

Sense of coherence in adults with congenital heart disease in 15 countries: Patient characteristics, cultural dimensions and quality of life

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Background

Previous studies have found that sense of coherence (SOC) is positively related to quality of life (QoL) in persons with chronic conditions. In congenital heart disease (CHD), the evidence is scant.

Aims

We investigated (i) intercountry variation in SOC in a large international sample of adults with CHD; (ii) the relationship between demographic and clinical characteristics and SOC; (iii) the relationship between cultural dimensions of countries and SOC; and (iv) variation in relative importance of SOC in explaining QoL across the countries.

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Methods

APPROACH-IS was a cross-sectional, observational study, with 4028 patients from 15 countries enrolled. SOC was measured using the 13-item SOC scale (range 13–91) and QoL was assessed by a linear analog scale (range 0–100).

Results

The mean SOC score was 65.5 ± 13.2 . Large intercountry variation was observed with the strongest SOC in Switzerland (68.8 ± 11.1) and the lowest SOC in Japan (59.9 ± 14.5). A lower SOC was associated with a younger age; lower educational level; with job seeking, being unemployed or disabled; unmarried, divorced or widowed; from a worse functional class; and simple CHD. Power distance index and individualism vs collectivism were cultural dimensions significantly related to SOC. SOC was positively associated with QoL in all participating countries and in the total sample, with an explained variance ranging from 5.8% in Argentina to 30.4% in Japan.

Conclusion

In adults with CHD, SOC is positively associated with QoL. The implementation of SOC-enhancing interventions might improve QoL, but strategies would likely differ across countries given the substantial variation in explained variance.

Keywords

Heart defects • congenital • nursing • positive psychology • resilience • salutogenesis • sense of coherence • quality of life

Implications for practice

- Knowledge of the association between patient characteristics and sense of coherence (SOC) may assist clinicians in identifying patients at risk for lower SOC.
- Congenital heart disease programs can include SOC-enhancing techniques as they develop educational and psychological initiatives.
- Patients can be assisted with identifying appropriate resources to cope with stressors.
- Patients should be empowered to use these resources, in order to view the world as more comprehensible, manageable, and meaningful.

Introduction

Chronic cardiovascular conditions can have a severe impact on afflicted individuals. However, whereas some patients struggle, others seem to cope well. This disparity is poorly understood. Therefore, a call has been made to perform studies investigating the underlying mechanisms fostering a process of successful coping in patients with a chronic disease.¹ Unraveling such mechanisms is crucial as they may identify potential targets and strategies for interventions to improve health and well-being in patients.

To understand why some people do well and others do not, Aaron Antonovsky developed the salutogenic theory.² In his theory, he abandoned the prevailing pathogenic model that focused on the origins of disease and ill-being, and introduced the salutogenic model focusing on the origins of health and well-being.³ According to Antonovsky, the answer to his question is to be found in a strong sense of coherence (SOC), which represents the central construct of the salutogenic model. SOC is:

a global orientation that expresses the extent to which one has a pervasive, enduring though dynamic feeling of confidence that (a) the stimuli deriving from one's internal and external environments in the course of living are structured, predictable, and explicable; (b) the resources are available to one to meet the demands posed by these stimuli; and (c) these demands are challenges worthy of investment and engagement.³ (p. 19)

Hence, people with a strong SOC perceive the world as (a) comprehensible, (b) manageable, and (c) meaningful. People with a strong SOC are able to cope better with the stressors in life, such as living with a chronic condition.⁴

SOC may be open to modification based on its three components and, therefore, can be seen as a viable intervention target.⁵ Healthcare professionals could strengthen patients' comprehensibility by providing accessible and understandable information and education. Furthermore, to enhance manageability, it would be important to encourage patients to be aware and make use of available resources. In addition, supporting patients to take control and to participate in decision-making could enhance their meaningfulness.³ Indeed, studies have found that SOC is a malleable construct that can change over time.⁵ Furthermore, several intervention studies have shown that SOC is modifiable using different methods, such as talk-therapy groups, lifestyle intervention programs, and patient education courses.^{6–8}

Congenital heart disease (CHD) is a chronic cardiovascular condition that includes structural abnormalities of the heart and/or great intrathoracic vessels, that have or could result in functional impairment,⁹ and comprises a wide spectrum of simple, moderate and complex heart defects.¹⁰ Studies investigating SOC in persons with CHD have been performed in Australia,^{11–13} Belgium,^{14–18} Denmark,^{19,20} Germany^{21,22} and Japan.²³ Eight out of the 13 studies investigated the relationship between SOC and quality of life (QoL), all of which confirmed a positive relationship,^{12–14,16,17,20–22}. Since SOC is actionable, these study findings make SOC a possible target for intervention to indirectly improve the QoL of individuals with CHD.

