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Transition from Paediatric to Adult Healthcare: Readiness and Experiences of Canadian Adolescents and Young Adults Diagnosed with Epilepsy in Childhood

Alison S.B. Dasiewicz, *The University of Western Ontario*

Supervisor: Dr. Kathy Nixon Speechley, *The University of Western Ontario*

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Abstract

Documenting the transition from paediatric to adult healthcare is important to guide the delivery of developmentally appropriate healthcare. The objectives of this research were to quantify the transition readiness of adolescents and young adults (AYAs) diagnosed with epilepsy in childhood, explore its association with individual and familial factors, and describe transfer/transition experiences for AYAs and parents. Families and physicians were followed for ten years as part of the Health-Related Quality of Life in Children with Epilepsy Study and provided report using questionnaires.

The distribution of transition readiness indicates much room for improvement. Age was the only factor significantly associated with transition readiness, however intriguing trends were observed for several factors. Transfer/transition experiences were not specific to epilepsy, with both negative and positive experiences represented. As the first of its kind, this research began to uncover factors influencing transition readiness and experiences and suggests potentially fruitful avenues for future research.

Keywords

Epilepsy, transfer, transition, transition readiness, Transition-Q, self-management, adolescent, cohort study, survey, questionnaire

Co-Authorship Statement

All chapters of this thesis were written by myself, Alison Dasiewicz, to partially fulfill the requirements for the degree of Master of Science in Epidemiology and Biostatistics. The research presented in this thesis is part of the Health-Related Quality of Life in Children with Epilepsy Study (HERQULES). The long-term follow-up phase of HERQULES is funded by the Canadian Institutes of Health Research Operating Grant MOP-64311, Principal Investigator: Dr. Kathy Nixon Speechley. I was involved in most aspects of the final follow-up of HERQULES, the 10-year follow-up, including: development of the research question, selection of a transition readiness measure, creating questions in the transfer/transition section of the questionnaires, assembling and mailing questionnaire packages, data entry, data analysis, and creating summary reports. My thesis supervisor, Dr. Kathy Nixon Speechley, provided guidance and feedback in all aspects of this thesis research. Feedback on the thesis was also provided by Dr. Guangyong Zou, Dr. Carol Camfield, and other members of the HERQULES research group.

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List of Abbreviations

AED(s) – Anti-epileptic drugs(s)

AYA(s) – Adolescent(s) and young adult(s)

GP – General Practitioner / Family doctor

HERQULES – Health-Related Quality of Life in Children with Epilepsy Study

HRQL – Health-related quality of life

ILAE – International League Against Epilepsy

PCMCH – Provincial Council for Maternal and Child Health

Chapter 1 : Introduction

Children with a chronic disease being followed by paediatric healthcare providers will have to move to adult healthcare as they grow into adulthood. In the past, common practice for moving patients from paediatric to adult healthcare was little more than a referral letter from the paediatrician, referred to as transfer. This approach is quick, easy, and initially inexpensive for clinicians and the healthcare system but may result in lack of patient preparation for transfer and subsequent poor long-term patient outcomes.[1] In response, clinicians and researchers are advocating for a planned, gradual move from paediatric to adult healthcare, referred to as transition. The goal of transition is to prepare adolescents to take responsibility for their healthcare in the adult-oriented setting.[2]

The stage of life when healthcare transfer occurs is often characterized by change. In addition to developmental changes, adolescents and young adults (AYAs) may be moving away from home and/or engaging in post-secondary studies. Lifestyle and disease management can suddenly go from routine to erratic. After transfer to adult care, parental involvement in disease management can end suddenly with the adolescent unprepared to take charge of his/her healthcare.[3] In addition, the paediatric and adult healthcare systems are often contrasting with respect to environment, care culture, and resources.[4] All of these changes and differences can make it challenging for AYAs to access healthcare effectively. If an adolescent is not adequately prepared for the transfer from paediatric to adult healthcare and its consequences, there is the potential for unfavorable health consequences resulting from loss of contact with the healthcare system and poor disease management.[5]

The measurement of how prepared adolescents are to make the final move to adult healthcare, referred to as transition readiness, can be useful in facilitating the transition process. Keeping an inventory of adolescents' self-management skills and health knowledge in the form of transition readiness allows healthcare providers to identify adolescents and their families who may need additional support and in which areas. Transition readiness is also used to evaluate the efficacy of transition programs and is considered to be an indicator of future transition success.[5, 6, 7] However, the importance

that researchers and clinicians place on transition readiness is not yet fully supported by the quality of the evidence. Few studies have been done to identify predictors or correlates of transition readiness.[8–18] In addition, the method of measuring transition readiness is not consistent in the literature. There is a need for a measure that is validated and adaptable across chronic diseases.[19]

Each chronic condition presents unique challenges during transition. The solution is thought to be a global transition framework that can be adapted to meet the challenges of all chronic diseases with the help of disease-specific research.[19] Individuals diagnosed with epilepsy in childhood are a unique population about whom little research on transition has been conducted. This population is highly heterogenous in terms of symptoms, severity, and prognosis. Although many children diagnosed with epilepsy stop having seizures before adulthood, there is still a risk of having seizures again in the future. In addition, children with epilepsy are at an increased risk of several chronic comorbidities which may be difficult to treat due to a lack of healthcare system resources and lack of attention from the patient without the presence of an eminent issue like seizures.[20] Therefore, it is important that AYAs with epilepsy are prepared to manage their healthcare independently in the adult system.

The aims of this research were to report the level of preparation of AYAs with epilepsy to manage their health as adults, assess its relationship with individual and familial factors, and to provide an overview of transfer/transition experiences for a diverse sample of AYAs with epilepsy in a Canadian healthcare setting where essential health services are theoretically accessible to everyone.

1.1 Objectives

The overall goal of this thesis is to describe the transition readiness and experiences of AYAs with epilepsy and their families in Canada. To our knowledge, this study is the first of its kind in the country. We hope that the results will inform further development and improvement of transition programs for Canadian AYAs with epilepsy as an essential step in achieving better long-term outcomes for people living with epilepsy.

The specific objectives are as follows:

1. Document the readiness of AYAs with epilepsy to transition from paediatric to adult healthcare in Canada.
2. Explore the associations of individual and familial factors with transition readiness for these AYAs with epilepsy.
3. Describe the experience of transfer/transition from paediatric to adult healthcare for these AYAs with epilepsy and their parents/caregivers.

Chapter 2 : Background

2 Chapter Overview

This chapter provides the context necessary to understand the importance of transition from paediatric to adult healthcare for adolescents with epilepsy. A brief overview of the clinical features and epidemiology of epilepsy are provided. The definition of transition and examples are provided to facilitate understanding of the potential for negative outcomes in the absence of formal transition. The current state of transition literature is reviewed, including the evidence in support of transition. Lastly, the concept of transition readiness is introduced.

2.1 Epilepsy: A Chronic Disease

The term *epilepsy* encompasses a group of neurological disorders that have in common an abnormal tendency for excess neuronal activity resulting in unprovoked convulsive and/or absence and/or focal seizures.[21] The various types and syndromes of epilepsy have diverse natural histories. Clinical characteristics such as cause, age of onset, seizure frequency, ability to achieve seizure control, and type and severity of comorbidities vary widely.[22] For example, benign partial epilepsy of childhood with centrotemporal spikes (BECTS) is one of the least severe types of epilepsy. Seizures often occur during sleep and are easily controlled using anti-epileptic drugs (AEDs). Children often stop having seizures before age 16 years, cognitive impairment is rare, and social outcomes are best for those with this epilepsy syndrome.[23,24] On the other hand, severe myoclonic epilepsy of infancy (also referred to as Dravet syndrome) is characterized by frequent and prolonged seizures early in childhood that are difficult to control and are often associated with behavioural difficulties like hyperactivity as well as permanent, moderate to profound cognitive impairment.[25]

Epilepsy is the most common neurological disorder in children. The average incidence of epilepsy in Nova Scotian children aged 1 to 10 years was estimated to be 46/100,000 person-years.[26] Based on the National Longitudinal Survey of Children and Youth, the prevalence of epilepsy in Canada was estimated as 5/1,000 children aged 13 years and

younger.[27] Many children with epilepsy will achieve control of their seizures, however there is a significant risk of having another seizure in early adulthood or later in life. A population-based study found that 74% of children diagnosed with epilepsy will experience 2 years seizure-free, a length of time commonly taken by physicians as an indication to stop AED use.[28] However, 25% of this subgroup had another seizure up to 5 years later. According to the International League Against Epilepsy (ILAE), an individual is considered to no longer have epilepsy when he/she has been seizure-free for 10 years and off of AEDs for 5 years.[29] In this case, their epilepsy is considered to be "resolved". However, this definition includes the possibility that the individual could have another seizure later in life.[29]

The effects of epilepsy go beyond the chronic nature of seizures and may have lifelong consequences. Individuals diagnosed with epilepsy in childhood are more likely to have a poorer quality of life than those with other chronic diseases.[30,31] They are also more likely to experience unfavorable outcomes such as problems with social relationships and obtaining education,[32] unemployment,[33] and single parent pregnancy than their healthy peers.[32] Children with epilepsy are also more likely to have difficulties with attention, memory, Attention Deficit Hyperactivity Disorder (ADHD),[34] depression, and anxiety.[35] Therefore, caring for an individual who has epilepsy requires attention to both the immediate and long-term effects.

2.2 Transition from Paediatric to Adult Healthcare

2.2.1 Definition and Potential for Loss of Contact

The terms *transfer* and *transition* have distinct meanings in the context of healthcare. Transfer is defined as the administrative task of moving a patient with a chronic medical condition from child-centered care to adult-centered care within the healthcare system, usually in the form of a referral letter.[36] Transition is defined by the Society for Adolescent medicine as "the purposeful, planned movement of adolescents and young adults with chronic physical and medical conditions from child-centered to adult-oriented health-care systems."[37]

The goal of transition as stated by the American Academy of Pediatrics, the American Academy of Family Physicians, and the American College of Physicians is "to maximize lifelong functioning and potential through the provision of high-quality, developmentally appropriate healthcare services that continue uninterrupted as the individual moves from adolescence to adulthood."[38]

Children who are diagnosed with epilepsy are often followed by a paediatric neurologist. If a paediatric neurologist believes that an adolescent patient with epilepsy will require follow-up by a specialist into adulthood, the adolescent is transferred from the paediatric neurologist to an adult neurologist at or before age 18 years. This transfer must occur before age 18 because generally in Canada's universal healthcare system, a paediatric specialist cannot bill for services provided to someone who is age 18 years or older. The adult neurologist practice may be in a different geographic location than the paediatric neurologist practice. A paediatric neurologist may also refer the adolescent directly back to their family physician for epilepsy-related care if their epilepsy is less severe and uncomplicated.

Paediatric and adult practices can be very different. Paediatric practices tend to have a culture of family-centered care and share the responsibility for patient care with parents. Paediatric specialists use a developmental approach to healthcare, while adult care is more likely to focus on the specific problem that is present at the time of the visit.[4] Scheduling appointments for follow-up or to make up for missed appointments is typically the responsibility of clinic staff or parents in a paediatric clinic, whereas in an adult care setting this is largely the patient's responsibility.[39] In the paediatric setting, external resources and multi-disciplinary clinicians are more readily available to the patient.[2] In an adult practice, patients must take most of the responsibility for accessing care, health education, adhering to their medications, and decision-making.[40] In addition, it is not uncommon for physicians who treat adults to feel unprepared to deal with certain types of epilepsy.[41] They typically do not receive training in a developmental approach to treatment (which paediatricians are required to do) and expect to see patients only when there is a new problem.[42] For individuals diagnosed with a chronic disease in childhood, an abrupt change of moving from paediatric to adult practices could negatively affect the

access to and efficacy of the healthcare they receive if they are not adequately prepared.[2] It is not uncommon for adult neurologists to provide only a single evaluation on a referral-only basis rather than continued follow-up.[20] Consequently, if a patient loses contact with his/her adult neurologist, it can be difficult to access specialist care.

AYAs diagnosed with epilepsy in childhood face an increased risk of ADHD and depression.[43] The treatment of these chronic conditions concurrently with epilepsy requires the coordination of several different healthcare resources, which is more difficult to accomplish in the adult healthcare setting.[19, 44] Healthcare providers themselves report that there is less coordination of care in the adult healthcare system when a patient requires care from more than one healthcare provider and less availability of resources like subspecialist consultations, social work, and psychiatry.[40, 45]

Epilepsy is episodic and the lifestyle changes that characterize young adulthood could trigger seizure activity. Young adults may be moving away from home and/or attending post-secondary school. Chronic lack of sleep, fatigue, changes in diet, stress, and consumption of alcohol and illicit drugs could play a role in their lives and each may be potential seizure triggers.[46, 47] A follow-up survey of adults diagnosed with epilepsy in childhood and their families conducted 20-30 years after they were diagnosed asked participants if they could identify the cause of their epilepsy.[48] Of these adults and families, 60% identified a diagnostic cause(s) that was discordant with what could be identified from their medical records (including idiopathic) and had been explained to parents/caregivers around the time of diagnosis. Of the causes identified by adults and families that were discordant, 33% were deemed implausible by the investigators, such as “an electrical storm outdoors” or “medication used for head lice.” This study provokes the concern that adults diagnosed with epilepsy in childhood may not remember details about their medical history that could be important for avoiding seizure triggers or helpful when recounting their medical history to healthcare providers in an emergency situation.

Clearly there is substantial potential for inadequate self-management of health and loss of contact with the adult healthcare system after transfer from child- to adult-oriented

healthcare. Several professional medical organizations have recognized this and developed transition recommendations aimed at directing a transition process that will increase adolescents' capacity for independence, self-advocacy in the healthcare system, disease management, and timely access to appropriate contacts in the adult healthcare system.

2.2.2 Transition Recommendations

Professional medical organizations have published recommendations advocating for transition as a crucial component of healthcare for AYAs with a chronic disease. They include the Society for Adolescent Medicine[49]; Canadian Paediatric Society[2]; American Academy of Pediatrics, American Academy of Family Physicians, and American College of Physicians – American Society of Internal Medicine[38]; and the American Academy of Pediatrics[50]. These recommendations are not disease-specific, reflecting the prevailing opinion that many core elements of transition apply to all chronic diseases. In fact, the American Academy of Pediatrics recommends that all adolescents be encouraged to take on increasing responsibility for their healthcare as part of family-centered care.[51] Common elements characterizing recommendations for a successful transition include:

- Services and health education should be provided in accordance with age and developmental stage.
- The patient should be given the opportunity to take on increasing responsibility for disease-management and interaction with healthcare providers.
- Health education should be both general and disease-specific. It should address current concerns as well as concerns that could arise in the future as a result of the specific disease.
- A written transition plan should be developed in the patient's early teens.
- An up-to-date medical summary should be provided to the patient.
- The patient's progress within in the transition framework should be evaluated several times.
- There should be direct communication between paediatric and adult healthcare providers.

- A joint appointment with the patient should be attended by both the paediatric physician and adult physician before transfer occurs.

In Ontario, the recommendations provided by the Provincial Council for Maternal and Child Health (PCMCH) to healthcare providers follow the Good2Go Shared Management Model used at the Hospital for Sick Children in Toronto.[19] This model includes all of the elements mentioned above. The PCMCH has also called for the use of a validated transition readiness measure to guide the intensity of transition planning on an individual level as one of the top priorities for the next few years.[19]

Formal transition programs and physician practices implement these recommendations to varying degrees. One study examined five well-known formal transition programs in Canada and their implementation of transition recommendations[52] and found that only two of the five implemented all nine recommendations of the Canadian Pediatric Society and the Society for Adolescent Medicine: ON TRAC[53] and the Good2Go Shared Management Model.[54] Only three of the five formal transition programs serve individuals with epilepsy: ON TRAC, the Good2Go Shared Management Model, and the Be Your Own Boss program.[55] These programs do not accommodate adolescents with epilepsy that is in remission yet who still have comorbidities that may continue to affect them in adulthood. According to the 2005-2006 National Survey of Children with Special Health Care Needs in the United States, only 41% of parents reported that youth had experienced at least a minimum standard of transition care (defined as parental report of two out of four of the following: physician discussed transfer or adult healthcare needs or health insurance, physician usually/always encouraged that the child take responsibility for their health).[56] Further investigation is needed to determine the extent to which transition recommendations are being implemented and how general transition guidelines can be modified for application to all paediatric chronic illnesses.

2.3 The Evidence for Transition

It is difficult to evaluate the benefits of transition due to the heterogeneity of existing transition programs. For example, one transition clinic in the United Kingdom consists of a joint consultation with the adolescent, the paediatric neurologist, and the adult neurologist where verification of diagnosis, review of medications, and discussion of how epilepsy can affect future health and lifestyle are the focus.[57] Alternatively, the ON TRAC transition model is designed to begin as early as 10 years of age involving multidisciplinary care during regular clinic appointments, six recommended content areas of education, a transition checklist, resources for adolescents (ex. a portable health planner), and gradually phasing out parental involvement in decision-making.[53] The method of measuring transfer/transition success also varies widely in the literature. Examples include patient satisfaction with transfer, disease-specific measures of disease management, or clinical patient outcomes.

Assessing the effectiveness of transition requires a clear description of what it means to have clinically meaningful transition success. The definition of transition implies a multi-faceted goal of optimizing health literacy, health outcomes, and quality of life. In the literature, the success of transition programs is evaluated using measures of disease management, health literacy, transition readiness, patient satisfaction, and healthcare access. It has been suggested that to make a comprehensive case that transition is more beneficial than transfer, three domains must be addressed in the literature: experience of transition care, patient health outcomes, and cost to the healthcare system.[5] These three categories will be addressed in the following sections.

