

Title: Psychological Functioning in Pediatric Patients with Single Ventricle Congenital Heart Disease: A Meta-Analysis and Systematic Review Protocol

Authors: Amanda D. McCormick, Megan M. Wilde, Christine E. Charpie, Kate M. Saylor, Sunkyung Yu, Melissa K. Cousino

Introduction: Mental health is an important yet understudied area of care for patients with congenital heart disease. Through limited studies, it is known that children and adults with congenital heart disease have increased incidence of mental health disorders when compared to their peers [1-3]. Some studies estimate that over half of adult patients with congenital heart disease have significant symptoms of a mood or anxiety disorder [3], although it is very likely that these symptoms are underrecognized. It is also known that in adults with congenital heart disease, depression is responsible for the variability of self-reported health status of patients, including physical functioning [4]. A prior review and meta-analysis of patient with complex congenital heart disease showed an increased risk of internalizing and externalizing behavior problems, however this review was not specific to the single ventricle population [5]. A recent review and meta-analysis of patients with children and adults with single ventricle physiology found worse health-related quality of life outcomes in this population [6], however currently less is known about psychological functioning specifically in the pediatric single ventricle population. The aim of this systematic review is to summarize and meta-analyze the existing literature of psychological outcomes in pediatric single ventricle patients. It is hypothesized that pediatric patients with single ventricle heart disease will have an increased risk of internalizing and externalizing problems as compared to their peers.

Inclusion Criteria: Quantitative studies published in English of pediatric patients (0 – 25 years of age) with single ventricle congenital heart disease if they examine psychological functioning as reported by the patient or the parent/caregiver. Exclusion criteria included qualitative studies and studies where more than half of the patients were older than 25 years of age. Studies that primarily evaluated neurodevelopmental or quality of life/health related quality of life that also included a quantitative measure of psychological functioning were included.

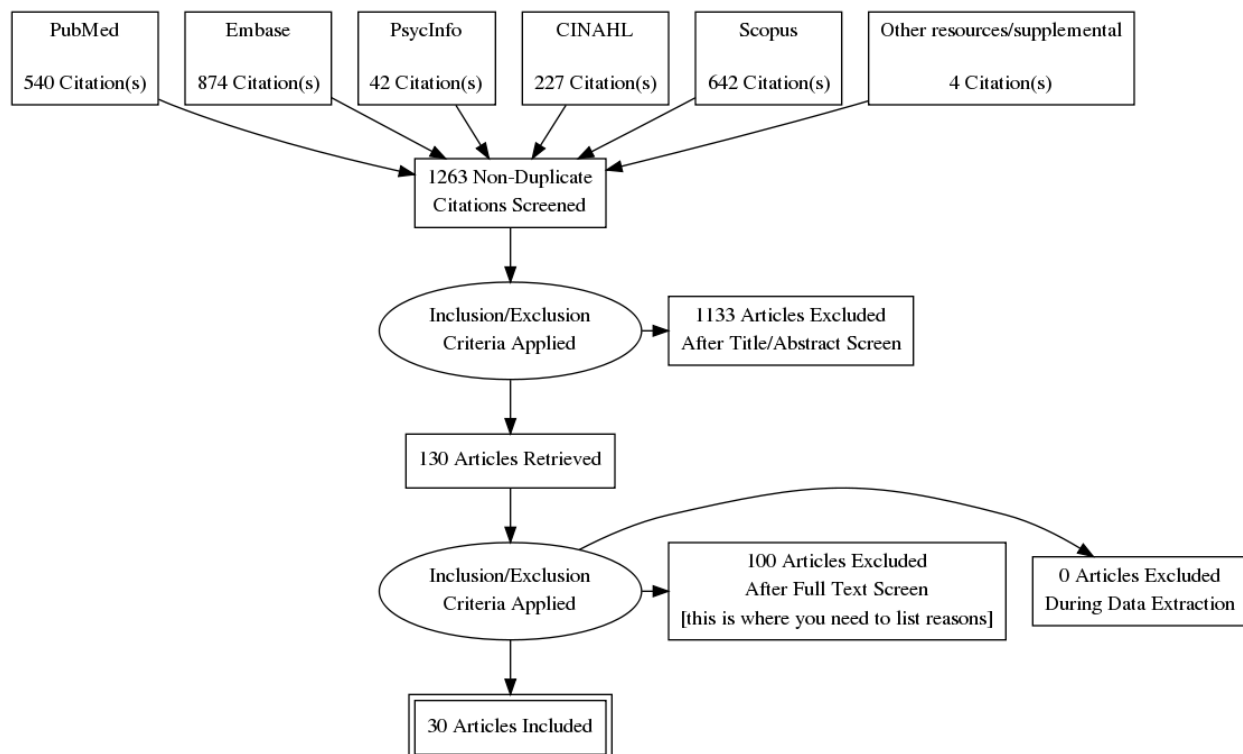
Search Strategy: The review team met with the informationist (KS) in early 2020. After discussion of included patients and outcomes, a search strategy was crafted to inform the selection of potential databases, concepts, and search terms. The databases selected for this project include PubMed, Embase, CINAHL, PsycInfo, and Scopus. When available, publication limits were applied to exclude commentaries and book chapters. Reviews were included in the initial search for evaluation of additional articles through the ancestry method, but were not included at manuscript review. The final searches were run on 4/21/20 and 6/15/20. Rayyan was used to manage citations and to identify and remove duplicates.

