


Parental Preferences Regarding Outpatient Management of Children with Congenital Heart Disease

Sarah K. Luthy¹  · Sunkyung Yu² · Janet E. Donohue² · Emefah Loccoh² · Caren S. Goldberg² · Jimmy C. Lu²

Received: 29 May 2015 / Accepted: 24 August 2015 / Published online: 5 September 2015
© Springer Science+Business Media New York 2015

Abstract The objective of this study was to identify patterns of health care usage among children with congenital heart disease (CHD) and determine predictors for contacting cardiologists for routine care. Parents of children with CHD completed surveys, indicating which provider they would contact first for 12 concerns. Predictors for preference for cardiologist evaluation were identified by multivariable logistic regression. Surveys were completed by 307 of 925 (33.2 %) parents. Median patient age was 9.4 years [interquartile range (IQR) 5.1–14.4 years] with a median of 1 cardiac surgery (IQR 1–3). Most parents agreed primary care physicians (PCPs) could identify problems related to CHD (70.0 %) and when to refer to cardiologists (89.6 %). More felt PCPs best understood their values (63.2 vs. 29.6 %, $P < 0.001$) and were more accessible (63.5 vs. 33.6 %, $P < 0.001$) than cardiologists. Parents preferred first evaluation by PCPs for 9 of 12 concerns. Preference for cardiology evaluation was independently associated with the number of cardiac catheterizations [adjusted odds ratio (AOR) for ≥ 2 catheterizations 2.4, 95 % confidence interval (CI) 1.1–4.9], belief the cardiologist better knew the child's medical history (AOR 2.4, 95 % CI 1.3–4.6), and provider accessibility (AOR 3.6, 95 % CI 1.8–7.4). Parents of CHD patients reported

close alignment with PCPs and would contact PCPs first for most routine care. However, some populations continue to contact cardiologists for routine care. Further study is needed to clarify best practices for clinician and parent education.

Keywords Congenital heart disease · Parental preference · Primary care · Pediatric cardiology · Coordination of care

Abbreviations

AOR	Adjusted odds ratio
CI	Confidence interval
CHD	Congenital heart disease
IOM	Institute of Medicine
IQR	Interquartile range
PCP	Primary care physician
PedsQL	Pediatric Quality of Life Inventory™

Introduction

Children with congenital heart disease (CHD) are a subset of a larger group of children with chronic medical conditions that may cause physical, social, or emotional impairment [7, 12, 21]. Prior research indicates that children with chronic medical conditions use an increasing proportion of health-care resources and experience greater morbidity and mortality compared to their peers [1, 3, 6, 15, 20]. Like all children, those with chronic medical conditions need routine health maintenance, but they also require long-term surveillance of their underlying diseases. Primary care physician (PCP) leadership in care coordination and shared decision-making, such as in the context

Electronic supplementary material The online version of this article (doi:10.1007/s00246-015-1257-7) contains supplementary material, which is available to authorized users.

✉ Sarah K. Luthy
sarah.luthy@childrenscolorado.org

¹ Section of Hospital Medicine, Children's Hospital Colorado, 13123 E. 16th Avenue, B302, Aurora, CO 80045, USA

² Division of Pediatric Cardiology, C. S. Mott Children's Hospital, Ann Arbor, MI, USA

of a medical home, leads to better clinical outcomes, fewer unmet needs, and decreased resource utilization among children with chronic medical conditions [2, 17, 18]. Despite the evidence supporting PCP involvement, however, how to balance the care of such children among subspecialists and PCPs, and how patient and parental preferences affect this interchange, are still being evaluated [11, 22]. Research on the management of children with CHD has yielded conflicting results. Parents in some studies preferred that cardiologists manage their children's care [25], while children with CHD relied on PCPs for most medical needs in other studies [10, 13].

We sought to evaluate perceptions of parents of children with CHD regarding their children's management and determine factors associated with increased preference for management by pediatric cardiologists compared to PCPs. In a contemporary cohort, we hypothesized that parents would prefer that pediatric cardiologists evaluate issues related to the cardiovascular system, but that the severity of the underlying cardiac defect would be associated with increased preference for cardiologist management of even non-cardiac concerns.

Methods

Study Design

This study was a cross-sectional survey of parents or guardians of children with CHD who were evaluated in the outpatient clinic at the Congenital Heart Center at C.S. Mott Children's Hospital. The Institutional Review Board at the University of Michigan approved this study.

