Patient empowerment during the transition to adulthood in young persons with chronic conditions

Mariela Acuña Mora

UNIVERSITY OF GOTHENBURG

Institute of Health and Care Sciences
Sahlgrenska Academy, University of Gothenburg

Department of Public Health and Primary Care
KU Leuven

Gothenburg 2021

Cover illustration: Jonathan Ramírez Cabezas

Patient empowerment during the transition to adulthood in young persons with chronic conditions
© Mariela Acuña Mora 2021
mariela.acuna.mora@gu.se

ISBN 978-91-8009-024-7 (PRINT)
ISBN 978-91-8009-025-4 (PDF)
http://hdl.handle.net/2077/65151

Printed in Borås, Sweden 2020
Printed by Stema Specialtryck AB, Borås

SVANENMÄRKET
Trycksak
3041 0234

For my parents

Para mi mamá y papá

ABSTRACT

During the transition to adulthood and adult care young persons with chronic conditions (CCs) are in need of developing the skills necessary to manage their condition, communicate with the healthcare provider, participate in the decision-making process and become autonomous. In order to help them develop such skills, patient empowerment has been suggested as a relevant approach. Through patient empowerment, young persons can become active partners in care and mobilize the resources they need to achieve their goals. Despite its relevance in adolescent health and care, patient empowerment has not been investigated thoroughly in this group. The aim of this doctoral thesis was to explore patient empowerment in young persons with CCs during the transition to adulthood.

Study I was a descriptive review that aimed to inventory the definitions and measurements of patient empowerment, appraise the conceptual and methodological rigor of published studies and to identify correlates of patient empowerment in persons with CCs. Study II was a cross-sectional study that described the development and psychometric evaluation of the Gothenburg Young Persons Empowerment Scale (GYPES). Study III was also a cross-sectional study measuring the level of patient empowerment in young persons with congenital heart disease (CHD) and examining potential correlates of patient empowerment. Study IV was a longitudinal study that aimed to examine the direction of effects between patient empowerment and other patient-reported outcomes.

The findings in this thesis show that there is no consensus on a definition of patient empowerment and available studies have used instruments that on occasion are not meant to measure patient empowerment. Given the limitations of previous instruments, GYPES was developed to measure patient empowerment. Results show the scale is valid and reliable in a sample of young persons with CHD and diabetes mellitus. Research has assessed a broad scope of correlates of patient empowerment, but most of this research has been in cross-sectional studies and the direction of the investigated associations is not yet clear from the available evidence. In young persons with CHD, patient empowerment was correlated with transition readiness and communication skills. Moreover, results from this doctoral thesis indicate that patient empowerment leads to improved communication skills in young persons with CHD.

From the findings of this thesis, it is possible to conclude that patient empowerment is relevant in improving communication skills. These skills are fundamental, as they are an initial step in becoming an active partner of the care process. Additionally, GYPES can be used to compare the level of patient empowerment across different CCs and contexts. Researchers should make an effort to achieve more consensus on definitions and dimensions that comprise patient empowerment to facilitate the comparison of the available evidence.

Keywords: chronic conditions, cross-sectional study, descriptive review, instrument development, longitudinal research, patient empowerment, transition, young persons

ISBN 978-91-8009-024-7 (PRINT)
ISBN 978-91-8009-025-4 (PDF)
http://hdl.handle.net/2077/65151

SAMMANFATTNING PÅ SVENSKA

Under övergången till vuxenlivet och vuxensjukvården behöver unga personer med långvariga tillstånd utveckla färdigheter som krävs för att hantera deras sjukdom, kommunicera med vårdgivaren, delta i beslutsprocesser och utveckla sin självständighet. För att hjälpa ungdomarna att utveckla dessa färdigheter har empowerment (egenmakt) visat sig vara betydelsefullt. Genom att stärka empowerment kan unga bli aktiva partners i vården och därmed mobilisera de resurser de behöver för att uppnå sina mål. Trots att det är relevant för ungdomars hälsa och vård har betydelsen av empowerment inte närmare studerats i denna grupp. Syftet med denna doktorsavhandling var att studera empowerment hos unga personer med långvariga tillstånd under övergången till vuxen livet.

Studie I var en beskrivande litteraturstudie där syftet var att granska och kartlägga definitioner av empowerment, kartlägga instrument avsedda att mäta empowerment, bedöma den konceptuella och metodologiska noggrannheten i publicerade studier samt att identifiera faktorer som samverkar med empowerment hos personer med långvariga tillstånd. Studie II var en tvärsnittsstudie som beskrev utvecklingen och den psykometriska utvärderingen av Gothenburg Young Persons Empowerment Scale (GYPES). Studie III var en tvärsnittsstudie som mätte graden av patient empowerment hos unga personer med medfödda hjärtfel och undersökte möjliga samverkande faktorer till patient empowerment. Studie IV var en longitudinell studie som syftade till att undersöka riktningen av effekterna mellan patient empowerment och andra patientrapporterade data.

Resultaten från studie I visar att det inte finns konsensus om definitionen av empowerment och att tidigare studier ibland har använt instrument som inte är avsedda att mäta empowerment. Av det skälet utvecklades GYPES (Gothenburg Young Persons Empowerment Scale). GYPES består av 15 frågor och är utvecklat för att mäta empowerment hos unga med långvariga tillstånd. I studie II visade den psykometriska utvärderingen att skalan är giltig och tillförlitlig då den testades av unga personer med hjärtfel och unga personer med typ 1 diabetes. Resultatet från studie III och IV visar att empowerment hos unga med hjärtfel var associerat med graden av beredskap och mognad att överföras till vuxensjukvården samt förmåga att kommunicera med sjukvårdspersonalen.

Resultaten från denna avhandling indikerar att empowerment spelar en viktig roll när det gäller unga personers förmåga att kommunicera med sjukvårdspersonalen. Dessa färdigheter är betydelsefulla då de är ett första steg till att bli en aktiv partner i vårdprocessen. Dessutom kan GYPES användas för att jämföra graden av empowerment vid olika långvariga tillstånd hos unga. I ett forskningssammanhang är det betydelsefullt att sträva efter att nå samstämmighet avseende definitioner och dimensioner av empowerment för att underlätta jämförelsen mellan resultat från olika studier och mellan olika grupper.

LIST OF PAPERS

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

I

Acuña Mora M, Sparud-Lundin C, Moons P, Bratt EL. Definitions, instruments and correlates of patient empowerment: a descriptive review. *Submitted.*

II

Acuña Mora M, Luyckx K, Sparud-Lundin C, Peeters M, van Staa A, et al. Patient empowerment in young persons with chronic conditions: psychometric properties of the Gothenburg Young Persons Empowerment Scale (GYPES). *PLoS One.* 2018; 13(7): e0201007.

III

Acuña Mora M, Sparud-Lundin C, Burström Å, Hanseus Å, Rydberg A, Moons P, Bratt EL. Patient empowerment and its correlates in young persons with congenital heart disease. *European Journal of Cardiovascular Nursing.* 2019; 18(5): 389-398.

IV

Acuña Mora M, Sparud-Lundin C, Moons P, Bratt EL. Patient empowerment and patient-reported outcomes: what is the direction of effect? *In manuscript.*

CONTENTS

ABBREVIATIONS 13

DEFINITIONS 14

INTRODUCTION 15

BACKGROUND 16

- Patient empowerment 16
  - Conceptual mapping of patient empowerment 17
  - Available measurements of patient empowerment 22
- Patient empowerment and chronic conditions 23
  - Chronic conditions 23
  - Balancing adolescence and a chronic condition 27

RATIONALE 36

AIM 37

- Overall aim 37
- Specific aims 37

METHODS 38

- Description of study designs 38
  - Descriptive reviews 38
  - Cross-sectional study designs 39
  - Longitudinal study designs 39
- Included studies 40
  - Study I 40
  - Study II 42
  - Study III 48
  - Study IV 51
- Ethical considerations 55

RESULTS 57

- An array of definitions and limited evidence on correlates of patient empowerment (Study I) 57
  - Study characteristics 57

Conceptual and methodological rigor 57

Correlates of patient empowerment 58

GYPES: a valid and reliable measure of empowerment (Study II) 58

Phase I: Face and content validity 58

Phase II: Content validity, factorial validity, internal consistency and responsiveness 59

Phase III: Content validity, factorial validity, internal consistency and responsiveness 61

Transition readiness and communication: significant correlates of empowerment (Study III) 62

Level of empowerment 62

Correlates of patient empowerment 62

Patient empowerment and its predictive value of communication skills (Study IV) 66

Changes in scores between data collections 66

Temporal relationships 66

DISCUSSION 67

How do we define and measure patient empowerment? 67

Is patient empowerment the way to improve other outcomes of relevance for health and wellbeing? 70

What is the evidence on patient empowerment during the transition to adulthood? 74

Methodological considerations 77

CONCLUSIONS 79

IMPLICATIONS FOR CARE 81

FUTURE PERSPECTIVES 83

ACKNOWLEDGEMENTS 84

REFERENCES 87

APPENDIX 103

STUDY I-IV

ABBREVIATIONS

CCs	Chronic Conditions
CFA	Confirmatory Factor Analysis
CFI	Comparative Fit Index
CHD	Congenital Heart Disease
DES	Diabetes Empowerment Scale
GYPES	Gothenburg Young Persons Empowerment Scale
IPQ	Illness Perception Questionnaire
MDES	Making Decisions Empowerment Scale
PedsQL	Pediatric Quality of Life Inventory
RCT	Randomized Controlled Trial
RMSEA	Root Mean Square Error of Approximation
RTQ	Readiness for Transition Questionnaire
SEM	Structural Equation Modeling
SRMR	Standardized Root Mean Square Residual
STEPSTONES	Swedish Transition Effects Project Supporting Teenagers with chrONic mEdical conditionS.
QoL	Quality of Life
WHO	World Health Organization

DEFINITIONS

Adolescence	The phase of life stretching between childhood and adulthood. It encompasses elements of biological growth and major social role transitions [1].
Adolescents and youth/young persons	Adolescents are individuals within the age range of 10-24 years and youth/young persons between the ages of 15-24 years [1].
Chronic conditions	Conditions that last or are expected to last twelve months or more and result in functional limitations and/or the need for functional limitations and/or the need for ongoing medical care [2].
Health	The experience of physical and psychological well-being. Good health and poor health do not occur as a dichotomy but as a continuum. The absence of disease or disability is neither sufficient nor necessary to produce a state of good health [3].
Patient empowerment	An enabling process or outcome that arises from communication with the healthcare professional and a mutual sharing of resources over information relating to illness, which enhances the patient's feelings of control, self-efficacy, coping abilities and ability to achieve change over time [4].
Transfer	Event or series of events through which adolescents and young adults with chronic physical and medical conditions move their care from pediatric to an adult health care environment [5].
Transition	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 [5].

INTRODUCTION

As an adult with asthma and who was diagnosed at an early age, I found it difficult to become involved in the care of my illness as I grew up. This became more accentuated as an adolescent and young adult and stemmed from a combination of factors that led to my not feeling confident in managing my illness, participating in the decision-making process or even asking questions about my disease. My experience is similar to that described by young persons with chronic conditions (CCs) in recent years. Research has found that young persons with CCs have difficulties assuming responsibility, lack sufficient knowledge and do not feel confident in their skills to manage their condition or adulthood [6-8]. Besides dealing with a CC, young people have to deal with different tasks associated with adolescence, such as developing a sense of self, planning for the future and becoming autonomous, all while undergoing a series of physical changes [9].

Patient empowerment has been suggested as a relevant approach in the care of persons with CCs [10]. While most research has involved adults, the relevance of this construct in adolescent health and medicine has been highlighted [11]. Through patient empowerment, it is possible to help the young person foster psychosocial skills that will help them in adolescence, during the transition to adulthood and the transfer to adult care. Empowered young persons can be defined as those that actively participate in care and are aware of the multiple factors influencing their health. Furthermore, they are able to recognize when they need assistance and feel confident to help others going through a similar situation [12].

Previous research has mostly involved adults and there is little evidence on patient empowerment in young persons with CCs. Is it possible to assume that this evidence is relevant for young persons with CCs? Furthermore, what is the role of patient empowerment during the transition to adulthood and how is it associated with other transition-related variables? Given these gaps in knowledge, this thesis aims to explore patient empowerment in young persons with CCs during the transition to adulthood.

BACKGROUND

Health and illness are core concepts in nursing science. However, given that nursing is a health-oriented discipline, focus should mostly be placed on the experience of health. The work nurses do is defined in terms of maintaining health or bringing a state of health to the individual [13]. Health can be seen as the lens through which nurses approach different tasks, such as when making assessments, designing interventions or evaluating care plans [13].

In nursing literature, health has been described in different ways, for instance, as optimum wellness, being whole or maximizing development of an individual's potential [14]. Health is understood as the experience of both physical and psychological well-being and it does not only entail the absence of disease [3]. It is a resource that is highly valued and prioritized in society, and allows people to function and undertake day-to-day activities [15]. It is multidimensional, influenced by different determinants and it is experiential. For this reason, health can be experienced and interpreted differently by each person. The patients' experiences have considerable influence on their health-related behavior and participation in the care process [16]. Thus, even when two people have the same disease, the perceived burden and experience of this disease can differ greatly. The experience can also be determined by whether the person has been diagnosed with an acute or a long-term condition or even whether the CC was diagnosed at birth (i.e. congenital) or later on in life.

Nurses can help patients achieve health by implementing strategies or by promoting certain outcomes. In this doctoral thesis, it is understood that one way to achieve health is by fostering empowerment in persons with CCs.

Patient empowerment

The origins of empowerment are associated with the Brazilian Paulo Freire, an educator who thought students should be more involved in the learning process in order to achieve better outcomes. According to Freire, students should critically question issues affecting them and participate in the decision-making process [17]. Additionally, empowerment involves not only individual change but also structural change (i.e. changing the conditions that affect the status of the individual). In this context, the role of the educator is to help the person become an actor in their learning process and eventually in their own life and society.

The concept of empowerment in healthcare is referred to as patient empowerment. It was introduced in the field of health promotion with the aim of increasing patient participation. The Ottawa Charter refers to patient empowerment as the main focus of health promotion [18]. Empowerment involves not only improving self-efficacy and health behaviors, but also targeting individual, group and structural change [17].

Conceptual mapping of patient empowerment

When revising the literature, there are many definitions available for empowerment and sometimes, it is better understood by its absence (e.g. alienation, powerlessness, helplessness, oppression) [19]. Empowerment originates from the Latin "*potere*", which can be translated as "*to be able*" or "*to enable*" [20]. These two translations add a layer of complexity to the concept, as it allows the reader to interpret empowerment as a process, an outcome or both [21]. When understood as a process it is associated with the professional relinquishing power and helping the person find solutions to the issues affecting them. As an outcome, it refers to the person taking over responsibility, becoming more autonomous and being aware of the situation he or she is in [20]. Empowerment involves a relationship with others, which makes it a transactional concept. It is also dynamic and democratic because of the redistribution of power that occurs and that should lead to social justice and the potential improvement of the individual [20].

Given the duality of the initial concept (i.e. empowerment), definitions across the literature will focus on defining patient empowerment as an outcome, process or both. For instance, if patient empowerment is considered as a process, Aujoulat [10] defines it as "*a process of communication and education between professionals and patients, in which knowledge, values and power are shared, which is seen as a process of personal transformation*". On the other hand, patient empowerment as an outcome can be understood as "*the capacity shown by patients with chronic disease to accept their illness and to develop and use specific coping strategies in order to regain a sense of control*" [22]. See Table 1 for additional definitions of patient empowerment.

Table 1. Definitions of patient empowerment

Definition	Author (s)
"*The process that enables patients to exert more influence over their individual health by increasing their capacities to gain more control over issues they themselves define as important*"	Castro [23]
"*Helping patients discover and develop the inherent capacity to be responsible for one's own life*"	Funnell [24]
"*An iterative process in which a person who lacks power sets a personally meaningful goal oriented toward increasing power, takes action toward that goal, and observes and reflects on the impact of this action, drawing on his or her evolving self-efficacy, knowledge and competence related to the goal*"	Cattaneo [25]
"*Empowerment is a process by which people, organizations, and communities gain mastery over issues of concern to them and psychological empowerment is a feeling of control, a critical awareness of one's environment and an active engagement in it*"	Zimmerman [26]
"*The motivation and capacity that patients can use to participate in decision-making, thus creating the opportunity to shift the balance of power in their relationship with the health professionals*"	Fumagalli [27]

Even when there is an array of definitions on patient empowerment, many with shared attributes, their origins are uncertain and sometimes not clear enough to differentiate the concept from other constructs in healthcare. Nevertheless, Small and colleagues [4] have proposed a definition of patient empowerment based on a literature review. They define patient empowerment as "*an enabling process or outcome that arises from communication with the healthcare professional and a mutual sharing of resources over information relating to illness, which enhances the patient's feelings of control, self-efficacy, coping abilities and ability to achieve change over time*" [4]. This definition accounts for the duality of the concept and considers the transactional, dynamic and democratic attributes associated with empowerment.

Besides providing a definition for patient empowerment, Small and co-authors [4] propose five dimensions that comprise patient empowerment. These dimensions are based on a series of qualitative interviews that involved adults with CCs. The dimensions are:

1. Knowledge and understanding: the level of disease-related knowledge the patients need in order to feel in control of their health and lives
2. Personal control: each patient should have the ability to manage their disease outside of the clinic. This involves having strategies to stay in control and be able to communicate

3. Identity: this entails how much the illness influences the lives of the patients and their sense of self
4. Shared decision-making: is the feeling of being able to make personal decisions together with the healthcare provider and having the opportunity to participate in the decision-making process
5. Enabling others: is the ability to share with others individual experiences and coping strategies, as well as motivating others who are going through a similar situation

Antecedents of patient empowerment

Working from a patient empowerment approach entails a paradigm shift. This shift involves the healthcare provider allowing the patient to take "the driver seat" in the care process. It means letting the patient guide the care process, while the healthcare professional becomes a partner, and a source of support, information and education. To be able to facilitate patient empowerment, there are certain antecedents that need to be fulfilled.

First, as Gibson [20] mentions, health belongs to the individual and it is the person who is responsible for his or her health. Individuals have the capacity to make decisions and act upon them but require information and help to do so [20, 28]. Second, the promotion of empowerment involves relationships that are based on mutual trust and respect [21]. Healthcare providers need to be aware and respectful of the patients' beliefs and trust in the decisions they make, as well as their capability to accept responsibility and act for themselves [29]. The third antecedent is related to active participation and motivation. An unmotivated person will not engage in the activities that facilitate empowerment. Fourth, healthcare providers need to surrender their need for control and promote a partnership with the patient [28]. They need to adopt a perspective that replaces a paternalistic view, where healthcare providers are the ones with all the knowledge [20]. Fifth, literature suggests that a person-centered care approach is required in order to facilitate patient empowerment. This approach to care involves individualizing care according to the individual needs, desires and circumstances of each person, not only focusing on health related aspects, but rather planning beyond the illness [23]. Figure 1 provides an overview of these antecedents and their influence over the process of empowerment.

Empowering processes

The process of empowerment is not linear, and while individuals may feel like they are moving forward in developing their skills, it is also possible for them to take a step back in the process (See Figure 1). This usually starts with an awareness phase and ends with an action [30]. The process of empowerment also entails a patient-healthcare provider interaction that is based on collaboration, negotiation and participatory decision-making [20]. Empowering processes found in the literature that cover some of the previously mentioned aspects are person-centered interventions, shared decision-making models, motivational interviewing, counselling and health coaching [28]. In health promotion, Freire's three-step approach has served as the basis for empowering education interventions. This approach comprises: 1) listening to understand the issue; 2) participatory dialogue about the issues; and 3) action or positive changes that people envision during the dialogue [17].

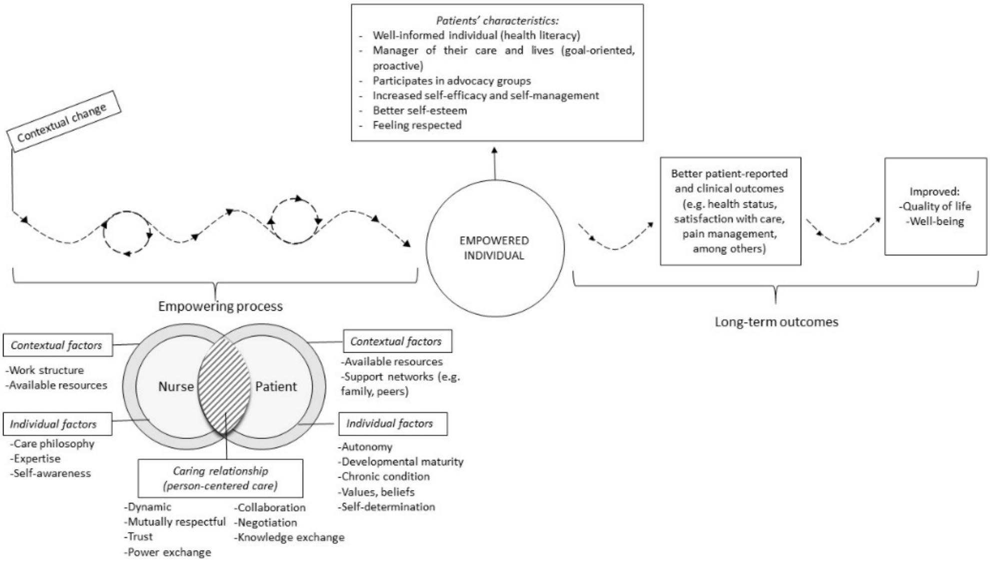


Figure 1. Empowerment model [12].

Characteristics of an empowered person

As shown in Figure 1, there are certain individual characteristics associated with an empowered individual. These characteristics should not be understood as a checklist that all individuals need to fulfill, but rather, they represent some of the characteristics that have been associated with an empowered individual [26].

According to Falk-Rafael [31] changes associated with empowerment are related to three different dimensions: changes in self, changes in behaviors and changes in relationships. Changes in self can be interpreted as obtaining higher levels of self-efficacy, self-esteem and increased autonomy. Changes in self are associated with altering one's self-evaluation or self-image. A successful empowering process leads to an integrated self, which means the person comes to terms with their threatened sense of security and identity [23]. Changes in behaviors relate to increased knowledge and skills development, which eventually lead to healthier choices. Changes in relationships are an outcome of patients becoming more involved in their care [31]. Changes in self, behaviors and relationships are the precursors to better patient-reported and clinical outcomes [12]. The characteristics shown in Figure 1 are representative of these changes.

Patient empowerment aims to help people gain control of their lives [32]. Assertiveness and confidence to speak up for themselves and for others who are experiencing unfair treatment, are characteristics of an empowered individual [31]. They also develop a sense of mastery, a sense of control, feelings of hope and social justice [20]. Being empowered leads to individuals who are purposeful and goal-oriented. St-Cyr-Tribble [30] identified as indicators of empowerment: awareness of one's life situation, own strengths and needs, increase in self-esteem, decrease in negative feelings, well-informed decision making, learning and developing skills, taking action, developing relationships with the social support and networks available and improvements in living conditions.

Knowledge is an essential aspect of empowerment. Patients need to have sufficient and adequate knowledge to make informed-decisions, define strategies to achieve change and solve problems. Knowledge is associated with health literacy, which refers to the ability to undertake knowledge-based tasks [33]. Literature on patient empowerment and health literacy suggests these concepts are closely related [34, 35]. If a person is aiming to achieve adequate disease management, having adequate knowledge and judgement skills is not enough; the motivation to participate in healthcare-related decisions is also necessary [36].

Patient empowerment and other outcomes

As shown in Figure 1, patient empowerment has been associated with patient-reported and clinical outcomes. The effects this construct may have over other outcomes can be immediate or occurs over time. The most frequent outcomes that have been investigated in relation to this construct are self-efficacy and self-management, concepts that are on occasion used as synonyms [23]. As mentioned in the previous section, empowerment is associated with changes in self and particularly with improvements in self-efficacy [37-41]. Self-efficacy has similar attributes to empowerment. In contrast, self-management is associated with the ability to self-monitor, actively participate in self-care and problem-solve [42]. However, these concepts are linked to the personal level, while empowerment has psychological, social and political elements. Additionally, empowerment describes care practices and characteristics of the healthcare system [43].

Studies aiming to identify other patient-reported outcomes associated with empowerment have found positive correlations with capacity to cope with negative feelings [37], disease-related knowledge [44, 45], adaptation to CC [28], health-related quality of life (QoL) [46], adherence [45, 47], healthy behaviors [48], stress reduction [40], health status [28, 39], QoL [28, 40, 49] and well-being [37]. Additionally, higher empowerment led to a decrease in diabetes distress [50] and healthcare utilization [46]. Research has also identified clinical outcomes associated with empowerment; among them, it is possible to mention positive correlations with glycemic and metabolic control [38, 46, 50, 51] and viral suppression [47].

Direct benefits or outcomes associated with empowerment relate to the improvement of individual skills. However, it is also possible to obtain indirect benefits, by involving caregivers, peers and healthcare providers and having better healthcare access and services [52]. Empowerment is also associated with improvements in the healthcare system. Preliminary evidence indicates that empowering interventions are cost-effective [53, 54], an outcome that may be associated with the idea that empowered individuals are better informed, capable of making healthier choices and adherence, which eventually leads to a decrease in healthcare costs [43].