A complete search strategy is included in Appendix A.

Source of Evidence Selection:

Article selection will be based on the inclusion/exclusion criteria described above and will include review of title and abstract, followed by a full-article review. Two reviewers (AM and MW) reviewed all articles at the abstract level, with 251 articles reviewed by both reviewers for reliability and the remainder reviewed by one of the two reviewers for inclusion. At the full-article review level, articles were reviewed for inclusion by two or three reviewers (AM, MW, CC/MC) for inclusion and risk of bias. See Figure 1 for PRISMA flowchart of review process.

Figure 1. Review process



Details will be provided in a table for all articles identified for exclusion in the final synthesis of this review.

Data extraction:

The data extraction form will extract the following key information from each article:

1. Author(s)
2. Year of publication
3. Population and sample size
 - a. Mean age/range, sex, congenital heart disease diagnosis/% of sample with single ventricle heart disease, presence of control group
4. Measures of psychological functioning
 - a. Reporter of measure
5. Summary of results

Meta-Analysis: The full-text article review level included 12 articles that utilized the Achenbach Child Behavior Checklist (CBCL), which is widely used to detect behavioral and emotional problems in children and adolescents [7]. Six of the twelve articles provided adequate data for inclusion in the meta-analysis, either with raw or t-score data. These articles will be meta-analyzed for evaluation of total score, internalizing score, and externalizing score.

References

1. DeMaso, D.R., et al., *Psychiatric Disorders in Adolescents With Single Ventricle Congenital Heart Disease*. Pediatrics, 2017. 139(3).
2. Luyckx, K., et al., *Development and persistence of depressive symptoms in adolescents with CHD*. Cardiol Young, 2016. 26(6): p. 1115-22.
3. Kovacs, A.H., et al., *Depression and anxiety in adult congenital heart disease: predictors and prevalence*. Int J Cardiol, 2009. 137(2): p. 158-64.
4. Ko, J.M., et al., *Clinical and Psychological Drivers of Perceived Health Status in Adults With Congenital Heart Disease*. Am J Cardiol, 2018. 121(3): p. 377-381.
5. Karsdorp, P.A., et al., *Psychological and cognitive functioning in children and adolescents with congenital heart disease: a meta-analysis*. J Pediatr Psychol, 2007. 32(5): p. 527-41.
6. Marshall, K.H., et al., *Health-Related Quality of Life in Children, Adolescents, and Adults With a Fontan Circulation: A Meta-Analysis*. J Am Heart Assoc, 2020. 9(6): p. e014172.
7. Achenbach, T., *Manual for the Child Behavior Checklist/4-18 and 1991 Profile*. 1991, Burlington, VT: University of Vermont, Department of Psychiatry.

Appendix A.

1.

"Univentricular Heart"[Mesh] OR "Hypoplastic Left Heart Syndrome"[Mesh] OR "Tricuspid Atresia"[Mesh] OR "Fontan Procedure"[Mesh] OR Fontan[tw] OR "Tricuspid Atresias"[tw] OR "Tricuspid Valve Atresia"[tw] OR "Absent Right Atrioventricular Connection"[tw] OR "Hypoplastic Left Heart Syndrome"[tw] OR "Univentricular Heart"[tw] OR "Single Ventricle"[tw]

2.

"Pediatrics"[Mesh] OR "Child"[Mesh] OR "Adolescent"[Mesh] OR "Young Adult"[Mesh] OR child[tw] OR children[tw] OR Childhood[tw] OR "school age"[tw] OR youth[tw] OR pediatric[tw] OR adolescent[tw] OR adolescents[tw] OR adolescence[tw] OR teen[tw] OR teens[tw] OR teenager[tw] OR teenagers[tw] OR "young adult"[tw] OR Pediatric[tw] OR Pediatrics[tw] OR paediatric[tw] OR paediatrics[tw]

3.

"Quality of Life"[Mesh] OR "Psychology"[Mesh] OR "psychology" [Subheading] OR "Mental Health"[Mesh] OR "Mental Disorders"[Mesh] OR "Adaptation, Psychological"[Mesh] OR "Behavioral Symptoms"[Mesh] OR "Psychological Distress"[Mesh] OR "Anxiety"[Mesh] OR "Affect"[Mesh] OR Psychosocial[tw] OR "Mental health" [tw] OR "Mental illness" [tw] OR Coping[tw] OR "Quality of life"[tw] OR Depression[tw] OR depressed[tw] OR Anxiety[tw] OR Mood[tw] OR moods[tw] OR Trauma[tw] OR Stress[tw] OR stressed[tw] OR Distress[tw] OR Distressed[tw] OR wellbeing[tw] OR well-being[tw] OR coping[tw] OR Psychology[tw]

1 AND 2 AND 3

Limits: English