier for patient populations (Kinsman & Doehring, 1996; Paff, Alexandru-Abrams, Muhonen, & Loudon, 2018; Young et al., 2014).

Skin health, a concern among 31% of patients in this study, is vital, specifically for those who experience spinal abnormalities causing a loss of sensation and motor function and who are reliant on assistive devices due to the increased risk for developing pressure ulcers (Liptak & Samra, 2010). In Liptak et al. (2016), skin concerns were one of the leading organ system conditions affecting adults with SB. In this same study, pressure ulcers were linked to increased emergency department visits (Liptak et al., 2016). As such, appropriate prevention and care, including pressure relief strategies, skin assessments, proper wound management, and assessments of assistive devices, are essential in preventing further complications (Liptak & Dosa, 2010; Liptak et al., 2013). In this retrospective study, it is unclear whether skin health concerns were mentioned by the patient/caregiver in the clinical setting or identified as an area of concern by the health care provider. Future research that distinguishes between patient/caregiver and provider concerns may help to better target patient-focused skin/pressure ulcer prevention and care strategies.

Although sexual/reproductive health and family planning was not one of the most common concern areas, it is an important area to note considering the younger mean age of this population, the potential for sexual dysfunction in males (Liptak & Dosa, 2010), and the larger number of female SB patients within the TLC clinic who are likely to have unique concerns regarding their reproductive health (puberty, sexuality, pregnancy, childbirth, and menopause) (Berndl, Nosek, & Waddington, 2020). A recent study by Akre et al. (2015) confirms that adolescents and young adults with SB report common concerns, challenges, and questions regarding their sexuality, fertility, and romantic relationships (Akre, Light, Sherman, Polvinen, & Rich, 2015). People with SB also have difficulty finding answers to their questions and desire information directly from their physicians (Akre et al., 2015). Adult health care providers within the general adult health care system often lack the knowledge and experience to effectively care for people with SB and can be uncomfortable or ambivalent when discussing specific health care topics (Binks et al., 2007). Women with SB report being perceived as asexual by their healthcare providers (Streur et al., 2019), while physicians report several barriers to care, including lack of formal training, knowledge gaps, and lack of comfort and time during appointments (Streur, Schafer, Garcia, & Wittmann, 2018). Meanwhile, health care providers within the TLC program are knowledgeable and interested in caring for the specific sexual, reproductive, and family planning needs of this population. The smaller percentage of concerns in this area may reflect that sexual and reproductive health concerns discussed more heavily in follow-up visits once patients feel more comfortable and have established a relationship with the TLC program team. Even so, an opportunity exists to direct more attention to and empower and educate youth and adults about their sexual health to encourage healthy physical and psychosocial development, successful transition to adult care (Akre et al., 2015), and prevent suboptimal outcomes, in line with expert guidelines and recommendations (Houtrow AJ & Roland M, 2020).

Lastly, orthopaedic concerns were found among 29% of the sample, with spinal curvature concerns (scoliosis and kyphosis) being the most prevalent. Again, this is unsurprising as orthopaedic problems, including abnormal development of the vertebrae leading to scoliosis and kyphosis, are common among people with SB (Liptak et al., 2013; Verhoef et al., 2004; Werhagen, Gabrielsson, Westgren, & Borg, 2013), where scoliosis was reported by 53% of patients in this study. As scoliosis can progress throughout adulthood in SB (Liptak et al., 2013), continued monitoring and management for orthopaedic concerns is needed, and regular visits with an orthopedist (or other qualified providers) are indicated (Mitchell et al., 2004). The TLC program is well-positioned to fulfill this need and function as a coordinator and manager of orthopaedic concerns.

Two concerns that fell below the threshold set for determining the most “common” concerns but are worth noting due to their clinical implications include bone health and mental health. The lack of bone health concerns among this cohort may actually reflect suboptimal screening in the initial clinical appointment and point to the ‘quiet’ nature of poor bone health, whereby only fractures or significant complications may bring attention to any bone-related issues. On the other hand, since a significant portion of patients were taking vitamin D at the time of consultation, it is also plausible that osteoporosis or fracture risk was considered well-managed among patients transitioning into the program during this period and therefore not frequently identified as a concern. Osteoporosis and bone health issues are common among people with SB and more common in adults with SB than in the general population (Liptak et al., 2013; Valtonen, Goksör, Jonsson, & Mellström, 2006). People with a higher level of neurological impairment and youth are especially at an increased risk of fractures (potentially due to inexperience navigating the physical environment and changes in bone mineral density during adolescence) (Dosa, Eckrich, Katz, Turk, & Liptak, 2007; Marreiros, Loff, & Calado, 2012). As such, TLC programming must consider bone health in treating and rehabilitating patients with SB and specifically consider risk factors for fractures and low bone mineral density in future clinical encounters (Marreiros et al., 2012; Trinh et al., 2017; Valtonen et al., 2006).

