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ORIGINAL RESEARCH

Health Service Utilization Patterns Among Adults With Congenital Heart Disease: A Population-Based Study

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BACKGROUND: Several studies have examined hospitalizations among patients with adult congenital heart disease (ACHD). Few investigated other services or utilization patterns. Our aim was to study service utilization patterns and predictors among patients with ACHD.

METHODS AND RESULTS: We identified 11 653 patients with ACHD aged ≥18 years (median, 47 years), through electronic records of 2 large Israeli healthcare providers (2007–2011). The association between patient, disease, and sociogeographic characteristics and healthcare resource utilization were modeled as recurrent events accounting for the competing death risk. Patients with ACHD had high healthcare utilization rates compared with the general population. The highest standardized service utilization ratios (SSRs) were found among patients with complex congenital heart disease including primary care visits (SSR, 1.53; 95% CI, 1.47–1.58), cardiology outpatient visits (SSR, 5.17; 95% CI, 4.69–5.64), hospitalizations (SSR, 6.68; 95% CI, 5.82–7.54), and days in hospital (SSR, 15.37; 95% CI, 14.61–16.12). Adjusted resource utilization hazard increased with increasing lesion complexity. Hazard ratios (HRs) for complex versus simple disease were: primary care (HR, 1.14; 95% CI, 1.06–1.23); cardiology outpatient visits (HR, 1.40; 95% CI, 1.24–1.59); emergency department visits (HR, 1.19; 95% CI, 1.02–1.39); and hospitalizations (HR, 1.75; 95% CI, 1.49–2.05). Effects attenuated with age for cardiology outpatient visits and hospitalizations and increased for emergency department visits. Female sex, geographic periphery, and ethnic minority were associated with more primary care visits, and female sex (HR versus men, 0.89 [95% CI, 0.84–0.94]) and periphery (HR, 0.72 [95% CI, 0.58–0.90] for very peripheral versus very central) were associated with fewer cardiology visits. Arab minority patients also had high hospitalization rates compared with the majority group of Jewish or other patients.

CONCLUSIONS: Healthcare utilization rates were high among patients with ACHD. Female sex, geographic periphery, and ethnicity were associated with less optimal service utilization patterns. Further research should examine strategies to optimize service utilization in these groups.

Key Words: adult congenital heart disease ■ healthcare service utilization ■ mortality ■ population-based study

A dramatic demographic shift in the congenital heart disease patient population has taken place over the past decades. A once primarily pediatric population, now mainly consists of patients with adult congenital heart disease (ACHD),[1–5] presenting new challenges to health systems worldwide in terms of care organization and resource allocation. The need for lifelong follow-up, disease- and procedure-related

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CLINICAL PERSPECTIVE

What Is New?

- This large cohort study is among the first to report health service utilization patterns among patients with adult congenital heart disease.
- Inpatient and outpatient service utilization rates are significantly higher than in the general population among all adult congenital heart disease lesion complexity groups.
- While older age is associated with greater healthcare utilization (emergency department visits excepted), the increase in cardiology visits or hospital admission rates, with higher disease complexity, diminishes with age.
- Female sex, geographic periphery, and ethnic minority were associated with more primary care visits, female sex and periphery were associated with fewer outpatient cardiology visits, and Arab minority were associated with high hospitalization rates.

What Are the Clinical Implications?

- Adult congenital heart disease is a rapidly growing patient population and healthcare requirements of these patients are expected to further increase as the population ages.
- We found less optimal service utilization patterns among female, periphery, and ethnic minority patients in a framework of universal coverage for primary to tertiary care. Greater disparities may be found in health systems based on private insurance.
- Characterization of healthcare utilization patterns carries important policy-making and infrastructure planning implications, as more experts in this field and centers of excellence for the treatment of this population will be needed in the upcoming years.

Nonstandard Abbreviations and Acronyms

ACHD	adult congenital heart disease
SSR	standardized service utilization ratio

late-onset complications, albeit early repair, as well as age-related acquired comorbidities, entail high health resource utilization among patients with ACHD throughout adult life.[6,7] A recent systematic review reported an increase in the number of hospitalizations and outpatient clinic visits among patients with ACHD over the past decade.[8] Healthcare requirements of patients with ACHD are expected to further increase, as long as the ACHD population continues to expand and age.

Data on ACHD patient healthcare utilization are paramount to informed policy making. However, most of the available information is limited to hospitalizations.[6,9–15] Data for additional services are scarce[11,15,16] and rarely population based.[11] Moreover, most published studies originate from North America and Western Europe. Reports from other regions of the world are limited[17,18] and analyses of utilization patterns and associated factors are scarce.

Our aims were to assess inpatient and outpatient healthcare utilization patterns among patients with ACHD; to examine the relationship of patient, disease, and sociogeographic characteristics to health service utilization; and to compare health services utilization between patients with ACHD and the general population.

METHODS

The authors declare that all supporting data are available within the article and its online supplementary files. The study was conducted in Israel in a framework of national health insurance with universal coverage for primary to tertiary care.[19] The broad benefit package, covering physician consultations, hospitalization, imaging, laboratory services, and medication is publically funded through taxes with limited copayment or no additional charge.

We identified patients with ACHD through electronic records of the 2 largest (of 4 existing) healthcare providers (Maccabi and Clalit health services), covering 77% of the population at the time of the study. Patients aged ≥18 years, with at least 1 documented congenital heart lesion or a specific congenital heart malformation repair procedure (Table S1), insured by the participating providers between January 2007 and December 2011, were included. The institutional review boards of the Sheba Medical Center and the participating healthcare providers approved the study. No informed consent was required.

From 17 637 identified patients with ACHD, we excluded 628 patients who switched providers during the data collection period, to avoid overlap of deidentified records from different providers. We also excluded 1809 patients for whom a diagnosis of congenital heart defect could not be ascertained based on the available data, and 3547 patients for whom disease complexity could not be determined based on diagnosis codes. Eighty-eight percent of the patients completed 5 years of follow-up and 92% completed at least 4 follow-up years. A unique identifier was used to link information sources (eg, chronic diagnoses, hospitalization, and clinic visits) within provider, at the patient level. All data were consolidated into a single database following coding unification and consistency checks.

The healthcare services examined included primary care and cardiology outpatient visits, emergency department (ED) visits, and hospital admissions. Maternity ward hospitalizations were excluded. Only face-to-face patient-doctor encounters were included. Several visits to the same specialty clinic (general practice or cardiology) on the same day were counted once. Data limitation prevented distinction between specialized ACHD clinics and other outpatient cardiology visits.

The *International Classification of Diseases, Ninth Revision* (*ICD-9*), was the main coding system used. Other diagnosis codes were converted to *ICD-9*. Congenital heart disease complexity was categorized based on the 32nd Bethesda Conference report as simple, moderate, severe, or unclassifiable by a hierarchal algorithm according to the most severe congenital heart defect.[3]

Comorbidities were summarized with the Charlson comorbidity score.[20] Geographic periphery is represented by an index based on standardized distance and accessibility of the participant residence locality, to a central economic center.[21] The index created by the central bureau of statistics (2015 version) ranges between 1 (most peripheral) and 10 (least peripheral locality). Residence in Arab localities was used as a surrogate for identification of Arab minority patients.

Statistical Analysis

Data were analyzed with SAS version 9.4 (SAS Institute Inc.). Age-adjusted rates of baseline characteristics by sex and congenital disease complexity were calculated by a direct method with the entire cohort as the reference group. Direct adjustment was selected as a method free of assumptions inherited in multivariable models. Rates were compared while controlling for age (grouped as: 18–24, 25–44, 45–64, and >64 years) with the Cochran-Mantel-Haenszel general association test.[22] To account for left skewed visit and inpatient day distributions, age-adjusted frequencies per 5 years are presented as least square geometric mean rates at mean age.

Negative binomial models with a time logarithm offset were used to assess adjusted relative rates for days in hospital. The Cox proportional hazard model was used for survival multivariable analysis. Adjusted cumulative predicted mortality rates were computed for men, intermediated complexity of congenital heart disease, and mean values of other variables in the model (healthcare provider, age, periphery index, ethnicity, noncardiac congenital abnormalities, and baseline comorbidity) unless used for stratification. The validity of the proportional hazard assumption for the variables of interest was tested by including a time-dependent explanatory variable for each, to test the assumption of no time-dependent effect. No violation was found.

