Original Article

Psychological Well-being in Adults with Congenital Heart Disease: Testing the Predictive Value of Illness Identity

Caroline Andonian[1,2], Sebastian Freilinger[1], Harald Kaemmerer[1], Jürgen Beckmann[2,3]

[1]Department of Paediatric Cardiology and Congenital Heart Defects, German Heart Center Munich, Technical University Munich, [2]Department of Sport and Health Sciences, Chair of Sport Psychology, Technical University Munich, Munich, Germany, [3]Faculty of Health and Behavioural Sciences, School of Human Movement and Nutritions Sciences, The University of Queensland, Brisbane, Australia

Abstract

Background: Due to advances in medical care and treatment of congenital heart disease (CHD), the number of adults with CHD (ACHD) is constantly growing. The psychological situation of ACHD has recently received increasing attention. There is evidence that adaptation to CHD may be affected by psychological factors, especially in how patients integrate their illness into their identities. The present study examined illness identity as a mediator of the association between a self-rated health and emotional distress among ACHD. **Materials and Methods:** The study used a cross-sectional design. A sample of 229 ACHD (38 ± 12.5 [18–73] years; 45% female) provided background data and completed three questionnaires on self-rated health (EuroQol group's visual analog scale), illness identity (Illness Identity Questionnaire), and emotional distress (Hospital Anxiety and Depression Scale) at the German Heart Center Munich. Serial multiple mediator models were tested using PROCESS macro for SPSS. **Results:** Perceived health had a direct and indirect effect on emotional distress which was mediated by illness identity ($P < 0.05$). Compared to all other dimensions of illness identity, engulfment fully mediated the relationship between self-rated health and emotional distress, when adjusted for sociodemographic and clinical confounders. The model explained 42% of variance in total emotional distress ($R^2 = 0.416$). The extent of emotional distress did not differ as a function of CHD complexity. **Conclusions:** Illness identity emerged as a strong mediating factor between a patient's self-rated health and psychological outcomes. More importance needs to be directed toward assessing a patient's health perception and psychological state, independently of cardiac severity. Based on present findings, targeted psychocardiological interventions should include psychoeducational components and emotion-focused strategies.

Keywords: Adults, congenital heart disease, illness identity, psychocardiology, psychological situation

Introduction

Due to technological and medical advances, over 90% of patients with congenital heart disease (CHD) reach adulthood.[1] However, many adults with CHD (ACHD) face ongoing medical complications and psychosocial disadvantages.[2] In recognition of this fact, increasing attention has been devoted to patient-reported outcomes in ACHD in terms of quality of life (QOL) and related psychological outcomes.[3] Existing findings indicate that ACHD presents a significantly higher risk for developing psychological disorders.[4-7] This is alarming given the fact that chronic emotional distress is known to negatively influence cardiovascular health even leading to premature mortality.[8] However, the current data can not sufficiently explain the

variability in their psychological functioning. This gap in knowledge may contribute to the striking lack of psychosocial support for this patient population.[9]

Most of the literature demonstrates that psychological functioning in ACHD is irrespective of their objective physical condition indicating that symptoms experienced by patients are relevant factors to consider.[10] Illness identity provides a novel framework for understanding how patients integrate their illness into their sense of self. One can differentiate

Access this article online

Quick Response Code:

Website:
www.heartmindjournal.org

DOI:
10.4103/hm.hm_32_21

Address for correspondence: Caroline Andonian, MSc,
Department of Pediatric Cardiology and Congenital Heart Disease, German Heart Center Munich, Technical University Munich, Munich, Germany.
E-mail: andonian@dhm.mhn.de

How to cite this article: 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;5:45-51.

between four different states of illness identity: rejection, engulfment, acceptance, and enrichment. Previous research has demonstrated a link between negative illness identity states and emotional distress in ACHD.[11,12] Yet, there have been no mediational studies to prove the predictive value of illness identity for psychological adjustment and long-term health outcomes in ACHD. A necessary step is to identify modifiable risk factors that predict emotional distress in ACHD. Therefore, the aim of the present study was to ascertain whether illness identity is a reliable predictor of emotional outcomes to detect patients at risk and consequently establish appropriate psychotherapeutic interventions.

