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ORIGINAL ARTICLE

Distance to Care, Rural Dwelling Status, and Patterns of Care Utilization in Adult Congenital Heart Disease

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Abstract

Many patients with adult congenital heart disease (ACHD) do not receive guideline-directed care. While distance to an ACHD center has been identified as a potential barrier to care, the impact of distance on care location is not well understood. The Oregon All Payer All Claims database was queried to identify subjects 18–65 years who had a health encounter from 2010 to 2015 with an International Classification of Diseases-9 code consistent with ACHD. Residence area was classified using metropolitan statistical areas and driving distance was queried from Google Maps. Utilization rates and percentages were calculated and odds ratios were estimated using negative binomial and logistic regression. Of 10,199 identified individuals, 52.4% lived < 1 h from the ACHD center, 37.5% 1–4 h, and 10.1% > 4 h. Increased distance from the ACHD center was associated with a lower rate of ACHD-specific follow-up [< 1 h: 13.0% vs. > 4 h: 5.0%, adjusted OR 0.32 (0.22, 0.48)], but with more inpatient, emergency room, and outpatient visits overall. Those who more lived more than 4 h from the ACHD center had less inpatient visits at urban hospitals (55.5% vs. 93.9% in those < 1 h) and the ACHD center (6.2% vs. 18.2%) and more inpatient admissions at rural or critical access hospitals (25.5% vs. 1.9%). Distance from the ACHD center was associated with a decreased probability of ACHD follow-up but higher health service use overall. Further work is needed to identify strategies to improve access to specialized ACHD care for all individuals with ACHD.

Keywords Congenital heart disease · Health outcomes · Epidemiology · Access to care · Rural health

Introduction

Adults with congenital heart disease (ACHD) in the United States (US) comprise a growing population with a high rate of healthcare utilization [1–4]. ACHD guidelines recommend ACHD specialist consultation for the majority of patients with CHD, with regular ACHD follow-up recommended for those with moderate-complex CHD [5]. Most US ACHD centers are located in urban population areas, leaving a significant proportion of patients at a distance from care [6]. Gaps in ACHD care are common in US ACHD patients and vary by region, with a higher rate of lapses in care in Western states than in other areas of the US [7]. While the reason for this is unknown, geographic challenges, including physical distance from the ACHD center, may be an important factor.

Rural dwelling individuals in particular face barriers in accessing healthcare, including a lack of transportation and limited healthcare provider availability [8, 9]. Travel distance is known to impact hospital choice for elective

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procedures, as CHD patients are more likely to pick a local center for non-cardiac surgery even if it lacks CHD expertise [10]. This "distance gap" may need to be addressed to meet the goal of delivering guideline-compliant ACHD care. The impact of physical distance and travel time on care utilization in ACHD is not well understood, but has important implications for developing systems of care to expand access for this complex population. In this analysis, we sought to determine whether or not distance to ACHD care and rural dwelling status affects care utilization, including utilization of ACHD-specific services. We utilized the Oregon All Payer All Claims database, a large administrative dataset representing approximately 94% of Oregon residents, including both public and privately insured patients.

Materials and Methods

The Oregon All Payer All Claims (Oregon APAC) database in the years 2010–2015 was queried to identify patients with an International Classification of Diseases-9 (ICD-9) code consistent with congenital heart disease (codes 745–747). Those younger than 18 years and older than 65 years in 2010 were excluded from further analyses. In identifying congenital heart disease, we considered diagnosis codes from any year and any claim in the dataset. We applied a hierarchical algorithm to (1) exclude patients who only had evidence of diagnoses with low sensitivity/specificity and (2) classify remaining patients into one of 13 major defect subgroups based on the codes. Generic codes for "other congenital heart abnormalities" (746.9, 745.9, 746.89) were excluded. This algorithm was previously validated in a university hospital population, but due to concern for low specificity in the general population, those with only the ICD code 394.0 (mitral stenosis, $N=698$) were omitted from the analyses. Importantly, 48% of individuals identified with this code in the sample had an age ≥ 50 years, suggesting that they were unlikely to have Fontan physiology, as they would have been categorized by the published algorithm [11, 12]. Based on commonly accepted severity categories, subgroups were defined as either mild or moderate to severe [13]. For the cohort of patients identified as having ACHD, all codes from all claims from all years were collected and used for analysis.

