Cardiac Denial and Expectations Associated With Depression in Adults With Congenital Heart Disease

Geoffrey D. Huntley, MD[a], Kristen M. Tecson, PhD[b], Sandeep Sodhi, MD[c], Joshua Saef, MD[c], Kamila S. White, PhD[d], Philip A. Ludbrook, MD[c], Ari M. Cedars, MD[e,*], and Jong Mi Ko, MA[e]

Depression in adults with congenital heart disease is highly prevalent and strongly associated with adverse prognosis. Better management of risk factors for depression may improve clinical outcomes in this population. We conducted a single-site, cross-sectional study of 78 adults with congenital heart disease followed at Washington University School of Medicine. Data considered in the analyses included retrospectively obtained clinical information and patients' self-assessed psychosocial functioning and health status. To identify the clinical and psychosocial variables associated with depression, we built a stepwise multivariate model to measure the relative contribution of these variables to depression status. The prevalence of depression in our sample was 26%. Our model accounted for approximately 67% of the variability in depression scores. The final model consisted of the Cardiac Denial of Impact Scale, expectations domain of Barriers to Care, and the energy and social domains of the Rand 36-Item Short Form Health Survey. Clinical variables did not predict variability in depression scores. In conclusion, greater cardiac denial and negative expectations of the healthcare team were associated with increased depression symptoms in ACHD. **© 2019 Elsevier Inc. All rights reserved.** **(Am J Cardiol 2019;123:2002−2005)**

Extraordinary advances in cardiology and cardiac surgery have significantly increased the number of adults living with congenital heart disease (CHD) in recent decades.[1,2] As the physical health of adults with CHD (ACHD) has improved, there is increasing emphasis on improving psychological health due to its known impact on health outcomes and quality of life.[3] ACHD have multiple risk factors for poor psychological health.[4,5] In North America, recent data suggest that the prevalence of depression is relatively high in ACHD and has been shown to independently predict worse clinical outcomes, in particular worse self-perceived health status, worse quality of life, shorter event-free survival, and increased risk of death or hospitalization due to heart problems.[3,6−8] Just as ACHD benefit from specialized clinical care for their cardiac disease, their unique cohort of life stressors may require specialized psychological care. As such, it is fundamentally important to identify variables associated with depression. In the present study, we explored the relation between demographic, clinical, and psychosocial variables and depression in a cohort of ACHD.

Methods

The data collection protocol and study participants have been previously described.[3] We conducted a single-site, cross-sectional study of ACHD followed at the Center for ACHD at Washington University School of Medicine in St. Louis between April 2013 and October 2014. The study was approved by the institutional review boards of Washington University School of Medicine, the University of Missouri in St. Louis, and Baylor University Medical Center. Informed consent was obtained from each patient. Participants were asked to complete a series of validated psychometric questionnaires and clinical variables were abstracted from their medical records (Supplementary Table 1). All clinical variables were collected within 9 months of enrollment. Psychometric questionnaires utilized include the Beck Depression Inventory-Version II (BDI-II), the Beck Anxiety Inventory, the Cardiac Denial of Impact Scale (CDIS[9]), the RAND Short Form-36 (SF-36), and the Barriers to Care Questionnaire (BTC[10]). Responses to questionnaires were scored according to established algorithms.

Continuous variables are presented as means with standard deviations or medians [quartile 1, quartile 3], if skewed. Categorical variables are presented as frequency (percentage). We assessed differences in psychometric measurements and clinical variables between subjects who were and were not depressed with two-sample *t* tests (or Wilcoxon Rank Sum tests, as appropriate) and chi-square tests. Due to the small number of patients with depression, we elected to evaluate psychometric and clinical variables' associations with the continuous measure of depression symptoms (BDI-II), instead of the dichotomized version. As such, we selected variables having a significant association with depression in bivariate analyses and considered them jointly in a multivariable model. Although 2 anxiety measures were found to be

[a]Department of Internal Medicine, The University of Texas Southwestern Medical Center, Dallas, Texas; [b]Baylor Heart and Vascular Institute, Dallas, Texas; [c]Department of Cardiology, Washington University School of Medicine, St. Louis, Missouri; [d]Department of Psychology, University of Missouri, St. Louis, Missouri; and [e]Division of Cardiology, Department of Internal Medicine, The University of Texas Southwestern Medical Center, Dallas, Texas. Manuscript received February 22, 2019; revised manuscript received and accepted March 8, 2019.

