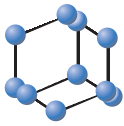


## REVIEW ARTICLE


**BENTHAM  
SCIENCE**

# Changes in Care- A Systematic Scoping Review of Transitions for Children with Medical Complexities



Tara Mantler<sup>1,\*</sup>, Kimberley T. Jackson<sup>2</sup>, Jessi Baer<sup>2</sup>, Jenna White<sup>2</sup>, Bridget Ache<sup>2</sup>, Katie Shillington<sup>1</sup> and Nokuzola Ncube<sup>1</sup>

<sup>1</sup>School of Health Studies, Faculty of Health Sciences, Western University, London, Canada; <sup>2</sup>Arthur Labatt Family School of Nursing, Faculty of Health Sciences, Western University, London, Canada

**Abstract: Background:** Children with medical complexity (CMC) and their parents are affected physically and mentally during transitions in care. Coordinated models of care show promise in improving health outcomes.

**Objective:** The purpose of this scoping review was to examine research related to CMC and their parents and transitions in care. The aim was 3-fold: (1) to examine the extent, range, and nature of research activity related to the impact of transitions on physical and mental health for CMC and their parents; (2) to summarize and disseminate research findings for key knowledge users; and (3) to identify research gaps in the existing literature to inform future studies.

**Methods:** Twenty-three sources were identified through database searches and five articles met the inclusion criteria of CMC (multi-organ involvement or technology-dependent) (or parents of CMC) transitioning from hospital to alternate levels of care where outcome measures were physical or mental health-related.

**Results:** Numerical analysis revealed substantial variation in methodological approaches and outcome measures. Content analysis revealed two themes for parents of CMC during this transition: (1) emotional distress, and (2) high expectations; and three themes for CMC: (1) improved health, (2) changes in emotion, and (3) disrupted relationships.

**Conclusion:** The findings from this scoping review reveal for parents, transitions in care are fraught with emotional distress and high expectations; and for CMC there are improvements in quality of life and emotional health post-hospital to home transitions when collaborative models of care are available. This review serves as an early attempt to summarize the literature and demonstrate a need for further research.

**Keywords:** Children with medical complexity, CMC, transitions in care, physical health, mental health, caregivers.

## 1. INTRODUCTION

In Ontario alone, there are over 15,000 children with medical complexity (CMC) - defined as children with chronic conditions with multiple organ system involvement and/or who are technology-dependent [1, 2]. While CMC currently represents less than 1% of pediatric patients, this number is growing due to medical advancements which are creating a new generation of survivors of childhood-onset diseases/injuries [3]. To illustrate, from 1993 to 2002, the infant mortality rate among extremely low gestational age pre-term infants decreased from 256 per 1000 live births to 114 per 1000; during the same time, the rates of cerebral

palsy (a condition often meeting the criteria for CMC) increased from 44.4 per 1000 to 100 per 1000 [4]. Pre-term mortality has continued to decrease over the last decade and morbidity has been on the rise contributing to increased numbers of CMC [5].

CMC, in Canada, utilizes approximately 1/3 of pediatric healthcare spending, or \$419 million annually, and 41% of pediatric hospital expenditures [3, 6]. The pediatric hospital expenditures only convey part of the story. Additional costs are borne by CMC and their parents stemming from their often numerous and fragmented interactions with systems including poor quality of life [7], dissatisfaction with care [8-10], and poor health outcomes [11, 12]. Parents report a lack of support in the community [6, 13] leading to frequent, unnecessary hospital readmissions [14]. Re-hospitalization places immense strain on the parents, contributing to familial

\*Address correspondence to this author at the School of Health Studies, Faculty of Health Sciences, Western University, London, Canada; Tel: 519-661-2111; E-mail: [tara.mantler@uwo.ca](mailto:tara.mantler@uwo.ca)

### ARTICLE HISTORY

Received: August 14, 2019  
Revised: September 05, 2019  
Accepted: November 26, 2019

DOI:  
10.2174/1573396316666191218102734



CrossMark

and support separation (impacting family bonding, cohesiveness, and roles) [15], increased financial strain (e.g., lost wages due to work absenteeism and costs associated with care provision for other children) [16, 17], and physical and psychological effects such as muscle aches, fatigue, difficulty concentrating, depression, and/or anxiety [18].

Transitions are a frequently occurring event for CMC and can be sometimes attributed to fragmented and disjointed healthcare systems, particularly as care moves from the hospital to the community [15, 19]. Transitions, which are defined as a movement between two relatively stable periods [20], occur across the continuum of care from birth to adulthood, within and between settings, and between providers [19]. These are critical junctures for CMC as they lead to parental and child stress and anxiety, increased responsibilities, new provider relationships, and potential gaps in care [15, 19].

The difficulties faced by CMC and their parents in terms of fragmented care and increased transition frequency are compounded by inadequate financial support. In the Canadian context, spending on services to support CMC is often divided between several ministries (health, social services, and education) which leads to disjointed funding and planning [21]. This leaves CMC vulnerable to insufficient care coordination [21], medical errors [12], adverse physical and mental health outcomes for caregivers [22], and financial burden for parents [9]. Care coordination for CMC is extremely challenging due to the lack of system or service level integration and limited by the vast continuum of care required and the episodic nature of the care received (i.e., a different care provider for each issue [23, 24]). This can contribute to perceptions of care that is disjointed and of poor quality [8-10].

The need for integration has been highlighted frequently in the literature as imperative to assist CMC and their parents in navigating the labyrinth of care and services [8, 25]. Despite calls for integration, efforts have failed and resulted in suboptimal, and oftentimes, unsuccessful transitions of care from the hospital, ultimately leading to re-admission [3, 8]. To that end, the goal of this systematic scoping review was to examine research activity related to CMC and their parents and transitions in care. Specifically, the purpose was 3-fold: 1) to examine the extent, range, and nature of research activity related to the impact of transitions on physical and mental health for CMC and their parents; 2) to summarize and disseminate research findings for key knowledge users; and 3) to identify research gaps in the existing literature to inform future studies.

## 2. METHODS

### 2.1. Criteria for Considering Studies for Review

Studies describing the experiences of CMC or their parents and the transition in care were included in this systematic scoping review. The following inclusion criteria were used: (1) target population was CMC (multi-organ involvement or technology-dependent) or their parents; and (2) reported physical or mental health outcomes associated with the transition from hospital to home care. For the purpose of this scoping review, CMC was operationalized as any child

with multi-organ system involvement and/or technology-dependent. Studies were excluded if not all children had medical complexity.

