Cardiology in the Young

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Real-world healthcare utilization in adult congenital heart disease: a systematic review of trends and ratios

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Review Article

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Abstract

Background: As a result of medical advances, the adult congenital heart disease population is rapidly expanding. Nonetheless, most patients remain prone to increased morbidity and mortality. Therefore, long-term medical resource use is required. This systematic review aims to present the trends over the past decades of medical resource utilization in adult congenital heart disease as well as its current status, with a focus on hospitalizations, emergency department visits, outpatient cardiology visits, and visits to other healthcare professionals. *Methods:* MEDLINE (Pubmed), Embase, and Web of Science were searched for retrospective database research publications. The ISPOR checklist for retrospective database research was used for quality appraisal. Trends over time are explored. *Results:* Twenty-one articles met the inclusion criteria. All but one of the studies was conducted in Western Europe and North America. The absolute number of hospitalizations has been increasing over the last several decades. This increase is highest in patients with mild lesions, although these numbers are largely driven by hospitalizations of patients with an atrial septal defect or a patent foramen ovale. Meanwhile, outpatient cardiology visits are increasing at an even higher pace, and occur most often in geriatric patients and patients with severe lesions. Conversely, the number of hospitalizations per 100 patients is decreasing over time. Literature is scarce on other types of healthcare use. *Conclusion:* A strong rise in healthcare utilization is noticed, despite the mitigating effect of improved efficiency levels. As the population continues to grow, innovative medical management strategies will be required to accommodate its increasing healthcare utilization.

The worldwide birth prevalence of congenital heart disease stabilized over the last two decades to 9.2 per 1000 births.[1] Survival into adulthood improved remarkably, as a result of medical progress and a more advanced organization of care. In addition, survival in adults with congenital heart disease has benefited from general healthcare improvements. The largest mortality reductions were observed in patients with severe lesions.[2] Consequently, the adult patient population is rapidly expanding. Marelli et al reported an 85% increase in prevalence between 1985 and 2000, [3] and a further 57% increase in the first decade of the 21st century.[4] Adult patients now outnumber pediatric patients by two to one.[4]

The majority of adults with congenital heart disease cannot be considered to be cured. The disease is characterized by a chronic course and morbidity and mortality risks are still high compared to the general population.[5,6] Therefore, adult congenital heart disease programs have been developed to periodically follow up on patients, in order to detect potential health problems at an early stage and to deal appropriately with recurrent care needs.[7] The increased prevalence together with the need for follow-up is expected to lead to an increase in healthcare utilization.

A good understanding of healthcare utilization of adult patients is necessary to develop appropriate healthcare structures. One recent scoping review[8] already provided a brief description of some aspects of healthcare utilization but a comprehensive systematic literature overview is lacking. Therefore, the current systematic review aims to present the trends over the past decades of real-world medical resource utilization as well as its current status, with a particular focus on hospitalizations, emergency department visits, outpatient cardiology visits, and visits to other healthcare professionals.

Materials and methods

Search strategy

Three bibliographical databases, MEDLINE (via Pubmed), Embase (via embase.com) and Web of Science Core Collection (via Web of Science), were searched for scientific publications as of 19 December, 2017. The applied search strategy consisted of patient population-related keywords (adult, grown-up, congenital, heart, cardiac, disease, defect, achd, and guch) in combination

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with keywords to identify relevant articles on resource utilization (health, healthcare, utilization, resources, hospitalization, admission, readmission, and visits). The search strategy was modified to the particular databases' properties and can be found in Supplementary Table S1.

Eligibility criteria

Studies were eligible for this review if they complied with the following inclusion criteria:

- The publication describes a population of patients in a real world context. Adulthood is defined as 18 years and older, or patients who have already transitioned from the pediatric clinic to the specialized adult congenital heart center. Age at transition differs across countries, and it is always individual dependent.
- The publication includes a description of medical resource utilization. Resource utilization is defined as hospitalization, emergency department visits, outpatient cardiology visits, or other healthcare use (such as general practitioner visits).
- Medical resource utilization is reported as a rate (e.g., a number of hospitalizations per patient year), a percentage (e.g., % of patients who are hospitalized during a given time period) or absolute numbers at more than one point in time (e.g., number of hospitalizations in 2010 and 2011).
- A retrospective database research was conducted.
- The manuscript was written in English, Dutch, or French.

Studies with a mixed sample of pediatric and adult patients were excluded if the reported results were not age stratified. Studies were also excluded if the patient population was defined based on a specific complication (e.g., patients with arrhythmia or patients with heart failure). However, studies describing medical resource utilization stratified for the lesion's complexity grade (e.g., single ventricle patients) were included. No restriction was applied on the date of publication. The corresponding author was contacted if the full text article was not accessible online.