Available measurements of patient empowerment

There are several instruments available to measure patient empowerment and some are disease, context or age-group specific [43, 55]. To date, there is no consensus on which instrument can be considered the "gold standard" for measuring patient empowerment. Systematic reviews that have identified the number of instruments associated with patient empowerment found between 13 to 50 questionnaires associated with the construct [56, 57]. In 2015, a systematic review that included 19 instruments measuring patient empowerment came to the conclusion that the quality of the instruments was poor to fair and that many important psychometric properties remained untested [55]. Another finding of this review was the lack of conceptual clarity among the instruments, with some overlapping with other constructs or using different terms when describing patient empowerment [55].

The lack of conceptual consensus in patient empowerment is supported by the different domains covered in the instruments, which include control, self-capacity, identity, advocacy, power, participation, self-esteem, positive relationships, coping strategies, decision-making and the ability to set objectives [43, 58]. Additionally, reviews trying to categorize the instruments according to their items and domains have found different themes. For example, the review by Barr and co-authors [55] had the four following themes: 1) patient states, experiences and capacities; 2) patient actions and behaviors; 3) patient self-determination within the healthcare relationship; and 4) developing patient skills, whereas the review by Pekonen and colleagues [56] included patient capacities, patient knowledge, patient behavior and support from others. This heterogeneity of the domains included in the instruments complicates the comparison of findings across different studies, as different measurements can capture different aspects of patient empowerment based on their theoretical and conceptual groundwork [55, 59]

Researchers should focus on developing instruments of patient empowerment that have a clear theoretical and conceptual background, as well as good psychometric properties. Preferably, instruments should be generic [60], despite the call from some authors against this. A generic scale facilitates the comparison of empowerment scores across different groups and makes it possible to develop empowering interventions that are inclusive of different groups.

Patient empowerment and chronic conditions

As previously mentioned, empowerment has been introduced in health promotion, but the concept evolved as a way of approaching the care of persons with CCs. In 1991, Funnell [61] was one of the first authors to suggest patient empowerment in the care of persons with diabetes mellitus.

Chronic conditions

CCs account for 70% of the global burden of disease [62]. In Sweden, care for this group accounts for approximately 80-85% of all healthcare costs [63]. Additionally, they carry societal costs such as lower wages, workforce participation and, labor productivity, early retirement and disability [64]. To manage the care of this growing population, healthcare systems have to go beyond a traditional approach (focused on acute onset of illness and of short duration) and also start implementing models that ensure accessibility, continuity of care and collaboration between healthcare professionals [62, 65]. Healthcare systems also need to involve the individual in the care process more and acknowledge the relevance of considering them as a member of the healthcare process. Furthermore, persons with CCs also need to have the skills to manage their condition and be able to navigate the healthcare system, all while balancing tasks associated with different life phases. The aforementioned tasks can be achieve by implementing a person-centered approach and patient-empowering strategies [66, 67].

There are different definitions for CCs across the literature. Hwang and colleagues [68] consider CCs as those that last 12 months or more and result in functional limitations and the need for ongoing medical care. For Bernstein and colleagues [69] CCs are permanent, leave residual disability, are non-reversible and require rehabilitation. Variations in the definitions arise from including different criteria in relation to the duration/latency, need for medical attention, effect on function, non-contagious nature, departure from well-being and pathology of the CC [70]. For the purpose of this thesis, CCs are defined as those that last or are expected to last twelve months or more and result in functional limitations and/or the need for functional limitations and/or the need for ongoing medical care [2]. This definition was selected given that when reviewing the literature, the majority of studies use definitions or criteria that establish a CC should last more than 12 months. Additionally, this definition highlights the need for continuity of care.

Approximately one in three adults live with a CC or multiple CCs [71] and they are the leading cause of mortality in Europe [65]. Country-specific prevalence rates indicate that in Denmark, 65.6% of their population has one CC and in Sweden, around 50% of the population has a CC [2, 63]. The increase in the prevalence of CCs is characteristic of an epidemiological transition, where rates of infectious diseases decrease and these are no longer the major cause of mortality as they are replaced by CCs [72, 73]. The epidemiological transition, and hence the changes in prevalence patterns of CCs, have been caused by improvements in the medical field, such as better diagnostic techniques, new and enhanced treatments and the implementation of better public health policies [73, 74]. Additionally, the increment of certain CCs has been associated with an ageing population, changes in health behaviors and genetic predispositions [65].

Young persons with chronic conditions

The aforementioned medical improvements have also led to decreased mortality in children with conditions that were previously considered to incur a short life expectancy, such as congenital heart disease, cystic fibrosis and spina bifida [75]. Nowadays, children with these conditions are expected to reach adulthood. Although the survival of children with certain CCs has increased in recent years, the prevalence of these has remained quite stable and does not entirely account for the increase of CCs among children and youths [75]. It has been suggested that an increase in the incidence of asthma, obesity, diabetes, mental health illnesses and neurodevelopmental disorders may be the cause of the rise in prevalence of CCs [76-78].

The prevalence of children living with CCs varies considerably when reviewing the literature. These variations account for the different criteria used when identifying cases of children with CCs, as some studies do not include mild disorders or children with physical limitations [79]. It is estimated that between 15-40% of children and young people live with a CC [79, 80]. During adolescence, the most common CCs to be reported are asthma, diabetes, allergies, cerebral palsy, heart defects and mental disorders [76, 81, 82].

Congenital heart disease: an excellent sample case of chronic conditions

Congenital heart diseases (CHD) can be defined as a "*gross structural abnormality of the heart or intrathoracic great vessels that is actually or potentially of functional significance*" [83]. They are an example of a CC that is usually diagnosed early on, sometimes during prenatal follow-up, and that requires long-term follow-up. Given the heterogeneity of the disorders, care needs can vary greatly across patients. This patient group is therefore a suitable example to use when discussing CCs in children and young persons.

CHD are the most common birth defects [84] with a worldwide prevalence of approximately 9.1 per 1000 live births [79]. In Europe, it is around 7.2 per 1000 live births [84]. Survival of children with CHD has increased in recent decades and today, more than 90% are expected to reach adulthood, a percentage that is expected to increase with continuous medical advancements [85-87].

These defects can be categorized based on their pathophysiology (e.g. left-to-right shunt lesions/acyanotic, or right-to-left shunt/cyanotic or obstructive lesions) or by their complexity (e.g. mild, moderate and severe) [88]. CHD can comprise small septal defects and minor valvar obstructions or be as complex as involving several blood vessels or a combination of different defects. Treatment for CHD will also vary according to its complexity; some patients may not require any interventions, while others could undergo catheter or invasive interventions or need pharmacological treatment. Additionally, after surgical repair, some CHD are associated with sequelae and long-term complications [85].

Some studies have found a lower QoL in children and young persons with CHD [89, 90] while others have found that their QoL was similar to those of their healthy peers or even better [91, 92]. Adverse neurodevelopmental outcomes, such as attention deficit disorder/ attention deficit hyperactivity disorder and intellectual disability, have been found to be higher in children with complex CHD [93]. Moreover, the odds of reporting worse health, more days of school absence, more emergency visits or worse academic outcomes are higher in children with CHD [93, 94]. Some children and young persons with CHD engaged in health-damaging behaviors [95]. Additionally, for some living with CHD entailed not being able to fulfill expected cultural roles and they experienced body image issues related to their scars [95, 96].

Some patients with CHD struggled to reconcile their diagnosis and had anxiety about the future. This anxiety was expressed in relation to career planning, building a family or even their life expectancy [95]. Nonetheless, this anxiety started to decrease as they accepted their CHD. They recognized they wanted to control their disease, rather than the other way around, and this required them to assimilate more information so they could adapt their routines [95]. For some young people, their CHD eventually became something that contributed to their personal strength and helped in their maturity [95].

The level of disease-related knowledge, treatment, need for follow-up or even possible signs of complications varied [97]. In many instances, children reached adolescence with little understanding of their CHD, and this was associated with a lack of direct discussion or parental overprotection [98]. Additionally, many had misconceptions about their CHD [8, 99, 100] or some were not interested in learning about their CHD [96]. Young persons with CHD have expressed their need for more information and not only related to their defect, but also in relation to alcohol, drugs, smoking, piercing, pregnancy and contraception [101]. Moreover, some young persons are interested in learning more about self-management and self-care, improving their communication skills, learning about the significance of regular follow-up and cultivating a positive attitude toward their illness [102].

Balancing adolescence and a chronic condition

Growing up with a CC places an additional burden on the young person. Adolescence is already a sensitive period and living with a CC entails having to balance the needs and tasks of adolescence with those of the CC. Adolescents state that living with a CC is a balancing act, between trying to achieve normalcy and being capable of managing their illness [103]. As Yeo [104] writes, "*adolescents with CC have the same developmental needs as their healthy peers*". Therefore, attention should be paid to both the developmental and health needs of this group.

Adolescence: an important developmental stage

Adolescence is considered as a transition period from childhood to adulthood that comprises a series of physical, cognitive, social and emotional changes. Societies usually define this period in terms of age and social roles and studies will refer to different age ranges to define this stage [9]. Defining the period that comprises adolescence varies according to the source. The World Health Organization (WHO) considers adolescence as the period between the ages of 10-19 years [105]. However, some authors consider this period should extend from 10-24 years [1]. Nevertheless, as Arnett [106] mentions, the period between 18-25 years can be understood as emerging adulthood, which sometimes can extend up to 30 years of age.

Puberty is considered the beginning of adolescence. It involves a cascade of endocrine changes that eventually lead to sexual maturation and reproductive capability. Moreover, it includes great physical growth and considerable brain maturation changes [107]. Cognitive changes associated with adolescence relate to moving from concrete thinking towards abstract thinking. During this period, it is also a time for adolescents to gradually start planning for the future and learning how to control their impulses [108]. At the end of this period, they are capable of delaying gratification and have an interest in moral reasoning [9].

Besides the physical and cognitive changes this group will experience, there are social and emotional changes that occur simultaneously. One of the most important is the development of the individual's identity. Adolescence is a time when young people tend to feel strange or insecure about themselves and their bodies. It is also during this period that they tend to evaluate themselves in relation to others and are subject to being influenced by peers [109]. Parental influence takes a step back and adolescents tend to rely more on their friends. Another important aspect is that at the beginning of this developmental stage, they will start having a desire for independence and will test rules and boundaries. It is expected that by the end, they will be self-reliant and independent [110].

During adolescence, there are different developmental tasks to be accomplished, such as consolidating identity, achieving independence from parents, establishing new, more mature relationships, acquiring a set of values and finding a vocation [109, 111]. Nevertheless, some of these tasks are not reached during adolescence and continue during the period known as emerging adulthood [106]. To achieve the aforementioned tasks, adolescents need to be in an environment that allows them to express and explore who they are, but they also need to have the resources necessary to reach them. Additionally, the expectations related to each one of those tasks can vary greatly depending on the sociocultural context they live in.

From a life course perspective, adolescence is a foundation for future health [9]. As stated by Viner and Macfarlane [112] "*adolescence is a time when new health behaviors are laid down—behaviors that track into adulthood and will influence health and morbidity throughout life*". This period is when adolescents begin to experiment with alternative health behaviors to those of their parents and that have up until this point had the biggest influence on them. It is the period when smoking, alcohol consumption, experimenting with drugs and risky sex behaviors can occur [113]. Additionally, health behaviors related to exercise and food intake are also established during this stage [112].

Living with a chronic condition during adolescence

The effect a CC condition has on this developmental stage varies according to the disease severity, need for treatment, patient-related aspects (psychosocial skills level) and interpersonal aspects (healthcare availability, family resources) [114]. CCs can have physical, emotional, social, educational and vocational effects on the adolescents. CCs that lead to malnutrition or chronic inflammation have been found to cause short stature and delay puberty [104, 114], while other conditions, such as some types of heart defects, involve surgical interventions, leaving scars that may cause body and self-esteem issues during adolescence.

Some CCs may also lead to poor school performance, which results from absenteeism due to medical appointments or hospitalizations. In the long term, this can affect the young person's prospects of entering the job market and achieving financial independence [115]. Effects on mental health vary across illnesses but for some, the level of anxiety and depressive disorders is similar to that of healthy peers. However, for other CCs, such as sickle-cell disease, it was found to be associated with behavioral problems and mental and emotional difficulties [116].

Adolescents may also experience social isolation and difficulties establishing friendships. Social support is crucial during adolescence. It is considered a key aspect of healthy youth and can improve QoL. For young persons with CCs it has been suggested that including close friends and peers in the process of disease management can potentially lead to better disease adaptation [117].

The fact that adolescents tend to experiment more, can also affect their disease management and health status [115]. Studies have identified that adolescents with CCs engage in risky health behaviors as much as their healthy peers [118-120]. A study undertaken in Sweden found that adolescents with CCs are triple troubled, this is the result of living with a CC, engaging in risky health behaviors and having less protective factors [121].

Research describing the lived experiences of CC in adolescents has found this experience to have positive and negative aspects and that while for some the CC has no effect on their daily lives, for others it is challenging [122]. For the latter it has been associated with having a lower health-related QoL, resilience and social support [117]. Among the positive aspects that young persons associated with a CC were the support and stability provided by family and peers [103]. They also mentioned that having a CC made them more caring, loving and understanding [122].

Transitioning to adulthood and to adult care

During adolescence, different transitions start (e.g. to adulthood, to adult care, to working life, among others). These transitions involve different tasks and require different skills and resources from the adolescent. Meleis [123] has proposed a middle-range theory on transition, in which she considers transitions are triggered by critical events and can be defined as "*a passage from one life phase, condition, or status to another…Transition refers to both the process and outcome of complex person-environment interactions. It may involve more than one person and is embedded in the context and the situation*"[123]. There are four types of transitions: 1) developmental (e.g. adolescence, becoming a parent); 2) situational (e.g. immigration, widowhood, moving out of abusive relationships, joining the workforce, changing jobs); 3) health/illness (e.g. becoming ill, recovering from illness, transition to outpatient or home care, changes in treatments); and 4) organizational (e.g. changes in leadership, introduction of new policies or guidelines, changes in environment).

Adolescents with CCs experience a developmental transition - the movement from adolescence towards adulthood - but they also go through a health/illness transition, which is the process of going from pediatric to adult care. In turn, this entails going through an organizational transition, whereby the adolescent experiences changes in leadership, the introduction of new policies and changes in the care environment. Hence, adolescents with CCs experience three simultaneous transitions (i.e. developmental, health/illness, and organizational) that require sufficient support and resources for them to proceed as smoothly as possible.

Healthcare transitions

In the literature, the developmental and health/illness transitions of young persons with CCs are usually referred to as "*transition of care*". There are over 14 definitions available for transition. In the literature, the definitions used by the Society for Adolescent Medicine [124], the American Academy of Pediatrics [125] and the definition proposed by Knauth Meadows [5], are the most frequently used. The latter defines transition 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*". She also proposes that transfer of care is the "*event or series of events through which adolescents and young adults with chronic physical and medical conditions move their care from pediatric to an adult health care environment*" [5]. Her definitions on transition and transfer are the ones used in this thesis when referring to either concept. This is because the definitions highlight the health/illness and developmental transitions, provide an outcome of the transition process and differentiate clearly between transition and transfer. The concepts of transition and transfer are sometimes used interchangeably in the literature. However, it is important to make the distinction between both concepts, since transition is a process that requires time, and continues even after the young person has been already established in adult health care (i.e. after transfer). The focus of this doctoral thesis is on the transition to adulthood and to adult care.

According to the Society for Adolescent Medicine [126] there are several fundamental principles that ought to be considered when working towards improving the transition process:

- Services need to be age and developmentally appropriate.
- Transition programs should address common aspects of adolescence and not only disease-specific concerns.

- In order to prevent dependency, psychosocial delay and developmental difficulties, enhancing autonomy, increasing a sense of personal responsibility and self-reliance are important.

Transition aims to provide uninterrupted, flexible, comprehensive, age and developmentally appropriate care [127]. Ideally, transition preparation should start early on, if possible, before the young person reaches adolescence [127]. While there is no consensus on when this process should start, there is clarity in that it should continue even after the adolescent has turned 18 years and is already in adult care.

Viner [11] proposes that ensuring an effective transition involves three elements: 1) a cultural shift in staff attitudes and training; 2) healthcare systems need to implement transition policies and programs; and 3) young persons need to be trained and empowered so they can become active partners in their own transition. Additionally, he suggests that transition can be successful when it is recognized as an essential component of adolescent medicine, when there is a healthcare provider coordinating the process, and there is a written individualized healthcare transition plan that is regularly reviewed and updated [11].

Transition programs are the interventions targeting the transition process. The curriculum of these interventions should include information related to the CC (treatment, complications, risks, disease management, among others), the adolescence stage, lifestyle issues, skills training and career and vocational planning [5]. The ultimate goal of transition programs is to optimize QoL and maximize the potential of young people [5]. These programs should be developed in light of different cultural and family dynamics, as well as in line with the available healthcare resources (i.e. healthcare professionals involved, healthcare organization).

Studies that have assessed the effectiveness of transition interventions have mostly been conducted on a single CC, the majority of these including patients with type 1 diabetes mellitus [128-130]. These interventions have shown to have positive effects over clinical outcomes, such as HbA1c [130-132], glycemic control [130, 131], graft success [133] remission rates [134], adherence [135], appointment attendance in adult care [130, 135-137], length of stay [130, 132], follow-up [138] and re-admissions [130]. Improvements in patient-reported outcomes have been found in self-efficacy [139], health behaviors [136], well-being [140], autonomy [140], disease-related knowledge [130, 141, 142], self-management [141, 142], transition readiness [130, 143] and QoL [144].

However, when assessing the outcomes that previous transition programs have evaluated, the studies focused more on health outcomes rather than outcomes that reflect a holistic definition of transition (i.e. one that addresses medical, psychosocial and vocational issues) [131]. Consequently, more research that measures psychosocial aspects is needed, especially of outcomes that have also been associated with improvements in QoL and health status, such as patient empowerment [28, 40, 49].

Inadequate transition planning has negative outcomes. Patients may be lost to follow-up, have an increased financial and emotional burden and develop comorbidities [145], as well as higher rates of healthcare use, poor clinical outcomes and low patient satisfaction [146]. Lack of transition preparation places the young persons at a risk for lower than expected health literacy, delays in securing specialty care, problems with treatment adherence and excess morbidity and even mortality [130].

Adolescents' transition experiences and needs

Research investigating adolescents' experiences and needs during the transition process has found that some young persons did not have enough time to develop self-care and self-management skills [147, 148]. These were important for them to feel confident in assuming responsibility for their care or communicating directly with the healthcare provider. The adolescents' level of involvement varied; while some had already started to assume some responsibility and could ask the healthcare professional questions, others continued to rely on their parents and were concerned about the shift in responsibility [8, 148, 149]. Some adolescents were of the opinion that they should start gradually taking on responsibility from the age of 15 years [7].

For some young people, having their parents involved during transition was important, because they provided support and a sense of security. Parents have been shown to provide emotional support, medication and appointment reminders, provide advice on medical and non-medical decisions and transportation [150]. While the shift in responsibility is inevitable, providing young people with information earlier and giving them time to decide and assume responsibility in a step by step process is needed to improve the experience of transition [151, 152].

Some young persons suggested that being given time alone with the healthcare providers at an early stage is a good first step towards the adolescents actively participating in their own care [7]. Having the healthcare professional address the young person directly was also a strategy suggested to increase involvement [101]. As the burden of responsibility for disease management differs between pediatric and adult care, adolescents need to get used to healthcare providers expecting to meet the young person alone being capable of answering for themselves.

The importance of patient empowerment during the transition to adulthood

Adolescence is a time when psychosocial skills are developed that will help the individual in adulthood. It is therefore not surprising that adolescence research has previously touched on the concept of empowerment. Within this research field (not focusing on those with CCs or in a healthcare context), the concept is known as youth empowerment.

Youth empowerment has two perspectives. The first mostly focuses on the actions that young people take in order to create change in organizational and societal policies, values and structures. In this context, empowering adolescents should lead to individuals who are critical thinkers, who actively participate in day-to-day activities and who are involved in building communities that are more equitable [153]. The second perspective focuses on increasing empowerment in order to provide adolescents with the skills necessary to decrease the effects of multiple risk factors. This perspective aims to increase a range of potential protective factors that will help them achieve better well-being [154].

Previous youth empowerment interventions have found improvements in social skills [154-156], coping skills [154] and problem behaviors [154]. Additionally, youth empowerment is also associated with developmental outcomes, including increased self-efficacy, self-awareness, positive identity development, positive social bonding, awareness of organizational operations and interpersonal relations and a sense of purpose [153]. Empowerment is also a concept associated with interpersonal and community outcomes, such as improved relationships with adults, political efficacy and group engagement [153, 157].

As the previous information shows, empowerment has been applied in a context outside of healthcare. It is therefore possible to argue that patient empowerment, when implemented in a healthcare context, has potential ramifications outside of the health/illness perspective and associates with the developmental stage of adolescence, leading to better developmental outcomes. Thus, patient empowerment is a necessary aspect of the transition process.

Patient empowerment was introduced in transition literature as a way of increasing responsibility, accountability and self-determination [158]. Some authors indicate that transition needs to focus on empowering young people so they are capable of managing their own transition, self-managing their own condition and having an active role as an independent adult in the adult healthcare system [159-161]. Hait [162] stated that the necessary traits for a successful transition are empowerment, self-efficacy and self-determination.

For adolescents, being empowered is an important factor in their recovery and entails being listened to, being understood, taking control and making decisions for themselves [163]. Moreover, young persons have stated they feel empowered when they have the opportunity to see the healthcare providers by themselves and when they can communicate comfortably [160, 164]. Adolescents experience empowerment in different ways, which is partly influenced by their relationships with adults and their expectations from the adolescents. Young people have said that their power to act is limited by the context and that their decisions are only acceptable when they are in line with the ones preferred by the adults [165]. These experiences are important to consider because they are incongruent with what one would expect from an empowering experience. Adolescents have expressed uncertainty around their skill level and are in need of better transition preparation that allows them to manage their disease better [102, 166]. Patient empowerment is a possible solution to this issue.

Patient empowerment and other transition outcomes

Figure 1 (page 20) shows patient empowerment is a precursor for improvements in other patient-reported outcomes. The construct is associated with assuming more responsibility, becoming more autonomous and increased patient participation. It is therefore possible to expect that empowerment is associated with other measured outcomes during transition, such as transition readiness, self-efficacy and self-management. Many of these outcomes have been significantly associated with empowerment in other contexts (i.e. not transition related) [39, 153]. For instance, a cross-sectional study that included young people found that empowerment mediated the relationship between psychological factors and well-being, mental health and recovery [37].

Studies have identified low health literacy in adolescents with CCs [167, 168]. This low health literacy can have a negative impact on health behaviors and health outcomes. Empowered adolescents are well informed about aspects related to illness, treatment, education, employment and risk factors, among others. Patients with more knowledge can feel more comfortable participating in the shared decision-making process, are expected to have healthier behaviors, improved health outcomes and are capable of navigating the healthcare system [36, 166, 169]. As a result, improvements in the previously mentioned variables are also expected. Considering that empowerment includes changes to self, one could possibly expect that for adolescents struggling with their sense of identity and their illness, becoming empowered, will enable them to accept their disease and view it in a more positive light.

RATIONALE

According to Meleis [13], nurses are in charge of assisting individuals and families so that they feel capable of caring for themselves and in empowering them with "*knowledge and experience to care for themselves and to manage their symptoms and their life transitions by fully utilizing available resources and creating new resources*". For this reason, patient empowerment is a central concept in nursing science.

Research in the field of patient empowerment is mostly theoretical in nature, with limited empirical evidence having tried to advance this field [30, 157]. Theoretical papers have made an important contribution in trying to explain the construct of empowerment, its antecedents and consequences. However, more work still needs to be done to bring conceptual clarity and further empirical evidence that evaluates aspects of the available models of empowerment. Patient empowerment is understood as a process, an outcome or both and the definition used in this thesis interprets as having both. However, this doctoral thesis primarily focuses on patient empowerment as an outcome (i.e. a measurable construct).

In order to delimit the research field of this thesis, its focus is on patient empowerment in the context of transition to adulthood and adult care of young persons with CCs. Patient empowerment is an important aspect of transition that has not been investigated thoroughly in the group of young persons with CCs. While it is theoretically sound to have patient empowerment as a component of transitional care, there is little to no evidence that supports this claim. So far, there is no evidence about the level of empowerment of young persons, its association with other transition-related outcomes or even the direction of effects between patient empowerment and other patient-reported outcomes. Thus, the four studies included in this doctoral thesis aim to contribute with empirically based knowledge that will help in the process of understanding the boundaries of patient empowerment and its relevance within the field of adolescent health and care.

AIM

Overall aim

The overall aim of this thesis was to explore patient empowerment in young persons with chronic conditions during the transition to adulthood.

Specific aims

Study I	(i) To inventory the definitions and measurements of patient empowerment; (ii) to appraise the conceptual and methodological rigor of published studies; and (iii) to identify correlates of patient empowerment in persons with chronic conditions
Study II	To describe the development and psychometric evaluation of the Gothenburg Young Persons Empowerment Scale (GYPES)
Study III	To measure the level of empowerment in young persons with congenital heart disease and to examine its correlates
Study IV	To examine the direction of effects between patient empowerment and other patient-reported outcomes

METHODS

The present thesis comprises four papers that consider individuals with CCs, with the majority of the studies including young persons with CHD. The studies cover a range of methods and statistical analyses that are described in Table 2 and in the next sections.