Age was calculated as the difference between 2010 and year of birth (or between the calendar year of claims for utilization analyses). The patient's geographic area of residence was classified as rural or urban using the designations by the Oregon Office of Rural Health, which defines rural as any geographic areas ten or more miles from the centroid of a population center of 40,000 people or more (https://www.ohsu.edu/oregon-office-of-rural-health/about-rural-and-frontier-data) [14]. These designations were matched by ZIP code, which is a static variable in the APAC dataset based on the most recent reported patient address. Driving distance to Oregon Health & Science University, which is the only accredited ACHD center in Oregon, was calculated using the centroid of the static ZIP code and queried from Google Maps (drive times estimated at mid-day during the working week) [15]. Insurance type was classified using APAC-provided categories; patients could have multiple coverage types and were counted if at least one claim appeared in any year with that coverage. ACHD specialty providers were identified using probabilistic matching methods to find and review both full and partial matches to a list of names and National Provider Identifiers (NPIs); any claim from these providers classified the patient as an ACHD specialty patient. Visits to ACHD providers included both visits at the main campus in Portland Oregon and at satellite centers in Bend, OR and Eugene, OR. Hospital type was determined by reviewing the names of billing facilities, and patients were classified as "urban" if they had visited any hospital in an urban area, then "other Oregon," and then out of state if only out-of-state hospital(s) appeared in their claims. Rural and frontier hospitals were categorized according to Oregon Office of Rural Health guidelines (https://www.ohsu.edu/oregon-office-of-rural-health/facilities-and-services), and critical access hospitals were identified by Center for Medicare and Medicaid Services designation. Comorbidities were determined using the SAS version of *Clinical Classifications Software (CCS) for ICD-9-CM* [16]. Guideline-indicated annual ACHD follow-up and annual echocardiography were defined as per the 2018 AHA/ACC Guideline for the Management of Adults With Congenital Heart Disease [17]. Diagnosis groups with guideline-indicated annual ACHD follow-up were Eisenmenger syndrome/cyanotic, single ventricle/Fontan, transposition of the great arteries (TGA), conotruncal abnormalities, and Ebstein anomaly. Diagnosis groups with guideline-indicated annual echocardiography and/or electrocardiography were Eisenmenger syndrome/cyanotic, single ventricle/Fontan, TGA, and Ebstein anomaly.

Visits were classified as outpatient, emergency department (ED), or inpatient using the Health Care Group (HCG) codes provided by APAC [18]. Inpatient episodes were identified using HCG codes, and if multiple overlapping or consecutive (next day) date ranges existed, these were counted as a single episode. Certain hospitalizations—for bone marrow or organ transplants, perinatal conditions, observation, or chemotherapy—were identified and excluded using Healthcare Effectiveness Data and Information Set (HEDIS) definitions [19]. Cardiac admissions were identified using ICD-9 codes that appeared either as the primary diagnosis or as the secondary diagnosis if the primary diagnosis was an ACHD code (i.e., 745–747). Outpatient and ED visits were considered only if they occurred outside of inpatient date ranges, with the exception of ED visits resulting in inpatient admissions, which were defined as ED claims with inpatient claims on the same or next day. Similarly, outpatient visits were counted only if they did not occur on the same day an ED visit. Multiple claims from the same day were counted as a single episode. A patient was considered to have annual specialist visits or annual testing if they had a visit or test in every year in which they appeared in claims data.

Descriptive statistics (counts and percentages) and Pearson's chi-squared testing were used to compare demographic characteristics of patients in three groups defined by driving time from the center of the patient's ZIP code to the ACHD center (< 1 h, 1 to 4 h, and > 4 h). The probability that a patient accessed a given type of care in a year was calculated as a percentage. For this percentage, a patient was counted once in the numerator for each year with at least one claim for the visit type or procedure in question and once in the denominator for every year with a claim of any type in the period 2010 to 2016. Odds ratios (ORs) and their 99% confidence intervals (CIs) were calculated using logistic regression with one observation per person per year and clustered variance estimates were used to account for the correlation between multiple years for the same person. Confidence intervals for percentages were calculated from predicted values in the crude logistic model to take advantage of the clustered estimates. Adjusted estimates were derived from a logistic model that included age and disease severity (mild vs moderate to severe), as well as patient-level variables for comorbidities that differed between the three driving distance groups and by home location.

Data management and descriptive statistics were completed using SAS software version 9.4 for Windows (SAS Institute, Cary, NC). Utilization analyses were completed with Stata/IC software version 15 for Windows (StataCorp, College Station, TX). This study was approved by the Institutional Review Board at Oregon Health and Science University.

