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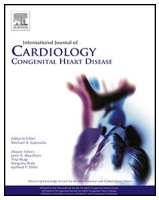

Excellent medium to long term outcomes after cardiac surgery for moderate and complex congenital heart disease, regardless of geographic location☆

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HIGHLIGHTS

- Congenital heart disease patients require lifelong follow-up.
- Long-term survival is excellent in both local and non-local patients.
- Non-local patients do not experience worse post-surgery outcomes, out to 21 years.

ABSTRACT

Objective: To compare the outcomes for repaired tetralogy of Fallot and Fontan patients who must travel from regional Victoria and interstate, in order to receive specialist congenital heart disease (CHD) surgery and ongoing care, with those of local patients.
Methods: This retrospective study included 332 patients who underwent tetralogy of Fallot (ToF) repair and 159 patients who underwent a Fontan procedure at Royal Children's Hospital (RCH) Melbourne between 2003 and 2017. Data was obtained from the National CHD Registry, linked with National Death Index data, and follow-up data from the Australian and New Zealand Fontan Registry.
Results: Equivalent outcomes were observed between location groups in both cohorts for all of the main outcomes of interest. Repaired ToF subjects were aged 0.76 years (IQR 0.52–3.33) at operation and 10.2 years (IQR 5.46–14.9) at last follow-up, whilst Fontan subjects were aged 4.94 (IQR 4.27–5.66) years at operation and 14.2 years (IQR 11.3–16.4) at last follow-up. Mortality rates were extremely low and did not significantly differ between geographic groups, with 10-year survival in the repaired ToF cohort 98.0 % in the City group, 98.1 % in the Regional group, and 98.8 % in the Interstate group; and 97.8 %, 92.3 %, and 97.5 % in the Fontan cohort, respectively.
Conclusions: In the Australian setting and with adequate planning and local follow-up options, patients travelling from regional areas or interstate for their CHD operations have similar outcomes, out to 21 years, compared to patients living locally.

1. Introduction

Congenital Heart Disease (CHD) is a leading cause of infant mortality [1]. The incidence of moderate and severe CHD is approximately 6 in every 1000 live births [2]. Advances in early detection, surgical interventions and management mean that the large majority of such CHD patients survive into adulthood, however many experience late complications [3,4] and require lifelong specialised follow-up [5].

Australia is a vast country with large rural and remote areas. CHD surgical services are provided in relatively few large urban centres, with many children travelling from rural areas or from interstate, to have surgery. Two examples are repair of tetralogy of Fallot and the Fontan procedure. The Royal Children's Hospital in Melbourne routinely provides cardiac surgery for all of the children in the state of Victoria, South Australia, Tasmania and the Northern Territory. In general, rural and remote Australians experience poorer health outcomes and a reduced

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life expectancy than that of their metropolitan counterparts [6]. In the National Congenital Heart Disease Online Survey, 40 % of respondents reported the need to travel over 200 km to receive specialist care [7].

Tetralogy of Fallot (ToF) is the most common defect in children born with cyanotic heart disease, accounting for approximately 10 % of CHD [8], occurring in 3 of every 10, 000 live births [9]. Tetralogy of Fallot has a variable phenotype, with differing severity between diagnostic subgroups, such as 'simple' ToF, to the more severe forms such as ToF with pulmonary atresia [10].

Long-term survival in repaired ToF is >90 % at 30 years [11], however life-long specialist CHD care has been reported as being important in the ongoing management of ToF patients [12–14]. Reinterventions are common in repaired ToF patients, though lower rates of reintervention have been reported following transatrial transpulmonary repair [10]. ToF patients still experience high morbidity however, with a US study finding adults with congenital heart disease such as ToF are hospitalised four to eight times more often than the general population, mainly for cardiac-related issues [13]. Arrhythmias and sudden cardiac death remain important late sequalae for patients with repaired ToF [15]. The most common causes of death in adults with repaired ToF are sudden cardiac death and heart failure [15–17].