### 2.2. Search Strategy for Identification of Studies

Electronic databases (PubMed, CINAHL, Scopus and PsychINFO) were accessed from January to April 2019 searching the following three concepts: CMC, transitions in care, and health outcomes (see appendix A). No publication date parameters were set, and the search was restricted to articles written in English. The search yielded 23 articles; articles were excluded if: (1) they did not align with the inclusion criteria based on titles and abstracts ( $n=13$ ), or (2) they were duplicated articles ( $n=1$ ). Next, the remaining eligible articles ( $n=9$ ) were screened in full, and references were searched to determine if there were any additional eligible articles. Five articles were included in this review. (Fig. 1). Appendix B provides a list of excluded studies and the rationale for exclusion.

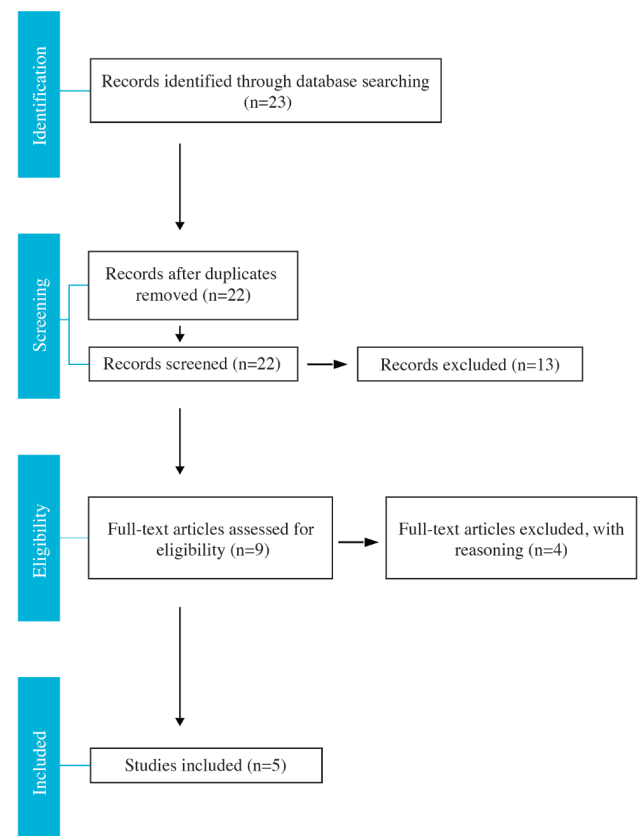


Fig. (1). Scoping review.

### 2.3. Data Extraction

Data were extracted into a customized table created by the research team (see Table 1). A narrative review [26] was conducted which included extracting information such as author(s), year of publication, country of origin, aim and purpose, sample size, demographics, methodology, intervention description, mental health outcomes, and physical health outcomes. No modification of the original data was performed in this review.

### 2.4. Bias Susceptibility

To assess bias, the Strengthening the Reporting of Observational Studies in Epidemiology (STROBE) was used [27]. The following variables were evaluated: confounding variable control, external validity, eligibility criteria, sample size, and losses (see Appendix C). All studies provided defi-

nitions for CMC, while four studies completely defined the transition being analyzed [28-32]. Physical health measures were detailed in two of the studies by measuring the hospitalization rates of participants both before and after the intervention [28-32]. Of the five articles, only one article failed to measure mental health outcomes [28-32].

**Table 1. Data extraction.**

Author(s) Year Country of Origin	Aims/Purpose, Sample Size, Demographics	Methodology	Outcomes
<b>Kids</b>			
Palfrey, Sofis, Davidson, Liu, Freeman, and Ganz  2004  Boston, Massachusetts, USA	The goal of this study was (1) to characterize CSHCN; (2) to examine parental satisfaction of the PACC intervention; (3) to examine the impact of hospitalizations and emergency department episodes; and (4) to assess the impact on parental work days lost and children’s school days lost for CSHCN before and during intervention.  Sample size: N=150 (T1) N at 2 years = 117 (T2)  Age: 0-5: 55.6% 6-18: 44.4%  Gender: Male: 66.7% Female: 33.3%  Ethnicity: White: 62.4% Non-White 37.6%	Quantitative methods: Pearson ( $x^2$ )  Longitudinal  Data were collected at baseline (T1) and 2 years post-intervention (T2)  To assess differences between groups: $x^2$ test for categorical data and independent sample t-test for continuous data.  To assess the differences across time periods: McNemar test for categorical data and paired t-test for continuous data.  Intervention:  Implementation of a community-based medical home model for children with complex health care needs  Comparator: N  Duration: 2 years	Measure: Children’s hospitalization rates  Findings:  There was a significant decrease in emergency hospitalizations of children (58% prior to PACC vs. 43% after PACC intervention)
Kirk  2008  North-West England, United Kingdom	The goal of this study was to analyze how young people with complex healthcare needs experienced different transitions  Sample size: N=28  Gender: Male: 17 (61%) Female: 11 (39%)  Age: 9 ages 8-11 11 ages 12-15 8 ages 16-19	In-depth qualitative interviews  Inductive coding, thematic coding analysis using Grounded Theory principles  Time points for data collection: 1 interview  Intervention: N  Comparator: N	Going into the unknown, going into a different world, disrupted relationships and ways of working  Young people could feel reluctant to become involved in therapies and procedures; young peoples’ impairments did not prevent them from involvement in health-related decision making; parents continued to negotiate service support and liaise with professionals on their child’s behalf

(Table 1) Contd....