2.3.1 Transfer and Transition Experiences

Many studies have collected transition experiences of AYAs affected by chronic diseases like diabetes, organ transplant, and juvenile idiopathic arthritis that have contributed to the foundation for a global transition framework.[58,59] A key theme that emerged from these studies was that AYAs thought preparedness, together with age, was important in transfer timing. Lack of preparedness for the transfer led AYAs to feel as if responsibilities and changes were piling up in their lives and that healthcare providers did

not care about them. On the other hand, feeling prepared while still in paediatric care led AYAs to feel frustrated that they were being treated like children when they felt ready to be treated like adults.[59] This illustrates that transfer is not a challenge for all adolescents. It highlights the need for implementation of a transition readiness measure that allows healthcare providers to focus transition resources on patients and families that need it most.[19]

Another common theme among AYAs was change in relationships and care culture.[59] Some AYAs enjoyed being more involved in decision-making in the adult environment. Others found the change to be intimidating or tedious, feeling that adult healthcare providers did not really care about them and they were not yet interested in taking full responsibility for their health.

One study documenting young adults' experiences of transfer found that many considered transfer to adult care as "no big deal", yet they consistently reported transfer as being more stressful than they first thought.[40] They were not prepared for the change in environment or to forge a new relationship with their adult care provider. Almost all young adults reported that they were not involved in decisions regarding transfer. This study also found that parents commonly feel they were not ready to relinquish control of their child's health because they were not able to trust that their child was ready to take on the responsibility. Healthcare providers commonly reported that paediatrics patients and their families were "spoilt" and they did not know what to do when the responsibility of managing their health on their own was suddenly put on them in the adult system.[40]

Examining the transition experiences of AYAs, parents, and healthcare providers affected by epilepsy would provide insight into how a global transition framework should be adapted to produce epilepsy-specific transition programs. However, research on the experiences and needs of those with epilepsy is sparse. The literature that does exist on this topic is restricted to the most severe epilepsy syndromes which are a small minority of those diagnosed with epilepsy in childhood. In fact, it is common that most adolescents with epilepsy participating in a transition program have less severe epilepsy. For example, in one study, only 22 out of 97 adolescents (23%) attending an epilepsy

transition clinic had severe cognitive impairment.[60] Individuals with more severe forms of epilepsy have vastly different needs as moderate cognitive impairment is often a consequence and prognoses for seizure control are worse.[61]

For AYAs diagnosed with epilepsy in childhood who have cognitive impairment, transfer was sometimes initiated because of an emergency. Parents had no prior preparation for transfer and did not receive adequate preparation because the need for transfer was immediate and urgent.[61] For those with Dravet syndrome, there was on average an eight month gap between the last consultation with the paediatrician and the first consultation with the adult specialist.[62] This study also found that 40% of caregivers were not satisfied with the transition preparation they received. Little evidence exists for the transfer and transition experiences of the vast majority of adolescents with less complicated epilepsy.

2.3.2 Patient Health Outcomes

A systematic review found 10 studies that evaluated transition as an intervention and measured post-transfer patient health as an outcome.[1] Eight of these studies compared a transition group with a comparator group (either a transfer group or a group with less intensive transition procedure). Healthcare outcomes were measured by markers of disease management like Hb1Ac in those with diabetes, rate of complications, or by documenting healthcare access such as number of missed follow-up appointments. Only one study measured quality of life as an outcome and none documented social outcomes. Six of these eight studies found a statistically significant benefit to at least one patient health outcome for those in the transition group. The longest follow-up period in the eight studies was three years.

One study not included in the aforementioned systematic review found that for adolescents with diabetes, patients who either transferred directly from paediatric to adult healthcare or who transferred from a paediatric clinic to a young adult clinic at another hospital had lower attendance rates in their receiving setting and higher rates of returning to a general practitioner (GP) than those who had a more intensive transition clinic at the same hospital/diabetes center.[63] This study also showed that those who met their adult

physician before the final transfer were more satisfied with their transfer/transition experience.

Another study compared nine young adult kidney transplant recipients who were directly transferred to adult care to twelve who participated in a transition clinic.[64] The transition clinic included a joint consultation with a youth worker, paediatric nephrology physicians and nurses, and adult nephrology physicians and nurses. An individual consultation was followed by a consultation with the patient's family. Six of nine (66%) of the transfer group experienced transplant failure compared to zero in the transition group. However, for reasons not stated by the authors, the follow-up time was much longer for the transfer group than the transition group (40 months versus 26 months) which may have skewed the results in favour of the transition group.

A similar study of kidney transplant patients was conducted retrospectively using administrative data.[65] The proportions of death and transplant failure were reported for a transfer group and a transition group who received their transplant at the same hospital. The transition procedure included a verbal briefing of the patient's history to the adult healthcare team by the paediatric healthcare team and involvement of a clinic coordinator who scheduled the first few appointments in adult healthcare. Patients were also encouraged to return to the paediatric clinic for social visits. During a two-year post-transplant follow-up, there were three deaths (9%) and seven transplant failures (21%) in the transfer group compared to zero deaths and zero transplant failures in the transition group.

In a study of adolescents with inflammatory bowel disease, a statistically significantly smaller proportion of those who had participated in a transition clinic experienced hospital admission and surgery compared to those who had not participated.[66] Medication adherence was considered unsatisfactory for the cohort as a whole, but a larger proportion of the transition group was fully adherent. It was not clear if this difference was clinically significant.

The relationship between transfer, transition, and patient outcomes is not yet fully understood. The results of existing studies suggest that transition may be more beneficial for patients than transfer, although long-term outcomes and social outcomes have yet to be investigated.

2.3.3 Cost of Transition

Recent studies suggest that the cost to the healthcare system is similar for transition and transfer, if not lower for transition. [65, 67, 68] The higher short-term costs of transition may be balanced by the higher long-term costs of transfer due to the larger number of physician visits and medical procedures for those who transfer. The higher long-term cost per patient for those who transfer may be a result of poorer disease management.[65]

2.4 Transition Readiness

Transition readiness has been defined as "the capacity of the adolescent and those in his or her primary medical system of support (family and medical providers) to prepare for, begin, continue and finish the transition process." [69] This conceptual definition is often operationalized through measurement of disease-management skills, healthcare system utilization knowledge and skills, and health knowledge in the form of a checklist or self-administered questionnaire. Thus far, transition readiness has been measured using a single question, a self-administered questionnaire/checklist that may be validated or not, and by parent/caregiver- or AYA-report. Measuring transition readiness several times over the course of transition allows the intensity and content of the transition procedure and the timing of the final transfer to be tailored to the individual. Finding an appropriate transition readiness measurement tool and using it to improve the transition process is one of the top priorities called for by the Provincial Council for Maternal and Child Health (PCMCH) of Ontario.[19] There is a need for consistent use of one transition readiness measurement tool that is appropriate for adolescents with a chronic disease and has been tested for reliability and validity.

Transition readiness is often used as an outcome for evaluating the effectiveness of transition programs.[6, 70, 71] The underlying assumption is that greater pre-transfer readiness will contribute to transfer/transition success, but evidence justifying this

assumption is limited. The ideal way to assess this relationship would be to conduct a longitudinal study with measurement of pre- and post-transition readiness measurements, long-term patient outcomes, and healthcare access patterns. This type of study would require a great deal of time, money, and effort. We identified ten studies that investigated relationships of factors with transition readiness, including some short-term indicators of transfer/transition success.

A cross-sectional study involving 192 AYAs aged 16 to 25 years (response rate 95%) with inflammatory bowel disease aimed to discover demographic correlates of transition readiness as measured using the TRAQ (TRAQ properties found in Table 3.1).[8] Data were collected over a period of two years from AYAs who attended an outpatient appointment with a transition coordinator at a children's hospital, prior to the introduction of formal transition programming at the hospital. Differences in transition readiness between age groups were tested using chi-square and simple linear regression. When grouped into ages 16 to 17 years and 18 to 25 years, the proportion of AYAs who had acquired 90% of the TRAQ skills were not significantly different. The positive relationship between age and TRAQ score was significant in simple linear regression with no covariates. Females acquired statistically significantly more TRAQ skills than males as tested by ANCOVA controlling for age. Time since diagnosis was not significantly correlated with the number of TRAQ skills acquired. Pearson correlation between the number of TRAQ skills acquired and a physician's assessment of disease activity was not statistically significant.

Another cross-sectional study explored the relationship of self-perceived readiness to transfer with nineteen individual factors.[9] All adolescents receiving treatment for a somatic chronic condition from any paediatric department of one hospital were contacted, resulting in 1087/3648 participants aged 12 to 19 years (response rate 30%). Examples of chronic conditions included were immunity disorders, neurological conditions, and connective tissue diseases. Readiness to transfer was assessed by a single question "Do you think that you are ready to transfer to adult care?" with response options on a 4-point Likert scale. All data were collected through AYA self-report. Covariates were grouped into the following domains: disease-related factors, effect of condition on health and life,

self-management, and attitude towards transition. Two multivariable models were tested: one with all covariates included and a final model which included only those covariates with $p < 0.05$. The following covariates had a significant relationship at $p < 0.05$ with feeling ready to transfer in the final model: age; non-Dutch surname; prescribed medications, diet or exercise; school/work absenteeism due to illness; self-efficacy skills required for hospital visits; general independence score during consultations; attitude towards transfer; and transfer often discussed during consultations.

Post-transfer attendance, treatment adherence, and health outcomes were investigated for correlation with self-management skills in a study of 71 paediatric liver transplant recipients aged 11 to 20 years and 58 of their parents (response rate not reported).[10] Health outcomes included transplant rejection, frequency of hospital admission, and blood test results. The study did not distinguish between AYAs who had transferred and AYAs who were still in a paediatric care setting. Self-management skills were assessed by the Transition Readiness Survey (TRS); a combined score of the number of self-management skills acquired, level of disease-specific knowledge, and level of psychosocial adjustment through AYA self-administered questionnaire and provider-administered questions. Parent report of their knowledge of their child's disease-management regimen and their perception of child's self-management skills was assessed through self-administered questionnaire. The psychometric properties of the AYA and parent versions of the TRS are reported in Table 3.3. The Spearman rank correlations between AYA TRS total score and age, and AYA TRS total score and treatment adherence were significant and positive, while those between health outcomes and AYA/parent TRS total scores were not significant. The Pearson product correlation between total AYA TRS total score and rate of clinic attendance was not significant.

One study investigated potential correlations of factors with transition readiness in a sample of 48 AYAs who were enrolled in a transition clinic for adolescent kidney transplant recipients and their parents (response rate 89%).[11] A distinction was not made in the analysis between AYAs who had formally transferred and those who had not. Transition readiness was measured using AYA and parent versions of the Readiness for Transition Questionnaire (RTQ) through AYA and parent self-report. Psychometric

properties of the RTQ are presented in Table 3.2. The outcome for analyses was a composite of 2 questions from the RTQ that represent the AYA's perception of their readiness to assume responsibility for their health and to transfer. Correlations between the outcome and gender, ethnicity, and time since transplant were not significant. Hierarchical simple regression was also used. In the final model, barriers to medication adherence, medication knowledge, calling in medication refills, clinic appointment adherence, and teen-parent relationship quality had a significant relationship with the composite outcome at $p < 0.05$ while age was not significant.

One study found that a higher average transition readiness score was associated with higher odds of believing they were able to manage their own healthcare in a multivariable simple regression model, controlling for age and gender.[12] Data from two samples of AYAs aged 16 to 25 years, surveyed two years apart, were aggregated. AYAs were receiving care at one of the following specialty clinics: adolescent medicine, cystic fibrosis, diabetes, or myelodysplasia/spina bifida. The first sample was a convenience sampling of 36 AYAs and 21 of their parents who were already participating in a study of patient-centered information delivery (100% response rate). The second sample was 43 AYAs and 31 of their parents recruited in the clinics and by mail (35% response rate). Transition readiness was measured using the TRAQ (TRAQ properties found in Table 3.1). This study also found that age was significantly positively correlated with transition readiness and that females had a higher score than males on average.

Using a pre-/post- design, one study evaluated the effectiveness of implementing two case-based training sessions for clinicians about transition for improving the self-management skills acquisition of AYAs with inflammatory bowel disease.[13] The study compared the proportion of AYAs who were mostly or fully responsible for each of ten self-management behaviours before the training sessions were implemented (294/358 AYAs, response rate 82%) to after two years of sessions had been completed (142/156 AYAs, response rate 91%). Relationships between the proportion of participants responsible for each of the behaviours and time were tested using Fisher's exact test of independence and simple logistic regression. This study did not make a distinction between AYAs who had transferred and those still being cared for in the paediatric

setting. After the two cycles of training sessions, there was no significant increase in the proportion of AYAs who reported taking most or full responsibility for any of the ten behaviours measured in the study.

A six-year longitudinal study included 606 young adults aged 18 to 25 years without cognitive impairment who had been treated for a paediatric chronic somatic condition at the same hospital (response rate < 60%).[15] Self-efficacy and transfer experiences were measured using the On Your Own Feet Self-Efficacy Scale (OYOF-SES[72]) and the On Your Own Feet Transfer Experiences Scale (OYOF-TES[15]). The OYOF-SES was created and pilot-tested in Dutch. Transfer satisfaction was determined by the question "How satisfied are you with the overall process of your transfer to adult care on a scale ranging from 1 to 10?" Although not explicitly stated, it seems that not all participants had transferred to adult care and only those who had transferred were included in the analysis. Two stepwise linear regression models (n = 293) were performed with the following covariates: sex, self-efficacy, pre-transfer attitude towards transfer, perceived importance of discussing transition, HRQL, self-efficacy change, HRQL change, type of adult healthcare setting, independence score during consultation, independent behaviours during consultation, and patient-centeredness of current healthcare providers. In the final model, the following variables were significant at $p < 0.05$ with transfer experiences: gender, attitude towards transition, self-efficacy change, and HRQL change. In the final model, the following variables were significant at $p < 0.05$ with transfer satisfaction: gender, importance of discussing transition, type of adult healthcare setting, independent behaviours during consultation, and patient-centeredness of current healthcare providers.

One study recruited 114 AYAs aged 17 to 19 years with type I diabetes from an outpatient diabetes clinic at a hospital, a private hospital, and a diabetes care center (response rate not stated).[16] Data were collected from AYAs by mail and online questionnaires twice: once during the last six months of high school and once in the fall following high school graduation (time interval ranged from 91 to 311 days). It was not specified whether AYAs had transferred to adult care. Linear mixed effects regression was used to model the relationships of time, who the AYA was living with, diabetes-specific self-efficacy, and worry about hypoglycemia with the outcome of diabetes

management (total score on a 24-item scale of management behaviours). Self-efficacy and measures of disease management are both parts of the transition readiness concept. They found that disease management became statistically significantly better over time, disease management did not differ based on whether the AYA lived with parents or not, and that AYAs who had higher diabetes self-efficacy also had better disease management.

One cross-sectional study investigated if type of chronic condition was associated with transition readiness as measured by the TRAQ (scale properties found in Table 3.1) and self-perceived transition readiness as measured by the Readiness for Transition Questionnaire (RTQ teen version properties found in Table 3.2).[17] The sample included 163 AYAs aged 12 to 22 years who were patients of a chronic disease outpatient clinic or a teen health center at the same hospital (response rate not reported). Data were collected using self-administered questionnaires that were distributed at a clinic appointment and completed either in the clinic or elsewhere. Structural equation modeling was used to incorporate both measures of transition readiness as outcomes. The TRAQ scores of AYAs with autism spectrum disorder, spina bifida, type I diabetes, and turner syndrome were compared to those of AYAs who had not been diagnosed with a chronic condition. AYAs with autism spectrum disorder, spina bifida, and turner syndrome had on average significantly lower TRAQ scores than AYAs who had not been diagnosed with a chronic condition. The average TRAQ scores of AYAs with type I diabetes were not significantly different from those of AYAs who had not been diagnosed with a chronic condition. These relationships persisted after sensitivity analyses by path analysis with age, sex, racial minority, mother's education level, family composition, and health literacy.

The only study of transition readiness of adolescents with special healthcare needs (defined as having a chronic condition requiring greater use of healthcare resources than a typical person[73]) in Canada was a validation study of the Self- Management Skills Assessment Guide (scale properties found in Table 3.4).[18] The sample included 49 AYAs who were either patients at a neurology clinic or participating in the Well on Your Way Youth Transition Program at Alberta Children's Hospital, and their parents. Both groups were convenience samples with a response rate of 65%. Mean total Self- Management

Skills Assessment Guide scores were not reported for AYAs or parents. Pearson correlation of mean AYA and parent scores indicated a significant, positive, moderate correlation between parent and child transition readiness. Mean score was not significantly correlated with age and mean scores of age groups 11-13, 14-16, and 17-18 were not significantly different. This result is in contrast to the common finding in the literature that transition readiness is positively correlated with age.

Of the aforementioned factors, clinic attendance, treatment adherence, health outcomes, and disease management could be considered as indicators of a successful transition. Of these factors, adherence was the only factor significantly associated with transition readiness. The following factors were found to have a significant relationship with transition readiness in at least one of the aforementioned studies: age, sex, proxy for ethnicity, prescribed therapies/treatments, treatment adherence, barriers to treatment adherence, clinic appointment adherence, self-efficacy skills, discussion of transfer with healthcare providers, attitude or beliefs about transfer, measures of independence, school or work absenteeism, AYA-parent relationship quality, disease management outcomes, and type of chronic condition. It is evident that the results for most of the relationships that have been investigated are inconclusive. Additionally, the aforementioned studies have some common limitations: use of convenience sampling, outcomes not validated, very small sample size, analyzed cross-sectional data only, and/or recruited from a single-center. Our research will begin to shed light on the relationship between transition readiness and individual and familial factors and address some of the limitations discussed here.

Chapter 3 : Methods

3 Chapter Overview

This chapter describes the source of the data for this research, the Health-Related Quality of Life in Children with Epilepsy Study (HERQULES). A general description of the recruitment methodology used in HERQULES and the two phases of the study are reported. The data collection strategy for the 10-year follow-up of HERQULES is then recounted. A description of the measures used in this research is also given. The chapter concludes with an outline of the data analysis plan.