Results

A total of 10,199 individuals with a diagnosis code consistent with ACHD were identified, 52.3% of whom were women. The most common diagnosis category was shunt lesions (32.1%), followed by bicuspid aortic valve (24.8%). 40.9% had moderate-severe complexity CHD, with the most common diagnosis in that group being conotruncal abnormalities (16.3% of the total sample), followed by coarctation of the aorta (8.4% of the total sample) (Table 1). Consistent with Oregon APAC as a whole, data on race and ethnicity were missing in the majority, at 64% and 67%, respectively. Of those with an indicated race, 86.0% were Caucasian, 5.8% were African American, 2.2% were American Indian or Alaska Native, 2.2% were Asian, 0.4% were Native Hawaiian or Pacific Islander, and 3.5% were listed as other.

Table 1 ACHD diagnoses represented in the cohort

ACHD diagnosis	Total number (%)
Mild complexity ($N=6018$)	
Shunt lesion	3276 (32.1)
Bicuspid aortic valve	2532 (24.8)
Pulmonic stenosis	210 (2.1)
Moderate-severe complexity ($N=4181$)	
Conotruncal abnormality	1666 (16.3)
Single ventricle/Fontan	375 (3.7)
Coarctation of the aorta	855 (8.4)
Anomalous coronary artery	554 (5.4)
Transposition of the great arteries	343 (3.4)
Subaortic stenosis	107 (1.1)
Ebstein anomaly	85 (0.8)
Atrioventricular septal defect	71 (0.7)
TAPVR/PAPVR	67 (0.7)
Eisenmenger/cyanotic	58 (0.6)

TAPVR/PAPVR total anomalous pulmonary venous return, partial anomalous pulmonary venous return

Individuals who lived farther from the ACHD center were more likely to have Medicaid (> 4 h: 44.2%, 1–4 h: 32.5%, < 1 h: 26.5%) and less likely to have commercial insurance (> 4 h: 42.5%, 1–4 h: 48.3%, < 1 h: 55.5%) (Table 2). As expected, these individuals were also more likely to live in a rural area (> 4 h: 66.0%, 1–4 h: 55.6%, < 1 h: 9.8%) vs. an urban area. Hypertension, coronary artery disease, and stroke were more common in those that lived > 1 h from the ACHD center (Table 2). Rhythm disorders and diabetes were not significantly associated with home location. No association between gender or disease severity and home location was seen in this cohort.

Association Between Distance to Care and Care Utilization

Living farther from the ACHD center was associated with a decreased likelihood of ACHD clinic follow-up [> 4 h: 5.0%, 1–4 h: 11.6%, < 1 h: 13.0%, adjusted odds ratio for far vs. near = 0.32 (0.22, 0.48)] (Table 3). The rates of guideline-indicated follow-up were significantly lower in those who lived farther away [> 4.0 h: 5.6%, 1–4 h: 11.5%, < 1 h: 11.0% of patients, OR 0.40 (0.21, 0.75)]. Of note, the percentage of patients with a diagnosis requiring annual follow-up by the ACHD guidelines was lower in the group living > 4 h from the ACHD center than in the group living < 1 h from the ACHD center (18.8% vs. 24.9%) [5].

While individuals living far from the ACHD center were less likely to access ACHD care, they were higher utilizers

Table 2 Characteristics of patients with health care claims consistent with adult congenital heart disease (ACHD) by driving distance from the ACHD center