Patients with a functionally single ventricle undergo the Fontan

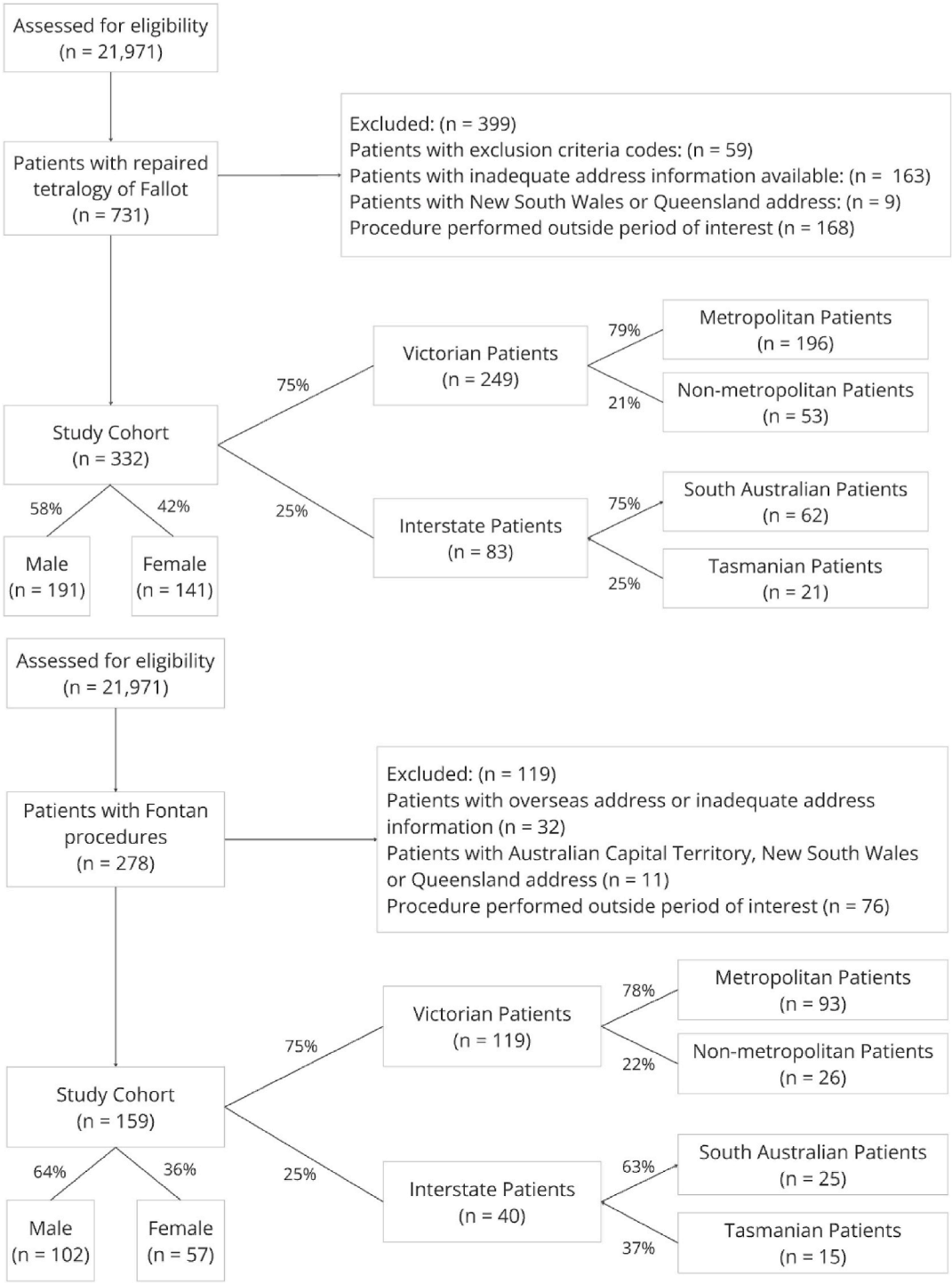

Fig. 1. a: Repaired tetralogy of Fallot cohort selection.b: Fontan cohort selection.

procedure. In the past 2 decades these have all been extra-cardiac conduit type Fontans [18]. Long-term survival is excellent in Fontan patients, however nearly half of the Fontan population experience a major adverse event in the 15 years following surgery, with hypoplastic left heart syndrome patients experiencing the highest rates of adverse events [19]. Potential adverse events include arrhythmia, thromboembolism, heart failure, hepatic dysfunction, transplantation or death [20–23].