However, given that the prior reports were single-center studies with varying methodologies conducted in a handful of countries, intercountry comparisons are questionable and results cannot be generalized to non-studied countries. Indeed, we do not know how SOC varies across different countries and whether the association between SOC and QoL is equally strong in all countries. Furthermore, it can be assumed that SOC is culture sensitive. In other words, characteristics of culture may impact on the development of SOC. Therefore, the aims of the present study were (i) to explore intercountry variation in SOC scores in a large sample of adults with CHD from Europe, Asia, Australia, North America, and South America; (ii) to examine the relationship between demographic and clinical characteristics and SOC; (iii) to investigate the relationship between cultural dimensions of countries and SOC above and beyond patient characteristics; and (iv) to evaluate to what extent the relative importance of SOC in explaining variance in QoL differs across the countries.

Methods

Study population and procedure

This analysis is part of APPROACH-IS (Assessment of Patterns of Patient-Reported Outcomes in Adults with Congenital Heart disease – International Study), a cross-sectional study in which data were collected in 15 countries, including Argentina, Australia, Belgium, Canada, France, India, Italy, Japan, Malta, the Netherlands, Norway, Sweden, Switzerland, Taiwan, and the USA.²⁴ Patients were eligible for study inclusion if they met the following criteria: (i) diagnosis of CHD; (ii) age ≥ 18 years; (iii) diagnosis established before adolescence; (iv) continued follow-up at a CHD center or included in a national/regional registry; and (v) physical, cognitive, and language capabilities required to complete self-report questionnaires. Patients with prior heart transplantation or primary pulmonary hypertension were not eligible.

Two recruitment strategies have been employed.²⁴ In the first strategy, centers randomly selected eligible patients from their institution's database. These patients received a study package by mail. Patients were asked to complete the questionnaires within two weeks. Various approaches were used to maximize response rates (e.g., mail or telephone reminders). A second recruitment strategy was to consecutively approach eligible patients at outpatient clinics. Patients who consented completed surveys during their clinic visit and returned them to the research assistant or data collection officer. Patients were also given the opportunity to return surveys by self-addressed stamped envelope.

Informed consent was obtained from each patient and the study protocol conforms to the ethics guidelines of the 1975 Declaration of Helsinki as reflected in a priori approval by the institution's human research committee. The rationale for APPROACH-IS and its methodology is detailed in a dedicated methods paper.²⁴

Measures

Demographic data were collected through self-report questionnaires. Medical data were obtained through chart review.

SOC was assessed using the 13-item Orientation to Life Questionnaire also known as the 13–SOC scale, evaluating the degree of meaningfulness, comprehensibility, and manageability.³ The questionnaire consists of four meaningfulness items (e.g. “How often do you have the feeling that there's little meaning in the things you do in your daily life?”), five comprehensibility items (e.g. “Do you have the feeling that you are in an unfamiliar situation and don't know what to do?”), and four

manageability items (e.g. “How often do you have feelings that you're not sure you can keep under control?”). Items are answered on a 7-point semantic differential scale, in which response options range from 1 (very seldom or never) to 7 (very often). Scores range from 13 to 91 with higher scores reflecting higher levels of SOC. There are no cut-offs for high, moderate or low SOC. The validity and reliability of this scale are well established in the literature, also in persons with CHD.^{25,26} The Cronbach's alpha of the 13–SOC scale was 0.85 (Supplemental Table 1). The alpha was 0.80 in all countries, except for India, where the alpha was 0.68. This instrument is applicable to different cultures around the globe.²⁷

QoL was defined as the degree of overall life satisfaction that is positively or negatively influenced by individuals' perception of certain aspects of life important to them, including matters both related and unrelated to health.²⁸ This definition was based on a thorough conceptualization.²⁹ It reflects the overall QoL of an individual, which is broader than health-related QoL,³⁰ as this latter construct is often referring to the health status or functional abilities of individuals.³¹ In line with this definition, QoL was measured by a linear analog scale (LAS). This instrument measured QoL on a vertically oriented line graded with indicators from 0 (worst imaginable QoL) to 100 (best imaginable QoL), and is valid and reliable for its use in persons with CHD.³²

Scores on the dimensions of national culture (scale from 0 to 100) were based on extensive research conducted by Hofstede.³³ The Hofstede model includes 6 dimensions: a power distance index (higher scores reflect higher levels of acceptance that power is distributed unequally in society), individualism versus collectivism (high scores reflect individualistic societies), masculinity versus femininity (higher scores reflect more masculine societies directed toward achievement and success), uncertainty avoidance index (higher scores reflect societies that are more rigid in beliefs and behaviors), long-term orientation versus short-term normative orientation (thriftiness and perseverance are associated with higher scores), and indulgence versus restraint (higher scores are observed in societies that foster gratification of human drives related to enjoying life and having fun).³³ The scores on these dimensions of national culture are represented in Supplemental Table 2.