Subject Selection

Potential subjects from all patients who had been seen for return visits at the outpatient clinic at the Congenital Heart Center in 2011 were included. We reviewed International Classification of Diseases, Ninth Revision (ICD-9), to exclude potential subjects who had no congenital heart disease (such as evaluation only for murmur or syncope). We used patients seen in 2011 to ensure that children were at least 2 years old at the time of enrollment, and thus had sufficient time to establish relationships with their PCPs and pediatric cardiologists. Investigators reviewed the medical records to confirm eligibility prior to recruiting potential subjects. Subjects were eligible if their children were <18 years old at the time of the study, had CHD that was confirmed by an echocardiogram, and had a PCP. Potential subjects were excluded if their primary language was not English, if their children had subsequently been discharged from the Congenital Heart Center, or if their children were

deceased. Surveys were mailed to potential subjects with enclosed, pre-addressed, postage-paid envelopes from December 2013 to March 2014. Subjects received no compensation for participating. Informed consent was obtained from all individual participants included in this study.

Survey Design

The survey consisted of 57 questions and required 15–20 minutes to complete (Supplementary data). The first part of the questionnaire asked which of the child's medical providers—PCP, pediatric cardiologist, or another provider—parents would contact first to manage 12 health-related concerns. The concept of first contact is a characteristic of primary care, as defined by the Institute of Medicine (IOM) [9]. The concerns were distributed among preventive care and acute symptom management to represent the comprehensive scope of care that PCPs may provide children with CHD. We chose the nature of the concerns, five of which were potentially referable to the cardiovascular system (sports clearance, tachypnea, exercise intolerance, chest pain, and syncope), based on the literature and clinical experience [13, 25]. Fourteen questions related to care coordination, knowledge of medical issues, accessibility, and the family-centered nature of care, were developed based on studies that used the IOM definition of primary care to evaluate the quality of pediatric care [4, 5, 19]. Subjects assessed their children's overall health, completed the physical functioning section of the Pediatric Quality of Life Inventory™ (PedsQL) version 4.0, with a scaled score calculated from 0 to 100 [23], and quantified their children's health-care utilization over the last year and duration of their relationships with their current medical providers.

Clinical Data Collection

Demographic information and CHD diagnoses were collected from medical records. To compare participants and non-participants, CHD severity was categorized as mild (isolated defects), moderate, or severe (including transposition of the great arteries and single-ventricle lesions) (Table 1). Children with multiple mild lesions were categorized as moderate; children with two or more moderate lesions were categorized as severe.

Data Analysis

Data are presented as frequency (percentage) or median (interquartile range [IQR]) as appropriate. Categorical variables were compared using one-sample binomial test or Mantel–Haenszel Chi-square test. Correlation between continuous variables was evaluated using Spearman's correlation coefficient. Higher cardiologist preference was

Table 1 Congenital heart disease diagnoses as confirmed by echocardiogram classified by severity

Mild	Moderate	Severe
Aortic insufficiency	Anomalous left coronary artery from the pulmonary artery	Double inlet left ventricle
Aortic stenosis	Aortopulmonary window	D-transposition of the great arteries
Atrial septal defect	Atrioventricular septal defect	Hypoplastic left heart syndrome
Branch pulmonary artery stenosis	Cor triatriatum	Shone complex
Coarctation of the aorta	Double outlet right ventricle	Single ventricle
Double-chamber right ventricle	Ebstein anomaly	Tricuspid atresia
Mitral insufficiency	L-transposition of the great arteries	Multiple—moderate
Mitral stenosis	Pulmonary atresia	
Partial anomalous pulmonary venous connections	Tetralogy of Fallot	
Pulmonary insufficiency	Total anomalous pulmonary venous connections	
Pulmonary stenosis	Truncus arteriosus	
Tricuspid regurgitation	Multiple—mild	
Ventricular septal defect		

defined as contacting a cardiologist first for ≥ 3 concerns, the cohort median. Univariate comparisons between higher versus lower cardiologist preference were made in demographics, CHD features, health-care utilization, perception of health, and attitudes toward providers, using Chi-square test or Fisher's exact test for categorical variables, Mantel-Haenzel Chi-square test for ordinal variables, and Wilcoxon rank-sum test for continuous variables. Variables from univariate analysis with P value < 0.05 were included in multivariable logistic regression using backward elimination method with improvements in the model fit evaluated by the model Chi-square statistics. Unadjusted (OR) and adjusted odds ratios (AOR) and 95 % confidence intervals (CI) for variables associated with higher cardiologist preference are reported. To avoid collinearity regarding ease of access, we combined "responds fastest to telephone calls" and "easier to see" into one variable. A P value < 0.05 was considered statistically significant. All analyses were performed using SAS v9.3 (SAS Institute, Cary, NC).