Each service utilization was modeled as a recurrent event accounting for the competing risk of death as suggested by Andersen et al.[23] The approach combines a marginal model (based on counting process) of recurrent events with analysis of competing risk to estimate the subdistribution hazard with the PHREG procedure in SAS. The cumulative number of recurrent events was computed by the Ghosh and Lin multiplicative marginal means model.[24] The proportional hazard assumption was violated for age groups in models for primary care visits, ED visits, and hospitalizations. Stratifying these models by age yielded similar estimates as nonstratified models.

To enable model comparability, we used the same fixed variable set in all models, including age, sex, congenital heart disease complexity, health service provider, geographic periphery, ethnicity according to locality, and comorbidities.

In a separate analysis, standardized service utilization (SSR) and standardized mortality ratios were calculated in reference to the general population. The reference rates for the general population were extrapolated from the population sample of the Hadera District Study.[25] SSR and standardized mortality ratios were computed with the SAS STDRATE procedure matching for age, sex, and ethnicity among patients with ACHD aged 25 to 74 years (the age range of the reference population).

RESULTS

The study cohort comprised 11 653 patients with ACHD (52% women). Median patient age at baseline was 47 years, with 24% of women and 20% of men >64 years. The most common congenital heart lesions were atrial septal defect among women and aortic valve stenosis or insufficiency among men (Table 1).

More than 1 heart defect was recorded for 18% of the women and 15% of the men (Table 2). Seven percent of the patients had an additional noncardiac congenital anomaly. Genetic syndromes included Down (46 patients) and Marfan (22 patients) syndromes (Table S2).

The congenital defect was complex in 5%, intermediate in 21%, and simple in 74% of the patients (Table 2). Patients with complex heart defects were younger on average, more frequently women (56% compared with 52% among patients with a simple congenital defect), and more likely to have multiple congenital heart defects and additional noncardiac anomalies (Table 2; $P<0.0001$ for all). Increasing congenital heart disease complexity was associated with increasing age-adjusted prevalence of acquired

Table 1. Distribution of Congenital Heart Defects Among 5551 Men and 6102 Women With ACHD

	All	Men	Women
	N (%)	n (%)	n (%)
Atrial septal defect	3539 (30.4)	1341 (24.2)	2198 (36.0)
Aortic valve stenosis/insufficiency	3032 (26.0)	1999 (36.0)	1033 (16.9)
Anomalies of the aorta	937 (8.0)	512 (9.2)	425 (7.0)
Ventricular septal defect	1654 (14.2)	714 (12.9)	940 (15.4)
Mitral valve stenosis/insufficiency	1090 (9.4)	411 (7.4)	679 (11.1)
Atrioventricular septal defect	775 (6.7)	274 (4.9)	501 (8.2)
Pulmonary valve anomaly	415 (3.6)	196 (3.5)	219 (3.6)
Patent ductus arteriosus	547 (4.7)	169 (2.3)	378 (6.2)
Tetralogy of Fallot	385 (3.3)	201 (3.6)	184 (3.0)
Common/single ventricle	201 (1.7)	82 (1.5)	119 (2.0)
Ebstein anomaly of tricuspid valve	146 (1.3)	58 (1.0)	88 (1.4)
Transposition of great arteries	77 (0.7)	35 (0.6)	42 (0.7)
Other defects	1209 (10.4)	553 (10.0)	654 (10.7)

ACHD indicates adult congenital heart disease. Patients can have >1 defect; therefore, the percentage can add up to >100%.

cardiac morbidity (Table 2), including ischemic heart disease (17% among complex versus 15% among simple congenital heart patients), heart failure (17% versus 7%), and arrhythmia (28% versus 17%, respectively). Chronic comorbidities such as diabetes mellitus, hypertension, and hyperlipidemia, on the other hand, were more prevalent among patients classified as intermediate, whereas the prevalence of stroke or transient ischemic attack was inversely related to disease complexity (Table 2). The corresponding crude rates are presented in Table S3.

During the data collection period (mean, 5.3 years [SD, 1.0 years]; 61 354 patient-years), 858 (7.4%) patients died, a rate 5.4 times higher (95% CI, 5.0–5.8) than expected in the general population matched by age, sex, and ethnicity. Standardized mortality ratios were 5.7 for simple and intermediate and 10.7 for complex congenital heart disease. Older age (hazard ratio [HR], 4.74 [95% CI, 2.30–9.76] for age 45–64 years, and HR, 23.8 [95% CI, 11.75–48.36] for age >64 years versus age 18–24 years), male sex (HR, 1.32 [95% CI, 1.12–1.56] versus female sex), complex congenital heart disease (HR, 1.94 [95% CI, 1.38–2.72] versus simple disease), and Arab minority (HR, 1.42 [95% CI, 0.96–2.11] versus Jewish) were associated with increased multivariable-adjusted mortality risk. The associated cumulative adjusted mortality rates by age, sex, and disease complexity are presented in Figure 1. The adjusted 5-year cumulative mortality probabilities were 5.5 (95% CI, 4.1–7.0) for simple, 5.2 (95% CI, 3.5–6.9) for intermediate, and 12.3 (95% CI, 7.2–17.1) for complex disease; 5.3 (95% CI, 3.6–6.9) for men and 3.6 for women (95% CI, 2.4–4.8); and ranged between 2% (95% CI, 0.5–3.2) for patients aged 18 to 24 years to 34% (95% CI, 25.2–41.7) for patients older than 64 years.

Health Services Utilization

Health service utilization rates are presented in Table 3. Most of the patients visited a primary care physician at least once in 5 years, with a median of 32 visits per 5 years. Cardiology outpatient visits were documented for 77% of the patients, and 53% were hospitalized during the follow-up period.

Service utilization rates were higher among patients with ACHD than in the general population, across congenital heart disease complexity (Figure 2). The largest differences were observed for patients with a complex lesion, including primary care (SSR, 1.53; 95% CI, 1.47–1.58), cardiology outpatient visits (SSR, 5.17; 95% CI, 4.69–5.64), hospital admissions (SSR, 6.68; 95% CI, 5.82–7.54), and days in hospital (SSR, 15.37; 95% CI, 14.61–16.12). Rates of visits to the ED were higher among patients with ACHD with complex disease and lower than in the general population for patients with simple or intermediate complexity congenital heart disease (Figure 2).

Healthcare resource utilization among patients with ACHD increased with age in each complexity category (Figure S1), except for ED visits not resulting in hospitalization, for which the multivariable-adjusted HR declined with increasing age (Table 4; Figure S2).

Compared with simple congenital heart disease, complex lesions were associated with the highest adjusted healthcare utilization rates (Table 4; Figure S3) with hazards 14% higher for primary care visits, 40% for cardiology visits, and >70% higher for hospitalization and in-hospital days. Further examination revealed an age and disease complexity interaction for outpatient cardiology visits (Table S4). While the relationship between congenital heart disease complexity and cardiology outpatient visits followed a dose response