Table 2. Overview of included studies

Studies	Study design	Sample	Data analysis
I	Descriptive review	Persons with CCs	Descriptive statistics, summative content analysis
II	Cross-sectional study	Young persons (14-25 years) with CHD and T1DM	Cognitive interviews, confirmatory factor analysis and internal consistency coefficients
III	Cross-sectional study	Young persons (14-18 years) with CHD	Univariate and multivariate linear regression analyses
IV	Longitudinal study	Young persons (16-17 years) with CHD	Paired sample t-tests and cross-lagged analyses

Description of study designs

This doctoral thesis involves three types of study design: a descriptive review, a cross-sectional study and a longitudinal study. These three designs are briefly described in the next section, with the aim of providing information related to the qualities of each design, possible output and scientific relevance.

Descriptive reviews

This type of review aims to determine the extent to which a set of empirical studies in a specific field supports any patterns to previous theories, methodologies or findings [170]. They involve structured and systematic search methods, they have a broad scope of questions, quality appraisal of the included studies is not necessary, and data is usually synthesized through content analysis and/or frequency analysis [170].

A descriptive review was included as part of the present doctoral thesis (Study I) because it allows us to describe the chosen research area, identify patterns in the evidence, question the available literature and provide specific research questions that need to be addressed in the future.

Cross-sectional study designs

Of the included studies in this doctoral thesis, two used a cross-sectional study design (Study II and III). This type of study collects data at one time point or over a short period. They provide a "snapshot" of the outcome and variables of interest at a particular point in time and they usually have a descriptive focus. However, they can also be the first step in evaluating the associations between variables before starting a longitudinal study [171]. These studies are a good opportunity to gain a broad base of knowledge from the variables of interest [172].

By using this study design, it was possible to evaluate the psychometric properties of an instrument to measure patient empowerment (Study II) and it was also possible to identify potential correlates of this construct (Study III). For these purposes, the data was analyzed by structural equation modeling (SEM) (Study II) and regression analyses (Study III), respectively. Regarding the identification of potential correlates in Study III, it is important to remember that cross-sectional data does not allow us to determine which variable is having an effect on another (i.e. predicting the value of another variable). Cross-sectional study designs only allow us to establish correlations or associations between variables but causal inference is not possible [172].

Longitudinal study designs

In Study III, potential correlates of patient empowerment were identified by implementing a cross-sectional study design. However, in light of these studies' limitations when it comes to establishing the predictive value of the variables, a longitudinal study was necessary in order to analyze these correlations further (Study IV).

A longitudinal study involves following participants over time and undertaking continuous or repeated measurements of the variables of interest. Lavrakas [173] states that the significance of this type of study "*stems from the fact that the knowledge, skills, attitudes, perceptions, and behaviors of individual subjects usually develop, grow, and change in essential ways over a period of time*". Through these studies it is possible to establish a sequence of events, follow change over time, correct for the "cohort effect" and identify the direction of effects between variables [171].

When analyzing longitudinal data, there are four important considerations: 1) the linked nature of the data for each participant; 2) the presence of fixed and dynamic variables; 3) potential for differences in time intervals between data instances; and 4) the presence of missing data [171]. Statistical analyses therefore need to account for these aspects and this done upon undertaking the SEM in Study IV.

Included studies

Study I

The purpose of this study was: (i) to inventory the definitions and measurements of patient empowerment; (ii) to appraise the conceptual and methodological rigor of published studies; and (iii) to identify correlates of patient empowerment in persons with CCs.

Study design

To achieve the aforementioned aim, a descriptive review was undertaken. A systematic search was performed in PubMed, CINAHL, Scopus and PsycInfo. In order to identify all possible relevant articles, no limits on publications dates were used during the search.

Eligibility criteria

Publications were included based on their study population and study design. The population of interest was persons with CCs of all ages. Given that the review aims to identify correlates of patient empowerment, only quantitative study designs were included, independently of whether they use an experimental, quasi-experimental or non-experimental design. Publications also needed to have been written in English, Swedish or Spanish.

Review criteria

The researchers developed seven review criteria to appraise the conceptual and methodological rigor of the included studies. These criteria are based on the criteria developed by Gill and Feinstein [174] for QoL and adapted for patient empowerment. The criteria include aspects related to the definition of patient empowerment used by the articles, the domains/dimensions that comprise the construct, the instrument(s) used to measure patient empowerment, a consistent use of the construct and the type of scoring. Additional information on the review criteria is included in the manuscript of Study I.

Study selection process and data extraction

Publications were included after a two-step selection process. The first step involved revising titles and abstracts by using EndNote software. The second step was the revision of the full-text articles. Both steps were undertaken by the author of this thesis.

Once articles were selected, the data extraction was performed by filling out a form in a Microsoft Access Database. The database was pilot-tested by all the researchers involved before beginning the formal process of data extraction. Data retrieved from the articles included: year of publication, country where the study was performed, study design (classified according to the Joanna Briggs Institute [175]), definition of empowerment, dimensions or domains of patient empowerment, study aim, instrument used to measure patient empowerment, sample size, age range, study duration, type of CC, and correlates assessed. Data extraction was undertaken by all the researchers involved in the study.

Data analysis

Descriptive statistics were used to summarize the studies' characteristics. To indicate how well the articles fulfilled the review criteria a summary score was calculated. This summary score was calculated by adding up the criteria each individual article fulfilled and dividing it by the number of criteria that were applicable for that article. The scores were multiplied by a 100, so the scores range between 0-100.

The identified correlates were categorized in order to determine trends in the included studies and facilitate understanding of the aspects covered by the available evidence. This categorization was done through summative content analysis [176].

Study II

The objective of this study was to assess the psychometric properties of the Gothenburg Young Persons Empowerment Scale (GYPES).

Development of GYPES

GYPES was developed in line with the theoretical foundations of Small and colleagues [4]. The development of the scale consisted of three stages. Stage one involved dividing the researchers into two groups. Each group created potential items based on literature and existing instruments and the other created items based on the definitions of the dimensions of empowerment and on clinical experiences. All items were formulated in English. Stage two involved combining all potential items (n=44) and selecting three items that were representative for each dimension. Three items per dimension were considered appropriate in order to avoid response burden from future respondents. In stage three the selected items (n=15) were translated from English to Swedish. A forward translation technique was conducted by the two research members whose first language is Swedish. This process led to having an English and Swedish version of the GYPES.

Evaluation of GYPES

The evaluation involved three phases. Phase 1 included an assessment of content and face validity and Phases 2 and 3 were undertaken to assess the scale's content validity, factorial validity, internal consistency and responsiveness.

Phase I

Sample

Cognitive interviews were conducted in two pediatric cardiology outpatient units and one pediatric outpatient diabetes unit in Sweden [177]. To participate in the interviews, the young people had to be between the ages of 14-19 years and had a scheduled appointment in one of the selected units during the period the interviews were held (November-December 2015). The total sample included 9 young persons (6 patients with CHD, 3 patients with type 1 diabetes mellitus).

Data collection

Participants were asked to respond the scale along with one of the researchers and provide feedback on the comprehensibility and interpretability of the items and response options. They were also asked to provide suggestions that could improve the scale.

Analysis

The participants' responses and suggestions were discussed by the interviewers and the researchers. Items were rephrased if the adolescents expressed difficulties in understanding an item.

Phase II

Study design and sample

This study used a cross-sectional study design and was part of the STEPSTONES project (Swedish Transition Effects Project Supporting Teenagers with chrONic mEdical conditionS). Participants were selected from the Swedish Registry of Congenital Heart Disease [178]. Eligible participants had to fulfill the following criteria: 1) had a confirmed diagnosis of CHD; 2) were between 14-18 years; and 3) were receiving follow-up at one of the participating pediatric cardiology outpatient centers in Sweden [177]. Patients were excluded if they had a cognitive and/or physical disability that did not allow them to answer the questionnaire, had undergone heart transplantation or had not provided assent to participate.

Five hundred ninety-three patients met the inclusion criteria and were sent a set of questionnaires. Of the 593, 202 patients returned the completed questionnaires, corresponding to a response rate of 34.1%. The mean age of the participants was 15.7±1.2 years and 45% were girls. Demographic characteristics of the sample are provided in Table 3.

Table 3. Sample characteristics of included studies

	Study II		Study III	Study IV
	Phase II	*Phase III*		
Sample size	202	273	202	132
Sex (%)				
Male	111 (55)	60 (22.0)	111 (55)	80 (60.6)
Female	91 (45)	213 (78.0)	91 (45)	52 (39.4)
Mean age (±SD)	15.7 (1.2)	19.9 (3.7)	15.7 (1.2)	NA
Education (%)				
Junior high school	116 (57.4)	42 (15.4)	116 (57.4)	NA
Senior high school	86 (42.6)	121 (44.3)	86 (42.6)	NA
College/University	0	110 (40.3)	0	NA
Disease complexity				
Mild	63 (31.2)	NA	63 (31.2)	17 (13.2)
Moderate	84 (41.6)	NA	84 (41.6)	80 (62.0)
Severe	55 (27.2)	NA	55 (27.2)	32 (24.8)

NA: non-applicable

Procedure

Each potential participant received a package containing information related to the study, an informed consent, a set of questionnaires and a pre-addressed envelope. Participants were asked to return the complete questionnaires and sign the informed consent in the pre-addressed envelope. To minimize non-response, a modified Dillman procedure was used [179]. Three weeks after dispatch of the documents, non-respondents were sent a personalized reminder letter. After five weeks, a second reminder was sent as well as a new set of questionnaires. The last reminder was sent after seven weeks. Persistent non-responders were contacted by telephone and asked if they had received the questionnaires, if they wanted to participate and if they needed a new set of questionnaires. Data collection for this study ran from January 25th until August 31st 2016.

Data analysis and description of statistical methods

The psychometric evaluation of GYPES was based on content validity, factorial validity, internal consistency and responsiveness. To assess content validity, the proportion of missing values and invalid scores was calculated. Factorial validity was evaluated through confirmatory factor analysis (CFA). Reliability was evaluated by calculating the Cronbach's alpha coefficient as a measure of internal consistency.

IBM SPSS Statistics for Windows version 22 was used to calculate the Cronbach's alpha coefficient, floor and ceiling effects and the proportion of missing values and invalid scores. To assess the factor structure, Mplus version 7 software was used. Once the psychometric properties were assessed, the scale was revised and tested again in Phase III.

Factorial validity: Confirmatory factor analysis

CFA is part of the SEM analysis and aims to test an a-priori model that indicates the associations between observed variables and underlying factors or theoretical constructs [180]. CFA can be used for different purposes; in this paper, it was used to confirm the factor structure and quality of GYPES [180]. CFA models comprised observed variables and latent factors.

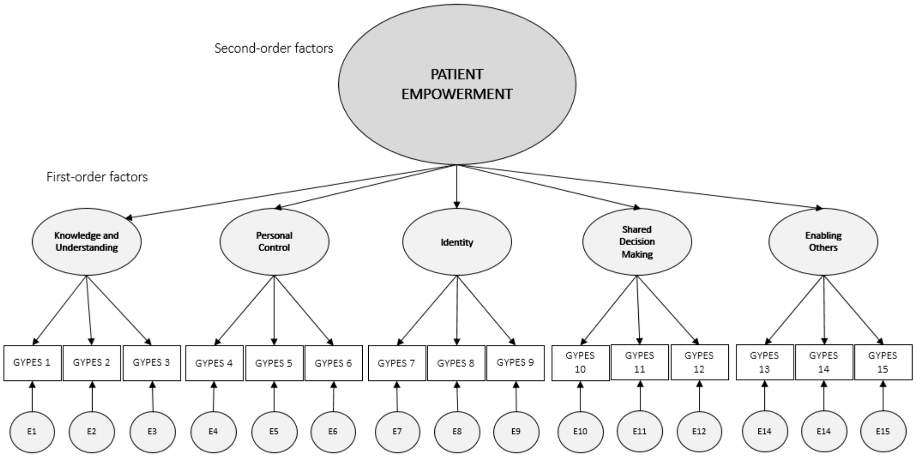


Figure 2. Model tested through confirmatory factor analysis.

As shown in Figure 2, the observed variables in this study are the items included in the scale and the latent factors are the dimensions, which comprise the items, as well as the overall construct of patient empowerment. The dimensions (e.g. knowledge and understanding, personal control, identity, shared decision-making and enabling others) are first-order factors. The second factor (e.g. patient empowerment) is considered a second-order factor. This entails that the construct of patient empowerment causes the first-order factors and can only be measured indirectly through the observed variables of these [181].

The output of CFA includes path coefficients (standardized regression weights), model fit, error variances, latent variable variances and modification indices. Path coefficients are also known as factor loadings and it is expected that they are large, statistically significant and in the right direction [182]. Factor loadings serve to identify whether a specific item (i.e. observed variable) is problematic and whether it may be necessary to replace, revise or eliminate it. Modification indices are statistics that provide information on how the model fit can be improved by allowing additional parameters to be estimated [180]. However, any changes to the model should be theoretically justified [183].

The model fit of SEM models is assessed with different indices. The most frequently reported indices are the chi-square goodness of fit (x^2), root mean square error of approximation (RMSEA), standardized root mean square residual (SRMR) and comparative fit index (CFI). The chi-square goodness of fit (x^2) is expected to be non-significant ($p \geq 0.05$). The RMSEA and SRMR are absolute fit indices that estimate how well the pre-established model reproduces the data. A RMSEA ≤ 0.06 and a SRMR ≤ 0.08 are desirable [184]. The CFI is a comparative index and compares the chi-square value of the hypothesized model with the chi-square of null model, where all variables are uncorrelated [182]. A larger CFI is desirable.

Reliability: Cronbach's alpha

Reliability was assessed with a measure of internal consistency. Internal consistency refers to the degree to which the items of an instrument are correlated with each other and thus measure the same underlying construct [182, 185]. This measurement is important in instruments that intend to measure underlying constructs, such as the dimensions comprised in GYPES and the overall construct of patient empowerment.

In this study, internal consistency was evaluated through the Cronbach's alpha coefficient, which is considered an adequate measure. A low Cronbach's alpha is an indicator of a lack of correlation among the items, whereas a high coefficient indicates the opposite [185]. The suitable Cronbach's alpha ranges between 0.70-0.95. This coefficient was calculated for each of the subscales and for the overall scale.

Responsiveness: floor and ceiling effects

Responsiveness is the ability of a questionnaire to detect important changes over time [185]. Usually, this psychometric property is best measured with longitudinal data. However, with cross-sectional data is it possible to calculate floor and ceiling effects, which are indicators of limited responsiveness, as they show that changes cannot be measured. Floor and ceiling effects were present if more than 15% of the participants had the lowest or highest score [185].

Phase III

Study design and sample

A cross-sectional study was conducted in young persons with type 1 diabetes mellitus as part of the "Better Transition in Diabetes project". Eligible participants had to be members of the Dutch Diabetes Association and Stichting èèndiabetes (a Dutch foundation for young persons with type 1 diabetes mellitus). Participants also had to be between the ages of 12-25. A total of 273 young people took part in the study. Demographic characteristics are shown in Table 3. The mean age of the participants was 19.9±3.7 years and 22% were boys.

Procedure

The patient organizations disseminated information about the study through their networks and posted a call on their Facebook pages. The Dutch Diabetes Association also sent an email to its members and a reminder two weeks later. Participants responded to the questionnaires using a web-based system and to encourage participation, five €50 gift vouchers were put to be won in a raffle for the respondents. Data collection started mid-June and lasted until mid-September 2016.

Scale translation

The revised version of GYPES was translated from English to Dutch following a forward-backward translation process, which involved two translators (one bilingual in Dutch and English) [186]. The translated version was tested for face validity with two mothers and a 16 year-old adolescent with epilepsy.

Statistical analyses

The same statistical analyses as in Phase II were used (pages 44-47). The scale's final version is provided in Appendix I.

Study III

This study aimed to measure the level of empowerment in young persons with CHD and to examine its correlates. To achieve this aim, data from the cross-sectional study described in Study II, Phase II was used (pages 43-44). Information related to the study design, setting, sample and data collection is therefore the same as the one described above.

Measurements: outcome variable

The outcome variable (i.e. patient empowerment) was measured using the GYPES. As previously mentioned, the scale allows calculation of both a total score and subscale scores for each dimension. The total score goes from 15 to 75, with higher scores indicating a higher level of empowerment. In this study, version I of Study II was used. During the psychometric analyses, this version had one item in the identity dimension with a low factor loading (0.185). To account for this, weighted scores were calculated for both the total and subscale scores. These were obtained by multiplying the scale scores by the respective factor loadings. This approach acknowledges the relative contribution of each item to the scale score.

Measurements: potential correlates

Participants' age and sex were retrieved from the background information questionnaire. CHD complexity was obtained from medical records and classified according to Task force 1 of the 32nd Bethesda Conference [88] as mild, moderate or severe.

QoL is understood as the "*degree of overall life satisfaction that is positively or negatively influenced by the individuals' perception of certain aspects of life important to them, including matters both related and unrelated to* health" [187]. QoL was measured with a 10 cm linear analog scale from 0 (worse imaginable QoL) to 100 (best imaginable QoL). Participants were asked to rate their overall QoL by marking whichever point on the scale indicates their QoL in their opinion. A linear analog scale has previously been used to measure QoL and shown to be valid, reliable and responsive [188].

Health behaviors are "*activities a person undertakes to maintain or improve health and prevent diseases*" [189]. This variable was measured using the Health Behavior Scale-CHD, which assesses alcohol consumption, tobacco use, dental care and physical activity. The questionnaire includes 15 items that allow the calculation of a substance use score (0-100), dental hygiene score (0-100) and total health risk score (0-100). A higher score indicates engaging in riskier behaviors [189]. Only the total health risk score was included in the analyses.

Illness perceptions are "*mental representations and personal ideas that people have about an illness*" [190] and these were measured using the Brief-Illness Perception Questionnaire (Brief-IPQ) [190]. The questionnaire includes 9 items that assess cognitive illness representations (consequences, timeline, personal control, treatment control and identity), emotional representations (concern and emotions) and illness comprehensibility. The last item in the scale assesses causal representation with an open-ended response that asks about possible causal factors in the illness. The Brief-IPQ allows the calculation of subscale scores and a total score. The total score was used in this study, with a higher score indicating a worse perception of the illness.

Knowledge about CHD was measured with the Knowledge Scale for Adults with Congenitally Malformed Hearts (Kno-CoMH) [191]. The scale includes 19 items that measure four dimensions: 1) general knowledge; 2) medical treatment; 3) endocarditis prophylaxis; and 4) contraceptives and pregnancy. A higher score indicates more knowledge in that particular dimension. General knowledge was the dimension included in the analyses.

Patient-reported health is a multidimensional construct conceptualized into domains of physical, emotional and social functioning and well-being. To measure this variable, participants were asked to answer the Pediatric Quality of Life Inventory 4.0 (PedsQL 4.0). The PedsQL 4.0 offers a generic and cardiac module, both of which were included in the study [192, 193]. The generic module has 23 items that cover the domains of patient-reported health. The scale allows the calculation of four subscores: 1) physical functioning; 2) emotional functioning; 3) social functioning; and 4) school functioning. Additionally, it is possible to calculate a psychosocial health summary, a physical health summary and a patient-reported summary score [192]. The latter score is the one included in the analyses. Higher scores indicate better patient-reported health. The PedsQl 4.0 cardiac module has 27 items that covers six scales: 1) heart problems and treatment; 2) treatment; 3) perceived physical appearance; 4) treatment anxiety; 5) cognitive problems; and

6) communication with the healthcare provider and others. The cardiac module does not include a total score. Higher scores in the scales indicate less problems [193]. All the scales, except for treatment, were included in this study. This scale was excluded because the majority of the patients are not under a pharmacological treatment regime.

Transition readiness is understood as the "*adolescent's readiness to assume complete responsibility for their healthcare and their readiness to transfer to adult medical care*" [194]. To measure this variable, the Readiness for Transition Questionnaire (RQT) was used [194]. The RTQ includes 20 items, of which only two were used in the present study. These items are: "*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 transition from pediatric care to adult care?*" [194]. The items are measured using a 4-point Likert scale (not at all ready, somewhat ready, mostly ready and completely ready). The score ranges from 2 to 8 and a higher score indicates a higher perceived readiness for healthcare responsibility and readiness for transfer to adult care. The RTQ overall score is the one included in this study.

Information regarding the validity and reliability of the aforementioned questionnaires can be found in Appendix II.

Data analysis and description of statistical methods

Statistical analyses were performed using IBM SPSS Statistics for Windows version 22 (Armonk, NY: IBM Corp). Descriptive statistics were expressed in absolute numbers, percentages, means and standard deviations. To assess potential correlates of patient empowerment, univariate and multivariate linear regression analyses (enter method) were performed. All tests were two-sided and significance level was established at $p<0.05$.

Linear regression

Linear regression analyses are done to judge the magnitude and quality of the relationship between a continuous dependent variable and one (univariate linear regression) or more independent variables (multivariate linear regression) [195]. When including several independent variables (i.e. multivariate), the researcher is making inferences about the effect of a specific variable while controlling the effect of the remaining variables in the model.

When undertaking this type of analysis, there are certain assumptions that must be evaluated and met to ensure the results are valid [195]. The first assumption is that there is independence of observations, which is checked with the Durbin-Watson statistic. The second assumption is that there needs to be a linear relationship between the dependent variable and the independent variables. The linear relationship can be evaluated by inspecting a scatterplot or a partial regression plot; if the relationship is not linear, the data needs to be transformed. The third assumption is that the data needs to show homoscedasticity, which is done by plotting the studentized residuals (i.e. errors) against the unstandardized predicted values. Homoscedasticity is related to the variance of the dependent and independent variables and how this should be the same across all the values of the plot. The fourth assumption relates to multicollinearity. Independent variables in linear regressions should not be highly correlated or the estimates will not be as precise [195]. These correlations can be assessed by calculating the Tolerance/Variance inflation factor. The fifth assumption is that there should not be significant outliers, as these can cause the model to be biased. The sixth and final assumption is that the residuals are approximately normally distributed. This is done by inspecting a normal P-P plot or normal Q-Q plot of the studentized residuals [195, 196].

The coefficient β and its significance level are the values reported when undertaking linear regression analyses. This coefficient is the "*expected change in outcome if X changes by one unit and all other variables are held constant*" [197]. The coefficient β also indicates the direction of the effect.

Study IV

This study aimed to assess the direction of effects between patient empowerment and other patient-reported outcomes.

Study design and sample

This study uses data from a larger study that aims to evaluate the effectiveness of a transition program to empower young persons with CHD. The study design involves a randomized controlled trial embedded in a longitudinal, observational study and includes participants from seven pediatric cardiology outpatient centers. The randomized controlled study included patients from two centers. Participants at these centers were randomly assigned to either an intervention group (i.e. transition program) or a comparison group (i.e. usual care). The longitudinal, observational study included five centers whose participants served as part of the control group (i.e. usual care).

Participants across the seven centers were followed for a period of two and a half years and were asked to answer a set of questionnaires at three different time points (baseline-T0, follow-up 1-T1 and follow-up 2-T2). Given the purpose of Study IV, only data from the comparison and control groups were included in the analyses. Data from the intervention group was excluded because the transition program aims to empower adolescents with CHD and the data is influenced by the effects of the intervention. Additionally, only data from T0 and T1 were included, as there were insufficient patients with data at T2 at the moment of the analyses. Additional information on the study design and the transition program can be found in a related article [198].

Participants were eligible if they: a) had a CHD diagnosis; b) were age 16; c) Swedish speaking; and 4) literate. They were excluded if they had a cognitive or physical impairment that prevented them from answering the questionnaires or had a heart transplantation [198]. Based on sample calculation for the larger study, 63 patients were needed in each group (i.e. intervention, comparison, control). To account for 10% dropout rate, 70 patients in each group were recruited, aiming for a total sample size of 210 young people [198]. For this study, the total sample comprised 140 patients. However, by the time the data analyses were performed, information on both T0 and T1, was only available for 132 patients.

Procedure

Data collection occurred when the participants were 16 and 17 years old. Eligible participants were sent a package containing information about the study, an informed consent, a set of questionnaires and a pre-addressed envelope. To minimize non-response, reminders were sent after 2, 4 and 6 weeks from when the package was originally sent. Reminders sent after 2 and 4 weeks involved a personal letter and a new set of questionnaires, respectively. The last reminder was done by phone and the young people were asked if they had received the questionnaires, were willing to participate and if they needed another set of questionnaires. In order to increase engagement, participants who answered the questionnaires received a voucher for the cinema.

Measurements

Patient empowerment was measured with the GYPES [177]. Information regarding this questionnaire is provided in the description of Study II (pages 42-48).

Patient-reported health, *communication skills*, *QoL* and *transition readiness* were measured with the PedsQL 4.0 (generic and cardiac)[192, 193], a linear analog scale [188] and the RTQ [194], respectively. As with GYPES, information about scoring and psychometric properties of the instruments are provided in the description of Study III (pages 48-50) and Appendix II.

Data analysis and description of statistical methods

All statistical analyses were performed using IBM SPSS Statistics version 25 and AMOS version 26. Descriptive statistics were expressed in absolute numbers, percentages, means and standard deviations. Paired sample t-tests were undertaken to compare the mean score differences between the two measurements. The direction of effects between patient empowerment and other patient reported outcomes was assessed with cross-lagged models.

Cross-lagged analysis

Cross-lagged analysis is used to test the effect two or more variables have on each other over time. They are considered "*crossed*" because they estimate the association from one variable to another and vice-versa. They are considered "*lagged*" due to the fact that they measure these relationships across different time points [199]. Cross-lagged analysis is "*widely used to examine the stability and relationships between variables over time to better understand how variables influence each other over time*" [199].

