

# Continued follow-up care

*- during transition and transfer of young people with  
congenital heart disease*

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# Abstract

Although life-long follow-up care and transfer from paediatric to adult facilities are recommended for most young people with congenital heart disease, a substantial proportion present with discontinued follow-up care, which is associated with adverse outcomes and requires active prevention.

**The overall aim** of this thesis was to scrutinize discontinuation of follow-up care among youths with congenital heart disease and gain a deeper understanding of associated factors on patient, hospital, and healthcare system levels, as well as provide increased clarity around the concept of discontinuation of follow-up care.

**The methods applied** include register and survey-based statistical analyses (I & III), individual semi-structured interviews with patients and healthcare providers subjected to qualitative content analysis (II & III) and a conceptual inquiry of the concept of discontinuation in published literature using a modified summative content analysis and a Delphi-inspired expert evaluation (IV).

**Findings:** Study I showed a relatively low proportion of discontinuation in the included centres, clear differences in proportions of discontinuation across hospitals and an association between paediatric outpatient volumes and continued follow-up care after transfer. The difference in proportions of discontinuation across the hospitals indicates the relevance of hospital-related factors influencing discontinuation of follow-up care. In Study II, patients' perceptions and experiences of factors affecting continued follow-up care after transfer included three main categories, "Motivation for follow-up care", "Participation in care and sense of connectedness with health care provider" and "Care accessibility". Study III showed a low proportion of no follow-up at two Belgian centres. Furthermore, healthcare providers' perceptions and experiences of factors perceived to affect continued follow-up care after transfer included the categories "Care structure", "Care processes" and "Patients' characteristics and circumstances". In Study IV, three different types of discontinuation of follow-up care emerged from the analysis of published

literature, including “Untraceability”, “Lost to follow-up care” and “Gap(s) in follow-up care”, the latter two being distinguished from one another through time intervals versus time periods and whether patients were currently engaged in care or not.

**In conclusion,** a plethora of factors affecting continuity of patient care was raised, including patient, hospital, and healthcare system factors, which likely trigger each other, creating synergetic effects. Agreement between patients and health care providers regarding relevant factors was high and many of the factors raised could possibly be tackled through clear structures, provision of sufficient health care provider resources, competencies, and skills, as well as transitional care interventions. Furthermore, expansion of the concept of discontinuation of follow-up care to include personal experiences as well as informational and relation aspects, is deemed mandatory.

**Keywords:** adolescent; continuity of patient care; cross-sectional study; expert evaluation; heart defects, congenital; individual interviews; logistic regression; patient transfer; qualitative descriptive study; qualitative content analysis; summative content analysis; young adult







# Sammanfattning på Svenska

Överföring från barn till vuxensjukvård är på många vis komplext då det innefattar dels en praktisk och fysisk förflyttning av vård och behandling, dels ett avslut av etablerade vårdrelationer och dels ger upphov till en förändring i vårdkultur och miljö för patienten. Under överföringsperioden tacklar ungdomar med långvarig sjukdom dessutom en utvecklingsrelaterad förändring när de går från att vara barn och ungdom till att vara vuxen. Den utvecklingsrelaterade förändringen är en process som på många sätt kan kompliceras av ett långvarigt sjukdomstillstånd eller en funktionsvarriation. Dessa ungdomar behöver utveckla kunskap och förmåga att i möjligaste mån självständigt hantera alla aspekter av sin hälsa, sin vård och sin behandling.

Ungdomar med långvarig sjukdom, så som till exempel medfödda hjärtfel riskerar att utveckla komplikationer och sämre hälsa om de inte följer den rekommenderade medicinska uppföljningen, som ibland är livslång. Avbrott i den medicinska uppföljningen under och efter överföring från barn till vuxensjukvård är ett känt problem bland unga personer med medfött hjärtfel och kräver förebyggande insatser. Faktorer som kan påverka avbrott i uppföljningen har beskrivits i tidigare forskning och mestadels innefattat patientrelaterade faktorer, så som biologiskt kön, sjukdomens svårighetsgrad eller pågående behandling. Troligtvis finns det även aspekter kopplade till vård, behandling samt hälso-och sjukvården i stort som skulle påverka ungdomarnas möjligheter och inställning till fortsatt uppföljning efter överföring från barn till vuxensjukvård. Vilka faktorer som skulle kunna vara betydelsefulla har studerats närmare i denna avhandling.

Det övergripande syftet med avhandlingen har varit att få en ökad förståelse för vilka faktorer som skulle kunna påverka avbrott i uppföljningen för unga personer med medfött hjärtfel i Sverige och Belgien, men också att ur ett teoretiskt perspektiv problematisera och belysa den konceptuella betydelsen av avbrott i uppföljningen, beskriva hur avbrott kan definieras och vad det egentligen innebär.

Den första studien undersökte andelen avbrott bland unga personer med medfött hjärtfel i Sverige, och visade att vid de sju universitetssjukhusen, så var det knappt 7% av ungdomarna som inte hade haft ett första besök på vuxenhjärtmottagning inom fem år. Studien visade också att det fanns stora skillnader i andelen avbrott mellan de sju sjukhusen, vilket skulle kunna indikera att sjukhusfaktorer påverkar. Studien antydde också ett samband mellan barnhjärtmottagningarnas patientvolym och andelen avbrott, där större patientvolym vid respektive barnhjärtmottagning ökade chansen för fortsatt uppföljning.

I den andra studien genomfördes individuella intervjuer med unga personer som deltagit studie 1, där de fick berätta om vilka faktorer som de själva tror kan påverka hurvida en ung person väljer att fortsätta sin uppföljning eller inte. Tre kategorier av faktorer som upplevdes kunna påverka identifierades, ”Motivation till uppföljning”, ”Känsla av tillhörighet och att få vara delaktig i sin vård” samt ”Vården tillgänglighet”. Deltagarna belyste särskilt vikten av tillräcklig kunskap om sin sjukdom och vikten av fortsatt uppföljning, samt att känna ett tydligt syfte med uppföljningen. Andra faktorer som kunde påverka var pågående behandling och symtom, likväl som vårdpersonalens bemötande och aktiva stöd för att ungdomen ska komma till mottagningen.

I den tredje studien undersöktes först andelen avbrott bland unga personer med hjärtfel vid två universitetssjukhus i Belgien, enligt samma metodologi som studie 1. Resultatet visade att vid de två inkluderade belgiska universitetssjukhusen, så var det drygt 5% av ungdomarna som inte hade haft ett första besök på vuxenhjärtmottagningen inom fem år. Därefter intervjuades kardiologer, sjuksköterskor samt administrativ personal vid både barnkardiologiska mottagningar samt specialistmottagningar för vuxna med medfödda hjärtfel, så kallade adult congenital heart disease (ACHD) mottagningar, i både Sverige och Belgien. Syftet med intervjuerna var att undersöka vilka fak-

torer som vårdpersonalen ser som möjliga stödjande faktorer respektive barriärer för fortsatt uppföljning efter överföring. Vårdpersonalen berättelser bekräftade fynden i studie 2, och deltagarna beskrev hur patientens individuella förutsättningar och omständigheter kan påverka. Till exempel den pågående transitionen till vuxenlivet, motivation och kunskap om sin sjukdom eller sjukdomens svårighetsgrad och pågående behandling. De lyfte även betydelsen av vårdens och mottagningens struktur och organisation så som bemaning, integrerade eller helt specialiserade ACHD mottagningar, och fysiskt avstånd mellan barn och vuxenkliniken. De lyfte också betydelsen av vårdprocesser och vårdrelationer, som till exempel samarbete mellan barn och vuxen, uppsökande verksamhet, specifika insatser för att underlätta den unges transition till vuxenlivet och överföringen till vuxensjukvården, vårdpersonalens kompetenser och administrativa rutiner.

I den fjärde studien undersöktes begreppet avbruten uppföljning utifrån ett teoretiskt och konceptuellt perspektiv. En kvalitativ analys utav termer och definitioner för avbruten uppföljning i den vetenskapliga litteraturen genomfördes, varpå tre olika typer av avbrott kunde identifieras. Med hjälp utav en expertpanel utvärderades de tre typerna av avbrott och konceptuella definitioner formulerades. De tre typerna var, "Glapp i uppföljningen" vilket konceptuellt kan definieras som att tidsintervallet mellan uppföljningsbesök inom en specifik vårdkontext har överskridits, men patienten är förnärvarande under uppföljning, "Avbruten eller utebliven uppföljning" vilket konceptuellt kan definieras som att patienten under en definierad tidsperiod och vårdkontext inte erhållit uppföljning samt att patienten förnärvarande inte är under uppföljning, och slutligen "Ospårbarhet", vilket konceptuellt kan definieras som att patienten inte har gått att kontakta. Det vill säga att uppföljning varken kan bekräftas eller dementeras.

Avhandlingen i sin helhet beskriver avbruten uppföljning i samband med överföringsperioden från barn till vuxensjukvård som ett komplext fenomen med stark koppling till både utvecklingsrelaterade och organisatoriska förändringar men också som ett fenomen som skulle gynnas av en breddad definition, och inte enbart inkludera det mätbara perspektivet kring om uppföljningen har genomförts eller inte utan även inkludera patientens subjektiva upplevelse av kontinuerlig vård. Patienter och vårdpersonal i de inkluderade studierna belyser en lång rad faktorer av möjlig betydelse för

fortsatt uppföljning på så väl patient, sjukhus och sjukvårdssystemnivå, med tyngdpunkt på vårdstrukturer och vårdprocesser som kan skapa förutsättningar för unga patienters kunskap och motivation till fortsatt uppföljning efter överföring.





# Nederlandsalige samenleving

De overgang van de kinder- naar de volwassenenzorg is in veel opzichten complex omdat het een praktische en geografische verplaatsing van zorg en behandeling met zich meebrengt, een verbreking van gevestigde zorgrelaties en aanleiding geeft tot een organisatorische transitie voor de patiënt. Jongeren met een chronische ziekte krijgen tijdens de overgangperiode ook te maken met een ontwikkelingsgerelateerde transitie, van kindertijd naar volwassenheid, een proces dat op veel manieren kan worden bemoeilijkt door chronische ziekte of handicap. Deze jongeren moeten de kennis en vaardigheden ontwikkelen om zelfstandig alle aspecten van hun gezondheid, zorg en behandeling te managen. Jongeren met een chronische ziekte, zoals aangeboren hartafwijkingen, riskeren complicaties en een slechtere gezondheid als ze hun medische opvolging als volwassene niet voortzetten. Onderbrekingen in de medische opvolging tijdens en na de overgang van kindergeneeskunde naar volwassenenzorg is een bekend probleem bij jongeren met een aangeboren hartafwijking en vereist preventieve maatregelen. Factoren die onderbrekingen in de follow-up kunnen beïnvloeden, zijn beschreven in eerder onderzoek en omvatten meestal patiëntgerelateerde factoren, zoals geslacht, ziektecomplexiteit of lopende behandeling. Waarschijnlijk zijn er ook aspecten die verband houden met zorg, behandeling en gezondheidszorg in het algemeen die van invloed zijn op de kansen en houding van jongeren ten aanzien van voortgezette follow-up na de overgang van kinder- naar volwassenengezondheidszorg, welke factoren die van belang kunnen zijn, zijn onderzocht in meer detail in dit proefschrift.

Het algemene doel van het proefschrift was om een beter begrip te krijgen op factoren die van invloed kunnen zijn op onderbrekingen in de follow-up van jonge mensen met aangeboren hartafwijkingen in Zweden en België, maar ook om het conceptuele begrip te vergroten van wat een onderbreking

eigenlijk betekent. De eerste studie onderzocht het aandeel onderbrekingen onder jongeren met hartafwijkingen in Zweden, en toonde aan dat in de zeven universitaire ziekenhuizen iets minder dan 7% van de jongeren niet binnen vijf jaar voor het eerst een hartkliniek voor volwassenen had bezocht. Uit het onderzoek bleek ook dat er grote verschillen waren in het aantal onderbrekingen tussen de zeven ziekenhuizen, wat erop zou kunnen wijzen dat ziekenhuisfactoren van invloed zijn. De studie suggereerde ook een verband tussen de patiëntvolumes van de kinderhartklinieken en het percentage onderbrekingen, waar grotere patiëntvolumes de kans op voortgezette follow-up vergrootten.

In het tweede onderzoek zijn individuele interviews afgenomen met jongeren uit onderzoek 1, waarbij zij moesten vertellen welke factoren volgens hen zelf van invloed kunnen zijn op de keuze van een jongere om de follow-up voort te zetten of niet. Deelnemers beschreven de beslissing om de follow-up al dan niet voort te zetten als veelzijdig. Er werden drie overkoepelende categorieën van factoren geïdentificeerd die de keuze zouden kunnen beïnvloeden: "Motivatie voor follow-up", "Gevoel te kunnen participeren in de zorg en het gevoel erbij te horen" en "beschikbaarheid van zorg". De deelnemers benadrukten vooral het belang van voldoende kennis over hun ziekte en het belang van voortzetting van de follow-up, evenals het gevoel een duidelijk doel voor de follow-up te hebben. Ook de lopende behandeling en eventuele klachten kunnen van invloed zijn, evenals de behandeling door het verzorgend personeel en de actieve ondersteuning om naar de raadplegingen te komen.

In de derde studie werd eerst het percentage onderbrekingen bij jongeren met hartafwijkingen in België onderzocht, volgens dezelfde methodologie als studie 1, waaruit bleek dat in de twee Belgische universitaire ziekenhuizen amper 6% van de jongeren geen raadpleging had binnen vijf jaar na de overgang naar de hartkliniek voor volwassenen. Cardiologen, verpleegkundigen en administratief personeel werden vervolgens geïnterviewd in zowel klinieken voor kindercardiologie als klinieken voor volwassenen met een aangeboren hartaandoening in zowel Zweden als België, om te onderzoeken welke factoren het zorgpersoneel als mogelijke bevorderende en belemmerende factoren voor verdere follow-up na overdracht zien. De verhalen van het zorgpersoneel bevestigden de factoren die werden benadrukt door de Zweedse jongvolwassenen in studie 2 en beschreven hoe de individuele omstandigheden en omstandigheden van de patiënt van invloed kunnen zijn, zoals de transitie naar volwassenheid,



motivatie en kennis van hun ziekte of ziektecomplexiteit en huidige behandeling. Ze benadrukken echter ook het belang van de structuur en organisatie van de zorg en opvang, zoals de personele bezetting, de geïntegreerde of een puur volwassen raadpleging, of de afstand tussen de kinder- en volwassenenkliniek. Ze benadrukken ook het belang van zorgprocessen en zorgrelaties, zoals samenwerking tussen kind en volwassene, outreachende activiteiten, transitiegerelateerde interventies, de competentie van het verzorgend personeel of administratieve routines.

In het vierde onderzoek is het begrip onderbroken follow-up vanuit een conceptueel perspectief onderzocht. Er is een kwalitatieve analyse uitgevoerd van termen en definities voor onderbroken follow-up in de wetenschappelijke literatuur, waarbij drie verschillende typen onderbreking konden worden geïdentificeerd. Met behulp van een expertpanel zijn de drie typen onderbrekingen geëvalueerd en zijn conceptuele definities opgesteld. De drie typen waren: "Gap in follow-up", wat conceptueel kan worden gedefinieerd als het tijdsinterval tussen follow-upbezoeken binnen een specifieke zorgcontext, is overschreden, maar de patiënt is momenteel onder follow-up; "onderbroken of ontbrekende follow-up", wat conceptueel kan worden gedefinieerd als een patiënt die gedurende een bepaalde periode en de zorgcontext niet is opgevolgd en de patiënt momenteel niet onder follow-up staat; evenals "Ontraceerbaar", wat conceptueel kan worden gedefinieerd als de patiënt die niet kon worden gecontacteerd vanwege een gebrek aan informatie. Dit betekent dat de follow-up niet kan worden bevestigd of ontkend.

Dit proefschrift beschrijft onderbroken follow-up tijdens de overdrachtsperiode als een complex fenomeen met een sterke samenhang met zowel ontwikkelingsgerelateerde als organisatorische transities en als een fenomeen dat baat zou hebben bij een bredere aanpak waarin ook de subjectieve beleving van continue zorg van de patiënt wordt meegenomen. Patiënten en gezondheidszorgswerker in de opgenomen onderzoeken benadrukken een breed scala aan factoren die mogelijk van belang kunnen zijn voor voortgezette follow-up op zowel patiënt-, ziekenhuis- als zorgsysteemniveau, met de nadruk op zorgstructuren en zorgprocessen die voorwaarden kunnen scheppen voor de kennis van jonge patiënten en motivatie om de follow-up na de overdracht voort te zetten.



# List of papers

This thesis is based on the following studies, referred to in the text by their roman numerals.

- I. Skogby S, Moons P, Johansson B, Sunnegårdh J, Christersson C, Nagy E, Winberg P, Hanséus K, Trzebiatowska-Krzynska A, Fadl S, Fernlund E, Kazamia K, Rydberg A, Zühlke L, Goossens E, Bratt E-L.  
Outpatient volumes and medical staffing resources as predictors for continuity of follow-up care during transfer of adolescents with congenital heart disease.  
*International journal of cardiology*. 2020;310:51-7.
- II. Skogby S, Goossens E, Johansson B, Moons P, Bratt E-L.  
Qualitative study of facilitators and barriers for continued follow-up care as perceived and experienced by young people with congenital heart disease in Sweden.  
*BMJ Open*. 2021;11:e049556.
- III. Skogby S, Goossens E, Johansson B, Moons P, Bratt E-L.  
Facilitators and barriers for continued follow-up care after transfer - from a healthcare providers' perspective.  
*Manuscript*.
- IV. Skogby S, Bratt E-L, Johansson B, Moons P, Goossens E.  
Discontinuation of follow-up care for young people with complex chronic conditions: conceptual definitions and operational components.  
*BMC Health Services Research*. 2021;21:1343.



# Abbreviations

ACHD	Adult Congenital Heart Disease
Adole7C	AdolesCents reCeiving Continuous Care for Childhood-onset Chronic Conditions
CCCs	Complex Chronic Conditions
CHD	Congenital Heart Disease
CONCOR	CONgenital COR Vitia
DCO	Data Collection Officer
FTE	Full-Time Equivalent
HCP	Health Care Provider
HLHS	Hypoplastic Left Heart Syndrome
NHS	National Health Service
OR	Odds ratio
$p$	$p$ -value
SPSS	Statistical Package for Social Science
SSH	Social Security Health care systems
SWEDCON	Swedish Congenital Heart Disease Registry



# Definitions

Adolescence	“A developmental period ranging from age 10 to 24 years that is characterized by biological growth and social role transitions” (1).
Complex chronic conditions	“Any medical condition that can be reasonably expected to last at least 12 months (unless death intervenes) and to involve either different organ systems or one organ system severely enough to require specialist paediatric care and probably some period of hospitalization in a tertiary care centre” (2).
Congenital heart disease	“A structural abnormality of the heart and/or intrathoracic great vessels that is actually or potentially of functional significance” (3).
Continuity of care	<i>“how one patient experiences care over time as coherent and linked; this is the result of good information flow, good interpersonal skills, and good coordination of care” (4).</i>

*Informational continuity* refers to “the use of information on past events and personal circumstances to make current care appropriate for each individual” (5).

*Relational continuity* refers to “an ongoing therapeutic relationship between a patient and one or more providers” (5).

*Management continuity* refers to “a consistent and coherent approach to the management of a health condition that is responsive to a patient’s changing needs” (5).

Discontinuation of follow-up care	No agreed upon definition exists. In this thesis considered as an umbrella term for describing a broken chain of appropriate follow-up care.
Emerging adulthood	A life stage in-between the adolescent period and the period of adulthood, ranging from 18-29 years of age and encompassing the end of the adolescent period and the entry of stable adulthood (6).
Transfer	“The event or series of events through which adolescents and young adults with chronic physical and medical conditions move their care from a paediatric to an adult health care environment” (7).
Transition	<p>“A passage from one life phase, physical condition, or social role to another, resulting in a temporary disconnectedness of the normal way of living, which demands an adjustment of the person and the environment” (8-10).</p> <p>“The process by which adolescents and young adults with chronic childhood illnesses are prepared to take charge of their lives and their health in adulthood” (7).</p>
Transitional care	“The provision of interventions that attends to the medical, psychosocial, and educational/vocational needs of adolescents as they move from being a dependent child towards an independent adult, with the aim of preparing the adolescents to take charge of their lives and their health in adulthood” (10).
Transition program	“A set of coordinated transitional care interventions that are provided in a structured albeit individualized way, to support the process of the transition to adulthood and achieve the outcomes of transition” (10).







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# Preface

## BORN WITH CONGENITAL HEART DISEASE

Gustav was born with multiple large ventricular septal defects requiring surgical intervention. His surgery in childhood was complicated and future need of surgery could not be ruled out. Gustav therefore received follow-up care at paediatric cardiology. At 17 years of age, he was told “this will be your last visit to paediatric cardiology” and that he would be summoned to the adult clinic within three years’ time. However, Gustav never showed up for his first appointment at the adult clinic. Reminders were sent several times, but Gustav could not be reached.

Alice was born with a cyanotic heart defect, tetralogy of fallot. After repeated surgeries, she was followed up in paediatric cardiology and eventually transferred to adult care at age 18. Alice was recommended a first visit to the specialist Adult Congenital Heart Disease (ACHD) clinic within 2 years. However, 6 years passed before Alice attended her first appointment. By the time of her first appointment, Alice had developed pulmonary regurgitation leading to right ventricle dysfunction and she required a surgical intervention.

Sonja was born with Hypoplastic Left Heart Syndrome. Sonja received palliative surgery in three steps as a child and was then followed up at paediatric cardiology every 6 months. She transferred to ACHD care at 18 and was recommended follow-up at the ACHD clinic every year. After transfer, she showed poor compliance with her medication regime and frequently missed her appointments. Her follow-up intervals eventually exceeded 3 years.

These are fictive stories, illustrating different types of potentially dangerous discontinuations of follow-up care that may occur during transition and transfer of young people with congenital heart disease.





# Introduction

## SECURE, LIFELONG, APPROPRIATE MEDICAL SURVEILLANCE

As a clinical nurse specialist working with pre-and postoperative care for children with CHD, I feel passionate about the care trajectories of patients and their families, but also about young patients being guided in their transition towards independent self-care management and transfer to adult care. This thesis is my contribution to the knowledge base on continued and discontinued follow-up care during transition and transfer of care among young people with congenital heart disease. It is also an attempt to contribute to safeguarding all efforts and investments made by patients, families, and healthcare providers during childhood, as well as future appropriate, lifelong medical surveillance and lifelong quality of life for this group of patients.

## INTERNATIONAL RESEARCH PROJECT AND DOUBLE DOCTORAL DEGREE

My doctoral training was performed as a double degree in collaboration between the University of Gothenburg, Sweden and Katholieke Universiteit Leuven, KU Leuven, Belgium.

