ORIGINAL RESEARCH

CONGENITAL HEART DISEASE

The Influence of Illness Perception and Coping on Anxiety in Adults With Congenital Heart Disease

Marie-Joëlle Marcil, BSc,[a,b] Cylia Houchi, BSc,[a,b] Kishani Nadarajah, MD,[a] Paul Khairy, MD, PhD,[a,c]
Geneviève A. Mageau, PhD,[d] Marie-France Marin, PhD,[e] Mariève Cossette, MSc,[f] Marie-Pierre Dubé, PhD,[a,c]
Marie-A. Chaix, MD, PhD,[a,c] François-Pierre Mongeon, MD, SM,[a,c] Annie Dore, MD,[a,c] Blandine Mondésert, MD,[a,c]
Reda Ibrahim, MD,[a,c] Judith Brouillette, MD, PhD[a,b]

ABSTRACT

BACKGROUND Up to one-half of adults with congenital heart disease (CHD) experience psychological distress, including anxiety.

OBJECTIVES This paper sought to: 1) assess the contribution of illness perception in explaining anxiety symptoms beyond sociodemographic and medical variables in adults with CHD; and 2) investigate the potential mediating effect of coping style.

METHODS CHD adult patients were recruited at Montreal Heart Institute between June 2019 and April 2021 for this cross-sectional study. Participants responded to self-reported questionnaires (Hospital Anxiety and Depression Scale, Brief Illness Perception Questionnaire, and Brief COPE). Medical characteristics (CHD complexity, NYHA functional class, and cardiac devices) were collected from medical records. We conducted hierarchical multiple linear regression and mediation analyses.

RESULTS Of the 223 participants (mean age 46 ± 14 years, 59% women), 15% had clinically significant anxiety symptoms. Medical and sociodemographic variables explained 15% of the variation in anxiety symptoms. Adding illness perception explained an additional 18% of the variation in anxiety. This R^2 change was significant ($F[1,188] = 49.06$, $P < 0.0001$). Illness perception explained more variance (18%) than medical and sociodemographic variables combined. A more threatening perception of illness was associated with greater anxiety symptoms ($\beta = 0.45$, $P < 0.0001$). Furthermore, illness perception was associated with coping, which was linked to reduced anxiety symptoms. Coping response style accounted for 20% of the total effect of illness perception on anxiety.

CONCLUSIONS Illness perception and coping are associated with anxiety in adults with CHD. Future initiatives should assess whether targeting these potentially modifiable factors effectively prevents or mitigates anxious symptoms in adults with CHD. (JACC Adv 2023;2:100425) © 2023 The Authors. Published by Elsevier on behalf of the American College of Cardiology Foundation. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/).

From the [a]Research Center, Montreal Heart Institute, Montréal, Québec, Canada; [b]Department of Psychiatry and Addiction, Faculty of Medicine, Université de Montréal, Montréal, Québec, Canada; [c]Department of Medicine, Faculty of Medicine, Université de Montréal, Montreal, Québec, Canada; [d]Department of Psychology, Faculty of Arts and Science, Université de Montréal, Montréal, Québec, Canada; [e]Department of Psychology, Faculty of Social Sciences and Humanities, Université du Québec à Montréal, Montréal, Québec, Canada; and the [f]Montreal Health Innovations Coordinating Centre, A Division of the Montreal Heart Institute, Montréal, Québec, Canada.

ABBREVIATIONS AND ACRONYMS

ACHD = adult congenital heart disease

B-IPQ = Brief Illness Perception Questionnaire

CHD = congenital heart disease

HADS-A = anxiety subscale of the Hospital Anxiety and Depression Scale

ICD = implantable cardioverter-defibrillator

MHI = Montreal Heart Institute

Medical and surgical advances have decreased childhood mortality and have improved the prognosis of individuals with congenital heart disease (CHD).[1] As a result, adults with CHD represent a growing population. Since the physical health of adults with CHD has markedly improved, attention has increasingly focused on psychological health and quality of life.[2] Between 3%[3] and 54%[4] of adults with CHD will suffer from an anxiety disorder at some point in their lives, which can negatively impact their quality of life.[5] Psychological distress, including anxiety, has been associated with lower quality of life and poorer cardiac prognosis.[2,6] Thus, efforts to reduce anxiety are a priority and could also impact the overall health of adults with CHD.

Studies on anxiety in adults with CHD have largely focused on associations with various medical and sociodemographic factors. However, there is little evidence that these clinical characteristics play a significant role. For example, among 25 medical characteristics, only the NYHA functional class, which categorizes patients' limitations according to their symptoms (eg, dyspnea, palpitations, anginal pain, and fatigue) during ordinary activity and at rest, was found to influence emotional functioning in adults with CHD.[7] Fewer studies have focused on psychological factors associated with anxiety in adults with CHD. There is increasing evidence that the subjective experience of illness, such as illness perception, is associated with anxiety in adults with CHD and other forms of heart disease.[8,9] Furthermore, medical and sociodemographic characteristics are rarely modifiable, providing limited guidance as to what can be targeted in the future to reduce anxiety. Therefore, identifying modifiable factors, such as illness perception and coping strategies, is important to guide future initiatives to reduce anxiety.

Illness perception refers to the cognitive and emotional representations each patient constructs about their illness. These beliefs are based on concrete and abstract sources of information (eg, experiences related to their illness, interpretation of medical knowledge related to the illness, reactions of others).[10,11] Coping refers to an individual's behavioral, cognitive, or emotional responses to minimize the distress caused by a perceived threatening situation.[12] Coping responses can be grouped into 3 categories: problem-focused, emotion-focused, and avoidant. Problem-focused coping responses involve directing efforts toward the stressful situation,

aiming to manage the problem causing the distress (eg, active coping).[12] Emotion-focused coping responses target the individual's emotional response rather than the stressful situation, aiming to manage the distress generated by the stressful situation.[12] For example, trying to find humor in stressful situations can aid individuals ease the emotional burden they feel, but it does not address the situation that caused these emotions. Avoidant coping responses refer to cognitive or behavioral efforts to disengage from the stressful situation to neutralize its associated distress (eg, denial, substances use).[12,13] According to the Common Sense model of illness, illness perception influences coping responses to the illness, which both contribute to psychological distress.[11,14] To our knowledge, no research has combined the study of illness perception and coping associated with anxiety in adults with CHD.