Author(s) Year Country of Origin	Aims/Purpose, Sample Size, Demographics	Methodology	Outcomes
<p>Cohen, Lacombe-Duncan, Spalding, MacInnis, Nicholas, Narayanan, Gordon, Margolis, and Friedman 2012 Toronto, Ontario, Canada (SickKids) and Brampton, Ontario, Canada (Brampton Civic Hospital) and Orillia, Ontario, Canada (Orillia Soldier's Memorial Hospital)</p>	<p>The goal of this study was to evaluate the outcomes of community-based complex care clinics integrated with a tertiary care facility</p> <p>Sample size: N=81</p> <p>Gender: Male: 64% Female: 36%</p> <p>Mean age (SD): 5.8 (4.7)</p>	<p>Mixed methods Longitudinal</p> <p>Caregiver's participated in semi-structured interviews</p> <p>Data were collected at baseline, 6 months, and 12 months</p> <p>Change in summary score over the period of follow up were compared using non-parametric analysis, the Friedman test</p> <p>Content analysis informed by the Institute for Health Care Improvement framework</p> <p>Intervention: Clinics used a co-management model with primary care providers and tertiary care affiliated nurse practitioners. Clinics were conducted weekly at each site and the focus of the visits was on care coordination, complex symptom management, and goal setting.</p> <p>Comparator: N</p>	<p>Measure: The average number of children's inpatient hospital days</p> <p>Findings: There was a significant decrease in the overall inpatient hospital days (<math>p = .0005</math>). Mean (SD) of 11.7 (21.4) pre intervention and 3.7 (11.3) post intervention.</p> <p>Measure: Health-related quality of life (QOL)-the PedsQLTM, a widely used generic measure of HR-QOL, was used in children <math>\geq 2</math> years</p> <p>Findings: Child quality of life improved between baseline and 6 months in two of five PedsQL domains [Social domain (<math>p=.01</math>) and Emotional domain (<math>p=.003</math>)]</p> <p>Measure: Health-related QOL- the Caregiver Priorities &amp; Child Health Index of Life with Disabilities (CPCHILD) measures the ease of care, comfort, health, and well-being of children with severe disabilities and was administered for children <math>\geq 1</math> year</p> <p>Findings: Child QOL improved between baseline and 1 year in two domains [Health Standardization Section (<math>p=.04</math>) and Comfort and Emotions (<math>p=.03</math>)], but the total CCHILD score decreased between baseline and 1 year (<math>p=.003</math>)</p> <p>3. Measure: Health related QOL (SF-36)</p> <p>Findings: Parental quality of life did not significantly change over the course of the study in any of the eight SF-36 domains</p>
<b>Parents</b>			
<p>Kirk and Glendinning 2003 North England, United Kingdom</p>	<p>To explore the experiences of families caring at home for a technology-dependent child; and to identify perceived problems and good practice in the purchasing, delivery, and coordination of services.</p> <p>Sample size: N=24 families N=3 hospitals N=38 healthcare professionals (purposive sampling)</p> <p>Families Mothers: 23 Fathers: 10 N=33</p>	<p>In-depth qualitative interviews (constant comparative)</p> <p>Inductive coding &amp; systemic comparison</p> <p>Time points for data collection: 1 interview</p>	<p>Social activity was restricted; required to do complex procedures and as a result anxiety and stress were reported (<i>i.e.</i>, exhaustion); over reliance on parents to communicate and coordinate services; parents felt the emotional aspects of caregiving were neglected by professionals</p>
<p>Manhas and Mitchell 2012 Calgary, Alberta, Canada</p>	<p>To contextualize the experience of transition from hospital to home care by examining the perceptions of 26 involved adults</p> <p>N=3 family members</p>	<p>Qualitative methods; in person and phone semi-structured interviews</p> <p>Case study (2 children)</p> <p>Qualitative thematic analysis of interview transcripts</p> <p>Time points for data collection: multiple (not specified)</p> <p>Intervention: N Comparator: N</p>	<p>Parental theme – key informants (not children); Transition focused on shifting considerable responsibility to mothers, which challenged mothers with demands and expectations of extraordinariness - leading to isolation; Transition was filled with loss, which challenged families with concomitant grief and uncertainty</p>

## 2.5. Collating, Summarizing, and Reporting of Results

Numerical and conceptual analyses were undertaken for this review [26]. Both analyses were undertaken independently by two graduate level research assistants, and subsequently, results were compared. The numerical analysis identifies the extent, nature, and distribution of studies included in this review by tabulating findings across all included studies. Initially, for the concept analysis, an inductive content analysis was undertaken in which emergent themes based on the totality of the research were independently identified by two graduate level researchers. Subsequently, the two independent graduate researchers compared emergent themes and in an iterative process supported by the co-investigators, were able to establish consensus on the emerging themes. All emerging themes were agreed upon by the graduate researchers and co-investigators.

## 3. RESULTS

### 3.1. Numerical Analysis

#### 3.1.1. Study Design

Of the five studies, one used a quantitative research approach [30], three utilized a qualitative approach [28, 29, 31], and one mixed-methods [32]. With respect to the study design, three of the studies were longitudinal. Of these longitudinal studies, one was retrospective (2 years post-intervention) [30], another collected data at three specific time periods (pre-, mid-, and post-intervention) [32], and the other was a case study which utilized semi-structured interviews but did not specify the frequency or duration of the data collection [31]. Two of the studies used cross-sectional data collection *via* in-depth interviews [29, 30]. There were no control groups used in any of the studies.

#### 3.1.2. Geographic Location and Patient Population

Two of the five studies were conducted in Canada [31, 32], two in the United Kingdom [29, 30], and one in the United States [30]. Three of the studies focused on CMC participated [28, 29, 32]. Specifically, 117 children were studied by Palfrey [30], of which 65 (55.6%) were aged zero to five years. Additionally, the mean age of the 81 children studied by Cohen [32] was approximately six years, and Kirk [28] studied 28 children between eight to 19 years old. Most children included in these three studies were between the ages of zero to six years old.

In addition, the study conducted by Palfrey [30] was the only study to explicitly reference ethnicity and noted that most of the children were white (62.4%) compared to non-white (37.6%). Cohen [32] noted the suburban hospital served a population comprised of 60% visible minorities (most commonly South Asian or Afro-Caribbean descent) while the rural hospital served a cohort of mainly Caucasians; however, ethnicity is not reported in the study sample. Only one study collected data on population density [31], where they stated transitions were from an urban specialist hospital to a small city regional hospital and then to a rural home. Only two of the five studies reported socioeconomic status with both noting approximately equal representation [28, 32].

Among the studies focused on parents of CMC, 36 family members participated [30, 31]. Specifically, 33 parents were studied by Kirk and Glendinning [29], of which 10 were male and 23 were female. In contrast, Manhas and Mitchell [31] did not specify parents, rather they stated that three family members were included in their study, two of which were the case studies' mothers. Neither of the two studies specified the ages of the participating family members.

