Transfer of Congenital Heart Patients from Paediatric to Adult services in England.

Ferran Espuny Pujol [1], Rodney Franklin [2], Sonya Crowe [1], Katherine L Brown [3], Lorna Swan [4], Christina Pagel [1], Katherine English [5].

1. Clinical Operational Research Unit, Department of Mathematics, University College London, UK.
2. Paediatric Cardiology Department, Royal Brompton and Harefield NHS Trust, London, UK.
3. Cardiorespiratory Department, Great Ormond Street Hospital for Children NHS Foundation Trust, London, UK.
4. Scottish Adult Congenital Heart Disease Service, Golden Jubilee Hospital, Glasgow, UK.
5. Department of Adult Congenital Cardiology, Leeds General Infirmary, Leeds, UK.

Corresponding Author:

Dr Katherine English, Department of Adult Congenital Cardiology, Leeds General Infirmary, Leeds, LS1 3EX, UK. kateenglish@nhs.net

Keywords: Heart Defects, Congenital; Quality of Health Care, Epidemiology, Health Care Economics and Organizations

Word Count: 3000 words

Abstract

Objective: This study assessed transfer of patients from paediatric cardiac to adult congenital heart disease (ACHD) services in England, and factors impacting on this process.

Methods: We undertook a retrospective cohort study utilizing a population-based linked dataset (LAUNCHES QI dataset) that includes all patients born between 1987 and 2000 recorded as having a CHD procedure in childhood. We used HES to identify transfer from paediatric to ACHD services between the ages of 16 and 22 years.

Results: Overall, 63.8% of a cohort of 10 298 patients transferred by their 22nd birthday. Estimated probability (% (95% CI)) of transfer by age 22 was 96.5% (95.3,97.7), 86.7% (85.6,87.9), and 41.0% (39.4,42.6) for severe, moderate, and mild CHD respectively. 42 patients (0.4%) died after age 16 but prior to transfer. Multivariable odds ratios in the moderate and severe CHD groups up to age 20 showed significantly lower likelihood of transfer in females (0.87 (0.78,0.97)), missing ethnicity data (0.31 (0.18,0.52)) patients, those from deprived areas (0.84 (0.72,0.98)), and moderate (compared to severe) CHD (0.30 (0.26,0.35)). Odds of transfer was lower for horizontal compared to vertical care model (0.44 (0.27,0.72)). Patients who did not transfer had a lower probability of a further NCHDA procedure between ages 20 and 30 compared to those who did transfer; 12.3% (5.1,19.6) vs 32.5% (28.7,36.3).

Conclusions: The majority of patients with moderate or severe CHD in England transfer to adult services in a timely fashion. Patients who do not transfer undergo fewer elective CHD procedures over the following decade.

Key Messages

What is already known about this subject?

Previous studies have reported high rates of loss to follow up at the point of transfer to ACHD services. Gaps in care are associated with worse outcomes.

What does this study add?

This study demonstrates that transfer from paediatric to ACHD services in England for patients with more complex congenital heart disease is highly effective, with a step-wise reduction in transfer rates in moderately complex and mildly complex patients. However, it demonstrates clear differences in practice between centres with a vertical and horizontal model of delivering care. Patients who do not transfer undergo fewer interventional or surgical procedures during the following decade.

How might this impact on clinical practice?

The UK model of specialised service provision with ACHD services closely affiliated with paediatric cardiology centres facilitates transfer of moderately and severely complex patients. Horizontal and vertical model centres clearly have different transfer policies with more patients from horizontal models (stand-alone paediatric hospitals) transferring later and failing to ultimately transfer at all. More work is required to understand the value of on-going care in adulthood for patients with simple lesions. Barriers to transfer for ethnic minorities and those from deprived areas should be further assessed and addressed.

Introduction:

Survival after paediatric cardiac surgery and catheter interventions for congenital heart disease (CHD) in the UK is excellent and the vast majority of children undergoing treatment for even complex anatomy now reach adulthood. (1,2)

Because these patients are at increased risk of late cardiac complications including arrhythmia, pulmonary hypertension, heart failure, endocarditis and premature death, (3-7) long term follow up in adult congenital heart disease (ACHD) services is recommended. (8) Patients lost to specialist ACHD follow up have an increased risk of premature death, and do not benefit from standard interventions designed to optimise cardiac function and longevity. (9,10)

Between the 1960's and 1980's paediatric cardiac surgery was provided in many small units across England, before becoming more concentrated in a smaller number of higher volume centres. Many of the current designated ACHD programmes have developed in conjunction with those centres, but until the late 1990's the relatively small number of complex ACHD patients, and scarcity of expertise in ACHD meant that services were fragmented. Often patients were referred to general adult cardiology services, and as many as 30% of patients were lost to follow up at the point of transfer. (11) A formal structure for health care services for ACHD patients is now well established in the UK, supported by the publication of National Service Standards and Specifications in 2016. (12) An entire section is dedicated to Transition, including standards for a structured transition programme beginning at age 12 with transfer from paediatric to specialised ACHD care from age 16.

