

Transition from Child Health Care to Adult Health Care for Adolescents with Congenital Heart Disease

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TRANSITION FROM CHILD HEALTHCARE TO ADULT HEALTHCARE FOR ADOLESCENTS WITH CONGENITAL HEART DISEASE

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Stockholm 2018

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Published by Karolinska Institutet.
Printed by E-Print AB 2018
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ISBN 978-91-7676-961-4



**Karolinska
Institutet**

Transition from child healthcare to adult healthcare for adolescents with congenital heart disease THESIS FOR DOCTORAL DEGREE (Ph.D.)

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Stockholm 2018

There is a need for preparation before the shift to adult health care

“So, you don’t just stand there, not knowing what to do...”

(quote from a 17-year-old-girl in study I

ABSTRACT

The life-prospects for adolescents with congenital heart disease (CHD) have improved, and today more than 90% survive to adulthood and need to continue their care into adulthood. During adolescence, the adolescents need to step by step take over the responsibility for their care from their parents. This shift in responsibility seems to be difficult for both the adolescent and the parents. Many adolescents have described themselves as ill prepared for the transfer, and it has also been shown that adolescents often lack knowledge about their heart condition and the need for medical follow-up. Further, the parents might need support in how to hand over this responsibility to their child.

The overall aim of this thesis was to examine the transition for adolescents with congenital heart disease and their parents prior to the transfer from pediatric health care to adult health care

The thesis consists of four studies. The participants were adolescents 14–18 years old and their parents. The adolescents were under follow-up care at one of four university hospitals in Sweden: Gothenburg, Lund, Stockholm, and Umeå.

The studies were based on individual interviews (I), focus group interviews (II), and questionnaires (III and IV).

Study I The aim was to identify and describe the needs of adolescents with CHD and their parents during the transition before transfer to adult cardiologic health care. The results revealed one theme for the adolescents, *Safety and control*, indicating a need for continuity and knowledge and a need of taking responsibility. The results also revealed one theme for the parents, *Safety and trust*, indicating a need for continuity and for shifting responsibility.

Study II The aim was to explore what adolescents with CHD view as important in the preparation for the transfer to adult care. The results revealed one main category, *Becoming a manager of the condition*, and the four subcategories of *Sufficient knowledge about the health*, *Be a participant in the care*, *Parental support*, and *Communicate with others about the health*.

Study III The aim was to (i) describe the level of readiness for transition in adolescents with CHD, (ii) to compare the level of readiness for transition as assessed by the adolescents with parental assessments, and (iii) to study potential correlates of transition readiness in adolescents with CHD. The results showed that the adolescents' level of readiness for transition increased with age. Further, there was a significant difference in adolescents' and their parents' scoring, where the adolescents scored higher on overall readiness compared to their parents. Additional, empowerment and illness perception (patient's overall perception about their disease) were associated with higher transition readiness.

Study IV The aims of the study were to (i) study parents' levels of uncertainty related to the transfer from pediatric to adult care in adolescents with CHD and (ii) to identify any factors that might correlate with parents' levels of uncertainty. The results showed that the mothers scored significantly higher on the uncertainty scale than the fathers and that there was a negative correlation between parental uncertainty and the adolescents' overall readiness.

SVENSK SAMMANFATTNING

Medfödda hjärtfel är den vanligaste missbildningen och den drabbar 0.8–1 % av barn som föds idag. Det är en stor variation på svårighetsgraderna som ofta delas in i enkla, medelsvåra och komplicerade hjärtfel. De senaste decennierna av medicinsk utveckling och förbättrat omhändertagande har inneburit att överlevnaden för denna patientgrupp har ökat avsevärt. Idag överlever fler än 90 % av barnen 18 års ålder. Även om överlevnaden har förbättrats så kvarstår många gånger behovet av fortsatta medicinska kontroller, medicinsk behandling och kirurgi i vuxen ålder. Detta innebär att ungdomen behöver fortsatt vård inom vuxensjukvården när hen fyller 18 år. Skiftet mellan barnsjukvård och vuxensjukvård kan vara problematisk för många ungdomar och deras föräldrar; många ungdomar och föräldrar upplever sig vara oförberedda inför överflyttningen. Tidigare studier visar även att ungdomar har bristande kunskap om sitt hjärtfel och behov av fortsatta medicinska kontroller, vilket kan medföra att dessa unga vuxna uteblir från planerade kontroller.

Under uppväxten behöver ungdomarna få möjlighet att lära sig om sitt hjärtfel och betydelsen av detta samt stegvis ta över ansvaret för sin hälsa från sina föräldrar. Många föräldrar uttrycker osäkerhet hur de ska stödja sitt barn under denna process. Likaså finns det undersökningar som beskriver att föräldrarna har svårt att lämna över och låta ungdomen ta ansvaret för sin hälsa.

Denna avhandling syftar till att studera transitionen inför överflyttning till vuxensjukvården för ungdomar med medfödda hjärtfel och deras föräldrar.

I avhandlingens fyra delstudier har ungdomar i åldern 14–18 år som har medfött hjärtfel och deras föräldrar vid fyra barnhjärtcentra i Sverige deltagit i intervjuer och/eller besvarat frågeformulär.

Syftet med **studie I** var att identifiera och beskriva vilka vårdbehov som ungdomar med medfödda hjärtfel och deras föräldrar har under transitionen inför överflyttning till vuxensjukvården. Individuella intervjuer med 13 ungdomar och 12 föräldrar genomfördes. Resultatet visade att ungdomarna hade behov av mer kunskap om sitt tillstånd. Det visade även på betydelsen av goda relationer med vårdgivaren och kontinuitet, men även på en medvetenhet om behovet att ta ett ökat ansvar om sin hälsa. Det framkom hur viktiga föräldrarna var under transitionen för att stödja ungdomen. Studien visade även att föräldrarna upplevde behov av eget stöd under transitionen, kring hur de på bästa sätt skulle kunna hjälpa sin ungdom att ta över ansvaret för sin hälsa.

I **studie II** genomfördes fyra fokusgruppsintervjuer med ungdomar vid fyra barnhjärtcentra i Sverige. Undersökningen syftade till att undersöka vad ungdomarna uppfattade vara viktigt i förberedelsen inför överflyttningen till vuxensjukvården. Resultatet av studien visade att ungdomarna önskade sig mer kunskap om hälsorelaterade frågor samt att vara involverade i sin vård. Det ansågs även angeläget att informationen som ges av vårdgivaren, måste vara saklig och tydlig. Ungdomarna ansåg att förberedelsen inför överflyttningen behövde påbörjas upp till tre år i förväg samt att det var viktigt att få träffa den nya vårdgivaren. Föräldrarna beskrevs vara viktiga under transitionen för att stödja ungdomarna vid kommunikation med vårdgivaren, men även för att tolka informationen vid läkarbesöket. Det visade sig att ungdomar kan behöva stöd när de talar med sin omgivning om sitt tillstånd.

I **studie III** besvarade 157 ungdomar och deras föräldrar från fyra barnhjärtcentra i Sverige ett antal frågeformulär. Syftet med denna studie var att (i) beskriva hur redo ungdomarna var att ta över ansvar för sin hälsa och vård, för överflyttning till vuxensjukvården samt föräldrarnas delaktighet i vården, (ii) jämföra ungdomars och föräldrars skattningar, samt (iii) studera faktorer som påverkar ungdomars uppfattning om att vara redo för överflyttning. Resultatet visade att med stigande ålder ökar ungdomens ansvar över sin hälsa och de upplever sig alltmer redo för överflyttning till vuxensjukvården samtidigt som föräldrarnas delaktighet minskar. Det sågs en skillnad mellan ungdomarnas och föräldrarnas skattning, ungdomar upplevde sig mer redo för överflyttning än deras föräldrar.

Syftet med **studie IV** var att (i) studera föräldrarnas upplevelse av osäkerhet inför ungdomens överflyttning till vuxensjukvården samt (ii) vilka faktorer som kan påverka detta. Föräldrar (351) vid fyra barnhjärtcentra deltog i denna enkätstudie. Resultatet visade att mammorna upplevde sig mer osäkra än papporna. En fjärdedel av föräldrarna uppgav ingen osäkerhet medan nästan var tionde förälder uppgav sig vara extremt osäker. Ungdomens ålder, kön eller svårighetsgrad hade ingen inverkan på föräldrarnas upplevelse av osäkerhet. Däremot minskade föräldrarnas osäkerhet när de upplevde att ungdomen var redo att föras över till vuxensjukvården. Undersökningen visade även att många föräldrar inte hade börjat tänka på den kommande överflyttningen.

Sammanfattningsvis visar resultaten att ungdomarna med medfödda hjärtfel och deras föräldrar upplever kontinuiteten och den goda relationen med sina vårdgivare som betydelsefull. Det finns ett behov hos ungdomarna av mer kunskap om det specifika hjärteftet och hälsorelaterade frågor, men även information om den nya vårdgivaren. Det framkom även att ungdomar upplever det svårt att kommunicera om sin sjukdom. Föga förvånande så upplever ungdomar sig mer redo för överflyttning med stigande ålder. Många föräldrar upplever behov av eget stöd om hur de bäst stödjer sin ungdom. Det sågs en stor variation i föräldrarnas osäkerhet inför överflyttningen; det är viktigt att ta reda på vilka föräldrar som upplever stor osäkerhet och ge dem adekvat stöd, då föräldrarnas osäkerhet kan ha en negativ effekt på ungdomens utveckling av självständighet. Föräldrarna spelar en nyckelroll för ungdomen i att kunna utveckla självständighet, egenvård, att kommunicera med vårdgivaren samt förberedelse inför överflyttningen. Resultatet visar att det finns utrymme och stora möjligheter för utveckling av förberedelserna inför överflyttningen till vuxensjukvården.

