Høgskulen på Vestlandet

Masteroppgave

MKS590

Predefinert informasjon

Startdato: 07-05-2019 09:00
Sluttdato: 29-05-2019 14:00
Eksamensform: Masteroppgave
SIS-kode: 203 MKS590 1 MÅ 2019 VÅR
Intern sensor: (Anonymisert)

Termin: 2019 VÅR
Vurderingsform: Norsk 6-trinns skala (A-F + Bestått)

Deltaker

Kandidatnr.: 407

Informasjon fra deltaker

Antall ord: 13302
Egenerklæring: Ja

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Sense of coherence and illness perceptions in adults with congenital heart disease

Oppleving av samanheng og oppfatning av sjukdom hjå vaksne med medfødd hjartefeil

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Innleveringsdato: 29.05.2019

Eg stadfestar at arbeidet er sjølvstendig utarbeida, og at referansar/kjeldetilvisingar til alle kjelder som er brukt i arbeidet er oppgitt. Jf. Forskrift om studium og eksamen ved Høgskulen på Vestlandet, § 10.
Acknowledgements

I would like to thank my supervisors Professor Tone M. Norekvål at Western Norway University of Applied Sciences, Bergen, and Professor Philip Moons at KU Leuven, Belgium. Working together with you have been a joy and a great inspiration. The visit to Leuven and the trips to Bergen have been among the highlights.

Without all the participants and collaborators of APPROACH- IS, this study would not be possible. Thank you.

Submission
From this study an article is being planned submitted to the European Journal of Cardiovascular Nursing. The article is therefore written to meet the criteria of this journal. Cross-lagged analysis on the longitudinal data from the Belgian part of the study will be added in the final script sent for publication. The members of the APPROACH- IS and the steering committee will be cited as co-authors as per publication strategy of the APPROACH- IS consortium. The total number of authors will be around 30.
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Sense of coherence and illness perceptions in adults with congenital heart disease.

Abstract

Background: Sense of coherence (SOC) and illness perceptions have been shown to be resources for a better life in people with congenital heart disease (CHD). Understanding the relationship of these modifiable factors could help inform interventions aimed at intentionally improving these.

Aims: To determine the relationship between illness perceptions, SOC, and their sub-dimensions in adults with CHD at an international level.

Methods: This study was a part of Assessment of Patterns of Patient-Reported Outcomes in Adults with Congenital Heart Disease - International Study (APPROACH-IS), a cross sectional study, with 4028 patients from 15 countries. The self-report questionnaires, 13 item Orientation to Life Questionnaire (SOC-13) and Brief Illness Perception Questionnaire (Brief IPQ), were administered and patient characteristics on sex, age, complexity of heart disease, ethnic background, educational level, employment status, marital status, self-reported New York Heart Association (NYHA) assessment, and parental status was collected. Descriptive statistics and correlational analyses were performed.

Results: Pearson product-moment correlation coefficient showed a moderate, negative relationship between illness perceptions and SOC ($r = -.426, n = 3955, p < .01$). A less threatening perception of illness was associated with higher levels of SOC. Multiple regression analysis showed that the following sub-dimensions of illness perceptions predicted SOC: emotional representations ($beta = -.307, p < .001$), coherence ($beta = -.157, p < .001$), consequences ($beta = -.092, p < .001$), treatment control ($beta = -.082, p < .001$), identity ($beta = -.079, p < .001$), and timeline ($beta = -.062, p < .001$).
Conclusion: Illness perceptions appear to be an actionable concept for improving SOC. This approach could be considered when making routines for follow-up of adults with CHD.

Abstrakt

Bakgrunn: Oppleving av samanheng (OAS) og oppfatning av sjukdom (illness perceptions) har blitt vist å vere ressursar for eit godt liv hjå menneske med medfødd hjartefeil. Kunnskap kring samanhengen mellom desse faktorane kan hjelpe lage lage tiltak som kan styrkje pasienten sine ressursar.

Hensikt/ mål: Å bestemme samanhengen mellom oppfatning av sjukdom, OAS, og deira underdimensjonar hjå vaksne med medfødd hjartefeil på internasjonalt nivå.

Metode: Studien er ein del av Assessment of Patterns of Patient-Reported Outcomes in Adults with Congenital Heart Disease - International Study (APPROACH-IS), ein tverrsnittstudie med 4028 deltakarar frå 15 land. Sjølvrapporteringsskjemaa, 13 item Orientation to Life Questionnaire (SOC-13) og Brief Illness Perception Questionnaire (Brief IPQ), samt demografiske data om kjønn, alder, kompleksiteten til hjartefeilen, etnisk bakgrunn, utdanningsnivå, yrkesstatus, sivil status, sjølvrapportert New York Heart Association (NYHA)-klasse og tal på born vart brukt for denne studien. Det vart gjort deskriptiv analyse og analyse av samanheng.

Resultat: Det var ein moderat negativ samanheng mellom oppfatning av eigen sjukdom og OAS, \( r = -0.426, n = 3955, p < 0.01 \). Ei mindre trugande oppfatning av sjukdom hadde samanheng med sterkare OAS. Multippel regresjonsanalyse viste at dei følgjande dimensjonane av oppfatning av sjukdom kunne foreseie OAS: emotional representations \((\beta = -0.307, p < 0.001)\), coherence \((\beta = -0.157, p < 0.001)\), consequences \((\beta = -0.092, p < 0.001)\), treatment control \((\beta = -0.082, p < 0.001)\), identity \((\beta = -0.079, p < 0.001)\), and timeline \((\beta = -0.062, p < 0.001)\).

Konklusjon: Oppfatning av sjukdom synast vere eit handlingsretta konsept for å styrkje OAS. Ei slik tilnærming kan vurderast når det skal lagast rutinar for oppfølging av vaksne med medfødd hjartefeil.
2 Introduction

Congenital heart disease is the most common congenital malformation and occur in slightly less than 1% of all live births (van der Linde et al., 2011). Survival rates 25 years after first surgery are 95%, 89%, and 72% for persons with a simple, moderate and complex CHD respectively (Spector et al., 2018). The mortality rate in the CHD population is still higher than in the normal population, but it has decreased the last decades, resulting in the number of adults with CHD surpassing the number of children with CHD (Khairy et al., 2010; Marelli et al., 2014; McCracken et al., 2018). This has led to increased knowledge about possible medical complications such as arrhythmia, heart failure and endocarditis, in addition to non-cardiac complications related to dysfunction in the kidneys, lungs and liver e.g. (Baumgartner et al., 2010; Lui et al., 2017). There is also a heightened risk of psychosocial problems like depression, anxiety, and loneliness in this group (Kovacs & Utens, 2015). The understanding of CHD as a lifelong and chronic disease along with the combination of improved treatment and knowledge about complications, may explain the interest in patient reported outcomes (PROs) for both children and adults with CHD. The use of PROs have also highlighted the patients subjective understanding and response to disease and treatment as an important part to consider in modern health care (Anker et al., 2014; Rumsfeld et al., 2013). Two such PROs are illness perceptions and sense of coherence (SOC).

SOC is a part of Aron Antonovsky´s theory on salutogenesis, and concerns the individual’s global orientation, a way of perceiving the world as comprehensible, manageable and meaningful (Antonovsky, 1979, pp. 123-124; 1987, pp. 16-19). The theory on illness representation or illness perceptions was created to understand the individual’s thoughts on own disease and how it affects coping and treatment adherence (Leventhal, Meyer, & Nerenz, 1980, p. 10; Leventhal, Nerenz, & Steele, 1984, pp. 219-220). Illness perceptions and SOC have both been studied in the adult congenital heart disease (ACHD) group and both are related to health outcomes (Eaton, Wang, & Menahem, 2017; Muller, Hess, & Hager, 2014; O'Donovan, Painter, Lowe, Robinson, & Broadbent, 2016; Rassart et al., 2017; Schoormans et al., 2014). A possible relationship between SOC and illness perceptions has to our current knowledge not been studied in the ACHD group. Previous research on other groups have found a relationship between the two (Alsen & Eriksson, 2016; Langeland et al., 2013; Lo Sterzo & Orgeta, 2017). Interestingly, sub-dimensions of illness perceptions predict changes
in SOC (Langeland et al., 2013). Interventions aimed at improving illness perceptions have shown promising results (Broadbent, Ellis, Thomas, Gamble, & Petrie, 2009; Christensen, Frostholm, Ornbol, & Schroder, 2015; Dalili & Bayazi, 2019; Rahimi Kordshooli, Rakhshan, & Ghanbari, 2018; Siemonsma et al., 2013). With illness perceptions appearing as an actionable concept, it could represent a possible pathway to increase SOC. A better understanding of these PROs can be used to improve treatment and identify modifiable factors for better health in this patient population (Moons & Norekval, 2006; Rassart et al., 2017).

3 Background

3.1 Living with congenital heart disease

Children and adults with CHD form a heterogeneous group. In addition to the obvious variation in age, there are major differences between the heart diseases and how they affect the functioning of the heart and the individual’s life. In the United States, one fourth of the children with CHD will undergo invasive treatment during their first year of life (Mozaffarian et al., 2016). Some children and adults will need several invasive procedures, lifelong treatment and follow up. Others have simple defects that spontaneously heal or require short term follow up after surgery (Baumgartner et al., 2010; Warnes et al., 2008).

