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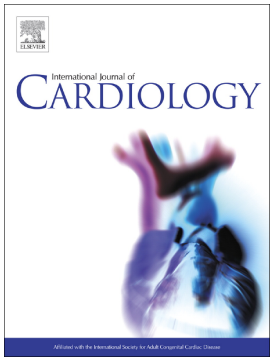
Rationale, design and methodology of APPROACH-IS II: International study of patient-reported outcomes and frailty phenotyping in adults with congenital heart disease



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PII: S0167-5273(22)01018-X

DOI: https://doi.org/10.1016/j.ijcard.2022.06.064

Reference: IJCA 30418

To appear in: *International Journal of Cardiology*

Received date: 20 April 2022

Revised date: 22 June 2022

Accepted date: 27 June 2022

Please cite this article as: L. Van Bulck, A.H. Kovacs, E. Goossens, et al., Rationale, design and methodology of APPROACH-IS II: International study of patient-reported

outcomes and frailty phenotyping in adults with congenital heart disease, *International Journal of Cardiology* (2022), https://doi.org/10.1016/j.ijcard.2022.06.064

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Rationale, design and methodology of APPROACH-IS II: international study of patient-reported outcomes and frailty phenotyping in adults with congenital heart disease

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ABSTRACT

Background: In recent years, patient-reported outcomes (PROs) have received increasing prominence in cardiovascular research and clinical care. An understanding of the variability and global experience of PROs in adults with congenital heart disease (CHD), however, is still lacking. Moreover, information on epidemiological characteristics and the frailty phenotype of older adults with CHD is minimal. The APPROACH-IS II study was established to address these knowledge gaps. This paper presents the design and methodology of APPROACH-IS II.

Methods/design: APPROACH-IS II is a cross-sectional global multicentric study that includes Part 1 (assessing PROs) and Part 2 (investigating the frailty phenotype of older adults). With 53 participating centers, located in 32 countries across six continents, the aim is to enroll 8,000 patients with CHD. In Part 1, self-report surveys are used to collect data on PROs (e.g., quality of life, perceived health, depressive symptoms, autonomy support), and explanatory variables (e.g., social support, stigma, illness identity, empowerment). In Part 2, the cognitive functioning and frailty phenotype of older adults are measured using validated assessments.

Discussion: APPROACH-IS II will generate a rich dataset representing the international experience of individuals in adult CHD care. The results of this project will provide a global view of PROs and the frailty phenotype of adults with CHD and will thereby address important knowledge gaps. Undoubtedly, the project will contribute to the overarching aim of improving optimal living and care provision for adults with CHD.

Keywords: Congenital Heart Disease, Frailty phenotype, Patient-Reported Outcomes

1. BACKGROUND

Congenital heart disease (CHD) is the most common form of congenital defect among newborns, with a global birth prevalence of 9.4 per 1,000.(1) As the life expectancy of patients with CHD is increasing, the population is substantially growing and aging, especially in higher-income countries.(2) Because patients with CHD remain at increased risk for comorbidities, they require lifelong follow-up to optimize outcomes. The epidemiological characteristics and healthcare needs of the 'emerging' group of older adults with CHD should be identified for optimal care planning. In addition, the focus has expanded from improving longevity to also enhancing patient-reported outcomes.(3)

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It is essential to understand the outcomes and experiences from patients' perspectives, namely via patient-reported outcomes (PROs). PROs are defined as *"any report of the status of a patient's health condition, health behaviour, or experience with health care that comes directly from the patient, without interpretation of the patient's response by a clinician or anyone else".(4)* PROs are related to a broad range of patient outcomes, such as mortality and resource use.(5, 6) The original APPROACH-IS study, which ran from 2013 until 2015, investigated PROs among adults with CHD around the globe.(7-9) This earlier study identified intercountry variation in PROs and detected associations both at the individual and contextual level.(10) Most variance in PROs could be explained by individual medical, demographic, behavioral, psychological, and social factors. Little variance could be explained by country-level characteristics. Indeed, APPROACH-IS has already answered some important initial questions.

However, gaps in our knowledge base remain.(9) Although a broad list of PROs and explanatory factors were included in the original APPROACH-IS study, only part of the variation in PROs could be explained, indicating a need to investigate the association between PROs and additional explanatory factors.(10) Moreover, a particular type of PRO, experiences with health care, remain largely uninvestigated in adults with CHD, leaving questions unanswered about the quality of care, the geographical variation and predictors of patient-reported experiences with care. Furthermore, the initial APPROACH-IS study included patients from 13 high-income and two middle-income countries.(7) An understanding of PROs in patients living in low- and middle-income countries, in comparison to high-income countries, is still lacking.