This work was funded by the Baylor Health Care System Foundation and the Department of Internal Medicine at the University of Texas Southwestern Medical Center.

See page 2005 for disclosure information.

*Corresponding author: Tel: 214-645-7500; fax: 214-645-7501.

E-mail address: Ari.cedars@utsouthwestern.edu (A.M. Cedars).

0002-9149/© 2019 Elsevier Inc. All rights reserved.
https://doi.org/10.1016/j.amjcard.2019.03.011

significantly associated with depression, we did not consider them for inclusion in the multivariable model considering the well-known relation between anxiety and depression, and this study is intended to identify lesser known relations. Additionally, we did not consider the emotional well-being and role limitation due to emotional problems domains of SF-36 for inclusion in the multivariable model as they likely directly measure symptoms of depression. We used the Akaike information criterion as the condition to identify the optimal model.[11] This method not only takes into account the model's fit, but also its complexity. This is a data-driven model and we did not make the decision to include/exclude domains of the SF-36 (except for the 2 abovementioned domains) or BTC; variables that were not part of the optimal combination were not included in the final model. Due to skew in residuals from the regression using raw BDI-II scores, we employed a log-transformation to achieve normality. We also identified 1 high leverage point and subsequently removed it from the analysis. There were no further concerning issues in the residuals after making these 2 modifications. We then calculated the standardized coefficients as well as the semipartial ω^2 values for each covariate in the model to evaluate its magnitude of impact and the proportion of total variation accounted for by the covariate, above and beyond the others. Hypothesis tests were conducted assuming a 2-sided alternative and a type I error rate of 5%. Analyses were performed in SAS version 9.4 (Cary, North Carolina).

Results

Of 105 ACHD who participated in this study, 78 (74%) completed the questionnaires; 20 (26%) had elevated depressive symptoms (BDI-II >13). Analyses to test for differences between patients who completed the questionnaires and those who did not showed no differences in patient age (MCOM = 45.04 vs MNC = 40.17 years, $p = 0.2$), gender (chi-square [1, n = 103] = 3.66, $p = 0.5$), or total years of education (MCOM = 15.18 vs MNC = 14.58 years, $p = 0.3$).

In unadjusted analyses (Table 1), clinical variables did not differ significantly by depression status, but several psychometric measures did. Those with depression had higher scores on CDIS, anxiety sensitivity index, and anxiety score, and all of each domain of BTC (skill, marginalization, expectations, knowledge and beliefs, and pragmatics) and SF-36 (physical functioning, physical limitations, emotional limitations, energy, emotional well-being, social functioning, pain, and general health. The final multivariable model (Table 2) included the CDIS, BTC—expectations, and the domains of energy and social functioning of the SF-36. This model accounted for 67% of observed variability in BDI scores. In the final model, 3%, 3%, 4%, and 6% of the total variability in BDI score was explained by CDIS, BTC—expectations, energy domain, and social domain, respectively, above and beyond all others.

Discussion

We explored the relation between demographic, clinical, and psychological variables and depression in a cohort of 78 ACHD. We found that while depressive symptoms did not differ based on clinical or demographic variables in the multivariate model, 67% of the variability in BDI-II scores was explained by 4 psychometric variables: cardiac denial, BTC—expectations, and social and energy domains of SF-36. Our study is the first to demonstrate that greater denial and negative expectations of the healthcare team are associated with depression in ACHD.

To our knowledge, the present study is the first to find a positive association between denial and depressive symptoms in ACHD. The CDIS measures the degree to which a patient minimizes emotional distress due to heart disease. It may seem logical that those with a stronger tendency to deny the impact of cardiac disease will report decreased depression. Indeed, higher levels of denial have been reported to improve psychological outcomes in patients who underwent coronary artery bypass grafting and STEMI, respectively.[12,13] On the contrary, denial was positively associated with depression in our study cohort. It has been hypothesized that the process of living with a serious chronic disease from birth leads to the development of coping mechanisms, such as denial.[14] Our finding suggests that prolonged denial of cardiac disease may be maladaptive to overall emotional health. Alternatively, patients with depression may develop denial to avoid perseverating on the effect of their heart disease. Future studies are needed to replicate our findings and further elucidate the relation between depression and denial in ACHD.