Children and adolescents with CHD have an increased risk of developmental delays, such as reduced cognitive and metacognitive abilities, fine and gross motor problems and lack of emotional control (Gerstle, Beebe, Drotar, Cassedy, & Marino, 2016; Marino et al., 2012; Sarrechia et al., 2016). However, studies on children in primary school find that these children consider themselves to be as competent as their peers. Being aware of their physical limitations, they instead focus on their own possibilities and their ability to adapt, in order to participate and be together with other children (Birks, Sloper, Lewin, & Parsons, 2006; Bjorbaekmo & Engelsrud, 2008; Miatton, De Wolf, Francois, Thiery, & Vingerhoets, 2007). Studies on adolescents and young adults find more contrasting experiences, such as wanting to be normal, but still being able to appreciate living with a chronic disease (Apers et al., 2016; Berghammer, Dellborg, & Ekman, 2006). Even among those with Fontan circulation due to an uni-ventricular heart, considered to be one of the most complex types of CHD, focus
on possibilities and happiness over being oneself have been discovered to be major themes (Berghammer, Brink, Rydberg, Dellborg, & Ekman, 2015). In adults with CHD, this struggle between wanting to be normal and accepting one’s own life as different, is found to be even more explicit (Claessens et al., 2005). Defining oneself as a survivor, and being able to recognize the positive influence of living with a CHD, despite the obvious negative consequences of the illness, illustrates the struggle of normalisation (Keir, Bailey, Lee, Kovacs, & Roche, 2018; Overgaard, King, Christensen, Schrader, & Adamsen, 2013). In a study concerning how adults with CHD experience physical activity, the willingness and ability to participate in physical activity was described as balancing a slackline (Bay, Lamas, Berghammer, Sandberg, & Johansson, 2018). This balancing act between the wish to be normal on one side, and accepting illness as part of oneself on the other, could be of interest in the context of SOC and illness perceptions. They could both be seen as factors or personal characteristics affecting the ability to balance, to be satisfied with one’s own life, to have a good quality of life (QoL).

The first study on QoL in people with CHD was published in 1974. Since then the number of studies have been increasing, with the subject gaining more interest the last decade. The total number of studies on QoL in children and adults with CHD reached 234 in 2014, with more than 30 studies in 2014 alone (Bratt & Moons, 2015). Presumably all this research should have given us a clear understanding of this topic. The answer however, is not that straightforward. The different definitions of QoL and methods used vary to such a degree that comparing findings give little meaning (Apers, Luyckx, & Moons, 2013; Bratt & Moons, 2015; Moons, Budts, & De Geest, 2006). Some studies on children and adolescents with CHD find a better QoL for this group compared to peers (Apers, Moons, et al., 2013; Reiner, Oberhoffer, Ewert, & Muller, 2018). Other studies find a worse QoL for this group (Amedro et al., 2015; Mellion et al., 2014; Werner, Latal, Valsangiacomobuechel, Beck, & Landolt, 2014). Studies on adults with CHD also find both worse and better, as well as similar QoL compared to a control group (Apers, Luyckx, & Moons, 2013). Even though reaching a conclusion regarding QoL is not possible at the present, taking into account experiences of living with CHD, interventions aimed at enhancing coping with this chronic condition should be of obvious value.

Adolescence and childhood have been suggested as the ideal time for interventions targeting illness perceptions and SOC, because identity and future orientation start developing at this
age (Fortenberry et al., 2014; Luyckx, Goossens, Van Damme, & Moons, 2011; Massey, Gebhardt, & Garnefski, 2008). This does of course not mean interventions aimed at adults are futile. More knowledge on medical complications, psychosocial problems and negative and stressful experiences in the CHD group, in addition to a heightened risk of developmental delays, have highlighted the need for multidisciplinary and lifelong care, as well as care not only for the disease, but for the whole patient.

3.2 Theoretical framework

3.2.1 Salutogenesis and sense of coherence

The American sociologist Aron Antonovsky was interested in how people stay healthy. During the late 60s and early 70s, he was taking part in a study on adaptation in women after menopause. Some of the participants in this study were World War II concentration camp survivors. Results did unsurprisingly reveal that most of these women showed poor adaptation. A minority of women did however claim to adapt well and have a reasonably good life even after this terrible experience. In a stroke of genius, Antonovsky turned his attention to this fact and started wondering how this could be. Where did this strength come from? The salutogenic question started taking form (Antonovsky, 1979, pp. 5-7).

What brings health? To answer this, Antonovsky developed the theory on salutogenesis and SOC (Antonovsky, 1979, 1987). Antonovsky did not think of salutogenesis as a substitute for the traditional paradigm on pathogenesis, but as a complementary theory (Antonovsky, 1979, pp. 35-36, 55). He thought of a person´s health being somewhere along a continuum ranging from dis-ease to ease (health), instead of being either ill or healthy. The idea was that this perspective should take into account all factors pushing a person´s health either way on the continuum, instead of narrowing down the search for causes for a specific disease (Antonovsky, 1979, pp. 39, 56-57).

SOC was Antonovsky´s answer to the salutogenic question. SOC represent a person´s view of the world as comprehensible, manageable and meaningful (Antonovsky, 1987, pp. 16-19). Antonovsky (1987, p. 152) suggested that a person with a strong SOC had a better chance of maintaining or even improving his health in the face of adversity, than a person with a weak
SOC. This leads to the question on whether SOC can be improved intentionally. Antonovsky (1987, pp. 123-126) proposed this is the case. Upon meeting patients, practitioners and nurses could improve the patient’s SOC not only temporarily, but also with a more long-lasting effect (Sullivan, 1989). If the patient experiences the meeting with her practitioner as meaningful and comprehensible there is a possibility for an enhanced SOC.

According to Antonovsky, another factor affecting health, is general resistance resources (GRRs). A general resistance resource is a characteristic of a person, group or society that can help manage tension, when facing a stressor. Effective management of tension prevents stress and the possibility of movement towards the dis-ease end of the continuum. Successful management of tension even has the possibility of being salutary and could contribute to the actualisation of potential resources, thus helping explain why humans seek stressful situations voluntary (Antonovsky, 1979, pp. 95-96, 99). GRRs cover a broad spectrum; from physical conditions such as the immune system, to material resources (money), knowledge, intelligence, social support, personal attitudes and religion (Antonovsky, 1979, pp. 103-118). Antonovsky defined a stressor as a demand on the organism that require a non-automatic response or action. This demand could come from the external or internal environment (Antonovsky, 1979, p. 72). He considered these stressors to be present in varying degrees more or less all the time in everybody’s lives. Because of this he thought it more important to emphasise GRRs and their part in the salutogenic question.

3.2.2 Illness perceptions and related aspects to Antonovsky’s theory

Illness perceptions, originally named illness representations, was a part of Leventhal et al.’s (1980, pp. 10, 18; 1984, pp. 219-224) theory on how people cope with health threats and adhere to medical treatment. The general idea was that illness perceptions are formed by a person´s symptoms, information from external sources and personal experience with disease (Leventhal et al., 1980, p. 18). Together these form a cognitive framework that the patient construct to make sense of own illness and how to cope with it. Illness representations initially consisted of four major components: identity of the illness, beliefs on cause, consequences and beliefs on duration. Additionally, beliefs on control or a cure, and emotions are also considered to play an important role (Leventhal et al., 1984, pp. 223, 232-233; Petrie & Weinman, 2006). Both the abstract labels and the concrete symptoms that form these
attributes have effect on a person’s behaviour and coping. These coping mechanisms are constructed and evaluated repeatedly, thus changing experience and knowledge (Leventhal et al., 1984, pp. 25-26, 221, 232). In order to create a stable change in what the authors call the self-regulatory system, a patient need to know what to expect, both on a concrete (symptoms) and abstract (knowledge) level, and how to act in certain situations, for example in recognising symptoms and knowing what to do if they appear. Finally, the patient need concrete criteria in order to evaluate the outcomes (Leventhal et al., 1980, pp. 11, 20, 23; Leventhal et al., 1984, p. 242). From this holistic approach, it becomes clear that a change in illness perceptions is not achieved at a whim. Antonovsky (1987, p. 124) held the same for true regarding SOC.

The two theories emphasise the personal perspective on either illness or health. Both theories diverge from the biomedical model. This model being concerned with the breakdown of the body and the separation of body and mind (Giddens & Sutton, 2013, p. 442). Perhaps it makes for a small wonder that these theories or models of understanding health and illness was made by a sociologist (Antonovsky), psychiatrist (Leventhal) and psychologist (Nerenz). Both theories have its roots within the tradition of symbolic interactionism, concerned with how the individual interpret the social world and its own disease and health situated in this (Giddens & Sutton, 2013, p. 453).

There are seemingly also other related aspects between some of the components of the two theories of SOC and illness perceptions. These are most obvious in Leventhal et al.’s (1980, p. 23; 1984, pp. 242-244) explanation of change in the self-regulatory system. Knowing what symptoms to expect have a similarity to the comprehensibility component of SOC, perceiving inner or outer stimuli as making sense and not stemming from chaos. Knowing how to act in certain situations and manageability, having resources to deal with stimuli also seems to reflect sides of the same coin. Regarding the third component of SOC, meaningfulness, it is difficult to find one particular parallel in the theory of illness perceptions. A person’s view of a challenge as worthy of engagement, seems to be intertwined with all the major components of illness representations (Antonovsky, 1987, pp. 16-19; Leventhal et al., 1984, pp. 242, 247). However, these theoretical speculations suggest there could be a relationship between SOC and illness perceptions.
3.3 Previous research

The relationship between SOC and other PROs have been investigated in children and adults with CHD. A stronger SOC is related to better social, emotional, and school functioning (Apers, Luyckx, Rassart, et al., 2013). There is increasing evidence of a relationship between SOC and QoL in children and adolescents with CHD (Apers, Luyckx, et al., 2015; Apers, Luyckx, Rassart, et al., 2013; Apers, Moons, et al., 2013; Neuner et al., 2011; Wang, Hay, Clarke, & Menahem, 2014). Also among adults with CHD there is some evidence of this relationship (Eaton, Wang, & Menahem, 2017; Muller, Hess, & Hager, 2014). Young adults with a strong SOC have been found to have more knowledge on their disease, feel sense of control, have good social support and experience negative life events as a meaningful challenge (Apers et al., 2016). In the study, these findings are understood in the context of the components of SOC, comprehensibility, manageability and meaningfulness. The authors suggest that SOC can be improved by increasing knowledge, and by empowering and involving the individual in decision-making (Apers et al., 2016). With the evidence of a relationship between SOC and QoL, interventions aiming at intentionally enhancing SOC ought to be a focus of future research (Moons & Norekval, 2018).