LIST OF SCIENTIFIC PAPERS

- I. **Adolescents with Congenital Heart Disease and Their Parents Needs Before Transfer to Adult Care**
Åsa Burström, Maria Öjmyr-Joelsson, Ewa-Lena Bratt, Bo Lundell, Margret Nisell
Journal of Cardiovascular Nursing, 2016, Vol 31, sid. 399-404.
- II. **Adolescents with congenital heart disease: their opinions about the preparation for transfer to adult care**
Åsa Burström, Ewa-Lena Bratt, Björn Frenckner, Margret Nisell, Katarina Hanséus, Annika Rydberg, Maria Öjmyr-Joelsson
European Journal of Pediatrics, 2017, May 16.
- III. **Ready for transfer to adult care?**
 - **A triadic evaluation of transition readiness in adolescents with congenital heart disease and their parents**
Å. Burström, M. Acuna Mora, M. Öjmyr Joelsson, C. Sparud-Lundin, A. Rydberg, K. Hanseus, B. Frenckner, M. Nisell, P. Moons, E-L. Bratt
In Manuscript
- IV. **Parents' self-reported experiences of uncertainty related to transfer from pediatrics to adult care in adolescents with congenital heart disease**
Å. Burström, M. Acuna Mora, M. Öjmyr Joelsson, C. Sparud-Lundin, A. Rydberg, K. Hanseus, B. Frenckner, M. Nisell, P. Moons, E-L. Bratt
In Manuscript

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LIST OF ABBREVIATIONS

CHD	Congenital Heart Disease
ACHD	Adult Congenital Heart Disease
GUCH	Grown-Up Congenital Heart Disease
HCP	Health Care Professionals
RTQ	Readiness for Transition Questionnaire
LAS	Linear Analogue Scale
HBS-CHD	Health Behavior Scale-Congenital Heart Disease
QoL	Quality of Life
HRQoL	Health Related Quality of Life
GYPES	The Gothenburg Young Persons Empowerment Scale
B-IPQ	Brief-Illness Perception Questionnaire
KnoCoHM	Knowledge Scale for adults with Congenitally Malformed Hearts

LIST OF DEFINITIONS

CHD	“A gross structural abnormality of the heart or the intrathoracic great vessels that is actually or potentially of functional significance” (Warnes et al. 2001)
Transition as a health care intervention	“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” (Knauth et al. 2006)
Transfer	“An event or series of events through which adolescents and young adults with chronic physical and medical conditions move their care from a pediatric to an adult health care environment” (Knauth et al. 2006)

1 INTRODUCTION

Congenital heart disease (CHD) is the most common congenital malformation. Most CHD are detected prenatally or during the first year of life. CHD has different levels of severity and different impacts on health and life. It varies from minor defects that can self-heal to complex defects that need several surgeries and interventions and that have an extensive impact on the patient's life. The intervals for medical check-ups are determined by the severity of the disease.

Throughout childhood, the parents are the advocate for the child and are responsible for the child's health and healthcare, but during adolescence the child needs to be more involved in order to be able to learn to take over full responsibility for their own health and care when they reach adulthood. In my work as a nurse at a pediatric outpatient clinic for patients with CHD, I have often heard parents' concerns about the transfer to adult care.

How to facilitate adolescence and their parents in the transfer to adult care has captured my interest. Today there is no structured way to prepare adolescents for this transfer of care, and there is no evidence for how to best perform the transition for the adolescent. Research is needed to explore the adolescents' and their parents' needs and how they perceive the transition process. Furthermore, research is needed to explore the degree of transition readiness and factors that are associated with readiness in order to facilitate the adolescent during the transfer of care.

The research projects included in this thesis are part of the STEPSTONES project (Swedish Transition Effects Project Supporting Teenagers with chrONic mEdical conditionS). The project involves the Pediatric Cardiology Centers at Astrid Lindgren Children's Hospital, Karolinska University Hospital, The Queen Silvia Children's Hospital in Gothenburg, Skåne University Hospital in Lund, and the University Hospital in Umeå.

2 BACKGROUND

2.1 CONGENITAL HEART DISEASE

A congenital heart disease (CHD) has a prevalence of 0.8–1.1 cases/1000 live births [1-3]. CHD is defined as “*a gross structural abnormality of the heart or the intrathoracic great vessels that is actually or potentially of functional significance*” [4]. Depending on the complexity of the defect, it is classified into simple, moderate, or severe CHD as defined by the 32nd Bethesda conference [4]. Developments in cardiac surgery, medical interventions, pharmacological treatments, and general care have led to improved survival rates. Up to 90 percent of all adolescents born with CHD now survive to adulthood [5], and this has resulted in a growing population of adults with CHD [6]. Today there are more adults living with CHD than children [7]. Despite such progress in medical treatments in infancy and childhood, many patients with repaired CHD have health issues that need further treatment during adolescence and adulthood [8]. This means that when the adolescents reach adulthood, many need to be transferred from pediatric health care to adult care for adults with CHD. The intervals between medical follow-ups depend on the severity of the CHD and the impact of the condition and range from approximately every 6 months to every 5 years [8-10].

The transfer to adult care is normally planned at age 15–18 years [11]. In Sweden, the health-care system requires the transfer to adult healthcare at the age of 18 at the latest. This transfer means that the adolescents will move from a pediatric cardiology healthcare setting to an adult healthcare setting for adults with CHD, Grown Ups with Congenital Heart disease (GUCH), or to a non-specialized setting with a physician who has knowledge in the specialty.

2.2 THEORETICAL FRAMEWORK

This thesis is based on the transition theory described by Meleis et al. [12]. In this theory, transitions are the normal changes and processes that everyone will experience over the course of their life. For an individual, different changes in health status might result in improved health and well-being or expose the patient to risks for illness, and this can start up a process of change and transition. These transitions are experienced individually, and persons who undergo and experience a transition are often vulnerable and might experience stress that can affect their health [12, 13]. By identifying and understanding the meanings and conditions of the transition, the health care professionals (HCPs) can facilitate and reduce barriers, which might result in an improved care for the patient and family and ensure healthier transitions [12].

Many times, an individual experiences multiple transitions at the same time [12]. This is true in this thesis as well, and the adolescents in these studies experienced different transitions that were many times concurrent with each other. These include *Developmental transitions* in which the adolescents experience natural developmental milestones, e.g. leaving childhood and entering adolescence, *Situational transitions* such as moving from one grade to the next or changing schools, *Health/illness transitions* in which some of the adolescents might have a new diagnosis or require medical intervention, and *Organizational transitions* in which the adolescent changes healthcare setting and acquires new caregivers [12].

To understand the patients' experiences during the different transitions, it is necessary to identify personal and environmental conditions that can be barriers or that can facilitate a healthy transition or outcome [12]. Three conditions are described– *personal conditions*, meaning the need for preparation and knowledge about the change, *community conditions*, including family support and relevant information from trusted HCPs, and finally *societal conditions*, such as when the individual experiences the transition as stigmatizing [12].

A successful transition is characterized by both process and outcome indicators. Because a transition is a process that develops over time, it allows HCPs to identify the process indicators (e.g. being involved in the care and coping) that can lead the patient towards health or towards vulnerability. The outcome indicators in the transition theory are mastery of new skills and fluid integration [12].

In this thesis, “transition of care” is defined as: “*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*” Knauth et al. [14]. The “transfer of care” is defined as: “*An event or series of events through which adolescents and young adults with chronic physical and medical conditions move their care from a pediatric to an adult health care environment*” Knauth et al. [14].

According to Rosen et al., this transition in healthcare should be “a purposeful, planned process that addresses the medical, psychosocial and educational/vocational needs of adolescents and young adults with chronic physical and medical conditions as they move from child-centered to adult-oriented health care systems” [15].

2.3 PREVIOUS RESEARCH ON TRANSITION AND TRANSFER

2.3.1 Adolescence- The Developmental Transition

Concurrent with the healthcare transition prior to the healthcare transfer, adolescents experience several biological and psychosocial developments. Puberty starts with changes in the whole body, including body height, body composition, hormonal changes, and metabolic changes. In addition, their social maturity develops and continues through young adulthood. The maturity process often includes intense emotions, sensation-seeking behavior, and risk taking as well as the desire not to be different from others [16]. During the developmental transition from early adolescence to late adolescence, the adolescents go from concrete thinking towards abstract thinking and develop a sense of social autonomy [17]. Friends come to play a more important role during adolescence, although the parents remain essential for support [18]. It is a time when adolescents strive to achieve independence and to separate from their parents [19]. This process of developing autonomy is important for the adolescents to achieve self-efficacy and independence [20]. For an adolescent with a chronic disease, the normal development toward autonomy might be negatively affected [16, 21, 22].

Adolescence is a time to explore and to take risks to a certain extent, which is a natural process [16]. However, for an adolescent with CHD it is vital to have a good understanding of health-promoting behavior and what can be harmful for their health [8]. This is information that is important for HCPs to discuss with the adolescents during the transition to adult care [23].

2.3.2 Parenting an adolescent during the transition to adulthood and transfer to adult care

The healthcare transition for adolescents with chronic conditions coincides with processes of becoming independent from one's parents [24], and these adolescents need to gradually increase their responsibility over their health situation and over the management of their own healthcare. They are dependent on their parents' support when taking over the responsibility for their healthcare [24], but the child should be involved early on in their healthcare together with their parents and HCPs in order to help them take on such responsibilities as efficiently as possible [25]. Coyne et al. argue for the importance of giving the child opportunities to participate in shared decision-making so that they can practice self-advocacy and self-management. This is important for building up the competences needed to make their own decisions when they turn 18 [25]. As the adolescent is practicing for taking over the

responsibility for their own healthcare, the parents need to change their role from a manager to a consultant and advocate in order to facilitate their child's transition from dependence to independence. The HCPs need to facilitate the parents and the adolescents during this process in order to promote the adolescent's autonomy and to provide expertise when needed [26, 27]. The parenting style during the independence process can affect the adolescent's perceived health. In a study from Belgium, one third of the adolescents with CHD reported overprotective parents, which might affect the adolescents' perceptions of their health [28]. It has been shown that democratic parenting (high on regulation and responsiveness, low on psychological control) has the best outcome in perceived health and quality of life for adolescents with CHD [29].