Furthermore, as adults with CHD are ageing, many will encounter disability, morbidity and a state of frailty, thereby increasing susceptibility for adverse outcomes and premature mortality.(11) To maintain longevity and quality of life, an understanding of variables associated with prognosis, comorbidity and mortality will enable us to map specific healthcare needs.(11) Frailty phenotype refers to a distinct clinical syndrome that classifies patients as non-frail, pre-frail, or frail based upon the assessment of five criteria: weakness, slow walking speed, unintentional weight loss, exhaustion, and low physical activity.(12) Growing evidence suggests that independent of age and comorbidity, frailty phenotyping can guide risk prediction in chronically ill patients.(13) Unfortunately, our current

knowledge about epidemiological characteristics, frailty phenotype and healthcare needs of ageing adults with CHD is very limited.(13)

These factors led to the decision to proceed with a second APPROACH-IS study, with an expanded list of PROs and explanatory variables and for which data from patients living in low- and middle-income countries are included. Moreover, APPROACH-IS II will also deliver much-needed empirical data describing the clinical and epidemiological characteristics of the emerging population of older adults with CHD. The paper aims to describe the design and methodology of the APPROACH-IS II study.

2. STUDY OBJECTIVES

The aims of APPROACH-IS II are (i) to increase our understanding of PROs in adults with CHD by enrolling adults with all types of CHD from low-, middle- and high-income countries and including a novel set of potential explanatory variables; and (ii) to assess the profile and healthcare needs of older adults with moderate to complex CHD, with a particular focus on frailty.

3. DESIGN AND METHODS

The project has a cross-sectional global multicentric design and consists of Part 1 (PROs) and Part 2 (frailty phenotype) (see Figure 1). All participating centers contribute data to Part 1, and data collection for Part 2 is optional (and likely depends upon local research resources).

PLEASE INSERT FIGURE 1 ABOUT HERE

3.1 Part 1: PROs

Data collection procedures

In Part 1 of the study, patients are asked to complete a set of self-reported surveys. Participating centers can recruit patients using one of four recruitment strategies:

1. Eligible patients can be approached consecutively at outpatient clinics for adults with CHD. Consecutive sampling is a technique in which every eligible case is selected until the required sample size is achieved. Following informed consent, patients may complete surveys while in the clinic (on paper or online) or at home (online or by returning paper surveys in a pre-addressed and pre-stamped envelope).

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2. Eligible patients can be randomly selected from the institution's database and receive a study package, including an information letter, two copies of the informed consent form, surveys, and an addressed envelope, by mail. Reminders can be sent out to non-responders.
3. Eligible patients can be identified from the institution's database and receive an email with a link to the online survey (i.e., REDCap). Informed consent can be obtained at an outpatient clinic, over the telephone, or online before the completion of the surveys. Reminders can be used to increase the response rate.
4. Eligible patients can be identified from the institution's database and receive a phone call to complete the survey. Informed consent can be obtained at an outpatient clinic visit, by phone or online. The option to collect data over the phone is limited to centers with a population with low (written) health literacy levels.

In addition to study surveys, clinical medical data are collected from the medical records of each study participant, overseen by a member of the medical team. The anatomic complexity and current physiological stage are measured and categorized according to the ACHD anatomical and physiological classification (see Table 1).(14)

Participating centers are responsible for the local data collection process. Data collection began in August 2019, was paused in all centers from March 2020 until June 2020 due to the COVID-19 pandemic, and will be completed by August 2022. In some centers, data collection was paused longer or again at a later date, depending on the local pandemic situations. Details regarding decision-making to pause the data collection process due to COVID-19 have been published elsewhere.(15)

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Sample

Participants are eligible if they fulfill the following criteria: (i) diagnosed with CHD, defined as: "*a gross structural abnormality of the heart and/or intra-thoracic great vessels that is actually or potentially of functional significance (including mild, moderate, and complex heart defects)*"(16); (ii) aged 18 years or older at the date of study entry; (iii) diagnosed with CHD before the age of 10 years; (iv) followed at an adult CHD center or included in a national/regional registry; (v) demonstrated physical, cognitive and language abilities required to complete self-report questionnaires. Patients are excluded if they received a heart transplantation before study participation.

The recruitment goal is 200 patients per center. This goal is determined based on a survey sample size calculation and feasibility for all participating centers. The survey sample size calculation takes into account the margin of error (measure of accurateness), size of the population and alpha level.(17) With a recruitment goal of 200 patients per center and an alpha level of 95%, the margin of error is around 7% for analyses of the PROs, which is acceptable. In addition, in the previous APPROACH-IS study, the sample size of 200 per center proved feasible for larger and smaller centers.(7) Although 53 centers are participating in APPROACH-IS II, we realize that centers located in low- and middle-income countries might have difficulties achieving this recruitment goal due to fewer patients in adult CHD care; as such, we estimate a total sample size of 8,000 patients for Part 1 of the project.

Variables

An overview of the core battery of questionnaires included in Part 1 of the study, their interpretation and psychometric properties are presented in Table 1.

