Received: 13 December 2018 | Revised: 4 March 2019 | Accepted: 21 April 2019

DOI: 10.1111/chd.12789

ORIGINAL ARTICLE

Congenital Heart Disease WILEY

Ancillary referral patterns in infants after initial assessment in a cardiac developmental outcomes clinic

Sonia A. Monteiro MD[1] | Faridis Serrano PhD[1] | Rocky Tsang MD[1] | Eboni Smith Hollier MD[1] | Danielle Guffey MS[2] | Lisa Noll PhD[1] | Robert G. Voigt MD[1] | Nancy Ghanayem MD[1] | Lara Shekerdemian MD[1]

[1]Department of Pediatrics, Baylor College of Medicine, Houston, Texas

[2]Dan L Duncan Institute for Clinical and Translational Research, Baylor College of Medicine, Houston, Texas

Correspondence
Sonia A. Monteiro, MD, Department of Pediatrics, Baylor College of Medicine 8080 North Stadium Drive, Houston 77054, TX.
Email: samontei@texaschildrens.org

Abstract

Objective: Neurodevelopmental impairment is common after surgery for congenital heart disease (CHD) in infancy. While neurodevelopmental follow-up of high-risk patients has increased, the referral patterns for ancillary services following initial evaluation have not been reported. The aim of this study is to describe the rates and patterns of referral at the initial visit to our outcomes clinic of patients who underwent surgery for CHD during infancy.

Outcomes Measures: The Cardiac Developmental Outcomes Program clinic at Texas Children's Hospital provides routine longitudinal follow-up with developmental pediatricians and child psychologists for children who required surgery for CHD within the first 3 months of life. Demographic, diagnostic, and clinical data, including prior receipt of intervention and referral patterns at initial presentation, were abstracted from our database.

Results: Between April 2013 and May 2017, 244 infants under 12 months of age presented for initial evaluation at a mean age of 7 ± 1.3 months. At presentation, 31% (76/244) were referred for either therapeutic intervention (early intervention or private therapies), ancillary medical services, or both. Referral rates for low-risk (STAT 1-3) and high-risk (STAT 4-5) infants were similar (28 vs. 33%, $P = .48$). Referrals were more common in: Hispanic white infants ($P = .012$), infants with non-cardiac congenital anomalies ($P = .001$), history of gastrostomy tube placement ($P < .001$), and infants with prior therapy ($P = .043$). Infants of non-English speaking parents were three times more likely to be referred (95% CI = 1.5, 6.4; $P = .002$).

Conclusions: At the time of presentation, nearly 1 in 3 infants required referral. Referral patterns did not vary by traditional risk stratification. Sociodemographic factors and co-morbid medical conditions increased the likelihood of referral. This supports the need for routine follow-up for all post-surgical infants regardless of level of surgical complexity. Further research into the completion of referrals and long-term referral patterns is needed.

KEYWORDS

ancillary services, CHD, early intervention, neurodevelopmental outcome, referral

1 | INTRODUCTION

It has been estimated that up to one-half of all children with congenital heart disease (CHD) may have subsequent, usually subtle, neurodevelopmental impairment. While the neurodevelopmental and academic achievement outcomes for these children at both preschool and school ages are not related to the underlying cardiac anatomy or type of surgery,[1,2] children with a history of CHD are 50% more likely to require special education services than those without CHD.[3] Studies performed in the general population report that only 10% of children between 9 and 24 months of age with developmental delay receive early intervention and therapeutic services, and less than 30% receive interventions prior to school entry.[4,5] Milder neurodevelopmental deficits often seen in children with CHD may not be obvious and therefore may be under recognized by parents, pediatric cardiologists, or primary pediatric health care providers. In the absence of targeted screening, many children who are affected may not receive adequate interventions prior to beginning school.

Developmental follow-up programs for children with CHD can play a vital role in the timely identification of subtle developmental delays and neurodevelopmental disorders at an early age. In turn, this can provide an opportunity for early referral for developmental services, such as "early intervention" (EI), speech and language therapy, physical therapy, or occupational therapy, to improve ultimate neurodevelopmental outcomes. In 2012, the American Heart Association (AHA) published guidelines recommending that all high-risk infants and children with CHD undergo formal developmental evaluation and be referred for early intervention.[6] Additionally, these guidelines outlined the need for scheduled longitudinal neurodevelopmental re-evaluation and recommended that referrals for additional interventions and therapies be made when a developmental disorder was identified. A recent survey regarding the implementation of the 2012 AHA guidelines among primary care providers reported that only 21% of participants were aware of the guidelines.[7] Most referrals for a developmental evaluation made by this group of providers occurred after a failed routine childhood screening test or on the basis of a self-reported concern from a parent. Only 22% of referrals were made proactively due to a history of heart surgery.