In the pediatric healthcare setting, the parents of children with chronic conditions are essential for their child's contact with HCPs. When adolescents are transferred to adult healthcare, they need to be responsible for their own healthcare. Therefore, it is essential that the adolescent has developed sufficient self-management skills before the transfer to adult healthcare [30]. However, there is a delicate balance between promoting independence and autonomy and the need for parental support in preventing complications in adolescents with special healthcare needs [31, 32]. It has been described that parents of adolescents with diabetes report that supporting independence might not mean withdrawal of all the support. Many adolescents need support in decision making in further health care management [33].

The healthcare transfer should be preceded by a transition process in which the adolescent and their family are prepared for the event in order to increase the adolescent's self-management [8, 15, 34]. However, many parents express uncertainty about how to support their adolescents during this transition in an appropriate way [35]. Mishel et al. [36] have defined uncertainty as the inability to determine the meaning of events, and uncertainty occurs in a situation where the decision-maker is unable to assign definite values to objects and events and/or is unable to accurately predict the outcomes of the events. This uncertainty should be distinguished from parental stress because uncertainty is due to environmental events [37], while parental stress is a psychological distress that parents might experience while meeting the demands of their parental role [38].

2.3.3 Getting ready for the transfer

Healthcare transition is "more than crossing a road" [39]. It is a time of preparing and of learning about one's health condition and maturing before taking over the responsibility of one's health from one's parents and transferring to adult healthcare [39]. For adolescents with

CHD, the healthcare transition means that they need to be prepared to leave pediatric healthcare and enter adult healthcare [40]. Bridgett et al. argue for the importance of holistic person-centered care in order to facilitate adolescents during this transition. It is important for the adolescents to be able to discuss the challenges that they meet in their life as they work to develop self-management [41]. A transition can be considered successful for an individual when he or she is adjusted to the new situation and demonstrates the skills and behaviors needed to manage the new situation [12].

The concern about preparation prior to the transfer of care for adolescents with chronic diseases is not a new phenomenon, and it is an area of growing interest among HCPs in many different disciplines [15, 20, 29, 34, 42, 43]. Still, it is an important area that has to be improved. One way is to use a formal transition program. According to the American Heart Association [8] a formal transition program should be person-centered, age and developmentally appropriate, flexible, and include comprehensible healthcare. The program should include education about medical conditions that is age appropriate. Further, it should promote skills in communication, decision making, self-care, and self-advocacy. The program should foster a sense of control over health, healthcare decisions, and psychosocial environment and should promote increased personal and medical independence [8]. The goals are similar to the transition outcomes described by Fair et al [44], and these transition outcomes include understanding the characteristics of the conditions, understanding health insurance, attending medical appointments, having a medical home, avoiding unnecessary hospitalization, having a social network, and having satisfactory quality of life (QoL). Parents of a child with a chronic condition face the challenge of handing over the responsibility for the child's health management to their child, which is important for developing healthcare autonomy in the child. According to Beacham et al.[33], the development of autonomy in children is central for the development of self-care management. Healthcare autonomy means that the patient is able to evaluate options, make decisions, define health-related goals, have the confidence to stand by their decisions, and develop strategies to meet their health-related goals. Healthcare autonomy is usually one of the last contexts in which autonomy is expressed, and it typically develops in late adolescence [33].

To be able to fulfill the goals of a successful transition and transfer to adult care [8, 15, 34], the transition characteristic during the process should be evaluated in order to determine if

intervention is necessary. It is acknowledged that adolescents with low transition readiness are at risk of significant negative outcomes [45, 46].

Over the last years, several instruments, including both generic [47-49] and disease-specific transition instruments [32], have been developed in order to assess adolescents' readiness for transition. Different chronic diseases have different demands, and the instruments contain items covering self-management and self-advocacy [47] and knowledge about the condition, medication, adherence, nutrition, self-management, informed reproduction, school, insurance, ongoing support, and new providers [48]. Some instruments come with a parental proxy versions [31, 32]. Gilleland and co-workers [31] address the parents' involvement in the adolescents' healthcare as a factor in avoiding non-compliance in medication and healthcare and as having an impact on the transfer of responsibility to the adolescent.

2.4 RATIONAL FOR THE STUDY

Developments within medicine and caring science have created new skills and knowledge and have increased the life expectancies of children and adolescents with CHD, and these adolescents need to be prepared to take over the responsibility for their life and health as they transition into adulthood. During the same period in life, the parents of these children need to shift their role of being the one with full control and to hand over responsibility to their child and to take on a more supportive role.

This progress in care and treatment strategies necessitates knowledge of how to meet and support adolescents and parents during the transition to adulthood and the transfer to adult care.

Today, transfer from pediatric care to adult care often occurs without any structure or plan and is not always straightforward. Adolescents are thus often unprepared to take over responsibility for their own care and have insufficient knowledge about their disease, its treatment, and the importance of uninterrupted medical follow up. To improve the transfer in care and to develop and implement effective transition programs, more knowledge about adolescents' and parents' experiences and needs is crucial.

3 AIM

3.1 OVERALL AIM

The overall aim of this thesis was to examine the transition for adolescents with congenital heart disease CHD and their parents prior to the transfer from pediatric health care to adult health care.

3.2 SPECIFIC AIMS

Study I was to identify and describe the needs of adolescents with CHD and their parents during the transition before transfer to adult cardiologic health care.

Study II The aim was to explore what adolescents with CHD view as important in the preparation for transfer to adult care.

Study III The aim was to (i) describe the level of readiness for transition in adolescents with CHD, (ii) to compare the level of readiness for transition as assessed by the adolescents with parental assessments, and (iii) to study potential correlates of transition readiness in adolescents with CHD.

Study IV The aim was to (i) study parent's levels of uncertainty related to the transfer from pediatric to adult care in adolescents with CHD and (ii) to identify any factors that might correlate with parents' level of uncertainty.

4 MATERIALS AND METHODS

4.1 DESIGN

This thesis consists of four studies (Table 1). The study designs were explorative and descriptive, and both qualitative and quantitative methods were used to fulfill the aim of the thesis.

Table 1. Overview of the designs for the studies in the thesis.

	Study I	Study II	Study III	Study IV
Focus	To identify and describe the needs during transition before the transfer to adult care in adolescents with CHD and their parents	To explore what the adolescents view as important in the preparation for the transfer to adult care	To describe the readiness for transition to adult care in adolescents with CHD	To study parent's level of uncertainty related to transfer from pediatric to adult health care
Design	Explorative Descriptive	Explorative Descriptive	Cross-sectional Descriptive	Cross-sectional Descriptive
Data collection	Individual interviews	Focus group interviews	Questionnaires	Questionnaires
Sample	Adolescents 16–18 years old and their parents	Adolescents 14–18 years old with CHD	Adolescents 14–18 years old and their parents	Parents of adolescents with CHD
Data analysis	Content analysis	Content analysis	Statistical analysis	Statistical analysis

4.2 SETTINGS

Study I was a single-center study performed in the pediatric outpatient clinic at Astrid Lindgren Children's Hospital. In Studies II–IV, the participants were recruited from pediatric outpatient clinics at four university hospitals (Gothenburg, Lund, Stockholm, and Umeå) in Sweden.

4.3 PARTICIPANTS

The participants in this thesis were adolescents with CHD under medical follow up in one of the participant centers and their parents. In Study I, the participants were recruited in one outpatient clinic. The participants in Study II were recruited from one of the four university hospitals in Sweden. The participants in the cross-sectional studies (III and IV) were identified via SWEDCON [50] (Table 2).

Table 2 Demographic data for the participants in the thesis.

	Study I	Study II	Study III	Study IV
Adolescents, n	13	17	157	n.a
Age, mean (\pmSD)		15.8 (1.6)	15.7 (1.1)	n.a
<i>Range</i>	16–18 years	14–18 years	14–18 years	n.a
Sex, n				
<i>Female</i>	7	10	72	n.a
<i>Male</i>	6	7	85	n.a
Complexity of the CHD				
<i>Mild</i>			46	n.a
<i>Moderate</i>	8	2	70	n.a
<i>Severe</i>	5	15	41	n.a
Parents, n				
<i>Mothers</i>	7	n.a	157	189
<i>Fathers</i>	5	n.a	157	162
Age, mean (\pmSD)				
<i>Mothers</i>	n.a	n.a	46.9 (4.9)	46.9 (5.3)
<i>Fathers</i>	n.a	n.a	49.3 (5.5)	49.3 (5.5)

n.a - not applicable

4.3.1 Study I

Inclusion criteria for this study were adolescents aged 16–18 years, born with a moderate to severe CHD, and fluent in Swedish. Additionally, one parent was invited to participate in an interview.

The recruitment of the participants was performed with the assistance of a specialist nurse at the outpatient clinic who identified 20 adolescents fulfilling the inclusion criteria. First the families were approached with oral information about the study. If the adolescent was interested in additional information about the study, they received written information along with the consent form and a pre-stamped envelope. One participant was excluded due to having recent surgery. In total, 13 adolescents and 12 parents participated. The interviews were performed at the hospital except for two, which were performed at the parent's place of work.

4.3.2 Study II

The participants were recruited from four hospitals. The inclusion criteria were adolescents aged 14–18 years with a moderate or complex CHD and who were Swedish speaking and able to participate in group discussions. Purposive sampling was used, and recruitment to the study was made by a nurse or physician at each respective center. They were asked to select 6–8 adolescents with equal gender and age distributions who fulfilled the inclusion criteria. The possible informants received an information letter about the study along with a consent form. When consent was received from the adolescents and parents, the adolescents were invited to participate in the focus group interview. The adolescents were informed that the interview was voluntary and that we were not necessarily going to talk about their personal situation, but rather their opinion about the transition to adult care.