The BTC—expectations measures the degree to which patients' negative expectations about interactions and care from their healthcare team work as barriers to obtain care (a low score signifies greater barrier). A negative relation between depression and BTC—expectations found in our study implies that patients with more depressive symptoms were more likely to identify negative expectations of their healthcare team as a major impediment to receive medical care. Although the BTC scale does not directly measure whether a patient's expectations are met, these findings suggest important areas of future research and interventions for adult CHD healthcare teams. Based on the association between lower patient expectations (i.e., skepticism) and worse mental health, higher rates of nonadherence, and less healthcare utilization and a direct correlation between patient satisfaction and patient expectations, increasing patient satisfaction may improve health outcomes.[15–17] Future studies and interventions focusing on identifying and improving aspects of the healthcare team that are most important to ACHD, and on how improving expectations might affect clinical outcomes, in particular in mental health, may improve overall health of ACHD. Given that many adult CHD clinicians report insufficient institutional support to focus heavily on patient satisfaction, increased focus on patient satisfaction in all parts of the healthcare experience may require the involvement of healthcare teams at both institutional and clinic levels.[18]

Expectedly, the social domain of SF-36 had the strongest association with depression. These findings support data reported by Kovacs et al[19] who found that loneliness, patient-perceived physical health status, and fear of negative evaluation explained 45% of the variance in BDI-II scores in a cohort of 225 ACHD. Similar to the present study, Kovacs et al[20,21] found no association between

Table 1
Patient characteristics by depression categorization (n = 78)

Variable	Depressed (n = 20)	Not depressed (n = 58)	p value
Male	8 (47%)	24 (51%)	0.7900
Age (years)	40.5 [30, 48.5]	43 [32, 51]	0.5030
White	15 (88%)	45 (96%)	0.2699
Married/cohabitating	10 (59%)	28 (60%)	0.5775
Belong to a religion	11 (65%)	40 (85%)	0.1727
Smoker	1 (6%)	1 (2%)	0.4496
Prior noncardiac surgery[3,11]	14 (82%)	35 (74%)	0.7401
Cirrhosis	0 (0%)	2 (4%)	1.0000
Implantable cardioverter-defibrillator	6 (35%)	5 (11%)	0.0844
Permanent pacemaker	9 (53%)	12 (26%)	0.1098
Cardiac valve disease	5 (29%)	18 (38%)	0.7121
Cyanotic	2 (12%)	4 (9%)	0.6432
Lesion cyanotic	9 (53%)	22 (47%)	0.5026
Health knowledge[1,3]	0.62 (± 0.08)	0.62 (± 0.09)	0.9875
Compliance[0,4]	70.84 (± 15.72)	72.63 (± 13.63)	0.6311
Number, cardiac surgeries[0,1]	2 [1, 2]	2 [1, 3]	0.7337
Noncardiac diagnoses[1,3]	4 [3, 6]	3 [2, 6]	0.2076
Hospital admissions	0.25 [0.14, 0.68]	0.2 [0.05, 0.5]	0.2247
Perceived stress scale	33 [31, 35]	31 [29, 34]	0.0797
Cardiac denial of impact score	25 [23.5, 27.5]	20.5 [17, 23]	<0.0001
Barriers to care: skill[0,1]	87.5 [73.21, 92.86]	96.43 [89.29, 100]	0.0075
Barriers to care: marginalization	90.91 [73.86, 96.59]	100 [95.45, 100]	0.0001
Barriers to care: expectations[0,1]	87.5 [76.79, 100]	100 [89.29, 100]	0.0136
Barriers to care: knowledge and beliefs[0,1]	100 [90.63, 100]	100 [100, 100]	0.0034
Barriers to care: pragmatics[0,1]	87.5 [72.22, 91.67]	91.67 [86.11, 97.22]	0.0107
Anxiety sensitivity index[0,3]	46.5 [30.5, 62]	16 [9, 27]	<0.0001
Social support score	5.54 [4.71, 6.96]	6.25 [5.75, 6.92]	0.1352
Social support: significant other	6.88 [5.25, 7]	6.88 [6, 7]	0.6593
Social support: family	6.13 [3.75, 7]	6.25 [6, 7]	0.2742
Social support: friends	5 [4, 7]	6 [5.5, 7]	0.1458
Anxiety score[0,1]	15 [10.5, 24.5]	5 [2, 7]	<0.0001
Physical functioning domain[1,0]	65 [40, 90]	85 [60, 95]	0.0088
Physical limitations domain[1,0]	25 [0, 75]	100 [75, 100]	0.0001
Emotional limitations domain	33.33 [0, 66.67]	100 [100, 100]	<0.0001
Energy domain	35 [25, 50]	65 [55, 80]	<0.0001
Emotional well-being domain	56 [40, 62]	86 [76, 92]	<0.0001
Social domain	50 [37.5, 75]	100 [87.5, 100]	<0.0001
Pain domain	72.5 [38.75, 90]	90 [70, 100]	0.0083
General health domain	46.56 (± 24.84)	64.4 (± 18.24)	0.0010
Encounters per year[0,6]	1.4 [0.61, 1.96]	0.92 [0.56, 1.57]	0.4031