Data extraction

Two reviewers (R.W. and A.W.) independently reviewed all titles and abstracts of candidate publications. Discrepancies were discussed until consensus was reached. Eligible full texts were screened by the former (R.W.). The following predetermined information was retrieved from the included studies:

- General information: study year, country, funding, conflict of interest, study design, database, study's time period.
- Population characteristics: lesion type, sample size, age, gender, disease distribution, complexity distribution (mild, moderate, severe, undefined), complexity definition (e.g., 32nd Bethesda conference), morbidity, and mortality.
- Clinical characteristics: e.g., teaching status, tertiary center, acute center, rural/urban center.
- Medical resource utilization: hospitalization, emergency department visits, outpatient physician visits, and other healthcare seeking behaviour.

In order to improve the comparability between studies, we recalculated the increase in medical resource utilization over time to a "percentage increase per year". We stratified results for mild, moderate, and severe lesions as defined by Task Force 1 of the 32nd Bethesda Conference.[9]

Quality appraisal

As the selected studies were based on retrospective database research, the ISPOR checklist for retrospective database studies[10] was used for quality appraisal. This checklist was initially developed for, but it is not limited to, medical claims or encounter-based databases. Studies based on disease registries or national survey databases can potentially be assessed as well.[10] A score of "1", "0" or "not applicable" was assigned to each question. The average score is an indication of the quality of the study. Two authors (R.W. and A.W.) independently assessed one article. Discrepancies were discussed until consensus was reached by defining the assessment criteria (Supplementary Table S2). The former (R.W.) subsequently assessed all remaining articles.

Results

Figure 1 displays the selection process' flow chart. After removal of duplicate records, 1605 articles were screened by title and abstract. Seventy-five articles were found eligible for full text screening. One article was not screened, as the full text could not be retrieved.[11] Two additional articles were found by the snowball search method.[12,13] Three articles reported on the same study population.[14–16] Only one of these three articles, covering all relevant information, was selected for inclusion.[14] Twenty-one articles in total fitted the inclusion criteria (Table 1).

Study characteristics

The included studies were published between 1999 and 2017. Respectively nine, four, and seven studies originated in the United States of America, Canada, and Western Europe. Only one Chinese study was included. The majority of the studies (71%) reported on hospitalizations; one third of the included studies reported on outpatient cardiology visits; five studies examined emergency department visits; one study described visits to other healthcare providers. Most studies included samples with a broad range of CHD types. Two studies focused on patients with a single ventricle,[17,18] one study focused on patients with tetralogy of Fallot,[19] and one study focused on patients with a single ventricle, tetralogy of Fallot, or corrected transposition of the great arteries (mainly atrial switch).[20]

We identified four methods in the reporting of medical resource utilization. The first two categories reflect the prevalence of the growing population and its impact on the broad community. The last two categories focus on the care provided to the adult congenital heart disease population and reflect the evolution of medical management (Table 2):

(i) in absolute numbers: the total number of instance hospitalizations in a defined time period (usually a year).
(ii) per 100,000 general population: this measurement reflects for instance the impact of adult congenital heart disease-related hospitalizations on the total population in a defined time period. The absolute number of hospitalizations (numerator) is divided by the total population (denominator) and multiplied by 100,000.
(iii) per 100 patients: this measurement divides for instance the absolute number of hospitalizations (numerator) by the number of adult congenital heart disease patients (denominator) in a defined time period and is then multiplied by 100.