Statistical analyses

Descriptive statistics (i.e., means and standard deviations) were performed to evaluate SOC and QoL worldwide. If the data were not normally distributed, medians and interquartile ranges (IQR) were calculated. One-way ANOVA with pairwise comparisons using the Tukey post hoc test was conducted to investigate whether SOC differed between countries. The association of patient characteristics and SOC was explored using general linear mixed models (GLMM), which is a form of multilevel analysis. We used demographic and clinical characteristics as fixed effects, and country as a random effect. To assess the relationship between dimensions of culture and SOC, GLMM was performed as well with patient characteristics and dimensions of national culture as fixed effects, and country as random effect. To investigate the relationship between SOC and QoL, multivariable linear regression analyses were performed. Using a hierarchical approach, in the first step, we included demographic and clinical variables that were significant correlates of SOC. In the second step we included SOC, which allowed us to investigate a change in explained variance. The assumptions for linear regression analyses were met. Data analysis was performed using IBM SPSS Statistics for Windows, version 25 (IBM Corp., Armonk, NY, USA). Ridgeline plots were made using RStudio, version 1.1.463³⁴. A p-level < 0.05 was used as the cut-off for statistical significance, and statistical tests were two-sided. Post hoc power analyses were performed using G*Power Version 3.1.9.3.

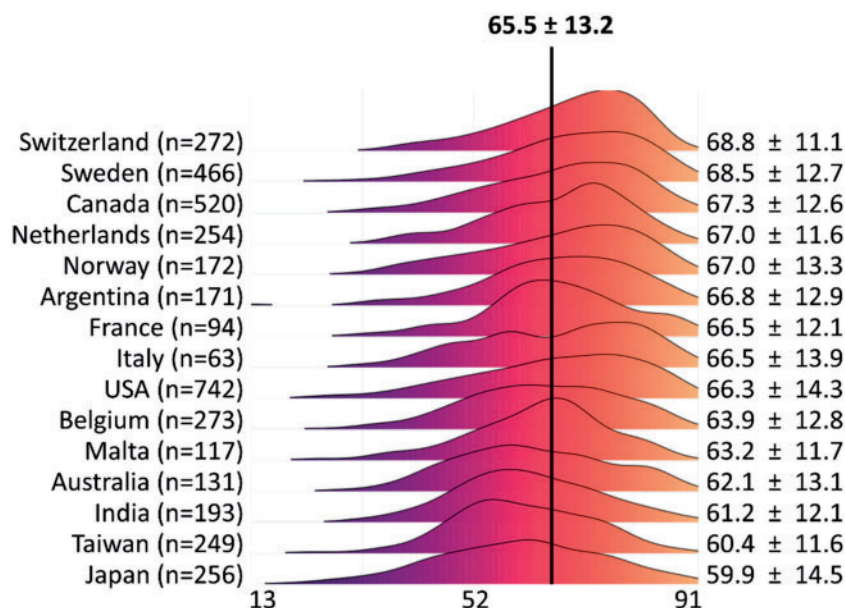


Figure 1 Sense of coherence in adults with congenital heart disease from 15 countries. The numbers represent the mean score \pm standard deviation.

Results

Sample characteristics

In total, 4028 adults with CHD were recruited. Patients had a median age of 32 years (IQR: 25–42y), 53% were women, 26% had CHD of simple complexity, 49% had CHD of moderate complexity, and 25% had complex CHD.³⁵ Full data on all variables of interest were available for 3910 patients (97%). More specifically, data on SOC were missing for 55 patients (1.4%) and 76 patients (1.9%) had missing data on QoL. The proportion of missing, however, ranged from 0.4 to 4.5% for SOC and from 0.0 to 4.5% for QoL, in the respective countries. Given the low proportion of missing values, no imputation of missing values has been performed.

