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SYSTEMATIC REVIEW AND META-ANALYSIS

Discontinuity of Cardiac Follow-Up in Young People With Congenital Heart Disease Transitioning to Adulthood: A Systematic Review and Meta-Analysis

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BACKGROUND: The majority of people born with congenital heart disease require lifelong cardiac follow-up. However, discontinuity of care is a recognized problem and appears to increase around the transition to adulthood. We performed a systematic review and meta-analysis to estimate the proportion of adolescents and emerging adults with congenital heart disease discontinuing cardiac follow-up. In pooled data, we investigated regional differences, disparities by disease complexity, and the impact of transition programs on the discontinuity of care.

METHODS AND RESULTS: Searches were performed in PubMed, Embase, Cinahl, and Web of Science. We identified 17 studies, which enrolled 6847 patients. A random effects meta-analysis of single proportions was performed according to the DerSimonian-Laird method. Moderator effects were computed to explore sources for heterogeneity. Discontinuity proportions ranged from 3.6% to 62.7%, with a pooled estimated proportion of 26.1% (95% CI, 19.2%–34.6%). A trend toward more discontinuity was observed in simple heart defects (33.7%; 95% CI, 15.6%–58.3%), compared with moderate (25.7%; 95% CI, 15.2%–40.1%) or complex congenital heart disease (22.3%; 95% CI, 16.5%–29.4%) (P=0.2372). Studies from the United States (34.0%; 95% CI, 24.3%–45.4%), Canada (25.7%; 95% CI, 17.0%–36.7%), and Europe (6.5%; 95% CI, 5.3%–7.9%) differed significantly (P=0.0004). Transition programs were shown to have the potential to reduce discontinuity of care (12.7%; 95% CI, 2.8%–42.3%) compared with usual care (36.2%; 95% CI, 22.8%–52.2%) (P=0.1119).

CONCLUSIONS: This meta-analysis showed that there is a high proportion of discontinuity of care in young people with congenital heart disease. The highest discontinuity proportions were observed in studies from the United States and in patients with simple heart defects. It is suggested that transition programs have a protective effect.

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Key Words: care gaps ■ continuity of care ■ heart defects, congenital ■ lapse of care ■ lost to follow-up ■ meta-analysis ■ systematic review

Congenital heart disease (CHD) is a birth defect that has evolved from an often lethal disorder to a chronic lifespan condition. Survival rates have substantially increased in recent decades, yielding >90% of children with CHD surviving into adulthood to date.[1,2] Consequently, the need for affected individuals to receive cardiac follow-up across their lifespan has grown. During childhood, patients with CHD are typically cared for in pediatric cardiology, whereas adult congenital heart disease (ACHD) programs are in place to provide medical follow-up during adulthood. By the end of adolescence or at emerging adulthood, patients

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CLINICAL PERSPECTIVE

What Is New?

- This systematic literature review on discontinuity of care in patients with congenital heart disease at the transitional age identified 17 studies.
- Discontinuity proportions ranged from 3.6% to 62.7%, with a pooled estimated proportion of 26.1%.
- European studies showed significantly lower proportions of discontinuity than American or Canadian studies; transition programs show the potential to reduce discontinuity of care.

What Are the Clinical Implications?

- The high proportion of discontinuity of care urges implementation of preventative interventions to keep patients under cardiac follow-up.
- Transition programs could be implemented as a way to improve continuity of care.

Nonstandard Abbreviations and Acronyms

ACHD	adult congenital heart disease
NOS	Newcastle-Ottawa Scale
Q	Cochran's Q for heterogeneity
QM	Cochran's Q for moderation

are assumed to transfer their care from pediatrics to adult-care facilities.[3]

Although continuous cardiac care is recommended for most individuals with CHD, a substantial proportion present with care gaps.[4] In the period of adolescence and emerging adulthood, patients are particularly vulnerable to developing such care gaps because they are undergoing physiological, psychological, and social changes and are changing providers and sometimes institutions. It has been shown that such interruptions in care are detrimental, because a substantial proportion of patients who were lost to follow-up present with complications of hemodynamic importance.[5] In addition, lapses in care are associated with triple the likelihood of needing an urgent surgical or catheter-based intervention.[6]

Transition programs are hypothesized to play a role in preventing patients from failing to continue regular follow-up.[7] Such programs can be effective, because transition curricula inform patients about the rationale for ongoing follow-up, and teach and empower them to navigate through the medical system.[8] This is necessary because adolescents and young adults lack knowledge about the need for lifelong cardiac care.[9,10]

Reported proportions of patients discontinuing care vary substantially across studies. This may be because of differences in definition and operationalization. In addition, there seems to be an important variability across countries and by disease complexity. Because of the lack of pooled data, the precise magnitude of the problem remains unknown and the impact of influencing factors is unclear. For instance, it is not known to what extent transition programs are effective in retaining patients in follow-up.[11] We therefore conducted a systematic literature review and meta-analysis aiming (1) to estimate the proportion of discontinuity of cardiac follow-up around the globe, (2) to investigate whether discontinuity of cardiac follow-up differs by the complexity of the heart defect, (3) to explore regional differences, and (4) to evaluate whether transition programs yield a lower discontinuity proportion than usual care.

METHODS

The authors declare that all supporting data are available within the article and its online supplementary files. Because the present study was a systematic literature review using published material, no approval from an Ethics Committee was needed.

Literature Sources and Searches

We performed a systematic literature search in PubMed, Embase, Cinahl, and Web of Science from their inception to April 6, 2020. The search strings that were used in the 4 databases are detailed in Table S1. The search was complemented by the snowball technique, whereby we screened reference lists of relevant publications. Authors who were contacted could provide additional references. Gray literature (eg, theses, unpublished data) was not deemed to be suitable for inclusion. The review and reporting are in line with the Preferred Reporting Items for Systematic Reviews and Meta-Analyses statement.[12] The review is registered at PROSPERO (CRD42020182413).

Eligibility Criteria

Studies that met the following criteria were considered eligible for inclusion: (1) entire study population or a subset of adolescents (aged 10–24[13]) or emerging adults (aged 18–29[14]) with CHD (ie, studies reporting discontinuity over the entire life spectrum were excluded); (2) quantitative research designs; (3) discontinuation of care (irrespective of definition or operationalization used) as primary, secondary, or ancillary end point; (4) published in English, Spanish, French, Dutch, German, or Swedish, because these are the languages that the authors master; and (5) available online (e-pub ahead of print) or in print. Studies were excluded if they (1)

studied adults with CHD over the entire age spectrum, or (2) did not report the size of the study population (ie, denominator was lacking). Only full articles were eligible for inclusion. Conference abstracts were excluded because they do not include enough details on the precise definition and measurement of discontinuity of care and could not be appraised in terms of the methodological rigor.

Intervention studies in which the effects of transition were evaluated were also included. However, in our review and meta-analysis, we used the data from the control/usual care groups. By doing so, we try to avoid comparative bias through interventions. Data from the intervention groups/transition programs were only used when comparing the discontinuity proportions of studies that evaluated the implementation of transition programs (research aim 4).

Study Selection and Data Extraction Process

The Preferred Reporting Items for Systematic Reviews and Meta-Analyses flowchart is shown in Figure 1. The search resulted in 1290 records. After removing duplicates, 898 references were evaluated based on title and abstract. A total of 859 references were not deemed relevant to the research question. The full text of the remaining 39 references was evaluated and an additional 25 publications were excluded (reasons mentioned in flowchart). Three additional articles were identified through the snowball method and by authors who we contacted. Eventually, 17 studies were included in this systematic review.[6,15–30] We used Rayyan as a web application to assist in the selection process.[31]

The data were extracted by 2 authors (P.M./E.G.) and compared. Discordances were discussed until consensus was reached. If required data were missing from the article, the authors were contacted by email to obtain the necessary information.

Quality Assessment of Individual Studies

We used the NOS (Newcastle-Ottawa Scale) to assess the methodological quality and risk of bias in the included articles.[32] This scale uses an 8-point classification. The critical appraisal was performed by 2 researchers (P.M./E.G.), and consensus meetings were held to discuss the items over which the reviewers were not in agreement. Items that were not relevant to the purpose of this study were indicated as "not relevant."