Persons with the same disease can have very different perceptions of their illness. These perceptions are related to a wide variety of health outcomes such as survival, symptoms, depression and QoL (Chilcot, Wellsted, & Farrington, 2011; Dickens et al., 2008; Kaptein et al., 2015; van der Have et al., 2013). In the ACHD group, illness perceptions are related to QoL and symptoms of depression and anxiety (O'Donovan, Painter, Lowe, Robinson, & Broadbent, 2016; Rassart et al., 2017; Schoormans et al., 2014). Individuals with Fontan circulation, a complex CHD, have more negative illness perceptions than individuals with isolated atrial septal defect (ASD) and ventricular septal defect (VSD) (Holbein et al., 2018).

There is limited evidence of a relationship between SOC and illness perceptions. Strong negative associations have been found between SOC and illness perceptions in a group of patients one year after myocardial infarction. (Alsen & Eriksson, 2016). A correlation between SOC and the sub-dimensions: coherence, consequences, and emotional representations of illness perceptions have been found in a group of patients with psoriasis.
Also in this study were worse perceptions of illness related to lower scores for SOC. Illness coherence, consequences, and emotional representations predicted changes in SOC from baseline to follow up (Langeland et al., 2013). In caregivers of relatives with dementia, a similar relationship between SOC and dimensions of illness perceptions, to that of patients with psoriasis was found (Langeland et al., 2013; Lo Sterzo & Orgeta, 2017). Together these findings suggest that sub-dimensions of illness perceptions might be natural targets for interventions aiming to change not only illness perceptions but also SOC.

In order to increase our understanding of the relationship between illness perceptions and SOC, this association needs to be investigated in larger international samples.

4 Aim

To further explore potential actionable factors within a salutogenic framework, the aim of the present study was to determine the relationship between illness perceptions, SOC, and their sub-dimensions in adults with CHD at an international level.

5 Methods

5.1 Design and setting

This study is a part of Assessment of Patterns of Patient-Reported Outcomes in Adults with Congenital Heart Disease – International Study (APPROACH-IS). APPROACH-IS is a cross-sectional study with participants from 15 countries across 5 continents. These countries include Argentina, Australia, Belgium, Canada, France, India, Italy, Japan, Malta, Norway, Sweden, Taiwan, the Netherlands, and the United States. A standardized protocol was developed and the study was conducted in collaboration with the International Society for Adult Congenital Heart Disease (ISACHD) (Apers, Kovacs, et al., 2015).
5.2 Sample

In this study, 4028 adults with CHD, from 24 participating centres, were enrolled. The inclusion criteria were: 1) diagnosis of CHD, defined as a structural abnormality of the heart or intra-thoracic great vessels that is present at birth and is actually or potentially functionally significant, 2) 18 years of age or older, 3) diagnosis established before the age of 10 (i.e., before adolescence to warrant sufficient experience of living with CHD), 4) continued follow-up at CHD centre or included in a national/regional registry and, 5) physical, cognitive and language capabilities required to complete the self-report questionnaires. Exclusion criteria were: 1) prior heart transplantation, 2) primary pulmonary hypertension or, 3) impaired cognitive abilities.

5.3 Measurements

Two self-report questionnaires were used to measure SOC and illness perceptions. 

SOC-13

The 13-item orientation to life questionnaire (SOC-13) was used to measure SOC. A semantic differential scale ranging from 1 (very seldom or never) to 7 (very often) with a total score from 13 to 91, was used to measure comprehensibility ("Has it happened in the past that you were surprised by the behaviour of people whom you thought you knew well?, Do you have the feeling that you are in an unfamiliar situation and don’t know what to do?, Do you have very mixed-up feelings and ideas?, Does it happen that you experience feelings that you would rather not have to endure? When certain events occurred, have you generally found that: you overestimated or underestimated their importance- you assessed the situation correctly?")(five items), manageability ("Has it happened that people you counted on disappointed you?, Do you have the feeling that you are being treated unfairly?, Many people, even those with a strong character, sometimes feel like losers in certain situations. How often have you felt this way in the past?, How often do you have feelings that you are not sure you could control?")(four items) and meaningfulness ("Do you have the feeling that you really don’t care about what is going on around you?, Until now your life has had: no clear goals - very clear goals and purpose?, Doing the things you do everyday is: a source of deep
pleasure and satisfaction - a source of pain and boredom?, How often do you have the feeling that there is little meaning in the things you do in your daily life?" (four items). A higher score is implying a stronger SOC.

The SOC scale has been found to have a high internal consistency with a Cronbach alpha score ranging from 0.70 to 0.92 (Eriksson & Lindstrom, 2005). For this study the Cronbach alpha score was 0.85 for SOC-13. The strong internal consistency supports the argument that the different items in the scale measure the same construct.

**Brief IPQ**

The brief illness perception questionnaire (Brief IPQ) uses a 8 item scale ranging from 0 to 10, with a higher score indicating a more threatening view of the illness. The 8 items include consequences ("How much does your illness affect your life?"), timeline ("How long do you think your illness will continue?"), personal control ("How much control do you feel you have over your illness?"), treatment control ("How much do you think your treatment can help your illness?"), identity ("How much do you experience symptoms from your illness?"), concern ("How concerned are you about your illness?"), illness coherence ("How well do you feel you understand your illness?"), and emotions ("How much does your illness affect you emotionally?, e.g., does it make you angry, upset, or depressed?"). In addition, there is an item on perceived causal factors. This last item is not rated from 0 to 10, but instead ask the respondent to rate the three most influential factors causing the illness.

### 5.4 Data collection

The eligible participants were selected either randomly from the institution’s database or consecutively approached at the outpatient clinics. The participants selected randomly were mailed a package containing an information letter, a copy of the survey package, an informed consent form (if necessary) and an addressed, pre-stamped return envelope. Mail or telephone reminders were used to increase response rates. Patients at the outpatient clinics could return the forms during their visit or later via mail. Data from medical records on CHD diagnosis, disease complexity, surgical history, history of anxiety, mood disorder or other psychiatric
diagnosis were collected and recorded by a member of the research team. Data collection commenced in April 2013 and were completed in March 2015.

5.5 Statistical analysis

Descriptive statistics was used to calculate SOC and illness perceptions. Correlation analyses was performed to compute a possible relationship between SOC, illness perceptions and their sub-dimensions, measuring Pearson product-moment correlation coefficient. Due to the fact that sub-dimensions of illness perceptions have been shown to predict changes in SOC, hierarchical multiple regression was performed with these sub-dimensions as independent variables (Langeland et al., 2013). It was adjusted for sex, age, complexity of heart defect, country of origin and self-reported NYHA-class. Histograms, scatterplots, normal P-P plots were generated, and collinearity diagnostics performed to ensure no violation of normality, linearity, homoscedasticity, outliers, independence of residuals, multicollinearity or singularity. The data was analysed using IBM SPSS Statistics, version 25 (Armonk, NY).

6 Ethical considerations

Autonomy is included in the first ethical principle of the Belmont report, Respect for persons. Being autonomous means each person has the right of self-determination. If a person is to give informed consent, three elements must be considered. The person needs sufficient information. This information must then be comprehended by the person. Finally, consent must be given voluntarily. The first ethical principle of the report also states that persons with reduced capacity for autonomy has the right to protection ("The Belmont report : ethical principles and guidelines for the protection of human subjects of research," 1979). According to this, all participants were handed written information and a written consent form if required. Patients with impaired cognitive abilities were excluded.

Beneficence is the second principle of the Belmont report. It states that researchers have an obligation not to harm, non-maleficence, and to maximize benefits and minimize harms ("The Belmont report : ethical principles and guidelines for the protection of human subjects of research," 1979).
research," 1979). The benefit for the participants is that by providing information and increasing knowledge, treatment can ultimately get better. In order to reduce costs for participants, the survey forms were handed in at their local clinic or mailed via pre-stamped envelopes. In order to maintain participant confidentiality no personal health information were sent from participating centres to the coordinating centre. A patient identification code was created for each participant. This was done to ensure that data was not exploited, and used against participants.

*Justice* is the final principle in the Belmont report. It concerns the distribution of benefits and burdens of research and how subjects are selected as eligible participants. In this study participant selection from eligible patients was done randomly from each institution’s database or through consecutively approaching them in the outpatient clinic.

The study protocol was approved by the Institutional Review Board of the University Hospitals Leuven/ KU Leuven. Participating centres gained ethical approval if required. The Norwegian part of the study was approved by the Regional committees for medical and health research ethics (ref.nr.: 2013/1332). The study follows the recommendations of the Declaration of Helsinki II (Apers, Kovacs, et al., 2015; "World Medical Association Declaration of Helsinki: ethical principles for medical research involving human subjects," 2000).

7 Results

7.1 Patient characteristics

Among the 4028 participants, the median age was 32 years and 53% were women. A majority of 74% were white or Caucasian, 64% had part-time or full-time work, 51% were married or living with a partner, and 60% had no children. Fifty-four percent reported being in NYHA-class I, and 49% had a CHD of moderate complexity (Table 1 in article).
7.2 Relationship between illness perceptions and SOC

There was a moderate, negative relationship between illness perceptions and SOC ($r = -.426$, $n = 3955$, $p < .001$). Higher levels of SOC was associated with lower levels of illness perceptions.

There was a moderate negative relationship between all three components of SOC and illness perceptions. For comprehensibility ($r = -.386$, $n = 3871$, $p < .001$), for manageability ($r = -.365$, $n = 3899$, $p < .001$), and for meaningfulness ($r = -.366$, $n = 3894$, $p < .001$).