We aimed to: 1) quantify the contribution of illness perception in explaining variance in anxiety symptoms in adults with CHD beyond medical-social-demographic variables; and 2) investigate the potential mediating role of coping in the association between illness perception and anxiety.

METHODS

STUDY DESIGN AND SAMPLE. Potentially eligible candidates were recruited from the Montreal Heart Institute (MHI) Adult Congenital Heart Disease (ACHD) Center (Québec, Canada) between June 2019 and April 2021. Participants were required to be 18 years old or older, diagnosed with CHD (see Supplemental Table 1 for CHD diagnosis and complexity of disease), and followed at the MHI ACHD Center. Participants who received a heart transplant and those with a language barrier or a medical condition that prevented them from accurately completing questionnaires were excluded. Patients had the options to complete the questionnaires in the waiting room or at home, on paper or online via a secure electronic platform, and in French or in English, according to their preferences. For the latter option, participants were assigned a unique identification number and a password to log into the platform. Participants had a deadline of 1 month to submit their responses. This study was approved by MHI Ethics and Research Committees. All patients provided written informed consent to participate in the study.

MEASURES. Sociodemographic and medical variables. Two team members independently collected medical information from participants' medical records, including the CHD diagnosis to

determine disease complexity, NYHA functional class, presence of a permanent pacemaker or implantable cardioverter-defibrillator (ICD), presence of chronic diseases other than CHD (defined as any condition that persists over several years and requires long-term medical management, eg, hypertension, cancer), age and sex. For disease complexity, we classified the CHD diagnosis as simple, moderate, or high complexity, according to the American Heart Association's classification.[15] Participants with multiple types of congenital heart defects were classified according to the diagnosis with the greatest complexity. To minimize errors in medical data collection, comparisons were performed between 2 independent data sets to identify discrepancies. When discordant data were found, medical records were consulted for verification. To assess economic status, participants were asked to report their annual family income.

Anxiety. Anxiety was assessed by the anxiety subscale of the Hospital Anxiety and Depression Scale (HADS-A).[16] This questionnaire evaluates anxiety symptoms over the past 7 days. No somatic symptoms are assessed to prevent confounding anxiety-related somatic symptoms with those of illness (eg, fatigue). The HADS-A consists of 7 items evaluated on a 4-point scale (0-3). The total score ranges from 0 to 21. Symptoms can be classified as nonclinical (score ≤ 7), probable (score 8-10), or clinically symptomatic (score ≥ 11).[16]

Illness perception. The Brief Illness Perception Questionnaire (B-IPQ)[17] was used to assess participants' perception of their illness. The questionnaire contains 8 items rated between 0 and 10, which assess cognitive and emotional representations of the disease (see Supplemental Table 2 for details). A ninth item asks participants to list 3 causes they believe are responsible for their illness, which was not considered in the present study. Scores range from 0 to 80, with a higher score reflecting a greater perception of threat from the illness.

Coping. Coping was assessed using the Brief COPE questionnaire,[18] which covers 14 coping responses. The Brief COPE has 28 items rated on a 4-point response scale. The 14 coping responses were classified into 3 types: problem-focused, emotion-focused, and avoidant (see Supplemental Table 3 for details). The score indicates the extent to which respondents usually use each coping style.

STATISTICAL ANALYSES. Sample size calculation. The sample size was calculated to detect a correlation of 0.2 between anxiety symptoms and illness perception using a 2-sided significance level of 0.05 and 80%

TABLE 1 Sociodemographic and Medical Characteristics of Participants (N = 223)	
Age (y)	46 ± 14
Women	131 (59)
Annual family income[a] ($ CAN)	
<$15,000	15 (7)
$15,000-$24,999	17 (8)
$25,000-$34,999	15 (7)
$35,000-$54,999	34 (16)
$55,000-$74,999	32 (15)
$75,000-$99,999	28 (14)
$\geq$1,00,000	66 (32)
CHD complexity	
Mild	42 (19)
Moderate	134 (60)
Complex	47 (21)
NYHA functional class	
I	153 (69)
II	53 (24)
III	17 (8)
IV	0 (0)
Pacemaker or ICD	52 (23)
Chronic disease	159 (71)

Values are mean $\pm$ SD or n (%). [a]n = 207.

CHD = congenital heart disease; ICD = implantable cardioverter-defibrillator.

power. This estimate is conservative, considering that a previous study reported that generalized anxiety and illness perception are highly correlated ($r = 0.50$) among cardiac patients.[19] Taking into account the presence of adjustment variables in the model, an inflation parameter linked to the correlation between the adjustment variables and illness perception was included in the calculation (the greater the correlation, the larger the sample size). Based on previous studies and substantive knowledge, it was estimated that the multiple correlation coefficient between illness perception and adjustment variables should not exceed 0.20, such that an inflation parameter of 1/(1-0.20) = 1.25 was used. In this context, a sample size of 240 participants was required to detect a correlation of 0.2 between anxiety symptoms and illness perception.

Missing data. Only participants with complete data for each of the variables studied were considered in multiple linear regression analyses. A prorated score was calculated for participants with 25% or less of the items missing from the B-IPQ or any of the 3 types of coping from the Brief-COPE. Considering that HADS-A scores cannot be prorated, all 7 items required answers. The final population for analysis included 223

TABLE 2 Descriptive Statistics of Participants' Scores on Questionnaires About Anxiety Symptoms, Illness Perception, and Coping Response Styles

Questionnaires	Theoretical Range	n	Mean ± SD	Median	Q1-Q3
Anxiety symptoms (HADS-A)	0-21	223	6.3 ± 3.8	6	4-8
Illness perception (B-IPQ)	0-80	215	36.6 ± 12.1	36	26-46
Brief COPE					
Problem-focused	8-32	221	21.1 ± 4.1	21	18-24
Emotion-focused	12-48	221	26.4 ± 4.3	26	24-29
Avoidant	8-32	222	13.2 ± 3.1	13	11-15

B-IPQ = Brief Illness Perception Questionnaire; HADS-A = anxiety subscale of the Hospital Anxiety and Depression Scale.

participants whose anxiety scores could be calculated (no missing items on HADS-A).