Four patient-reported outcomes are included: perceived health status, psychological distress, quality of life, and patients' perception of providers' autonomy support. Perceived health status is measured using the 12-item shortened and adapted version of the RAND-36.(18) It is a disease-generic measure of eight health domains: physical functioning, role participation with physical health problems, bodily pain, general health, vitality, social functioning, role participation with emotional health problems, and mental health. Psychological distress is assessed using the Patient Health Questionnaire-8 (PHQ-8) for depression (19) and the General Anxiety Disorder-7 (GAD-7) for anxiety.(20) The PHQ-8 includes eight of the nine criteria of the DSM–IV (Diagnostic and Statistical Manual of Mental

Disorders-IV) diagnosis of depressive symptoms; it does not include the item about suicidal or self-injurious ideation.(19) Quality of life is determined using a 0 – 100 linear analog scale.(21) Our experience measure is the brief Health Care Climate Questionnaire, which has been modified for use with adults with CHD and assesses patients' perceptions of the degree to which their team of healthcare providers is supporting their autonomy (versus them taking control).(22) The questionnaire is based on self-determination theory, which proposes that individuals tend to feel more competent when they are autonomously motivated.(23)

The survey packet includes additional explanatory variables. The Stigma Scale for Chronic Illnesses measures multifaceted (both enacted and internalized) stigma.(24) The Illness Identity Questionnaire assesses four illness identity dimensions (i.e., engulfment, rejection, acceptance and enrichment).(25) Empowerment, the capacity of individuals to become responsible for their health,(26) is assessed by the Gothenburg Empowerment Scale,(27) which has five dimensions: identity, knowledge and understanding, personal control, shared-decision making, and enabling others (i.e. peers with similar conditions).(28) Healthcare use (i.e., hospitalizations, visits to the general practitioner, medical specialist or emergency department in the last 12 months) was captured using a healthcare measure that has previously been used with adults with CHD; a distinction is made between healthcare use related to CHD vs. other diseases/symptoms.(29) The Multidimensional Perceived Social Support Scale (MSPSS) assesses perceived social support, which refers to how individuals perceive family, friends and significant others as sources available to provide psychosocial, materialistic and overall support during times of need.(30) Parental involvement is measured using a modified version of the MSPSS, in which the items reflect perceived social support by parents. Social media and advance care planning are measured using survey items developed by the Steering Committee based on existing surveys.(31) Information about basic demographic variables are also collected. Surveys for Part 1 were carefully selected based on their validity, reliability and availability in different languages.

PLEASE INSERT TABLE 1 ABOUT HERE

3.2 Part 2: Healthcare needs of older CHD patients

Sample

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For Part 2, additional inclusion criteria are (i) age of 40 years or older, and (ii) CHD diagnosis of moderate or great complexity.(14) Patients who meet these additional inclusion criteria can complete study procedures for both Part 1 and Part 2. Centers that are collecting data for Part 2 are asked to aim to enroll (i) 20 adults aged 40-50 years, (ii) 20 adults aged 51-60 years, and (ii) 20 adults older than 60 years. The estimation of the sample size for Part 2 was 800 patients; 21 centers are participating in Part 2 of the project, but we realize that many centers will encounter difficulties enrolling patients in the older cohorts.

Data collection procedure

In Part 2 of the study, patients participate in several assessments carried out by a research assistant during a patient visit at an outpatient clinic.

Variables

Two primary outcomes, namely cognitive functioning and frailty phenotype, are included. Cognitive functioning is assessed using the Montréal Cognitive Assessment (MoCA).(32) The MoCA assesses different cognitive domains, including attention, concentration, executive functions, memory, language, visuospatial skills, abstraction, calculation and orientation. Frailty phenotype is assessed using the Fried method.(12) This method consists of five parts: self-report questions about unintentional weight loss, exhaustion and physical activity, an assessment of weakness performed using a handgrip dynamometer, and a walk test. In addition, the Charlson Comorbidity Index is determined based on medical files for every participant to provide information on the presence and burden of comorbidities.(33) Table 2 contains an overview of the included variables in Part 2.

PLEASE INSERT TABLE 2 ABOUT HERE

4. DATA ANALYSIS

Descriptive statistics (means, standard deviations, medians and interquartile ranges) will be calculated and compared between participating centers. Multilevel analyses will be performed because the data have a hierarchical nature. More specifically, data will be organized at the level of (1) the individual patient, (2) the center, and (3) the country. Data of individual patients are nested within the center and country levels (i.e., aggregate units). General and generalized linear mixed models will be used to analyze continuous, and binomial or count variables, respectively. Additional analyses will be

performed to determine the psychometric properties of the surveys. Frailty phenotypes will be calculated based on the Fried method. Multilevel multivariable analyses will be performed by grouping patients with comparative phenotypes and exploring trends and associations in terms of frailty phenotype, comorbidity burden, and healthcare consumption.