3.1 Study Design and Sample

The data used in this research are part of the Health-Related Quality of Life in Children with Epilepsy Study (HERQULES). HERQULES is a prospective, multi-center cohort study of children across Canada newly diagnosed with epilepsy and their parents/caregivers. Data were collected from families and physicians six times over approximately ten years following diagnosis. Participants were recruited using a two-stage sampling strategy which has been shown to achieve high physician response rates by engaging a group of participating physicians as recruiters of their peers.[74] Beginning in 2004, paediatric neurologists were recruited from across Canada. All practicing paediatric neurologists who were treating children with epilepsy in Canada were invited to participate in the study. Paediatric neurologists then approached parents/caregivers of eligible patients at consecutive appointments over approximately 18 months.

Inclusion criteria were: new diagnosis of epilepsy seen for the first time by the participating paediatric neurologist; epilepsy was diagnosed when the patient was aged 4 to 12 years; the parent/caregiver participating in the study was the primary caregiver; and the participating parent/caregiver had to have sufficient English language proficiency to complete the questionnaire. Exclusion criteria were: previous diagnosis of a progressive neurological disorder and previous diagnosis of other major comorbidity that would affect quality of life (ex. significant cognitive impairment).

In the first phase of HERQULES, parents/caregivers were asked to complete mailed questionnaires as soon as possible after their child's diagnosis (hereafter referred to as baseline), 6 months, 1 year, and 2 years later. For each child whose parent had consented, paediatric neurologists were also asked to complete a form providing clinical information at baseline and at each of the three follow-ups over the first two years. Approval was obtained from research ethics boards governing each of the participating paediatric neurologists' centers across Canada.

In the second phase of HERQULES, the long-term follow-up, families were followed-up again at approximately 8- and 10-years post-diagnosis. Adolescent self-report was introduced at the 8-year follow-up. Adolescents were eligible to provide report at the 8-year follow-up if their family was still enrolled in HERQULES at the 2-year follow-up, if they were aged 11 years or older, and if the adolescent's parent/caregiver had given consent for them to participate. Data were collected at the 8- and 10-year follow-ups from the adolescents and young adults (referred to as AYAs when discussing the second phase of the study) by mailed or online questionnaire. Data from their parents/caregivers and their physicians who were providing care for epilepsy were collected by mailed questionnaire. Given the pre-existing relationship with the families that was established in the first phase of HERQULES, approval received from the Western University Health Science Research Ethics Board covered all of the participants from across Canada. Ethics approval for changes made to the written materials, study protocol, and questionnaires for the 10-year follow-up (primarily to facilitate the research described in this thesis) was obtained from the Western University Health Science Research Ethics Board with an amendment (Appendix A).

3.2 Data Collection Strategy

The outcomes assessed in this thesis are from the 10-year follow-up. Analysis also includes data collected at baseline (shortly after diagnosis). AYAs were aged 12 to 22 years at the time of questionnaire mailing for the 10-year follow-up.

The Tailored Design Method was adapted for use in this study, which has been successful in achieving high response rates.[75] The Tailored Design Method is an evidence-based

method of survey design that has been demonstrated to maximize response rates and data quality in survey research studies. It was applied in the design of all written materials given to paediatric neurologists, parents/caregivers, and AYAs; the scheduling of contacts including mailing of questionnaires and reminders; telephone communication; and tokens of appreciation for participating.

Questionnaire packages were sent to all parents/caregivers who were still enrolled in the study at the 8-year data collection point (n = 215) and to all children whose parent/caregiver had, at the 8-year data collection point, given verbal consent for their child to participate (n = 176), with the exception of one family whose child was not able to complete the questionnaire on their own. Questionnaire packages/emails were sent in December, 2015.

Parents/caregivers were sent packages containing the letter of information, questionnaire, return envelope, and a $20 gift card as a token of appreciation for participation. AYAs who requested paper questionnaires were sent packages containing the letter of information, questionnaire, and return envelope. AYAs who requested online questionnaires were sent an email containing the letter of information and a link to the questionnaire. The online questionnaire was split into Part A and Part B such that participants had the option of completing Part B immediately or later. Gift cards were mailed to AYAs after their completed questionnaires were received. One of two versions of the questionnaire was distributed to AYAs based on age at the time of mailing/email: 11-17 years and 18+ years old.

One to two weeks after the initial package was sent, a reminder postcard was sent to parents and AYAs if their completed questionnaire had not been received. Approximately two weeks after reminder postcards were sent, reminder packages were sent to participants whose completed questionnaires had not yet been received. Those receiving mailed questionnaires were sent a reminder letter, a second copy of the questionnaire, and a return envelope. Those receiving online questionnaires were sent a reminder email with a link to the questionnaire. If a completed questionnaire had not been received after six weeks, the study coordinator attempted to contact the participant by phone to confirm

their interest in participating in the study. Attempts were made to contact participants who were not reachable by phone or whose packages/emails were returned to sender. All questionnaires were reviewed for quality control.

Physician forms (Appendix B) were sent for all AYAs on whom we had pre-existing consent to release clinical information. In instances where it was discovered that an AYA had changed physicians, an attempt was made to obtain a physician form from the new physician.

3.3 Measurement

The parent and AYA questionnaires at the 10-year follow-up were composed of several validated measures of AYA and family characteristics. There were also questions that provided a broader picture of the lives of the AYAs and their families, such as living situation, household income, education, and other sociodemographic information. To address the objectives of this thesis, we added an additional section on transfer/transition to the parent and AYA questionnaires. To address the first objective, we added a measure of transition readiness, the Transition-Q, to the AYA questionnaire. For the second objective, we selected a number of variables that were measured at baseline and the 10-year follow-up to include in multivariable regression. For the third objective, we added questions to the parent and AYA questionnaires that asked about their transfer/transition experience, current epilepsy status, and healthcare access. Most of the questions were developed specifically for this thesis research based on a literature review. Some questions were modeled after those found in the 2009-2010 National Survey of Children with Special Health Care Needs.[76]

3.3.1 Transition Readiness

A literature review was conducted to identify existing measures of transition readiness. The following criteria were used to evaluate published transition readiness measures: disease-specificity, psychometric evaluation, pilot testing, characteristics of the test sample (age, clinical diagnosis), applicability of item content to target population, language of administration, mode of administration (interviewer-administered or self-

administered), extent of adoption by researchers, stakeholder involvement in development, and practicality of completion time for research purposes.

The intention was to choose a measure that would be useful to both clinicians and researchers. Therefore, an ideal measure for use in this research would have the following characteristics: not disease-specific, satisfactory psychometric evaluation, pilot tested in a population that has a similar disease course to epilepsy, all items applicable to the HERQULES population, developed and tested in English, self-administered questions only, widely used by researchers, developed in consultation with clinicians and AYAs who were pre- and post-transfer, and perceived as short in length.

The following scales were considered for use in this research: Transition Readiness Survey[10], Readiness for Transition Questionnaire[11], Self-Management Skills Assessment Guide[18], TRAQ[77], Transition-Q[78], Am I ON-TRAQ[79], UNC TRxANSITION[80], Self-Care Independence Scale[81], Good2Go Readiness Checklist[82], Readiness Questionnaire[83], Client Questionnaire[60], Readiness assessment by McPherson et al.[84], and the Transition Readiness Questionnaire[85]. Tables 3.1 to 3.4 summarize the properties of the above scales.

The Transition-Q was selected as the measure best suited for use in HERQULES. The Transition-Q is not disease-specific, has sound psychometrics, was pilot-tested in AYA cancer survivors, all items were deemed applicable to the HERQULES population, it was developed and tested in English, it is a self-administered questionnaire, it was developed in consultation with clinicians and pre-/post-transfer AYAs, and can be completed in a few minutes.

3.3.2 Properties of the Transition-Q

Permission to use the Transition-Q in this research was obtained from the developers.[78] The development and validation of the scale was not part of this thesis, but are described here to highlight its properties and applicability to the HERQULES sample. The Transition-Q was initially developed within the context of childhood cancer survivors transitioning from paediatric care to adult long-term follow-up care. There are clearly

differences between the chronic conditions of childhood cancer and epilepsy, but similar to living with epilepsy, surviving childhood cancer entails living with risks of relapse and of developing associated conditions later in life.[86]

Development of the Transition-Q began with an investigation of psychological factors that affect transition success for survivors of childhood cancer. A list of themes was produced based on interviews of 38 survivors of childhood cancer including those who were pre-transition, successful post-transition, and unsuccessful post-transition AYAs.[87] Based on the emerging themes three constructs were identified, one of which was self-management skills. Potential scale items were developed and then reviewed by seventeen stakeholders including paediatric physicians, adult physicians, nurses, social workers, a psychologist, a neuropsychologist, and a childhood cancer survivor.[86]

Validity and reliability were first evaluated in a sample of childhood cancer survivors aged 15 to 26 years who were pre- or post-transfer using Rasch Measurement Theory analysis.[86] Item response thresholds, item fit statistics, item locations, and item stability were analysed to determine validity. Reliability was evaluated using the Person Separation Index and test-retest reliability was evaluated between one week and two months after first completion. Based on the results of validity and reliability testing, the response options were changed from a four-point ("strongly disagree" to "strongly agree") to a 3-point Likert scale.

The self-management skills scale that became the Transition-Q had a Cronbach's alpha of 0.81 and test-retest reliability of 0.90, indicating acceptable reliability. Differential item functioning was not detectable for age at diagnosis or gender. Item fit statistics (residuals between -2.5 and 2.5, item chi-squares were not significant) and item locations (not skewed, small gaps between items) were satisfactory.

Validity and reliability of the Transition-Q were further examined in a sample of youth aged 12 to 18 years with diverse chronic conditions who were recruited from McMaster Children's Hospital in Hamilton, Ontario.[78] Pilot testing was again conducted consisting of two rounds of cognitive interviews. A field test was also conducted on the 19-item scale. The Flesch-Kincaid test indicated a reading level below grade six for fourteen

items. One item had a reading level of grade 10.1, which was revised to lower it. Two rounds of cognitive interviews were conducted with 23 and 9 participants, respectively, aged 12 to 18 years. The cognitive interviews determined how easily the instructions and items were understood and the thought process behind answering the items. Participants also suggested revisions. As a result, three items were dropped, two items were added, and nine items were revised.

The resultant 18-item scale was field tested with 337 participants aged 12 to 18 years with a chronic disease, 10 of whom had a diagnosis of epilepsy. Rasch Measurement Theory analysis was again used, reporting item response thresholds, item fit statistics, item targeting, item stability, Person Separation Index, dependency, chi-square test of model fit, and test-retest reliability at least two weeks after first completion. As a consequence of the item response threshold and item fit results, response options were changed to “never”, “sometimes”, and “always” and four items were dropped, respectively. The final version of the Transition-Q had 14 items with response options “never” = 0, “sometimes” = 1, and “always” = 2 scoring. Item locations were not skewed and had a few small gaps, indicating that the items likely cover most of the topics that measure the construct of self-management. Properties of the Transition-Q are also summarized in Table 3.3. The final version of the Transition-Q used in this research is shown in Appendix C.

3.3.2.1 Transition-Q Scoring

Item responses (“never” = 0, “sometimes” = 1, and “always” = 2) are summed to create a raw score. Possible total raw scores range from 0 to 28. The raw score is then transformed to a Rasch-based person measure score. This is a nonlinear transformation based on the Rasch model that depends on the person item location distributions.[88] Transformation of raw scores was done using a table provided by the developers. The transformed score ranges from 0-100, the range is not continuous. A higher score indicates greater transition readiness; exhibiting more self-management skills with higher frequency.

3.3.3 Individual and Familial Factors

The individual and familial factors examined in this research were collected at the baseline assessment which was done as soon as possible after diagnosis (referred to as "at baseline") and the 10-year follow-up. Age at diagnosis, sex, epilepsy type at diagnosis, and severity of epilepsy at diagnosis were collected from the physician form at baseline. Status of epilepsy care, time since last seizure, last time AEDs were taken, health-related quality of life (HRQL), depressive symptoms, and anxiety were collected from AYAs at the 10-year follow-up. Physical comorbidities; developmental, behavioural, or psychological comorbidities; with whom the AYA lives; family demands; family adaptation; parental/caregiver household income; and parents'/caregivers' highest level of education were collected from parents/caregivers at the 10-year follow-up. Data were also collected from parents/caregivers at the 10-year follow-up about services their child was receiving or had received in the past: medication for behavioural or emotional problems, speech or occupational therapy, and extra help with schoolwork or placement in special class for children with learning difficulties. Table 3.5 shows the constructs that were collected using validated scales.

3.3.4 Development of Questions to Capture Transition Experiences

Review of the literature was conducted to identify essential topics for development of the questions that would capture the experience of transferring from paediatric to adult healthcare. This review produced a list of nine topic areas of interest: AYAs' knowledge regarding long-term risks associated with epilepsy,[60, 89] the nature of discussion with a healthcare professional about transfer or transition[19], participation in a formal transition program,[1] receipt of written information about transfer/transition and medical history summary,[38, 90, 91] meeting with the adult neurologist before the final transfer/care coordination,[50, 92] attendance at an appointment with the adult neurologist after referral and final transfer,[90, 93] involvement of a professional other than physician ns (ex. Nurse, social worker) in transfer/transition,[49, 90, 94] availability of resources or support for

families,[2, 95, 96] and positive or negative transfer/transition experiences of the families.[3, 40, 97]

While there were many interesting and important areas that were desirable to include in the transfer/transition section of the questionnaires, it was evident that several essential pieces of information needed to be gathered to make our results comparable to those in the literature. To better understand the AYAs' situations and provide a context for interpreting the transition readiness data, information needed to be collected on the following: AYAs' last visit to a physician for an epilepsy-related reason, if transfer had ever been discussed with them, status of epilepsy care (still with paediatric neurologist, transferred to adult neurologist, no longer seeing a neurologist, etc.), and last time AEDs were taken.

From the lists of identified topics above, nine multiple-choice questions and three open-ended questions were developed and included on the AYA questionnaire (Appendix D). A subset of these questions were also included on the parent questionnaire in addition to two questions from the 2009-2010 National Survey of Children with Special Health Care Needs (Appendix D). It was decided that additional topic areas of interest would be too difficult to address in a self-administered questionnaire. For example, some of the topics would require complicated skip patterns. Skip patterns are often difficult for respondents to follow and this would result in lower data quality if questions were accidentally skipped by a large proportion of respondents.

3.4 Data Analysis

Data from parent questionnaires and physician forms were entered into SPSS Version 24 and converted to SAS data sets. Data from AYA questionnaires were entered into Microsoft Excel 2016 and converted into SAS datasets. All data analyses were conducted using SAS 9.4 for Windows (SAS Institute Inc., Cary, NC, USA).

Univariate analyses were conducted to describe the sample in terms of sociodemographic characteristics, parent and familial factors, and clinical features. Means and standard

deviations were reported for continuous variables and frequencies and proportions for categorical variables.

Internal consistency of the Transition-Q was assessed using Cronbach's alpha. Transition-Q scores were summarized using the sample mean, standard deviation, quartiles, range, and sample distribution. Differences in mean Transition-Q score across categories of the following categorical and binary variables were examined using ANOVA or Student's t-test: status of epilepsy care; developmental, behavioural, or psychological comorbidities; and receipt of medication or therapy for behaviour or emotional problems. The characteristics of AYAs with a Transition-Q score ≤ 10 and ≥ 90 were highlighted.

HERQULES provided an opportunity to gain preliminary perspective on the factors associated with transition readiness. Many constructs were measured in the HERQULES questionnaires at baseline and the five follow-ups. A construct was investigated for a relationship with Transition-Q score if a similar construct had been previously investigated for a relationship with transition readiness in the transition literature, was highlighted as possibly influencing adolescent healthcare autonomy acquisition in the literature,[98, 99] or was of investigator interest. Using these criteria, it was deemed appropriate to include certain constructs measured at baseline and the 10-year follow-up.

The following individual factors were chosen for examination by linear regression for a relationship with Transition-Q score: sex; age at diagnosis; age at 10-year follow-up; epilepsy severity at baseline; epilepsy type at diagnosis; time since last seizure; last time AEDs were taken; status of epilepsy care; HRQL; depressive symptoms; anxiety; physical comorbidities; developmental, behavioural, or psychological comorbidities; with whom the AYA lives; and past or current receipt of extra help with schoolwork or placement in special class for children with learning difficulties, speech or occupational therapy, and medication or therapy for behaviour or emotional problems. The following familial factors were chosen for examination by linear regression for a relationship with Transition-Q score: parental household income; parent highest level of education (the highest of primary caregiver and their live-in partner/spouse, if applicable); family

demands; and family adaptation. Data were examined by cross-tabulation of variables and determining correlation structure of independent variables.

We began by first examining the bivariable relationships between each covariate and Transition-Q score using simple linear regression models. Covariates with $p < 0.3$ were then selected for inclusion in the multivariable regression analysis. A liberal p-value of 0.3 was chosen to reduce the possibility of missing potentially important variables.[100] The conventional significance level of $p < 0.05$ was used in the multivariable analysis. Assumptions of normality and linearity in the final model were assessed using a Q-Q plot and Transition-Q score residuals plot.

Transfer/transition experiences were described by summarizing self- and parent-reported AYA contact with healthcare (Appendix D) and responses to the three open-ended transition experiences questions (Appendix D). Responses to the open-ended questions were coded by distinct theme. The number of times each theme was mentioned was recorded.

