Congenital Heart Disease

Tech Science Press

DOI: 10.32604/chd.2023.044874

ARTICLE

Loss to Specialized Cardiology Follow-Up in Adults Living with Congenital Heart Disease

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Received: 10 August 2023 Accepted: 27 September 2023 Published: 20 March 2024

ABSTRACT

Background: Much has been written about the loss to follow-up in the transition between pediatric and adult Congenital Heart Disease (CHD) care centers. Much less is understood about the loss to follow-up (LTF) after a successful transition. This is critical too, as patients lost to specialised care are more likely to experience morbidity and premature mortality. **Aims:** To understand the prevalence and reasons for loss to follow-up (LTF) at a large Australian Adult Congenital Heart Disease (ACHD) centre. **Methods:** Patients with moderate or highly complex CHD and gaps in care of >3 years (defined as LTF) were identified from a comprehensive ACHD database. Structured telephone interviews examined current care and barriers to clinic attendance. **Results:** Overall, 407 (22%) of ACHD patients (n = 1842) were LTF. The mean age at LTF was 31 (SD 11.5) years and 54% were male; 311 (76%) were uncontactable. Compared to adults seen regularly, lost patients were younger, with a greater socio-economic disadvantage, and had less complex CHD ($p < 0.05$ for all). We interviewed 59 patients (14%). The top 3 responses for care absences were "feeling well" (61%), losing track of time (36%), and not needing follow-up care (25%). **Conclusions:** A large proportion of the ACHD population becomes lost to specialised cardiac care, even after a successful transition. This Australian study reports younger age, moderate complexity defects, and socio-economic disadvantage as predictive of loss to follow-up. This study highlights the need for novel approaches to patient-centered service delivery even beyond the age of transition and resources to maintain patient engagement within the ACHD service.

KEYWORDS

Loss to follow-up; follow-up; adult congenital heart disease; lapse in care; gaps in care; care gaps; care continuity; ACHD; predictors

Nomenclature

ACHD	Adult Congenital Heart Disease
ADL	Activities of daily living
LTF	Lost to follow-up

1 Introduction

Adults with congenital heart defects experience longer and fuller lives credited to advances in prenatal screening and medical and surgical care. Now more adults live with ACHD than children, and they continue to survive, with complex medical and psychosocial needs [1–4]. An essential part of living longer with ACHD is the requirement for consistent life-long follow-up with specialized clinicians to recognize and address progressive changes associated with cardiac defects [5,6]. The frequency of specialist care generally varies from every one to three years, depending on the ACHD classification.

Loss to follow-up (LTF), also referred to as gaps in care or lapses in specialty care, are absences from a specialized congenital healthcare provider for three or more years [7]. Patients with gaps in their specialist care are more likely to experience higher morbidity and mortality rates than those with regular, guideline-directed care [8,9]. Without continuous specialized care, there is more preventable death, loss of cardiac function, reduced life expectancy, unnecessary intervention, and delays in timely intervention [10–13]. Ultimately these factors lead to unplanned or emergency treatment in hospitals, increased health resource utilization, and poor quality of life [14,15]. Loss to follow-up rates differs between regions ranging from 4% to 63% of people with ACHD internationally, predominantly reported after the transition from paediatric care [16–19]. No studies examine ACHD follow-up experiences post-transition, within a universal health care system, thus highlighting the research gap addressed by this Australian study. In this study, we identify the prevalence, causes, and predictors of gaps in specialist ACHD care in an outpatient cohort. We examine the proportion of patients lost to follow-up, the barriers to care, and the reasons for return to care.

2 Methods

2.1 Design

We conducted a single-centre cohort study of the largest Adult Congenital Heart Disease (ACHD) service, embedded in a tertiary referral centre in New South Wales, Australia.

2.2 Setting

The outpatient clinic receives medical referrals for adults with ACHD, from paediatric and adult generalists and specialists, encompassing a population of 8.16 million people over a geographical area of 801,150 km².

2.3 Participants

All adults registered to the ACHD clinic database with moderately or severely complex anatomy were included in the study. ACHD anatomical complexities were categorized according to the 2018 AHA Guidelines. Researchers examined database attendance records in May 2019 to identify patients who had not attended follow-up at the clinic for three years or more. Follow-up attendance was determined by the frequency of care plans documented in the medical records. Patients were excluded if they were non-English speaking, aged 16 years or younger, intellectually disabled or had ACHD anatomy classified as simple complexity. We also excluded people over 75 years as that group were confounded by other issues such as nursing home placement and difficulty travelling to appointments. This study was approved by the Human Research and Ethics Committee Sydney Local Health District (Protocol X18-0189 HREC/11/RPAH/625). Additionally, all people surveyed provided informed verbal consent at the time of telephone contact.

Loss to follow-up was defined as absence from any form of ACHD cardiology care for greater than 3 years or being unable to be located. Patients were telephoned at various times of the day and evening, across all weekdays, including weekends. Voicemail or text messages were left on valid numbers to locate and engage missing patients. Further hand searching of electronic and paper medical records for alternate contacts was undertaken for patients with invalid contact details.

Patients who gave verbal consent were invited to participate in the survey which was conducted by an experienced clinician not working at the ACHD clinic to reduce response bias. Telephone questionnaires were administered between May 2019 and May 2020 to understand current care, barriers to clinic attendance and to offer specialty care reconnection if desired. Participants without a current ACHD specialist were invited to return to the adult CHD clinic or another appropriate specialist centre. The inclusion process is shown in Fig. 1.

