Trial Protocol

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The role of family adaptation in the transition to adulthood for youth with medical complexity: a qualitative case study protocol

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Abstract

Background: For youth with medical complexity and their families, the transition to adulthood is a stressful and disruptive period that is further complicated by the transfer from relatively integrated and familiar pediatric services to more fragmented and unfamiliar adult services. Previous studies report that families feel abandoned, overwhelmed, and unsupported during transition. In order to provide better support to families, we need to understand how families currently manage transition, what supports they need most, and how key factors influence their experiences. The aim of this study is to understand how families of youth with medical complexity adapt to the youth's transition to adulthood and transfer to adult health care, social, and education services, and to explain how contextual factors interact to influence this process.

Methods: Informed by the Life Course Health Development framework, this study will use a qualitative explanatory case study design. The sample will include 10–15 families (1–3 participants per family) of youth with medical complexity (aged 16–30 years) who have lived experience with the youth’s transition to adulthood and transfer to adult services. Data sources will include semi-structured interviews and resources participants identified as supporting the youth’s transition. Reflexive thematic analysis will be used to analyze interview data; directed content analysis will be used for documentary evidence.

Discussion: While previous studies report that families experience significant challenges and emotional toll during transition, it is not known how they adapt to these challenges. Through this study, we will identify what is currently working for families, what they continue to struggle with, and what their most urgent needs are in relation to transition. The anticipated findings will inform both practice solutions and policy changes to address the needs of these families during transition. This study will contribute to the evidence base needed to develop novel solutions and advance policies that will meaningfully support successful transitions for families of youth with medical complexity.

Keywords: adolescents; case study; complex care; qualitative research; transfer; transition; young adults.

Introduction

Children and youth with medical complexity (YMC) are a diverse group of young people who share the following common characteristics: chronic health conditions with functional limitations, substantial health care use, and significant caregiving needs [1]. Their medical and caregiving needs profoundly impact them and their families. They often rely heavily on full-time support from family caregivers for basic activities, such as feeding, bathing, toileting, and managing medications and technological devices (e.g., machines for breathing and feeding). YMC also rely on the support of many health care, social, and education services and providers [2]. Furthermore, the complicated structuring of provincial and regional health care, social, and education systems in Canada adds additional complexity to the care of YMC and their families.
The extensive needs of YMC are reflected in the financial costs to health care systems, as well as in the caregiving burden for their families. While this group comprises just under 1% of the Canadian pediatric population [3], they account for one third of child health care spending in Ontario [2]. These costs do not include out-of-pocket expenses (e.g., for therapies, drugs, private home care) or wages lost to provide informal and unpaid caregiving, with over 50% of these families having a member who stopped working to care for their child [4]. For YMC with neurological impairment, more than half of their caregivers report distress associated with caregiving demands [3].

In Western developed countries, technological and medical advances of the last few decades have helped extend the lifespan of many YMC, leading to a new generation who are surviving into adulthood [5]. Along with the developmental transition to adulthood, their families are now being forced to navigate the transition from relatively familiar pediatric services to more unfamiliar adult services. The transition to adulthood and transfer to adult services is particularly challenging for YMC and their families because of the large number of services, providers, and relationships that will change [2, 6]. When transferring to adult health care, YMC and their families often experience service fragmentation and gaps in care delivery [7]. Rather than a gradual and seamless transition, families often experience an abrupt “transfer” of services that leaves them feeling abandoned, overwhelmed, and unsupported [8, 9].

A literature review about the experiences of YMC and their families with the transition to adulthood describes transition as a highly stressful and disruptive period [9]. Families reported feeling abandoned and alone, and both youth and families had unmet emotional and health care needs. The emotional toll of transition places immense burden not only on youth and their families, but also on their communities and the health care system, as overburdened families will be more challenged to meet caregiving demands. Family caregivers of YMC report that planning for transition requires dedicated time and effort [10, 11], yet they already spend an average of 44–52 h per week providing informal care in the home [3, 12]. Many do this while also managing other responsibilities, such as raising other children, caring for themselves and other family members, and maintaining the home. While previous studies report that families experience stress, abandonment, and uncertainty during transition, it is not known how they adapt to these challenges. This study will explore how families of YMC adapt to challenges and opportunities during transition and will be critical to informing future interventions aimed at supporting families during transition.