This thesis is situated within the international research project Adole7C (AdolesCents reCeiving Continuous Care for Childhood-onset Chronic Conditions). The Adole7C-project was based on international research collaboration and had the overall aim to examine the role of patient-, hospital- and healthcare system-related factors associated with discontinuation of care.



# Thesis structure

This thesis is structured around nine sections. **Section 1 “Background”** introduces the patient population and contextual factors, including, *complex chronic conditions* and *congenital heart disease*, as well as the period of *adolescence and emerging adulthood*. This section also defines and introduces the concepts of *transition* and *transfer of care*. **Section 2, “Continued follow-up care”**, introduces the research topic and provides an *overview of previous research*. It also attempts to put discontinuation in relation to *quality of care, transition and transfer* as well as the related concept of “*continuity of care*”. **Section 3, “Rational and aim”** describes rational and aim of the thesis. **Section 4, “Theoretical perspectives”** outlines *epistemological and ontological foundations*. Reflections about pre-understanding are presented and central concepts of *transfer, transition and quality of care* are related to *health care science*. **Section 5 “Methods”** describes the *overall context*, as well as *setting* of the thesis and provides an *overview of the included studies*. Both general and specific *study designs* are described. **Section 6 “Findings”** presents findings from individual studies, including *prevalence of discontinuation* and *hospital-related factors* from a Swedish multicentre perspective (Study I), *young CHD patients’ perceptions and experiences* of factors affecting continued follow-up care (Study II), *healthcare providers’ perceptions and experiences* of factors affecting continued follow-up care (Study III) and *conceptual definitions* and operational components for discontinuation of follow-up care among young people with complex chronic conditions (CCCs) (Study IV). The findings are then discussed in **Section 7 “Discussion”** in relation to previous research and theoretical perspectives. In **Section 8 “Methodological considerations”**, *strengths and weaknesses* of the studies are discussed. In **Section 9 “Looking ahead”**, the findings are summarized and concluded, with a reflection on *future research needs*, and *applications of current findings*.



# 1. Background

The number of young people with CHD requiring life-long follow-up care and transfer from paediatric to adult care is growing rapidly, due to increased survival rates (11-13). This raises new challenges for healthcare providers (HCPs) and healthcare systems. Failing to meet the needs of this population and failing to keep them in appropriate, continued follow-up care across the course of life, may result in adverse outcomes (14, 15), which would be a personal tragedy for afflicted patients as well as an increased cost for healthcare systems and society.

In this thesis, discontinuation of follow-up care is thought of as a disrupted chain of appropriate, medical follow-up care. Discontinuation is a prevalent issue among young people living with different types of CCCs (14, 16-34), and particularly among young people with CHD.

The period of transition to adulthood and transfer to adult care seems particularly vulnerable to discontinuation. Transfer and transition are therefore valuable perspectives when trying to understand the phenomenon of discontinuation within this particular context.

From an international perspective, discontinuation is a well-described issue and the consequences for young people with CHD can indeed be severe, including increased morbidity and need for urgent interventions (14, 15), which can affect both current and future health. Discontinuation could therefore be considered a matter of quality of care and patient safety, requiring active prevention. We know so far that patient-related factors, such as sex, CHD complexity, prescribed medications or insurance issues can affect the proportions of discontinuation of care (18, 20, 21, 24, 25, 27). However, to tailor effective preventive strategies, we still need in-depth knowledge of organizational fac-

tors of follow-up care delivery and their effect on discontinuation. To facilitate such knowledge, we need to investigate discontinuation of care from a multicentre perspective. This thesis therefore, explores the mechanisms behind discontinuation of follow-up care for young people with CHD from a multicentre perspective and attempts to provide guidance on the development of preventive strategies.

# COMPLEX CHRONIC CONDITIONS

Discontinuation of care could be considered a generic issue, with past descriptions generating from different types of CCCs, such as diabetes type 1 (29, 30), other endocrine disorders (31, 32), juvenile idiopathic arthritis (33, 34) or CHD (14, 16-28).

Within the field of adolescent health, a CCC can be defined as:

*“any medical condition that can be reasonably expected to last at least 12 months (unless death intervenes) and to involve either different organ systems or one organ system severely enough to require specialist paediatric care and probably some period of hospitalization in a tertiary care centre” (2).*

Survival of young people with CCCs has increased. For example, cystic fibrosis or complex CHD were previously considered fatal conditions (35). Today, in high income countries, up to 98% of children with CHD survive to adulthood (13) and life expectancy for patients with cystic fibrosis has risen from 30 to over 50 years of age (36). The increased survival of children with CCCs puts pressure on healthcare systems to deliver high quality care throughout the course of life, since most young people with CCCs suffer substantial risks of long-term complications and development of sequelae. Most of these young patients require life-long follow-up care, including transfer from paediatric-focused to adult-focused healthcare facilities.

# CONGENITAL HEART DISEASE

Congenital heart disease (CHD) is a perfect example of a complex chronic condition (CCC) and a condition with clinical heterogeneity. CHD is considered the most common congenital malformation in new-borns and can be defined as:

*“a structural abnormality of the heart and/or intrathoracic great vessels that is actually or potentially of functional significance” (3).*

## EPIDEMIOLOGY

The present thesis has been performed in high income European settings which have reported an overall birth prevalence of around 8 CHD cases/1.000 births (37). The prevalence of CHD differs across countries and regions, which is suggested to be related to unmet diagnostic capacities (37).

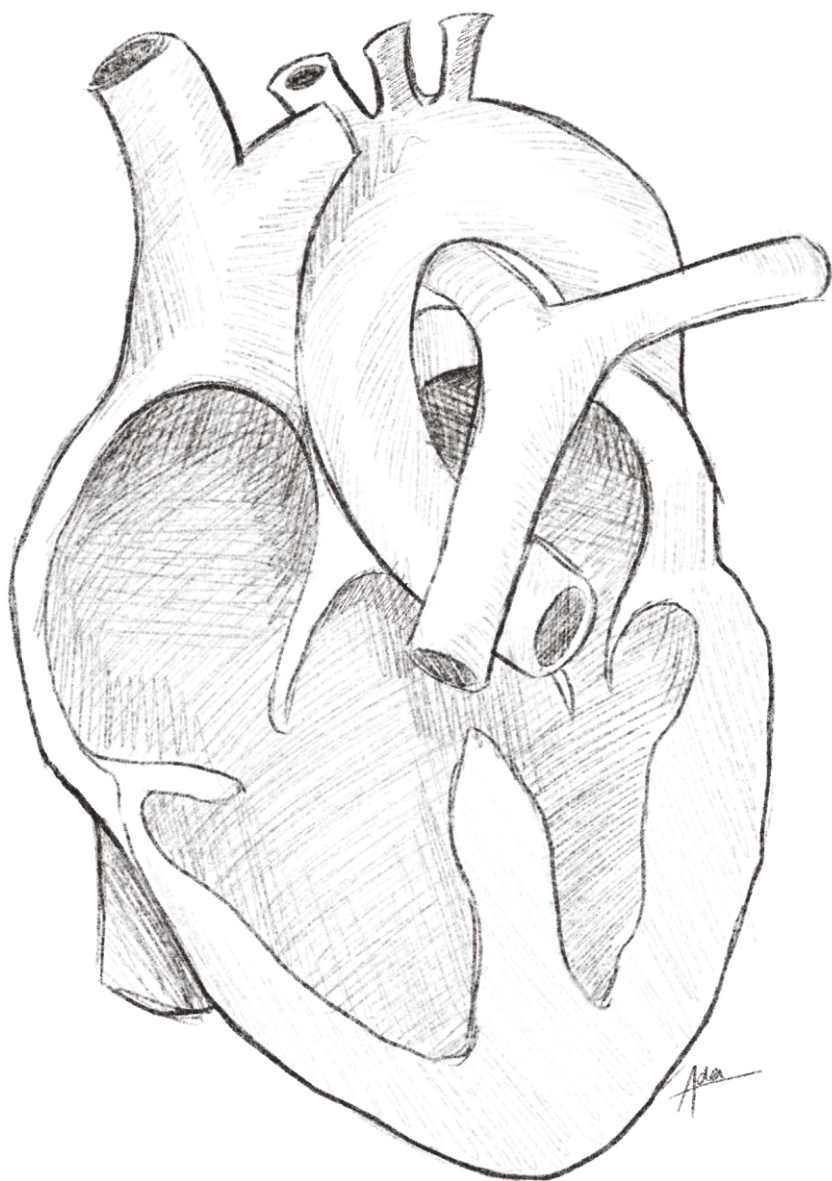
In recent years, the overall prevalence of CHD has increased, one suggested reason for this being better diagnostics of simple lesions (37, 38). In contrast, some types of severe CHD lesions have decreased over time, which is suggested to be related to improved prenatal detection, leading to an increased number of terminated pregnancies (37, 39).

# HISTORY

The accomplishments within paediatric cardiology care and surgery are nothing less than remarkable (40). Early descriptions of CHD dates to 1673 but the first intervention, surgical closure of a persistent ductus arteriosus, was performed approximately 250 years later in 1938. Extra cardiac surgery developed further with, for example, the Blalock-Taussig shunt procedure developed by Alfred Blalock, Helen Taussig and Vivien Thomas to increase blood flow to the lungs in patients with certain cyanotic CHD (41, 42). The later introduction of cardiopulmonary bypass during surgery allowed for a rapid development of intracardiac surgery, e.g. Åke Senning's atrial redirection procedure for d-transposition of the great arteries. Other important achievements in CHD care and treatment include development of cardiac catheterization interventional techniques, pacemaker treatment, prostaglandin treatment to stabilize patients with ductus-dependent circulation and not least, refined ultrasound techniques and other imaging modalities (42). For patients with single ventricle circulation, the Fontan procedure was developed in the late 1960s (43) increasing survival dramatically in very complex CHD. This treatment for patients with single ventricle was even further refined during the late 1980s, into a three-step surgical procedure with a final stage referred to as total cavo pulmonary connection (TCPC), which has the same physiological principle as the previous Fontan procedure, often referred to as Fontan circulation. In the 1980s the first paediatric heart transplantation became a reality. From the 1990s to date, techniques have developed even further, both within the surgical and interventional field, within diagnostics, as well as within pre- and post-surgical care (42). However, with increased possibilities and increased survival comes increased challenges and responsibility. *(Or as Uncle Ben put it; "with great power comes great responsibility" – Spiderman).* Much remains to improve and to investigate, to give children born with CHD not only a chance of survival but also prerequisites for long term quality of life as adults. The long-term prognosis for many CHD patients is still unknown, e.g. patients palliated with TCPC, for whom long term survival will only become clearer in the decades to come.

Thanks to these achievements in early survival, research now focuses on the potential complications and problems later in life.





# CLASSIFICATION

There is broad variation in CHD defects, as patients with mild defects can be almost asymptomatic while complex defects can cause severe symptoms and limitations in daily life. There is also great clinical heterogeneity within defects. Two patients born with the same defect can respond differently to treatment, experience different degrees of symptoms and limitations, and suffer from different complications or comorbidities. This creates a broad spectrum of disease severity and consequently, anatomical classifications only provide one dimension of the patient's true clinical state. To consider the full picture, classifications considering both anatomical circumstances and physiological impact are being developed, but still require further refinement before being implemented in clinical practice and research (44).

With the present thesis and its included studies, we are limited to information on anatomical diagnosis of patients. For this reason we are relying on one of the most frequently used anatomical classifications for CHD as described by Task Force 1 of the 32<sup>nd</sup> Bethesda conference from 2001 (45), and a modified version described in the 2008 guidelines from the American College of Cardiology/American Heart Association (46). According to this classification, CHD defects have either mild, moderate, or severe complexity (45).

It is common that patients are diagnosed with more than one congenital heart defect. In such case it becomes necessary to determine which defect should be considered primary along with its respective complexity. To determine the primary defect, the CONCOR classification has been used in the present thesis. CONCOR is based on the classification from Task Force 1 of the 32<sup>nd</sup> Bethesda conference (45) and further provides a hierarchy of defects (47, 48).

# FOLLOW-UP CARE

The advances in care and treatment have resulted in increased overall survival rates for CHD patients, as to date, in high-income countries, up to 98% survive to adulthood (12, 13). However, for the vast majority, the risk of developing complications later in life is still substantial, even after successful surgery and despite long asymptomatic intervals. The risk of complications is even suggested to be substantial for patients with mild defects (49). To prevent complications, most of these patients require life-long follow-up care (14, 15, 50).

International guidelines describe both a recommended level of care as well as recommended time intervals between follow-up visits based on the respective anatomical complexity of the heart defect (45, 46, 50-52). During childhood, care for children with CHD is recommended to be provided within paediatric cardiology services. When approaching adulthood, adolescents should be transferred to adult care facilities to receive care appropriate for their age and developmental level but also to receive care from healthcare providers (HCPs) trained in adult CHD care (46, 53). For this purpose, adult congenital heart disease (ACHD) clinics have been established in many high-income countries.

Adolescents should be transferred to a level of care deemed appropriate based on their needs and CHD complexity. However, irrespective of CHD complexity, it is recommended that an ACHD specialist sees all adolescents after transfer to adult care to determine the most appropriate setting, level, and frequency of follow-up care in adulthood (50). According to a consensus of international experts (45, 46, 51), there are three levels of follow-up care within the adult setting: specialist care, shared care, and non-specialist care.

**Specialist care:** follow-up provided by ACHD specialist cardiologists at a tertiary care centre. Specialist care should be provided to all patients with complex CHD defects. For patients with complex CHD defects, follow-up is recommended every 6-12 months (45, 46, 51).

**Shared care:** follow-up provided by a general cardiologist in collaboration with a specialist ACHD cardiologist. This level suits patients with mild to moderate CHD defects. For patients with moderate CHD defects, follow-up is recommended every 1-2 years (45, 46, 51).

**Non-specialist care:** follow-up provided by a general cardiologist or general practitioner. This level of care is deemed appropriate for patients with mild CHD defects as they have a low risk of long-term complications. For patients with mild CHD defects, follow-up is recommended every 3-5 years (45, 46, 51).

# ADOLESCENCE & EMERGING ADULTHOOD

Discontinuation of follow-up care has been frequently reported during the period of transition to adulthood and transfer to adult care (54). When transfer to adult care is generally initiated, patients are still going through adolescence, with all its associated challenges. During the first years in adult care, a period of emerging adulthood is entered, introducing new challenges. Here follows a description of adolescence and emerging adulthood, to illustrate these specific stages in life.

## ADOLESCENCE

During adolescence, “the transitional age” you are no longer a child but not yet an adult. It is a time for physical maturation, social role transition, a search for identity and in the end, finding your own way through life (55).

Adolescence includes physical growth, puberty, sexual maturation, brain maturation, cognitive development, as well as social and emotional development. Puberty is generally considered the starting point of adolescence (56). Looking back in time, physical and social-role maturity has been synchronised and the beginning and end of adolescence has been relatively clear. Today, puberty generally starts earlier and the completion of social transitions, such as education, employment or starting a family are pushed forward, making the end of adolescence less clear (1, 56). Adolescence has often been considered to unfold between 10-19 years of age and the term “young people” extends to 24 years of age. Given today’s earlier start of puberty and delayed social maturity, a more appropriate definition of adolescence today is 10-24 years of age (1, 56) and the term “emerging adulthood” extending to 29 years of age (6). However, at what point a young person becomes an adult with responsibility varies across countries, settings, and cultures.

Within this thesis, adolescence is defined as:

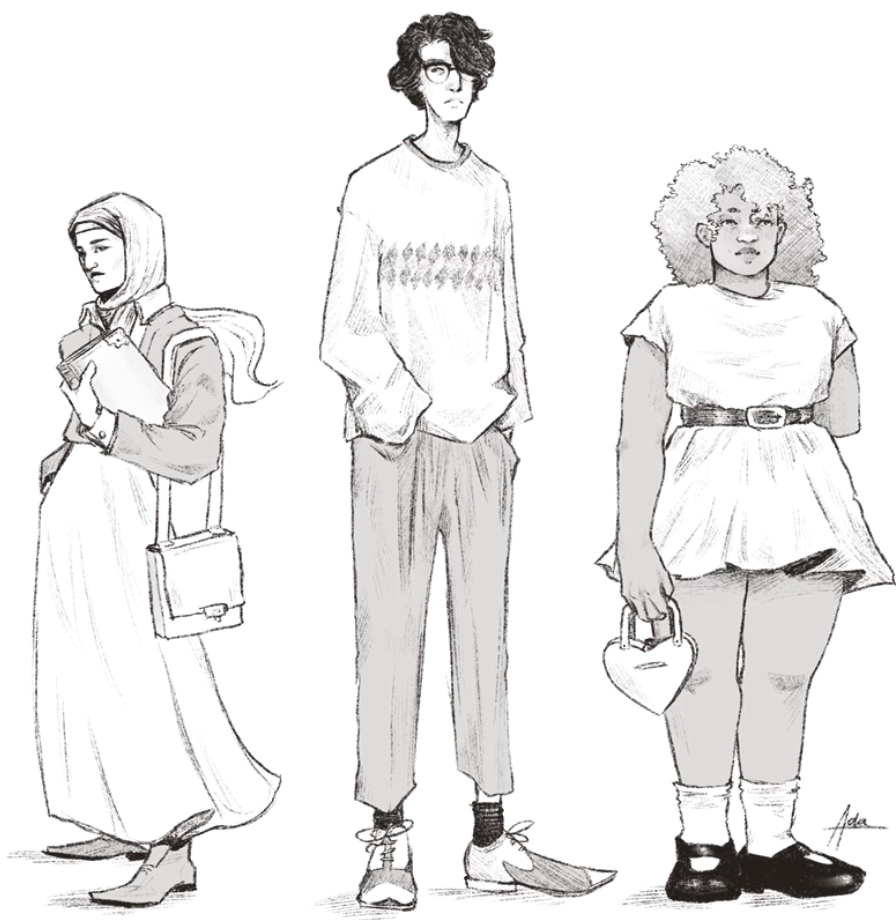
*“A developmental period ranging from age 10 to 24 years that is characterized by biological growth and social role transitions” (1).*

The focus of this thesis lies both on the adolescent period of 10-24 years (1) and the period of emerging adulthood, stretching 18-29 years (6).

If briefly summarizing adolescence, the first phase occurs approximately between 10-14 years of age and is highly characterized by puberty, physical growth, and development. Cognitively, the ability to think in abstract terms improves. However, the focus still lies in the present and less thought is given to the future. Struggles with identity and body image are common, as well as increased sexual interest. Relationships with peers become important and conflicts with parents can occur, along with mood swings and testing of limitations (56).

At approximately 15-19 years of age, physical growth of girls slows down as that of boys continues. Cognitive, abstract abilities increase further, and you start to reflect on life, meaning and future. Struggling with body image and thoughts about not being normal can still be an issue. At this stage, adolescents can be quite self-involved and sometimes struggle with poor self-identity. Love and sex are of increased interest and emotional control improves. Your desire to be independent increases, and the roles of parents will change (56).

At the age of 20-24 years, physical development of girls is often complete, whereas boys continue their physical development, adding on muscle mass, height, and weight. Thoughts about the future, inner experiences and moral reasoning become of more interest. Emotions, sense of identity and sexual identity are more stable. Relationships with peers are still important and you develop independence and self-reliance (56).



# EMERGING ADULTHOOD

Emerging adulthood can be defined as a life stage “in-between” adolescence and adulthood, ranging from 18-29 years of age. Emerging adulthood encompasses the end of adolescence as well as the entry into stable adulthood (6). Emerging adults have reached physical and sexual maturity and are no longer minors, but compared to adults, they can still experience instability. They might still be in education, perhaps combining education with work, or moving between different jobs or between different romantic relationships (6).

# BRAIN DEVELOPMENT

In the context of adolescence and emerging adulthood, it is important to consider the development of the brain. In general terms, brain maturation starts with motor and sensory systems and ends with the prefrontal cortex (57). The adolescent brain has fast maturing intellectual capacity but there is an imbalance between intellectual capacity and impulsive control, especially in emotionally tense situations. This can cause young people to make unfortunate decisions in emotional situations, regardless of cognitive capacity. This is a result of the limbic system being more advanced than the prefrontal control (57). The maturation of the brain can continue well beyond the age of 20, but there is quite a large variation across individuals and across sexes (58).



# COMPLEX CHRONIC CONDITIONS DURING ADOLESCENCE

Adolescents with complex chronic conditions (CCCs) face the standard developmental tasks of adolescence and emerging adulthood, simultaneously with CCC management. It is fair to say that management of a CCC during adolescence is challenging, both for the adolescents and family.

The CCC can affect adolescent development, and vice versa (59). For example, priority issues between education and illness management might arise and school absence due to treatment or hospitalization might have a negative impact on educational achievements and later cause difficulties reaching financial independence in adulthood (59). A CCC can be an obstacle for an adolescent when trying to achieve autonomy, but also when trying to develop identity, since experimenting with life styles and social networks can be challenging (60) due to physical limitations or life-style restrictions, for example. A CCC can have effects on both puberty and growth, depending on the type of condition. Puberty can be delayed and growth can be slow, sometimes leading to increased psychosocial difficulties (59) and fear of not being normal, which is already common during adolescence (56). Mental health issues can sometimes be linked to a CCC but the evidence differs across studies and conditions. Emotional problems have been described, such as depression, low self-esteem, and loneliness. There are also descriptions of struggles with identity and self-image as well as sexual difficulties, for example due to scars or stomas (59).

During adolescence, young people often engage in risky health-related behaviours. Adolescence is a phase of life where future health patterns and health-related behaviours, such as physical exercise, alcohol and tobacco use, food habits and sexual practices are established, which will have an impact on future health as an adult (56, 61). Experimenting behaviours are a natural part of adolescence but for young people with CCCs, risky behaviours may have long-term effects on future health and life. It has been suggested that health

risk behaviours are more common in adolescents with chronic conditions, as compared to healthy peers (62, 63).

Nevertheless, it is difficult to estimate the impact of a CCC on life and health, and how this potential impact is perceived by the individual. Research on experiences of adolescents living with chronic conditions confirms that these often have some sort of impact. For example, adolescents with CCCs can experience a sense of disruption from normal life and feel uncomfortable, both in their bodies and in the world. It is, however, important to acknowledge that not all experiences are negative and that positive perspectives are needed to maintain and promote health (64).

Adolescence should nevertheless be considered a vulnerable period, especially when living with a CCC. To prevent long-term complications, the healthcare needs of young people need to be considered. One goal of adolescence and emerging adulthood is the establishment of independence and self-reliance. Life trajectories such as education, employment, physical and mental health, as well as relationships are to some extent dependent on individual resources, but also affected by chronic disease and physical or intellectual disability (65). Adolescents and emerging adults with CCCs need to develop knowledge, skills, a level of autonomy and decreased dependency on parents in order to manage their health independently and to optimize their future quality of life (7). This is a quest requiring both motivation and support.