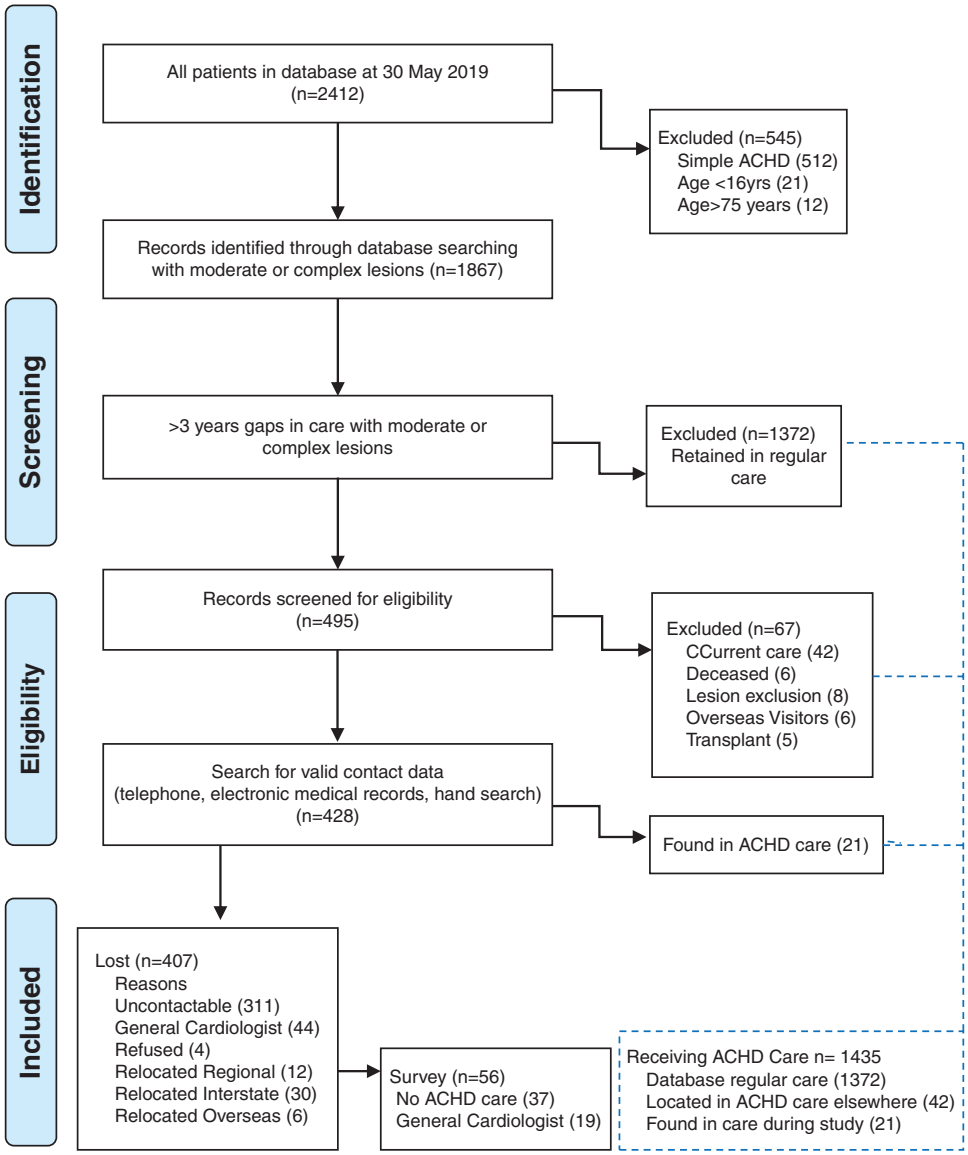

Figure 1: Flowsheet with screening for gaps in care, eligibility and inclusion for survey

2.4 Survey Tool

Data were collected directly from the database, electronic medical record (eMR) and survey tool. Demographic and clinical characteristics of the patients were collected, including gender, age at last clinic review, socio-economic status, geographical location and ACHD anatomy. Socio-economic status was sourced from the Socio-Economic Indexes for Areas (SEIFA) by the Australian Bureau of Statistics that ranks areas in Australia according to relative socio-economic advantage and disadvantage [20]. An area

with a high score on this index has a relatively high incidence of advantage and a relatively low incidence of disadvantage. Geographical remoteness was derived from the Accessibility/Remoteness Index of Australia (ARIA) which categorizes areas according to their distance from "service centers" [21]. ARIA defines five categories of remoteness from very remote, remote, outer regional, inner regional to major city by utilizing postcodes. Remote and very remote categories were condensed.

The survey tool was adapted from the HEART-ACHD trial published by Gurvitz et al. [18], to assess reasons for returning to or abstaining from specialized ACHD care. The self-reported survey of approximately ten (10) minutes duration was administered over the telephone to consenting people. The survey examined demographics (7 fields; gender, nationality, ethnicity, education level, English fluency, language spoken at home, independence in ADL), ACHD care arrangements (5 fields with yes, no or numeric responses); reasons for absence from care (Likert 6 points scale with 19 items). People were asked to rate their reasons for ceasing ACHD care from 5 strongly agree to 1 strongly disagree and 0 as not relevant if no choices were appropriate. Patients who had returned to general cardiology care were asked what prompted this to occur (Likert 6 points scale with 14 items). The results are reported as frequencies.

2.5 Statistical Analysis

Data were analyzed using IBM SPSS Statistics version 27. Data are presented as means and standard deviation (SD) or frequency and proportions. Characteristics of the subjects were compared using *t*-tests for continuous variables if normally distributed, Mann-Whitney if non-normally distributed and categorical data were analyzed using Pearson's $\chi 2$ test and two sample z tests (https://epitools.ausvet.com.au/ztesttwo).

The predictors of loss to follow-up were examined with univariable and multivariable analyses using Cox Hazard regression. The Cox Regression was modelled with the event LTF from the last clinic review date, time as the years in care and covariates included gender, geographical remoteness, socio-economic disadvantage and ACHD lesion. Age was not included as a covariate as it linked to the "event" and forms part of the model and consequently every iteration of analysis shows age as having a very strong relationship. All variables were retained in the model to identify the multivariate predictors.

