

Predictors of Care Gaps in Adolescents With Complex Chronic Condition Transitioning to Adulthood

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abstract

CONTEXT: Breaks in the delivery of health care (ie, [health]care gaps) occur in a large proportion of young people transitioning to adulthood. Developing interventions that prevent adolescents from dropping out of the medical system, as they leave pediatric care, requires an understanding of determinants of care gaps.

OBJECTIVE: To ascertain determinants of care gaps in young people with chronic conditions as they transition to adulthood by performing a systematic literature search.

DATA SOURCES: MEDLINE, CINAHL, and Embase were queried for pertinent peer-reviewed publications.

STUDY SELECTION: Primary quantitative or mixed methods studies that aimed to identify determinants of care gaps in young people (aged 10–25 years) diagnosed with complex chronic conditions and written in English, French, or Dutch were selected. Ten publications satisfied these criteria.

DATA EXTRACTION: For each publication, determinants of care gaps and quantitative results were extracted. Determinants were categorized into 4 groups using thematic analysis. Quantitative results were standardized, and raw data were converted into odds ratios.

RESULTS: Overall, 11 risk factors and 9 protective factors for care gaps were identified. All factors were related to patient characteristics. Demographics, disease-related characteristics, health care services use, and patient health behaviors and beliefs were significant determinants of care gaps in adolescents with chronic conditions.

LIMITATIONS: Large variability in study methods, statistical techniques, and study populations resulted in inconsistent study findings.

CONCLUSIONS: This systematic review identified patient-related determinants of care gaps. Unfortunately, the internal and external validity of the study findings are limited, warranting future prospective, multilevel studies that address remaining knowledge gaps.



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As a result of improved medical therapies and diagnostic tools, together with better survival rates for infants with life-threatening conditions, the prevalence of chronic health conditions in childhood has increased substantially in Western countries.^{1,2} The estimated prevalence ranges from 3.5% to 35.3%.³ An emerging population of special interest for health care delivery is young people with complex chronic conditions (CCCs). CCCs are defined as “any medical condition that can be reasonably expected to last at least 12 months (unless death intervenes) and to involve either several different organ systems or one organ system severely enough to require specialty pediatric care and probably some period of hospitalization in a tertiary care center.”⁴ CCCs represent a specific subgroup of patients, accounting for ~10% of pediatric admissions in the United States. The most frequent types of CCCs are cardiovascular, congenital, neuromuscular, respiratory, and oncological.⁵ In general, young people with CCCs disproportionately use health care resources because of their medical fragility. Indeed, these patients have significantly higher hospitalization rates, readmissions, greater use of technology-dependent devices, use more prescribed medications, and have higher inpatient mortality rates, yielding a financial and organizational burden for the current healthcare system.⁵⁻¹¹

Because most young people with CCCs are at risk for developing long-term complications, lifelong specialized care is mandatory.^{6,12,13} During childhood, care is generally provided at pediatric services, but as these young patients transition to adulthood, transfer to adult-oriented care services is recommended.^{12,13} This health care transition is an important but challenging life event,

¹⁴ and successful transfer to adult care is paramount for young people with CCCs.¹⁵

Gaps in this transitional process occur in 7% to 21% of young people with acromegaly,¹⁶⁻¹⁸ 9% to 17% of HIV-infected adolescents,¹⁹ 11% to 24% of patients with type I diabetes mellitus,²⁰ and 7% to 76% of young people with congenital heart disease.²¹ This break in the delivery of health care (ie, care gaps) as adolescent patients leave pediatric care is associated with increased morbidity rates, long-term complications, number of hospitalizations, need for urgent interventions or reinterventions, and higher rates of health-risk behaviors.^{17,20-30} Therefore, developing measures to prevent such care gaps are of utmost importance.

To develop tailored preventive interventions or health care system reforms, a comprehensive understanding of the determinants of care gaps is required first. Care gaps can be predicted by patient-related factors, factors related to the level of care organization within hospitals, and factors associated with the overall health care system. To date, no comprehensive overview of determinants of care gaps has been made available. As a first step, we carried out a systematic review of the available research. The aim of the present review, then, was to systematically search the literature for determinants of care gaps in young people with CCCs transitioning to adulthood.