Figure 3 shows the cross-lagged model tested. In this model, patient empowerment can be considered as X0 and X1 being measured at T0 and T1 and variable Y0 and Y1 as the other patient-reported outcomes measured (e.g. patient-reported health, QoL, transition readiness, communication skills) also at those same time points. This model was replicated four times, because we individually assessed the direction of effects between patient empowerment and transition readiness, patient-reported health, communication skills and QoL.

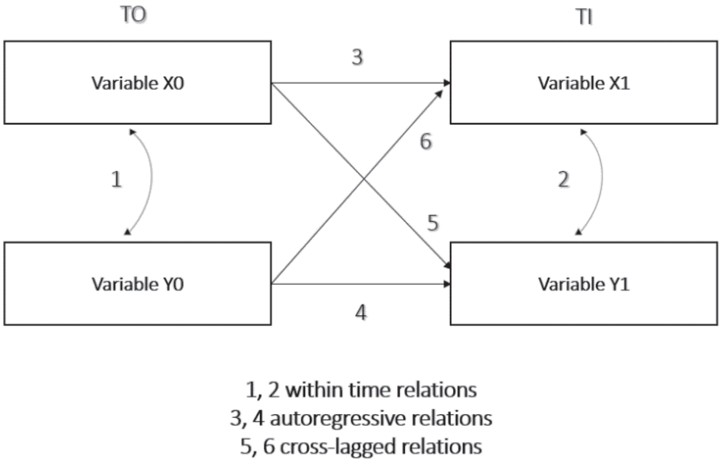


Figure 3. Example of a cross-lagged model.

As shown in Figure 3 the model includes within time relations (1 and 2), autoregressive relations (3 and 4) and cross-lagged relations (5 and 6). Autoregressive effects are the temporal stability of the construct over time. The closer the coefficient is to one, the more stable the rank order of the individual from one time point to the other [200]. A lower coefficient then indicates there is more variance and the construct is less stable. Including the autoregressive relations in the model is important because the variance of Y_2 predicted by X_1 is residual variance, as the levels of Y_1 are being controlled in the model [201, 202].

It is not possible to establish causality from cross-lagged models. In general, no statistical model can establish causality [202]. The results of cross-lagged analyses provide causal predominance, which means determining which variable influences another one without experiencing a reciprocal influence [199]. This causal predominance can be determined by inspecting the standardized coefficients of the cross-lagged paths. If the coefficient from X1 on Y2 is large and the effect of Y1 on X2 is small or zero, then X is predicting the effect of Y later on.

The output of a cross-lagged analysis includes the different coefficients from the different relations included in the model, their significance and model fit indices. The model fit indices reported are the same ones when determining model fit of a CFA model (See page 46 for information on the model fit indices). However, in this study the estimates calculated are based on a saturated model and no model fit statistics are offered, as a saturated model gives a perfect model fit [181]. All tests were two-sided and significance level was established at $p<0.05$. Missing data was managed through Full Information Maximum Likelihood method [180].

Ethical considerations

All the studies included in the thesis are compliant with the ethical principles of the Declaration of Helsinki from the World Medical Association [203]. According to this document, medical research should protect life, health, dignity and integrity [203]. Special precautions should be taken to protect the privacy and confidentiality of the research participants' personal information. To ensure confidentiality of the participants, a study identification number was given to each one. Participants' personal data and study-related information is stored in locked cabinets and digital documents are password protected and kept on a secure server.

Ethical approval was received for Studies II (phase I and II)-IV from the Regional Ethical Review Board in Gothenburg (Diary no. 953 13 and 931 15) (See Appendix III). Given that Study I is a descriptive review, there was no need for an ethical application. Phase III of Study II was undertaken in The Netherlands, a country where there is no need for ethical approval when undertaking studies that do not include treatment of patients or do not evaluate related healthcare [204]. For Studies II (phase II)-IV, an information letter with details about the project (e.g. purpose, confidentiality, potential risks), a written consent and a set of pre-coded questionnaires was sent to each eligible participant. Contact details of the principal investigator and project coordinator were also included in the project information, in case eligible participants had any questions regarding the project. To ensure that participants would not send the questionnaires together with the informed consent they received two pre-addressed envelopes, one for the questionnaires and one for the informed consent.

Patients included in the studies were between the ages of 14-25. According to Swedish regulations, young persons between the ages of 15-18 can assent to participate in studies without requiring parental approval [205]. Assent is the principal requirement for pediatric research and a young person's refusal to participate should be respected [206]. Even when the young people were asked to provide assent, an informed consent form was included in the information package for the guardians to approve their child's participation.

Participants in Studies II (phase II)-IV were offered a cinema ticket for every time they returned a set of questionnaires. This was offered as recognition for the contribution the participants made to the research outcomes [207]. Some researchers are against providing any sort of research participation incentives. This is because they think it may undermine the voluntary participation of the study and may compromise the scientific integrity of the research [207]. However, the participants in our study received a low-cost incentive and previous research has found that modest financial incentives do not negatively influence study participation [208].

The studies included are supported by funding from the Swedish Heart-Lung Foundation (grant 20150535), Swedish Research Council for Health, Working Life and Welfare-FORTE (STYA-2015/0003), Swedish Children Heart Association, Swedish Research Council (2015-02503), FNO (101.325) and the Diabetes Fund (2015.30.1852). Additionally, the doctoral position was funded by the Institute of Health and Care Sciences, Sahlgrenska Academy, University of Gothenburg.

RESULTS

An array of definitions and limited evidence on correlates of patient empowerment (Study I)

An initial literature search resulted in 11 142 publications; after removing duplicates, 9 349 were screened for eligibility. After screening titles and abstracts, 327 were read in full-text. A total of 76 articles were included in the analyses.

Study characteristics

The majority of studies were of a cross-sectional nature (n=53, 69.7%) and only 23 (30.3%) had a longitudinal or experimental study design. In regards to the CCs included in the studies, psychiatry (n=33) and endocrinology (n=20) corresponded to the two medical specialties with the highest number of publications. Within these specialties, schizophrenia, schizotypal and delusional disorders (e.g. schizophrenia, psychosis), mood and affective disorders (e.g. depression, bipolar disorder, affective disorder) and diabetes mellitus were the most frequent CCs. There were 28 studies that included more than one diagnostic group, but only six of these studies had included CCs from different medical specialties.

Conceptual and methodological rigor

Of the 76 included studies, 49 had included a definition for patient empowerment. The definitions corresponded to 29 different authors, as some of the authors proposed more than one definition for patient empowerment. The most frequent definitions used were those proposed by Rappaport (n=7) [209], Funnell and Anderson (n=7) [24], Zimmerman (n=6)[26] and Corrigan (n=6)[210].

In the included studies, 38 different instruments were used to measure patient empowerment. Eight studies measured empowerment with more than one instrument and seven used self-developed scales. Nevertheless, the majority of the studies used one of two instruments, the Making Decisions Empowerment Scale (MDES) developed by Rogers [211] and the Diabetes Empowerment Scale (DES) developed by Anderson and co-authors [212]. These scales were used in 24 and 19 studies, respectively. The MDES was developed for users of mental health services, whereas the DES was developed for those with diabetes mellitus.

The latter has been modified and it is used in other CCs, such as rheumatic and heart disease.

All the included articles were evaluated in light of the previously mentioned review criteria. The mean summary score was 63.38±22.59. The least fulfilled criteria were: 1) providing a reason for selecting a particular instrument (n=61); 2) providing domains/dimensions of patient empowerment (n=33); 3) providing a definition for patient empowerment (n=27); and 4) using the concept consistently (n=24).

Correlates of patient empowerment

Correlates covered by the included publications were collected in four categories: 1) sociodemographic characteristics; 2) clinical outcomes; 3) patient-reported outcomes; and 4) patient-reported experiences. Overall, over 50 different correlates were identified, with the majority falling under the category of patient-reported outcomes (n=181) and sociodemographics (n=122).

The most frequent studied associations were participants' sex (n=15), educational level (n=15), age (n=21), employment (n=15), psychiatric symptoms (n=12), self-care (n=18) and QoL (n=18). An analysis of the most frequent associations (i.e. investigated in more than five studies) showed that not all of them are significant. QoL, health status, stigma, self-efficacy and self-esteem were the variables that always had a significant association with patient empowerment.

GYPES: a valid and reliable measure of empowerment (Study II)

Phase I: Face and content validity

Cognitive interviews with the young persons with CHD and type 1 diabetes led to modification of the responses in the 5-point Likert scale (strongly disagree, disagree, neither agree nor disagree, agree and strongly agree) and the re-wording of one item in the "enabling others" dimension. This was done because the participants said that the response categories were slightly vague and confusing and two items in the "enabling others" dimension were difficult to differentiate.

Phase II: Content validity, factorial validity, internal consistency and responsiveness

The proportion of missing values in the cross-sectional study involving young persons with CHD ranged between 0.5 and 3.4, with the majority of missing items from the "shared decision-making" and "enabling others" dimensions.

GYPES's factorial validity was assessed through CFA. The model tested is the one shown in Figure 2 (page 45). This model showed adequate fit (*d*¦: 80; $\chi 2$: 154.948, $p<0.0001$; CFI: 0.916; RMSEA: 0.068; SRMR: 0.069). When examining the factor loadings of the individual items with their respective dimensions, item nine in the "identity" dimension ("*My condition does not stop me from living the life I want*") with a low factor loading (0.185, $p= 0.014$). However, the remaining factor loadings ranged from 0.475 to 0.892 (See Table 4). Another model that did not include item 9 was estimated and the model fit improved (*d*¦: 67; $\chi 2$: 128.464, $p<0.0001$; CFI: 0.930; RMSEA: 0.067; SRMR: 0.058). A second-order factor model was also evaluated but the model did not converge properly. This could have been related to the identity dimension only including two items.

Cronbach's alpha coefficients for the dimensions ranged between 0.521-0.751, while for the overall scale it was 0.819. None of the participants had the lowest possible score and only 1.5% scored the highest possible value (i.e. 75).

Based on the aforementioned results, GYPES was modified and evaluated again in Phase III. On statistical grounds, item 9 was rephrased (revised item "*I have given my condition a place in my heart*"). Additionally, to improve consistency and understanding, items 7 ("*My condition is a part of who I am*"), item 11 ("*I actively participate in discussions about my health*") and item 14 ("*I feel comfortable sharing with others about my condition*") were rephrased (revised items "*My condition is part of who I am as a person*"; "*I actively participate in discussions with my healthcare providers about my health*"; and "*I am able to give helpful advice to people who are struggling with their condition*").

Table 4. Factor loadings for both versions of GYPES

Factors and items	Phase II[a] Initial version of GYPES	Phase III[b] Final version of GYPES
Knowledge and Understanding		
1[cd]. I know and understand *my condition*	0.585	0.706
2[cd]. I know what to do to stay healthy	0.744	0.843
3[cd]. I know when to contact healthcare providers for *my condition*	0.531	0.517
Personal control		
4[cd]. I have the skills to manage *my condition* in daily life	0.475	0.762
5[cd]. I have a sense of control over my health	0.876	0.627
6[cd]. I am active in maintaining my health	0.621	0.385
Identity		
7[c]. M*y condition* is a part of who I am	0.590	
7[d]. *My condition* is a part of who I am as a person		0.406
8[cd]. Living with *my condition* makes me stronger as a person	0.892	0.541
9[c]. *My condition* does not stop me from living the life I want to live	0.185	
9[d]. I have given *my condition* a place in my heart		0.714
Decision making		
10[cd]. I am capable to express to my healthcare providers what is important to me	0.727	0.679
11[c]. I actively participate in discussions about my health	0.609	
11[d]. I actively participate in discussions with my health care providers about my health		0.869
12[cd]. I am capable to make decisions about my health and health care together with the healthcare providers	0.773	0.772
Enabling others		
13[cd]. I have the skills to support other young people with *my condition*	0.761	0.783
14[c]. I feel comfortable sharing with others about *my condition*	0.531	
14[d]. I am able to give advice to people who are struggling with their *condition*		0.941
15. I can help other people by sharing how I keep myself well	0.808	0.649

[a] Five factor model
[b] Five factor model with error correlations within factors
[c] Congenital heart disease
[d] Diabetes
All factor loadings were significant at p<0.0001, except for item 9 in Phase II (p=0.014)

Phase III: Content validity, factorial validity, internal consistency and responsiveness

There were no missing values for the GYPES items when evaluating the new version.

CFA evaluating the five-factor structure indicated an adequate model (*df*: 80; χ2: 235.375, p<0.0001; CFI: 0.897; RMSEA: 0.084; SRMR: 0.059). However, CFI did not reach the threshold and it was decided to revise the model according to a suggestion from the modification indices. The revised model included the same five-factor structure and an error correlation between items 6 and 15. This was the largest error correlation from the modification indices and while the two items belong to a different dimension, theoretically, it can be expected that individuals who are more actively involved in their care also feel more capable of sharing their experiences and coping strategies with other people. This new model had an adequate model fit across all indices (*df*: 79; χ2: 201.950, p<0.0001; CFI: 0.919; RMSEA: 0.076; SRMR: 0.057). The factor loading from this model ranged from 0.385 to 0.941 (see Table 4).

Lastly, a second-order factor model was also evaluated, which was found to have an adequate fit across all indices (*df*: 84; χ2: 222.788, p<0.0001; CFI: 0.908; RMSEA: 0.078; SRMR: 0.061). The factor loadings of each first-order factor in relation to the global empowerment factor had values exceeding 0.660 (knowledge and understanding= 0.729; shared decision-making= 0.660; identity= 0.777; personal control= 0.950; and enabling others= 0.617). These results support the calculation of an overall empowerment score as well as subscale scores.

This new version of GYPES had good internal consistency, with Cronbach's alpha ranging from 0.609 to 0.833 for the dimensions and 0.858 for the overall scale. Only 2.2% obtained the highest possible score and none of the participants had the lowest scoring.

Transition readiness and communication: significant correlates of empowerment (Study III)

Level of empowerment

The mean level of empowerment in the participants was of 54.5±10.5. The lowest subscale score was for "enabling others" (10.0±3.3), while personal control (12.6±2.4) had the highest score. For the remaining scales, the scores were 11.6±2.7 for knowledge and understanding, 10.8±3.1 for shared decision-making and 10.2±3.2 for identity.

Correlates of patient empowerment

When analyzing the association between patient empowerment and every potential correlate individually, the construct was associated with older age, better QoL, higher transition readiness, better health behaviors, a less threatening view of their illness and improvements in perceived physical appearance, treatment anxiety, cognitive problems, communication and patient-reported health (see Table 5). However, when including other variables in the model, only higher transition readiness and fewer problems communicating with the healthcare provider were still significantly associated when controlling for the other variables (see Table 5).

QoL, transition readiness, perceived physical appearance, treatment anxiety, cognitive problems, communication and patient-reported health, health behaviors, illness perception and heart problems were univariately associated with the scores of the dimensions of "knowledge and understanding", "personal control", and "shared decision-making". Additionally, age was also associated with "knowledge and understanding". The "enabling others" scores were correlated with CHD knowledge, transition readiness, perceived physical appearance, treatment anxiety, cognitive problems and patient-reported health. Scores for the "identity" dimension were not significantly correlated with any of the included variables (see Table 5).

Multivariate analyses indicated that transition readiness and communication were the variables still having a significant effect over the scores of "knowledge and understanding", "shared decision-making" and "enabling others". Moreover, age was significant for "knowledge and understanding", illness perceptions for "personal control" and heart problems for "enabling others" (see Table 5).

Table 5. Univariate and multivariate linear regression analyses

Correlates	Sex	Age	DC		QoL	KNO	RTQ	IP	HB	HP	PPA	TA	CP	COM	PRH
			Mild	Moderate											
GYPES (total score)															
Univariate															
B	0.40	0.85	-0.74	-0.05	0.09	0.21	1.56	-0.16	-0.10	0.05	0.07	0.07	0.07	0.13	0.14
(SE)	(0.98)	(0.42)	(1.29)	(1.21)	(0.03)	(0.13)	(0.25)	(0.04)	(0.05)	(0.03)	(0.02)	(0.02)	(0.02)	(0.02)	(0.03)
β	0.03	0.14	0.05	0.00	0.23	0.12	0.40	-0.28	-0.14	0.12	0.23	0.26	0.27	0.45	0.31
Multivariate															
B	-0.95	0.31	-0.59	0.61	0.02	0.09	1.05	-0.06	-0.04	-0.06	0.02	-0.01	0.02	0.10	-0.02
(SE)	(0.96)	(0.42)	(1.28)	(1.16)	(0.03)	(0.13)	(0.30)	(0.05)	(0.04)	(0.04)	(0.03)	(0.03)	(0.03)	(0.03)	(0.06)
β	-0.07	0.05	-0.04	0.05	0.04	0.05	0.28	-0.10	-0.06	-0.15	0.05	-0.04	0.08	0.36	-0.04
Knowledge and Understanding															
Univariate															
B	0.22	0.30	0.15	-0.04	0.02	0.03	0.43	-0.04	-0.03	0.01	0.01	0.02	0.02	0.03	0.03
(SE)	(0.24)	(0.01)	(0.31)	(0.29)	(0.01)	(0.03)	(0.06)	(0.01)	(0.01)	(0.01)	(0.01)	(0.01)	(0.00)	(0.01)	(0.01)
β	0.07	0.21	0.04	-0.01	0.19	0.07	0.47	-0.31	-0.20	0.15	0.21	0.23	0.26	0.41	0.29
Multivariate															
B	-0.02	0.25	0.19	0.07	0.00	0.00	0.27	-0.02	-0.02	-0.01	0.00	-0.00	0.01	0.02	-0.02
(SE)	(0.23)	(0.10)	(0.30)	(0.27)	(0.01)	(0.01)	(0.07)	(0.01)	(0.01)	(0.01)	(0.01)	(0.01)	(0.01)	(0.01)	(0.02)
β	-0.01	0.18	0.06	0.03	0.03	0.05	0.30	-0.17	-0.10	-0.06	0.00	-0.03	0.11	0.30	-0.14
Personal control															

Univariate															
B	0.11	0.11	0.39	0.26	0.03	0.02	0.31	-0.05	-0.03	0.03	0.02	0.02	0.02	0.03	0.05
(SE)	(0.23)	(0.01)	(0.30)	(0.28)	(0.01)	(0.03)	(0.06)	(0.01)	(0.01)	(0.01)	(0.01)	(0.00)	(0.00)	(0.01)	(0.01)
β	0.03	0.08	0.11	0.08	0.31	0.05	0.34	-0.38	-0.15	0.30	0.26	0.24	0.33	0.41	0.43
Multivariate															
B	-0.32	0.07	0.23	0.08	0.01	0.02	0.14	-0.03	-0.01	0.00	0.00	-0.01	0.01	0.01	0.01
(SE)	(0.24)	(0.10)	(0.31)	(0.28)	(0.01)	(0.03)	(0.08)	(0.01)	(0.01)	(0.01)	(0.01)	(0.01)	(0.01)	(0.01)	(0.02)
β	-0.10	0.05	0.07	0.03	0.10	0.05	0.15	-0.22	-0.08	0.04	0.00	-0.10	0.08	0.16	0.08
Identity															
Univariate															
B	-0.36	0.19	-0.98	-0.34	0.01	0.07	0.03	0.00	0.00	-0.01	0.01	0.01	-0.00	0.01	-0.00
(SE)	(0.28)	(0.11)	(0.36)	(0.34)	(0.01)	(0.04)	(0.08)	(0.01)	(0.01)	(0.01)	(0.01)	(0.01)	(0.01)	(0.01)	(0.01)
β	-0.09	0.12	-0.24	-0.09	0.04	0.12	0.03	-0.03	0.00	-0.04	0.06	0.12	-0.01	0.07	-0.01
Multivariate															
B	-0.46	0.05	-0.85	0.01	0.00	0.00	0.03	-0.00	0.00	-0.01	0.00	0.01	0.00	0.01	0.01
(SE)	(0.33)	(0.15)	(0.44)	(0.40)	(0.01)	(0.04)	(0.11)	(0.02)	(0.02)	(0.01)	(0.01)	(0.01)	(0.01)	(0.01)	(0.02)
β	-0.12	0.03	-0.21	0.00	0.00	0.01	0.03	-0.02	0.02	-0.10	0.04	0.06	0.00	0.09	0.05
Shared decision-making															
Univariate															
B	0.41	0.00	0.30	0.16	0.03	-0.01	0.47	-0.06	-0.04	0.02	0.02	0.02	0.03	0.04	0.04
(SE)	(0.32)	(0.14)	(0.42)	(0.40)	(0.01)	(0.04)	(0.08)	(0.01)	(0.02)	(0.01)	(0.01)	(0.01)	(0.01)	(0.01)	(0.01)
β	0.09	0.00	0.06	0.04	0.21	-0.01	0.38	-0.33	-0.19	0.17	0.22	0.22	0.29	0.47	0.29
Multivariate															
B	0.09	-0.08	0.21	0.31	0.00	0.00	0.35	-0.03	-0.02	-0.01	0.00	-0.01	0.01	0.04	-0.04
(SE)	(0.33)	(0.14)	(0.44)	(0.40)	(0.01)	(0.04)	(0.10)	(0.02)	(0.02)	(0.01)	(0.01)	(0.01)	(0.01)	(0.01)	(0.02)
β	0.02	-0.04	0.04	0.07	0.02	0.01	0.28	-0.17	-0.09	-0.07	0.03	-0.10	0.14	0.42	-0.24

Enabling others															
Univariate															
B	-0.20	0.12	-0.72	-0.36	0.02	0.10	0.31	-0.02	-0.02	-0.00	0.02	0.02	0.01	0.03	0.03
(SE)	(0.34)	(0.15)	(0.45)	(0.42)	(0.01)	(0.05)	(0.09)	(0.01)	(0.02)	(0.01)	(0.01)	(0.01)	(0.01)	(0.01)	(0.01)
β	-0.04	0.06	-0.14	-0.08	0.12	0.16	0.24	-0.11	-0.06	-0.02	0.15	0.18	0.15	0.32	0.16
Multivariate															
B	0.59	-0.05	-0.57	-0.06	-0.00	0.05	0.28	0.02	0.01	-0.03	0.01	-0.00	0.00	0.03	0.02
(SE)	(0.37)	(0.16)	(0.50)	(0.45)	(0.01)	(0.05)	(0.12)	(0.02)	(0.02)	(0.01)	(0.01)	(0.01)	(0.01)	(0.01)	(0.02)
β	-0.13	-0.02	-0.12	-0.01	-0.01	0.08	0.22	0.09	0.03	-0.24	0.10	-0.03	0.00	0.30	0.14

DC: disease complexity; QoL: quality of life; KNO: knowledge; RTQ: transition readiness; IP: illness perceptions; HB: health behaviors; HP: heart problems; PPA: perceived physical appearance; TA: treatment anxiety; CP: cognitive problems; COM: communication; PRH: patient-reported health

Level of significance			
NS	<0.05	<0.01	<0.001

Patient empowerment and its predictive value of communication skills (Study IV)

Changes in scores between data collections

There were changes in the mean scores of all the five variables. Communication skills, patient empowerment, patient-reported health and transition readiness increased between measurements whereas QoL decreased. However, these changes were only significant for patient-reported health (T0: 78.9, T1: 83.9, $p \leq 0.01$), QoL (T0: 82.6, T1: 76.9, $p \leq 0.01$) and transition readiness (T0: 5.1, T1: 5.6, $p \leq 0.01$).

Temporal relationships

Of the four models evaluated through cross-lagged analyses, three within time correlations were significant. One at T0 between patient empowerment and QoL and the association between patient empowerment and transition readiness was significant at both T0 and T1. All the autoregressive relations were significant in the four models, which means the participants rank order was stable across data collections.

Lastly, one cross-lagged path was found to be significant. Patient empowerment at T0 predicted communication skills at T1 ($\beta = 0.28$), meaning a higher level of empowerment at T0 was related to an increase in communication skills. Therefore, young persons with higher levels of empowerment had better communication skills at T1.

DISCUSSION

This thesis explored patient empowerment in young persons with CCs during the transition to adulthood. To the best of my knowledge, this is one of the first to focus on patient empowerment during this period of life. The findings indicate that patient empowerment has mainly been researched in adult participants and very little research is available that involves adolescents and young people. Given that most of the research has focused on adults, there were no available instruments that had been previously validated in young people. Additionally, most of the available instruments are meant to be used in specific CCs.

As previous theoretical models and authors have proposed, research suggests that patient empowerment is associated with patient-reported and clinical outcomes. Nevertheless, there is limited evidence from longitudinal studies that can help us better comprehend patient empowerment. This thesis provides results in relation to the longitudinal association between patient empowerment and other outcomes. More specifically, there was evidence that a higher level of patient empowerment was associated with improvements in communication skills. The findings will be discussed in more detail in the following section:

How do we define and measure patient empowerment?

From the available evidence, there is no consensus on a definition of patient empowerment. Study I shows that the literature uses a range of definitions (n=35). According to these definitions, patient empowerment is a multidimensional construct, associated with gaining control over one's life, a sense of agency, feeling competent, taking personal responsibility, establishing goals and feeling motivated. It is not only associated with the level of influence, choice and control in an individual context, but also within the community. Many of these defining characteristics are in line with what has previously been written on patient empowerment, and in fact with the definition used in this doctoral thesis. Even the most frequent definitions found in the articles by Rappaport [19], Corrigan [210], Zimmerman [26] and Anderson and Funnell [24, 212] have similarities with Small's definition [4]. However, unlike Small and colleagues' definition, theirs do not embrace the duality of the construct, recognize the role of the healthcare provider, and mostly focus on control. Additionally, not all the definitions include knowledge as an important aspect of patient empowerment, which is recurrently considered a vital component of this construct [25].