Statistical Analysis

The proportion of patients with discontinuity of care is expressed in absolute numbers and percentages. Depending on the methodology used, some studies have a proportion of patients that were untraceable. We therefore performed a sensitivity analysis by computing a worst-case scenario, in which untraceable patients are assumed not to be in follow-up.

To determine an overall estimate of the discontinuity of care proportion, we used a random effects meta-analysis of single proportions according to the DerSimonian-Laird method. We did this because heterogeneity was anticipated, based on prior reviews of the literature.[4,7] To stabilize variances, study data were first transformed using the logit transformation. Heterogeneity between studies was assessed with the Cochran's Q test, and its magnitude was evaluated by the I^2 statistic.[33] To explore whether differences in the definition of discontinuity or the place of recruitment (pediatrics or adult care) were sources of heterogeneity, we performed moderator analyses (expressed in Cochran's Q for moderation [QM]). Further, we conducted analyses by region (United States, Canada, or Europe) and by complexity of the heart defect (simple, moderate, or complex)[34] to investigate whether region or complexity moderated the outcome. Publication bias was evaluated by visually inspecting the funnel plot and by using the Egger's test of asymmetry applied to the funnel plot. All statistical analyses were performed with the metafor and meta packages in Rstudio.[35]

RESULTS

Characteristics of Selected Studies

The 17 studies in this systematic review enrolled a total of 6847 patients with CHD (Table). Ten studies were performed in the United States,[6,18–22,24,25,27,28] 5 studies in Canada,[15,16,23,26,29] 1 in Belgium,[17] and 1 in Sweden.[30] Different concepts were used to express the phenomenon under study. Eight studies formulated it in terms of continuity of care,[15,16,18,22,23,27,29,30] and 7 studies in terms of discontinuity of care.[6,17,19,21,24,26,28] Two studies provided 2 definitions, both reflecting continuity and discontinuity of care, which were not completely complementary to each other.[20,25] The operational definitions used in the different studies are described in the Table. These definitions could be categorized into 3 groups: "lacking any cardiac visit in a period of 4 to 5 years after transfer,"[15–17,20,27,30] "time intervals of more than 2 or 3 years between visits (or similar, depending on complexity of CHD),"[6,18,19,22,24–26,28,29] and "discontinuity of care over a period of 12 years or longer during adolescence and emerging adulthood."[21,23] The different methodological approaches are graphically expressed in Figure 2.[6,15–30] Eleven out of the 17 studies[6,16,19,21–24,26–29] reported on the full study population, whereas the remaining 6 studies[15,17,18,20,25,30] had a group of patients who were untraceable. Thirteen studies recruited patients in pediatric cardiology and

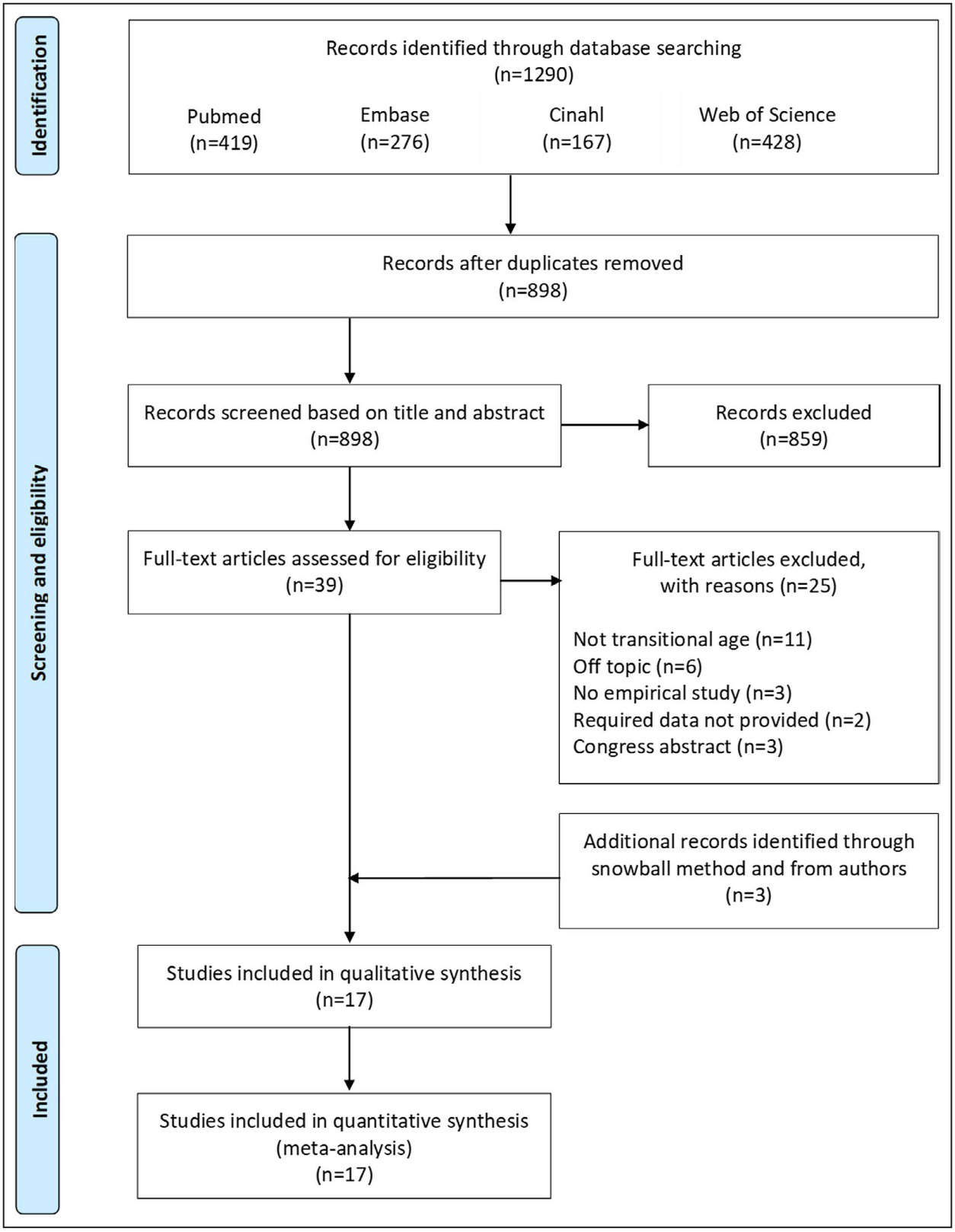

Figure 1. PRISMA flowchart of article selection.
PRISMA indicates preferred reporting items for systematic reviews and meta-analyses.