	Overall	Distance from the ACHD center			
		< 1 h	1–4 h	> 4 h	*P*-value*
Total, *N* (%)	10,199 (100)	5343 (52.4)	3827 (37.5)	1029 (10.1)	–
Female, *N* (%)	5336 (52.3)	2801 (52.4)	1966 (51.4)	569 (55.3)	0.08
Age, *N* (%)					
18–24	1443 (14.2)	775 (14.5)	504 (13.2)	164 (15.9)	< 0.0001
25–34	1893 (18.6)	1086 (20.3)	616 (16.1)	191 (18.6)	
35–44	1665 (16.3)	940 (17.6)	570 (14.9)	155 (15.1)	
45–54	2256 (22.1)	1156 (21.6)	892 (23.1)	208 (20.2)	
55–65	2942 (28.9)	1386 (25.9)	1245 (32.5)	311 (30.2)	
Disease severity, *N* (%)					
Mild	6018 (59.0)	3119 (58.4)	2272 (59.4)	627 (59.4)	0.26
Mod-severe	4181 (41.0)	2224 (41.6)	1555 (40.6)	402 (39.7)	
Insurance type, *N* (%)					
Medicaid	3126 (30.7)	1429 (26.5)	1242 (32.5)	455 (44.2)	< 0.0001
Medicare	939 (9.2)	527 (9.9)	361 (9.4)	51 (5.0)	< 0.0001
Dual Medicaid/Medicare	989 (9.7)	470 (8.8)	418 (10.9)	101 (9.8)	0.003
Commercial	5255 (51.5)	2968 (55.5)	1850 (48.3)	437 (42.5)	< 0.0001
Self	2977 (29.2)	1698 (31.8)	1046 (27.3)	233 (22.6)	< 0.0001
Home geographic area, *N* (%)					
Urban	6870 (67.4)	4821 (90.2)	1700 (44.4)	349 (33.9)	< 0.0001
Rural/Frontier	3329 (32.6)	522 (9.8)	2127 (55.6)	680 (66.0)	
Comorbidities, *N* (%)					
Rhythm disorder	5329 (52.3)	2755 (51.6)	2030 (53.0)	544 (52.0)	0.34
Hypertension	5013 (49.2)	2443 (45.7)	2042 (53.4)	528 (51.3)	< 0.0001
Diabetes	2907 (28.5)	1481 (27.7)	1127 (29.4)	299 (29.1)	0.18
CAD	2445 (24.0)	1119 (20.9)	1054 (27.5)	272 (26.4)	< 0.0001
Stroke	2142 (21.0)	1014 (19.0)	865 (22.6)	263 (25.6)	< 0.0001
Heart failure	1748 (17.1)	863 (16.2)	690 (18.0)	195 (19.0)	0.02

**p* value from chi-squared test of association. Lower *p* values suggest that at least one group, defined by distance from the ACHD center, differs from the others

of non-ACHD-specific care, including outpatient, inpatient, and ED visits. 76.5% of individuals living > 4 h from the center had at least one outpatient visit per year with any provider vs. 65.4% in those living 1–4 h from the ACHD center and 63.8% in the group living < 1 h from the center [OR adjusted for age, CHD severity, and comorbidities = 1.80 (1.58, 2.04)] (Table 3). Emergency department (ED) visits were slightly more common [> 4 h: 26.9% per year; 1–4 h: 24.3%; < 1 h: 21.8% per year, aOR 1.21 (1.16, 1.52)], as were inpatient hospitalizations [> 4 h: 14.3% per year; < 1 h: 11.8% per year, aOR 1.24 (1.06, 1.39)] in those who lived farther away. ED to hospital admissions was more than twice as frequent in those who lived > 4 h away than in those who lived < 1 h away [2.2% per year vs. 0.8% per year, aOR 2.32 (1.62, 3.32)]. These analyses were repeated in a sample limited to those with moderate-severe CHD ($N = 4917$), with similar results (data not shown).

Individuals living farther from the ACHD center were more likely to access inpatient or emergency care at rural/frontier or critical access hospitals (Table 4), with critical access hospitals accounting for 8.8% of inpatient admissions and 20.5% of emergency department visits for those who lived > 4 h away vs. 0.7% of inpatient visits and 2.0% of emergency department visits for those who lived < 1 h away. 18.2% of inpatient admissions occurred at the ACHD center for those who lived < 1 h away vs. 15.9% for those who lived 1–4 h away and 6.2% for those who lived > 4 h away. Similarly, ED visits occurred more commonly at the ACHD center for those who lived < 1 h (7.9%) vs. those who lived 1–4 h (1.0%) or > 4 h away (0.3%).

After adjustment for age, CHD severity, and comorbidities, individuals who lived farther away were not more likely to have had a catheterization, or electrophysiology procedure, or an electrocardiogram or echocardiogram (Table 3).

Table 3 Prevalence of healthcare utilization for patients with adult congenital heart disease (ACHD) by distance from the ACHD center in Oregon, All Payer All Claims, 2010 to 2015.