Analyses. To assess: 1) whether respondents' characteristics differed significantly from non-respondents'; and 2) whether respondent characteristics and scores differed significantly between participants recruited before the onset of the COVID-19 pandemic and those recruited after Mann-Whitney tests (for continuous or ordinal variables) and chi-square tests (for categorical variables) were used.

A 3-step hierarchical multiple linear regression analysis was performed using the HADS-A score as the dependent variable. In the first step, adjustment variables were entered into the model, which were variables generally recognized as being associated with anxiety: 1) age; 2) sex; 3) annual family income; and 4) presence of chronic illness. In the second step, medical variables were introduced: 1) CHD complexity; 2) NYHA functional class; and 3) presence of a permanent pacemaker or ICD. In the third step, the B-IPQ score was introduced. Changes in the coefficient of determination (R^2) were assessed after introducing a new cluster of variables to the model. Extra sum-of-squares F-tests were performed to determine if changes in R^2 were statistically significant.

A multiple mediator model[20] was conducted assuming that the B-IPQ score is modeled as influencing the HADS-A score directly, as well as indirectly through using multiple linear regressions with 3 coping types as mediator variables: 1) problem-focused; 2) emotion-focused; and 3) avoidant. The analysis was adjusted for sociodemographic variables associated with anxiety, based on the results of the hierarchical regression model. The 95% CIs were estimated using the percentile bootstrap method. Lower and upper limits of the CI for indirect effects were defined as the values corresponding to the 2.5th

and 97.5th percentiles of the distribution of 5,000 bootstrap estimates of indirect effects.

Statistical tests were 2-sided at a significance level of 0.05. All statistical analyses were performed using SAS version 9.4 (2022, SAS Institute Inc).[21]
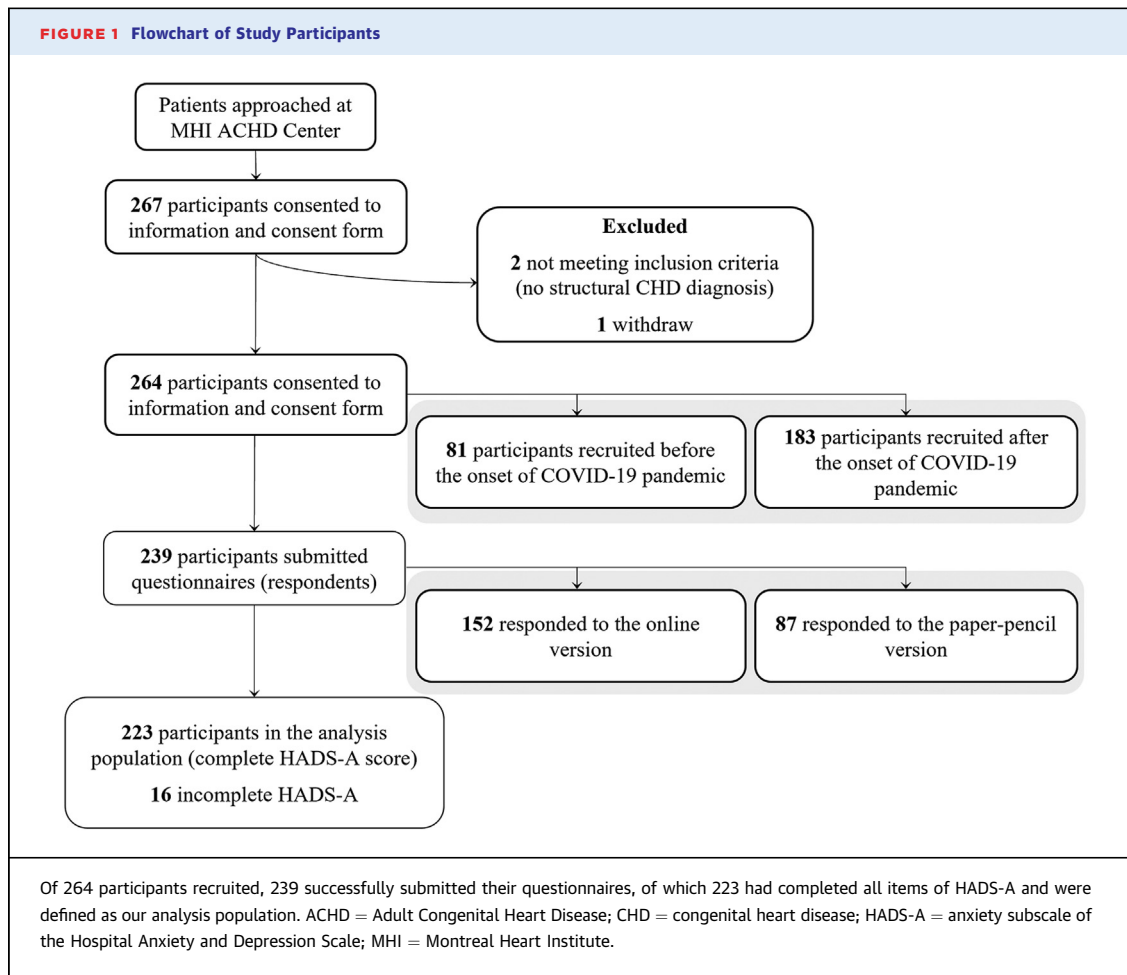
RESULTS

PARTICIPANTS' CHARACTERISTICS. Descriptive statistics for all variables of interest are presented in **Tables 1 and 2**. Among 267 recruited patients, 1 withdrew, and 2 were excluded due to the non-congenital nature of their heart disease (see participants flowchart, **Figure 1**). Of the remaining 264 patients, 239 (91%) responded to the questionnaires. Sixteen (7%) participants had incomplete HADS-A data such that analyses were performed on the remaining 223 participants (age 46 ± 14 years, 59% women) (**Table 1**). The type of CHD was mild, moderate, and complex in proportions of 19%, 60%, and 21%, respectively. Most participants were asymptomatic during ordinary physical activities and at rest (69%, NYHA functional class I) or had mild symptoms during physical activities (24%, NYHA functional class II). There was no significant difference in medical or sociodemographic characteristics between respondents and non-respondents (**Table 3**). Participants recruited before and after the onset of the COVID-19 pandemic reported similar anxiety symptoms (**Table 3**) (HADS-A score pre- and per-pandemic: 6.0 ± 3.8 vs 6.5 ± 3.8, $P = 0.272$, Hedges's g = 0.13). However, they differed in their NYHA functional class (chi-square: [2, n = 223] = 7.3, $P = 0.026$) and ICD permanent pacemaker status (chi-square: [1, n = 223] = 4.2, $P = 0.040$).