Similarly, mental health concerns were less frequent among this group. Only 15% of the cohort had a specific concern relating to affective disorders, and only a small number of other mental health concerns were identified by this study. However, mental health problems are frequent and undertreated in people with SB (Kritikos, Smith, & Holmbeck, 2020; Liptak et al., 2013) A recent study by Dicianno, Kinback, et al. (2015) found that over 25% of adult participants (including younger and older adults) had depressive symptoms, which was comparable to rates in younger people with SB. The authors also estimated that the rate of participants with a history of depression could have been as high as 46% (B. Dicianno et al., 2015). Furthermore, The Mental Health Guidelines for the Care of People with Spina Bifida by Kritikos et al. (2020) summarize that due to the social, cognitive, physical, and neuropsychological challenges surrounding SB, people with SB are at risk for symptoms of depression, anxiety, and lower quality of life than the general population (Kritikos et al., 2020). Although the TLC program can screen and provide interventions for mental health issues at the initial patient consultation, specific, concrete strategies could be integrated into future initial clinic visits and follow-up appointments to meet the guidelines proposed by Kritikos et al. (2020).

Associations

None of the hypothesized associations were significant after accounting for multiple comparisons using the Bonferroni correction. This finding (or lack thereof) is a limitation and may be due to the small sample size. However, several variables reached significance before the application of the Bonferroni correction. Because this study is exploratory, and since limited research exists regarding the associations between health care concerns and specific patient factors, readers should be aware of which associations would have reached significance (if not for accounting for multiple comparisons) to inform future larger-scale studies.

In this study, it was hypothesized that pain would be significantly associated with sex as previous research has shown that women reported higher pain intensity or are more sensitive to pain and are more likely to report pain compared to men (Fillingim, King, Ribeiro-Dasilva, Rahim-Williams, & Riley, 2009; Lidal et al., 2019). However, pain was not significantly associated with sex after adjusting for multiple comparisons. This finding aligns with research by Werhagen, Hutling, and Borg (2010), who found that pain was not associated with gender, completeness of injury, or neurological level in adults with SB (Werhagen et al., 2010).

Although age was weakly correlated with concerns about diet and sexual/reproductive health and family planning, the associations were not significant after applying the Bonferroni correction. However, the initial correlation between age and diet may suggest an increase in diet-related concerns later in adulthood or certain nutrition and weight-management concerns being specific or more common during select periods in adulthood (Mcpherson et al., 2020). Sexual/reproductive health and family planning may have also correlated with age since most concerns in this category related to family planning, which included concerns around birth control and SB prevention. Some patients also had concerns or questions about fertility and pregnancy/childbearing, which would most likely have been more common concerns for patients of reproductive age. The relatively young mean age further increases the likelihood of such an association in this sample.

Skin health was also no longer significantly associated with neurological level after controlling for multiple comparisons. However, it may be worthwhile to investigate this association in future studies. Kim et al. (2015) found that pressure ulcers were more common in people with myelomeningocele than in people with other SB types, potentially due to differences in ambulation (Kim et al., 2015). After adjusting for SB type, the SB clinic participants attended, and age, the authors also found that in addition to wheelchair use, urinary continence, shunt presence, upper orthopaedic surgery, recent surgery, and male sex, lesion level was significantly associated with the presence of pressure ulcers, where higher lesion level (i.e., a greater level of impairment) corresponded to an increase in the risk of having a pressure ulcer (Kim et al., 2015; Liptak & Dosa, 2010). Higher lesion level generally correlates with increased motor and sensory loss and wheelchair use, potentially explaining the association between functional level and skin health in this study (Kim et al., 2015; Liptak & Dosa, 2010).

Neurological level was not significantly associated with pain and physical activity/fitness after using the Bonferroni correction. Currently, there is a paucity of research on pain in people (including adults) with SB (Alriksson-Schmidt et al., 2018). Specifically, more research is needed to determine the factors associated with pain, the nature of pain, and effective pain management strategies for adults with SB (Alriksson-Schmidt et al., 2018; Clancy, McGrath, & Oddson, 2007). In this study, those with a lower neurological lesion may experience less sensory loss (Liptak & Dosa, 2010) and thus, may be at a greater risk of experiencing condition-related pain due to increased sensation. However, in a study on pain in children and adolescents with SB, pain variables, including pain frequency, intensity, location, and duration, were not associated with neurological lesion level (Clancy et al., 2007). As such, future research, with larger samples, may consider studying the factors that associate with pain in SB and whether neurological level can predict aspects of pain in adults with SB.

Lastly, the research examining the association between neurological level and physical activity is scant. However, some studies have looked at the association between physical activity and other factors. In a study exploring psychosocial correlates of physical activity among youth with SB, only the variable ‘perception of competence’ was positively associated with participating in physical activity (Marques, Maldonado, Peralta, & Santos, 2015). In a study of youth with SB who used wheelchairs by Bloemen et al. (2020), being older and non-ambulatory was negatively associated with physical activity. In this study, the association between neurological level and physical activity/fitness was weak and not significant after controlling for multiple comparisons. However, such an association may appear in studies with larger samples.

Similar to Marques et al. (2015), in this study, this association may be explained by greater preservation of mobility and ambulation in people with lower-level neurological lesions and a greater ability to participate in physical activity. Again, neurological level typically determines extent of sensory and motor loss (Liptak & Dosa, 2010), and higher-level lesions are associated with number of secondary conditions (Verhoef et al., 2004). Those with higher-level lesions and hydrocephalus (i.e., more severe impairment) are also likely to have lower functional independence, especially in locomotion (Verhoef et al., 2006). As physical activity/fitness concerns were often concurrent with pain or weight management concerns in this population, this finding may also point to an even greater need for physical activity among patients with a higher neurological level. More research is needed to investigate this association.