Results

Patient Characteristics

Surveys were sent to 925 eligible subjects from December 2013 to March 2014. The overall response rate was 38.4 %; 310 consented to participate and 45 declined. A total of 307 completed surveys were returned, which comprised the cohort for analysis. Demographic data are presented in Table 2. Ninety-nine (32.2 %) children had mild CHD, 129 (42.0 %) moderate, and 79 (25.7 %) severe. Respondents'

children had more severe CHD compared to those who did not respond ($P = 0.01$), but there was no significant difference in gender, age, presence of single-ventricle physiology, arrhythmias, or cardiac transplants between respondents and non-respondents. Participants' children underwent a median of 1 cardiac surgery (IQR 1–3) and 1 cardiac catheterization (IQR 0–2).

Perception of Health

Parents reported few limitations in the physical functioning portion of the PedsQL (median scaled score 84.4, IQR 59.4–100). Although most parents described their children's CHD as having no impact or only a small impact on their health (226, 73.6 %), only 196 (63.8 %) agreed their children were as healthy as other children, and only 163 (53.1 %) felt they could be treated like other children when ill. In the previous year, parents called/visited their children's PCPs a median of three times (IQR 2–5), compared to 1 call/visit (IQR 1–2) to their cardiologists. Perceived CHD severity more closely correlated with the number of cardiologist contacts ($r = 0.49$, $P < 0.001$) than PCP contacts ($r = 0.20$, $P = 0.001$).

Relationship with Health-Care Providers

Children had been followed by their PCPs and cardiologists a median of 6 years (IQR 3–11 and 3–10, respectively). The majority of parents reported confidence in their PCP's ability to determine whether a problem is related to CHD, knew when to refer, and knew the results of the last cardiology appointment (Fig. 1). Cardiologists had more

Table 2 Subject demographic characteristics ($N = 307$)

	N (%) or median (IQR)
Male sex	178 (58.0)
Age (years)	9.4 (5.1–14.4)
Caucasian race	257 (83.7)
Insurance	
Private	184 (59.9)
Public	49 (16.0)
Both public and private	74 (24.1)
Total household income	
Less than \$25,000	34 (11.1)
\$25,000–\$50,000	41 (13.4)
\$50,000–\$75,000	65 (21.2)
\$75,000–\$100,000	62 (20.2)
More than \$100,000	96 (31.3)
Unknown	9 (2.9)
Primary diagnosis	
Multiple—minor	42 (13.7)
Tetralogy of Fallot	35 (11.4)
Atrioventricular septal defect	30 (9.8)
Ventricular septal defect	25 (8.1)
Multiple—major	25 (8.1)
Aortic stenosis	22 (7.2)
Coarctation of the aorta	21 (6.8)
D-transposition of the great arteries	20 (6.5)
Atrial septal defect	16 (5.2)
Hypoplastic left heart syndrome	16 (5.2)
Other	55 (17.9)
Single-ventricle physiology	45 (14.7)
Number of cardiac surgeries	
0	47 (15.3)
1	130 (42.3)
≥ 2	130 (42.3)
Number of cardiac catheterizations	
0	124 (40.4)
1	89 (29.0)
≥ 2	86 (28.0)
≥ 1 non-cardiac surgery	122 (39.7)
Followed by ≥ 1 non-cardiac subspecialist	150 (48.9)

IQR interquartile range

frequently defined their roles than PCPs (75.2 vs. 59.3 %, $P < 0.001$). More parents felt that the PCP best understood their values/beliefs compared to the cardiologist (Fig. 2). They felt the PCP and cardiologist were comparable in their knowledge of the child's medical history and ability to explain issues, and felt similarly comfortable asking them questions. However, most parents preferred to discuss the emotional and financial aspects of CHD with cardiologists. Parents reported that PCPs were more accessible

than cardiologists, with respect to time to get to clinic ($P = 0.01$), rapidity of responding to telephone calls ($P < 0.001$), and ease of being seen in clinic ($P < 0.001$).