The identified definitions in Study I indicate that patient empowerment is understood as an outcome, a process or both, which is in line with the duality of the concept [20]. However, the different interpretations available complicate the comparability of results and raise the question of whether the available evidence is actually providing information on the same construct [213]. Across the literature, there are plenty of examples of a lack of consistent use of patient empowerment. This has been mentioned by previous authors [23] and is highlighted by the findings of Study I, which showed a percentage of the included articles intended to measure patient empowerment yet used other instruments that are related to other constructs, such as the Patient Activation Measure [214]. There were also articles that used patient empowerment interchangeably with self-efficacy and self-management [23, 43]. Palumbo [215] mentions that patient empowerment is a multifaceted concept that could have different meanings for different people, which could explain why it is confused with other concepts. Indeed, there are aspects of patient empowerment that overlap with other constructs, such as patient enablement, patient activation and patient participation [23]. However, there are critical aspects that differentiate patient empowerment from these.

The lack of consistent use brings into question whether all the available evidence on patient empowerment can actually be said to be related to this construct. From the results of this thesis, it is plausible to argue that it is not. Is this a problem associated only with patient empowerment? Possibly not, considering that this construct is used interchangeably with others, as mentioned previously. It seems patient empowerment is sometimes used as a buzzword across the literature [29] and there is little effort from the authors to elucidate how they understand the construct (i.e. definition, dimensions). It is important to remember that this construct was adopted from another field and that it has been the subject of extensive research [165]. Nevertheless, literature has focused a great deal on analyzing the concept [23, 29, 31, 43] and little has been done on concept development. Conceptual and theoretical research is critical for the development of a cohesive body of knowledge [216], otherwise the construct will continue to be used inconsistently. As Beecher [217] states, concept development is critical to the credibility of research and without it, available evidence may be based on false assumptions.

As there are many available definitions of patient empowerment, there is a variety of instruments available for measuring this construct [43, 55, 56]. There are also studies available that use instruments that are not necessarily meant to measure patient empowerment. Findings from Study I show that some of the included articles used the Patient Activation Measure, Coping Questionnaire, Patient Self-Advocacy Scale, 10-Item Self-Esteem Scale, among others. The aforementioned instruments are not designed to measure patient empowerment, yet researchers have used them in relation to this construct. Even when the findings of these studies may provide relevant information, they only explain a side of what is relevant for patient empowerment [25].

Findings from Study I indicate that the two most common instruments, the MDES [211] and the DES [212], are disease-specific, meant to be used with mental health services users and diabetes, respectively. Some authors, such as Zimmerman [26], have suggested that patient empowerment is disease-specific [20]. According to Zimmerman, patient empowerment manifests in different perceptions and skills and therefore, instruments that attempt to measure this construct are difficult to develop and should not be generic. While there are individual and contextual factors that influence patient empowerment [26], it might not be the outcome or the skills associated with it that are different but rather, the skills level. For instance, if patient empowerment is expressed differently between young persons and adults, it could be associated with the fact that they are in two different life stages, with different developmental characteristics. Study I found that previous research has tried to determine which variables can influence patient empowerment, but the data is limited. However, a qualitative study found that, despite significant variations in participants' age, culture, CC and socioeconomic status, the understanding and experience of this construct were similar [163]. This consequently supports the idea of developing a generic instrument that can help facilitate the comparison of studies across different CCs.

The MDES and DES have other aspects that limit their applicability in other contexts. The MDES measures 15 attributes associated with patient empowerment but with a special focus on community activism and control [211]. Additionally, the MDES did not follow an explicit a-priori theoretical framework and is not of high quality [58]. Unlike the MDES, DES was developed to measure psychosocial self-efficacy [212], yet it is used across literature as a measurement for patient empowerment. A meta-analysis identified DES as one of the most commonly used instruments in studies evaluating empowering interventions [213]. Included studies used DES to either measure patient empowerment or self-efficacy. Hence, there are an important number of studies that use this instrument to measure two different constructs.

While there is a need to develop a new instrument that measures patient empowerment in light of the theoretical and practical limitations of the DES and MDES, there is also a lack of instruments with good psychometric properties, as identified in previous systematic reviews [55-57, 60] and the majority of these have only been tested in adults [218]. Study II describes the development of

GYPES, a generic instrument with the goal of measuring patient empowerment in young persons. The need for a generic measure that is valid and reliable for use across different contexts, has been mentioned previously [55].

Unlike other available instruments, GYPES' theoretical foundation is clear. The operationalization of patient empowerment is facilitated by the fact that Small and colleagues provide not only a definition of patient empowerment, but also five dimensions that comprise the construct [4]. Their definition was the outcome of a literature review and the dimensions resulted from qualitative interviews with persons with CCs. Both aspects are associated with important information that is needed when developing an instrument [182]. Their definition and dimensions, as well as further review of the literature, helped provide a thorough understanding of the underlying construct. Pekonen [56] mentioned that an instrument aiming to measure patient empowerment should include items related to four elements: 1) patient's capacities; 2) patient's knowledge; 3) patient's behaviors; and 4) support by others. These elements are based on available definitions of patient empowerment. GYPES items cover all aspects related to those elements. GYPES's results indicate that the scale is preliminarily valid and reliable. However, the scale needs to be further evaluated in order to investigate all other psychometric properties that were not included in Study II. For instance, assessment of properties that need more than one time-point measurement, such as test-retest reliability, can help determine the stability of the measurement [182]. Additionally, there are certain properties that can be assessed by comparing groups, such as interpretability. This psychometric property also facilitates translating a quantitative value into a qualitative category [185] (i.e. low, medium, high level of patient empowerment), which is now not possible to determine.

Is patient empowerment the way to improve other outcomes of relevance for health and wellbeing?

Previous literature and proposed theoretical models suggest a strong association between patient empowerment and other patient-reported outcomes. Moreover, they suggest patient empowerment can be understood as a mediating variable [37, 219] or an intermediate outcome [28]. In this doctoral thesis, patient empowerment is understood as an intermediate outcome, rather than a mediating one, given the broad scope of the construct. Intermediate outcomes represent an important step in the causal chain between exposure and an outcome [220]. Patient empowerment involves the reinforcement of important psychosocial skills, such as identifying personal needs, defining strategies to achieve goals, solving problems and seeking information [30, 49]. Additionally, this construct entails the individual has the necessary skills to influence aspects related not only to health, but also other determinants of the individual's life goals [26]. Therefore, it is plausible to expect patient empowerment to lead to improvements in other important variables of a person's life, such as QoL, patient-reported health or clinical outcomes.

Studies I, III and IV provide information about the different relations between patient empowerment and other outcomes, such as QoL, glycemic control, transition readiness, patient-reported health, adherence, level of symptoms and sociodemographic characteristics. Findings from these studies were not always significant, independently of whether they were analyzed in a cross-sectional or longitudinal study. In many cases, certain correlations were expected to be significant, based on the characteristics associated with patient empowerment. However, even correlations that are significant in certain studies prove to be non-significant in others. For instance, in Study I, HbA1c was investigated in relation to patient empowerment in eight studies, yet it was only significant in 62.5% of these studies. This difference in significant correlations also appeared to be the case in Studies III and IV. In Study III, patient empowerment was correlated in univariate analyses with age, QoL, transition readiness, illness perceptions, health behaviors, treatment anxiety, physical appearance, cognitive, communication skills and patient-reported heath. Once other variables were included in the model, only transition readiness and communication skills continued to be significant. On the other hand, in Study IV there were no significant within time correlations between these variables.

The lack of significant correlations could be related to the different characteristics of the studies, such as the number of participants, the participants' age and the quality of the studies. For the articles included in Study I, it could also be due to how the concept was operationalized. The instrument used determines the empirically observable manifestations of patient empowerment [55, 59] and is it possible that if the authors had used another instrument, the associations may have been significant.

As found in Study I, research evaluating correlates of patient empowerment has been mostly cross-sectional. These studies have assessed a broad range of correlates, including patient-reported outcomes, clinical outcomes, patient-reported experiences and sociodemographic characteristics. Nevertheless, the absence of sufficient studies using a longitudinal design limits how far the available evidence can explain the theoretical associations between patient empowerment and other variables.

An empowerment model is presented in the background to this thesis (Figure 1, page 20). Such a model is based on previous theoretical articles and empirical evidence. The right section of the model proposes that patient empowerment leads to better patient-reported and clinical outcomes and later on to improvements in QoL and well-being. In order to test some of the associations in the model, longitudinal research is needed. Results from Study IV allowed the determination of the predictive value of patient empowerment over four patient-reported outcomes (e.g. QoL, patient-reported health, transition readiness and communication skills). Study IV found that a higher level of patient empowerment led to better communication skills. This was the only significant cross-lagged effect of the four models that were tested in Study IV. These findings indicate that the relationship between patient empowerment and communication skills is as presented in Figure 4, scenario A. While some authors have considered communication skills to be an indicator of patient empowerment [28], results from Study IV show that this is not the case and that patient empowerment is a determinant of communication skills.

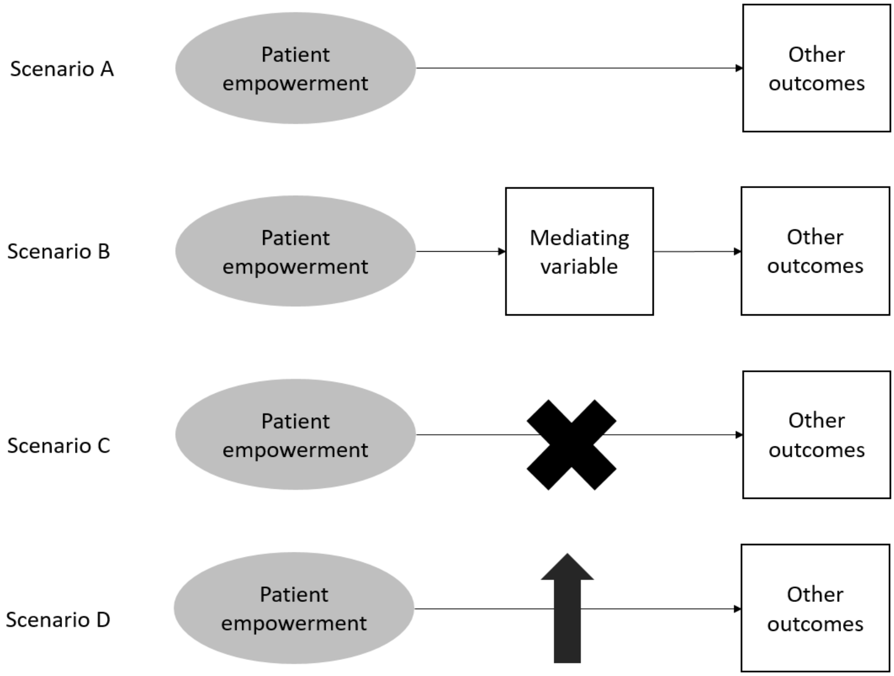


Figure 4. Potential longitudinal associations between patient empowerment and other outcomes

Patient empowerment is a transactional concept because it implies a relationship with another person, in this case between the patient and the healthcare provider [20]. This relationship is based on mutual respect and trust [21] and involves the healthcare provider assisting the patient in developing the skills to participate in care. It is therefore reasonable that patient empowerment predicts communication skills. Existing definitions on patient empowerment have also understood it as a process of communication [10] and even the definition used in this thesis understands it as an outcome and process arising from communication with the healthcare provider [4]. Through constant communication, individuals have several opportunities to express their needs, ask questions and discuss different paths of treatment with the healthcare provider. Moreover, patient empowerment entails changes in self and behaviors [31]. These changes, such as increased confidence, self-efficacy and actively participating in care, can influence the individual's dialogue skills.

Study IV also showed three cross-lagged relations that were not significant. These relations were expected to be significant based on previous research and models, yet our findings support the opposite. However, this does not mean that these relationships cannot be significant in future studies but it does raise the question as to what the actual relationships between these variables could be. Figure 4 shows additional scenarios. In scenario B, it is possible that the relationship between patient empowerment and other variables is mediated by a third variable that is not accounted for in the models. Palumbo [215] mention that research on the mediating variables between patient empowerment and other outcomes is limited. As mentioned previously, little work has been done on concept development in patient empowerment, which also influences what can be concluded about this construct and other variables. It could nevertheless be plausible that these variables are not in fact related to each other and that patient empowerment does not lead to improvements in transition readiness, QoL and patient- reported health (Figure 4, scenario C). A final potential scenario (Figure 4, scenario D) is that the level of patient empowerment of the included participants was not high enough for it to lead to improvements over other outcomes. Available research has not established if there is a necessary level of patient empowerment that should be reached before improvements over other outcomes are noted. Nevertheless, the aforementioned scenarios assume that patient empowerment is the variable leading to improvements over other outcomes and it is possible that the relationship is inverse, i.e. the other outcome predicts the value of patient empowerment. In this case, the direction of the arrows in scenarios A, B and D should be reversed.

So far, it has been assumed by the majority of the literature and this doctoral thesis that patient empowerment is associated with improvements over other outcomes. However, it could be that patient empowerment does not result in actions that are in line with the health practices expected by healthcare providers [165]. Patient empowerment entails giving the patients appropriate information and the opportunity to decide for themselves and act according to what they consider is best. Thus it is always possible that the patient's health behaviors are not health promoting. We should not assume that healthy choices are synonymous with rational choices [29].

The lack of significant relations in Studies I, III and IV could be also explained by this paradox. Patient empowerment involves dialogue, establishing goals and a way to achieve them. However, the priorities of the patient may differ from those of the healthcare provider [165]. Moreover, being healthy may have different meanings for different people [16]. These facts add another layer of complexity to the relationship between patient empowerment and other outcomes. Additionally, patient empowerment is not linear; it is an iterative process that is ongoing and fluctuating [218] and this possibility of constant change can influence the effect patient empowerment has over other outcomes.

What is the evidence on patient empowerment during the transition to adulthood?

Study I found that the majority of publications involved adults and very few studies included persons under the age of 18 years. A further assessment of those studies including young people found that none were related to the field of transition. Studies III and IV therefore add additional evidence for a particular group with an evidence gap and are some of the first studies to evaluate patient empowerment in the context of transition.

Patient empowerment was introduced early in the care of persons with CCs, but this has not been translated to research involving young people. Why has research mostly focused on adults? Perhaps one potential answer is that until recently, adolescents' health had not been the primary focus of different stakeholders. While great focus has been placed on children's health, policies targeting the health of young people have been limited. This recent change came about once the adolescent stage was acknowledged as the foundation of future health and that resources for healthy adolescent growth and emotional development would yield large benefits in the future [221]. Another explanation could be that young persons with CCs were not considered to have an active role in their care, due to their parents shouldering a great part of the responsibility. This perspective might have changed, however, once research on the transition to adulthood increased and the need to involve young persons in their care was highlighted.

Young people can benefit from patient empowerment. Their involvement in care will give them a voice and a significant role in their care. As Grealish [37] mentions, young people are often the recipients rather than partners in care. Patient empowerment allows them to develop the skills they need to manage their illness and their lives and become independent. Considering adolescence is a stage in which young people develop the skills that will help them during adulthood [1], empowering them will benefit them at this time.

Evidence on patient empowerment in relation to transition to adulthood is limited. Available studies have described empowering interventions [222, 223] and only two studies have attempted to evaluate patient empowerment [139, 224]. However, these studies rather than measuring the construct in itself, used measures that reflect targets or components of patient empowerment, which are: self-efficacy, health-related transition competence and patient activation [139, 224].

A study that investigated this construct in young people with cancer identified it was correlated with health-related QoL, as well as with autonomy, self-awareness and perception of social support [225]. As in Study III, QoL was correlated with patient empowerment but this correlation was no longer significant when including other variables in the model. Moreover, in Study IV this association was only significant at T1. As mentioned in the previous section, there could be different reasons for the lack of significant correlations. In this particular case, it could be associated with the age and maturation of the participants or that a higher level of patient empowerment or QoL is needed.

Another study that included adolescents with type 1 diabetes found patient empowerment was related to executive functions, disease duration and metabolic control [226]. This study focused more on disease-specific outcomes but it did provide evidence on the association between patient empowerment and executive functions. These are understood as increased reasoning, self-awareness, behavioral organization and ability to multitask. These variables were not measured in the studies, yet they could be associated with aspects related to transition readiness and health behaviors, which were significant in Study III, but not Study IV.

In Study III and IV there is evidence that patient empowerment is correlated with communication skills and that it leads to improvements of these variables in the future. Communication is an essential part of the transition process, allowing young people to articulate their needs, make their voices heard and discuss with the healthcare providers how the transition process should proceed. A systematic review [227] identified that young people had difficulties communicating with the healthcare providers about sensitive topics and were concerned about confidentiality. Adolescents also find it difficult to ask questions that might reveal poor health behaviors [228]. Discussing sensitive topics such as sexuality, drug use or other types of risky health behaviors is relevant, particularly with young persons with a CC. This group engages in risky behaviors, as much as their healthy peers and they should be aware of the consequences these behaviors can have on their CC [119, 120]. Through patient empowerment, a relationship based on trust and mutual respect is established [21] that allows the young person to feel comfortable, but also to find ways of communicating with the healthcare provider.

Patient-provider communication is a complex dyadic interaction and plays a key role in preparing young people for adulthood and adult care [229]. While some adolescents have previously expressed feeling comfortable about asking questions, others do not have this ability [147, 148]. Hence, there is a need for young people to be empowered so they feel capable of asking questions and participating in the care process. Moreover, developing their communication skills will facilitate their participation in adult care once they are transferred.

While the interaction between patient-provider is usually dyadic, in the case of young persons with CCs, the parents are also part of the interaction. This triadic interaction affects whom the information is directed to and who answers the questions posed by the healthcare providers. Parents of adolescents with CCs are usually highly involved in the care of their children and have a great deal of responsibility [148, 150]. However, during transition it is expected that a responsibility shift will gradually occur and parents will take a step back in the care of their children [7, 148]. This change should occur as the young person grows and it is encouraged by the healthcare provider through different strategies, such as addressing the young person directly [101]. This is a strategy that adolescents appreciate, as they want to be the main partner in health-communication [229]

By communicating with the healthcare providers adolescents can build a relationship with them and also learn to achieve autonomy, aspects that have been highlighted by adolescents as important [227]. Autonomy is closely related not only to the developmental stage of adolescence [109] but also to the transition process [126]. It is possible to say that through patient empowerment, young people can achieve autonomy by developing communication skills.

Findings from the included studies in this doctoral thesis indicate only significant associations between patient empowerment and transition readiness and communication skills. Aspects related to these correlations have been discussed previously. However, it is also worth considering whether patient empowerment is potentially associated with other outcomes measured during transition. Empowered young people are likely to understand their care and mobilize resources to achieve goals [225]. Moreover, patient empowerment increases their capacity to think critically [26]. One could therefore expect it to be associated with other outcomes that are usually measured during transition, such as self-management, satisfaction with transition services, disease-related knowledge, self-efficacy and self-management. Available research already shows that it is frequently significantly associated with self-efficacy, as shown in Study I.

It is worth considering that the correlations between patient empowerment and other outcomes in young persons could be influenced by their interpretation of health. Spencer [165] mentions that this group's understanding of health is usually in reference to "fun" and not in line with what adults would interpret as being healthy. Additionally, a young person's priorities might not be in line with what healthcare providers expect. Adolescents have their own understandings of health and through patient empowerment they gain skills but also the confidence to decide for themselves [29, 31].

Young people go through a series of physical, cognitive and social changes [9], changes that can influence the level of patient empowerment and the changes in self, behaviors and relationships that it involves [31]. Adolescence is a complex stage that poses several challenges to the young person and this context can potentially influence their process of patient empowerment. On the other hand, patient empowerment can be the opportunity for a young person to go through this developmental stage with fewer complications.

Methodological considerations

The results of the four studies included in this doctoral thesis should be assessed in light of certain methodological limitations. Studies II, III and IV included only two CCs, diabetes and CHD. Even when patient empowerment is not considered a disease-specific construct, it is important to consider the generalizability of the results when only two CCs have been investigated. The majority of the participants included in the thesis were above 16 and under 18 years of age. Moreover, only Swedish-speaking participants were included in Studies II (Phase II), III and IV. The aforementioned aspects influence the external validity of the studies, as the extent to which the results can be inferred to a larger sample is limited by the participants' age and the CC included.

Studies II (Phase II) and III had a low response rate. Despite the efforts of the study collaborators, it was not possible to increase the number of questionnaires received. This low response rate could have an effect on internal and external validity because the sample variance could be understated and the results might not be applicable in other situations.

Study I helped identify a broad range of correlations between patient empowerment and other outcomes but the quality of the studies was not assessed, so these correlations should be interpreted cautiously. Moreover, given the broad range of definitions and instruments used by the included articles in Study I, it is possible to conclude that not all the correlates found are in fact related to patient empowerment. This limitation could have been minimized if the articles had been selected based on their conceptualization and/or instrument of patient empowerment. However, this would limit the number of eligible articles, and due to previous knowledge of the heterogeneity of the available evidence, it would not have been the most suitable approach. A further assessment of the instruments used would have been helpful in capturing how the articles understood patient empowerment.

Study II did not evaluate all the psychometric properties suggested by the COSMIN checklist [230], so there are aspects related to validity and reliability that have to be assessed in future studies. Additionally, a forward-backward translation method was not used when translating the Swedish version of GYPES.

Study III had a hypothesis-generating focus and a broad range of variables in relation to patient empowerment were included. However, a more specific model with fewer variables could have provided different results. Given the nature of the data (i.e. cross-sectional), it is not possible to establish causal relationships. The questionnaires included in this study had been previously validated but the version of GYPES that was included here is the version used in Study II, Phase II, which was not the final version of the scale from Study II, Phase III. This version had certain psychometric limitations that can influence the internal validity of the study but this was managed by calculating weighted scores.

For SEM a minimum of 200 participants is recommended, an aspect that was not possible to fulfill in Study IV. In order to compensate for the number of participants, only two variables (measured at two time points) were included in the models. This limitation affects the internal validity of the study because it was not possible to evaluate models that are more complex and the estimates and relationships found could have been different. Moreover, there were dropouts at T1, which meant there was no complete data set for all the participants.

CONCLUSIONS

Available evidence uses a variety of definitions and instruments to measure patient empowerment. This lack of consensus on both a definition and instrument means that current literature provides heterogeneous evidence. Moreover, a proportion of this evidence is at risk of not being related to patient empowerment at all, due to the use of instruments that are not meant to measure patient empowerment or the use of the construct interchangeably with others. GYPES was developed to compensate for the lack of availability of a valid and reliable instrument to measure patient empowerment. The scale allows for comparisons across a wide range of CCs and the studies showed that it is valid and reliable. Additionally, GYPES its one of the first available instruments that has been validated in young persons with CCs.

Although there are studies measuring different correlates, few assess the same correlation. This means that, despite the large number of correlates evaluated, there is little evidence that can support the majority of them. Most of the studies assessing correlations are also of a cross-sectional nature, meaning the extent of the evidence is limited in providing information regarding a potential predictive effect of patient empowerment on other variables or vice versa. Such models need to be further tested and potentially revised on the basis of longitudinal, empirical evidence.

The findings of the included studies in this doctoral thesis give evidence that patient empowerment leads to improvements in the communication skills of young persons with CCs. These skills are related to communicating with the healthcare provider but could also improve this group's communication with other people outside of a healthcare context. Through improved communication skills young people can express their needs and ask questions in relation to their CC, but also discuss aspects related to adulthood, education, sexuality and relationships.

Overall, there is limited research involving young people and patient empowerment. The evidence is even more limited when trying to assess this construct within the context of the transition to adulthood and adult care. Although patient empowerment is a relevant construct for young people, it seems it has mostly been studied in adults. Empowering young people can potentially facilitate the transition to adulthood and improve their participation in adult care. However, further research is needed in order to better understand the relevance of this construct in young persons with CCs going through transition.

IMPLICATIONS FOR CARE

Nurses are in a unique position to facilitate the transition to adulthood and adult care by providing person-centered care to young persons with CCs. Given this role, they can also potentially be responsible for empowering them. However, before this can happen, nurses and other healthcare providers need to be aware of the implications and important moderators that can potentially influence the level of patient empowerment.

Healthcare providers interested in empowering young persons should initially identify their own caring philosophy and whether it fits with patient empowerment. Person-centered care as an approach to care can facilitate the process of patient empowerment, by building a relationship, establishing common goals and identifying barriers and resources. It is worth noting that healthcare providers need to be willing to relinquish control and accept the individual responsibility and accountability patients have for their health.

Moreover, healthcare providers ought to be aware of the contextual and societal factors that can influence the process of empowering the patients. This knowledge will be valuable when determining goals along with the patient. Identifying such factors can be facilitated by implementing HEADSS psychosocial interviewing.

Special interest should be placed on understanding what being healthy means to young people, as this can potentially influence patient empowerment's effect on other variables. Occasionally, their interpretation of health will differ from that of the healthcare providers and this should be discussed with the patient.