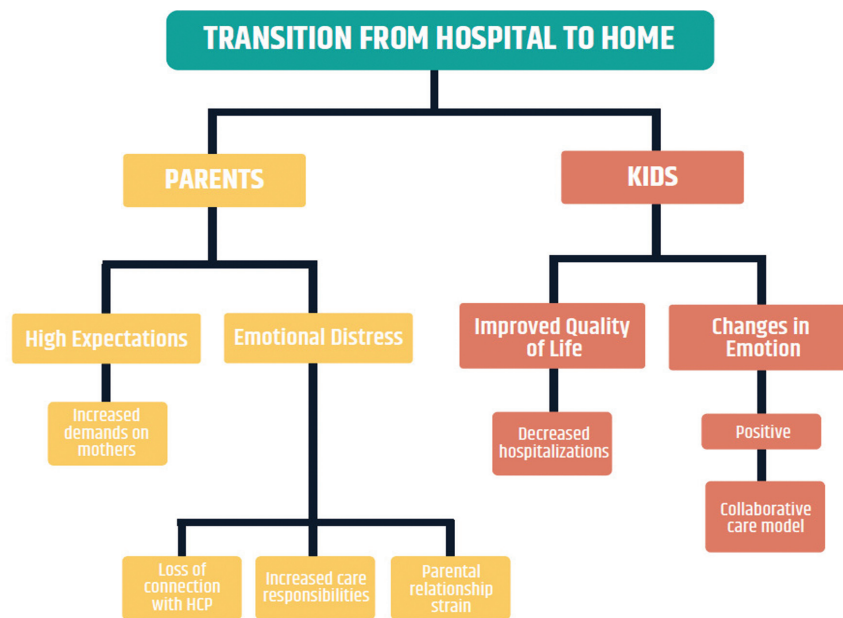
#### 3.1.3. Interventions

Two of the studies that examined children focused on integration at the community-level to provide comprehensive care for children [28, 32]. One study integrated community-based complex care clinics with a tertiary care center using a co-management model and evaluated the outcomes for CMC [32]. Clinics were conducted weekly and care plans were developed on-site by the Nurse Practitioner (NP), in partnership with parents, with the focus of care coordination, symptom management, and goal setting [32]. The second study created an integrated system of care for CMC (built upon principles of family-centered care and the medical home model (*i.e.* preventative care, assurance of ambulatory and inpatient care 24 hours per day, continuity of care, appropriate use of referrals and specialty consultations, interactions between health care, school, and community agencies, and a central database that contains all pertinent health information) [30]. The final study examining CMC used in-depth interviews with the experience of transition from pediatric to adult services and from parental care to self-care [28].

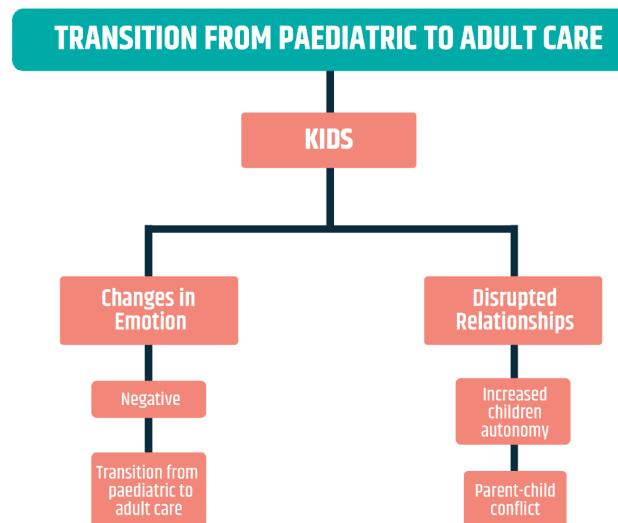
Of the studies that explored the parents of CMC, both focused on the parental perspectives of children during transition periods utilizing in-depth [29] and semi-structured interviews [31]. Manhas and Mitchell [31] specifically examined parents' perspectives on the transition from hospital to home care in ventilator-dependent children. Kirk and Glendinning [29] examined transition on a broader scale by exploring the experiences of parents identifying the perceived barriers and supports surrounding service delivery and coordination.

### 3.2. Conceptual Analysis of Outcome Measures

Analysis of these studies revealed findings that were organized into two themes parental themes and three CMC themes. For parents, the emergent themes were: (1) emotional distress - which addresses the emotional challenges such as anxiety and stress that parents face when caring for their children at home, and (2) high expectations - which addresses the increased demand and reliance on parents to coordinate and provide care. The themes for the children included: (1) improved health - which addresses a decrease in hospitalizations and improved quality of life when transition is coordinated by a care team, (2) disrupted relationships - which addresses the changes in the relationships children have with their parents and healthcare providers as the CMC become more independent with managing their care, and (3) changes in emotions - which addresses the CMC emotions during the transition process (See Figs. 2 and 3). Each theme will be discussed in turn.



**Fig. (2).** Transitions from hospital to home. (A higher resolution / colour version of this figure is available in the electronic copy of the article).



**Fig. (3).** Transitions from paediatric to adult care. (A higher resolution / colour version of this figure is available in the electronic copy of the article).

### 3.2.1. Parents - Emotional Distress

The process of transition from hospital to home-based care resulted in increased distress for parents. Mahan and Mitchell [31] identified during the transition, parents lost their connections with familiar healthcare providers in the hospitals and had to create new relations with home care providers which lead to feelings of grief and uncertainty. They also identified increased responsibility as it was challenging to find and maintain at-home caregivers. This increased the responsibility of the parents leading to both stress and exhaustion. The increased responsibility was echoed by Kirk and Glendinning [29] who found that parents felt healthcare providers placed too much emphasis on learning technical caregiving skills while the emotional aspects of care for CMC were neglected. Parents felt obligated to learn and perform technical tasks (*i.e.* changing tracheostomy tubes, administering intravenous infusions, *etc.*) which at

times were painful to the CMC causing parental distress. Emotional distress also stemmed from increased strain within the parental relationship. Parents reported that their social activities were restricted due to the large equipment they were required to carry around with their children and the need to administer scheduled treatments and therapies [29]. These demanding tasks placed strain on the relationship between the parents due to their inability to spend time together [29].

### 3.2.2. Parents - High Expectations

Parents felt there were considerable expectations placed upon them in terms of care coordination. Parents reported feeling like their own service coordinators because of the over-reliance on parents to communicate information between the various healthcare providers [29]. Transitions increased the demands on the mothers especially, who had the bulk of the responsibility [31]. Manhas and Mitchell [31 pp.

228] reported in their findings that, “transition focused on shifting considerable responsibility to mothers, which challenged them with demands and expectations of extraordinariness.”

### 3.2.3. Children - Improved Quality of Life

Improved quality of life when transitions were effectively supported between hospital and home care was a reoccurring theme. Palfrey [30] reported that the hospitalization rate of CMC ( $n=117$ ) dropped from 58% to 43% ( $p<.01$ ) one year after the implementation of a medical home. Cohen [32] showed that there was a significant decrease ( $p=0.005$ ,  $n=81$ ) in the overall inpatient hospital days from 11.7 (21.4) [Mean (SD)] to 3.7 (11.3) [Mean (SD)] when community based complex care clinics were integrated with tertiary care hospitals. In these two studies, NPs played a large role in the coordination of care between the acute care and community settings which contributed to fewer hospital admissions [28, 32], and reduced the burden on the parents [30].