Nearly, 15% of participants had clinically significant anxiety symptoms (HADS-A ≥11), 17% had subclinical symptoms (HADS-A = 8-10), and 68% had anxiety symptoms considered within the normal spectrum (HADS-A ≤7). **Table 2** presents descriptive statistics for the psychological questionnaire scores.

CONTRIBUTION OF ILLNESS PERCEPTION TO ANXIETY SYMPTOMS. The results of hierarchical multiple regression are presented in **Table 4**. The first model, which includes adjustment variables, explained 9% of the variance in the anxiety score. Introducing the additional medical variables explained a further significant 6% of the variance in anxiety symptoms ($\Delta R^2 = 0.06$, $F(6,197) = 2.20$, $P = 0.045$).

Introducing illness perception contributed significantly to the model, explaining an additional 18% variance ($\Delta R^2 = 0.18$, $F(1,188) = 49.06$, $P < 0.0001$). The third model accounted for 33% of the variance in the HADS-A score.

FIGURE 1 Flowchart of Study Participants

Of 264 participants recruited, 239 successfully submitted their questionnaires, of which 223 had completed all items of HADS-A and were defined as our analysis population. ACHD = Adult Congenital Heart Disease; CHD = congenital heart disease; HADS-A = anxiety subscale of the Hospital Anxiety and Depression Scale; MHI = Montreal Heart Institute.

The final model indicated that age, family income, and illness perception were significantly associated with anxiety symptoms. Younger respondents, lower family income, and those perceiving their illness as more threatening tended to report more anxiety symptoms. For each SD increase in the B-IPQ score (more threatening disease perception), the anxiety score generally increased by 0.45 SD, when other adjustment and medical variables were held constant ($\beta = 0.45$, $P < 0.0001$). For each SD increase in age, the HADS-A score generally decreased by 0.23 SD, when other variables in the model were held constant ($\beta = -0.23$, $P = 0.0009$). For every 1 SD increase in annual family income, the HADS-A score decreases significantly by an average of 0.12 SD ($\beta = -0.12$, $P = 0.048$). No medical variables were significantly associated with anxiety in the final model. The NYHA functional class variable was significantly associated with anxiety in the second model ($P = 0.020$) but not in the final model ($P = 0.054$).

MEDIATING ROLE OF COPING. Figure 2 illustrates the results of the mediation analysis. The mediation model was adjusted for age and family income, which were significantly associated with anxiety in the first regression model. Mediation analyses showed that illness perception had a significant direct positive relation ($\beta = 0.37$, 95% CI: 0.261-0.486]) with anxiety symptoms, and a significant indirect positive path through coping ($\beta = 0.09$, 95% CI: 0.027-0.159]), when controlling for age and family income. The indirect path via coping accounted for nearly 20% of the total effect of illness perception on anxiety symptoms ($\beta = 0.46$). More specifically, problem-focused coping was the only coping category with a significant indirect path ($\beta = 0.05$, 95% CI: 0.014-0.102]). Avoidant coping was significantly associated with illness perception ($\beta = 0.14$, 95% CI: 0.0002-0.275]) and anxiety ($\beta = 0.31$, 95% CI: 0.191-0.435]), although no significant indirect path was observed ($\beta = 0.04$, 95% CI: −0.002 to 0.100]).

TABLE 3 Comparison of Sociodemographic, Medical, and Psychological Characteristics of Respondents and Nonrespondents, and Respondents Recruited Before and After the Onset of the COVID-19 Pandemic

	Respondents (n = 232)	Nonrespondents (n = 30)	P Value	Recruited Before COVID-19 (n = 61)	Recruited After COVID-19 (n = 162)	P Value
Age (y)	46 ± 14	47 ± 13	0.647	49 ± 16	45 ± 13	0.075
Women	137 (59)	13 (43)	0.102	33 (54)	98 (60)	0.387
CHD complexity			0.553			0.072
Mild	44 (19)	7 (23)		11 (18)	31 (19)	
Moderate	140 (60)	15 (50)		31 (51)	103 (64)	
Complex	48 (21)	8 (27)		19 (31)	28 (17)	
NYHA functional class			0.670			0.026
I	158 (68)	21 (70)		34 (56)	119 (73)	
II	56 (24)	8 (27)		19 (31)	34 (21)	
III	18 (8)	1 (3)		8 (13)	9 (6)	
IV	0 (0)	0 (0)		0 (0)	0 (0)	
PPM or ICD	55 (24)	10 (33)	0.251	20 (33)	32 (20)	0.040
Chronic disease	166 (72)	22 (73)	0.838	44 (72)	115 (71)	0.866
Annual family income ($ CAN)				(n = 59)	(n = 148)	0.129
<$15,000				7 (12)	8 (5)	
$15,000-$24,999				6 (10)	11 (7)	
$25,000-$34,999				2 (3)	13 (9)	
$35,000-$54,999				10 (17)	24 (16)	
$55,000-$74,999				12 (20)	20 (14)	
$75,000-$99,999				8 (14)	20 (14)	
≥$1,00,000				14 (24)	52 (35)	
Anxiety symptoms (HADS-A)				6.0 ± 3.8	6.5 ± 3.8	0.272
Illness perception (B-IPQ)				37.4 ± 12.1	36.3 ± 12.1	0.348

Values are mean ± SD or n (%).

B-IPQ = Brief Illness Perception Questionnaire; CHD = congenital heart disease; HADS-A = anxiety subscale of the Hospital Anxiety and Depression Scale; ICD = implantable cardioverter-defibrillator; PMP = permanent pacemaker.

DISCUSSION

Main findings of this detailed study on anxiety in adults with CHD include the following: about one-third of participants had subclinical or clinical anxiety symptoms; illness perception is the most important explanatory factor that is significantly associated with anxiety symptoms above and beyond medical and sociodemographic variables; and the indirect link from illness perception to anxiety via coping, particularly problem-focused coping, was significant, suggesting partial mediation (**Central Illustration**).