METHODS

A systematic literature review was performed. The review and reporting are in line with the instructions and recommendations of the 2009 PRISMA (Preferred Reporting Items

for Systematic Reviews and Meta-Analyses) statement.³¹

Search Strategy

Three bibliographic databases, MEDLINE, CINAHL, and Embase, were queried for relevant peer-reviewed publications from inception to September 2014. A distinctive search string was created for each respective database through repeated brainstorm sessions (see Supplemental Table 5). These were verified by a biomedical librarian and were supplemented by the snowball searching technique (ie, screening of reference lists of relevant publications).

Eligibility Criteria

Publications were selected if the candidate paper met the following criteria: (1) reported on a primary study that included quantitative or mixed-methods designs, (2) the study population comprised young people (aged 10–25 years) diagnosed with CCCs^{4,32,33} transferring to adult care, (3) 1 of the study aims was to identify determinants of care gaps, and (4) published in English, French, or Dutch peer-reviewed journals. Qualitative studies, editorials, published comments, and letters to the editor were excluded. No restrictions on the date of publication were made.

For the purpose of this literature review, a (health) care gap was conceptually defined as “any type of discontinuation of the care process in which the time lapse between mandatory follow-up visits exceeds the period of time defined by disease-specific recommendations.” The notion of a care gap includes concepts found in literature such as lapse(s) of care,²⁵ loss/lost to follow-up,^{34,35} unsuccessful transfer or transition,³⁶⁻³⁸ and cessation of follow-up.²¹

Study Selection

A total of 1718 candidate records were initially identified in the

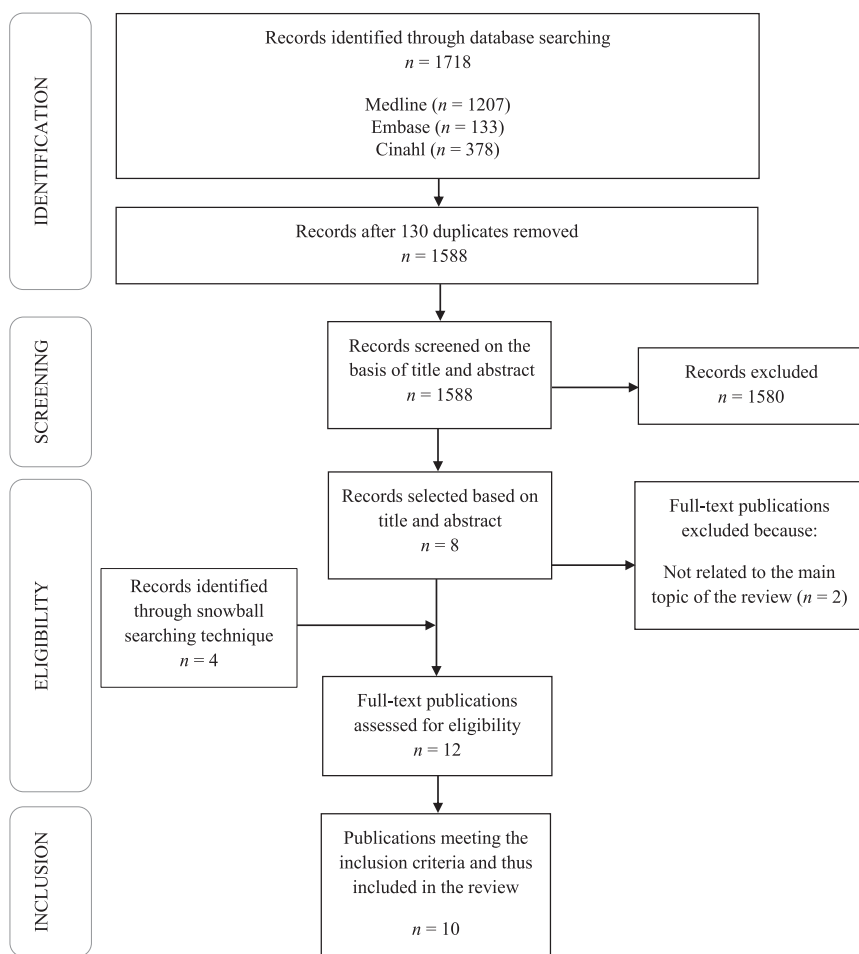


FIGURE 1
Flow diagram of the study selection process.

respective databases. After exclusion of 130 duplicate papers, the titles and abstracts of 1588 records were screened using RefMan software version 12.0 (Thomas Reuters). This screening was independently performed by 2 of the authors (E.G., L.B.). Finally, 8 full-text publications were independently selected for eligibility assessment by 2 of the authors (E.G., L.B.). At this stage, 4 publications were added to the 8 through the use of the snowball technique. Twelve publications were finally identified for eligibility. Two publications, however, were excluded after a detailed review of the full text of the paper, leaving 10 publications to be included in this review (see Fig 1).