Young people have mentioned that a shift in responsibility should be gradual, which is something one could expect to reflect on when discussing their process of becoming empowered. This transfer of control and power should be made gradually. Moreover, the process of patient empowerment is not linear and it might be that some young people need to take a few steps back before feeling capable of assuming more responsibility. Empowering this group should therefore be planned according to the needs of the young individuals and reassessed as it progresses. During this process, different tools can be used, for example the shared decision-making model, which highlights the stepwise process of becoming responsible for your care and life.

Encouraging active participation from young people during care visits starts by creating a safe environment that assures them they can ask and discuss any subject without being judged. Engaging young people in care is also determined by how adolescent-friendly the services are, meaning healthcare providers should approach them through common interests and by implementing strategies that are relevant to this age group.

Parents have a very important role during the transition to adulthood and adult care and nurses should consider involving them in the process of empowerment. This acknowledges the importance of the job they have done with the young person and also that their knowledge is an important resource. Their involvement also means that the parents will not become a barrier, but a source of support during the shift in responsibility.

Clinically measuring the level of patient empowerment can help determine how the person perceives their participation in care. GYPES is a short scale that can be used clinically to measure this outcome. Besides providing a total score for the level of patient empowerment, GYPES has five dimensions and assessing these and the individual items can provide further guidance on which areas are problematic for the young person and should be targeted.

FUTURE PERSPECTIVES

This doctoral thesis highlights the lack of longitudinal research on patient empowerment, indicating that longitudinal studies should be prioritized. Current conceptual models on patient empowerment need longitudinal, empirical evidence to generate better understanding of what this concept entails. Additionally, concept development work is needed to help clarify the dimensions that comprise the construct, its effect on other variables, and its association with other similar constructs, such as self-efficacy or self-management.

Future research should also evaluate the association between patient empowerment and other transition outcomes. Not only patient-reported outcomes but also clinical outcomes are important to assess, given their relevance to healthcare utilization and costs. While there is preliminary evidence that patient empowerment leads to improved communication skills, this association should be further assessed in other groups. Moreover, variables that might affect the predictive effect of patient empowerment should also be considered in future conceptual models. In addition, special focus should be given to ways that healthcare providers can help improve communication skills. Such strategies should be evaluated and designed according to the target group.

Research that highlights the role of patient empowerment in young people and its potential benefits should increase. This construct is relevant not only for managing disease, but also for helping young people manage the transition to adulthood much better. Additionally, future research should also be oriented towards discussion with young people about their perspectives on the process of patient empowerment and how this can be facilitated.

This doctoral thesis does not include the perspectives of parents or healthcare providers. In order to have a full understanding of the process of patient empowerment in young persons, research should acknowledge contextual factors. Besides this, individual factors such as cultural traditions and ethnic background are variables of interest, because power and responsibility can be greatly influenced by these aspects.

GYPES needs to be further tested in order to gain additional evidence on the psychometric properties of the scale in other groups, but also to help compare the level of patient empowerment across different groups and help determine a low, medium and high level of patient empowerment.

ACKNOWLEDGEMENTS

During my five years as a PhD fellow, I have had the opportunity to meet wonderful and inspiring people, and this journey was definitely much better because of them. Many people have contributed to this doctoral thesis, either by giving me words of encouragement, guiding me in the world of research or participating in the studies. I cannot name every single person that has been with me on this road but there are some individuals that I would like to acknowledge.

First, I would like to thank the wonderful supervisors that I was lucky enough to have. *Ewa-Lena*, *Carina* and *Philip*, you have inspired me in so many ways. From each one of you I have learned so many things, that I do not think it is possible to count them. I will be forever grateful that you saw potential in me and allowed me to join STEPSTONES. You changed my life and the accomplishments that I have achieved so far are greatly because I have had your support the entire way. Thank you for this!

The first PhD in STEPSTONES, *Åsa* when I started, I was very uncertain about what I was doing and what my role was in the project. However, knowing that you were there and had a better idea of what was happening gave me peace of mind. I have seen you work clinically and in research, and in both situations I have learned from you. Your knowledge about patients with CHD was always something that I admired. Thank you for sharing your knowledge with me.

Markus, when you joined STEPSTONES you became my research sibling. It was reassuring to know I could count on you to discuss the project and our papers and it was always fruitful and interesting. We could always rely on each other along the way. We have a similar sense of humor and that was always a treat. I hope you know that I will always be available to continue having our discussions and being each other's support (besides my drawer will always have candy).

Sandra, you have always believed in me and supported me along the way. Through every moment that I doubted myself or was uncertain, you would always lift me up. I am very grateful for your kind words of support and for always being there to have a kind-hearted talk.

To the relatively new PhD students in STEPSTONES, *Mikaela* and *Salma*, you are very lucky to be part of such a well-organized and fun research group. We have not shared a lot, but I hope you know that I am here for you.

Ulrica, thank you for always being there. I knew that if I ever needed to get advice about work or someone to hear me out about my life, you would always be there, ready for some coffee with 3-second milk in it.

Lina, we started at the same time and I must say that it was nice to share most of this path with you. I really enjoyed that we could always bounce ideas around about anything. I learned a lot about qualitative research because of you and having your comments/suggestions was always something that improved my output. You guided me when I needed help and listened me when I just needed to vent.

To my amazing unicorns, *Fouke* and *Liesbet*, words cannot express the joy you have brought to my life. You made going to Belgium fun. You welcomed me and my loud laugh with open arms. There is no way this PhD would have been the same without you. We have grown and learned together and you inspire me in many ways. We have been each other's rocks when things have gotten difficult. I cherish every moment we have shared and I look forward to the many memories we will create. I love you girls!

Eva, you are an amazing researcher. I am at awe at your skills and your knowledge and I feel privileged to be able to learn from you and to have had your support and guidance in this path.

I would like to thank all my *fellow doctoral students* at the Institute of Health and Care Sciences and the Department of Public Health and Primary Care. As doctoral students, we share a common experience and we have at some point had discussions about life and research in the lunch room. Those discussions have been encouraging, part of a highly-valued knowledge exchange and fun. I would like to especially thank *Jenny*, *Calle* and *Ramona*; we have always shared a good laugh and have had discussions that have helped me decide which way to go sometimes.

To my fellow PhD mom, *Leila*, you know what it is to balance this hectic research life with a toddler, all while being in a foreign country. You make me laugh and give me food; there is nothing better than that. I am so thankful for your presence in my life.

I would like to thank my *parents*. When I told them that I was starting a PhD, they were shocked and a bit saddened, because this meant that their only child was not coming back home. However, they also showed how proud they were of me and have been my rock, even at a distance. My parents are my biggest driver to continue pursuing my dreams. They have taught me that everything requires sacrifice and hard work and that nothing should be taken for granted. They have taught me to be grateful and always to be humble. My success is their success because their struggles, allowed me to enjoy many privileges. I love them and I thank them for their love and support.

If there is someone who has seen me struggle, it is my husband *Fahad*. He has seen me go through a range of emotions that highlight the highest and the lowest points as a PhD fellow. When I have doubted myself, he has always reminded me of my abilities and always has faith in me. If there is someone that believes in me unconditionally, it is him. Thank you for dealing with the joy, but most of all the insanity that came along. Just like me, you also survived this PhD. I love you!

To my beautiful daughter *Amira*, you came along when I was half-way through my PhD and while you may not remember how it was to have a PhD mom, I will tell you this: you helped me achieve balance. You gave me another purpose and brought more light into my life. Thank you for always being so well-behaved during the conferences, video calls and supervisions that you tagged along on.

To my aunt, *Lalita* and my cousin, *Robert*, your love and constant support has been so valuable. Knowing that I could always count on you is priceless for me.

Irly, you are my best friend, the closest thing that I have to a sister. Despite the distance, you have been a great support during my PhD. You have been there when it has been difficult, but also when it has been joyful. You believe in me and your words of encouragement at times, have been that extra push I have needed.

I would like to thank those that financially enabled me to pursue my PhD, the *Institute of Health and Care Sciences* for my PhD fellowship and all the funding institutions that support STEPSTONES, the *Swedish Heart-Lung Foundation*, the *Swedish Children Heart Association*, the *Swedish Research Council for Health, Working Life and Welfare*, the *Swedish Research Council* and *PK Hedborg Foundation*. Lastly, thank you to the *University of Gothenburg* and *KU Leuven*, for giving me the opportunity to be double degree PhD fellow.

REFERENCES

1. Sawyer SM, Azzopardi PS, Wickremarathne D, Patton GC. The age of adolescence. Lancet Child Adolesc Health. 2018;2(3):223-8. doi: 10.1016/S2352-4642(18)30022-.
2. Hvidberg MF, Johnsen SP, Davidsen M, Ehlers L. A nationwide study of prevalence rates and characteristics of 199 chronic conditions in Denmark. Pharmacoecon Open. 2019;4(2). doi: 10.1007/s41669-019-0167-7.
3. Boyd KM. Disease, illness, sickness, health, healing and wholeness: exploring some elusive concepts. Med Humanit. 2000;26(1):9-17.
4. Small N, Bower P, Chew-Graham CA, Whalley D, Protheroe J. Patient empowerment in long-term conditions: development and preliminary testing of a new measure. BMC Health Serv Res. 2013;13:263. doi: 10.1186/1472-6963-13-263.
5. Knauth Meadows A, Bosco V, Tong E, Fernandes S, Saidi A. Transition and transfer from pediatric to adult care of young adults with complex congenital heart disease. Curr Cardiol Rep. 2009;11(4):291-7.
6. Iversen E, Kolltveit BH, Hernar I, Mårtensson J, Haugstvedt A. Transition from paediatric to adult care: a qualitative study of the experiences of young adults with type 1 diabetes. Scand J Caring Sci. 2019;33(3):723-30.
7. Asp A, Bratt EL, Bramhagen AC. Transfer to adult care-experiences of young adults with congenital heart disease. J Pediatr Nurs. 2015;30(5):e3-10. doi: 10.1016/j.pedn.2015.05.025.
8. Burström Å, Öjmyr-Joelsson M, Bratt EL, Lundell B, Nisell M. Adolescents with congenital heart disease and their parents: needs before transfer to adult care. J Cardiovasc Nurs. 2016;31(5):399-404.
9. Sawyer SM, Afifi RA, Bearinger LH, Blakemore S-J, Dick B, Ezeh AC, et al. Adolescence: a foundation for future health. Lancet. 2012;379(9826):1630-40. doi: 10.1016/S0140-6736(12)60072-5.
10. Aujoulat I, d'Hoore W, Deccache A. Patient empowerment in theory and practice: polysemy or cacophony?. Patient Educ Couns. 2007;66(1):13-20.
11. Viner RM. Transition of care from paediatric to adult services: one part of improved health services for adolescents. Arch Dis Child. 2008;93(2):160-3.
12. Acuña Mora M, Sparud-Lundin C, Bratt EL, Moons P. Empowering young persons during the transition to adulthood. In: Betz CL, Coyne I, editors. Transition from pediatric to adult healthcare services for adolescents and young adults with long-term conditions: an international perspective nurses' roles and interventions. The Netherlands: Springer Nature; 2020. p.19-41.
13. Meleis A. Theoretical nursing: development and progress. (6th ed) China: Wolters Kluwer; 2018. Chapter 6, The discipline of nursing: perspective and domain; p. 85-105.
14. Jones SP, Meleis A. Health is empowerment. Adv Nurs Sci. 1993;15(3):1-14.

15. McCartney G, Popham F, McMaster R, Cumbers A. Defining health and health inequalities. Public Health. 2019;172:22-30. doi: 10.1016/j.puhe.2019.03.023.
16. Seidlein AH, Salloch S. Illness and disease: an empirical-ethical viewpoint. BMC Med Ethics. 2019;20(1):5. doi: 10.1186/s12910-018-0341-y.
17. Wallerstein N, Bernstein E. Empowerment education: Freire's ideas adapted to health education. Health Educ Q. 1988;15(4):379-94.
18. World Health Organization. The Ottawa Charter for health promotion [Internet]. Geneva, Switzerland: WHO;1986 [2020 Feb 11]. Available from: https://www.who.int/healthpromotion/conferences/previous/ottawa/en/
19. Rappaport J. Terms of empowerment/exemplars of prevention: toward a theory for community psychology. Am J Community Psychol. 1987;15(2):121-48.
20. Gibson CH. A concept analysis of empowerment. J Adv Nurs. 1991;16(3):354-61.
21. Ryles SM. A concept analysis of empowerment: its relationship to mental health nursing. J Adv Nurs. 1999;29(3):600-7.
22. Bulsara C, Styles I, Ward AM, Bulsara M. The psychometrics of developing the patient empowerment scale. J Psychosoc Oncol. 2006;24(2):1-16.
23. Castro EM, Van Regenmortel T, Vanhaecht K, Sermeus W, Van Hecke A. Patient empowerment, patient participation and patient-centeredness in hospital care: a concept analysis based on a literature review. Patient Educ Couns. 2016;99(12):1923-39.
24. Funnell MM, Anderson RM. Empowerment and Self-Management of Diabetes. Clin Diabetes. 2004;22(3):123-7.
25. Cattaneo LB, Chapman AR. The process of empowerment: a model for use in research and practice. Am Psychol. 2010;65(7):646-59.
26. Zimmerman MA. Psychological empowerment: issues and illustrations. Am J Community Psychol. 1995;23(5):581-99.
27. Fumagalli LP, Radaelli G, Lettieri E, Bertele P, Masella C. Patient empowerment and its neighbours: clarifying the boundaries and their mutual relationships. Health Policy. 2015;119(3):384-94.
28. Bravo P, Edwards A, Barr PJ, Scholl I, Elwyn G, McAllister M, et al. Conceptualising patient empowerment: a mixed methods study. BMC Health Serv Res. 2015;15(1):252. doi: 10.1186/s12913-015-0907-z.
29. Rodwell CM. An analysis of the concept of empowerment. J Adv Nurs. 1996;23(2):305-13.
30. St-Cyr Tribble D, Gallagher F, Bell L, Caron C, Godbout P, Leblanc J, et al. Empowerment interventions, knowledge translation and exchange: perspectives of home care professionals, clients and caregivers. BMC Health Serv Res. 2008;8:177. doi: 10.1186/1472-6963-8-177.
31. Falk-Rafael AR. Empowerment as a process of evolving consciousness: a model of empowered caring. Adv Nurs Sci. 2001;24(1):1-16.
32. Hawks JH. Empowerment in nursing education: concept analysis and application to philosophy, learning and instruction. J Adv Nurs. 1992;17(5):609-18.

33. Camerini A-L, Schulz PJ. Health literacy and patient empowerment: separating con-joined twins in the context of chronic low back pain. PloS One. 2015;10(2):e0118032-e. doi: 10.1371/journal.pone.0118032.
34. Crondahl K, Eklund Karlsson L. The nexus between health literacy and empowerment: a scoping review. SAGE Open. 2016;6(2). doi: 10.1177/2158244016646410.
35. Sak G, Rothenfluh F, Schulz PJ. Assessing the predictive power of psychological empowerment and health literacy for older patients' participation in health care: a cross-sectional population-based study. BMC Geriatr. 2017;17(1):59. doi: 10.1186/s12877-017-0448-x.
36. Schulz PJ, Nakamoto K. Health literacy and patient empowerment in health communication: the importance of separating conjoined twins. Patient Educ Couns. 2013;90(1):4-11.
37. Grealish A, Tai S, Hunter A, Emsley R, Murrells T, Morrison AP. Does empowerment mediate the effects of psychological factors on mental health, well-being, and recovery in young people?. Psychol Psychother. 2017;90(3):314-35.
38. Kuo CC, Lin CC, Tsai FM. Effectiveness of empowerment-based self-management interventions on patients with chronic metabolic diseases: a systematic review and meta-analysis. Worldviews Evid Based Nurs. 2014;11(5):301-15.
39. Kärner Köhler A, Tingström P, Jaarsma T, Nilsson S. Patient empowerment and general self-efficacy in patients with coronary heart disease: a cross-sectional study. BMC Fam Pract. 2018;19(1):76. doi: 10.1186/s12875-018-0749-y.
40. Moattari M, Ebrahimi M, Sharifi N, Rouzbeh J. The effect of empowerment on the self-efficacy, quality of life and clinical and laboratory indicators of patients treated with hemodialysis: a randomized controlled trial. Health Qual Life Outcomes. 2012;10:115. doi: 10.1186/1477-7525-10-115.
41. Sit JW, Chair SY, Choi KC, Chan CW, Lee DT, Chan AW, et al. Do empowered stroke patients perform better at self-management and functional recovery after a stroke? A randomized controlled trial. Clin Interv Aging. 2016;11:1441-50.
42. Cuthbert CA, Samawi HH, Hemmelgarn BR, Cheung WY. Effectiveness and components of self-management interventions in adult cancer survivors: a protocol for a systematic review and planned meta-analysis. Syst Rev. 2018;7(1):238.
43. Cerezo PG, Juvé-Udina ME, Delgado-Hito P. Concepts and measures of patient empowerment: a comprehensive review. Rev Esc Enferm USP. 2016;50(4):667-74.
44. Akhter K, Bunn C, Graffy J, Donald S, Ward C, Simmons D. Empowerment-based education for established type 2 diabetes in rural England. Pract Diab. 2017;34(3):83-8.
45. van den Berg JJ, Neilands TB, Johnson MO, Chen B, Saberi P. Using path analysis to evaluate the healthcare empowerment model among persons living with HIV for antiretroviral therapy adherence. AIDS Patient Care STDS. 2016;30(11):497-505.

46. Wong CKH, Wong WCW, Wan EYF, Wong WHT, Chan FWK, Lam CLK. Increased number of structured diabetes education attendance was not associated with the improvement in patient-reported health-related quality of life: results from Patient Empowerment Programme (PEP). Health Qual Life Outcomes. 2015;13:126. doi: 10.1186/s12955-015-0324-3.
47. Wilson TE, Kay ES, Turan B, Johnson MO, Kempf MC, Turan JM, et al. Healthcare empowerment and HIV viral control: mediating roles of adherence and retention in care. Am J Prev Med. 2018;54(6):756-64.
48. Tang TS, Funnell MM, Brown MB, Kurlander JE. Self-management support in "real-world" settings: an empowerment-based intervention. Patient Educ Couns. 2010;79(2):178-84.
49. Shin S, Park H. Effect of empowerment on the quality of life of the survivors of breast cancer: the moderating effect of self-help group participation. Jpn J Nurs Sci. 2017;14(4):311-9.
50. Rossi MC, Lucisano G, Funnell M, Pintaudi B, Bulotta A, Gentile S, et al. Interplay among patient empowerment and clinical and person-centered outcomes in type 2 diabetes. The BENCH-D study. Patient Educ Couns. 2015;98(9):1142-9.
51. Cortez DN, Macedo MML, Souza DAS, Dos Santos JC, Afonso GS, Reis IA, et al. Evaluating the effectiveness of an empowerment program for self-care in type 2 diabetes: a cluster randomized trial. BMC Public Health. 2017;17(1):41. doi: 10.1186/s12889-016-3937-5.
52. Wallerstein N. What is the evidence on effectiveness of empowerment to improve health? [Internet]. Copenhagen, Denmark: WHO Regional Office for Europe; 2006. [cited 2020 April 10]. Available from: https://www.euro.who.int/__data/assets/pdf_file/0010/74656/E88086.pdf
53. Johnsen AT, Eskildsen NB, Thomsen TG, Grønvold M, Ross L, Jørgensen CR. Conceptualizing patient empowerment in cancer follow-up by combining theory and qualitative data. Acta Oncol. 2017;56(2):232-8.
54. Lian J, McGhee SM, So C, Chau J, Wong CKH, Wong WCW, et al. Five-year cost-effectiveness of the Patient Empowerment Programme (PEP) for type 2 diabetes mellitus in primary care. Diabetes Obes Metab. 2017;19(9):1312-6.
55. Barr PJ, Scholl I, Bravo P, Faber MJ, Elwyn G, McAllister M. Assessment of patient empowerment--a systematic review of measures. PloS One. 2015;10(5):e0126553-e. doi: 10.1371/journal.pone.0126553.
56. Pekonen A, Eloranta S, Stolt M, Virolainen P, Leino-Kilpi H. Measuring patient empowerment – a systematic review. Patient Educ Couns. 2020;103(4):777-87.
57. Herbert RJ, Gagnon AJ, Rennick JE, O'Loughlin JL. A systematic review of questionnaires measuring health-related empowerment. Res Theory Nurs Pract. 2009;23(2):107-32.
58. Cyril S, Smith BJ, Renzaho AM. Systematic review of empowerment measures in health promotion. Health Promot Int. 2016;31(4):809-26.

59. Eskildsen NB, Joergensen CR, Thomsen TG, Ross L, Dietz SM, Groenvold M, et al. Patient empowerment: a systematic review of questionnaires measuring empowerment in cancer patients. Acta Oncol. 2017;56(2):156-65.
60. McAllister M, Dunn G, Payne K, Davies L, Todd C. Patient empowerment: the need to consider it as a measurable patient-reported outcome for chronic conditions. BMC Health Serv Res. 2012;12:157. doi: 10.1186/1472-6963-12-157.
61. Náfrádi L, Nakamoto K, Csabai M, Papp-Zipernovszky O, Schulz PJ. An empirical test of the Health Empowerment Model: does patient empowerment moderate the effect of health literacy on health status?. Patient Educ Couns. 2018;101(3):511-7.
62. Nuño R, Coleman K, Bengoa R, Sauto R. Integrated care for chronic conditions: the contribution of the ICCC Framework. Health Policy. 2012;105(1):55-64.
63. Myndigheten för vård och omsorganalys. Stöd på vägen: en uppföljning av satsningen på att förbättra vården för personer med kroniska sjukdomar. Sweden: Vårdanalys; 2018.
64. Brennan P, Perola M, van Ommen GJ, Riboli E. Chronic disease research in Europe and the need for integrated population cohorts. Eur J Epidemiol. 2017;32(9):741-9.
65. Busse TR BM, Scheller-Kreinsen, D, Zentner, A. Tackling chronic disease in Europe: strategies, interventions and challenges. United Kingdom: European Observatory on Health Systems and Policies; 2010.
66. Ekman I, Swedberg K, Taft C, Lindseth A, Norberg A, Brink E, et al. Person-centered care--ready for prime time. Eur J Cardiovasc Nurs. 2011;10(4):248-51.
67. Anderson RM, Funnell MM. Patient empowerment: reflections on the challenge of fostering the adoption of a new paradigm. Patient Educ Couns. 2005;57(2):153-7.
68. Hwang W, Weller W, Ireys H, Anderson G. Out-of-pocket medical spending for care of chronic conditions. Health Aff. 2001;20(6):267-78.
69. Berstein AB HE, Moss AJ, Allen KF, Siller AB, Tiggle RB. Health care in America: trends in utilization. Hyattsville, Maryland: National Center for Health Statistics. 2003.
70. Goodman RA, Posner SF, Huang ES, Parekh AK, Koh HK. Defining and measuring chronic conditions: imperatives for research, policy, program, and practice. Prev Chronic Dis. 2013;10:E66. doi: 10.5888/pcd10.120239.
71. Hajat C, Stein E. The global burden of multiple chronic conditions: a narrative review. Prev Med Rep. 2018;12:284-93.
72. Murray CJ, Barber RM, Foreman KJ, Abbasoglu Ozgoren A, Abd-Allah F, Abera SF, et al. Global, regional, and national disability-adjusted life years (DALYs) for 306 diseases and injuries and healthy life expectancy (HALE) for 188 countries, 1990-2013: quantifying the epidemiological transition. Lancet. 2015;386(10009):2145-91.
73. McKeown RE. The epidemiologic transition: changing patterns of mortality and population dynamics. Am J Lifestyle Med. 2009;3(1 Suppl):19s-26s.

74. Bunker JP. The role of medical care in contributing to health improvements within societies. Int J Epidemiol. 2001;30(6):1260-3.
75. Perrin JM, Anderson LE, Van Cleave J. The rise in chronic conditions among infants, children, and youth can be met with continued health system innovations. Health Aff. 2014;33(12):2099-105.
76. Miller GF, Coffield E, Leroy Z, Wallin R. Prevalence and costs of five chronic conditions in children. J Sch Nurs. 2016;32(5):357-64.
77. Van Cleave J, Gortmaker SL, Perrin JM. Dynamics of obesity and chronic health conditions among children and youth. JAMA. 2010;303(7):623-30.
78. Mayer-Davis EJ, Dabelea D, Lawrence JM. Incidence trends of type 1 and type 2 diabetes among youths, 2002-2012. N Engl J Med. 2017;377(3):301. doi: 10.1056/NEJMc1706291.
79. van der Lee JH, Mokkink LB, Grootenhuis MA, Heymans HS, Offringa M. Definitions and measurement of chronic health conditions in childhood: a systematic review. JAMA. 2007;297(24):2741-51.
80. Kennedy A, Sloman F, Douglass JA, Sawyer SM. Young people with chronic illness: the approach to transition. Inter Med J. 2007;37(8):555-60.
81. Llanes-Álvarez C, Andrés-de Llano JM, Álvarez-Navares AI, Pastor-Hidalgo MT, Roncero C, Franco-Martín MA. Trends in psychiatric hospitalization of children and adolescents in spain between 2005 and 2015. J Clin Med. 2019;8(12):2111.
82. Määttä H, Hurtig T, Taanila A, Honkanen M, Ebeling H, Koivumaa-Honkanen H. Childhood chronic physical condition, self-reported health, and life satisfaction in adolescence. Eur J Pediatr. 2013;172(9):1197-206.
83. Mitchell SC, Korones SB, Berendes HW. Congenital heart disease in 56,109 births incidence and natural history. Circulation. 1971;43(3):323-32.
84. Dolk H, Loane M, Garne E. Congenital heart defects in Europe: prevalence and perinatal mortality, 2000 to 2005. Circulation. 2011;123(8):841-9.
85. Ávila P, Mercier LA, Dore A, Marcotte F, Mongeon FP, Ibrahim R, et al. Adult congenital heart disease: a growing epidemic. Can J Cardiol. 2014;30(12 Suppl):S410-9.
86. Mandalenakis Z, Rosengren A, Skoglund K, Lappas G, Eriksson P, Dellborg M. Survivorship in children and young adults with congenital heart disease in Sweden. JAMA Intern Med. 2017;177(2):224-30.
87. Moons P, Bovijn L, Budts W, Belmans A, Gewillig M. Temporal trends in survival to adulthood among patients born with congenital heart disease from 1970 to 1992 in Belgium. Circulation. 2010;122(22):2264-72.
88. Warnes CA, Liberthson R, Danielson GK, Dore A, Harris L, Hoffman JI, et al. Task force 1: the changing profile of congenital heart disease in adult life. J Am Coll Cardiol. 2001;37(5):1170-5.
89. Amedro P, Dorka R, Moniotte S, Guillaumont S, Fraisse A, Kreitmann B, et al. Quality of life of children with congenital heart diseases: a multicenter controlled cross-sectional study. Pediatr Cardiol. 2015;36(8):1588-601.