The interviews were performed in a conference room at each hospital.

The intention was to have 6–8 participants in each group; however, due to late dropouts there were only 3–6 adolescents in the four groups.

4.3.3 Study III

Study III was a cross-sectional study including adolescents and their parents. Data from the Swedish registry of congenital heart disease (SWEDCON) [50] were used to identify eligible patients. Inclusion criteria for the study were adolescents aged 14–18 years with CHD and in need of medical follow-up in adulthood. The participating adolescents were under active

follow-up at one of four university hospitals in Sweden. Exclusion criteria were syndromes affecting cognitive abilities, heart transplantation, acquired heart diseases, illiteracy, or being non-Swedish speaking.

4.3.4 Study IV

Data from the Swedish registry of congenital heart disease (SWEDCON) [50] were used to identify eligible patients. The inclusion criteria were being a parent to an adolescent aged 14–18 years who was born with CHD and who was under active follow-up at one of the four participating university hospitals in Sweden. Parents were excluded if their child had CHD associated with syndromes that affected cognitive abilities, had undergone a heart transplant, or had a non-structural heart disease or if the parents were illiterate or non-Swedish speaking. In total, 1,186 eligible parents of 593 adolescents fulfilled the inclusion criteria.

4.4 DATA COLLECTION

4.4.1 Study I

Data were collected through semi-structured individual interviews with adolescents and their parents. Individual interviews are an often-used method in qualitative research for collecting data. The method was chosen in order to be able to explore the participants' perspectives [51]. The success in acquiring rich data depends upon the interviewer's skills and the interviewee's willingness to share their opinions and experiences and how deeply they engage in the interview process [52]. It has previously been reported that adolescents want to be involved in their own healthcare [53], which makes it important to ask them about their needs during the transition to adult care.

An interview guide was constructed by the research team to support the researcher in exploring and illuminating the issues of interest [51, 54]. All interviews were performed at the hospital except for two on request from a parent and an adolescent.

4.4.2 Study II

Data were collected with semi-structured focus group interviews with adolescents at four university hospitals.

Focus group interviews are a method used to explore the range of opinions [55, 56] and are useful when the subject can best be understood through discussions [57]. The participants do not just respond to the questions from the researcher, but also based on the answers given by

others in the group [56]. In the focus group interviews in this study, the adolescents were asked to discuss *what* was important in the preparation for transfer, *how* the preparation should be performed, *when* the preparation should start, and *when* the transfer should take place.

An interview guide was developed by the research team to be a support to the interviewer during the interviews. The interviews lasted 50–65 minutes. Sociodemographic characteristics were collected from the patients' medical records.

4.4.3 Study III

To fulfill the aim of Study III, we used one questionnaire to assess readiness for transition and additional questionnaires to study correlates that have been described to be important during the transition in order to identify any factors that might have an impact on transition readiness [8].

Data were collected between January 25, 2016, and August 3, 2016. The questionnaires were sent to all 593 eligible adolescents and 1,186 parents. Study information and the informed consent form were sent together with the questionnaires along with a pre-addressed envelope. To increase the number of responders, a special structured approach was used [58]. If no response was received after three weeks, a reminder letter was sent. The non-responders received a new set of questionnaires if still no response was received after five weeks. Finally, after seven weeks the non-responders were contacted by telephone and were asked if they had received the questionnaires, if their address was correct, and if they had an interest in participating in the study.

The study information, consent form, and questionnaires were distributed by a company with experience in administrating research projects.

4.4.3.1 Questionnaires measuring readiness for transition

Readiness for transition was evaluated using the *Readiness for Transition Questionnaire (RTQ)* developed by Gilleland et al [31]. They acknowledged the transition to adult care both as a process as well as an event, and transition readiness was defined as “the adolescents’ readiness to assume complete responsibility for their healthcare and their readiness to transfer to adult medical care.” The instrument was originally created to be used among adolescents with kidney transplants and to assess adolescents’ perception about their readiness for transition and transfer to adult care [31], but has been used in other chronic diagnosis [59].

The questionnaire was found to also be appropriate for this study population, and it had the advantage that it came with a proxy version to be answered by the parents.

The RTQ was translated into Swedish by researchers in the STEPSTONES project at Gothenburg University using standardized principles [60] and included forward and backward translation in a 2-stage procedure. Four Swedish-speaking translators, and who were researchers with knowledge in the field, performed the forward translation independently [60]. Discrepancies were resolved between their versions. Furthermore, a backward translation into English was performed by an authorized translator and native speaker of English, and further discrepancies were resolved [60]. One item related to healthcare insurance was removed because the Swedish healthcare system covers all persons independent of their age. An additional response option of “not relevant” was added to the items related to adolescents’ responsibility and parental involvement after the face validity was evaluated. This option was included because there were items related to medication and frequent contact with HCPs that are not relevant for all patients with CHD.

The modified version of the questionnaire consists of three domains – *adolescents’ health behavior and responsibility*, *parental involvement*, and *adolescents’ transition readiness*. The adolescents’ responsibility domain contains nine items covering aspects related to taking blood samples, managing medication, renewing prescriptions, communicating about the disease, communicating with HCPs, and attending medical appointments. The parental involvement domain consists of the same nine items. The adolescents’ responsibility and parental involvement domains are rated on a 4-point Likert scale (not at all responsible/involved; somewhat responsible/involved; mostly responsible/involved; completely responsible/involved), and the additional option “not applicable”. A total mean score for adolescents’ responsibility and parental involvement was calculated if there were no more than four items missing per individual in each domain. The additional option “not applicable” for each of the 18 items was counted as a missing value.

The total overall transition readiness includes two items – “Overall, how ready do you think you are to assume complete responsibility for your healthcare?” and “Overall, how ready do you think you are to be transferred from pediatric health care to adult health care?” The scores are based on a 4-point Likert scale (not at all ready; somewhat ready; mostly ready; completely ready). Internal consistency was confirmed in adolescents with kidney transplants, with Cronbach α values >0.7 . [31].

4.4.3.2 Additional questionnaires and measurements

The *Knowledge Scale for Adults with Heart Malformations (KnoCoHM)* was used to assess disease-specific knowledge. The questionnaire was developed to measure young adults' knowledge about their heart defect and heart-related condition, their medication, and possible complications and implications due to the heart condition [61]. Before using the questionnaire in the present study, the questionnaire was discussed in the research group and with medical experts to determine the relevance of the instrument. After face validation, some minor changes were made regarding wording because some expressions were adult oriented. Further items related to endocarditis were removed because the medical recommendations had changed [62].

The questionnaire includes 19 items with four domains – *general knowledge, medical treatment, endocarditis prophylaxis, and pregnancies and contraceptives*. For this study, only the domain of general knowledge was used to calculate a knowledge score. This domain includes a total of 11 items regarding knowledge about the condition, treatment, endocarditis, physical activity, and heredity. A higher score (ranging from 0 to 20) reflects a higher level of knowledge.

The *Health Behavior Scale–Congenital Heart Disease (HBS-CHD)* is a self-rating questionnaire developed and used for assessing health behavior among adolescents with CHD. The questionnaire contains 15 items about physical activity, drugs, alcohol, cigarettes, and endocarditis [63]. A total score is calculated (score 0-100), and higher scores denote worse health behavior [63].

Quality of life (QoL) was measured using a vertical linear analogue scale (LAS) with a range from 1 to 100. QoL is defined as “the degree of overall life satisfaction that is positively or negatively influenced by individuals' perception of certain aspects of life important to them, including matters both related and unrelated to health” [64, 65]. The participants were asked to specify on the scale how they perceive their current QoL. The endpoints were 0 = “worst imaginable QoL” and 100 = “best imaginable QoL”. Higher scores imply a better perceived QoL [66]. The LAS scale measuring QoL was used in a study on adults with CHD and was described as easy to use [64, 66].

The *Pediatric Quality of Life inventory 4.0 (PedsQL 4.0)* [67] is a questionnaire that provides information on patients' reported health-related quality of life (HRQoL). It is a generic module of the PedsQL and contains 23 items in four domains – physical functioning, emotional functioning, social functioning, and school functioning – that are measured on a

five-point Likert scale (never, almost never, sometimes, often, and almost always). Higher total scores indicate greater HRQoL.

The *Gothenburg Young Persons Empowerment Scale-Chronic Heart Disease (GYPES-CHD)* is an empowerment scale developed by Acuna et al. (Acuna Mora et al., under review). Empowerment is defined as “an enabling process or outcome that arises from communication with healthcare professionals and from mutual sharing of resources and information related to an illness that enhances the patient’s feeling of control, self-efficacy, coping abilities, and abilities to achieve changes in their condition.” The instrument consists of five dimensions of empowerment – knowledge and understanding, personal control, identity, shared decision-making, and enabling others. Further, it has 15 items with total scores ranging between 15 and 75 points with higher scores indicating a higher level of empowerment (Acuna Mora et al., under review).

The *Brief-Illness Perception Questionnaire (B-IPQ)* [68, 69] was used to measure illness perception. The B-IPQ is a nine-item questionnaire covering cognitive and emotional illness perceptions, including timeline, personal control, treatment control, identity of disease, illness coherence, emotional response, and concern. A total score is calculated on eight items, each with a score of 0–10 points. The last item in the instrument is an open-ended question about the causal factors for the illness, and a score for this cannot be calculated. The total score is 0–80 points, and a higher total score represents a more threatening view of the disease.