Data Extraction

For each of the 10 included publications, the following predetermined set of variables was extracted by 1 author (E.G.): first author, year of publication, setting, study design, method of data collection, sample characteristics, operational definition of the primary outcome, and kind of statistical analyses employed. Furthermore, for each paper, the determinants of care gaps and the quantitative results of the respective statistical tests were extracted. Results obtained through qualitative study designs or analysis techniques as part of a mixed methods design were excluded, however.

Using thematic analysis, we categorized determinants of care gaps into 4 groups, those that related to (1) demographic characteristics, (2) disease-related characteristics, (3) health care services use, and (4) patient behavior. Quantitative study results were standardized, and *P* values were converted into odds ratios and 95% confidence intervals, using a Web-based calculator.³⁹ Because some studies reported insufficient data, we were unable to calculate effect sizes for these studies.

Assessment of Methodologic Rigor

Published studies vary with regard to methodologic rigor, but this can be assessed quantitatively. Because all selected articles reported on studies that had an observational, nonrandomized design, the methodologic quality of each study was critically appraised by using an adapted version of the Methodological Index for Non-Randomized Studies (MINORS) instrument.⁴⁰ The original MINORS instrument comprises 12 items, of which the first 8 are applicable to noncomparative studies. The 4 remaining items are to be evaluated in studies using a comparative study design. After critically revising all MINORS items, we deleted 2 items from the original instrument because they were not applicable to our included studies. These were item 5 (“unbiased [blind/double-blind] assessment of the study endpoint”) and item 7 (“attrition of study participants <5%”). Furthermore, although the original MINORS instrument only assesses the appropriateness of applied statistical analysis techniques for comparative studies, we decided to include this item to evaluate all included studies. Each study was rated on the respective MINORS items as “not reported” (0), “reported but inadequate” (1), or “reported and adequate”

(2).⁴⁰ The use of this adapted MINORS instrument produces a global score calculated as the sum of the scores on the respective list of relevant items (maximum value of 14) for noncomparative studies and (maximum value of 20) for comparative studies (see Supplemental Table 6).

RESULTS

Study Characteristics

The 10 selected publications reported results on studies of 4 types of CCCs: sickle cell disease,^{36, 41} congenital heart disease,^{21,25,35, 37, 42,43} congenital adrenal hyperplasia,³⁴ and juvenile idiopathic arthritis.³⁸ Two of these studies explicitly stated that patients diagnosed with a neurodevelopmental condition were excluded.^{37,43} The majority of studies (80%) took place in the United States^{25,36,37,41,43} or Canada,^{35,38,42} whereas only 2 of the studies (20%) were conducted in Europe.^{21, 34} Sample sizes ranged from 18 to 922 young people (median: 227). Although most papers did not report the ethnic composition of their study sample,^{25,34,35,37,38,42} the majority included samples that were exclusively African American^{36,41} or white/Caucasian.^{21,38}

Seven studies used a retrospective descriptive design,^{21,25,34,36, 38,41, 42} 2 studies used a cross-sectional descriptive design,^{37,43} and 1 study used a limited mixed methods approach, combining a matched case-control quantitative study with a qualitative interview study.³⁵ Most studies (70%) collected data on care gaps and on potential determinants based on a retrospective review of medical records or clinical databases.^{21,34–38,41} Some studies used patient self-reports such as surveys, telephone interviews, or structured face-to-face interviews to collect these data.^{21,25,35, 37,41,43} One study analyzed a province-wide administrative database of young CCC

patients.⁴² Single-center studies were most common,^{21,25,34–36,38} although 4 studies used a multicenter study in design^{37,41–43} (see Supplemental Table 7).