There was also a weak to moderate negative relationship between SOC and the following subdimensions of illness perceptions: consequences ($r = -.364$, $n = 3934$, $p < .001$), identity ($r = -.346$, $n = 3907$, $p < .001$), concern ($r = -.257$, $n = 3925$, $p < .001$), coherence ($r = -.252$, $n = 3918$, $p < .001$), and emotional representation ($r = -.446$, $n = 3926$, $p < .001$).

Table 2

Correlation matrix of SOC, illness perceptions, and their sub-dimensions

<table>
<thead>
<tr>
<th></th>
<th>SOC</th>
<th>SOC1</th>
<th>SOC2</th>
<th>SOC3</th>
<th>IPQ</th>
<th>Consequences</th>
<th>Timeline</th>
<th>Personal control</th>
<th>Treatment control</th>
<th>Identity</th>
<th>Concern</th>
<th>Coherence</th>
<th>Emotional representations</th>
</tr>
</thead>
<tbody>
<tr>
<td>SOC</td>
<td>-</td>
<td></td>
<td></td>
<td></td>
<td></td>
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<td></td>
<td></td>
</tr>
<tr>
<td>SOC1</td>
<td>.907**</td>
<td>-</td>
<td></td>
<td></td>
<td></td>
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<td></td>
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<td></td>
<td></td>
<td></td>
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<tr>
<td>SOC2</td>
<td>.817**</td>
<td>.709**</td>
<td>-</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
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<td></td>
<td></td>
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</tr>
<tr>
<td>SOC3</td>
<td>.822**</td>
<td>.570**</td>
<td>.579**</td>
<td>-</td>
<td></td>
<td></td>
<td></td>
<td></td>
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<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>IPQ</td>
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<td>-.386**</td>
<td>-.365**</td>
<td>-.366**</td>
<td>-</td>
<td></td>
<td></td>
<td></td>
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<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Consequences</td>
<td>-.364**</td>
<td>-.318**</td>
<td>-.306**</td>
<td>-.308**</td>
<td>.349**</td>
<td>-</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Timeline</td>
<td>.009</td>
<td>.001</td>
<td>.002</td>
<td>.001</td>
<td>.457**</td>
<td>.349**</td>
<td></td>
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</tr>
<tr>
<td>Personal control</td>
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<td>-.310**</td>
<td>-.306**</td>
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<td>-.338**</td>
<td>.460**</td>
<td></td>
<td></td>
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<td></td>
</tr>
<tr>
<td>Treatment control</td>
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<td>-.315**</td>
<td>-.316**</td>
<td>-.314**</td>
<td>.314**</td>
<td>.461**</td>
<td>.463**</td>
<td>.462**</td>
<td></td>
<td></td>
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<td></td>
</tr>
<tr>
<td>Identity</td>
<td>-.346**</td>
<td>-.299**</td>
<td>-.303**</td>
<td>-.303**</td>
<td>.726**</td>
<td>.569**</td>
<td>.554**</td>
<td>.212**</td>
<td>.886**</td>
<td></td>
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</tr>
<tr>
<td>Concern</td>
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<td>-.255**</td>
<td>-.318**</td>
<td>-.192**</td>
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<td>.475**</td>
<td>.435**</td>
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<td>.740**</td>
<td></td>
<td></td>
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</tr>
<tr>
<td>Coherence</td>
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<td>-.231**</td>
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<td>-.244**</td>
<td>.321**</td>
<td>.892**</td>
<td>.806**</td>
<td>.197**</td>
<td>.386**</td>
<td>.479**</td>
<td>.658**</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Emotional representations</td>
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<td>-.415**</td>
<td>-.398**</td>
<td>-.555**</td>
<td>.741**</td>
<td>.826**</td>
<td>.322**</td>
<td>.226**</td>
<td>.855**</td>
<td>.596**</td>
<td>.370**</td>
<td>.322**</td>
<td></td>
</tr>
</tbody>
</table>

**. Correlation is significant at the 0.001 level (2-tailed).
*. Correlation is significant at the 0.05 level (2-tailed).

SOC1= comprehensibility, SOC2= manageability, SOC3= meaningfulness
7.3 Relationship between illness perceptions sub-dimensions and SOC with sub-dimensions

Table 2 in the corresponding article presents the results of the hierarchical multiple regression analysis. After controlling for sex, age, complexity of heart defect, country of origin and self-reported NYHA-class, illness perceptions sub-dimensions explained an additional 18% of variance in SOC ($R^2$ change $= .180$, $F$ change $= 115.501$, $p < .001$). The model as a whole explained 29% of variance in SOC ($F = 114.290$, $p < .001$). Six of the sub-dimensions of illness perceptions were statistically significant as predictors for SOC, comprehensibility, manageability and meaningfulness. Of the sub-dimensions emotional representations ($beta = -.307$, $p < .001$), coherence ($beta = -.157$, $p < .001$), and consequences ($beta = -.092$, $p < .001$) recorded the highest beta values.

8 Discussion

8.1 Discussion of results

This cross-sectional study examined the association between illness perceptions, SOC and sub-dimensions in adults with CHD. It is to our knowledge the first study to do this. The results show that there was a moderate negative relationship between illness perceptions and SOC. In other words, that worse illness perceptions were related to a lower SOC. The fact that the strength of the relationship was moderate might not be surprising. After all, SOC represent a fairly stable and global view on the world, whereas illness perceptions represent a more contemporary view of a specific disease or condition. The two instruments measure two different experiences. The relationship found simply states that the two are somehow connected and that they possibly affect each other.

The difference between the three dimensions of SOC and their relationship to illness perceptions were small, with comprehensibility having the strongest relationship. In patients with myocardial infarction, a stronger relationship between the subscales of SOC and illness perceptions was found, but also in this study did comprehensibility account for the largest variation in illness perceptions, explaining 32% (Alsen & Eriksson, 2016). Even though the
authors of this study consider illness perceptions of fatigue a chronic condition one year after infarction, the question of whether the length of the disease or condition affects the relationship naturally raises. The discussion on splitting SOC into subscales in empirical studies follows in the chapter on measurement issues.

Concerning the correlation between SOC and the subscales of illness perceptions our findings are similar to that of previous research. Emotional representations and SOC showed the strongest relationship in all three studies (Langeland et al., 2013; Lo Sterzo & Orgeta, 2017). Illness coherence, consequences and emotional representations predicted improvement in SOC after a patient education programme. The possibility to consider illness coherence and emotional representations as components of GRRs was thus suggested (Langeland et al., 2013). This would imply that at least parts of illness perceptions are important in managing tension and that it is connected to SOC.

The relationship between total SOC and the different items in Brief IPQ have interesting parallels with findings from interviews with adolescents with CHD and their measured SOC. The participants with a strong SOC experienced less stress than those with a lower SOC (Apers et al., 2016). This is in line with our finding of the relationship between emotional representations and SOC. Antonovsky (1987, p. 149) hypothesised that a person with a strong SOC would indeed experience different emotions than a person with a weak SOC in response to the same stressor. After emotional representations, illness coherence had the highest beta score. This finding is in line with previous research showing a relationship between the two, and the predictive ability of coherence on SOC (Apers et al., 2016; Langeland et al., 2013). It has been proposed that illness coherence is related to SOC through the restructuring of illness meaning, thus disrupting or contributing to coherence between experience, and past and new understanding of own illness (Baarnhielm, 2005). This idea has a parallel to the finding that comprehensibility is more important than meaningfulness and manageability for changes in SOC, leading to the possible suggestion that a high comprehensibility is a prerequisite for a high meaningfulness (Alsen & Eriksson, 2016; Bergman, Malm, Ljungquist, Bertero, & Karlsson, 2012). This is in opposition to Antonovsky’s (1987, pp. 21-22) hypothesis that meaningfulness is the most important factor of the three. It is worth noting that coherence in our study is self-perceived. It does not necessarily reflect good knowledge and understanding. A major point however, is that illness coherence is a resource for SOC.
More perceived consequences and symptoms (identity) were related to lower SOC. These findings can be seen as a parallel to more consequences and symptoms being related to poorer QoL in adults with CHD (O'Donovan et al., 2016; Rassart et al., 2017; Schoormans et al., 2014). This is in contradiction to experiences of adolescents with CHD, where the participants with a strong SOC reported more symptoms and greater consequences than those with a weak SOC (Apers et al., 2016). The dynamic suggested at play, is that in order to perceive the challenge as worthy of engagement, the stressor must also be considered as significant (Helgeson, Reynolds, & Tomich, 2006). In other words, the experienced impact of disease or disaster, affect the effort seen as meaningful to invest in solving the problem. This is then considered as an expression of a cognitive process rather than distress. A possible, but somewhat unsatisfactory explanation to the opposing results, is that the wording in the Brief IPQ causes the answer to reflect distress instead of cognitive processing and acceptance of the condition. Another explanation could be that these experiences changes from adolescence to adulthood. The duration of the illness might actually affect the relationship between identity, consequences and SOC in adults with CHD differently than for adolescents with CHD. Time passed since trauma (i.e. cancer, war, natural disasters) play an important role in moderating the effect of benefit of health outcomes (Helgeson et al., 2006). In our study a more chronic perception of timeline was in fact related to a stronger SOC. Living with CHD could very well prove to alter the relationship between SOC, illness identity and illness consequences differently from living after a traumatic life event. Leventhal et al. (1984, p. 236) considered illness representations to be more frequently shaped by our understanding of illness as an acute condition instead of a chronic one. A gradual change in perception of timeline from acute to a more chronic view of illness could very well explain the different finding in adults with CHD. Suggesting a mechanism behind the change of the relationship between illness consequences, identity and SOC from adolescence to adulthood is beyond the aim of this thesis.