This study used the LAUNCHES QI ('Linking Audit and National datasets in Congenital Heart Services for Quality Improvement') dataset (13), to examine transfer of patients from paediatric to adult congenital heart services in England.

Methods

Dataset

Information on patients with CHD, and their utilisation of health care services in England and Wales, is not available in a single dataset. Since April 2000, the main source of information on 30-day outcomes following therapeutic congenital cardiovascular procedures in the UK has been the mandatory, National Congenital Heart Disease Audit (NCHDA).(14) As part of the LAUNCHES QI research project, a combined dataset for understanding patient journeys through secondary and tertiary health care systems was built to explore variation across services and identify priorities for QI.(13) NCHDA was linked with two national validated registries; 'PICANet' for admissions to paediatric intensive care units (15) and 'ICNARC-CMP' for admissions to adult intensive care units (16); and with death registrations from Office for National Statistics (ONS), and hospital episode statistics (HES) for routine NHS administrative data on hospital admissions, accident and emergency (A&E) attendances, and outpatient (OP) appointments in England.(17) Using the LAUNCHES QI dataset, this retrospective study examines transfer from paediatric services to ACHD services in a large cohort of patients, and factors affecting successful transfer.

Patient Selection

10,326 patients from the LAUNCHES QI dataset born between 1/4/1987 and 31/3/2000 and aged over 16 years at the time of data collection were studied. Selection is shown in Figure 1.

Baseline characteristics were determined including birth cohort (patients divided into two groups; - those born 1987/88-1993/94, and those born 1995/96-1999/2000), sex, ethnicity and deprivation quintile. Complexity classification (mild, moderate, severe) in accordance with current European Society Cardiology guidelines was assigned to each patient using NCHDA diagnostic and procedural categories and HES ICD-10 diagnostic codes (see supplementary material and Tables S4-S6).(18) Patients were grouped by whether their paediatric cardiology centre before age 16, employed a horizontal (paediatric services in a separate children's hospital, with affiliated ACHD service on different hospital site) or vertical (paediatric cardiac services and ACHD services within the same hospital site) model of care. Deprived (Q1,2) / Non-deprived (Q3,4,5) status was assigned according to postcode derived Index of multiple deprivation (IMD). (19) Further details are given in the supplementary methods (Tables S2-S3).

Main Outcome Measures

Primary outcome was evidence that transfer from paediatric to ACHD services had occurred, and was assigned when the patient was seen in cardiology out-patients or admitted electively as a cardiology in-patient in a recognised UK specialist ACHD centre or a recognised affiliated outreach centre before their 22nd birthday. (11)

Many patients with mild complexity lesions are purposefully discharged during childhood as they are not considered to require lifelong on-going follow up. Those with mild lesions referred on for adult follow up may only need to be seen every 4-5 years so we determined that data collection from age 16-22 should capture the overwhelming majority of patients. However, all patients with moderate or severely complex conditions would be expected to be seen at least every 2 years with transfer to specialist adult services primarily at ages 16-19 years. (8) We therefore studied a sub-group of patients with moderate or severe disease (n=5824) up to their 20th birthday to minimise the effects of right-censoring of available data and purposeful discharge in the mildly complex group.

Death after age 16 but before transfer was a competing risk to transfer. Life status was ascertained using ONS mortality registry; patients with missing life status (no linkage to ONS) were censored at their last known visit.

We explored factors which may affect transfer, including birth cohort, sex, ethnicity, deprivation and paediatric model of care.

Finally, we examined whether failure to transfer was associated with increased mortality or differences in further procedures in the decade from age 20 to age 30.

Statistical analyses

Patient characteristics and outcomes are first described using counts and percentages.

Conditional Probability Functions (CPFs) were fitted to estimate probability of transfer subject to being alive. (20) CPF differences between groups were assessed using Pepe-Mori tests for all pairwise comparisons. (21) CPFs are expressed as average % (95% CI).

Single variable and multivariable logistic regressions were used to explore factors potentially affecting transfer including birth cohort, age at transfer, sex, ethnicity, diagnostic complexity, socioeconomic deprivation, and service model for severe and moderate complexity patients. Kaplan-Meier and CPFs were used to estimate their probability of death and reintervention, respectively, during ages 20-30 by transfer status by age 20.

Results

Patient Characteristics are shown in Table 1.