Intercountry variation in sense of coherence

The total sample of adults with CHD had a mean SOC of 65.5 (± 13.2) (Figure 1). Patients from Japan had the lowest mean SOC (59.9 ± 14.5) and patients from Switzerland had the highest mean SOC (68.8 ± 11.1), representing a disparity of almost 9 points. One-way ANOVA showed that SOC differed significantly between countries ($F=13.7$, $\eta^2=0.05$, $p<0.001$). Significant results from post hoc tests are included in Supplemental Table 3. The countries with the lowest scores on SOC (i.e. India, Taiwan, and Japan) differed significantly from most other countries.

Patient characteristics and sense of coherence

Table 1 describes the demographic and clinical characteristics as correlates of SOC, using multivariable mixed models. Younger age;

lower educational level; job seeking, being unemployed or disabled; unmarried, divorced or widowed; poorer New York Heart Association (NYHA) functional class; and simple heart lesions were associated with lower SOC. These characteristics explained 13.8% of the variance of SOC, and country as random effect explained 2.9% of the variance (Table 1).

Cultural dimensions and sense of coherence

The GLMM showed that power distance index (Estimate=-0.10; SE=0.04; $p=0.033$) and individualism versus collectivism (Estimate=0.07; SE=0.03; $p=0.04$) significantly explained SOC, above and beyond demographic and clinical characteristics. More specifically, when power is more equally distributed in society and when societies are more individual (less collectivistic), people tend to have a stronger SOC. No statistical relationship with other cultural dimensions and SOC has been found.

Relationship between sense of coherence and quality of life

Patients had a mean QoL of 78.3 (± 16.6). SOC was positively correlated with QoL in this sample of adults with CHD ($r=.498$, $p<.001$). When adjusted for patient demographic and clinical characteristics that were significantly associated with SOC (Table 1), SOC was a significant correlate of QoL, explaining 17.2% of its variance, while patient characteristics explained 17.4% of the variance (total model: adjusted $R^2=34.5\%$) (Figure 2). Adjusted for patient characteristics, SOC was a significant correlate of QoL in all countries. The variance of QoL explained by SOC, however, varied substantially across the countries. In Japan, 30.4% of the variance of QoL was explained by SOC, whereas in Argentina, this was only 5.8%. There were five countries where the explained variance was more than 20%: Japan,

Table 1 Demographic and clinical characteristics as correlates of sense of coherence in adults with congenital heart disease.

	Estimate (SE)	P-value
Sex		
Men	0.6 (0.4)	0.159
Women	#	
Age	0.1 (0.02)	<0.001
Educational level		
Less than high school	-2.9 (1.0)	0.003
High school	-2.3 (0.5)	<0.001
College degree	-1.5 (0.6)	0.009
University degree	#	
Employment status		
Part-time or full-time work	#	
Homemaker or retired	-0.1 (0.8)	0.909
Job seeking, unemployed, or disability	-4.9 (0.7)	<0.001
Full-time student	0.9 (0.8)	0.238
Other	0.9 (0.8)	0.271
Marital status		
Never married	-2.1 (0.5)	<0.001
Married or living with partner	#	
Divorced or widowed	-2.5 (0.9)	0.009
Other	-15.4 (6.9)	0.026
Being religious/spiritual	0.07 (0.4)	0.874
Patient-reported NYHA assessment		
Class I	13.0 (0.1)	<0.001
Class II	8.4 (1.1)	<0.001
Class III	5.1 (1.3)	<0.001
Class IV	#	
Anatomical complexity of heart defect		
Simple	-1.4 (0.6)	0.018
Moderate	0.05 (0.5)	0.917
Complex	#	
R^2	16.9%	
Semipartial R^2 country differences	2.9%	
Semipartial R^2 patient characteristics	13.8%	
Power	100%	

NYHA: New York Heart Association; #: reference group

Norway, Taiwan, Sweden, and Italy. Four countries had an explained variance of less than 10%: Argentina, India, Switzerland, and Canada. In Belgium, Switzerland, USA, and Australia, the patient characteristics were substantially more explanatory than SOC. Conversely, QoL was mainly explained by SOC, rather than patient demographic and clinical characteristics in Taiwanese patients.

Discussion

In this large international study, comprising over 4000 patients from 15 countries, we found that the mean SOC score in adults with CHD was 65.5. However, large intercountry variation was observed, with

the strongest SOC in Switzerland and the lowest SOC in Japan. Patient characteristics associated with a lower score on SOC were younger age; lower educational level; job seeking, being unemployed or disabled; unmarried, divorced or widowed; worse NYHA functional class; and simple heart lesions. Patients living in countries characterized by a lower power distance and a higher individualism tend to have higher SOC scores. SOC was positively associated with QoL in the total sample and in the participating countries separately, indicating that SOC is a universally meaningful concept.