90. Drakouli M, Petsios K, Giannakopoulou M, Patiraki E, Voutoufianaki I, Matziou V. Determinants of quality of life in children and adolescents with CHD: a systematic review. Cardiol Young. 2015;25(6):1027-36.

91. Reiner B, Oberhoffer R, Ewert P, Müller J. Quality of life in young people with congenital heart disease is better than expected. Arch Dis Child. 2019;104(2):124-8.

92. Apers S, Moons P, Goossens E, Luyckx K, Gewillig M, Bogaerts K, et al. Sense of coherence and perceived physical health explain the better quality of life in adolescents with congenital heart disease. Eur J Cardiovasc Nurs. 2013;12(5):475-83.

93. Razzaghi H, Oster M, Reefhuis J. Long-term outcomes in children with congenital heart disease: national health interview survey. J Pediatr. 2015;166(1):119-24.

94. Oster ME, Watkins S, Hill KD, Knight JH, Meyer RE. Academic outcomes in children with congenital heart defects: a population-based cohort study. Circ Cardiovasc Qual Outcomes. 2017;10(2). doi: 10.1161/CIRCOUTCOMES.116.003074.

95. Chong LSH, Fitzgerald DA, Craig JC, Manera KE, Hanson CS, Celermajer D, et al. Children's experiences of congenital heart disease: a systematic review of qualitative studies. Eur J Pediatr. 2018;177(3):319-36.

96. Chiang YT, Chen CW, Su WJ, Wang JK, Lu CW, Li YF, et al. Between invisible defects and visible impact: the life experiences of adolescents and young adults with congenital heart disease. J Adv Nurs. 2015;71(3):599-608.

97. Van Deyk K, Pelgrims E, Troost E, Goossens E, Budts W, Gewillig M, et al. Adolescents' understanding of their congenital heart disease on transfer to adult-focused care. Am J Cardiol. 2010;106(12):1803-7.

98. Moceri P, Goossens E, Hascoet S, Checler C, Bonello B, Ferrari E, et al. From adolescents to adults with congenital heart disease: the role of transition. Eur J Pediatr. 2015;174(7):847-54.

99. Jackson JL, Tierney K, Daniels CJ, Vannatta K. Disease knowledge, perceived risk, and health behavior engagement among adolescents and adults with congenital heart disease. Heart Lung. 2015;44(1):39-44.

100. Yang HL, Chen YC, Wang JK, Gau BS, Moons P. An evaluation of disease knowledge in dyads of parents and their adolescent children with congenital heart disease. J Cardiovasc Nurs. 2013;28(6):541-9.

101. Burström Å, Bratt EL, Frenckner B, Nisell M, Hanséus K, Rydberg A, et al. Adolescents with congenital heart disease: their opinions about the preparation for transfer to adult care. Eur J Pediatr. 2017;176(7):881-9.

102. Chen CW, Su WJ, Chiang YT, Shu YM, Moons P. Healthcare needs of adolescents with congenital heart disease transitioning into adulthood: a Delphi survey of patients, parents, and healthcare providers. Eur J Cardiovasc Nurs. 2017;16(2):125-35.

103. Ferguson P, Walker H. ‘Getting on with life’: resilience and normalcy in adolescents living with chronic illness. Int J Incl Educ. 2014;18(3):227-40.

104. Yeo M, Sawyer S. Chronic illness and disability. BMJ. 2005;330(7493):721-3.

105. World Health Organization. The seconde decade: improving adolescent health and development [Internet]. Geneva, Switzerland: Department of Child and Adolescent Health and Development; 2001 [cited 2020 March 2]. Available from: https://apps.who.int/iris/bitstream/handle/10665/64320/WHO_FRH_ADH_98.18_Rev.1.pdf;jsessionid=A5E38A03DF6F451CAF7EA0B267EC27AC?sequence=1
106. Arnett JJ. Emerging adulthood: the winding road from the late teens through the twenties. 2nd ed. New York: Oxford University Press; 2014.
107. Patton GC, Viner R. Pubertal transitions in health. Lancet. 2007;369(9567):1130-9.
108. Christie D, Viner R. Adolescent development. BMJ. 2005;330(7486):301-4.
109. Colver A, Dovey-Pearce G. The relationship of adolescent behaviors to adolescent brain changes and their relevance to the transition of adolescents and young adults with chronic illness and disability. In: Hergenroeder A, Wiemann C, editors. Healthcare transition: building a program for adolescents and young adults with chronic illness and disability: Springer Nature; 2018. p.15-19.
110. Sawyer SM, Drew S, Yeo MS, Britto MT. Adolescents with a chronic condition: challenges living, challenges treating. Lancet. 2007;369(9571):1481-9.
111. Hazen E, Schlozman S, Beresin E. Adolescent psychological development: a review. Pediatr Rev. 2008;29(5):161-7.
112. Viner R, Macfarlane A. Health promotion. BMJ. 2005;330(7490):527-9.
113. Bonomo Y, Proimos J. Substance misuse: alcohol, tobacco, inhalants, and other drugs. BMJ. 2005;330(7494):777-80.
114. Michaud PA SJ, Viner R. The adolescent with a chronic condition: epidemiology, developmental issues and health care provision. Geneva, Switzerland: World Health Organization; 2007.
115. Suris JC, Michaud PA, Viner R. The adolescent with a chronic condition. Part I: developmental issues. Arch Dis Child. 2004;89(10):938-42.
116. Poku BA, Caress AL, Kirk S. Adolescents' experiences of living with sickle cell disease: an integrative narrative review of the literature. Int J Nurs Stud. 2018;80:20-8.
117. Santos T, de Matos MG, Marques A, Simões C, Leal I, Machado MdC. Adolescent's subjective perceptions of chronic disease and related psychosocial factors: highlights from an outpatient context study. BMC Pediatr. 2016;16(1):211. doi: 10.1186/s12887-016-0748-x.
118. Goossens E, Fieuws S, Van Deyk K, Luyckx K, Gewillig M, Budts W, et al. Effectiveness of structured education on knowledge and health behaviors in patients with congenital heart disease. J Pediatr. 2015;166(6):1370-6.e1. doi: 10.1016/j.jpeds.2015.02.041.
119. Surís JC, Michaud PA, Akre C, Sawyer SM. Health risk behaviors in adolescents with chronic conditions. Pediatrics. 2008;122(5):e1113-8.
120. Weitzman ER, Salimian PK, Rabinow L, Levy S. Perspectives on substance use among youth with chronic medical conditions and implications for clinical guidance and prevention: A qualitative study. PLoS One. 2019;14(1):e0209963.

121. Nylander C, Seidel C, Tindberg Y. The triply troubled teenager--chronic conditions associated with fewer protective factors and clustered risk behaviours. Acta Paediatr. 2014;103(2):194-200.
122. Venning A, Eliott J, Wilson A, Kettler L. Understanding young peoples' experience of chronic illness: a systematic review. Int J Evid Based Healthc. 2008;6(3):321-36.
123. Meleis A. Transitions theory: middle range and situation specific theories in nursing research and practice. New York, United States: Springer; 2010.
124. Blum RW, Garell D, Hodgman CH, Jorissen TW, Okinow NA, Orr DP, et al. Transition from child-centered to adult health-care systems for adolescents with chronic conditions. A position paper of the Society for Adolescent Medicine. J Adolesc Health. 1993;14(7):570-6.
125. American Academy of Pediatrics, American Academy of Family Physicians, American College of Physicians-American Society of Internal Medicine. A consensus statement on health care transitions for young adults with special health care needs. Pediatrics. 2002;110(6 Pt 2):1304-6.
126. Rosen DS, Blum RW, Britto M, Sawyer SM, Siegel DM. Transition to adult health care for adolescents and young adults with chronic conditions: Position paper of the society for adolescent medicine. J Adolesc Health. 2003;33(4):309-11.
127. Sable C, Foster E, Uzark K, Bjornsen K, Canobbio MM, Connolly HM, et al. Best practices in managing transition to adulthood for adolescents with congenital heart disease: the transition process and medical and psychosocial issues: a scientific statement from the American Heart Association. Circulation. 2011;123(13):1454-85.
128. Hart LC, Patel-Nguyen SV, Merkley MG, Jonas DE. An evidence map for interventions addressing transition from pediatric to adult care: a systematic review of systematic reviews. J Pediatr Nurs. 2019;48:18-34.
129. Schmidt A, Ilango SM, McManus MA, Rogers KK, White PH. Outcomes of pediatric to adult health care transition interventions: an updated systematic review. J Pediatr Nurs. 2020;51:92-107.
130. Gabriel P, McManus M, Rogers K, White P. Outcome evidence for structured pediatric to adult health care transition interventions: a systematic review. J Pediatr. 2017;188:263-9.e15. doi: 10.1016/j.jpeds.2017.05.066.
131. Crowley R, Wolfe I, Lock K, McKee M. Improving the transition between paediatric and adult healthcare: a systematic review. Arch Dis Child. 2011;96(6):548-53.
132. Holmes-Walker DJ, Llewellyn AC, Farrell K. A transition care programme which improves diabetes control and reduces hospital admission rates in young adults with type 1 diabetes aged 15-25 years. Diabet Med. 2007;24(7):764-9.
133. Weissberg-Benchell J, Shapiro JB. A review of interventions aimed at facilitating successful transition planning and transfer to adult care among youth with chronic illness. Pediatr Ann. 2017;46(5):e182-e7.

134. Otto C, Tárnok A, Erős A, Szakács Z, Vincze Á, Farkas N, et al. Planned transition of adolescent patients with inflammatory bowel disease results in higher remission rates. J Pediatr Nurs. 2019;45:62-6.
135. Allemang B, Allan K, Johnson C, Cheong M, Cheung P, Odame I, et al. Impact of a transition program with navigator on loss to follow-up, medication adherence, and appointment attendance in hemoglobinopathies. Pediatr Blood Cancer. 2019;66(8):e27781.
136. Grant C, Pan J. A comparison of five transition programmes for youth with chronic illness in Canada. Child Care Health Dev. 2011;37(6):815-20.
137. Vaikunth SS, Williams RG, Uzunyan MY, Tun H, Barton C, Chang PM. Short-term outcomes following implementation of a dedicated young adult congenital heart disease transition program. Congenit Heart Dis. 2018;13(1):85-91.
138. Chu PY, Maslow GR, von Isenburg M, Chung RJ. Systematic review of the impact of transition interventions for adolescents with chronic illness on transfer from pediatric to adult healthcare. J Pediatr Nurs. 2015;30(5):e19-27.
139. Schmidt S, Herrmann-Garitz C, Bomba F, Thyen U. A multicenter prospective quasi-experimental study on the impact of a transition-oriented generic patient education program on health service participation and quality of life in adolescents and young adults. Patient Educ Couns. 2016;99(3):421-8.
140. Colver A, McConachie H, Le Couteur A, Dovey-Pearce G, Mann KD, McDonagh JE, et al. A longitudinal, observational study of the features of transitional healthcare associated with better outcomes for young people with long-term conditions. BMC Med. 2018;16(1):111. doi: 10.1186/s12916-018-1102-y.
141. Mackie AS, Islam S, Magill-Evans J, Rankin KN, Robert C, Schuh M, et al. Healthcare transition for youth with heart disease: a clinical trial. Heart. 2014;100(14):1113-8.
142. Mackie AS, Rempel GR, Kovacs AH, Kaufman M, Rankin KN, Jelen A, et al. Transition intervention for adolescents with congenital heart disease. J Am Coll Cardiol. 2018;71(16):1768-77.
143. Seeley A, Lindeke L. Developing a transition care coordination program for youth with spina bifida. J Pediatr Health Care. 2017;31(6):627-33.
144. Hilderson D, Moons P, Van der Elst K, Luyckx K, Wouters C, Westhovens R. The clinical impact of a brief transition programme for young people with juvenile idiopathic arthritis: results of the DON'T RETARD project. Rheumatology. 2016;55(1):133-42.
145. Everitt IK, Gerardin JF, Rodriguez FH, 3rd, Book WM. Improving the quality of transition and transfer of care in young adults with congenital heart disease. Congenit Heart Dis. 2017;12(3):242-50.
146. Betz CL, Ferris ME, Woodward JF, Okumura MJ, Jan S, Wood DL. The health care transition research consortium health care transition model: a framework for research and practice. J Pediatr Rehabil Med. 2014;7(1):3-15.

147. Brown M, Higgins A, MacArthur J. Transition from child to adult health services: A qualitative study of the views and experiences of families of young adults with intellectual disabilities. J Clin Nurs. 2020;29(1-2):195-207.
148. Coyne I, Sheehan A, Heery E, While AE. Healthcare transition for adolescents and young adults with long-term conditions: qualitative study of patients, parents and healthcare professionals' experiences. J Clin Nurs. 2019;28(21-22):4062-76.
149. Tuchman LK, Slap GB, Britto MT. Transition to adult care: experiences and expectations of adolescents with a chronic illness. Child Care Health Dev. 2008;34(5):557-63.
150. Nguyen T, Henderson D, Stewart D, Hlyva O, Punthakee Z, Gorter JW. You never transition alone! Exploring the experiences of youth with chronic health conditions, parents and healthcare providers on self-management. Child Care Health Dev. 2016;42(4):464-72.
151. van Staa AL, Jedeloo S, van Meeteren J, Latour JM. Crossing the transition chasm: experiences and recommendations for improving transitional care of young adults, parents and providers. Child Care Health Dev. 2011;37(6):821-32.
152. Sonneveld HM, Strating MM, van Staa AL, Nieboer AP. Gaps in transitional care: what are the perceptions of adolescents, parents and providers?. Child Care Health Dev. 2013;39(1):69-80.
153. Jennings LB, Parra-Medina DM, Hilfinger-Messias DK, McLoughlin K. Toward a critical social theory of youth empowerment. J Community Pract. 2006;14(1-2):31-55.
154. Morton MH, Montgomery P. Youth empowerment programs for improving adolescents' self-efficacy and self-esteem:a systematic review. Res Soc Work Pract. 2013;23(1):22-33.
155. Eisman AB, Zimmerman MA, Kruger D, Reischl TM, Miller AL, Franzen SP, et al. Psychological empowerment among urban youth: measurement model and associations with youth outcomes. Am J Community Psychol. 2016;58(3-4):410-21.
156. Zimmerman MA, Eisman AB, Reischl TM, Morrel-Samuels S, Stoddard S, Miller AL, et al. Youth empowerment solutions: evaluation of an after-school program to engage middle school students in community change. Health Educ Behav. 2018;45(1):20-31.
157. Úcar Martínez X, Jiménez-Morales M, Soler Masó P, Trilla Bernet J. Exploring the conceptualization and research of empowerment in the field of youth. Int J Adolesc Youth. 2017;22(4):405-18.
158. Betz CL. Facilitating the transition of adolescents with chronic conditions from pediatric to adult health care and community settings. Issues Compr Pediatr Nurs. 1998;21(2):97-115.
159. Rosen DS. Transition of young people with respiratory diseases to adult health care. Paediatr Respir Rev. 2004;5(2):124-31.
160. Burke L, Kirkham J, Arnott J, Gray V, Peak M, Beresford MW. The transition of adolescents with juvenile idiopathic arthritis or epilepsy from paediatric health-care services to adult health-care services: a scoping review of

the literature and a synthesis of the evidence. J Child Health Care. 2018;22(3):332-58.
161. Christie D, Viner R. Chronic illness and transition: time for action. Adolesc Med State Art Rev. 2009;20(3):981-7, xi.
162. Hait E, Arnold JH, Fishman LN. Educate, communicate, anticipate-practical recommendations for transitioning adolescents with IBD to adult health care. Inflamm Bowel Dis. 2006;12(1):70-3.
163. Agner J, Braun KL. Patient empowerment: A critique of individualism and systematic review of patient perspectives. Patient Educ Couns. 2018;101(12):2054-64.
164. Grande SW, Longacre MR, Palmblad K, Montan MV, Berquist RP, Hager A, et al. Empowering young people living with juvenile idiopathic arthritis to better communicate with families and care teams: content analysis of semistructured interviews. JMIR Mhealth Uhealth. 2019;7(2):e10401. doi: 10.2196/10401.
165. Spencer G. Young people and health: towards a new conceptual framework for understanding empowerment. Health. 2014;18(1):3-22.
166. Heery E, Sheehan AM, While AE, Coyne I. Experiences and outcomes of transition from pediatric to adult health care services for young people with congenital heart disease: a systematic review. Congenit Heart Dis. 2015;10(5):413-27.
167. Sansom-Daly UM, Lin M, Robertson EG, Wakefield CE, McGill BC, Girgis A, et al. Health literacy in adolescents and young adults: an updated review. J Adolesc Young Adult Oncol. 2016;5(2):106-18.
168. Sanders LM, Federico S, Klass P, Abrams MA, Dreyer B. Literacy and child health: a systematic review. Arch Pediatr Adolesc Med. 2009;163(2):131-40.
169. Stewart KT, Chahal N, Kovacs AH, Manlhiot C, Jelen A, Collins T, et al. Readiness for transition to adult health care for young adolescents with congenital heart disease. Pediatr Cardiol. 2017;38(4):778-86.
170. Paré G, Trudel M-C, Jaana M, Kitsiou S. Synthesizing information systems knowledge: a typology of literature reviews. Inform Manag. 2015;52(2):183-99.
171. Caruana EJ, Roman M, Hernández-Sánchez J, Solli P. Longitudinal studies. J Thorac Dis. 2015;7(11):E537-E40.
172. Levin KA. Study design III: Cross-sectional studies. Evid Based Dent. 2006;7(1):24-5.
173. Lavrakas P. Encyclopedia of Survey Research Methods. 2008.
174. Gill TM, Feinstein AR. A critical appraisal of the quality of quality-of-life measurements. JAMA. 1994;272(8):619-26.
175. Joanna Briggs Institute. Levels of evidence [Internet]. [cited 2020 April 7]. Available from: https://joannabriggs.org/jbi-approach.html.
176. Hsieh H-F, Shannon SE. Three approaches to qualitative content analysis. Qual Health Res. 2005;15(9):1277-88.
177. Acuña Mora M, Luyckx K, Sparud-Lundin C, Peeters M, van Staa A, Sattoe J, et al. Patient empowerment in young persons with chronic conditions:

Psychometric properties of the Gothenburg Young Persons Empowerment Scale (GYPES). PLoS One. 2018;13(7):e0201007. doi: 10.1371/journal.pone.0201007.
178. Uppsala Clinical Research Center. SWEDCON [Internet]. 2016. [cited 2020 April 8].Available from: http://www.ucr.uu.se/swedcon/.
179. Dillman D. Mail and other self-administered questionnaires. In: Rossi P, Wright J, Anderson A, editors. Handbook of survey research. New York: Academic Press; 1983.
180. Bowen NK. Structural Equation Modeling. Cary: Oxford University Press Incorporated; 2011.
181. Kline R. Principles and practices of strucutal equation modeling. 4th ed. United States: Guilford Publications; 2016.
182. Polit D, Yang F. Measurement and the measurement of change: Wolters Kluwer; 2016.
183. Jackson DL, Gillaspy JA, Purc-Stephenson R. Reporting practices in confirmatory factor analysis: an overview and some recommendations. Psychol Methods. 2009;14(1):6-23.
184. Hu Lt, Bentler PM. Cutoff criteria for fit indexes in covariance structure analysis: conventional criteria versus new alternatives. Struct Equ Modeling. 1999;6(1):1-55.
185. Terwee CB, Bot SD, de Boer MR, van der Windt DA, Knol DL, Dekker J, et al. Quality criteria were proposed for measurement properties of health status questionnaires. J Clin Epidemiol. 2007;60(1):34-42.
186. Wild D, Grove A, Martin M, Eremenco S, McElroy S, Verjee-Lorenz A, et al. Principles of good practice for the translation and cultural adaptation process for Patient-Reported Outcomes (PRO) measures: report of the ISPOR Task Force for Translation and Cultural Adaptation. Value Health. 2005;8(2):94-104.
187. Moons P, Van Deyk K, Marquet K, Raes E, De Bleser L, Budts W, et al. Individual quality of life in adults with congenital heart disease: a paradigm shift. Eur Heart J. 2005;26(3):298-307.
188. Moons P, Van Deyk K, De Bleser L, Marquet K, Raes E, De Geest S, et al. Quality of life and health status in adults with congenital heart disease: a direct comparison with healthy counterparts. Eur J Cardiov Prev R. 2006;13(3):407-13.
189. Goossens E, Luyckx K, Mommen N, Gewillig M, Budts W, Zupancic N, et al. Health risk behaviors in adolescents and emerging adults with congenital heart disease: psychometric properties of the Health Behavior Scale-Congenital Heart Disease. Eur J Cardiovasc Nurs. 2013;12(6):544-57.
190. Broadbent E, Petrie KJ, Main J, Weinman J. The brief illness perception questionnaire. J Psychosom Res. 2006;60(6):631-7.
191. Rönning H, Arestedt K, Erik Nielsen N, Swahn E, Strömberg A. development and psychometric evaluation of the Knowledge Scale for Adults with Congenitally Malformed Hearts. J Cardiovasc Nurs. 2013;28(3):228-37.
192. Petersen S, Hägglöf B, Stenlund H, Bergström E. Psychometric properties of the Swedish PedsQL, Pediatric Quality of Life Inventory 4.0 generic core scales. Acta Paediatr. 2009;98(9):1504-12.

193. Sand P, Kljajić M, Sunnegårdh J. The reliability of the Pediatric Quality of Life Inventory 3.0 Cardiac Module™ for Swedish children with congenital heart defects. Nordic Psychol. 2013;65(3):210-23.
194. Gilleland J, Amaral S, Mee L, Blount R. Getting ready to leave: transition readiness in adolescent kidney transplant recipients. J Pediatr Psychol. 2012;37(1):85-96.
195. Field A. Discovering Statistics using IBM SPSS Statistics: Sage Publications Ltd.; 2013.
196. Machin D, Campbell MJ, Walters S. Medical statistics a textbook for the health sciences. 4th ed. West Sussex, England : John Wiley & Sons Ltd; 2007.
197. Heinze G, Wallisch C, Dunkler D. Variable selection - A review and recommendations for the practicing statistician. Biom J. 2018;60(3):431-49.
198. Acuña Mora M, Sparud-Lundin C, Bratt EL, Moons P. Person-centred transition programme to empower adolescents with congenital heart disease in the transition to adulthood: a study protocol for a hybrid randomised controlled trial (STEPSTONES project). BMJ Open. 2017;7(4):e014593. doi: 10.1136/bmjopen-2016-014593.
199. Kearney M. The SAGE Encyclopedia of Communication Research Methods [Internet]. Thousand Oaks, California: SAGE Publications, Inc; 2017. [cited 2020 April 14]. Available from: http://sk.sagepub.com/reference/the-sage-encyclopedia-of-communication-research-methods.
200. Hamaker EL, Kuiper RM, Grasman RP. A critique of the cross-lagged panel model. Psychol Methods. 2015;20(1):102-16.
201. Kuiper RM, Ryan O. Drawing conclusions from cross-lagged relationships: re-considering the role of the time-interval. Struct Equ Modeling. 2018;25(5):809-23.
202. Selig P, Little T. Autoregressive and cross-lagged panel analysis for longitudinal data. In: Laursen B, Little T, Card N, editors. Handbook of developmental research methods: Guilford Press; 2012.
203. World Medical Association. Declaration of Helsinki: ethical principles for medical research involving human subjects. JAMA. 2013;310(20):2191-4.
204. Central Committee on Research Involving Human Subjects. Your research: is it subject to the WMO or not? [Internet]. [cited 2018 July 12] Available from: https://english.ccmo.nl/investigators/legal-framework-for-medical-scientific-research/your-research-is-it-subject-to-the-wmo-or-not
205. CODEX;. Regler och riktlinjer för forskning: forskning som involverar barn 2020 [Internet]. Sweden; [updated 2020 April 12; cited 2020 Oct 28] Available from: http://www.codex.vr.se/manniska1.shtml.
206. Wendler DS. Assent in paediatric research: theoretical and practical considerations. J Med Ethics. 2006;32(4):229-34.
207. Zutlevics T. Could providing financial incentives to research participants be ultimately self-defeating?. Res Ethics. 2016;12(3):137-48.
208. Resnik DB. Bioethical issues in providing financial incentives to research participants. Medicoleg Bioeth. 2015;5:35-41.

