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***FAST*TRACK CLINICAL RESEARCH**
Congenital heart disease

Last year of life of adults with congenital heart diseases: causes of death and patterns of care

Liesbet Van Bulck [1,2], Eva Goossens [1,3], Lucas Morin [4,5], Koen Luyckx [6,7], Fouke Ombelet [1,8,9], Ruben Willems [10], Werner Budts [11,12], Katya De Groote [13], Julie De Backer [14], Lieven Annemans [10], Stéphane Moniotte [15], Michèle de Hosson [14], Arianne Marelli[16], Philip Moons [1,17,18]*, and BELCODAC consortium

[1]Department of Public Health and Primary Care, KU Leuven – University of Leuven, Kapucijnenvoer 35 (box 7001), 3000, Leuven, Belgium; [2]Research Foundation Flanders (FWO), 1000, Brussels, Belgium; [3]Faculty of Medicine and Health Sciences, Centre for Research and Innovation in Care, University of Antwerp, 2000, Antwerp, Belgium; [4]Inserm CIC 1431, University Hospital of Besançon, 25000, Besançon, France; [5]Inserm U1018, High-Dimensional Biostatistics for Drug Safety and Genomics, CESP, 94800, Villejuif, France; [6]Department of Psychology and Educational Sciences, KU Leuven – University of Leuven, 3000, Leuven, Belgium; [7]Unit for Professional Training and Service in the Behavioural Sciences (UNIBS), University of the Free State, 9300, Bloemfontein, South Africa; [8]Division of Neurology, University Hospitals Leuven, 3000, Leuven, Belgium; [9]Lab of Neurology, VIB – KU Leuven Centre for Brain and Disease Research, 3000, Leuven, Belgium; [10]Department of Public Health and Primary Care, Ghent University, 9000, Ghent, Belgium; [11]Division of Congenital and Structural Cardiology, University Hospitals Leuven, 3000, Leuven, Belgium; [12]Department of Cardiovascular Sciences, KU Leuven – University of Leuven, 3000, Leuven, Belgium; [13]Department of Pediatric Cardiology, Ghent University Hospital, 9000, Ghent, Belgium; [14]Department of Adult Congenital Cardiology, Ghent University Hospital, 9000, Ghent, Belgium; [15]Division of Pediatric and Congenital Cardiology, Department of Paediatrics, Cliniques universitaires Saint-Luc, 1000, Brussels, Belgium; [16]McGill Adult Unit for Congenital Heart Disease Excellence (MAUDE Unit), McGill University Health Center, H3A 0G4, Montréal (Quebec), Canada; [17]Institute of Health and Care Sciences, University of Gothenburg, 41346, Gothenburg, Sweden; and [18]Department of Pediatrics and Child Health, University of Cape Town, 7700, Cape Town, South Africa

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Abstract

Aims Although life expectancy in adults with congenital heart diseases (CHD) has increased dramatically over the past five decades, still a substantial number of patients dies prematurely. To gain understanding in the trajectories of dying in adults with CHD, the last year of life warrants further investigation. Therefore, our study aimed to (i) define the causes of death and (ii) describe the patterns of healthcare utilization in the last year of life of adults with CHD.

Methods and results This retrospective mortality follow-back study used healthcare claims and clinical data from BELCODAC, which includes patients with CHD from Belgium. Healthcare utilization comprises cardiovascular procedures, CHD physician contacts, general practitioner visits, hospitalizations, emergency department (ED) visits, intensive care unit (ICU) admissions, and specialist palliative care, and was identified using nomenclature codes. Of the 390 included patients, almost half of the study population (45%) died from a cardiovascular cause. In the last year of life, 87% of patients were hospitalized, 78% of patients had an ED visit, and 19% of patients had an ICU admission. Specialist palliative care was provided to 17% of patients, and to only 4% when looking at the patients with cardiovascular causes of death.

Conclusions There is a high use of intensive and potentially avoidable care at the end of life. This may imply that end-of-life care provision can be improved. Future studies should further examine end-of-life care provision in the light of patient's needs and preferences, and how the healthcare system can adequately respond.

* Corresponding author. Tel: +32 16 37 33 15, Fax: +32 16 33 69 70, Email: philip.moons@kuleuven.be