Figure 1 depicts hypothetical pathways among self-rated health, illness identity, and psychological distress. We hypothesized that a patient's self-rated health status would be related to psychological adjustment (measured by depression and anxiety) and that illness identity would mediate this relationship in different ways. Given the inconsistent findings on the relationship between objective medical parameters and psychological adjustment, it was also investigated on how cardiac disease severity and illness identity interact to influence the relationship between perceived health and emotional distress. The identification of mechanisms that affect psychological well-being in ACHD is necessary to establish adequate psychocardiological interventions for this patient population.

MATERIALS AND METHODS

Population
The present study was part of the nationwide, cross-sectional MERLIN-CHD initiative which investigates the health situation of ACHD in Germany. Study participants were recruited at the German Heart Center Munich, which covers a large spectrum of ACHD ranging from simple to complex CHD. Inclusion criteria were (1) confirmed diagnosis of CHD;[13] (2) participant age 18 years and older; (3) necessary physical, cognitive, and language capabilities to complete self-report questionnaires.

Procedures
The study was approved by the ethical committee in June 2019 (158/19 S). The protocol is in line with ethical guidelines

Figure 1: Conceptual framework

established by the Declaration of Helsinki.[14] Informed consent was obtained from all participants. All patients received a study package consisting of (1) study information, (2) informed consent, and (3) set of questionnaires. Medical records were reviewed for each patient for verified medical information. Data on primary CHD diagnosis, disease complexity, and functional status were extracted and recorded separately.

Measures
Four domains were measured: (1) demographics, (2) self-rated health, (3) illness identity, and (4) emotional distress. All outcome variables were assessed by standardized questionnaires.

Demographic and clinical information
Patient demographic information was obtained with a background questionnaire. Medical data (CHD diagnosis, functional status, surgical status, cyanosis) were gathered using medical chart reviews. CHD diagnosis was divided into three groups according to the classification of CHD severity by Warnes *et al.*[15]

Independent variable: Perceived health status
Self-rated health was assessed as part of the EQ-5D-5L[16] which provides a simple, generic measure of a patient's perceived health status. For reasons of feasibility, this study used the EuroQol group's visual analog scale (EQ-VAS) as a single-item approach for the measurement of a patient's perceived health status. The EQ-VAS seeks the respondents' overall rating of their health.[17] It is presented as a vertical scale, marked from 0 ("the worst health you can imagine") to 100 ("the best health you can imagine"). Studies generally report high levels of validity and reliability compared to multi-item questionnaires (Cronbach's alpha = 0.87).[18] Satisfactory psychometric properties to assess both, perceived health status and QOL, specifically among ACHD, have been documented by Moons *et al.*[19]

Mediator variable: Illness identity
The Illness Identity Questionnaire (IIQ) reflects how individuals integrate their chronic illness into their sense of self. Items are rated on a 5-point Likert scale ranging from 1 – strongly disagree to 5 – strongly agree. The 25-item IIQ assesses four different illness identity states: rejection (5 items), engulfment (8 items), acceptance (5 items), and enrichment (7 items). To maintain high equivalence between the original English questionnaire and the translated German version, forward and back translation was performed by two independent bilingual translators.[12] Cronbach's alpha values for ACHD were 0.79 for rejection, 0.88 for acceptance, 0.93 for engulfment, and 0.90 for enrichment. All factor correlations are below 0.8 and indicate high discriminant validity.[12]