Table. Methodological Characteristics of the Included Studies

Author, y	Country	Sample Size for Analysis	#Simple Defects*	#Moderate Defects*	#Complex Defects*	Concept	Operationalization	Age Range of Outcome Assessment
Reid, 2004[15]	Canada	234	…	…	234	Successful transfer†	At least 1 visit to an ACHD center of the Canadian Adult Congenital Heart network	18–22 y
Yeung, 2008[6]	USA	158	…	88	57	Lapse of care‡	Intervals of >2 y between the last pediatric and the first adult CHD visit	18 y–§
Mackie, 2009[16]	Canada	292	NR	NR	NR	Cardiology follow-up†	Any outpatient assessment by a cardiologist documented by billing data	18–22 y
Goossens, 2011[17]	Belgium	785	268	444	73	No follow-up‡	Currently not being in cardiac care was confirmed by self-report	16–25 y
Norris, 2013[18]	USA	158	…	86	67	Retention in care†	Any cardiology clinic visit within 2 y of the study interview	19–28 y
Gurvitz, 2013[19]	USA	922	234	447	206	Gaps in cardiology care‡	A >3-y interval between any cardiology appointments (internal medicine, pediatric, or adult congenital cardiology)	18 y–§
Goossens, 2015[20]	USA	230	94	75	61	Cardiac follow-up†	At least 1 outpatient visit documented or self-reported	18–23 y
						Not being in cardiac follow-up‡	Complete cessation of cardiac care confirmed	
Bohun, 2016[21]	USA	229	77	102	50	Lost to follow-up‡	Not seen by any provider in the institution	18 y–§
Harbison, 2016[22]	USA	33	11	16	6	Successful transfer†	Appointment with an adult cardiac provider within 2 y following the last pediatric cardiology visit.	18–20 y
Goossens, 2018[23]	Canada	2630	…	…	2630	Cardiac surveillance†	At least 1 visit to any specialized pediatric or CHD cardiologist documented in the Quebec CHD database	12–24 y
Hergenroeder, 2018[24]	USA	30 CG	…	10	19	Lapse of care‡	Intervals of >3 y between the last pediatric and the first adult CHD visit	18–28 y
Kollengode, 2018[25]	USA	58	16	26	11	Maintenance of care†	At least 1 ambulatory assessment by a cardiac provider within the same healthcare system <3 y after index visit	18 y–§
						Loss of follow-up‡	No documented ambulatory visits with a cardiac provider within the same healthcare system for 3 y at the time of chart review	
Mackie, 2018[26]	Canada	63 CG	…	49	14	Excess time‡	The time interval of >3 mo between the final pediatric visit and the first adult visit minus the recommended time interval between these visits suggested by the pediatric cardiologists	18–20.5 y
Vaikunth, 2018[27]	USA	67	5	38	24	Successful transfer†	At least 1 visit in the ACHD clinic at the adult hospital	18–23 y

(*Continued*)

Table. Continued

Author, y	Country	Sample Size for Analysis	#Simple Defects*	#Moderate Defects*	#Complex Defects*	Concept	Operationalization	Age Range of Outcome Assessment
Gaydos, 2020[28]	USA	54 [CG]	9	21	13	Lost to follow-up[‡]	Persistent absence from cardiac care for at least 6 mo beyond the recommended follow-up time and without an upcoming visit scheduled or documentation of external transfer of care.	18–20.5 y
Mondal, 2020[29]	Canada	279	137	104	38	Successful transfer[†]	Attendance at the Adult Congenital Cardiac Clinic within 2 y of discharge from pediatric cardiology	17–26 y
Skogby, 2020[30]	Sweden	630	228	309	93	Continuity of care[†]	At least 1 cardiac follow-up visit within the 5-y period after intended transfer documented in the medical records or self-reported	18–23 y

ACHD indicates adult congenital heart disease; CG, control group; and NR, not reported/not retrievable.
*Derived from the articles or provided by the authors does not always sum up to the total sample size.
†Formulated in terms of continuity.
‡Formulated in terms of discontinuity.
§Upper age limit not determined.

followed them up in adult care,[15–18,20–22,25–30] whereas 4 studies recruited patients in adult care.[6,19,23,24] Three of the included studies evaluated the impact of a transition program on continuity of care, 1 of which used a randomized controlled trial design,[26] and 2 studies used a pre–posttest design.[24,28]

Quality and Publication Bias Assessment

Based on the NOS, the overall quality of the studies was moderate to high (Table S2). Ten of the 17 studies obtained a maximum score. These studies analyzed data on the entire cohort or relied on databases. Assessment of outcomes was the criterion that 7 studies did not fulfill because they (partly) used self-report to determine whether patients were in follow-up or not. Three studies did not fulfill the criterion of adequacy of follow-up because they had a substantial proportion of patients that were untraceable (Table S2).

Figure S1 represents the funnel plot for included studies. Both the funnel plot and the Egger's test (P=0.2205) did not indicate asymmetry, meaning there is no evidence of publication bias.

Discontinuation of Care

The proportions of discontinuity of care range between 3.6% and 62.7%. Random effects meta-analysis showed that the pooled estimated proportion of discontinuation of care was 26.1% (95% CI, 19.2%–34.6%) (Figure 3).[6,15–30] There was substantial heterogeneity between the studies (Q=600, P<0.0001, I^2=97%), confirming that the random-effects model is preferred above a fixed-effects model. Moderator analysis demonstrated that the type of definition did not significantly impact the proportion of discontinuation of care (QM=2.79, P=0.2476). On the other hand, the place of recruitment (pediatrics or adult care) seemed to be a significant moderator (QM=4.80, P=0.0284) (recruitment pediatrics 21.7% [95% CI, 13.2%–33.7%]; recruitment adult care 43.7% [95% CI, 34.3%–53.5%]). The sensitivity analysis showed that in the worst case, when all untraceable patients are assumed not to be in follow-up, the pooled estimated proportion of discontinuation of care was 31.9% (95% CI, 24.8%–40.0%; Q=568, P<0.0001, I^2=97%). In this scenario, the pooled estimate was not moderated by the definition (QM=0.67; P=0.7141) or the place of recruitment (QM=2.02; P=0.1550).

Disease Complexity

The proportions of patients with discontinuity of care varied across different levels of heart defect complexity. The pooled estimated proportion of patients with simple heart defects was 33.7% (95% CI, 15.6%–58.3%) (Figure 4).[17,19–22,25,27–30] For people with moderately complex defects, the proportion was 25.7% (95%

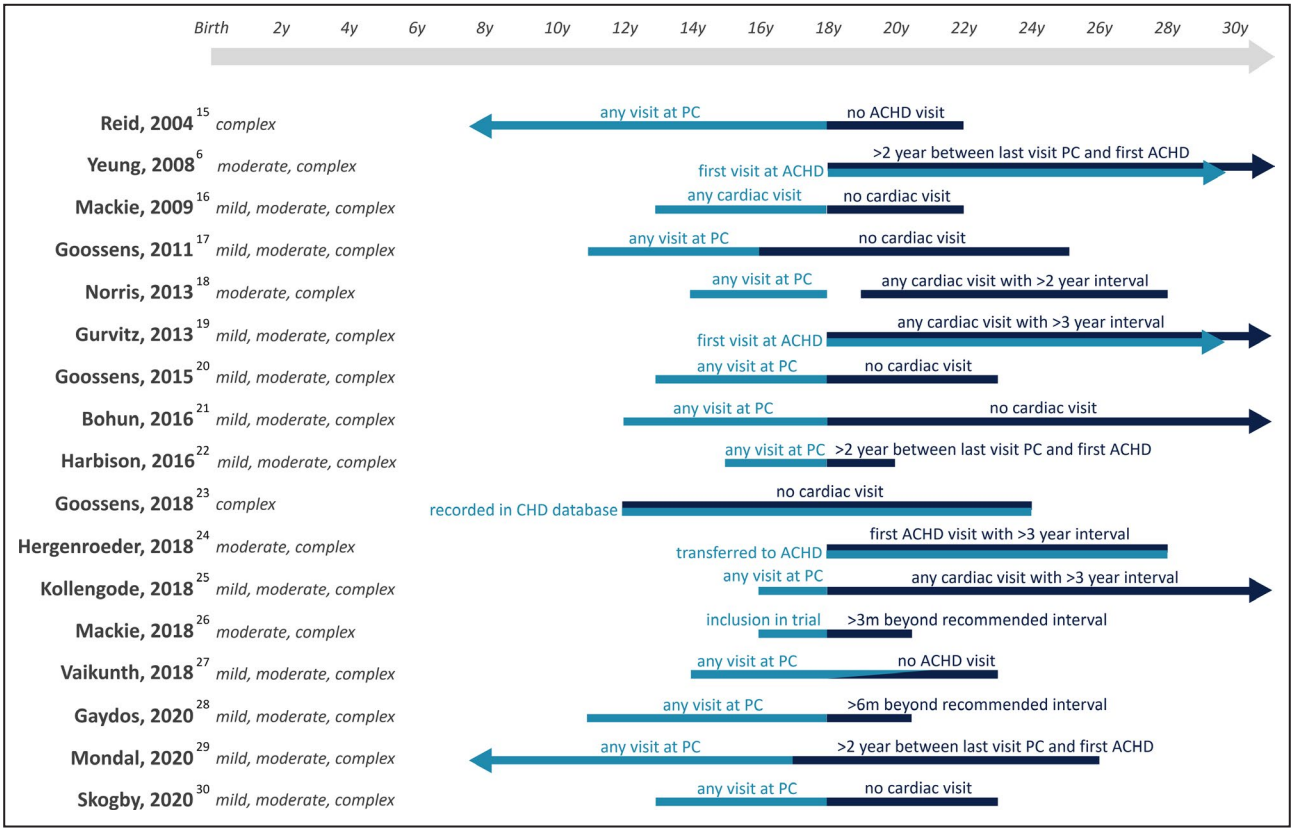

Figure 2. Graphical depiction of the inclusion of eligible patients (light blue) and the assessment of discontinuation of care (dark blue) in the 17 included studies.
ACHD indicates adult congenital heart disease; CHD, congenital heart disease; and PC, pediatric cardiology.