Current research and transition interventions often focus on assessing and developing transition readiness by building youth self-management skills [13–15]. However, YMC frequently have significant functional limitations that prevent them from being able to meaningfully manage their own care. Interventions that focus on self-management are often not realistic for YMC and do not adequately address the intensive involvement of families in their care. There is an urgent need for interventions that meet the unique needs and characteristics of this group. The development of tailored, adaptive, and effective interventions needs to be informed by an understanding of: (i) how families currently manage transition; (ii) what supports they need most; and (iii) how key contextual factors influence the transition process. This protocol paper describes a research study that will explore how families of YMC adapt to the youth’s transition to adulthood and transfer to adult services and will seek to explain how contextual factors influence this process.

This paper may be of interest to clinicians, researchers, and health care leaders who want to stay informed about current developments in research on health care transition and transition to adulthood. Beyond transparent reporting of research design and methods, this paper also makes two key contributions to the field of transition research: (1) methodological insights and (2) conceptual clarifications. The methodological strategies described may be of interest to researchers who want to investigate complex, dynamic social phenomena, such as health care transition, as well as those who wish to pursue meaningful family engagement or integrate theory throughout a qualitative case study. Furthermore, the clearly explicated definitions of key theoretical and transition-related concepts found herein may be helpful to researchers and clinicians alike.

**Theoretical framework**

The Life Course Health Development (LCHD) framework, which is underpinned by complex adaptive systems [16], aims to provide a comprehensive description of how health develops over the life span [17]. LCHD was selected as a guiding framework for this study because it: (1) incorporates developmental and temporal concepts that are central to the transition to adulthood, and (2) addresses the multidimensional nature of transition and the influence of environmental factors on youth and family adaptation. The LCHD framework consists of seven interrelated principles:
health development, unfolding, complexity, timing, plasticity, harmony, and thriving.

Applying the principles of the LCHD framework [17], YMC and their families can be understood as individuals that interact dynamically with their physical, natural, and social environments (complexity). Over time, these interactions shape youth and family health development, which unfolds continuously over the life course. During the transition to adulthood, youth and their families face numerous adaptive challenges that are related to time-sensitive processes (timing), such as changes in family roles and health care, social, and education services. These challenges can have anticipated, yet unpredictable and non-linear effects, which are influenced by: prior states of health development (e.g., health stability); the timing and social structuring of environmental exposures (e.g., the timing of transfer); and the harmony of physical, psychological, social, and cultural processes (e.g., the alignment of physical and psychological maturation with social and cultural expectations about adulthood). Plasticity enables youth and families to respond to these challenges and pursue desired activities and goals in adulthood (thriving).

In this study, the LCHD framework will provide structure for the constructs and factors to be investigated. In accordance with the relational ontology of complex adaptive systems, this study will use methods of data collection and analysis that explore context, relationships, patterns, and processes (e.g., interviews, pattern matching, within- and cross-case analyses). The principles of LCHD also informed the development of propositions, research questions, the interview guide, and the initial conceptual framework, which will subsequently guide data analysis and interpretation.

**Objectives**

The aim of this study is to understand how families of YMC adapt to the youth’s transition to adulthood and transfer to adult services, and to explain how contextual factors interact to influence this process.

**Research questions**

1. How do families of YMC adapt to challenges and opportunities posed by the youth’s transition to adulthood and transfer to adult health care, education, and social services?
2. How do contextual factors interact to influence this adaptive process?
3. What are families’ recommendations for support during transition and transfer?
4. How can health professionals support families during transition and transfer?

