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Parental perception of functional status and impact on the family of children with congenital heart surgery☆

Richard U. Garcia [a,*,1], Sanjeev Aggarwal [a], Girija Natarajan [b]

[a] Division of Cardiology, Carman and Ann Adams Department of Pediatrics, Children's Hospital of Michigan, Wayne State University School of Medicine, Detroit, Michigan, USA
[b] Division of Neonatology, Carman and Ann Adams Department of Pediatrics, Children's Hospital of Michigan, Wayne State University School of Medicine, Detroit, MI, USA

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ABSTRACT

Aims: To assess the functional status and the family impact of children with congenital heart defects (CHD), using the parental Functional Status II (FS-II) and the Impact on Family (IOF) questionnaires.
Methods: In this prospective observational study, parents of children who underwent surgery for CHD during the first year of life completed the FS-II and the IOF questionnaires. Genetic syndromes and prematurity <32 weeks were exclusion criteria. The FS-II generates a total score and age specific general health (GH), activity (A) and, responsiveness (R) subscales. The IOF generates a total scale and financial and sibling subscales.
Results: Our cohort (n = 100), comprised 54% males; the median (IQR) age was 32 (10–56) months. Eighteen children had age-specific scores in the 1–2 SD below mean range (n = 17) or more than 2 SD below the mean (n = 1) for "ill children". There were significant negative correlations between the total FS-II and total IOF ($r = -0.35$, $p < 0.001$) and financial IOF ($r = -0.35$, $p < 0.001$). RACHS-1 category of CHD 4–6 was associated with higher likelihood of lower functional status.
Conclusions: More complex CHD was associated with lower functional status, which correlated with a greater impact on the families. Parental questionnaires may be useful instruments for developmental surveillance in this population.

1. Introduction

Over the last few decades, advances in management of infants with congenital heart disease (CHD) have resulted in increasing survival rates. Survivors of complex CHDs, however, are now recognized to be at risk of neurodevelopmental sequelae, possibly related to intrinsic cardiac physiology, anesthesia, cardiopulmonary bypass, low birth weight, coexisting genetic and extracardiac anomalies and hospital stay [1–4]. The 2012 American Heart Association (AHA) Scientific Statement emphasizes that the prevalence and severity of developmental disorders increases with severity of CHD. It classifies neonates or infants requiring open heart surgery and children with other cyanotic lesions not requiring open heart surgery during the neonatal period or infancy as "high risk" for developmental disorders [1]. As part of developmental surveillance, it is recommended to "elicit and attend" to parents' concerns about their child's development [1]. The value of parental concerns as screening tools to detect and address developmental problems has been previously well-described [5].

The National Institute of Child Health and Human Development (NICHD) workshop on follow-up of high risk infants underscored the advantages of assessment of the functional strengths and challenges of children [6]. These include the need for a minimal database, the ability to assess typical performance, and ease of administration [7]. The NICHD statement also describes family/environmental characteristics and external supports that may be associated with outcomes and the need to measure outcomes from the family's perspectives in high-risk populations [6]. The Functional Status-II (FS-II) questionnaire is a validated measure for children with chronic illnesses, as reported by the parent (The Functional Status II R Measure is copyrighted by R.E.K.Stein, C.K. Riessman and D.J. Jessop, 1981, 1991) [8]. Scores have been shown to be unaffected by maternal psychological adjustment but related to the child's hospitalizations, clinical rating and overall behavioral adjustment to illness [9]. The parental FS-II questionnaires have been demonstrated to correlate with formal classifications of impairment by developmental testing in preterm infants and to be useful in perinatal hypoxic–ischemic encephalopathy and in tracheotomy-dependent patients [10,11]. Data on functional status of children with CHD, by the parents' perception, are scarce.

☆ All Authors take responsibility for all aspects of the reliability and freedom from bias of the data presented and their discussed interpretation.

* Corresponding author at: Division of Cardiology, Carman and Ann Adams Department of Pediatrics, Children's Hospital of Michigan, Wayne State University School of Medicine, 3901 Beaubien Blvd, Detroit, MI 48201-2119, USA.
E-mail address: rgarcias@dmc.org (R.U. Garcia).
[1] Children's Hospital of Michigan, Detroit, MI.

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There is increasing recognition that families of children with chronic illnesses may cope differently and that the impact on the family may modulate health-related outcomes. In addition to early identification of developmental deficits and therapies, family-level interventions may improve functional outcomes of children with CHD. A systematic review of 25 published studies on parental coping and quality of life of families of children with CHDs concludes that coping depends on individual and familial factors beyond the severity of the child's condition [12]. Fewer psychosocial resources and lower levels of support may be risk factors for higher psychological distress and lower well-being for both parent and the child [12]. The experiences, needs and ways of coping in families of children with CHD varied widely [12]. The majority of the included studies used quantitative methods and validated parental tools, including the Impact on the Family (IOF) scale [12–14]. The impact on the family of having a child with CHD requiring surgery during infancy and whether this correlates with the parental perception of the child's functional status is currently unknown. Our study seeks to address these knowledge gaps.

Our aims were to describe the functional status and the impact on the family, as reported by parents using the FS-II and IOF questionnaires and their correlation, in a cohort of children (1 month-6 years) with CHD who underwent surgical repair during infancy (≤12 months of age). We also investigated factors associated with FS-II more than 1 standard deviation (SD) below the mean in this population.