Although the importance of lifelong follow-up in CHD patients has been well reported [24,25], the influence of geographic location on outcomes has not been investigated in this population. Given the particular importance of lifelong follow-up and management in moderate and severe complexity patients with ToF [12–14] and Fontan circulations [26,27], the objective of this project is to compare the outcomes for repaired ToF and Fontan patients who must travel long distances in order to receive specialist CHD surgery, with those of more local patients.

2. Methods

2.1. Ethics statement

The study was approved under the National CHD Registry project by the Sydney Local Health District – RPAH Zone Human Research Ethics Committee, under ethics protocol 2019/ETH07472. Data was collected under a waiver of consent for retrospective data. National Death Index Linkage was approved by the Australian Institute of Health and Welfare Human Research Ethics Committee, under ethics protocol EO2023/3/1327.

2.2. Cohort selection

This project involved a retrospective study from the National CHD Registry, linked with data from the National Death Index [28]. The National CHD Registry collects information on patient demographics, diagnoses and procedures from each participating centre's clinical systems [29].

2.3. Repaired ToF cohort

We performed a retrospective study of 332 consecutive patients (Fig. 1a, Table 1a) who underwent complete ToF repair using the transatrial-transpulmonary approach at Royal Children's Hospital Melbourne, between 2003 and 2017 (chosen to allow at least 5 years of follow-up since surgery). Patients who underwent a shunt operation prior to complete repair were also included in the analyses. Patients with pulmonary atresia and/or genetic syndromes (e.g. DiGeorge) were excluded.

Table 1a
Repaired tetralogy of Fallot cohort demographics.

	n	% of total cohort
Sex		
Male	191	58
Female	141	42
Birth year		
1985–1989	4	1
1990–1994	15	5
1995–1999	30	9
2000–2004	30	9
2005–2009	64	19
2010–2014	134	40
2015–2019	55	17
Location Measure		
City	196	59
Regional	53	16
Interstate	83	25
Index of Relative Socio-economic Advantage and Disadvantage (IRSAD) Quintile		
1 (most disadvantaged)	64	19
2	52	16
3	61	18
4	77	23
5 (most advantaged)	78	23

2.4. Fontan cohort

We performed a retrospective study of 159 consecutive patients (Fig. 1b, Table 1b) who underwent a Fontan procedure at Royal Children's Hospital Melbourne, between 2003 and 2017, enriched with data from the Australian and New Zealand Fontan Registry [21]. A chart review was conducted on 20 patients in this cohort that did not have follow-up data available in the Australian and New Zealand Fontan Registry, to ensure outcomes of interest were not missed.

2.5. Location categorisation

We compared three groups; metropolitan Melbourne ('City') patients, non-metropolitan ('Regional') Victorian patients, and 'Interstate' patients travelling from Tasmania or South Australia for treatment. The location groups were classified in accordance with Australian Statistical Geography Standard (ASGS) Edition 3 [30]. Remoteness areas are derived from the Accessibility/Remoteness Index of Australia (ARIA) [31], which classifies Australia into 5 zones on the basis of relative access to services; major cities, inner regional, outer regional, remote and very remote [32]. For the purpose of this study, metropolitan or 'City' Victorian patients were those in the major city category, whilst non-metropolitan or 'Regional' Victorian patients were those in any of the other four categories. Patients were categorised using their suburb and postcode to determine their Statistical Area Level 2, then using the ABS look up tables to match the SA2 to either a City or Regional classification [33]. Addresses are collected routinely as part of the National CHD Registry minimum dataset, extracted from the electronic medical record at each participating site. We did not collect Aboriginal and Torres Strait Islander status. Socioeconomic status was categorised in accordance with the Australian Bureau of Statistics 2021 Socio-Economic Indexes for Areas (SEIFA) Index of Relative Socio-economic Advantage and Disadvantage (IRSAD) [34].