To determine if attrition bias was a concern, characteristics were compared between families who completed a questionnaire at baseline and 10-year follow-up and families who completed a questionnaire at baseline but did not complete a questionnaire at the 10-year follow-up. A family was classified as lost to follow-up at the 10-year follow-up if they completed the baseline questionnaire but not the 10-year follow-up questionnaire. The following individual and familial variables were available both on the baseline and 10-year follow-up questionnaires and included in the attrition analysis: child sex, severity of epilepsy, epilepsy type at diagnosis, family demands, family adaptation, parental household income, relation of primary caregiver to child (biological parent vs. other relation, biological mother vs. other relation), and primary caregiver's highest level of education (the highest of primary caregiver and their live-in partner/spouse, if applicable).

Table 3.1: Properties of transition readiness measures found in literature search

Properties	TRAQ[77]	Am I ON TRAC[79]	Good2Go Readiness Checklist[82]	Client Questionnaire[60]
Disease-specific?	No	No	No	Yes – epilepsy
Psychometric evaluation	Principal component factor analysis. Total Cronbach's alpha = 0.93 after removal of 4 items.	Principal component analysis. Knowledge scale Cronbach's alpha = 0.84	None	None
Pilot testing	Ethnographic interviews with 15 youth aged 16-23	See above.	None	Limited – frequencies of responses reported
Test sample	n = 192, 100% response rate, 3% missing data Not clear if any youths with epilepsy in the sample.	n = 200, aged 12-19, 93.5% response rate, 10% of respondents recruited from neurology	None	n = 97, 22 cases of proxy report, aged 16-18
Applicability to AYAs with epilepsy	Some items not applicable to Canadian population, HERQULES age range.	All items applicable	All items applicable	All items applicable
Available/Tested in English?	Yes	Yes	Yes	Yes
Mode of administration	Self-administered	Self-administered	Self-administered	Self-administered or proxy by caregiver
Use by other researchers	Yes	Used in transition programs in Alberta, Canada and the United Kingdom	The Hospital for Sick Kids (Toronto, ON) Good 2 Go program only.	Unknown

Properties	TRAQ[77]	Am I ON TRAC[79]	Good2Go Readiness Checklist[82]	Client Questionnaire[60]
Stakeholder involvement in development	Youth and transition healthcare providers rating of content and face validity	Not stated.	Not stated.	Paediatric and adult neurologists
Length	29 items	25 items	26 items	12 items
Scoring	Two domains, non-additive scoring	Two domains with independent scores: knowledge (additive scoring) and behaviour (raw score and cut-off score)	Not scored	Not scored

Table 3.2: Properties of transition readiness measures found in literature search

Properties	Readiness Questionnaire[83]	Readiness for Transition Questionnaire[11]	Transition Readiness Questionnaire[85]	Readiness assessment by McPherson et al.[84]
Disease-specific?	Yes – cystic fibrosis	Yes – kidney transplant	Items themselves were not, but scoring was – HIV	Yes – sickle cell disease
Psychometric evaluation	n = 36 AYAs. Compared scores to a readiness rating of 1-4 given by healthcare providers known to the AYAs.	Internal consistency: Cronbach's alpha = 0.79.	None	None
Pilot testing	See above.		None	None
Test sample	See above.	n = 54 AYAs enrolled in a transition clinic, 89% response rate	51 parent-child pairs of clinical research participants. Child participants aged 9-25.	None
Applicability to AYAs with epilepsy	Some disease-specific items not applicable	All items applicable except for two.	Items mostly applicable	Difficult to judge based on description given. Likely not applicable given disease-specificity.
Available/Tested in English?	Yes	Yes	Yes	Yes
Mode of administration	Self-administered questionnaire	Self-administered questionnaire	Interviewer-administered, parent and AYA report.	Self-administered questionnaire, AYA report.
Use by other researchers	Unknown	Unknown	Yes	Unknown

Properties	Readiness Questionnaire[83]	Readiness for Transition Questionnaire[11]	Transition Readiness Questionnaire[85]	Readiness assessment by McPherson et al.[84]
Stakeholder involvement in development	Semi-structured interviews with 35 paediatric and adult healthcare professionals and patients (adult care).	Not stated.	Not clear, "designed by investigators."	Not stated.
Length	24 items	22 items	Six main categories with total of 5 sub-items.	9 readiness assessment items split into 2 categories (readiness and transition knowledge), 3 items on opinions on improving transition.
Scoring	Two subscales scores and a total score, additive scoring.	Two sections with the same items: one for their own responsibilities and one for their parents' responsibilities. Only 2 items are used in scoring	Overall score Categories not equally weighted.	Separates score for both readiness assessment categories. The opinions on improving transition section involved ranking a list of options.

Table 3.3: Properties of transition readiness measures found in literature search

Properties	Transition Readiness Survey (TRS)[10]	Transition-Q[78]	UNC TRxANSITION[80]	Self-Care Independence Scale[81]
Disease-specific?	Yes – liver transplant	No	No	Yes – cystic fibrosis
Psychometric evaluation	AYA & Parent: Non-parametric principal component analysis identified 4 domains accounting for 32.5% response variance. <u>AYA version</u> Domain Cronbach's alpha range: 0.68-0.81 Overall Cronbach's alpha: 0.85 <u>Parent version</u> Domain Cronbach's alpha range: 0.18-0.75 Overall Cronbach's alpha: 0.75 AYA-Parent domain score correlations range -0.26 to 0.58 Higher TRS scores correlated with higher medication adherence.	n = 337 AYAs aged 12-18 with asthma, blood disorder, cancer, cerebral palsy, cystic fibrosis, diabetes, eating disorder, epilepsy, cardiac disorder, inflammatory bowel disease, or mental health disorder. Flesch-Kincaid grade level was 2 to 7. Internal consistency: Cronbach's alpha = 0.85. Test-retest reliability: intra-class correlation coefficient = 0.92 at 21 days.	Inter-rater reliability: weight kappa = 0.71 (0.64, 0.77). Internal consistency: correlations of all items with total score >0.42, range of correlation between domain scores and overall score = 0.34-0.74. Score relationship with age, linear regression β=1.08.	Cronbach's alpha = 0.93 Test-retest reliability (n = 35) = 0.81

Properties	Transition Readiness Survey (TRS)[10]	Transition-Q[78]	UNC TRxANSITION[80]	Self-Care Independence Scale[81]
Pilot testing	None	Two rounds: 1. Field test of full scale 2. Cognitive interviews	Two rounds: 1. 92 adolescents with IBD and kidney conditions 2. 36 adolescents with IBD and kidney conditions	None
Test sample	58 parent-AYA pairs. AYAs were 11+ yrs old, greater than 6 months post-transplant.	1. n = 250, aged 15-26, cancer survivors, 4% missing data 2. n = 7, aged 16-22, cancer survivors	128 adolescents with IBD, kidney conditions, sickle cell, SLE, and diabetes.	n = 76 of 105 children and adolescents with cystic fibrosis
Applicability to AYAs with epilepsy	Not all items applicable	All items applicable	All items applicable	Items not applicable.
Available/Tested in English?	Yes	Yes	Yes	Yes
Mode of administration	First section: parent and/or AYA report, self-administered. Second section: healthcare provider-administered.	Self-administered questionnaire	Semi-structured interview, training required for interviewer.	Parent report, self-administered questionnaire.
Use by other researchers	Unknown	Yes	Yes	Unknown

Properties	Transition Readiness Survey (TRS)[10]	Transition-Q[78]	UNC TRxANSITION[80]	Self-Care Independence Scale[81]
Stakeholder involvement in development	Paediatric specialist, psychologist, transplant surgeon, social worker, dietician, and transplant nurse coordinator were involved	Interviews with 38 pre- and post-transfer Canadian cancer survivors. Feedback from 17 healthcare providers and parents of childhood cancer survivors.	Interviews with healthcare transition experts, transition coordinators, patients, adolescent specialists, other healthcare professionals.	Clinicians and researchers developed items, evaluated for relevance by 10 healthcare professionals.
Length	38 self-administered items 4 healthcare provider-administered items	14 items	33 items	44 items
Scoring	Different for each item, max score 126, higher score = greater skills acquisition. Also domain-specific scores.	Single domain, additive scoring with transformation.	Requires verification against medical record. 10 domains, possible scores in each domain are 0-1. Total score range 0-10.	Additive, overall score

Table 3.4: Properties of transition readiness measures found in literature search

Self-Management Skills Assessment Guide[18]			
Properties		**Properties**	
Disease-specific?	No	Available/Tested in English?	Yes
Psychometric evaluation	Agreement of parent and AYA mean scores: 0.54. Internal consistency: Cronbach's alpha for AYA = 0.89, for parent = 0.93. AYA mean scores were higher than parent scores. Correlation of Scales of Independent Behaviour scores (adaptive functioning, parent report) with parent and AYA readiness scores: parent = 0.74, AYA = 0.44. Flesch-Kincade reading level for AYA version = 4.9, for parent version = 5.9.	Test sample	Convenience sample of 32 AYA neurology patients (21 had epilepsy) and 17 AYAs participating in a transition program and their parents. AYA had to be diagnosed with special healthcare needs.
Pilot testing	None	Mode of administration	AYA and parent report
Use by other researchers	Designed for use in transition programs at Alberta Children's Hospital	Stakeholder involvement in development	Not clear, items from transition readiness checklists used in Canadian tertiary-care hospitals.
Applicability to AYAs with epilepsy	All items applicable. Some seem to have low face validity.	Length	21 items
		Scoring	Likert scale response options 1-5. Additive scoring?

Table 3.5: Properties of validated scales used in HERQULES

Factor	Measure	Properties
Epilepsy severity at baseline	Global Assessment of the Severity of Epilepsy scale (GASE)[101]	Range of 1 to 7 from "Not at all severe" to "Extremely severe."
Health-Related Quality of Life (HRQL)	Quality of Life in Epilepsy Inventory: QOLIE-AD-48[102] QOLIE-31-P[103]	Measures the health-related quality of life for individuals with epilepsy. QOLIE-AD-48 for AYAs 11-17 years old, Patient Weighted Quality Of Life In Epilepsy (QOLIE-31-P) for AYAs 18+ years old. Range is 1 to 100, higher score indicates better HRQL.
Depressive symptoms (CESD ≥ 16)	Center for Epidemiolgic Studies Depression scale (CES-D)[104]	Measures symptoms of depression. Higher score indicates greater number and frequency of depression symptoms. Score of 16 or higher indicates mild to significant symptomatology.
Anxiety	STAI-Y6[105]	Measures current level of anxiety. Range is 20 to 80, higher score indicates higher anxiety.
Family demands	Family Life Events and changes scale FILE[106]	Measures family stress by counting events and changes. Range is 0 to 71, higher score indicates more stress on the family.
Family adaptation	Family APGAR[107]	Measure of family functioning Range is 0 to 20, higher score indicates greater satisfaction with family functioning.

Chapter 4 : Results

4 Chapter Overview

This chapter presents the findings of this research. The sample characteristics of AYAs and their families are described followed by Transition-Q score distributions and a summary of the AYAs' status of epilepsy care. Then the results of the bivariable and multivariable linear regression analyses are reported followed by a summary of the healthcare access patterns of AYAs and the transfer/transition experiences of the AYAs and their parents/caregivers. The chapter concludes with the findings of the attrition analysis comparing families who completed the 10-year follow-up to families who were lost to follow-up.

4.1 Sample Characteristics

The results reported here represent the 131 AYAs and 121 of their parents/caregivers who returned completed questionnaires during the 10-year follow-up of HERQULES. A detailed participant flow chart of parents and AYAs are depicted in Figure 4-3 and Figure 4-4, respectively. Of the 455 families who were eligible to participate at baseline, 373 (82%) returned completed baseline questionnaires. When youth self-report was introduced at the 8-year follow-up, there were 220 youth eligible to provide self-report. Twelve youth were ineligible because they were unable to complete the questionnaire on their own due to significant cognitive impairment. Of those 220, 154 (70%) returned completed questionnaires. At the 10-year follow-up, the sixth and final data collection point, questionnaires were sent to 176 AYAs and 215 parents/caregivers. The 10-year follow-up return rates were 74% for AYAs (131/176) and 81% for parents/caregivers (173/215). The overall response rate at the 10-year follow-up for AYAs was 60% (131/220). Of the 131 AYAs who returned completed questionnaires at the 10-year follow-up, 121 had a parent/caregiver return a completed questionnaire. AYA individual and familial characteristics are summarized in Table 4.1 and Table 4.2, respectively. AYAs ranged in age from 12 to 23 years with a mean of 18 years and standard deviation of 2.6 (Only one individual was age 12 years. Initial enrollment in HERQULES occurred over a period of three years, thus follow-up times are approximate. The final follow-up

for this youth occurred eight years after diagnosis). The sex ratio was almost equal with 52% being female. Just over 80% of the AYAs were attending school. The majority of AYAs had a job; 37% had a part-time job and 12% had a full-time job. Of those aged ≥18 years, 58% of AYAs were in college or university, and 91% either had a job or were in school. At baseline, the average severity of epilepsy as reported by paediatric neurologists was between "Somewhat severe" and "A little severe", 5.6 on a scale of 1 to 7. The range of severity of epilepsy at baseline was 3 "Quite severe" to 7 "Not at all severe." At the 10-year follow-up, half of AYAs had a past or current diagnosis of developmental, behavioural, or psychological comorbidities as reported by the parent/caregiver (a composite of the diagnoses listed in the "Other diagnoses" section of Table 4.1).

The vast majority of participating parents/caregivers were the AYA's biological mother at 109 (90%) with the remainder being 5 biological fathers, 2 step-mothers, 4 adoptive mothers, and 1 female guardian. Most parents/caregivers were living with a partner or spouse (86%) and 80% were married. Parents/caregivers ranged in age from 35 to 69 years. Regarding parent/caregiver highest level of education, 73% had completed college or university and 12% of parents/caregivers had completed high school. Almost two thirds of parents/caregivers were working full time and half of parents/caregivers had a yearly household income of $100,000 or more.

The majority of AYAs indicated that they no longer receive care for epilepsy or seizures from a physician (68%). Table 4.3 presents the status of epilepsy care for all AYAs and age subgroups. Of those who were aged ≥18 years, 14 (22%) had transferred to an adult neurologist and still receive epilepsy care from them and 4 (6%) transferred to an adult neurologist but no longer receive epilepsy care from them. There were 6 (5%) AYAs who were receiving epilepsy care from a GP, 5 of whom were aged ≥18 years. All 11 (9%) AYAs who were receiving epilepsy care from a paediatric specialist were aged ≤ 17 years, as expected based on healthcare system stipulations for paediatric services. Of the 35 AYAs who indicated they were taking AEDs at the 10-year follow-up, 16 (46%) indicated that they had transferred and still receive epilepsy care from their adult specialist, 1 AYA indicated that they transferred but no longer receive epilepsy care from

their adult specialist, and surprisingly 3 (9%) indicated that they no longer receive care for epilepsy from a physician.

4.2 Transition-Q Scores

Cronbach's alpha for the Transition-Q was 0.88, indicating high internal consistency. The distribution of Transition-Q scores was slightly left-skewed, with scores clustered towards higher transition readiness. However, the overall average Transition-Q score of 64 was mediocre given its theoretical range of 0 to 100. The mean Transition-Q score of AYAs aged ≤ 17 years was 57, lower than the mean score of 72 for those aged 18 years or older. The distributions of Transition-Q scores for AYAs aged 11-17 years and aged 18+ years are shown in Figures 4-1 and 4-2, respectively. In the context of AYAs' status of epilepsy care, mean Transition-Q score was lowest for those who receive care from a paediatric specialist (53), followed by those who had transferred to adult healthcare but no longer see their adult specialist (57). Those who no longer receive epilepsy care and those who had transferred to adult healthcare and still receive epilepsy care from their adult specialist had the highest average Transition-Q scores (67 and 66, respectively). None of these differences in mean Transition-Q scores by category of epilepsy care status were significantly different as tested by ANOVA. Mean Transition-Q score was slightly higher for females than for males (68 vs. 62) but not significantly different ($p = 0.106$). On average, AYAs who had been diagnosed with a developmental, behavioural, or psychological comorbidity had lower Transition-Q scores than those who had not (70 versus 60, one-sided $p = 0.003$). AYAs who had ever received medication or therapy for behavioural or emotional problems had slightly lower Transition-Q scores on average than those who had not, but this difference was not significant (62 versus 67, one-sided $p = 0.1$).

4.3 Bivariate Analyses

The bivariate analyses for Transition-Q score with the independent variables are presented in Table 4.4 for individual characteristics and Table 4.5 for familial characteristics. Seven individual factors had a significant relationship with Transition-Q score at $p < 0.05$: 1. age at diagnosis ($p < 0.001$); 2. epilepsy type at diagnosis ($p = 0.01$);

3. developmental, behavioural, or psychological comorbidities (p=0.007); 4. age at ten year follow-up (p <0.001); 5. HRQL (p = 0.008); 6. extra help with schoolwork or placement in special class for children with learning difficulties (past or current) (p = 0.003); and 7. speech or occupational therapy (past or current) (p = 0.0005). None of the familial factors assessed had a significant relationship with Transition-Q score at $p < 0.05$ (parent/caregiver household income, parent/caregiver highest level of education attained, family demands, or family adaptation).

4.4 Multivariable Analysis

A variable was selected for inclusion in the multivariable regression with Transition-Q score if the bivariate analysis resulted in a p-value no larger than 0.3. The individual factors that met this criterion were: age at diagnosis; epilepsy syndrome type at diagnosis; time since last seizure; last time AEDs were taken; developmental, behavioural, or psychological comorbidities; status of epilepsy care; age at ten year follow-up; sex; HRQL; with whom the AYA lives; extra help with schoolwork or placement in special class for children with learning difficulties (past or current); speech or occupational therapy (past or current); and medication or therapy for behaviour or emotional problems (past or current). The only familial factor that met this criterion was family adaptation. Moderate, significant correlations were observed between age at diagnosis and age at 10-year follow-up (Pearson correlation 0.94, $p < 0.001$), and time since last seizure and last time AEDs were taken (Spearman correlation 0.57, $p < 0.001$). It was decided that age at diagnosis and last time AEDs were taken should not be included in the multivariable analysis to avoid effects of colinearity.