Preference for PCP Versus Cardiologist Management

Parents preferred to contact the PCP first for 9 out of 12 concerns (Fig. 3). A majority preferred first evaluation by the cardiologist for exercise intolerance, chest pain, and sports clearance. Parents preferred first evaluation by the cardiologist for a median of 3 concerns (IQR 1–5). In univariate analysis, demographic characteristics, parental perception of the child's health, and connection with the provider were significantly associated with preferring cardiology evaluation for ≥ 3 concerns (Table 3). Degree of physical limitation reported in the physical functioning portion of the PedsQL was not associated with cardiologist preference. Time to travel to the providers' clinics, clarification of roles by providers, the number of other subspecialists involved in the child's care, household income, and duration of the relationships with providers were not associated with greater cardiologist preference. In multivariable analysis (Table 4), the following variables remained independently associated with increased cardiologist preference: the number of cardiac catheterizations, the belief that the cardiologist had the best knowledge of the child's medical history, and provider accessibility.

Discussion

In our study, parents of children with CHD reported confidence in their children's PCPs and preferred to contact them first for most routine care, including some concerns potentially related to the cardiovascular system. However, provider accessibility, the belief that the cardiologist has the best knowledge of the child's medical history, and the number of prior cardiac catheterizations were independently associated with increased preference for cardiologist management. These data in a contemporary cohort suggest increasing focus on the medical home has improved parental perceptions of PCPs, and may inform discussions on delineation of physician roles.

Provider accessibility and knowledge of a patient's medical history are significant measures of consumer satisfaction and primary care quality, particularly among parents of children with chronic illnesses [4, 8, 14, 19]. One measure that may be related to accessibility, the time it takes to travel to a provider's clinic, was not statistically significant, suggesting that trust in the provider and ease of communication may be more important than matters of convenience.

Fig. 1 Parent-reported care coordination between primary care physician and cardiologist (N = 307)

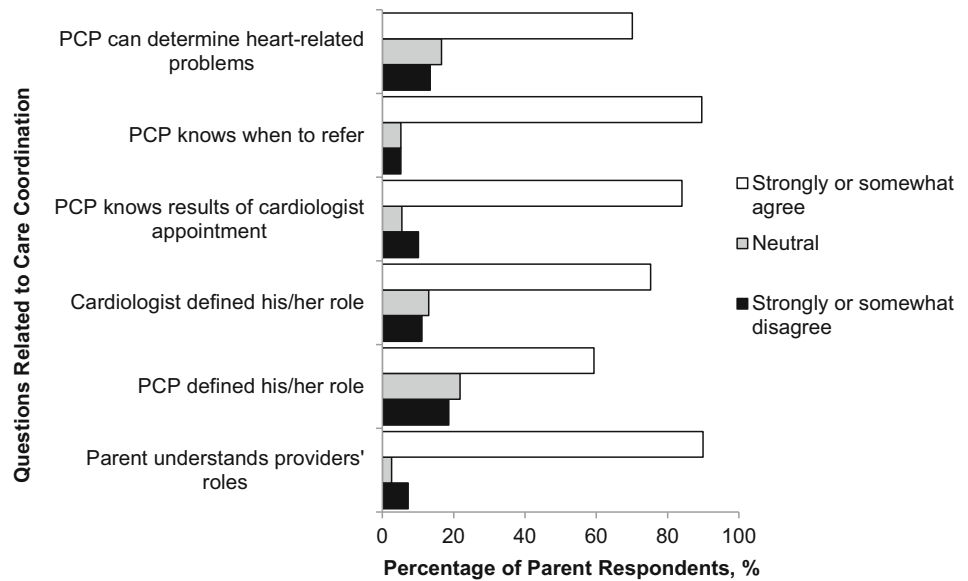
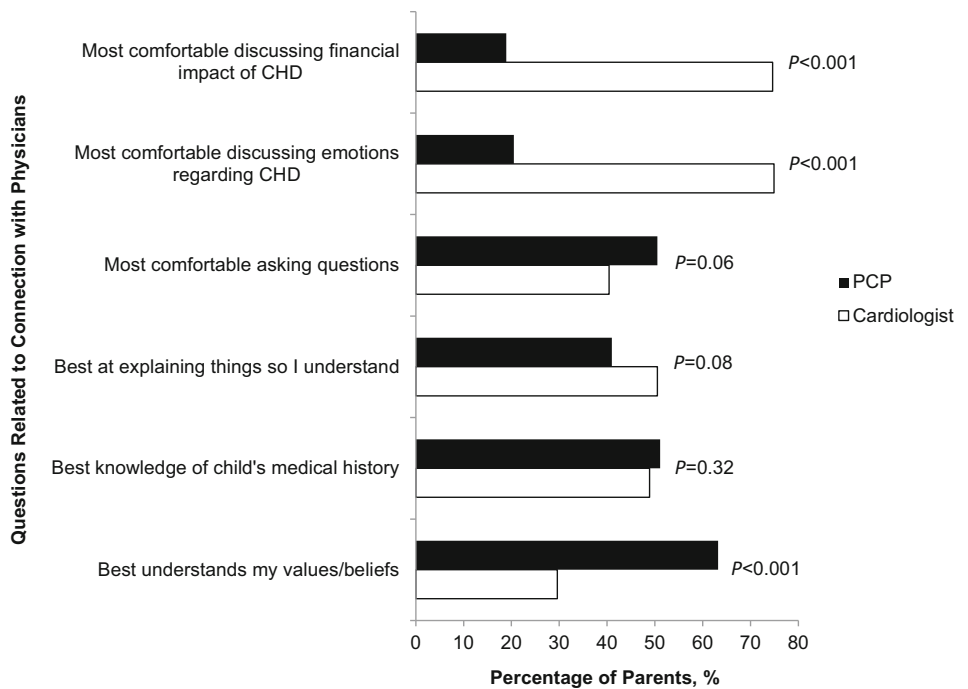