Table 1b
Fontan cohort demographics.

	n	% of total cohort
Sex		
Male	102	64
Female	57	36
Birth year		
2001–2001	2	1
2002–2003	12	8
2004–2005	31	19
2006–2007	35	22
2008–2009	37	23
Location Measure		
City	93	58
Regional	26	16
Interstate	40	25
Index of Relative Socio-economic Advantage and Disadvantage (IRSAD) Quintile		
1 (most disadvantaged)	21	13
2	28	18
3	36	23
4	43	27
5 (most advantaged)	31	19
Primary Ventricular Morphology		
HLHS	47	30
(Other) Right Ventricle Dominant	26	16
Left Ventricle Dominant	71	45
Indeterminant/Biventricular	15	9

2.6. Definition of outcomes

Survival was determined by an analysis of mortality data received in National Death Index linkage, as well as deaths recorded in hospital electronic medical records. Long-term survival (1-year, 5-year, 10-year) was compared amongst the groups. We evaluated age at surgery, primary pathology and age at last follow-up. We reasoned that admissions due to major cardiac complications in these groups would be at RCH, given the specialised nature of these treatments, especially in the paediatric age range. We examined hospital records for defibrillator or pacemaker implantation, incidence of arrhythmias post-procedure, and frequency and length of follow-up at RCH or its visiting clinics. Routine care received locally for interstate patients could not be ascertained, however if patients were having serious post-operative complications they would return to RCH for intervention. Repaired ToF cohort-specific outcomes include heart failure, endocarditis and reoperation, whilst the Fontan cohort-specific outcomes include heart transplantation, major thromboembolic event, protein-losing enteropathy (PLE), and reintervention or re-operation.

2.7. Statistical analysis

All analyses were performed using R Statistical Software v 4.3.0. Data cleaning was completed using the tidyverse package(22). Significant differences between the three geographical groups, for each of the two conditions (ToF, Fontan) were investigated using one-way ANOVA and then independent samples *t*-test with Bonferroni adjustment for pairwise comparisons, for continuous variables, and Kruskal-Wallis Tests for categorical variables. Statistical significance was set at a two-sided p-value <0.05. Continuous variables were presented by mean and standard deviations or medians with interquartile ranges, and categorical variables were summarised by frequencies. Mortality was shown using Kaplan-Meier estimates.

3. Results

3.1. Repaired Tetralogy of Fallot cohort outcomes

3.1.1. Survival

A total of 7 patients died during follow-up: 3 City Victoria, 3 Regional Victoria, 1 Interstate (Table 2). A log-rank test of the survival distribution showed no evidence of a statistically significant difference between the location groups, $\chi^2(2) = 0.9$, $p = 0.6$. Kaplan-Meir overall estimates of survival at 1, 5 and 10 years did not differ significantly between location groups (Table 2).

3.1.2. Other outcomes of interest

There was no significant difference between location groups for other outcomes of interest (Table 2). There were 6 patients with documented arrhythmia, and a total of 10 patients required pacemaker/ICD implantation. There were 70 patients who required reoperation, 21.7 % of the total cohort. There were no recorded admissions for endocarditis in the Registry. One case of heart failure was recorded in the Registry data.

3.2. Follow-up at Royal Children's Hospital Melbourne

3.2.1. Age at last follow-up and length of follow-up at Royal Children's Hospital Melbourne

There was no significant difference between location groups for age at last follow-up or length of follow-up at Royal Children's Hospital (Table 3).

3.2.2. Frequency of follow-up at Royal Children's Hospital Melbourne

As might be expected, there was a significant difference in the median number of post-ToF repair follow-up appointments at Royal Children's Hospital between the location groups ($F(2) = 12.94$, $p < 0.001$))

Table 3
Measures of follow-up at Royal Children's Hospital Melbourne of 332 patients who underwent tetralogy of Fallot Repair at Royal Children's Hospital between 2003 and 2017.

Follow-up measure	City Victoria	Regional Victoria	Interstate	Total	p value
Median age at last follow-up at RCH (years)	9.95 (IQR 4.66–14.5)	11.9 (IQR 8.79–15.5)	9.11 (IQR 3.56–14.1)	10.2 (IQR 5.46 14.9)	0.0716
Median length of follow-up at RCH (years)	6.86 (IQR 2.36–10.1)	8.44 (IQR 6.01–10.9)	6.87 (IQR 0.91–10.4)	7.02 (IQR 2.36–10.5)	0.221
Median number of follow-up visits at RCH, n (SD)	13 IQR 6–25)	15 (IQR 11–28.5)	4 (IQR 3–13)	12 (IQR 4–22)	<0.001

Table 2
Outcomes of 332 patients who underwent tetralogy of Fallot Repair at Royal Children's Hospital between 2003 and 2017.