# CONGENITAL HEART DISEASE DURING ADOLESCENCE

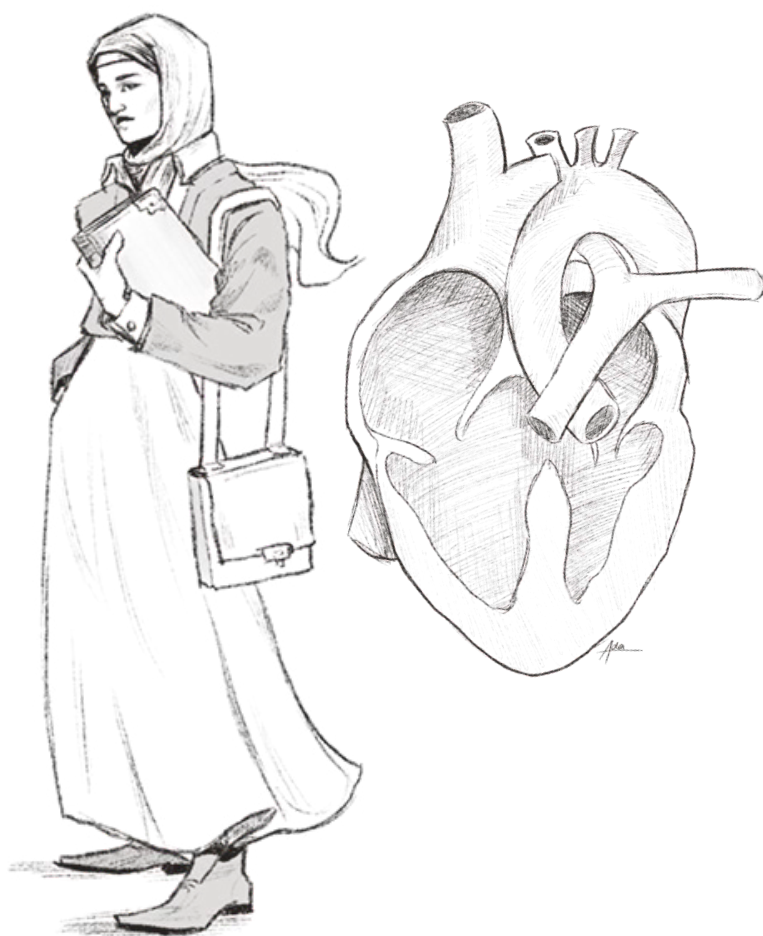
The epidemiology of congenital heart disease (CHD) is changing, with more and more children surviving into adulthood, adding to the growing population of adult people with CHD (11-13). To provide appropriate care across the course of life, the aging population with CHD must be given priority.

Being born and growing up with CHD is challenging in many ways, but also a journey characterized by great heterogeneity. Consequently, the experiences of children and adolescents will differ, and no description will fit all. Some adolescents are likely to experience few effects and consequences of their CHD, while others may feel deeply affected and limited.

Some children and adolescents with CHD have been reported to experience their CHD as disruptive in the sense that the disease interfered with their daily lives, sometimes putting life on hold and challenging self-perception. Struggles with accepting the condition have also been described especially when feeling well, sometimes leading to a denial of the disease and neglecting to keep informed about it. Stressful memories of previous surgeries and interventions are also described, as well as fear of needing future surgery and fear of death. Uncertainty about the prognosis of the condition and their future life could be very demanding and tiring for young people. In addition, trying to minimize damage and avoid future complications by adapting a healthy lifestyle could reduce the feeling of living “a normal life”. Feelings of frustration and sometimes shame over limited physical functionality as well as struggles with body image are described and sometimes even social exclusion due to physical limitations or deviating appearances (66). This well illustrates a very challenging road for a young person with CHD. Studies have indicated that quality of life is an issue (67) and some dimensions of quality of life are lower for this population than health peers (68).

In contrast, some studies describe how quality of life among adolescents with CHD is even better than that of healthy controls due to higher sense of co-

herence (69). Young people with CHD have also been described as developing a will to fight the disease, feeling proud over their achievements and lucky to be alive. Descriptions of adaptation to former limitations, a switch of focus from limitations towards possibilities, appreciation for life and increased personal strength and maturity because of the condition (66) cast a different light on the journey of adolescents with CHD.



# TRANSITION & TRANSFER

As this thesis falls within the specific context of transition to adulthood and transfer of care from paediatric to adult health care facilities, the following section provides an overview of the concepts of transition and transfer of care in relation to young people with CCCs.

## DEFINITIONS

Transition and transfer of care are two closely related concepts, both highly relevant to young people with CCCs. However, in scientific literature, transfer and transition are often used interchangeably, and it is important to clearly distinguish them from one another.

Transition can generally be defined as:

*“a passage from one life phase, physical condition, or social role to another, resulting in a temporary disconnectedness of the normal way of living, which demands an adjustment of the person and the environment” (8-10).*

In the more specific context of young people with chronic disease, transition can also be defined as:

***“A process by which adolescents and young adults with chronic childhood illness are prepared to take charge of their lives and their health in adulthood” (7, 70).***

In contrast, transfer of care, is defined as:

***“The event or series of events through which adolescents and young adults with chronic physical and medical conditions move their care from a paediatric to an adult health care environment” (7, 70).***

# TRANSITION

Transition is a concept related to change and development. The middle range theory of transition by Meleis (9) further describes transition as a multidimensional concept, including elements of process, time and perception. Transitions starts as soon as change is anticipated and are triggered by critical events or changes occurring within the life of the individual or environment (9). Transitions should be considered complex processes, often characterized by a feeling of disruption (71). When a transition process has been completed, it implies that a period of greater stability has been reached (71).

The theory of transition by Meleis further includes different *types* and *patterns* of transitions, *properties* of the transition experience, *facilitators and inhibitors*, *patterns of response*, *outcome indicators* and *nursing therapeutics* (72).

## TYPES

Schumacher and Meleis (8) have presented a typology including four different types of transition. The first is *developmental transition* characterized by developmental change, for example going through adolescence into adulthood. The second is *situational transition*, characterized by situational change, such as starting a new job, moving to another country or changes in relationships or marital status. The third type is *health/illness transition* which includes for example becoming ill or recovering and regaining health, but also changes in health care levels, such as going from hospitalization to outpatient care, or changes in treatment strategies. The last type of transition is *organizational transition*, including changes in the environment, for example new leadership, or policies (8).

Adolescents with CCCs, are experiencing a *developmental transition*, going from childhood, a life phase during which they are mainly being cared for, towards adulthood, a stage in life characterized by autonomous management of life, health and care (8). These adolescents are also experiencing an *organizational transition* as they are asked to change healthcare provider and setting (8). This organizational transition is linked to their transfer of care from the paediatric to adult focused facilities, as the transfer itself results in change of environment and routines.

## PATTERNS

In terms of patterns, different complexities of transitions exist. For example, a transition can occur as a single event or multiple transitions can occur at the same time. Multiple transitions can also occur in a sequence or predictable order (72).

## PROPERTIES

The properties of transition include awareness and recognition of the transition, the degree of engagement in the transition, the perceived change and difference, how the transition is experienced over time, and critical events of the transition (72).

## FACILITATORS AND INHIBITORS

There are also facilitators and inhibitors of a transition process, which can be described as events or circumstances that either support or complicate a healthy transition process, for example preparation and knowledge about the transition, the perceived meaning of the transition, socioeconomic status, cultural beliefs and attitudes, but also community and societal conditions (72).

## RESPONSE AND OUTCOME INDICATORS

In transition theory, responses to the transition process can be described in terms of process and outcome indicators. Process indicators inform about the patient's current direction towards either a healthy or unhealthy transition outcome. Indicators of a healthy transition include feeling connected, interacting, and developing confidence and coping. Outcome indicators include the ability to master the new skills needed to manage the new situation and the development of a new identity (72).

# TRANSFER OF CARE

Since young people with CCCs cannot be considered cured, the care of their disease must continue in a manner tailored to their age-specific healthcare needs, throughout childhood, adolescence, adulthood and not least old age. Continuous follow-up care during the course of life requires a successful transfer of care between different healthcare providers and settings, such as between paediatric and adult-focused care.

As previously mentioned, transfer of care is defined as:

*“**The event** or series of events through which adolescents and young adults with chronic physical and medical conditions **move their care** from a paediatric to an adult health care environment (7, 70).*

Transfer is thereby different from transition, the latter involving an inner process of change and development. Transfer is limited to external events. However, the transfer itself can result in environmental changes and circumstances which can trigger a transition process, in such cases referred to as an organizational transition (8).

Transfer from paediatric to adult care facilities not only allows young people to receive care appropriate for their age and developmental level, but also provides the opportunity to receive care from HCPS specialized in CHD management during adulthood (46, 53). Transfer of care is also recommended as the adult setting is more experienced in addressing issues related to adult life, such as independent living or family planning.

Transfer towards adult care for young people with CCCs should ideally be coordinated jointly between the paediatric and adult providers (7). After transfer from paediatric to adult care, the design of the health care system in most contexts expects young people to have reached a certain level of independence and self-care management. In adult care, the young person often is or soon will be legally an adult, which from a health care perspective comes with certain rights and expectations and often certain limitations for parents.



In addition, the adult health care settings often provide more individual-based care in comparison to family-focussed approaches often seen in paediatric care (73).

Bearing in mind that the period of adolescence and emerging adulthood in most cases continues beyond the point of transfer and transition to adult care, it seems reasonable to assume that support in the process of attaining independence and self-care management skills is needed. Hence, transfer to adult care should occur only after successful completion of a preparatory transition process during which the young person is being prepared for independent self-care management (7).

# TRANSFER & TRANSITION FOR YOUNG PEOPLE WITH CONGENITAL HEART DISEASE

The most appropriate transfer destination for most young patients with CHD is an ACHD program. Indeed, as previously mentioned, international guidelines recommend transfer of care for a vast majority of patients (46, 50, 51, 53). It is furthermore recommended that all CHD patients should attend at least one visit to an ACHD program to receive recommendation on follow-up level and intervals (53). The timing of transfer differs across settings. Ideally, the developmental stage of patients should be considered when deciding on the timing of transfer. However, some studies suggest that a delayed transfer until 18 or 19 years of age is the most appropriate course (74).

As the transfer from paediatric to adult care approaches, adolescents often perceived it as a natural step in their care process (75-77) and many feel calm and positive about transfer but need to adjust to the new environment (77). Some adolescents even feel that they have outgrown the paediatric setting and feel uncomfortable with the paediatric unit, which is often attended by toddlers and infants (77). However, some studies report worry or anxiety among adolescents with CHD about the upcoming transfer, such as anxiety related to the new adult environment and the new adult HCPs. The strong bond that they have developed with paediatric HCPs can contribute to an anticipation about changing HCPs when they transfer (76, 78). For some patients, meeting with the adult HCPs in advance has eased concerns and increased their sense of security (78).

In preparation for transfer, young people emphasize the importance of sufficient knowledge about their condition but also about the transfer itself, the new HCPs, how follow-up care will be organized in the future and who to contact for questions or concerns (75, 78). Receiving sufficient information could ease their worries about transfer and increase their sense of security (78).



To prepare for the transfer and transition, adolescents also considered it important to be involved in process. They felt preparation needed to start well in advance and be a gradual process with guidance in achieving self-responsibility (78).

During transition and transfer, the relationship between the young person and their parents changes. The young person's responsibility increases and the parents responsibility decreases (76). Readiness for transition as well as level of responsibility have been shown to increase in young patients as they get older. However, young people often feel more ready for transfer than their parents (79). Hence, it is important to acknowledge the parents' position during the transition process, as parents play an important role in providing support practically, for example reminders or advice but also emotionally (75).

# TRANSITIONAL CARE

When trying to facilitate transfer to adult care and ease transition into adulthood, we talk about transitional care. Transitional care can be defined as:

*“the provision of **interventions** that attends to the **medical, psychosocial, and educational/vocational needs** of adolescents as they move from being a dependent child towards an independent adult, with the aim to **prepare the adolescents to take charge** of their lives and their health in adulthood” (10).*

During a transition process and through transitional care, the young person is being prepared to take charge of life as well as health. The transition process is partly educational in that a curriculum should be provided to help young people understand their diagnosis, medical history, and its impact on current and future life, as well as how to navigate in the adult health care system. The education should aim to increase self-management skills and sense of control. The transition process and transitional care interventions should continue until the young person has reached a sufficient level of self-care management and can take over full responsibility for health and independently navigate the adult health care system (7).

## OUTCOMES OF TRANSITIONAL CARE

How do we know that our transitional care efforts have been successful? When can we talk about successful transition and transfer of care?

There is a plethora of possible outcomes for transition and transfer provided in the literature. Two prior studies have attempted to reach consensus on the matter using a Delphi methodology (80, 81) and the results were recently summarized and adapted in a global consensus statement on transition and transfer for adolescents with CHD (10). Ten outcomes were presented, including: *quality of life, self-management skills, knowledge of your disease, continuity of patient care, appropriate health care consumption, disease control, access to and receiving coordinated care and peer interaction* (10).

Some of these aspects are related to the individual, for example *quality of life*, *self-management skills* and *knowledge of your disease* (10, 80). Some outcomes are more related to health care delivery, such as *continuity of patient care* and *coordinated care* (10, 80).

*Continuity of care*, which could be considered a health care delivery-related aspect, is in this context often referred to as attendance of medical appointments (80). Continuity of care is a highly emphasized outcome in this matter, and it is an essential indicator for successful transition and transfer of care (81). The fact that many young patients with CHD are found to have a lack of continuity of care (i.e. discontinuation) after transfer, including no follow-up care whatsoever or receiving follow-up at an inappropriate care level is often used as an argument for the need to implement or improve transitional care interventions (7).

The link between (dis)continued follow-up care and transition and transfer will be further elaborated on in the upcoming section.







## 2. Continued follow-up care

The continuum of follow-up care has two extremes, continued follow-up care at one end and discontinued follow-up care at the other. This thesis considered continued follow-up care as the norm, to strive for, and discontinued follow-up care as a patient safety hazard, requiring prevention. The following section introduces continued and discontinued follow-up care, including related concepts, definitions, prevalence and associations. Discontinued follow-up or discontinuation will also be described in relation to quality of care, transition and transfer.

### CONTINUITY OF CARE

*“Continuity of care”* is an established concept which could be considered linked to *continued follow-up care* and a valuable perspective for this thesis. *Continuity of care* as a concept can be somewhat challenging to grasp. According to the Oxford English dictionary, the word continuity is defined as:

*“the state or quality of being uninterrupted in sequence or succession”* (82).

However, the meaning of continuity of care is often presumed in the absence of clear definitions (4) and the distinction between continuity of care and related concepts remains vague (83). As mentioned earlier, within the context of transitional care, continuity of care is often referred to as attendance of

medical appointments. However, more comprehensive perspectives on the concept do exist (4, 5).

Historically, definitions for continuity of care have shifted. Initially, around 1950, continuity of care was mostly considered related to patients having personal care providers. Later, around 1970, the focus shifted towards relatedness between past and present care, as well as care being coordinated and uninterrupted. Multidimensional models were also introduced at this point. Then came an era where continuity of care was instead considered a measurable concept and defined in terms of patients seeing the same physician. Around 1990, multidimensional models returned and the experiences and views of patients became central parts of continuity of care and its definitions (83). It is important to acknowledge that continuity of care can take different forms depending on situational contexts, such as type of HCP or patient populations (4). Indeed, definitions are shaped by the setting in which they are developed and used, leading to differences across different types of healthcare settings. However, Reid, Haggerty and colleagues (2002-2003) defined continuity of care in a way that bridges disciplines and organizations (4, 5).

**Continuity** overall is defined as:

*“how one patient experiences care over time as coherent and linked; this is the result of good information flow, good interpersonal skills, and good coordination of care”* (4).

Reid, Haggerty and colleagues further describe three different types of continuity:

**Informational continuity**, referring to *“the use of information on past events and personal circumstances to make current care appropriate for each individual”* (5).

**Relational continuity**, referring to *“an ongoing therapeutic relationship between a patient and one or more providers”* (5).

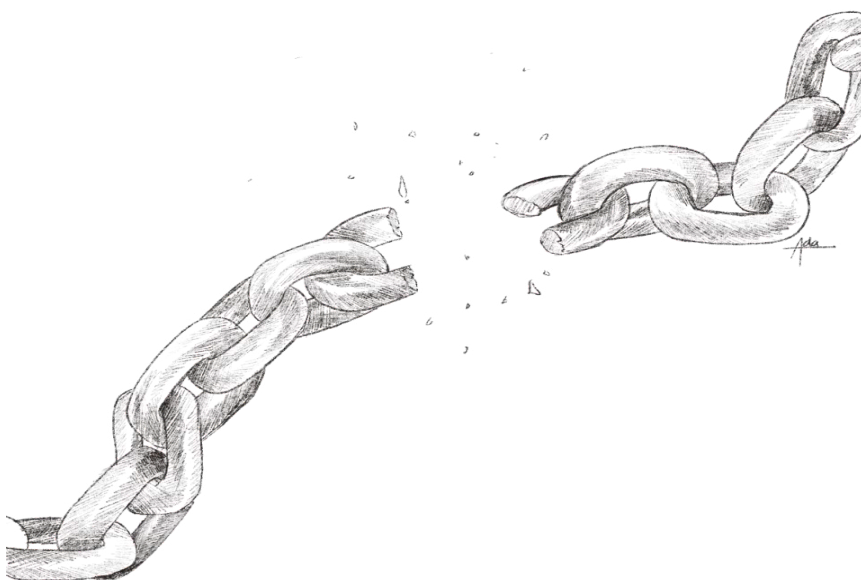
**Management continuity**, referring to *“a consistent and coherent approach to the management of a health condition that is responsive to a patient’s changing needs”* (5).

*Continuity of care*, as defined by Reid, Haggerty and colleagues also requires the presence of two core elements. First, “*care of an individual patient*” and second, “*care delivered over time*” (5), which means that patients’ experience of continuity and not only that of providers or objectively measurable events, should be centrally placed, and that a “*longitudinal or chronological dimension of continuity*” is important (5). These two core elements help to distinguish continuity of care from other attributes of care, such as quality of care or coordination of care (5). In addition, the three different types of continuity can be viewed from either a person-focused or a disease-focused perspective (4). Despite these core elements and specific types of continuity, there are still disparities between healthcare settings. Different healthcare settings emphasize different types and aspects of continuity (5). For example, from the mental healthcare setting, coordination of care and *relational continuity* might be emphasized, but in contrast, within a disease management setting, logical and timely delivery of care (i.e. *management continuity*) might be more emphasized (5).

The experience of the individual patient is central in continuity of care and the unit of measurement should primarily be the individual (5). However, the concept does extend beyond the individual experience and objective evaluation is also deemed mandatory (84). This makes the measurement of this concept interesting since it needs to capture both objective information and the individual’s experience. Clearly no single measure can capture all aspects of continuity of care. Existing measures are often based on patterns of care delivery, which do not capture the experience of the individual (4).

Continued follow-up care after transfer for young people with CCCs is desired to maintain health and quality of life. When assessing continued follow-up care among young people with CCCs the individual experience of continuity of care would be crucial to consider to gain a deeper understanding. Most previous studies on discontinued follow-up care for young people with CHD have been concerned with patterns of care delivery (14, 16-28), which, in the light of Haggerty’s definition of continuity of care could be seen as a form of *management continuity* (5).

# DISCONTINUATION OF FOLLOW-UP CARE



In this thesis, discontinuation of follow-up care is thought of as a disrupted chain of continued, appropriate medical follow-up care. For example, follow-up care could be completely lacking, the patient could have long time gaps between follow-up visits or follow-up could be provided in an inappropriate health care setting, based on the patient's medical needs. In relation to continuity of care, as described by Reid, Haggerty and colleagues (2002-2003) the approach of the present thesis could indeed be considered mostly related to management continuity (5). Currently, there is lack of conceptual clarity on the concept of discontinuation of follow-up care and the heterogeneity among operational definitions in the literature is high, complicating research comparability (85). For example, common terms used in the literature are "Lost to follow-up", "Lapse in care" and "Gap in care", among many others. In this thesis, *discontinuation of follow-up care* is considered an umbrella term, covering the many different terms and concepts presented in the literature.



# PREVALENCE & ASSOCIATIONS OF DISCONTINUATION

Discontinuation of follow-up care is not only a concept relevant to the population of patients with CHD, but has also been described among young people with diabetes type 1 (29, 30), other endocrine disorders (31, 32), and juvenile idiopathic arthritis (33, 34), for example. However, many prior studies have focused on the investigation of this concept in young people with CHD (14, 16-28), as does this thesis.

One of the first descriptions of discontinuation of follow-up care among young patients afflicted by CHD is from 2004 and was published by Reid and colleagues (27). This study was performed in Canada and studied young adults with moderate to complex CHD. In total, 47% of their sample failed to attend ACHD care after transfer (27). After 2004, the number of studies on this subject has increased, and discontinuation of follow-up care is today a highly recognized issue in research, with the affected percentage of adolescents and emerging adults with CHD ranging from 3.6-62.7% (14, 16-28, 86, 87) and a pooled estimated percentage of 26.1% (54).

To provide some examples, Yeung and colleagues (2008) reported 63% of their sample experience “lapses of care” referring to a time interval greater than 2 years between the last paediatric and first adult visits (14).

Mackie and colleagues (2009) reported on “loss of follow-up care” after the 6<sup>th</sup>, 13<sup>th</sup> and 18<sup>th</sup> birthday as occurring in 28%, 47% and 61% of their sample from Quebec, Canada, respectively (25).

In 2011, Goossens and colleagues reported on a Belgian single centre study and described 7.3% “No follow-up care” in their sample of mild, moderate, and complex CHD (20).

In 2013, Gurvitz and colleagues published one of the few multicentre studies on the topic, reporting 42% gaps in cardiology care, and referring to more than three years between any type of cardiology appointments (21).

Kollengode and colleagues reported in 2018 on an American single centre study where 37.6% of patients with CHD had been “lost to follow-up” for a period of more than 3 years (24).

In 2020, Mondal and colleagues reported on a Canadian single centre study where only 3.6% of all patients failed to attend a first adult visit within two years (87).

As we can see, the reported percentage of patients vary greatly from one scientific peer-reviewed paper to another. In addition, we can observe the use of many different terms and operationalizations for the variable of (dis)continuity of follow-up care, making comparisons of percentages very challenging.