Table 2. Characteristics of 11 653 Patients With ACHD and CHD

	Sex			Disease complexity			
	Men	Women	*P* Value	Simple	Intermediate	Complex	*P* Value
No. (%)	5551 (48)	6102 (52)		8637 (74)	2457 (21)	559 (5)	
Age, median (IQR), y	46 (30–61)	47 (32–64)	<0.0001	48 (32–63	44 (30–62)	34 (25–55)	<0.0001
No. of heart defects, n (%)							
1	4705 (84.8)	5006 (82.0)	0.0002	7804 (90.4)	1589 (64.7)	318 (56.9)	<0.0001
2	703 (12.7)	874 (14.3)		731 (8.5)	683 (27.8)	163 (29.2)	
3+	143 (2.7)	222 (3.6)		102 (1.2)	185 (7.5)	78 (14.0)	
Other congenital anomaly (n of body systems affected), n (%)							
0	5229 (94.2)	5664 (92.8)	0.02	8115 (94.0)	2272 (92.5)	506 (90.5)	<0.0001
1	302 (5.4)	405 (6.6)		493 (5.7)	171 (7.0)	43 (7.7)	
2+	20 (0.3)	33 (0.5)		29 (0.3)	14 (0.6)	10 (1.8)	
Cardiac morbidity, n (%)*							
Arrhythmia	916 (17.0)	1154 (18.4)	0.03	1512 (17.1)	431 (18.0)	127 (27.7)	<0.0001
Valve disease	387 (7.1)	604 (9.7)	<0.0001	527 (6.1)	351 (14.4)	113 (23.8)	<0.0001
Ischemic HD	997 (18.6)	749 (11.8)	<0.0001	1313 (14.8)	364 (15.4)	69 (16.6)	0.42
Heart failure	472 (8.9)	436 (6.8)	<0.0001	630 (7.1)	203 (8.5)	75 (16.9)	<0.0001
Other HD	924 (17.1)	888 (14.2)	<0.0001	1176 (13.4)	501 (20.9)	135 (27.7)	<0.0001
Other morbidity, n (%)*							
Diabetes mellitus	728 (13.4)	808 (12.9)	0.35	1148 (12.9)	339 (14.4)	49 (12.3)	0.08
Hypertension	1773 (32.6)	1981 (31.7)	0.2	2799 (31.5)	829 (35.1)	126 (28.9)	0.0004
Hyperlipidemia	1619 (29.6)	1718 (27.8)	0.02	2512 (28.4)	712 (30.2)	113 (26.9)	0.07
Stroke/TIA	729 (13.5)	846 (13.6)	0.82	1289 (14.6)	240 (10.2)	46 (10.2)	<0.0001
Kidney failure	30 (0.6)	14 (0.2)	0.004	34 (0.4)	9 (0.4)	1 (0.2)	0.9
Residence locality ethnicity, n (%)*							
Jewish/other	3849 (69.3)	4246 (69.9)	0.36	5992 (69.6)	1732 (70.7)	371 (67.1)	0.05
Arab	403 (7.1)	389 (6.5)		565 (6.6)	167 (6.6)	60 (9.1)	
Mixed	1293 (23.6)	1444 (23.6)		2056 (23.8)	554 (22.7)	127 (23.8)	
Residence locality peripherality, n (%)*							
Very peripheral	121 (2.2)	106 (1.8)	0.33	174 (2.0)	47 (1.9)	6 (0.9)	0.02
Peripheral	710 (12.7)	749 (12.4)		1052 (12.3)	334 (13.5)	73 (13.0)	
Medium	996 (17.9)	1097 (18.1)		1546 (18.0)	443 (17.9)	104 (16.6)	
Central	1140 (20.6)	1314 (21.6)		1890 (22.0)	451 (18.0)	113 (17.2)	
Very central	2576 (46.7)	2813 (46.2)		3950 (45.8)	1177 (48.2)	262 (49.3)	

ACHD indicates adult congenital heart disease; CHD, congenital heart disease; HD, heart disease; IQR, interquartile range; and TIA, transient ischemic attack.

*Age-adjusted percentage.

pattern for all age groups, the differences diminished with age, so that patients 45 years or older had a similar cardiology visit hazard across disease complexity categories. No other significant interaction was detected, although an attenuation of relative risks with age was noted among patients with complex disease for recurrent hospitalization.

Healthcare utilization patterns varied by sex (Table 3). Women were likely to visit a general practitioner more often (multivariable adjusted HR, 1.23; *P*<0.0001) and a cardiologist less often (adjusted HR, 0.89; *P*<0.0001). Women were also more likely to visit an ED (HR, 1.25; *P*<0.0001) but less likely to be admitted (HR, 0.90; *P*<0.0001), once maternity hospitalizations were excluded (Table 4; Figure S4).

Peripheral locality residence was associated with more primary care visits, fewer cardiology clinic visits (up to 28% less for very peripheral locations), fewer ED visits, and more days in intensive care (Table 4).

Compared with Jewish and other localities, residents of Arab localities were younger (median, 37 versus 47 years) and more likely to have a complex disease (6% versus 5%). Arabs had higher adjusted recurrent primary care visit rates (HR, 1.38), ED visits (HR, 1.24), and hospitalizations (HR, 1.54) (Table 4).

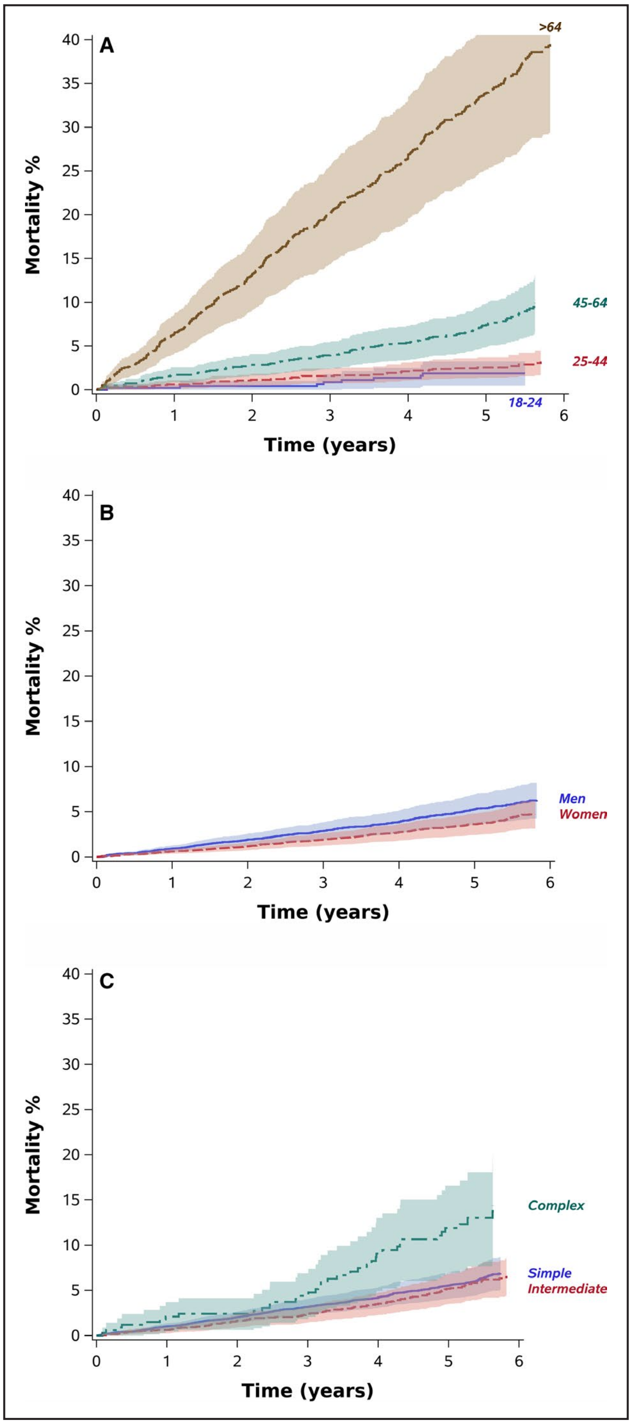

Figure 1. Cumulative multivariable-adjusted mortality rates by age (A), sex (B), and congenital heart disease complexity (C).

Color bands represent 95% CIs. The fixed values used included men, non-Arab ethnicity, intermediate congenital disease complexity, and mean values of age, geographic periphery, number of noncardiac congenital defects, and baseline comorbidity score.

The distance to the nearest specialized ACHD clinic, contributed little in multivariable analysis beyond periphery (HR, 1.0 for all services).

Sensitivity Analysis

As atrial septal defect and patent foramen ovale share the same *ICD-9* code (745.5), we repeated the analysis excluding 2955 (25%) patients identified solely by this code. Following exclusion, SSRs were identical and multivariable-adjusted service utilization estimated hazards were similar to those estimated before exclusion of these patients (Table S5).