Regression Analysis

None of the identified patient factors were significantly associated with total number of concerns after accounting for multiple comparisons. Sex, type of SB, and ambulatory status could not explain the number of concerns identified by patients. Furthermore, age and functional level (although significant predictors of total number of concerns in the log-transformed regression) did not explain total number of concerns such that the results can be applied to the clinical setting. Therefore, there were no identifiable patient groups that had more health concerns over others. Many patients also presented to the clinic with a similar number of concerns. This conclusion appears to reinforce the need for patient-centred, individualized care for all people with SB, regardless of patient characteristics or other factors. In defining health care transition, multiple organizations have emphasized the need for individually oriented care for patients and their families (American Academy of Pediatrics et al., 2002; Rosen et al., 2003). Liptak & Dosa (2010) noted that multiple factors, including the severity of one's condition, their support system, and the availability of community resources, influence the transition to adult health care (Liptak & Dosa, 2010). The model proposed by Holmbeck et al. (2021) on the transition to adult care in youth with SB suggests that several factors, including executive functioning, family cohesion, peer support, mental health, socioeconomic status, and self-management skills, predict medical adherence and ultimately the types and number of health complications. As such, it is possible that the number of conditions a patient presents with at the initial clinical encounter results from a complex interaction between the patient and their environment and is specific to the individual and their family, necessitating an individualized approach to care, such as the one provided by the TLC program. Furthermore, Young et al. (2009) documented that among Canadian adolescents and adults with cerebral palsy, acquired brain injury, and SB, people with SB had the lowest-rated health status and that global health ratings were poor in adults with SB compared to adolescents with SB (Young et al., 2006). This may help explain the large average number of presenting health concerns at the initial clinical consultation.

Considerations and Future Directions

SB is a unique and important population to study, and only recently has research begun to emerge on the specific transition and lifelong care needs of this population. SB can be an extremely complex condition and typically presents a complex transition challenge (Holmbeck et al., 2021). Adolescents and adults must manage the transfer of multiple areas of care to multiple new health care providers, all while learning to self-manage their condition and comorbidities, advocate for themselves, and coordinate these diverse health care services (Holmbeck et al., 2021). This transition proves to be even more complex for those who have reduced cognitive functioning and are heavily reliant on caregivers (Holmbeck et al., 2021). While SB has its unique challenges and clinical presentations, people with chronic childhood-onset disabilities share a common need for planned and coordinated transition care, as well as ongoing support throughout adulthood, including rehabilitative, medical, and social services (American Academy of Pediatrics et al., 2002; Binks et al., 2007). Thus, the results of this study, particularly the need for individualized, coordinated care, may apply to people with other chronic disabilities of childhood-onset.

Interestingly, in evaluating health and health care utilization among adults with SB from a multidisciplinary adult clinic in the United States, Liptak et al. (2016) found that many participants still experienced difficulty with accessing health care due to inadequate medical resources or for other unspecified reasons. Although programs such as the TLC program may be helpful, it is possible that barriers to accessing care still exist, which may continue to adversely affect health outcomes in adults with SB. Despite recent efforts to develop and evaluate transition programs and models, much of the published research is unreliable due to the methodologic and design limitations within the studies and the lack of information provided for replication and program development (Betz et al., 2018). While program development is still in its infancy, research on optimal processes and outcomes of transition programming is critical to understand what is needed in transition programs to achieve optimal health through adolescence and adulthood for those with childhood-onset disabilities.

Future Research

As this was a retrospective study evaluating patient needs at the initial clinical encounter with the TLC program, this study only reflects patient concerns at the initial point of contact, during the transition or re-initiation of care with the TLC clinic. Future research will need to determine whether these patient needs have been addressed and if patients are satisfied with the services provided by the TLC program. Other initiatives include retrospective evaluations of the concerns and needs of other TLC populations (i.e., cerebral palsy) and prospective evaluations of patient needs (thus providing an opportunity to separate patient/caregiver-reported concerns from health care provider concerns). Since this study could not address the third aim of the IHI Triple Aim Framework (reduce per capita costs of health care) (Berwick et al., 2019), future research should also include an economic evaluation of the TLC program to address this aim. Additional research initiatives within the TLC may involve evaluating changing patient needs over time and evaluating differences in concerns of patients lost to follow-up versus those who continue to access services. Future research should also consider mixed-methods or qualitative study designs to fully capture patient and family needs, experiences, and voices. Ultimately, this research will contribute to an evaluation of the TLC program as a process of quality improvement.