Beyond just a reduction in hospitalization, Cohen [32] reported improved quality of life (QOL) for CMC between baseline and at six months in the social ( $p=.01$ ) and emotional domains ( $p=0.003$ ). There was also an improvement in the CMC QOL between baseline and one year in the Health Standardization ( $p=0.04$ ) and the Comfort and Emotions ( $p=.04$ ) sections of the CPCHILD tool despite an overall decrease in the total CPCHILD score. In general, the hospitalization rate of CMC decreased while QOL improved.

### 3.2.4. Children - Disrupted Relationships

In the study by Kirk [28], CMC reported how their relationships changed with their parents and healthcare providers as they transitioned from pediatric to adult healthcare; CMC was expected to be more independent with their care and decision making. Transfer from parental to self-care was influenced by their own readiness to accept responsibility, which at times created conflicts between the children and their parents [28]. One participant in Kirk [28] described her interaction with healthcare providers in adult services with regard to taking her medication. She reported the healthcare provider informed her of the benefit of taking her medication but the decision on whether to take the medication or not was solely hers [28]. In contrast, in the pediatric service, this communication was between the healthcare provider and the parent [28]. Even though CMC became more involved with their care, parents still continued to play major roles in negotiating support services and coordinating care with healthcare providers which contributed to a disruption in the relationship [28].

### 3.2.5. Children - Change in Emotions

CMC experienced positive and negative emotional changes during the transition. Positive emotional changes were reported by Cohen [32] when a collaborative model of care was utilized. The child QOL improved between baseline and 6 months in the Emotional domain ( $p=.003$ ) of the PedsQL tool used for children  $\geq 2$  years. Similarly, this study also showed an improved QOL in the Comfort and Emotions domains ( $p=.03$ ) of the CPCHILD tool between baseline and 1 year for children  $\geq 1$  year, even though there was a decrease in the total score [32].

Regarding the transition between pediatric and adult care, CMC expressed negative emotions. For example, they described pediatric services as protective, warm, bright, and colorful environments [28]. In contrast, they described adult environments as threatening, depersonalizing, and gloomy [28]. CMC also described their experience as shocking, filled with uncertainty and unhappiness, and like it was moving into a different world [28]. Participants reported feelings of loss as they had to end their relationships with their pediatric healthcare providers and felt uncertainty in having to build new relationships with adult healthcare providers [28]. In addition, CMC at times felt reluctant in managing their therapies due to unpleasant sensations such as smell [28]. This reluctance to take control of their health care contributed to a disrupted relationship with their parents.

## 4. DISCUSSION

The purpose of this scoping review was to examine the extent, range, and nature of research activity related to the impact of transitions on physical and mental health for CMC and their parents. The major findings of this scoping review revealed that for parents, transitions in care are fraught with emotional distress and high expectations; and for CMC there are improvements in quality of life and emotional health post-hospital to home transitions when collaborative models of care are available. However, transitions between pediatric and adult care for CMC result in disrupted child-parent relationships as well as negative emotions for CMC in terms of interactions with the adult healthcare system.

Emotional distress and high expectations that parents described in this scoping review are echoed in other studies where parents navigated multiple, and often fragmented, healthcare systems. Kelly [9], in a case study application of the medical home model, reported a large contributor of stress for parents is the fragmented coordination of multiple service systems. Furthermore, the breadth of demanding healthcare needs of CMC is also associated with stress on caregivers [33]. Raina [33] applied structural equation modeling with data from a large cohort ( $n=68$ ) of caregivers of children with cerebral palsy and found that caregiving demands and child behavior (both directly and indirectly through their effects on self-perception and family function) strongly influenced the physical and psychological health of the caregiver. Similarly, Lach [22] through a survey-based study (conducted jointly by Statistics Canada and Human Resources Social Development Canada) of caregivers of children with neurodevelopmental disorders found that caregivers more frequently report poorer health (*e.g.*, increased frequency of chronic conditions such as asthma, arthritis, and migraines) and psychosocial problems (*e.g.*, poorer social support and activity limitations). In addition, poor QOL and dissatisfaction of care were reported by Russel and Simon [7], in a review of the CMC literature, and Cohen [8], during the development of an integrated complex care model. These findings collectively support our identified themes of emotional distress and high expectations.

The theme of improvements in child QOL post coordinated care model implementation was also revealed in other studies. The process of being admitted to the hospital is a severe disruption in the life of a child and is typically anxi-



ety-provoking and even traumatic time for children as they are separated from the security and stability of the home and placed in the foreign hospital environment [34]. Children are particularly vulnerable during this transition due to their dependence on others, their cognitive and emotional limitations, and the resulting disturbance/distortion the hospital experience can have on their developmental processes (e.g., impacting their psychosocial development and their level of physical activity) [34]. Research indicates that the implications of hospitalization (i.e., behavioral, developmental, or psychological difficulties) gradually reduce post-discharge if the transition home is successful [34]. However, chronically ill children or CMC who are continually readmitted to the hospital suffer from additional anxiety and long-term adjustment problems as they fall behind normal development and lack the stability of a routine [34]. Therefore, consistent with the findings of this review, prolonged admittance in hospital and repeated transitions into hospital can have a negative effect on a child's overall QOL.

Research evidence supports the difficult transfer from pediatric to adult healthcare described. In Hopper's [35] case study analysis, a 27-year-old woman with severe asthma and sickle cell disease identified several challenges of her experience transitioning from pediatric to adult healthcare, some of which included: a lack of understanding of the adult healthcare system and the different expectations of an adult patient, lack of coordination among adult physicians, and a general feeling that the healthcare system no longer understood her needs. This patient also referenced being impacted by two opposite assumptions: 1) that because she was young she "did not know about her disease" and 2) that because she was an adult she should be independent from her parents, "the adult providers wanted my mom to step back, but transition was a very hard time for me, and I needed my mom to help out" [37 pp. 252, 251]. Hopper's [35] results substantiate this review's finding of disturbed relationships as young adults negotiate the transition to adult care, their new role, and the adapting roles of their caregivers in this new context. Another study illustrating transition difficulty was Rutishauser's [36] cross-sectional study comparing pre- and post-transfer young adults' identified barriers to a successful transition into adult healthcare. Rutishauser [36] found that the most important barriers to a successful transition to adult healthcare were: feeling at ease with the pediatrician, negative emotions regarding the transfer, such as anxiety, and lack of information [36]. This study echoes the theme of this review; negative emotions regarding transfers in the absence of collaborative care, thus substantiating this finding.

Unfortunately, literature evaluating parents and CMC during the transition period from hospital to alternate levels of care is scant; therefore, comparable findings are difficult to obtain. Of note, a Cochrane systematic review [37] is being completed which may provide insight into outcome measures of concern to this scoping review including CMC and parental health, QOL, and number of healthcare encounters.