CI, 15.2%–40.1%).[6,17–22,24–30] For complex heart defects, the pooled discontinuity proportion was 22.3% (95% CI, 16.5%–29.4%) (Figure 4).[6,15,17–30] However, the moderator effect was not statistically significant (QM=2.88, *P*=0.2372).

Regional Differences

The pooled estimated proportion of discontinuity is significantly different across regions (QM=15.83, *P*=0.0004). Studies conducted in the United States yielded a pooled estimated proportion of 34.0% (95% CI, 24.3%–45.4%) (Figure 5).[6,15–30] Canadian data showed a pooled estimated proportion of 25.7% (95% CI, 17.0%–36.7%). The pooled estimated proportions from European studies were 6.5% (95% CI, 5.3%–7.9%). (Figure 5).

For the sensitivity analysis, we investigated regional differences in studies that solely recruited patients at pediatrics, since the place of recruitment was found to be a significant moderator. Even in studies that recruited patients at pediatrics, significant differences between the United States, Canada, and Europe were observed (QM=6.89, *P*=0.0320). Regional differences were confirmed within simple (QM=13.90, *P*=0.0002), and moderate heart defects (QM=10.58, *P*=0.0011). A borderline significant moderator effect of region was found in complex heart defects (QM=3.26, *P*=0.0710). These sensitivity analyses indicate that the regional differences were not confounded by variation in place of recruitment and disease complexity across the regions.

Impact of Transition Programs

The pooled estimated proportion of discontinuity of care in the intervention groups of the 3 studies that evaluated the impact of a transition program was 12.7% (95% CI, 2.8%–42.3%) (Figure 6).[24,26,28] In the control groups, this proportion was 36.2% (95% CI, 22.8%–52.2%). This difference did not reach statistical significance (QM=2.53, *P*=0.1119).

DISCUSSION

For people with chronic conditions, the transition to adulthood is a vulnerable period for discontinuity of care.[4] To date, 17 empirical studies on discontinuity of care in young people with CHD have been published. The pooled estimated proportion of

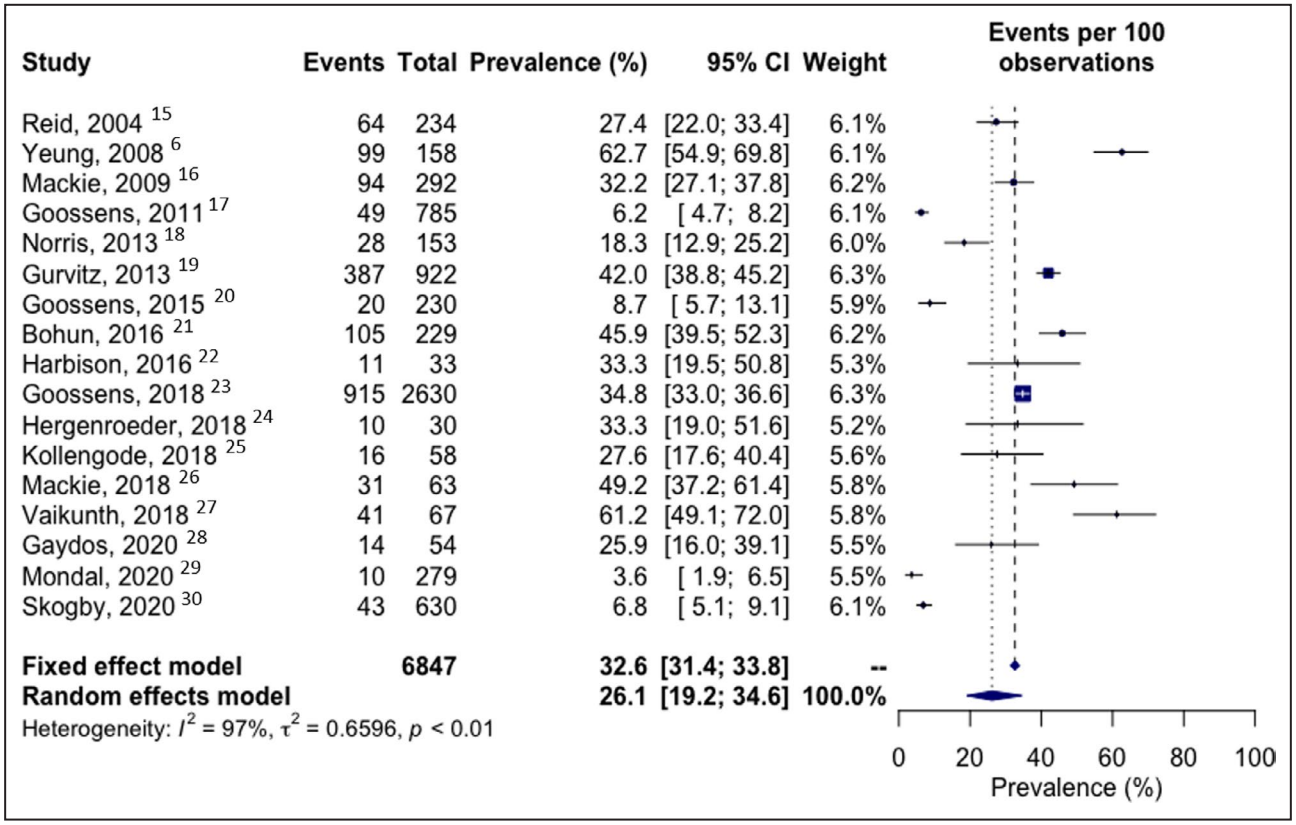

Figure 3. Forest plot for discontinuity of care in people with congenital heart disease at the transitional age.

discontinuity in these studies was 26.1%. In a worst-case scenario, the pooled estimate was 31.9%. It can be expected that the true discontinuity proportion is situated within this range. The results are summarized in Figure 7.

As expected, great heterogeneity across the studies was found. Sources of this heterogeneity were inherent to the methodology used (ie, the place of recruitment). However, even when uniform methodologies were used, variability across centers could be observed. For instance, 2 multicenter studies found ranges between 21%–61%[19] and 0%–12.7%[30] in the participating centers. This suggests that organizational factors in different hospitals may play a role. Indeed, a study including 7 centers in Sweden showed that higher outpatient volumes at pediatric outpatient clinics were associated with better continued follow-up care after transfer.[30] When untraceable patients were included in the analysis (ie, worst-case scenario), the outpatient volumes at ACHD were also predictive.[30] The relationship between outpatient volumes and continuity of care could be explained by the fact that centers with high outpatient volumes are more likely to provide full-time dedicated staff for their patients with CHD compared with centers with lower volumes, where staff often need to combine caring for patients with CHD with caring for patients with other conditions.[30] Moreover, it is believed that dedicated administrative staff and program managers also play a critical role in keeping patients in care.[30]