Whole cohort outcomes (n = 10,298) are shown in Table 2 and Figure 2.

63.8% of the whole cohort transferred to ACHD services by their 22nd birthday. Rates of transfer are determined by complexity. Only 0.4% (n=42) died prior to successful transfer. In 22.2% (n=2,287) of the whole cohort, there were insufficient years of follow up in the data set to ascertain their status by their 22nd birthday, but they had not died or been transferred at the point of censoring.

The estimated probability (%(95% CI)) of transfer by 22nd birthday (calculated to take account of competing risk of death and right censoring of data) was 68.3% (67.3,69.3) for the whole cohort, 96.5% (95.3,97.7) in the severely complex group, 86.7% (85.6,87.9) in the moderate group, and only 41.0% (39.4,42.6) in the mild complexity group.

Moderate and Severe patients

Transfer and estimated probability (CPF) of transfer by age 20 for the moderate and severe cohort overall and according to our pre-determined factors are shown in Table 3 and Figure S2.

Of the moderate and severely complex patients (n= 5,820), 81.6% (n=4,747) were known to have transferred to adult services, 0.4% (n=26) died without transfer occurring, and 10.5% (n=611) were known to be alive but had not transferred by their 20th birthday. 436 patients (7.5%) did not have enough years of data to fully assess outcome at their 20th birthday. Estimated probability of transfer in the group as a whole at age 20 was 84.7% (83.7,85.7).

Single variable and multivariable odds ratios (OR (95% CI)) are shown in Table 4. In the multivariable model, moderate complexity (OR = 0.30 (0.26, 0.35), p=0.000) was the most important factor determining non-transfer; followed by; missing ethnicity (OR = 0.31 (0.18, 0.52) p=0.000); horizontal model of care (OR = 0.44 (0.27, 0.71) p=0.001); deprived area (OR = 0.84 (0.72, 0.98) p=0.023); and female sex (OR = 0.87 (0.78, 0.98) p=0.014).

Model of care

The multivariable analysis demonstrates that model of care is an important factor in determining transfer. The estimated probability of transfer (% (95%CI)) in the whole cohort at age 22 was 68.8%

(67.6, 67.0) in the vertical model and 56.1% (54.5, 57.7) in the horizontal model. In the moderate/severe subgroup, estimated probability of transfer at age 20 was 89.3% (88.2, 90.4) in the vertical model and 78.3% (76.5, 80.1) in the horizontal model. (See table 3) The timing and rate of transfer by model is shown in Figure 3.

Transfer occurs significantly earlier in patients in a vertical model than in a horizontal model. Transfer by complexity in each model is shown in Figs 3c and 3d, demonstrating that the timing of transfer is mostly determined by model of care rather than by complexity of the patient.

Patients who have NOT transferred by age 20

Of the 611 severe/moderate cohort who had not transferred by age 20 (Table 3), 155 (25.4%) subsequently transferred between age 20-22. 107 (69.0%) of these were from horizontal centres, and 129 (83.2%) were moderate rather than severe complexity.

Of the 283 severe/moderate cohort who were known to have not transferred by age 22 (Table 2), 57.6% were from horizontal centres despite these patients only making up 42.1% of the overall severe/moderate cohort, demonstrating a shortfall in transfer for patients from horizontal centres even up to age 22. Between ages 16 and 22, 26 of these 283 patients were only seen in cardiology at paediatric centres, and a further 89 patients had either an inpatient or outpatient episode in general adult cardiology. Of the remaining 168 patients, it was not possible to identify whether they were sent any cardiac appointments (and failed to attend), or were never sent appointments.

Outcomes in relation to transfer status

Despite complex congenital heart disease, probability of death in both groups remained very low and was not impacted by transfer status. (Fig 4a) 2.4% (0.8,4.0) vs 3.9% (3.1,4.8). Patients transferred by age 20 had significantly higher probability of undergoing a further NCHDA procedure between the ages of 20-30.(Fig 4b) 12.3% (5.1,19.6) vs 32.5% (28.7,36.3).

Discussion

Life-long specialist ACHD follow up is appropriate for all but the least complex of congenital heart lesions detected in childhood, so, as patients enter their teens, the process of transition begins. Transition programmes for adolescent patients aim to reiterate the importance of long-term care and to empower patients to take ownership of their own healthcare decisions. Effective Transition

programmes improve the chance of transfer to adult care, (9) which usually occurs at age 16-18 depending on the individual needs and comorbidities of the patient. Rate of transfer is only one measurement of effectiveness, and does not reflect other aspects of quality of a transition programme, which cannot be captured in routine data collection.