To date, the frequency and nature of referrals for infants routinely followed in neurodevelopmental clinics specific to CHD have not been comprehensively described. The aim of this study is to describe the frequency and referral patterns at the initial visit to our dedicated cardiac neurodevelopmental outcomes clinic and explore the association of developmental and ancillary medical needs with demographic and diagnostic factors.

2 | METHODS

2.1 | Patients

The Cardiac Developmental Outcomes Program (CDOP) clinic at Texas Children's Hospital was launched in April 2013. Our program was developed to offer routine longitudinal neurodevelopmental follow-up by board-certified developmental behavioral pediatricians and child psychologists for all infants who undergo surgery for CHD during the first 3 months of life (primary clinic cohort). The clinic population does not include medically complex children with significant medical needs (eg, those with tracheostomy, major congenital syndromes, or major chromosomal anomalies known to be associated with neurodevelopmental impairment). The CDOP clinic also provides neurodevelopmental evaluations and longitudinal monitoring for "ad hoc" referrals of children and adolescents with CHD and neurodevelopmental concerns from pediatric cardiologists and other providers, but these patients will not be discussed further. Patients in the primary clinic cohort all receive an inpatient consultation by a developmental behavioral pediatrician prior to hospital discharge and are then scheduled for their first CDOP clinic appointment at 6 months of age, which is the visit of interest for this study. The timing of the first visit is with the consideration that a significant proportion of these patients require prolonged hospitalization or repeat hospitalization for staged surgery, within the first 6 months of life. However, infants exhibiting neurodevelopmental concerns at the time of their inpatient consultations are referred to early intervention or private therapeutic services prior to hospital discharge, so that interventions can be accessed prior to the initial clinic visit at 6 months of age. Additionally, the program has a dedicated Institutional Review Board approved database to serve as a platform for research.

2.2 | Neurodevelopmental tests

At the first CDOP clinic visit at around 6 months of age, each infant undergoes a comprehensive clinical and neurological examination and standardized developmental testing using the Capute Scales performed by a developmental behavioral pediatrician. The Capute Scales consist of the Clinical Linguistic and Auditory Milestone Scale (CLAMS) and the Cognitive Adaptive Test (CAT).[8] The CLAMS evaluates receptive and expressive language, while the CAT evaluates nonverbal visual motor problem solving. The CAT and the CLAMS each provide a developmental quotient (DQ). The DQ is calculated by dividing the infant's age equivalent in months from the neurodevelopmental evaluation by their chronologic age and multiplying by 100. Infants are considered to be at risk or suspect if their DQ falls between 75 and 85. A score in this range would warrant further close longitudinal monitoring and referral in some cases. A DQ < 75 is considered delayed and would necessitate referral to early intervention or private therapeutic services. For some infants falling in the at-risk or suspect range, referrals for services may be turned down by families or deferred until the follow-up visit. In addition, to qualify for early intervention programs in the state of Texas, infants and toddlers less than 18 months of age must have development that is 25% delayed (equivalent to a DQ < 75) compared to their corrected age. Some infants with additional medical diagnoses may be eligible for interventions, even if they do not meet this 25% delay cutoff, including those with a known genetic syndrome and those with gastrostomy tubes requiring feeding therapy.

2.3 | Referrals for additional services

Referrals for additional developmental interventions, including state-funded birth to 3 years early intervention (EI) programming and private therapies (speech, occupational, and/or physical) as well as ancillary referrals to other medical services (eg, Audiology, Ophthalmology, and Gastroenterology) are made by the developmental behavioral pediatrician during CDOP clinic visits. Early intervention programming has a family-centered approach and involves the development of an Individualized Family Service Plan to create goals and monitor progress. Possible interventions include speech/language, occupational, and/or physical therapy, as well as services provided by an early childhood special educator, who provides parental support and works together with parents and caregivers in the home setting to incorporate developmentally stimulating activities into their everyday routines and daily activities to promote each child's development. Referrals for ancillary medical services are made when specific concerns are reported by parents during the visit or when the neurodevelopmental evaluation and physical examination elicit concerns that require further subspecialty medical evaluation, including concerns about vision or hearing.