The finding that there was no relationship between personal control and SOC is somewhat surprising. The results did after all show a relationship between treatment control and SOC. The question then becomes if personal control of illness is clearly distinguishable from an overall sense of control and therefore does not affect SOC. This appears counterintuutive. Adolescents with CHD and a strong SOC experienced a higher degree of personal control than those with a lower SOC. Those with a lower SOC also reported more worries (Apers et al., 2016). This also contradicts our finding that illness concern is not related to SOC.
Comparing our results with findings from interviews with adolescents with CHD is of course far from unproblematic. The divergence on what appears to be central parts of the theory of SOC and salutogenesis is still somewhat surprising.

Of the 8 items in Brief IPQ, the following 6 was found to have a significant relationship with SOC: consequences, timeline, treatment control, identity, coherence, and emotional representations. Personal control and concern was not related to SOC. Although scarce, the existing evidence points to a relationship between SOC and illness perceptions and that sub-dimensions of illness perceptions predict changes in SOC (Alsen & Eriksson, 2016; Langeland et al., 2013; Lo Sterzo & Orgeta, 2017). Together with our findings, this suggests that interventions aimed at improving the 6 mentioned sub-dimensions of illness perceptions could lead to an enhanced SOC.

8.2 Discussion of methods

8.2.1 Methodological issues

There are several issues regarding the design and method of this study. First, causality cannot be inferred due to the cross-sectional design. Even though there is a relationship between the variables, both were measured at the same time, making temporality an issue. It is not possible to decide if one of the variables precede the other and therefore is causing the effect on the other. Second, the pragmatic inclusion of participating centres, including one centre from most countries might have an effect on representativeness. The 24 participating centres in the study were chosen because of the size of their patient population and resources to collect data. It is possible that the selected centres, because of their size, resources and willingness to participate, somehow affects attending patients differently from smaller and less resourceful centres. The Norwegian centre selected, is the only centre in Norway performing surgical treatment of these patients, and is therefore a natural choice. The selection of centres do however raise questions about the generalizability of the results and makes it hard to conclude whether differences observed are related to country or the actual centres. Also of notice is the fact that no centre from the African continent participated in the study. With no centre from the African continent, and about 2/3 of participating centres from western industrialised countries, the sample is probably biased if the target population is meant to be adults with congenital heart disease across the globe. Even though the pragmatic
choice of centres can be argued with, it has the strength that centres from non-western countries were deliberately selected. Thus it can be argued that the population in these countries benefits from the study. Still, this could have been achieved even better, had more centres been located in non-western countries. Third, further selection bias cannot be ruled out because persons physically or mentally incapable of filling out self-report questionnaires were excluded from the study. There is the chance that nonresponse bias will affect results. With the in-clinic continuous approach there is no record of response rate or possibility to compare background data from responders and non-responders. A comparison between participants and non-participants from the Swedish branch of APPROACH-IS showed significant, but small differences between the groups, concluding that the two are comparable (Berghammer, Mattsson, Johansson, Moons, & Dellborg, 2017). Fourth, and in relation to the former issue, the study does not use a control group. This makes it impossible to investigate a possible difference between the participants with CHD and the general population. Fifth, differential item functioning, that people from different groups have a tendency towards answering a questionnaire in a certain way, was not verified. The SOC-13 has however been confirmed to be cross culturally applicable (Eriksson & Lindstrom, 2005). The Brief-IPQ have also been translated to several languages, validated and used in populations with different diagnoses, although it has not been validated as cross culturally applicable (Broadbent et al., 2015).

The study also has several strengths. First, the uniform method measuring SOC and illness perceptions in the large international sample makes it possible to compare similarities and differences between the centres and to some extent the countries. Second, the large number of participants strengthens the statistical power and the statistical conclusion validity. Third, sampling was conducted using either random sampling from the centre database or through consecutive sampling, approaching attending patients. The aim of both methods was to get a representative sample from the accessible population and reduce sample bias. This increases the external validity and supports the generalizability of the inferred relationship to a target population. External validity is also supported by use of centres across the world, making this a multisite study, enhancing the generalizability (Polit & Beck, 2017, pp. 229-230, 254-256). Fourth, the two PROMs, SOC and illness perceptions, are measured using self-report questionnaires created based on the theories of Antonovsky and Leventhal et al. (Antonovsky, 1987; Broadbent, Petrie, Main, & Weinman, 2006; Eriksson & Lindstrom, 2005; Leventhal et al., 1980; Leventhal et al., 1984; Luyckx et al., 2012). The solid conceptualization of the
variables enhances construct validity. Fifth, a strength regarding the variables in the study, is the low number of missing values thus minimizing the potential of impact on results and increasing reliability.

8.2.2 Measurement issues

SOC-13
Only a few longitudinal studies using SOC-13 have been performed in order to calculate test-retest-reliability of the scale. Test-retest-reliability illustrates if the measure is reproducible, on two different occasions on the same people and refers to the stability of the test. Most of the studies show a moderate correlation, with one exception showing very strong correlation (Eriksson & Lindstrom, 2005; Hansen, Kristensen, Cederlund, & Tromborg, 2017). The high internal consistency reported on SOC-13 support the argument that the different items measure the same underlying trait or construct (Eriksson & Lindstrom, 2005; Polit & Beck, 2017, p. 307). Reliability is concerned with the quality of a measure and to which degree it is free form measurement error (Polit & Beck, 2017, pp. 303-304). Overall, SOC-13, has been found to be a reliable instrument (Eriksson & Lindstrom, 2005; Luyckx et al., 2012).

Face validity, concerning how SOC-13 appears to measure the construct, and consensual validity, how scientists agree on the properties of the instrument, have been found to be moderate and weak respectively. The SOC scale have been used in different cultures, populations, and scientific disciplines. SOC-29 and SOC-13 have been used in at least 49 languages. In addition to the original 29 item and 13 item questionnaires, at least 15 modified versions existed in 2005, implying that there is debate regarding the SOC scale. The instrument’s ability to predict other outcomes, predictive validity, is relatively good (Eriksson & Lindstrom, 2005; Eriksson & Mittelmark, 2017; Polit & Beck, 2017, pp. 310, 312).

Construct validity deals with the fundamental question of what is actually being measured with the instrument (Polit & Beck, 2017, p. 315). Differing results on the correlation between the SOC scales and the theoretical construction has been found (Eriksson & Lindstrom, 2005). It has been suggested that SOC is a multidimensional rather than a uni-dimensional concept, as Antonovsky proposed (Antonovsky, 1987, p. 19; Eriksson & Mittelmark, 2017, p. 99). No common single factor have been found in the different items measuring SOC, implying that SOC consists of different dimensions (Sandell, Blomberg, & Lazar, 1998). The
construction of SOC make it possible to theoretically divide SOC in three components and discuss their relationship. If the aim is to empirically measure correlation between the components, the questionnaire would have to be structured differently, because items from one component would have to be entirely different from items from another component. Thus Antonovsky claimed that the empirical use of the three components or sub dimensions would probably not give any meaning and considered this a theme for future work (Antonovsky, 1987, pp. 86-88; Eriksson & Lindstrom, 2005). It has been argued that predictability is unnecessary for a strong SOC and good health, and thus is an inappropriate part of SOC-scales. Removing the items most related to predictability from SOC-29 and SOC-13 strengthens the relationship to measures of physical health (Flensborg-Madsen, Ventegodt, & Merrick, 2005). The argument against this is that it would be contradictory to how Antonovsky emphasised consistency and predictability as a necessity to shape a strong SOC (Antonovsky, 1979, p. 187; Eriksson & Mittelmark, 2017, p. 102) This example, along with the fact that there is debate concerning the dimensional aspect of SOC, shows that the concept of SOC and the dynamics between the different dimensions is still to be understood and agreed upon completely. Despite this, the SOC-13 is considered a valid instrument (Eriksson & Lindstrom, 2005).

**Brief IPQ**

Test-retest reliability have been found to be ranging from moderate to good (Broadbent et al., 2006; de Raaij, Schroder, Maissan, Pool, & Wittink, 2012; Hallegraeff, van der Schans, Krijnen, & de Greef, 2013). Due to the single item form, internal consistency is not possible to measure (Wanous, Reichers, & Hudy, 1997). This is because only one item measures each dimension of illness perceptions, making it of limited value to measure consistency across items (Broadbent et al., 2015; Polit & Beck, 2017, p. 307). Some studies do however report Cronbach alpha scores on modified subscales of the Brief IPQ. The authors suggesting that the instrument better fits a two-factor structure than the original three-factor structure, and that the cognitive and emotional representations are not completely distinct factors (de Rooij et al., 2018; Zhang et al., 2017).

The Brief IPQ has been confirmed to have good predictive validity. Predictive validity refers to whether the instrument, in this case the Brief IPQ, can predict a criterion measured in the future (Broadbent et al., 2006; Broadbent et al., 2015; Polit & Beck, 2017, pp. 311-312). Concurrent validity is examined by assessing associations between the instrument and
criterions measured at the same time (Polit & Beck, 2017, p. 312). Concurrent and discriminant validity have both been found to be good for the Brief IPQ (Broadbent et al., 2006; Broadbent et al., 2015). Discriminant validity is used to distinguish Brief IPQ from other similar measures and make sure that they are not measuring the same construct (Polit & Beck, 2017, pp. 317-318). There has been some debate regarding content validity, to which degree the Brief IPQ adequately measure the construct. This has to do with the relevance of the items in the instrument and whether the items are balanced and comprehensive enough to cover the entire construct (Polit & Beck, 2017, p. 310) It has been argued that the instrument needs improvement in order to do so (Broadbent, Kaptein, & Petrie, 2011; French, Schroder, & van Oort, 2011; van Oort, Schroder, & French, 2011). Although the above mentioned concerns over the instrument, the Brief IPQ is frequently used and considered to be valid and reliable (Broadbent et al., 2015).