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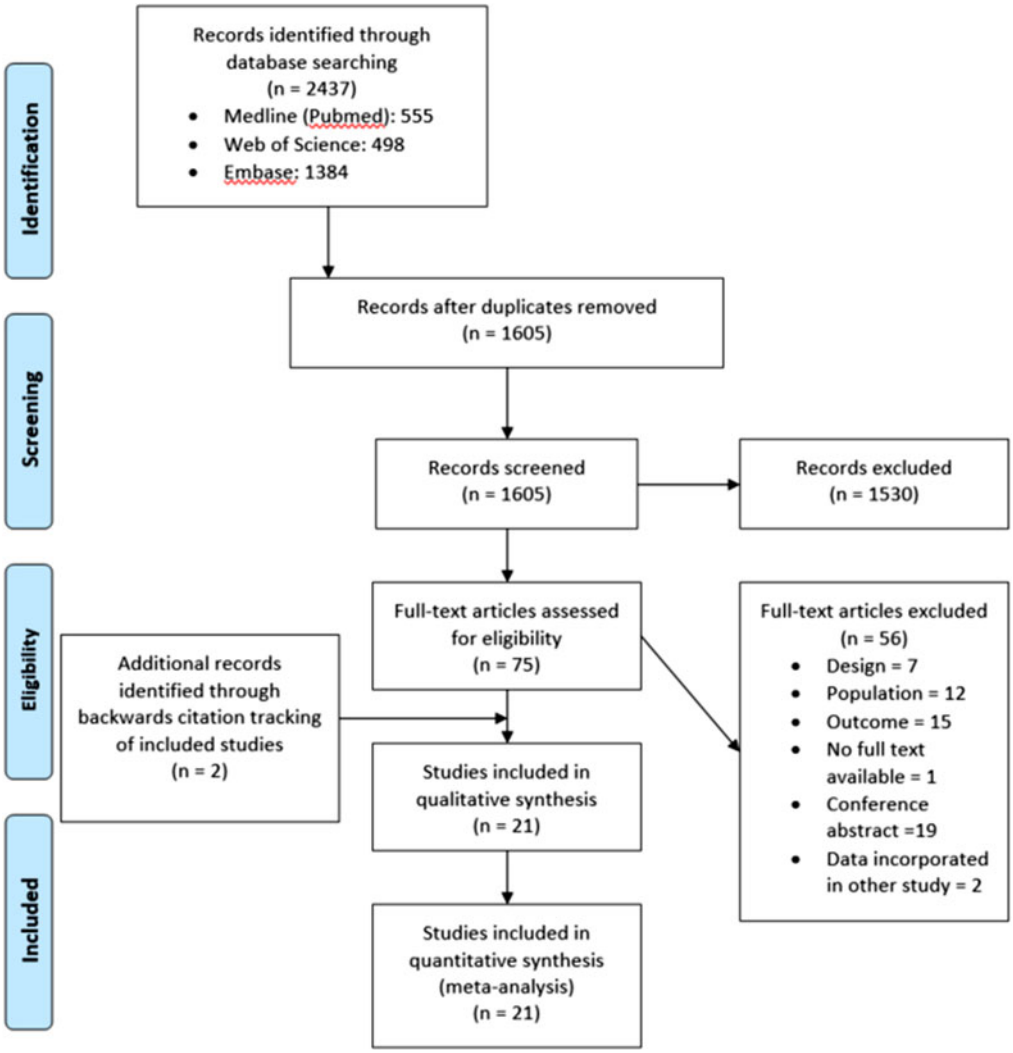

Figure 1. Prisma flow diagram.

(iv) per 100 patient years: this measurement is similar to the "per 100 patient" –measurement but also takes mortality into account.

Hospitalization

Absolute numbers

Most studies described a rapidly growing hospitalization burden in adults with congenital heart disease. In the last two decades, hospitalization increased 3.45–10.56% per year. The exclusion of patients with an atrial septal defect or a patent foramen ovale shows more modest, though still high, estimates up to 7.66% per year.[21–25] Some evidence indicated a declining growth rate of hospital admissions in more recent times.[18,19]

Hospitalization numbers of patients with mild lesions increased 8.09–12.63% per year.[21,24,25] As previously mentioned, these numbers are largely driven by hospitalizations of patients with an atrial septal defect or a patent foramen ovale (for defect closure). Excluding these patients, reported yearly increases were 3.13–7.47%.[21,24] One study from the United Kingdom of moderate and severe lesion-related hospitalizations found small yearly increases of only 0.42%.[26] All other studies reported a yearly 2.86–6.92% rise in the hospitalization of patients with moderate and severe lesions.[17–19,21,24–26] Hospitalization numbers of patients with an unclassified lesion increased 3.41–10.22% per year.[21,24–26] A growing proportion of all adult congenital heart disease-related hospitalizations can be attributed to the hospitalization of patients with mild lesions, with the proportion increasing by 1.04% per year in absolute terms. However, when excluding patients with an atrial septal defect and a patent foramen ovale, the proportion remained invariable over time with mild lesions accounting for 39% of all hospitalizations.[24]

Hospitalizations of geriatric (+65) patients increased at a higher yearly pace (5.26% versus 1.95 and 2.66% in age categories 18–39 and 40–64 years, respectively).[23] Additionally, the number of single ventricle patients admitted to pediatric hospitals is decreasing (–5.57% per year) while admissions to general hospitals are increasing (+4.61% per year).[17]

Per 100,000 general population

Males across different age categories (25–44, 45–64 and 65+ years) were significantly less frequently hospitalized than females in 2004 (12.4 versus 14.5, 11.9 versus 12.0, and 6.4 versus 7.2, respectively).[22] A Chinese study reported lower hospitalization rates

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Table 1. Overview of the included studies