Dependent variable: Psychological functioning
The Hospital Anxiety and Depression Scale (HADS) was used to assess perceived emotional distress in terms of depressive and anxiety symptoms.[20,21] Cronbach's alphas indicate high internal consistency for all measures (0.83 to 0.87.) The HADS
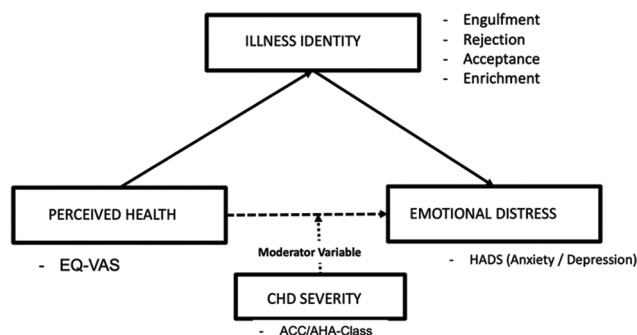
is a 14-item self-report questionnaire which combines 7-item subscores for depression (HADS-D) and anxiety (HADS-A). Each item is scored on a 4-point Likert scale ranging from 0 (not present) to 3 (considerable). The item scores are summed to provide subscale scores on the HADS-D and the HADS-A which may range from 0 to 21. Studies most commonly employ a cut-off point of ≥8 for each of the respective subscales to indicate a probable case.[22]

Statistical analysis

All statistical calculations. Were carried out using SPSS Statistics 27.0 (IBM, 2020). Descriptive statistics were calculated measuring univariate coefficients of central tendency and distribution. To evaluate the applied instruments, psychometric properties of the underlying scales were analyzed by measuring reliability and minimal selectivity. Pearson product-moment correlations were applied to test bivariate correlations between the main variables. To test direct and indirect effects of perceived health on emotional distress, serial multiple mediator models were used in accordance with procedures described by Hayes and the PROCESS macro for SPSS. Direct and indirect (i.e., mediated) effects were assessed by evaluation of 95% confidence intervals produced by bootstrapping (bootstrapping = 5000). Absolute correlation coefficients did not exceed 0.8 which provides no indication for multicollinearity. For all tests, the statistical significance level was set at $P < 0.05$.

RESULTS

Patient characteristics

Sociodemographic and clinical characteristics are presented in Table 1. Out of 300 distributed questionnaires, 241 questionnaires were returned (response rate: 80.3%). After exclusion for reasons of ineligibility or incompleteness, the remaining 229 ACHDs were retained for final analysis (45% female and 55% male). The mean age of ACHD was 38.2±12.5 [18-73] years.

The severity of CHD was determined according to the Warnes *et al*. classification system as simple ($n = 56$, 24.5%), intermediate ($n = 88$, 38.4%), and severe ($n = 85$, 37.1%).[15] Patients were subclassified according to their functional status based on their symptomatic restrictions (100). 218 patients were in FC I/II (95.2%), 9 patients in FC III (3.9%), and 2 in FC IV (9%).

Bivariate correlations between main study variables

Bivariate correlations among the study variables are presented in Table 2. Higher self-rated health was associated with functional illness identity dimensions and lower emotional distress. Functional illness identity states were also inversely correlated with emotional distress. Older age was consistently associated with higher emotional distress, as well as lower self-rated health.

Multiple mediator model

The suggested conceptual model explained 42% of the variance in total emotional distress (adjusted $R^2 = 0.416$). The highest

Table 1: Sociodemographic and clinical characteristics of adults with congenital heart disease

Variables	Value
Age (years)	38.2±12.5 (18-73)
Gender (*n*=229), *n* (%)	
Female	103 (45.0)
Male	126 (55.0)
Marital status (*n*=221), *n* (%)	
Married	94 (42.5)
Divorced	4 (1.8)
Engaged	42 (19.0)
Single	80 (36.2)
Widowed	1 (.5)
Level of education completed (*n*=217), *n* (%)	
No schooling completed	11 (5.1)
Primary school degree	55 (25.3)
Secondary school degree	60 (27.6)
Vocational/polytechnic degree	28 (12.9)
General university entrance qualification	63 (29.0)
Financial standing (*n*=223), *n* (%)	
Poor	21 (9.4)
Fair	61 (27.4)
Good	141 (63.2)
Functional Class (*n*=229), *n* (%)	
I/II	218 (95.2)
III	9 (3.9)
IV	2 (.9)
Severity code of CHD (*n*=214), *n* (%)	
Simple	56 (24.5)
Intermediate	88 (38.4)
Severe	85 (37.1)
Leading diagnosis (*n*=229), *n* (%)	
Complex congenital heart defects	75 (32.8)
Posttricuspid shunts	18 (7.9)
Left heart malformation	44 (19.2)
Right heart malformation	39 (17.0)
Pretricuspid shunts	35 (15.3)
Other	18 (7.9)

CHD=Congenital heart disease

coefficients of determination were found for depression and anxiety through the mediator variable engulfment (depression $R^2 = 0.564$, anxiety $R^2 = 0.405$). In contrast, rejection indicated the weakest coefficients of determination, especially in explaining the variance of anxiety ($R^2 = 0.339$).