Quality Assessment of Research Methodology

The MINORS global quality score ranged from 15 to 16 of 20 for comparative studies,^{35,36} and from 7 to 12 of 14 for noncomparative studies.^{21,25,34,37, 38,41–43} With the exception of 1 study,⁴¹ all eligible patients were included, follow-up periods were adequate, and a clear explanation was provided of the criteria used to evaluate the outcome of care gaps. Eight studies (80%) collected data on determinants retrospectively.^{21,34–36, 38,41,42,44} None of the studies performed a power analysis with a prospective calculation of the required sample size. The statistical analyses used to identify determinants of care gaps were adequate in 6 studies (60%).^{21,25,35,37,42,43} These studies used multivariate logistic regression analyses to account for potential confounders. Four studies, however, only performed 2-group comparisons,^{34,36} did not correct for multiple statistical tests,^{34,36,38, 41} calculated correlation coefficients,⁴¹ or performed only univariate analysis³⁸ (see Supplemental Table 6).

Definitions of Care Gaps in the Literature

A broad range of definitions of care concepts has been used in the selected publications (Supplemental Table 8). All studies provided an operational definition. However, the level of setting specification, time intervals, and data sources varied greatly across studies. Some studies gave highly detailed information on these components, whereas others did not. Instead, the specifications could indirectly be derived from the study methodology or results section. Only 1 study failed to

provide any kind of information on the time interval considered for the determination of a care gap.⁴¹ Detailed information on the concepts, operational definitions, and specifications is provided in Supplemental Table 8.

Determinants of Care Gaps in the Literature

Forty-five potential determinants of care gaps have been investigated. Factors that could increase or decrease the likelihood of experiencing care gaps were also identified. These factors can be considered as “risk factors” or “protective factors,” respectively. Overall, 11 risk factors and 9 protective factors were identified. An overview of the determinants of care gaps identified in our systematic review is provided in Tables 1, 2, 3, and 4.

Demographics

Several relevant patient demographic characteristics stood out in the literature. Living independently from parents,²⁵ male gender,^{21,42} lower family income,³⁵ and greater travel distance to closest adult specialized clinic^{34,36} were identified as significant risk factors for care gaps. However, nonsignificant results were reported in some studies for the latter 3 factors. Older age of the patient at the last pediatric visit was reported to be either a risk factor³⁶ or a protective factor.³⁷ Ethnicity,^{36, 41,43} type of health care insurance,^{36,41} location of residence (urban/rural),⁴² age at diagnosis,²⁵ and educational level attained^{38,43} were not significantly related to care gaps in any of the reviewed papers (Table 1).

Disease-Related Characteristics

Milder disease activity (eg, low active joint number in patients with juvenile idiopathic arthritis), severity (eg, SC/Sβ⁺ genotype in SCD patients), and complexity (eg, mild complexity of a congenital heart defect) were

significant risk factors for care gaps in several studies.^{25,34,36, 38,42,43} The study of Goossens and colleagues, however, did not find a significant effect of disease complexity on care gaps. For patients with congenital heart disease, not having undergone heart surgery also increased the risk of care gaps.²¹ However, having at least 1 comorbid condition was reported to be protective against the occurrence of care gaps.³⁷ Candidate determinants that were analyzed but not significant were residual hemodynamic problems identified at the most recent echocardiography, medication use, and an implanted pacemaker or cardiac defibrillator³⁵ (Table 2).

Health Care Services Use

The patients' pattern of using various health care services also predicted to some extent whether they would experience care gaps. For example, significant risk factors for care gaps included fewer outpatient visits in pediatric care over the 3-year period before transfer,^{34, 42} the last visit taking place outside a university hospital,⁴² childhood hospitalizations,⁴² and a history of at least 1 missed appointment.³⁵ Furthermore, the multicenter study of Gurvitz et al (2013)⁴³ found a significant geographic difference among hospital locations (ie, states of Colorado, Oregon, and Washington) on whether the care gaps occurred. By contrast, protective factors against care gaps were having a written referral to a specific professional who would provide adult follow-up care³⁵ and attending the first or second outpatient visit in an adult clinic (ie, good early attenders^{34,41}; Table 3).