8.3 Epistemological and ontological framework

The aim of this study, to investigate a possible relationship between SOC and illness perceptions, puts it within a tradition of epidemiological research, with roots in empirism and positivism (Thornquist, 2003, pp. 78-79). Experience is the source for knowledge for empirists and positivists. Within this tradition, the scientist or subject, were considered to be a passive receiver of observations. August Comte, the founder of positivism, emphasised the importance and meaning of studying possible correlations between (social) phenomena (sociology), in line with this study (Giddens & Sutton, 2013, p. 11). Several ideas can be traced back to positivism in this study, but it also breaks with central ideas and research ideals of this tradition.

In previous research on children and adults with congenital heart disease, different methods have been used to measure QoL (Bratt & Moons, 2015). Not surprisingly this has led to different results. In APPROACH-IS, QoL as well as SOC and illness perceptions, was measured in the same way in all the countries and participating centres. This has made it possible to investigate a possible relationship as well as international differences. The findings will present future researchers with data to compare against. The positivist principle of unified science and the researcher as a passive or neutral subject, is to be glimpsed between the lines here (Thornquist, 2003, pp. 39-41). Forssen et al. (2011) argues that this ideal have
contributed in making scientists less responsible for the choices that are made in the research process. They argue that researchers ought to explain their choices from an ethical, social and political perspective. Such a stance is in opposition to the positivist idea of a neutral subject, and it highlights that choices made during research are based on ontological and epistemological assumptions on scientific knowledge, whether deliberate and intentional, or not. Today, the modified version of positivism, post-positivism, still strive for objective knowledge and truth, while recognising that absolute objectivity is impossible to obtain (Polit & Beck, 2017, pp. 9-10). It has been claimed that post-positivism made positivism redundant, and that it is anachronistic for nursing researchers to defend their work from a positivist stance. While there is a variety of competing paradigms, none have been able to supplant the scientific method and the principle of falsification of post-positivism (Corry, Porter, & McKenna, 2018). In experimental and quasi-experimental studies the randomized controlled study (RCT) and the testing of the null hypothesis is still the gold standard. The use of theories in order to create a hypothesis or 0-hypothesis stems from Poppers hypothetic-deductive method. This is in contrast with earlier positivist principles. Poppers idea was that a hypothesis can never be proved, only falsified. We can never be certain that a certain event is always going to cause that which we have experienced in the past (Thornquist, 2003, p. 64).

In this study, the two PROs, SOC and illness perceptions, have been measured using self-report questionnaires created based on the theories of Antonovsky and Leventhal and colleagues (Antonovsky, 1987; Broadbent et al., 2006; Eriksson & Lindstrom, 2005; Leventhal et al., 1980; Leventhal et al., 1984; Luyckx et al., 2012). Critics claim the way questionnaires such as these simplify and arrange experience in a way that removes the original meaning. It has been argued that there is a qualitative difference between for example weak and strong pain, and that putting pain into a scale therefore is impossible. If there is a qualitative difference between a strong and a weak SOC, worse or better illness perceptions, it does not give any meaning putting them in an increasing or decreasing order. Comte however, emphasized that research on social phenomena should be useful in a practical way for man (Thornquist, 2003, pp. 50-51, 71-73). Simplifying SOC and illness perceptions into numbers, thus making an investigation of a relationship possible, could be justified, if one deems the practical usefulness of research good enough. It is basically the only alternative when investigating a possible relationship between the variables in 4000 participants.
From the above it should be clear that the theories used today all build on earlier theories. The current theories are not static but a constant subject to discussion, modification and amendment. As long as no theory for everything exists, we need different theories for different purposes. The aim of the study decides which theory and method is better suited to answer the research questions, not the other way around.

9 Conclusion

We examined the relationship between illness perceptions and SOC in adults with CHD in a large international sample. Our analyses show that less threatening illness perceptions are related to stronger SOC. There was also a relationship between SOC and 6 of the sub-dimensions of illness perceptions: consequences, timeline, treatment control, identity, coherence, and emotional representations. Consequences, coherence and emotional representations were the strongest predictors of SOC.

10 Implications and future research

10.1.1 Implications on the individual level

According to Antonovsky (1987, pp. 94-100) the formation of SOC start at infancy. By early adulthood he considered the SOC to have reached a fairly stable point (Antonovsky, 1987, p. 107). Later research have shown that the development of SOC might not be this straightforward and that SOC is prone to changes also later in life (Drageset, Espehaug, Hallberg, & Natvig, 2014; Feldt et al., 2011; Nilsson, Holmgren, Stegmayr, & Westman, 2003; Silverstein & Heap, 2015). This has led to the emergence of questions on how and at what age SOC could best be intentionally improved (Moons & Norekval, 2018; Sullivan, 1989; Super, Wagemakers, Picavet, Verkooijen, & Koelen, 2016). SOC have been widely studied in the adolescent population (Braun-Lewensohn, Idan, Lindstrom, & Margalit, 2017, p. 124). Adolescence is a time were youth with CHD probably develop their own understanding of their disease. Previous research have shown that the understanding of own medical history are important in order to create meaning for adolescents. The subjective
cognitive and emotional aspects of living with CHD might be considered a resource for dealing with adversity and stress related to CHD (Aujoulat et al., 2014). Childhood and adolescence have been suggested as an appropriate time for interventions dealing with coping in patients with chronic conditions (Apers, Luyckx, et al., 2015; Aujoulat, Mustin, Martin, Pelicand, & Robinson, 2017, p. 339; Moons & Norekval, 2006).

10.1.2 Implications on group level

In order to speak about SOC on a group level, Antonovsky (1987, p. 176) considered a certain duration of a stable social context, and group consciousness, to be prerequisites. To speak of all adults with CHD as a group in this sense give no meaning. While Antonovsky considered the family, friends and close colleagues as groups where it could be possible to speak of a collective SOC other possibilities exist. Local branches of patient organisations and social media could both provide meeting grounds and might create an opportunity for some sort of cultural production. In this context cultural production include collective myths, language, humour etc. for the group (Antonovsky, 1987, p. 176). If the individuals face similar stressors, share their emotions, thoughts on cause and how to deal with this threat, it might be possible that the group develop GRRs available for collective use, as well as common illness perceptions and a collective SOC. Antonovsky himself raised the question of representativeness of the cultural production of the group and whether this actually give a true picture. In the case of social media the algorithms favour statements that get reactions, causing these statements to get more views even though they doesn’t necessarily represent the majority’s view. In such a case representativeness most arguably become an issue. How to deal with problems such as extremism and the polarization of debate in part caused by internet algorithms, is a major issue of our modern society. Providing answers for this is certainly beyond the scope of this thesis.

Understanding more about the complex relationship between SOC, GRRs and illness perceptions would be a first step in order for this knowledge to have practical usefulness on a group level.
10.1.3 Implications on health service

Interventions aimed at intentionally improving illness perceptions have showed positive results in different patient populations (Broadbent et al., 2009; Christensen et al., 2015; Lee, Chien, Hung, & Chou, 2015; Petrie, Cameron, Ellis, Buick, & Weinman, 2002). The same holds true for interventions aimed at strengthening SOC in different populations (Heggdal & Lovaas, 2017; Kekalainen, Kokko, Sipila, & Walker, 2018; Odajima, Kawaharada, & Wada, 2017; Tan, Chan, Wang, & Vehvilainen-Julkunen, 2016). Interestingly mindfulness-based cognitive therapy have been shown to have a positive effect on SOC in atrial fibrillation patients and on illness perceptions in patients with rheumatoid arthritis (Dalili & Bayazi, 2019; Malm et al., 2018). These findings support the argument that SOC and illness perceptions are related to each other, and that they are altered and affected through somewhat similar mechanisms. Even though there is some evidence on the direction of the relationship between illness perceptions and SOC, it is still not possible to draw strong conclusions on whether change in one precedes and causes the change in the other, or the effect is bidirectional (Alsen & Eriksson, 2016; Langeland et al., 2013). Knowing that SOC and illness perceptions are related to a variety of clinical outcomes highlights the fact that modern healthcare needs to address and consider more than clinical signs and symptoms. The subjective experience of living with a chronic condition such as CHD ought to be routinely assessed and examined along with i.e. blood pressure and echocardiography.

Studies on salutogenesis or SOC in the hospital setting still uses a perspective emphasising risk instead of resources (Dietscher, Winter, & Pelikan, 2017, p. 292). This is perhaps a reminder that a change of perspective is much needed, and that less severe, or "good" illness perceptions can indeed be viewed as a resource along with a strong SOC. More research is needed on the organisational structure of health care in communities and hospitals, and the impact it has on SOC (Dietscher et al., 2017, p. 293). This would also be true for illness perceptions.

10.1.4 Implications on education

Different educational programmes on salutogenesis exist for healthcare workers. These emphasise the ability to translate theoretical knowledge on salutogenesis into practical skills and use (Vinje, Ausland, & Langeland, 2017, pp. 307-308). The parallel between illness
perceptions and the salutogenic understanding of each individual in their own context, and the activation of resources for adaptive coping, should be rather obvious. The change of perspective from pathogenesis to salutogenesis and the importance of the subjective experience can hardly be overstated. This knowledge ought to be an integral part in the education of healthcare workers.

10.2 Future research

Due to the fact that illness perceptions and SOC both were measured only at one time, temporality becomes an issue, making it possible only to speculate about the direction of the relationship between the two. Future research should measure the two at different time points, thus making it possible to decide a direction of the relationship. Interventions aimed at intentionally improving SOC and/or illness perceptions could take such knowledge into account. Further studies are needed in order to establish effective methods with this goal in mind.
11 References


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doi:10.1136/archdischild-2017-314211


Coping with illness reality: the relationship between illness perceptions and sense of coherence in adults with congenital heart disease

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To be submitted to the European Journal of Cardiovascular Nursing

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Abstract

Background: Sense of coherence (SOC) and illness perceptions have been shown to be resources for a better life in people with congenital heart disease (CHD). Understanding the dynamics of these modifiable factors could help inform interventions aimed at intentionally improving these.