Different interactions between the paths self-rated health – illness identity (α) and illness identity – emotional distress (β) were analyzed, and respective regression weights are depicted in Table 3. All effects were significant indicating that self-rated health and illness identity met the criteria for mediational analysis. Moderated mediation models were tested, such that clinical variables were applied as moderators and removed as model covariates. Neither of the moderated mediation models produced significant effects ($P > 0.05$), suggesting that the association between all three variables did not differ as a function of CHD severity.

Table 2: Bivariate Pearson correlations

	1	2	3	4	5	6	7
1. Age							
2. Rejection	0.005						
3. Acceptance	−0.088	−0.486**					
4. Engulfment	0.118	0.376**	−0.415**				
5. Enrichment	−0.047	−0.281**	0.365**	−0.135*			
6. EQ-5D VAS	−0.272**	−0.205**	0.324**	−0.616**	0.213**		
7. HADS anxiety	0.164*	0.388**	−0.389**	0.613**	−0.259**	−0.489**	
8. HADS depression	0.223**	0.304**	−0.401**	0.685**	−0.338**	−0.662**	0.671**

$*P<0.05$, $**P<0.01$. VAS=Visual Analog Scale, HADS=Hospital Anxiety and Depression Scale

Table 3: Mediation models for the relationship between perceived health status (EQ-VAS) and emotional distress (Hospital Anxiety and Depression Scale)

Mediation models	Coefficient A VAS-illness Id (α^*)	Coefficient B illness Id-HADS (β^*)	Direct effect C VAS-HADS (c^*)	Indirect effect C' (CI) ($c^{*'}$)
3.1 Rejection anxiety	−0.2161**	0.3352**	−0.3961*	−0.0724 (−0.1311-−0.0254)
3.2 Engulfment anxiety	−0.06160**	0.5163**	−0.2021	−0.3180 (−0.4369-−0.2062)
3.3 Acceptance anxiety	0.3220**	−0.2744**	−0.3549*	−0.0884 (−0.1611-−0.0338)
3.4 enrichment anxiety	0.2200**	−0.1721*	−0.4709**	−0.0379 (−0.0791-−0.0064)
3.5 Rejection depression	−0.2161**	0.2056**	−0.5755**	−0.0444 (−0.0880-−0.00124)
3.6 Engulfment depression	−0.6160**	0.4508**	−0.3857**	−0.2777 (−0.3784-−0.1846)
3.7 Acceptance depression	0.3220**	−0.2414**	−0.5183**	−0.0777 (−0.1383-−0.0317)
3.8 Enrichment depression	0.2200**	−0.2193**	−0.6188**	−0.0482 (−0.0894-−0.0141)

$*P<0.05$, $**P<0.01$, Model shows standardized regression weights demonstrating that illness identity dimensions mediate the association between perceived health (EQ-VAS) and emotional distress (HADS); α^*, β^*, c^*, $c^{*'}$=Standardized regression coefficient. VAS=Visual Analog Scale, HADS=Hospital Anxiety and Depression Scale, CI=Confidence interval, EQ-VAS= visual analog scale

After illness identity was included into the model, perceived health (x) significantly predicted illness identity, which, in turn, significantly predicted emotional distress (y). Coefficient A was negative in the association between self-rated health and maladaptive illness identity ($\alpha_{eng}^* = -0.06160$; $\alpha_{rej}^* = -0.2161$, $P < 0.001$) but turned positive in interaction with anxiety and depression ($\beta_{eng}^* = 0.5163$, $\beta_{rej}^* = 0.3352$, $P=0.001$). Accordingly, high self-rated health predicted higher values in functional illness identity dimensions (acceptance and enrichment), which consequently lead to lower scores on emotional distress. On the contrary, low self-rated health leads to higher values in dysfunctional illness identity states (rejection and engulfment), which resulted in higher emotional distress ($P < 0.05$).