Patient Behavior

Certain patient behaviors helped patients avoid care gaps. Significant protective factors were greater independence in attending appointments,³⁷ belief that follow-up should be continued in specialized adult care,³⁷ higher levels of

TABLE 1 Demographic Determinants of Care Gaps

	Significant Risk Factors	Significant Protective Factors	Nonsignificant Results Reported
Living independently from parents	OR = 4.1; 95% CI 1.7–10.1 ²⁵		—
Male	OR = 1.80; 95% CI 1.02–3.17 ²¹		OR = 0.57; 95% CI 0.57–1.23 ³⁶
	OR = 1.52; 95% CI 1.05–2.20 ⁴²		OR = 1.23; 95% CI 0.61–1.98 ³⁴
			OR = 2.15; 95% CI 0.91–2.6 ³⁸
			41a,43a
Higher family income		OR = 0.87 per increase in annual salary of \$10 000; 95% CI 0.77–0.98 ³⁵	^{37a}
Older age at last pediatric visit	OR = 10; 95% CI 1.71–58.43 ³⁶	OR = 1.29; 95% CI 1.10–1.51 ³⁷	
Travel distance to closest adult-specialized center	OR = 7.71; 95% CI 1.53–38.83 ³⁶		OR = 0.56; 95% CI 0.27–1.17 ³⁷
	$P = .03$ (χ^2 14 vs 24 miles) ^{34a}		$P = .34$ ^{35a}
			21a
Ethnicity			$P = .67$ ^{36a}
			41a
Type of healthcare insurance			OR = 0.51; 95% CI 0.56–1.21 ³⁶
			41a
Type of residence (urban, rural)			42a
Age at diagnosis			OR = 1; 95% CI 1.0–1.1 ²⁵
Educational level			OR = 0.49; 95% CI 0.46–1.0838 ^{43a}

^a Insufficient data were reported, and thus raw data could not be converted into odds ratio (ORs) and 95% confidence intervals (CIs).

self-efficacy,⁴¹ abstaining from substance use,⁴⁰ and full compliance with antibiotic prophylaxis regimens.³⁷ Nonsignificant determinants were self-reported family functioning,³⁷ health beliefs,³⁷ 36-item Short Form Health Survey scores,³⁷ self-rated activity restrictions,³⁷ general preference to self-care,³⁷ self-reported expected frequency of visits in an adult clinic,³⁷ treatment adherence,⁴¹ and knowledge of disease name⁴³ (Table 4).

DISCUSSION

Over recent decades, survival rates have improved substantially for children diagnosed with a complex chronic condition (CCC).^{1,2} However, to guarantee improved quality

of life, enhanced health status, and prevention of complications in this growing population, the provision of continuous care is mandatory. International guidelines stress the importance of providing uninterrupted, age- and developmentally appropriate health care to patients with CCCs throughout their life.^{6,12,13} Achieving continuity of care appears to be challenging in adolescents and young adults diagnosed with CCCs, however.

Typically, adolescence is the phase of life in which authority is challenged and risk-taking behaviors are more common, posing additional risks for patients with CCCs.^{45,46} These developmental transitions, in combination with the required transfer of health care setting, embody an important challenge for

TABLE 2 Disease-Related Determinants of Care Gaps

	Significant Risk Factors	Significant Protective Factors	Nonsignificant Results Reported
Milder disease severity, complexity, or activity	OR = 0.26; 95% CI 0.07–0.95 ³⁶		^{21a}
	OR = 4.31; 95% CI 0.77–24.14 ³⁴		
	OR = 4.14; 95% CI 2.17–7.87 ⁴²		
	OR = 2.3; 95% CI 1.2–4.5 ²⁵		
	OR = 2.67; 95% CI 1.16–6.16 ³⁸		
	OR = 2.2–4.1, $P < .0001$ ⁴³		
≥1 Comorbid condition		OR = 3.13; 95% CI 1.13–8.67 ³⁷	OR = 1.12; 95% CI 0.62–2.04 ³⁵
No previous heart surgery performed (CHD)	OR = 5.97; 95% CI 3.04–11.72 ²¹		
Residual hemodynamic problems at last echocardiography examination			OR = 1.69; 95% CI 0.93–3.07 ³⁵
Medication use			OR = 1.48; 95% CI 0.84–2.59 ³⁵
Implanted pacemaker or automatic implantable cardiac defibrillator			OR = 8.00; 95% CI 0.84–75.81 ³⁵

CHD, congenital heart disease.