Limitations

The major limitation of this study was its retrospective nature. Due to the study design, specific data elements were not consistently reported within patient medical charts (as multiple standard clinic data collection forms were available and filled by various team members). Therefore, standardizing the data collection and extraction of particular data elements of interest was challenging and resulted in the exclusion of specific variables, including communication and hip status, from this initial study. Furthermore, there was no method to distinguish between concerns reported by the patient or caregiver and concerns raised by the physician. This study had a significant proportion of female patients, where other studies with large sample sizes have found a near equal representation of male and female participants (Sawin et al., 2015) and only a slightly higher birth-prevalence rate among females (Lary & Edmonds, 1996). To determine whether the non-significant associations and low percentage of variance explained in the regression model (with age and neurological level) were due to lack of statistical power, a post hoc power analysis was conducted. The analysis revealed that this study had a limited sample size and was underpowered. Some of the less common concerns identified by this study may have also been more prominent concerns in a study with a larger sample size. The Bonferroni correction is a conservative approach and although it may reduce the chance of a type I error, it decreases the chance of detecting significance and possibly increases the risk of type II error. This is particularly relevant considering large number of statistical tests performed in this initial study and the resulting small significance values. In the regression analysis (due to the choice of backward stepwise regression), the small sample size and low power may also have caused underfitting (causing important predictors of total number of concerns to be left out). This study also has an inherent selection bias in including only TLC patients in this study. Those with less pressing concerns may never have been referred for comprehensive management, thus introducing the possibility of over-estimating the health care needs of this population. Lastly, this study did not consider other social characteristics such as ethnicity, education level, or employment status. While these factors could conceivably be related to patient concerns, the limited information available within patient medical charts meant that these data elements could not be reliably extracted or assessed.

In this sample, age was not normally distributed, most patients were 19 years of age, and very few patients were older than 60. As such, the most pressing concerns of this cohort, as revealed in this study, may not reflect the topmost concerns of the entire SB population in this particular geographical area and may be more applicable to those around the typical age of transition (18-21 years of age). Currently, limited research exists on the concerns, characteristics, functional abilities, and participation of older adults with SB (Lidal et al., 2019). The literature is especially limited in the concerns of those over 50 years of age (Lidal et al., 2019). The TLC program is a relatively new clinical service and as such, may not be as accessible to older people who have long passed the age of transition and are not aware of the service. It is also possible that people with SB continue to experience a high mortality rate into adulthood (Oakeshott et al., 2009), and consequently, there are fewer older people with SB who can access the TLC program.

On the contrary, it is plausible that those who survive later into adulthood are higher functioning adults and thus, require fewer health and social services. Liptak et al. (2016) found that older adults with SB have higher rates of hospitalization and fewer interactions with primary care compared to younger people with SB, which could further explain the smaller population of older adults with SB within the TLC program during this particular period. Ultimately, although this study attempted to identify the concerns of all TLC patients with SB, more research is needed to conclude whether these concerns apply to older adults with SB.

Chapter 6: Conclusion

This study identified many common health concerns in adolescents and adults with SB initiating care in the TLC program. In particular, it established the significance of the TLC's role in care coordination, medication management, and facilitating assistive device use. It also identified common SB-related health conditions of concern, most notably, neurogenic bladder. At the initial clinical consultation, patients frequently presented with multiple health concerns. However, this study could not identify specific patient factors, such as sex, age, or neurological level, that contributed to the most common health concerns or each patient's total number of concerns. As such, there is a need for continuous and individualized care for people with SB throughout the lifespan, in line with expert recommendations (American Academy of Pediatrics et al., 2002). It is important that physiatrists and other health care providers within the TLC clinic continue to address well-documented concerns, including neurogenic bladder and bowel, mobility, social, and pain concerns. Consideration also must be given to less frequently discussed topics (identified in the literature as significant in this population), such as sexual health, mental health, and bone health. This research is the first critical step in TLC quality improvement and program evaluation, and in addressing the aims of the IHI Triple Aim Framework (improving the experience of care, improving the health of populations, and reducing health care costs). The findings of this study will inform further growth and development of the TLC clinic such that the program aligns with patient needs and optimizes their transition from paediatric to adult care. This study can also help inform priority setting in developing other transition efforts and programming to improve the standard of care for this population more globally.