We observed that nearly 32% of participants reported mild, moderate, or severe anxiety symptoms (HADS-A ≥8). This result is similar or lower than the prevalence of anxiety symptoms found in 2 recent North American studies (35%[22] and 42%[23]). Nevertheless, the average severity of anxiety symptoms we observed (6 ± 4) is similar to what these 2 studies (6 ± 4[22], 7 ± 4[23]) reported, as well as an international study (6 ± 4[24]).

Our findings are consistent with other studies on adults with CHD and heart failure, which reported that patients' characteristics explained 3%[8] to 11%[25]

of anxiety symptoms. Our study also provides further insight into the individual contribution of medical and sociodemographic characteristics in explaining anxiety variance among CHD patients. We found that medical characteristics contributed significantly to explain variance in anxiety, above sociodemographic characteristics. However, we found that illness perception accounted for a greater proportion of explained variance in anxiety than medical characteristics. In the present study, illness perception explained 18% of anxiety symptoms and was a stronger predictor of anxiety symptoms than the patient's characteristics. Past studies showed that illness perception in adults with CHD explained 22% of anxiety symptoms[8] and 34% in patients with heart failure.[9] Likewise, both cited studies found that illness perception explained more variance than patients' characteristics. Our findings support the importance of considering illness perception in relation to anxiety in adults with CHD.

Other studies have reported how subjective experience of health and illness is associated with psychological distress and quality of life in adults with

TABLE 4 Hierarchical Multiple Linear Regression Analysis for Anxiety Symptoms Reported by Adults With Congenital Heart Disease

|	Model 1 (n = 207)				Model 2 (n = 207)				Model 3 (n = 199)			
	B	β	95% CI	P Value	B	β	95% CI	P Value	B	β	95% CI	P Value
Adjustment variables												
Sex (women vs men)	0.54	0.14	−0.13 to 0.41	0.293	0.55	0.15	−0.13 to 0.42	0.290	0.28	0.07	−0.17 to 0.32	0.555
Age	−0.07	−0.24	−0.37 to 0.11	0.0004	−0.08	−0.29	−0.44 to 0.14	0.0002	−0.06	−0.23	−0.37 to 0.10	0.0009
Family income	−0.34	−0.17	−0.31 to 0.04	0.011	−0.30	−0.16	−0.29 to 0.02	0.021	−0.24	−0.12	−0.25 to 0.001	0.048
Medical variables												
Complexity of CHD								0.605				0.720
Moderate vs mild					−0.57	−0.15	−0.50 to 0.19	0.388	−0.50	−0.13	−0.45 to 0.19	0.418
Severe vs mild					−0.10	−0.03	−0.48 to 0.42	0.903	−0.41	−0.11	−0.53 to 0.31	0.610
NYHA functional class								0.020				0.054
II vs I					0.04	0.01	−0.32 to 0.35	0.950	−0.77	−0.20	−0.51 to 0.10	0.194
III vs I					2.70	0.72	0.20 to 1.23	0.006	1.52	0.40	−0.08 to 0.88	0.105
Pacemaker or ICD (yes vs no)					0.29	0.08	−0.25 to 0.41	0.645	−0.12	−0.03	−0.34 to 0.27	0.836
Chronic illness (yes vs no)					0.68	0.18	−0.14 to 0.50	0.267	0.42	0.11	−0.18 to 0.40	0.455
Subjective medical variable												
Illness perception (B-IPQ)									0.14	0.45	0.32-0.58	<0.0001
R^2		0.09				0.15				0.33		
ΔR^2						0.06				0.18		
Extra sum of squares F						2.20				49.06		
P value						0.045				<0.0001		

Unstandardized (B) and standardized (β) regression coefficients are presented.
B-IPQ = Brief Illness Perception Questionnaire; CHD = congenital heart disease; ICD = implantable cardioverter-defibrillator.

CHD. Specifically, these studies observed that perceived health correlates strongly with anxiety and depression,[25,26] and predicts depressive symptoms, while medical characteristics are not predictive.[27] While cardiac diagnosis itself was not directly asso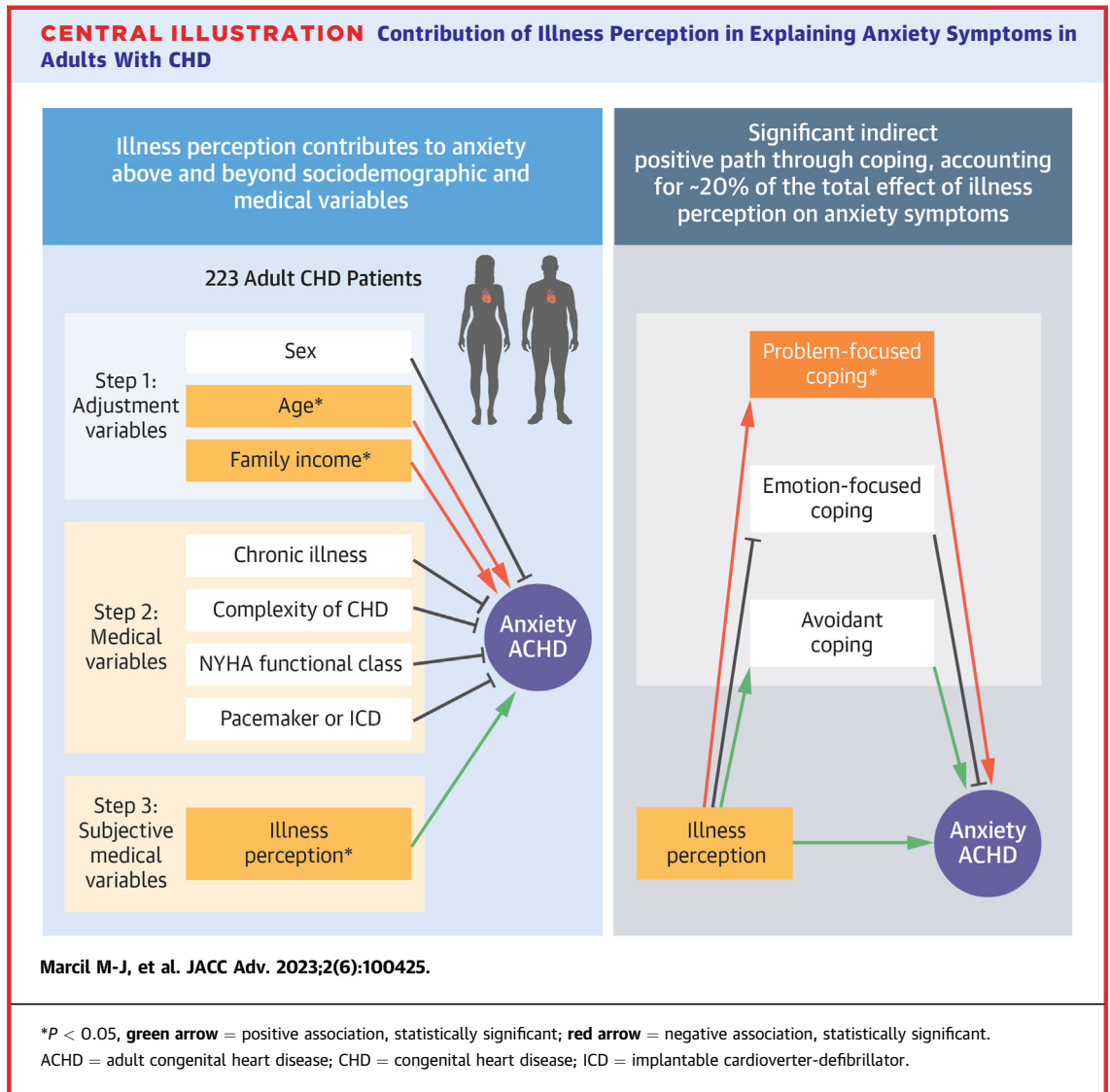ciated with quality of life, it was associated with illness perception, which in turn was associated with quality of life.[28]