Within the scoping review itself, there are additional limitations that warrant discussion. Specifically, each emerging theme is based upon three or fewer studies, which is an important consideration when interpreting findings. Also, the

varying transitions examined within this scoping review (i.e., between hospital and home and between child and adult care) also need to be accounted for when understanding the study's findings. Finally, the quality of evidence included in this scoping review merits discussion. There was a lack of control groups, and lack of consistent measurement tools used across studies, which does not lead to robust conclusions regarding the relationship between CMC/parents of CMC and health outcomes. The lack of conclusive findings is hampered not only by the limited number of studies but also methodological issues within the studies including, small sample sizes, lack of control groups, variation in age ranges, lack of ethnic diversity, diversity in research methodology; and lack of intervention studies. A significant gap in this literature area is that the main group of study participants were CMC who had limited abilities to respond to questions or complete study tools. Parents, therefore, played a great role in completing tools that addressed issues faced by the children.

However, regardless of the limitations of this review, meaningful findings were revealed which suggest: 1) collaboration between acute care centers and community resources may improve health and quality of life for CMC, and 2) further awareness of the expectations and emotional challenges parents and CMC face may be beneficial. It is essential to understand that this review, while not providing exhaustive conclusions, does contribute intriguing findings regarding CMC, their parents and the transition between hospital and home-based care. Arguably, the most important finding of this scoping review is that it underlines the lack of existing research in the area of transition for CMC and their parents from acute care hospitals to community and home care settings. Further research that evaluates the physical and psychosocial health outcomes for CMC and their parents during this transition is essential. Furthermore, continued research is also needed in areas that evaluate the effectiveness of models of care that address transition coordination for CMC and their parents. The availability of research data would help facilitate changes in policy and practices to help improve the health and QOL of CMC and their parents.

## CONCLUSION

Despite the limitations of this review, the significance of the findings cannot be ignored. The growing number of CMC and the corresponding cost escalation across multiple sectors (i.e., health, social services, and education) is a substantial motivating factor for system improvement across the spectrum of care, from hospital to the community [9]. Presently, several policy barriers including a lack of information integration, home care, primary care, and allied health provider shortages, and insufficient community resources/inter-professional care centers impact the delivery of coordinated, inter-disciplinary, family-centered care for CMC, particularly during the transition process [9]. These barriers affect CMC's transition experience and access to the level of community-based care required to keep them out of the hospital and in their communities. This review provides a summary of the issues faced by CMC and their caregivers during the transition process and can inform and guide policy maker's allocation of resources and formulation of resolutions that address these concerns and support parents.



Healthcare providers have an important role as patient advocates to push for legislation that will improve health outcomes and support these children and their parents. Healthcare providers in the hospital and community need to be cognizant of the emotional toll the transition process has on CMC and their parents so they can incorporate support measures into their practice.

**LIST OF ABBREVIATIONS**

- CCMS = Comprehensive Case Management Service
- CMC = Children with Medical Complexity
- CPCHILD = Caregiver Priorities & Child Health Index of Life with Disabilities
- NP = Nurse Practitioner
- PedsQLTM = Pediatric Quality of Life Inventory
- SD = Standard Deviation

**CONSENT FOR PUBLICATION**

Not applicable.

**FUNDING**

None.

**CONFLICT OF INTEREST**

The authors declare no conflict of interest, financial or otherwise.

**ACKNOWLEDGEMENTS**

The PIs (TM, KJ) designed the review, conducted the analysis, and contributed to the writing of this manuscript. KS, NN, JW, and BA contributed to the literature search, data extraction, analysis. JW, JB, and BA contributed to the writing of this manuscript.

**APPENDIX A**

**Search Strategy**

The following three concepts were searched in combination using AND. To facilitate a broad search, various terms for each concept were combined using OR.

- 1) Technology dependent kids OR complex care kids OR pediatrics OR technology-dependent OR children with medical complexity OR parents OR caregivers OR guardians.
- 2) Transition OR community care OR integrated care OR community complex care OR hospital OR home OR primary OR tertiary.
- 3) Mental health OR physical health OR health OR quality of life.

**APPENDIX B**

**Inclusion and Exclusion Rationale**

**Table 2. List of studies review and rationale for inclusion/exclusion.**

Author	Rationale
Altman, Woolfenden, and Breen (2017) [38]	Target population not aligned with eligibility criteria.
Boroughs and Dougherty (2009) [39]	Physical and mental health outcomes not included.
Breneol, S., Belliveau, J., Cassidy, C., & Curran, J. A. (2017) [40]	Physical and mental health outcomes not included.
Cohen <i>et al.</i> (2012) [41]	Included: meets criteria.
Cohen (1999) [32]	Physical and mental health outcomes not included.
Cooper and Centrone (2014) [42]	Physical and mental health outcomes not included.
Elias, Murphy, Council on Children with Disabilities (2012) [43]	Physical and mental health outcomes not included.
Fleming (2004) [44]	Transition & physical and mental health outcomes not included.
Hoffman and Larson (2018) [45]	Physical and mental health outcomes not included.
Kingsnorth <i>et al.</i> (2015) [25]	Physical and mental health outcomes not included.
Kirk (2008) [28]	Included: meets criteria.
Kirk and Glendenning (2004) [29]	Included: meets criteria.
Leyenaar, O'Brien, Leslie, Lindenauer, Mangione-Smith (2017) [46]	Physical and mental health outcomes not included.

MacGregor and Roeher Institute. (2000) [47]	Transition from hospital to home care not included.
Manhas and Mitchell (2012) [31]	Included: meets criteria.
Mendes (2013) [48]	Physical and mental health outcomes not included.
Noyes, Brenner, Fox, and Guerin (2014) [49]	Physical and mental health outcomes not included.
Palfrey <i>et al.</i> (2004) [30]	Included: meets criteria.
Price, McCloskey, and Brazil (2018) [50]	Physical and mental health outcomes not included.
Runciman and McIntosh (2003) [51]	Physical and mental health outcomes not included.
Teare (2008) [52]	Physical and mental health outcomes not included.

## APPENDIX C

### Bias Susceptibility

**Table 3. Bias susceptibility.**

Reference first author:	Palfrey [30]	Kirk [28]	Cohen [32]	Kirk and Glendenning [29]	Manhas [31]
Children with Medical Complexities Definition	+	+	+	+	+
Transition definition	+	+	+	±	+
Physical health outcomes	+	-	+	-	-
Mental health outcomes	-	±	+	±	±
Control of confounders	-	-	-	-	-
External Validity	±	±	+	-	±
Eligibility Criteria	+	+	+	+	±
Inclusion and exclusion criteria	+	+	+	+	+
Sample size and losses	+	+	+	+	+
Statistical analysis	+	-	+	N/A	N/A

(+) yes- indicates that the information was considered; (-) no - indicates that the information was not considered; (±) partially- indicates part of the criteria was met; (?) unclear- indicates the information provided lacked clarity; and (NA) indicates not applicable.