Superscripts indicate missing data.
Medians were reported in [quartile 1, quartile 2].

depression and clinical or demographic factors, and a high prevalence of depression (33%). Factors that lead to decreased social functioning in ACHD include delayed progression into adulthood, impaired peer relations, and restricted employment opportunities.

Our findings on the relation between the energy domain and depression are consistent with recent data which demonstrated that energy level or patient-perceived physical capacity rather than objective physical capacity was associated with psychological outcomes.[22] Overall, these findings

Table 2
Model summary for the Beck Depression Index

Parameter	Estimate	Standard error	*t* value	Pr > |*t*|	Standardized estimate	Semipartial ω^2
Intercept	4.514	0.620	7.28	<0.001		
Cardiac denial of impact	0.036	0.013	2.8	0.007	0.207	0.032
Barriers to care—expectations	−0.016	0.006	−2.73	0.008	−0.197	0.030
Energy domain	−0.012	0.004	−3.09	0.003	−0.321	0.040
Social domain	−0.013	0.003	−3.76	<0.001	−0.377	0.061

add to the evidence suggesting interventions that enhance social interaction and independence can be beneficial in the adult CHD clinical practice.

There are limitations to the present study. The surveys utilized were self-reported, and, thus, are subject to the biases of missing data or poor recall. When structured psychiatric interviews are used for assessment, more patients are found to be depressed compared with self-report surveys, so this study may actually underestimate the true prevalence of depression in our sample.[19,23,24] In addition, the patients were recruited during regular outpatient visits due to convenience, so data may not be generalizable. Furthermore, patients receiving psychotherapy for depression or medications for either mood or cardiac disease, which may alter depressive symptoms, were not recorded. Lastly, conclusions about causation cannot be made in this cross-sectional study and it is unclear if findings with statistical significance are clinically relevant.

In conclusion, greater cardiac denial and negative expectations of the healthcare team are associated with elevated depression symptoms in ACHD. This study adds to the evidence that a greater focus on addressing mood disorders and improving the healthcare experience for ACHD may improve both clinical and psychological outcomes.

Disclosures

The authors have no conflicts of interest to disclose.

Supplementary materials

Supplementary material associated with this article can be found in the online version at https://doi.org/10.1016/j.amjcard.2019.03.011.