DISCUSSION

This study based on a large ACHD patient population is among the first to report rates and patterns of both inpatient and outpatient health service utilizations and characteristics associated with utilization in this patient group. Compared with the general population, we found high healthcare utilization rates among patients with ACHD, including outpatient primary and cardiology care, ED visits, and hospitalizations, across congenital heart disease complexity. As expected, the highest SSRs were observed among patients with a complex disease, as previously reported for primary care consultations,[11,26] and hospital admissions.[11,14] Multiple congenital heart and other defects and higher age-adjusted prevalence of ischemic heart disease, heart failure, and arrhythmia, found among patients with complex disease, likely contributed to higher healthcare utilization rates among this group. Nonetheless, in the current study, healthcare utilization rates among patients with simple or intermediate disease were also substantially higher than in the general population. This finding is in accord with the high mortality risk in the current study and previously reported increased long-term morbidity and mortality in this patient group.[27,28]

Most of the studies among patients with ACHD focused on inpatient services and few on outpatient care and therefore were not able to analyze utilization patterns. One exception is the study by Mackie et al,[11] which examined healthcare resource utilization in Quebec (1996–2000). Compared with that earlier study, our patients were older (median, 47 versus 43 years) with a higher proportion of men (48% versus 42%). These and health organization–related differences between the 2 studies may explain higher cardiology outpatient visit rates in our study (77% versus 55%), while rates of primary care (99% versus 91%) and hospitalizations (53% versus 51% at 5 years) were of similar magnitude.

The majority of the 11 563 patients in our study had a simple congenital heart disease and 5% had a complex congenital heart disease, a rate lower than previously reported for specialized clinics or hospitalized patients. The inclusion of patients less likely to be hospitalized in adulthood or being under follow-up

Table 3. Health Services Utilization by Sex and CHD Complexity

	Sex			Disease Complexity			
	Men	Women	*P* Value	Simple	Intermediate	Complex	*P* Value
Used the service at least once,* n (%)†							
Outpatient primary care	5472 (98.6)	6031 (98.8)	0.13	8531 (98.8)	2426 (98.7)	546 (97.8)	0.08
Outpatient cardiology	4298 (77.4)	4717 (77.2)	0.49	6643 (76.5)	1947 (79.1)	425 (76.0)	0.004
ED only visits	3071 (56.2)	3525 (57.0)	0.36	4863 (55.6)	1394 (57.7)	339 (64.5)	<0.0001
Hospital admissions	2910 (53.3)	3227 (52.1)	0.17	4526 (51.7)	1296 (53.6)	315 60.3)	<0.0001
Intensive care unit	520 (9.6)	444 (7.1)		625 (7.1)	249 (10.4)	90 (16.2)	
Frequency per 5 y,‡ geometric mean (SD)							
Primary care visits	26.5 (2.5)	34.1 (2.3)		30.6 (2.4)	29.0 (2.4)	31.0 (2.4)	<0.0001
Outpatient cardiology visits	4.2 (2.6)	4.0 (2.5)		4.0 (2.5)	4.2 (2.5)	5.0 (2.5)	0.002
ED only visits	1.9 (2.0)	2.1 (2.1)		2.0 (2.0)	2.0 (2.1)	2.2 (2.2)	<0.0001
Hospital admissions	2.6 (2.6)	2.5 (2.5)		2.5 (2.5)	2.5 (2.5)	3.2 (2.7)	0.26
Days in hospital	8.7 (4.2)	8.3 (4.2)		8.4 (4.2)	8.0 (4.1)	11.5 (4.7)	0.14
Days in intensive care unit	7.5 (3.9)	9.2 (4.1)		8.8 (4.1)	6.8 (3.6)	8.3 (4.1)	0.03

CHD indicates congenital heart disease; and ED, emergency department visits not ending in hospitalization.
*Used the service at least once in the data collection period (2007–2011).
†Adjusted for age.
‡Age-adjusted geometric mean per 5 years among patients who used the service.

of specialized clinics in population studies leads to a higher proportion of patients with simple disease and therefore a lower proportion of patients with a complex disease. The prevalence of severe cardiac lesions reported by Mackie et al[1] was 8%, in line with our results. However, differences in classification method preclude direct comparison of this group between the studies.

Of all services examined, hospitalizations exert the highest costs. Increased survival of patients with ACHD increase hospitalization burden.[6,9,10,13] Based on 6 US state inpatient databases, most hospitalizations among patients with ACHD are cardiac related.[29] Acute kidney failure, bacterial infection, anemia, and procedure-related complications were identified as the main drivers of longer hospital stay.

We found the association between congenital heart disease complexity and healthcare utilization to be age dependent. In agreement with a previous report,[11]

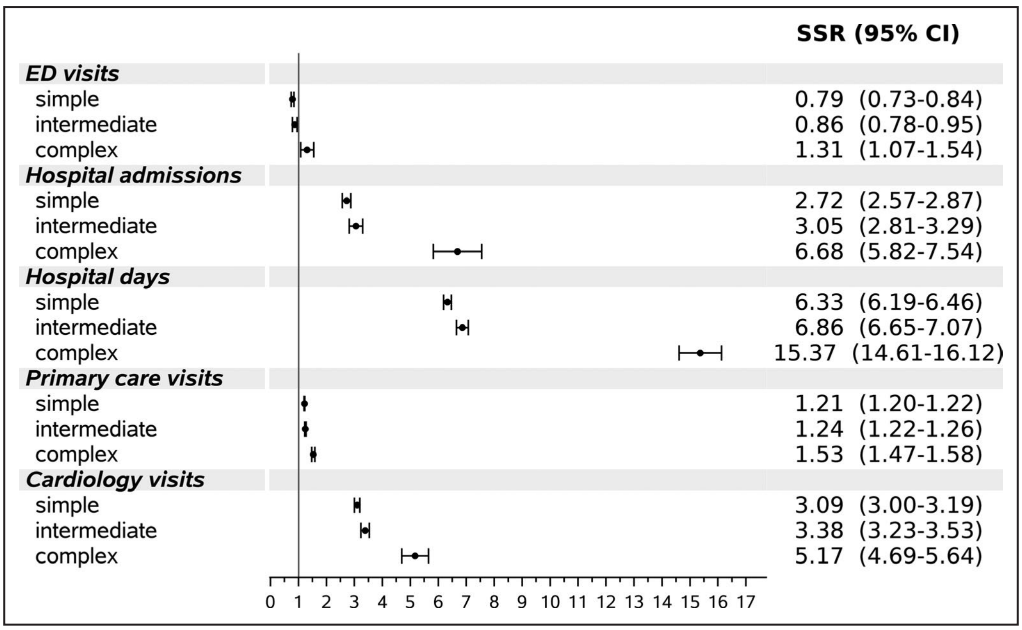

Figure 2. Standardized service utilization rates (SSRs) by congenital heart disease complexity. Rates matched for age, sex and ethnicity among patients with adult congenital heart disease aged 25 to 74 years compared with the general population (extrapolated from the population sample of the Hadera District Study). ED indicates emergency department.

Table 4. Multivariable-Adjusted Recurrent Health Service HR Among Patients With ACHD