Healthcare system factors are also alleged to impact on continuity of care, but this has not been investigated to date.[4,30] The present meta-analysis allowed us to estimate the pooled proportions for different countries/regions. The United States had the highest proportion of discontinuity, while European studies revealed a significantly lower one. It could be hypothesized that this disparity is because of differences in access to health care. For example, Belgium and Sweden have universal access to health care, in contrast to the United States. However, access is probably not a strong protective factor, as suggested by several Canadian studies, which demonstrated high proportions of discontinuity even though Canada has universal access to health care. Another explanatory factor could be the adoption of a systematic transfer to adult care. A survey among US and European centers on transfer practices[36] revealed that 68% of the US centers and 81% of the European centers systematically transfer patients from pediatric cardiology to ACHD (data on file). If US centers transfer patients to adult care, this transfer is mandatory in only 16% of the centers, whereas it is mandatory in 85% of the European centers. A recent European survey showed a slight increase in

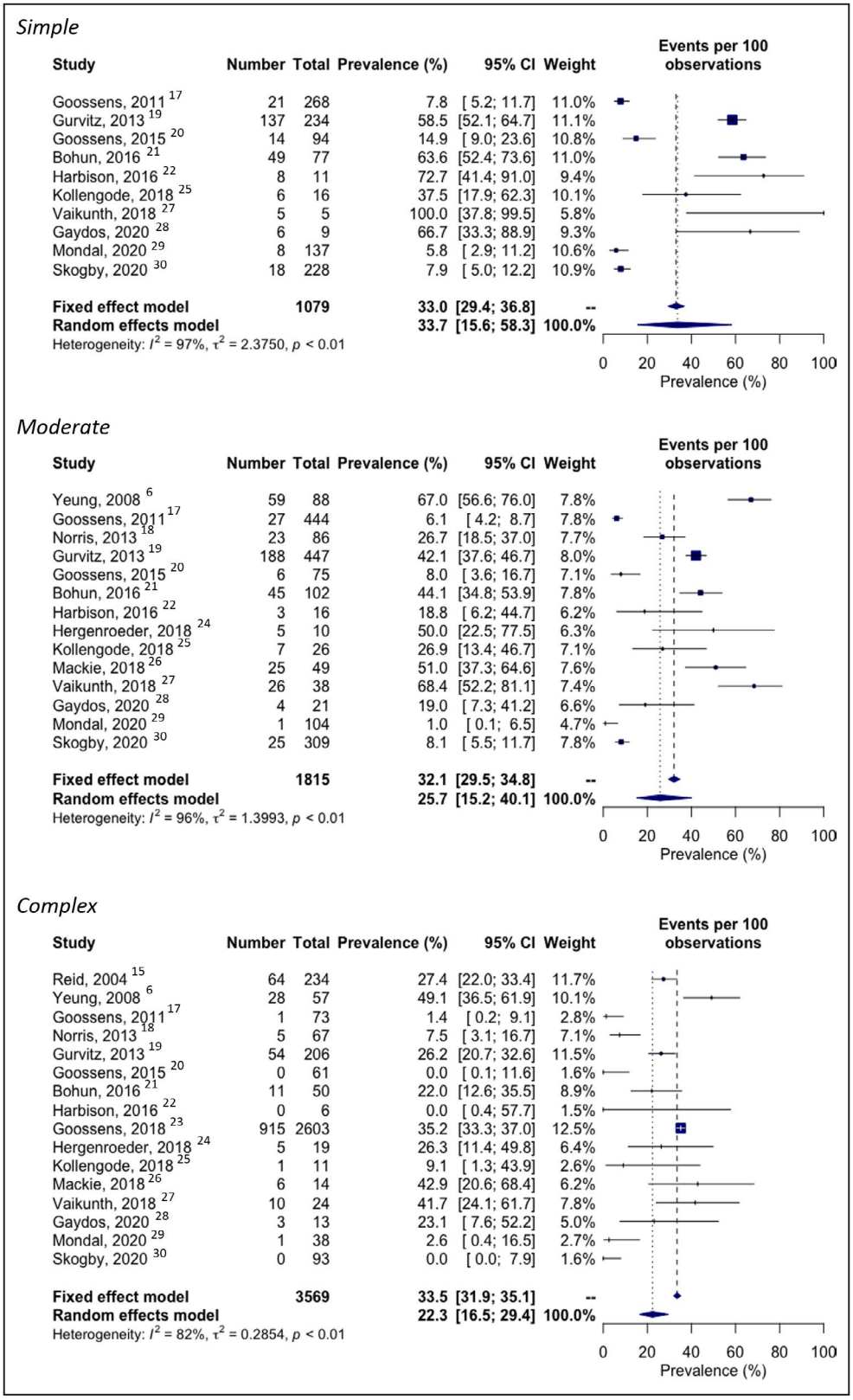

Figure 4. Forest plots for discontinuity of care in people with congenital heart disease at the transitional age, by complexity of the heart defect.

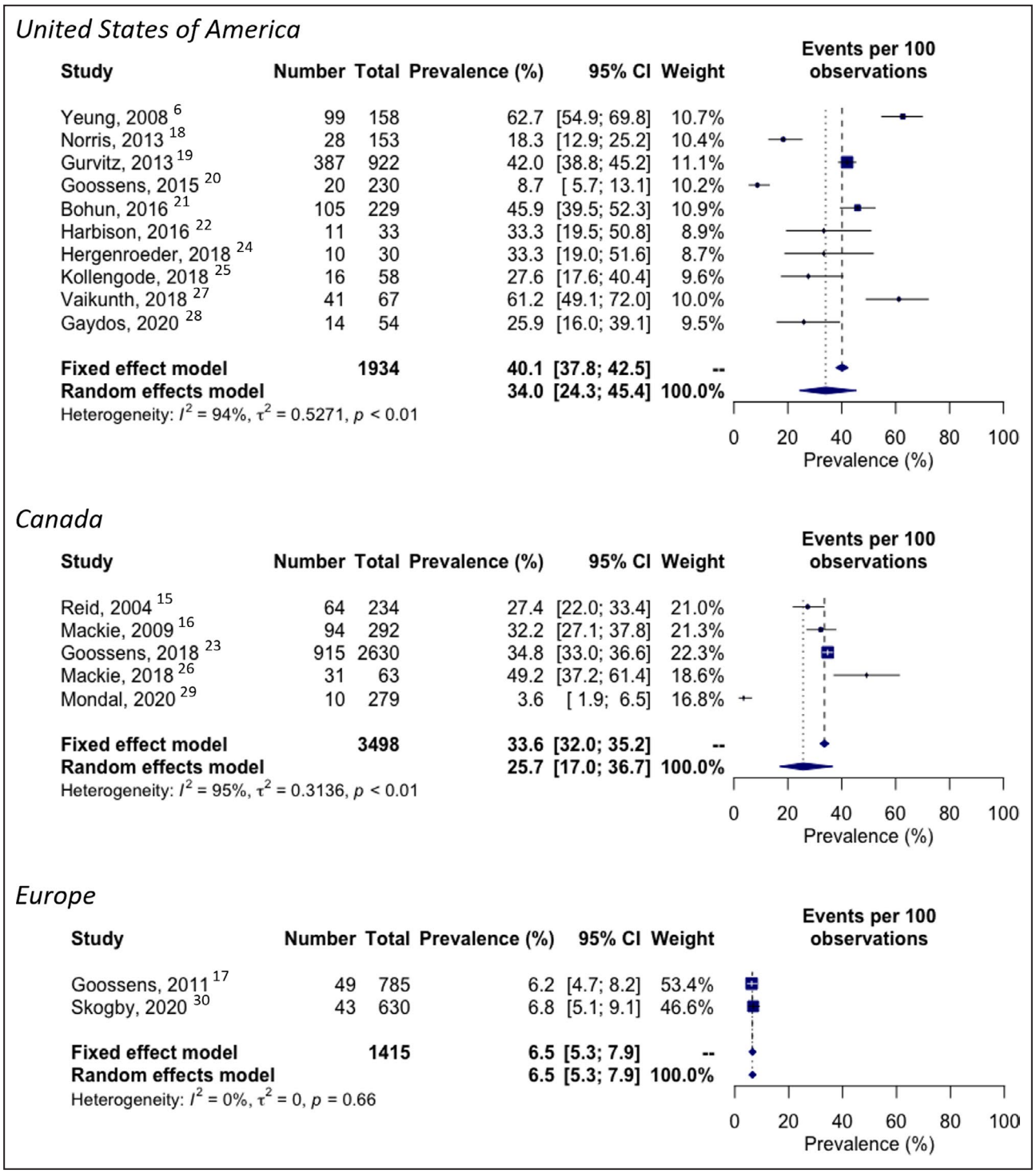

Figure 5. Forest plots for discontinuity of care in people with congenital heart disease at the transitional age, by region of the study.