Qualitative comments from the participant survey were analyzed thematically and patterns reported.

3 Results

In May 2019, a total of 2412 records were extracted from the database. The records were screened to exclude all patients with simple complexity disease or in current care. Four hundred and ninety-five people were identified as being lost to follow-up and are the subjects of this study, as shown in Fig. 1.

3.1 Prevalence of Loss to Follow-Up

Of people with care gaps in our database, 407 were lost to follow-up. This translates to 22% of our cohort.

3.2 Characteristics of Lost to Follow-Up

We compared the patients with LTF to those in current care and found differences between the groups in age, socio-economic status, remoteness and disease complexity (Table 1). Patients LTF were younger (30.9, SD 11.4 years) than those in current care (35.0, SD 12.65 years) based on age at their last clinic visit ($p < 0.001$). There were no differences in gender between the groups; males with LTF and those in current care were 53.8% and 54.5% ($p = 0.87$), respectively. Geographical remoteness scores showed some differences between the groups, with the majority of people in both groups residing close to a major city (80.1 % *vs.* 83.5%, $p = 0.16$). A higher proportion of LTF compared to current care resided in remote/very remote areas (1.4% *vs.* 0.2%, $p = 0.003$). We found that patients LTF had lower relative

socio-economic advantage and disadvantage indices (6.0 SD 2.98) compared to those in the current care group (6.6 SD 2.77, $p = 0.002$). In the moderate ACHD category, more people were LTF than in current care (84% *vs.* 74.8%, $p < 0.001$), while in the severe category, more were in current care than LTF (25.2% *vs.* 16%, $p < 0.001$).

Table 1: Cohort characteristics (n = 1,842)

	Current Care n = 1435 N (%)	LTF n = 407 N (%)	Difference (95% CI)	*p* value
Age at last visit (years) [a]				<0.001
Mean (SD)	35.0 (12.65)	30.9 (11.45)	4.1 (2.75, 5.78)	
Years since last ACHD Visit				<0.001
Mean (SD)[x]	0.5 (1.47)	8.6 (4.03)	−8.06 (−8.46, −7.66)	
Male[b]	778 (54.5%)	219 (53.8%)	0.70% (−4.78, 6.18)	0.866
Socio-Economic Status [c,d]				
Mean (SD)	6.6 (2.77)	6.0 (2.98)	0.60 (0.24, 0.95)	0.002
Remoteness [d]				
Major City	1176 (83.5%)	233 (80.1%)	3.4% (−1.35, 8.15)	0.16
Inner Regional	192 (13.6%)	45 (15.5%)	1.90 (−2.47, 6.27)	0.39
Outer Regional	37 (2.6%)	9 (3.1%)	0.50 (−1.54, 2.54)	0.63
Remote/Very remote	3 (0.2%)	4 (1.4 %)	1.20 (0.40, 2.00)	0.003
ACHD Complexity				
Moderate	1074 (74.8 %)	342 (84 %)	9.2% (4.56, 13.84)	0.001
Severe	361 (25.2 %)	65 (16 %)	9.2% (4.56, 13.84)	0.001

Notes: [a] Cohen's d 0.332. [x] Cohen's d −3.52. [b] Transgender (n = 4). [c] Socio-Economic Status using Index of Relative Socio-economic Advantage and Disadvantage (lowest to highest); Cohen's d 0.212. [d] Available complete sample: Current Care n = 1403, LTF n = 292.

3.3 Predictors of Loss to Follow-Up

Predictors of loss to follow-up in our cohort are detailed in Table 2. The same variables were predictive in both the univariable and multivariable analyses. People who lived in remote/very remote areas were 3.12 (95% CI: 1.15, 8.43) times more likely to be lost to follow-up than those living in a major city. The risk of becoming lost to follow-up reduced by 6% (95% CI: 0.90, 0.98) for every one-point increase in the level of socio-economic advantage ($p = 0.002$). People with moderately complex lesions were 1.46 (95% CI: 1.06, 1.99) times more likely to be LTF than those with severely complex lesions. Table 2 reports results calculated from cases with complete data only to enable comparison between univariate and multivariate analyses. Appendix 1 provides supplementary univariate analysis of all cases, demonstrating consistent predictors of LTF, irrespective of missing data for gender, SES and geographical remoteness.

Table 2: Predictors of Loss to Follow-Up

	Univariable regression		Multivariable regression[a]	
Factors	H.R. (95% CI)	*p*	H.R. (95% CI)	*p*
Gender				
Female	0.98 (0.78–1.23)	0.87	0.96 (0.76, 1.21)	0.750
Male (ref)				

(Continued)

Table 2 (continued)

	Univariable regression		Multivariable regression[a]	
Geographical Remoteness		0.057		0.166
Inner Regional	1.11 (0.81–1.53)	0.51	1. (0.73, 1.39)	0.953
Outer Regional	1.29 (0.66–2.51)	0.46	1. (0.56, 2.16)	0.790
Remote/Very remote	3.75 (1.39–10.08)	0.009	3.12 (1.15, 8.43)	0.025
Major City (ref)				
Socio-Economic Status	0.94 (0.89–0.97)	<0.001	0.94 (0.90, 0.98)	0.002
ACHD Anatomy				
Moderate	1.47 (1.07–2.01)	0.016	1.46 (1.06, 1.99)	0.019
Severe (ref)				

Notes: [a] Adjusted for all other variables in the table.