	Primary Care Visits	Outpatient Cardiology Visits	ED Only Visits*	Hospitalizations	Days in Hospital	Days in ICU
	HR (95% CI)	HR (95% CI)	HR (95% CI)	HR (95% CI)	RR (95% CI)	RR (95% CI)
Age (reference=18–24 y), y†						
25–44		1.29 (1.17–1.43)			1.26 (1.05–1.52)	0.91 (0.37–2.22)
45–64		2.12 (1.92–2.34)			1.93 (1.60–2.33)	2.26 (0.90–5.74)
>64		2.63 (2.36–2.93)			4.73 (3.90–5.72)	3.56 (1.38–9.18)
Women (reference =men)	1.23 (1.19–1.27)	0.89 (0.84–0.94)	1.25 (1.16–1.35)	0.90 (0.83–0.97)	0.80 (0.73–0.87)	0.78 (0.53–1.17)
Disease complexity (reference=simple)						
Intermediate	0.97 (0.93–1.01)	0.99 (0.93–1.06)	0.96 (0.87–1.06)	1.02 (0.92–1.13)	0.88 (0.79–0.98)	0.60 (0.36–0.99)
Complex	1.14 (1.06–1.23)	1.40 (1.24–1.59)	1.19 (1.02–1.39)	1.75 (1.49–2.05)	1.71 (1.42–2.05)	1.50 (0.64–3.54)
Residence locality peripherality (reference=very central)						
Central	0.98 (0.94–1.02)	0.94 (0.87–1.00)	0.69 (0.62–0.77)	1.04 (0.94–1.14)	1.08 (0.97–1.200)	1.64 (0.99–2.72)
Medium	1.04 (0.99–1.09)	0.99 (0.91–1.06)	0.93 (0.83–1.03)	1.10 (0.99–1.22)	1.15 (1.02–1.29)	1.23 (0.69–2.21)
Peripheral	1.16 (1.10–1.24)	0.89 (0.81–0.99)	0.68 (0.60–0.77)	1.01 (0.89–1.16)	1.08 (0.92–1.25)	2.58 (1.30–5.10)
Very peripheral	1.15 (1.01–1.31)	0.72 (0.58–0.90)	0.71 (0.56–0.92)	1.10 (0.83–1.46)	1.28 (0.89–1.85)	1.04 (0.22–4.96)
Residence locality ethnicity (reference=Jewish)						
Arab	1.38 (1.28–1.49)	1.09 (0.96–1.25)	1.24 (1.06–1.45)	1.54 (1.29–1.85)	1.02 (0.84–1.24)	0.78 (0.31–1.93)
Mixed	0.96 (0.93–1.00)	0.92 (0.86–0.99)	1.15 (1.04–1.26)	1.03 (0.94–1.13)	0.97 (0.87–1.08)	1.05 (0.65–1.68)
Other congenital defects	1.15 (1.10–1.20)	1.10 (1.01–1.20)	1.18 (1.03–1.36)	1.03 (0.88–1.20)	0.86 (0.76–0.99)	1.28 (0.62–2.65)
Charlson comorbidity score	1.09 (1.08–1.10)	1.07 (1.06–1.09)	1.12 (1.09–1.16)	1.19 (1.17–1.22)	1.32 (1.28–1.35)	1.32 (1.14–1.54)

Relative mean number of days in hospital or days in intensive care unit (ICU) were computed with negative binomial count models (see Methods). ACHD indicates adult congenital heart disease; HR, hazard ratio (for recurrent visits and hospitalization, computed with Cox proportional hazard models accounting for the competing risk of death) (see Methods); ICU, intensive care unit; and RR, rate ratio.

*Emergency department (ED) visits—visits to the ED not leading to hospitalization.

†Models were stratified by age groups when the proportional hazard assumption was violated (primary care visit, ED visits, and hospitalization). In addition to the variables in the table models were also adjusted for health service provider, geographic periphery index, ethnicity, other congenital defects, and baseline comorbidities.

the increase in cardiology visits or hospital admission rates, with higher disease complexity, diminished with age as acquired comorbidities play a greater role in healthcare demands.

Women visited a general practitioner more often and a cardiologist less often than men, and while they were also more likely to visit an ED, they were less likely to be hospitalized. These differences were not explained by congenital disease complexity or comorbidity in a multivariable model and may stem from sex-related differences in healthcare utilization present in the general population. Our finding of higher mortality rates among men is supported by data from the European Heart Survey on ACHD.[30] Higher hospitalization rates among women compared with men, reported by others,[13] may be explained by the inclusion of maternity-related hospitalizations.

Less optimal utilization patterns found in geographic periphery and minority localities, albeit universal access to care, may reflect lower accessibility of specialized ACHD care related to distance, language, and health literacy. Similar utilization patterns were reported by others for Israeli Arabs as well as for other minority and low socioeconomic population groups.[31]

General cardiologists treating patients with ACHD were reported to deviate from guidelines more often and have higher rates of adverse patient sequels.[32]

Strengths and Limitations

The use of electronic patient records enabled adequate sample size and population-based assessments of healthcare utilization rates and patterns among patients with ACHD. A challenge of recurrent event analysis is the possible interaction of a death with recurrent events, which may result in biased estimates.[24] Accounting for this is one of the strengths of the current study.

A number of limitations deserve notice. First, because of data limitations, we could not distinguish between general cardiology and specialized ACHD clinic visits recommended by current guidelines. While the distance to the nearest specialized clinic, as a proxy, contributed little beyond peripheral residency in multivariable analysis, differential access to specialized clinics among minority and peripheral population sections cannot be ruled out. Evaluations of SSR and standardized mortality ratios were limited to patients aged 25 to 74 years, which hindered any conclusion regarding younger or older patients with ACHD.

Second, *ICD-9* codes have been generally used for identification and classification of patients with ACHD in studies based on administrative data (for review see reference 33), albeit recognized limitations.[34,35] The single code (745.5) shared by atrial septal defect and patent foramen ovale may lead to overestimation of simple congenital heart disease prevalence and healthcare resource utilization among them, when patent foramen ovale is incidentally detected. In the current study, 25% of the patients were identified solely by this code, yet results of a sensitivity analysis, excluding these patients, were similar to the main results. Additionally, the accuracy of identification and classification of patients with ACHD based on *ICD-9* codes was criticized, particularly among patients with low disease complexity.[35] Deidentified data in our study preclude direct assessment of misclassification in the current study.

Last, data were collected through December 2011. Nevertheless, the overall structure of the Israeli healthcare system, including the number of facilities and services provided, have not significantly changed since then. While diagnostic and therapeutic techniques are constantly evolving, no substantial changes have occurred over the past decade that would have rendered our results irrelevant.

CONCLUSIONS

Patients with ACHD had high healthcare utilization rates compared with the general population. Female sex, geographic periphery, and ethnic minority were associated with higher primary care visit rates, and female sex and periphery with lower outpatient cardiology visit rates. This outpatient service pattern was associated with higher hospitalization rates among Arabs and higher relative number of days in the intensive care unit for periphery. Greater disparities may be found in health systems based on private insurance. Further research should examine health policies and their implementation according to current guidelines in different parts of the patient population and in different settings.

APPENDIX

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Disclosures

None.

Supplementary Material

Tables S1–S5
Figures S1–S4

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SUPPLEMENTAL MATERIAL

Table S1. Congenital heart disease diagnosis code groups (a) and congenital heart disease related procedures used to identify congenital lesions (b)

a. Heart defect group	ICD-9 code
Aortic valve stenosis/insufficiency	746.3, 746.4
Transposition of great arteries (TGA)	745.1
Atrial Septal Defect (ASD)	745.5
Mitral valve stenosis/insufficiency	746.5, 746.6, 746.7
Pulmonary Valve anomaly (PV) anomaly	746.0
Anomalies of the Aorta	747.1, 747.2
Ebstein anomaly of Tricuspid Valve	746.2
Tetralogy of Fallot (TOF)	745.2
Common/single ventricle	745.3, 746.01, 746.1
Ventricular septal defect (VSD)	745.4
Endocardial cushion	745.6
Patent ductus arteriosus (PDA)	747.0
Other defects	648.51, 648.52, 648.53, 746.9, 745.0, 745.11, 745.7, 747.3, 747.40, 747.49, 745.8, 745.9, 746., 746.8

b. Procedure	
PROSTH REP HRT SEPTA NOS	35.50
PROS REP ATRIAL DEF-OPN	35.51
PROS REPAIR ATRIA DEF-CL	35.52
PROST REPAIR VENTRIC DEF	35.53
PROS REP VSD	35.55
GRFT REPAIR HRT SEPT NOS	35.60
GRAFT REPAIR ATRIAL DEF	35.61
GRAFT REPAIR VENTRIC DEF	35.62
HEART SEPTA REPAIR NOS	35.70
ATRIA SEPTA DEF REP NEC	35.71
VENTR SEPTA DEF REP NEC	35.72
ENDOCAR CUSHION REP NEC	35.73
TOT REPAIR TETRAL FALLOT	35.81
TOT COR TRANSPOS GRT VES	35.84
INTERAT VEN RETRN TRANSP	35.91
CONDUIT RT VENT-PUL ART	35.92
CONDUIT ARTIUM-PULM ART	35.94
CAVAL-PULMON ART ANASTOM	39.21

Table S2. Distribution of non-cardiac congenital defects.

	N	(%)
Neurologic	22	(0.2)
Cranio-Face	129	(1.1)
Respiratory	35	(0.3)
Gastrointestinal	24	(0.2)
Genitourinary	210	(1.8)
Muscoskeletal	297	(2.5)
Genetic syndromes		
Marfan syndrome	22	(0.2)
Down syndrome	46	(0.4)

Table S3. Characteristics and health service utilization frequency of 11,653 ACHD patients with congenital heart disease.