We observed a significant positive association between increasing NYHA functional class and anxiety in the second model. Similarly, studies reported that a higher functional class was associated with a

FIGURE 2 Mediation Analysis Between Illness Perception, Coping Strategies, and Anxiety
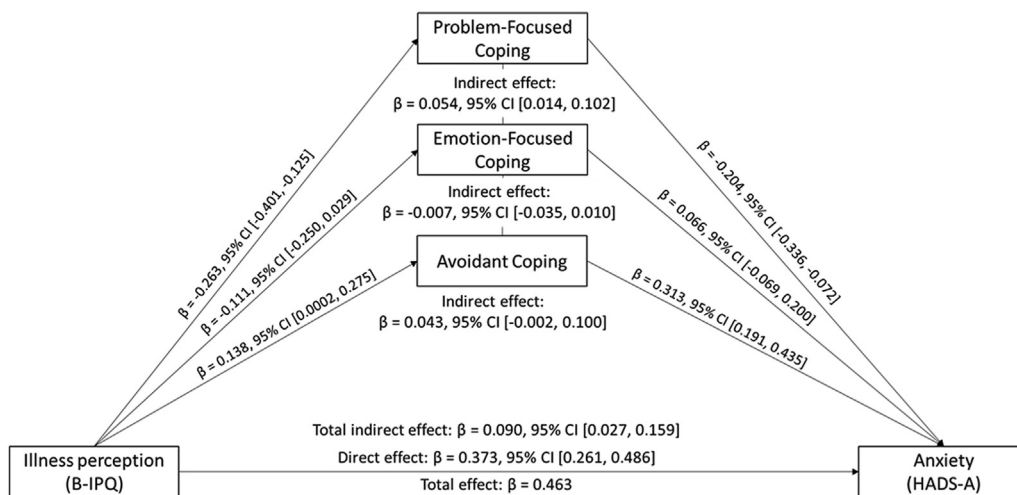
Multiple mediation model showing the direct path from illness perception (independent variable) to anxiety symptoms (dependent variable) and indirect path through problem-focused, emotion-focused and avoidant coping (mediators), adjusted for age and family income (n = 198). Unstandardized coefficients and 95% CIs are reported. B-IPQ = Brief Illness Perception Questionnaire; HADS-A = anxiety subscale of the Hospital Anxiety and Depression Scale.

CENTRAL ILLUSTRATION Contribution of Illness Perception in Explaining Anxiety Symptoms in Adults With CHD

Marcil M-J, et al. JACC Adv. 2023;2(6):100425.

*$P < 0.05$, **green arrow** = positive association, statistically significant; **red arrow** = negative association, statistically significant.
ACHD = adult congenital heart disease; CHD = congenital heart disease; ICD = implantable cardioverter-defibrillator.

significant increase in symptoms of psychological distress, including anxiety symptoms and lower quality of life.[7,8,25,29] Interestingly, the NYHA functional class has a subjective component as it is derived from the patient's perception of limitations during daily activities.[30] Nevertheless, NYHA functional class has been documented to correlate with objective measures of cardiopulmonary capacity in adults with CHD.[31] Norozi et al[29] found that NYHA functional class was associated with anxiety in adults with CHD but not VO$_2$ max, an objective measure of the cardiopulmonary capacity. After introducing illness perception into the final model, the NYHA functional class was no longer significantly associated with anxiety. This suggests that illness perception is more strongly associated with anxiety than NYHA classification.

Our findings support the Common-Sense theoretical framework, which suggests an association between illness perception, coping responses, and psychological distress, such as anxiety.[11,14,32] We found that illness perception was indirectly related to anxiety through coping. Adults with CHD who perceive their illness as more threatening experienced more anxiety. Furthermore, a more threatening perception of their illness was associated with less positive coping, which was also linked to anxiety levels.

We found that a more threatening perception of illness was associated with less use of problem-focused coping and more avoidant coping, which were associated with higher anxiety symptoms. When decomposing the total indirect effect, we observed that only the indirect effect of problem-

focused coping was statistically significant. A meta-analysis[32] documented the unique effects of the dimensions of illness perception and coping on psychological distress. The authors reported that problem-focused coping was associated with less psychological distress and that the avoidant coping style was associated with more distress. Our results indicate a similar relation between these 2 coping styles and anxiety. Unlike the meta-analysis, however, we did not observe a significant association between emotion-focused coping and anxiety. These inconsistencies may be attributable to differences in the classification of coping. In our study, emotion-focused coping included emotional venting, emotional support, humor, acceptance, religion, and self-blame of the Brief COPE. In the meta-analysis, only emotional venting was considered in the Brief COPE, along with related constructs from different coping instruments.