3.4 Re-Engagement and Survey

Telephoning, digital medical record and hand searches determined the remaining care status and survey inclusions accordingly as shown in Fig. 1. More than 1500 phone calls were placed in search of lost patients and to engage the survey participants, with 3–5 phone attempts required to connect with 75% of people, 4–9 call attempts to reach 23%, and 10–15 call attempts for the remaining 1%. Fifty-six eligible participants were invited to complete the survey. Further analysis was undertaken in the survey sub-group who responded to our phone calls. Call duration averaged 10 minutes, however 39% of calls were longer due to participant comments (20–30 minutes).

3.5 Survey Characteristics

We stratified the survey responses into “no ACHD care” (n = 37) and “general cardiology” (n = 19) summarized in Table 3.

Table 3: Survey characteristics comparison of survey respondants by care type

Factors	No Care (37) N (%)	General Cardiology Care (19) N (%)	*p* value
Age at last visit (years)[a] Mean (SD)	27.5 (8.7)	33.0 (12.4)	0.086
Years since last ACHD Visit Mean (SD)	8.1 (3.9)	8.16 (3.9)	0.964
Males	22 (59.5%)	5 (26.3%)	0.025
Education			0.364
Nil or Incomplete School	2 (5.4%)	2 (10.5 %)	
School	14 (37.8%)	2 (10.5%)	
Certificate	12 (32.4%)	7 (36.8%)	
Graduate Diploma	1 (2.7%)	0 (0%)	
Bachelor Degree	5 (13.5%)	5 (26.3%)	
Advanced Diploma	1 (2.7%)	1 (5.3%)	
Postgraduate Studies	2 (5.4%)	2 (10.5%)	

(Continued)

Table 3 (continued)

Factors	No Care (37) N (%)	General Cardiology Care (19) N (%)	*p* value
English Proficiency			0.291
Not well	0 (0%)	1 (5.3%)	
Well	1 (2.7%)	0 (0%)	
Very well	36 (97.3%)	18 (94.7%)	
Independence[b] Mean (SD)	6.7 (0.93)	6.8 (0.71)	0.791
Socio-Economic Status[c] Mean (SD)	5.69 (2.53)	6.8 (2.83)	0.148
Remoteness			0.945
Major City	30 (83.3%)	16 (84.2%)	
Inner Regional	3 (8.3%)	1 (5.3%)	
Outer Regional	2 (5.6%)	1 (5.3%)	
Remote/Very remote	1 (2.8%)	1 (5.3%)	
Nationality			0.443
Australian	34 (91.9%)	19 (100%)	
New Zealand	2 (5.4%)		
Southern European	1 (2.7%)		
Ethnicity			0.532
Australian-Caucasian	28 (75.7%)	13 (68.4%)	
South East Asian	–	1 (5.3%)	
North East Asian	2 (5.4%)	1 (5.3%)	
Southern European	2 (5.4%)	1 (5.3%)	
Maori	2 (5.4%)	–	
Middle Eastern	3 (8.1%)	2 (10.5%)	
South American	–	1 (5.3%)	
ACHD Complexity			0.556
Moderate	32 (86.5%)	17 (89.5%)	
Severe	5 (13.5%)	2 (10.5%)	

Notes: [a] Cohen's d 0.435. [b] Independence (1 = totally dependent, 4 = minimal assistance, 6 = modified independence, 7 = independent). Cohen's d −0.069. [c] Socio-Economic Status using Index of Relative Socio-economic Advantage and Disadvantage (1= lowest, 10 = highest); Cohen's d 0.499.

3.6 Self-Reported No ACHD Care

Thirty-seven people were absent from ACHD care citing positive health status as a reason; 62% stated they felt well, 35% had lost track of time and 24% believed they no longer needed follow-up. A further 24% stated their GP did not recommend any ongoing cardiac care and 19% perceived their condition to be fixed, as seen in Fig. 2. Several people were worried about receiving bad news (11%), had moved away from the ACHD centre (11%) and had been recommended to have care every 3 years (19%). There were several statements that did not resonate with the cohort and deemed "not applicable" outlined in Appendix 2.

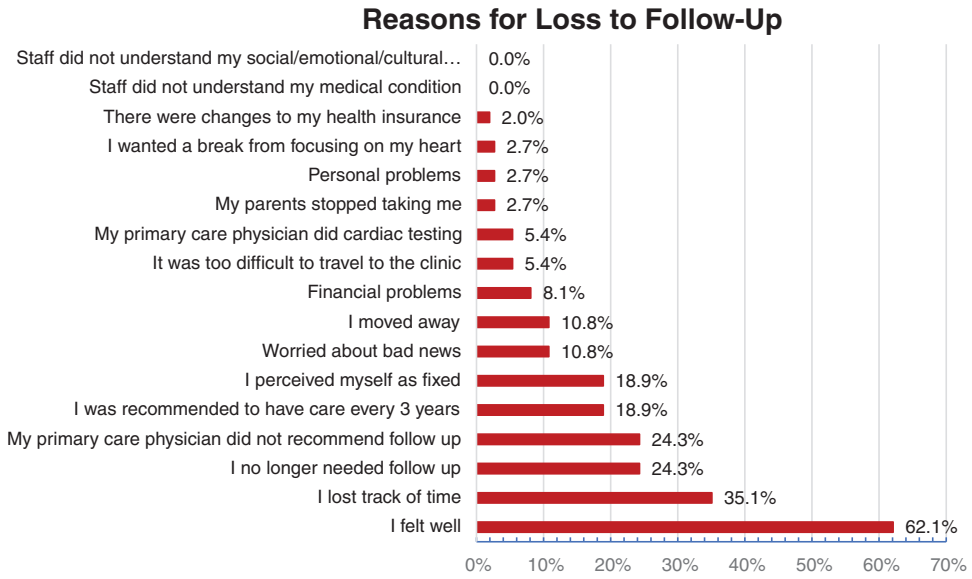

Figure 2: Reasons for loss to follow-up

% Agree and Strongly Agree with reasons for loss to follow-up statements.