Structured Graphical Abstract

Key Question

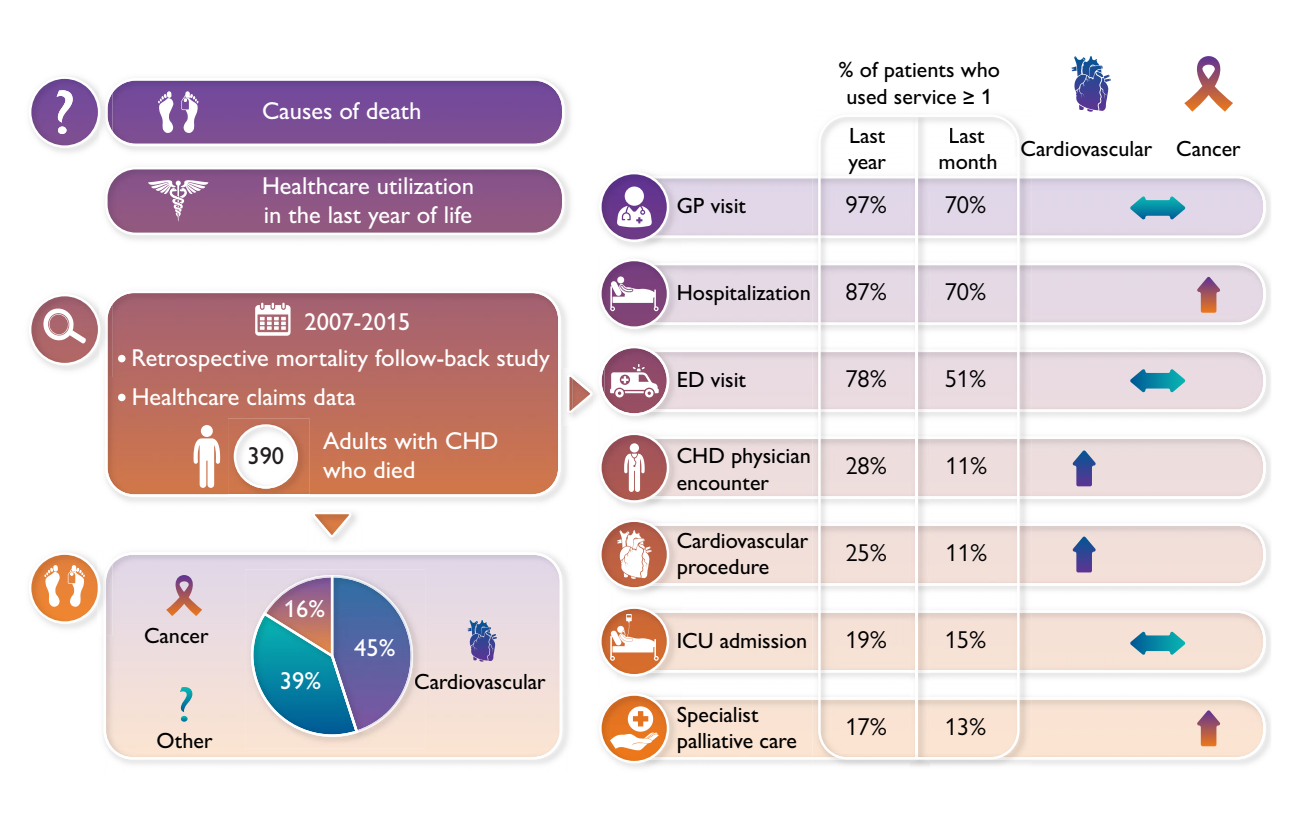
The study aimed to define causes of death and describe patterns of healthcare utilization in the last year of life of adults with congenital heart diseases (CHD).

Key Finding

- 45% died from a cardiovascular cause.
- In the last year of life: 87% of patients were hospitalized, 78% of patients had an emergency department (ED) visit, 19% of patients had an intensive care unit (ICU) admission, and 17% of patients received specialist palliative care.

Take Home Message

A tendency towards intensive and potentially avoidable care is noticed at the end of life. Future studies should further examine end-of-life care provision in the light of patient's needs and preferences, and how the healthcare system can adequately respond.



CHD, congenital heart disease; ED, emergency department; GP, general practitioner; ICU, intensive care unit. A horizontal arrow indicates no significant difference between the two groups and a vertical arrow indicates significantly more healthcare use for this group in the last year of life.

Keywords Cause of death • Congenital heart disease • End-of-life • Healthcare utilization • Hospitalizations • Palliative care

Introduction

Over the past five decades, patients with congenital heart disease (CHD) have experienced a considerable increase in their life expectancy.[1] For instance, nowadays, 97% of children with CHD reach adulthood, compared with 85% two decades ago.[2,3] Despite these improvements, a large fraction of adults with CHD remains symptomatic and at high-risk for premature death (median age at death is still below 50 years).[4] Long-term sequalae, such as heart failure and arrhythmias, become increasingly frequent as patients age, with a negative impact on the quality of life and overall survival.[5]

Indeed, the trajectories that patients follow towards death depend upon the cause of death and presence of long-term sequalae or co-morbidities.[6,7] It is likely that there will be a group of patients characterized by sudden death, a group with a rapid decline (typically for cancer patients), and a group of patients with intermittent serious episodes of complications (typically for patients with some form of organ failure).[8,9] Prior studies that have reported the causes of death of adults with

CHD have used historical cohorts,[9–11] were single centre studies,[12] or have been performed outside of Europe.[13]

Given the burden of symptoms and risk of premature death of adults with CHD, it is argued that there is a need for adequate palliative and end-of-life care.[5] Palliative care can be defined as '*an approach that improves the quality of life of patients and their families facing the problems associated with life-threatening illness, through the prevention and relief of suffering*'.[14] Palliative care can be complementary to standard medical care and can be introduced at any stage of the illness.[15] In contrast, end-of-life care can be defined as '*the process of supporting patients who are in their final months of life and their relatives*'.[16]

There are well-documented reasons to believe that adults with CHD have specific needs as they near the end of life, which are not comparable with the needs of patients who die from acquired heart diseases.[17] They not only die at a younger age,[4] the presentation of symptoms and cardiac deterioration of CHD patients is also diverse and often atypical because of the heterogeneity of the heart disease and subsequent surgical and transcatheter interventions.[18] Therefore, it is challenging to define a prognosis and identify the triggering event for deterioration of the situation.[18,19]

Although the implementation of palliative care in cardiology has gained substantial traction in recent years[20] and recommendations for advance care planning and end-of-life care for adults with CHD were discussed in guidelines by ESC working groups,[21,22] palliative and end-of-life care for adult CHD patients still is an understudied area. Up to now, the existing empirical studies mainly focus on preferences regarding advance care planning.[19,23–26] One study has investigated healthcare utilization at the end of life and showed that many patients were admitted to an intensive care unit (ICU) during their last month of life, hinting at a lack of adequate end-of-life care.[27] To date, no empirical studies have described the current outpatient healthcare utilization or specialist palliative care of adults with CHD. Given these gaps in the body of knowledge, the present study aimed to (i) identify the causes of death, and (ii) describe the patterns of healthcare utilization in the last year of life of adults with CHD.