5. PARTICIPATING CENTERS

Centers are eligible to take part if (i) participation is feasible in terms of infrastructure and clinical research resources, and (ii) patient volume is sufficient to support the recruitment of an adequate number of patients. As shown in Figure 2, 53 centers across 32 countries are participating in Part 1 of the study. A total of 21 centers, across 15 countries, are also participating in Part 2 of the study. The full list of participating centers is available in the supplementary material (eTable 1).

PLEASE INSERT FIGURE 2 ABOUT HERE

6. PROJECT MANAGEMENT

The University of Leuven (KU Leuven, Belgium) is the coordinating center of APPROACH-IS II and is responsible for the general management and administration, as carried out by the international project coordinator (LVB). All aspects of this international study are overseen by the Steering Committee (PM, EG, KL, AK) that makes substantive decisions and has final responsibility for scientific conduct. All participating centers have a local principal investigator, responsible for the study execution in their center.

Standard Operating Procedures (SOPs) were developed to standardize processes during the preparatory phase, data collection and data management, and to ensure the use of a uniform methodology. The Open Science Framework (https://osf.io/) is used as a platform to store and share relevant documents about the project. For survey distribution, data entry and data hosting, REDCap (Research Electronic Data Capture) was used, which is a secure, web-based software platform designed to support data capture for research studies.(34) The use of REDCap for the APPROACH-IS II project has been described in more detail elsewhere.(34) Project information, updates, and data collection progress are available on the study website (www.approach-is.net), and via information flashes regularly distributed to the consortium.

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7. TRANSLATIONS

Given the global geographic distribution of this project, survey documents were required in 22 different languages. If there was no available translation for a measure, the local team undertook this process using a standardized academic translation protocol, based on WHO guidelines.(35) This protocol includes a forward translation, a backward translation, pre-testing in a few patients, proofreading, finalization and documentation. No substantial changes to the English version of the survey were permitted.

8. ETHICAL ISSUES

The Institutional Review Board of the University Hospitals Leuven/KU Leuven (i.e., the coordinating center) approved the main study protocol of APPROACH-IS II and each participating center obtained local ethics approval for study execution. Written informed consent is obtained from all participants as required; in some regions, legislation stipulates that written informed consent is unnecessary for survey studies. The project is conducted in accordance with the declaration of Helsinki. The protocol is registered at ClinicalTrials.gov (NCT04902738).

Data management is conducted in accordance with current worldwide privacy regulations. Participant confidentiality is a priority in this project. The coordinating center did not collect patient names, medical record numbers, or dates of birth. Only non-identifiable information was made available to other participating centers.

9. COVID-19

The APPROACH-IS II study has faced unexpected challenges and made several adaptations due to the COVID-19 pandemic.(15) Enrollment was paused during the first months of the coronavirus outbreak in all participating centers, because of a potential risk of biased results (March till June 2020). Data collection was also paused locally by many participating centers during outbreaks of the virus, as this typically entailed a shift in clinical care and research priorities as well as a reduction in routine outpatient visits.

As increased levels of anxiety and depression have been documented in the general population during the pandemic, we realized that data collected peri-pandemic could be at risk of bias.(36) Therefore, in three participating centers, namely Leuven (Belgium), Oslo (Norway) and Seoul (South

Korea) in which data collection was completed before the first COVID-19 outbreak, a second measurement wave was set up one year after the first measurement wave.(36) Published results revealed that, fortunately, differences between pre- and peri-pandemic PROs were very small and clinically negligible on a group level and no differences were observed between patients who had and had not been infected with COVID-19.(36) Hence, we may be relatively confident that the results of this project will not be biased by the pandemic.

10. DISCUSSION

Optimization of quality of life remains a healthcare priority for the growing and ageing population of patients with CHD. However, important questions remain unresolved. International variation in PROs is only partly understood (10) and an elucidation of PROs in patients living in low- and middle-income countries is currently lacking. In addition, our field currently lacks data on the frailty phenotype and healthcare needs of ageing adults with CHD.(13) The APPROACH-IS II project will contribute to addressing these respective issues. With 53 participating centers located in 32 countries, APPROACH-IS II will be one of the largest collaborations on PROs worldwide and will generate a large study sample of around 8,000 patients. As high-, middle- and low-income countries from six continents are represented, the data will reflect cultural and regional diversities of the adult CHD population. The project employs a robust and uniform methodology, which will generate and safeguard reliable data.

This project has some limitations. First, the project has a cross-sectional research design, which will not allow to determine the direction of effects. Using advanced techniques, it will be possible to make causal inferences on this large database. Moreover, in three centers a second measurement wave has been set-up that generated longitudinal data. Second, only patients who are physically or mentally capable of completing surveys are included. Indeed, this impacts the generalizability of the results. Third, although high-, middle- and low-income countries are represented in this study, still a larger number of high-income countries are included, leading to their overrepresentation.