	< 1 h (Near) % (99% CI)	1–4 h (Mid) % (99% CI)	> 4 h (Far) % (99% CI)	Mid vs Near Adjusted OR* (99% CI)	Far vs Near Adjusted OR* (99% CI)
Outpatient visit	63.8 (62.6, 65.0)	65.4 (64.0, 66.7)	76.5 (74.4, 78.6)	1.00 (0.93, 1.09)	1.80 (1.58, 2.04)
ACHD clinic visit					
Any visit	5.6 (5.0, 6.3)	4.9 (4.2, 5.6)	1.9 (1.1, 2.7)	0.90 (0.74, 1.09)	0.30 (0.19, 0.45)
Guideline-indicated[a]	11.0 (9.2, 12.8)	11.5 (9.2, 13.7)	5.6 (2.3, 8.9)	1.05 (0.79, 1.41)	0.40 (0.21, 0.75)
At least once in 2010-2015[b]	13.0 (11.8, 14.2)	11.6 (10.3, 13.0)	5.0 (3.2, 6.7)	0.91 (0.77, 1.09)	0.32 (0.22, 0.48)
Emergency department (ED)	21.8 (20.8, 22.7)	24.3 (23.1, 25.5)	26.9 (24.5, 29.4)	1.12 (1.03, 1.22)	1.21 (1.06, 1.39)
ED to inpatient admission	0.8 (0.7, 1.0)	1.5 (1.3, 1.8)	2.2 (1.6, 2.9)	1.71 (1.32, 2.21)	2.32 (1.62, 3.32)
Inpatient admissions					
All cause	11.8 (11.2, 12.5)	13.1 (12.2, 13.9)	14.3 (12.5, 16.0)	1.01 (0.92, 1.11)	1.08 (0.92, 1.26)
Primary cardiac	5.7 (5.3, 6.2)	6.6 (6.1, 7.2)	7.2 (6.1, 8.4)	1.04 (0.93, 1.17)	1.09 (0.90, 1.31)
Cardiac contributor	8.7 (8.1, 9.3)	9.6 (8.8, 10.3)	9.7 (8.3, 11.1)	0.99 (0.89, 1.09)	0.95 (0.80, 1.12)
Cardiac procedures					
Catheterization	3.9 (3.5, 4.2)	4.7 (4.3, 5.2)	4.8 (4.0, 5.7)	1.07 (0.95, 1.22)	1.11 (0.90, 1.36)
EP study/ablation	0.68 (0.53, 0.83)	0.78 (0.59, 0.97)	0.75 (0.40, 1.10)	1.09 (0.78, 1.51)	1.04 (0.62, 1.74)
Pacemaker/ICD	0.42 (0.31, 0.53)	0.58 (0.43, 0.72)	0.48 (0.20, 0.75)	1.26 (0.87, 1.80)	1.00 (0.53, 1.89)
Cardiac diagnostic testing					
Electrocardiography	34.3 (33.3, 35.4)	34.7 (33.5, 35.9)	36.8 (34.3, 39.2)	0.93 (0.87, 0.99)	1.02 (0.91, 1.14)
Echocardiography	27.4 (26.5, 28.3)	31.6 (30.5, 32.7)	30.0 (27.9, 32.1)	1.20 (1.12, 1.28)	1.08 (0.97, 1.21)
Guideline-indicated[c]	33.3 (29.1, 37.5)	32.1 (27.2, 37.0)	29.4 (19.7, 39.0)	0.94 (0.70, 1.24)	0.86 (0.53, 1.42)
At least once 2010-2015[b]	70.8 (69.2, 72.4)	77.0 (75.3, 78.8)	74.0 (70.4, 77.5)	1.37 (1.20, 1.57)	1.09 (0.89, 1.35)
N patients	5343	3827	1029		
N patient-years	24,278	17,378	4419		

Values are expressed as the percent (99% CI) of patients per year with one or more events, unless otherwise noted

*Adjusted odds ratio is adjusted for age, CHD severity, and patient-level comorbidities of hypertension, coronary artery disease, stroke, and heart failure

[a]Indicates the percentage of patients in a given year who should have an ACHD visit and do. Denominator is those for whom guidelines indicate annual follow-up (n, prevalence, 99% CI) < 1 h $n=1330$, 24.9% (23.4, 26.4); 1 to 4 h $n=858$, 22.4% (20.7, 24.2); > 4 h $n=193$, 18.8% (15.8, 22.1)

[b]Percentage of patients rather than patient-years

[c]Indicates the percentage of patients in a given year who should have an ACHD visit and do. Denominator is those for whom guidelines indicate annual testing (n, prevalence, 99% CI) < 1 h $n=385$, 7.2% (6.3, 8.2); 1 to 4 h $n=278$, 7.3% (6.3, 8.4); > 4 h $n=52$, 5.1% (3.5, 7.1)

Table 4 Inpatient and emergency department visits by hospital type and distance (in driving time) from the ACHD center