The multivariable analysis with Transition-Q score as the dependent variable is presented in Table 4.6. The only variable significant at p-value ≤ 0.05 was age at 10-year follow-up which had a positive relationship with Transition-Q score when controlling for the other independent variables. The overall model was significant with $p = 0.001$. With a r-square of 0.50, 50% of the variance in Transition-Q score was explained by this multivariable model. The Q-Q plot and Transition-Q score residuals plot confirmed assumptions of normality and linearity, respectively.

4.5 Transfer/Transition Experience

Parent and AYA responses to the contact with healthcare questions and the three open-ended transfer/transition experience questions asking parents/caregivers and AYAs what they liked, did not like, and suggestions for improvement about their transfer/transition experience (Appendix D) are summarized here.

4.5.1 AYA Contact with Healthcare

There were two sets of questions asking for descriptions of AYA contact with healthcare providers: discussion about transfer/transition and last visit to a physician for an epilepsy- or seizure-related reason (Appendix D). The same proportion of AYAs and parents/caregivers indicated that a physician or other healthcare provider had spoken to them about transfer/transition (19% and 20%, respectively). However, only 9 parent-AYA dyads both answered "yes." Of the AYAs who reported that a physician had spoken to them about transfer, there were 4 (21%) under age 18 (ages 14 to 17). The majority of AYAs and parents/caregivers who reported that their physician had not talked to them about transfer/transition thought that a discussion about transfer/transition would not be helpful to them (71% and 81%, respectively). The latter question had a high rate of missing data at 36%. Almost all parents/caregivers (95%) and 100% of AYAs whose physician had talked to them about transfer thought that it had been either somewhat or very helpful.

One third of parents reported that their child's healthcare providers had discussed their child's healthcare needs in adulthood as they were growing up. Of the 81 (69%) parents who reported that their child's healthcare providers had not discussed their child's healthcare needs in adulthood, 24 (36%) indicated that such a discussion would have been helpful to them. Parents were also asked how frequently their child's healthcare providers encouraged their child to take responsibility for his/her healthcare needs. One third of parents reported "never", 19% reported "sometimes", 21% reported "usually", and 24% reported "always."

One third of AYAs reported going to a physician in the last two years for reasons related to epilepsy or seizures. Of these, 29 (78%) also reported that they were currently taking

AEDs. Of the subgroup of AYAs who were aged ≥18 years, 21 (31%) had seen a physician in the last two years for reasons related to epilepsy or seizures. Of the AYAs who reported they went to a physician for epilepsy or seizures in the last two years, none had last visited an emergency room or urgent care clinic for epilepsy or seizures. One AYA had seen a paediatric neurologist after reaching the age of 18 years. This is noteworthy because according to current standard practice in Canada, patients aged ≥18 years who require continued follow-up should have transferred to adult healthcare. He/she was aged 20 years, last visited a paediatric neurologist less than six months ago, and his/her last seizure was less than six months ago.

4.5.2 Parent/Caregiver Response Themes

There were 17 parents/caregivers who reported either that their child had transferred/transitioned to an adult neurologist who still provides epilepsy care for them or that they transferred/transitioned but no longer receive care from their adult neurologist. They were therefore eligible to give a response to the three open-ended questions about their child's transfer/transition experience. Of these, 15 responded to at least one of the three questions. Parent/caregiver responses were highly polarized; responses were either very positive or very negative with few responses falling between these extremes. Parent/caregiver responses tended to be longer than AYA responses and sometimes had more than one theme per response.

Parents/caregivers liked that their child's adult neurologist did not make them feel rushed, that their child was comfortable with their new neurologist, and that their child's adult neurologist only treated patients with epilepsy. Three parents/caregivers complimented the adult neurologist's attitude, for example: "she was always understanding and helpful." One parent/caregiver liked that their child's paediatric neurologist set up the first adult neurologist appointment for them.

Parent/caregiver responses to what they did not like about their child's transfer experience were more homogenous than their responses to what they liked. Three common themes emerged: losing control of their child's health, issues with the change in location of healthcare providers, and difficulty communicating with the adult team.

Parents/caregivers did not like being left out of their child's healthcare. Two parents/caregivers expressed worry that their child was not prepared to handle the responsibility on his/her own. Parents/caregivers expressed difficulty contacting the adult neurologist, a delay in scheduling the first appointment with the adult neurologist, and two parents/caregivers reported that the transfer/transition had not been discussed at all. One parent commented that their child's adult neurologist had not discussed how having epilepsy could affect their child in adulthood. Two parents/caregivers commented that the adult neurologist was located much farther from their home than the paediatric neurologist and one commented that parking expenses became an issue because their child's adult physicians were now spread over multiple locations.

The most common response to what would have made their child's transition experience better was that nothing would have made it better. The other themes that were mentioned included a joint meeting before the transfer, the ability to choose the adult neurologist, adult neurologist's review of their child's medical history, and the option of having the same health team in the adult system.

4.5.3 AYA responses

There were 23 AYAs who reported that they transferred to an adult neurologist and still receive epilepsy care from him/her or that they transferred but no longer receive care from their adult neurologist and were therefore eligible to give a response to the three open-ended questions. Of these, 21 responded to at least one of the three questions.

The theme most commonly mentioned by AYAs in response to what they liked about their transition was that they felt their adult neurologist and/or adult team treated them well (5 / 21 responses). These five responses referred to the manner in which the adult neurologist or adult team acted towards them. For example, "The doctor was very friendly and upfront and seemed to care about my well-being" and "Everyone treated me with respect and wanted the same goal." Four AYAs said that they liked being treated like an adult and that they were gaining more independence.

The theme most commonly mentioned by AYAs in response to both what they did not like about their transition experience and what could have made their transition experiences better was “nothing” (7/15 responses and 6/17 responses, respectively). AYAs disliked losing their relationship with the paediatric team, having to wait a long time for their first adult neurologist appointment, losing extra services, and had difficulty contacting their adult neurologist. One stated that transfer was a “tedious and time-consuming task.” Another said he/she transferred only because their parents wanted them to. One AYA said that he/she changed neurologists several times “because some acted like they didn’t care.”

To improve transfer/transition experiences, AYAs suggested a joint meeting with their paediatric and adult neurologists, shortening the time between the last paediatric appointment and the first appointment with the adult neurologist, and being able to keep the same health services.

4.6 Attrition Analysis

To shed light on possible attrition bias, an analysis was conducted comparing families who were lost to follow-up to families who completed the 10-year follow-up. Of the 373 parents/caregivers who completed the baseline questionnaire, 200 (54%) did not complete the 10-year questionnaire and were lost to follow-up. Table 4.7 shows the results of the attrition analysis. Neither age at diagnosis nor epilepsy type at diagnosis were significantly different. Child’s sex was also not significantly different. The familial factors were all significantly different. On average, family adaptation score was higher and family demands score was lower for families who completed the 10-year follow-up. Families who were lost to follow-up generally had a lower household income and lower level of education.

Table 4.1: AYA individual characteristics †

n = 131 AYAs	
Sex (male)*: Frequency (%)*	63 (48.1)
Age at diagnosis: *mean (SD), median, [range]*	7.5 (2.4), 7.0, [3, 12]
Age at 10-year follow-up: *mean (SD), median, [range]*	17.8 (2.6), 18.0, [12, 23]
Epilepsy severity at baseline: *mean (SD), median, [range]*	5.6 (1.1), 6.0, [3, 7]
Epilepsy syndrome type at baseline*: Frequency (%)*	
Primary generalized	18 (14)
Absence	36 (28)
Simple/complex partial	33 (25)
Benign epilepsy of childhood with rolandic spikes (BECRS)	16 (12)
Secondarily generalized	14 (11)
BECRS + secondarily generalized	12 (9)
Undetermined	2 (2)
Time since last seizure*: Frequency (%)*	
< 6 months	13 (10)
6 months – 1 year	4 (3)
1 – 2 years	7 (5)
2 – 5 years	18 (14)
5 – 10 years	57 (44)
10+ years	20 (15)
Don't remember	11 (9)
Last time AEDs were taken*: Frequency (%)*	
Currently	35 (28)
< 6 months ago	1 (1)
1 – 2 years ago	2 (2)
2+ years ago	61 (49)
Never taken AEDs	11 (9)
Don't remember	14 (11)
Status of epilepsy care (AYA report)*: Frequency (%)*	
Transferred and still receives epilepsy care from adult neurologist	19 (15)
Transferred but no longer receives care	5 (3)
Receives epilepsy care from paediatric specialist	11 (9)
Receives epilepsy care from GP	5 (4)
Does not receive epilepsy care	84 (68)
Transition-Q score: *mean (SD), median, [range]*	65.0 (19.1), 65.0, [0, 100]

n = 131 AYAs	
Health-related quality of life: *mean (SD), median, [range]*	77.8 (16.0), 82.0, [25, 97]
CES-D ≥ 16*: Frequency (%)*	50 (39)
Anxiety: *mean (SD), median, [range]*	37.1 (13.5), 35.0, [20, 70]
Physical comorbidities*: Frequency (%)*	
No	98 (82)
Yes, diagnosed before epilepsy	5 (4)
Yes, diagnosed after epilepsy	15 (13)
Yes, some diagnosed before and some after epilepsy	1 (1)
With whom the AYA lives*: Frequency (%)*	
Parent	106 (90)
Other relative	4 (3)
Non-relative	8 (7)
Other Diagnoses	
Developmental delay*: Frequency (%)*	17 (14)
Learning disability*: Frequency (%)*	40 (33)
Attention deficit disorder or attention deficit hyperactivity disorder*: Frequency (%)*	24 (20)
Autism, pervasive developmental disorder, or Asperger's syndrome*: Frequency (%)*	3 (3)
Oppositional defiant disorder*: Frequency (%)*	7 (6)
Conduct disorder*: Frequency (%)*	2 (2)
Depression*: Frequency (%)*	17 (14)
Anxiety*: Frequency (%)*	27 (23)
Services Received, Past or Current	
Extra help with schoolwork or placement in special class for children with learning difficulties*: Frequency (%)*	73 (66)
Speech or occupational therapy*: Frequency (%)*	27 (25)
Medication or therapy for behaviour or emotional problems*: Frequency (%)*	27 (26)

† All variables have less than 10% missing data, except for variables assessing services received on which there were 15% to 19% missing data. Data collected at 10-year follow-up unless otherwise specified.

Figure 4-1: Transition-Q scores of AYAs aged 11-17 years

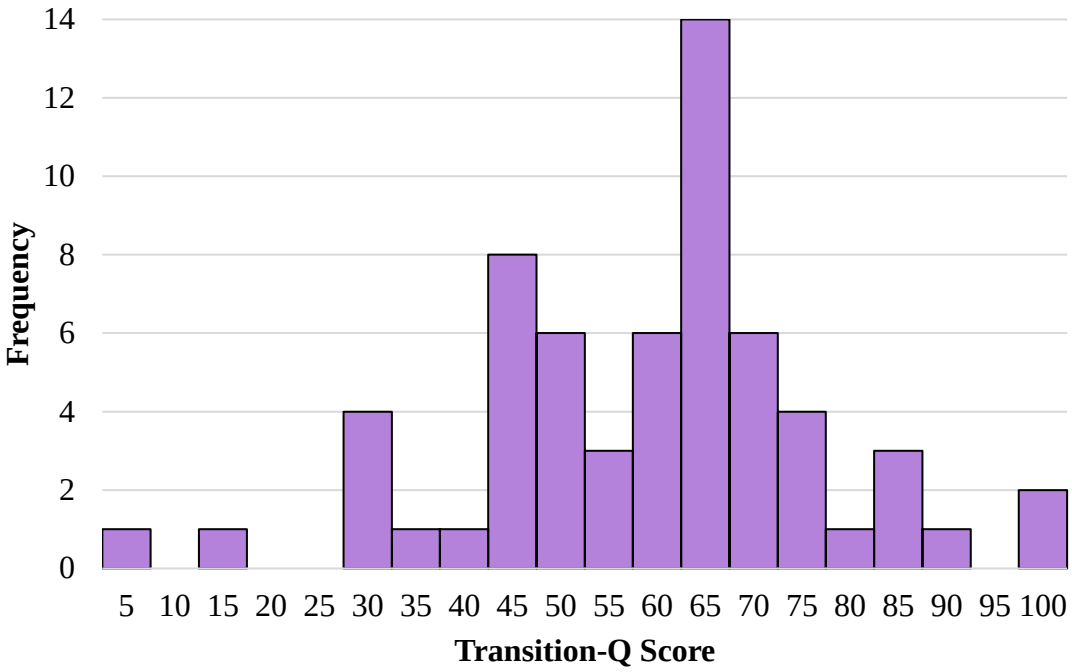


Figure 4-2: Transition-Q scores of AYAs aged 18+ years

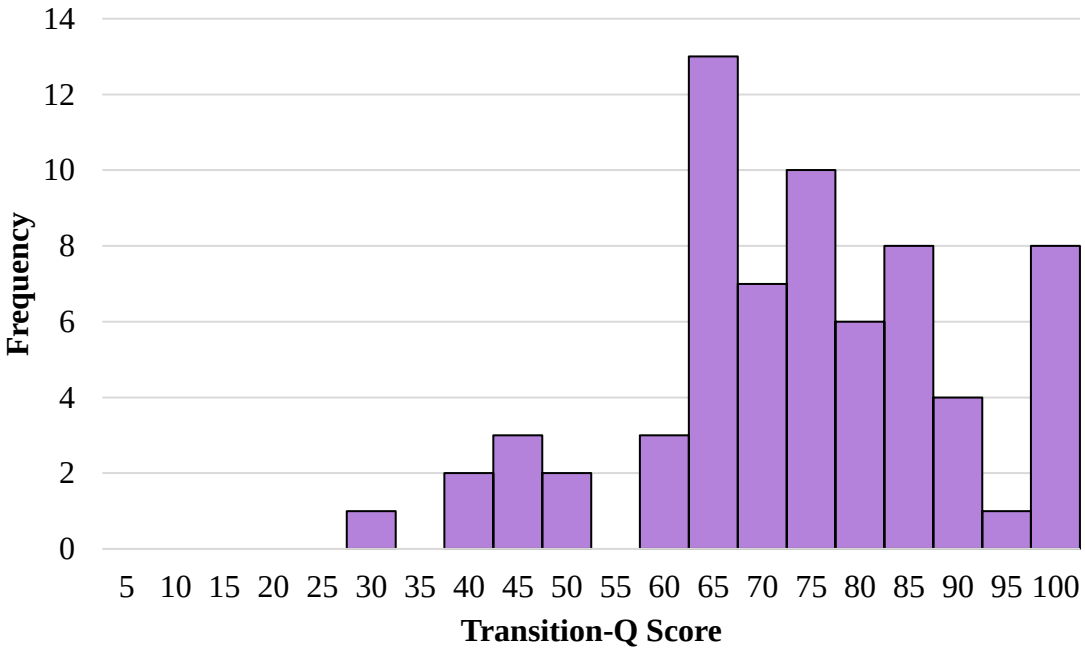


Table 4.2: AYA familial characteristics, 10-year follow-up†

n = 131 AYAs	
Household income*: Frequency (%)*	
< $20,000 – 39,999	15 (12)
$40,000 – $59,000	10 (9)
$60,000 – $79,999	15 (13)
$80,000 – $99,999	17 (14)
$100,000 – $149,999	28 (24)
$150,000+	33 (28)
Parent highest level of education attained*: Frequency (%)*	
High school	12 (10)
Vocational/technical training	18 (15)
College/university	64 (53)
Masters or PhD	26 (22)
Family demands: *mean (SD), median, [range]*	8.4 (5.9), 6.0, [0, 27]
Family adaptation: *mean (SD), median, [range]*	14.6 (4.0), 15.0, [5, 20]

† All variables have less than 10% missing data

Table 4.3: Status of epilepsy care by age group

Status of epilepsy care (AYA report)	All AYAs Frequency (%) n = 124 7 missing	AYAs ≤ 17 Frequency (%) n = 59 4 missing	AYAs 18+ Frequency (%) n = 65 3 missing
Transferred and still receives epilepsy care from adult neurologist	19 (15)	5 (8)	14 (22)
Transferred but no longer receives care	4 (3)	0	4 (6)
Receives epilepsy care from paediatric specialist	11 (9)	11 (9)	0
Receives epilepsy care from GP	6 (5)	1 (2)	5 (8)
Does not receive epilepsy care	84 (68)	42 (71)	42 (65)

Table 4.4: Bivariate regression analysis of individual factors with Transition-Q score †

n = 131 AYAs	Coefficient	P-value
Individual		
Sex (male=0)	5.4	0.1
Age at diagnosis	3.0	<0.0001
Age at 10-year follow-up	3.1	<0.0001
Epilepsy severity at baseline	-0.02	0.9
Epilepsy syndrome type at diagnosis		0.01
Benign epilepsy of childhood with rolandic spikes (BECRS)	Ref	
Primary generalized	-9.3	0.1
Absence	2.5	0.7
Simple/complex partial	-6.2	0.3
Secondarily generalized	-8.1	0.2
BECRS + secondarily generalized	3.3	0.6
Undetermined	32.8	0.02
Time since last seizure		0.2
< 6 months	Ref	
6 months – 1 year	3.1	0.8
1 – 2 years	-6.4	0.5
2 – 5 years	-5.2	0.5
5 – 10 years	2.2	0.7
10+ years	0.2	0.9
Don't remember	-14.1	0.07
Last time AEDs were taken		0.3
Currently	Ref	
< 6 months ago	-19.3	0.6
1 – 2 years ago	-7.5	0.6
2+ years ago	6.1	0.1
Never taken AEDs	8.3	0.2
Don't remember	-3.9	0.5
Status of epilepsy care (AYA report)		0.2
Transferred and still receives epilepsy care from adult neurologist	Ref	
Transferred but no longer receives care	-9.6	0.3
Receives epilepsy care from paediatric specialist	-11.2	0.1
Receives epilepsy care from GP	0.76	0.9
Does not receive epilepsy care	2.86	0.5
Health-related quality of life	0.3	0.008

n = 131 AYAs	Coefficient	P-value
CES-D ≥ 16	-3.1	0.4
Anxiety	-0.08	0.6
Physical comorbidities		0.7
No	Ref	
Yes, diagnosed before epilepsy	-5.8	0.5
Yes, diagnosed after epilepsy	4.2	0.4
Yes, some diagnosed before and some after epilepsy	-8.8	0.7
Developmental, behavioural, or psychological comorbidities	-9.6	0.007
With whom the AYA lives		0.2
Parent	Ref	
Other relative	0.9	0.9
Non-relative	13.6	0.05
Services Received, Past or Current		
Extra help with schoolwork or placement in special class for children with learning difficulties	-10.9	0.003
Speech or occupational therapy	-14.5	0.0005
Medication or therapy for behaviour or emotional problems	-5.2	0.2

† All variables have less than 10% missing data, except for variables assessing services received on which there were 15% to 19% missing data.