Study (Quality score)	Country	Study period	Sample	Cohort's disease complexity distribution	Outcome	Main results
Agarwal 2016a[31] (0.65)	USA	2006–2012	72,090 ED visits	32.4% mild* 42.5% moderate/severe* 25.1% unclassified* ASD and PFO excluded if isolated	Change in the absolute number of ED visits Proportion hospitalization after ED visit	+6.59 to +10.24%/year 66.9–70.1% Admission rate decreases over time among patients with a mild and a complex lesion
Agarwal 2016b[21] (0.70)	USA	2003–2012	195,306 hospitalizations	65.9% mild * 24.6% moderate/severe* 9.5% unclassified*	Change in the absolute number of hospitalizations Change in the proportion of hospitalized patients first attending the ED	+3.13 to +10.12%/year +0.60 to +1.65%/year mild without ASD and PFO: non-significant
Billett 2007[22] (0.47)	UK	1995–2004	2766–3851 hospitalizations per year	Unknown	Change in the hospitalization rate per 100,000 population Change in the absolute number of hospitalizations	+1.96 to +5.27%/year +4.22%/year males < females
Briston 2016[26] (0.55)	USA	2002–2012	14,307–14,915 discharges per year	71.75% moderate* 28.25% severe*	Change in the absolute number of hospitalization (discharges)	−0.11 to +1.02%
Cedars 2016[20] (0.60)	USA	1996–2014	352 patients	45% TOF 36% TGA of which 68% atrial switch 19% SV	Hospitalization rate per 100 patient years Relative risk of hospitalization versus general population	39–72 4–8
Collins 2016[17] (0.71)	USA	2001–2011	8,330 hospitalizations	100% SV	Change in the absolute number of hospitalizations Change in the hospitalization to general hospitals Change in the hospitalization to pediatric hospitals	+3.06%/year +4.61%/year −5.57%/year
Cui 2017[12] (0.65)	CHN	2007–2011	15,919 patients	Unknown	Hospitalization rate per 100,000 population Change in the hospitalization rate per 100,000 population	0.1–7.3 Females > males 0 to +2.25%/year
Engelfriet 2005[14] (0.43)	EU	1998–2003	4109 patients	21% ASD 15% VSD 20% TOF 13% aortic coarctation 9% TGA 7% Marfan 5% Fontan circulation 9% cyanotic defect	Outpatient cardiology visits per 100 patient years	114–192
Gatzoulis 1999[32] (0.35)	CAN	1987–1997	570 patients	Unknown	Change in the absolute number of outpatient cardiology visits	+7.57 to +21.89%/year
Islam 2016[23] (0.65)	CAN	2003–2012	23,749 patients	All CHD except isolated patent ductus arteriosus No stratification to severity for adults available.	Change in the absolute number of hospitalizations Proportion of hospitalized patients needing multiple hospitalizations Change in hospitalization rate per 100,000 population Change in the hospitalization rate per 100 ACHD population Hospitalization rate per 100 ACHD population	+1.95 to 5.26%/year 40.9% +1.7 to 2.66%/year −4%/year 3.9–5.5

Kempny 2016[33] (0.75)	UK	1991–2010	4461 patients	43% mild* 36% moderate* 19% severe* 2% undefined*	Change in the absolute numbers of outpatient cardiology visits Change in absolute numbers of outpatient cardiologist appointments made Change in the absolute numbers of non-attendance	+8.22%/year +9.40%/year +13.24%/year
Mackie 2007[30] (0.76)	CAN	1996–2000	22,096 patients	92.01% others* 7.99% severe*	Hospitalization rate per 100 ACHD population Proportion of patients attending the ED (median visits) Relative risk patients with a severe lesion versus other ACHD patients Proportion of patient having an outpatient cardiology visit (median visits) Relative risk patients with a severe lesion versus other ACHD patients Proportion of patients attending the general practitioner (median visits) Relative risk patients with a severe lesion versus other ACHD patients Proportion of patients attending a specialist outpatient visits (median visits) Relative risk patients with a severe lesion versus other ACHD patients	20.8–35.4 67.9% (3 visits) 1.09 54.8% (4 visits) 1–2.39 91.1% (15 visits) 0.91 87.2% (10 visits) 1.06
Moons 2001[29] (0.73)	BEL	1997	192 patients	20.1% TOF, 18.8% Eisenmenger, 10% coarctation of the aorta, 10% aortic valve stenosis, 40% others	Hospitalization rate per 100 ACHD population Proportion of the population requiring a hospitalization	23 20.3%
Mylotte 2014[13] (0.81)	CAN	1990–2005	7943–10036 patients per year	17.5% severe 40.6% shunts 41.9% others Severe is defined as endocardial cushion defect, TOF, truncus arteriosus, TGA, and HLHS/SV	Proportion of patients having an outpatient cardiology visit Subgroups with a higher likelihood of attending a cardiologist Subgroups with a higher likelihood of attending specialized care centers Change in outpatient cardiology visits per 100 ACHD population (1990–1997) Change in outpatient cardiology visits per 100 ACHD population (1997–2005)	24.7% (70% in a specialized center, 27% in a non-referral center and 4.1% was mixed care) Younger age, female, severe complexity lesion, one or more comorbidities Younger age, severe complexity lesion +1.3 to +2.1%/year +5.6 to +7.4%/year
O'Leary 2013[25] (0.55)	USA	1998–2010	953,246 hospitalizations	58.5–66.2% mild* 31.5–43.5% isolated secundum ASD or PFO 11.9–9.7% unclassified* 29.6–24.1% severe*	Change in the absolute numbers of hospitalization Change in the proportion of ACHD admission of all CHD admissions	+6.74 to +15.78%/year +1.12%/year