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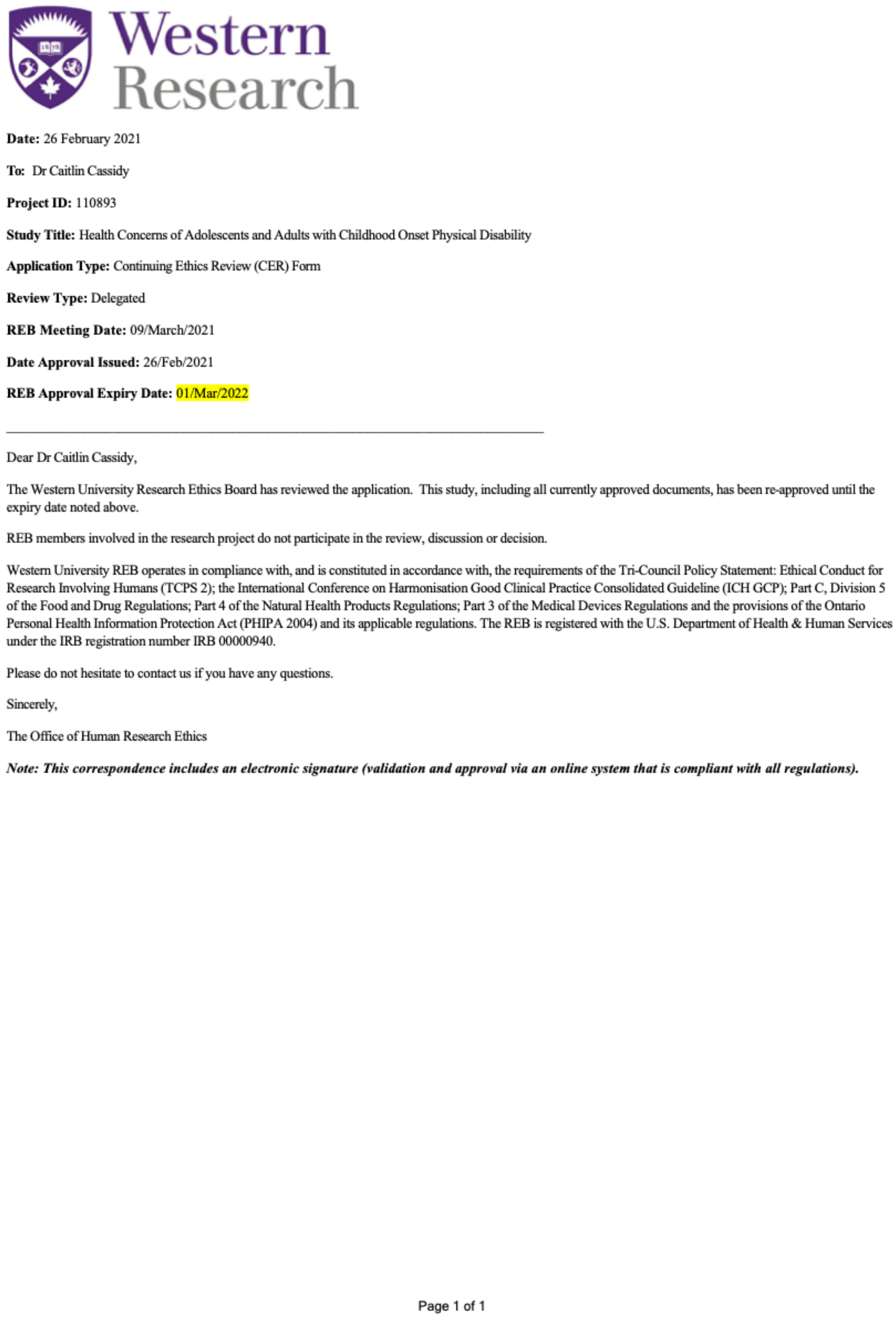
Appendix A: Ethics Approval



Western
Research

Date: 26 February 2021

To: Dr Caitlin Cassidy

Project ID: 110893

Study Title: Health Concerns of Adolescents and Adults with Childhood Onset Physical Disability

Application Type: Continuing Ethics Review (CER) Form

Review Type: Delegated

REB Meeting Date: 09/March/2021

Date Approval Issued: 26/Feb/2021

REB Approval Expiry Date: 01/Mar/2022

Dear Dr Caitlin Cassidy,

The Western University Research Ethics Board has reviewed the application. This study, including all currently approved documents, has been re-approved until the expiry date noted above.

REB members involved in the research project do not participate in the review, discussion or decision.

Western University REB operates in compliance with, and is constituted in accordance with, the requirements of the Tri-Council Policy Statement: Ethical Conduct for Research Involving Humans (TCPS 2); the International Conference on Harmonisation Good Clinical Practice Consolidated Guideline (ICH GCP); Part C, Division 5 of the Food and Drug Regulations; Part 4 of the Natural Health Products Regulations; Part 3 of the Medical Devices Regulations and the provisions of the Ontario Personal Health Information Protection Act (PHIPA 2004) and its applicable regulations. The REB is registered with the U.S. Department of Health & Human Services under the IRB registration number IRB 00000940.