4.4.4 Study IV

The participants in Study IV were parents of adolescents with CHD aged 14–18 years. To fulfill the aim in study IV, which was to study the uncertainty among the parents in relation to the coming transfer of care for their child, we used an instrument developed specifically for the study. The Parental Uncertainty Scale was sent to the parents, along with the questionnaires to the adolescents in Study III. Sociodemographic data were collected along with the questionnaire, and information about the adolescents’ age, sex, and disease complexity was collected from the medical records.

4.4.4.1 The Parental Uncertainty Scale

The Parental Uncertainty Scale is a linear analogue scale (LAS) with a vertically oriented line ranging from 0 (Not at all uncertain) to 100 (Extremely uncertain).

Face validity was tested on four parents of children with CHD in two hospitals. The parents understood the wording, but a few suggestions for changes were made. After this, the scale was changed from 0–10 to 0–100 because the original scale was considered too narrow and changing the wording from “parents” to “you as a parent”.

4.4.4.2 Additional questionnaire

RTQ was used for assessing Adolescents overall readiness [31]. As described in a previous paragraph, the transition readiness questionnaire contains three domains, adolescents’ responsibility, Parental involvement and Overall transition readiness. In study IV it was only the Overall readiness mean score that was calculated.

4.5 DATA ANALYSIS

Table 3. Data analysis at a glance.

Data analysis	Study I	Study II	Study III	Study IV
Content analysis	X	X		
Descriptive statistics			X	X
T-test			X	
Mann–Whitney U-test			X	X
Chi-square			X	X
Wilcoxon signed-rank test			X	X
Univariate regression analysis			X	X
Multivariate regression analysis			X	
Intra class correlation (ICC)				X

4.5.1 Study I

The individual interviews (Study I) were transcribed verbatim and analyzed using qualitative content analysis with an inductive and manifest approach in order to provide knowledge and understanding of the phenomenon and to describe the participants' experiences [70, 71]. The analysis started with reading and reviewing the texts to get a sense of the whole material [70, 72]. The text was sorted into meaning units that were condensed and coded, and the codes were sorted into subcategories and categories [72]. To express the underlying meaning of the categories, two themes were formulated, one for the adolescents and one for the parents. For illustrating the categories, quotations from the interviews were presented. Qualitative content analysis is a method that describes differences and similarities in the data and can also be used for understanding the meaning of the communication [73]. To achieve trustworthiness and credibility, the analysis was performed together with the other co-authors. When there were differences in opinions about the categories, we went back to the text in order to find agreement in the way the data were labeled and categorized [72].

4.5.2 Study II

The focus group interviews were transcribed verbatim and analyzed using qualitative content analysis with an inductive and manifest approach [70, 71]. The analysis started with reading and reviewing the texts to get a sense of the whole material [70, 72]. An open coding was performed, and notes were written in the text. The codes were compared to find similarities and differences. Comparisons were made between the groups and within the groups [56, 73], and the analysis went back and forth between the text and the codes [72]. The codes were grouped and categories were created, and the categories were then included in a main category that described the categories' meaning. Additionally, quotations from the dialogues in the different groups were used to illustrate the categories [56]. The analysis was performed together with the other co-authors [72].

4.5.3 Study III.

The descriptive statistics were expressed in absolute numbers and percentages and as means and standard deviations. When comparing the study group and non-responders regarding age the t-test was used. Non-parametric analysis was used as appropriate for analyzing not normally-distributed data. Between-group comparisons were conducted using the Mann–Whitney U-test. For comparison between more than two groups, the Kruskal-Wallis test was used. The Wilcoxon signed-rank test was used for comparing dyads (paired tests). To explore correlates of adolescents' perceived overall readiness, adolescents' readiness, and parental

involvement, univariate linear regression analyses were performed. The multivariate model (enter method) was based on univariate variables with $p < 0.1$. The assumptions of linearity of residuals, absence of multicollinearity, and independence of observations were met by assessing Durbin–Watson statistics, the variance inflation factor, and normal probability plots (P-P plots). All tests were two-sided, and the level of significance was established at $p < 0.05$. To estimate the effect size, Cohen’s d was calculated (small effect size=between 0.10 and 0.30; medium effect size=0.30 and 0.50 and large effect size= 0.50 or higher) [74].

IBM SPSS statistics for Windows version 24 (Armonk, NY: IBM Corp.) was used to perform the statistical analysis.

4.5.4 Study IV

The descriptive statistics were expressed in numbers and percentages or as means and standard deviations. Non-parametric analysis was used as appropriate according to the distribution of the data. Between-group comparisons were conducted using Mann–Whitney U-tests. The Wilcoxon signed-rank test was used when comparing between dyads (paired). The chi-square test was used on a nominal level. Cohen’s d was calculated for testing the effect size of significant values of significant values [74]. An intra-class correlation (ICC, two-way random, absolute agreement) was calculated in order to analyze the agreement in perceived uncertainty by the parents (dyads) [75]. To explore potential correlates of perceived uncertainty, univariate linear regression analysis was performed. The assumptions of linearity of residuals and independence of observations were confirmed by assessing Durbin–Watson statistics and normal probability plots (P-P plots). The parental perceived overall readiness mean score [31] was calculated on two items (19 and 20) of the RTQ. The level of significance was established at $p < 0.05$.

IBM SPSS statistics for Windows version 24 (Armonk, NY: IBM Corp.) was used to perform the statistical analysis.

4.6 ETHICAL CONSIDERATIONS

All studies were performed according to Helsinki declaration [76]

The study (I) was approved by the regional ethics review board in Stockholm, Dnr: 2012/1803-31/4 and (study II-III) the regional ethics review board in Gothenburg, Dnr: 953-13. Research using children as subjects can be ethically problematic because their ability to judge risk and to assess consequences is limited [77]. They are considered to be a vulnerable group, and thus extra sensitivity is required. Before performing research on adolescents, the researcher need to be knowledgeable about research ethics and sensitive to potential risk and benefits [78]. The study information given must be appropriate for their age so that they understand the purpose of the study and their rights to decide if they want to participate in the studies [79, 80].

Before the interview studies the adolescents and the parents were informed both verbally and in writing about the study and were asked for consent. In study III and IV only written information was provided along the consent. The participants were informed that participation was voluntary and that they had the right to cease participation at any time.

5 FINDINGS

5.1 STUDY I

In all, 13 adolescents and 12 parents participated in individual interviews. The results from the interviews with the adolescents resulted in the theme *Safety and Control*. The theme depicted the meaning of three categories – *Change of relationship*, *Knowledge and information*, and *Daily living*.

The themes for the parents were *Safety and Trust*. The theme depicted the meaning of two categories – *Change of relationship* and *Daily living*.

5.1.1 Change of relationship-adolescents

The coming transfer to adult care was considered to be a natural step for the adolescents, and it came with a change in the relationship to the parents and to the HCPs. Although the adolescents started to take more responsibility in their health management, they still needed support from their parents. The continuity, to have met the same pediatrician over the years, had generated a feeling of trust and security. The adolescents expressed a wish to be able to meet the adult health care team before the transfer, so they could be better prepared for the transfer.

5.1.2 Knowledge and information-adolescents

The parents were important for receiving medical information, although some of the adolescents claimed that they tried to pose the questions themselves. One adolescent stated that information that can be important for you when you get older must be given at a younger age, e.g. about pregnancies.

5.1.3 Daily living-adolescents

The parents played an important role for the adolescents in this study. It was also important to learn about how to deal with limitations in the daily life.

5.1.4 Change of relationship-parents

Similar to the adolescents, continuity was repeatedly mentioned. For the parents, the health care transition meant that their role was changing. One parent expressed a sense of loss of not being needed any longer as a mother to a child with a heart malformation, but at the same time it could be relief to shift the responsibility. They also expressed a need for support in

learning how to facilitate the adolescent in taking over the responsibility for their care. Some parents expressed worries about changing health care environment for the child, but at the same time one parent expressed confidence that the adult care setting would probably have the same expertise as the pediatric setting.

5.1.5 Daily living-parents

In daily living, it was important for the parents to be available for the child and to encourage the child to participate in normal activities.

5.2 STUDY II

In all, 17 adolescents (10 girls and 7 boys) participated in four focus group interviews. The age of the participants was 14–18 years old (mean age 15.8 years), and there were 3–6 participants in each group. The analysis resulted in one main category, *To become a manager of the CHD*, and the four sub-categories of *Sufficient knowledge about the health*, *Be a participant in the care*, *Parental support*, and *Communication with others about the health*.

5.2.1 Sufficient knowledge about the health

The adolescents wanted information about their CHD and other health-related information. The groups discussed how the information could be difficult to understand, e.g. becoming exhausted when playing sports, and how sometimes the information was exaggerated, e.g. information about sex and alcohol.

5.2.2 Be a participant in their care

The adolescents discussed learning how to handle self-management, e.g. taking their medication. To increase the involvement in the care, the adolescents wanted the physician to address the questions to them and not their parents. This could be supportive and helped them to be more reflective about their health management. However, the older participants wanted a dialogue with the physician, and not a monolog.

5.2.3 Parental support

Parental support was described to be crucial. The parents were important in encouraging them to participate in the consultations and in interpreting the medical information for them. To meet the physician alone during the consultation was discussed in all groups. For the older adolescents, this was mentioned as important. However, it was comforting to have one's parents waiting in the waiting room. Such meetings had to be introduced to the adolescent

and the parent in advance. Some of the adolescents felt totally unprepared for such meetings, however, and refused to be without their parents.

5.2.4 Communication with others about the health

To learn how to communicate with others was described as a challenge. Many of the younger adolescents described how they became frustrated when they could not explain their condition to others, e.g. if they got tired and had to lie down to rest. They got annoyed when people got worried when they just needed a short break.

The participants in the focus groups shared their opinions in a friendly way, and they expressed how good it was to discuss things with others who shared the same experiences.