2. Materials and methods

This prospective study was performed in the cardiology clinic at Children's Hospital of Michigan. Approval was obtained from the Institutional Review Board at Wayne State University. Parents of children with CHD, aged 1 month to 6 years, were approached for written informed parental consent during a routine visit between November 2013 and October 2014. Children in whom surgical repair was not undertaken in infancy (≤12 months of age), diagnosed genetic syndromes or extreme prematurity (<32 weeks gestational age) were excluded. A single investigator (RG) administered the age-specific FS-II and the IOF questionnaires. The sample size of 100 was a convenience sample based on the number of yearly clinic visits that we have in the eligible age range Electronic medical records were reviewed to obtain data on the CHD, gestational age, insurance and details of the cardiac surgical procedure done during infancy and cardiac medications at the clinic visit. The cardiac surgical procedure was classified using the Risk-Adjusted Congenital Heart Surgery (RACHS)-1 scoring, in which higher categories are associated with higher risk of mortality [15].

The validated 43-item FS-II questionnaire by Stein and Jessop addresses elements such as communication, mobility, mood, energy, sleeping, and eating [8]. Parents respond to each question on a 3-point categorical Likert scale. The FS-II is administered in two parts; in the first part, the parent rates the functional limitation of the child or the extent of difficulty with specific behaviors; in the second part, the parent rates the extent (fully, partly, or not at all) to which the difficulty with specific behaviors is due to the ongoing illness. Scoring is based on the subset of items chosen by the parent indicating that the child's functional status was partially or fully impacted as a result of illness, which we defined as "CHD-related" in this study. Higher scores indicate a more favorable functional status. The FS-II questionnaire generates a total score and 2 component scores—general health (GH) in all age groups and responsiveness (R), activity (A) or interpersonal functioning (IPF), depending on the age group. The questionnaire provides means ± SD for the total score for all ages and for children <1 year, 1 year–23 months, 2–3 years, and 4 years and older. Stein and Jessop have validated the instrument using data from 732 children aged 2 weeks to 16 years who were either chronically ill or well [8]. In this study, similar to previous studies we used means and SD for "ill children" to define decreased functional status as any (total, GH or R/A/IPF) score more than 1 SD below the age-specific means. We decided to use means for "ill children" for the analysis since patients born with CHD have a chronic condition that requires lifelong medical care.

The IOF Scale is a 27-item scale which assesses financial burdens on families as well as emotional concerns and positive outcomes, using a 4-point Likert scale [13,14]. It is reported to have high internal consistency and construct validity [14]. The total IOF scales correlate with maternal psychiatric symptoms, poor education and public assistance and poor child health and adjustment and increased child hospitalizations. The instrument has one robust factor representing overall family impact of the child's chronic condition, and two subscales for financial and sibling impact [13].

In addition, parents completing the questionnaires were asked about their race, educational achievement (completed college degree, high school graduate or less), employment and household structure (single parent, both parents, single parent with other parent involved, foster care or other caregiver) and therapies (occupational, physical and speech) and special educational services that their child was receiving at the time of the clinic visit.

Statistical analysis: FS-II scores were described as mean ± SD or median (IQR). Bivariate comparisons between groups of children with and without decreased functional status as defined were conducted using the t test for continuous normally distributed variables, Mann–Whitney U test for skewed variables and χ^2 test for categorical variables. To investigate the association between clinical factors and decreased functional status, we performed binary logistic regression analysis, including RACHS-1 category, age, prematurity (<37 weeks gestational age) and parental education as potential confounders. Pearson correlation was used to examine the correlation between FS-II and IOF scales. SPSS software (version 21.0) was used for statistical analysis.

3. Results

We approached parents of 102 consecutive eligible patients, of whom 2 refused. The cohort (n = 100) comprised 54% males, 35% black, 45% white and 20% children of other races. Among enrolled children, 14% were born preterm and 3% were multiples. Table 1 describes the cardiac diagnoses and initial surgical procedures. The median (IQR) age of the initial procedure was 0.6 (0.3–4) months. The proportion of children who underwent a single procedure was 65%, whereas 19% underwent two and 16% had three or more procedures prior to enrollment. The median (IQR) ages at second and the most recent procedure were 7 [6–10] and 4.8 (0.6–10.5) months respectively. At the time of initial surgery, 57% of children were on public insurance.

At the time of FS-II administration, the median (IQR) age was 32 (10–56) months and median (IQR) time elapsed from last surgery was 18 (4–44) months. Cardiac medications were required in 36% of children, 74% of children lived in 2-parent households, 24% with a single parent and 1 child each was in foster care and with a single parent and no involvement of the other parent. During the study interview, 48% of parents reported that they had completed high school. In 44% of families, both parents were employed; one parent was employed in 43% and for 13% of children, both parents were unemployed. The proportion of children with public insurance at the time of the study was 62%; 6 children who had private insurance initially had changed to public insurance and 1 child switched to private insurance at the latter time point. Twenty three children were receiving occupational or physical therapy. One patient required ventilation, another was on tube feeding and 7 children had a G-tube. The caregivers reported a median (IQR) of 1 [1–2] hospitalization of 5 [2–15] days duration within 6 months prior to assessment in 25% of children.