	Inpatient visits			Emergency department visits		
Hospital type, *N* visits (%)	< 1 h	1–4 h	> 4 h	< 1 h	1–4 h	> 4 h
Urban	4281 (93.9)	2698 (74.8)	603 (55.5)	10,011 (88.6)	4493 (51.0)	917 (40.8)
ACHD center	830 (18.2)	574 (15.9)	67 (6.2)	888 (7.9)	88 (1.0)	7 (0.3)
Rural/Frontier	88 (1.9)	508 (14.1)	277 (25.5)	751 (6.6)	1891 (21.5)	769 (34.3)
Critical access	31 (0.7)	190 (5.3)	96 (8.8)	224 (2.0)	1925 (21.9)	460 (20.5)
Out of state	158 (3.5)	209 (5.8)	110 (10.1)	312 (2.8)	499 (5.7)	99 (4.4)
Total	4558 (100.0)	3605 (100.0)	1086 (100.0)	11,298 (100.0)	8808 (100.0)	2245 (100.0)

Impact of Rural Home Location

Of the 3329 individuals who lived in a rural location, 522 (15.7%) lived within 1 h of the ACHD center, 2127 (63.8%) lived within 1–4 h, and 680 (20.4%) lived > 4 h from the ACHD center (Table 2). In age and distance-adjusted analyses, rural home location was association with increased rates of ACHD visits and outpatient, inpatient, and ED visits (Fig. 1). After additional adjustment for comorbidities more common in rural patients (hypertension, coronary artery disease, stroke, and heart failure), this association was no longer significant for ACHD center visits, ED visits, and inpatient admissions. The association between rural home location and outpatient visits persisted [rural: 72.3%, urban: 62.4%; aOR 1.34 (1.22, 1.48)], as did the association between home location and ED to inpatient admissions [rural: 2.2%, urban: 0.8%; aOR 1.92 (1.45, 2.55)] (Supplementary Table 1).

Discussion

To our knowledge, this is the first study to comprehensively examine the impact of distance to an ACHD center on access to ACHD care in the US. Our findings are threefold. First, nearly half of the Oregon ACHD population lives one hour or more from the state's only accredited ACHD center and therefore face significant geographic barriers in accessing ACHD care. Second, those who live farther from the ACHD center are less likely to access ACHD-specific care, but are paradoxically higher utilizers of outpatient, inpatient, and emergency department services overall, even after adjustment for age, CHD disease complexity, and comorbidities. Third, rural location did not have as large an impact on location of care delivery as driving distance after adjustment for comorbidities which were more prevalent in the rural population.

While this study is limited to a single-state population, our findings have broad implications, as an estimated 45% of the US ACHD population lives at least one hour away from an ACHD center [6]. Importantly, 10% of the Oregon ACHD population lives more than 4 h from an ACHD center, a higher percentage than is estimated nationally [6]. This makes Oregon residents an ideal population in which to study challenges in access to care related to geographic distance. Importantly, there is a nationwide US shortage of ACHD physicians, with the majority of ACHD centers located in major urban areas and a high degree of regional variation in the density of ACHD cardiologists [20]. Because of these regional differences, ACHD physician shortages are likely to disproportionately affect individuals in certain areas, including those who live outside major population centers.

We utilized an administrative dataset which is nearly comprehensive of the state's population and therefore were able to obtain data on a large number of ACHD patients accessing care across the health system. Our study is near comprehensive of care delivered in the state of Oregon between 2010 and 2015, as > 94% of individuals accessing the healthcare system are represented in the Oregon APAC database. However, the dataset does not include individuals who did not receive care during the study period. Oregon's topography, which includes a large geographic area which is remote from the state's major urban population center, presents a significant challenge for the delivery of ACHD care. Additionally, the state has high rates of income inequality and disparities in educational attainment, factors which also