2.4 | Data collection

Demographic, diagnostic, and clinical data relating to the infants' initial hospitalization and developmental evaluations completed in the CDOP clinic visit are maintained in our IRB approved database. Data related to primary hospitalization include the cardiac diagnosis and STAT (The Society of Thoracic Surgeon-European Association for Cardiothoracic Surgery) category of surgical complexity. Each surgical procedure is designated a STAT category of 1-5 with the surgery carrying the highest risk of mortality receiving a score of 5.[9] Data specific to developmental follow-up include age at initial clinic visit and referrals to developmental (eg, early intervention (EI) programs and/or private physical therapy, occupational therapy, and/or speech/language therapy) and ancillary medical (eg, audiology, dietician, and ophthalmology) services. Additionally, information relating to ongoing or previous developmental interventions and private therapies prior to the first developmental clinic visit is also recorded at the first clinic visit.

2.5 | Analysis

Patient and clinical characteristics were summarized using mean with standard deviation, median with 25th and 75th percentiles, and frequency with percentage as appropriate. The summary statistics were stratified by referral and prior therapy and compared using two-sample *t* test, namely Wilcoxon rank sum test and Fisher's exact test or Chi-square test. Independent logistic regression is used to assess the association between characteristics and the odds of referral. Statistically significant ($P < .05$) characteristics are combined in a multivariable logistic regression model. All statistical analyses were performed using Stata version 15.1 (Stata Corp, College Station, TX).

3 | RESULTS

Between April 2013 and May 2017, 244 infants who underwent surgery for CHD during the first 3 months of life presented to the CDOP Clinic for their first assessment. The mean age at the first clinic visit was 7 ± 1.3 months. Infants were predominantly male (58%). At the time of their initial evaluation, 51 (21%) infants were previously receiving or had been evaluated for EI, private therapies, or ancillary services. Of the infants who were already receiving prior interventions, 22 (43%) required referrals for additional services, compared with 28% of those without prior referrals ($P = .043$). Figure 1 illustrates the breakdown of the new referrals to either EI, private therapy, and/or ancillary medical services in the subset of infants who had prior interventions. Infants with additional congenital anomalies were more likely to have received additional services (O.R. 2.6; 95% CI = 1.3, 4.9; $P = .004$) as well as those with a gastrostomy tube (O.R. 8.2; 95% CI = 3.6, 18.7; $P < .001$).

At the completion of their initial neurodevelopmental evaluation by a developmental behavioral pediatrician, a referral for one or more of EI, private therapy, and/or ancillary medical services was placed for 76 (31%) infants (Figure 2). Thirty-two (42%) of the referrals were for EI alone, and 52 (68%) were referred to EI and a combination of other services.

In total, there were 118 referrals in 76 infants following the initial evaluation in our clinic. Specific referrals are detailed in Table 1. Of the 118 referrals, 52 (44%) were to EI and 21% were to private therapies, including physical, occupational, and/or speech therapies. In addition, there were 41 referrals (35%) made to 9 different ancillary medical services, including audiology, medical genetics, physical medicine and rehabilitation, gastroenterology, ophthalmology, neurology, endocrinology, neurosurgery, and ENT.

The referral patterns for additional services of CDOP patients, based on infant characteristics are detailed in Table 2. There was

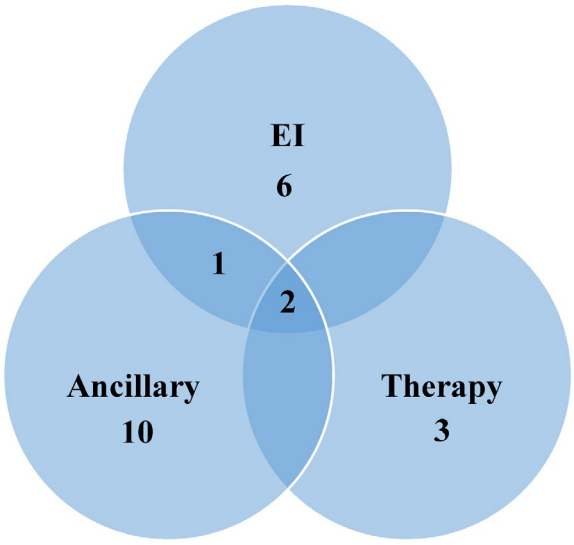

FIGURE 1 Venn diagram showing the subset of infants who had prior therapies ($n = 51$) but required additional referrals to early intervention (EI), therapy, and/or ancillary services