5.3 STUDY III

Out of the total of 593 included adolescents and 1,186 parents, 202 adolescents (response rate 34%) and 415 parents accepted participation in the study. There were 157 complete triads (adolescent, mother, and father) and 37 dyads (adolescent and mother or father). Eight adolescents participated without their parents, and 52 parents participated without their adolescent.

The participants in this study was the 157 complete triads.

The adolescents' overall readiness mean score was 2.7 (± 0.9) (range 2–4) and it increased with age with age ($p=0.021$). The adolescents' responsibility mean score was 2.4 ± 0.7 , and the reported score increased with age ($p = 0.001$), while the parental involvement score (mean 3.6 ± 0.6) decreased with age, but not significantly ($p = 0.087$). There was a significant difference between the adolescents' overall transition readiness mean score 2.7 (± 0.9) and the mother's and father's reported overall readiness scores (2.4 ± 1.0 ; $p=0.001$ vs. 2.4 ± 0.9 ; $p = 0.001$). There was no significant difference seen in adolescents' responsibility score (2.4 ± 0.7) compared to their mothers (2.4 ± 0.8 ; $p=0.583$) or fathers (2.5 ± 0.8 ; $p = 0.119$). There was a significant difference in parental involvement score, with the adolescents (3.6 ± 0.6) scoring significantly higher compared to their fathers (3.2 ± 0.8 ; $p=0.001$, Cohen's $d=0.56$), but no difference was seen between the adolescents and their mothers (3.6 ± 0.5 ; $p=0.811$).

The regression analysis showed that an increased level of adolescents' overall transition readiness score was associated with higher mothers' and fathers' overall transition readiness scores (mothers $\beta = 0.277$, $p = 0.002$; fathers $\beta = 0.281$, $p = 0.001$) and with older age ($\beta = 0.109$, $p = 0.055$). Further, higher levels of empowerment ($\beta = 0.146$, $p = 0.026$) and lower

scores on the B-IPQ ($\beta = -.142, p = 0.047$) were associated with higher levels of overall transition readiness. Furthermore, the adolescents' responsibility scores were associated with older age ($\beta = 0.168, p = 0.039$) and higher reported adolescent responsibility scores by mothers ($\beta = 0.367, p = 0.003$) and fathers ($\beta = 0.29, p = 0.02$). Adolescents' perceived parental involvement score was positively associated with mothers perceived parental involvement ($\beta = 0.215, p = 0.023$) and negatively associated with adolescents' age ($\beta = -0.212, p = 0.025$). No association was found regarding general knowledge, QoL, HRQoL or health behavior.

5.4 STUDY IV

Three hundred fifty-one parents (189 mothers and 162 fathers) to adolescents participating in Study III out of 1,186 eligible parents participated in this study. The parents filled in the Parental Uncertainty LAS.

The parents' reported uncertainty scores regarding their adolescent's transfer to adult healthcare were on average 42.5 ± 30.1 . The distribution of uncertainty scores displayed a wide range in the perceived uncertainty. Twenty-four percent had a very low level of uncertainty (score 0–10) and seven percent had a very high level (score 91–100).

When testing the agreement in uncertainty between the matched parents, there was a highly significant ICC agreement of 0.737 (95% confidence interval 0.58–0.84, $p = 0.005$).

However, mothers generally significantly higher level of uncertainty than fathers (46.1 ± 32.4 vs 41.2 ± 33.3 ; $p = 0.03$, Cohen's $d = 0.15$). The univariate regression analysis showed that increased levels of adolescents' overall transition readiness as perceived by the parents, was associated with a lower level of uncertainty ($\beta = -0.6, p = 0.001$). No association was found regarding adolescents' sex, age, CHD complexity, or the parents' age.

Additionally, the results revealed that a third of the parents (mothers 26%; fathers 36%) to adolescents with CHD had not started to think about the future transfer. The proportion of parents reporting "I have not thought about the transfer" did not differ between being a parent of a boy or a girl (mothers, $p = 0.47$; fathers, $p = 0.49$), nor did the complexity of the disease show any significant differences for mothers ($p = 0.84$), although it did for fathers ($p = 0.037$). With increasing age of the adolescent, mothers and fathers reported a decrease in "I have not thought about the transfer" ($p = 0.001$ vs $p = 0.001$).

6 DISCUSSION

6.1 CHANGES IN READINESS DURING THE TRANSITION

The adolescents described the transfer to adult care as a natural step when one grows up (Study I). The adolescents were starting to become aware that they need to take over responsibility for their health management from their parents (Studies I and II), which has also been described previously [81]. Some of the adolescents described how they started to ask their questions themselves during the medical appointments. Many adolescents wanted their own time with their physician, but they emphasized the importance that such time was planned in advance, otherwise they might feel forced and unprepared (Study II). The adolescents need to be encouraged and offered to have their own time with the physician, which has been reported to be important in developing independence [82]. This suggests the importance of involving the adolescent in the transition process and of preparing the family for the changes.

The results in this study showed that adolescents' perceived transition readiness increased with age (Study III), and this phenomenon has also been described in studies of adolescents with other chronic conditions [83], heart disease [84, 85], and liver transplants [32].

Further, greater empowerment and experiencing a less threatening view of the disease were associated with a higher transition readiness (Study III).

6.2 BARRIERS AND FACILITATORS

6.2.1 Continuity

Continuity seemed to be important during the transition. Many of the adolescents and the parents spoke about the gratitude, trust, and the feeling of safety they experienced by meeting the same physician since the first examination (study I). A good relationship with the HCPs seemed to help the adolescents to be involved during the consultations and helped in the communication at the clinical appointments (Studies I and II). The adolescents spoke about the knowledge the HCPs had and how important it was that the physician paid interest in them as persons (Study I). Asp et al. confirm this in a study with young adults with CHD who were transferred to adult healthcare. Continuity in healthcare providers is important when building trust in a caregiver and to be able to vent their concerns and questions [93], and continuity makes it easier to be involved in one's own healthcare management [94].

However, some differences in attitudes towards transitional care exist among pediatric and adult care providers which can be a barrier for continuity [95].

Many parents and some adolescents expressed sorrow for having to leave the pediatric surrounding (Study I). Feeling at ease with the pediatric HCP has been described as a possible barrier for a successful transfer, and this barrier seems to be more evident for the parents [96]. However, having a good relationship is an important opportunity for the pediatric HCP to engage the adolescent in the transition process. One father explained that they had experienced such good care that he was convinced that the new caregiver would also give good care (Study I). Continuity is probably one of the more important factors for a successful transition, and the HCPs can educate the adolescent stepwise during the transition. Also, the adolescent can feel encouraged when they feel that the HCPs have a grasp of the entire picture.

6.2.2 Communication

Communication seems to be an important and difficult issue for the adolescents (II). It sometimes appeared to be a barrier and sometimes a facilitator to gaining knowledge and sharing information about the condition during the transition [12]. The adolescents' communication skills seem to develop with age. The result in Study III showed an increased level of self-management and responsibility, which included communicating with HCPs and explaining their condition to others. Some adolescents described how they used their parents to ask their questions during the consultations when they were younger. With time this changed, and the parents started to facilitate them by initiating questions, saying things like, "You had a question about" (Study I). The communication skills seemed to be difficult for younger adolescents. Some of the adolescents found it difficult to explain their condition, for example, to their teachers, and they became annoyed when they could not make themselves understood. This has also been described by adolescents with other chronic conditions [86]. This can perhaps be partly explained by adolescents having difficulties understanding the meaning of information given during the consultations, e.g. about physical activity (Study II). It has been reported that adolescents find it important that teachers are informed about the condition [87], and in many of the pediatric cardiology healthcare settings in Sweden this kind of support is provided by the nurses. One can discuss if there are additional ways for the adolescent in their communication with their school in order to promote self-management and independence. Betz et al. describe the importance of finding strategies for how parents and HCPs can support adolescents in communicating about their health [86].

6.2.3 Knowledge

Disease-specific knowledge is important during the transition. According to Van Deyk et al., low levels of disease-related knowledge in adolescents with CHD can lead to less motivated individuals [88]. In a study on adolescents with CHD, Van Deyk et al. found that fewer than 50 percent knew the name of their heart condition or the reasons for medical follow up [88]. According to Clarizia et al., adolescents with more knowledge about their condition demonstrate better understanding about the transition [89]. However, in Study III, general knowledge was not associated with transition readiness. But at the same time, the adolescents in Studies I and II expressed a wish for learning more about the heart condition, health-related concerns, and the transition. They stated however, that it was important that the information given was not exaggerated but accurate, e.g. about health-related issues (Study II). For the HCP, it can be a challenge to give information to the adolescents. There is information about treatment and health conditions that adolescents need to understand and learn, but it has to be given in an age-appropriate way [90].

6.2.4 Changing responsibility

The adolescents spoke about how they were learning as they got older to take more responsibility in the meetings with the HCP and in learning to manage their health (Studies I and II), which was confirmed in the results in Study III. With older age, the adolescents' responsibilities increased. One can argue about the importance of assessing the transition readiness and adolescents' responsibility for the health in adolescents. When using transition readiness instruments, the HCPs can identify topics that need to be supported. Meleis et al. describe that it is important to monitor outcome indicators to ensure that the transition is developing towards a healthy transition [12]. A transition questionnaire is a self-reported, subjective instrument that can give indications on what direction the transition readiness is developing in, but it is not an objective measure and it does not show if the adolescent is managing their self-care. HCPs caring for adolescents with chronic diseases should also be aware that although adolescents might have knowledge, e.g. about medication, they might take their medication only when it is more convenient and not when they are supposed to [91]. Further, although the adolescents perceive high self-management skills, this might not be accurate [32].