Our data demonstrate a very high rate of transfer to specialist ACHD services in England for patients with severe and moderate lesions, with an estimated probability of transfer of 96.5% for severely complex patients and 86.7% for moderately complex patients by their 22nd birthday. Only 1.3% of severely complex and 6.0% of moderately complex patients are identified as being lost to follow up at this point with small numbers of patients with unknown outcomes due to incompleteness of their timelines. Previous studies from Canada and the USA show higher proportions of patients being lost to follow-up. (22, 23) Despite these successes, overall, 10.5% of our moderate and complex patient cohort in England did not transfer to specialist adult congenital services by their 20th birthday, with very small numbers of patients continuing to transfer after the age of 20. Gaps in care and lack of regular specialist follow-up are likely to have a detrimental impact on long-term outcomes. (24) It is important that patients in our cohort who do not transfer by age 20 undergo significantly fewer NCHDA procedures in the subsequent decade (Fig 4b), suggesting they may be missing out on standard interventions offered relatively routinely to patients under active follow up.

When we focus on those with moderate and severe complexity, various factors were found to be important for transfer. In our cohort women were slightly less likely to transfer than men, the reasons for this are unclear. Our cohort was unbalanced with regard gender split at baseline with more men than women. This gender imbalance in complex congenital heart disease is well described, and the differences we see may merely reflect subtle differences in patient complexity not captured by our severity groupings.

Transition programmes develop over time, responding to local factors and the changing needs of patients, and as such we may expect to see an increase in effectiveness over time. However, we did not demonstrate any differences in the effectiveness of transfer between our two birth cohorts.

Social deprivation was a significant determinant of failure to transfer care in this study as has been previously reported. (12, 24) While we did not demonstrate concern with any specific ethnic groups, it is notable that patients with ‘missing’ ethnicity data were less likely to transfer. Our experience is that a ‘missing’ ethnicity code is more likely to occur in patients from ethnic minority backgrounds. It is likely that there is overlap and interaction between these two factors, as patients from ethnic minority communities are more likely to reside in areas of higher deprivation. (25)

How we organise care does appear to have a marked impact on both timing and eventual rate of transfer. In England there are two models; vertical model (care from infancy to death in the same institution) and horizontal model (where paediatric care and adult care are in two separate institutions). In our study, patients from a horizontal model were less likely to transfer to adult services by their 22nd birthday, regardless of complexity.

The optimal age for transfer to adult services for individual patients varies depending on their maturity, other health needs and patient preference, but most authors recommend transfer between 16 and 18 years.(26,27) Later transfer may be appropriate in patients with complex needs remaining under the care of multiple paediatric specialties, but this may restrict autonomy of the young adult in relationships with both medical care-givers and parents, and limit access to expert advice regarding sexual and reproductive health, more commonly the domain of adult practitioners. Later transfer may also pose difficulties in the event of acute admissions as access to in-patient facilities tends to be determined by age. Conversely, vertical model units transfer the majority of severe and moderately complex patients by age 17, and almost all by age 18. This approach may not necessarily be in the best interests of patients with complex needs, or low levels of maturity, and may reflect a lack of institutional flexibility in how care is best provided. These discussions aside, it remains more likely that patients from a horizontal model will be lost to follow up at their 22nd birthday.

In our cohort, only 37% of patients with mild lesions, as defined by the ESC guidelines, (18) were transferred to ACHD services by age 22. From this dataset it cannot be determined if this low rate of transfer was due to clinically appropriate planned discharge or not. There is increasing evidence that unrepaired, and even repaired, mild lesions, do carry an excess of cardiovascular and respiratory morbidity in later life, (28,29) such that it could be argued all of these patients should stay under life-long follow up to facilitate access to specialist care and advice regarding non-cardiac surgery, future pregnancy, contraception and genetic risk, and endocarditis. This is balanced against personal and healthcare costs of well patients receiving arguably unnecessary follow up. Our data suggest that a large proportion of patients with mild lesions are discharged prior to adulthood, or never transfer, and their needs and ways to meet these needs, should be studied in more detail.

It is noteworthy, that of the total cohort of 10,298 patients, only 166 patients (1.6%) died between the ages of 16 and 22 years, with 42 of those (0.4% of total) dying without transfer to ACHD. In contrast to historic cohorts the life expectancy curves for patients born with congenital heart disease now much more closely mimic those of the general population. (30) The extremely good prognosis

for the vast majority of teenagers with congenital heart disease is another driver for timely transfer through to adult services in all patients so they can build and develop relationships with their adult team likely to be looking after them for many years to come.

Limitations

Our cohort consisted of patients undergoing a surgical or interventional cardiac procedure as a child. Patients with congenital heart disease who did not undergo a procedure, were not included. However, the study was likely to capture almost all of those with moderate or severe disease who survived to adulthood.