**Methods**

**Study design**

The transition to adulthood and transfer to adult services for YMC is a contemporary phenomenon situated within temporal, physical, and sociocultural contexts. In health services research, qualitative methods offer a powerful approach for understanding complex human interactions and the influence of multiple contextual factors [18]. Case study methodology is used to investigate contemporary phenomena within their real-world contexts [19]. For these reasons, a qualitative explanatory case study was selected as the best design for this study.

**Patient and public involvement**

Two parent partners with lived experience of the phenomenon of interest are co-investigators on this study. Parent partners were recruited during the study design stage and will be involved in all phases of the research, at varying levels of involvement of their choosing. Parent partners have critically reviewed the research protocol and study materials. Drawing on their lived experiences as parents and advocates of YMC, they have contributed to refining conceptual definitions and methods for sampling, recruitment, and data collection, as well as piloting and refining the interview guide. They will collaborate in ongoing research activities such as recruitment, data analysis, results interpretation, and dissemination. Parent partners will have a central role in knowledge translation; they will: (i) distill key actionable messages, (ii) interpret how this knowledge can be used, and (iii) share these messages with parent communities and lay audiences. At each stage of the project, they will select their roles from a menu of options for involvement (the “Engagement Matrix” can be found in Supplemental Material 1) and will be compensated at a rate of $25 per hour.

**Theoretical propositions**

Theoretical propositions are hypothetical explanations about the potential or likely outcomes of the research, based on empirical literature, existing theories, and personal and professional experiences [19, 20]. Propositions strengthen a case study by increasing its feasibility and creating the basis for generalizable findings that extend beyond the case study. Informed by a literature review on the experiences of YMC and their families with the transition to adulthood [9] and the LCHD framework [17], the following propositions will guide this study:

1. Families of YMC will have the capacity and motivation to adapt to challenges and opportunities posed by transition and will do so to achieve their goals.
2. Each family’s transition experiences will encompass multiple interrelated transitions within the health care, social, and education sectors.
(3) Unique combinations of factors will have non-linear, synergistic, and counteractive effects on family adaptation.
(4) Family recommendations for support will reflect areas where they face the greatest adaptive challenges.

The case under study

The “case” is a real-world phenomenon occurring within a bounded context and is defined based on the original research questions [19, 20]. The case in this study is “family adaptation” in response to the youth’s transition to adulthood and transfer to adult services. Within Yin’s [19] approach to case study, the researcher can choose to conduct a single-case study (single context) or a multiple-case study (multiple contexts). For this study, the context of transition and transfer will be consistent across all units of analysis; thus, a single-case study will be conducted.

Setting and participants (case binding)

“Binding the case” refers to clarifying and setting boundaries around the case in order to keep the study feasible in scope [19, 20]. Case binding is akin to setting inclusion and exclusion criteria in quantitative studies in that it can bind a case by time, place, definition, and other contextual boundaries [19, 20]. One way to bind the study is to clearly define the specific concepts that are used in the case. These definitions, when supported by the literature, will also add to the overall quality of the case study by increasing its construct validity [19]. The following concepts will be used to bind the case: YMC, family, adaptation, transition, and transfer (see Table 1 for definitions).

The spatial binding of the case will be the Canadian province of Ontario. This geographical boundary will allow for homogeneity of the context (i.e., same provincial jurisdiction), thereby allowing for the study of a “typical” case in Ontario. The case will be bound temporally to families’ experiences with any stage of the youth’s transition to adulthood and adult services, including pre-planning, preparation, in process, or completion. These phases are fluid conceptualizations that may change as new data are collected and analyzed. See Figure 1 for a visual representation of the case and case study binding.

Sampling

Sampling in qualitative studies uses purposeful strategies to identify individuals and other data sources that can provide insight on the topic being studied [21]. This study will use a combination of criterion, snowball, and maximum variation sampling strategies.

Criterion sampling seeks to identify participants or information sources that meet predetermined criteria [21]. The sample will include English-speaking families of YMC aged 16–30 years living in Ontario. Because the transfer to adult health care typically occurs at age 18 in Ontario, we will also require that participants have experience with any stage of the youth’s transition process. Specific eligibility criteria for YMC and families are based on the conceptual definitions found in Table 1.