For the direct pathways, after controlling for demographic and clinical variables, self-rated health was inversely related to emotional distress. Schematic illustrations of the effects of perceived health on emotional outcomes through the mediator variable engulfment are depicted in Figures 2 and 3. All submodels showed significant negative effects, except for engulfment anxiety ($c_{eng}^* = -0.2021$, $P > 0.05$) [Figure 3]. Throughout the entire analysis, the relationship between self-rated health (x) and emotional distress (y) was partially mediated by illness identity. Engulfment fully mediated the association between self-rated health and anxiety. In accordance with previous calculations, all indirect effects were negative with anxiety engulfment showing the highest values.

Summary of analyses

In sum, illness identity constituted a significant predictor of outcome in the relationship between self-rated health and emotional distress satisfying the criteria for a mediator. High self-rated health was related to increase in functional illness identity states, which in turn lead to less emotional distress. In contrast, low subjective health was associated with increase in dysfunctional illness identity dimensions which consequently compromised psychological functioning. Compared to all other dimensions, engulfment showed the strongest effects on psychological outcomes. Objective medical parameters were unrelated to psychological outcomes and therefore did not qualify for moderation testing.

DISCUSSION

The present study offers valuable insights into mediating psychological factors associated with emotional distress in ACHD. To our knowledge, there have been no mediation studies on the recently developed concept of illness identity. The present findings support the proposed conceptual framework, suggesting that illness identity mediates the relationship between individual perceptions of health and emotional distress among ACHD. The mediating effect varied between different dimensions of illness identity with engulfment presenting the highest correlations to emotional distress. The present study further echoed previous findings

that did not confirm a relationship between objective medical parameters and psychological functioning in ACHD.[23] Furthermore, it corroborates findings by Callus *et al*. who confirmed that a patients' subjective disease severity rating had a profound effect on psychological outcomes. The present study expands Callus *et al*.'s findings in several significant ways.[23]

The concept of illness identity provides a new perspective on life with CHD. Existing literature focused on illness perceptions[3] but failed to explore how patients incorporate their illness into their identities.[24] In the present study, illness identity accounted for unique differences in emotional distress among ACHD. Consistent with prior findings, dysfunctional illness identity states were associated with higher emotional distress, while functional illness identity states correlated with lower emotional distress. Engulfment fully mediated the relationship between self-rated health and emotional distress regardless of potential confounders. In ACHD, the predictive role of engulfment had thus far only been associated with the occurrence of excessive health-care encounters.[25] However, in preceding studies, no mediational analysis could be performed due to insufficient data variability. Therefore, underlying mechanisms of psychological well-being in ACHD could not be scrutinized.[3]

The constellation of depression and anxiety in highly engulfed patients is especially alarming due to a 3-fold increased risk of all-cause mortality.[8] Engulfment was defined as the degree to which a patient's self-concept is dominated by his or her CHD. It is claimed that engulfment is determined by both, the individual's perception of his or her illness and the individual's perception of his or her self.[26] Closer inspection of the IIQ revealed that engulfment-related items mainly refer to the impact of illness on the self without considering subjective perceptions of illness. Exemplary items of the IIQ read as follows, i.e., "My illness has a strong impact on how I see myself," "It seems as if everything I do, is influenced by my illness." However, the present results provide evidence that individual perceptions of the severity and permanence of their CHD may play an equally important role in either hampering or promoting ways of adapting to CHD. Future research should acknowledge both dimensions of illness experience to further extend the currently introduced framework of illness identity in ACHD.