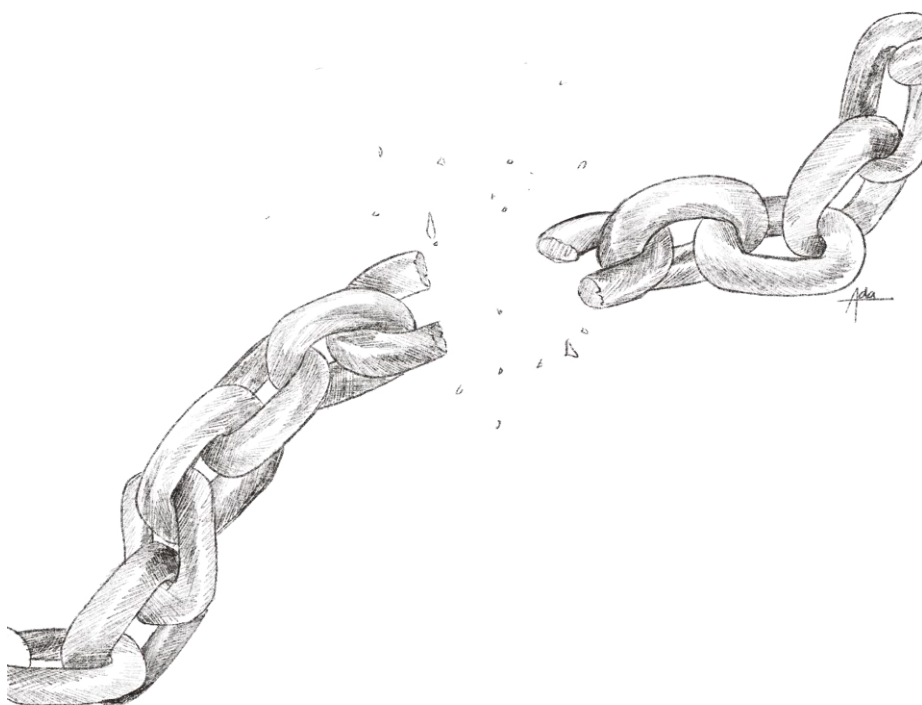
In addition to percentages of patients discontinuing follow-up care, several of these prior studies have investigated associated factors. So far, mainly association related to the individual (so-called patient-related factors) have been investigated and identified (85). For young people with CHD, it has been shown that male sex, lower level of CHD complexity and no prior cardiac interventions can be associated with discontinuation, but also lack of prescriptive medications, and insurance issues (18, 20, 21, 24, 25, 27).

However, it is likely that in addition to patient-related factors, hospital and even health care system-related factors may play a role in discontinuity of follow-up care in this patient population. Indeed, the percentage of patients discontinuing differ widely and the multicentre study from the U.S.A by Gurvitz and colleagues (2013) showed differences across hospitals (21), indicating the potential influence of hospital-related factors.

Reports on patients’ own perceptions or experiences of factors affecting continued or discontinued follow-up care after transfer are few (14, 21, 88) but include factors such as losing track of time, (21) being told follow-up is not needed (14), changing or losing insurance (14, 21), unstable housing, missed appointment letters, and anxiety about cardiac health (88).

In addition to the associated factors described above, discontinuation of follow-up care is also associated with adverse outcomes, such as increased morbidity and the need for urgent interventions (14, 15).

For example, Yeung and colleagues reported that patients who experienced a “lapse in care” of more than 2 years were three times as likely to require an urgent cardiac intervention (14). Cordina and colleagues reported a higher risk of clinical complications in patients who had been cared for by general cardiologists than in patients cared for by CHD-cardiologists (15). This increased risk of morbidity makes discontinuation of follow-up care a highly relevant issue and highlights the need of effective preventative strategies.





# DISCONTINUATION & QUALITY OF CARE

Since discontinuation of follow-up care has shown to be associated with adverse outcomes for young people with CHD (14, 15), it could be considered a matter of quality of care and patient safety.

High quality care is safe, timely, effective, efficient, equitable (89) and person centred. High quality care considers the patients' desires and values and provides the right type of care to the right patient at the right time (90). When follow-up care for young people with CHD is disrupted, not delivered in a timely manner or not offered within an appropriate health care setting, it could indeed be considered a matter of deficient care quality.

Quality of care should be seen as a multidimensional concept. When trying to improve quality of care, several different levels need to be considered, including the broad level of regulations and finances, the level of organization (such as the hospital or clinic), the level of microsystems functioning within an organization (such as an outpatient care team), and also the level of the individual patient (90). Successful delivery of continuous follow-up care for young people with CHD could most probably be related to all these levels.

Patient safety is a relatively new, fast growing and highly valued discipline in a world where health care delivery and health care systems are becoming more and more complex. The modern perspective on patient safety shifts our focus from the individual HCP committing an error towards the health care systems. It views most errors as preventable and often related to defective design of the health care system. Patient safety can be defined as:

*“a discipline in the health care sector that applies safety science methods toward the goal of achieving a trustworthy system of health care delivery. Patient safety is also an attribute of health care systems; it minimizes the incidence and impact of, and maximizes recovery from, adverse events” (91).*

Discontinuation of follow-up care could be considered a matter of patient safety, as it is associated with adverse outcomes (14, 15). For example, discontinuation of follow-up care includes not only complete absence of follow-up care, but also follow-up care provided with inadequate timing or provided within an inappropriate setting. Catastrophic and major clinical complications have been shown to be more common in CHD patients who were cared for by general cardiologists than by cardiologists specialized in (A)CHD (15). This is an example of how the clinical setting and organisation of follow-up care can lead to discontinuation of appropriate follow-up care and in the long run raise a patient safety issue. A health care system focussing on patient safety issues attempts to minimise the incidence of adverse events, which would include ensuring appropriate continuous follow-up care when recommended.

## DISCONTINUATION & TRANSITION and TRANSFER

The period of transition to adulthood and transfer of care could be considered as particularly vulnerable for discontinuation of follow-up care, firstly, because of the physical movement between health care settings, secondly, because of the developmental process of going from adolescence towards adulthood and thirdly, because of the newly appointed responsibility for health and self-care management.

Bearing in mind that the period of adolescence and emerging adulthood continues in most cases beyond the point of transfer to adult care, transition, and thereby the risk of discontinuation of follow-up care, are probably relevant in both the paediatric and adult health care setting.

There is some indication that transitional care interventions can affect the prevalence of discontinuation of follow-up care. For example, Hergenroeder and colleagues (2018) evaluated a new transition program for young people with CHD and reported that control patients had prolonged time intervals between last paediatric and first adult visits, as well as an increased risk of

lapse in care during transfer when compared to patients receiving a transition program intervention (23). Mackie and colleagues (2018) tested a nurse-led transition intervention and reported that usual care patients had longer time intervals between last paediatric and first adult visit than intervention patients (86). For patients participating in a transition intervention, Gaydos and colleagues (2020) reported lower rates of lost to follow-up than the controls (17). However, in-depth understanding is needed to determine the actual impact of transitional care interventions on discontinuation of follow-up care, a task which extends beyond the scope of this thesis. Nevertheless, it cannot be ignored that continued follow-up care is often argued to be a central outcome indicator of successful transfer and transition (7, 10, 80).

Considering the specific context of the present thesis covering the period of transition to adulthood and transfer to adult care, Meleis' transition theory could provide a valuable perspective. Discontinuation is therefore viewed through the lens of Meleis' transition theory (72), and "Discontinuation" and "Transition" are considered linked. This reflective exercise is described below in the form of a modified version of Meleis' model of transition theory (Figure 1).

In the adult setting, patients are expected to navigate the health care system, to attend follow-up care visits and independently manage health and care. In the unfortunate event of discontinuation of follow-up care after transfer to adult care, discontinuation of follow-up care could be seen, from the perspective of transition theory as an indication of a patient's insufficient role mastery of a new "adult care" situation (72), and thereby as an *outcome indicator* for an unhealthy or unsuccessful transition process (Figure 1).

For a young person with CHD, the transition *pattern* could be considered as "multiple", with two types of transition occurring simultaneously: one developmental transition going from childhood towards adulthood and one organizational transition going from paediatric to adult care facilities. The event of transfer could be considered a *property* of the organizational transition, in the form of a *critical event* (72) (Figure 1).

The patient-, hospital-, and health care system-related factors possibly influencing discontinuation of follow-up care, which is being investigated in this

thesis, could be considered *transition conditions* at different levels, which either function as *inhibitors* or *facilitators* for a healthy and successful transition (72).

*The personal level* in the transition theory, could be considered linked to patient-related factors possibly affecting discontinuation of follow-up care, for example the patient's level of understanding their CHD condition and the respective need for life-long follow-up.

*The community level* in the transition theory could, be considered linked to hospital-related factors possibly affecting discontinuation of follow-up care, for example the respective availability of transition programs.

*The society level* in the transition theory, could be considered linked to health care system-related factors possibly affecting (dis)continued follow-up. For example, out-of-pocket cost for ACHD care (72) (Figure 1).

Transitional care interventions provided to facilitate successful transition and continuity of care after transfer could be seen as part of (health care) *therapeutics* within the transition model. Indeed, not only nurses encounter young patients in transition. Other HCPs, such as physicians or administrative staff, also play important roles in a patient's journey and their actions might influence transfer and transition outcomes. Therefore, within this modified version of Meleis' transition model (Figure 1), nursing therapeutics is referred to simply as *therapeutics*, in a more generic sense (72).

Indeed, this reflective exercise, relating the concept of discontinuation of follow-up care to Meleis' theory of transition is not to be understood as a development of the model, but rather as a contextual frame for discontinuation. This respective middle-range theory of transition incorporates a much richer perspective on the transition process than presented here. Discontinuation of follow-up care should therefore be seen as one of many relevant outcome indicators of an unhealthy transition process.

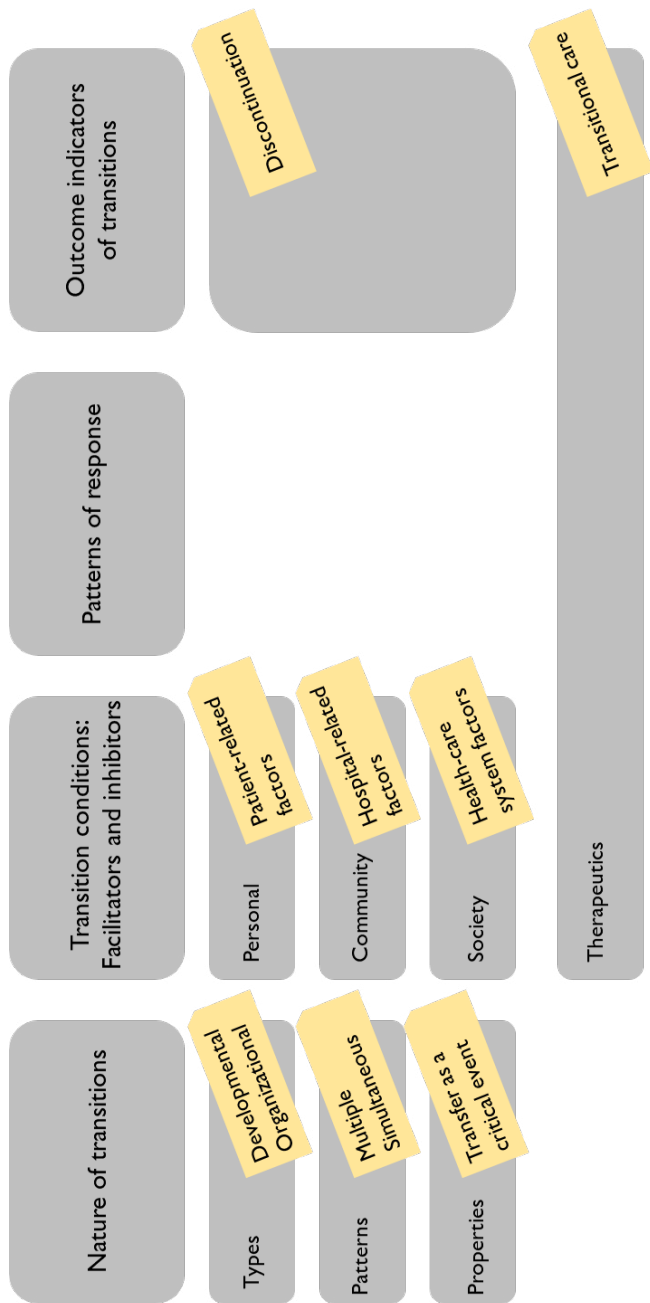


Figure 1. Modified model of Meleis et al., 2000 Transition theory



# 3 Rationale & Aim

Young people born with CCCs, such as CHD, may risk long-term complications and therefore require life-long follow-up care and transfer towards adult care facilities. Healthcare professionals and healthcare systems ought to be prepared to provide appropriate life-long medical surveillance to maximize potential and lifelong functioning of these young people. Although life-long follow-up care is required and recommended for most patients with CHD (46, 50, 51, 53), a substantial proportion presents with discontinuation of follow-up care (14, 16-28, 87), which is associated with adverse outcomes (14, 15), and therefore a matter of quality of care and patient safety, requiring active prevention. The period of transition to adulthood and transfer from paediatric to adult care seems particularly vulnerable for discontinuation of follow-up care and should be considered when approaching this phenomenon. From the perspective of the transition theory developed by Meleis (9), discontinuation of follow-up care could be considered an outcome indicator for an unsuccessful transition process.

In relation to discontinuation of follow-up care, mainly patient-related factors have been studied so far (85). However, it can be assumed that hospital- and healthcare system-related factors may also play a role in its occurrence. The possible association between discontinuation of follow-up care and hospital-related factors should be investigated as well as the possible impact of healthcare system factors. Patient-related factors are richly described from a quantitative perspective (18, 20, 21, 24, 25, 27). However, young people's experiences and perceptions of discontinuation of follow-up care and its associated factors have been less described (14, 21, 88) and could provide further direction for qualitative research and intervention. In addition, a more in-depth understanding of the conceptual and operational definitions of the concept of discontinuation of follow-up care is currently lacking (85) and could ease interpretation of future study results as well facilitate study comparability.

Most previously reported studies on discontinuation of follow-up care in patients with CHD are performed in either Canada or the United States of America (14, 16-28, 87) and few studies have been published from other types of health care systems. In addition, most of our current knowledge on discontinuation of follow-up care is retrieved from single centre studies, which do not account for inter-institutional variations, which is a prerequisite if you want to investigate hospital- and health care system-related factors.

Considering the current insights and identified knowledge gaps, this thesis includes four studies, performed in Swedish and Belgian settings, with the aim of exploring factors affecting discontinuation of follow-up care after transfer in young people with CHD. Discontinuation of follow-up care will be investigated, firstly from a multicentre perspective, secondly from the perspective of young patients, (to investigate their view on barriers and facilitators for continued follow-up care), thirdly from the perspective of HCPs (to investigate their view on barriers and facilitators for continued follow-up care) and fourthly, from a conceptual perspective, to enhance conceptual clarity of this concept from a generic perspective.



The overall aim of this thesis, and its included studies was to explore discontinuation of follow-up care among young people with CHD by addressing the following set of research questions:

From a multicentre perspective, what is the proportion of patients with CHD who continue or discontinue follow-up care after transfer?

*(Study I and III)*

From a multicentre perspective, which hospital-related factors could be associated with discontinuation of follow-up care in young people with CHD?

*(Study I)*

What are the barriers and facilitators for continued follow-up care after transfer, as perceived by young adults with CHD?

*(Study II)*

What are the barriers and facilitators for continued follow-up care after transfer, as perceived by HCP in paediatric cardiology and ACHD care?

*(Study III)*

What are the terms being used in published literature to describe discontinuation of follow-up care for young people with CCCs and how are these terms defined and operationalized?

*(Study IV)*



# 4. Theoretical perspectives

Ontology and epistemology are two closely related concepts. Our ontology describes the nature of reality, what reality is. Epistemology describes our view of knowledge, what can be known about our reality and how do we obtain such knowledge.

A paradigm can be described as an ontological perspective or view of the world. The positivist paradigm dates far back and simply stated assumes that there is *one truth* of the world, driven by natural causes, a truth which can be investigated and is independent of human observations, meaning that the truth is not created within the human mind. Objectivity is highly valued within this paradigm and personal beliefs are considered to be sources of bias. Pure positivism is rarely implemented in modern day research. The post-positivist paradigm acknowledges that total objectivity is not realistic, but does still strive towards objectivity, and wants to determine the “probable” true state of the world. These two paradigms put emphasis on the objective perspective and mainly use quantitative research methods, often including hypothesis testing, quantification, or prediction (92).

The constructivist or naturalistic paradigm sees the truth of the world as being mentally constructed. Truth is therefore deemed subjective and interpreted within people’s minds. Truth is investigated through an interactive process between the researcher and those being researched. The focus is often to understand lived experiences and qualitative methods are favoured including

subjective and non-quantifiable data, narratives and in-depth seeking of understanding (92).

The paradigm referred to as pragmatism puts emphasis on the research question at hand rather than methodological approaches or philosophical perspectives. The type of research question determines the direction of the research. The methodological approach which will provide the best evidence to answer the research question is considered the most appropriate (92).

In the present thesis, both the objective truth and the subjective truth are considered and highlighted, and the nature of the investigated phenomenon is acknowledged to be complex. The present thesis is inspired by the pragmatic paradigm, and the direction and methodological approaches of this thesis and included studies have been guided by the research questions at hand.

In the quantitative measures and inquiries of this thesis, the objective truth was sought and as researchers we strove for objectivity and independence from the data. Within the qualitative inquiries of this thesis, reality was considered constructed and interpreted in the interaction between researchers and participants, and while we as researchers were striving for objectivity, we also acknowledged that we were part of the generation of research results.

In this thesis, discontinued follow-up care is considered both as an objective, a measurable phenomenon, but also as a phenomenon affected by subjective experience. Factors affecting continued follow-up care are considered to be partly measurable, but also strongly affected by subjective experiences and human interactions. In line with the field of health care science, which will be further described below, this thesis also considers subjective perceptions as crucial to understanding the mechanism behind discontinuation of follow-up care.

Experiences and perceptions cannot be objectively observed, they can only be described through interpretations. These interpretations are affected by prior knowledge and understanding of the phenomenon which requires active reflection to ensure neutrality and avoid bias. In this project, the research group contributed with different perspectives and prior knowledge of the

phenomenon. My own preunderstanding and professional experience included mostly inpatient paediatric cardiology care, which has given me a good understanding of patient experiences and care trajectories before transition to adulthood and transfer to adult care. However, I had no previous professional experience of either outpatient care or ACHD care. On the one hand, this could have enabled a more open-minded and neutral view of the stories of patients and health care providers during this project, but on the other, it could also be considered a limitation since experiences of care delivery in the adult setting could ease understanding of perceptions and experiences of patients and HCPs.

# THEORETICAL REASONING

From a theoretical perspective, discontinuation of follow-up care is viewed in this thesis firstly as a health-related event and the study of its prevalence and determinants could therefore be considered an epidemiology (93). Secondly, discontinuation of follow-up care is viewed as a matter of quality of care and patient safety and an issue related to health care delivery at large.

Transition and transfer of care could be considered contextual factors, since discontinuation of follow-up care can exist without the elements of transfer or transition. However, in the context of young people with CCCs transferring their care from paediatric to adult care and thereby going through an organizational transition from paediatric to adult focused facilities, discontinuation of follow-up care could be seen as an outcome indicator of an unsuccessful transition process. The theory of transition (72) is therefore seen as a useful perspective when interpreting perceptions and experiences of this phenomenon and is used within this thesis as a theoretical basis for interpretation and discussion of the findings.

Transition theory and quality of care are both concepts of high relevance to the scientific discipline of health and care sciences, which serves as an overall context for the present thesis.

Health care science is a relatively young discipline that has grown immensely over the past decades. Initially, this discipline was referred to as nursing science but the label was perceived to be limiting for researchers without nursing or care-related backgrounds so the discipline was renamed health care science (94). Currently no established definition for health care science exists and its delimitation towards other disciplines is somewhat vague. Initially, definitions of health care science focused on the professional inheritance of the researchers performing the research and later shifted towards focusing on the type of scientific research problems being investigated (94).

The borders of health care science towards other disciplines can be somewhat difficult to determine. Health care science and medical science share many attributes, including some scientific methods but they are somewhat different

in terms of theoretical approaches and in their view of knowledge. To provide an example, in medical science, “health” more often refers to the physical dimension and is more often measured objectively and quantitatively. In contrast, from the perspective of health care science, “health” always encompasses more than the physical dimension, and the patients’ subjective perceptions and experiences of “health” are considered just as central as any objective measure (94). It is nevertheless worth mentioning that in recent decades, biopsychosocial perspectives on health have been more common in medical science too.

Health care science is a broad field and addresses, for example, issues and interventions related to health, illness, and care, as well as health care organization and strategies to maintain and improve health. The perspective of quality of care within the present thesis fits well with the more organizational dimensions of health care science. Examples of scientific questions within health care science are the perceptions and experiences of patients, as well as issues and measures related to health, illness, health care organization or preventive strategies (94). Exploring discontinuation of follow-up care in terms of prevalence, associated factors, and conceptual characteristics is therefore considered relevant for the field. Within the present thesis, the theoretical perspective of transition further contributes to a more subjective perspective, in line with the values of health care science.





# 5. Methods

## OVERALL CONTEXT

This doctoral position was a double degree obtained in collaboration between the Institute of Health and Care Sciences, University of Gothenburg, Sweden and the Department of Public Health and Primary Care, Academic Center for Nursing and Midwifery, KU Leuven, Belgium.

The studies, presented and performed within this thesis, are part of the Adole7C-project (AdolesCents reCeiving Continuous Care for Childhood-onset Chronic Conditions). The Adole7C-project is a bilateral research project and a collaboration between Sweden and South Africa, with the overall aim to examine the role of patient-, hospital- and healthcare system-related factors associated with discontinuation of follow-up care. Although the Adole7C-project is generic in nature, CHD was selected as the primary focus of this thesis, since the prevalence of discontinuity of care is particularly high within this patient group and can have a detrimental impact on both the current and future health of this population.

The present thesis describes results from the Swedish part of the Adole7C-project, with a focus on young people with CHD. The thesis also includes a Belgian perspective on discontinuation of follow-up care, as well as an investigation of perceptions and experiences of Swedish patients and Swedish and Belgian HCPs. Included in this thesis is also a descriptive conceptual study comprising young people with different types of CCCs.

# SETTING

The included studies have primarily been performed in Sweden and Belgium. - two countries which together could serve as good examples of European health care settings. The following section provides a brief overview of the two countries in terms of health care system and organization of CHD care.

## HEALTH CARE SYSTEMS & ORGANIZATION OF CARE

To understand how care is structured and provided in a country, one should consider the overall health care system, as well as its specific characteristics. The National Health Services systems (NHS), also referred to as the Beveridge model, and the Social Security Health care system (SSH), also referred to as the Bismarck model, are two major types of health care systems. The major difference between the two models mainly relates to the funding principles of health care services, influence and power over health care and accessibility (95). The overall health care system provides a structure, but it is often the details which determines whether the system will be successful. Such details could be local resources, price regulations or out-of-pocket costs for patients.

# SWEDEN

## HEALTH CARE SYSTEM

Sweden applies the Beveridge model with tax funded health care. Budget responsibility and major influence and power over health care lies with the government. The structure of different health care levels is pyramid shaped, with access to specialist care dependent on referral from lower levels of care (95). Health care in Sweden is organized in 21 different regions of varying size and geography, providing health care for the population. Certain subspecialties, e.g paediatric cardiac surgery and ACHD surgery, are centralized on a national level to ensure sufficient concentration of competence.

