Received: 13 November 2020 | Revised: 26 May 2021 | Accepted: 15 June 2021

DOI: 10.1111/jan.14949

ORIGINAL RESEARCH:
EMPIRICAL RESEARCH – QUANTITATIVE

JAN Leading Global Nursing Research | WILEY

Illness identity in adults with congenital heart disease: Longitudinal trajectories and associations with patient-reported outcomes and healthcare use

Liesbet Van Bulck[1,2] | Eva Goossens[1,2,3] | Silke Apers[4] | Philip Moons[1,5,6] | Koen Luyckx[7,8]

[1]Department of Public Health and Primary Care, KU Leuven – University of Leuven, Leuven, Belgium

[2]Research Foundation Flanders (FWO), Brussels, Belgium

[3]Faculty of Medicine and Health Sciences, Centre for Research and Innovation in Care, Division of Nursing and Midwifery, University of Antwerp, Antwerp, Belgium

[4]Department of Gynaecology and Obstetrics, University Hospitals Leuven, Leuven, Belgium

[5]Institute of Health and Care Science, University of Gothenburg, Gothenburg, Sweden

[6]Department of Paediatrics and Child Health, University of Cape Town, Cape Town, South Africa

[7]Department of School Psychology and Development in Context, KU Leuven – University of Leuven, Leuven, Belgium

[8]UNIBS, University of the Free State, Bloemfontein, South Africa

Correspondence
Philip Moons, Department of Public Health and Primary Care, KU Leuven – University of Leuven, Kapucijnenvoer 35 (box 7001) B-3000 Leuven, Belgium.
Email: philip.moons@kuleuven.be

Funding information
This work was supported by the Research Fund - KU Leuven through grant OT/11/033; by the Research Foundation Flanders through grants G0B3514N, G097516N, 1154719N and 12E9819N; and by the Research Council KU Leuven through grant c14/15/036.

Abstract

Aims: The aims of this study were to investigate the development of illness identity, the degree to which the disease is integrated into one's identity, by identifying trajectory classes in adults with congenital heart disease (CHD) and to describe these classes in terms of age, sex, disease complexity, patient-reported outcomes (PROs) and healthcare use.

Design: This three-wave observational cohort study was carried from 2013 till 2015 and includes 276 adults (median age: 34 years; 54% men) with CHD.

Methods: Illness identity entails four dimensions: engulfment, rejection, acceptance and enrichment. PROs included perceived health status, quality of life and psychological distress. Latent class growth analysis, analysis of variance, Poisson regression and negative binomial regression analyses were performed.

Results: Illness identity was relatively stable over time. The identified classes were meaningfully different in terms of age, disease complexity, PROs and healthcare use. Patients who did not reject their disease, patients who were not overwhelmed or patients who accepted their disease over time reported better health status and quality of life and less psychological distress. Less hospitalizations and visits to the general practitioner and medical specialist were reported by patients who were not overwhelmed or patients who accepted their disease over time. Patients with low rejection and high enrichment scores over time reported more visits at the general practitioner.

Conclusion: These findings indicate that illness identity should be taken into account when trying to understand and optimize PROs and healthcare use of adults with CHD.

Impact: This study scrutinizes the development and clinical meaningfulness of illness identity measured over time for adults with CHD. Illness identity was found to be stable over time. Moreover, the illness identity trajectories differed in terms of PROs and healthcare use, showing that measuring and intervening upon illness identity could be a potential pathway to optimize PROs and healthcare use.

KEYWORDS
anxiety, congenital heart defects, depression, health services research, nursing, psychology, quality of life

1 | INTRODUCTION

Survival of patients with congenital heart disease (CHD) has improved remarkably over the past decades, introducing a growing and aging population of adults with CHD (Mandalenakis et al., 2017; Moons et al., 2010). However, despite surgeries in childhood, the majority of patients has residual abnormalities and complications and cannot be considered to be cured (Engelfriet et al., 2005). This has two important consequences for the healthcare system.

The healthcare perspective for these patients has widened with increased attention for aspects of optimal living, expressed in research on patient-reported outcomes (PROs) such as quality of life (QOL), perceived health and psychosocial functioning (Tzelepis et al., 2015). PROs have shown to be of clinical relevance in cardiac populations because they independently predict cardiovascular events, hospitalizations and mortality (Mommersteeg et al., 2009).

Also, long-life follow-up for this rapidly growing population causes an exponential rise of healthcare use of these patients. Over the past decades, the absolute number of hospitalizations and outpatient cardiology visits has expanded, showing high and changing healthcare demands for this population. These changes will put an increased burden on the healthcare system and society (Willems et al., 2019). To reduce this, a good understanding of the predictors of high healthcare use is needed.

For both PROs and healthcare use, differences between patients with CHD are not sufficiently understood yet. Some patients experience substantial difficulties related to their disease resulting in unfavorable PROs and high healthcare use, whereas others succeed in coping with the disease-related challenges (Moons et al., 2018; Schoormans et al., 2016). Therefore, a good understanding of the modifiable predictors of PROs and healthcare use is needed. One modifiable predictor that has shown to be related to both PROs and healthcare use in adults with CHD is "illness identity" (Van Bulck et al., 2019).

Previous literature has indicated that illness identity may play a key role in regulating PROs and healthcare use in adults with CHD (Andonian et al., 2020; Oris et al., 2018; Van Bulck et al., 2018). However, so far, these associations are only investigated in cross-sectional research, leaving questions about how illness identity may predict these variables over time unanswered (Van Bulck et al., 2019). Indeed, there is a need for longitudinal research to examine the development and stability of illness identity in adults with CHD and to explore whether illness identity over time could be a helpful tool to understand why certain patients do better in terms of PROs and healthcare use than others.