As in any similar study, the dataset had limited granularity and was subject to the limitations of coding and hospital information systems throughout England.

Right-censoring of follow up data for later births limited some data analyses, with not all patients reaching an event end-point or age end-point within the study time. Competing risks analysis (CPFs estimation) was performed to minimise this impact.

Recording of ethnicity was not complete, limiting the completeness of our analyses into the impact of ethnicity (white/non-white differences were found significant in single analyses and narrowly not significant in multivariable analysis, but the missing ethnicity was found significant).

There are likely to be other patients born during our study period who had procedures in childhood prior to the NCHDA being set up in the late 1990s, so those born between 1987 and 1997-2000 are likely to be under represented.

Conclusion

Overall, transfer of severe and moderately complex congenital heart patients to specialist adult services in England is extremely effective. Future initiatives should focus on effective care planning for those at increased risk of loss to follow-up. These include transition programmes, co-designed with partners from non-white groups, and deprived areas to address barriers to transfer. Care-givers in both horizontal and vertical models should consider the demonstrated differences between the models of care and whether changes should be made to their current programmes. Those in

horizontal models should note evidence of lower numbers successfully transferring overall, and further invest in robust links with their ACHD partners.

Finally, careful thought should be given to the needs of those with minor lesions in whom there may be increased late morbidity.

Competing Interests:

There are no Competing Interests to Declare.

Patient and Public Involvement

The LAUNCHES QI programme was constituted with an independent advisory board with both a patient representative and parent representative recruited through national patient charities.

Contributorship Statement

All authors planned the overall study design and analysis. FEP undertook the statistical analysis. RF used his coding expertise to allocate patients to complexity groupings, with clinical assistance from LS, KE. All authors were involved in writing and approving the final manuscript.

Funding Statement

This study was funded by The Health Foundation. Grant # 685009.

Ethics

This study was approved by the North of Scotland Research Ethics Committee. Trial # 18/NS/0106.

References

1. Brown KL, Crowe S, Franklin RC, et al. Trends in 30-day mortality rate and case mix for paediatric cardiac surgery in the UK between 2000 and 2010. Open Heart 2015;14:2(1):e000157.
2. Raissadati A, Nieminen H, Jokinen E, et al. Progress in late results among paediatric cardiac surgery patients: a population based 6-decade study with 98% follow up. Circulation 2015;131:347-53.
3. Hernández-Madrid A, Paul T, Abrams D, et al. Arrhythmias in congenital heart disease: a position paper of the European Heart Rhythm Association (EHRA), Association for European Paediatric and Congenital Cardiology (AEPC), and the European Society of Cardiology (ESC) Working Group on Grown-up Congenital heart disease, endorsed by HRS, PACES, APHRS, and SOLAECE. Europace 2018;20(11):1719-1753.
4. Brida M, Gatzoulis MA. Pulmonary arterial hypertension in adult congenital heart disease. Heart. 2018;104(19):1568-1574.
5. Leusveld EM, Kauling RM, Geenen LW et al. Heart failure in congenital heart disease: management options and clinical challenges. Expert Rev Cardiovasc Ther. 2020;18(8):503-516.
6. Vincent LL, Otto CM. Infective Endocarditis: Update on Epidemiology, Outcomes, and Management. Curr Cardiol Rep. 2018;20(10):86. doi: 10.1007/s11886-018-1043-2.
7. Yu C, Moore BM, Kotchetkova I et al Causes of death in a contemporary adult congenital heart disease cohort. Heart 2018;104(20):1678-1682.
8. Moons P, Bratt E, De Backer J et al. Transition to adulthood and transfer to adult care of adolescents with congenital heart disease: a global consensus statement of the ESC Association of Cardiovascular Nursing and Allied Professions (ACNAP), the ESC Working Group on Adult Congenital Heart Disease (WG ACHD), the Association for European Paediatric and Congenital Cardiology (AEPC), the Pan-African Society of Cardiology (PASCAR), the Asia-Pacific Pediatric Cardiac Society (APPCS), the Inter-American Society of Cardiology (IASC), the Cardiac Society of Australia and New Zealand (CSANZ), the International Society for Adult Congenital Heart Disease (ISACHD), the World Heart Federation (WHF), the European Congenital Heart Disease Organisation (ECHDO), and the Global Alliance for Rheumatic and Congenital Hearts (Global ARCH). European Heart Journal 2021;42: 4213–4223. https://doi.org/10.1093/eurheartj/ehab388
9. Wray J, Frigiola A, Bull C. Loss to specialist follow-up in congenital heart disease; out of sight, out of mind. 2013 Apr 1; 99(7): 485–490)