3.7 Self-Reported General Cardiology Care

General cardiology care was motivated by several factors shown in Fig. 3 with recommendation from other health care providers (32%) and a desire to prevent potential problems (32%) being the top responses for statement agreement. The onset of new symptoms or health problems (16%), an interest in getting pregnant (11%), concern about deterioration (5%) and an emergency department visit (5%) were the only other affirmative responses.

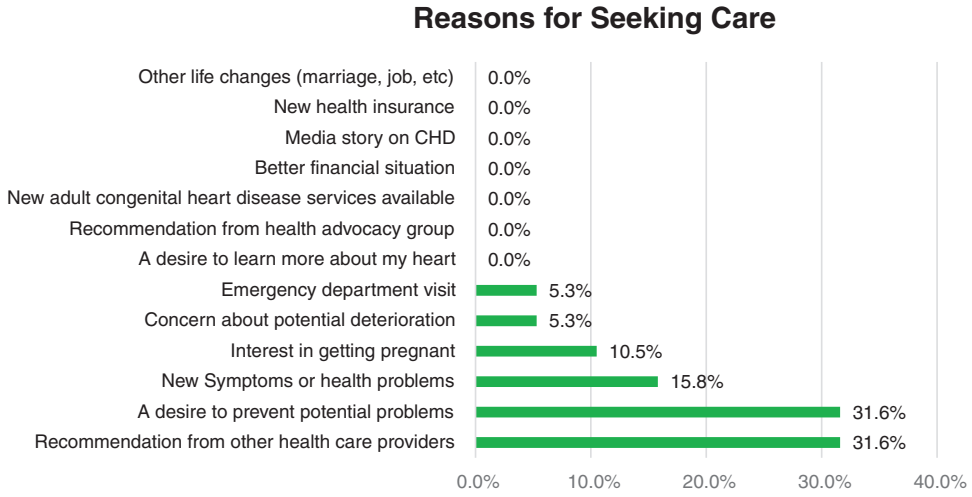

Figure 3: Reasons for returning to general cardiology care

% Agree and Strongly Agree with the return to care statements.

As with the "no care" group, a number of statements did not resonate with the cohort and deemed "not applicable" outlined in Appendix 3.

Self-reports showed that 34% were being seen by a general cardiologist, the remaining 66% were not under any care. People reported independently finding a general cardiology specialist, with some selected because they were local or offered private rooms consultations which were preferable for some to a public clinic. Several people shared their fears of surgery, previous bad experiences, lack of understanding, the difficulty of navigating their own way back to care, re-engaging in generalist care rather, as an alternative to no ongoing cardiac care in Appendix 4.

4 Discussion

Our study examined the prevalence and predictors of LTF in an Australian outpatient ACHD population, finding that 22% of patients with moderate or severely complex ACHD were lost. Whilst in the lower range when compared to European, Canadian, and American studies reporting LTF at 4–61% [17,18,22–24], it is concerning that one-fifth of people did not access guideline-based care.

Predictors for loss to follow-up included younger age, moderately complex lesions, socio-economic disadvantage, and geographical remoteness which are consistent with other studies [10,18,25–27]. In our ACHD cohort LTF occurred in young people in their early 30s who had already transitioned from pediatric care to our ACHD centre. Although the transition is a well-recognized time of risk for LTF to occur [27–30] our data suggest that measures are important throughout life to maintain engagement in ACHD care.

Our study highlights that even adults with a high school education, living close to a major city, and with English language skills are not being retained in care. People with moderately complex lesions were more likely to experience LTF compared to people who had more severe disease. The reasons for this observation are likely multifactorial, due to periods of "wellness" between interventions, less health anxiety, and non-ACHD clinicians providing care. Gender did not influence loss to follow-up in our study, although previous studies have shown higher care gaps in males [24,29]. Low SES scores predicted LTF, likely reflecting the impact of disadvantage in terms of people's health literacy and social resources. Geographic remoteness was a predictive factor for people in living remote/very remote locations although the majority resided in major cities. Australia is not densely populated, and patients travel considerable distances that exceed many European countries' land borders to access specialized services [31,32]. This may be related to the fact that we only considered patients who had been seen at least once at our ACHD service-it is likely that some children had been lost to the system before reaching adult care and thus unrepresentative of the broader ACHD population in Australia, particularly considering the health disparities in rural and remote areas and amongst Aboriginal and multi-cultural people [33].

In our cohort, people did not remain engaged in ACHD care due to feeling well, because of a perception that care was not needed or losing track of time. Worldwide these are recurrent themes that reflect challenges in transition preparation, ACHD knowledge for patients as well as primary health care teams, and the need for lifelong care messaging. Lack of patient transfer by retiring clinicians was an important factor highlighted in survey comments, that led to a perception that no care was needed and subsequent failure to maintain care continuity. There is a strong likelihood that as many as 20% of lost people will eventually present as emergency admissions and be at risk for catastrophic complications due to a lack of close monitoring in specialty care [13]. Missed referral opportunities at both primary and specialist health levels to return to or retain ACHD care were highlighted by over a third of people.

Conversely, we found most people returned to care due to a recommendation from other health care providers. The ACHD health promotion role of community-based providers such as dentists, youth health

and community nurses, pharmacists, and primary health may be under-recognized. For example, identification of ACHD during a health encounter could trigger an action to promote re-engagement or a referral to the nearest ACHD centre. Whilst studies have examined the economic and health benefits of shared care relationships with ACHD specialists, these have been at the medical specialist level [34,35]. In Australia, the design and implementation of an ACHD health pathway framework could provide information for general practice teams, together with patients, at the point of care to guide decisions and facilitate local ACHD service referrals. Any CHD history would activate the pathway, then recommend (via electronic record or visual aid) management, referrals, education, and resources for any patient.