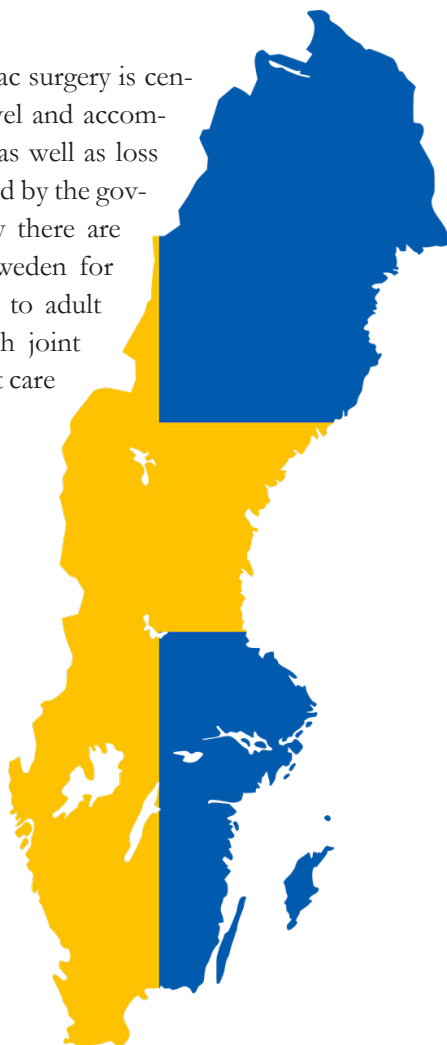
All health care services for children are free of charge until 18 years of age, including inpatient care, outpatient care, and prescribed medications. Some regions extend the free-of-charge period until 20 years of age. Out-of-pocket costs for adult health care services vary depending on the level of care, and minor variations also exist across regions in the country. Outpatient care expenses exceeding 112 euros per 12-month period are fully covered by the government. Cost for medications exceeding 228 euros per 12-month period is fully covered by the government. Inpatient care costs 10 euros per day, regardless of type of treatment, with a maximum cost of 145 euros per month.

## CONGENITAL HEART DISEASE CARE

All new-born children in Sweden are examined by a physician before maternity ward discharge, most commonly, by the paediatric service, which in combination with antenatal screening programs during pregnancy and pulse oximetry screening programs for new-born children (96, 97), makes Sweden a country with ample resources for early detection of CHD. A child born in Sweden will also receive around 12 visits to the governmental child healthcare centre, from birth up until 5 years of age. During the first year of life a paediatrician attends three of those visits.

If diagnosed with CHD in Sweden, the child will, if needed, receive paediatric cardiology follow-up care until 18 years of age. At approximately 18 years of age, the young person will be transferred to adult cardiology care. In Sweden, there are seven tertiary paediatric cardiology centres and seven tertiary specialist ACHD centres. In addition, there are also “shared care” establishments, where follow-up care is performed in collaboration between a regional hospital and a specialist centre as well as “satellite centres” where cardiologists from specialist centres have “out-reach” consultations in regional hospitals. Shared care and satellite clinics occur in both the paediatric and adult care settings.

Paediatric and complex ACHD cardiac surgery is centralized in two national centres. Travel and accommodation expenses for the families, as well as loss of income for parents, is partly covered by the government insurance system. Currently there are no formal transition programs in Sweden for transferring patients from paediatric to adult care facilities but local routines with joint meetings between paediatric and adult care are common.



# BELGIUM

## HEALTH CARE SYSTEM

Belgium applies a Bismarck inspired model which is based on a compulsory health insurance which covers 99% of the population (98). Compared to Sweden and the Beveridge model, the Belgian government has less influence over health care. In addition, access to the different care levels is more parallel shaped than Sweden's hierarchical structure for care access (95).

Health care is mainly funded through social contributions, meaning people pay a percentage of their income to the government, which then finances insurance. The contribution is proportional to the person's level of income and one part is paid by their employer. The actual amount that patients must pay for health care in Belgium varies between patients. Most health care costs are covered by compulsory insurance but there are some exceptions. Consequently, most Belgian citizens choose a non-compulsory hospitalization insurance, to cover additional costs. National fees for different types of care are established and that fee is mostly automatically reimbursed to patients based on national agreements between the government and health insurance companies. However, part of the fee is charged to patients in the form of "co-payments". The co-payment is generally a fixed amount that patients pay upfront for a specific health care service, for example a medical consultation or a blood sample. The size of the co-payment can vary, depending on type of provider and patient's level of reimbursement. Patient's level of reimbursement is mostly affected by social status and income (98). In general, the co-payment for an outpatient consultation to a medical specialist ranges from 3-12 euros, depending on the patient's level of reimbursement. If examinations are needed on top of the consultation, such as an echocardiography or a blood sample, co-payment fees for these examinations are added. Consequently, an average out of pocket cost for ACHD follow-up is difficult to estimate. There are protective mechanisms in place to avoid large health care costs for patients, such as a maximum amount. The size of the maximum cost is based on household income as well as social status, for example chronic conditions (98).

## CONGENITAL HEART DISEASE CARE

Like Sweden, a child born in Belgium will receive around 12 visits to the governmental agency for children and family (48). These visits, in combination with high quality antenatal screening programs during pregnancy and paediatrician examination of the new-born within 5 days after birth, provide ample resources to detect CHD at an early stage (48).

Belgium has seven tertiary paediatric cardiology centres. Paediatric cardiac surgery is not centralized in Belgium and currently performed at four of the paediatric cardiology centres. Transfer from paediatric to adult care is standard practice in Belgium, but there is no formal age for transfer across the country. At the University Hospitals Leuven, for example, young people are transferred to adult care at 16 years of age, if their cardiac condition is medically stable (20). In Belgium, there are six ACHD centres and like paediatric care, adult cardiac surgery is not centralized. In addition, there are also shared care establishments and satellite centres in both the paediatric and adult care setting. There is currently one hospital in Belgium providing a formal transition program for transferring patients from paediatric to adult care facilities (i.e. University Hospital of Ghent).



# STUDY DESIGNS

This thesis can most accurately be described as a multiple-method research project. When it comes to study-specific design, the included studies were of descriptive, retrospective cross-sectional designs as well as descriptive qualitative designs. Data collection methods include registry and medical file data, surveys, individual interviews, and a review of scientific literature. Apart from documented medical file and registry data, data sources included patients, HCPs, and researchers within the field. Both quantitative and qualitative analysis methods were used, including statistical analysis, qualitative content analysis (99) and modified summative content analysis (100). This section will go deeper into these respective designs and methods in general and then in more detail for each individual study.

## DESCRIPTIVE & RETROSPECTIVE STUDIES

A descriptive study aims to observe and describe a phenomenon and to provide an accurate description of its natural occurrence, for example its characteristics, circumstances or frequencies (101). A retrospective study design goes back in time and starts with identification of a phenomenon which exists in the present and then attempts to link the present phenomena to events or phenomena which occurred in the past. It can also be described as an examination of correlation between a dependent variable from the present and independent variables from the past (101). The strength of a descriptive retrospective study lies in the opportunity to observe the natural occurrence of a phenomenon and the limitations mainly lie in the lack of casual conclusions.

# CROSS-SECTIONAL STUDY DESIGNS

Two studies in the present thesis, include cross-sectional elements in their design (I and III). A cross-sectional design implies data collection at a given point in time, and allows the captures of a phenomenon or an outcome at a specific time point (101). The nature of a cross-sectional study is often descriptive, with the purpose of determining the prevalence of an outcome at a given time point, and to capture associations between the outcome and hypothesized risk factors (102). A cross-sectional design is static in the sense that it provides no information with regard to the effects of time upon variables, nor does it provide information on causal relationships between variables (103). It is, however, an appropriate design for describing the current state of a phenomenon or outcome under study (101) and for the assessment of associations between variables (103).

# DESCRIPTIVE QUALITATIVE STUDY DESIGNS

Within qualitative research, part of the aim is to understand and to describe reality from the view of study participants. Many qualitative designs clearly adhere to a qualitative research tradition and philosophy, such as ethnography or phenomenology, while others claim no particular adherence and are often referred to as descriptive qualitative designs (101). Qualitative descriptive studies are often described as closer to the data and having a lower level of interpretation, than more traditional qualitative methods (104). The major strength of a qualitative design is the in depth knowledge of participants' perceptions but one limitation is the lack of generalizable conclusions.



# OVERVIEW OF STUDIES

The present thesis includes 4 research studies, presented below in Table 1.

TABLE 1. OVERVIEW OF STUDIES

STUDIES	DESIGN	SETTING	SAMPLE	DATA COLLECTION	DATA ANALYSIS	
I	Retrospective	Sweden 7 university hospitals	Young people with CHD	Registries	Descriptive statistics	
	Cross-sectional			Medical files	Multivariable logistic regression	
				Surveys		
II	Qualitative descriptive	Sweden 7 university hospitals	Young people with CHD	Individual Interviews	Qualitative content analysis	
III	Qualitative descriptive	9 university hospitals from Sweden & Belgium	HCPs in paediatric cardiology & ACHD clinics	Interviews	Qualitative content analysis	
	<i>Retrospective</i>					
	<i>Cross-sectional element</i>			<i>Young people with CHD</i>	<i>Registries and medical files</i>	<i>Descriptive statistics</i>
IV	Descriptive conceptual	Literature	Young people with CCCs	Systematic literature review	Summative content analysis	
		International research community	Experts within the field of CCCs	Surveys		

# STUDY SPECIFIC METHODS

## STUDY I

### AIM

The specific aims of the first study (105) were to:

- Investigate the proportion of CHD patients with no follow-up after leaving paediatric cardiology.
- Determine the transfer destinations of patients who continued follow-up care after leaving paediatric cardiology.
- Identify possible variation across centres in the proportions of patients with no follow-up.
- Investigate if there is an association between the outpatient volumes at centre level and the proportion of no follow-up.
- Investigate if medical staffing resources in paediatric and adult cardiology outpatient clinics are related to the proportion of no follow-up (105).

### SETTING AND POPULATION

A descriptive, observational, multicentre study was performed, comprising seven university hospitals in Sweden, located in *Gothenburg, Linköping, Lund, Örebro, Stockholm, Umeå and Uppsala*. Both the paediatric cardiology clinic and ACHD clinic in each location participated (105).

Participants comprised adolescents born in the period 1991-1993. Further requirements for inclusion were 1. A CHD diagnosis, matching the following definition:

*“structural abnormalities of the heart and/ or intrathoracic great vessels that are actually or potentially of functional significance” (3).*

2. Attendance and formal registration of outpatient assessment in paediatric cardiology, within 5 years preceding the intended transfer to ACHD care, which in Sweden is planned around 18 years of age.

The considered time-period for paediatric outpatient assessment was January 1<sup>st</sup> 2005 – December 31<sup>st</sup> 2011, depending on the year of birth (105). Reasons for exclusion include no CHD, written dismissal from further cardiac follow-up, death, heart transplantation or relocation outside of Sweden before study inclusion (105).

## PROCEDURE

Data collection began in 2016. At that point, participants were 23-25 years of age and would therefore be recommended to be in follow-up at adult focused facilities. Data collection officers (DCOs) at each ACHD clinic determined follow-up status of included participants through administrative hospital systems and medical files. Additional variables were collected, including primary and secondary CHD diagnosis, complexity of the CHD lesion, previously performed surgery, and catheter-based intervention, as well as sex and year of birth (105).

Participants with unknown follow-up status were contacted for a self-report of adult outpatient assessment after transfer. In case of no response, the Swedish Registry of Congenital Heart Disease (SWEDCON) was searched for any documented adult outpatient assessments (105).

Each participating clinic, both paediatric and ACHD, completed a hospital factor survey for the period 2008-2012. The survey comprised questions about staffing resources, characteristics of the hospital, the clinic as well as care and transfer processes (105).

## DEFINITIONS

### COMPLEXITY

The primary CHD defect was used for categorization, based on a modified version of CONgenital COR Vitia hierarchy (CONCOR) (48). Anatomical complexity was determined as either mild, moderate, or severe complexity, in line with Task Force 1 of the 32<sup>nd</sup> Bethesda Conference (45).

### FOLLOW-UP STATUS

The observation window reached five years beyond intended transfer at 18 years of age. Follow-up status was determined as either “In follow-up care”, “Not in follow-up care” or “Untraceable”. If participants had at least one cardiac outpatient visit during the five-year period that was either documented or self-reported by the participants, they were considered “In-follow-up care”. If there were no documented outpatient visits and a self-reported statement of no outpatient visits, participants were considered “Not in follow-up care”. When the follow-up status could not be determined based on registries or self-reports, participants were considered “Untraceable” (105).

### TRANSFER DESTINATIONS

In line with the three levels of care as described by Deanfield (51), the level of care after transfer was determined as either specialist care, shared care, or non-specialist care. Specialist care was defined as follow-up provided by an ACHD cardiologist or paediatric cardiologist, performed at a tertiary health care facility. Shared care was defined as care provided in collaboration between an ACHD cardiologist and general cardiologist. Non-specialist care was defined as care provided by a general cardiologist or general practitioner (105).

## OUTPATIENT VOLUMES AND MEDICAL STAFFING RESOURCES

The average number of outpatient visits to each participating outpatient clinic was collected to demonstrate outpatient volumes. Within the paediatric setting, CHD patients, consultant referrals as well as screenings were included. In the adult setting, only CHD patients were represented (105). In the present study, cardiologists and fellows available for CHD outpatient assessments were considered “Medical staff”.

## DATA ANALYSIS AND STATISTICAL METHODS

### CHI-SQUARE TEST AND FISHER'S EXACT TEST

When searching for differences between groups regarding categorical/nominal variables like “Follow-up care”, the Chi-square test and Fisher’s exact test can be useful. Both the Chi-square test and Fisher’s exact test considers the relationship between the observed and the expected counts in a cross tabulation (106).

The chi-square test is a non-parametric test which is appropriate for use on nominal data, unequal sample sizes, or when the assumptions of either normal distribution, equal variance or homoscedasticity are violated (107). A chi-square test can be calculated manually or by using statistical software. In the present study, analysis was performed using IBM SPSS Statistics for Windows, version 24 (IBM Inc., Armonk, NY, USA). Interpretation of the chi-square test can be made by the Chi-square-value and respective p-values (107). In the present study, a p-value of  $< 0.05$  was considered statistically significant. In this case, a significant p-value would indicate that there is a statistically significant difference between the observed and the expected counts in the table and thereby a statistically significant difference between groups.

When the expected count in a table cell is less than 5, Fisher's exact test can be an option. Fisher's exact test is similar to the Chi-square test, it is non-parametric and therefore suitable for use in categorical/nominal data. However, a difference to the Chi-square test is that the Fisher's exact test calculates probabilities. Simply put, the test generates all possible tables with the same row and column total as the original table and calculates the probability of each table to occur. If a generated table has the same or lower probability of occurring as the original table, this indicates differences across groups. In the end, the sum of all probabilities of generated tables provide the probability of observing a table supporting the alternative hypothesis, which in this case is statistically significant differences between groups (108).

The Fisher's exact test is mathematically complex and requires the use of statistical software. Initially a Fisher's exact test was only considered appropriate for 2x2 tables. However, with the help of modern technology and advanced statistical software it can also be applied to larger tables (108), often using an extension of Fisher's exact test called the Fisher-Freeman-Halton test. In the present study, the Chi-Square test was used to compare patient characteristics between hospitals and Fisher's exact test was used for comparing proportions of discontinuation of follow-up care across hospitals.

## LOGISTIC REGRESSION ANALYSIS

When using regression analysis, dependence of one variable upon another independent variable is investigated. The nature of the dependent variable affects the choice of the respective regression method. In the present study, the dependent variable is categorical, and therefore not suitable for linear regression. The alternative for a categorical outcome is logistic regression analysis, which calculates the odds ratio of an outcome given certain information from independent variables. In contrast to linear regression, which uses least-squares estimate, logistic regression models the probability of a specific outcome. Results are expressed in terms of odds-ratio (OR) and confidence intervals (CI). The OR indicates change of odds for a specific outcome based on one unit change in the independent variable (109).

In the present study, logistic regression was applied to determine if there was an association between *medical staffing resources* or *outpatient volumes* and *continued follow-up care* after transfer. Within the regression model, one scale-step corresponds to 500 paediatric outpatient visits or 50 adult outpatient visits (105). Medical staffing was expressed in terms of full-time equivalence (FTE). To compare FTE across clinics, a ratio calculation was used, determining FTE per 1,000 outpatient visits (105).

# STUDY II

## AIM

The specific aim of the second study was to describe factors affecting (dis)continued follow-up care after transfer, as perceived and experienced by young people with CHD.

## SETTING AND POPULATION

In the present study, individual interviews were conducted with participants from the cohort of Study I. The ambition was to understand, from the perspective of the young people, which factors affected or hypothetically could have affected whether they to continue follow-up care or not after transfer. In the present study, both participants with and without follow-up care after transfer are represented. Participants were recruited from all seven university hospitals in Sweden. The cohort from study I comprised 654 patients, born in 1991-1993 and diagnosed with CHD. All participants with a follow-up status of either “In follow-up care” or “No follow-up care” in Study I, were considered eligible for inclusion in the present study. A mixed purposive sampling technique was used (110).

## INCLUSION OF PARTICIPANTS “IN FOLLOW-UP CARE”

Inclusion of participants “In follow-up care” began with a maximum variation approach (111, 112), where three key dimensions were considered. The ambition was to attain similar proportions of participants within the three key dimensions, as reported in Study I. The key dimensions were: sex, CHD-complexity, and clinical location. These dimensions were selected because previous studies reported them to be associated with discontinuation (20, 21, 24, 25, 105, 113). As a last step, after the above selection, a purposeful random sample was taken among available participants (110-112).



## INCLUSION OF PARTICIPANTS “NOT IN FOLLOW-UP CARE”

Inclusion of participants characterized as “Not in follow-up care” was a convenience sample (111, 112), due to the limited number of participants considered to be “Not in follow-up care” (110).

“Untraceable” participants were excluded, as well as participants with cognitive impairment and participants with difficulties to speak and understand Swedish (110).

## PROCEDURE AND ANALYSIS

Invitations to participate in the study were sent by post. Participants provided written informed consent and interviews were conducted by phone when convenient for the participants. A semi-structured interview guide was used during the interviews. The guide contained open-ended questions about their transfer of care and factors perceived to affect continued follow-up. With the consent of the participant, all interviews were audio recorded. Interviews were then transcribed verbatim in preparation for analysis. Data were analysed using an inductive approach to qualitative content analysis, as described by Graneheim and Lundman (99, 110). Qualitative content analysis adheres to constructivism, and its assumption that knowledge is generated through interaction. Qualitative content analysis is a systematic and structured analysis of data, which does not rely on any philosophical foundation. An inductive approach was used in this study, meaning that prior assumptions and existing theory were not applied to the analysis. The manifest content, which refers to the clear and obvious components of the data were inquired.

# STUDY III

## AIM

The overall aim of the third study was to explore patient-, hospital-, and healthcare system-related factors that could affect (dis)continued follow-up care after transfer among young people with CHD, as perceived and experienced by paediatric cardiology and ACHD HCPs acting in low-prevalence settings for discontinuation of follow-up care.

## DESIGN

A descriptive qualitative design was adopted, including individual interviews with HCPs to investigate factors that could affect continued follow-up care, as perceived and experienced by paediatric and adult HCPs in low prevalence settings for discontinuation of follow-up care. A cross-sectional element was added to this study to confirm the “low prevalence status” of included settings.

## SETTING AND POPULATION

Participants in the individual interviews were HCPs acting in either paediatric cardiology or ACHD outpatient care at nine European university hospitals, with seven from Sweden, located in *Gothenburg, Linköping, Lund, Örebro, Stockholm, Umeå* and *Uppsala* and two from Belgium, located in *Leuven* and *Ghent*. Eligible participants were either cardiologists, nurses, or administrative staff, working in either ACHD outpatient care or paediatric cardiology outpatient care at one of the nine university hospitals. Additional eligibility criteria included having worked at least a year at the respective outpatient clinic and having direct contact with young patients with CHD.

## DATA COLLECTION

Interview data were collected between 2021-2022 in both Sweden and Belgium. Eligible participants were approached for participation after contact with their superior. They were informed about the purpose of the study and provided consent for participation. Semi-structured interviews were performed digitally or by telephone at a time convenient for the participant. A semi-structured interview guide with open ended questions was used and interviews were audio recorded after permission from the participants.

Data was collected in two stages. In the first stage, ACHD HCPs were interviewed, and the data were analysed. Based on the results of the ACHD interviews, the interview guide was complemented to target specific findings and confirm and extend the results from the ACHD interviews. Interviews were then held with paediatric cardiology HCPs and analysis was performed.

## DATA ANALYSIS

Interview data was subjected to qualitative content analysis as described by Graneheim and Lundman (99). Analyses included both inductive and deductive approaches (114). First, the ACHD interviews were analysed inductively, generating categories and subcategories. Secondly, the paediatric HCP interviews were coded deductively based on the subcategories identified from the preceding ACHD interviews. Any data not fitting the pre-existing ACHD categories would have been inductively analysed and additional categories and subcategories created.

## PROCEDURE FOR CONFIRMATION OF LOW PREVALENCE SETTINGS

To confirm that both Sweden and Belgium could be considered low prevalence settings, percentages of no follow-up and transfer destinations were reported as hospital setting characteristics in the present study. Swedish data on percentages of no follow-up and transfer destinations from Study I were also included in this third study. Swedish data were collected from 2016-2017. New Belgian data on percentages of no follow-up and transfer destinations were collected in 2020-2021 for the purpose of this third study, using methodology similar to that of study I, that was applied in the Belgian setting (105).

Included patients in both Sweden and Belgium were born between 1991-1993, attended paediatric outpatient care from 2005-2011 and were diagnosed with CHD defined as:

*“structural abnormalities of the heart and/ or intrathoracic great vessels that are actually or potentially of functional significance” (3).*

Reasons for exclusion were written dismissal from follow-up by cardiologist or either death, prior heart transplantation or relocation outside of Sweden or Belgium before study inclusion (105). Data was retained from medical files and registries using DCOs at each site. Collected variables included follow-up status and transfer destination. An observation window of five years after intended transfer was considered. Additional variables, including primary and secondary CHD diagnosis, complexity of CHD lesion, as well as sex and year of birth, were collected (105). Patients with unknown follow-up status were contacted for a self-report of adult outpatient assessments after transfer.

The patients' primary CHD defect was used for categorization and determined based on a modified version of the CONgenital COR Vitia hierarchy (CONCOR)(48). Anatomical complexity was determined as either mild, moderate, or severe, in line with Task Force 1 of the 32<sup>nd</sup> Bethesda Conference (45).

Follow-up status was determined as either “In follow-up care”, “Not in follow-up care” or “Untraceable”. A documented or self-reported follow-up

visit with any type of cardiologist within five years post-transfer was considered “In-follow-up care”. If there were no documented or self-reported cardiac follow-up visits within 5 years post-transfer, participants were considered “Not in follow-up care”. If follow-up status could not be determined through medical files and the patient could not be reached for self-report, the patient was considered “Untraceable” (105).