Despite these limitations, we are confident that APPROACH-IS will address knowledge gaps about PROs and the frailty phenotype of adults with CHD worldwide.

11. FUNDING

This work is supported by KU Leuven – University of Leuven, Research Foundation Flanders through grants 1159522N and 12E9819N, Swedish Heart and Lung Foundation through grant 20190525, the University Research Council of the Aga Khan University in Karachi in Pakistan, Maltese Cardiac Society, UnIC (Unidade de Investigação Cardiovascular da FMUP) supported the costs of the translations into Portuguese used in Portugal, Children's Heart Unit Fund (www.chuf.org.uk), Newcastle upon Tyne through their funding of the CHUF Fontan nurse specialist. This study was partially supported by Ricerca Corrente funding from the Italian Ministry of Health to IRCCS Policlinico San Donato.

12. ACKNOWLEDGEMENTS

We want to thank all APPROACH-IS II participants and all individuals at the participating centers who are making substantial contributions to this project.

13. DECLARATION OF INTEREST

Declarations of interest: none

14. APPROACH-IS II CONSORTIUM

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FIGURES

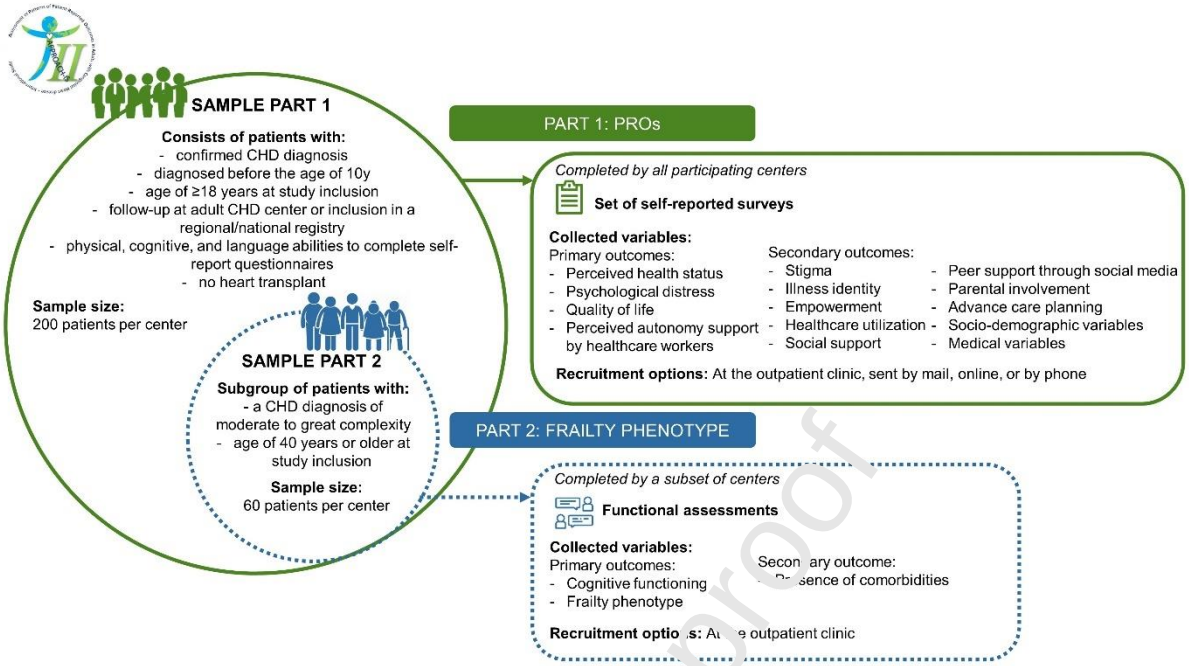

Figure 1: Structure of the APPROACH-IS II project

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Figure 2: Geographic distribution of the APPROACH-IS II participating centers

Legend. Yellow dots indicate centers that are participating in Part 1 and Part 2 of the study (n=21).

Pink dots indicate centers that are participating in Part 1 only (n=32).