(*Continued*)

Table 1. (*Continued*)

Study (Quality score)	Country	Study period	Sample	Cohort's disease complexity distribution	Outcome	Main results
Opotowsky 2009[24] (0.65)	USA	1998–2005	35,992–72,656 hospitalizations per year	54–63% mild* 27–35% moderate/severe*	Change in the absolute numbers of hospitalization Change in the proportion of patients with a mild lesion over all ACHD hospitalizations Change in the proportion of patients with a severe lesion over all ACHD hospitalizations Proportion of hospitalized patients first attending the ED	+6.46 to +17.46%/year +1.04%/year −1.01%/year 41.7%
Padrutt 2017[34] (0.58)	SWZ	1996–2015	1725 patients	35% mild* 43% moderate* 22% severe*	Change in the absolute number of outpatient cardiology visits	+11.42%/year
Schmidt 2016[19] (0.65)	USA	2000–2011	15,553 hospitalizations	100% TOF	Change in the absolute numbers of hospitalization	+0.86 to +4.53%/year
Tabtabai 2015[18] (0.65)	USA	2000–2011	11,068 hospitalizations	35.6% SV55% TA13.2% HLHS	Change in the absolute numbers of hospitalization	+3.35 to 6.68%/year
Tutaral 2014[28] (0.57)	UK	2000–2012	5887 patients	44.0% mild* 46.4% moderate* 7.5% severe* 2.2% unclassified*	Hospitalization rate per 100 patient years Outpatient cardiology visits per 100 patient years	9.49–16.40 122.46–159.44
Verheugt 2010[27] (0.53)	NL	2001–2006	5798 patients	40% mild* 49% moderate* 11% severe*	Hospitalization rate per 100 patient years Proportion of ACHD patients in need of a hospitalization 5-year hospitalization rate age 20–30 5-year hospitalization rate age 70–80 Relative risk hospitalization rate per 100 patient years compared to the general population (for age 30+)	30.8 50% 11% 68% 2–3

*=defined by the 32nd Bethesda Conference. ASD: atrium septal defect; BEL: Belgium; CAN: Canada; CHD: congenital heart disease; CHN: China; EU: Europe; HLHS: hypoplastic left heart syndrome; NL: the Netherlands; SV: single ventricle; SWZ: Switzerland; TA: tricuspid atresia; TGA: transposition of the great arteries; TOF: Tetralogy of Fallot; UK: United Kingdom; VSD: ventricle septal defect; USA: United States of America

Table 2. Percentage increase in hospitalizations per year and hospitalization rates, stratified for lesion's complexity grade

	Unclassified	Mild	Moderate	Severe	Total cohort	References
Absolute numbers	+3.41 to +10.22%	+8.09 to +12.63% +3.13 to +7.47%*	+0.42 to +6.92%	+0.42 to +6.92%	+3.45 to +10.56% +3.99 to +7.66%*	17–19,21–26
Per 100 patient years	NDA	NDA	TOF: 39	TGA: 42 SV: 72	9.49–30.8	20,27,28
Per 100 patients	20.8	20.8	20.8	35.4	3.9–23 −4%	23,29,30
Per 100,000 general population	NDA	4.2–7+2.25%	4.2–7+2.25%	−0.3+0.0%	4.3–36 +1.7 to +5.27%	12,22,23
Relative Risk vs general population	2.02	2.02	2.02–4	3.44–8	2–3	20,27,30

Mild/moderate/severe classification is based, in most papers, on the 32nd Bethesda Conference classification.
Percentages are stated as yearly increases.
*Exclusion of patients with an atrial septal defect or a patent foramen ovale.
TGA: transposition of the great arteries; TOF: tetralogy of Fallot; SV: single ventricle;
NDA: no data available

Table 3. Percentage increase in outpatient cardiology visits per year and number of outpatient cardiology visits, stratified for lesion's complexity grade

	Unclassified	Mild	Moderate	Severe	Total cohort	References
Absolute numbers	NDA	NDA	NDA	NDA	+8.22 to +11.42%	32–34
Per 100 patient years	Marfan: 137	ASD: 145 VSD: 114	TOF: 142 CoA: 127	TGA: 152 Fontan: 192 Cyanotic: 186	122–159	14,28
Per 100 patients	+2.1% (1990–1997) +7.1% (1997–2005)	2.1% (1990–1997) 7.1% (1997–2005)	2.1% (1990–1997) 7.1% (1997–2005)	1.3% (incl. TOF; 1990–1997) 5.6% (incl. TOF; 1997–2005)	1.4% (1990–1997) 7.4% (1997–2005)	13
Relative Risk	NDA	NDA	NDA	Versus other lesions 18–40 y: 2.39 41–64 y: 1.67 65+ y: 1.00	Versus general population: 2.24	30