	Sex		p	Disease complexity			p
	Men	Women		Simple	Intermediate	Complex	
Cardiac Morbidity N (%)							
Arrhythmia	916 (16.5)	1154 (18.9)	0.0007	1512 (17.5)	431 (17.5)	127 (22.7)	0.007
Valve disease	916 (16.5)	1154 (18.9)	<.0001	527 (6.1)	351 (14.3)	113 (20.2)	<.0001
Ischemic HD	997 (18.0)	749 (12.3)	<.0001	1313 (15.2)	364 (14.8)	69 (12.3)	0.2
Heart failure	472 (8.5)	436 (7.1)	0.006	630 (7.3)	203 (8.3)	75 (13.4)	<.0001
Other HD	924 (16.6)	888 (14.6)	0.002	1176 (13.6)	501 (20.4)	135 (24.2)	<.0001
Other Morbidity N (%)							
Diabetes mellitus	728 (13.1)	808 (13.2)	0.8	1148 (13.3)	339 (13.8)	49 (8.8)	0.005
Hypertension	1773 (31.9)	1981 (32.5)	0.5	2799 (32.4)	829 (33.7)	126 (22.5)	<.0001
hyperlipidemia	1619 (29.2)	1718 (28.2)	0.2	2512 (29.1)	712 (29.0)	113 (20.2)	<.0001
Stroke/TIA	729 (13.1)	846 (13.9)	0.2	1289 (14.9)	240 (9.8)	46 (8.2)	<.0001
Kidney failure	30 (0.5)	14 (0.2)	0.006	34 (0.4)	9 (0.4)	1 (0.2)	0.7
Residence locality ethnicity N (%)							
Jewish / Other	3849 (69.4)	4246 (69.8)	0.2	565 (6.6)	167 (6.8)	60 (10.8)	0.003
Arab	403 (7.3)	389 (6.4)		5992 (69.6)	1732 (70.6)	371 (66.5)	

	Sex					Disease complexity							
	Men		Women		p	Simple		Intermediate		Complex		p
Mixed	1293	(23.3)	1444	(23.8)		2056	(23.9)	554	(22.6)	127	(22.8)	
Residence locality Peripherality N (%)												
Very peripheral	121	(2.2)	106	(1.7)	0.3	174	(2.0)	47	(1.9)	6	(1.1)	0.01
Peripheral	710	(12.8)	749	(12.3)		1052	(12.2)	334	(13.6)	73	(13.1)	
Medium	996	(18.0)	1097	(18.0)		1546	(18.0)	443	(18.1)	104	(18.6)	
Central	1140	(20.6)	1314	(21.6)		1890	(21.9)	451	(18.4)	113	(20.3)	
Very Central	2576	(46.5)	2813	(46.3)		3950	(45.9)	1177	(48.0)	262	(47.0)	
Service utilization Frequency/5 years Median (IQR)												
Primary care visits	28	(15-49)	36	(21-58)	<.0001	33	(18-55)	32	(16-54)	32	(17-57)	0.04
Outpatient Cardiology visits	3	(1-7)	3	(1-6)	0.03	3	(1-6)	3	(1-7)	3	(1-8)	0.0001
ED only visits	0	(0-2)	1	(0 -2)	<.0001	1	(0-2)	0	(0-2)	1	(0-2)	0.06
Hospital admissions	1	(0-2)	1	(0-2)	0.8	1	(0-2)	1	(0-2)	1	(0-3)	0.002
Days in hospital	7	(3-20)	6	(2-19)	0.0004	7	(2-19)	7	(2-19)	10	(2-33)	0.002
Days in ICU	0	(0-0)	0	(0-0)	<.0001	0	(0-0)	0	(0-0)	0	(0-1)	<.0001

ED-emergency department, HD- heart disease; ICU-intensive care unit, IQR-interquartile range; TIA-transient ischemic attack

Table S4. Health service utlization hazard under age and congenital heart disease complexity interaction.

		Primary care visits		Cardiology outpatient visits	ED visits	Hospitalization
Age	**Complexity** (ref=simple)	**HR**	**(95% CI)**	**HR (95% CI)**	**HR (95% CI)**	**HR (95% CI)**
18-24	Complex	0.92	(0.86-0.97)	1.50 (1.33-1.69)	1.01 (0.82-1.26)	1.59 (1.17-2.18)
	Intermediate	0.96	(0.92-1.01)	1.15 (1.04-1.27)	0.90 (0.75-1.08)	0.90 (0.62-1.32)
25-44	Complex	1.04	(1.00-1.09)	1.21 (1.12-1.31)	1.11 (0.98-1.27)	1.45 (1.24-1.69)
	Intermediate	1.00	(0.98-1.02)	1.03 (0.97-1.09)	0.93 (0.85-1.01)	0.98 (0.86-1.11)
45-64	Complex	0.99	(0.93-1.06)	1.10 (0.98-1.23)	1.22 (0.99-1.50)	1.42 (1.19-1.69)
	Intermediate	0.99	(0.97-1.02)	1.02 (0.97-1.06)	1.00 (0.88-1.14)	1.04 (0.94-1.15)
>64	Complex	0.97	(0.91-1.04)	1.03 (0.92-1.15)	1.11 (0.86-1.44)	1.35 (1.11-1.64)
	Intermediate	0.98	(0.95-1.01)	0.99 (0.94-1.04)	0.99 (0.87-1.13)	0.94 (0.86-1.04)
P for Interaction		0.18		0.0004	0.5	0.99

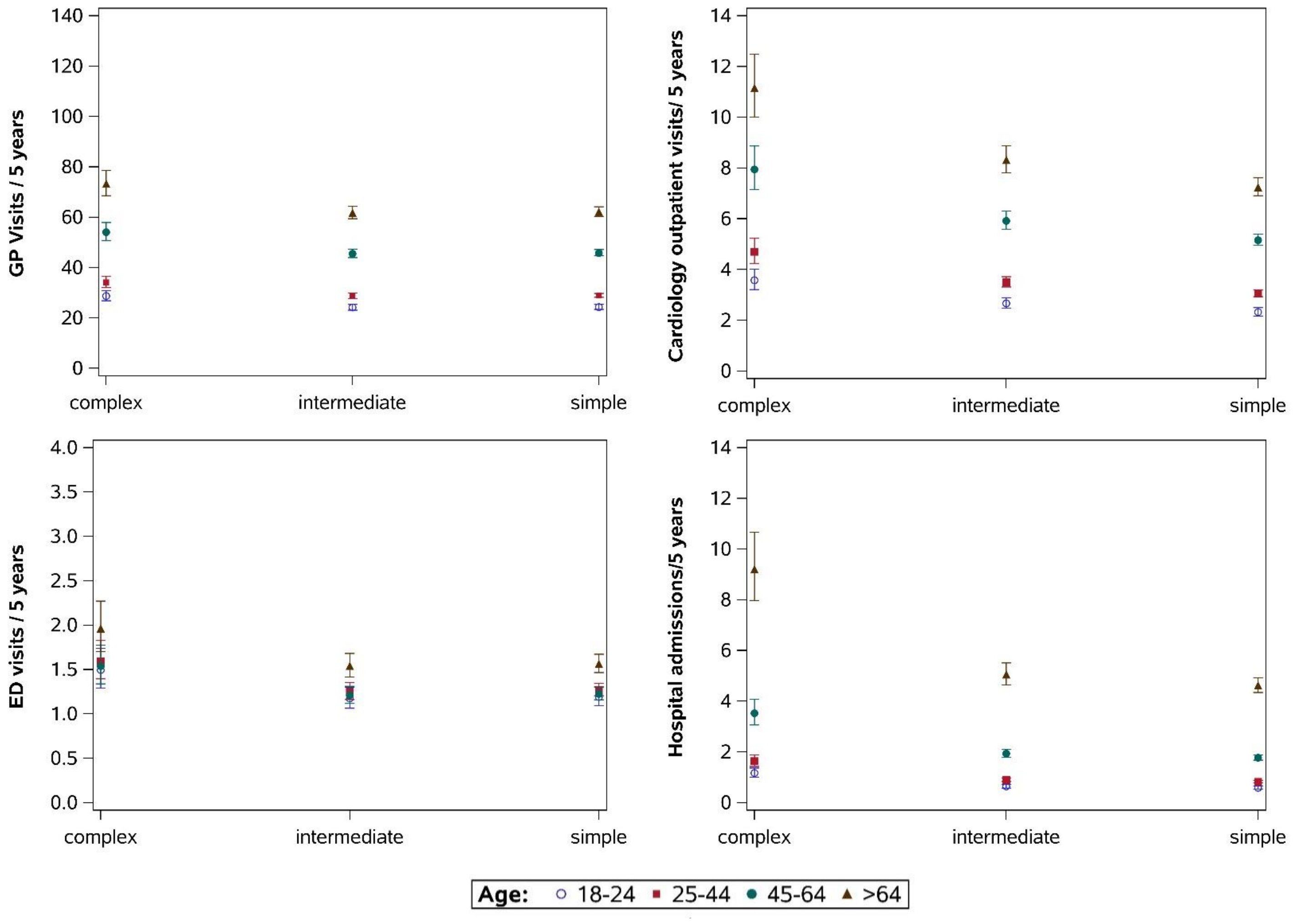
CI- confidence interval, ED- emergency department, HR- hazard ratio in strata under interaction. Adjusted for: sex, residence peripherality, ethnicity, other congenital defects, comorbidity, health service provider, geographical periphery index, ethnicity, other congenital defects, and baseline comorbidity.