Outcome	City Victoria (n = 196)	Regional Victoria (n = 53)	Interstate (n = 83)	Total (n = 332)	p value
Overall mortality, n	5	2	1	8	0.6
1-year survival (%)	98.5 (95 % CI, 96.8–100)	98.1 (95 % CI, 94.5–100)	98.8 (95 % CI, 96.5–100)	98.5 (95 % CI, 97.2–99.8)	0.4
5-year survival (%)	98.0 (95 % CI, 96.0–99.9)	98.1 (95 % CI, 94.5–100)	98.8 (95 % CI, 96.5–100)	98.2 (95 % CI, 96.8–99.6)	0.8
10-year survival (%)	98.0 (95 % CI, 96–99.9)	98.1 (95 % CI, 94.5–100)	98.8 (95 % CI, 96.5–100)	98.2 (95 % CI, 96.8–99.6)	0.9
Median age at procedure (years)	0.82 (IQR = 0.52–2.92)	0.62 (IQR = 0.45–6.25)	0.75 (IQR = 0.52–2.50)	0.76 (IQR = 0.52–0.3.33)	0.436
Pacemaker and Implantable cardioverter defibrillator (ICD) Implantation, n	5	2	3	10	0.840
Arrhythmia, n	2	1	3	6	0.332
Endocarditis, n	0	0	0	0	N/A
Re-operation (complete repair), n	43	12	15	70	0.736
Heart Failure, n	1	0	0	1	N/A

(Table 3). There is evidence that the median number of follow-up visits at RCH in City patients is higher than the Interstate patients (p = < 0.001). The median number of follow-up visits at RCH for Regional patients was higher than for the Interstate patients (p = < 0.001). There was weak evidence of a difference between the City and Regional groups (p = 0.088). There were 2 patients in the cohort with no recorded follow-up at Royal Children's Hospital Melbourne.

3.3. Fontan cohort outcomes

3.3.1. Survival

A total of 8 patients died during follow-up: 4 City Victoria, 2 Regional Victoria, 2 Interstate (Table 4)). A log-rank test of the survival distribution showed no evidence of a statistically significant difference between the location groups, $\chi^2(2) = 0.5$, p = 0.8. Kaplan-Meir overall estimates of survival at 1, 5 and 10 years did not differ significantly between location groups (Table 3).

3.3.2. Other outcomes of interest

There was no significant difference between location groups for the other outcomes of interest (Table 4). 14 patients had recorded incidences of arrhythmia, and a total of 10 patients required pacemaker or ICD implantation. 43 patients underwent reintervention, with 12 patients undergoing Fontan revision or conversion. One case of heart transplantation was recorded in the Registry data. There were 18 cases of thromboembolism, and 3 of protein-losing enteropathy.

3.4. Follow-up at Royal Children's Hospital Melbourne

3.4.1. Age at last follow-up at Royal Children's Hospital Melbourne

There was a significant difference in the median age at last follow-up appointment at Royal Children's Hospital between the location groups (F(2) = 4.307, p = 0.0151)) (Table 5). Pairwise t-tests with Bonferroni adjustment showed that the median age at last follow-up at RCH in City patients is higher than the Interstate patients (p = 0.052), and that the median age at last follow-up at RCH in Regional patients is higher than the Interstate patients (p = 0.024). There was no evidence of a difference between the City and Regional groups (p = 0.922). There were 2 patients in the cohort with no recorded follow-up at Royal Children's Hospital Melbourne.

Table 5
Measures of follow-up at Royal Children's Hospital Melbourne of 159 patients who underwent a Fontan Procedure at Royal Children's Hospital between 2003 and 2017.