2 | BACKGROUND

Illness identity is defined as "*the degree to which a condition is integrated into one's identity*" and encompasses four dimensions: engulfment, rejection, acceptance and enrichment (see Figure S1) (Charmaz, 1995; Oris et al., 2016). The first two dimensions represent dysfunctional illness identity dimensions and induce maladaptive responses, such as excessive concern or avoidance. Engulfment refers to the degree to which the disease dominates one's identity and daily life. Patients with high engulfment scores mainly define themselves in terms of their disease as they pay a lot of attention to their restrictions in daily life (Morea et al., 2008; Oris et al., 2016). Rejection captures the degree to which the disease is seen as unacceptable to the self (Adams et al., 1997; Oris et al., 2016). Patients with high rejection scores are usually characterized by suboptimal self-management and adherence and refuse to see their disease as part of their identity (Oris et al., 2016; Tilden et al., 2005). Acceptance and enrichment represent more adaptive dimensions of illness integration. Individuals who accept their disease find the right balance between devoting attention to their illness and other aspects of their identity (Adams et al., 1997; Morea et al., 2008; Oris et al., 2016). Enrichment is the degree to which the disease enriches one's sense of self, enables one to grow as a person and results in positive life changes (Oris et al., 2016; Senol-Durak, 2013).

Illness identity has been investigated in several patient populations during the past years, that is, young adults with refractory epilepsy (Luyckx et al., 2018), adolescents with celiac disease (Meyer & Lamash, 2021), youth with type 1 diabetes (Oris et al., 2016; Raymaekers et al., 2020), patients with systemic diseases (Oris et al., 2016) and adults with CHD (Andonian et al., 2020; Oris et al., 2016). In all these populations, associations between the four illness identity dimensions and clinical parameters, PROs and healthcare utilization were found, indicating that illness identity could be a valuable concept that has the potential to support clinical practice. In adults with CHD, the illness identity dimensions were shown to be related to depression, anxiety, self-reported health, hospitalizations and visits at the general practitioner and medical specialist (Andonian et al., 2020; Oris et al., 2018; Van Bulck et al., 2018, 2019). However, further studies ought to investigate the potential of illness identity further, before it can be used in interventions or guidelines to ameliorate clinical care.

3 | THE STUDY

3.1 | Aims

The aims of this study were (a) to identify classes of individuals with CHD characterized by similar longitudinal trajectories of illness identity dimensions and (b) to describe these classes in terms of age, sex, disease complexity, PROs and healthcare use.

3.2 | Design

The present study is a secondary data analysis that used the data of the Belgian branch of the APPROACH-IS project (Apers et al., 2015). The Belgian branch of APPROACH-IS is a longitudinal three-wave observational cohort study with 1-year intervals (T_{2013}, T_{2014} and T_{2015}). A total of 276 patients response rate (RR: 69%), 255 patients (RR: 74%) and 229 patients (RR: 80%) responded on T_{2013}, T_{2014} and T_{2015}, respectively. A more detailed description of the recruitment process can be found in Figure S2 (Van Bulck et al., 2018).

3.3 | Participants

Patients have been selected randomly from the database of adults with CHD of the University Hospitals Leuven in Belgium. Patients were eligible if they were (a) diagnosed with CHD, according to the definition of Mitchell and colleagues (Mitchell et al., 1971), (b) born before 1991, (c) diagnosed before the age of 10 years (to warrant ample experience of living with CHD), (d) in follow-up at the University Hospitals Leuven, (e) Dutch speaking and (f) demonstrating language, cognitive and physical proficiency to complete self-reported surveys. Patients were excluded if they had (a) prior heart transplantation or (b) isolated pulmonary hypertension (Apers et al., 2015).

3.4 | Data collection

The study was carried out from 2013 till 2015. Eligible participants were asked to fill out a set of self-reported surveys. Patients received a study package by postal service, including a study information letter, an informed consent form, a survey package and an addressed prestamped envelope. To increase the response rate, a modified Dillman approach was used (Dillman, 1983).

3.5 | Variables and instruments

Illness identity was measured at all three waves. PROs (i.e., perceived health status, QOL and psychological distress) and healthcare use were measured at T_{2015}. More information about the instruments used to measure PROs and healthcare use can be found in Table 1.

3.5.1 | Validity, reliability and rigour of the instruments

The psychometric properties of the instruments used are described in Table 1 as well.

3.6 | Ethical considerations

Ethics approval was obtained from the ethics research committee UZ/KU Leuven. Procedures were in accordance with the declaration of Helsinki. Written informed consent was obtained from each patient. The protocol was registered at ClinicalTrials.gov (NCT02150603).

3.7 | Data analyses

First, to identify trajectory classes of the illness identity dimensions over time (Research Aim 1), latent class growth analysis (LCGA) was conducted. LCGA is a person-centered approach that summarizes longitudinal information by classifying individuals into trajectory classes (Muthen & Muthen, 2000). Each class consists of individuals showing similar development of illness identity over time and is characterized by unique initial levels (intercepts) and rates of change (slopes). To identify the optimal number of classes within the data, LCGA models for one to four classes ran for each illness identity dimension separately. The best-fitting model was selected based on several fit indices (i.e., the Bayesian Information Criterion [BIC], entropy (*E*) and the Lo–Mendell–Rubin Loglikelihood Ratio Test [LMR-LRT]) and the size of the classes. Supplementary material contains additional information on the LCGA (Annex S1).

Second, to describe the classes in terms of age, sex, disease complexity and PROs (Research Question 2), Kruskal–Wallis H, chi-square tests and multivariate analyses of variance with pairwise comparisons using Gabriel's test procedure (i.e., post hoc tests) were performed. An expectation–maximization algorithm was performed to estimate the missing PROs. To investigate to what extent the trajectory classes differed from each other in terms of healthcare use, multivariate Poisson (for hospitalizations and visits to medical specialists) and negative binomial regression analyses (for visits to general practitioners) were performed, adjusting for age, disease complexity and PROs.

LCGA was conducted in Mplus 8.0. The remaining analyses were conducted in SPSS version 26 with the significance level at $p \leq .05$ (two tailed).

4 | RESULTS

4.1 | Sample characteristics

Table 2 shows the sociodemographic variables, derived through self-report, and clinical variables, derived through chart review of the medical records, of the study sample at baseline (T_{2013}). Age ranged from 22 to 78 years, 46.0% of patients were women, almost all patients (96.0%) finished at least high school and most patients (63.3%) worked full time. Disease complexity was categorized following the classification of Task Force 1 of the 32nd Bethesda conference, as simple (33.7%), moderate (54.3%) or great (12.0%) (Warnes et al., 2001).