10. Mylotte D, Pilote L, Ionescu-Ittu R, Abrahamowicz M, Khairy P, Therrien J, Mackie AS, Marelli A. Specialized adult congenital heart disease care: the impact of policy on mortality. Circulation 2014;129:1804-12.
11. Heery E, Sheehan AM, While AE, Coyne I. Experiences and Outcomes of Transition from Paediatric to Adult Health Care Services for Young people with Congenital Heart Disease: A systematic Review. Congenital Heart Disease 2015:10;412-427.
12. https://www.england.nhs.uk/wp-content/uploads/2018/08/Congenital-heart-disease-standards-and-specifications.pdf
13. Espuny Pujol F, Pagel C, Brown KL, et al. Linkage of National Congenital Heart Disease Audit data to hospital, critical care and mortality national datasets to enable research focused on quality improvement. Accepted BMJ Open April 202210.1017/S1047951100012208
14. NICOR | Congenital Heart Disease in Children and Adults (Congenital audit). https://www.nicor.org.uk/national-cardiac-audit-programme/congenital-heart-disease-in-children-and-adults-congenital-audit/
15. Universities of Leeds & Leicester. PICANet – Paediatric Intensive Care Audit Network for the UK and Ireland. https://www.picanet.org.uk.
16. Harrison DA, Brady AR, Rowan K. Case mix, outcome and length of stay for admissions to adult, general critical care units in England, Wales and Northern Ireland: the Intensive Care National Audit & Research Centre Case Mix Programme Database. *Critical Care* 2004;**9**:S1. doi:10.1186/cc3745
17. Herbert A, Wijlaars L, Zylbersztejn A, *et al.* Data Resource Profile: Hospital Episode Statistics Admitted Patient Care (HES APC). *Int J Epidemiol* 2017;46:1093–1093i. doi:10.1093/ije/dyx015
18. Baumgartner H, De Backer J, Babu-Narayan SV, et al. Eur Heart J 2021;42(6):563-645. doi: 10.1093/eurheartj/ehaa554. 2020 ESC Guidelines for the management of adult congenital heart disease.
19. https://www.gov.uk/government/collections/english-indices-of-deprivation
20. M. Pintilie. 'Competing Risks: A Practical Perspective'. Wiley 2006. ISBN: 978-0-470-87069-3
21. Pepe MS, Mori M. Kaplan-Meier, Marginal or Conditional Probability Curves in Summarizing Competing Risks Failure Time Data? Statistics in Medicine 1993;12:737-751.
22. Reid GJ, Irvine MJ, McCrindle BW et al. Prevalence and correlates of successful transfer from pediatric to adult health care among a cohort of young adults with complex congenital heart defects. Pediatrics 2004;113:e197-205. doi: 10.1542/peds.113.3.e197

23. Kollengode MS, Daniels CJ, Zaidi AN. Loss of follow-up in transition to adult CHD: a single-centre experience. Cardiol Young 2018;28(8):1001-1008
24. Kempny A et al. Determinants of outpatient clinic attendance amongst adults with congenital heart disease and outcome. Int J Cardio 2016;203:245-50.
25. Knowles RL, Ridout D, Crowe S, et al. Ethnic and socioeconomic variation in incidence of congenital heart defects. Arch Dis Child 2016;0:1–7. doi:10.1136/archdischild-2016-311143
26. Yassaee A, Hale D, Armitage A, et al. The impact of age of transfer on outcomes in the transition from pediatric to adult health systems: a systematic review of reviews. J Adolesc Health 2019;64:709–720
27. Moons P, Pinxten S, Dedroog D, et al. Expectations and experiences of adolescents with congenital heart disease on being transferred from pediatric cardiology to an adult congenital heart disease program. J Adolesc Health 2009;44:316–322
28. Saha P, Potiny P, Rigdon J, et al. Substantial Cardiovascular Morbidity in Adults With Lower-Complexity Congenital Heart Disease. Circulation 2019;139(16):1889-1899. doi: 10.1161/CIRCULATIONAHA.118.037064.
29. Goldberg JF. Long-term Follow-up of "Simple" Lesions--Atrial Septal Defect, Ventricular Septal Defect, and Coarctation of the Aorta. Congenit Heart Dis 2015;10(5):466-74. doi: 10.1111/chd.12298.
30. Khairy P, Ionescu-Ittu R, Mackie A et al. Changing Mortality in Congenital Heart Disease. JACC 2010. 56:1149-1157