In line with Deanfield (2003), the level of care after transfer was determined either specialist care, shared care or non-specialist care. Specialist care was defined as follow-up care provided by an ACHD cardiologist at a tertiary health care facility. Shared care was defined as care provided in collaboration between an ACHD cardiologist and general cardiologist. Non-specialist care was defined as care provided by a general cardiologist or general practitioner (105). For the present study, patients remaining in paediatric cardiology were reported as a separate level.

# STUDY IV

## AIM

The specific aims of the fourth study were to:

- Provide an overview of terms and definitions used to describe discontinuation of follow-up care for young people with CCCs.
- Clarify operational components of discontinuation of follow-up care.
- Develop conceptual definitions and propose terms for discontinuation of follow-up care.
- Perform an expert-based evaluation of developed terms and definitions for discontinuation of follow-up care (115).

## DESIGN

The present study includes a systematic literature search for data collection, an inductive summative content analysis and an expert evaluation using a Delphi-inspired survey.

## LITERATURE SEARCH

We first included publications identified in a previous systematic review, to update the search, the same previously developed systematic literature search string (85) was applied in PubMed. Publication dates from 1<sup>st</sup> October 2014 to 29<sup>th</sup> October 2018 were considered. Inclusion criteria were:

- primary research
- young people (10-25y) with CCCs included
- the aim included an investigation/determination of discontinuation of follow-up care (partly)
- papers were published in English

Eligibility of publications was assessed independently by the first and last author and differences discussed. The final sample included 40 publications of which 16 publications came from the updated search strategy, 7 publications were included through application of snowball-sampling and 7 publications were retrieved from additional resources. Moreover, 10 publications originated from the previous systematic review and the primary search of the applied string (115).

## DATA ANALYSIS

A modified summative content analysis approach was applied, inspired by Hsieh and Shannon (100). Through summative content analysis, we can explore usage of words in texts and better understand contextual usage of words and content. Both quantification of words or content, and interpretation of the underlying meaning are included in summative content analysis (100). The present analysis was performed on a manifest level and with an inductive approach. However, interpretation is always present when conducting a qualitative analysis (99, 115).

The respective analysis comprised seven steps divided into two phases.

Phase 1 started with a **retrieval of terms and their appurtenant operational definitions** from the included publications, followed by **coding of definitions based on operational components**. In the present study, operational components were thought of as parts of an operational definition which provides instructions for measurement. The first phase was completed through **clustering of the codes into categories** (115).

In phase 2, definitions were compared, starting by **grouping definitions** which represented the same terms and then making a **comparison of the codes** assigned to each definition. The comparison was performed both within groups and across groups. The process of comparing definitions was iterative and **resulted in preliminary types of discontinuations**. “A type of” discontinuation was a recurring pattern of operational components. The analysis was completed by **formulating preliminary conceptual definitions** and **suggesting suitable terms** to function as a label for each preliminary type of discontinuation. The conceptual definitions were based on included operational components (115).

## EVALUATION

The proposed conceptual definitions were evaluated using an expert panel of 12 international researchers within the field. Identified types of discontinuation and conceptual definitions were presented to the expert panel through a survey. The expert panel provided comments on the conceptual definitions and then suggested suitable terms for each type. Percentage of agreement among the experts was calculated and considered by the research group together with free text comments. If more than 70% of experts suggested the same term for a definition, agreement was considered as reached. The input from the expert panel resulted in further refinements of types, conceptual definitions and terms (115).



# ETHICAL CONSIDERATIONS

The studies included in the present thesis were performed in accordance with the 2013 Helsinki declaration from the World Medical Association (116). This declaration is a statement of ethical principles for medical research which involve human participants. The declaration states responsibility of the researchers to protect the health, wellbeing and rights of study participants, including privacy, integrity, dignity, confidentiality and the right to self-determination (116).

Study I, II and the Swedish cross-sectional part of Study III have been approved by the regional Ethics Review Board (application numbers: 632-15 and T917-16). The local ethics review boards of the University Hospital Leuven in Belgium and the University Hospital in Ghent (number: S61011) approved the Belgian cross-sectional parts of Study III.

Regarding Swedish HCP interviews in Study III, the Swedish ethical review authority waived the need for ethical approval (application number: 2021-04576). HCP interviews in Belgium (i.e. Study III) and expert surveys (Study IV) did not require ethical approval.

Integrity and confidentiality of study participants in Study I, II, III and IV were ensured through pseudonymization of all data material. Sensitive information was appropriately stored, documents containing personal data were kept in locked cabinets and computer files stored on secured servers. For participant-reported information and individual interviews, the informed consent of each participant was obtained.

Epidemiology is the study of the distribution and determinants of health-related states or events, and the application of study results to the control of disease and other health related problems (93, 117). The studies on prevalence and associated factors for discontinuation of follow-up care (i.e. Study I and III) could be considered to be partly epidemiological studies. They are based on data from registries and medical files and collected with ethical approval

but without the patient's informed consent. We have a defined patient population of adolescents and emerging adults with CHD. The main principle is to ensure the confidentiality of participants. However, by requiring an informed consent for an epidemiological study, there is risk of losing or having insufficient data, which might result in lack of power to answer the research question. Gathering data that might not sufficiently address the research question would be unethical. The benefits of the obtained knowledge from the present studies are considered to outweigh the risks of participating for the patient population.

Most prior studies on discontinuation of follow-up care for young people with CCCs have been performed in high-income countries. In these countries, such as Sweden and Belgium, access to care is something that we take for granted. However, access to care is still a challenge in many parts of the world. It is therefore important to highlight countries with remaining challenges and limited resources for care delivery. Unfortunately, such studies are not part of the present thesis, but they were part of the Adole7C project from which the present thesis originates.

In Study II, patients with cognitive impairment or difficulties in speaking Swedish were not included due to resource limitations. This could be considered an ethical issue. Indeed, patients with cognitive impairment and or language deficiency could be hypothesised to be even more vulnerable to discontinuation, given difficulties in processing information and or attaining independence. Their experiences and perception of factors affecting continued follow-up care could therefore provide valuable information. Inclusion of these individuals in future studies is therefore imperative.

The studies included in the present thesis were supported by research grants provided by the Swedish Heart-Lung Foundation (grant number 20150535 and 20160393) and the Swedish Research Council for Health, Working Life and Welfare-FORTE (grant 2016-07259). The doctoral position was funded by the Institute of Health and Care Sciences, Sahlgrenska Academy, the University of Gothenburg, Sweden. The findings of the studies within this thesis have been or will be published in peer-reviewed journals and presented at national and international conferences.





# 6. Findings

## SUMMARY OF INDIVIDUAL STUDY FINDINGS

### STUDY I

In this first study, the proportion of CHD patients with no follow-up after leaving paediatric cardiology was assessed, as well as transfer destinations of patients who continued follow-up care.

Variation across centres in terms of patient characteristics and proportions of patients with no follow-up care was sought, as well as the possible influence of outpatient volumes and medical staffing resources on continued follow-up care (105). The total number of participants in the study was 654, of which almost 60% were male. Mild, moderate, and severe complexity lesion constituted 36.9%, 48.9% and 14.2% of the total sample respectively.

Across the seven participating hospitals, a significant difference was observed in terms of CHD complexity ( $\chi^2=21.971$ ;  $p < 0.038$ ) and patients having had previous interventions ( $\chi^2 = 23.007$ ;  $p < 0.001$ ) (105).

As much as 89.7% of the participants continued follow-up care after the intended transfer. The overall proportion of no follow-up in the present sample was 6.6%. Furthermore, 3.7% of participants were found to be untraceable,

meaning that their follow-up status could not be determined by the resources at hand. Of the 89.7% who continued follow-up care after intended transfer, the vast majority (83.9%) received specialist ACHD follow-up and only one patient (0.2%) remained in a paediatric cardiology setting. The remaining patients received follow-up care within a shared care or non-specialist setting; and for a hand full of patients (0.8%), the level of care was unclear (105).

Across the seven hospitals, a significant variability in percentages with no follow-up care was observed ( $F = 14.880$ ;  $p < 0.014$ ), ranging from 0% to 12.7%. The number of outpatient visits ranged from 400-8.400 in the paediatric settings and from 120-1.100 in the adult settings. The multivariable logistic regression model concluded that the number of paediatric outpatient visits was significantly associated with continued follow-up care after intended transfer ( $OR = 1.061$ ; 95%  $CI = 1.001-1.124$ ). The higher the number of paediatric outpatient visits, the higher the odds of continued follow-up care after intended transfer. No significant association was found between continued follow-up care and the number of outpatient visits to the adult clinics. However, if including untraceable patients in the model, the number of outpatient visits to the paediatric clinic remained significantly associated with continued follow-up ( $OR = 1.072$ ; 95%  $CI = 1.021-1.125$ ) and also the number of outpatient visits to the adult clinics was shown to be associated with continued follow-up care ( $OR = 1.036$ ; 95%  $CI = 1.000-1.073$ ) (105).

The medical staffing resources at the clinics ranged from 0.8-3.0 FTE in the paediatric settings and from 0.25-2.75 FTE in the adult settings. The calculated ratio linking FTE to outpatient visits ranged from 0.4-2.3 FTE/1,000 visits in the paediatric settings and from 2.1-6.1 FTE/1,000 visits in the adult settings. No significant association was found between medical staffing and continued follow-up care (105).

## STUDY II

Of the 16 young adults being interviewed, 50% were female, 13 had continued follow-up care five years after intended transfer, and 3 had no follow-up care after intended transfer. Furthermore, 31% had mild complexity lesions, 50% had moderate complexity lesions, and 19% had severely complex lesions. The findings are presented as a whole, with an integration of perceptions from participants with and without follow-up care. This approach was deemed appropriate since there were few differences in the perceptions of participants with and without follow-up care (110). The qualitative content analysis resulted in three main categories, 'Motivation for follow-up care', 'Participation and connectedness with HCP' and 'Care accessibility', all supported by subcategories (110).

### MOTIVATION FOR FOLLOW-UP CARE

Being motivated facilitated continued follow-up, while lack of motivation was perceived as a barrier. Some participants described follow-up care as natural and important, and that they wanted to detect changes and possible interventional needs by attending follow-up. Perceiving clear purpose with follow-up and having sufficient knowledge of their condition increased motivation. Lack of perceived purpose reduced motivation and was reinforced by a low degree of symptoms and not receiving any news. Information about CHD and risks associated with discontinuation could increase motivation, as well as adherence to ongoing treatment. Struggles with acceptance or having a high degree of fear decreased motivation. In contrast, experiencing a manageable degree of fear could be facilitating. Trust in the system was deemed a barrier, as some patients did not feel comfortable about questioning lack of follow-up and years could pass before they contacted the clinic. Attitudes towards follow-up care could also be influenced by parents, both in a positive and negative direction (110).

## PARTICIPATION IN CARE AND SENSE OF CONNECTEDNESS WITH HEALTH CARE PROVIDERS

Newfound responsibility could be challenging, and forgetfulness could be a barrier to continued follow-up. Active invitation, reminders and encouragement from the clinic and family were facilitating, especially when appointments were missed. Sufficient information was important, including information on follow-up intervals, contact information and booking procedures. Upcoming transfer often created anxiety, which could be eased through the provision of information and by meeting adult staff beforehand. The period in paediatric care and continuity in paediatric HCPs created emotional bonds. Transfer of care was perceived to rupture such bonds. Quickly establishing a new relationship with adult HCPs was facilitating for continued follow-up. Patients in follow-up described a shallower connection with adult HCPs than paediatric HCPs, a stricter manner from adult HCPs, a more stressful pace in adult care and less regard for integrity, anxiety, as well as emotional and mental issues in the adult setting. They also described a sense of not belonging and feeling misplaced, particularly at integrated clinics. The sense of not belonging related to a mix of patients with other conditions, a mix with older patients and being questioned by receptionists about whether they were supposed to be there (110).

## CARE ACCESSIBILITY

Distance and time allocated to travel was a barrier, as was temporary accommodation while studying and feeling limited by the locations of ACHD clinics. Unexpected patient fees were also an issue. Facilitators were flexibility regarding time and day of follow-up, as well as careful planning of follow-up according to personal preferences and where possible in collaboration with other HCPs (110).



# STUDY III

The overall percentages of no-follow-up in Sweden and Belgium were 6.6% and 5.2% respectively, with both countries considered low prevalence settings. Percentages between the included clinics ranged from 0-12.7%. HCPs described factors perceived to affect continued follow-up care. The qualitative content analysis resulted in 3 main categories of factors, including 3-4 subcategories per main category (Figure 2).

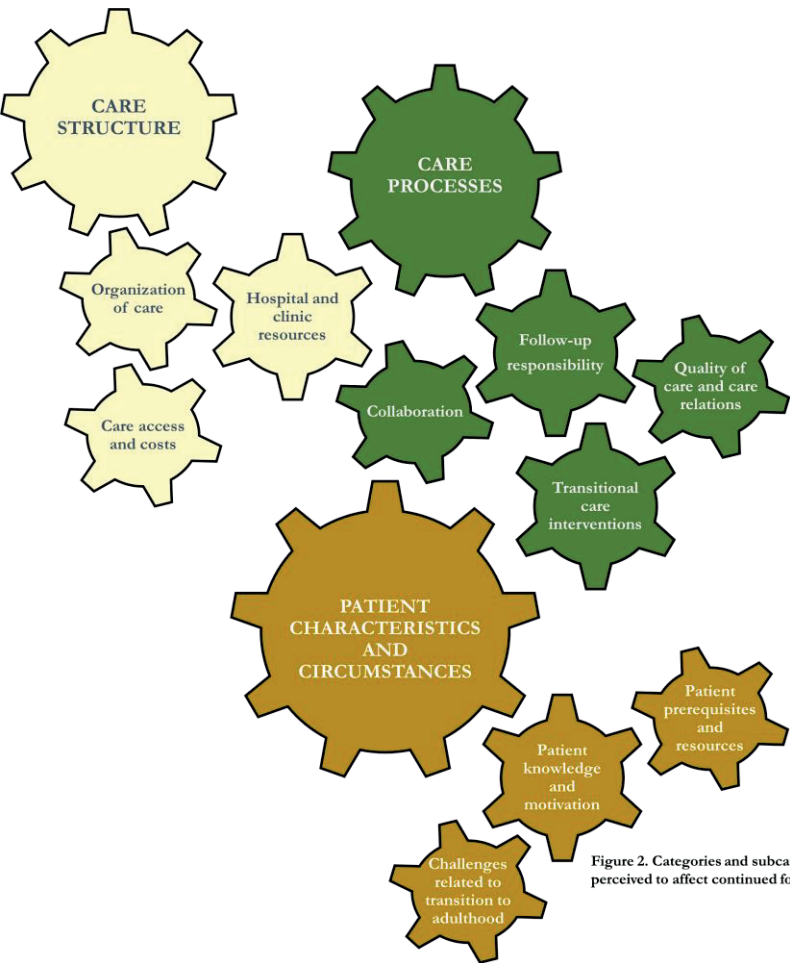


Figure 2. Categories and subcategories of factors perceived to affect continued follow-up care after transfer

## PATIENT CHARACTERISTICS AND CIRCUMSTANCES

The category “Patient characteristics and circumstances” describes challenges related to patient’s ongoing *transition to adulthood* as possible barriers to continued follow-up care, for example lack of independence, varying maturity or geographical mobility and unstable work situations. *Patients’ knowledge and motivation* was also raised as a factor affecting continued follow-up, were for example lack of knowledge about CHD and the importance of follow-up could be a barrier. Individual *patient resources and prerequisites* were also of potential influence, including CHD complexity or level of social support.

## CARE PROCESSES

*Quality of care* was highlighted as influential, including medical management continuity and *HCP competence*. ACHD competence was highlighted, as was competence about adolescence and emerging adulthood. *Care relationships* were considered influential, for example, strong bonds with paediatric HCP could be an issue and HCP continuity and a warm welcome from adult HCP could be facilitating. The patient’s own *responsibility* to attend care could be a barrier while active *responsibility* from HCPs could be protective, for example issuing invitations and reminders as well as taking active measures with patients who missed appointments to ensure future attendance. In addition, *transitional care interventions* were emphasized as being protective, including the provision of information and education, but also the availability of joint clinics and conferences as well as structured transition programs bridging paediatric and adult settings. *Collaboration*, in particular collaboration between paediatric and adult care, was emphasized as protective in facilitating not only continued follow-up but also transitional care interventions.

## CARE STRUCTURE

HCPs considered structure of care as influential for continued follow-up after transfer. Factors possibly complicating things were varying *access to care* and *high patient costs* and *clinic organization*, in terms of geographical location and distance between paediatric and adult care, as well as integrated ACHD clinics. Full time dedicated staff and adequate *resources* were considered protective, as this meant having sufficient time and an adapted physical environment to care for patients but also a reasonable ratio of patients/administrative staff.

No conflicting factors were found between HCP from paediatric versus adult care, nor between HCP from Sweden versus Belgium. However, factors were sometimes described differently or emphasized to different degrees or from different perspectives.

## STUDY IV

Discontinuation of follow-up care can conceptually be described as “Lost-to follow-up care”, “Gap in follow-up care” or “Untraceability” with *clinic visit*, *time*, and *context* as central operational components or characteristics (115).

The overall aim of the fourth study was to enhance the conceptual clarity of discontinuation of follow-up care. This study included a systematic literature search, a modified summative content analysis, input of an expert panel and research group discussions to develop and evaluate conceptual definitions for discontinuation of follow-up care (115).

In total, 40 publications were included from the systematic literature search and 47 terms and definitions were retrieved. The modified summative content analysis yielded a hierarchy of operational components (Figure 3 panel B). Five preliminary types of discontinuations of follow-up care were presented to the expert panel (Figure 3 panel A). Agreement between panellists was reached for three types of discontinuation. However, free text comments and further discussion within the research group resulted in omitting two initial types and reformulating the three remaining types. Omission was due to overlap between concepts and consideration of certain components as more contextual. In the final stage, three types of discontinuations of follow-up care were identified (Figure 3 panel A) (115).

**Panel A, Preliminary conceptual definitions, expert panel agreement rates and final conceptual definitions**

Preliminary Type and term	Preliminary Conceptual definition	Agreement rate	Final Type and term	Final Conceptual definition
<b>Lost to follow-up care*</b>	No show or not being seen for a clinic visit within a defined time period and within a defined context*	50 %	<b>Lost to follow-up care*</b>	No visit within a defined time period and within a defined context, and the patient is currently no longer engaged in follow-up care*
<b>Retention in care*</b>	Attending a clinic visit within a defined time period and within a defined context*	91 %	Considered to overlap with Lost to follow-up care and therefore omitted. *	
<b>Gap in follow-up care*</b>	A defined time interval between clinic visits within a defined context*	42 %	<b>Gap in follow-up care*</b>	Exceeded time interval between clinic visits within a defined context, and the patient is currently engaged in follow-up care*
<b>Unsuccessful transfer*</b>	Not attending a clinic visit within a defined context after transfer*	75 %	Considered a contextual factor and therefore omitted*	
<b>Untraceability*</b>	Failure to make contact due to lack of information*	83 %	<b>Untraceability*</b>	Failure to make contact due to lack of contact information*

**Panel B, Operational components**



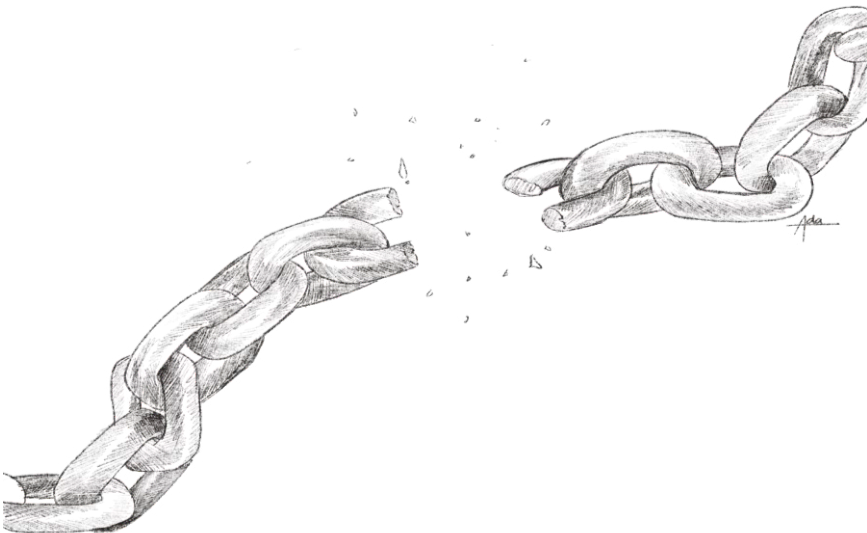
\* Skogby S, Bratt E-L, Johansson B, Moons P, Goossens E. Discontinuation of follow-up care for young people with complex chronic conditions: conceptual definitions and operational components. BMC health services research. 2021 2021/12/15;21(1):1343.

**Figure. 3 Conceptual definitions and operational components**



## 7. Discussion

In this thesis, (dis)continued follow-up care after transfer has been explored among young people with CHD. The findings of this thesis' conceptual inquiry indicated that discontinuation of follow-up care among young people with CCCs is a concept mainly concerned with follow-up care delivery within appropriate time frame and health care contexts. Previous research has suggested that discontinuation of follow-up care is associated with certain patient-related factors. Less research has been published on hospital or health care system related factors affecting continued follow-up care. To the best of my knowledge, this thesis is one of the first to address hospital and health care system related factors affecting continued follow-up care after transfer in patients with CHD. In addition to the conceptual inquiry described above, this thesis presents percentages of no follow up after transfer among young CHD patients in Sweden and Belgium, as well as barriers and facilitators for continued follow-up as perceived and experienced by both patients and HCPs.



# FACTORS AFFECTING CONTINUED FOLLOW-UP

The theory or hypothesis that hospital and health care system factors (in addition to patient related factors) can affect continued follow-up care seems even more probable based on the findings of this thesis. Indeed, the first study reported hospital differences in percentages of patients affected by discontinuation, which indicates the influence of hospital factors (105). In addition, both the patients in Study II and HCPs in Study III raised a plethora of hospital-related factors and some health care system factors perceived to affect continued follow-up care after transfer (110).

As previously stated, mainly patient-related factors affecting continued follow-up care have been described in previous literature (85). Considering the joint findings of Study II and III patient-, hospital- and health care system-related factors are all raised, which could be considered a valuable addition to the current knowledge base.

Patient-, hospital-, and health care system-related factors should most probably be thought of as linked. For example, “transitional care interventions”, which is a hospital-related factor, is most probably related to the patient-related factor “patient knowledge and motivation”, since transition programs both in theory (7, 10) and as described by HCPs in Study III include educational curricula, aimed at increasing patient knowledge. Knowledge was also perceived by patients in Study II to increase sense of purpose and motivation for follow-up care (110). Based on this reasoning, factors at different levels most likely interact. It is therefore important for clinical practice to acknowledge the possible links between patient, hospital and health care system levels and the hypothesized advantage of including all levels when designing preventive strategies.