Fig. 2 Parent-reported connection with primary care physician versus cardiologist (N = 307)

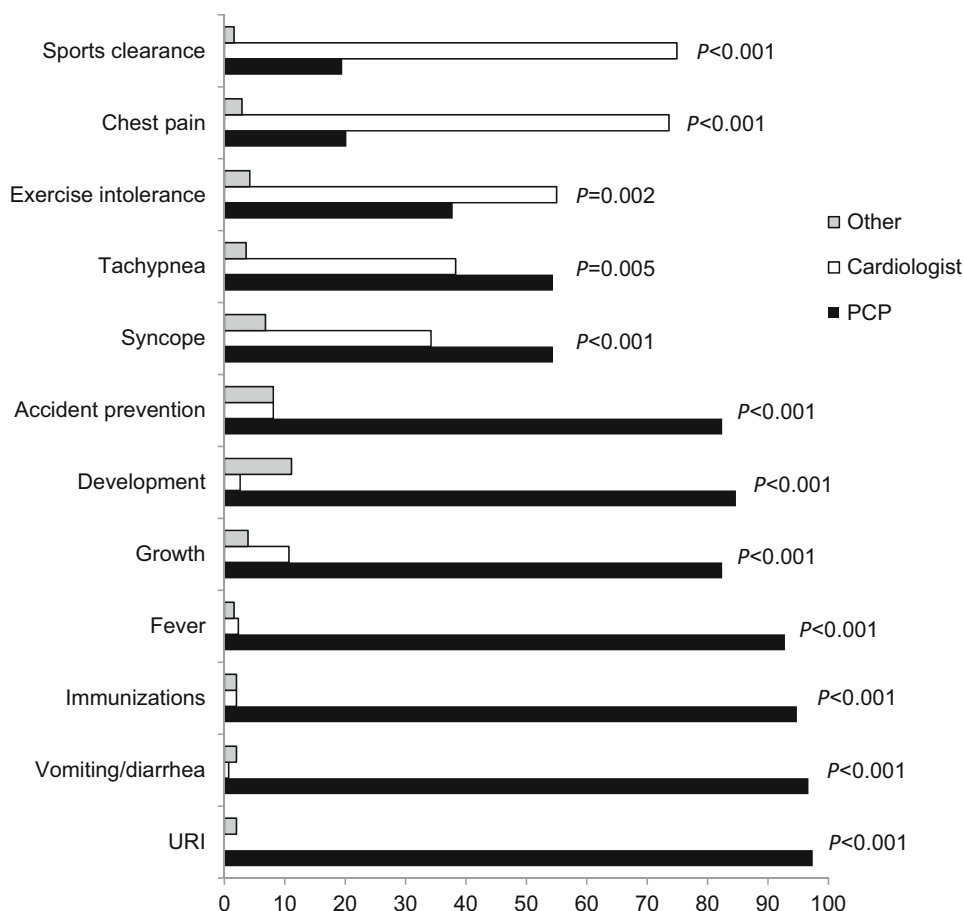


In prior studies, CHD severity and involvement of other subspecialists in a child’s care predicted increased cardiologist preference [13, 24, 25]. However, among the variables that may indicate more severe CHD or poorer overall health, only the number of cardiac catheterizations was statistically significant in multivariable analysis. It is possible that the number of catheterizations better reflected severity than measures such as the number of surgeries or hospitalizations. Cardiac catheterizations are performed by cardiologists and can be either an invasive diagnostic procedure or an intervention. Multiple catheterizations may

imply more severe CHD, as patients may need to tolerate more invasive tests, or undergo multiple interventions, due to complications or persistent disease. This may lead to a stronger clinical relationship with the cardiologist, or reflect a child with residual effects from CHD that require closer monitoring by a cardiologist. Multiple surgeries, on the other hand, may strengthen relationships with the surgeon rather than the cardiologist.