TABLE 1 Overview of the properties of instruments

Variable	Instrument	Structure of the instrument	Reliability	Validity	Use in cardiac samples	Interpretation
Illness identity	Illness Identity Questionnaire (IIQ) (Oris et al., 2016)	- 25 statements - 5-point Likert scale ranging from strongly disagree to strongly agree - Consists of 4 subscales: • rejection scale with 5 items • acceptance scale with 5 items • engulfment scale with 8 items • enrichment scale with 7 items	Supported (Oris et al., 2016) Cronbach α values in present study for T_{2013}, T_{2014} and T_{2015} respectively: • 0.75, 0.77 and 0.78 for rejection • 0.83, 0.81 and 0.83 for acceptance • 0.92, 0.92 and 0.93 for engulfment • 0.95, 0.94 and 0.95 for enrichment	Supported (Oris et al., 2016)	Yes, in adults with CHD (Oris et al., 2018)	- Mean score per subscale - Higher scores = higher level of rejection, acceptance, engulfment or enrichment
Perceived health status	EuroQoL-Visual Analogue Scale (EQ-VAS) (EuroQol Group, 1990)	Ranges from 0 (worst imaginable health state) to 100 (best imaginable health state)	Supported (Moons, Van Deyk, et al., 2006)	Supported (Moons, Van Deyk, et al., 2006)	Yes, in adults with CHD (Moons et al., 2018)	Higher scores = better perceived health
	12-item Short-Form Health Survey version 2 (SF−12v2) (Ware et al., 2009)	Ranges from 0 to 100 on 8 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	Supported (Ware, Kosinski, & Keller, 1996)	Supported (Ware, et al., 1996)	Yes, in adults with CHD (Moons et al., 2018)	Higher scores =better perceived health
Quality of life (QOL)	Linear Analog Scale (LAS) (Moons, Budts, & De Geest, 2006)	Vertically oriented, 10-centimeter line graded, ranges from 0 (worst imaginable QOL) to 100 (best imaginable QOL)	Supported (Moons, Van Deyk, et al., 2006)	Supported (Moons, Van Deyk, et al., 2006)	Yes, in patients with CHD (Moons, Van Deyk, et al., 2006)	Higher scores =better QOL
	Satisfaction with Life Scale (SWLS) (Diener, Emmons, Larsen, & Griffin, 1985)	- Comprises of 5 statements with a response ranging from strongly disagree to strongly agree - Total score ranges from 5 (extremely dissatisfied) to 35 (extremely satisfied)	Supported (Moons, Van Deyk, et al., 2006) Cronbach α value of present study: 0.89	Supported (Moons, Van Deyk, et al., 2006)	Yes, in patients with CHD (Moons, Van Deyk, et al., 2006)	- Score of 20 = neutral point - Higher scores =better QOL
Psycho-logical distress	Hospital Anxiety and Depression Scale (HADS) (HADS; (Spinhoven et al., 1997; Zigmond & Snaith, 1983))	- 14 items - 4-point Likert scale with values ranging from 0 to 3 - Total score ranges from 0 to 42	Supported (Zigmond & Snaith, 1983) Cronbach α value of present study: 0.86	Supported (Bjelland, Dahl, Haug, & Neckelmann, 2002)	Yes, in adults with CHD (Moons et al., 2018)	Higher scores =more psychological distress

(Continues)

TABLE 1 (Continued)

Variable	Instrument	Structure of the instrument	Reliability	Validity	Use in cardiac samples	Interpretation
Healthcare use	Patient-Reported Inpatient and outpatient Utilization Survey (PRIUS) (Van Bulck et al., 2018)	- 3 items, recall time frame of 6 months, healthcare use that is related to the heart disease - Only variables that have shown to be significantly related to illness identity in a previous study on the same dataset were included. (Van Bulck et al., 2018)	NA	NA	Yes, in adults with CHD (Van Bulck et al., 2018)	Higher number =more healthcare use

Abbreviations: NA, not applicable.

References:

Bjelland, I., Dahl, A. A., Haug, T. T., & Neckelmann, D. (2002). The validity of the Hospital Anxiety and Depression Scale. An updated literature review. *J Psychosom Res*, *52*(2), 69–77.

Diener, E., Emmons, R. A., Larsen, R. J., & Griffin, S. (1985). The Satisfaction with Life Scale. *J Pers Assess*, *49*(1), 71–75.

EuroQol Group. (1990). EuroQol--a new facility for the measurement of health-related quality of life. *Health Policy*, *16*(3), 199–208.

Moons, P., Budts, W., & De Geest, S. (2006). Critique on the conceptualisation of quality of life: a review and evaluation of different conceptual approaches. *Int J Nurs Stud*, *43*(7), 891–901. 10.1016/j.ijnurstu.2006.03.015

Moons, P., Kovacs, A. H., Luyckx, K., Thomet, C., Budts, W., Enomoto, J.,... the International Society for Adult Congenital Heart, D. (2018). Patient-reported outcomes in adults with congenital heart disease: Inter-country variation, standard of living and healthcare system factors. *Int J Cardiol*, *251*, 34–41. 10.1016/j.ijcard.2017.10.064

Moons, P., Van Deyk, K., De Bleser, L., Marquet, K., Raes, E., De Geest, S., & Budts, W. (2006). Quality of life and health status in adults with congenital heart disease: a direct comparison with healthy counterparts. *Eur J Cardiovasc Prev Rehabil*, *13*(3), 407–413.

Oris, L., Luyckx, K., Rassart, J., Goubert, L., Goossens, E., Apers, S.,... Moons, P. (2018). Illness Identity in Adults with a Chronic Illness. *J Clin Psychol Med Settings*, *25*(4), 429–440. 10.1007/s10880-018–9552–0

Oris, L., Rassart, J., Prikken, S., Verschueren, M., Goubert, L., Moons, P.,... Luyckx, K. (2016). Illness Identity in Adolescents and Emerging Adults With Type 1 Diabetes: Introducing the Illness Identity Questionnaire. *Diabetes Care*, *39*(5), 757–763. 10.2337/dc15-2559

Spinhoven, P., Ormel, J., Sloekers, P. P., Kempen, G. I., Speckens, A. E., & Van Hemert, A. M. (1997). A validation study of the Hospital Anxiety and Depression Scale (HADS) in different groups of Dutch subjects. *Psychol Med*, *27*(2), 363–370. 10.1017/S0033291796004382

Van Bulck, L., Goossens, E., Luyckx, K., Oris, L., Apers, S., & Moons, P. (2018). Illness Identity: A Novel Predictor for Healthcare Use in Adults With Congenital Heart Disease. *J Am Heart Assoc*, *7*(11), e008723. 10.1161/jaha.118.008723

Ware, J., Jr., Kosinski, M., & Keller, S. D. (1996). A 12-Item Short-Form Health Survey: construction of scales and preliminary tests of reliability and validity. *Med Care*, *34*(3), 220–233. 10.1097/00005650-199603000–00003

Ware, J. E., Kosinski, M., Turner-Bowker, D. M., Sundaram, M., Gandek, B., & Maruish, M. E. (2009). *User's Manual for the SF-12v2 Health Survey Second Edition*: QualityMetric, Incorporated.