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Table 1. Details and psychometric properties of the surveys used in Part 1 of APPROACH-IS II

Variable	Source	Measurement tool	# items	Validity	Reliability	Use in cardiac population	Interpretation
Socio-demographic variables - Age - Sex - Marital status - Number of children - Cultural background/ethnicity - Educational level - Employment status - Religion	Self-report	Survey developed by Steering Committee	13	NA	NA	NA	NA
Medical variables - New York Heart Association functional class - Height and weight	Self-report	Survey developed by Steering Committee	3	NA	NA	NA	NA
- Diagnosis of CHD - History of cardiac surgeries / interventions - Number of cardiac admissions (over past 5 years) - Number of cardiac outpatient visits (over past 5 years)	Chart review	Form developed by Steering Committee	12	NA	NA	NA	NA
- Aortopathy - Arrhythmia - Concomitant valvular heart disease - End-organ dysfunction - Exercise capacity - Hypoxemia/hypoxia/cyanosis - NYHA functional classification system (physician assessment) - Pulmonary hypertension - Shunt (hemodynamically significant shunt) - Venous and arterial stenosis	Chart review	Form based on the ACHD Anatomical and Physiological Classification System (14)	21	NA	NA	NA	NA

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Primary outcomes							
Perceived health status	Self-report	12-item shortened version of the RAND-36 (18)	12	Supported (37)	Supported (37)	Yes, in adults with CHD (7)	Composite physical (PCS) and mental health (MCS) scores are computed. Scores range from 0 (lowest health level) to 100 (highest health level).
		Linear Analogue Scale Health Status (LAS HS) (21)	1	Supported (21)	Supported (21)	Yes, in adults with CHD (21)	Scores range from 0 (worst imaginable health state) to 100 (best imaginable health state).
Psychological distress	Self-report	Patient Health Questionnaire 8 (19)	8	Supported (38)	Supported (38)	Yes, in adults with CHD (39)	Scores range from 0 to 24. Scores of ≥10 indicate depression.
		General Anxiety Disorder 7 (20)	7	Supported (38)	Supported (38)	Yes, in adults with CHD (39)	Scores range from 0 to 21. Scores of 5, 10, and 15 are taken as cut-off points for mild, moderate and severe anxiety.
Quality of life	Self-report	Linear Analog Scale Quality of Life (LAS QOL) (21)	1	Supported (21)	Supported (21)	Yes, in adults with CHD (21)	Scores range from 0 (worst imaginable quality of life) to 100 (best imaginable quality of life)
Perceived autonomy support by health workers	Self-report	Modified Health Care Climate Questionnaire (22)	6	Supported (40)	Supported (40)	Yes, in patients with cardiovascular disease (41)	Each of the 6 items is scored from 1 to 7. Scores are calculated by averaging the individual item scores. Higher average score represents a higher level of perceived autonomy support.

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Secondary outcomes							
Stigma	Self-report	Chronic Illness Stigma Scale (CISS) (24)	8	Supported (24)	Supported (24)	No, used in patients with chronic disease, but not yet used in cardiac populations	Scores range from 8 to 40. Higher scores indicate higher levels of perceived stigma.
Illness identity	Self-report	Illness Identity Questionnaire (IIQ) (25)	25	Supported (42)	Supported (42)	Yes, in adults with CHD (25)	Consists of five-item rejection scale, seven-item enrichment scale, five-item acceptance scale and eight-item engulfment scale. A mean score is calculated per subscale. Higher scores indicate more rejection, enrichment, acceptance or engulfment.
Empowerment	Self-report	Gothenburg Empowerment Scale (GES generic v1.1) (27)	15	Supported (43)	Supported (43)	Yes, in adolescents with CHD (43)	Scores range from 15 to 75. Higher score reflects a higher level of empowerment.
Healthcare utilization	Self-report	Patient-Reported In- and outpatient Utilization Scale (PRIUS) (29)	16	NA	NA	Yes, in adults with CHD (29)	Higher numbers indicate more healthcare use.
Perceived Social Support	Self-report	Multidimensional Perceived Social Support Scale (MSPSS) (30)	12	Supported (44)	Supported (44)	Yes, in adults with CHD (45)	Scores range from 12-84. Higher score indicates greater social support perceived by an individual.
Social media to connect with peers	Self-report	Survey developed by the Steering Committee	3	NA	NA		NA

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Parental Involvement	Self-report	Adapted version of the items, retrospectively reflecting perceived social support by parents of the MSPSS (30)	5	NR	NR	Scores range from 5-35. Higher score indicates greater parental support in childhood and adolescence.
Thinking about the future (advance care planning)	Self-report	Survey developed by the Steering Committee	5	NA	NA	NA

Legend. CHD: congenital heart disease, NA: not applicable, NR: not reported

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Table 2. Variables included in Part 2 of APPROACH-IS II

Variable	Source	Measurement tool	# items	Validity	Reliability	Use in cardiac population	Interpretation
Cognitive functioning	Assessment by research assistant	Montréal Cognitive Assessment (32)	30	Supported (32)	Supported (32)	Yes, in adolescents and young adults with CHD (46)	Scores range from 0 to 30. Scores of <26 indicate cognitive dysfunction.
Frailty phenotype (i.e., non-frail/pre-frail/frail)	Assessment by research assistant	Fried method (12): 1. Unintentional weight loss: self-reported 2. Exhaustion: self-reported 3. Low physical activity level: self-reported 4. Weakness: test performed using a handgrip dynamometer, assessment performed by a research assistant and adjusted for sex and body mass index 5. Slow walking speed: based on time to walk 15 feet, assessment performed by a research assistant and adjusted for sex and standing height	5	Supported (12, 47)·(48)	Supported (47)	Yes, in patients with cardiac disease (48)	Frail: when ≥3 criteria are positive Pre-frail: 1 or 2 criteria are positive Robust or non-frail: no criterion is positive
Presence and burden of comorbidities	Chart review	Charlson Comorbidity Index (33)	19	Supported (49)	Supported (49)	Yes, in adults with CHD (50)	Comorbidities range from 1 to 6 points. The final score is obtained via the summation of applicable points and ranges from 0 (no disease burden) to 29 (maximal disease burden).