It may seem counterintuitive that persons with a simple heart defect have a lower SOC than persons with a complex defect, when adjusted for other demographic and clinical characteristics. However, this is in line with Antonovsky's theory, which states that repeated exposure to various stressful events during life and the successful management of those events help an individual to develop generalized resistance resources, and consequently, to strengthen that individual's SOC.³ Since growing up with a simple heart defect can be assumed to be less stressful than growing up with a moderate or complex defect, people with simple defects are not as much confronted with experiences that enhance comprehensibility, manageability, and meaningfulness.³⁶ As a consequence, they may have developed a lower SOC than people with a moderate or complex heart defect.

We found a significant relationship between SOC and the power distance index and individualism vs collectivism, respectively. These cultural dimensions may partly explain why there is an intercountry variation in SOC, as we found in the present study. In a prior study, we have investigated if dimensions of national culture are also related to patients' QoL.³⁵ In that study, we found that cultural dimensions were not associated with variation in QoL after adjustment for patient characteristics, although large intercountry variation in QoL existed.³⁵ This suggests that national culture is not directly impacting on QoL, but may indirectly have an impact through SOC.

The contribution of SOC to QoL differed between the countries, ranging from 5.8% in Argentina to 30.4% in Japan. Indeed, our findings highlight the varying importance of SOC across countries. For instance, although patients from Switzerland appear to have a high SOC, the contribution of SOC to the QoL of Swiss patients is marginal. Patient demographic and clinical characteristics obviously played a bigger role in understanding QoL in Switzerland. Divergent observations were seen for patients from Japan and Taiwan, demonstrating a relatively low SOC compared to the other countries in this study, yet SOC contributed for 25–30% to their QoL. These findings suggest that a country-specific approach is needed to understand the degree to which patients' QoL may be impacted through education and psychological interventions targeting SOC. Based on the results of this study, it is possible that SOC-focused interventions would have greater impact in countries like Japan, Taiwan, Norway, Sweden, and Italy rather than Switzerland, Argentina, India, or Canada. The results of India are quite unique, as neither SOC nor patient demographic/clinical characteristics were strong correlates of QoL. The relatively poor patient-reported outcomes in patients from India,³⁷ and the weak explanatory power of SOC and patient characteristics warrant further research. It may be that other factors (e.g., stigma or illness identity) are more explanatory for QoL in India, and perhaps by extension in other low and lower middle-income countries. This prompted inclusion of other potential explanatory variables in the same phase of the APPROACH-IS II project, currently underway.

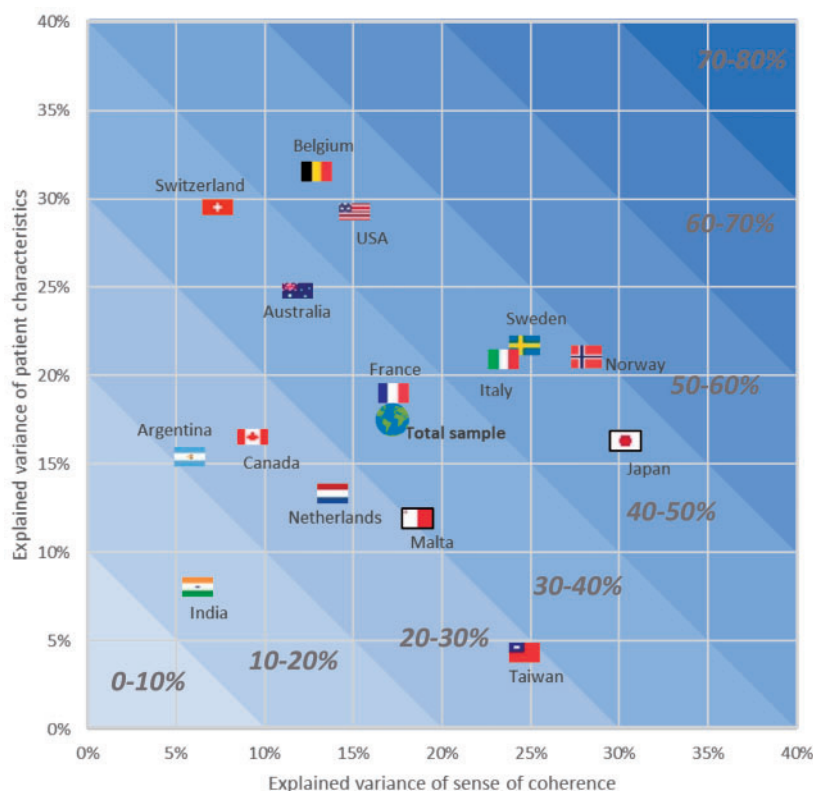


Figure 2 Explained variance of sense of coherence and patient characteristics in relation to quality of life in adults with congenital heart disease from 15 countries.