Mild/moderate/severe classification is based, in most papers, on the 32nd Bethesda Conference classification.
Percentages are stated as yearly increases.
ASD: Atrial Septal Defect; CoA: Coarctation Aorta; TGA: Transposition of the Great Arteries; TOF: Tetralogy of Fallot; VSD: Ventricular Septal Defect;
ACHD: adults with congenital heart disease; NDA: no data available.

per 100,000 population, but hospitalization was also remarkably more frequent in females (7.3 versus 4.3).[12]

The hospitalization rate increased 3.3–5.3% per year over an 8-year period, depending on age category and gender. The hospitalization rate in geriatric males increased only by 2% per year.[22] A slower trend was observed by Islam et al[23], reporting a 1.7, 1.0, and 2.5% yearly increase in age categories 18–39, 40–64 and 65+ years, respectively, and by Cui et al[12], who reported a 2.1% yearly increase in the total cohort.

Per 100 patients

A little more than one in five Belgian adults with congenital heart disease was hospitalized over a one-year timeframe; 27.9% of these patients had multiple hospitalizations.[29] Other research reported hospitalizations in half of all Dutch patients, of which 60% needed multiple hospitalizations over a 5-year timeframe.[27] Per 100 patients, 21.4 and 23 hospitalizations were reported amongst Canadian and Belgian patients, respectively.[29,30] On the contrary, another Canadian study reported a hospitalization rate of only five hospitalizations per 100 patients. Substantially, fewer patients needed multiple hospitalizations over a longer timeframe (41% over 10 years). In addition, they found a yearly 4% decrease in hospitalization rate.[23] Adults with congenital heart disease required 108% more hospitalizations than the general population. The hospitalization rate in severe complexity patients was 244% higher.[30]

Per 100 patient years

The hospitalization rate per 100 patient years is generally higher than the hospitalization rate per 100 patients, as it accounts for mortality. A Dutch study reported 30.8 hospitalizations per 100 patient years,[27] but a British study reported hospitalization rates two to three times lower.[28] The hospitalization rate increased with increasing age[27,28] and higher complexity of the lesion, with single ventricle patients recording 72 hospitalizations per 100 patient years, eight times higher than the general population.[20] Moreover, a two-to-threefold higher hospitalization risk for the overall cohort is reported.[27]

Emergency department

In a study by Mackie et al,[30] 67.9% of the patients visited the emergency department over a 5-year timeframe, with a median of three visits. The likelihood to attend an emergency department was 9% higher in patients with severe or moderate lesions compared to mild and unspecified lesions.[30] However, the gap might be gradually closing as the absolute number of emergency department visits

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in patients with mild (7.05% per year) and unspecified (10.24% per year) lesions was growing more rapidly between 2006 and 2012 compared to patients with severe or moderate lesions (6.59% per year).[31] 70 and 66.9% of patients with mild lesions and severe lesions, respectively, attending the emergency department between 2006 and 2012 were subsequently hospitalized. Hospitalization rates after emergency department visit relatively declined by approximately 1.30% per year in both patients with severe and a mild lesions. Patients with severe lesions were however more often transferred to another hospital.[31]

Conversely, over 40% of patients hospitalized between 1998 and 2005 visited the emergency department a priori,[21,24] and this proportion is increasing over time for both patients having either a mild or a severe lesion. The significant proportional increase in patients with mild lesions can be entirely attributed to the vast increase in hospitalized patients with an atrial septal defect or a patent foramen ovale, admitted through the emergency department.[21]

Outpatient cardiology visits

Absolute numbers

A major increase in the number of outpatient visits over the last three decades is generally noted by single-center studies (Table 3). A Canadian study reported an 11.20% yearly increase of outpatient visits to their clinic between 1987 and 1997.[32] A British study reported a yearly 8.22% increase between 1991 and 2010. However, little growth in outpatient visits was observed between 2006 and 2010.[33] A Swiss study reported similar results: the number of outpatient visits substantially increased until 2005. Thereafter, little growth was observed until 2010, after which further sharp growth was observed.[34] Moreover, an even higher increase was noticed in the number of appointments made in the United Kingdom but clinicians there also encountered a rise in no-shows.[33]

Per 100 patients

Outpatient visits to specialized centers per 100 patients increased steadily (1.4% per year) before the inflexion point in 1997. Afterwards, a yearly 7.4% increase per 100 patients was noted.[13] A quarter of all Canadian patients made an outpatient cardiology visit in 2005, of which 70% were to a specialized center.[13] Younger patients and patients with severe lesions are more prone to make an outpatient visit to a specialized center. On the other hand, younger patients make less outpatient cardiology visits in general (not only to specialized centers), as do patients with less severe lesions.[13] However, an interaction between age and severity was noticed.[30] Patients with severe lesions were 139% more likely to visit outpatient cardiology services in young adulthood (18–40 years) compared to other patients. A decrease was noticed in middle-aged patients (41–64 years) but the likelihood remained 67% higher. No significant difference was observed in late adulthood.[30]