### REFERENCES

- [1] Dewan T, Cohen E. Children with medical complexity in Canada. *Paediatr Child Health* 2013; 18(10): 518-22. <http://dx.doi.org/10.1093/pch/18.10.518> PMID: 24497777
- [2] Law M, Rosebaum P. Service coordination for children and youth with complex needs. *CanChild Cent Child Disabil Res* 2004.
- [3] Cohen E, Berry JG, Camacho X, Anderson G, Wodchis W, Guttmann A. Patterns and costs of health care use of children with medical complexity. *Pediatrics* 2012; 130(6): e1463-70. <http://dx.doi.org/10.1542/peds.2012-0175> PMID: 23184117
- [4] Vincer M, Allen A, Joseph K, Stinson D, Scott H, Wood E. Increasing prevalence of cerebral palsy among very preterm infants: a population-based study. *Pediatric* 2006.
- [5] Anderson JG, Baer RJ, Partridge JC, *et al.* Survival and major morbidity of extremely preterm infants: a population-based study. *Pediatrics* 2016; 138(1)e20154434 <http://dx.doi.org/10.1542/peds.2015-4434> PMID: 27302979
- [6] Southwestern Ontario Maternal, Newborn Child and Youth Network. Transitions in Care: Medically Fragile and Technology Dependent Children. CCKO Strateg 2018.
- [7] Russell CJ, Simon TD. Care of children with medical complexity in the hospital setting. *Pediatr Ann* 2014; 43(7): e157-62. <http://dx.doi.org/10.3928/00904481-20140619-09> PMID: 24977678
- [8] Cohen E, Bruce-Barrett C, Kingsnorth S, Keilty K, Cooper A, Daub S. Integrated Complex Care Model: Lessons Learned from Inter-organizational Partnership. *Heal Q* 2011.
- [9] Kelly AM, Kratz B, Bielski M, Rinehart PM, Blum R. Implementing transitions for youth with complex chronic conditions using the medical home model. *Pediatrics* 2002; 110(6 Pt 2): 1322-7. PMID: 12456952
- [10] Srivastava R, Stone BL, Murphy NA. Hospitalist care of the medically complex child. *Pediatr Clin North Am* 2005; 52(4): 1165-1187, x. <http://dx.doi.org/10.1016/j.pcl.2005.03.007> PMID: 16009262
- [11] Sacchetti A, Sacchetti C, Carraccio C, Gerardi M. The potential for errors in children with special health care needs. *Acad Emerg Med* 2000; 7(11): 1330-3. <http://dx.doi.org/10.1111/j.1553-2712.2000.tb00484.x> PMID: 11073487
- [12] Slonim AD, LaFleur BJ, Ahmed W, Joseph JG. Hospital-reported medical errors in children. *Pediatrics* 2003; 111(3): 617-21. <http://dx.doi.org/10.1542/peds.111.3.617> PMID: 12612245
- [13] Cohen E, Jovcevska V, Kuo DZ, Mahant S. Hospital-based comprehensive care programs for children with special health care needs: a systematic review. *Arch Pediatr Adolesc Med* 2011; 165(6): 554-61. <http://dx.doi.org/10.1001/archpediatrics.2011.74> PMID: 21646589