Methods

Data source

We used the BELgian COngenital heart disease Database combining Administrative and Clinical data (BELCODAC), which includes administrative and clinical data of patients with CHD in Flanders and Brussels. More details about this database have been described previously.[28] In short, BELCODAC comprises data on healthcare utilization from 2006 to 2015 and clinical data (e.g. information about diagnosis, comorbidities, and interventions) from the same period and before. Data were derived from five organizations, namely three Belgian hospitals (i.e. University Hospital Leuven, Saint-Luc University Hospital, and Ghent University Hospital), the national statistical office (i.e. Statistics Belgium), and the authority in charge of maintaining all healthcare claims from the seven health insurance funds in the country (i.e. InterMutualistic Agency). These different datasets were linked at the patient level under the supervision of the Belgian Data Protection Authority, and included a four-layer pseudonymization system to safeguard the identity of all patients. Overall, BELCODAC comprises 18 510 children and adults with CHD.[28]

Study design and population

For the purpose of the present study, we conducted a mortality follow-back study in adult patients who died between 1 January 2007 and 31 December 2015, and who had lived in Belgium during their last year of life. The timeline and flowchart of this study are presented in Supplementary material online, *Figures S1 and S2*.

A total of 390 patients died in the observation period, 51% of which were women, and the median age at the time of death was 55 years (*Table 1*). Most patients had a CHD of moderate complexity (46%), followed by mild complexity (43%) and severe complexity (11%), as defined by Stout *et al.*[29] The most common heart defect in patients who died was Type 2 atrial septal defect ($n = 88$; 23%). In Supplementary material online, *Table S1*, the variables and codes used to identify clinical characteristics in BELCODAC are described.

Outcome definitions

Causes of death were derived from the International Classification of Diseases and Related Health Problems, 10th revision (ICD-10) codes reported as the underlying cause of death on the death certificates of patients, which are filled out by a certifying doctor for every death that takes place in Belgium. The categories and corresponding ICD-10 codes are reported in Supplementary material online, *Table S2*.

Sudden death, defined as '*a death, non-violent and not explained otherwise, occurring <4 h from the onset of symptoms*',[30] was identified by LVB based on the clinical interpretation of available data. For patients who had no healthcare utilization in the last 10 days of life, except for the day of death, the underlying cause of death, the clinical profile, and the provision of care in the last year of life was checked to evaluate whether or not death was most likely sudden and unexpected. Reasons to believe that a patient did not die suddenly were a registered chronic disease, residence in a nursing home, use of incontinence equipment, regular hospitalizations in the last year of life, or regular visits at the general practitioner (GP) or specialist in the last 6 months of life. Ambiguous cases were discussed within the team (L.V.B., P.M., E.G.) until an agreement was made.

Healthcare utilization includes contacts with a GP, contacts with a CHD physician, hospitalizations, visits to the emergency department (ED), cardiovascular procedures, admissions to the ICU, and specialist palliative care, which can be both inpatient and outpatient. Healthcare utilization was based on nomenclature codes derived from healthcare claims data. Detailed information about the variables and nomenclature codes used are described in Supplementary material online, *Table S3*.

Statistical analysis

Sample characteristics and causes of death of all patients who died ($n = 390$) were analysed descriptively and were presented as median and quartiles for continuous variables and as frequencies and percentages for categorical variables.

Healthcare utilization was analysed for the 327 patients who had a cause of death and died non-suddenly and non-accidently, as these patients could have had end-of-life care needs and death could have been expected. Healthcare utilization in the last 12 months of life was analysed descriptively and logistic regression analyses were used to examine differences between patients who died due to either a malignant or a cardiovascular cause of death. For contacts with a GP, hospital days, and ED visits, the mean monthly days with an event were reported for every month in the last year of life and modelled using binomial regressions with generalized estimating equations. For the analyses of GP and ED visits, only the days that patients were not hospitalized (starting from the second day of the hospital admission) were taken into account, as patients usually do not have GP contacts or ED visits during a hospital admission. For contacts with CHD physicians, cardiovascular

Table 1 Demographic and clinical characteristics of the study population

Sample size	390
Sex, women	200 (51%)
Age at time of death, years, median (Q1–Q3)	55 (40–73)
Disease complexity	
Mild	166 (43%)
Moderate	181 (46%)
Complex	43 (11%)
Primary CHD diagnosis	
Univentricular physiology	12 (3%)
Tricuspid atresia	2 (1%)
Tetralogy of Fallot	30 (8%)
Pulmonary atresia with ventricular septal defect	1 (0%)
Double outlet right ventricle	8 (2%)
Double inlet left ventricle	2 (1%)
Truncus arteriosus	2 (1%)
Transposition of the great arteries	10 (3%)
Congenitally corrected transposition of the great arteries	6 (2%)
Coarctation of the aorta	28 (7%)
Atrioventricular septal defect	20 (5%)
Atrial septal defect type 1	12 (3%)
Ebstein malformation	4 (1%)
Pulmonary valve abnormality	25 (6%)
Aortic valve abnormality	30 (8%)
Aortic abnormality	6 (2%)
Left ventricular outflow tract obstruction	1 (0%)
Atrial septal defect Type 2	88 (23%)
Ventricular septal defect	31 (8%)
Mitral valve abnormality	15 (4%)
Pulmonary vein abnormality	10 (3%)
Other	47 (12%)
Heart failure	163 (42%)
Diabetes	52 (13%)
PM/ICD implantation	32 (8%)

CHD, congenital heart disease; ICD, implantable cardioverter-defibrillator; PM, pacemaker.

procedures, admissions to the ICU, and specialist palliative care, it was considered inappropriate to calculate the mean monthly days for each item, because of excessive zeros in the data. Therefore, for these types of healthcare utilization, the proportions of patients using this healthcare service at least once per month for the last 12 months of life were reported, together with exact confidence limits for the proportions.