Snowball sampling will allow people both enrolled in and external to the study to identify new participants [21]. This method will be essential to obtain the desired sample size and to access hard-to-reach populations (e.g., those living in rural or remote areas). Snowball sampling will also be used to identify additional participants from the same family. Ideally, multiple participants from each family will be recruited in order to elicit perspectives from a variety of familial roles.

Maximum variation sampling is defined as the selection of participants that differ based on some predetermined criteria [21]. We will aim to explore a wide range of experiences (e.g., positive/negative) and contexts (e.g., urban/rural settings) to gain a broad understanding of transition for YMC and their families in Ontario. After the minimum sample size has been obtained, additional theoretical sampling will continue with the specific aim of identifying missing experiences and testing emerging themes in new contexts [21].

Table 1: Definitions of key concepts binding the case.

<table>
<thead>
<tr>
<th>Concept</th>
<th>Definition</th>
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<tbody>
<tr>
<td>Youth with medical complexity</td>
<td>In this study, youth with medical complexity will be defined as youth or young adults (aged 16–30) who: (1) have a chronic health condition; (2) rely on medical technology (e.g., feeding tube, tracheostomy, mechanical ventilation, long-term IV therapies/nutrition, etc.), OR are completely physically dependent, OR require daily or near daily skilled care/monitoring; and (3) are (or were) cared for by five or more health care providers/teams while in the child health care system. This definition is based on Cohen et al.’s [1] definitional framework and the Provincial Council for Maternal and Child Health’s [28] standard operational definition for children who are the focus of the Complex Care Kids Ontario Strategy.</td>
</tr>
<tr>
<td>Family</td>
<td>Family will be defined as a group of biologically, legally, or socially related people who self-identify as family and play a significant role in the youth’s well-being [29]. Individual participants may include YMC and their biological, adoptive, or foster mothers, fathers, and other family members (e.g., siblings, grandparents, other primary caregivers), aged 16 and older.</td>
</tr>
<tr>
<td>Adaptation</td>
<td>Adaptation will be defined as the process by which families respond to environmental challenges and opportunities in an attempt to achieve their goals [17].</td>
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<tr>
<td>Transition</td>
<td>The transition to adulthood will be defined as a developmental process in which youth and their families experience physical, psychological, social, and cultural shifts associated with becoming an adult.</td>
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<tr>
<td>Transfer</td>
<td>Embedded within the transition to adulthood are multiple transfers to adult health care, education, and social services, which refer to events in which children’s providers “hand over” care to adult providers [30].</td>
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</table>
Documentary evidence will be collected as a secondary data source used to inform, enrich, and supplement the interviews with additional contextual information. Documentary evidence will include tools and resources identified by interview participants that influenced their adaptation to transition. Demographic data such as gender, age, ethnicity, and socioeconomic status will also be collected.

All data will be imported into NVivo 12 qualitative data analysis software. Interviews will be audio-recorded and transcribed verbatim. All identifying information will be removed and names will be replaced with a placeholder or pseudonym. All data will be managed digitally, and files will be encrypted and password protected. LL will collect and manage all data. Data collection began in March 2021 and is expected to continue until the end of 2021.

Data analysis

Data will be analyzed concurrently with recruitment and data collection. Each family will constitute an “embedded unit,” allowing for analysis of how family adaptation differs between families. Data analysis will take a hybrid approach that employs both deductive and inductive strategies. The original theoretical propositions will be used to deductively identify analytic priorities and preliminary codes, while new codes will be generated inductively from the data. This hybrid approach will allow for a focused analysis linked to the original study propositions and research questions, while also allowing for unexpected and serendipitous discoveries to be made. Data will first be analyzed within each family unit and subsequently across family units (“embedded analysis”) [21].