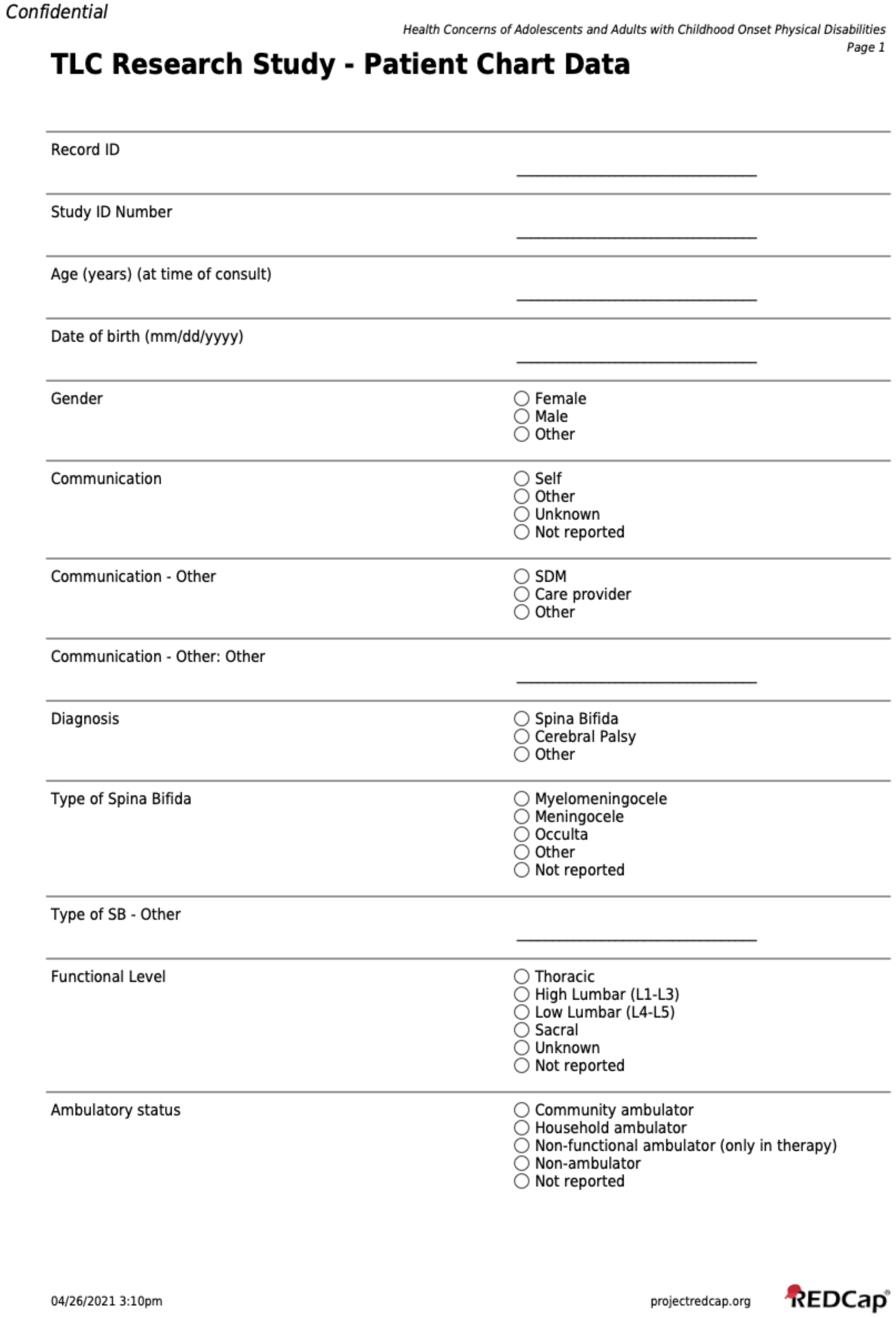
Please do not hesitate to contact us if you have any questions.

Sincerely,

The Office of Human Research Ethics

Note: This correspondence includes an electronic signature (validation and approval via an online system that is compliant with all regulations).

Appendix B: REDCap Data Extraction Tool



Confidential

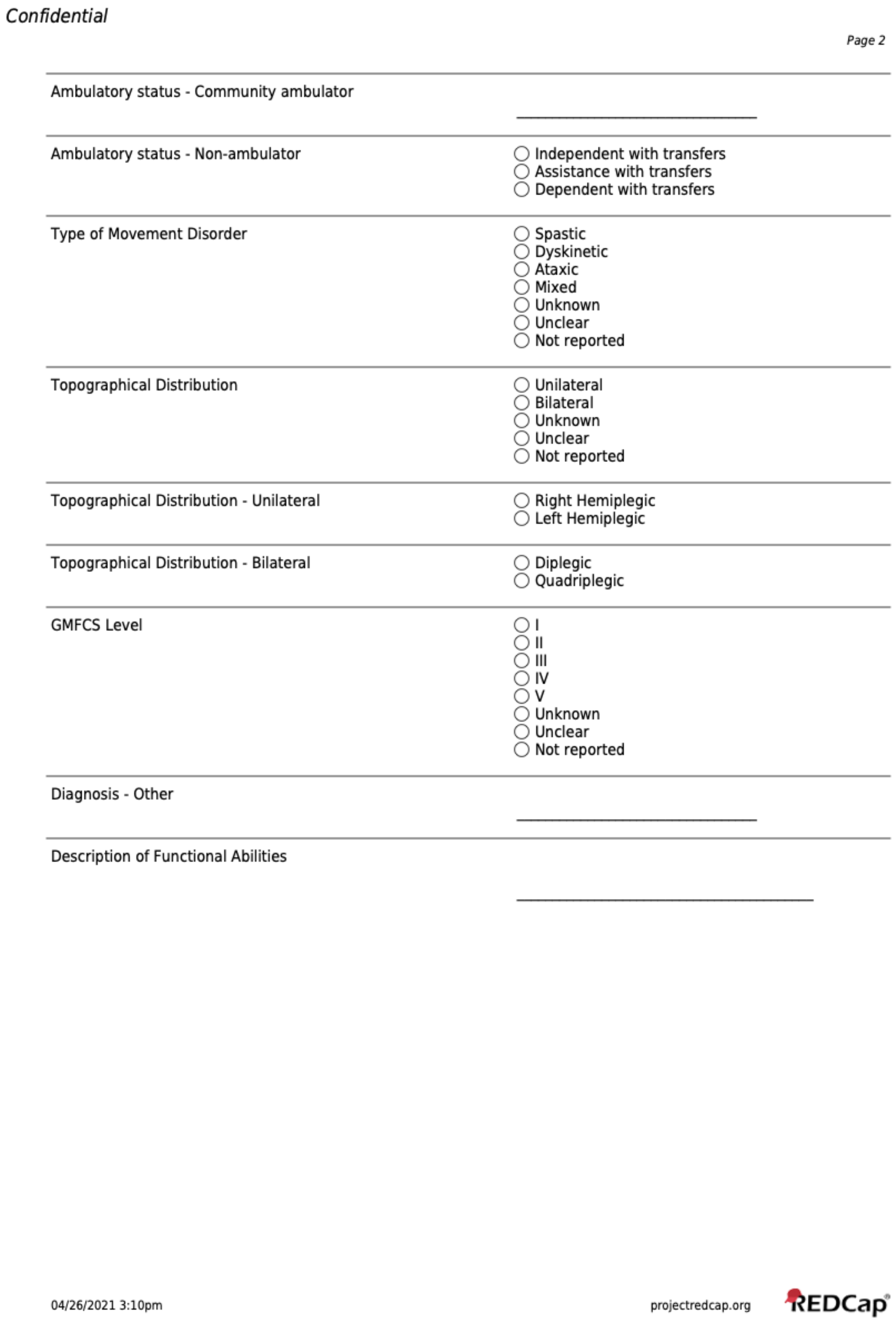
Health Concerns of Adolescents and Adults with Childhood Onset Physical Disabilities
Page 1