Ethical approval

Approval of the ethics committee from University Hospitals Leuven (file numbers S59859 and S59858), Ghent University Hospital (file numbers 2017/0436 and 2017/0437), and Saint-Luc University Hospitals (file numbers 2017/26JUI/333 and 2017/26JUI/332) was obtained. At this governmental level, approval was obtained from the Statistical Supervisory Committee (SA2/STAT-MA-2017-021), the Sectoral Committee of Social Security and of Health (CSSSS/17/184 and SCSZG/17/184), and the Sectoral Committee of the National Register. The study is legally exempt from the obligation to obtain written informed consent from each patient. More information can be found elsewhere.[28]

Results

Causes of death

The distribution of causes of death is depicted in *Figure 1*. While 55 (14%) patients died due to a sudden, accidental, or violent cause of death, most patients ($n = 327$; 84%) had a non-sudden death. A total of 28 (7%) patients had an accidental or violent cause of death (including 14 deaths due to intentional self-harm), 15 (4%) patients had a sudden cardiac death, and 12 (3%) patients died suddenly with a noncardiac cause of death reported. For eight patients (2%), the cause of death was missing or unclear (for six patients due to death outside of Belgium).

Almost half of the study population ($n = 174$; 45%) died from a cardiovascular cause, of which 15 (4%) patients had a sudden cardiac death. Of the patients who died due to a cardiovascular cause, 30% ($n = 51/174$) had the CHD as reported cause of death and 14% ($n = 25/174$) died due to a stroke.

In addition, 64 (16%) patients died from cancer, with the most common cancers being lung cancer ($n = 13/64$; 20%) and breast cancer ($n = 7/64$; 11%). The remaining 104 patients died from respiratory diseases ($n = 24$; 6%), non-cardiovascular congenital malformations ($n = 19$; 5%), digestive diseases ($n = 12$; 3%), endocrine, nutritional and metabolic disorders ($n = 12$; 3%), infectious and parasitic diseases ($n = 9$; 2%), diseases of the nervous system ($n = 7$; 2%), and other reasons ($n = 21$; 5%).

As reported in *Table 2*, a large majority of patients with severe CHD lesions ($n = 32/43$; 74%) died from a cardiovascular cause of death, compared with only half of patients ($n = 88/174$; 50%) with moderate lesions and only 31% ($n = 54/173$) of the patients with mild lesions. A quarter of the patients ($n = 41/173$; 24%) with mild lesions died due to a malignant cause of death. These proportions were lower for patients with moderate ($n = 21/174$; 12%) and severe ($n = 2/43$; 5%) CHD lesions. The causes of death for each CHD diagnosis can be found in Supplementary material online, *Table S4*. For patients with an atrioventricular septum defect, Ebstein malformation, a valve abnormality, aortic abnormality, left ventricular outflow tract obstruction, Type 2 atrial septal defect, ventricular septal defect, and pulmonary vein abnormality, proportionally more patients died due to a malignant or other cause of death than to a cardiovascular cause of death.

Healthcare utilization

Contacts with the general practitioner

Almost all patients (97%) had a GP encounter in the last year of life and 70% had a GP encounter in the last month of life. The proportions of patients having had GP visits in the last year were not significantly different for the two groups (*Figure 2*). However, the number of GP visits in the last month of life significantly differed for the two groups, with patients with a malignant cause of death reporting twice as many visits (*Figure 3*).

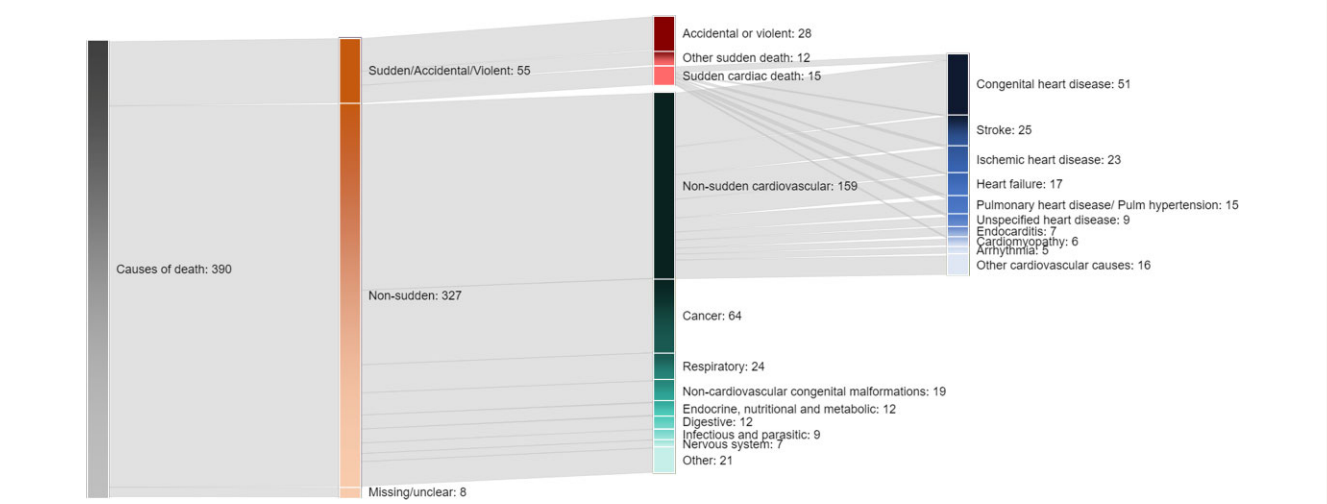


Figure 1 Causes of death of adults with CHD ($n = 390$).