Table 4.5: Bivariate analysis with Transition-Q score, familial factors†

n = 131 AYAs	Coefficient	P-value
Household income		0.8
< $20,000 – 39,999	Ref	
$40,000 – $59,000	4.1	0.6
$60,000 – $79,999	6.4	0.3
$80,000 – $99,999	-1.8	0.8
$100,000 – $149,999	3.8	0.5
$150,000+	6.6	0.2
Parent highest level of education attained		0.5
High school	Ref	
Vocational/technical training	-5.0	0.4
College/university	2.3	0.6
Masters or PhD	3.3	0.6
Family demands	0.04	0.9
Family adaptation	0.6	0.2

† All variables have less than 10% missing data.

Table 4.6: Multivariable regression analysis with Transition-Q score

n = 104	Coefficient	P-value
Individual		
Sex (male=0)	2.6	0.5
Age at 10-year follow-up	3.1	<0.001
Epilepsy type at diagnosis		0.1
Benign epilepsy of childhood with rolandic spikes (BECRS)	Ref	
Primary generalized	-1.9	0.8
Absence	4.9	0.4
Simple/complex partial	-0.2	0.9
Secondarily generalized	-0.7	0.9
BECRS + secondarily generalized	3.2	0.7
Undetermined	43.5	0.007
Time since last seizure		0.7
< 6 months	Ref	
6 months – 1 year	-11.2	0.4
1 – 2 years	-4.8	0.6
2 – 5 years	-6.3	0.5
5 – 10 years	-6.4	0.5
10+ years	-0.6	0.9
Don't remember	-12.7	0.2
Status of epilepsy care (AYA report)		0.5
Transferred and still sees adult neurologist	Ref	
Transferred but no longer receives care	2.8	0.8
Seen by paediatric specialist	1.5	0.8
Seen by GP	-15.1	0.1
Does not receive epilepsy care	2.0	0.7
Health-related quality of life	0.3	0.1
Developmental, behavioural, or psychological comorbidities	-5.1	0.3
With whom the AYA lives		0.6
Parent	Ref	
Other relative	-8.6	0.3
Non-relative	-3.0	0.7
Family adaptation	0.5	0.3
Services Received, Past or Current		
Extra help with schoolwork or placement in special class for children with learning difficulties	1.9	0.7
Speech or occupational therapy	-4.0	0.4
Medication or therapy for behaviour or emotional problems	0.5	0.9

Table 4.7: Attrition analysis, baseline compared with 10-year follow-up

Variable		Mean for LTF families or Count	Mean for non-LTF families or Count	p (t-test) or p (χ^2)*
Child's sex:	Male	106	93	0.7
	Female	98	80	
Epilepsy severity at diagnosis		5	5	0.4
Epilepsy type at diagnosis				0.4
Primary generalized		26	20	
Absence		55	44	
Simple/complex partial		63	43	
Benign epilepsy of childhood with rolandic spikes (BECRS)		24	26	
Secondarily generalized		28	18	
BECRS + secondarily generalized		11	19	
Undetermined		4	3	
Family adaptation score		13	15	0.001
Family life events score (family demands)		11	8	0.001
Parental household income				<0.001
< $20,000 – 39,999		38	11	
$40,000 – $59,000		42	28	
$60,000 – $79,999		34	33	
$80,000 – $99,999		26	35	
$100,000 – $149,999		11	9	
$150,000+		31	49	
Relation of caregiver to child:				0.01
Biological parent		184	164	
Other relationship		27	9	
Relation of caregiver to child: mother				0.03
Biological mother		170	153	
Other relationship		41	20	
Parent highest level of education attained				0.001
< 8 years		2	3	
8 – 12 years		24	4	
High school		40	23	
Vocational/technical training		25	23	
College/university		86	90	
Masters or PhD		20	30	

*Mean and t-test p-values for continuous variables, count and χ^2 p-value for categorical variables

Figure 4-3: HERQULES Participant Flow Chart, Parent

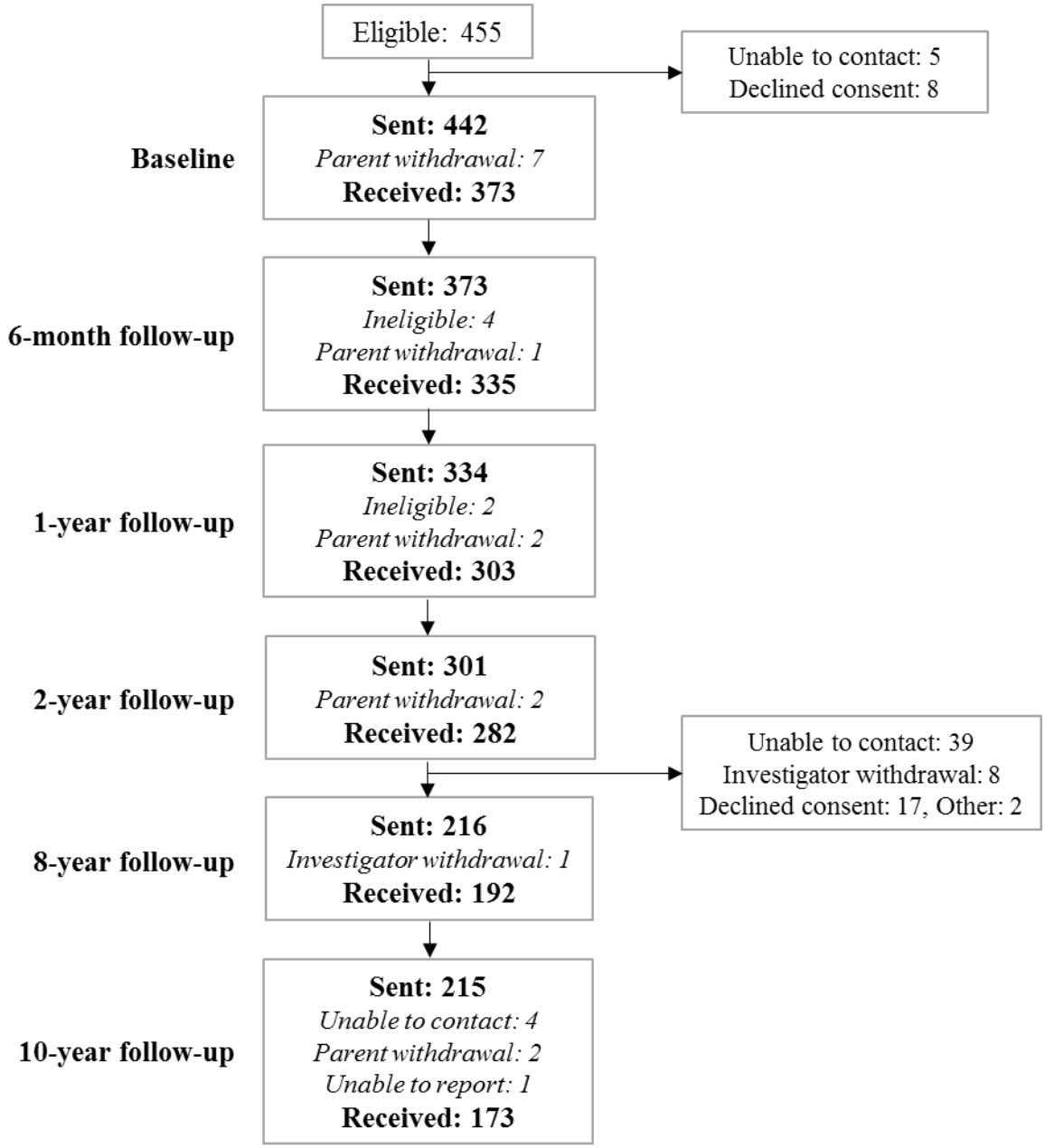
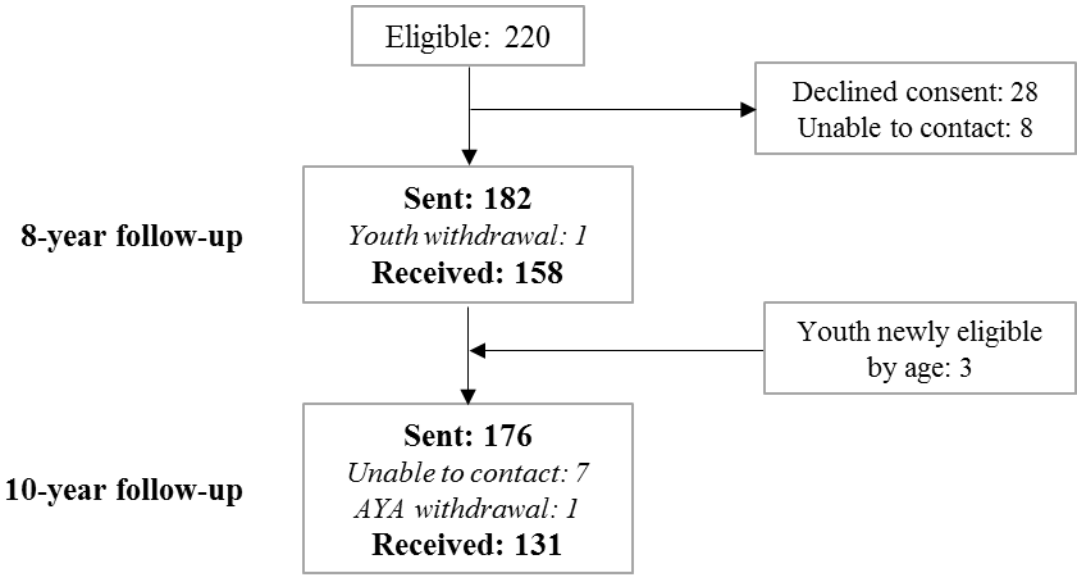


Figure 4-4: HERQULES Participant Flow Chart, AYA



Chapter 5 : Discussion

5 Chapter Overview

The final chapter discusses the results in the context of current literature and interprets key findings pertaining to each objective and their implications. A summary of key findings is then given. Next, the strengths and limitations of this research are highlighted followed by a discussion of the generalizability of the results and suggested directions for future research. Conclusions are presented in the last section.

5.1 Transition Readiness

The first objective of this thesis was to quantify the transition readiness of AYAs who were diagnosed with epilepsy in childhood. Transition-Q scores were left-skewed in our sample. The mean score observed in the sample used to validate the Transition-Q was 48, lower than our sample mean of 65.[78] The sample of n = 337 used to validate the Transition-Q included individuals aged 12 to 18 years with a chronic health condition (ex. Asthma, cerebral palsy, epilepsy). This sample had a much higher proportion of participants who were younger than 16 years of age than our sample, which may explain the lower average transition readiness.[78] Adolescents without a chronic condition were not included in this study for comparison. The age range of our sample of 12 to 23 extends several years beyond the age range of the sample in the validation study. Although differential item functioning tested in that study showed that item performance did not differ by age in their sample aged 12 to 18 years, measurement invariance could have been violated in our sample due to the presence of older individuals. This is currently the only published study that measures transition readiness with the Transition-Q. If we consider the TRAQ, one of the most commonly used transition readiness measures for research purposes, it typically has a mean score of 3 to 4 when tested in populations with chronic paediatric illnesses. [8, 12, 17] That range is the third quarter of the TRAQ's score range of 1 to 5. The mean transition readiness score we observed is also in the third quarter of the Transition-Q's score range of 0 to 100, suggesting that the mean transition readiness that we observed is similar to that observed in the literature with

chronic paediatric illnesses other than epilepsy. To date, a minimal clinically important difference has not been established for the Transition-Q to assist in interpreting scores. However, the broad distribution and low average transition readiness relative to the theoretical range suggest that there is much room for improvement through transition programming.

Although differences in average transition readiness between categories of epilepsy care status were not significantly different it is interesting to note that on average, AYAs who no longer receive care for epilepsy had the highest transition readiness. As adolescents with epilepsy are generally only referred to an adult neurologist if their epilepsy is still active, it is likely that those who did not transfer/transition to adult care and no longer see a physician for epilepsy have not had a seizure in many years. In fact, a post-hoc analysis showed that 79% of those who no longer receive care for epilepsy had not had a seizure in over five years and were statistically significantly less likely to have a developmental, behavioural, or psychological comorbidity. It has been found that on average, AYAs with a chronic condition have lower transition readiness than AYAs without a chronic condition[17], so perhaps those whose seizures were controlled earlier do not consider themselves to have a chronic condition, potentially contributing to their greater self-management abilities. The finding that AYAs who had been diagnosed with a developmental, behavioural, or psychological comorbidity had significantly lower transition readiness on average supports this idea. One possible mechanism could be that the presence of a chronic condition causes parents to take more responsibility for their child's health, further hindering the youth's ability to develop self-management skills.

It was also not surprising that on average, AYAs who transferred/transitioned to adult healthcare but no longer receive care for epilepsy had lower transition readiness than all other groups (except for those who receive epilepsy care from a paediatric neurologist). This could be because those who transferred but no longer receive care did not have the readiness skills necessary to maintain contact with their adult neurologist.

5.2 Factors Associated with Transition Readiness

The second objective was to explore the associations of individual and familial factors with transition readiness. The following individual variables were found to be significant in the bivariate analyses with Transition-Q score: age at diagnosis; epilepsy type at diagnosis; time since last seizure; last time AEDs were taken; developmental, behavioural, or psychological comorbidities; status of epilepsy care; age at 10-year follow-up, sex, HRQL, with whom the AYA lives; extra help with schoolwork or placement in special class for children with learning difficulties (past or current); speech or occupational therapy (past or current); and medication or therapy for behaviour or emotional problems (past or current). One familial variable, family adaptation, was found to be significant in the bivariate analyses with transition readiness. In the multivariable analysis, only age at 10-year follow-up retained a significant relationship with transition readiness.

As expected, we found that that level of transition readiness was positively associated with age. This result is consistent with several studies using the Transition-Q and other measures of transition readiness like the TRAQ, which have shown a positive relationship between age and transition readiness.[8, 9, 10, 77, 78] Sex had a weak but significant relationship with transition readiness in the bivariate analysis which disappeared when included in the multivariable analysis. Psychology and neurophysiology literature leans toward the theory that on average, girls outperform boys of the same age in most cognitive areas (other than spatial reasoning) during youth and adolescence.[108] This would suggest that perhaps girls would take more responsibility for their healthcare than boys and have a higher transition readiness score on average. Transition literature has not reached a consensus on this relationship with two studies showing no relationship[9, 11] and two showing that females have higher transition readiness. [8, 12]

Although two out of five clinical epilepsy factors were significant in the bivariate analyses (at $p < 0.05$ significance level), none of those included the multivariable analysis were significant at the conventional significance level of 0.05. It was expected that more severe epilepsy at diagnosis may make it more likely for parents to actively teach their

children about epilepsy and its management and to raise awareness of healthcare management in general. In addition, that having more immediate epilepsy concerns like a recent seizure would make AYAs more aware of healthcare responsibilities. However, these results suggest that the clinical features of epilepsy we examined do not have an independent effect on transition readiness. This is in agreement with the existing literature for AYAs with kidney transplant[11], irritable bowel disease[8], and several other diseases[9] in which time since diagnosis and transplant were found to have non-significant relationships in univariate and multivariable analyses. That being said, there are other clinical factors that need to be further investigated before concluding that they are not associated with transition readiness. It is possible that the severity of epilepsy at the AYAs' last visit with a neurologist may have been more important than severity at diagnosis, but this information was unknown for a large proportion of AYAs and was therefore not analysed.

Despite these nonsignificant findings, it is interesting to note the trends in coefficients for these epilepsy-related variables in the bivariate analyses. Those who took AEDs in the past 6 months had the lowest transition readiness. Transition readiness increased with increasing time since last seizure. In addition, those diagnosed with BECRS + secondarily generalized seizures had the highest transition readiness of the recorded epilepsy types (those of undetermined type had much higher transition readiness, but since there were only three observations in this category, results are not reliable). These trends may merit further investigation in future research especially considering their usefulness for clinicians as predictors of transition readiness.

Neither past/current extra help with schoolwork or placement in special class for children with learning difficulties; speech or occupational therapy; nor medication or therapy for behaviour or emotional problems were significant in the multivariable analysis. That extra help with schoolwork or placement in special class for children with learning difficulties did not have a significant relationship with transition readiness is contrary to the previous finding that those in special education had statistically significantly lower odds of being ready for transition.[9] It is possible that placement in a special class affects transition readiness but needing extra help with schoolwork does not, and combining

them in a single variable obscured the individual effect. This may account for the discrepancy. Initially, it seemed reasonable to speculate that those who are receiving these services may have higher transition readiness given their greater exposure to more healthcare services. Weakening of the relationships with transition readiness of each of the "services received" variables after multivariable analysis may indicate that this is not the case. Of course, it could be that exposure to additional services is not as relevant a contributor to transition readiness as is coordination among services. No other studies have investigated service use not directly related to the chronic disease under study (ex. For comorbidities).