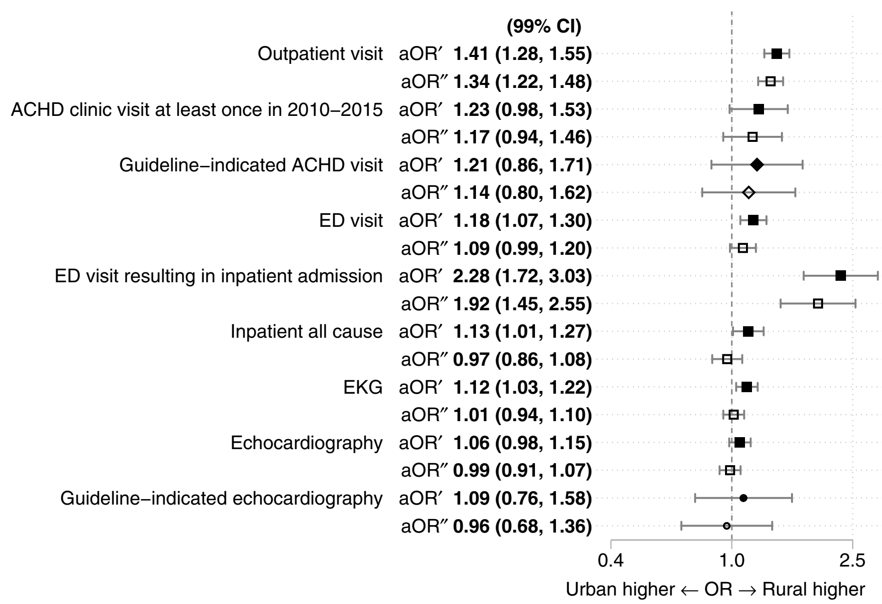

Fig. 1 Odds of health care utilization by rural vs. urban home location. Solid. Analyses adjusted for age, CHD severity, and distance to the ACHD center. Hollow. Analyses adjusted for age, CHD severity, distance to the ACHD center, and patient-level comorbidities of hypertension, coronary artery disease, stroke, and heart failure

constitute barriers to care for many patients [21]. Oregon has a lower percentage of uninsured patients than many states, minimizing the impact of lack of insurance on access to care [22]. This makes Oregon an ideal state in which to study healthcare utilization in the ACHD population.

Home location is associated with cardiovascular disease outcomes, including mortality, in non-ACHD cardiac populations [23]. Rural dwelling patients are less likely to receive evidence-based medications for heart failure, a factor which may lead to an increased risk of adverse outcomes [24, 25]. Geographic challenges in accessing outpatient subspecialty follow-up are postulated to be an important driver of this association, as rural patients with heart failure are less likely to have office-based physician visits, but more likely to utilize inpatient or emergency department services [24, 25]. Similarly, rural patients with myocardial infarction are less likely to receive timely high-quality cardiac care, likely at least in part related to distance from care [26].

While distance to the ACHD center and rural home location are correlated in Oregon, more than a third of individuals who live > 4 h from the accredited ACHD center live in a small city or other urban area. Therefore, we chose to examine both distance to care and rural home location separately in our analyses. The findings were similar, likely because of the significant overlap between distance to care and rurality in our state. Importantly, however, in our analyses, distance was more strongly associated with the probability of ACHD follow-up than rural home location alone. This suggests that in Oregon, physical distance presents a more significant barrier to ACHD follow-up than other factors associated with US rural life, such as socioeconomics, comorbidity burden, cultural factors, internet access, and educational attainment, among others [9, 27–29].

Notably, outpatient, inpatient, and emergency department care utilization were higher in those who lived farther from the ACHD center, even after adjustment for age, disease complexity, and comorbidities. Inpatient admission from the emergency department was more than twice as likely for those patients than for urban dwelling individuals or those who lived close to the ACHD center. This finding could be explained in several ways. Emergency department providers may be hesitant to discharge ACHD patients due to a lack of access to local specialty expertise or uncertainty about follow-up. Alternately, patient factors, such as unmeasured cardiac and non-cardiac comorbidities, or systems factors, such as local care practices or availability of short-term interval outpatient follow-up, could contribute.

Patients living at a distance from ACHD care were more likely to utilize rural or critical access hospitals and less likely to utilize urban or ACHD hospitals for inpatient and emergency department care. These hospitals are small and located at a distance from larger referral hospitals and typically do not have cardiology consultative services. As such, they are not optimized to deliver care to ACHD patients. Further work is needed to determine if differences in patterns of hospital utilization are associated with adverse outcomes in this population.

Only a small percentage of ACHD patients in Oregon had ACHD center follow-up over the study period. This parallels previous data that demonstrate a high rate of gaps in care in ACHD, especially in the Western states [7, 30]. Regional variability in access to ACHD care may be a factor, as could be patient travel time, financial constraints, or insurance (either lack of or insurance with coverage that directs care away from ACHD specialists) [20]. In our population, physical distance was strongly associated with access to ACHD care. Interestingly, this finding conflicts with a previous study which found that travel time was not predictive of ACHD clinic attendance in a non-US-based population [31]. This difference could be related to unmeasured health systems factors, such as referral patterns or restrictive insurance plans, or to the magnitude of the travel distance in a large state like Oregon.

Importantly, individuals living > 1 h from the ACHD center tended to be older. Not surprisingly, they also had a higher rate of hypertension and coronary artery disease, which may in part explain higher rates of healthcare utilization overall. They did not have a higher rate of comorbidities typically associated with higher degree of ACHD complexity, such as rhythm disorders and heart failure. This finding highlights the fact that individuals who live close to and remote from the ACHD center in Oregon are distinct populations with different healthcare needs. Importantly, there was no significant difference in ACHD disease severity by distance from the ACHD center. Future work should focus on optimizing systems of care to effectively care for all ACHD patients, inclusive of geographic variation in patient characteristics and healthcare needs.