Follow-up measure	City Victoria	Regional Victoria	Interstate	Total	p value
Median age at last follow-up at RCH (years)	14.2 (IQR 11.6–16.2)	15.0 (IQR 13.7–17.6)	13.6 (IQR 7.04–16.3)	14.2 (IQR 11.3–16.4)	0.0151
Median length of follow-up at RCH (years)	8.90 (IQR 6.48–11.0)	10.4 (IQR 8.43–11.9)	7.38 (IQR 0.45–10.9)	9.12 (IQR 6.44–11.3)	0.0022
Median number of follow-up visits at RCH, n	30 (IQR 17–44)	29 (IQR 25–39)	10 (IQR 4.5–36)	28 (IQR 13–42)	0.0057

3.4.2. Length of follow-up at Royal Children's Hospital Melbourne

There is strong evidence of a significant difference in the length of post-Fontan follow-up at Royal Children's Hospital between the location groups (F(2) = 6.395, p = 0.0022) (Table 5). Pairwise t-tests with Bonferroni adjustment showed that the average length of follow-up at RCH was higher in City patients than Interstate patients (p = 0.0125),

Table 4
Outcomes of 159 patients who underwent a Fontan Procedure at Royal Children's Hospital between 2003 and 2017.

Outcome	City Victoria (n = 93)	Regional Victoria (n = 26)	Interstate (n = 40)	Total (n = 159)	p value
Overall mortality, n	4	2	2	8	0.8
1-year survival, %	100 (95 % CI, 100–100)	100 (95 % CI, 100–100)	100 (95 % CI, 100–100)	100 (95 % CI, 100–100)	N/A
5-year survival, %	98.9 (95 % CI, 96.9–100)	92.3 (95 % CI, 82.6–100)	97.5 (95 % CI, 98.8–100)	97.5 (95 % CI, 95.1–99.9)	0.1
10-year survival, %	97.8 (95 % CI, 94.9–100)	92.3 (95 % CI, 82.6–100)	97.5 (95 % CI, 92.8–100)	92.9 (95 % CI, –)	0.4
Median age at procedure (years)	4.80 (IQR = 4.26, 5.52)	4.70 (IQR = 4.11–5.33)	5.39 (IQR (4.85–6.19)	4.94 (IQR = 4.27–5.67)	0.364
Arrhythmia, n	8	1	5	14	0.479
VT, n	0	0	1	1	0.368
SVT, n	6	1	1	8	0.368
Undefined, n	2	0	3	5	0.368
Heart Transplant, n	0	1	0	1	N/A
Re-intervention, n	27	8	8	43	0.505
Re-intervention: Coil embolisation of aortopulmonary collateral arteries	4	1	0	5	0.559
Re-intervention: Fenestration closure	12	2	2	16	0.437
Re-intervention: Pacemaker insertion	4	0	0	4	0.279
Re-intervention: Fontan takedown	0	0	1	1	0.368
Re-intervention: Tricuspid valve repair	0	0	1	1	0.368
Re-intervention: Common atrioventricular valve repair	0	1	0	1	0.151
Re-intervention: Pulmonary artery reconstruction	0	1	0	1	0.151
Re-intervention: Other	2	0	0	2	0.545
Re-intervention: Fontan revision, n	5	3	4	12	0.778
Thromboembolism, n	12	3	3	18	0.573
Protein-Losing Enteropathy, n	1	1	1	3	0.623

SVT: supraventricular tachyarrhythmia; VT: ventricular tachycardia.

and that the average length of follow-up at RCH was higher in Regional than Interstate patients ($p = 0.0036$). There was no evidence of a significant difference in average length of follow-up at RCH between City and Regional patients ($p = 0.599$).

3.4.3. Frequency of follow-up at Royal Children's Hospital Melbourne

There is evidence of a difference in the median number of post-Fontan procedure follow-up appointments at Royal Children's Hospital between the location groups ($F(2) = 5.352$, $p = 0.0057$) (Table 5). Pairwise t-tests with Bonferroni adjustment showed that the median number of follow-up visits at RCH in City patients is higher than the Interstate patients ($p = 0.007$), and that the median number of follow-up visits at RCH in Regional patients is higher than the Interstate patients ($p = 0.037$). There was no evidence of a significant difference between the City and Regional groups ($p = 1.00$).

3.4.4. Socioeconomic status

It was investigated whether socioeconomic status was more important than geographic location group in influencing outcomes. No evidence was found of an effect of socioeconomic status on either cohort for the outcomes of interest.