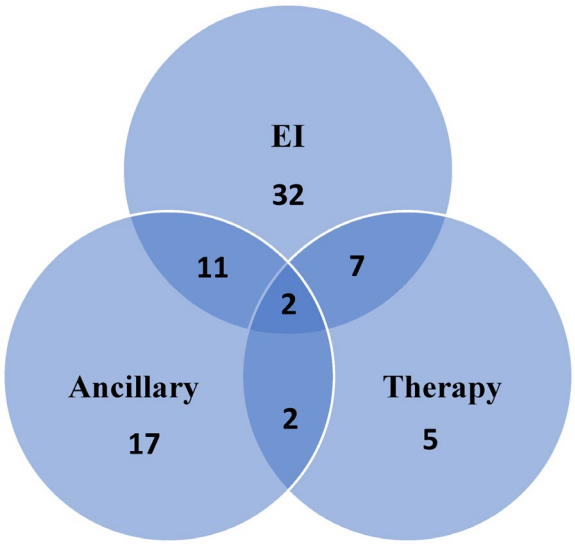

FIGURE 2 Venn diagram illustrating number of infants referred (*n* = 76) during their initial neurodevelopmental assessment in our cardiac developmental outcomes clinic to early intervention (EI), therapy, and ancillary services

TABLE 1 Breakdown of EI, therapy, and ancillary services referrals following initial neurodevelopmental evaluation of 76 infants in our CDOP clinic

Referrals	*N*
EI	52
PT-Speech	4
PT-Occupational	9
PT-Physical	12
Audiology	11
Medical genetics	9
Ophthalmology	4
Neurosurgery	4
Swallow study	4
Gastro & nutrition	3
Endocrinology	3
Physical medicine & rehab	2
ENT	1
Sleep medicine	0
Neurology	0
Total	118

Abbreviations: EI, early intervention; ENT, ear, nose and throat; PT, private therapy.

no difference in median STAT category between those infants who were and were not referred (*P* = .47). Referred patients were more likely to have a non-English speaking parent, to have at least one non-cardiac congenital anomaly, to have a gastrostomy tube, and to have been already receiving therapy prior to the initial clinic appointment. These characteristics were included in a multivariable logistic regression model, and in this model, infants with non-English speaking parents (O.R. 3.1; 95% CI = 1.5, 6.4; *P* = .002), a non-cardiac congenital anomaly (O.R. 2.5; 95% CI = 1.4, 4.7; *P* = .003), and a gastrostomy tube (O.R. 4.3; 95% CI = 1.8, 9.9; *P* = .001) were more likely to be referred for additional services. Receipt of prior therapies was no longer significantly associated with referral for additional services in the multivariable analysis.

4 | DISCUSSION

This report is the first to delineate referral patterns at initial presentation for neurodevelopmental assessment in the first year of life for infants who have undergone cardiac surgery. At the time of presentation, nearly one in three infants required referrals following evaluation in our dedicated cardiac developmental outcomes clinic. Referral patterns did not vary by mortality risk (STAT category). Thus, our study provides evidence supporting the routine follow-up of all post-surgical infants with CHD regardless of level of surgical complexity.

We were encouraged that 21% of infants seen at CDOP were already receiving early intervention or private therapeutic services at the time of their first developmental clinic visit. This observation suggests that primary pediatric health care providers are identifying some children with a history of CHD and referring them to appropriate interventions. Implementation of the 2006 American Academy of Pediatrics policy recommending developmental surveillance during all primary care encounters has increased identification of all children at risk, including children with CHD.[10,11] As primary pediatric health care providers (PCP) become more familiar with the American Heart Association guidelines specific to children with CHD, the frequency of referrals made by primary care providers should continue to increase. Many parents of infants with CHD prefer to contact their PCP first for health-related concerns, including those regarding development[12] and rely on their PCP to assist in securing needed interventions. Of note, receipt of early intervention or private developmental therapies prior to initial formal developmental testing at the CDOP clinic did not negate the need for additional referrals for other therapies or ancillary medical services in nearly half of those infants. Thus, developmental follow-up programs can provide needed assistance in coordination of care and ensuring that needs do not go unmet or overlooked.