While the optimal strategy for expanding access to ACHD care has yet to be defined, there are several promising options. Telehealth is likely to serve an important role, especially for those who do not live in geographic proximity to an ACHD center. ACHD patients in general appear to be receptive to the idea of electronic health delivery, but its efficacy in recruiting and retaining patients in ACHD care has yet to be studied [32]. Off-site or satellite clinics have been historically utilized in many ACHD programs in the US, especially by those who care for patients in a large geographic territory. These clinics have the benefit of providing specialty care in a location closer to home, but are difficult to operate at a large scale, given the travel requirements for ACHD providers and challenges in delivering advanced services such as cardiac imaging in a community setting. Lastly, the hub-and-spoke model of care, in which local providers, typically cardiologists, work closely with ACHD centers to deliver comprehensive care, has significant potential if it is executed correctly [33]. Factors which strongly impact the success of hub-and-spoke care include the ease of sharing of medical records between health systems, the ease and extent of communication between ACHD and local providers, and the openness of patients to pursue a shared model of care.

In Oregon, individuals who live farther from the ACHD center were more likely to have Medicaid insurance. As an expanded Medicaid state, Oregon has a low rate of uninsurance and a higher rate of Medicaid utilization than some states. Nonetheless, our results parallel to previous paper, which showed a higher rate of uninsurance in individuals living farther from ACHD centers, highlight that this population is one with a higher degree of health vulnerability due to socioeconomic and other differences [6]. As noted above, future efforts to expand access to ACHD care should include programs addressing geographic barriers (e.g., satellite clinics or telehealth). They should also address socioeconomic barriers, as differential access to telehealth may exacerbate barriers to care. Importantly, some rural populations face specific challenges in access to virtual health technologies and therefore, telehealth alone is unlikely to be an optimal solution for this population [29].

There are several important limitations of this analysis. We were unable to adjudicate ACHD diagnoses in this administrative dataset. ICD-based billing codes are not always accurate, especially in those with mild disease [11, 12, 34]. We have previously shown that inaccuracies can be related to the codes themselves (e.g., lack of specificity) or to miscoding or misclassification, which may be more common among those without CHD-specific knowledge [12]. While the accuracy of CHD billing codes in a university hospital population is relatively good, especially for those with moderate-complex disease, the accuracy of billing codes in the community may be lower [12]. For instance, our study revealed a smaller than expected number of individuals with atrioventricular septal defects (AVSD), perhaps because of misclassification as other shunt lesions.

We took several steps to maximize accuracy in the selection of our study cohort. First, we excluded individuals older than 65 years, as they have a lower probability of having actual CHD [12]. Second, we utilized a well-validated algorithm to categorize ACHD diagnoses and eliminated nonspecific billing codes where possible. Third, we repeated our analyses in a cohort limited to those with moderate-complex CHD, and the results were not significantly changed.

This study was limited to individuals who had at least one healthcare claim during the study period and thus this study does not include individuals who did not access the healthcare system. Most ACHD patients retain some access with the healthcare system, even if they are lost to ACHD-specific follow-up [7, 30]. Therefore, our dataset is likely to represent a sizable percentage of individuals with ACHD in the state of Oregon. Finally, our data do not allow for analysis of potential confounders that may be influenced by home location, such as education, physical activity, health literacy, or tobacco use.

In conclusion, our study demonstrates that individuals living farther from the ACHD center in Oregon are less likely to access ACHD-specific care, but paradoxically more likely to access healthcare in general. Distance appears to be a more important predictor of lack of ACHD follow-up than rural home location. Importantly, the population living farther from the ACHD center has distinct characteristics, including a higher rate of Medicaid insurance, older age, and a higher rate of overall healthcare utilization. Future work is needed to improve systems of care in ACHD, with a focus on minimizing geographic and socioeconomic disparities in access to ACHD care.

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Data Availability The data that support this study will be available from the corresponding author at reasonable request. To minimize the chances of sharing data that will be could be used to identify patients, location data (zip code, hospital location) will not be shared.

Declarations

Conflict of interest The authors have no competing interests to declare.

Ethical Approval The Department of Veterans Affairs did not have a role in the conduct of the study; in the collection, management, analysis, or interpretation of data; or in the preparation of the manuscript. The views expressed in this article are those of the authors and do not necessarily represent the views of the Department of Veterans Affairs or the U.S. Government.

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