Table 2 CHD complexity and causes of death

Causes of death	Total ($n = 390$)	Mild ($n = 173$)	Moderate ($n = 174$)	Severe ($n = 43$)
Cardiovascular	174 (45%)	54 (31%)	88 (50%)	32 (74%)
Cancer	64 (16%)	41 (24%)	21 (12%)	2 (5%)
Other	144 (37%)	74 (43%)	62 (36%)	8 (19%)
Missing	8 (2%)	4 (2%)	3 (2%)	1 (2%)

The number of GP encounters significantly increased in the last months of life (*Figure 3*). The patterns of GP contacts over time were different for patients who died due to a cardiovascular or malignant cause of death (*Figure 3*).

Hospitalizations

A total of 815 hospital admissions in the last year of life were found for all patients, of which almost half (374; 46%) were preceded by an ED visit. A total of 87 and 70% of patients were hospitalized at least once in the last year and month of life, respectively. The proportion of patients who were hospitalized in the last year of life was significantly higher for patients who died due to a malignant cause of death compared with patients with a cardiovascular cause of death (*Figure 2*). However, the number of hospital days in the last month of life was not significantly different for these two groups.

Patients spent more days hospitalized towards the end of life (*Figure 3*). The patterns of mean monthly hospital days over the last year were similar for patients who died due to cardiovascular and malignant causes of death (*Figure 3*).

Visits to the emergency department

In the last year and month of life, 78 and 51% of patients had at least one ED visit. The proportion of patients who had an ED visit in the last year of life and the mean number of ED visits in the last month were not different for patients who died due to a cardiovascular or malignant cause of death (*Figures 2 and 3*).

In the last month of life, the number of ED visits increased (*Figure 3*). Patients who died from a malignant or cardiovascular disease followed different patterns throughout the last year of life (*Figure 3*) that were characterized by a mean monthly number of ED visits of 0.5 for patients with a malignant deaths and 0.1 for cardiovascular deaths.

Contact with a congenital heart disease physician

The proportions of patients with at least one CHD contact in the last year and month of life were 28 and 11%, respectively (*Figures 2 and 3*). A significant difference between the proportion of patients with CHD physician contacts during the last year and last month of life was found for patients with cardiovascular and malignant causes of death (*Figures 2 and 3*).

The proportion of patients having a CHD physician contact was higher in the last month of life compared with 1 year before death, but an increase around the 5th and 6th month before death could be noticed as well (*Figure 3*).

Cardiovascular procedures

A total of 25 and 11% of patients had a cardiovascular procedure (e.g. heart surgery or catheter-based heart intervention) in the last year and month of life, respectively (*Figures 2 and 3*). Compared with patients with a malignant cause of death, significantly more patients who died from a cardiovascular disease had procedures in the last year and month of life (*Figures 2 and 3*).

There was an increase in patients receiving a cardiovascular procedure towards the end of life, for all patients and for patients who died due to a cardiovascular cause (*Figure 3*).

Admissions to the intensive care unit

A total of 19% had an ICU admission in the last year of life and 15% had an ICU admission in the last month of life (*Figures 2 and 3*). These proportions were not significantly different for patients who died due to a malignant or cardiovascular cause.