Patient characteristics are: Age, educational level, employment status, marital status, complexity of the heart defect, and New York Heart Association functional class. The shades represent the total explained variance of patient characteristics and SOC. Numbers of the explained variances are given in supplemental table 4.

The present study identified correlates of SOC in people with CHD. Knowledge of these correlates may assist clinicians in identifying patients at risk for lower SOC. Subsequently, adult CHD programs may wish to consider include SOC-enhancing techniques as they develop educational and psychological initiatives.⁶⁻⁸ Based on theoretical insights from Antonovsky's salutogenic theory and previously published studies on this topic, Super and coworkers³⁸ argue of two mechanisms that can be influenced to strengthen a person's SOC. The first refers to assisting people with identifying appropriate resources to cope with stressors (i.e., the behavioral mechanism). The second refers to training people in viewing the world as more comprehensible, manageable, and meaningful (i.e., the perceptual mechanism). To influence these mechanisms, empowerment can be used to facilitate the use of appropriate resources and reflection can assist people in becoming aware of their beliefs and assumptions³⁸ and could enhance persons' self-efficacy.³⁹ Indeed, empowering patients with CHD is of paramount importance and interventions to increase the level of empowerment are currently under study.^{40,41}

Methodological considerations

APPROACH-IS has several strengths. More than 4000 patients from 15 countries were included; a high degree of complete data was obtained; and valid and reliable instruments to comprehensively

assess SOC and QoL in an international setting were used. Indeed, 11 languages were represented in APPROACH-IS.

However, we have to consider some methodological limitations inherent to APPROACH-IS.^{24,35} First, APPROACH-IS is a cross-sectional study, and thus causality cannot be determined.²⁴ Second, in most participating countries, only one center partook in the project. This might hamper the representativeness of our sample.³⁵ Third, a possible selection bias cannot be ruled out, since patients with physical or mental incapacities preventing them from completing the questionnaires were excluded.²⁴ However, a comparison of participants and nonparticipants in the Swedish branch of APPROACH-IS revealed only small differences in demographic and clinical data.⁴² Fourth, we did not collect data in a control group. Hence, we cannot state to what extent SOC in our patients is better or worse than that of the general population. The scores of SOC in the Swedish cohort of APPROACH-IS corresponded closely to the mean scores of the general population samples in Sweden (median=69).⁴³ However, prior studies in Belgium¹⁶ and Germany²² showed that patients with CHD had a stronger SOC than their counterparts in the general population. Fifth, reasons for intercountry variation in SOC are still poorly understood. It could be that SOC in persons with CHD is dependent on the standard of living,³⁷ or healthcare system factors.⁴⁴ This is an avenue for future research. Sixth, in CHD, the

implementation of SOC-enhancing interventions has not been studied. Although interventions have shown to be effective in other cardiac populations,⁴⁵ we do not know yet to what extent improving SOC would improve QoL in persons with CHD. Seventh, the mechanism of impact is not fully understood yet. It might be possible that the relationship between SOC and QoL is fully or partially mediated by other factors, such as coping, as has been demonstrated in other studies.⁴⁶

Conclusions

SOC in patients with CHD varied substantially across the different countries, with the strongest SOC in Switzerland and the lowest SOC in Japan. Patient characteristics associated with a lower SOC were younger age; lower educational level; job seeking, being unemployed or disabled; unmarried, divorced or widowed; worse functional status; and simple heart lesions. Some dimensions of national culture showed to be related to SOC. SOC was positively associated with QoL, independent of demographic/clinical characteristics. However, the explained variance of SOC in QoL differed between the countries. The results have implications regarding targeted strategies to enhance SOC as a means of improving QoL in adults with CHD.

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Conflict of interest

The authors have no conflicts of interest to declare.

Trial registration

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References

- de Ridder D, Geenen R, Kuijer R, et al. Psychological adjustment to chronic disease. *Lancet* 2008;**372**: 246–255.
- Moons P and Norekval TM. Why some people do well and others don't. The role of sense of coherence in disease adaptation. *Eur J Cardiovasc Nurs* 2018;**17**: 672–674.
- Antonovsky A. *Unraveling the mystery of health: How people manage stress and stay well*. San Francisco: Jossey-Bass, 1987.
- Eriksson M and Lindstrom B. Antonovsky's sense of coherence scale and the relation with health: a systematic review. *J Epidemiol Community Health* 2006;**60**: 376–381.
- Feldt T, Leskinen E, Koskenvuo M, et al. Development of sense of coherence in adulthood: a person-centered approach. The population-based HeSSup cohort study. *Qual Life Res* 2011;**20**: 69–79.
- Langeland E Riise T, Hanestad BR, et al. The effect of salutogenic treatment principles on coping with mental health problems A randomised controlled trial. *Patient Educ Couns* 2006;**62**: 212–219.