Aim: To determine the relationship between illness perceptions, SOC, and their sub-dimensions in adults with CHD at an international level.

Methods: This study was a part of Assessment of Patterns of Patient-Reported Outcomes in Adults with Congenital Heart Disease - International Study (APPROACH-IS), a cross-sectional study, with 4028 patients from 15 countries. The self-report questionnaires, 13 item Orientation to Life Questionnaire (SOC-13) and Brief Illness Perception Questionnaire (Brief IPQ), were administered and patient characteristics on sex, age, complexity of heart disease, ethnic background, educational level, employment status, marital status, self-reported New York Heart Association (NYHA), and parental status was collected. Descriptive statistics, correlational, and hierarchical multilevel analyses were performed.

Results: Multiple regression analysis showed that the following dimensions of illness perceptions predicted SOC: emotional representations (beta = -.307, p < .001), coherence (beta = -.157, p < .001), consequences (beta = -.092, p < .001), treatment control (beta = -.082, p < .001), identity (beta = -.079, p < .001), and timeline (beta = -.062, p < .001).

Conclusion: Illness perceptions appear to be an actionable concept for improving SOC. This approach could be considered when making programmes for follow-up of adults with CHD.
1 Introduction

A good life being a goal in itself, patient reported outcomes (PROs) are gaining interest because they have been found to be associated with quality of life (QoL), clinical outcomes and even mortality (1). Together with the recognition of congenital heart disease (CHD) as a lifelong chronic disease, this has led attention to subjective well-being. The use and understanding of different PROs recognizes the patient as an important stakeholder in treatment and management of CHD, and promotes a more holistic approach from health care workers (2). Understanding how different PROs are related may help inform and improve everyday work for the patient. Two such PROs are illness perceptions and sense of coherence (SOC).

SOC describes a persons global view of the world as comprehensible, manageable and meaningful (3). Through SOC a person has the potential to maintain or even improve health by the successful use of resources to cope with the stressors of everyday life, or more chaotic situations and life events (3). The concept of coping is also a central part in the theory on illness representation or illness perceptions (4). The aim is to get a better understanding of how people interpret a health threat and how this affects coping and adherence to medical treatment. The theory is that illness perceptions are created by a persons bodily experience or symptoms, information from external sources (friends, family and healthcare workers), and finally from personal experience with illness (4). Together these illness representations form a cognitive framework constructed by the patient to make sense of the illness (5).

In adults with congenital heart disease (ACHD) both illness perceptions and SOC have been related to different health outcomes (6-10). There is however limited evidence of a relationship between SOC and illness perceptions (11-13). In patients with myocardial
Infarction a strong negative association between illness perceptions, SOC, and all three sub-dimensions was found (12). In carers for relatives with dementia, weak to moderate negative associations between sub-dimensions of illness perceptions (illness consequences, illness identity, illness coherence and emotional representations) and SOC have been found (13). In a patient education programme for people with psoriasis, illness coherence, emotional representations and illness consequences predicted change in SOC (11). This represents a possibility to consider these sub-dimensions of illness perceptions as resources for SOC.

Interventions aimed at improving illness perceptions have shown promising results (14-16). With illness perceptions appearing as an actionable concept, it could represent a possible pathway to increase SOC. A better understanding of these PROs can be used to improve treatment and identify modifiable factors for better health in this patient population (8, 17). The relationship between illness perceptions and SOC have not been examined in the ACHD-group and not in a large-scale international study. Therefore, the aim of this study was to determine the relationship between illness perceptions, SOC, and their sub-dimensions in adults with CHD at an international level.

2 Methods

2.1 Study population and procedure

The present study is part of an international study, the Assessment of Patterns of Patient-Reported Outcomes in Adults with Congenital Heart Disease – International Study (APPROACH-IS) (18). This is a cross-sectional study with participants from 15 countries across 5 continents. These countries include Argentina, Australia, Belgium, Canada, France, India, Italy, Japan, Malta, Norway, Sweden, Taiwan, the Netherlands, and the United States.
A standardized protocol was developed and the study was conducted in collaboration with the International Society for Adult Congenital Heart Disease (ISACHD).

In this study, 4028 adults with CHD, from 24 participating centres, were enrolled. The inclusion criteria were: 1) diagnosis of CHD, defined as a structural abnormality of the heart or intra-thoracic great vessels that is present at birth and is actually or potentially functionally significant, 2) 18 years of age or older, 3) diagnosis established before the age of 10 (i.e., before adolescence to warrant sufficient experience of living with CHD), 4) continued follow-up at CHD centre or included in a national/regional registry and, 5) physical, cognitive and language capabilities required to complete the self-report questionnaires. Exclusion criteria were: 1) prior heart transplantation, 2) primary pulmonary hypertension or, 3) impaired cognitive abilities. The eligible participants were selected either randomly from the institution’s database or consecutively approached at the outpatient clinics and received an information letter, a copy of the survey package, and an informed consent form (if necessary).

Data on patient characteristics were collected from medical records and patient self-reports and included age, sex, marital status, presence of children, educational level, employment status, self-reported NYHA-class, CHD diagnosis, and complexity of CHD (18).

2.2 Measurement

Two self-report questionnaires were used to measure SOC and illness perceptions.

*The 13-item orientation to life questionnaire (SOC-13)*

The 13-item orientation to life questionnaire (SOC-13) were used to measure SOC. A semantic scale ranging from 1 (very seldom or never) to 7 (very often) with a total score from 13 to 91, was used to measure comprehensibility (five items), manageability (four items) and
meaningfulness (four items). A higher score implying a stronger SOC. In a systematic review, the SOC scale was found to be valid, reliable, cross culturally applicable and with internal consistency ranging from 0.70 to 0.92, measured by Cronbach’s α (19). High internal consistency support the argument that the different items in SOC-13 measure the same underlying trait or construct. For this study the Cronbach’s α was 0.85.

*The brief illness perception questionnaire (Brief IPQ)*

The brief illness perception questionnaire (Brief IPQ) uses a 9 item scale ranging from 0 to 10, with a higher score indicating a more threatening view of the illness. The 9 items include consequences, timeline, personal control, treatment control, identity, concern, emotions, illness comprehensibility and perceived causal factors. The Brief IPQ have been confirmed to have good predictive validity as well as concurrent validity and discriminant validity (20, 21). The Brief IPQ has been found to be sensitive to change. Overall the Brief IPQ is found to be valid and reliable (21).

### 2.3 Statistical analysis

Descriptive statistics was used to calculate SOC and illness perceptions. Correlation analyses were performed to compute a possible relationship between SOC, illness perceptions and their sub-dimensions, measuring Pearson product-moment correlation coefficient. Due to the fact that sub-dimensions of illness perceptions have been shown to predict changes in SOC (11), hierarchical multiple regression was performed with these sub-dimensions as independent variables. It was adjusted for sex, age, complexity of heart defect, country of origin and self-reported NYHA-class. Histograms, scatterplots, normal P-P plots were generated, and collinearity diagnostics performed to ensure no violation of normality, linearity, homoscedasticity, outliers, independence of residuals, multicollinearity or singularity. The data was analysed using IBM SPSS Statistics, version 25 (Armonk, NY).
2.4 Ethical approval

The study protocol was approved by the Institutional Review Board of the University Hospitals Leuven/ KU Leuven. Participating centres gained ethical approval if required. The investigation conforms with the principles outlined in the Declaration of Helsinki II (18, 22).

3 Results

3.1 Patient characteristics

Among the 4028 participants, the median age was 32 years and 53% were women. A majority of 74% were white or Caucasian, 64% had part-time or full-time work, 51% were married or living with a partner, and 60% had no children. Fifty-four percent reported being in NYHA-class I, and 49% had a CHD of moderate complexity (Table 1).

3.2 Relationship between illness perceptions and SOC

Table 2 presents the results of the hierarchical multiple regression analysis. After controlling for sex, age, complexity of heart defect, country of origin and self-reported NYHA-class, illness perceptions sub-dimensions explained an additional 18% of the variance in SOC ($R^2$ change = .180, $F$ change = 115.501, $p < .001$). The model as a whole explained 29% of the variance in SOC ($F = 114,290$, $p < .001$). Six of the sub-dimensions of illness perceptions were statistically significant as predictors for SOC, comprehensibility, manageability and meaningfulness. Of the sub-dimensions emotional representations ($beta = -.307$, $p < .001$), coherence ($beta = -.157$, $p < .001$), and consequences ($beta = -.092$, $p < .001$) recorded the highest beta values.
4 Discussion

This cross-sectional study examined the relationship between illness perceptions, SOC and sub-dimensions in adults with CHD. To our knowledge this is the first study to do this. The findings suggest illness perceptions as an actionable concept that can guide healthcare interventions aimed at improving SOC.