Differences between our results and those from previous studies may reflect differences in our study populations. Unlike previous studies on preferences of parents of

Fig. 3 Parents' preferred providers for evaluation of 12 health-related concerns ($N = 307$)



children with CHD [25], we did not exclude potential subjects based on the severity of CHD or the number of times they had been seen by their cardiologists. By including children with mild CHD that has not required surgical repair and who require less than annual cardiology follow-up, subjects in our study could be more inclined to seek the majority of their care from PCPs. However, the population responding the survey had more severe CHD than non-responders, suggesting there may be a true increase in preference for first contacting the PCP.

Unlike previous studies [13], we included children with other medical conditions, a growing cohort in this population, which may have increased the likelihood that our subjects would seek care from PCPs. Perrin et al. showed children with chronic diseases with higher medical morbidity scores received the majority of their care from generalists, whereas their peers with lower morbidity scores received most of their care from subspecialists [16]. As the physician coordinating care, PCPs may have the greatest awareness of multiple concurrent medical issues and how best to triage concerns, thus leading to greater preference for PCP management. This hypothesis is supported by our finding that having the greatest knowledge of a child's

medical history was associated with provider preference. Alternatively, managing multiple medical issues may cause the PCP to develop a more substantive relationship with the child/parents relative to other providers in a way that was not captured among the other variables.

Variables that were not significantly associated with provider preference may still influence patient outcomes and quality of care. For example, the duration of the relationship with a provider and factors related to the family-centered nature of care (e.g., understanding families' values and beliefs, explaining things in a way parents understand, or whether they feel comfortable asking providers questions) were not statistically significant, although they are used to assess primary care quality [4, 5, 19]. These qualities may also be valued more in situations not assessed in our questionnaire, such as the long-term management of a condition. Moreover, some factors may be essential elements of patient care for health-care providers. Although providers' definitions of their roles were not a significant factor associated with cardiologist preference among parents in our survey, PCPs have identified definition of roles as an important aspect of co-managing care of children with chronic illnesses with subspecialty providers [11].

Table 3 Predictors of increased cardiologist preference in univariate analysis

	OR (95 % CI)	P value
Demographic characteristics		
Public insurance	2.0 (1.0, 3.9)	0.049
Single ventricle	3.1 (1.5, 6.8)	0.002
Number of cardiac surgeries		0.002
0	1.0	
1	0.7 (0.4, 1.4)	
≥2	2.1 (1.0, 4.1)	
Number of cardiac catheterizations		<0.001
0	1.0	
1	2.4 (1.4, 4.2)	
≥2	2.9 (1.7, 5.3)	
Total lifetime hospitalizations		0.01
0 or 1	1.0	
≥2	2.1 (1.3, 3.5)	
Perception of health		
My child is as healthy as other kids	0.8 (0.7, 0.9)	0.005
When ill, he/she can be treated like other kids	0.8 (0.7, 1.0)	0.03
How severe do you think your child’s CHD is now?	1.2 (1.3, 2.3)	<0.001
Connection with cardiologist versus PCP		
Understands my values/beliefs	5.4 (3.0, 10.3)	<0.001
Best knowledge of my child’s medical history	3.6 (2.2, 5.9)	<0.001
Best explains things to me	2.7 (1.7, 4.4)	<0.001
Most comfortable asking questions	3.9 (2.3, 6.6)	<0.001
Comfortable discussing emotions regarding CHD	2.1 (1.2, 3.7)	0.01
Easier to call or see cardiologist	5.9 (3.3, 10.7)	<0.001

CI confidence interval, OR (unadjusted) odds ratio

Table 4 Predictors of cardiologist preference in multivariable analysis

	AOR (95 % CI)	P value
Number of cardiac catheterizations		0.02
0	1.0	
1	2.3 (1.2, 4.5)	
≥2	2.4 (1.1, 4.9)	
Connection with cardiologist versus PCP		
Best knowledge of my child’s medical history	2.4 (1.3, 4.6)	0.006
Easier to call or see cardiologist	3.6 (1.8, 7.4)	<0.001