7. Forsberg KA Björkman T, Sandman PO, et al. Influence of a lifestyle intervention among persons with a psychiatric disability: a cluster randomised controlled trial on symptoms, quality of life and sense of coherence. *J Clin Nurs* 2010;**19**: 1519–1528.
8. Fagermoen MS, Hamilton G and Lerdal A. Morbid obese adults increased their sense of coherence 1 year after a patient education course: a longitudinal study. *J Multidiscip Healthc* 2015;**8**: 157–165.
9. Mitchell SC, Korones SB and Berendes HW. Congenital heart disease in 56,109 births. Incidence and natural history. *Circulation* 1971;**43**: 323–332.
10. Warnes CA, Liberthson R, Danielson GK, et al. Task force 1: The changing profile of congenital heart disease in adult life. *J Am Coll Cardiol* 2001;**37**: 1170–1175.
11. Wang Q, Hay M, Clarke D, et al. The prevalence and predictors of anxiety and depression in adolescents with heart disease. *J Pediatr* 2012;**161**: 943–946.
12. Wang Q, Hay M, Clarke D, et al. Associations between knowledge of disease, depression and anxiety, social support, sense of coherence and optimism with health-related quality of life in an ambulatory sample of adolescents with heart disease. *Cardiol Young* 2014;**24**: 126–133.
13. Eaton SL, Wang QF and Menahem S. Determinants of quality of life in adults with CHD: an Australian cohort. *Cardiol Young* 2017;**27**: 1571–1576.
14. Luyckx K, Missotten L, Goossens E, et al. Individual and contextual determinants of quality of life in adolescents with congenital heart disease. *J Adolesc Health* 2012;**51**: 122–128.
15. Apers S, Luyckx K, Rassart J, et al. Sense of coherence is a predictor of perceived health in adolescents with congenital heart disease: a cross-lagged prospective study. *Int J Nurs Stud* 2013;**50**: 776–785.
16. Apers S, Moons P, Goossens E, et al. Sense of coherence and perceived physical health explain the better quality of life in adolescents with congenital heart disease. *Eur J Cardiovasc Nurs* 2013;**12**: 475–483.
17. Apers S, Luyckx K, Goossens E, et al. Sense of coherence in young people with congenital heart disease. *J Dev Behav Pediatr* 2015;**36**: 267–276.
18. Apers S, Sevenants L, Budts W, et al. Sense of coherence does not moderate the relationship between the perceived impact of stress on health and self-rated health in adults with congenital heart disease. *Eur J Cardiovasc Nurs* 2016;**15**: 529–536.
19. Overgaard D, Schrader AM, Lisby KH, et al. Patient-reported outcomes in adult survivors with single-ventricle physiology. *Cardiology* 2011;**120**: 36–42.
20. Berg SK, King C, Overgaard D, et al. Sense of coherence as a resource for quality of life in patients with congenital heart disease: the benefits continue into adulthood. *Eur J Cardiovasc Nurs* 2013;**12**: 567–568.
21. Neuner B, Busch MA, Singer S, et al. Sense of Coherence as a Predictor of Quality of Life in Adolescents With Congenital Heart Defects: A Register-Based 1-Year Follow-Up Study. *J Dev Behav Pediatr* 2011;**32**: 316–327.
22. Muller J, Hess J and Hager A. Sense of coherence, rather than exercise capacity, is the stronger predictor to obtain health-related quality of life in adults with congenital heart disease. *Eur J Prev Cardiol* 2014;**21**: 949–955.
23. Nio K. Sense of coherence in adolescents with congenital cardiac disease. *Cardiol Young* 2010;**20**: 538–546.
24. Apers S, Kovacs AH, Luyckx K, et al. Assessment of Patterns of Patient-Reported Outcomes in Adults with Congenital Heart disease - International Study (APPROACH-IS): rationale, design, and methods. *Int J Cardiol* 2015;**179**: 334–342.
25. Eriksson M and Lindstrom B. Validity of Antonovsky's sense of coherence scale: a systematic review. *J Epidemiol Community Health* 2005;**59**: 460–466.
26. Luyckx K, Goossens E, Apers S, et al. The 13-Item Sense of Coherence Scale in Dutch-Speaking Adolescents and Young Adults: Structural Validity, Age Trends, and Chronic Disease. *Psychol Belg* 2012;**52**: 351–368.