Patients who felt more negative emotions and experienced more consequences of their illness reported a lower SOC. Stronger perception of a coherent illness understanding and belief that treatment can control the disease were related to a stronger SOC. Interestingly, a more chronic view of illness were also related to a stronger SOC. Finally, more perceived symptoms were related to a lower SOC. There was no observed relationship between personal control, concern and SOC. This is in line with previous research, as is the relationship between emotional representations, illness coherence, consequences and SOC (11, 13). These three dimensions showed the strongest relationship with SOC in the present study and all three predicted change in SOC in patients with psoriasis undergoing an education programme (11). From a theoretical viewpoint it has been proposed that illness coherence is related to SOC through the restructuring of illness meaning, thus disrupting or contributing to coherence between experience, and past and new understanding of own illness (23). A parallel to this idea is the finding of comprehensibility as a more important predictor than manageability and meaningfulness for change in SOC (24). In young adults with CHD a coherent understanding of own disease appeared to be a more common experience among those with a stronger SOC (25). Good knowledge of the disease has also been shown to correlate with health-related QoL in adolescents with heart disease (26). The patient’s knowledge and subjective understanding of own condition appears to be a possible resource for SOC.
Emotional representations showed the strongest relationship to SOC among the illness perceptions sub-dimensions. Antonovsky (3) argued that a perception of less severe negative emotions is the result of the successful resolution of tension caused by a stressor by a person with a strong SOC. He further suggested that persons with a strong SOC were likely to experience different emotions than those with a weak SOC, and that upon facing a stressor, a person with a strong SOC would consider this a challenge, instead of feeling hopelessness and apathy (3). The finding of emotional representations as a predictor for SOC, does however imply that emotional state is also affecting SOC (11). It might well be that severe negative emotions have an influence on tension management when facing a stressor, causing a tendency to dysfunctional coping with the demand and a lower SOC in the long term. In support of our finding, both emotional representations and SOC have been found to be associated with symptoms of anxiety and depression (8, 27). Revealing severe emotional representations is the first step in order to help the patient deal with these and if necessary seek appropriate help and treatment.

Illness consequences are thought to reflect the perceived severity of the disease. The distinction between subjective perceptions of consequences and clinical markers observed by healthcare professionals are worth noting. Obviously, there is not necessarily a relationship. Whether the patient or the clinician holds the true answer is debatable. It does however seem plausible to suggest that there might exist dysfunctional perceptions worth uncovering. This probably holds true for all parts of illness perceptions and could help uncover misunderstandings between patient and healthcare worker. The impact of disease on everyday life covers a variety of possible factors that may include social life, ability to work or study, and physical limitations. In line with our findings, previous research has found a relationship between a felt impact of restrictions and limitations and SOC in adolescents with CHD, with
more limitations related to a lower SOC (28). In contradiction to this, another study on adolescents with CHD found that those with a stronger SOC actually reported a greater influence of the disease on daily life (25). The incongruent findings might partly be explained by the fact that in the latter study the adolescents with a strong SOC considered the heart disease to contribute to their positive outlook on life (25). This could lead on to speculations on whether the consequences in it self, or how they are viewed is more important. If severe consequences represent an obstacle seen as a worthy challenge instead of being overwhelming, this may explain the different findings. Again the crucial point of examining the patient’s ideas become evident.

The observed relationship between illness identity and SOC have also been shown in carers for patients with dementia, but not in patients with psoriasis (11, 13). In adults with CHD more perceived symptoms has been associated with poorer QoL (8). A possible explanation is that the perceived burden of symptoms influence QoL through SOC. In neither patients with psoriasis, or in dementia caregivers, was our finding of a relationship between timeline, treatment control and SOC replicated (11, 13). The contradictory results may reflect actual differences in symptoms, available treatment and the trajectory of the disease, that in turn would influence the patient’s perceptions of the illness. Maybe realising the chronic nature of the congenital heart disease represent an integration of the disease as a part of oneself. The struggle for normalisation and defining oneself as a survivor, thus contributing to a sense of purpose, have been found to be central themes in adults with CHD (29). Adults with CHD have probably already overcome many obstacles and experienced positive effect of medical and/or surgical treatment. The perception of available resources at hand, and the appreciation of life-saving/enhancing surgery may lead to stronger SOC (17).
Even though in line with previous research, the finding that there was no relationship between personal control and SOC is somewhat surprising (11, 13). Antonovsky (3) considered perceived resources to deal with stressors at the core of the manageability dimension of SOC. In adolescents with CHD it has been found that those with a strong SOC experienced a higher degree of personal control than those with a lower SOC. Those with a lower SOC also reported more worries (25). This also contradicts our finding that illness concern is not related to SOC. These divergences are difficult to explain.

Six of 8 dimensions of illness perceptions was found to have a significant relationship with SOC: consequences, timeline, treatment control, identity, coherence, and emotional representations. The understanding of illness perceptions sub-dimensions as a factor influencing SOC leads to the question on whether illness perceptions can be changed. Different interventions, also nurse-led, have proven that it is possible to intentionally modify illness perceptions (14-16). Together with the fact that illness perceptions are related to a variety of health outcomes, the clinical relevance for healthcare workers seems clear. The subjective reality of illness needs to be addressed during consultations with patients with ACHD. Setting up programmes aiming for a less threatening perception of illness has the potential to improve SOC.

*Methodological issues*

Strengths of the study include the large sample, low level of missing values and use of a uniform measuring method. In addition, the measures rely on well known, and solid theoretical conceptualisation. However, there are also issues that need to be considered. First, because of the cross-sectional design it was not possible to decide the direction of the relationship between illness perceptions sub-dimensions and SOC. This would require a
longitudinal study design. Second, the selection of participating centres due to size and resources, implies that the results can not be generalised to the entire ACHD population.

Adding to this point is the fact that the majority of the study population was white/Caucasian and came from western industrialised countries. Third, because of the lack of a control group, it was not possible to compare the results to the general population. Fourth, differential item functioning, whether people from different groups, i.e. people from different countries, respond to the items a certain way, can not be ruled out.

5 Conclusion

The present study showed that less threatening perceptions of illness are related to stronger SOC in adults with CHD in a large international sample. After controlling for patient characteristics, the following 6 sub-dimensions of illness perceptions showed substantial associations to SOC: consequences, timeline, treatment control, identity, coherence, and emotional representations. By modifying these perceptions it appears possible to strengthen SOC. As stated in a previous editorial in this journal, investigating the effects of interventions aiming at improving SOC, appears to be one way forward for nursing research within the field of cardiology (30).

6 Implications for practice

- Healthcare workers need to address and examine the patient’s illness perceptions in order to understand the subjective reality of the illness.
- In addition to uncovering misunderstandings, including the patient’s perspective could contribute to a better dialogue and inviting the patient to participate in decision-making.
• Longitudinal research is needed in order to conclude on the direction of the relationship between illness perceptions and SOC.

• Further research should aim to establish effective interventions for improving SOC.

Acknowledgements

The author(s) would like to thank the participants of APPROACH-IS.

Declaration of conflicting interests

The authors declare that there is no conflict of interests.

Funding

This work was supported by grants from the Vestfold Hospital Trust (196150), and by the Norwegian Nurses Association (19/0489).

The study protocol was registered at ClinicalTrials.gov: NCT02150603.
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Table 1 Patient characteristics

<table>
<thead>
<tr>
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<th>Total</th>
<th>n (%)</th>
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<tr>
<td>Female</td>
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<tr>
<td>Male</td>
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<td>1897 (47.3)</td>
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<td><strong>Median age in years</strong></td>
<td>4021</td>
<td>32 (IQR: 25-42)</td>
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<td><strong>Complexity of heart defect</strong></td>
<td>4028</td>
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<tr>
<td>Simple</td>
<td></td>
<td>1040 (25.8)</td>
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<tr>
<td>Moderate</td>
<td></td>
<td>1957 (48.6)</td>
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<tr>
<td>Complex</td>
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<td>1031 (25.6)</td>
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<td><strong>Ethnicity</strong></td>
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**Table 2** Results from hierarchical multiple regression analysis examining associations between illness perception sub-dimensions and SOC with sub-dimensions in adults with congenital heart disease

<table>
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<tr>
<th></th>
<th>SOC</th>
<th>Comprehensibility</th>
<th>Manageability</th>
<th>Meaningfulness</th>
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<td><strong>Consequences</strong></td>
<td>-.092 (-.633-.223)**</td>
<td>-.056 (-.269-.022)*</td>
<td>-.052 (-.162-.009)*</td>
<td>-.144 (-.309-.159)**</td>
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<tr>
<td><strong>Timeline</strong></td>
<td>.062 (.142-.391)**</td>
<td>.042 (.023-.137)**</td>
<td>.049 (.028-.121)**</td>
<td>.063 (.050-.142)**</td>
</tr>
<tr>
<td><strong>Personal control</strong></td>
<td>-.026 (-.241-.015)</td>
<td>-.025 (-.105-.011)</td>
<td>-.030 (-.094-.002)</td>
<td>-.017 (-.072-.022)</td>
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<tr>
<td><strong>Treatment control</strong></td>
<td>-.082 (-.508-.243)**</td>
<td>-.072 (-.207-.086)**</td>
<td>-.066 (-.157-.058)**</td>
<td>-.077 (-.173-.076)**</td>
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<tr>
<td><strong>Identity</strong></td>
<td>-.079 (-.588-.180)**</td>
<td>-.064 (-.230-.044)**</td>
<td>-.088 (-.227-.074)**</td>
<td>-.062 (-.180-.030)**</td>
</tr>
<tr>
<td><strong>Concern</strong></td>
<td>.012 (-.092-.188)</td>
<td>-.021 (-.101-.027)</td>
<td>.026 (-.016-.089)</td>
<td>.037 (.001-.104)*</td>
</tr>
<tr>
<td><strong>Coherence</strong></td>
<td>-.157 (-.120-.774)**</td>
<td>-.144 (-.462-.305)**</td>
<td>-.105 (-.289-.160)**</td>
<td>-.163 (-.409-.282)**</td>
</tr>
<tr>
<td><strong>Emotional representations</strong></td>
<td>-.307 (-.444-.113)**</td>
<td>-.295 (-.620-.469)**</td>
<td>-.301 (-.506-.382)**</td>
<td>-.206 (-.361-.240)**</td>
</tr>
<tr>
<td><strong>Change R²</strong></td>
<td>.180</td>
<td>.155</td>
<td>.138</td>
<td>.130</td>
</tr>
</tbody>
</table>

Adjusted for: age, sex, complexity of heart defect, and physical limitations.

***. Correlation is significant at the 0.001 level (2-tailed).

**. Correlation is significant at the 0.01 level (2-tailed).

*. Correlation is significant at the 0.05 level (2-tailed).