Considering the size of their coefficients in relation to their scale ranges in the multivariable model, HRQL had a very weak and non-significant relationship with transition readiness, and anxiety did not have a relationship with transition readiness. A previous study found a significant relationship between general health and disease impact-related well-being with transition readiness in bivariate analyses which were not significant after multivariable analysis.[9] That is the only study that has searched for a link between measures of general health/quality of life and transition readiness. Neither general anxiety nor depression have been investigated for a relationship with transition readiness in any other studies. Several studies have shown that for individuals with diabetes, having even sub-clinical depressive symptoms negatively affects their diabetes self-management. For example, one study showed that adults with diabetes who had major depression as measured by a self-report questionnaire had statistically significantly higher odds of performing self-care behaviours infrequently compared to those without major depression, for six out of ten self-care behaviours.[109] It has also been proposed that disease-specific psychological distress may have a stronger effect on self-management than general psychological distress. One review article proposed that for individuals with diabetes, disease-specific anxiety may have a stronger relationship with self-management than depression and general distress.[110] Our findings could be compatible with that proposal.

This research is the first to our knowledge to investigate comorbidities for a relationship with transition readiness. Research in adults with multimorbidity suggests that having

more than one chronic condition negatively affects self-management and/or patient outcomes, although evidence for this relationship is limited.[111, 112, 113] As self-management is one component of transition readiness and the component that the Transition-Q measures, we should expect to see relationships between transition readiness and physical comorbidities and with developmental, behavioural, or psychological comorbidities. Studies often quantify multimorbidity based on the number of comorbidities and the clinical characteristics of the comorbidities when investigating its relationship with self-management and patient outcomes, while we separated comorbidities and categorized physical comorbidity by the chronology of diagnosis in relation to epilepsy.

We did not find a significant relationship with transition readiness for any of the familial factors we investigated. While this research is the first to investigate familial factors for a relationship with transition readiness, a recently published Nova Scotian cohort study which followed children diagnosed with epilepsy for approximately 25 years found that those whose families were in the top two income quintiles at the time of epilepsy onset were significantly less likely to experience negative social outcomes like unemployment and not finishing high school.[114] In multivariable analysis, neither income nor parental education were associated with seizure outcome. In light of this result, it seems unlikely that the family factors examined in the Nova Scotian study and in this research would strongly influence transition readiness.

Overall, the results of the multivariable regression analysis were not as expected. Based on theory, limited available evidence, and clinical expertise, we expected to find several significant relationships with transition readiness. We had fewer observations per covariate in the multivariable analysis than recommended; 4 events per variable (due to dummy coding of non-ordinal categorical variables, although there were 12 factors being regressed on there were 26 covariates) versus the guideline of 10.[115] A small number of observations per variable in linear regression makes it more likely for the strength and significance of associations to be biased. Therefore, this type of bias may have obscured some existing relationships. Another possible explanation could be that of the several components of transition readiness, the Transition-Q measures only one: number and

frequency of self-management skills. The inclusion of other components like health literacy and disease-specific health knowledge may strengthen some associations. Of course, our findings may have deviated from the expected because no relationships actually exist between transition readiness and the non-significant covariates we tested.

One new transition readiness measure has been published since the 10-year follow-up of HERQULES, the STARx.[116] It is a generic, 18-item, AYA-report questionnaire. One cross-sectional study has used the STARx to quantify transition readiness.[14] One fifth (160/781) of attendees of a therapeutic camp for youth aged 6 to 16 years with a paediatric chronic condition including cerebral palsy, diabetes, cancer, and neurologic conditions responded by online questionnaire. This study explored associations between method used to learn about the transition process and transition readiness, an association which was not examined in our research.

This area of the literature is clearly undeveloped. Approximately eleven studies have investigated the relationship of transition readiness with individual factors, and none have investigated the relationship with familial factors. Adjustment for covariates was done in only a few of these studies. As the second study to conduct a preliminary analysis on factors associated with transition readiness and the first study to our knowledge to investigate the relationship of transition readiness with familial factors, this research provides an important contribution to transition literature.

5.3 Transfer/Transition Experiences

The third objective was to summarize the transfer/transition experiences of AYAs diagnosed with epilepsy in childhood and their parents/caregivers.

5.3.1 AYA Contact with Healthcare

It is encouraging that none of the AYAs who reported that they received care from a physician for reasons related to epilepsy or seizures within the past two years reported going to an emergency room or urgent care/walk-in clinic. They were accessing care from their paediatric or adult specialist, or their GP. As the vast majority of these AYAs were currently taking AEDs, it is likely that they received ongoing medical management

while being followed regularly for active epilepsy. For those not currently taking AEDs, this result suggests that when an epilepsy-related issue arose AYAs were able to access care from their regular healthcare providers, or that they had not experienced an epilepsy-related emergency. Interpretation of this result is somewhat limited because data were collected regarding only the last time healthcare was accessed and we do not know the reason for the visit. While these observations are potentially encouraging, more data are needed to determine healthcare access patterns of AYAs diagnosed with epilepsy in childhood.

Interestingly, six AYAs who reported that they no longer receive care from a physician for epilepsy or seizures had visited a physician for reasons related to epilepsy or seizures in the last two years. This reveals a potential concern: that AYAs whose epilepsy is in remission may no longer consider themselves as having epilepsy. This is a theme that has been observed informally while contacting HERQULES families. It is worrisome that some may not know how childhood epilepsy could affect them as an adult and they may be unable to recount their medical history accurately in an emergency. This phenomenon is a worthwhile topic for future research as it remains an uninvestigated barrier to effective healthcare access. Similarly perplexing was the observation that a few AYAs who indicated that they currently taking AEDs also reported that they no longer receive care for epilepsy from a physician.

Regarding discussion about transfer/transition with a physician, one study of AYAs with inflammatory bowel disease found that 9/29 (30%) AYAs reported that their paediatric physician had discussed transition to adult care with them, higher than the 19% we observed in our sample. This could be because most of our sample were considered to be in remission and thus did not require follow-up with an adult neurology specialist according to current conventional practice. It is somewhat worrying that although the evidence suggests that transition programming is beneficial for the future health of AYAs diagnosed with a chronic paediatric illness, the large majority of AYAs and parents/caregivers did not think that a discussion about transfer/transition would be/would have been helpful to them. It is possible that families were unfamiliar with the

concept of transitioning to adult care since formal transition programming was uncommon until very recently.

5.3.2 Parent/Caregiver Response Themes

Each theme that we identified based on AYA and parent responses to the open-ended questions were also observed in the transition literature for chronic paediatric conditions like diabetes, cystic fibrosis, and inflammatory bowel disease. The themes of wanting a joint meeting with paediatric and adult physicians, enjoying being treated as an adult, and wait time for an appointment with their adult neurologist as described by AYAs are well-documented.[40, 59, 117] For parents/caregivers, the themes of alienation and worrying that their child is not ready to manage his/her own healthcare are also well-recognized.[40, 59, 118]

Almost all studies on transfer/transition experiences of AYAs with epilepsy have focused on those who have severe epilepsy syndromes and cognitive impairment using only parent report of experiences.[61, 62, 118] One exception is a study in which 30 AYAs with epilepsy and 28 of their parents were interviewed.[95] The clinical characteristics of epilepsy for the sample were not reported. The most common themes identified by AYAs were difficulty understanding and communicating with their adult neurologist, which were not raised by our sample. Other experiences reported by that group of AYAs that were not raised in our sample were unwillingness to talk about epilepsy with their parents and reproductive concerns. Some similar themes were expressed in both, such as enjoying being treated as an adult. Parents expressed concerns that they did not know enough about their child's diagnosis prior to transfer, that their child would not be able to manage their own healthcare, and that physicians did not discuss their child's health in layman's terms, causing feelings of alienation.

The experiences reported by our sample were quite homogeneous and less detailed compared to those observed in other studies. This is likely because the vast majority of studies collected experiences by semi-structured interview while we collected experiences through written self-report. Interviewers can obtain more detailed responses with probing, which is much more difficult with a self-administered questionnaire.

We observed that the experiences reported by AYAs and parents/caregivers in our sample were not specific to epilepsy and had much in common with the experiences of AYAs with other chronic conditions. In the context of the literature, these findings are in agreement with the position of the PCMCH of Ontario, that a universal transition framework could be used for all chronic paediatric illnesses which can then be customized for the unique complications of each condition.[19] Although the majority of AYAs and parents thought that a discussion about transfer/transition would not have been helpful to them, this may have been because transition has not been widely adopted and thus its benefits may not be known to patients. It is evident from the Transition-Q scores that the number and frequency of self-management skills mastered by these AYAs is mediocre and may benefit from transition programming.

Our results support the literature in suggesting that transition practices are not yet implemented consistently across Canada. The AYA and parent responses showed that some AYAs with epilepsy have positive transfer/transition experiences and felt supported and comfortable during the process while others felt that they had been neglected by the adult healthcare system. Not to mention that many reported that transfer/transition had not been discussed with them. This is not surprising considering that only five formal transition programs exist in Canada[52] and Ontario is the only province with published provincial transition recommendations for adolescents with a chronic condition.[19]

5.4 Summary of Findings

The AYAs assessed in this study were doing well in terms of epilepsy outcome. The majority were no longer taking AEDs and had not had a seizure in over five years. Most AYAs were no longer receiving care for epilepsy from a physician. Almost one fifth had transferred/transitioned to adult healthcare. Although only a few years into adulthood, almost all were either in school or employed. They also appear to have favorable family environments: relatively few family demands on average, the majority had an annual parental household income of over $80,000, and over two thirds of parents had at least a college or university degree. Despite these seemingly positive circumstances, half of our sample had developmental, behavioural, or psychological, comorbidities; over one third

had potentially clinically relevant depression symptoms; and average transition readiness was mediocre.

In our sample, age was the most important factor affecting transition readiness while familial factors did not appear to have much influence. Trends in the bivariate analyses also suggested that early seizure control and living with relatives may result in better transition readiness; having a comorbid condition may impair transition readiness; and epilepsy type at diagnosis may influence transition readiness. There were several concerns of insufficient contact with healthcare providers. Similarly, the representation of both positive and negative transfer/transition experiences may further indicate inconsistency in implementation of transition recommendations across Canada.

5.5 Strengths and Limitations

One of the strengths of this work is the HERQULES sample itself. As HERQULES was a prospective cohort study that followed families for 10 years since the time of diagnosis of epilepsy in children aged 4 to 12 years of age, we were in an ideal position to observe the transition readiness and experiences of AYAs with a wide range of epilepsy outcomes. As a multi-center study that contacted all paediatric neurologists in Canada, the recruitment method of the baseline sample was strong. Adapting the Tailored Design Method[75] for surveys and having multiple times of contact with the same study coordinator established a relationship with the families that may have made them more likely to continue participating at each follow-up.

Another strength is that as HERQULES included children with a wide range of epilepsy types and severity, the 10-year follow-up sample had a high proportion of AYAs who had grown out of their epilepsy which is representative of the outcome of most AYAs diagnosed with epilepsy in childhood. Transition research thus far has mostly been concerned with very burdening/limiting diseases and this work provides a perspective on the transition readiness and experiences of the majority of AYAs diagnosed with epilepsy in childhood who are currently severely under-represented in the literature.

We used a reliable, validated, disease-general scale to measure transition readiness, which follows the recommendations of several professional medical organizations. The Transition-Q is currently being used in multiple paediatric health centers across Ontario.[119, 120] Contributing to the use of this scale will facilitate comparison between future studies that use the Transition-Q, enhancing the applicability of previous and future studies of transition readiness.

One of the limitations of this work was that the self-administered questionnaire was not an ideal method of collecting information on such an amorphous topic as transfer/transition experiences. Among AYAs who moved from paediatric to adult healthcare, we would have preferred to be able to differentiate between AYAs who were transitioned and AYAs who were transferred to adult healthcare. This would have been cumbersome on a questionnaire because it would require a list of possible transition elements which the AYA would have to read through and select as part of a complex skip pattern. Classifying transfer/transition experiences based on a written response would also be difficult because the individual's meaning could not be clarified by probing, especially considering that AYA descriptions of transfer/transition experiences were extremely short.

The sample size available for the multivariable regression analysis puts some restriction on its interpretation. The regression coefficients and p-values are likely unpredictably biased as the number of participants per variable is below ten. Additionally, we would have liked to examine the most recent clinical features of epilepsy in the multivariable regression analysis. However, only a small proportion of AYAs had recent data from physician forms available for analysis and too much of these data were missing to justify inclusion in the analysis.

As the attrition analysis showed, our results were likely affected by attrition bias. The families who participated in the 10-year follow-up were relatively advantaged in terms of income and level of family functioning. This could partially explain the lack of significant associations in the multivariable analysis. Another possible explanation for this could be that the Transition-Q quantifies one of several domains that make up the

concept of transition readiness (self-management skills). It is possible that the "full picture" of transition readiness[121] was not represented in the outcome, thus limiting the degree to which significant associations were observed.

5.6 Generalizability of Results

The outcomes of our sample were typical of those observed in other longitudinal studies of adults diagnosed with epilepsy in childhood. One longitudinal Nova Scotian study of a cohort of children diagnosed with epilepsy in childhood reported outcomes for those with rolandic epilepsy (also known as BECRS) 30 years after diagnosis.[23] There were 42 participants, all of whom were considered to be in "terminal remission" at the 30-year follow-up. Virtually all of those with rolandic epilepsy were employed, 3 (10%) were diagnosed with attention-deficit/hyperactivity disorder, and 10 (31%) had a behaviour disorder. The follow-up of this cohort is substantially longer than the HERQULES follow-up and their reports were of specific subgroups of the cohort, however epilepsy and social outcomes still appear similar.

The Connecticut Study of Epilepsy followed an original sample of 613 children newly diagnosed with epilepsy for nine years and reported the outcomes of 277 families.[122] In HERQULES, we observed that ten years after their diagnosis of epilepsy, 18% had ever been diagnosed with a physical comorbidity, 14% with depression, 23% with anxiety, 20% with attention deficit disorder or attention deficit hyperactivity disorder, 6% with oppositional defiant disorder, 2% with a conduct disorder, 14% with a developmental delay, and 3% with autism, pervasive developmental disorder, or Asperger's syndrome. Our findings are generally comparable to the Connecticut study, however we observed twice the proportion of depression and half the proportion of developmental delay. The Connecticut study also reported that 64% of their sample had not had a seizure for five years or more and 31% were taking AEDs, comparable to the 59% and 28% that we observed.

The Dutch Study of Epilepsy in Childhood followed 413 individuals diagnosed with epilepsy in childhood recruited from multiple centers for 15 years. Our sample had better social outcomes; one third of their sample was employed compared to half of our sample

and half of their sample who were aged 15-24 years were in school compared with 80% in our sample.[123] The clinical epilepsy outcomes were similar to those for the HERQULES sample. It was found that almost half of participants had not had a seizure in the last 13 years of follow-up and one third were still being treated for seizures at the end of follow-up.[124]

As those who could not complete the questionnaire on their own were excluded from participating in the study, AYAs with severe cognitive impairment were not represented in this research. Although our sample is not representative of the entire population of AYAs diagnosed with epilepsy in childhood, those who have severe cognitive impairment would nevertheless require a different tool to measure transition readiness especially if they require a caregiver to make healthcare decisions for them. Thus, it is reasonable to assume that our results are only generalizable to AYAs diagnosed with epilepsy in childhood who do not have severe cognitive impairment.

5.7 Future Research

This research uncovered several trends that are worthy of further exploration. The multivariable regression analysis showed that many of the factors we investigated had interesting patterns of association with transition readiness. Based on our results, it may be fruitful to pursue further research in this area involving clinical features of illnesses, the living arrangement of adolescents, and the presence of comorbidities. We also found that some families experienced a smooth transfer while others were quite disjointed. Future research may explore which components of transition produce the best outcomes for patients and their families so that a unified approach can be developed and implemented across health disciplines and consistently across the country.

Ideally, future studies on the factors affecting transition readiness would recruit adolescents before transition has begun and follow them until after the final transfer has taken place. One obstacle to widespread adoption of formal transition programming is the lack of knowledge about which components of transition have the most impact on transition readiness and patient outcomes. Therefore, it is imperative that future research investigate the link between components of transition, transition readiness, and patient

outcomes. To do this, the procedural details of each adolescent's transfer/transition and/or participation in a formal transition program should be recorded to distinguish those who were transferred from paediatric to adult healthcare from those who transitioned. Long-term follow-up is also important for collecting data on social outcomes like education, employment, and family structure as social outcomes are essential for assessing transition success. A key contributor to the lack of knowledge regarding the key components for successful transition is that a standard transition readiness measure has not been adopted by researchers and clinicians. Widespread use of a reliable transition readiness measure with sound psychometric properties and applicability to all chronic conditions is needed to be able to make concrete progress in understanding the impact of transition programming on long-term patient outcomes and the role that transition readiness plays.

Literature on diabetes and depression has suggested that disease-specific anxiety or distress may have a much stronger impact on transition readiness than depression and general anxiety. It would be interesting to examine the relationship of epilepsy-specific anxiety with transition readiness. We have access to this construct in the HERQULES data in the form of epilepsy impact and seizure worry on the QOLIE-AD-48 and QOLIE-31-P HRQL scales, respectively, so an opportunity exists to investigate this in future analyses.