In contrast to all other forms of illness identity, the global model fit for rejection displayed the lowest scores within the present study. This could be explained by inherent psychometric weaknesses for the rejection dimension confirmed in an earlier validation study.[12] From a psychological standpoint, rejection might be used by patients who perceive their illness as an external threat to their identity and engage in avoidance behaviors aimed at reducing the seriousness of their CHD.[13] Indeed, avoidance has often been shown to be unrelated to depressive and anxious symptoms in the general population which could potentially explain the weak correlations within the present study.[27]

The present findings are consistent with the shifting perspectives model of chronic illness by Paterson.[28] In their model, the perception of reality, not reality itself, influences how patients adjust to their chronic illness. This is strongly reflected by present results which found significant correlations with subjective health ratings regardless of objective parameters. Furthermore, the authors describe chronic illness experience as a continually shifting process, in which either illness or wellness prevails in a patient's perception. Similarly, the current findings indicate that perceptions of high disease severity have patients pay increased attention to the burden associated with their CHD. In contrast, perceptions of low disease severity allow a shift from "victim of circumstances to creator of circumstances" enabling patients to take ownership of their health and actively manage their chronic condition as a consequence of functional illness integration.[28]

Implications for clinical practice

Given the fact that subjective perception and not objective parameters predict emotional outcomes in ACHD, medical professionals should pay particular attention to educating patients on their condition. Especially in light of the striking supply deficits in ACHD,[29] patients would benefit from ongoing assessments of their illness identity within comprehensive cardiac care. Based on the present results, especially engulfed patients may benefit from emotion-focused interventions by restructuring and transforming problematic emotions on the basis of a caring, client-centered relationship.[30] Although psychotherapeutic interventions have been shown to improve emotional distress in adults with acquired heart disease,[31] there are currently no evidence-based interventions to address psychological well-being in ACHD. However, a recent pilot study conducted by Kovacs *et al*. demonstrated significant effects of cognitive-behavioral group intervention (called ACHD-CARE Program, $n = 42$) on psychological distress in ACHD.[29] Large-scale trials are strongly encouraged to eventually establish manualized interventions for ACHD.

Limitations

Some caution is warranted in the interpretation of current findings. First, due to the cross-sectional design, the direction of the relationship between predictor variables and psychological outcomes cannot be determined. Within the present study, the relationship between self-rated health, illness identity, and psychosocial adjustment might not be one directional and all variables reflect instantaneous, interactional processes. Longitudinal research is needed to further disentangle the pathways. Second, it remains unclear whether illness identity is dynamic or fixed in nature. In accordance with the shifting perspectives model of chronic illness,[28] some patients might assume one predominant dimension but not a static entity. Further influencing factors need to be explored to develop a more accurate understanding of illness identity. Third, disease severity, derived from the Warnes *et al*. classification system, was selected as a central parameter.[15] However, the informational value of this categorization might be limited, and various other indicators of CHD severity need to be considered
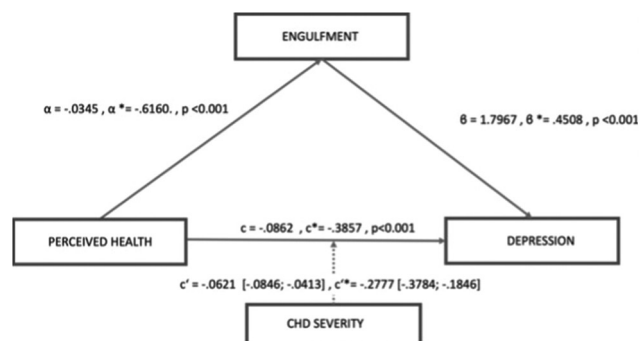
Figure 2: Schematic illustration of the effect of perceived health (visual analog scale) on depression (Hospital Anxiety and Depression Scale-D) through the mediator variable engulfment
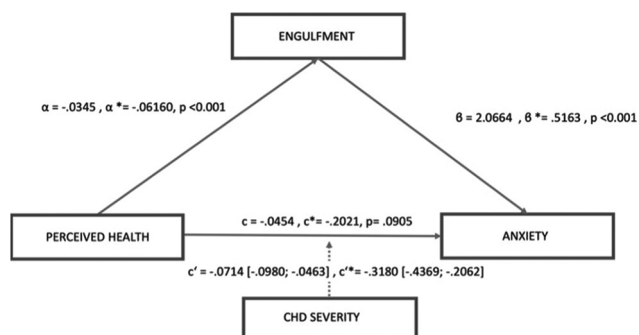