The decision to return to ACHD care was not influenced by concerns about health insurance and finances, appointment schedules, or the location of the ACHD centre which differs from the findings in North America [18]. We found that several causes of LTF in USA and Canadian studies were not perceived as relevant by participants. Finances and health insurance were not reasons for an absence from care and this difference may be attributed to the universal health care system present in Australia.

5 Limitations

Our sample is drawn from a single-site study albeit from the largest ACHD service in NSW. The LTF proportion in our study may be linked to the high volume of ACHD outpatient centre which have been shown to be predictive of care continuity [36]. It is also possible that the LTF rates are underestimated as the true prevalence of ACHD is imprecise in the absence of a national database [37–39]. Our data describes only people seen in our service and possibly does not reflect those lost from paediatric to adult care.

6 Conclusion

Retention to consistent, lifelong specialized ACHD care positively influences morbidity and mortality. Loss to follow-up has previously been demonstrated in the teenage and early 20's population, attributed to the inadequate transition from paediatric to adult services. This study identifies that even after a successful transition; younger age, moderate complexity defects, SES disadvantage, and geographical remoteness are predictors of loss to follow-up. Fear, lack of specific health knowledge, and communication contribute to LTF. Conversely, stronger connections with knowledgeable general practitioners and uptake of consumer-orientated digital applications promoting ACHD health are potentially protective pillars [40]. Opportunities for novel communication and engagement modalities and maintaining connections between patients and providers must be explored to enhance the retention of specialty ACHD care beyond the period of transition.

Acknowledgement: None.

Funding Statement: The authors received no specific funding for this study.

Author Contributions: The authors confirm contribution to the paper as follows: study conception and design: Cordina and Celermajer; data collection: Dickson; analysis and interpretation of results: Dickson & Fethney; draft manuscript preparation: Dickson. All authors reviewed the results and approved the final draft.

Availability of Data and Materials: The datasets are not publicly available due to ethical considerations, though can be made available upon reasonable request.

Ethics Approval: Human Research and Ethics Committee Sydney Local Health District (Protocol X18-0189 HREC/11/RPAH/625). Additionally, all people surveyed provided informed verbal consent at the time of telephone contact.

Conflicts of Interest: The authors declare that they have no conflicts of interest to report regarding the present study.

References

1. Moons P, Bovijn L, Budts W, Belmans A, Gewillig M. Temporal trends in survival to adulthood among patients born with congenital heart disease from 1970 to 1992 in Belgium. Circ. 2010;122(22):2264–72.
2. Mylotte D, Pilote L, Ionescu-Ittu R, Abrahamowicz M, Khairy P, Therrien J, et al. Specialized adult congenital heart disease care: the impact of policy on mortality. Circ. 2014;129(18):1804–12. doi:10.1161/CIRCULATIONAHA.113.005817.
3. Khan A, Gurvitz M. Epidemiology of ACHD: what has changed and what is changing? Prog Cardiovasc Dis. 2018;61(3):275–81. doi:10.1016/j.pcad.2018.08.004.
4. Australian Institute of Health and Welfare. Congenital heart disease in Australia. Cat. no. CDK 14. Canberra: AIHW; 2019.
5. Stout KK, Daniels CJ, Aboulhosn JA, Bozkurt B, Broberg CS, Colman JM, et al. AHA/ACC guideline for the management of adults with congenital heart disease. J Am Coll Cardiol. 2018. doi:10.1016/j.jacc.2018.08.1029.
6. Brida M, Šimkova I, Jovović L, Prokšelj K, Antonová P, Olga Balint H, et al. European society of cardiology working group on adult congenital heart disease and study group for adult congenital heart care in central and south eastern european countries consensus paper: current status, provision gaps and investment required. Eur J Heart Fail. 2021;23(3):445–53. doi:10.1002/ejhf.2040.
7. Heery E, Sheehan AM, While AE, Coyne I. Experiences and outcomes of transition from pediatric to adult health care services for young people with congenital heart disease. Syst Rev. 2015;10(5):413–27. doi:10.1111/chd.12251.
8. Cordina R, Nasir Ahmad S, Kotchetkova I, Eveborn G, Pressley L, Ayer J, et al. Management errors in adults with congenital heart disease: prevalence, sources, and consequences. Eur Heart J. 2017;39(12):982–9. doi:10.1093/eurheartj/ehx685.
9. Baumgartner H. Does frequently inadequate adult care threaten the outcome of congenital heart disease after successful paediatric treatment? Eur Heart J. 2018;39(12):990–2. doi:10.1093/eurheartj/ehy035.
10. Gerardin J, Raskind-Hood C, Rodriguez F, Hoffman T, Kalogeropoulos A, Hogue C, et al. Lost in the system? Transfer to adult congenital heart disease care—challenges and solutions. Congenit Heart Dis. 2019;14(4):541–8. doi:10.1111/chd.12780.
11. Fernandes SM, Khairy P, Fishman L, Melvin P, O'Sullivan-Oliveira J, Sawicki GS, et al. Referral patterns and perceived barriers to adult congenital heart disease care: results of a survey of U.S. pediatric cardiologists. J Am Coll Cardiol. 2012;60(23):2411–8. doi:10.1016/j.jacc.2012.09.015.
12. Iversen K, Vejlstrup NG, Sondergaard L, Nielsen OW. Screening of adults with congenital cardiac disease lost for follow-up. Cardiol Young. 2007;17(6):601–8. doi:10.1017/S1047951107001436.
13. Kaemmerer H, Fratz S, Bauer U, Oechslin E, Brodherr-Heberlein S, Zrenner B, et al. Emergency hospital admissions and three-year survival of adults with and without cardiovascular surgery for congenital cardiac disease. J Thorac Cardiovasc Surg. 2003;126(4):1048–52. doi:10.1016/S0022-5223(03)00737-2.
14. Mackie AS, Tran DT, Marelli AJ, Kaul P. Cost of congenital heart disease hospitalizations in Canada: a population-based study. Can J Cardiol. 2017;33(6):792–8. doi:10.1016/j.cjca.2017.01.024.
15. Apers S, Kovacs AH, Luyckx K, Thomet C, Budts W, Enomoto J, et al. Quality of life of adults with congenital heart disease in 15 countries: evaluating country-specific characteristics. J Am Coll Cardiol. 2016;67(19):2237–45. doi:10.1016/j.jacc.2016.03.477.
16. Mackie AS, Ionescu-Ittu R, Therrien J, Pilote L, Abrahamowicz M, Marelli AJ, et al. Children and adults with congenital heart disease lost to follow-up: who and when? Circ. 2009;120(4):302–9.
17. Wray J, Frigiola A, Bull C. Network, adult congenital heart disease research network, 2013, Loss to specialist follow-up in congenital heart disease; out of sight, out of mind. Heart. 2013;99(7):485–90.