Legend. CHD: congenital heart disease.

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Highlights

- The variability of patient-reported outcomes in adults with congenital heart diseases (CHD) is not fully understood yet.
- Available information on the frailty phenotype of older adults with CHD is minimal.
- The research project APPROACH-IS II will establish these knowledge gaps.
- APPROACH-IS II is a cross-sectional global study in which 53 centres from 32 countries participate.

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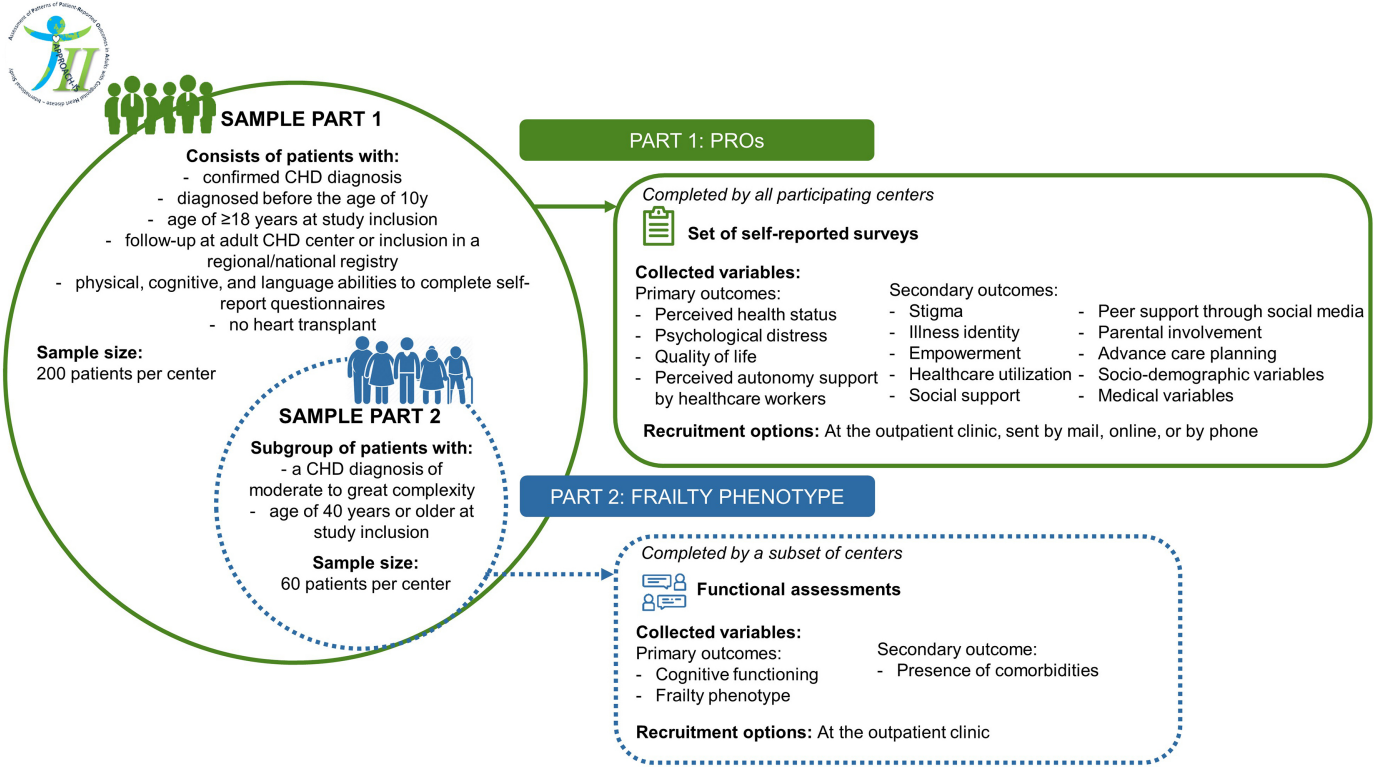
SAMPLE PART 1
Consists of patients with:
- confirmed CHD diagnosis
- diagnosed before the age of 10y
- age of ≥18 years at study inclusion
- follow-up at adult CHD center or inclusion in a regional/national registry
- physical, cognitive, and language abilities to complete self-report questionnaires
- no heart transplant
Sample size:
200 patients per center
SAMPLE PART 2
Subgroup of patients with:
- a CHD diagnosis of moderate to great complexity
- age of 40 years or older at study inclusion
Sample size:
60 patients per center
PART 1: PROs
Completed by all participating centers
Set of self-reported surveys
Collected variables:
Primary outcomes:
- Perceived health status
- Psychological distress
- Quality of life
- Perceived autonomy support by healthcare workers
Secondary outcomes:
- Stigma
- Illness identity
- Empowerment
- Healthcare utilization
- Social support
- Peer support through social media
- Parental involvement
- Advance care planning
- Socio-demographic variables
- Medical variables
Recruitment options: At the outpatient clinic, sent by mail, online, or by phone
PART 2: FRAILTY PHENOTYPE
Completed by a subset of centers
Functional assessments
Collected variables:
Primary outcomes:
- Cognitive functioning
- Frailty phenotype
Secondary outcome:
- Presence of comorbidities
Recruitment options: At the outpatient clinic