27. Eriksson M and Mittelmark MB. The Sense of Coherence and Its Measurement. In: MB Mittelmark, S Sagy, M Eriksson, et al. (eds) *The Handbook of Salutogenesis*. Cham (CH):Springer, 2017, pp.97–106.
28. Moons P, Van Deyk K, Marquet K, et al. Individual quality of life in adults with congenital heart disease: a paradigm shift. *Eur Heart J* 2005;**26**: 298–307.
29. Moons P, Budts W and De Geest S. Critique on the conceptualisation of quality of life: a review and evaluation of different conceptual approaches. *Int J Nurs Stud* 2006;**43**: 891–901.
30. Bratt EL and Moons P. Forty years of quality-of-life research in congenital heart disease: Temporal trends in conceptual and methodological rigor. *Int J Cardiol* 2015;**195**: 1–6.
31. Moons P. Why call it health-related quality of life when you mean perceived health status? *Eur J Cardiovasc Nurs* 2004;**3**: 275–277.
32. Moons P, Van Deyk K, De Bleser L, et al. Quality of life and health status in adults with congenital heart disease: a direct comparison with healthy counterparts. *Eur J Cardiovasc Prev Rehabil* 2006;**13**: 407–413.
33. Hofstede G, Hofstede GJ and Minkov M. *Cultures and organizations: Software of the mind*. New York:McGraw-Hill USA, 2005, p.550.
34. RStudio Team. *RStudio: Integrated development for R*. Boston, MA:RStudio, Inc., 2015.
35. Apers S, Kovacs AH, Luyckx K, et al. Quality of Life of Adults With Congenital Heart Disease in 15 Countries: Evaluating Country-Specific Characteristics. *J Am Coll Cardiol* 2016;**67**: 2237–2245.
36. Moons P and Norekval TM. Is sense of coherence a pathway for improving the quality of life of patients who grow up with chronic diseases? A hypothesis. *Eur J Cardiovasc Nurs* 2006;**5**: 16–20.
37. Moons P, Kovacs AH, Luyckx K, et al. Patient-reported outcomes in adults with congenital heart disease: Inter-country variation, standard of living and healthcare system factors. *Int J Cardiol* 2018;**251**: 34–41.
38. Super S, Wagemakers MA, Picavet HS, et al. Strengthening sense of coherence: opportunities for theory building in health promotion. *Health Promot Int* 2016;**31**: 869–878.
39. Thomet C, Moons P, Scherzmann M, et al. Self-efficacy as a predictor of patient-reported outcomes in adults with congenital heart disease. *Eur J Cardiovasc Nurs* 2018;**17**: 619–626.
40. Acuña Mora M, Sparud-Lundin C, Bratt E-L, et al. Person-centred transition programme to empower adolescents with congenital heart disease in the transition to adulthood: a study protocol for a hybrid randomised controlled trial (STEPSTONES project). *BMJ Open* 2017;**7**: e014593.
41. Acuña Mora M, Sparud-Lundin C, Burstrom A, et al. Patient empowerment and its correlates in young persons with congenital heart disease. *Eur J Cardiovasc Nurs* 2019;**18**: 389–398.
42. Berghammer MC, Mattsson E, Johansson B, et al. Comparison of participants and non-participants in patient-reported outcome surveys: the case of Assessment of Patterns of Patient-Reported Outcomes in Adults with Congenital Heart disease - International Study. *Cardiol Young* 2017;**27**: 427–434.
43. Hendriks T, Nilsson M and Westman G. Sense of coherence in three cross-sectional studies in Northern Sweden 1994, 1999 and 2004 – patterns among men and women. *Scand J Public Health* 2008;**36**: 340–345.
44. Van Bulck L, Luyckx K, Goossens E, et al. Patient-reported outcomes of adults with congenital heart disease from eight European countries: scrutinising the association with healthcare system performance. *Eur J Cardiovasc Nurs* 2019;**18**: 465–473.
45. Malm D, Fridlund B, Ekblad H, et al. Effects of brief mindfulness-based cognitive behavioural therapy on health-related quality of life and sense of coherence in atrial fibrillation patients. *Eur J Cardiovasc Nurs* 2018;**17**: 589–597.
46. Kristofferzon ML, Engstrom M and Nilsson A. Coping mediates the relationship between sense of coherence and mental quality of life in patients with chronic illness: a cross-sectional study. *Qual Life Res* 2018;**27**: 1855–1863.