AOR adjusted odds ratio, CI confidence interval

Our study highlights several potential areas for future research, particularly in the area of care coordination. Although 89.9 % of parents reported that they understood what roles individual medical providers played in their children’s care, only 59.3 and 75.2 % of parents agreed that PCPs and cardiologists had clarified their respective roles (Fig. 1). Given that physicians have identified definition of roles as an important aspect of co-managing care [11], further investigation is needed to determine whether parents’ understanding of roles is correct and whether role definition may help parents/patients and health-care

providers coordinate management. Only 70.0 % of parents in our survey felt PCPs were able to identify whether their children were experiencing heart-related problems and 84.0 % felt PCPs knew the results of their child’s last cardiology evaluation. We did not survey cardiologists or PCPs in this study; however, doing so may help identify unmet educational and communication needs that would improve diagnosis and sharing of information among providers.

There are several limitations to this study. First, this study was conducted at a single academic, tertiary care

center among patients who are predominantly Caucasian with private insurance. Although the demographic characteristics of our subjects are representative of our patient base, our results may not be generalizable to other clinical settings and populations. Second, our response rate was only 38.4 %. Although the demographic characteristics of study participants were representative of our patient population, participants may differ from non-participants in ways we were unable to quantify. Third, parents' responses regarding their children's medical history and health-care usage are subject to recall bias, although we limited our questions regarding health-care use to services received within the last year to minimize this effect. Fourth, the nature of this survey addressed parent-reported preferences, which may differ from true usage. Additional study is necessary to track patients' actual usage of the medical system and to target changes in education and best practices. Finally, our study did not assess whether preferences were appropriate to a child's medical condition. For example, some children may be unlikely to experience chest pain or tachypnea based on their CHD lesion, and initial evaluation may best be conducted by the PCP.

Conclusion

In this study of parents of over 300 children with CHD, parents expressed a high degree of confidence in their children's PCPs, who were more accessible than their cardiologists and had a better understanding of their values and beliefs. Further, they were confident in their PCPs' abilities to manage many medical issues, including some concerns that may be referable to the cardiovascular system. Provider accessibility, the belief that the cardiologist has the best knowledge of the child's medical history, and the number of prior cardiac catheterizations were independently associated with increased preference for cardiologist management. Further research is needed to identify methods for improving coordination of care and communication among subspecialists and generalists who care for children with chronic medical conditions in order to develop effective collaborative care models.

Compliance with Ethical Standards

Conflict of interest The authors declare that they have no conflict of interest.