- [14] Berry JG, Hall DE, Kuo DZ, *et al.* Hospital utilization and characteristics of patients experiencing recurrent readmissions within children's hospitals. *JAMA* 2011; 305(7): 682-90. <http://dx.doi.org/10.1001/jama.2011.122> PMID: 21325184
- [15] Leyenaar JK, Rizzo PA, Khodyakov D, Leslie LK, Lindenauer PK, Mangione-Smith R. Importance and Feasibility of Transitional Care for Children With Medical Complexity: Results of a Multistakeholder Delphi Process. *Acad Pediatr [Internet]* 2018; 18(1): 94-101. <http://dx.doi.org/10.1016/j.acap.2017.07.005> PMID: 28739535
- [16] Bristow S, Jackson D, Shields L, Usher K. The rural mother's experience of caring for a child with a chronic health condition: An integrative review. *J Clin Nurs* 2018; 27(13-14): 2558-68. <http://dx.doi.org/10.1111/jocn.14360> PMID: 29575208
- [17] Murphy KL, Kobayashi D, Golden SL, Nageswaran S. Rural and nonrural differences in providing care for children with complex chronic conditions. *Clin Pediatr (Phila)* 2012; 51(5): 498-503. <http://dx.doi.org/10.1177/0009922812436884> PMID: 22345634
- [18] Yigitalp G, Surucu H, Gumus F, Evinc E. Predictors of Caregiver Burden in Primary Caregivers of Chronic Patients. *Int J Caring Sci [Internet]* 2017; 10(3): 1168-77. Available from: [www.internationaljournalofcaringsciences.org](http://www.internationaljournalofcaringsciences.org)
- [19] Rosenbaum P. No Title. *Sick Kids* 2008.
- [20] March S, Keim-Malpass J. Transition experiences between hospital- and home-care for parents of children with hypoplastic left heart syndrome. *J Spec Pediatr Nurs* 2018; 23(3)e12225 <http://dx.doi.org/10.1111/jspn.12225> PMID: 30098124
- [21] Cohen E, Patel H. Responding to the rising number of children living with complex chronic conditions. *CMAJ* 2014; 186(16): 1199-200. <http://dx.doi.org/10.1503/cmaj.141036> PMID: 25316900
- [22] Lach L, Kohan D, Gamer R, Brehaut J, Miller A, Klassen A, *et al.* The health and psychosocial functioning of caregivers of children with neurodevelopmental disorders. *Disabil Rehabil* 2009. <http://dx.doi.org/10.1080/08916930802354948>
- [23] Matlow A, Wright J, Zimmerman B, Thomson K, Valente M. How can the principles of complexity science be applied to improve the coordination of care for complex pediatric patients?. *Qual Saf Heal Care* 2006. <http://dx.doi.org/10.1136/qshc.2005.014605>
- [24] Kingsnorth S, Lacombe-Duncan A, Keilty K, Bruce-Barrett C, Cohen E. Inter-organizational partnership for children with medical complexity: The integrated complex care model. *Child Care Heal Dev* 2015.
- [25] Kuo D, Goudie A, Cohen E, Houtrow A, Agrawal R, Carle A, *et al.* Inequities in health care needs for children with medical complexity. *Heal Aff* 2014. <http://dx.doi.org/10.1377/hlthaff.2014.0273>
- [26] Peters M, Godfrey C, Khalil H, McInerney P, Parker D, Soares C. Guidance for conducting systematic scoping reviews. *Int J Evid Based Heal* 2015. <http://dx.doi.org/10.1097/XEB.0000000000000050>
- [27] von Elm E, Altman DG, Egger M, Pocock SJ, Gøtzsche PC, Vandenbroucke JP. STROBE Initiative. The Strengthening the Reporting of Observational Studies in Epidemiology (STROBE) statement: guidelines for reporting observational studies. *Lancet* 2007; 370(9596): 1453-7. [http://dx.doi.org/10.1016/S0140-6736\(07\)61602-X](http://dx.doi.org/10.1016/S0140-6736(07)61602-X) PMID: 18064739
- [28] Kirk S. Transitions in the lives of young people with complex healthcare needs. *Child Care Heal Dev* 2008. <http://dx.doi.org/10.1111/j.1365-2214.2008.00862.x>
- [29] Kirk S, Glendinning C. Developing services to support parents caring for a technology-dependent child at home. *Child Care Health Dev* 2004; 30(3): 209-18. <http://dx.doi.org/10.1111/j.1365-2214.2004.00393.x> PMID: 15104576
- [30] Palfrey JS, Sofis LA, Davidson EJ, Liu J, Freeman L, Ganz ML. Pediatric Alliance for Coordinated Care. The Pediatric Alliance for Coordinated Care: evaluation of a medical home model. *Pediatrics* 2004; 113(5)(Suppl.): 1507-16. PMID: 15121919
- [31] Manhas K, Mitchell I. Extremes, uncertainty, and responsibility across boundaries: Facets and challenges of the experience of transition to complex, pediatric home care. *J Child Heal Care* 2012. <http://dx.doi.org/10.1177/1367493511430677>
- [32] Cohen E, Lacombe-Duncan A, Spalding K, *et al.* Integrated complex care coordination for children with medical complexity: a mixed-methods evaluation of tertiary care-community collaboration. *BMC Health Serv Res* 2012; 12(1): 366-77. <http://dx.doi.org/10.1186/1472-6963-12-366> PMID: 23088792
- [33] Raina P, O'Donnell M, Rosenbaum P, *et al.* The health and well-being of caregivers of children with cerebral palsy. *Pediatrics* 2005; 115(6): e626-36. <http://dx.doi.org/10.1542/peds.2004-1689> PMID: 15930188
- [34] Rokach A. Psychological, emotional and physical experiences of hospitalized children. *Clin Case Rep Rev* 2016; 2(4): 399-401. <http://dx.doi.org/10.15761/CCRR.1000227>
- [35] Hopper A, Dokken D, Ahmann E. Transitioning from pediatric to adult health care: the experience of patients and families. *Pediatr Nurs* 2014; 40(5): 249-52. PMID: 25929117
- [36] Rutishauser C, Sawyer SM, Ambresin AE. Transition of young people with chronic conditions: a cross-sectional study of patient perceptions before and after transfer from pediatric to adult health care. *Eur J Pediatr* 2014; 173(8): 1067-74. <http://dx.doi.org/10.1007/s00431-014-2291-9> PMID: 24610395
- [37] Comprehensive care programmes for children with medical complexity. *Cochrane Database Syst Rev* 2019; 5.
- [38] Altman L, Woolfenden S, Breen C. Kids GPS integrated care-leaving the ivory tower. *International Journal of Integrated Care* 2017; 17(3): A29. DOI: <http://doi.org/10.5334/ijic.3141>
- [39] Boroughs D, Dougherty JA. Care of technology-dependent children in the home. *Home Healthcare Now* 2009; 27(1), 37-42. doi: 10.1097/01.NHH.0000343784.88852.94.
- [40] Breneol S, Belliveau J, Cassidy C, Curran J. A. Strategies to support transitions from hospital to home for children with medical complexity: A scoping review. *Int J Nurs Stud* 2017; 72: 91-104. doi: 10.1016/j.ijnurstu.2017.04.011.
- [41] Cohen MS. Families coping with childhood chronic illness: A research review. *Families, Systems, & Health* 1999; 17(2): 149. DOI: <https://doi.org/10.1037/h0089879>
- [42] Cooper Jr, MT, Centrone WA. A medical home far away from home: if the medical home concept of team-based healthcare works for kids and their families in Peru, it will work in your community, too. *Contemporary Pediatrics* 2014; 31(10): 11-13.
- [43] Elias ER, Murphy N. A. Council on Children with Disabilities. Home care of children and youth with complex health care needs and technology dependencies. *Pediatrics* 2012; 129(5): 996-1005.
- [44] Fleming JW. Home health care for children who are technology dependent. New York: Springer Pub. Co. 2004.
- [45] Hoffman A, Larson I. A. Primary Care Preparation for Children with Medical Complexity for Emergency Encounters in the Community. *Pediatric Annals* 2018; 47(3): e102-e105. DOI: 10.3928/19382359-20180221-02 PMID: 29538782
- [46] Leyenaar JK, O'Brien ER, Leslie LK, Lindenauer PK, Mangione-Smith RM. Families' priorities regarding hospital-to-home transitions for children with medical complexity. *Pediatrics* 2017; 139(1): e20161581. DOI: <https://doi.org/10.1542/peds.2016-1581>
- [47] MacGregor D, & Roehner Institute. When kids belong, supporting children with complex medical needs at home and in the community. North York, ON: Roehner Institute 2000.
- [48] Mendes MA. Parents' descriptions of ideal home nursing care for their technology-dependent children. *Pediatric nursing* 2013; 39(2): 91.
- [49] Noyes J, Brenner M, Fox P, Guerin A. Reconceptualizing children's complex discharge with health systems theory: novel integrative review with embedded expert consultation and theory development. *Journal of Advanced Nursing* 2014; 70(5): 975-96.
- [50] Price J, McCloskey S, Brazil K. The role of hospice in the transition from hospital to home for technology-dependent children- A qualitative study. *J Clin Nurs* 2018; 27(1-2): 396-406. DOI: 10.1111/jocn.13941 PMID: 28658513
- [51] Runciman P, McIntosh J. Evaluation of the PATCH nursing service: partnership and training supporting children with complex needs at home. *Primary Health Care Research & Development* 2003; 4(4): 307-18.
- [52] Teare J. Caring for Children with Complex Disabilities. *Caring for Children with Complex Needs in the Community* 2008: 75-91.