4. Discussion

It has been suggested that geographic location may introduce gaps in care for CHD patients and thereby influence outcomes(64). There are 4 paediatric congenital heart units in Australia providing cardiac surgery. Outreach clinics are run from some of these centres. The paediatric cardiac programme in Melbourne provides outreach clinics to the state of Tasmania and many locations in regional Victoria however many rural and remote CHD patients are still required to travel large distances in order to receive specialist care. State governments offset the costs by providing funds to support patient travel and accommodation, though this may not compensate for time off work and school.

Previous studies have investigated the effect of distance from care centres on one-year mortality in CHD. A U.S. study on the effect of patient location and distance to treatment in children undergoing CHD surgery found there was no increased mortality in those in rural areas (67). Other studies also found overall mortality was not associated with distance from a surgical centre(68, 69). As these studies all reported on 1 year survival, little is known about the effect of distance to CHD centres on longer-term outcomes. This current study investigates the long-term outcomes of CHD patients after heart surgery, for up to 21 years, comparing three location groups.

Overall, the survival rates in both study cohorts are excellent, with only 2.4 % mortality at a median of 7.0 years post-procedure (8 of 332 patients) for the rToF cohort and 6.3 % mortality at a median of 9.1 years post-procedure (10 of 159 patients) for the Fontan cohort. The 1, 5 and 10-year mortality did not differ significantly between location groups in either disease cohort (Table 2, Table 4). Both rToF and Fontan patients have generally reported excellent long-term survival in the paediatric age range [10,19], and the consistently low mortality between location groups is a promising outcome.

Arrhythmic events increase the risk of heart failure and sudden cardiac death significantly [35]. Arrhythmias are common in patients with repaired ToF, with ventricular arrhythmias a key cause of mortality in repaired ToF [36]. A previous multicentre study of repaired ToF in adults reported 43 % of recruited patients had a sustained arrhythmia or arrhythmia intervention [37]. Although the risk of arrhythmias in patients with an extracardiac-conduit Fontan is lower than in atriopulmonary Fontan patients, arrhythmias remain a persistent and problematic issue [38]. No difference in prevalence of arrhythmia between location groups was observed in our study (Table 2, Table 4). Our cohort is relatively young however, and arrhythmia was rare overall.

Implantable cardiac devices such as pacemakers may be required in rTOF [39] and Fontan patients [19,40]. Pacemaker and defibrillator implantation are major complications in post-operative CHD patients, increasing morbidity and mortality and extending hospital stay [41]. No difference in pacemaker or defibrillator implantation rates between location groups was observed in the rToF cohort (Table 2) or the Fontan cohort (Table 4), in our study.

D'Udekem et al. reported that after 30 years of follow-up, 24 ± 5 % of rToF patients underwent re-operation [42]. 21.7 % of this rToF cohort underwent reoperation (Table 2). It is important to note however that the threshold for pulmonary valve replacement has evolved in the past 20 years. An ANZFR study revealed 22 % of Fontan patients required a reoperation after 16 years of age, whilst 23 % required more than 1 separate intervention. In this study, in a younger cohort of patients with median follow-up of approximately 10 years after operation, 27 % of the Fontan cohort have undergone reintervention, and 7.5 % have had Fontan revision (Table 4). None of this cohort underwent conversion. As with the rToF cohort, rates were similar between location groups for both reintervention and revision.

Repaired ToF patients are at increased risk of infective endocarditis and heart failure [16], but no admissions for endocarditis and only 1 for heart failure were captured in the rToF cohort (Table 2).

Thromboembolic events, protein-losing enteropathy and heart transplantation in the Australian and New Zealand Fontan cohort have already been well characterised as important late adverse events by the Australia and New Zealand Fontan Registry [19], but the lack of difference in prevalence in these adverse events between location groups is an important finding in this study.

Although survival following ToF repair and Fontan completion have both increased over time, both procedures are associated with complications requiring lifelong follow-up [43,44]. This study has found Interstate patients have lower frequency of follow-up at Royal Children's Hospital Melbourne in both cohorts, and lower length of follow-up at Royal Children's Hospital Melbourne in the Fontan cohort (Table 2, Table 4). The median age at last follow-up at Royal Children's Hospital Melbourne was also lower in the interstate group in the Fontan cohort (Table 3). Although the interstate patients may not be seen as often or for as long post-operatively at RCH, they are likely still receiving routine follow-up in their home state from their general practitioners or specialist care. The frequency of these routine visits is unknown, but we are unlikely to have missed major adverse outcomes, as these patients would have been re-referred to RCH if there were major problems. Whilst outcomes are already excellent for regional and interstate patients, telehealth and satellite clinics with CHD specialists make follow-up far more accessible for those patients who would otherwise need to travel long distances to RCH to receive this care.