TLC Research Study - Patient Chart Data

Record ID	______________________________
Study ID Number	______________________________
Age (years) (at time of consult)	______________________________
Date of birth (mm/dd/yyyy)	______________________________
Gender	○ Female ○ Male ○ Other
Communication	○ Self ○ Other ○ Unknown ○ Not reported
Communication - Other	○ SDM ○ Care provider ○ Other
Communication - Other: Other	______________________________
Diagnosis	○ Spina Bifida ○ Cerebral Palsy ○ Other
Type of Spina Bifida	○ Myelomeningocele ○ Meningocele ○ Occulta ○ Other ○ Not reported
Type of SB - Other	______________________________
Functional Level	○ Thoracic ○ High Lumbar (L1-L3) ○ Low Lumbar (L4-L5) ○ Sacral ○ Unknown ○ Not reported
Ambulatory status	○ Community ambulator ○ Household ambulator ○ Non-functional ambulator (only in therapy) ○ Non-ambulator ○ Not reported

04/26/2021 3:10pm

projectredcap.org



Ambulatory status - Community ambulator	______________________
Ambulatory status - Non-ambulator	○ Independent with transfers ○ Assistance with transfers ○ Dependent with transfers
Type of Movement Disorder	○ Spastic ○ Dyskinetic ○ Ataxic ○ Mixed ○ Unknown ○ Unclear ○ Not reported
Topographical Distribution	○ Unilateral ○ Bilateral ○ Unknown ○ Unclear ○ Not reported
Topographical Distribution - Unilateral	○ Right Hemiplegic ○ Left Hemiplegic
Topographical Distribution - Bilateral	○ Diplegic ○ Quadriplegic
GMFCS Level	○ I ○ II ○ III ○ IV ○ V ○ Unknown ○ Unclear ○ Not reported
Diagnosis - Other	______________________
Description of Functional Abilities	____________________________

Surgical History	☐ None ☐ Achilles ☐ Hamstrings ☐ Adductors/hip soft tissue ☐ Hip osteotomy ☐ Salivary Procedure ☐ Fundoplication ☐ Tib ant tendon transfer ☐ Tib post tendon transfer ☐ Scoliosis ☐ Bladder augmentation ☐ Mitrofanoff ☐ Cecostomy (c) tube ☐ MACE procedure ☐ VP shunt ☐ Tethered cord release ☐ Chiari decompression ☐ Other: Orthopedic ☐ Other: Non-Orthopedic ☐ Unclear ☐ Unknown ☐ Not reported
Surgical history - Other: Orthopedic Surgery	__________________________________
Surgical history - Other: Non-Orthopedic Surgery	__________________________________
Hip Status	○ Bilaterally Located ○ R Subluxed ○ R Dislocated ○ L Subluxed ○ L Dislocated ○ Other ○ Unknown ○ Unclear ○ Not reported
Hip status - Other	____________________________
Epilepsy History	○ Yes ○ No ○ Unknown ○ Unclear ○ Not reported
Epilepsy History - Yes	○ No seizure in >5 years ○ No seizure in 1-5 years ○ Seizure frequency > 1/day ○ Seizure frequency daily ○ Seizure frequency weekly ○ Seizure frequency monthly ○ Seizure frequency yearly ○ Seizure frequency < yearly ○ Other

Epilepsy History - Yes: Other ______________________________

Medication(s)