A great plethora of factors has been highlighted in this thesis. However, which of these factors are most important is difficult to determine within the context of this thesis.



The findings show many similarities with the few previous studies on young patients' perceptions or experiences of factors affecting continued follow-up care after the transfer, for example, in a previous study young patients report aspects such as feeling well, or being unaware that follow-up was needed (21) which is in line with the findings of Study II where patients highlight lack of symptoms or knowledge as well as insufficient information (110), among others. In another previous study, young patients report feeling out of place in the adult setting (88) which is in line with the description of patients in Study II reporting the sense of not belonging in the adult clinic as affecting continued follow-up care (110).

Also, the findings of Study III show similarities with the few previous studies on young patients' perceptions or experiences of factors affecting continued follow-up care after transfer. For example, in Study III, HCPs describe the issue with integrated clinics, which have been previously raised by patients, suggesting dedicated ACHD cardiologists and separation of younger and older patients (88).

The findings show many similarities with previous studies focusing on the transition process. For example, a Swedish study investigated adolescents' experiences of preparations before transfer (78). These adolescents raised aspects related to knowledge and care relationships as affecting their transition experience, similar to the factors raised by patients in Study II as affecting continued follow-up care after transfer. Another example is the fact that both Studies II and III highlighted continued parental support as important for continued follow-up care after transfer (110). Previous studies on patients' and parents' perceptions of the transition process (76, 118) and published recommendations for transitional care (10) highlight parental support, here as affecting the transition process.

There are similarities between HCP's perceptions of factors affecting continued follow-up care after transfer in study III and previous research on HCP's views on how to best manage transition to adult care (119). For example, HCPs in a previous study highlighted inadequate preparation and education, cultural differences between paediatric and adult care as well as parental influences as barriers to transition. This previous study recommends more support and education before transfer, and increased collaboration and

communication between the paediatric and adult settings to support transition (119). These are all aspects which are raised by HCPs in Study III as affecting continued follow-up care after transfer.

The similarities between factors identified in the present thesis as affecting continued follow-up care and factors described in previous research to affect transition and transfer could be considered to further confirm the link between transition, transitional care and the issue of discontinuation of follow-up care after transfer.



## SIMILARITIES BETWEEN THE PERCEPTIONS AND EXPERIENCES OF PATIENTS AND HEALTH CARE PROVIDERS

The similarities between the perceptions and experiences of patients and HCPs in the present thesis are striking but each contributes with a unique perspective. Below follows a discussion on a **selection** of factors. Factors from Studies II and III have been jointly considered and grouped in a way thought to represent **the most central elements of the findings of Studies II and III**. The identified groups are:

- Patient knowledge and motivation
- Personal care relationships and patients' sense of belonging
- Transitional care interventions and self-care management support
- Clinic organization, resources, and competencies

### PATIENT KNOWLEDGE AND MOTIVATION

Patients' level of disease-related knowledge, motivation, and sense of clear purpose with follow-up was highlighted by patients in Study II as facilitating for continued follow-up after transfer (110). In previous studies young patients describe being unaware that follow-up is needed as a reason for discontinuation, and a desire to prevent future complications as a common reason for returning to follow-up care (21). This begs the question, how do we encourage such motivation in young patients? It can certainly be a challenge, considering the ups and downs typical of adolescence and the transition to adulthood. Young people with CHD face the same standard developmental tasks as any other young individual or peer. This means that at around 15-19 years of age, (a period which includes the point of transfer for most health care settings in the world) young patients can be struggling with self-image and identity (56). Indeed, HCPs in Study III described how patients could set aside health and follow-up in favour of other more appealing aspects of life. For a young person struggling with self-image and striving for social acceptance, de-prioritising peers and social events in favour of a medical follow-

up visit, most probably requires both knowledge and a high level of motivation, as indicated by patients in Study II (110).

Patients in Study II described how knowledge of CHD and sensing a clear purpose with follow-up care increased their motivation to continue follow-up (110). This result could indicate the importance of HCPs providing CHD education early enough for patients to grasp the information and develop motivation for further follow-up. The process of transition and transitional care interventions, including education, also needs to continue until the young person can be considered capable of full, independent care responsibility (7). Education to increase disease-related knowledge is a central aspect of transition programs (10) and the activity as such has been shown to be successful in previous studies (120) also when included in transition programs (86). A delayed transfer, meaning older age at last paediatric visit has previously been reported as a protective factor for successful transfer, as well as the patient's belief that follow-up care should take place at an ACHD clinic (27). In the light of this thesis' findings, highlighting disease-related knowledge, a delayed transfer might provide more time for education and attaining knowledge and thereby developing motivation. Patients in Study II who were currently **in follow-up care** also described feeling a lack of motivation for their follow-up (110) which might increase their risk of future discontinuation of follow-up care. For HCPs to offer suitable support and prevent future discontinuation in clinical practice, there may be a need to inquire not only into patient's disease-related knowledge, but also their motivation.

In both studies II and III, CHD complexity was raised as a relevant factor for continued follow-up care (110). This is in line with previous research reporting severe disease complexity as protective for continued follow-up and mild disease complexity as a risk factor for discontinuation (21, 24, 25). Patients in Study II described ongoing treatment and experiencing CHD-related symptoms as increasing their motivation for follow-up (110). HCPs in Study III described how patients with complex CHD are prioritized for transitional care interventions and, because of their disease complexity, receive more frequent visits. A higher frequency of visits might provide more time to provide and reiterate information and education, and thereby possibly ease the development of personal motivation. Similarly, offering transitional care interventions should probably not be guided by CHD complexity only. Patients with

mild CHD or low number of interventions have been shown to be particularly vulnerable (20, 21, 24, 25, 27) and might need such interventions, regardless of their medical examinations.

## PERSONAL CARE RELATIONSHIPS & PATIENTS SENSE OF BELONGING

Patients in Study II described the importance of trusting the adult HCPs, feeling well treated and having a sense of belonging to their new adult clinic. The feeling of belonging and trusting the adult HCPs could be challenged by integrated clinics, stressful pace, and the perceived lack of competence or regard for emotional issues in HCP's. Differences between the paediatric and adult side in terms of the manner of treatment was highlighted and emotional attachment to paediatric HCPs was described (110). Perceived differences between paediatric and adult care have previously been reported, where in the adult setting patients describe feeling rushed and like a number in the system in contrast to paediatric setting (88). The HCPs in Study III also highlighted the importance of a personal care relationship and considering "the whole" patient. Young patients with CHD carry experiences of growing up with and conquering CHD, experiences which can most clearly affect their psychosocial wellbeing. Considering the whole patient and not setting aside or presuming mental health or emotional status is therefore vastly important (121). The patient-provider relationship and anticipation about the change of HCPs at the point of transfer has previously been highlighted as an important reason for why young people can feel anxious about transfer (76, 78). Meeting the adult staff before transfer was perceived as important both by HCPs in Study III and patients in Study II, in order to increase trust in adult HCPs and to feel secure (110). Introducing the ACHD HCPs to patient and family is also considered a critical component of transition programs (10). HCPs in Study III further raised the advantage of ACHD nurses functioning as transition coordinators for patients in paediatric care, and thereafter following patients through their transfer and onwards in adult care. This approach was thought to bridge the gap and provide security. Indeed, the environmental change that follows a transfer, including change in HCPs, often induces an organizational transition process, which can lead to perceived instability (8, 71). For some patients, maintaining an HCP relationship during transfer would most prob-

ably smooth the organizational transition. From a clinical perspective, the relationship between patients and HCPs should be carefully safeguarded and considered from a long-term perspective.

## TRANSITIONAL CARE INTERVENTIONS AND ACTIVE SELF-CARE MANAGEMENT SUPPORT

In Study III, transitional care interventions were frequently highlighted by HCPs as facilitating continued follow-up care. Activities such as education, active practice of self-care management or visiting adult clinics were often mentioned. Sometimes these factors were highlighted as protective and being actively used. Sometimes they were described as a desired element to implement or improve in practice. Transitional care interventions, when applied in a structured transition program, aim to support the transition to adulthood and to achieve transitional care outcomes. Transitional care outcomes include many different aspects, for example disease-related knowledge, as described in the section above, but also include continuity of care, referring to attendance of medical appointments (10). There are indications that transition programs can have a positive impact on continued follow-up care and other transitional outcomes, such as disease-related patient knowledge (17, 23, 86). Patients in Study II also raised aspects that could be related to transitional care, sometimes in the form of barriers, such as not actively participating or being engaged in their care or not receiving enough information. Sometimes these aspects were described as facilitators, such as meeting the adult staff before transfer (110). Even though only one of the included centres had adopted a formal transition program, transitional care interventions were frequently mentioned by HCPs in Study III as protective, and many times as currently used. HCPs in Study III highlighted the importance of structured processes in this regard, which could indicate the relevance of further developing transitional care and adopting structured transition programs in clinical practice. Considering the findings of Study II and III, the arguments for implementing transition programs could be considered strengthened.

Active self-care management support was highlighted by HCPs in Study III as highly protective for continued follow-up, including aspects such as active invitation, referral, reminders, calling patients who did not show up for scheduled appointments, sending motivational letters to non-attenders or contacting general practitioners to assist in motivating patients to attend clinic.

Support could also include individual care adaptations, such as offering appointments which suite patients' schedules. Patients from Study II confirmed the importance of self-care management support, highlighting active invitation and reminders. In case of missed appointments, extra encouragement and support was perceived by patients as particularly important (110). The latter was confirmed by HCPs who were convinced that calling patients who did not show up could be highly decisive for continued follow-up. Monitoring continued follow-up is also emphasized in the context of transition programs, highlighting the importance of active invitations, reminders and offering new appointments (10). HCPs in Study III particularly highlighted the administrative staff function in this regard, as knowing the patients and being highly available to them. Indeed, in Study I, one hypothesized explanation for the success of centre 4, which had 100% follow-up, was the dedicated administrative function, actively keeping track of patient attendance (105). In clinical practice, prioritizing and organizing administrative staff functions in a way that facilitates overview of patients and personal care relationships could most likely be protective for continued follow-up.

Both patients and HCPs mentioned parental and family support as facilitating. Parental attitudes could influence patient attitudes to care and continued parental encouragement after transfer could be imperative for some patients. This can indeed be related to transitional care interventions and transition programs, where parents play an important role. In a transition program, parental support is a crucial component in which parents should also be supported in their own transition from having primary responsibility for their child's care, towards assuming a more supportive role (10). This is a process which can be challenging and requires support (76, 118). In clinical practice, the attitudes of parents should be considered as they transition towards a more supportive role and their support needs should be addressed to facilitate continued follow-up for patients after transfer.

## CLINIC ORGANIZATION, RESOURCES AND COMPETENCIES

The organization of the outpatient clinic was perceived to affect continued follow-up care. One of the clearest examples was HCPs in Study III describing how integrated clinics (meaning for example mixing ACHD with general cardiology) could be a risk factor, leading to HCPs having to divide their time between different patient groups, which was perceived to reduce the HCP's competence and thereby quality of care. This issue concerned cardiologists, as well as nurses and administrative staff. The availability of integrated clinics was also raised by patients in Study II but then described in terms of waiting rooms being shared with other specialties (110). The issue with integrated clinics have been raised in previous studies, where young patients described that they felt out of place when clinics mix young and old patients and suggested dedicated ACHD cardiologists and described a desire to be cared for in a clinic with a younger clientele (88). Based on the findings of this thesis, the idea of having separate ACHD clinics is appealing, but it is probably difficult to implement in smaller settings from a resource point of view. Nevertheless, the issue of seeing few ACHD patients and thereby risking lack of competence and quality of care is clearly worth reflecting upon when planning clinic organization. When separate clinics and full-time dedicated staff are not an option for the hospital, perhaps increased collaboration between ACHD clinics, increased number of satellite clinics or increased shared care arrangements could reduce some of these negative effects.

Collaboration between the paediatric and adult clinic were highlighted as a facilitator in Study III and lack of collaboration as a barrier, often related to lack of interest or deficient personal relationships between HCPs. Moreover, in Study II, patients described a perceived difference in the manner of treatment between adult and paediatric HCPs (110). Similarly, previous research on HCPs views on transition (119) highlights cultural differences between paediatric and adult care as well as the importance of collaboration between paediatric and adult care. Increased collaboration and mutual agreement about how to organize transfer and transitional care interventions therefore seems highly important to facilitating continued follow-up care.

Clinic resources is another important factor, raised by HCPs in Study III. Shortage of cardiologists and limited clinic time were highlighted as barriers to continued follow-up, increasing waiting time for clinic visits. The issue of



nursing staff resources was also raised, highlighting the importance of nurses having time to contact patients and be available for advice. Lack of resources have also been described in previous research, where young patients describe busy cardiologists, overwhelmed administrative staff and difficulties scheduling their appointment (88). The importance of self-care management support has been described as imperative and discussed above, this is an intervention requiring resources but is most probably a worthwhile investment, considering the possible health care burden of increased morbidity possibly arising from discontinuation (14, 15). However, such health economic consequences still warrant investigation. Similarly, the relevance of the administrative staff function is evident in the findings of Study III. If administrative staff had a manageable number of patients, small enough to get an overview and thus structure administrative routines to perform manual searches for non-attending patients, we might reduce the number of lost patients and decrease discontinuation of follow-up care.

HCP competencies are further described in Study III in relation to the period of adolescence and emerging adulthood, highlighting that HCPs should have an understanding of the need for additional support and care adaptations during these periods. Specific training and competencies in adolescent health and how to communicate with adolescents for HCP's caring for adolescent patients has previously been highlighted as important (10, 122). In Study II it becomes clear that many young adults have persisting transitional needs after transfer (110) which further strengthens the argument of continued transitional care interventions after transfer and the importance of sufficient HCP competence in adolescent health and medicine to make a significant impact.

In summary, there is undoubtedly high agreement between HCPs, patients, and previous research thus strengthening the relevance of the findings and their application to future research design and clinical preventive strategies.

## WHAT ABOUT HEALTH CARE SYSTEM FACTORS?

Few factors were related to the health care system in studies II and III. However, some factors were actually mentioned, such as out-of-pocket costs for patients. In Study II, this was described as a barrier, when the cost was unexpected (110) but in Study III it was mainly described as a facilitator, with focus on generous health care insurance or tax-funded care with low patient fees. Financial and insurance issues have previously been described, sometimes as a non-significant factor (85) but also as a common self-reported reason for leaving follow-up care, especially among patients with more complex CHD (21). Belgium and Sweden have different health care systems, but HCPs from both countries highlighted the low out-of-pocket costs for care within their respective systems as a facilitator for continued follow-up. Based on current findings, the type of health care system is probably not decisive for continued follow-up care, but it could possibly be of influence, especially in combination with patients suffering financial difficulties. In clinical practice, patients' financial situation in combination with out-of-pocket costs could be a risk factor to be aware of.

## PERCENTAGE OF PATIENT DISCONTINUATION

Based on the findings of this thesis, discontinuation of follow-up care in young people with CHD seemed to be a relatively small issue, with only 6.6% in Sweden (105) and 5.2% in Belgium, as compared to many other parts of the world reported in many prior research papers, with percentages ranging between 3.6-62.7% (54). However, comparison of these percentages across studies is problematic, given the great heterogeneity of operational definitions for discontinuation of follow-up care. Moreover, the percentages reported for both Sweden and Belgium, do not represent complete national cohorts, and should therefore be interpreted with caution. Instead, I would consider these results as an indication, that follow-up within five years after transfer in these two countries (including only ACHD settings) seems relatively successful. In addition, several methodological limitations of these two investigations should be considered and these are discussed in the methodological considerations section.

## SIMILARITIES WITH OTHER TYPES OF CCCs

The present thesis concerns young people with CHD, which is considered a model case for CCCs. Nevertheless, it is not possible, based on the present thesis to determine whether the findings would also be applicable in other types of CCCs. However, some reflections can be made. Probably, some factors defined in the present thesis could be shared with other groups of CCC, given that most factors are not described by patients and HCPs as specifically being CHD-related. Most factors are more generic, such as motivation, receiving invitations or interactions and relationships with HCPs. Different types of CCCs and care trajectories have different impacts on daily life, so the relevance or impact of respective factors might differ across types of CCCs.

Patients in Study II raised the fact that a lack of symptoms and CHD not being part of daily life could be a barrier to continued follow-up care (110). When comparing this description, with for example young people with diabetes, who have to consider their disease several times a day, the effect on daily life could be hypothesized as greater for some diabetes patients than some CHD patients in which symptoms could sometimes be completely lacking. Consequently, this barrier might be less important in a population of young people with diabetes.

In a Swedish study investigating how young adults with severe asthma experienced their transition from paediatric to adult care, the young adults described a feeling of being left out of the system, not knowing where to turn (123). This could be related to the findings of Study III, where HCPs highlighted the importance of having a clearly defined adult provider. There is probably a vast difference between CCCs regarding the structure of care and the process of finding a suitable adult provider. For this reason, the relevance of a clearly defined adult provider, and thereby the “structure of care”, might be an even larger barrier for some groups of patients where the most appropriate adult destinations might be less clear.

In view of the above, I believe that some factors identified in this thesis could also possibly be considered relevant for other groups of CCCs, even though emphasis might differ and further inquiry is needed.



# FINDINGS THROUGH THE LENSE OF TRANSITION THEORY

From a theoretical perspective, discontinuation of follow-up care could be reflected upon as a possible outcome indicator of an unhealthy transition process. Considering the context of the present thesis, this could be considered a valuable perspective. Indeed, the findings of this thesis highlight factors which could be related to different domains of transition theory. Below follow some examples, also illustrated in Figure 4.

## TRANSITION PROPERTIES

The properties of transition include several different aspects. In the reflective exercise described in the background, the event of transfer from paediatric to adult care was thought of as a property of the transition process, in the form of a critical event. Another property of transitions is the individual's engagement in the transition process (72). In the context of this thesis, engagement could be related to the patients' attempts to learn more about the condition and ACHD team. This could be further related to the findings of Studies II and III in which patients' knowledge of the condition and their motivation are perceived by patients and HCPs to affect continued follow-up care (110).

## TRANSITION CONDITIONS AND THERAPEUTICS

Within transition theory, transition conditions, are facilitators or inhibitors at three different levels (i.e. personal, community and societal), affecting the transition process and outcomes (72). *Patient knowledge and motivation*, as described in the present thesis (i.e. Studies II and III), could be considered a transition condition, in the form of a facilitator at a personal level, facilitating continued follow-up care and thereby a healthy transition process. Transition

facilitators at a personal level could also include patients' socio-economic status (72), which has been raised in Study III, as well as cultural beliefs and attitudes (72). In Study III, HCPs raised the aspect of being exposed to a culture which prioritize health and trusts medical expertise, as well as the possible influence of parental attitudes, as acknowledged by patients in Study II (110).

*“Personal care relationships and patients’ sense of belonging”* are factors which could be considered transition conditions at a community level, as well as *“Transitional care interventions and active self-care management support”* and *“Clinic organization, resources, and competencies”*.

*“Transitional care interventions and active self-management support”* could also be related to nursing therapeutics, which within transition theory, are interventions aimed at supporting the transition process and affecting patients' responses to facilitators and inhibitors (72).

## ROLE INSUFFICIENCY

Transition theory highlight role insufficiency or insufficient role mastery, referring to difficulties in fulfilling the requirements or expectations of a new role, a situation which can occur if the patient is not properly prepared for transition (9). Mastery of the new situation is also considered an outcome indicator of a healthy transition process (72). For a young person with a CCC in the transition from childhood to adulthood, this new role would, for example, include independent self-care management, and in greater detail, include attendance of medical appointments. Patients in Study II talked about forgetfulness and a need for extra support during this transition period (110). HCPs in Study III talked about the vulnerable period of transition to adulthood, and about patients prioritizing other things above follow-up in striving for normality. These are aspects which could in fact be related to role insufficiency (9). We do expect different things from an adult patient than we do from a paediatric patient, and the paediatric care culture is known to differ from the adult care (73). Perhaps HCPs should consider what that new “ACHD-patient role” should include. Do we give these adolescents a fair chance to master that role, considering the rather limited support structures and interventions currently in place at some clinics, adolescent development

(56) and the fact that brain maturation continues long beyond the age of 20 years (57, 58) ? If not, perhaps supplementary mechanisms are needed. Support needs during transition and transfer are likely to differ from patient to patient as each life and CHD trajectory is unique.

Looking at the joint findings of the thesis, several factors raised by HCPs and patients in Studies II and III could most probably be tackled by adapting current structured transition programs. For example, “*Patient knowledge and motivation*”, could be assessed in transition consultations including educational curricula, psychosocial screenings and empowerment (10). “*Personal care relationships and sense of belonging*” are also factors which could probably, at least in part, be addressed by transition programs, especially if these are coordinated by ACHD nurses, which would also facilitate collaboration between paediatric and adult care. Other factors, such as “*Clinic organization, resources and competencies*” are in many ways prerequisites for transitional care interventions. Of course, not all factors identified in the present thesis are related to transition. Indeed, health care access and out-of-pocket costs for example, are factors without clear specific links to transition, and should instead be seen as factors affecting the healthcare organisation in general. However, these factors are also likely to be affected by the patients’ transition to adulthood, yielding increased geographical mobility and greater financial insecurity.

In addition to continuity of care, transition includes many other aspects that could be likened to “outcome indicators”, for example patient empowerment, quality of life or disease control (10). It is not the intention of this discussion to dismiss such aspects, rather the focus of this thesis has been on the aspect of continued follow-up care.