Zigmond, A. S., & Snaith, R. P. (1983). The hospital anxiety and depression scale. *Acta Psychiatr Scand*, *67*(6), 361–370. 10.1111/j.1600–0447.1983.tb09716.x

4.2 | Descriptive statistics of included variables

The mean, standard deviation, median and range of all included variables are reported in Table 3.

4.3 | Objective 1: Illness identity trajectories

Using LCGA, classes of illness identity dimensions were identified. A graphical presentation of the observed means of the classes for the illness identity dimensions on the three time points is provided in Figure 1a. A detailed description of how the most appropriate models have been selected can be found in Annex S2 and Table S1.

For engulfment, the model with three classes was selected (BIC = 1,374.6; *E* = 0.891; LMR-LRT *p* = .03). Class 1 (*n* = 28; 10.1%, mean intercept =3.614) was characterized by persistent high scores on engulfment (*Chronically high*). Class 2 (*n* = 74; 26.8%, mean intercept = 2.409) consisted of patients having moderate engulfment scores at the three time points (*Intermediate stable*). Class 3 represented the majority of the sample (*n* = 174; 63.0%, mean intercept = 1.309) and demonstrated low engulfment scores over time (*Consistently low*).

TABLE 2 Patient characteristics of the total sample at T_{2013} (n = 276)

Characteristics	n (%)
Median age at T_{2013}	34y (Q1= 28y; Q3 = 42y)[a]
Men	149 (54.0)
Highest level of education	
Less than high school	11 (4.0)
High school	131 (47.8)
College degree	80 (29.2)
University degree	52 (19.0)
Employment status	
Full-time paid job	174 (63.3)
Part-time paid job	47 (17.1)
Homemaker	8 (2.9)
Job seeking	5 (1.8)
Unemployed	2 (0.7)
Disability/government financial assistance	24 (8.7)
Retired	10 (3.6)
Other	5 (1.8)
Disease complexity, Task Force 1	
Simple	93 (33.7)
Moderate	150 (54.3)
Complex	33 (12.0)

[a]Interquartile range.

TABLE 3 Descriptive overview of the included variables

	T_{2013}	T_{2014}	T_{2015}
ILLNESS IDENTITY DIMENSIONS			
Rejection	2.6 (0.9)[a]	2.6 (0.9)[a]	2.6 (1.0)[a]
Acceptance	4.2 (0.8)[a]	4.2 (0.7)[a]	4.3 (0.7)[a]
Engulfment	1.8 (0.9)[a]	1.8 (0.8)[a]	1.8 (0.8)[a]
Enrichment	3.0 (1.2)[a]	3.1 (1.1)[a]	3.1 (1.1)[a]
PATIENT-REPORTED OUTCOMES			
Perceived health			
EuroQoL-Visual Analogue Scale			78.1 (13.8)[a]
Physical Component Summary			76.2 (19.6)[a]
Mental Component Summary			72.1 (16.5)[a]
Psychological distress			
Hospital Anxiety and Depression Scale			8.6 (5.8)[a]
Quality of life			
Linear Analogue Scale-Quality of Life			77.8 (13.5)[a]
Satisfaction With Life Scale			25.5 (5.6)[a]
HEALTHCARE USE			
Hospitalizations in past 6 months (reason related to the heart disease)			0 (3)[b] 8%[c]
Visits at general practitioner in past 6 months (reason related to the heart disease)			0 (24)[b] 28%[c]
Visits at medical specialist in past 6 months (reason related to the heart disease)			0 (10)[b] 50%[c]

[a]Mean and standard deviation are reported.

[b]Median and range are reported.

[c]Percentage of the sample with at least one hospitalization/visit at general practitioner/visit at medical specialist.

FIGURE 1 Trajectory classes of illness identity dimensions and their clinical relevance. Bolt results = significantly different; blurred results = not significantly different. (a) Graphical presentation of illness identity trajectories across three time points. Observed means are reported. (b) Trajectories in terms of age, sex and disease complexity. Medians and percentages are reported. = simple disease complexity; = moderate disease complexity; = great disease complexity. (c) Trajectories in terms of patient-reported outcomes. Means are reported. (d) Trajectories in terms of healthcare use (of the past 6 months; related to heart disease). Odds ratios and confidence intervals are reported (on a logarithmic scale). EQ-VAS = EuroQoL-Visual Analogue Scale; LAS-QOL = Linear Analogue Scale-Quality of Life; HADS = Hospital Anxiety and Depression Scale; PCS = Physical Component Summary; MCS = Mental Component Summary; SWLS = Satisfaction With Life Scale

Also, for rejection, three trajectory classes were identified (BIC =1,682.0; E = 0.803; LMR-LRT p =.0014). The first class (n = 22; 8.0%, mean intercept =4.294), which included a minority of patients, showed persistently high rejection scores (*Chronically high*). In Class 2 (n = 124; 44.9%, mean intercept =3.067), patients with moderate rejection scores that remained stable over time were included (*Intermediate stable*). Finally, the remaining patients (n = 130; 47.1%, mean intercept =1.928) had consistently low scores on the rejection subscale (*Consistently low*).

For acceptance, a two-class solution was selected (BIC = 1,489.1; E = 0.823; LMR-LRT p = .004). The first class was labelled "*Consistently high*" (n = 212; 76.8%; mean intercept = 4.484). These patients consistently accepted their disease as part of their identity. The second class (n = 64; 23.2%; mean intercept = 3.137; mean slope = 0.201) consisted of patients who demonstrated moderate acceptance levels at baseline and showed slightly better acceptance levels over time (*Intermediate increasing*).