- ☐ Amitriptyline (Elavil)
- ☐ Aripiprazole (Abilify)
- ☐ Baclofen (oral)
- ☐ Baclofen (intrathecal)
- ☐ Baclofen (unspecified)
- ☐ Botulinum Toxin
- ☐ Calcium
- ☐ Cannabis
- ☐ Carbamazepine (Tegretol)
- ☐ Citalopram (Celexa)
- ☐ Clonazepam
- ☐ Diazepam
- ☐ Dilantin
- ☐ Domperidone
- ☐ Duloxetine (Cymbalta)
- ☐ Escitalopram (Cipralex)
- ☐ Fluoxetine (Prozac)
- ☐ Gabapentin
- ☐ Iron
- ☐ Lacosamide (Vimpat)
- ☐ Lactulose
- ☐ Lamotrigine (Lamictal)
- ☐ Levetiracetam (Keppa)
- ☐ Levodopa
- ☐ Linaclotide (Constella)
- ☐ Melatonin
- ☐ Milk of magnesia
- ☐ Nabilone
- ☐ Nitrazepam
- ☐ Olanzapine (Zyprexa)
- ☐ Peg/peglyte/peg 3350/restoralax/lax-a-day
- ☐ Phenobarbital
- ☐ PPI
- ☐ Ranitidine
- ☐ Resotran
- ☐ Risperidone
- ☐ Scopolamine
- ☐ Serokot/Senna
- ☐ Seroquel (Quetlapine)
- ☐ Sertraline (Zoloft)
- ☐ Suppositories (Dulcolax, glycerin)
- ☐ Tizanidine
- ☐ Topiramate (Topamax)
- ☐ Trazodone
- ☐ Tylenol regularly scheduled
- ☐ Valproic acid
- ☐ Venlafaxine (Effexor)
- ☐ Vitamin D
- ☐ Zopiclone (Imovane)
- ☐ Other: PRN
- ☐ Other: Regular
- ☐ None
- ☐ Unknown
- ☐ Unclear
- ☐ Not reported

Medication - Other: PRN

____

Medication - Other: Regular

____

Presenting Concerns	○ Retrospective ○ Prospective
Presenting concerns - Retrospective	______________________________
Presenting concerns - Prospective - patient/caregiver	______________________________
Presenting concerns - Prospective - healthcare provider	______________________________
Physical Exam Results (presence of contractures)	☐ R Hip Flexion ☐ L Hip Flexion ☐ R Knee Flexion ☐ L Knee Flexion ☐ Scoliosis ☐ R Elbow Flexion ☐ L Elbow Flexion ☐ R Wrist Flexion ☐ L Wrist Flexion ☐ R Plantar flexion ☐ L Plantar flexion ☐ Other ☐ None ☐ Unknown ☐ Unclear ☐ Not reported
Physical Exam - Other	______________________________

Curriculum Vitae

Name:	Jessica Starowicz
Education	Western University London, Ontario, Canada MSc., Health and Rehabilitation Sciences 2019-2021
	The University of Western Ontario London, Ontario, Canada BHSc. Honors Specialization in Health Sciences 2015-2019
Honours and Awards	James Mackinnon Local 1059 Scholarship Award 2020
	Dean’s Honor List 2017, 2018, 2019
Related Work Experience	Research Project Coordinator St. Joseph’s Healthcare 2021
	Graduate Research Assistant Western University 2020-2021
	Graduate Teaching Assistant Western University 2020
	Graduate Fellow Western University 2019-2020
	Graduate Teaching Assistant Western University 2019
Certifications & Training	Introduction to Qualitative Analysis with NVivo Western Libraries, Western University 2021
	Sex and Gender in Primary Data Collection with Human Participants CIHR IRSC

2020

safeTALK
Centre for Teaching and Learning, Western University
2019

TCPS 2: CORE (Course on Research Ethics)
Government of Canada
2018

Workshop Participation

Volunteers-In-Progress Workshop
Western University
2019

Leadership Education Program (LEP) Participant
Western University
2017-2019

Memberships

Canadian Child Health Clinician Scientist Program
2019- present

Other Related Activities

Functional Electrical Stimulation (FES) Assistant
St. Joseph’s Health Care London
2020

Promoter
Living Well @ Western, Western University
2019

Welcome Team Member
IESC, Western University
2018-2019

Recreation Volunteer
Mount Hope for Long-Term Care, St. Joseph’s Health Care London
2018

Exercise Program Assistant
Canadian Centre for Activity and Aging, Western University
2018

Publications

<u>Submitted Manuscripts</u>

Starowicz, J., Pratt, K., McMorris, C., & Brunton, L. (Submitted April 9, 2021). Mental health benefits of physical activity in youth with cerebral palsy: A scoping review. *Physical and Occupational Therapy in Pediatrics.*

Conference Participation

Starowicz, J., Pratt, K., McMorris, C., & Brunton, L. (2020). Mental health benefits of physical activity in youth with cerebral palsy: A scoping review. AACPDM 74th Annual Meeting, American Academy for Cerebral Palsy and Developmental Medicine, Live Virtual Meeting, September 24-26, 2020 (Poster).

Starowicz, J., Pratt, K., McMorris, C., & Brunton, L. (2019). Mental health benefits of physical activity in youth with cerebral palsy: A scoping review. Child Health Symposium, Western University & Thames Valley Children's Centre, London, ON, May 10, 2019 (Poster).

Starowicz, J., Pratt, K., McMorris, C., & Brunton, L. (2019). Mental health benefits of physical activity in youth with cerebral palsy: A scoping review. Western Student Research Conference (WSRC), Western University, London, ON, March 29, 2019 (Poster).

Starowicz, J., Pratt, K., McMorris, C., & Brunton, L. (2020). Mental health benefits of physical activity in youth with cerebral palsy: A scoping review. Empowered Kids Ontario (EKO) Spring Symposium 2020, Toronto, Ontario, March 30-31, 2020 (Conference Cancelled due to COVID-19).