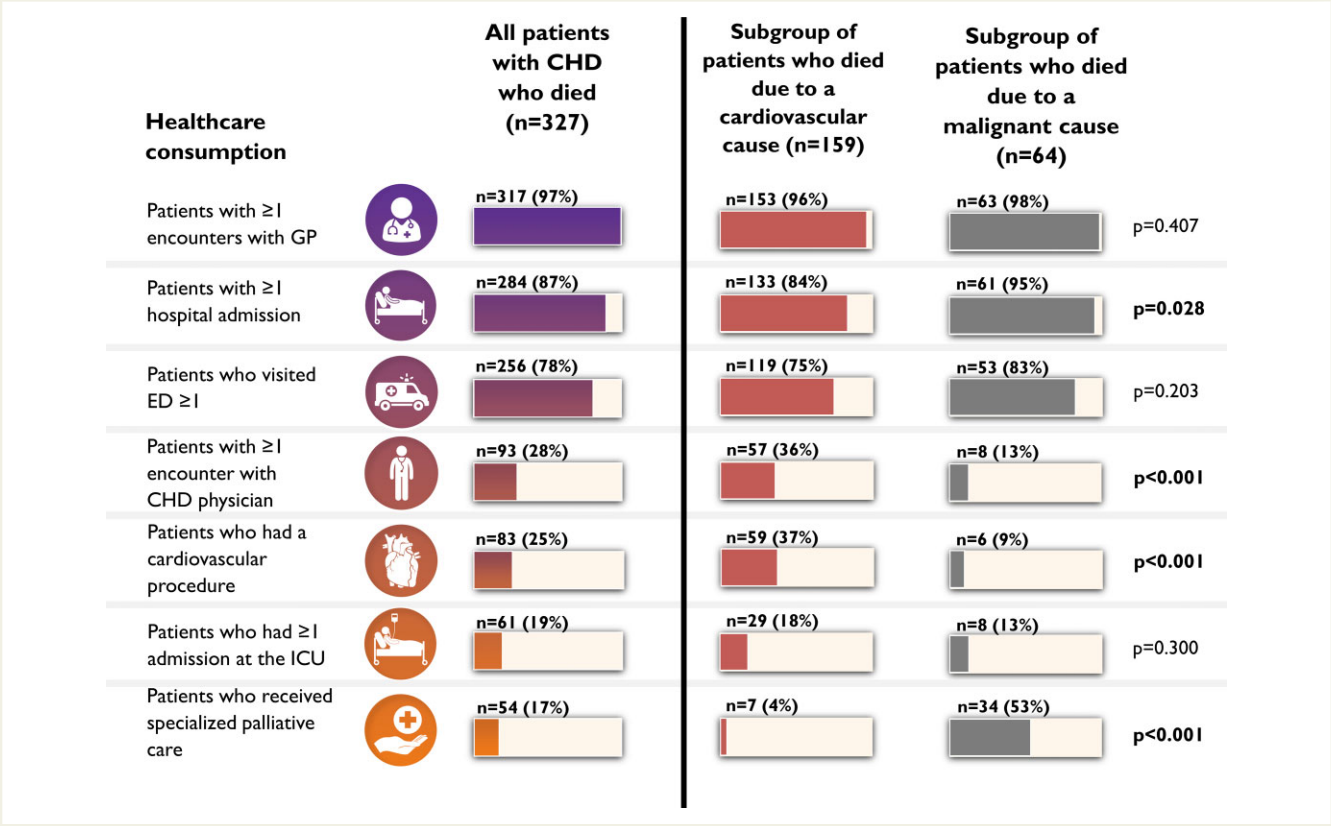


Figure 2 Healthcare utilization in the last year of life of adults with CHD ($n = 327$) and of subgroups of patients with a cardiovascular cause of death ($n = 159$) and malignant cause of death ($n = 64$). CHD, congenital heart disease; ED, emergency department; GP, general practitioner; ICU, intensive care unit.

When looking at the proportion of patients with admissions to the ICU, an increase in the last months of life was noticed. The trajectory over time was similar for patients who died due to a cardiovascular or malignant cause of death (*Figure 3*).

Specialist palliative care

A total of 17 and 13% of patients received specialist palliative care in the last year and month of life, respectively (*Figures 2 and 3*). Significantly more palliative care in both the last year and month of life was provided to patients with a malignant cause of death, as compared with patients with a cardiovascular cause of death. Of the latter group, 4% received specialist palliative care in the last month, whereas 41% of patients with a malignant cause of death received specialist palliative care in the last month of life.

Discussion

The last year of life of adults with CHD is an understudied domain. The present study had several notable findings. First, less than half of people with CHD died due to cardiovascular diseases. Second, we found high healthcare utilization in terms of hospital admissions, ED visits, cardiovascular procedures, and ICU admissions, which increased towards the last months of life. Third, use of specialist palliative care seemed very limited, especially for patients with a cardiovascular cause of death (*Structured Graphical Abstract*).

Our finding that less than half (45%) of the study population died due to a cardiovascular cause of death is in line with a recent study from the USA.[13] However, older studies reported markedly higher proportions of death due to cardiovascular causes. In studies from 2000 and 2007, it was stated that cardiovascular death accounted for 65[9] and 67%[31] of all deaths of patients with CHD, respectively. Earlier studies have reported that the causes of death differ by CHD complexity, with more cardiovascular death for those with severe lesions.[13] This was also confirmed in our study. The different proportions of cardiovascular deaths found in the literature could be due to subjectivity in reporting the causes of death, or by the changing profile of patients with mild and moderate CHD lesions in the last decade. For instance, a study published in 2010 described that over 70% of patients with a ventricular septal defect died due to a cardiovascular cause,[4] whereas in our study, only 32% of the patients with a ventricular septal defect died due to a cardiovascular cause. The changing profile is also noticeable when looking at the median age of the decedents. The median age in this study was 55 years, which is remarkably higher than the mean ages at death reported in previous studies.[4,9,32] Only in the recent study of Goldstein *et al.*,[13] a higher median age of 64.2 years was reported. The shifting cause of death from cardiovascular death to other causes and increasing ages could be the result of improved management of long-term sequelae and a better follow-up of patients with mild and moderate CHD during child- and adulthood.[13] The changing profiles might also have implications for the end-of-life and palliative care needs, which may be different to date than in the past. Care at the end of life should go beyond cardiovascular care, and interdisciplinary collaborations are increasingly