For enrichment, the model with three classes was most favourable (BIC = 1,836.0; E = 0.839; LMR-LRT p < .00001). The "*Consistently high*" class (n = 102; 37.0%; mean intercept = 4.147) consisted of patients that felt highly enriched over the three time points. The second class (n = 126; 45.7%; mean intercept = 2.768) had enrichment scores that were moderate and remained stable over time (*Intermediate stable*). Finally, the remaining participants (n = 48; 17.4%; mean intercept = 1.448) followed a trajectory of persistently low enrichment (*Chronically low*).

4.4 | Objective 2a: Trajectories in terms of sociodemographic and clinical variables

Kruskal–Wallis H and chi-square tests were performed to describe the illness identity classes in terms of age, sex and disease complexity. The distribution of age, gender and disease complexity for the

different trajectory classes is shown in Figure 1b and in Table S2. Age seemed to be a predictor for the classes of engulfment (p = .013; η^2 = 0.046) and rejection (p = .019; η^2 = 0.048). More specifically, patients with *Consistently low* engulfment scores were significantly younger than the other patients. Patients with *Chronically high* rejection scores were older compared with the other rejection classes. Furthermore, the classes of engulfment (p = .031; φ_c = 0.139) and enrichment (p = .029; φ_c = 0.190) significantly differed in terms of disease complexity. The majority of patients with simple and moderate lesion complexity had *Consistently low* engulfment scores. Moreover, most complex patients were located in the class with *Consistently high* enrichment scores.

4.5 | Objective 2b: Trajectories in terms of PROs

Multivariate analyses of variance with pairwise comparisons using Gabriel's test procedure (i.e., post hoc tests) were performed to describe the illness identity classes in terms of PROs. Figure 1c shows the mean values of perceived health status, QOL and psychological distress, measured using the EuroQoL-Visual Analogue Scale (EQ-VAS), Linear Analogue Scale-QOL (LAS-QOL), Hospital Anxiety and Depression Scale (HADS), 12-Item Short Form Survey (SF-12; Physical Component Summary [PCS] and Mental Component Summary [MCS]) and Satisfaction With Life Scale (SWLS) for all trajectory classes. The *F*-values and η^2-values of these associations can be found in Table S3.

The three trajectory classes of engulfment were significantly different from each other for all PROs. Patients with *Chronically High* engulfment scores reported lower perceived health, QOL and more psychological distress compared with patients with *Intermediate Stable* and *Consistently Low* scores on engulfment. Patients with *Consistently Low* engulfment scores reported opposite results and, hence, showed more favourable PROs. The η^2-values show large effect sizes for all PROs, except MCS for which a medium effect size was found.

The classes of rejection significantly differed from each other in terms of QOL (LAS-QOL), psychological distress (HADS) and physical health (PCS). More specifically, patients who did not reject their disease over time (*Consistently Low*) reported better QOL and less psychological distress compared with patients with *Intermediate Stable* rejection scores and reported better physical health compared with patients who reject their disease over time (*Chronically High*). In terms of effect sizes, η^2-values show small effect sizes for all PROs.

Perceived health (EQ-VAS), QOL (LAS-QOL and SWLS) and psychological distress (HADS) were significantly different for the two trajectory classes of acceptance. Patients that accepted their disease as part of their identity over time (*Consistently High*) reported better perceived health, better QOL and less psychological distress, compared with patients with an *Intermediate Increasing* level of acceptance. The η^2-values show small effect sizes for all PROs.

Physical health (PCS) was different for the three trajectory classes of enrichment. This score was higher in patients with *Chronically Low* enrichment scores, compared with patients with *Consistently High* enrichment scores and patients with *Intermediate Stable* enrichment scores. Small effect sizes (η^2) were found for all PROs, except for QOL and SWLS for which negligible effect sizes were found.

4.6 | Objective 2c: Trajectories in terms of healthcare use

Based on multivariable Poisson and negative binomial regression analyses, odds ratios and confidence intervals of healthcare use of the different classes of the four illness identity dimensions are reported in Figure 1d and Table S4.

Patients with low engulfment scores over time (*Consistently Low*) were less likely to be hospitalized and to visit a general practitioner in the past 6 months for a reason related to their health disease, compared with participants who were overwhelmed by their CHD over time (*Chronically High*).

For rejection, patients who did not reject their disease as part of their identity (*Consistently Low*) reported more visits at the general practitioner because of the CHD, compared with patients who tend to refuse to experience their disease as part of their identity (*Chronically High*).

Compared with individuals who accepted their disease over time (*Consistently High*), participants with *Intermediate Increasing* acceptance scores were more likely to be hospitalized, to visit a general practitioner and to visit a medical specialist, for a reason related to their CHD.

Patients who are highly enriched by their disease (*Consistently High*) were more likely to visit a general practitioner for a reason related to the CHD, compared with patients who did not experience positive life changes due to their disease (*Chronically Low*).

5 | DISCUSSION

To the best of our knowledge, this is the first study that investigated illness identity in adults with CHD using a longitudinal research design. This study aimed to identify classes of individuals with CHD characterized by similar trajectories and to describe these classes in terms of sex, age, disease complexity, PROs and healthcare use.

Overall, illness identity has shown to be a stable concept in adults with CHD. Previous research showed that illness identity dimensions are likely to change over the different stages of the illness trajectory (Oris et al., 2016), but patients with CHD in the present study received their diagnosis at birth or at least 10 years before inclusion in the study, which could explain the stability of the concept in this sample.

The identified trajectory classes did not differ from each other in terms of sex, which is in line with preceding studies (Andonian

et al., 2020; Luyckx et al., 2018; Oris et al., 2016, 2018). In the present study, patients with *Consistently low* engulfment scores were younger than the other patients, and patients with *Chronically high* rejection scores were older compared with the other rejection classes. Previous literature showed conflicting results about the association between age and illness identity dimensions (Andonian et al., 2020; Luyckx et al., 2018; Oris et al., 2016, 2018). In our sample, disease complexity was found to be significantly associated with classes of engulfment and enrichment, with complex patients situated in higher levels of engulfment and enrichment over time. This could be because patients with complex heart defects are more prone to experiencing physical limitations and illness symptoms are positively associated with engulfment and enrichment (Kovacs et al., 2005; Oris et al., 2018). For enrichment, it is stated that individuals have to experience a substantial impact of the disease to be able to grow as a person, which could also explain why patients with complex heart disease tend to have higher enrichment scores (Helgeson et al., 2006). Moreover, for certain patients, enrichment could be a coping mechanism to seek something positive in previous negative experiences (Reeve & Lincoln, 2002).