18. Gurvitz M, Valente AM, Borberg C, Cook S, Kay J, et al. Alliance for Adult Research in Congenital Cardiology (AARCC) and Adult Congenital Heart Association. Prevalence and predictors of gaps in care among adult congenital heart disease patients: HEART-ACHD (The Health, Education, and Access Research Trial). J Am Coll Cardiol. 2013;61(21):2180–4. doi:10.1016/j.jacc.2013.02.048.
19. Moons P, Skogby S, Bratt E, Zühlke L, Marelli A, Goossens E, et al. Discontinuity of cardiac follow-up in young people with congenital heart disease transitioning to adulthood: a systematic review and meta-analysis. J Am Heart Assoc. 2021;10(6):e019552.
20. Australian Bureau of Statistics. Socio-economic indices for areas (SEIFA) 2016; 2018.
21. Australian Bureau of Statistics . Accessibility/Remoteness Index of Australia (ARIA). Australian Bureau of Statistics. Available from: https://www.abs.gov.au/statistics/standards/australian-statistical-geography-standard-asgsedition-3/jul2021-jun2026/remoteness-structure/remoteness-areas [Accessed 2023].
22. Caruana M, Aquilina O, Grech V. Can the inevitable be prevented?–An analysis of loss to follow-up among grown-ups with congenital heart disease in Malta. Malta Med J. 2018;30(1):13–21.
23. Gaydos SS, Chowdhury SM, Judd RN, McHugh KE. A transition clinic intervention to improve follow-up rates in adolescents and young adults with congenital heart disease. Cardiol Young. 2020;30(5):633–40. doi:10.1017/S1047951120000682.
24. Moore JA, Sheth SS, Lam WW, Alexander AJ, Shabosky JC, Espaillat A, et al. Hope is no plan: uncovering actively missing transition-aged youth with congenital heart disease. Pediatr Cardiol. 2022;43(5):1046–53.
25. Goossens E, van Deyk K, Budts W, Moons P. Are missed appointments in an outpatient clinic for adults with congenital heart disease the harbinger for care gaps? Eur J Cardiovasc Nur. 2021;21(2):127–34. doi:10.1093/eurjcn/zvab012.
26. Ko JM, Yanek LR, Cedars AM. Factors associated with a lower chance of having gaps in care in adult congenital heart disease. Cardiol Young. 2021;31(10):1576–81. doi:10.1017/S1047951121000524.
27. Kollengode MS, Daniels CJ, Zaidi AN. Loss of follow-up in transition to adult CHD: a single- centre experience. Cardiol Young. 2018;28(8):1001–8. doi:10.1017/S1047951118000690.
28. Kempny A, Diller GP, Dimopoulos K, Alonso-Gonzalez R, Uebing A, Li W, et al. Determinants of outpatient clinic attendance amongst adults with congenital heart disease and outcome. Int J Cardiol. 2016;203:245–50. doi:10.1016/j.ijcard.2015.10.081.
29. Norris MD, Webb G, Drotar D, Lisec A, Pratt J, Akanbi F, et al. Risk factors associated with loss to follow-up in young adults with congeital heart disease. J Am Coll Cardiol. 2012;59(13):E790. doi:10.1016/S0735-1097(12)60791-8.
30. Sonneveld HM, Strating MMH, van Staa AL, Nieboer AP. Gaps in transitional care: what are the perceptions of adolescents, parents and providers? Child: care. Health Dev. 2013;39(1):69–80. doi:10.1111/j.1365-2214.2011.01354.x.
31. McGrath L, Taunton M, Levy S, Kovacs AH, Broberg C, Khan A, et al. Barriers to care in urban and rural dwelling adults with congenital heart disease. Cardiol Young. 2022;32(4):612–7. doi:10.1017/S1047951121002766.
32. Salciccioli KB, Oluyomi A, Lupo PJ, Ermis PR, Lopez KN. A model for geographic and sociodemographic access to care disparities for adults with congenital heart disease. Congenit Heart Dis. 2019;14(5):752–9. doi:10.1111/chd.12819.
33. Jackson JL, Morack J, Harris M, DeSalvo J, Daniels CJ, Chisolm DJ, et al. Racial disparities in clinic follow-up early in life among survivors of congenital heart disease. Congenit Heart Dis. 2019;14(2):305–10. doi:10.1111/chd.12732.
34. Willems R, Ombelet F, Goossens E, de Groote K, Budts W, Moniotte S, et al. Different levels of care for follow- up of adults with congenital heart disease: a cost analysis scrutinizing the impact on medical costs, hospitalizations, and emergency department visits. Eur J Health Econ. 2021;22(6):951–60. doi:10.1007/s10198-021-01300-5.
35. Hardy RY, Keller D, Gurvitz M, McManus B, Varda D, Lindrooth RC, et al. Patient sharing and health care utilization among young adults with congenital heart disease. Med Care Res Rev. 2021;78(5):561–71. doi:10.1177/1077558720945925.