Many children with CHD have additional chronic medical conditions, with the rate of non-cardiac comorbidities reported to be anywhere from 20% to 50%.[13,14] Comorbid medical conditions result in increased resource utilization following hospital discharge.[15] In our sample, infants with a history of a congenital anomalies were more likely to have received developmental interventions prior to their first clinic visit and were more likely to be referred for further interventions. Our findings support the need for close follow-up for infants with congenital anomalies. Additional factors increasing the likelihood of referral included gastrostomy tube placement and

TABLE 2 Referral patterns by infant characteristics

Variable	No Referral (*N* = 168)			Referral (*N* = 76)			*P* value
Maternal age, yrs	*N*	Mean	(SD)	*N*	Mean	(SD)	
	144	28.9	(6.0)	73	29.3	(6.0)	.646
Gestational age	*N*	Median	(IQR)	*N*	Median	(IQR)	
	168	38	(37.0, 39.0)	76	38	(37.0, 39.0)	.786
Age at first visit, days	168	6.7	(6.2, 7.5)	76	6.7	(6.1, 7.8)	.987
Age at initial surgery, days	168	0.4	(0.4, 1.4)	76	0.5	(0.3, 1.8)	.121
STAT category	168	4	(2.0, 4.0)	76	4	(2.0, 4.0)	.471
	N total	*N*	(%)	*N* total	*N*	(%)	
Gender	168			76			1.000
Male		98	(58.3)		44	(57.9)	
Female		70	(41.7)		32	(42.1)	
Language	168			76			.010
English speaking		147	(87.5)		56	(73.7)	
Non-English speaking		21	(12.5)		20	(26.3)	
Race/ethnicity	168			76			.125
Non-Hispanic White		77	(45.8)		26	(34.2)	
Hispanic White		61	(36.3)		41	(53.9)	
Black		16	(9.5)		6	(7.9)	
Asian		11	(6.5)		3	(3.9)	
Other		3	(1.8)		0	(0.0)	
Insurance type	168			76			.067
No insurance		1	(0.6)		2	(2.6)	
Public		79	(47.0)		45	(59.2)	
Private		88	(52.4)		29	(38.2)	
Congenital anomalies	168			76			.001
No		131	(78.0)		43	(56.6)	
Yes		37	(22.0)		33	(43.4)	
Prior therapy	168			76			.043
No		139	(82.7)		54	(71.1)	
Yes		29	(17.3)		22	(28.9)	
G Tube	168			76			<.001
No		157	(93.5)		57	(75.0)	
Yes		11	(6.5)		19	(25.0)	
Stat category	168			76			.480
1-3		69	(41.1)		27	(35.5)	
4-5		99	(58.9)		49	(64.5)	

Abbreviations: STAT, The Society of Thoracic Surgeon-European Association for Cardiothoracic Surgery category of surgical complexity; Values are *n* (%), mean (SD, standard deviation).
P values were determined by Fisher's exact test or Chi-square test for categorical comparisons, Wilcoxon rank sum test for median comparisons, and two-sample *t* test for mean comparisons.

primary language, and these continued to be significant in the multivariable model while other factors, including history of prior developmental therapy, were no longer significant.

It is important to note that many of the patients in our CDOP clinic return for subsequent follow-up visits, thus providing opportunities for further developmental evaluation and referrals for interventions as indicated. This may speak to the value of providing reassurance and anticipatory guidance to caregivers of children with congenital heart disease as part of a longitudinal program. Such longitudinal follow-up is particularly important, as it has been shown that while

motor deficits are commonly identified in early assessments of children with CHD, cognitive, and language impairments typically present at a later time.[16]

The limitations to our study include the fact that our CDOP clinic represents a routine, elective clinical service that relies upon family engagement, and willingness to attend for assessment and follow-up. Thus, while our clinic attendance rates are good, one cannot say with certainty that the clinic population is absolutely representative of the surgical population. Additionally, the data presented here relate to the initial developmental follow-up clinic visit, and developmental status can clearly change over time, requiring more detailed review at a later stage as the clinic matures further.

This study demonstrates the importance of routine developmental follow-up programs for children with congenital heart disease. Even at a very early age, a significant proportion of infants require additional services to address developmental delays, as well as ancillary medical services. This study also demonstrates the importance of following all children with congenital heart disease regardless of surgical complexity, as no clear cut "high risk" patients can be identified based upon surgical category.

CONFLICT OF INTEREST

The authors declare that they no conflicts of interest with the contents of this article.

AUTHOR CONTRIBUTIONS

Conceptualized and designed the study, collected data, prepared data for analysis, led interpretation of results, prepared manuscript, and approved the final manuscript as submitted: Monteiro

Advised on study design, contributed to drafting manuscript, critically reviewed and revised the manuscript, and approved the final manuscript as submitted: Serrano

Advised on study design, critically reviewed and revised the manuscript, and approved the final manuscript as submitted: Tsang

Contributed to study conception, critically reviewed and revised the manuscript, and approved the final manuscript as submitted: Hollier-Smith

Conducted statistical analyses, contributed to interpretation, and approved the final manuscript as submitted: Danielle Guffey.