Figure 3: Schematic illustration of the effect of perceived health on anxiety (Hospital Anxiety and Depression Scale-A) through the mediator variable engulfment

to draw valid conclusions on the impact of potential medical confounders. Fourth, this study was based on self-report measures and obtained results were therefore subject to biased responses. Furthermore, patients were not blinded to the aim of the study which might involve the risk of the frequently discussed Hawthorne effect.[32] Fifth, doubts could be raised regarding sufficient precision of the applied instruments for research and screening purposes. Even though the applied psychometric tools are high-quality screening instruments, neither of them can replace a structured clinical interview. We eagerly await studies to extend our findings to confirmed clinical disorders. Sixth, the included sample may not necessarily be fully representative of the population of ACHD. Although patients were equally distributed according to their CHD complexity, patients with higher functional limitations were underrepresented in the sample (FCIII and IV: 4.8%). Whether this underrepresentation has affected, the results of this study need to be clarified.

CONCLUSIONS

There have been several calls in the literature for the investigation of explanatory factors in the psychological adjustment of ACHD. The present study provides novel insights into clinical and psychosocial factors associated with emotional distress in ACHD. It strengthens the hypothesis that adaption to CHD is affected by psychological factors, especially how patients appraise their condition and integrate their illness into their identities. The present findings are crucial for guiding future research and, especially, clinical practice. CHD patients might benefit from early psychological support consisting of both, psychoeducational components to enhance the understanding of their condition, and emotion-focused strategies to address difficult thoughts and emotions associated with their CHD to facilitate adaptive ways of illness integration. This might be a useful starting point for establishing psychotherapeutic interventions for ACHD.

Conflicts of interest

There are no conflicts of interest.

REFERENCES

1. Zimmermann R, Schranz D, Ewert P, Kaemmerer H. Pulmonary arterial hypertension in congenital heart defects with shunt: A heterogeneous and complex constellation. Dtsch Med Wochenschr 2013;138:1244-6.
2. Kovacs AH, Sears SF, Saidi AS. Biopsychosocial experiences of adults with congenital heart disease: Review of the literature. Am Heart J 2005;150:193-201.
3. Holbein CE, Fogleman ND, Hommel K, Apers S, Rassart J, Moons P, *et al.* A multinational observational investigation of illness perceptions and quality of life among patients with a Fontan circulation. Congenit Heart Dis 2018;13:392-400.
4. Andonian C, Beckmann J, Biber S, Ewert P, Freilinger S, Kaemmerer H, *et al.* Current research status on the psychological situation of adults with congenital heart disease. Cardiovasc Diagn Ther 2018;8:799-804.
5. Westhoff-Bleck M, Briest J, Fraccarollo D, Hilfiker-Kleiner D, Winter L, Maske U, *et al.* Mental disorders in adults with congenital heart disease: Unmet needs and impact on quality of life. J Affect Disord 2016;204:180-6.
6. Roseman A, Kovacs AH. Anxiety and depression in adults with congenital heart disease: When to suspect and how to refer. Curr Cardiol Rep 2019;21:145.
7. Kovacs AH, Saidi AS, Kuhl EA, Sears SF, Silversides C, Harrison JL, *et al.* Depression and anxiety in adult congenital heart disease: Predictors and prevalence. Int J Cardiol 2009;137:158-64.
8. Watkins LL, Koch GG, Sherwood A, Blumenthal JA, Davidson JR, O'Connor C, *et al.* Association of anxiety and depression with all-cause mortality in individuals with coronary heart disease. J Am Heart Assoc 2013;2:e000068.
9. Kovacs AH, Grace SL, Kentner AC, Nolan RP, Silversides CK, Irvine MJ. Feasibility and outcomes in a pilot randomized controlled trial of a psychosocial intervention for adults with congenital heart disease. Can J Cardiol 2018;34:766-73.
10. Moons P, Luyckx K. Quality-of-life research in adult patients with congenital heart disease: Current status and the way forward. J Acta Paediatr 2019;108:1765-72.
11. Oris L, Luyckx K, Rassart J, Goubert L, Goossens E, Apers S, *et al.* Illness identity in adults with a chronic illness. J Clin Psychol Med Settings 2018;25:1-2.
12. Andonian C, Beckmann J, Ewert P, Freilinger S, Kaemmerer H, Oberhoffer-Fritz R, *et al.* Assessment of the psychological situation in

adults with congenital heart disease. J Clin Med 2020;9:779.