the proportion of centers that formally transfer patients to adult care.[37] The size of the country and the population density are factors that should be taken into account as well. For instance, Belgium is a small country with a population density (991 per mile^2) that is more than 10-fold that of the United States (92 per mile^2), Sweden (64 per mile^2), or Canada (11 per mile^2). Studies in the United States[20–22] and Belgium[17] did not find distance to the ACHD center to be a significant predictor for discontinuity of care. However,

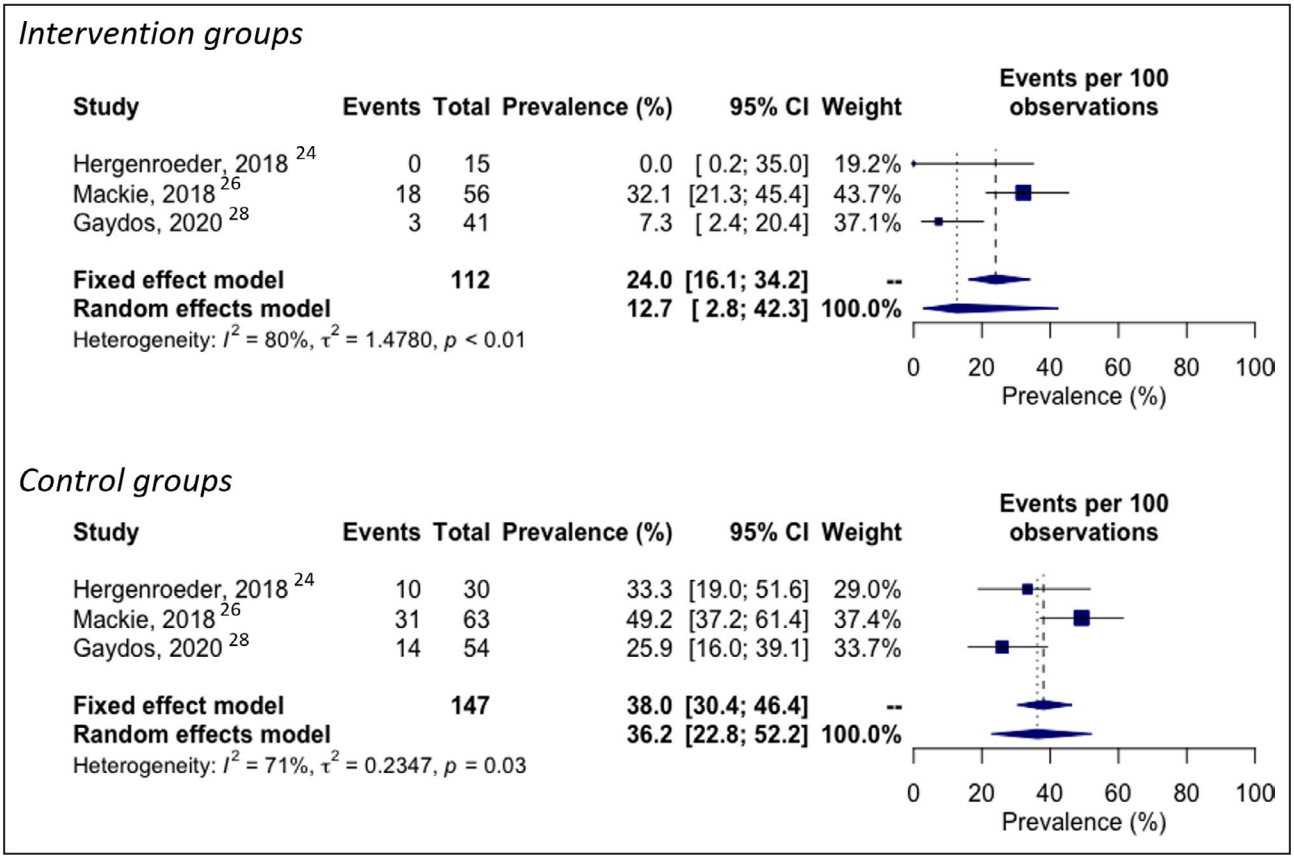

Figure 6. Forest plots for discontinuity of care in people with congenital heart disease following a transition program vs usual care.

in Canada, it appears to be more explanatory,[15,29] given the magnitude of the country. Nonetheless, accessibility to ACHD care remains an issue of concern. For instance, in the United States, ≈45% of the population is estimated to live >1 hour to an ACHD center, and 5.4% was living >4 hours away.[38] Such patients require specific attention to avoid discontinuity of care.

Proportions of discontinuity of care are higher in groups of patients with simple heart defects. This is in line with prior findings for CHD and other childhood-onset conditions, such as sickle-cell disease, adrenal hyperplasia, or juvenile idiopathic arthritis, where milder subtypes were also associated with more discontinuation.[4] Other patient-related factors found to increase the risk of discontinuation are male sex, lower socio-economic status, too young when transferred, fewer pediatric outpatient visits in the pretransfer period, last visit in a nonuniversity setting, missed appointments, poor health behaviors, and absence of written recommendation for follow-up care.[4]

Clinicians, managers, and administrators are urged to implement strategies for keeping patients in the system. A systematic and mandatory transfer to adult care and the implementation of transition programs would be beneficial. We found that the discontinuity proportion was systematically lower in groups of patients enrolled in a transition program than in patients receiving usual care. Admittedly, there were only 3 studies that compared patients with or without transitional care. Ongoing trials on transition programs in CHD will likely strengthen the body of knowledge.[8,39–41]

Methodological Considerations

The findings of this systematic review and meta-analysis should be interpreted in light of some methodological considerations. First, we studied discontinuity of care at the transitional age. Studies on discontinuity of care during childhood or throughout adulthood were not included. Consequently, the present review does not allow conclusions to be drawn regarding continuity of care over the entire lifespan. Second, only full research articles were eligible and conference abstracts were excluded because detailed information on the conceptualization and operationalization is lacking in such abstracts. Our search showed that there were only 2 conference abstracts that had not been