Table 1. Patient characteristics

	All *N*	All %	Severe & Moderate *N*	Severe & Moderate *%*
All	10,298		5,820	
Birth cohort				
Born between 1987/88 and 1993/94 (7 years)	3,293	32.0%	1,979	34.0%
Born between 1994/95 and 1999/2000 (6 years)	7,005	68.0%	3,841	66.0%
Sex				
Male	5,435	52.8%	3,389	58.2%
Female	4,863	47.2%	2,431	41.8%
Ethnicity				
White	8,590	83.4%	4,803	82.5%
Non-white	1,536	14.9%	953	16.4%
Black	332	3.2%	201	3.5%
Asian	897	8.7%	561	9.6%
Other	307	3.0%	191	3.3%
Missing	172	1.7%	64	1.1%
Area of residence deprivation				
Deprived area	4,431	43.0%	2,496	42.9%
IMD Q1 (most deprived)	2,391	23.2%	1,310	22.5%
IMD Q2	2,040	19.8%	1,186	20.4%
Non-deprived area	5,867	57.0%	3,324	57.1%
IMD Q3	1,954	19.0%	1,158	19.9%
IMD Q4	1,931	18.8%	1,094	18.8%
IMD Q5 (least deprived)	1,982	19.2%	1,072	18.4%
Model of care				
Vertical model (Same site adults and children)	6,040	58.7%	3,368	57.9%
Horizontal model (Different site adults and children)	4,258	41.3%	2,452	42.1%
Complexity score*				
Severe	1,454	14.1%	1,454	25.0%
Moderate	4,366	42.4%	4,366	75.0%
Mild	4,478	43.5%		

Note: *Complexity score: Severe includes (repaired/unrepaired): double outlet ventricle, functionally univentricular heart (with or without Fontan palliation), interrupted aortic arch, pulmonary atresia (all types), common arterial trunk (truncus arteriosus), heterotaxy syndromes, cyanotic congenital heart disease (unoperated/palliated), transposition of great arteries (except post-arterial switch); Moderate includes anomalous pulmonary venous connections, atrioventricular septal defects, coarctation of aorta, repaired tetralogy of Fallot, repaired transposition of great arteries with arterial switch, severe pulmonary valvar disease, aortic sub-/supravalvar stenosis, Ebstein anomaly; Mild includes isolated unrepaired small septal defects, repaired large septal defects, isolated mild aortic, pulmonary and mitral valvar disease.

Table 2 Outcomes for the 10,298 patients at their 22nd birthday, overall and by complexity group. The estimated probabilities (CPF) of transfer are conditional on survival of patients and take into account the mortality and censoring of patients.

	N	**Transfer to ACHD services** *N (row %)*	**Death w/o transfer** *N (row %)*	**Not transferred to ACHD services (alive)** *N (row %)*	**Outcome censored before age 22** *N (row %)*	**Estimated probability of transfer at age 22** *% (95% CI)*
All patients	*10,298*	6,567 (63.8%)	42 (0.4%)	1,402 (13.6%)	2,287 (22.2%)	68.3 (67.3,69.3)
Complexity						
Severe	*1,454*	1,329 (91.4%)	12 (0.8%)	19 (1.3%)	94 (6.5%)	96.5 (95.3,97.7)
Moderate	*4,366*	3,573 (81.8%)	15 (0.3%)	264 (6.0%)	514 (11.8%)	86.7 (85.6,87.9)
Mild	*4,478*	1,665 (37.2%)	15 (0.3%)	1,119 (25.0%)	1,679 (37.5%)	41.0 (39.4,42.6)

Table 3 Outcomes for the 5,820 Severe & Moderate cohort at their 20th birthday, overall and by group characteristics. The estimated probabilities (CPF) of transfer are conditional on survival of patients and take into account the mortality and censoring of patients.