We chose to consider illness perception as a whole to limit the number of variables included in our models to avoid statistical overfitting, convey a clear message, and facilitate translation to the clinical setting. However, the authors of the meta-analysis observed an inverse effect of particular illness perception dimensions on coping responses.[32] For example, a greater perceived control over the illness (less threatening) was associated with less distress. Nonetheless, the authors found that the greater the degree of control over the illness, the more emotional venting coping was used, resulting in greater distress. Future studies could investigate how individual illness perception dimensions are associated with coping in adults with CHD.

Our study showed that both illness perception and coping, which are modifiable variables, are associated with anxiety in adults with CHD, and could potentially be targeted in future efforts to reduce anxiety. A psychotherapeutic approach, including psychoeducation and cognitive behavioral techniques,[2] could be an interesting approach to modify inadequate perceptions of CHD and to encourage adaptive coping methods. Further study is required to determine if such psychological interventions are effective.

STUDY LIMITATIONS. The MHI ACHD Center is within a quaternary referral institution such that caution should be exerted in extrapolating results to the ACHD population at large and those living in other geographical areas and sociocultural contexts. Patients followed at the MHI ACHD Center are likely to have more complex CHD and higher symptom burden (including NYHA functional class) than the

average population of adults with CHD. Moreover, geographic and sociocultural variations have been documented in anxiety symptoms and illness perception in adults with CHD.[8,24,25,33]

Secondly, an inherent limitation of cross-sectional studies is that causal relationships between variables of interest and anxiety symptoms cannot be inferred.

CONCLUSIONS

This study highlights the important association between illness perception and anxiety in adults with CHD. Illness perception accounts for a greater proportion of variance in anxiety symptoms than medical and sociodemographic characteristics. Our findings suggest that clinicians should consider the subjective illness experience in the care of adults with CHD and engage in further dialogue to understand the patient's perspective, moderate it if it differs from objective facts, and address related questions. Moreover, illness perception is not only directly linked to anxiety symptoms, it also has an indirect association with anxiety symptoms through coping, particularly problem-focused coping. Perceptions of illness and coping strategies could be potential targets for future prevention and intervention strategies to prevent or mitigate anxiety symptoms.

ADDRESS FOR CORRESPONDENCE: Dr Judith Brouillette, Montreal Heart Institute, 5000 Bélanger Street, Montreal, Quebec H1T 1C8, Canada. E-mail: judith.brouillette@icm-mhi.org.

PERSPECTIVES

COMPETENCY IN PATIENT CARE: ACHD are an emerging population needing comprehensive health management, including psychosocial care. Nearly 15% of participants in our study had clinically significant anxiety symptoms, 17% had subclinical symptoms, and 68% had anxiety symptoms considered within the normal spectrum. Clinicians should be aware that ACHD with more severe anxiety symptoms were younger, had a lower income, had a worse NYHA functional class, and had a more threatening perception of their CHD. Illness perception explains more anxiety than the medical or sociodemographic variables. Therefore, clinicians should pay particular attention to how patients perceive their illness. The clinician is well-positioned to open a discussion with the patient about how they perceive their illness and attempt to adjust it if it deviates from the objective facts. Additionally, patients should be encouraged to use problem-focused coping responses, as it was the only coping category with a significant indirect path in the association of illness perception and anxiety symptoms.

TRANSLATIONAL OUTLOOK: We found that illness perception and coping strategies are modifiable factors associated with anxiety. Longitudinal studies are required to allow testing of causality between illness perception, coping, and anxiety. Future studies could aim to identify modifiable factors associated with illness perception, coping, and anxiety, in order to identify targets for interventions. Eventually, interventions might be evaluated to attenuate the threatening perception of the illness, to help prevent or mitigate anxiety symptoms in ACHD. Additionally, it would be pertinent to investigate how individual characteristics that influence perceptions of self and events, such as personality traits, are associated with illness perception, coping style, and anxiety.

REFERENCES

1. Khairy P, Ionescu-Ittu R, Mackie AS, Abrahamowicz M, Pilote L, Marelli AJ. Changing mortality in congenital heart disease. *J Am Coll Cardiol*. 2010;56(14):1149–1157. https://doi.org/10.1016/j.jacc.2010.03.085

2. Kovacs AH, Brouillette J, Ibeziako P, et al. Psychological outcomes and interventions for individuals with congenital heart disease: a scientific statement from the American Heart Association. *Circ Cardiovasc Qual Outcomes*. 2022;15(8): e000110. https://doi.org/10.1161/hcq.0000000000000110

3. Fogleman ND, Apers S, Moons P, et al. Regional variation in quality of life in patients with a Fontan circulation: a multinational perspective. *Am Heart J*. 2017;193:55–62. https://doi.org/10.1016/j.ahj.2017.07.019

4. Kasmi L, Calderon J, Montreuil M, et al. Neurocognitive and psychological outcomes in adults with dextro-transposition of the great arteries corrected by the arterial switch operation. *Ann Thorac Surg*. 2018;105(3):830–836. https://doi.org/10.1016/j.athoracsur.2017.06.055

5. Olatunji BO, Cisler JM, Tolin DF. Quality of life in the anxiety disorders: a meta-analytic review. *Clin Psychol Rev*. 2007;27(5):572–581. https://doi.org/10.1016/j.cpr.2007.01.015

6. Benderly M, Kalter-Leibovici O, Weitzman D, et al. Depression and anxiety are associated with high health care utilization and mortality among adults with congenital heart disease. *Int J Cardiol*. 2019;276:81–86. https://doi.org/10.1016/j.ijcard.2018.09.005

7. Jackson JL, Hassen L, Gerardo GM, Vannatta K, Daniels CJ. Medical factors that predict quality of life for young adults with congenital heart disease: what matters most? *Int J Cardiol*. 2016;202:804–809. https://doi.org/10.1016/j.ijcard.2015.09.116

8. Rassart J, Apers S, Kovacs AH, et al. Illness perceptions in adult congenital heart disease: a multi-center international study. *Int J Cardiol*. 2017;244:130–138. https://doi.org/10.1016/j.ijcard.2017.06.072

9. Morgan K, Villiers-Tuthill A, Barker M, McGee H. The contribution of illness perception to psychological distress in heart failure patients. *BMC Psychol*. 2014;2(1):50. https://doi.org/10.1186/s40359-014-0050-3

10. Leventhal H, Benyamini Y, Brownlee S, et al. Illness representations: theoretical foundations. In: Petrie KJ, Weinman JA, eds. *Perceptions of Health and Illness: Current Research and Applications*. Harwood Academic Publishers; 1997:19–45.