36. Skogby S, Moons P, Johansson B, Sunnegårdh J, Christersson C, Nagy E, et al. Outpatient volumes and medical staffing resources as predictors for continuity of follow-up care during transfer of adolescents with congenital heart disease. Int J Cardiol. 2020;310:51–7.
37. Nicolae M, Gentles T, Strange G, Tanous D, Disney P, Bullock A, et al. Adult congenital heart disease in Australia and New Zealand: a call for optimal care. Heart Lung Circ. 2019;28(4):521–9. doi:10.1016/j.hlc.2018.10.015.
38. Celermajer D, Strange G, Cordina R, Selbie L, Sholler G, Winlaw D, et al. Congenital heart disease requires a lifetime continuum of care: a call for a regional registry. Heart Lung Circ. 2016;25(8):750–4. doi:10.1016/j.hlc.2016.03.018.
39. Jenkins K, Botto LD, Correa A, Foster E. Public health approach to improve outcomes for congenital heart disease across the life span. J Am Heart Assoc. 2019;8(8):e009450. doi:10.1161/JAHA.118.009450.
40. Bassareo PP, Chessa M, Di Salvo G, Walsh KP, Mcmahon CJ. Strategies to aid successful transition of adolescents with congenital heart disease: a systematic review. Children. 2023;10(3):423.

Appendices

Appendix 1: Supplementry data (Univariable all cases)

	Univariable regression	
Factors	H.R. (95% CI)	*p*
Gender		
Female	0.98 (0.81–1.19)	0.87
Male (ref)		
Geographical Remoteness		0.057
Inner Regional		
Outer Regional	1.11 (0.81–1.53)	0.51
Remote/Very remote	1.28 (0.66–2.50)	0.46
Major City (ref)	3.75 (1.39–10.08)	0.009
Socio-Economic Status	0.94 (0.89–0.97)	<0.001
ACHD Anatomy		
Moderate	1.44 (1.11–1.88)	0.007
Severe (ref)		

Note: Supplementary data are presented here with all cases in the model for completeness and transparency as cases with data missing at random were excluded from the analysis presented in the main paper. Appendix 1 shows the univariable analysis including all cases, consistently identifies SES and moderate complexity lesions are predictors of LTF.

Appendix 2: Statements found not applicable by participants

Reason for Loss to Follow-Up	Not Applicable
I felt well	13.5%
I no longer needed follow up	16.2%
I perceived myself as fixed	29.7%
I lost track of time	43.2%
My primary care physician did not recommend follow-up	51.4%
I was recommended to have care every 3 years	54.1%
I moved away	59.5%
It was too difficult to travel to the clinic	64.9%
Worried about bad news	67.6%
Financial problems	67.6%
Personal problems	67.6%
My primary care physician did cardiac testing	70.3%
My parents stopped taking me	75.7%
I wanted a break from focusing on my heart	75.7%
Cardiology staff did not understand my medical condition	75.7%
Cardiology staff did not understand my social/emotional/cultural needs	75.7%
A specific, difficult experience relating to my heart care	76.5%
There were changes to my health insurance	78.4%

Appendix 3: Motives for seeking general care deemed not applicable

Reason for Seeking General Cardiology Care	Not Applicable
Recommendation from other health care providers	42.1%
A desire to prevent potential problems	47.4%
New Symptoms or health problems	63.2%
Interest in getting pregnant	78.9%
Concern about potential deterioration	63.2%
Emergency department visit	68.4%
A desire to learn more about my heart	78.9%
Recommendation from health advocacy group	84.2%
New adult congenital heart disease services available	84.2%
Better financial situation	84.2%
Media story on CHD	84.2%
New health insurance	78.9%
Other life changes (marriage, job, etc.)	84.2%

Appendix 4: Comments from survey participants

	General Cardiology to Care
No ACHD care recommendation	*One male expressed his concern after years of uncertainty- "I wasn't local so I wasn't told to go for follow-up, where to go or what to do… I stumbled upon a Dr …"*
Personalized environment	*The parent of a developmentally delayed young adult explained she "… maintained follow-up in rooms as it is easier than attending the clinic and more personal"*
Deterioration concern in non- ACHD care	*A parent who had diligently attended all of her daughter's paediatric appointments recalled "… she was very symptomatic...she was like an old person... could barely move, so breathless and tired…" stated her daughter is much better since treatment with the ACHD specialist*
New symptoms	*A female expressed her surprise at developing palpitations as "I was never referred to come back" (to the ACHD clinic) and eventually found a doctor via work colleagues*
	No ACHD Care
Felt well	*An asymptomatic male "…had one visit … was expecting to have a follow-up letter or reminder sent"*
	Female "…assumed if I needed follow-up it would be arranged".
	Male "as nothing changed on the tests … (I) became a bit complacent"
Fear	*Male "being scared of bad news...I'd been told at some stage I would need open heart surgery and new valve and my chest cut wide open..."*
Lost track of time	*Male "… had forgotten and … had no symptoms…did not need to hurry with follow-up"*
	Male "… after I turned 18 I had to make my own appointments and it just fell by the wayside. Without a reminder you just lose track of time"
	Female "I just got busy working…"
	Female "I knew I had to go but it went out of my mind…I didn't know how often I needed to attend"
	Mother of disabled male "...keeping up with things has been hard"
Not recommended Lack of adult to adult clincian transfer	*Male "...been drilled as a child what I needed …but I was not told by generalist Cardiologist that specific ACHD care was needed".*
	Female explains impact of retiring specialist "tried unsuccessfully to find a new doctor - no one understood my condition and they all said as I had no records … they can't really offer much advice!"
	"I feel lost"

Note: A concluding general open question was part of the survey to ensure any important issues were not missed.