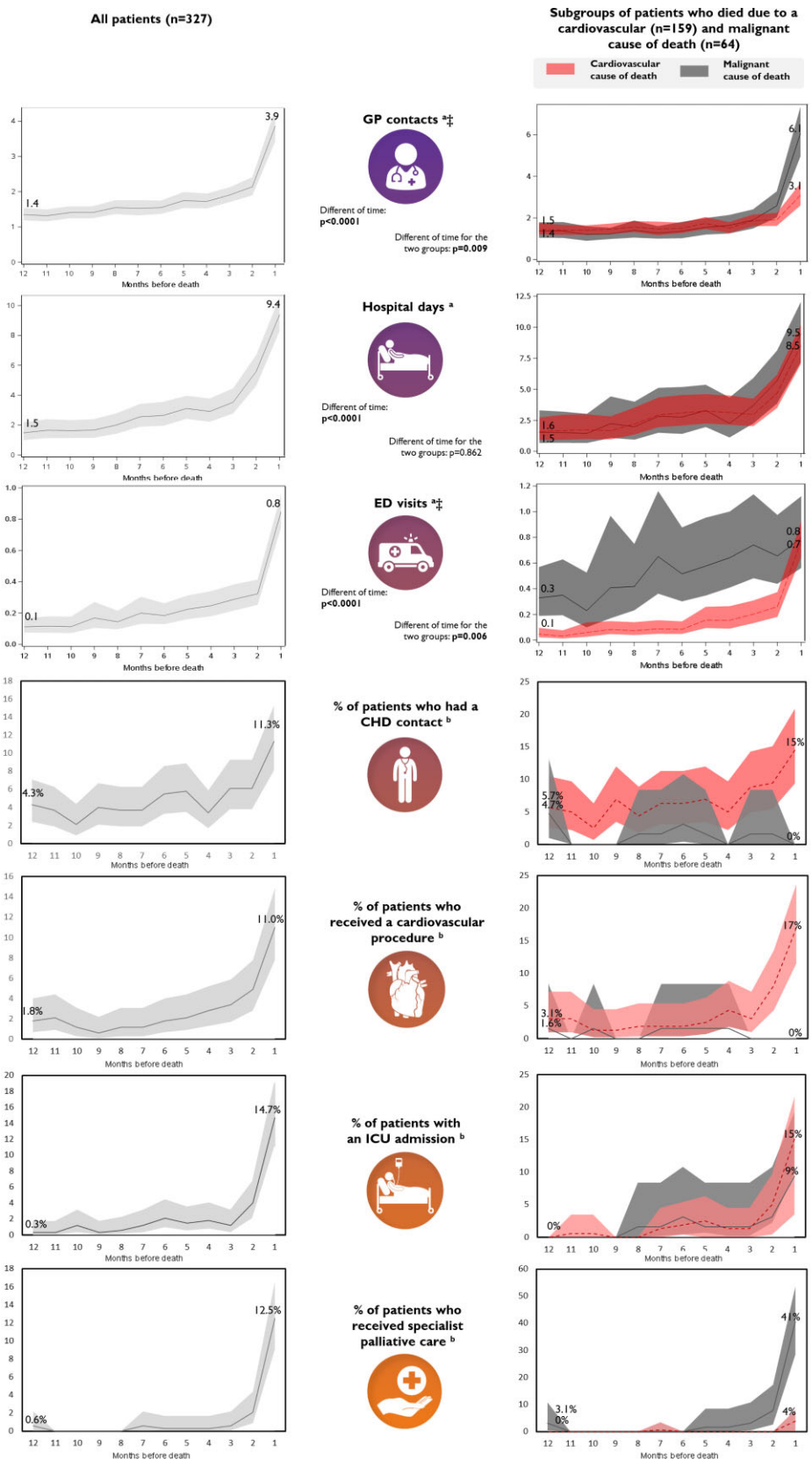


Figure 3 Healthcare utilization patterns of the last year of life in adults with CHD ($n = 327$). (*a*) Binomial regression model with generalized estimating equations (mean numbers of days with event per month over the last 12 months are shown); (*b*) Exact confidence limits for the proportion (proportions of patients who used health service at least once per month over the last 12 months are shown); lighter coloured area represents the 95% confidence interval. ‡Calculations are based on days that patients were not hospitalized. CHD, congenital heart disease; ED, emergency department; GP, general practitioner; ICU, intensive care unit.

needed, as patients with CHD do not exclusively die from their heart disease and many patients have comorbidities. Developing best practice models with an interdisciplinary approach regarding the CHD and comorbid conditions such as cancer and respiratory diseases could improve outcomes for this patient population.

It was also remarkable that 14/382 patients (3.6%) died due to intentional self-harm, of which at least 12 were confirmed suicides (12/382; 3.1%). The prevalence of psychiatric disorders is higher in adults with CHD than in the general population.[33] However, the suicide rate found in this study is much higher than the 0.56% suicide proportion found among all deaths in a Danish cohort of CHD patients.[34] The suicide rate per 100 000 inhabitants is higher in Belgium (18.3) than in Denmark (10.7).[35] However, that does not explain the five-fold higher percentage of suicides as cause of death in the patients in BELCODAC and, hence, this finding requires further investigation.

We observed potentially avoidable resource use in terms of (unplanned) hospital admissions, ED visits, cardiovascular procedures, and ICU admissions at the end of life, mainly in the last 3 months. This was in line with the existing literature. The only published study to date about end-of-life care for adults with CHD, which included 65 patients from the USA with moderate or severe CHD lesions, reported high healthcare utilization in the last month of life.[27] In that study, Steiner *et al.*[27] described that 39% of patients were hospitalized, 39% had an admission to the ICU, and 3% had a visit to the ED in the last month of life. In our study, a much higher number of patients (70%) were found to be hospitalized and to have had an ED visit (51%) in the last month of life. A lower proportion (15%) was admitted to the ICU in the last month of life. These discrepancies might originate from the differences between healthcare systems in Europe and USA or could be due to differences in the study methodologies and sample sizes, with the American study having a much smaller sample. Furthermore, the high resource use at the end of life was also not surprising because adults with CHD already use a lot of healthcare during their lives, much more than people from the general population.[36–38] A previous BELCODAC study reported that, in 2015, 21% of patients visited a physician for adults with CHD, 86% had a GP visit, 17% of patients had an ED visit, and 17% were hospitalized at least once.[38] On top of that, we know from studies in other chronic patient populations that healthcare utilization increases in the last months of life, mainly as a response to the presence of comorbidities and exacerbations at the end of life.[39] Referral to palliative care has shown to decrease the number of inpatient stays and ICU admissions at the end of life.[40] Indeed, to lower unnecessary healthcare utilization, such as hospital admissions and ED visits at the end of life, properly implementing palliative care for adults with CHD could be an important strategy. Patients with CHD, especially those with a cardiovascular cause of death, seem to have received only little specialist palliative care. In contrast to hospitalizations, ED visits, and ICU admissions, which may not necessarily be appropriate at the end of life, palliative care is typically seen as desirable at the end of life. Although palliative care is gaining recognition as a critical component of comprehensive care for adults with CHD early in the disease trajectory,[41] the actual implementation of palliative care in routine clinical practice remains limited. In a study in adults with CHD, who were not necessarily in the terminal phase of their lives, 91% had indicated that they were at least moderately willing to speak to a clinician specialized in palliative care and 3% indicated to have met one.[19] A qualitative study showed that adults with CHD are rather unfamiliar with the definition and elements of palliative care.[24] Palliative care was often related to the terminal phase and described as 'giving up'.[24] In a survey study among CHD healthcare providers, the providers reported low levels of personal palliative care knowledge.[25] Moreover, only 14–32% of CHD providers indicated that they would initiate referral to palliative care.[25] Furthermore, although the importance of early advance care planning has been described for adults with CHD,[21,42] CHD providers seem to be worried about the readiness of their patients for these discussions, particularly for patients who are still in good health.[25,43] When advance care planning is not routinely addressed, this may cause lack of referral to palliative care.[44] Compared with patients who died from a cardiovascular disease, a much higher proportion of patients who died from cancer received specialist palliative care. This is not surprising as end-of-life care needs of cancer patients have been studied thoroughly and receive much more attention.[45] A Western Australian study reported that, during the last year of life, 69% of decedents with cancer and only 14% of non-cancer decedents accessed specialist palliative care.[45] Compared with decedents with heart failure, decedents with cancer were 10 times more likely to receive specialist palliative care.[45]