5.8 Conclusions

Evidence is mounting that transition from paediatric to adult healthcare achieves superior long-term outcomes for adolescents with a chronic paediatric illness and their families compared with current standard practice. Yet, very few formal transition programs exist. Moreover, it is not known how widely transition recommendations are being applied or how prepared AYAs with a paediatric chronic condition are to manage their healthcare independently. To our knowledge, this is the first study to quantify the transition readiness and collect the transfer/transition experiences of AYAs diagnosed with epilepsy in childhood without significant cognitive impairment. Thus, this research provides the perspective of a large proportion of AYAs who are severely under-represented in the literature thus far.

Our sample of AYAs diagnosed with epilepsy in childhood were generally doing well ten years after their diagnosis. Most were in school or employed and over two thirds were no longer taking AEDs. The results are cautiously optimistic as our sample of AYAs were only a few years into adulthood; previous studies have shown that epilepsy issues often resurface and social outcomes are less positive than their healthy peers.[24, 32] On the other hand, their knowledge and application of self-management health skills was lacking as indicated by the mediocre average transition readiness. This may have implications for their future health. An estimated one third of adults with remitted epilepsy will have at least one seizure later in life.[28] We also know that developmental, behavioural, and psychological comorbidities which require monitoring by a physician and self-management are common in this population.[34, 35] In fact, a notably large proportion of our sample displayed clinically relevant depressive symptoms. A mastery of self-management skills would likely allow adolescents to manage their health effectively not only during this vulnerable time, but throughout their lives.

This research showed that age is likely an important factor affecting transition readiness. It may also be worthwhile for future research to investigate the roles of clinical features of epilepsy, living situation, and comorbidities in the development of transition readiness. Publication of studies currently being conducted by other researchers using the Transition-Q will allow further interpretation of the implications and clinical relevance of Transition-Q scores we observed.

Both parent and AYA report of transfer/transition experiences indicated that transition has not been widely implemented in Canada. The benefits of transition are likely not known to parents and AYAs and they may not even be aware of the concept of transition. The reported experiences were in agreement with those observed for AYAs with other chronic conditions and compatible with the widely-held viewpoint that a single transition framework could be implemented for all chronic conditions.

This work provided an initial perspective on the transition readiness and experiences of adolescents and young adults diagnosed with epilepsy in childhood. We hope that our findings will contribute new hypotheses for investigation in future research and inspire

the continued growth of transition programming for adolescents living with a paediatric chronic illness.

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Appendices

Appendix A: Ethics Approval Notice

Western Research

Research Ethics

Western University Health Science Research Ethics Board
HSREB Amendment Approval Notice

Principal Investigator: Dr. Kathy Speechley
Department & Institution: Schulich School of Medicine and Dentistry\Epidemiology & Biostatistics,Western University

Review Type: Expedited
HSREB File Number: 102819
Study Title: Health-related quality of Life in children with New-Onset Epilepsy: A Long-term Follow-up
Sponsor: Canadian Institutes of Health Research

HSREB Amendment Approval Date: September 14, 2015
HSREB Expiry Date: September 06, 2016

Documents Approved and/or Received for Information:

Document Name	Comments	Version Date
Change in Study Personnel	Incoming Research Assistants (S. Brar & A. Dasiewicz)	2015/08/27
Revised Western University Protocol		2015/08/27
Other	Cover Letter: Time 6 Parent Questionnaire	2015/08/19
Instruments	Time 6 Parent Questionnaire	2015/08/27
Other	Cover Letter: Time 6 Youth 11-17 Questionnaire	2015/08/19
Instruments	Time 6 Youth 11-17 Questionnaire	2015/08/27
Other	Cover Letter: Time 6 Youth 18+ Questionnaire	2015/08/17
Instruments	Time 6 Youth 18+ Questionnaire	2015/08/27
Other	Email/Cover Letter: Time 6 Youth 18+ Questionnaire	2015/08/19
Other	Email/Cover Letter: Time 6 Youth 11-17 Questionnaire	2015/08/19

The Western University Health Science Research Ethics Board (HSREB) has reviewed and approved the amendment to the above named study, as of the HSREB Initial Approval Date noted above.

HSREB approval for this study remains valid until the HSREB Expiry Date noted above, conditional to timely submission and acceptance of HSREB Continuing Ethics Review.

The Western University HSREB operates in compliance with the Tri-Council Policy Statement Ethical Conduct for Research Involving Humans (TCPS2), the International Conference on Harmonization of Technical Requirements for Registration of Pharmaceuticals for Human Use Guideline for Good Clinical Practice Practices (ICH E6 R1), the Ontario Personal Health Information Protection Act (PHIPA, 2004), Part 4 of the Natural Health Product Regulations, Health Canada Medical Device Regulations and Part C, Division 5, of the Food and Drug Regulations of Health Canada.

Members of the HSREB who are named as Investigators in research studies do not participate in discussions related to, nor vote on such studies when they are presented to the REB.

The HSREB is registered with the U.S. Department of Health & Human Services under the IRB registration number IRB 00000940.

Ethics Officer to Contact for Further Information

Erika Basile	Grace Kelly	Mina Mekhail	Vikki Tran
ebasile@uwo.ca	grace.kelly@uwo.ca	mmekhail@uwo.ca	vikki.tran@uwo.ca

This is an official document. Please retain the original in your files.

Appendix B: HERQULES Physician Form, 10-Year Follow-up

Health-related quality of Life in children with New-Onset Epilepsy: A Long-term Follow-up (HERQULES 2)

Study ID __ __ __ __ __ __ Date of Birth: ___ ___ ___ (dd / mmm / yyyy) **Time 6** Physician ID __ __ __

Please answer the following questions based on information from patient's most recent visit and return upon completion.

1. Date of patient's last visit: _ _ / _ _ / _ _ _ _ (dd / mm / yyyy)

2. Date form completed: _ _ / _ _ / _ _ _ _ (dd / mm / yyyy)

3. How long have you provided epilepsy care for this patient?

___ ___ Months OR __ __ Years

4. Seizure type(s):1) _____

2) _____

3) _____

5. Epilepsy syndrome: _____

6. Does this patient have convulsive status epilepticus?
☐ No
☐ Yes ⇨ Last episode _ _ / _ _ _ _ (mm / yyyy)

7. Does this patient have exclusive nocturnal seizures?
☐ No
☐ Yes

8. Does this patient have any family with epilepsy?
☐ No
☐ Yes

9. Number of AEDs ***currently***: _____

10. Number of AEDs ***ever tried***: _____

11. Does the patient have behavioural problems?
☐ No (normal)
☐ Yes → Please check one: ☐ mild ☐ moderate ☐ severe
Diagnosis _____

12. Does the patient have cognitive problems?
☐ No (normal)
☐ Yes → Please check one: ☐ borderline ☐ mild ☐ moderate ☐ severe
Diagnosis _____

13. Does this patient have motor problems?
☐ No
☐ Yes → Please check one: ☐ mild ☐ moderate ☐ severe
Diagnosis _____

14. Does this patient have any other neurological deficits?
☐ No
☐ Yes → Please specify: _____

15. Taking into account all aspects of this patient's epilepsy, how would you rate its severity at his/her last visit? Please check one answer.

☐ Extremely severe
☐ Very severe
☐ Quite severe
☐ Moderately severe
☐ Somewhat severe
☐ A little severe
☐ Not at all severe

16. Rate the following aspects of this patient's epilepsy at his/her last visit.

Check one box using the following 7-point scale: **1 = none or never** **7 = extremely frequent, severe or high**

	1	2	3	4	5	6	7
Frequency of seizures							
Intensity of seizures							
Falls or injuries during seizures							
Severity of post-ictal period							
Amount of antiepileptic drugs							
Side effects of antiepileptic drugs							
Interference of epilepsy or drugs with daily activities							

17. Is there any additional important information about this child's epilepsy not captured by our questions?

_____

_____

Please scan and email completed form now to **jane@herqules2.com**

OR

Return in postage paid envelope provided.

Version Date: June 19, 2012

Appendix C: Transition-Q

Self-Management Skills scale: These questions are about being in charge of your health. For each question, please circle only 1 answer.

	Never	Sometimes	Always
a) I answer a doctor's or nurse's questions.	0	1	2
b) I help to make decisions about my health.	0	1	2
c) I am in charge of taking any medicine that I need.	0	1	2
d) I talk to a doctor or nurse when I have health concerns.	0	1	2
e) I look for an answer when I have a question about my health.	0	1	2
f) I talk about my health condition to people when I need to.	0	1	2
g) I ask the doctor or nurse questions.	0	1	2
h) I speak to the doctor instead of my parent(s) speaking for me.	0	1	2
i) I summarize my medical history when I am asked to.	0	1	2
j) I contact a doctor when I need to.	0	1	2
k) I see the doctor or nurse on my own during an appointment.	0	1	2
l) I drop off or pick up my prescriptions when I need medicine.	0	1	2
m) I travel on my own to a doctor's appointment.	0	1	2
n) I book my own doctor's appointments.	0	1	2

Appendix D: Transfer/Transition Experiences Questions, Parent and AYA Versions

Parent Version	AYA Version
Section introduction: When this study began back in 2004, all of the participating families had children who were diagnosed with epilepsy by specialists who treat children with epilepsy (pediatric neurologists). Things have changed since then. Participants are older and some can no longer receive care from doctors who treat children. Also, some have outgrown their epilepsy and are not receiving health care for epilepsy.	*Section introduction:* When this study began back in 2004, all of the participating families had children who were diagnosed with epilepsy by specialists who treat children with epilepsy (pediatric neurologists). Things have changed since then. Participants are older and some can no longer receive care from doctors who treat children. Also, some have outgrown their epilepsy and are not receiving health care for epilepsy.
6.1. Did your doctors or other health care providers talk with you or your child about eventually seeing doctors or other health care providers who treat adults? ☐ No → Would a discussion about doctors who treat adults be helpful/have been helpful to you? ☐ No ☐ Yes ☐ Yes → How helpful was that? ☐ Not at all helpful ☐ Somewhat helpful ☐ Very helpful	7.2. Did your doctors or other health care providers talk with you about eventually seeing doctors or other health care providers who treat adults? ☐ No → Would a discussion about doctors who treat adults be helpful/have been helpful to you? ☐ No ☐ Yes ☐ Yes → How helpful was that? ☐ Not at all helpful ☐ Somewhat helpful ☐ Very helpful

Parent Version	AYA Version
6.2. Did your son's/daughter's doctors or other health care providers ever talk with you or your son/daughter about his/her health care needs in adulthood as he/she was growing up? ☐ No ⟶ Would a discussion about health care needs be/have been helpful? ☐ No ☐ Yes ☐Yes	No equivalent
6.3. How often do/did your son's/daughter's doctors or other health care providers encourage him/her to take responsibility for his/her health care needs, such as taking medication, understanding (his/her) health, or following medical advice? ☐ Never ☐ Sometimes ☐ Usually ☐ Always	No equivalent

Parent Version	AYA Version
6.4. We would like to learn about your son's/daughter's situation. Which of the following best describes his/her experience? ☐ Transferred from a paediatric specialist to an adult neurologist, who still provides care for epilepsy/seizures. ☐ Transferred from a paediatric specialist to an adult neurologist but no longer receives care for epilepsy/seizure from him/her. ☐ Still being cared for by a pediatric specialist. GO TO QUESTION 6.6 ☐ Receives care for epilepsy or seizures from a family doctor/general practitioner (GP). GO TO QUESTION 6.6 ☐ Does not receive care for epilepsy or seizures from any doctors now. GO TO QUESTION 6.6 ☐ None of the above. Please describe your experience:_____ _____ _____ _____ _____GO TO QUESTION 6.6	7.3. Which of the following best describes your experience? ☐ I transferred from a pediatric specialist to an adult neurologist, who still provides care for my epilepsy/seizures. ☐ I transferred from a pediatric specialist to an adult neurologist but I no longer receive care for epilepsy/seizures from him/her. ☐ I am still being cared for by a pediatric specialist. GO TO QUESTION 7.5 ☐ I receive care for epilepsy or seizures from a family doctor/general practitioner (GP). GO TO QUESTION 7.5 ☐ I do not receive care for epilepsy or seizures from any doctors now. GO TO QUESTION 7.5 ☐ None of the above. Please describe your experience:_____ _____ _____ _____GO TO QUESTION 7.5

Parent Version	AYA Version
6.5. Please tell us about your family's experience of transitioning from health care for children to health care for adults. a) What did you like about your experience? _____ _____ _____ b) What did you not like about your experience? _____ _____ _____ c) What would have helped to make your transition experience better? _____ _____ _____	7.4. Please tell us about your experience of transitioning from health care for children to health care for adults. a) What did you like about your experience? _____ _____ _____ b) What did you not like about your experience? _____ _____ _____ c) What would have helped to make your transition experience better? _____ _____ _____

Parent Version	AYA Version
No equivalent	7.5. Please think back to the last time you went to a doctor for any reason **related to epilepsy or seizures**. Approximately how long ago was that? (It is OK to provide your best guess.) ☐ Less than 6 months ago ☐ 6 months to less than 1 year ago ☐ 1 year to less than 2 years ago ☐ 2 years to less than 5 years ago → GO TO QUESTION 7.8 ☐ 5 years to less than 10 years ago → GO TO QUESTION 7.8 ☐ More than 10 years ago → GO TO QUESTION 7.8
No equivalent	7.6. What kind of doctor did you last see for any reason **related to epilepsy or seizures**? ☐ Pediatric neurologist (specialist who treats children with epilepsy) ☐ Adult neurologist (specialist who treats adults with epilepsy) ☐ Pediatrician (specialist who treats children) ☐ Family doctor/ general practitioner (GP) ☐ Emergency room doctor ☐ Urgent care / walk-in clinic doctor ☐ Other _____

Parent Version	AYA Version
No equivalent	7.7. Do you regularly/usually see this doctor (from Question 7.6) for epilepsy- or seizure-related care? ☐ No → What kind of doctor do you see regularly or usually about epilepsy or seizures? ☐ Pediatric neurologist (neurologist who treats children with epilepsy) ☐ Adult neurologist (neurologist who treats adults with epilepsy) ☐ Pediatrician ☐ Family doctor/ general practitioner (GP) ☐ Emergency room doctor ☐ Urgent care / walk-in clinic doctor ☐ Other _____ ☐ I don't see any doctor regularly/usually for epilepsy or seizures. ☐ Yes

Parent Version	AYA Version
6.6. Do you know when your son's/daughter's last seizure was? (It is OK to provide your best guess.) ☐ Less than 6 months ago ☐ 6 months ago to less than 1 year ago ☐ 1 year ago to less than 2 years ☐ 2 years ago to less than 5 years ago ☐ 5 years ago to less than 10 years ago ☐ 10 years ago or more ☐ I don't know	7.8. When was your last seizure? (It is OK to provide your best guess.) ☐ Less than 6 months ago ☐ 6 months ago to less than 1 year ago ☐ 1 year ago to less than 2 years ☐ 2 years ago to less than 5 years ago ☐ 5 years ago to less than 10 years ago ☐ 10 years ago or more ☐ I don't remember
No equivalent	7.9. Are you currently taking any medication(s) to treat epilepsy or seizures? ☐ No → When was the last time you took medication for epilepsy or seizures? ☐ Less than 6 months ago ☐ 6 months to less than 1 year ago ☐ 1 year to less than 2 years ago ☐ More than 2 years ago ☐ I have never taken medication(s) for epilepsy or seizures ☐ I don't remember ☐ Yes

Curriculum Vitae

Name	Alison Sonia Beatrice Dasiewicz
Post-secondary Education and Degrees	Western University London, Ontario, Canada 2014 – 2017 MSc. Epidemiology & Biostatistics University of Waterloo Waterloo, Ontario, Canada 2007 – 2012 BSc. (Honours Co-op)
Honours and Awards	Children's Health Research Institute, Lawson Health Research Institute Graduate Research Fellowship 2015 – 2016 Department of Epidemiology & Biostatistics Western Graduate Research Scholarship 2014 – 2016
Related Work Experience	Teaching Assistant Western University 2015 Research Assistant Western University 2014 – 2016

Presentations:

Alison S.B. Dasiewicz, Guangyong Zou, Carol S. Camfield, Kathy Nixon Speechley. Transition from Paediatric to Adult Health Care: Readiness and Experiences of Canadian Adolescents and Young Adults Diagnosed with Epilepsy in Childhood. *American Epilepsy Society Annual Meeting 2016, George R. Brown Convention Center, Houston, Texas. December 5, 2016.* (Poster presentation)

Alison S.B. Dasiewicz, Guangyong Zou, Carol S. Camfield, Kathy Nixon Speechley. Transition from Paediatric to Adult Health Care: Readiness and Experiences of Canadian Adolescents and Young Adults Diagnosed with Epilepsy in Childhood. *Canadian League Against Epilepsy Scientific Meeting 2016, Hotel Pur, Quebec City, Quebec. October 15, 2016.* (Poster presentation)

Alison S.B. Dasiewicz, Guangyong Zou, Kathy Nixon Speechley. Paediatric to adult health care transition readiness and experiences of Canadian adolescents diagnosed with epilepsy in childhood and their parents. *Canadian Society for Epidemiology and Biostatistics National Student Conference 2016, Brodie Centre – Bannatyne Campus, Winnipeg, Manitoba. June 10, 2016.* (Oral presentation, 1st prize Epidemiology Category)

Alison S.B. Dasiewicz, Guangyong Zou, Kathy Nixon Speechley. Transition from Paediatric to Adult Health Care: Readiness and Experiences of Canadian Adolescents and Young Adults Diagnosed with Epilepsy in Childhood. *Paediatrics Research Day, London Health Sciences Centre – Victoria Hospital, London, Ontario. May 18, 2016.* (Oral presentation, 2nd Prize Basic Sciences Category)

Alison S.B. Dasiewicz, Guangyong Zou, Kathy Nixon Speechley. Transition from Paediatric to Adult Health Care: Readiness and Experiences of Canadian Adolescents and Young Adults Diagnosed with Epilepsy in Childhood. *London Health Research Day, London Convention Centre, London, Ontario. March 29, 2016.* (Poster presentation)