209. Rappaport J. In praise of paradox: A social policy of empowerment over prevention. Am J Community Psychol. 1981;9(1):1-25.
210. Corrigan PW, Garman AN. Considerations for research on consumer empowerment and psychosocial interventions. Psychiatr Serv. 1997;48(3):347-52.
211. Rogers ES, Chamberlin J, Ellison ML, Crean T. A consumer-constructed scale to measure empowerment among users of mental health services. Psychiatr Serv. 1997;48(8):1042-7.
212. Anderson RM, Funnell MM, Fitzgerald JT, Marrero DG. The Diabetes Empowerment Scale: a measure of psychosocial self-efficacy. Diabetes care. 2000;23(6):739-43.
213. Werbrouck A, Swinnen E, Kerckhofs E, Buyl R, Beckwée D, De Wit L. How to empower patients? A systematic review and meta-analysis. Transl Behav Med. 2018;8(5):660-74.
214. Hibbard JH, Stockard J, Mahoney ER, Tusler M. Development of the Patient Activation Measure (PAM): conceptualizing and measuring activation in patients and consumers. Health Serv Res. 2004;39(4 Pt 1):1005-26.
215. Palumbo R. The bright side and the dark side of patient empowerment: co-creation and co-destruction of value in the healthcare environment. Switzerland: Springer Nature; 2017.
216. Rodgers BL, Jacelon CS, Knafl KA. Concept analysis and the advance of nursing knowledge: state of the science. J Nurs Scholarsh. 2018;50(4):451-9.
217. Beecher C, Devane D, White M, Greene R, Dowling M. Concept development in nursing and midwifery: an overview of methodological approaches. Int J Nurs Pract. 2019;25(1):e12702. doi: 10.1111/ijn.12702.
218. Jørgensen CR, Thomsen TG, Ross L, Dietz SM, Therkildsen S, Groenvold M, et al. What facilitates "patient empowerment" in cancer patients during follow-up: a qualitative systematic review of the literature. Qual Health Res. 2018;28(2):292-304.
219. Croom A, Wiebe DJ, Berg CA, Lindsay R, Donaldson D, Foster C, et al. Adolescent and parent perceptions of patient-centered communication while managing type 1 diabetes. J Pediatr Psychol. 2011;36(2):206-15.
220. Seuc AH, Peregoudov A, Betran AP, Gulmezoglu AM. Intermediate outcomes in randomized clinical trials: an introduction. Trials. 2013;14(1):78.
221. Patton GC, Olsson CA, Skirbekk V, Saffery R, Wlodek ME, Azzopardi PS, et al. Adolescence and the next generation. Nature. 2018;554(7693):458-66.
222. Bomba F, Herrmann-Garitz C, Schmidt J, Schmidt S, Thyen U. An assessment of the experiences and needs of adolescents with chronic conditions in transitional care: a qualitative study to develop a patient education programme. Health Soc Care Community. 2017;25(2):652-66.
223. Bergeron S, Noskoff K, Hayakawa J, Frediani J. Empowering adolescents and young adults to support, lead, and thrive: development and validation of an aya oncology child life program. J Pediatr Nurs. 2019;47:1-6.
224. Markwart H, Bomba F, Menrath I, Brenk-Franz K, Ernst G, Thyen U, et al. Assessing empowerment as multidimensional outcome of a patient education

program for adolescents with chronic conditions: A latent difference score model. PLoS One. 2020;15(4):e0230659. doi: 10.1371/journal.pone.0230659.
225. Kaal SEJ, Husson O, van Duivenboden S, Jansen R, Manten-Horst E, Servaes P, et al. Empowerment in adolescents and young adults with cancer: relationship with health-related quality of life. Cancer. 2017;123(20):4039-47.
226. Łuczyński W, Łazarczyk I, Szlachcikowska I, Kiernozek Ż, Kaczmarek A, Szylaj O, et al. The empowerment of adolescents with type 1 diabetes is associated with their executive functions. Biomed Res Int. 2019;2019:5184682.
227. Kim B, White K. How can health professionals enhance interpersonal communication with adolescents and young adults to improve health care outcomes?: systematic literature review. Int J Adolesc Youth. 2018;23(2):198-218.
228. Beresford BA, Sloper P. Chronically ill adolescents' experiences of communicating with doctors: a qualitative study. J Adolesc Health. 2003;33(3):172-9.
229. Monaghan M, Hilliard M, Sweenie R, Riekert K. Transition readiness in adolescents and emerging adults with diabetes: the role of patient-provider communication. Curr Diab Rep. 2013;13(6):900-8.
230. Mokkink LB, Terwee CB, Knol DL, Stratford PW, Alonso J, Patrick DL, et al. The COSMIN checklist for evaluating the methodological quality of studies on measurement properties: a clarification of its content. BMC Med Res Methodol. 2010;10(1):22.

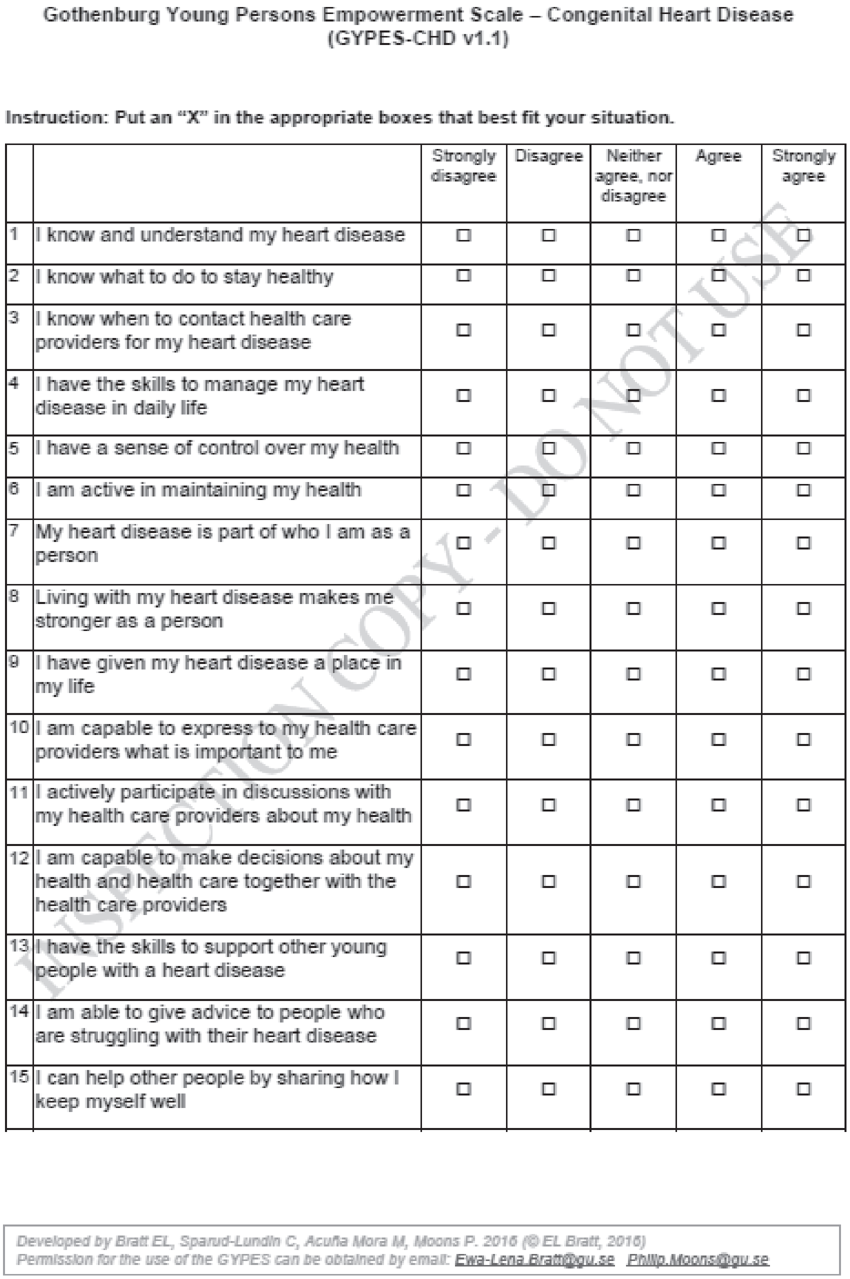
APPENDIX

I Revised version of the Gothenburg Young Persons Empowerment Scale

II Psychometric properties of the questionnaires used in the included studies

III Ethical approvals of the included studies

Appendix I. Revised version of the Gothenburg Young Persons Empowerment Scale



Gothenburg Young Persons Empowerment Scale – Congenital Heart Disease (GYPES-CHD v1.1)

Instruction: Put an "X" in the appropriate boxes that best fit your situation.

		Strongly disagree	Disagree	Neither agree, nor disagree	Agree	Strongly agree
1	I know and understand my heart disease	☐	☐	☐	☐	☐
2	I know what to do to stay healthy	☐	☐	☐	☐	☐
3	I know when to contact health care providers for my heart disease	☐	☐	☐	☐	☐
4	I have the skills to manage my heart disease in daily life	☐	☐	☐	☐	☐
5	I have a sense of control over my health	☐	☐	☐	☐	☐
6	I am active in maintaining my health	☐	☐	☐	☐	☐
7	My heart disease is part of who I am as a person	☐	☐	☐	☐	☐
8	Living with my heart disease makes me stronger as a person	☐	☐	☐	☐	☐
9	I have given my heart disease a place in my life	☐	☐	☐	☐	☐
10	I am capable to express to my health care providers what is important to me	☐	☐	☐	☐	☐
11	I actively participate in discussions with my health care providers about my health	☐	☐	☐	☐	☐
12	I am capable to make decisions about my health and health care together with the health care providers	☐	☐	☐	☐	☐
13	I have the skills to support other young people with a heart disease	☐	☐	☐	☐	☐
14	I am able to give advice to people who are struggling with their heart disease	☐	☐	☐	☐	☐
15	I can help other people by sharing how I keep myself well	☐	☐	☐	☐	☐

INSPECTION COPY - DO NOT USE

Developed by Bratt EL, Sparud-Lundin C, Acuña Mora M, Moons P. 2016 (© EL Bratt, 2016)
Permission for the use of the GYPES can be obtained by email: Ewa-Lena.Bratt@gu.se Philip.Moons@gu.se

Appendix II. Table with psychometric properties of included instrument

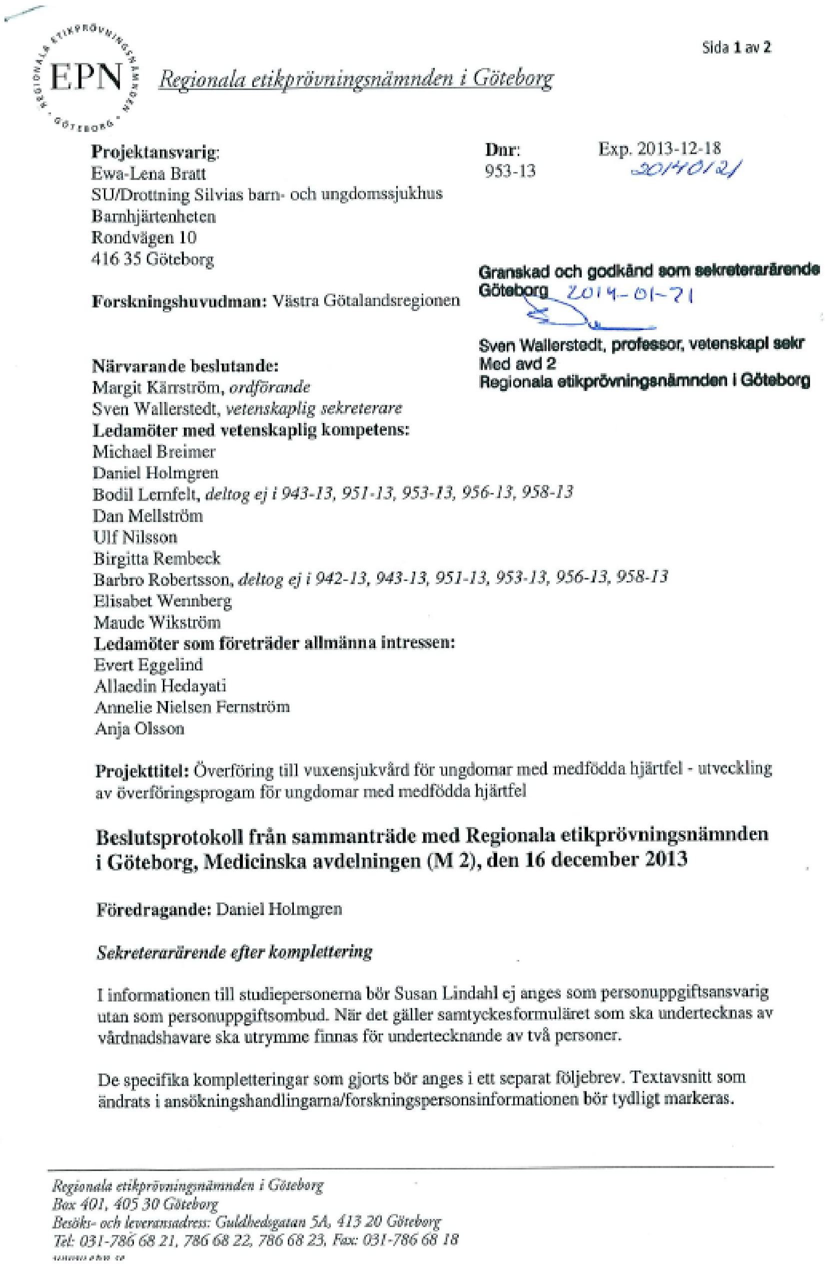
Psychometric properties of the questionnaires used in studies II-IV						
Variable	***Measurements***	***Items***	***Validity***	***Reliability***	***Responsiveness***	***Interpretation***
Primary outcome						
Patient empowerment	Gothenburg Young Persons Empowerment Scale (GYPES) [177]	15	Structural validity supported an adequate model fit of a five factor solution (df: 80; χ2: 222.79, $p<0.0001$; CFI: 0.908; RMSEA: 0.078; SRMR: 0.061) in young persons with type 1 diabetes mellitus. Face validity confirmed in young persons with CHD	Internal consistency confirmed in young persons with CHD (Chronbach's alpha=0.819) and type 1 diabetes mellitus (Chronbach's alpha=0.858)	Floor scores: 0%; Ceiling scores: 2.2%	Total score from 15-75. Higher score reflecting a higher level of patient empowerment
Secondary outcomes						
Transition readiness	Readiness for Transition Questionnaire (RTQ) [194]	26	Validity based on relationships with other variables confirmed in young persons with kidney transplant	Internal consistency confirmed in young persons with kidney transplant; α values over (Chronbach's alpha= 0.79 for overall RTQ score)	NR	Scores from 2-8. Higher scores denote increased readiness for transition

Knowledge on ConHD	Knowledge Scale for Adults with Congenitally Malformed Hearts (KnoCoMH) [191]	19	Validity was confirmed in adults with CHD in relation to discrimination ability, the relationship of the items and the construct of interest	Internal consistency (0.68 (general knowledge), 0.74 (medical treatment), 0.90 (endocarditis prophylaxis), 0.65 (contraceptive and pregnancy)) and test-retest reliability (0.41 (general knowledge), 0.47 (medical treatment), 0.65 (endocarditis prophylaxis), 0.65 (contraceptive and pregnancy)) confirmed in adults with CHD	NR	Scores are calculated by dichotomizing the answers (correct/incorrect) for each domain. Higher scores indicate more knowledge
Health behaviors	Health Behavior Scale-Congenital Heart Disease (HBS-CHD) [189]	15	Item content validity (0.60-1.0), scale content validity index (0.81) and validity based on relationships to other variables confirmed in adolescents with CHD	Reliability coefficient 0.57 for the overall scale. Stability not confirmed for all the subscales	Confirmed in adolescents with CHD by Guyatt's Responsiveness Index (0.95 for the overall scale)	Total health risk score from 0-100. Higher scores denote a higher risky behaviors

Patient-reported health	Pediatric Quality of Life Inventory 4.0 (PedsQL) generic module [192]	23	Convergent validity, measurement invariance and factor structure (df: 80; χ2: 698, p<0.0001; CFI: 0.92; RMSEA: 0.04; SRMR: 0.05) confirmed in pediatric populations	Test-retest reliability, and internal consistency (Chronbach's alpha= 0.90 for the total score) pediatric populations	Minimal clinically important differences reported for young people with CHD was 6.0 for the total score	Scores from 0 to 100. Higher scores indicate a better perceived health status
	Pediatric Quality of Life Inventory 4.0 (PedsQL) cardiac module [193]	27	Convergent validity was confirmed between the PedsQL 4.0 and PedsQL 3.0	Internal consistency measured by Chronbach's alpha was for majority of scales exceeded α values of 0.79	Minimal clinically important differences reported for young people with CHD ranged from 7.6 for symptoms to 12.6 for communication	There is no total score. There are 6 subscales scores, which range from 0 to 100. Higher scores indicate lower problems
Quality of life	Linear analog scale (LAS) [188]	1	Validity based on relationship with Satisfaction with Life Scale found to be highly correlated (ϱ=0.52). Test content confirmed in adults with CHD (100% of patients understood the wording and format)	Test-retest reliability confirmed in adults with CHD (0.65)	Confirmed in adults with CHD (Floor scores= 0%, ceiling scores=2.7%)	Score is from 100 (best imaginable health status) to 0 (worst imaginable health status)

			Validity has been evaluated in adolescents with CHD			
Illness perceptions	Brief Illness Perception Questionnaire (Brief-IPQ) [190]	9	Concurrent, predictive and discriminant validity confirmed in patients with renal disease, asthma, type and 2 diabetes mellitus, in relation to other variables.	Test-retest reliability in renal patients (range from 0.42-0.75)	NR	Total score ranges from 0-8. Higher scores indicate a more threatening view of the disease

Appendix III. Ethical approvals



EPN Regionala etikprövningsnämnden i Göteborg

Sida **1** av **2**

Projektansvarig:
Ewa-Lena Bratt
SU/Drottning Silvias barn- och ungdomssjukhus
Barnhjärtenheten
Rondvägen 10
416 35 Göteborg

Dnr:
953-13

Exp. 2013-12-18
20140121

Forskningshuvudman: Västra Götalandsregionen

Granskad och godkänd som sekreterarärende
Göteborg 2014-01-21

Sven Wallerstedt, professor, vetenskapl sekr
Med avd 2
Regionala etikprövningsnämnden i Göteborg

Närvarande beslutande:
Margit Kärrström, *ordförande*
Sven Wallerstedt, *vetenskaplig sekreterare*
Ledamöter med vetenskaplig kompetens:
Michael Breimer
Daniel Holmgren
Bodil Lernfelt, *deltog ej i 943-13, 951-13, 953-13, 956-13, 958-13*
Dan Mellström
Ulf Nilsson
Birgitta Rembeck
Barbro Robertsson, *deltog ej i 942-13, 943-13, 951-13, 953-13, 956-13, 958-13*
Elisabet Wennberg
Maude Wikström
Ledamöter som företräder allmänna intressen:
Evert Eggelind
Allaedin Hedayati
Annelie Nielsen Fernström
Anja Olsson

Projekttitel: Överföring till vuxensjukvård för ungdomar med medfödda hjärtfel - utveckling av överföringsprogam för ungdomar med medfödda hjärtfel

Beslutsprotokoll från sammanträde med Regionala etikprövningsnämnden i Göteborg, Medicinska avdelningen (M 2), den 16 december 2013

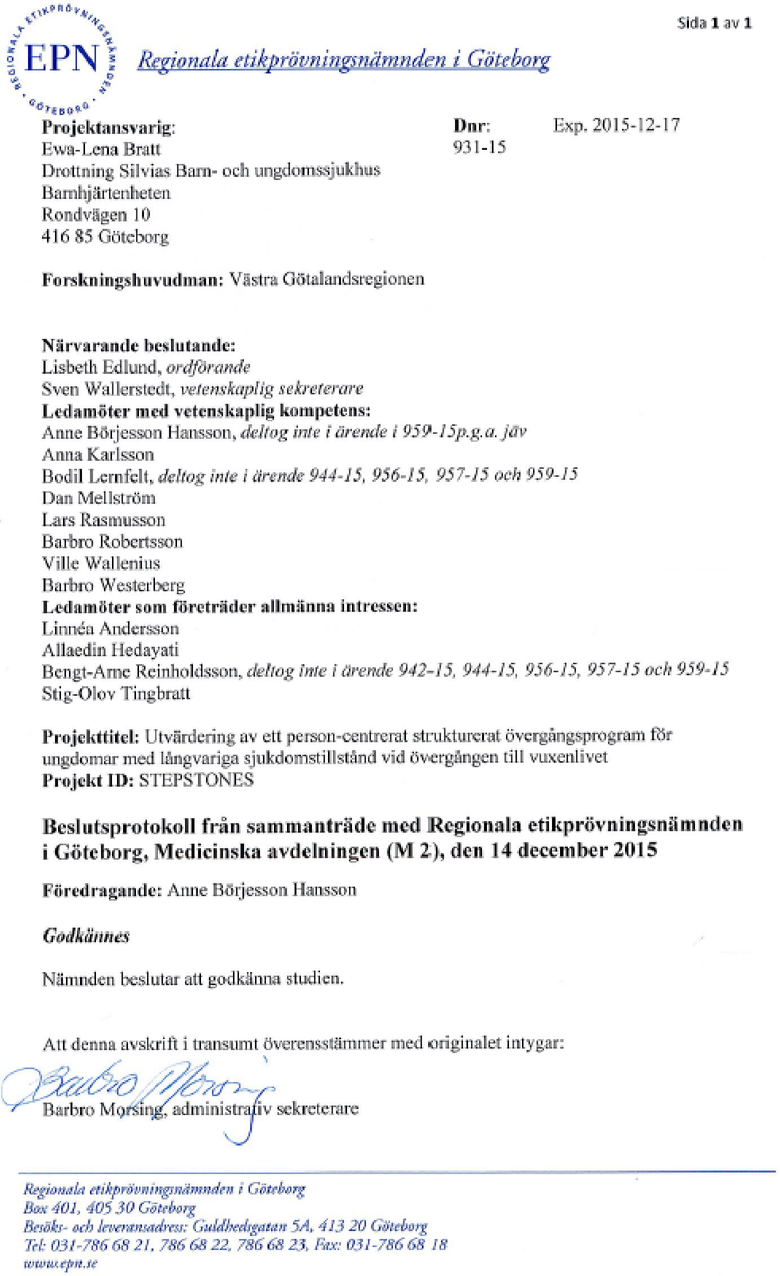
Föredragande: Daniel Holmgren

Sekreterarärende efter komplettering

I informationen till studiepersonerna bör Susan Lindahl ej anges som personuppgiftsansvarig utan som personuppgiftsombud. När det gäller samtyckesformuläret som ska undertecknas av vårdnadshavare ska utrymme finnas för undertecknande av två personer.

De specifika kompletteringar som gjorts bör anges i ett separat följebrev. Textavsnitt som ändrats i ansökningshandlingarna/forskningspersonsinformationen bör tydligt markeras.

Regionala etikprövningsnämnden i Göteborg
Box 401, 405 30 Göteborg
Besöks- och leveransadress: Guldhedsgatan 5A, 413 20 Göteborg
Tel: 031-786 68 21, 786 68 22, 786 68 23, Fax: 031-786 68 18



EPN *Regionala etikprövningsnämnden i Göteborg*

Sida **1** av **1**

Projektansvarig:
Ewa-Lena Bratt
Drottning Silvias Barn- och ungdomssjukhus
Barnhjärtenheten
Rondvägen 10
416 85 Göteborg

Dnr:
931-15

Exp. 2015-12-17

Forskningshuvudman: Västra Götalandsregionen

Närvarande beslutande:
Lisbeth Edlund, *ordförande*
Sven Wallerstedt, *vetenskaplig sekreterare*
Ledamöter med vetenskaplig kompetens:
Anne Börjesson Hansson, *deltog inte i ärende i 959-15p.g.a. jäv*
Anna Karlsson
Bodil Lernfelt, *deltog inte i ärende 944-15, 956-15, 957-15 och 959-15*
Dan Mellström
Lars Rasmusson
Barbro Robertsson
Ville Wallenius
Barbro Westerberg
Ledamöter som företräder allmänna intressen:
Linnéa Andersson
Allaedin Hedayati
Bengt-Arne Reinholdsson, *deltog inte i ärende 942-15, 944-15, 956-15, 957-15 och 959-15*
Stig-Olov Tingbratt

Projekttitel: Utvärdering av ett person-centrerat strukturerat övergångsprogram för ungdomar med långvariga sjukdomstillstånd vid övergången till vuxenlivet
Projekt ID: STEPSTONES

Beslutsprotokoll från sammanträde med Regionala etikprövningsnämnden i Göteborg, Medicinska avdelningen (M 2), den 14 december 2015

Föredragande: Anne Börjesson Hansson

Godkännes

Nämnden beslutar att godkänna studien.

Att denna avskrift i transumt överensstämmer med originalet intygar:

Barbro Morsing
Barbro Morsing, administrativ sekreterare