Figure 1

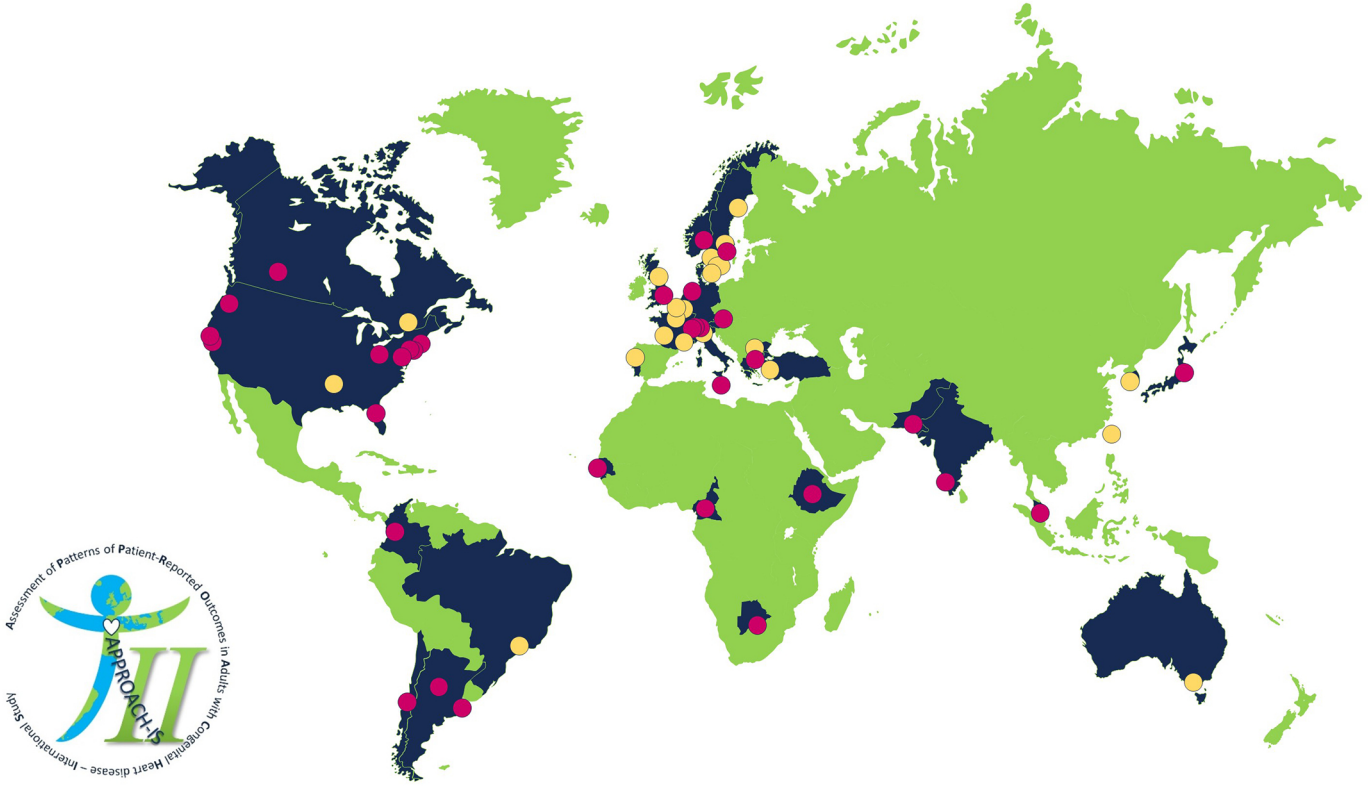
Assessment of Patterns of Patient-Reported Outcomes in Adults with Congenital Heart disease – International Study
APPROACH-IS

Figure 2