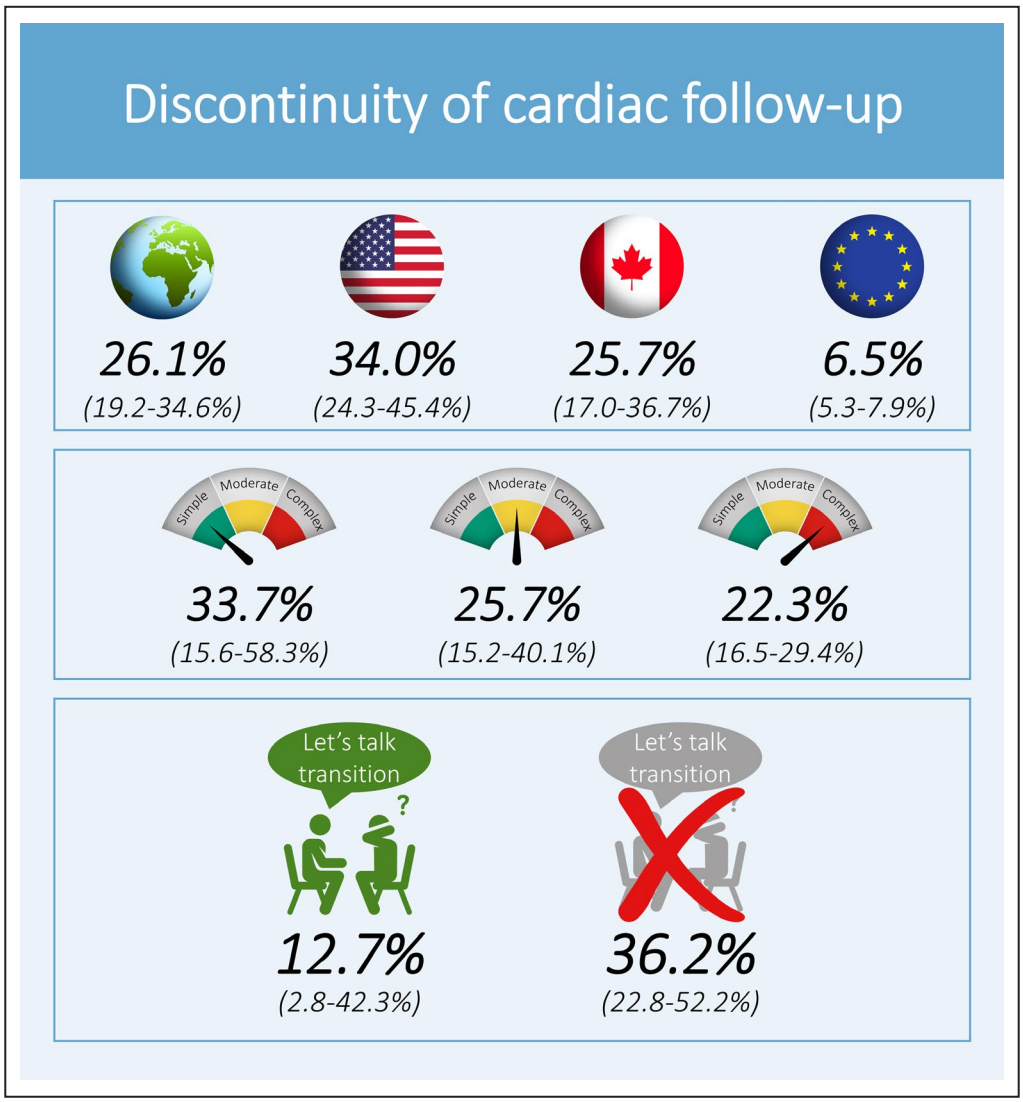

Figure 7. Discontinuity of care in people with congenital heart disease at the transitional age, globally, by region of the study, by complexity, and by implementation of transition programs.

published as a full article later on. Post hoc sensitivity analysis showed that our findings are not biased by excluding these conference abstracts. Third, we observed great heterogeneity between the studies, which highlights the importance of investigating sources for heterogeneity. Although some significant moderators were identified, there is still substantial heterogeneity that warrants further investigation. Factors that could be scrutinized in future studies are hospital-related factors, such as staffing, case-load, or features of transfer policies.[37,42] Fourth, some moderator analyses did not reveal a statistically significant effect, despite considerable disparities between groups. This is probably because of the fact that some subgroups are small and therefore the data are underpowered. More specifically, studies that evaluate the impact of transition programs on reducing discontinuity of care are scant. Fifth, there were 4 studies in which patients were enrolled at ACHD. These studies were able to assess the delay in transfer to adult care, but they did not have data on patients who did not make it to ACHD at all. Hence, the worst-case scenario is possibly a bit worse than what we were able to estimate. As a result, the range in which the true proportion of discontinuity lies is likely somewhat broader, with a higher upper limit. Sixth, we included studies irrespective of their quality assessment. The risk of bias, however, was low to moderate, and no studies with a high risk of bias were found. Seventh, Belgium and Sweden were the only European countries included in this review and these 2 countries are not necessarily representative of Europe. Consequently, more research in Europe is needed to test whether the relatively low discontinuity of care also holds true in other European countries. Eighth, we

could not pinpoint specific healthcare system factors in this meta-analysis. However, research on healthcare system factors that are protective or entail a risk of discontinuity of care is needed. International research using a uniform methodology is required to fill the current gaps in our knowledge.[43] From this perspective, it is important that robust studies on discontinuity of care also be conducted in South America, Asia, and Africa because to date, these regions are missing in the empirical body of knowledge. Specific funding to conduct such research in low-resource areas must be allocated.

CONCLUSIONS

In this systematic review and meta-analysis, we identified 17 studies that investigated discontinuity of care in young people with CHD at the transitional age. Our findings demonstrated a high proportion of discontinuity of care, with high heterogeneity across the studies. European studies showed significantly lower proportions of discontinuity than American or Canadian studies. A trend towards more discontinuity was observed for patients with simple heart defects. The high proportion of discontinuity of care here revealed urges implementation of preventative interventions. Transition programs show the potential to reduce discontinuity of care, although more research is needed to draw firm conclusions. The present review and meta-analysis should give new impetus to investigating and implementing interventions that reduce discontinuity of care in this vulnerable group of patients.

ARTICLE INFORMATION

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Disclosures

None.

Supplementary Material

Tables S1–S2
Figure S1

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SUPPLEMENTAL MATERIAL

Table S1. Search strings.

Pubmed (“Child”[Mesh: NoExp] OR child[tiab] OR child's[tiab] OR children[tiab] OR childhood[tiab] OR children's[tiab] OR kid OR kid's OR girl OR girls OR boy OR boys OR adolescents OR Adolescence OR teen OR teens OR teenager OR teenagers OR youth OR youths OR youngster*[tiab] OR adult child OR minors OR young adults OR young adul* OR emerging adul* OR junior high OR middle-school OR high-school OR juvenile OR juveniles OR “Pediatrics”[Mesh: NoExp] OR Pediatrics[tiab] OR Pediatric[tiab]) AND ("congenital heart"[All Fields] OR "congenital cardiac"[All Fields] OR "heart defects"[All Fields] OR Fallot[All Fields] OR ("transposition"[All Fields] AND "great arteries"[All Fields]) OR ("aortic coarctation"[MeSH Terms] OR ("aortic"[All Fields] AND "coarctation"[All Fields]) OR "aortic coarctation"[All Fields] OR "coarctation"[All Fields]) OR Eisenmenger[All Fields] OR "septal defect"[All Fields] OR "septal-defects"[All Fields] OR "atrial septal defect"[All Fields] OR "ventricular septal defect"[All Fields] OR "congenital aortic stenosis"[All Fields] OR "congenital pulmonary stenosis"[All Fields] OR univentricular[All Fields] OR "single ventricle"[All Fields] OR "hypoplastic left heart"[All Fields] OR "tricuspid atresia"[All Fields] OR "pulmonary atresia"[All Fields] OR "anomalous pulmonary venous"[All Fields] OR "truncus arteriosus"[All Fields] OR "ductus arteriosus"[All Fields] OR Fontan[All Fields] OR "double outlet"[All Fields] OR "double inlet"[All Fields] OR Ebstein[All Fields]OR “anomalous aortic”[All Fields] OR “anomalous coronary”[All Fields] OR “interrupted aortic”[All Fields] OR "congenital aortic valve"[All Fields] OR "congenital pulmonary valve"[All Fields]) AND (continuity of patient care[tiab] OR “continuity of care” OR “continuation of care” OR “discontinuation of care” OR “transition to adult care” OR “Care Continuum” OR “Care Continuity” OR “loss to follow up” OR “loss to follow-up” OR “lost to follow up” OR “lost to follow-up” OR “care gap” OR “care gaps” OR “gaps in care” OR “lapse of care” OR “lapses of care” OR “transfer of care” OR “transition to adult care” OR healthcare transition*[tiab] OR health care transition*[tiab] OR “lifelong care” OR “life long follow-up” OR “lifelong follow-up” OR “transitional care” OR “successful transfer” OR untraceable OR untraceability) AND (english[Language] OR spanish[Language] OR french[Language] OR dutch[Language] OR german[Language] OR swedish[Language])
EMBASE ('child' OR 'child':ti OR 'child':ab OR 'children':ti OR 'children':ab OR 'childhood':ti OR 'childhood':ab OR kid OR girl OR boy OR girls OR boys OR adolescents OR adolescence OR teen OR teens OR teenager OR teenagers OR youth OR youths OR minors OR young NEXT/1 adult OR young adults OR young adulthood OR youngster*:ti OR youngster*:ab OR adult child OR junior high OR 'middle school' OR 'middleschool' OR 'high-school' OR juvenile OR juveniles OR pediatrics OR 'pediatrics'/de OR 'pediatric':ti OR 'pediatric':ab OR 'pediatrics':ti OR 'pediatrics':ab) AND ('congenital heart disease'/exp OR 'congenital heart' OR 'congenital cardiac' OR heart NEAR/1 defect* OR ‘fallot’ OR transposition NEAR/1 “great arteries” OR aort* NEAR/1 coarct* OR eisenmenger OR septal NEAR/2 defect* OR congenit* NEAR/2 stenos* OR aort* AND near AND stenos* OR univentricul* OR 'single ventricle' OR 'hypoplastic left heart' OR (tricuspid OR pulmonar*) NEAR/1 atresia OR 'pulmonary vein malformation'/exp OR (anomalous AND pulmonary NEAR/1 (vein* OR venous)) OR (ductus OR truncus) NEAR/1 arteriosus OR fontan OR double NEAR/1 (inlet* OR outlet*) OR ebstein OR anomalous NEAR/1 (aort* OR coronar*) OR interrupt* NEAR/1 aort* OR congenital NEAR/2 valve*) AND (‘continuity of patient care’ OR ‘continuity of care’ OR ‘continuation of care’ OR ‘discontinuation of care’ OR 'transition to adult care' OR 'patient care' OR ‘care continuum’ OR ‘care continuity’ OR ‘loss to follow up' OR ‘lost to follow up' OR ‘loss to follow-up' OR ‘lost to follow-up' OR ‘care gap’ OR ‘care gaps’ OR ‘lapse of care’ OR ‘lapses of care’ OR ‘lapses in care’ OR ‘transfer of care’ OR 'healthcare transition' OR ‘health care transition’ OR ‘lifelong care’ OR ‘life-long care’ OR ‘lifelong