Analysis of interview data will take an interpretive approach, using Braun and Clarke’s [22] reflexive thematic analysis. Braun and Clarke describe six iterative phases to their approach: (1) familiarizing yourself with the data, (2) generating initial codes, (3) generating (initial) themes, (4) reviewing themes, (5) defining and naming themes, and (6) producing the report. Within this method, analysis can be both inductive and driven by a priori theoretical propositions [22], which aligns with the general analytic strategy of this study.

In contrast, analysis of documentary data, which may come in a wide variety of formats, will take a more literal approach, using directed content analysis [23]. Qualitative content analysis is used to analyze textual data (in verbal, print, or electronic form) and focuses on the characteristics of language, while paying close attention to the content and contextual meaning of the data [23]. “Directed” content analysis uses existing theory to guide analysis and aligns with the use of theoretical propositions in case study. Scope and feasibility are important considerations when analyzing documents, which may produce exceedingly large amounts of data. To avoid this situation, boundaries for the focused analysis of documentary evidence will be determined by theoretical propositions and interview findings. Following preliminary data analysis, themes from each data source will be converged for final data interpretation using case study-specific techniques of pattern matching, explanation building, time-series analysis, and logic models [19]. See Figure 2 for the methods flow chart.

Rigour

Yin [19] proposes four quality criteria that are specific to case study: construct validity, internal validity, external validity, and reliability.
Construct validity refers to the degree to which a case study’s measures reflect the concepts being studied. Internal validity refers to the strength of inferences made about causal relationships, while external validity pertains to the degree to which the study findings can be analytically generalized (i.e., applied to theoretically analogous situations beyond the study). Lastly, reliability concerns the degree to which the study findings can be replicated and can be judged by how explicitly the study procedures have been documented.

Construct validity will be enhanced through data triangulation (convergence of multiple data sources), clear conceptual definitions, and member checking (validation of findings with participants). Member checking will be performed by sharing preliminary findings/themes with all interested participants through mail or email and having them provide feedback. Multiple levels of analysis and examination of rival explanations will improve internal validity. Peer auditing will be done by having the study team and parent research partners provide feedback on interim interpretations, which will enhance both construct and internal validity. Use of a priori theory and study propositions to guide the study will contribute to both construct and external validity. Reliability will be ensured through detailed documentation of study procedures (including an audit trail), systematic data management, and linking of findings to the original study propositions.

Lastly, engaging in reflexivity is a widely accepted practice of “good” qualitative research. In case study research, reflexivity can help ensure the research remains inductive and is not too driven by the conceptual framework and propositions [20]. LL, who is a registered nurse who works in an acute pediatric medicine and complex care setting, will maintain a reflexive journal to keep an audit trail of the moments when researcher subjectivity influences particular study decisions or interpretations.

Ethical considerations

This study meets ethical standards set out by the Declaration of Helsinki, as well as the Tri-Council Policy Statement: Ethical Conduct for Research Involving Humans, based on the principles of respect for persons, concern for welfare, and justice [26]. This study received final ethical approval from the Hamilton Integrated Research Ethics Board (#1118) in October 2020. Participants will be provided with a written information sheet describing the study aims, procedures, risks and benefits. Informed consent will be obtained electronically and all efforts will be made to protect the privacy and confidentiality of all participants. A $50 honorarium will be provided for each interview completed and the results of the research will be shared with interested participants.

Dissemination

Dissemination of study findings will target specific audiences, including clinicians, families, researchers, and policy makers. Findings will be shared in open-access peer-reviewed journals and national and international conferences (e.g., Children’s Healthcare Canada). Knowledge translation strategies will also include a webinar, infographic, and video. This video will be publicly available online and shared via social media. It will offer ideas to clinicians for developing tools to support and empower families in their adaptation to transition and raise awareness of key issues among policy makers and the public. The study findings will also be submitted to and housed, open access, on the McMaster University Libraries MacSphere Institutional Repository.