Per 100 patient years

Engelfriet et al[14] reported an average of 144 visits per 100 patient years between 1998 and 2003 in the Netherlands. These results were confirmed by a study in the United Kingdom spanning the 2000 to 2012 time period. Young adults (20–40 years), middle-aged adults (41–60 years), and older patients (60+ years) undertook 122, 145, and 159 visits per 100 patient years, respectively.[28] Outpatient cardiology visits are associated with diseased complexity: single ventricle patients visited their cardiologist nearly 200 times per 100 patient years, while patients having a ventricular septal defect made 114 visits per 100 patient years.[14]

Other healthcare visits

The frequency of visits to a general practitioner was mentioned in only one Canadian study. Ninety-one percent of the included patients visited their general practitioner over a 5-year timeframe. Half of the patients made at least 15 visits (IQR 7–27). Patients with severe lesions were 9% (CI 4–13%) less likely to visit a general practitioner compared to other patients.[30] The same study reported on outpatient specialist visits, other than cardiology visits. 87% of the patients visited a specialist medical doctor with a median of 10 visits over 5 years (IQR 4–22). Severe complexity patients were 6% (CI 0–12%) more likely to visit other medical specialists compared to other patients.[30] Age was unrelated to both healthcare provider consultations (general practitioner and medical specialists).[30] Unfortunately, no trends of other types of healthcare consultations have yet been reported in retrospective database research.

Discussion

This systematic review summarizes the high and changing healthcare demands of adult congenital heart disease patients. We noticed high annual increases in the absolute number of hospitalizations and the hospitalization rate per 100,000 general population. These two measurements reflect the expanding patient population. Simultaneously, the hospitalization rate per 100 patient/patient-years appears to be decreasing over time. These measurements can be seen as proxies for improved medical management. Indeed, a strong increase in outpatient cardiology visits can be observed over the past 30 years.

Our findings are relevant from a health policy point of view. Even though undeniably remarkable improvements are being made, current long-term medical management can only address part of the healthcare needs of the described patient population. Currently observed trends in emergency department visits support this thesis: emergency department visits and unplanned hospitalizations are increasing substantially in adults with congenital heart disease. Tang et al[35] reported a 2.10% yearly increase in emergency department visits in the general population, while the increase in emergency department visits of adults with congenital heart disease is observed to be three to four times higher. Meanwhile, the proportion of hospitalized patients who first visited the emergency department is increasing. The number of hospitalizations after an emergency department visit is thus growing faster than hospitalization after an outpatient cardiology visit. In other words, the number of unscheduled hospitalizations is growing faster than the number of scheduled hospitalizations. An even closer, systematical follow-up at outpatient services may mitigate the high (unplanned) hospitalization burden. This is of paramount importance to maintain a sustainable healthcare system in a patient population expected to continue to grow,[36] although some evidence suggests a decreased pace of increasing hospitalization numbers in more recent times.

The growth in absolute numbers of hospitalizations is highest in patients with mild lesions. Hospitalization trends in patients with mild and moderate lesions are of at least comparable magnitude to those in patients with severe lesions, even after exclusion of patients with an atrial septal defect or a patent foramen ovale. Similar patterns can be observed in emergency department visits and related hospitalization. One explanation of the significantly higher hospitalization rate after an emergency department visit in patients with mild lesions might be a less established follow-up structure. Patients with more complex lesions have more

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outpatient contact points, consistent with published guidelines dictating a higher follow-up frequency with growing complexity.[37–39] It will be necessary to explore optimal contact frequency and referral strategies to improve long-term care of patients with less complex lesions, particularly since the positive effect of referral to specialized centers on mortality is mainly found in patients with severe lesions.[13] Nonetheless, the risk of an adverse outcome (such as a permanent loss of cardiac function) is also higher in patients with mild or moderate lesions managed at a non-specialized center. General cardiologists tend to deviate from the guidelines more often than pediatric and adult congenital heart disease-trained cardiologists.[40] Therefore, all patients should be seen by a specialized cardiologist at least once and thereafter, a shared care model with a general cardiologist should be established.[39]

The trends described above can also be explained by technology advances, which may explain for example the high hospitalization increase (after emergency department visit) in patients with an atrial septal defect or a patent foramen ovale. A substantial number of patients admitted with ischemic cerebrovascular events are expected to be categorized in the patent foramen ovale cohort. Although accessibility to percutaneous interventions improved since the turn of the century,[24] it is still debatable whether patent foramen ovale closure (in combination with antiplatelet therapy), anticoagulation therapy, or antiplatelet therapy alone is the preferred therapy after stroke. Patent foramen ovale closure may reduce the occurrence of subsequent ischemic stroke, but it may also cause atrial fibrillation and device complications.[41–43] However, from an economic perspective, patent foramen ovale closure appears to be the most cost-effective option.[44,45] Hence, we can expect a further increase of hospitalizations in patients with mild lesions.