How best to treat rural and remote patients with moderate or complex CHD is challenging because these patients often leave the familiarity of a tertiary referral centre such as Royal Children's Hospital Melbourne to receive care at local centres. This study shows that interstate patients, who have the greatest distance to travel to receive follow-up at the centre where they underwent their procedure, but with support of experienced local centres, do not experience poorer outcomes.

This study is limited by incomplete data capture - although the National CHD Registry is the largest of its kind, data were initially collected retrospectively from various clinical systems with potentially incomplete data fields and missing values. We are also limited by our ability to correctly classify location category on the basis of having the most current address information for individuals. As is the case with many studies of this kind, those lost-to-follow-up will not be represented, which presents an inherent selection bias. Some interstate patients may have had some complications treated locally – such as pacemaker implantation - but this seems very unlikely in those under 18 years old (the great majority of patients in this study), who much more likely would have received these treatments at RCH. Other outcomes such as liver pathology are not routinely collected in this Registry but may be an important sequala post-surgery. Future investigation into this cohort

includes assessing the impact of location on healthcare utilisation using data linkage. Future studies should incorporate information on the type of care interstate individuals with critical CHD are receiving once they stop receiving care at their tertiary referral centre. It may also be of interest to see how attitudes factor into follow-up adherence, if patients who must cover large distances to receive follow-up at the centre where their procedure was performed are less worried because they receive care locally, or are more worried as a result of having less surveillance post-procedure. Qualitative research into how location relates to decision-making regarding follow-up could provide great insight into this. Finally, as most late complications occur up to several decades after childhood heart surgery, longer term follow-up to examine the effect of geographic location on late outcomes will be important in the future.

5. Conclusion

There is no evidence that those travelling from regional areas or interstate for moderate and complex congenital heart surgery experience worse outcomes than those patients who reside locally, during medium to long term follow-up. This promising outcome suggests that non-local patients can expect the same outcomes as their metropolitan counterparts, assuming access to well-structured local support systems.

CRediT authorship contribution statement

Larissa Lloyd: Writing – review & editing, Writing – original draft, Visualization, Software, Project administration, Methodology, Investigation, Formal analysis, Data curation. **Calum Nicholson:** Writing – review & editing, Validation, Software, Funding acquisition, Data curation, Conceptualization. **Geoff Strange:** Writing – review & editing, Funding acquisition. **Rachael Cordina:** Writing – review & editing, Supervision, Data curation. **David S. Celermajer:** Writing – review & editing, Supervision, Funding acquisition, Conceptualization. **Michael M.H. Cheung:** Writing – review & editing, Validation, Supervision, Data curation.

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Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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Glossary

Arrhythmia a condition in which the heart beats with an irregular or abnormal rhythm

Endocarditis Endocarditis is inflammation of the endocardium; the inside lining of the heart chambers and heart valves

Fontan procedure The Fontan operation is a palliative surgical procedure performed in patients with a functional or anatomic single ventricle (also known as univentricular heart)

Protein-losing enteropathy a condition characterized by severe loss of serum protein into the intestine.

Tetralogy of Fallot A condition caused by a combination of four heart defects that are present at birth

Thromboembolism obstruction of a blood vessel by a blood clot that has become dislodged from another site in the circulation

Abbreviations

ARIA	Accessibility/Remoteness Index of Australia
ANZFR	Australia New Zealand Fontan Registry
ASGS	Australian Statistical Geography Standard
CHD	congenital heart disease
CI	confidence interval
ICD	implantable cardioverter-defibrillators
IRSAD	Index of Relative Socio-economic Advantage and Disadvantage
IQR	inter quartile range
NSW	New South Wales
RCH	Royal Children's Hospital
rTOF	repaired Tetralogy of Fallot
SA2	statistical area 2
SEIFA	(Socio-Economic Indexes for Areas)
SVT	supraventricular tachyarrhythmia
ToF	Tetralogy of Fallot
U.S.:	United States
VT	ventricular tachycardia

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