Discussion

This study is novel in that other studies on transition within this population have not explored how families manage and adapt to transition. Instead, previous studies have focused on narrative experiences and barriers and facilitators, which are not always linked to much-needed
solutions. While previous research has certainly been informative and valuable in creating an evidence base, the treatment of barriers and facilitators as static factors with unidirectional effects oversimplifies the complex nature of the problem. Informed by a complex adaptive systems approach, this study is directed toward the development of solutions that will be responsive to the dynamic and complex interactions of contemporary conditions.

Furthermore, to our knowledge, previous studies on transition for this group have not engaged people with lived experience as partners throughout the research process, including at the early stages of study design. There are many benefits of engaging people with lived experience in research, including: increased relevance and quality of research; improved translation of research into policy and practice; increased accountability and transparency of research; and the potential for new insights and discoveries [25]. The involvement of parent research partners in all phases of this study, from design to execution and knowledge translation, will provide a unique and valuable contribution to the field. It is anticipated that their involvement will lead to revelatory questions, interpretations, and insights that would otherwise remain unexplored.

Health care, social, and education systems face mounting demands to create effective evidence-based policies, informed by the perspectives of key stakeholders, that will meet the needs of this population. This study will deliver a unique contribution to the evidence base required to advance policies that will support the successful transition to adulthood and transfer to adult services for YMC and their families. Furthermore, the complexity and fragile health of this population means that they are most likely to experience adverse outcomes as a result of gaps in the system. In this way, they expose the underlying system flaws, and their health can be seen as an indicator of the overall function of our health care, education, and social systems; therefore, efforts to improve the health of YMC and their families have the potential to improve these systems of care for others who access these services.

Limitations

In Canada, health care, education, and social services are administered regionally and vary based on the needs and resources of different communities. It is not feasible to sample from every community in Canada. Only families living in Ontario will be sampled, and even then, not all regional jurisdictions will be represented. As a result, the findings from this research may be limited in addressing the needs of families whose contexts differ greatly from those included in the sample. However, it is also anticipated that the findings will reveal essential aspects of family adaptation that are common across various contexts, settings, and geographies. These findings will be very helpful in understanding how families of YMC adapt to transition and how they might respond to different interventions in the future.

Another limitation is that some contextual factors influencing family adaptation will not be modifiable. As it is predicted that some factors will interact in non-linear, synergistic, or counteractive ways, only targeting certain modifiable factors may not have the anticipated effect that is desired. In this study, it will be important to identify and differentiate between modifiable and non-modifiable factors to begin to understand the dynamic relationships at play. Future research beyond the scope of this study will be required to fully explicate these relationships by testing interventions delivered in real world settings.

Conclusions

There is an urgent need to better understand the priorities, gaps, and adaptive strategies related to transitioning YMC, from the perspectives of their families. This study will address a critical gap in the Canadian health care system. Improving the transition from child to adult services has been identified as a priority internationally, by key organizations such as the Canadian Pediatric Society [26]; the American Academy of Pediatrics, American Academy of Family Physicians, and American College of Physicians [6]; and the National Institute for Health and Care Excellence in the United Kingdom [27]. Novel insights from this study will inform the development of timely, tailored, and adaptive interventions that will capitalize on family and community strengths, and provide support where families need it most. Furthermore, by explicating the dynamic relationships between contextual factors influencing transition, future interventions will be able to target synergistic combinations of factors to optimize the process and outcomes of transition. The findings of this study will be crucial to building a future program of research aimed at developing and evaluating effective and meaningful interventions to support YMC and their families during transition.

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Author contributions: All authors contributed to study conception and design. LL developed the initial draft of the manuscript. All authors were involved in multiple iterations of critically revising the protocol and manuscript. All authors have accepted responsibility for the entire content of this manuscript and approved its submission.

Competing interests: Authors state no conflict of interest.

Informed consent: Informed consent was obtained from all individuals included in this study.

Ethical approval: The research related to human use has complied with all the relevant national regulations and institutional policies and was performed in accordance with the tenets of the Helsinki Declaration, and has been approved by the authors’ Institutional Review Board (Hamilton Integrated Research Ethics Board, 11184).

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