	N	**Transfer to ACHD services** *N (row %)*	**Death w/o transfer** *N (row %)*	**Not transferred to ACHD services (alive)** *N (row %)*	**Outcome censored before age 20** *N (row %)*	**Estimated probability of transfer at age 20** % (95% CI)
All severe and moderate complexity	5,820	4,747 (81.6%)	26 (0.4%)	611 (10.5%)	436 (7.5%)	84.7 (83.7,85.7)
Complexity						
Severe	1,454	1,303 (89.6%)	12 (0.8%)	67 (4.6%)	72 (5.0%)	93.5 (92.1,94.9)
Moderate	4,366	3,444 (78.9%)	14 (0.3%)	545 (12.5%)	364 (8.3%)	81.7 (80.5,83.0)
Birth cohort						
Born between 1987/88 and 1993/94	1,979	1,685 (85.1%)	15 (0.8%)	279 (14.1%)	0 (0%)	85.8 (84.3,87.3)
Born between 1995/96 and 1999/2000	3,841	3,062 (79.7%)	11 (0.3%)	332 (8.6%)	436 (11.4%)	83.9 (82.5,85.2)
Sex						
Male	3,389	2,799 (82.6%)	19 (0.6%)	338 (10.0%)	233 (6.9%)	85.7 (84.5,87.0)
Female	2,431	1,948 (80.1%)	7 (0.3%)	273 (11.2%)	203 (8.4%)	83.1 (81.5,84.7)
Ethnicity						
White	4,803	3,975 (82.8%)	20 (0.4%)	463 (9.6%)	345 (7.2%)	85.9 (84.8,86.9)
Non-white	953	731 (76.7%)	6 (0.6%)	130 (13.6%)	86 (9.0%)	79.9 (77.2,82.6)
Missing	64	41 (64.1%)	0 (0%)	18 (28.1%)	5 (7.8%)	65.9 (53.8,77.9)
Area or residence deprivation						
Deprived area	2,496	1,977 (79.2%)	11 (0.4%)	294 (11.8%)	214 (8.6%)	82.5 (80.9,84.1)
Non-deprived area	3,324	2,770 (83.3%)	15 (0.5%)	317 (9.5%)	222 (6.7%)	86.2 (85.0,87.5)
Model of care**						
Vertical model	3,368	2,959 (87.9%)	10 (0.3%)	249 (7.4%)	150 (4.5%)	89.3 (88.2,90.4)
Horizontal model	2,452	1,788 (72.9%)	16 (0.7%)	362 (14.8%)	286 (11.7%)	78.3 (76.5,80.1)

Note: **Model of care: Vertical if paediatric cardiac services and ACHD services are within the same hospital site; Horizontal if paediatric services are in a dedicated children's hospital, with an affiliated ACHD service on a different hospital site. Details in Supplementary Material.

Table 4: Odds ratios for transfer to ACHD services of severe and moderate patients between age 16 and 20th birthday, adjusting for covariates one at a time (single variable odds ratios) or together (multivariable odds ratios)

	Single variable	Multivariable
Birth cohort		
Born between 1987/88 and 1993/94	1.11 (0.87, 1.40)	
Born between 1994/95 and 1997/98 [REF]	1.00	
Sex		
Male [REF]	1.00	1.00
Female	0.85** (0.77, 0.94)	0.87* (0.78, 0.97)
Ethnicity		
White [REF]	1.00	1.00
Non-white	0.63* (0.40, 1.00)	0.68 (0.46, 1.01)
Missing	0.29*** (0.17, 0.51)	0.31*** (0.18 0.52)
Area of residence deprivation		
Non-deprived area [REF]	1.00	1.00
Deprived area	0.75*** (0.65, 0.85)	0.84* (0.72, 0.98)
Complexity		
Severe [REF]	1.00	1.00
Moderate	0.33*** (0.28, 0.38)	0.30*** (0.26, 0.35)
Model of care		
Vertical (same site) model [REF]	1.00	1.00
Horizontal (not same site) model	0.45** (0.26, 0.75)	0.44** (0.27, 0.71)

Notes: ***p<=0.001, **p<=0.01, *p<=0.05. The sample were 4,036 moderate and severe complexity patients born before 1998/99 (data covering all of their ages between 16 and 20) and alive at age 20 (two patients were excluded to allow clustering standard errors by last centre as child; see supplementary material): 3,425 were transferred to adult services, and 611 were not. The multivariable model includes only factors that were significant in the single variable analysis.

Figure 1: Inclusion / Exclusion Criteria

Figure 2. Whole cohort estimated probability of transfer if alive. Overall estimate (left) and by complexity (right) over the follow-up period between 16th and 22nd birthdays. The estimated probabilities conditional on survival of patients takes into account the mortality and right censoring of patients. **Note:** All complexity CPFs were significantly different pairwise (Pepe-Mori test p-values < 0.001).

Figure 3. Outcomes by model of care and complexity. Top left 3a: whole cohort by model of care. Top right 3b: Severe and moderate complexity by model of care. Bottom left 3c: severe and moderate patients in vertical model of care. Bottom right 3d: severe and moderate patients in horizontal model of care. The estimated probabilities conditional on survival of patients so take into account the mortality and censoring of patients. For each subfigure, the pairs of CPFs were significantly different (Pepe-Mori test p-values < 0.001).

Figure 4a. Kaplan Meier average % (95% CI) survival curves, by transfer status aged 20

Note: The Sample is a subgroup of 4,038 severe and moderate patients alive at age 20 and still followed by the dataset (born before 1998/99).

Figure 4b: Cumulative Probability Functions of undergoing a further NCHDA procedure between the ages of 20 and 30 by transfer status aged 20. The Sample for Figure 4.b is a subgroup of 3,391 severe and moderate patients alive at age 20 and still followed by the NCHDA dataset (born before 1997/98). The two CPFs were significantly different (Pepe-Mori test p-values < 0.001).