Collected data, critically reviewed and revised the manuscript, and approved the final manuscript as submitted: Noll.

Contributed to data interpretation, critically reviewed and revised the manuscript, and approved the final manuscript as submitted: Voigt.

Advised on study design, data interpretation, critically reviewed and revised the manuscript, and approved the final manuscript as submitted: Ghanayem and Shekerdemian.

ORCID

Sonia A. Monteiro https://orcid.org/0000-0003-3835-2637

REFERENCES

1. Brosig CL, Bear L, Allen S, et al. Preschool neurodevelopmental outcomes in children with congenital heart disease. *J Pediatr*. 2017;183:80-86.e1.
2. Mulkey SB, Bai S, Luo C, et al. School-age test proficiency and special education after congenital heart disease surgery in infancy. *J Pediatr*. 2016;178:47-54.e1.
3. Riehle-Colarusso T, Autry A, Razzaghi H, et al. Congenital heart defects and receipt of special education services. *Pediatrics*. 2015;136(3):496-504.
4. Rosenberg SA, Zhang D, Robinson CC. Prevalence of developmental delays and participation in early intervention services for young children. *Pediatrics*. 2008;121(6):e1503-e1509.
5. Silverstein M, Sand N, Glascoe FP, Gupta VB, Tonniges TP, O'Connor KG. Pediatrician practices regarding referral to early intervention services: is an established diagnosis important? *Ambul Pediatr*. 2006;6(2):105-109.
6. Marino BS, Lipkin PH, Newburger JW, et al. Neurodevelopmental outcomes in children with congenital heart disease: evaluation and management: a scientific statement from the American Heart Association. *Circulation*. 2012;126(9):1143-1172.
7. Knutson S, Kelleman MS, Kochilas L. Implementation of developmental screening guidelines for children with congenital heart disease. *J Pediatr*. 2016;176:135-141.e2.
8. Accardo PJ, Capute AJ. *The Capute Scales: Cognitive Adaptive Test/Clinical Linguistic & Auditory Milestone Scale (CAT/CLAMS)*. Baltimore, MD: Paul H. Brookes Publishing Co; 2005.
9. O'Brien SM, Clarke DR, Jacobs JP, et al. An empirically based tool for analyzing mortality associated with congenital heart surgery. *J Thorac Cardiovasc Surg*. 2009;138(5):1139-1153.
10. Pediatrics AAo. *Clinical Practice Guidelines & Policies*. 5th ed. Elk Grove, IL: American Academy of Pediatrics; 2005.
11. Council on Children With D, Section on Developmental Behavioral P, Bright Futures Steering C, Medical Home Initiatives for Children With Special Needs Project Advisory C. Identifying infants and young children with developmental disorders in the medical home: an algorithm for developmental surveillance and screening. *Pediatrics*. 2006;118(1):405-420.
12. Luthy SK, Yu S, Donohue JE, Loccoh E, Goldberg CS, Lu JC. Parental preferences regarding outpatient management of children with congenital heart disease. *Pediatr Cardiol*. 2016;37(1):151-159.
13. Massin MM, Astadicko I, Dessy H. Noncardiac comorbidities of congenital heart disease in children. *Acta Paediatr*. 2007;96(5):753-755.
14. Patel A, Costello JM, Backer CL, et al. Prevalence of noncardiac and genetic abnormalities in neonates undergoing cardiac operations: analysis of the society of thoracic surgeons congenital heart surgery database. *Ann Thorac Surg*. 2016;102(5):1607-1614.
15. Tuomela KE, Gordon JB, Cassidy LD, Johaningsmeir S, Ghanayem NS. Resource utilization associated with extracardiac co-morbid conditions following congenital heart surgery in infancy. *Pediatr Cardiol*. 2017;38(5):1065-1070.
16. Mussatto KA, Hoffmann RG, Hoffman GM, et al. Risk and prevalence of developmental delay in young children with congenital heart disease. *Pediatrics*. 2014;133(3):e570-e577.

How to cite this article: Monteiro SA, Serrano F, Tsang R, et al. Ancillary referral patterns in infants after initial assessment in a cardiac developmental outcomes clinic. *Congenital Heart Disease*. 2019;14:797–802. https://doi.org/10.1111/chd.12789