1. Marelli AJ, Ionescu-Ittu R, Mackie AS, Guo L, Dendukuri N, Kaouache M. Lifetime prevalence of congenital heart disease in the general population from 2000 to 2010. *Circulation* 2014;130:749–756.
2. Olsen M, Christensen TD, Pedersen L, Johnsen SP, Hjortdal VE. Late mortality among Danish patients with congenital heart defect. *Am J Cardiol* 2010;106:1322–1326.
3. Ko JM, Tecson KM, Al Rashida V, Sodhi S, Saef J, Mufti M, White KS, Ludbrook PA, Cedars AM. Clinical and psychological drivers of perceived health status in adults with congenital heart disease. *Am J Cardiol* 2018;121:377–381.
4. Kovacs AH, Silversides C, Saidi A, Sears SF. The role of the psychologist in adult congenital heart disease. *Cardiol Clin* 2006;24:607–618.
5. Cornett L, Simms J. At the "heart" of the matter: an exploration of the psychological impact of living with congenital heart disease in adulthood. *J Health Psychol* 2014;19:393–406.
6. Ko JM, Cedars AM. Depression in adults with congenital heart disease: prevalence, prognosis, and intervention. *Cardiovasc Innov Appl* 2018;3:97–106.
7. Diller G-P, Braütigam A, Kempny A, Uebing A, Alonso-Gonzalez R, Swan L, Babu-Narayan SV, Baumgartner H, Dimopoulos K, Gatzoulis MA. Depression requiring anti-depressant drug therapy in adult congenital heart disease: prevalence, risk factors, and prognostic value. *Eur Heart J* 2016;37:771–782.
8. Kourkoveli P, Rammos S, Parissis J, Maillis A, Kremastinos D, Paraskevaidis I. Depressive symptoms in patients with congenital heart disease: incidence and prognostic value of self-rating depression scales. *Congenit Heart Dis* 2015;10:240–247.
9. Fowers BJ. The cardiac denial of impact scale: a brief, self-report research measure. *J Psychosom Res* 1992;36:469–475.
10. Seid M, Sobo EJ, Gelhard LR, Varni JW. Parents' reports of barriers to care for children with special health care needs: development and validation of the barriers to care questionnaire. *Ambul Pediatr* 2004;4:323–331.
11. Akaike H. *Information Theory and an Extension of the Maximum Likelihood Principle*. New York, NY: Springer; 1998. p. 199–213.
12. Folks DG, Freeman AM, Sokol RS, Thurstin AH. Denial: predictor of outcome following coronary bypass surgery. *Int J Psychiatry Med* 1989;18:57–66.
13. Fang X, Albarqouni L, von Eisenhart Rothe AF, Hoschar S, Ronel J, Ladwig K-H. Is denial a maladaptive coping mechanism which prolongs pre-hospital delay in patients with ST-segment elevation myocardial infarction? *J Psychosom Res* 2016;91:68–74.
14. White KS, Pardue C, Ludbrook P, Sodhi S, Esmaeeli A, Cedars A. Cardiac denial and psychological predictors of cardiac care adherence in adults with congenital heart disease. *Behav Modif* 2016;40:29–50.
15. Fiscella K, Franks P, Clancy CM. Skepticism toward medical care and health care utilization. *Med Care* 1998;36:180–189.
16. Doescher MP, Saver BG, Franks P, Fiscella K. Racial and ethnic disparities in perceptions of physician style and trust. *Arch Fam Med* 2000;9:1156–1163.
17. Glickman SW, Boulding W, Manary M, Staelin R, Roe MT, Wolosin RJ, Ohman EM, Peterson ED, Schulman KA. Patient satisfaction and its relationship with clinical quality and inpatient mortality in acute myocardial infarction. *Circ Cardiovasc Qual Outcomes* 2010;3:188–195.
18. Rozenblum R, Gianola A, Ionescu-Ittu R, Verstappen A, Landzberg M, Gurvitz M, Jenkins K, Bates DW, Marelli AJ. Clinicians' perspectives on patient satisfaction in adult congenital heart disease clinics—a dimension of health care quality whose time has come. *Congenit Heart Dis* 2015;10:128–136.
19. Kovacs AH, Saidi AS, Kuhl EA, Sears SF, Silversides C, Harrison JL, Ong L, Colman J, Oechslin E, Nolan RP. Depression and anxiety in adult congenital heart disease: predictors and prevalence. *Int J Cardiol* 2009;137:158–164.
20. 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.
21. Kovacs AH, Moons P. Psychosocial functioning and quality of life in adults with congenital heart disease and heart failure. *Heart Fail Clin* 2014;10:35–42.
22. Ko JM, White KS, Kovacs AH, Tecson KM, Apers S, Luyckx K, Thomet C, Budts W, Enomoto J, Sluman MA, Wang J-K, Jackson JL, Khairy P, Cook SC, Subramanyan R, Alday L, Eriksen K, Dellborg M, Berghammer M, Johansson B, Mackie AS, Menahem S, Caruana M, Veldtman G, Soufi A, Fernandes SM, Callus E, Kutty S, Gandhi A, Moons P, Cedars AM. APPROACH-IS Consortium and International Society for Adult Congenital Heart Disease (ISACHD). Physical activity-related drivers of perceived health status in adults with congenital heart disease. *Am J Cardiol* 2018;122:1437–1442.
23. Westhoff-Bleck M, Briest J, Fraccarollo D, Hilfiker-Kleiner D, Winter L, Maske U, Busch MA, Bleich S, Bauersachs J, Kahl KG. Mental disorders in adults with congenital heart disease: unmet needs and impact on quality of life. *J Affect Disord* 2016;204:180–186.
24. Bromberg JI, Beasley PJ, D'Angelo EJ, Landzberg M, DeMaso DR. Depression and anxiety in adults with congenital heart disease: a pilot study. *Hear Lung J Acute Crit Care* 2003;32:105–110.