Although parental involvement (study III) decreased over the years with an increase in the adolescents' responsibility, the level of parental involvement was still considered high. This should be further explored. Huang et al. highlighted problems with to extensive parental

involvement. They reported that the more involved the parents are in their adolescents' healthcare, the less interest and control the adolescents take. Further, there is a risk for an abrupt decrease in parental involvement once the adolescent changes environment to adult care, which might lead to a lack of preparation in the adolescent for how to conduct a clinical appointment [92].

6.2.5 Parental support

Concurrent with the adolescent's transition, the parents to adolescents with CHD are also going through a transition. When adolescents with CHD grow up and take over responsibility for their own care from their parents, the parents might experience this as a loss of a certain role, namely losing the attention as a parent to a child with CHD (Study I). Some parents (Study I) explained that it could be experienced as a relief to hand over the responsibility, but it could also give a feeling of sadness of not being needed any longer. Some parents were concerned that new HCPs would not realize that the adolescents needed help and support in the beginning (Study I). This emphasizes the need to involve the parents during the process and to have them on board, especially considering how important parental support has been shown to be. They need to be reassured that the child will be cared for by competent HCPs and that the adolescents have the skills and confidence to manage their health on their own because this will help the parents to hand over responsibility to their child [97].

Although parental support has been argued to be important (Studies I and II), it can also be a barrier for developing independence. In a study among adolescents during transition, half of the group of adolescents perceived some level of parental overprotection [84]. When the parental monitoring is too comprehensive, it can seriously affect adolescents' peer relationship [87]. In Study III, there was a significant difference between the father and adolescent in parental involvement, with the father scoring lower than the adolescent, but no difference was seen between the mother and adolescent.

6.2.6 Parental uncertainty

HCPs must acknowledge the parents' perception of uncertainty during the transition process, and they should, in a structured way, inform the adolescents and the parents about the transition and the new caregivers. It is acknowledged that a coordinated transition might support the family and promote the transition experience [98], and information is an important aspect in reducing and preventing uncertainty [36].

There are reports describing parental concerns during the transition. After raising the child with a chronic condition, the parents meet new demands when they need to start to shift roles regarding responsibility for their child's healthcare management [99]. The adolescents described parental support as essential (Studies I and II). The adolescents used their parents to ask their questions during the medical appointments, and also needed them to explain different things regarding their heart condition (Studies I and II). This confirms the importance of taking the parents into account. They play a key role during the transition for the adolescents in becoming experts on their own condition [100].

The parents described how they felt insecure and uncertain, and questions were raised about how they could support their adolescent during the transition (Study I), which has been reported in previous studies with qualitative designs [29, 82]. The results in Study IV were inconsistent, with some parents scoring high on the Parental Uncertainty Scale while other parents perceived no uncertainty. Other parents had not yet started to think about the transfer and were therefore not worried. This result shows that there is a wide range among the parents. The parents scored on average 42.5 ± 30.1 on the Parental Uncertainty Scale, and there was a significant difference between the parents with mothers scoring higher. Further, there was no difference in uncertainty level according to the adolescent's age, sex, or CHD complexity. This might be seen as contradictory that there is no difference in the level of uncertainty with regards to the complexity of the disease, and it could be expected that parents of adolescents with more complex heart conditions experience more uncertainty. However, because patients with more complex heart conditions have more frequent medical follow-up [9] they might be better informed about the transfer and have had more possibilities to discuss the transfer with their HCPs.

Heath et al. report in a review article about parental stress and fear regarding the healthcare transition, and they emphasize that the adolescents' healthcare transition is a transition for the parents as well [100]. The parents described uncertainty regarding how much responsibility they could hand over to the adolescent as well as fears for complications [100]. Some parents might experience fears and concerns about "letting the child go". The uncertainty might be based on the feeling of being excluded during the transition process, but also to concerns that the child would be good cared for [96]. This is similar to findings described in Study I, where one mother raised the concern that they would take care of her son and that the HCPs would remember that he was a new adult, a beginner. Meeting the new caregiver could be one way to reduce uncertainty about the new setting. This was also emphasized in the interview studies in this thesis (Studies I and II) and has been described in previous studies [101]. Fear,

stress, and uncertainty among parents should be disclosed because these might affect the adolescents during the transition [8]. Suris et al. acknowledge the importance of supporting the parents during the transition and that good coordination between the pediatric healthcare team and the adult healthcare team is crucial elements for parents to be ready for the transition [102]. Although it is considered to be a natural step for adolescents with chronic conditions to grow up and to be transferred to adult care, both parents and their adolescents might feel insecure and uncertain about the transition process and the transfer of care [83, 103, 104].

6.3 METHODOLOGICAL DISCUSSION

This thesis contains both qualitative and quantitative studies to fulfill the overall aim. The trustworthiness in a qualitative study is dependent on the credibility, dependability, and transferability of the results.

Credibility refers to how well data and processes of analysis cover the intended aim as well as how the categories and themes cover the data. Credibility also deals with selection of context, participants and gathering data [72]. In the qualitative studies (Studies I and II), we used purposive sampling to get a variation of participants in age and sex. Nurses and physicians at each hospital were asked to identify eligible patients who fulfilled the inclusion criteria to invite for the study. On the one hand, this ensured that the eligible patients fulfilled the criteria, but there is risk of selection bias. In Studies I and II, the analysis was made with two (Study I) and three (Study II) co-authors to enhance credibility. Dependability refers to consistency over time. The interviews were semi-structured, and an interview guide was created by the research team, to be used as a support during the interviews. Transferability refers to what extent the findings can be transferred to other settings. The author can give suggestions, but it is the reader who decides whether the findings can be transferred to another context. The author must therefore give a clear description of the context and culture [72]. These criteria were fulfilled in both study I and II.

The interviews in Study I were performed at the location where the researcher was working, and this might have had an impact on the data collection. However, to reduce the risk for the adolescents not being able to speak freely, the author had not been part of the adolescents' care. To emphasize the fact that the interviewer was in the role of researcher, the interviewer wore private clothes, instead of the hospital uniform. Throughout the data collection (the interviews) and data analysis, reflective discussions were held with one of the co-authors in order to handle the pre-understanding of the author of this thesis. Reflexivity is an important

process and involves critical reflections on oneself in order to become aware of personal values and experiences that might affect the data collection and interpretation [105].

Focus group interviews were used in Study II, and the participants have certain common characteristics that help them to relate to a special topic in a group discussion [56]. When using this method, it is important for the participants to feel comfortable, respected, and free to share their opinions. [56]. There does not seem to be any consensus on how many participants there should be in a focus-group interview [105, 106], but to get a range and width in the discussions, it was planned to include 6–8 participants (both boys and girls and between 14 and 18 years of age). Due to late drop outs, we had between 3 and 6 participants in our groups. The group with only three participants was still interviewed, and despite the small number of participants the adolescents still discussed a lot. Late drop outs are an unwanted occurrence, and therefore it is recommended to invite more participants than needed to achieve a large enough group for discussion [107]. On the other hand, too many participants can limit the participants in sharing their opinions. Although small, the groups in the present study yielded rich data. A lot of effort was made to involve all of the adolescents [107], but it was clear that the older participants were more active. This was most likely due to better communication skills. There are differences in maturity in the age range (14–18 years) included in this study, but the interaction with the older adolescents encouraged the younger adolescents to participate in the discussions [108, 109]. The interviewees were informed about the voluntariness and confidentiality of participating, that there were no right or wrong answers, and that the topics that were discussed would be “kept in the room” so that no one would be afraid to be pointed out.

For the cross-sectional studies (III and IV), the SWEDCON register provided a unique prospect to identify all eligible patients with CHD who were expected to be transferred to adult care. However, there was a low response rate in study III and study IV which is a limiting factor regarding the external validity to which extent it can be generalized to other situations and to other people. With a low response rate, there is a danger that the responders and non-responders’ characteristics differ [110]. There was no difference in sex or disease complexity between the participants and the non-responders in Study III. However, there was a significant difference in mean age with the responders being slightly older (15.7 ± 1.1 years vs. 15.5 ± 1.1 years). Nonetheless, this difference was not clinically meaningful given the low Cohen’s d of 0.18. Only complete triads were included in study III, and this led to an additional reduction in the number of participants. Also in study IV, only parents with

participating adolescents in study III were included which led to an additional reduction in participants.

When contacting the non-responders to ask if they received their questionnaire, many had not received their study information and wanted a new letter. Others stated that their adolescent felt well and therefore did not want to participate. One must be aware that this procedure might affect the family. We were well aware of this and highlighted the voluntariness of participating.

The distribution of a questionnaire might affect the response rate [110]. However, there is conflicting opinions and no evidence supporting which mode (e-questionnaire or paper questionnaire) is the most successful [111]. Additionally, a questionnaire puts a cognitive demand on the responders, which might have had an impact on the response rate in the study [112]. There were several questionnaires (study III), and this might have led to a heavy demand for the adolescent. Although the response rate was low, the results give important insights into the adolescents' transition readiness.

There were limitations with the RTQ that need to be mentioned. The complexity of the disease and impact of the disease differ in the patient group as do the demands for self-management skills. Only 23 percent of the adolescents were taking medication, explaining why many answered "not applicable" on several items in the adolescents' responsibility domain and parental involvement domain. "Not applicable" was counted as missing data, and a mean score on the domains "adolescents' responsibility" and "parental involvement" was calculated if the respondent answered more than four items in both of those domains. The number of respondents who answered "not applicable" might have had an impact on the results for the adolescents' responsibility and parental involvement assessment, and this must therefore be taken into consideration when interpreting the results.

As discussed previously in this thesis, there are several readiness questionnaires that have been tested and are in use. However, the RTQ has the advantage of having a proxy version for parents to fill in, which was an important reason why it was chosen. In this research project, it was considered important to study the results of how the parents perceive the adolescent's transition readiness because this has an impact on the adolescent. Further, this questionnaire covers domains that are important for adolescents with chronic conditions in general.

