Preschool Neurodevelopmental Outcomes in Children with Congenital Heart Disease

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Objective To describe preschool neurodevelopmental outcomes of children with complex congenital heart disease (CHD), who were evaluated as part of a longitudinal cardiac neurodevelopmental follow-up program, as recommended by the American Heart Association and the American Academy of Pediatrics, and identify predictors of neurodevelopmental outcomes in these children.

Study design Children with CHD meeting the American Heart Association/American Academy of Pediatrics high-risk criteria for neurodevelopmental delay were evaluated at 4-5 years of age. Testing included standardized neuropsychological measures. Parents completed measures of child functioning. Scores were compared by group (single ventricle [1V]; 2 ventricles [2V]; CHD plus known genetic condition) to test norms and classified as: normal (within 1 SD of mean); at risk (1-2 SD from mean); and impaired (>2 SD from mean).

Results Data on 102 patients were analyzed. Neurodevelopmental scores did not differ based on cardiac anatomy (1V vs 2V); both groups scored lower than norms on fine motor and adaptive behavior skills, but were within 1 SD of norms. Patients with genetic conditions scored significantly worse than 1V and 2V groups and test norms on most measures.

Conclusions Children with CHD and genetic conditions are at greatest neurodevelopmental risk. Deficits in children with CHD without genetic conditions were mild and may not be detected without formal longitudinal testing. Parents and providers need additional education regarding the importance of developmental follow-up for children with CHD. *(J Pediatr 2017;183:80-6).*

Children with congenital heart disease (CHD) are at higher risk for neurodevelopmental problems than healthy children, across all time points in development, from infancy through adolescence.[1] Although IQ is often in the low average/average range, a characteristic pattern of mild deficits in multiple other domains, including motor and visual spatial skills, adaptive behavior, executive functioning, language, and social cognition, is common, and found in children with a wide range of CHD diagnoses.[2-7] Deficits are thought to be related to a number of factors including altered prenatal brain maturation,[8] comorbid genetic conditions,[9,10] perioperative and postoperative events,[11] socioeconomic status,[12] and parenting style.[13] As a result of these deficits, children with CHD are more likely than healthy children to require special education services,[14] resulting in a significant impact on them, their families and society.[15]

To promote early detection of delays and optimize outcomes, the American Heart Association (AHA) and the American Academy of Pediatrics (AAP) now recommend systematic evaluation of development in children with CHD throughout childhood.[1] Cardiac centers have begun to incorporate developmental follow-up programs as part of routine cardiac care.[16,17] We have previously reported developmental outcomes of children who were evaluated in our longitudinal developmental follow-up program over the first 3 years of life and found that 46% of patients were delayed in at least 1 domain (cognitive, language, or motor skills); feeding difficulty and medical and genetic comorbidities increased risk for delays.[10,18] The aim of this study was to summarize and identify predictors of neurodevelopmental outcomes for preschoolers who were seen as part of a longitudinal developmental evaluation program for children with CHD.

Methods

Children with CHD believed to be at high risk for developmental delay as defined by the AHA/AAP guidelines,[1] were recruited from the Herma Heart Center

1V	Single ventricle
2V	Two ventricles
AAP	American Academy of Pediatrics
AHA	American Heart Association
CPB	Cardiopulmonary bypass
DHCA	Deep hypothermic circulatory arrest
LOS	Length of stay
WPPSI	Wechsler Preschool and Primary Scales of Intelligence

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Developmental Follow-up Clinic at Children's Hospital of Wisconsin. Eligibility criteria and operation of the Herma Heart Center Developmental Follow-Up Clinic have been previously described.[10,16,18] Children were deemed to be at high risk for developmental delay and eligible for the clinic if they had any cardiac surgery as a neonate, surgery using cardiopulmonary bypass (CPB) in the first year of life, a cardiac defect resulting in cyanosis, or other comorbid conditions or complications such as prematurity, genetic syndromes, seizures, or cardiac arrest that placed them at higher risk for delay. Genetic testing was used to confirm a diagnosis when a genetic syndrome was suspected, but all patients did not routinely undergo genetic testing. All families whose children met the AHA/AAP high risk for delay criteria were contacted by letter and subsequently called to schedule a preschool evaluation. Children were seen for neurodevelopmental testing within the cardiology clinic; appointments lasted approximately 2-3 hours. Parents provided informed consent to have their child's data included in a databank approved by the Institutional Review Board at Children's Hospital of Wisconsin. No subjects were excluded based on race or other coexisting medical or genetic condition. Only children who spoke English were included, as tests were administered in English.