^a Insufficient data were reported, and thus raw data could not be converted into odds ratio (ORs) and 95% confidence intervals (CIs).

adolescents with CCCs. Previous studies show that a significant proportion of young people with CCCs experience care gaps, and some are even completely lost to follow-up.^{16–21}

Improving continuity of care for young people with CCCs requires tailored interventions or strategies to facilitate a smooth and continuous care process. An in-depth understanding of determinants of care gaps is, nevertheless, indispensable for developing such interventions. Because a comprehensive overview of determinants of care gaps is currently missing in literature, we performed a systematic review of all the existing relevant literature. This review identified 10 relevant publications that reported on

investigations of young people diagnosed with a CCC. Four types of CCCs were represented in these articles: sickle cell disease,^{36,41} congenital adrenal hyperplasia,³⁴ congenital heart disease,^{21,25,35,37,42,43} and juvenile idiopathic arthritis.³⁸ To our knowledge, no other types of CCCs have been investigated. The phenomenon of care gaps appeared to be most often studied in populations of young people with congenital heart disease.^{21,25,35,37,42,43}

A total of 11 risk factors and 9 protective factors within 4 groups were identified: (1) demographics, (2) disease-related characteristics, (3) use of health care services, and (4) patient behavior.

A milder disease was the only care-gap risk factor for which all studies unequivocally reported

significant results.^{25,34,36,38,42,43} Furthermore, having no or few comorbid conditions³⁷ and not having undergone heart surgery,²¹ which are both indirect indicators of milder disease, were significant determinants of care gaps. Hence, special attention should be given to patients diagnosed with milder types of CCCs because these patients seem to have an increased likelihood of experiencing care gaps. These patients might perceive their long-term risk for mortality and morbidity to be low compared with patients with moderate-to-complex conditions. Tailored interventions aiming to convince patients of the benefits of continued follow-up care could be implemented in practice as a preventive measure against care gaps.

A set of 5 characteristics related to demographics or the patient's living situation^{21,42} were identified as significant determinants of care gaps. Unfortunately, other studies have reported nonsignificant results for some of these demographic risk factors, leading to inconsistent research findings. These patient-related characteristics are less modifiable but could be used in practice when screening for patients at risk for care gaps. Finally, variables related to the health care consumption of patients⁴² increased the risk for care gaps. It is notable, however, that patients demonstrating better self-management skills were less likely to experience care gaps.^{37,41}

Overall, this review revealed that a rather limited set of patient-related characteristics is known to alter the risk for care gaps in young people with CCCs transitioning to adulthood and adult care. Most factors were related to demographics, the disease, or health care services use. Although most identified risk factors are modifiable to a limited extent, some protective factors such as guaranteeing that patients

leave pediatric care with a written referral to a specific professional who would be providing follow-up adult care, or checking whether patients attend their first outpatient visits in adult care, might be components of interventions aimed at preventing care gaps.

Although this review aimed to identify determinants of care gaps, no consensus on the conceptual definition of care gaps currently exists. None of the publications provided a conceptual definition, but they all operationalized this concept for the purpose of their study. This review, however, revealed a large heterogeneity of operational definitions on the concept of discontinuation of care. Synonyms used for care gaps⁴³ were as follows: uninterrupted care,⁴¹ lapse in care,²⁵ lack of follow-up,⁴² no follow-up,²¹ lost to follow-up,^{34, 35} and unsuccessful transition.³⁶ All studies provided a relatively detailed operational definition in terms of setting and frequency of care when determining the occurrence of care gaps. To enhance comparability of study results, consensus should be reached on both the conceptual and operational definitions of care gaps. Additional efforts should be made to define the uniform requirements for the operationalization of care gaps. Operational definitions of care gaps need to specify condition-specific time intervals and differentiate, if applicable, between levels of disease severity.

Although all studies aimed to identify determinants of care gaps, a wide variety of statistical techniques, research designs, and data collection methods were used. Determinants were identified using a variety of techniques such as 2-group comparisons, calculation of correlation coefficients, or uni- and multivariable regression analyses. From a methodologic point of view, there is an important difference in the robustness of results obtained