Some of the questionnaires in this thesis were created for an adult population and were used in an adult population (KnoCoHm, B-IPQ, and QoL-LAS) [61, 64, 69]. Before adding them

to Study III, they were discussed in the research group and with medical experts. The questionnaires were tested on adolescents for face validity to ensure that the adolescents understood the wordings.

The Parental Uncertainty Scale was created for study IV. The instrument was found to be easy to use, but it has not been tested in other groups of patients and can therefore not be compared to other groups of parents. Further, it is a single-item questionnaire, and one concern with a scale with a global question is to discern if the respondent understood the question that was being asked [112]. However, face validity confirmed that 100% of the parents understood the wording and the scale.

7 CONCLUSION

Continuity and a trustworthy relationship with the HCP are two of the most important facilitators for the adolescent in developing healthcare autonomy and promoting readiness for transition. It is also important to help the adolescent to develop communication skills, and continuity might be a facilitator when educating the adolescent.

It is important to include the adolescent as an active partner in the transition. The adolescent wants disease-specific and health-related information, but it should be given in a developmentally appropriate way. Further, there is room for improvement in preparing the adolescent for their new healthcare providers and GUCH.

The transition readiness increased with the adolescent's age, but age alone is not solely the only factor that determine when the adolescent is ready for transfer. Empowerment and illness perception are also related to transition readiness and therefore emphasis should be given to empowering adolescents and to supporting them in gaining personal control, increased awareness about their condition, and achieving self-management. Even though adolescents' and parents' perceptions differed in transition readiness, they still correlate and show that higher scoring by parents is associated with higher self-perceived transition readiness in adolescents.

There was a wide range of perceived level of uncertainty observed in parents. However, no significant differences were seen between the mothers and fathers of the same adolescent, nor did the adolescents' age, sex, or complexity of the disease seem to have an influence on levels of perceived uncertainty. There was a negative association between parental uncertainty and perceptions of adolescents' transition readiness. That is, parents who perceived their adolescents as being more ready perceived themselves as less uncertain. Further, many of the parents to the 17-year-old adolescents had not thought about the transition, which indicates that there is room for improvement in the preparation for the parents. Parents play a key role in supporting and facilitating their adolescent during the transition, and they might need support in shifting responsibility for their child's healthcare management.

8 CLINICAL IMPLICATIONS

The results of this research provide insight into adolescents' needs, expectations, and readiness for transition. There is a difference in adolescents' complexity of disease, age, and gender, but the need for preparation during transition is universal regardless of the complexity of the disease. The preparation needs to be structured, tailored, and person-centered in order to meet the individual's needs. The results from this research reveal evidence and factors that seem to be important when creating a transition program for adolescents with CHD.

The results provide information about the parental uncertainty regarding transfer and showed a variety in perceived uncertainty in the parent group. Parental uncertainty about the transfer is important to make visible because such uncertainty might obstruct parents' handing over the responsibility for health to their adolescent. Therefore, it might be important to assess parental uncertainty in order to be able to intervene when needed and to better prepare the parents for the transfer, and the Parental Uncertainty Scale might be useful in this matter.

It is important to consider how healthcare resources can be used most efficiently before implementing a transition program. It is therefore essential to explore the needs, the level of transition readiness in adolescents with CHD, and the factors associated with an increased level of readiness before designing a new program.

9 FUTURE RESEARCH

The results from this thesis suggest the following future research directions:

- To further develop and evaluate instruments for monitoring transition readiness in adolescents with CHD.
- To study the parental role during the transition.
- To evaluate the Parental Uncertainty Scale.
- To further evaluate the effects of transition programs because the level of evidence is low. Further research is needed, preferably in the form of randomized control trials with a longitudinal follow up.
- To evaluate the readiness for transition among adolescents with other chronic conditions.

10 ACKNOWLEDGEMENT

De här åren har varit otroligt lärorika på så många sätt. Det är många som på olika sätt varit med och bidragit till att den här avhandlingen blivit av. Jag vill speciellt tacka:

Alla ungdomar och föräldrar i Stockholm, Göteborg, Lund och Umeå som på olika sätt har deltagit i dessa studier. Tack för att ni så generöst delat med er av era erfarenheter och tagit er tid att besvara alla enkäter.

Jag vill fortsätta med att tacka min kvartett av handledare, utan er hade jag inte varit här idag. *Maria Öjmyr-Joelsson*, min huvudhandledare som introducerade mig och uppmuntrade att bli doktorand. Tack för att du trodde på min ide' och lotsade in mig på den här banan.

Margret Nisell, bihandledare, tack för din tid och stöd under alla dessa år. Alltid vänlig, hjälpsam, entusiastisk och positiv.

Björn Frenckner, bihandledare, med stort kunnande och generositet, som alltid tagit sig tid för mitt arbete och mina frågor.

Ewa-Lena Bratt, min bihandledare. Jag har alltid varit så imponerad av din entusiasm, generositet och kunnande som forskare. Du har lärt mig så mycket under de här åren. Du är ett föredöme på så många sätt.

Anders Ringner min kära mentor. Ditt stöd och uppmuntran har betytt mycket för mig under de här åren. Tack för alla dina goda, kloka och roliga råd. Jag är så tacksam för all hjälp och stöd.

Mina underbara kreativa STEPSTONES-CHD kollegor, jag är glad och tacksam att få vara en del av denna grupp.

Mariela Acuna Mora, omtänksam doktorand-kollega vid Göteborgs Universitet och medförfattare i studie III-IV. Samarbetet med dig har varit ett nöje och jag är så tacksam för hjälp med statistikens snåriga klurigheter. Jag ser fram emot vårt fortsatta samarbete i Stepstones-CHD.

Philip Moons. Scientific leader in the Stepstones-project, brilliant researcher and co-author in study III-IV. Thank you for generously sharing your knowledge and time.

Markus Saarijärvi och *Sandra Skogby*, doktorandkollegor i Göteborg, alltid lika trevligt att få arbeta med er.

Carina Sparud-Lundin, docent vid Sahlgrenska akademien, ingår i Stepstones forskargrupp med expertis även inom andra kroniska sjukdomar. Är även medförfattare till studie III-IV, tack för kloka reflektioner och synpunkter.

Helena Wigander and *Elin Öst*, mina underbara doktorandkollegor vid Astrid Lindgrens barnsjukhus. Ni har betytt mycket för mig under de här åren, är tacksam för all hjälp och uppmuntran när det var lite mycket för mig. Uppskattar även vårt snick-snack. Ser fram emot fortsatt samarbete med er.

Per Winberg, barnkardiolog på barnhjärtcentrum och medarbetare för Stepstones i Stockholm och med ett stort patientengagemang. Tack för all hjälp, goda råd, intresse, uppmuntran och att du alltid har tid att svara på frågor.

Bo Lundell, numera pensionerad barnkardiolog i Stockholm, medförfattare i studie I. Tack för all hjälp och stöd inför min doktorandregistrering.

Ett stort till ALLA mina underbara kollegor vid Barnhjärtcentrum; sjuksköterskor, barnkardiologer, biomedicinska analytiker och läkarsekreterare. Tack för all uppmuntran och glada tillrop! Jag är så glad och stolt att få jobba med er. Ser fram emot att åter få vara med på ett och annat skritt i fikarummet här framöver.

Vill även säga ett speciellt tack till min närmaste chef *Anna-Karin Hammarstedt* som stöttat och hjälpt mig på olika sätt och gjort det praktiskt möjligt att slutföra det här. Vill också tacka vår sektionschef på barnhjärtcentrum, *Gunnar Bergman* för ditt stöd och uppmuntran.

Anna Sandberg, Karolinska Institutet och Kvinnors och Barns Hälsa, tack för all administrativ hjälp under de här åren.

Annika Rydberg, Barnkardiolog i Umeå och *Katarina Hanseus*, Barnkardiolog i Lund, medförfattare i studie II -IV. Tack för lärorikt samarbete, uppmuntran och era många kloka och konstruktiva synpunkter.

Ann Asp sjuksköterska i Lund som bidragit på många sätt med datainsamlingen och input.

Birgitta Svensson sjuksköterska och doktorand i Lund, uppskattar dina kloka synpunkter och reflektioner.

Maryann Florbrant och *Liss Ingrid Schröder*, fina vänner och före detta kollegor vid barnintensivvårdsavdelningen på ALB. Vår botanisering bland restauranger i Stockholm har varit underbara (och goda) avbrott i tillvaron. Tack för ert intresse för min forskning och all uppmuntran. Det har betytt mycket för mig.

Min fina familj som alltid varit så viktig för mig. Mina systrar och bror, mina svågrar, svägerskor och alla syskonbarn, ni är så underbara. Mina älskade föräldrar som betytt och betyder så mycket för mig. Mina kära svärföräldrar Sture and Gun-Marie, som alltid stöttat och trott på mig under dessa år.

William Vernersson, min systerson som räddade mig från sammanbrott när min dator inte längre ville samarbeta, lagom till att kappan skulle sammanställas.

Min stora kärlek, Gunnar. Tack för att du hela tiden uppmuntrat mig och trott på min förmåga att jag kan. Ser fram emot att kunna läsa söndagsbilagan i SvD med dig igen. Mina kära, kloka och underbara barn, Kristoffer, Rebecka och Wilma, det bästa som hänt mig. Tack att ni haft tålamod med mig de senaste åren. Jag kommer alltid finnas här för er. Jag älskar er.

Vill även tacka alla generösa bidragsgivare: Stiftelsen Kronprinsessan Lovisas, Ebba Danelius stiftelse, Sällskapet Barnavård, Stiftelsen Samariten, Kvinnor och Hälsa. Stiftelsen Clas Groschinsky Minnesfond, Stiftelsen Kempe-Carlgrén. Jerringfonden, Hjärt-Lungfonden, Forte, Hjärtebarnsförbundet och Vårdförbundet för värdefullt ekonomiskt stöd.

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