Table S5. Sensitivity analysis- Multivariable adjusted recurrent health service hazard ratio- Excluding patients identified solely by ASD/PFO.

	Primary care visits	**Outpatient Cardiology visits**	**ED visits***	**Hospitalizations**
	HR (95% CI)	**HR (95% CI)**	**HR(95% CI)**	**HR(95% CI)**
Age (ref=18-24)				
25-44		1.27 (1.14-1.41)		
45-64		2.15 (1.93-2.40)		
>64		2.56 (2.27-2.89)		
Men (Ref=Women)	0.81 (0.78-0.84)	1.16 (1.09-1.23)	0.79 (0.72-0.86)	1.06 (0.97-1.16)
Disease complexity (Ref=Simple)				
Intermediate	0.98 (0.94-1.02)	0.98 (0.92-1.05)	0.98 (0.88-1.09)	1.02 (0.91-1.14)
complex	1.15 (1.07-1.24)	1.39 (1.22-1.57)	1.21 (1.03-1.42)	1.75 (1.48-2.07)
Residence locality Peripherality (Ref=Very central)				
Central	0.99 (0.95-1.04)	0.90 (0.83-0.97)	0.66 (0.58-0.74)	1.01 (0.89-1.14)
Medium	1.07 (1.01-1.13)	1.04 (0.95-1.13)	0.90 (0.79-1.02)	1.18 (1.04-1.35)
Peripheral	1.18 (1.10-1.27)	0.88 (0.79-0.99)	0.62 (0.54-0.72)	1.04 (0.90-1.22)
Very peripheral	1.26 (1.06-1.49)	0.86 (0.67-1.09)	0.79 (0.56-1.10)	1.07 (0.77-1.50)
Residence locality ethnicity (Ref=Jewish)				
Arab	1.40 (1.28-1.53)	1.16 (1.00-1.35)	1.26 (1.04-1.52)	1.49 (1.20-1.84)
Mixed	0.98 (0.93-1.03)	0.94 (0.87-1.01)	1.13 (1.00-1.26)	1.06 (0.95-1.19)
Other congenital defects	1.16 (1.10-1.22)	1.14 (1.04-1.25)	1.23 (1.04-1.44)	1.05 (0.87-1.26)
Charlson comorbidity score	1.10 (1.08-1.11)	1.08 (1.06-1.10)	1.13 (1.09-1.16)	1.21 (1.18-1.25)

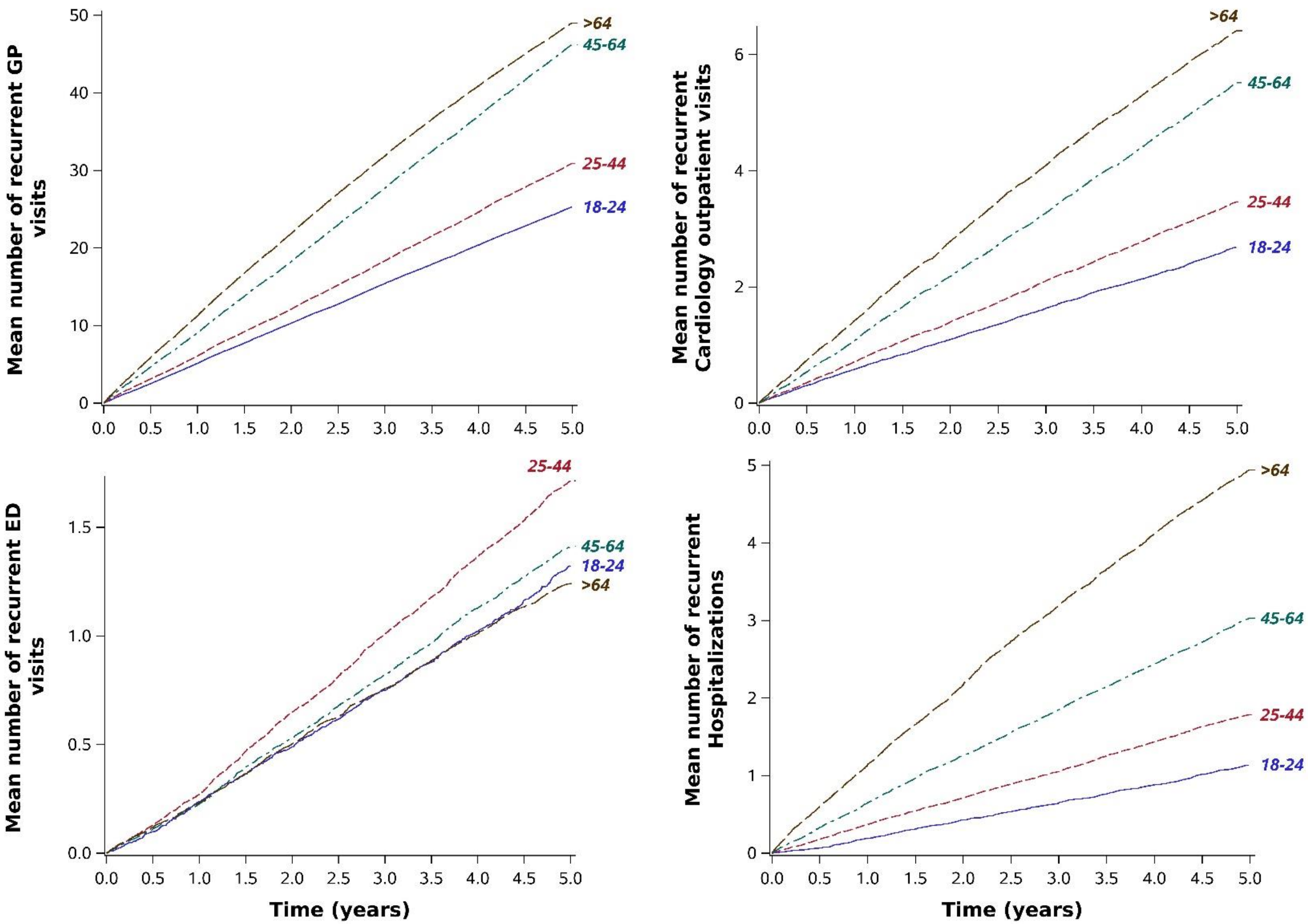
CI- confidence interval; ED-emergency department; HR- Hazard ratios for recurrent visits and hospitalization, computed with cox proportional hazard models accounting for the competing risk of death (see methods); ICU-intensive care unit; Ref-reference. Models were stratified by age groups when the proportional hazard assumption was violated (GP visit, ED visits and hospitalization). In addition to the variables in the table, models were also adjusted for: Health service provider, geographical periphery index, ethnicity, other congenital defects, and baseline comorbidity.