follow up' OR ‘lifelong follow-up' OR ‘transitional care’ OR ‘successful transfer’ OR untraceable OR untraceability)
AND
([english]/lim OR [spanish]/lim OR [french]/lim OR [dutch]/lim OR [german]/lim OR [swedish]/lim)

CINAHL

(((“Child”[Mesh: NoExp] OR child[tiab] OR child's[tiab] OR children[tiab] OR childhood[tiab] OR children's[tiab] OR kid OR kid's OR girl OR girls OR boy OR boys OR adolescents OR Adolescence OR teen OR teens OR teenager OR teenagers OR youth OR youths OR youngster*[tiab] OR adult child OR minor OR young adults OR young adul* OR emerging adul* OR junior high OR middle-school OR high-school OR juvenile OR juveniles OR “Pediatrics”[Mesh: NoExp] OR Pediatrics[tiab] OR Pediatric[tiab]))
AND
(((MH "Heart Defects, Congenital+") OR 'congenital heart' OR ‘congenital cardiac’ OR (heart N1 defect*) OR Fallot OR (transposition N1 ‘great arteries’) OR (MH "Aortic Coarctation") OR (aort* N1 coarct*) OR eisenmenger OR septal N2 defect* OR (congenit* N2 stenos*) OR (aort* N1 stenos*) OR univentricul* OR ‘single ventricle’ OR ‘hypoplastic left heart’ OR (tricuspid OR pulmonar*) N1 atresia OR (anomalous AND (pulmonary N1 (vein* OR venous))) OR ((ductus OR truncus) N1 arteriosus OR Fontan OR (double N1 (inlet* OR outlet*)) OR Ebstein OR (anomalous N1 (aort* OR coronar*)) OR (interrupt* N1 aort*) OR (congenital N2 valve*)))
AND
((continuity of patient care[tiab] OR “continuity of care” OR “continuation of care” OR “discontinuation of care” OR “transition to adult care” OR “patient care” OR “Care Continuum” OR “Care Continuity” OR “loss to follow up” OR “loss to follow-up” OR “lost to follow up” OR “lost to follow-up” OR “care gap” OR “care gaps” OR “transfer of care” OR healthcare transition*[tiab] OR health care transition*[tiab] OR “lifelong care” OR “life long follow-up” OR “lifelong follow-up” OR “transitional care” OR “successful transfer” OR untraceable OR untraceability))
AND LA
((english OR spanish OR french OR dutch OR german OR swedish))

Web of Science

ALL=(child OR child's OR children OR childhood OR children's OR kid OR kid's OR girl OR girls OR boy OR boys OR adolescents OR Adolescence OR teen OR teens OR teenager OR teenagers OR youth OR youths OR youngster* OR adult child OR minors OR young adults OR young adul* OR emerging adul* OR junior high OR middle-school OR high-school OR juvenile OR juveniles OR Pediatric*)
AND
ALL=("congenital heart" OR "congenital cardiac" OR "heart defects" OR Fallot OR "transposition" AND "great arteries" OR "aortic coarctation" OR "coarctation of the aorta" OR Eisenmenger OR "septal defect" OR "septal-defects" OR "atrial septal defect" OR "ventricular septal defect" OR "congenital aortic stenosis" OR "congenital pulmonary stenosis" OR univentricular OR "single ventricle" OR "hypoplastic left heart" OR "tricuspid atresia" OR "pulmonary atresia" OR "anomalous pulmonary venous" OR "truncus arteriosus" OR "ductus arteriosus" OR Fontan OR "double outlet" OR "double inlet" OR Ebstein OR “anomalous aortic” OR “anomalous coronary” OR “interrupted aortic” OR "congenital aortic valve" OR "congenital pulmonary valve")
AND
ALL=(“continuity of patient care” OR “continuity of care” OR “continuation of care” OR “discontinuation of care” OR “transition to adult care” OR “patient care” OR “Care Continuum” OR “Care Continuity” OR “loss to follow up” OR “loss to follow-up” OR “lost to follow up” OR “lost to follow-up” OR “care gap” OR “care gaps” OR “transfer of care” OR “healthcare transition*” OR “health care transition*” OR “lifelong care” OR “life long follow-up” OR “lifelong follow-up” OR “transitional care” OR “successful transfer” OR untraceable OR untraceability)
AND LANGUAGE: (English OR Dutch OR French OR German OR Spanish OR Swedish)

Table S2. Quality assessment of included studies using the Newcastle-Ottawa Scale (NOS) for cohort studies .

Study	Selection				Comparability	Outcome			Total quality score
	S1	S2	S3	S4	C1	E1	E2	E3	
Reid, 2004	*	NR	*	*	NR	—	*	—	4/6
Yeung, 2009	*	NR	*	*	NR	*	*	*	6/6
Mackie, 2009	*	NR	*	*	NR	*	*	*	6/6
Goossens, 2011	*	NR	*	*	NR	—	*	*	5/6
Norris, 2013	*	NR	*	*	NR	—	*	—	4/6
Gurvitz, 2013	*	NR	*	*	NR	—	*	*	5/6
Goossens, 2015	*	NR	*	*	NR	—	*	—	4/6
Bohun, 2016	*	NR	*	*	NR	*	*	*	6/6
Harbison, 2016	*	NR	*	*	NR	*	*	*	6/6
Goossens, 2018	*	NR	*	*	NR	*	*	*	6/6
Hergenroeder, 2018	*	NR	*	*	NR	*	*	*	6/6
Kollengode, 2018	*	NR	*	*	NR	—	*	—	4/6
Mackie, 2018	*	NR	*	*	NR	*	*	*	6/6
Vaikunth, 2018	*	NR	*	*	NR	*	*	*	6/6
Gaydos, 2020	*	NR	*	*	NR	*	*	*	6/6
Mondal, 2020	*	NR	*	*	NR	*	*	*	6/6
Skogby, 2020	*	NR	*	*	NR	—	*	*	5/6

S1 Representativeness of the exposed cohort; S2 Selection of the non-exposed cohort; S3 Ascertainment of exposure; S4 Outcome not present at start of study; C1 Comparability of cohorts on the basis of the design or analysis; E1 Assessment of outcome, E2 Long enough follow-up for outcome to occur; E3 Adequacy of follow-up; NR=Not relevant.

Figure S1. Funnel plot for the 17 included studies.