References

- Berry JG, Hall M, Hall DE, Kuo DZ, Cohen E, Agrawal R, Mandl KD, Clifton H, Neff J (2013) Inpatient growth and resource use in 28 children's hospitals: a longitudinal, multi-institutional study. *JAMA pediatrics* 167(2):170–177. doi:[10.1001/jamapediatrics.2013.432](https://doi.org/10.1001/jamapediatrics.2013.432)
- Boudreau AA, Perrin JM, Goodman E, Kurowski D, Cooley WC, Kuhlthau K (2014) Care coordination and unmet specialty care among children with special health care needs. *Pediatrics* 133(6):1046–1053. doi:[10.1542/peds.2013-2174](https://doi.org/10.1542/peds.2013-2174)
- Burns KH, Casey PH, Lyle RE, Bird TM, Fussell JJ, Robbins JM (2010) Increasing prevalence of medically complex children in US hospitals. *Pediatrics* 126(4):638–646. doi:[10.1542/peds.2009-1658](https://doi.org/10.1542/peds.2009-1658)
- Cassady CE, Starfield B, Hurtado MP, Berk RA, Nanda JP, Friedenber LA (2000) Measuring consumer experiences with primary care. *Pediatrics* 105(4 Pt 2):998–1003
- Chen AY, Schrager SM, Mangione-Smith R (2012) Quality measures for primary care of complex pediatric patients. *Pediatrics* 129(3):433–445. doi:[10.1542/peds.2011-0026](https://doi.org/10.1542/peds.2011-0026)
- Cohen E, Berry JG, Camacho X, Anderson G, Wodchis W, Guttmann A (2012) Patterns and costs of health care use of children with medical complexity. *Pediatrics* 130(6):e1463–e1470. doi:[10.1542/peds.2012-0175](https://doi.org/10.1542/peds.2012-0175)
- Feudtner C, Christakis DA, Connell FA (2000) Pediatric deaths attributable to complex chronic conditions: a population-based study of Washington State, 1980–1997. *Pediatrics* 106(1 Pt 2):205–209
- Garwick AW, Kohrman C, Wolman C, Blum RW (1998) Families' recommendations for improving services for children with chronic conditions. *Arch Pediatr Adolesc Med* 152(5):440–448
- Institute of Medicine Committee on the Future of Primary Care (1996) Defining Primary Care. In: Donaldson MS, Yordy KD, Lohr KN, Vanselow NA (eds) Primary care: America's health in a new era. National Academies Press, National Academy of Sciences, Washington
- Kuhlthau K, Ferris TG, Beal AC, Gortmaker SL, Perrin JM (2001) Who cares for medicaid-enrolled children with chronic conditions? *Pediatrics* 108(4):906–912
- McClain MR, Cooley WC, Keirns T, Smith A (2014) A survey of the preferences of primary care physicians regarding the comanagement with specialists of children with rare or complex conditions. *Clin Pediatr (Phila)* 53(6):566–570. doi:[10.1177/0009922814528035](https://doi.org/10.1177/0009922814528035)
- McPherson M, Arango P, Fox H, Lauver C, McManus M, Newacheck PW, Perrin JM, Shonkoff JP, Strickland B (1998) A new definition of children with special health care needs. *Pediatrics* 102(1 Pt 1):137–140
- Miller MR, Forrest CB, Kan JS (2000) Parental preferences for primary and specialty care collaboration in the management of teenagers with congenital heart disease. *Pediatrics* 106(2 Pt 1):264–269
- Miller AR, Condin CJ, McKellin WH, Shaw N, Klassen AF, Sheps S (2009) Continuity of care for children with complex chronic health conditions: parents' perspectives. *BMC Health Serv Res* 9:242. doi:[10.1186/1472-6963-9-242](https://doi.org/10.1186/1472-6963-9-242)
- Newacheck PW, Kim SE (2005) A national profile of health care utilization and expenditures for children with special health care needs. *Arch Pediatr Adolesc Med* 159(1):10–17. doi:[10.1001/archpedi.159.1.10](https://doi.org/10.1001/archpedi.159.1.10)
- Perrin JM, Kuhlthau KA, Gortmaker SL, Beal AC, Ferris TG (2002) Generalist and subspecialist care for children with chronic conditions. *Ambul Pediatr* 2(6):462–469
- Porterfield SL, DeRigne L (2011) Medical home and out-of-pocket medical costs for children with special health care needs. *Pediatrics* 128(5):892–900. doi:[10.1542/peds.2010-1307](https://doi.org/10.1542/peds.2010-1307)
- Raphael JL, Zhang Y, Liu H, Tapia CD, Giardino AP (2009) Association of medical home care and disparities in emergency care utilization among children with special health care needs. *Acad Pediatr* 9(4):242–248. doi:[10.1016/j.acap.2009.05.002](https://doi.org/10.1016/j.acap.2009.05.002)

19. Seid M, Varni JW, Bermudez LO, Zivkovic M, Far MD, Nelson M, Kurtin PS (2001) Parents' Perceptions of Primary Care: measuring parents' experiences of pediatric primary care quality. *Pediatrics* 108(2):264–270
20. Simon TD, Berry J, Feudtner C, Stone BL, Sheng X, Bratton SL, Dean JM, Srivastava R (2010) Children with complex chronic conditions in inpatient hospital settings in the United States. *Pediatrics* 126(4):647–655. doi:[10.1542/peds.2009-3266](https://doi.org/10.1542/peds.2009-3266)
21. Stein RE, Bauman LJ, Westbrook LE, Coupey SM, Ireys HT (1993) Framework for identifying children who have chronic conditions: the case for a new definition. *J Pediatr* 122(3):342–347
22. Stille CJ, Primack WA, Savageau JA (2003) Generalist-subspecialist communication for children with chronic conditions: a regional physician survey. *Pediatrics* 112(6 Pt 1):1314–1320
23. Varni JW, Burwinkle TM, Seid M, Skarr D (2003) The PedsQL 4.0 as a pediatric population health measure: feasibility, reliability, and validity. *Ambul Pediatr* 3(6):329–341
24. Westbom L, Kornfalt R (1991) Utilization of primary care versus specialized care in children with and without chronic illness. A population-based study. *Acta Paediatr Scand* 80(5):534–541
25. Young PC, Shyr Y, Schork MA (1994) The role of the primary care physician in the care of children with serious heart disease. *Pediatrics* 94(3):284–290