11. Hagger MS, Orbell S. A meta-analytic review of the common-sense model of illness representations. *Psychol Health*. 2003;18(2):141–184. https://doi.org/10.1080/088704403100081321

12. Carver C. Coping. In: Gellman MD, Turner JR, eds. *Encyclopedia of Behavioral Medicine*. Springer New York; 2013:496–500.

13. Balmores-Paulino RS. Avoidance coping strategies. In: Zeigler-Hill V, Shackelford TK, eds. *Encyclopedia of Personality and Individual Differences*. Springer International Publishing; 2018:1–3.

14. Leventhal H, Leventhal EA, Contrada RJ. Self-regulation, health, and behavior: a perceptual-cognitive approach. *Psychol Health*. 1998;13(4): 717–733. https://doi.org/10.1080/08870449808407425

15. American Heart Association, Stout KK, Daniels CJ, Aboulhosn JA, et al. 2018 AHA/ACC guideline for the management of adults with congenital heart disease: a report of the American College of Cardiology/American Heart Association Task Force on Clinical Practice Guidelines. *J Am Coll Cardiol*. 2019;73(12):e81–e192. https://www.jacc.org/doi/10.1016/j.jacc.2018.08.1029

16. Zigmond AS, Snaith RP. The hospital anxiety and depression scale. *Acta Psychiatr Scand*. 1983;67(6):361–370. https://doi.org/10.1111/j.1600-0447.1983.tb09716.x

17. Broadbent E, Petrie KJ, Main J, Weinman J. The brief illness perception questionnaire. *J Psychosom Res*. 2006;60(6):631–637. https://doi.org/10.1016/j.jpsychores.2005.10.020

18. Carver CS. You want to measure coping but your protocol' too long: consider the brief cope. *Int J Behav Med*. 1997;4(1):92–100. https://doi.org/10.1207/s15327558ijbm0401_6

19. Timmermans I, Versteeg H, Meine M, Pedersen SS, Denollet J. Illness perceptions in patients with heart failure and an implantable cardioverter defibrillator: dimensional structure, validity, and correlates of the brief illness perception questionnaire in Dutch, French and German patients. *J Psychosom Res*. 2017;97:1–8. https://doi.org/10.1016/j.jpsychores.2017.03.014

20. Preacher KJ, Hayes AF. Asymptotic and resampling strategies for assessing and comparing

indirect effects in multiple mediator models. *Behav Res Methods*. 2008;40(3):879-891.

21. *The SAS System for Windows, Release 9.2*. SAS Institute Inc; 2013.

22. Leslie CE, Schofield K, Vannatta K, Jackson JL. Perceived health competence predicts anxiety and depressive symptoms after a three-year follow-up among adolescents and adults with congenital heart disease. *Eur J Cardiovasc Nurs*. 2020;19(4): 283-290.

23. Gleason LP, Deng LX, Khan AM, et al. Psychological distress in adults with congenital heart disease: focus beyond depression. *Cardiol Young*. 2019;29(2):185-189.

24. Lévesque V, Laplante L, Shohoudi A, et al. Implantable cardioverter-defibrillators and patient-reported outcomes in adults with congenital heart disease: an international study. *Heart Rhythm*. 2020;17(5):768-776. https://doi.org/10.1016/j.hrthm.2019.11.026

25. 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. https://doi.org/10.1016/j.ijcard.2017.10.064

26. Andonian C, Freilinger S, Kaemmerer H, Beckmann J. Psychological well-being in adults with congenital heart disease: testing the predictive value of illness identity. *Heart Mind*. 2021;5(2):45.

27. Kovacs AH, Saidi AS, Kuhl EA, et al. Depression and anxiety in adult congenital heart disease: predictors and prevalence. *Int J Cardiol*. 2009;137(2):158-164. https://doi.org/10.1016/j.ijcard.2008.06.042

28. Holbein CE, Fogleman ND, Hommel K, et al. A multinational observational investigation of illness perceptions and quality of life among patients with a Fontan circulation. *Congenit Heart Dis*. 2018;13(3):392-400. https://doi.org/10.1111/chd.12583

29. Norozi K, Zoege M, Buchhorn R, Wessel A, Geyer S. The influence of congenital heart disease on psychological conditions in adolescents and adults after corrective surgery. *Congenit Heart Dis*. 2006;1(6):282-288. https://doi.org/10.1111/j.1747-0803.2006.00048.x

30. Raphael C, Briscoe C, Davies J, et al. Limitations of the New York Heart Association functional classification system and self-reported walking distances in chronic heart failure. *Heart*.

2007;93(4):476-482. https://doi.org/10.1136/hrt.2006.089656

31. Bredy C, Ministeri M, Kempny A, et al. New York Heart Association (NYHA) classification in adults with congenital heart disease: relation to objective measures of exercise and outcome. *Eur Heart J Qual Care Clin Outcomes*. 2018;4(1):51-58. https://doi.org/10.1093/ehjqcco/qcx031

32. Hagger MS, Koch S, Chatzisarantis NLD, Orbell S. The common sense model of self-regulation: meta-analysis and test of a process model. *Psychol Bull*. 2017;143(11):1117-1154. https://doi.org/10.1037/bul0000118

33. Van Bulck L, Goossens E, Luyckx K, et al. Healthcare system inputs and patient-reported outcomes: a study in adults with congenital heart defect from 15 countries. *BMC Health Serv Res*. 2020;20(1):496. https://doi.org/10.1186/s12913-020-05361-9

KEY WORDS anxiety, congenital heart defect, coping, illness perception

APPENDIX For supplemental tables, please see the online version of this paper.