The identified classes have shown to be clinically relevant in terms of PROs. The directions of the associations remain unknown. One can assume that these results highlight the negative effects of being overwhelmed by the disease or when rejecting the disease as part of the identity and the positive impact of accepting the disease on the health of patients. However, PROs can also impact the illness identity dimensions. That would explain why patients with low enrichment levels reported better physical health, as a certain severity and impact of the disease should be experienced, to be enriched by the disease (Helgeson et al., 2006). The findings of this longitudinal study are in line with prior cross-sectional research in patients with CHD that showed associations between illness identity and emotional distress (Andonian et al., 2020; Oris et al., 2018).

The classes were also meaningfully differentiated in terms of healthcare use. High engulfment scores or low acceptance scores seem to induce higher healthcare use, as engulfed patients or patients with intermediate acceptance scores may have a lower threshold to seek care (Van Bulck et al., 2018). Oppositely, it could also be that patients who regularly consume healthcare are more often confronted with their disease, which can induce engulfment or can oppose acceptance of the disease (Van Bulck et al., 2018). The finding that patients who reject their disease were less likely to visit a general practitioner is not surprising as these patients might more often cancel clinic visits or might not attribute certain complaints, for which they seek care, to their CHD. Moreover, when looking at the opposite direction of effects, if patients consume no healthcare related to their CHD, they are not often confronted with their disease and can easily neglect it. The positive association between visits at the general practitioner and enrichment scores could be explained because enriched patients experience more illness symptoms, which could lead to healthcare use (Oris et al., 2018).

5.1 | Clinical implications

Both Oris et al., (2018) and Andonian et al., (2020) have described illness identity as a possible helpful tool to understand how patients with CHD experience their disease and suggested to develop interventions to ameliorate clinical and patient-reported findings. The results of the present study provide additional insights that can contribute to the development of tailored interventions or to the routine measurement of illness identity in practice.

First, illness identity has been shown to be a relatively stable concept over time, showing that a one-time assessment of illness identity could already provide interesting information for clinical practice. Oppositely, frequent short-term measurements seem less useful. This could be good news in terms of feasibility of implementing such assessment in clinical practice. However, it could also be that no change over time was noticed, because the data were collected over a relatively short period of 3 years and all patients received their diagnosis at least 10 years before inclusion in the study. To examine the stability further, it would be interesting to look at a broader time frame and to include patients in different phases of the illness trajectory.

Second, when measuring illness identity in practice, the question will remain on who to target. Therefore, it is interesting to know that older patients are more prone to be overwhelmed by the disease or to reject the disease over time and that patients with more complex heart disease might have higher chances of being overwhelmed and enriched. If this is confirmed in future studies, older patients could be seen at risk for less favourable outcomes and should therefore receive more attention in the clinic. Moreover, patients with complex heart disease could also benefit from monitoring of the level of engulfment and enrichment.

Third, the associations between illness identity and PROs indicate that identifying illness identity trajectories could be a potential pathway for targeting patients with poor PROs. PROs could be enhanced when challenging maladaptive illness identity integration and inducing acceptance, for example, by referring patients to psychotherapy (Tilden et al., 2005) or by actively challenging the negative thoughts. This could be facilitated by administering the Illness Identity Questionnaire (IIQ) or by letting the patient draw a circle that symbolizes the identity (based on the Pictorial Representation of Illness and Self Measure - Drawing [PRISM-D] [Sándor et al., 2020]). These strategies to ameliorate illness identity integration can be part of tailored interventions to optimize PROs and reach a holistic approach towards care for patients with CHD.

Fourth, as the present study confirmed that illness identity is associated with healthcare use in adults with CHD, this could be taken into account when trying to understand why certain people consume more healthcare compared with others.

Finally, for all identity dimensions, a group of patients with less favourable scores over time could be identified, which shows that, although patients might have lived with their CHD for a long time, some patients still have trouble integrating their disease into their identity. These patients with a maladaptive illness identity require

additional support from healthcare practitioners, starting with the acknowledgement of the identity issues (Oris et al., 2018).

5.2 | Limitations and suggestions for future research

First, only three waves with a 1-year time interval were included, which represents a relatively short period of time. This could be one of the reasons why we found little change in the development of illness identity. Longitudinal research with a continuous follow-up would provide additional information on the development of illness identity over time. Second, data on illness identity, PROs and healthcare use were self-reported, which could have introduced single-reporter bias, recall bias or telescoping. However, self-report questionnaires are the most appropriate method to gather PROs, as patients' views should be captured, and to gather information about internal processes, such as illness identity. The recall time frame of 6 months for healthcare use is more valid than larger time frames (Bhandari & Wagner, 2006), and earlier research in chronic patients demonstrated that self-reported healthcare use correlated strongly with data from medical records (Severs et al., 2016). Third, due to the study design, significant differences among the classes cannot be interpreted in terms of causality. They provide a first insight into potentially relevant factors for clinical practice. To examine the predictors of illness identity or predictive value of illness identity over time, cross-lagged path analysis could be applied. Fourth, we could not distinguish between appropriate and inappropriate healthcare use. In the light of optimizing healthcare use in this population, it is important to take this into account in future studies. Fifth, only adults with CHD have been included in the study. It could be interesting for future studies to include children and adolescents with CHD, as these patients could have gone through different stages of the illness trajectory. Indeed, that way, the development of illness identity in patients with CHD can be understood even better. Sixth, there is limited generalizability, due to the single-center setting, exclusion of patients with severe cognitive/language difficulties and exclusion of patients who received their CHD diagnosis after the age of 10 years. It would be interesting to investigate illness identity in a multicentric international study and in other patient populations and thereby investigate to what extent the results can be generalized.