13. Thiene G, Frescura C. Anatomical and pathophysiological classification of congenital heart disease. Cardiovasc Pathol 2010;19:259-74.

14. Association WM. Ethical principles for medical research involving human subjects. Eur J Emerg Med 2001;8:221.

15. Warnes CA, Liberthson R, Danielson GK, Dore A, Harris L, Hoffman JI, *et al.* Task force 1: The changing profile of congenital heart disease in adult life. J Am Coll Cardiol 2001;37:1170-5.

16. Herdman M, Gudex C, Lloyd A, Janssen M, Kind P, Parkin D, *et al.* Development and preliminary testing of the new five-level version of EQ-5D (EQ-5D-5L). Qual Life Res 2011;20:1727-36.

17. Feng Y, Parkin D, Devlin NJ. Assessing the performance of the EQ-VAS in the NHS PROMs programme. Qual Life Res 2014;23:977-89.

18. de Boer AG, van Lanschot JJ, Stalmeier PF, van Sandick JW, Hulscher JB, de Haes JC, *et al.* Is a single-item visual analogue scale as valid, reliable and responsive as multi-item scales in measuring quality of life? Qual Life Res 2004;13:311-20.

19. Moons P, Van Deyk K, De Bleser L, Marquet K, Raes E, De Geest S, *et al.* Quality of life and health status in adults with congenital heart disease: A direct comparison with healthy counterparts. Eur J Cardiovasc Prev Rehabil 2006;13:407-13.

20. Zigmond AS, Snaith RP. The hospital anxiety and depression scale. Acta Psychiatr Scand 1983;67:361-70.

21. Norton S, Cosco T, Doyle F, Done J, Sacker A. The hospital anxiety and depression scale: A meta confirmatory factor analysis. J Psychosom Res 2013;74:74-81.

22. Olssøn I, Mykletun A, Dahl AA. The hospital anxiety and depression rating scale: A cross-sectional study of psychometrics and case finding abilities in general practice. BMC Psychiatry 2005;5:1-7.

23. Callus E, Utens EM, Quadri E, Ricci C, Carminati M, Giamberti A, *et al.* The impact of actual and perceived disease severity on pre-operative psychological well-being and illness behaviour in adult congenital heart disease patients. Cardiol Young 2014;24:275-82.

24. Mauthner OE, De Luca E, Poole JM, Abbey SE, Shildrick M, Gewarges M, *et al.* Heart transplants: Identity disruption, bodily integrity and interconnectedness. Health (London) 2015;19:578-94.

25. Van Bulck L, Goossens E, Luyckx K, Oris L, Apers S, Moons P. Illness identity: A novel predictor for healthcare use in adults with congenital heart disease. J Am Heart Assoc 2018;7:e008723.

26. Beanlands HJ. Engulfment among adults with chronic renal disease, a study of self-loss and its correlates (Doctoral dissertation, University of Toronto, Ontario, Canada,2001) 2001.

27. Blalock JA, Joiner TE. Interaction of cognitive avoidance coping and stress in predicting depression/anxiety. Cognit Ther Res 2000;24:47-65.

28. Paterson BL. The shifting perspectives model of chronic illness. J Nurs Scholarsh 2001;33:21-6.

29. Kovacs AH, Bandyopadhyay M, Grace SL, Kentner AC, Nolan RP, Silversides CK, *et al.* Adult Congenital Heart Disease-Coping And REsilience (ACHD-CARE): Rationale and methodology of a pilot randomized controlled trial. Contemp Clin Trials 2015;45:385-93.

30. Timulak L, McElvaney J. Emotion-focused therapy for generalized anxiety disorder: An overview of the model. J Contemp Psychother 2016;46:41-52.

31. Linden W, Phillips MJ, Leclerc J. Psychological treatment of cardiac patients: A meta-analysis. Eur Heart J 2007;28:2972-84.

32. Adair JG. The Hawthorne effect: A reconsideration of the methodological artifact. J Appl Psychol 1984;69:334.