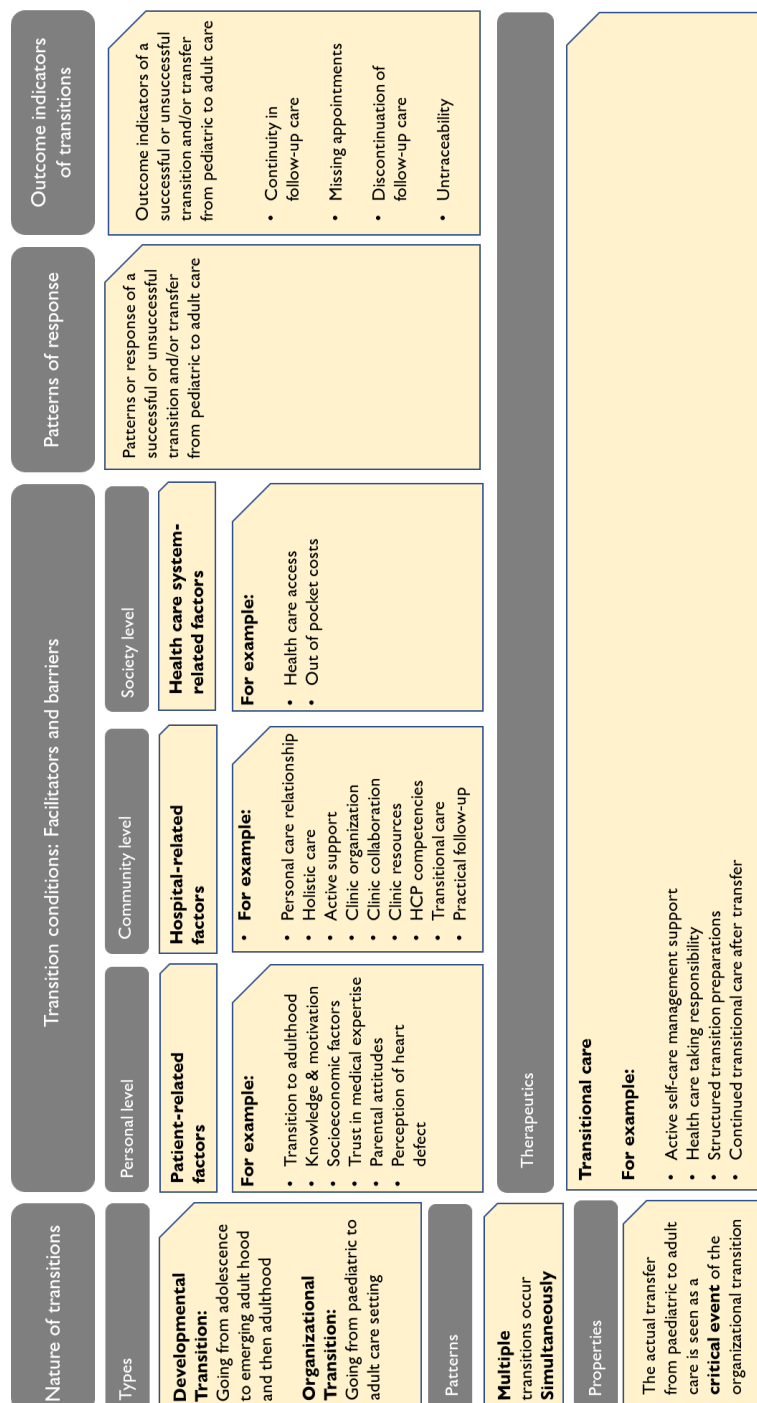


Fig 4. Modified model of Meleis et al., 2000 Transition theory.



# FINDINGS THROUGH THE LENSE OF “CONTINUITY OF CARE”

Discontinuation of follow-up care has been thought of as an “umbrella term” within this thesis, a term which covers all the different terms and operationalizations used in the current scientific literature. From a more conceptual perspective, discontinuation of follow-up care has been thought of as a broken or disrupted chain of continuous, appropriate follow-up care during life.

In the fourth study, included in this thesis, discontinuation of follow-up care was investigated from a more conceptual perspective, using scientific literature and expertise from researchers within the field. This study highlighted three main types of discontinuation of follow-up care: “*Lost to follow-up care*”, “*Gap in follow-up care*” and “*Untraceability*” (115). These three types are a representation of the concepts currently being addressed and reported in the scientific literature. These types, along with their appurtenant conceptual definitions, can guide us in the design and script of research studies investigating discontinuation of follow-up care. Hopefully, they can also contribute to a more homogeneous terminology and conceptual understanding of these concepts.

When contrasting the three types of discontinuation of follow-up care, as identified in Study IV, and the established concept of “continuity of care” it is fair to be critical, since there seems to be quite some difference between discontinuation and the established concept of “continuity of care”. Indeed, continuity of care, as defined by Reid, Haggerty, and colleagues (2002/2003) is distinguished from other concepts through two core elements, which conclude that continuity of care is a concept that 1. should be experienced by the patient and 2. should be inquired over a period of time (4, 5).

Haggerty and colleagues (2002) pointed out that “*continuity of care is how individual patients experience integration of services and coordination*” and unless we investigate the concept over time and attempt to “*understand the mechanisms through which care delivery over time improves outcome, continuity interventions may be misdirected or inappropriately evaluated*”(5).

The three main types of discontinuation of follow-up care identified in Study IV: “*Lost to follow-up care*”, “*Gap in follow-up care*” and “*Untraceability*” (115), do not include any measurement of patients’ experiences of this concept. These types are exclusively viewed and measured as objectively occurring phenomena. Regarding the aspect of continuity of care being investigated “over time”, the three main types of discontinuation of follow-up care in Study IV could perhaps be considered to include “time” as a central aspect. However, is investigating time between visits enough to consider the concept as being investigated “over time”? Perhaps “time” as defined by Reid, Haggerty, and colleagues (2002/2003) is a question of both length of time, contextual perspectives, and personal experience (4, 5). Indeed, Reid and colleagues (2002/2003) described continuity of care as follows:

*“Continuity of care is how one patient experiences care over time as coherent and linked; this is the result of good information flow, good interpersonal skills, and good coordination of care”* (4).

It is clear that the three types of discontinuation of follow-up care currently being investigated in the literature and reported in Study IV (115) differ significantly from this latter concept of continuity of care as defined by Reid et al. (4, 5).

Since “continuity of care” as defined by Reid, Haggerty and colleagues (2002/2003) puts such emphasis on the patients’ *experiences* of care (4, 5), it seems fitting to contrast the findings of the second study against continuity of care, since the second study reflects young adults’ perceptions and experiences of factors affecting continuity of care (110). After considering the stories of patients in Study II, there seem to be some aspects of similarity between their perceptions of factors affecting discontinuation and the characteristics of “continuity of care” (4, 5).

To provide some examples, *informational continuity* is one of the three types of continuity of care, whereby information from prior care is interpreted and actively used to provide appropriate care, but also to bridge both past, present, and future care. *Informational continuity* includes not only information transfer regarding medical circumstances, but also information on the patients' preferences, values, as well as past and current social contexts (4, 5). The participants of the second study described their experiences and perceptions of factors affecting continued follow-up care after transfer. Their experiences were reported using three main categories, one of which was "*Participation in care and sense of connectedness with health care provider*" which included the subcategory "*Safe and holistic care*". This subcategory describes the importance of adult HCPs having access to patients' medical information from the paediatric setting, as well as information about patients' specific health care needs (110). These are aspects which could definitely be considered related to "Informational continuity" (4, 5). In the subcategory "*Safe and holistic care*", participants also described the facilitating effect of the care-relationships for continued follow-up care, as well as the stress and anxiety which could arise from the disruption of care relationships at the point of transfer (110). These are aspects which could be considered linked to *relational continuity*, highlighting the importance of the care relationship in connecting and bridging care over time. In *relational continuity*, knowledge about the patient as a person is highlighted, as well as the ongoing relationship between a patient and a provider (4, 5). In the subcategory "*Practical and flexible follow-up*" placed under the main category "*Care accessibility*", participants described that coordination between different health care facilities could make follow-up care more practical and facilitate continued follow-up (110). This is a factor which could be considered related to *management continuity* (4, 5) which focuses on consistency of care across providers and settings. When *management continuity* is attained, required care is received in a timely manner and without duplication (4, 5).

The findings of Study III can also be viewed from the perspective of continuity of care. Perceptions and experiences of HCPs in Study III included factors which could be related to all three types of continuity of care (4, 5). For example, the proper handover of relevant information from paediatric to adult care was highlighted in Study III, an aspect which could definitely be

considered related to “*information continuity*” (4, 5). Aspects related to “*management continuity*” (4, 5) were also raised by HCPs, such as application of follow-up guidelines and reaching medical consensus between paediatric and adult care at the point of transfer. Also “*Relational continuity*” is frequently highlighted in the descriptions by HCPs, for example the ambition to bridge care during transfer through adult ACHD nurses functioning as transition coordinators, consulting patients in paediatric care, and later accompanying patients to the adult health care facility, thereby avoiding the harsh disruption of care relations at the point of transfer.

In summary, there seem to be several aspects related to the different types of continuity of care, as described by Reid, Haggerty and colleagues (4, 5), included in both patients’ perceptions and experiences in Study II and HCP’s perceptions and experiences in Study III. Considering these similarities, it might be fair to say that discontinuation of follow-up care, as we currently know it from published literature and as described in Study IV (115), is a concept which might require expansion and inclusion of individual experiences of patients, as well as informational and relational aspects of follow-up care delivery across health care settings? Indeed, based on the results from Studies II (110) and III, when contrasted in regard to continuity of care (4, 5), there seems to be a need to consider both informational, relational and management continuity in order to prevent discontinuation of follow-up care. However, one could ask, why should we follow the definition of continuity of care by Reid and colleagues? Well, firstly, the conceptual work of Reid and colleagues could be considered rigorous and broad since it covers different types of health care settings (4, 5). In addition, within the context of this thesis and from the perspective of health care science, the individual experiences of patients should be central, as from the perspective of health care science the patients’ subjective perceptions and experiences of health and care are considered as central as any other objective measurement of a concept such as discontinuation of follow-up care (94).





# 8. Methodological considerations

## STRENGTHS & LIMITATIONS

The included studies should be considered in the light of their individual strengths and limitations. Below follows a general reflection on these aspects in relation to the design of the thesis, followed by more specific reflections on validity and trustworthiness of the included studies.

The qualitative design of Studies II and III allows for a broader perspective of factors affecting continued follow-up care to be identified, in addition to previous quantitative measures. The multi centre perspective contributes to the body of literature which mainly report single centre inquiries. The conceptual work of Study IV attempts to contribute to future clarity regarding the concept of discontinuation. The thesis design does not allow for any causal conclusions to be drawn, which could be considered a limitation. However, the factors raised in this thesis, could indeed guide future inquiries, which is a valuable addition. During this research project it has proven difficult to quantitatively assess process-related aspects of care from a retrospective design, which has forced this thesis into a more qualitative approach. However, the qualitative approach is also considered a strength since it allows us to consider factors in the light of patients' and HCP's experiences.

# VALIDITY OF QUANTITATIVE INQUIRIES

The first study had a retrospective multi centre design using medical files, registries, and self-reports. The number of missing data was low, and most patients were identified as either “in” or “out of” follow-up care, which should be considered a strength. The fact that only ACHD settings were included in this study could be considered a limitation, affecting both internal and external validity. The lack of a complete national cohort and the inclusion of only university hospital settings, risks underestimating of the proportion of discontinuing patients from this group in Sweden as a whole. This would appear even more pertinent in light of the finding that larger paediatric outpatient volumes seem associated with continued follow-up care. The percentages of discontinuing patients reported in Study I should therefore not be considered representative for Sweden as a country. The fact that outpatient volumes and medical staffing resources were partially estimated at some centres in Study I could have further affected the validity of this study. The five-year observation window, which was applied in both Study I and the quantitative part of Study III, could lead to underestimating discontinuation of follow-up care percentages for patients with CHD of moderate to severe complexity, affecting validity. The fact that only 2 centres were included in the quantitative Belgian part of Study III could also be considered to reduce validity, since patients could be followed at other centres.

## GENERAL REFLECTIONS

The fact that only 600+ patients were identified in this first study was unexpected, considering the birth prevalence of CHD and the number of births in Sweden – we would have expected more to be eligible. However, because some patients are dismissed from further follow-up care during childhood, and only patients at the 7 university hospitals were included, the identified number of patients could be considered accurate. The hypothesized factors included in the hospital surveys in Study I, came back with very little variability, and few could be assessed as factors possibly associated with discontinuation. This lack of variability led to the decision to explore possible factors of relevance from a qualitative approach in Studies II and III.



# TRUSTWORTHINESS OF QUALITATIVE INQUIRIES

Trustworthiness can be assessed using positivistic or quantitative terms, such as validity and reliability, but there are other options, more related to the constructivist paradigm, such as credibility, dependability and transferability (124). Considering the use of qualitative content analysis (99) in Studies II and III and summative content analysis in study IV, the concepts of credibility, dependability and transferability seem most fitting to assess trustworthiness in these studies.

## STUDIES II AND III

### CREDIBILITY

Credibility concerns confidence in truth of data and its interpretation (124) and can be achieved through meticulous planning, execution and reporting. In studies II and III, clear descriptions of design, data collection, participants, sampling strategies, data collection and analysis techniques have been attempted to achieve credibility. Continuous discussions within the research group during data analysis have also contributed to credibility of the results. Quotation has been used in Studies II and III as an attempt to demonstrate how the reported categories corresponded with the data, which could be considered to strengthen the credibility of the studies (99).

In order to consider a sample representative, the conclusions reached from studying your sample should be the same as if you had studied the entire study population (125). A qualitative study can never reach generalizable conclusions, regardless of sample or analytic approach. Instead, a qualitative study provides us with experiences which can be considered representative within the larger population. However, clear and accurate sample selection and description is imperative and affects the trustworthiness of the results.

In Study II, participants from all seven university hospitals and both patients with and without follow-up care were included. The maximum variation approach regarding complexity and sex could be considered a supplementary contribution to credibility. However, only 3 out of 16 participants were without follow-up, which could be considered a limitation. The fact that the data collection for Study II took place approximately 10 years after the young adults had transferred their care could be considered to increase the risk of recall bias.

In Study III, HCPs from different professions, different hospitals and different countries were included, which strengthens the credibility. A limitation for both study II and III is that only university hospital settings were included.

Interviews in Study II and some interviews in Study III were performed by telephone, which could be considered a limitation. However, telephone interviews have been considered a valid alternative, lifting geographical boundaries (126).

## DEPENDABILITY

Dependability concerns data stability and reliability both over time and in between conditions (124) and also includes replication of data, analysis and results. A thorough methodological description has been attempted to demonstrate dependability. Furthermore, in Studies II and III, data collection procedures strove for consistency in using a semi-structured interview guide. Through semi-structured techniques, all relevant topics can be covered and participants can still speak freely (127).

## CONFIRMABILITY

Confirmability has to do with objectivity, that the researcher's own perspectives did not affect the data and that the results truly represent the view of the study participants (124). However, a researcher always influences both data collection and interpretation based on previous knowledge, experiences, and origin of discipline. Awareness of and descriptions of theoretical foundations and pre-understanding can ease interpretation of confirmability. Confirmability could also be strengthened through participants' confirmation of

results (99). If this is not an option, confirmation from co-researchers can be, which was the case in Studies II and III by virtue of discussions within the research group during the analysis process. Nevertheless, seeking confirmation from others is debatable from the perspective that multiple truths can exist related to subjective interpretation. This argument is sometimes described using the phrase “*methods become what they are in the hands of the researcher*” (128).

## TRANSFERABILITY

Applicability of results within a new setting or for a new group of patients can be described in terms of transferability (124). In Studies II and III we have attempted to demonstrate transferability through a rich description of the findings and the use of quotation, but also by providing cohort descriptions and participant characteristics (99). However, since only university hospital settings were included in Studies II and III and only patients who were able to speak Swedish were included in Study II, the application of the findings in other populations should be considered rather limited.

## STUDY IV

Regarding the fourth study (115), the systematic inclusion of publications could be considered to strengthen credibility. Inclusion of all different types of CCCs strengthened the transferability of the findings and the expert evaluation contributed to increased confirmability. The fact that two authors independently included publications and performed the different steps of the analysis, and thereafter discussed any differences, contributes to increased dependability (115, 124). A search string from a previous study (85) was reapplied, which could be considered a limitation since it did not cover all possible terms. That said, the previous study (85) and Study IV focused on the same phenomenon and patient population. Additionally, the search string was developed in collaboration with a librarian. Omission of certain terms was not deliberate, indeed many studies investigating discontinuation of follow-up care use specific words, such as “lapse in care” or “lost to follow-up”, but they also include more overarching terms, such as “continuity of care”, “transfer” or “transition”, terms that were included in the search string. Thus, although not all terms were included in the search strings, we still retrieved papers using terms that were not included. Since snowballing sampling techniques and additional resources of publications were used in addition to the updated search, I am not concerned that relevant publications have been missed. Only one bibliometric database was used for the updated search, which could be seen as a limitation, affecting credibility. An additional limitation is the low response rate (30%) of the expert surveys, which could affect credibility and confirmability.





# 9. Looking ahead

## CONCLUSION

Discontinuation of follow-up care among young people with CHD as investigated in published literature is mainly concerned with aspects of timing and context of follow-up care. Considering the contrast between the findings of Study IV and the established concept of “Continuity of care”, discontinuation could probably benefit from expansion to consider not only patients’ experiences of continuity of follow-up care but also to highlight informational and relational aspects of continuity in addition to today’s narrow and sole focus on management aspects.

Discontinuation in the investigated settings has been shown to be relatively low but a more comprehensive methodology is needed to confirm these findings.

A plethora of factors on patient-, hospital- and health care system-related levels have been highlighted by both patients and HCP within the framework of this thesis, and the recipe for success in keeping patients in follow-up could indeed be considered as multifactorial. In particular, hospital and process-related factors such as transitional care interventions, care relationships and clinic collaborations have been highlighted as affecting continued follow-up care although the identified factors most probably work in synergy. In addition, each patient is likely to be uniquely affected by each factor, making interventions in this area complex and probably best designed from a person-centred perspective.

Patients' perceptions of care delivery during transfer and transition seem to affect continued follow-up care, as well as personal motivation and continued self care-management support after transfer. We need to provide high quality transitional care to support transition and transfer related challenges and we need organizational structures and resources to support such care.

Based on the findings of the present thesis, discontinuation of follow-up care seems highly connected to transition to adulthood. Many of the identified factors, perceived by HCPs and patients to be affecting continued follow-up care, could be seen as facilitators or inhibitors of a transition process. Transition theory and transitional care could therefore possibly inspire and guide development of preventive strategies for continued follow-up. In addition, transition programs could probably bridge several of the highlighted barriers to continued follow-up care and could probably be a valuable instrument for prevention.

There is a high level of agreement between the HCPs, patients and previous research, which strengthens the relevance of the findings and their application to future research designs and clinical preventive strategies. However, based on the findings of this thesis, it is fair to say that further inquiries of factors affecting continued follow-up care after transfer are needed, especially when it comes to cause and effects.



# CLINICAL IMPLICATIONS

Clinical practices should, as previously mentioned, acknowledge the influence of patient-, hospital-, and health care system-related factors on continued follow-up care after transfer of young people with CHDs.

HCPs in paediatric cardiology and ACHD outpatient clinics have the opportunity to provide structured transitional care, which could potentially bridge many of the barriers to continued follow-up mentioned in this thesis and might also support patients in areas other than continued follow-up. To perform such interventions, sufficient staffing resources are needed, as well as sufficient skills and competence. Both nurses and cardiologists should make sure to attain sufficient knowledge on adolescent health and medicine to be able to provide holistic transitional care for this group of patients. Specific attention should be dedicated to supporting young patients in finding their own motivation to continue follow-up, offering long lasting personal relationships and providing early, sufficient, and repeated information.

Health care structures and resources should be considered with the transitional needs of patients in mind. Administrative staffing functions should be prioritized to identify non-attending patients at an early stage, and if possible, full-time dedicated staff and separate clinics for ACHD patients should be provided. Collaboration between paediatric and adult care regarding transitional care structure, as well as transitional care content and delivery, should be nurtured, and transitional care interventions should continue in the adult setting as standard practice rather than an exceptional intervention.

Clinical practice should also inquire what continued follow-up care might mean for the individual patient and as far as possible shape care in a manner that fits the individuals' needs and preferences.

# FUTURE PERSPECTIVES

Additional, multicentre international assessments of discontinuation of follow-up care are needed to confirm which hospital- and health care system-related factors have the most impact on discontinuation of follow-up care and to further understand the relationship between discontinuation and such factors in young people with CCCs.

The issue of discontinuation of follow-up care still needs to be determined from a broader perspective, including longer time periods covering both paediatric and adult care, but also including a broader range of care levels such as regional hospitals and primary care. National, register-based studies assessing continuity of care in national cohorts, from a life course perspective could facilitate this. In addition, a broader variety of patients should be studied in terms of their perceptions and experiences, including patients with functional variations, cognitive impairment, and language deficiencies.

Structure and process related factors of care have proven difficult to assess retrospectively. To further scrutinize the relationship between discontinuation and such factors, prospective study designs would be needed.

Patients' experiences of continuity of care and what it includes should be scrutinized, and our conceptual understanding of discontinuation further developed.

Considering the close bond between discontinuation of follow-up care and transitional practices, future research could include randomized control trials evaluating the effectiveness of transitional care interventions in terms of continued follow-up care.





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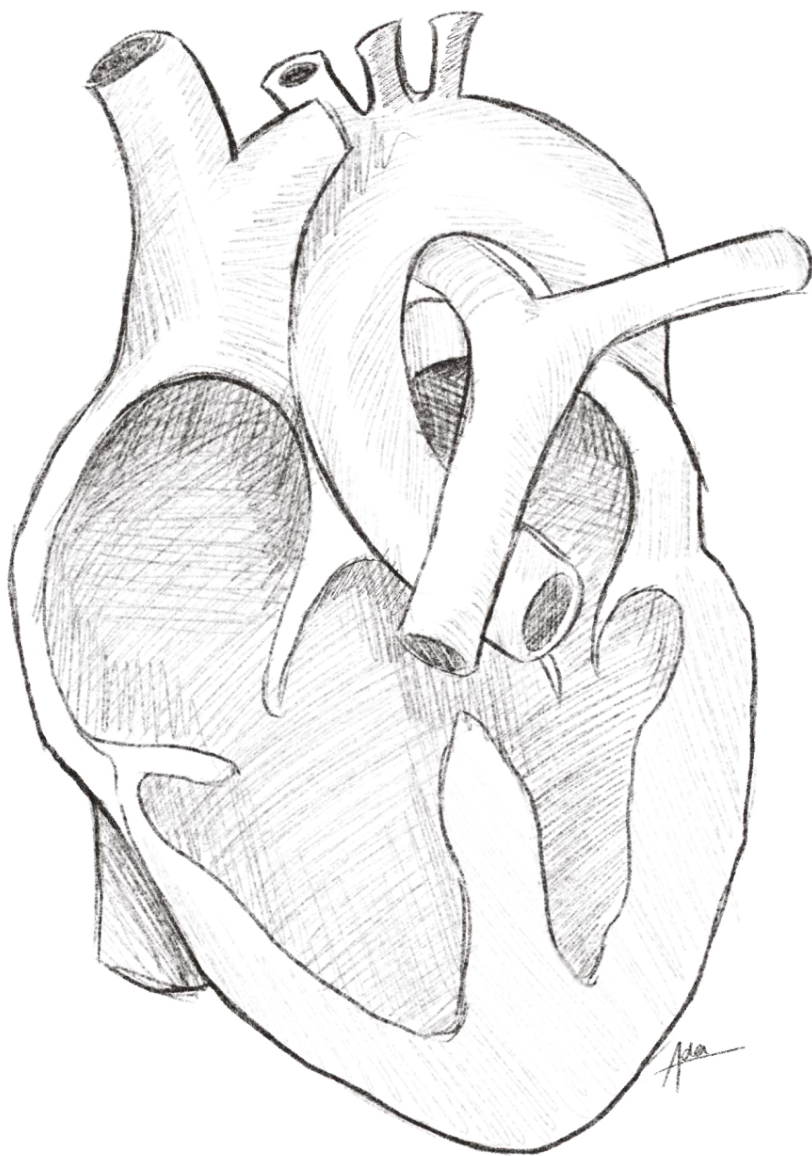
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