6 | CONCLUSION

This is the first study that examined the development of illness identity in adults with CHD, which has shown to be relatively stable over time. The identified classes were meaningfully differentiated in terms of age, disease complexity, PROs and healthcare use. These findings indicate that illness identity should be taken into account when trying to understand and optimize PROs and healthcare use of adults with CHD.

ACKNOWLEDGMENTS

The authors want to thank all participants of this study and all collaborators who made substantial contributions to the Belgian branch of APPROACH-IS.

CONFLICT OF INTEREST

The authors have no conflicts of interest to declare.

AUTHOR CONTRIBUTIONS

[(This will be published with your article. Please provide authors' initials as appropriate)].

Criteria	Author initials
Made substantial contributions to conception and design, or acquisition of data, or analysis and interpretation of data;	LVB, SA, PM, KL, EG
Involved in drafting the manuscript or revising it critically for important intellectual content;	LVB, SA, PM, KL, EG
Given final approval of the version to be published. Each author should have participated sufficiently in the work to take public responsibility for appropriate portions of the content;	LVB, SA, PM, KL, EG
Agreed to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.	LVB, SA, PM, KL, EG

CLINICAL TRAIL REGISTRATION NUMBER

ClinicalTrials.gov: NCT02150603.

PEER REVIEW

The peer review history for this article is available at https://publons.com/publon/10.1111/jan.14949.

ORCID

Liesbet Van Bulck https://orcid.org/0000-0001-8975-4455

TWITTER

Liesbet Van Bulck @BulckLiesbet
Eva Goossens @EvaGoosens_PhD
Philip Moons @MoonsPhilip

REFERENCES

Adams, S., Pill, R., & Jones, A. (1997). Medication, chronic illness and identity: the perspective of people with asthma. *Social Science & Medicine*, *45*(2), 189–201. https://doi.org/10.1016/S0277-9536(96)00333-4.

Andonian, C., Beckmann, J., Ewert, P., Freilinger, S., Kaemmerer, H., Oberhoffer-Fritz, R., Sack, M., & Neidenbach, R. (2020). Assessment of the psychological situation in adults with congenital heart disease. *Journal of Clinical Medicine*, *9*(3), 779. https://doi.org/10.3390/jcm9030779.

Apers, S., Kovacs, A. H., Luyckx, K., Alday, L., Berghammer, M., & Budts, W., …, International Society for Adult Congenital Heart, D (2015). Assessment of Patterns of Patient-Reported Outcomes in Adults with Congenital Heart disease - International Study (APPROACH-IS): Rationale, design, and methods. *International Journal of Cardiology*, *179*, 334–342. https://doi.org/10.1016/j.ijcard.2014.11.084.

Bhandari, A., & Wagner, T. (2006). Self-reported utilization of health care services: Improving measurement and accuracy. *Medical Care Research and Review*, *63*(2), 217–235. https://doi.org/10.1177/1077558705285298.

Charmaz, K. (1995). The body, identity, and self: Adapting to impairment. *Sociological Quarterly*, *36*(4), 657–680. https://doi.org/10.1111/j.1533-8525.1995.tb00459.x.

Dillman, D. A. (1983). Chapter 10 - Mail and other self-administered questionnaires. In P. H. Rossi J. D. Wright & A. B. Anderson (Eds.), *Handbook of Survey Research* (pp. 359–377). Academic Press. https://www.sciencedirect.com/science/article/pii/B9780125982269500161.

Engelfriet, P., Boersma, E., Oechslin, E., Tijssen, J., Gatzoulis, M. A., Thilén, U., Kaemmerer, H., Moons, P., Meijboom, F., Popelová, J., Laforest, V., Hirsch, R., Daliento, L., Thaulow, E., & Mulder, B. (2005). The spectrum of adult congenital heart disease in Europe: Morbidity and mortality in a 5 year follow-up period. The Euro Heart Survey on adult congenital heart disease. *European Heart Journal*, *26*(21), 2325–2333. https://doi.org/10.1093/eurheartj/ehi396.

Helgeson, V. S., Reynolds, K. A., & Tomich, P. L. (2006). A meta-analytic review of benefit finding and growth. *Journal of Consulting and Clinical Psychology*, *74*(5), 797–816. https://doi.org/10.1037/0022-006X.74.5.797.

Kovacs, A. H., Sears, S. F., & Saidi, A. S. (2005). Biopsychosocial experiences of adults with congenital heart disease: Review of the literature. *American Heart Journal*, *150*(2), 193–201. https://doi.org/10.1016/j.ahj.2004.08.025.

Luyckx, K., Oris, L., Raymaekers, K., Rassart, J., Moons, P., Verdyck, L., Mijnster, T., & Mark, R. E. (2018). Illness identity in young adults with refractory epilepsy. *Epilepsy and Behavior*, *80*, 48–55. https://doi.org/10.1016/j.yebeh.2017.12.036.

Mandalenakis, Z., Rosengren, A., Skoglund, K., Lappas, G., Eriksson, P., & Dellborg, M. (2017). Survivorship in children and young adults with congenital heart disease in Sweden. *JAMA Internal Medicine*, *177*(2), 224–230. https://doi.org/10.1001/jamainternmed.2016.7765.

Meyer, S., & Lamash, L. (2021). Illness identity in adolescents with celiac disease. *Journal of Pediatric Gastroenterology and Nutrition*, *72*(2), e42–e47. https://doi.org/10.1097/MPG.0000000000002946.

Mitchell, S. C., Korones, S. B., & Berendes, H. W. (1971). Congenital heart disease in 56,109 births incidence and natural history. *Circulation*, *43*(3), 323–332. https://doi.org/10.1161/01.CIR.43.3.323.

Mommersteeg, P. M., Denollet, J., Spertus, J. A., & Pedersen, S. S. (2009). Health status as a risk factor in cardiovascular disease: A systematic review of current evidence. *American Heart Journal*, *157*(2), 208–218. https://doi.org/10.1016/j.ahj.2008.09.020.

Moons, P., Bovijn, L., Budts, W., Belmans, A., & Gewillig, M. (2010). Temporal trends in survival to adulthood among patients born with congenital heart disease from 1970 to 1992 in Belgium. *Circulation*, *122*(22), 2264–2272. https://doi.org/10.1161/CIRCULATIONAHA.110.946343.