TABLE 3 Determinants of Care Gaps Related to Health Care Services Use

	Significant Risk Factors	Significant Protective Factors	Nonsignificant Results Reported
Lower number of pediatric outpatient visits during pretransfer period	$P = .01^{34a}$ OR = 1.15 per visit decrease; 95% CI 1.09–1.21 ⁴²		
Last follow-up visit outside of university hospital	OR = 1.63; 95% CI 1.02–2.61 ⁴²		OR = 2.09; 95% CI 0.71–6.13 ³⁵
Non-cardiac-related hospitalizations	OR = 1.93; 95% CI 1.18–3.17 ⁴²		
Cardiac-related hospitalizations without invasive procedures	OR = 2.22; 95% CI 1.36–3.62 ⁴²		
History of ≥ 1 missed appointments	OR = 13.0; 95% CI 3.3–51.7 ³⁵		
Geographic location of clinic	$P < .001$ (Colorado); $P < .002$ (Oregon); $P < .027$ (Washington) ^{43a}		
Written recommendation on type of professional performing adult care		OR = 0.4; 95% CI 0.2–0.8 ³⁵	
Keeping first and second appointments in adult care (ie, good early attenders)		OR = 5.2; 95% CI 1.25–21.57 ³⁴	
		$P < .05^{41a}$	

^a Insufficient data were reported, and thus raw data could not be converted into odds ratio (ORs) and 95% confidence intervals (CIs).

TABLE 4 Determinants of Care Gaps Related to Patient's Behavior

	Significant Risk Factors	Significant Protective Factors	Nonsignificant Results Reported
Patients' belief that follow-up should be performed at specialized adult clinic		OR = 3.64; 95% CI 1.34–9.9 ³⁷	
No substance use		OR = 0.18; 95% CI 0.07–0.50 ³⁷	
Compliance to antibiotic prophylaxis		OR = 4.23; 95% CI 1.48–12.06 ³⁷	
Greater independence in attending appointments		OR = 6.59; 95% CI 1.61–27.0 ³⁷	
Higher levels of self-efficacy		$P < .05^{41a}$	
Self-reported family functioning			37a
Health beliefs			37a
SF-36 scores			37a
Self-rated activity restrictions			37a
General preferences for self-care			37a
Patient-reported expected frequency of visits to adult clinic			37a
Treatment adherence			37a
Patient knows name of the condition			37a

SF-36, 36-item Short Form Health Survey.

^a Insufficient data were reported, and thus raw data could not be converted into ORs and 95% CIs.

through the use of repeated group comparisons without correction for multiple testing versus those obtained through the use of multivariate regression analyses. Furthermore, critical appraisal of the methodological rigor of the included studies revealed that most studies analyzed determinants retrospectively. Retrospective analyses, however, are characterized by the risk of missing information on specific factors not included in the previously established data set.

Despite the transparent, objective, and rigorous nature of the present review, some methodologic limitations should be noted. First, although relevant literature was searched for using rigorously developed search strings, only 10 of 1718 retrieved records (<0.5%) were deemed relevant. The inclusion of the search term “loss/lost to follow-up” most likely resulted in a large number of unrelated publications. This search term is often used in the abstract of articles within the context of attrition of participants from a longitudinal study. This search term was, however, indispensable for our purpose because it is often used as a synonym for care gaps. Furthermore, additional relevant references might have been overlooked when indexed in other databases or only

retrievable in the gray literature. Second, the development of forest plots or conducting a meta-analysis would have been highly valued when investigating determinants of care gaps. Unfortunately, these analysis techniques could not be applied because of the large variability in study designs, data collection methods, sample sizes, small number of studies investigating a specific factor, and the lack of sufficient raw data. Third, generalizability of study results is fairly low because only 4 types of CCCs were investigated; patients with neurodevelopmental impairments were explicitly excluded in most studies, and sample sizes were small to moderate.

Although this systematic review identified factors associated with care gaps, additional research is needed to address the remaining knowledge gaps. Prospective, multicenter, international study designs that analyze both clinical and administrative datasets are sorely needed to better understand the relationship between care gaps and patient-, hospital-, and health care system-related factors. Furthermore, to expand the internal and external validity of research findings summarized in this review, additional studies should be performed in larger

patient populations diagnosed with other types of CCCs.

CONCLUSIONS

This systematic literature review identified a set of determinants related to the characteristics of young people with CCCs that increase or decrease the risk of experiencing a care gap. All identified determinants were related to the individual patient, such as demographics, disease-related characteristics, use of health care services, and patient behavior. Previous studies demonstrate that the transition to adulthood is a vulnerable period for young people with CCCs and one characterized by care gaps. Strategies to prevent such gaps in the care process are urgently needed, but additional prospective, multicenter, international research projects are necessary to bolster the body of evidence. Furthermore, a uniform operational definition of a care gap is needed to increase the comparability of study findings.

ABBREVIATIONS

CCC: complex chronic condition
MINORS: Methodological Index
for Non-Randomized
Studies

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