Geriatric patients are a newly emerging patient population, with the highest hospitalization rates and the highest outpatient cardiology visits per 100 patient years. However, the literature indicates that they have fewer outpatient cardiology visits at specialized centers compared to younger adult patients. This might be caused by the fact that, although the first specific adult congenital care programmes were established in the sixties, the exponential growth in programmes was only observed between 1990 and 2000.[7] Geriatric patients might thus currently be followed up at a less specialized, less appropriate care level. A second valid explanation is the higher mortality risk in severe lesions, leading to higher proportions of mild lesions in the elder population.[46] Conversely, age-related comorbidities[47] might urge specialized follow-up in geriatric patients, irrespective of the lesion's complexity grade.

The reliance on medical resource use seems to be higher in females, although results on gender disparities are inconclusive. One explanation might be the impact of pregnancy on the cardiovascular and respiratory systems. Close pregnancy management is of utmost importance to limit health risks.[48] The higher hospitalization rate in females reflects the observed hospitalization patterns in the general population, even after exclusion of maternal stays.[49] Hence, pregnancy alone may not account for the differences in medical resource use. Second, biological sex differences (such as a lower body weight and narrower arteries in females) may impact morbidity and mortality in adulthood. Pulmonary hypertension for instance is known to be more frequent in females,[50,51] possibly affecting hospitalization patterns. Lower compliance to treatment in females is a less likely explanation, as data published by White et al[52] indicated lower compliance rates in males.

Literature on patients' visits to a general practitioner and non-cardiology specialists is scarce. We found only one retrospective database study examining general practitioner visits and other healthcare provider visits. About 91 and 87% of all patients did consult a general practitioner and other medical specialists over a 5-year period, respectively. Patients with severe lesions made less general practitioner visits and more specialist visits compared to other patients. In addition to retrospective database research, a Dutch cross-sectional survey study reported general practitioner visits in 40% of all patients in the past year. A wide range of additional medical healthcare providers were consulted by patients but only outpatient contact with a nurse, physical therapist, and thrombosis specialist occurred in more than 10% of all adults with congenital heart diseases.[53,54]

It is possible that research subjects and regions show linkages in these observations. Half of the studies on hospitalization were conducted in the United States of America, two in Canada, four in Europe, and one in Asia. Three of the studies on emergency department visits were conducted in the United States of America, and one in Canada. Three of the studies on outpatient cardiology visits were conducted in Canada, three in Europe, and none in the United States of America. Replication of research in different countries can provide us with new insights on how healthcare systems may impact patients' medical resource seeking. In fact, differences in hospitalization patterns even occur within one country, as we have noted in Canada.[23,30] At last, we found low hospitalization numbers in China compared to European and American hospitalization numbers,[12] possibly reflecting cultural, genetic, or healthcare system differences.

Limitations

Six limitations were identified. First, we did not analyze additional characteristics of hospitalization or outpatient visits. A hospitalization for instance can be characterized by the length of stay, the intensive care unit stay, and the number of interventions performed. There is a need to assess how the length of stay evolved over time or to assess in how many hospitalizations surgery occurred. A second limitation is that we did not link the change in, and frequency of, medical resource use to costs. The financial impact of this quickly evolving patient population on healthcare systems is of paramount importance and should be closely monitored. Third, several database studies included made use of ICD-9 classification to determine the study sample. It was therefore not feasible to distinguish between atrial septal defect and patent foramen ovale as they share a common code. Fourth, the interpretation of emergency department-related hospitalization rates must be made with caution. In some countries, it may reflect how the healthcare system is working, rather than reflecting the disease's severity. Fifth, future research should investigate in which department the hospitalizations took place (cardiology versus other). Sixth, we only included retrospective administrative database studies. Database studies have several disadvantages (e.g., data are not primarily gathered to answer the study's research questions, administrative data are prone to miscoding, and coding principles might change over time).[55] However, one of our inclusion criteria was on study design since databases contain real world data and is thus representative of the population of interest.[55]

Conclusion

Lifetime medical resource use in congenital heart disease patients can be plotted as a U-curve with very high resource use in early childhood[22,23,56] and higher resource use in late compared to

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middle and young adulthood. The fact that improved efficiency levels cannot compensate for a growing population calls for improved long-term care models and for determinants of the optimal care level. In this review, we focused on retrospective database research. Scientific knowledge generated by retrospective analyses of databases may enhance specialized cardiologists' abilities to determine the appropriate referral level for patients with varying complexity lesions. Furthermore, the scope of future research should expand beyond hospitals and explore the burden adults with congenital heart disease exerts on the broad healthcare sector.

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Conflicts of Interest. None.

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