Moons P., Kovacs A. H., Luyckx K., Thomet C., Budts W., Enomoto J., Sluman M. A., Yang H., Jackson J. L., Khairy P., Cook S. C., Subramanyan R., Alday L., Eriksen K., Dellborg M., Berghammer M., Johansson B., Mackie A. S., Menahem S., … International Society for Adult Congenital Heart Disease. (2018). Patient-reported outcomes in adults with congenital heart disease: Inter-country variation, standard of living and healthcare system factors. *International Journal of Cardiology*, *251*, 34–41. http://dx.doi.org/10.1016/j.ijcard.2017.10.064.

Morea, J. M., Friend, R., & Bennett, R. M. (2008). Conceptualizing and measuring illness self-concept: A comparison with self-esteem and optimism in predicting fibromyalgia adjustment. *Reseach in Nursing & Health*, *31*(6), 563–575. https://doi.org/10.1002/nur.20294.

Muthen, B., & Muthen, L. K. (2000). Integrating person-centered and variable-centered analyses: Growth mixture modeling with latent trajectory classes. *Alcoholism, Clinical and Experimental Research*, *24*(6), 882–891. https://doi.org/10.1111/j.1530-0277.2000.tb02070.x.

Oris, L., Luyckx, K., Rassart, J., Goubert, L., Goossens, E., Apers, S., Arat, S., Vandenberghe, J., Westhovens, R., & Moons, P. (2018). Illness identity in adults with a chronic illness. *Journal of Clinical Psychology in Medical Settings*, *25*(4), 429–440. https://doi.org/10.1007/s10880-018-9552-0.

Oris, L., Rassart, J., Prikken, S., Verschueren, M., Goubert, L., Moons, P., Berg, C. A., Weets, I., & Luyckx, K. (2016). Illness identity in adolescents and emerging adults with type 1 diabetes: introducing the illness identity questionnaire. *Diabetes Care*, *39*(5), 757–763. https://doi.org/10.2337/dc15-2559.

Raymaekers, K., Prikken, S., Vanhalst, J., Moons, P., Goossens, E., Oris, L., Weets, I., & Luyckx, K. (2020). The social context and illness identity in youth with type 1 diabetes: A three-wave longitudinal study. *Journal of Youth and Adolescence*, *49*(2), 449–466. https://doi.org/10.1007/s10964-019-01180-2.

Reeve, D. K., & Lincoln, N. B. (2002). Coping with the challenge of transition in older adolescents with epilepsy. *Seizure*, *11*(1), 33–39. https://doi.org/10.1053/seiz.2001.0574.

Sándor, Z., Látos, M., Pócza-Véger, P., Havancsák, R., & Csabai, M. (2020). The drawing version of the pictorial representation of illness and self measure. *Psychology and Health*, *35*, 1033–1048. https://doi.org/10.1080/08870446.2019.1707825.

Schoormans, D., Sprangers, M. A. G., van Melle, J. P., Pieper, P. G., van Dijk, A. P. J., Sieswerda, G. T., Hulsbergen-Zwarts, M. S., Plokker, T. H. W. M., Brunninkhuis, L. G. H., Vliegen, H. W., & Mulder, B. J. M. (2016). Clinical and psychological characteristics predict future healthcare use in adults with congenital heart disease. *European Journal of Cardiovascular Nursing*, *15*(1), 72–81. https://doi.org/10.1177/1474515114555819.

Senol-Durak, E. (2013). Stress related growth among diabetic outpatients: role of social support, self-esteem, and cognitive processing. *Social Indicators Research*, *118*(2), 729–739. https://doi.org/10.1007/s11205-013-0435-3.

Severs, M., Petersen, R. E., Siersema, P. D., Mangen, M. J., & Oldenburg, B. (2016). Self-reported health care utilization of patients with inflammatory bowel disease correlates perfectly with medical records. *Inflammatory Bowel Disease*, *22*(3), 688–693. https://doi.org/10.1097/MIB.0000000000000643.

Tilden, B., Charman, D., Sharples, J., & Fosbury, J. (2005). Identity and adherence in a diabetes patient: Transformations in psychotherapy. *Qualitative Health Research*, *15*(3), 312–324. https://doi.org/10.1177/1049732304272965.

Tzelepis, F., Sanson-Fisher, R. W., Zucca, A. C., & Fradgley, E. A. (2015). Measuring the quality of patient-centered care: Why patient-reported measures are critical to reliable assessment. *Patient Preference and Adherence*, *9*, 831–835. https://doi.org/10.2147/PPA.S81975.

Van Bulck, L., Goossens, E., Luyckx, K., Oris, L., Apers, S., & Moons, P. (2018). Illness identity: A novel predictor for healthcare use in adults with congenital heart disease. *Journal of the American Heart Association*, *7*(11), e008723. https://doi.org/10.1161/jaha.118.008723.

Van Bulck, L., Luyckx, K., Goossens, E., Oris, L., & Moons, P. (2019). Illness identity: Capturing the influence of illness on the person's sense of self. *Journal of Cardiovascular Nursing*, *18*(1), 4–6. https://doi.org/10.1177/1474515118811960.

Warnes, C. A., Liberthson, R., Danielson, G. K., Dore, A., Harris, L., Hoffman, J. I. E., Somerville, J., Williams, R. G., & Webb, G. D. (2001). Task force 1: The changing profile of congenital heart disease in adult life. *Journal of the American College of Cardiology*, *37*(5), 1170–1175. https://doi.org/10.1016/S0735-1097(01)01272-4.

Willems, R., Werbrouck, A., De Backer, J., & Annemans, L. (2019). Real-world healthcare utilization in adult congenital heart disease: A systematic review of trends and ratios. *Cardiology in the Young*, *29*(5), 553–563. https://doi.org/10.1017/s1047951119000441.

SUPPORTING INFORMATION

Additional supporting information may be found online in the Supporting Information section.

How to cite this article: Van Bulck, L., Goossens, E., Apers, S., Moons, P., & Luyckx, K. (2021). Illness identity in adults with congenital heart disease: Longitudinal trajectories and associations with patient-reported outcomes and healthcare use. *Journal of Advanced Nursing*, 00, 1–12. https://doi.org/10.1111/jan.14949