Figure S1. **Health service utilization (number of visits) by age and congenital disease complexity.**



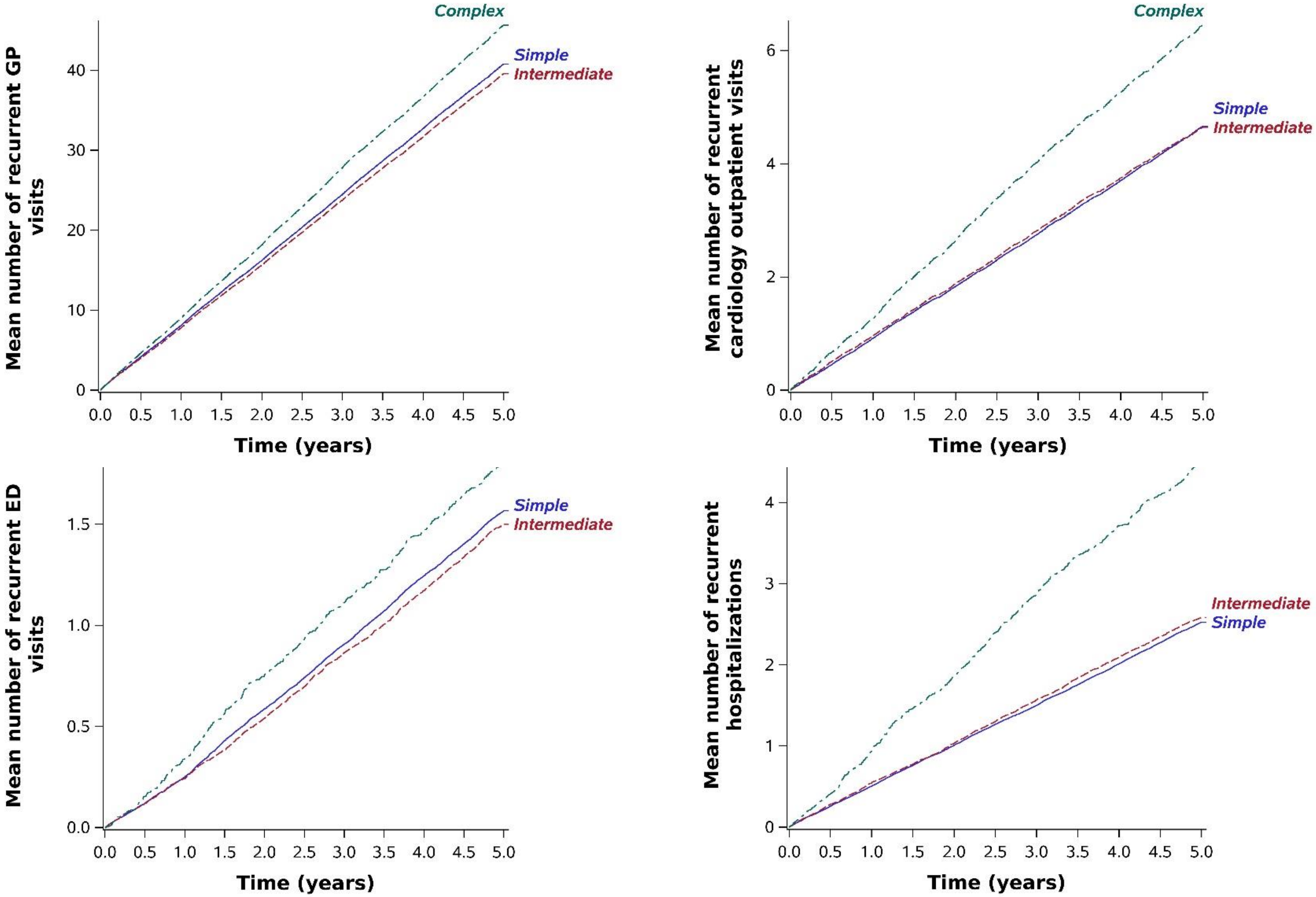
GP- general practice, ED- emergency department.

Figure S2. **Multivariable adjusted cumulative expected number of health service utilizations by age.**



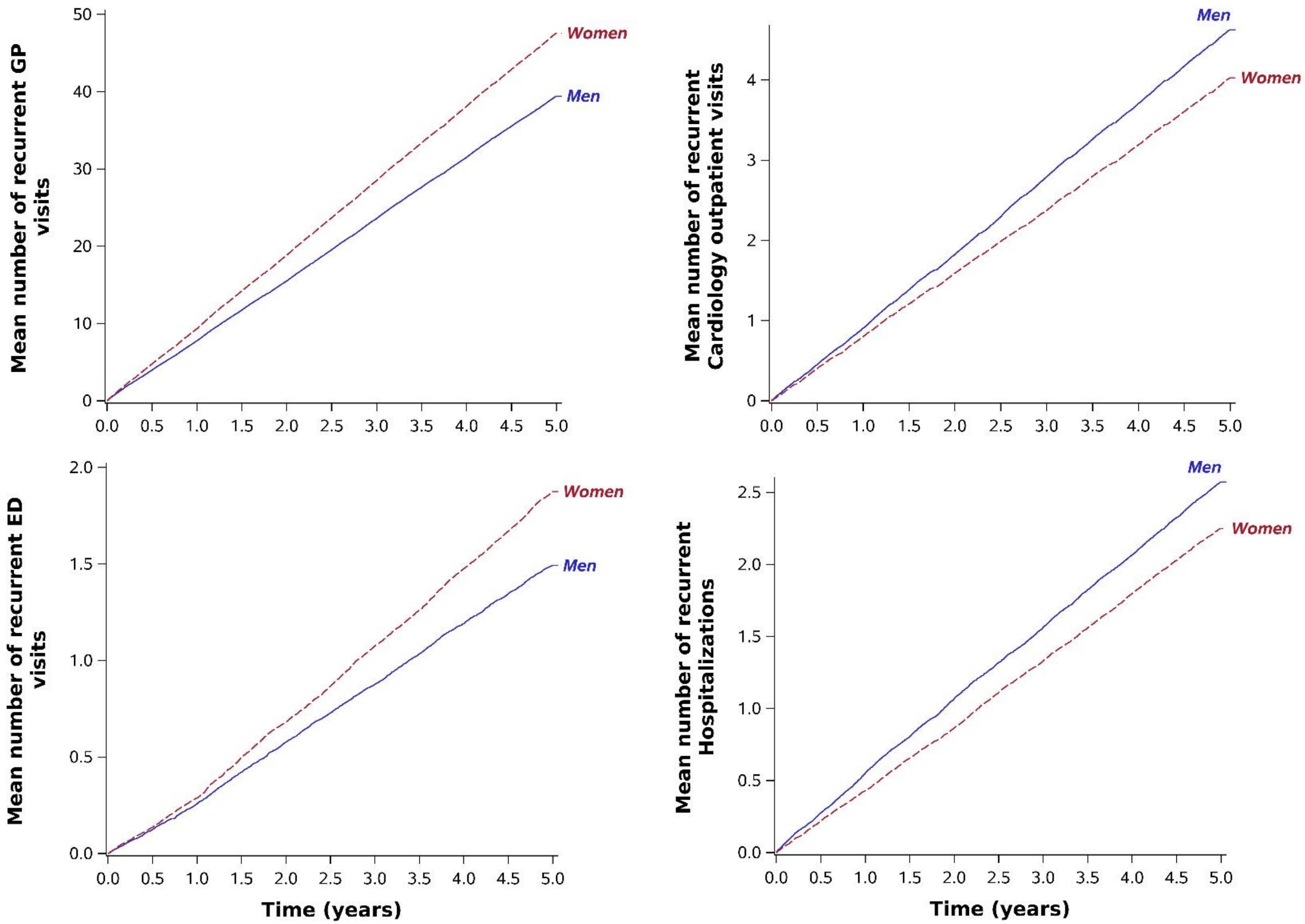
GP- general practice, ED- emergency department.

Figure S3. **Multivariable adjusted cumulative expected number of health service utilization by congenital heart disease complexity.**



GP- general practice, ED- emergency department

Figure S4. **Multivariable adjusted cumulative expected number of health service utilization by sex.**



GP- general practice, ED- emergency department.