Only one in four patients saw a CHD physician in the last year of life and 11% in the last month of life. When looking at the patients who died due to a cardiovascular disease, still only 36 and 15% had a CHD physician encounter in the last year of life and month of life, respectively. This finding raises the question whether CHD physicians are adequately involved in the final stage of life and whether the cardiovascular needs and symptoms are sufficiently taken into account at the end of life. It would be interesting to examine whether a closer engagement of CHD physicians at the end of life could reduce potentially avoidable resource use and improve referral to palliative care.

Hence, the findings of this study suggest that healthcare utilization at the end of life of adults with CHD requires more attention. Future studies should further examine the changing end-of-life care needs and trajectories towards death, and investigate how the healthcare system can adequately respond to these needs. Improving the quality of end-of-life care provision for adults with CHD will be an important health challenge for the upcoming years.

The present study has several strengths. First, this is the first study to report on healthcare utilization of outpatient healthcare services, cardiovascular procedures, and specialist palliative care in the last year of life of adults with CHD. Second, the study included data of a large number of patients who died in a 9-year period. Third, the study used data of a population-level database. The database included patients without preselection. As a result, the level of ascertainment bias is much lower than in registries that include only a subgroup of patients and this database also includes patients that are often missed. Fourth, whereas most existing databases use ICD diagnosis to identify patients, BELCODAC patients were selected in the hospital database and their diagnosis was confirmed by expert clinicians.

However, our results should also be interpreted with a number of limitations in mind. First, we could not distinguish whether a healthcare service was used for a reason related to the CHD or not, because this information was not available. In future studies, it would be interesting to look into more detail whether the high potentially avoidable resource use was related to the heart disease or not. Second, the underlying causes of death should be interpreted with caution. Unfortunately, only the primary cause of death was available in BELCODAC, and the accuracy and reliability of ICD-10 codes are known to be limited.[10,46] The disease profile of adults with CHD is often complicated at the end of life and, therefore, might be hard to interpret by the physician without CHD background who fills out the death certificate. Moreover, the investigators had to use proxies to pinpoint potentially sudden and unexpected deaths, without having access to the full electronic medical records. Inaccuracies may also occur due to incomplete

death certificates, diagnostic issues, or when the death had multiple causes.[10] Also, we did not have information about the place of death. Third, it is likely that the reported findings are not entirely generalizable to other countries and healthcare systems, as the study only uses data of patients from Belgium. Fourth, the data are subject to limitations because they are not collected for research purposes, but primarily for clinical and reimbursement purposes. As a consequence, some clinical data are not comprehensive enough, and the reliability of the data mainly depends on its impact on reimbursements. Finally, no data on patient-reported outcomes, preferences, or on the reasoning behind certain end-of-life care decisions were available.

Conclusion

High healthcare utilization in terms of hospital admissions, ED visits, ICU admissions, and cardiovascular procedures, was found, whereas the use of specialist palliative care was limited. This intensive and maybe sometimes avoidable care, especially for patients with a cardiovascular cause of death, suggests that care provision at the end of life for adults with CHD can be improved. Care at the end of life for adults with CHD should go beyond cardiovascular care, as less than half of the study population died due to a cardiovascular disease. Future studies should further examine the changing end-of-life care needs, patient's preferences, and investigate how the healthcare system can adequately respond.

Author contributions

L.V.B. conceived and designed the study, performed the statistical analysis, interpreted the data, drafted, and critically revised the manuscript. E.G., P.M., L.M., and K.L. interpreted the data and critically revised the manuscript. F.O., R.W., W.B., M.d.H., K.D.G., L.A., J.D.B., and S.M. acquired, analysed interpreted the data, and critically revised the manuscript. A.M. was essential while setting up the database and critically revised the manuscript. All authors gave approval for the final version of the manuscript and agree to be accountable for all aspects of the work.

Supplementary material

Supplementary material is available at *European Heart Journal* online.

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Conflict of interest: The authors declare that they have no conflict of interest regarding this article.

Data availability

Clinical data cannot be made publicly available because of privacy issues. However, additional results and aggregated findings are available on reasonable request.

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