Children completed a variety of neurodevelopmental measures that were selected based on developmental challenges that are commonly seen in children with CHD. In addition, parents completed several measures of child functioning and behavior. All measures are validated and have normative values based on a healthy population. New editions of some measures were published during the 4 years in which evaluations were completed; the testing protocol was updated to include the most current version of all measures at the time of assessment.

The full scale IQ score from the Wechsler Preschool and Primary Scale of Intelligence (WPPSI), Third or Fourth Edition[19,20] (mean: 100 ± 15) was used as a measure of cognitive functioning. The WPPSI-Fourth Edition full scale IQ score correlates .86 with the WPPSI-Third Edition full scale IQ score. The Letter-Word Identification, Applied Problems, and Spelling subtests of the Woodcock Johnson III Tests of Achievement[21] were used to assess prereading, premath, and prespelling skills, respectively (mean: 100 ± 15). The Developmental Test of Visual Motor Integration, Sixth Edition[22] was used to assess visual motor integration ability (mean: 100 ± 15). The Pegboard subtest of the Wide Range Assessment of Visual Motor Abilities[23] was used to assess fine motor skills (mean: 100 ± 15). The General Communication Composite score of the Children's Communication Checklist-2 (completed by parent)[24] was used as a measure of language skills (mean: 100 ± 15). The General Adaptive Composite score of the Adaptive Behavior Assessment System-Second Edition (completed by parent)[25] was used as a measure of adaptive behavior (mean: 100 ± 15). The Global Executive Composite score of the Behavior Rating Inventory of Executive Function-Preschool Version (completed by parent)[26] was used as a measure of executive functioning (mean T score: 50 ± 10; higher scores indicate more problems). The Total score of the Conners' Parent Rating Scale-Revised-Short Form (completed by parent)[27] was used as a measure of attention problems (mean T score: 50 ± 10; higher scores indicate more problems). The Total Problems score of the Child Behavior Checklist (completed by parent)[28] was used as a measure of child behavior problems (mean T scores: 50 ± 10; higher scores indicate more problems). The Total score of the Social Responsiveness Scale, First or Second Edition (completed by parent)[29,30] was used as a measure of child social problems (mean T scores: 50 ± 10; higher scores indicate more problems). The Total score is comparable across versions of this measure, as items are exactly the same. The second edition of this measure allows for administration across a wider age range, and 1 subscale name was changed.

For children who were too developmentally impaired to complete a task (n = 10), the lowest possible score for that test was assigned. Children who did not complete a task for other reasons (separation anxiety, n = 1; distractibility, n = 1; language delay, n = 1; fatigue, n = 4; oppositional behavior, n = 7) were excluded from analysis for tasks they did not complete.

Statistical Analyses

Sample characteristics and clinical variables are presented as medians with IQR (25th percentile-75th percentile) for continuous data and frequencies (%) for categorical data. Neurodevelopmental test scores were converted to standard z scores based on test norm means/SDs and compared with the population mean using a Wilcoxon signed rank test. Converting neurodevelopmental test scores to standard z scores, a common metric, allowed for comparison of scores across measures, as not all neurodevelopmental tests have the same scales. To adjust for multiple comparisons, a step-down Bonferroni procedure was used because it is less conservative than the Bonferroni in controlling for the family of hypotheses error rate.[31] To perform this adjustment, raw P value needs to be a number. Therefore, a P value of <.0001 was treated as .0001. Scores were classified as: normal (within 1 SD of test mean); at risk (1-2 SDs from test mean); or impaired (>2 SDs from test mean). A 1-sample proportion test was used to examine whether the observed percentages were different from the expected percentages for impaired (2.5%) and at risk (13.5%) categories. A Kruskal-Wallis test or Mann-Whitney-Wilcoxon test was used to compare test scores by group (single ventricle [1V] without genetic condition; 2 ventricles [2V] without genetic condition; CHD with genetic condition). A Cochran-Armitage trend exact test was used to examine the trend in proportions of the number of domains that fell in the normal, at risk, or impaired range for the genetic vs 1V and 2V nongenetic groups. Univariable and multivariable logistic regression analyses were used to assess the impact of patient and clinical factors on binary neurodevelopmental test scores (at risk/impaired vs normal). Predictors with P value of < .1 from the univariate analyses were included in the multivariable logistic regression models (1 model for each neurodevelopmental test) using a forward selection method. The following variables were included as predictors based on our previous findings and a review of the literature: sex, race (White, non-Hispanic, vs others), maternal education (completed beyond high school vs completed high school or less), prenatal

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