Cardiol Young 2001; 11: 301–313

ISSN 1047-9511

Original Article

A pilot study of expenditures on, and utilization of resources in, health care in adults with congenital heart disease

Philip Moons[1], Kaat Siebens[1], Sabina De Geest[1], Ivo Abraham[1,2], Werner Budts[3], Marc Gewillig[3]

[1]Center for Health Services and Nursing Research, School of Public Health, Katholieke Universiteit Leuven, Leuven, Belgium; [2]The Epsilon Group LLC, Charlottesville, VA, USA; [3]Department of Congenital Cardiology, University Hospitals of Leuven, Leuven, Belgium

Abstract *Background:* Congenital cardiac disease may be a chronic condition, necessitating life-long follow-up for a substantial proportion of the patients. Such patients, therefore, are often presumed to be high users of resources for health care. Information on utilization of resources in adults with congenital heart disease, however, is scarce. *Methods:* This retrospective pilot study, performed in Belgium, investigated 192 adults with congenital heart disease to measure the annual expenditures and utilization of health care and compared the findings with data from the general population. We also sought to explore demographic and clinical parameters as predictors for the expenditures. *Results:* Hospitalization was documented in 20.3% of the patients, with a median length of stay of 5 days. The overall payment by health insurance associations in 1997 was 1794.5 ECU per patient, while patients paid on average 189.5 ECU out-of-pocket. For medication, the average reimbursement and out-of-pocket expenses were estimated at 78 ECU and 20 ECU, respectively. Expenditures for patients with congenital heart disease were considerably higher than the age and gender-corrected expenditures for the general population (411.7 ECU), though this difference was accounted for by only one-eighth of the cohort of those with congenital heart disease. In general, higher expenditures were associated with abnormal left ventricular end-diastolic diameter, female gender, functional impairment and higher age, although the explained variance was limited. *Conclusion:* Our study has provided pilot data on the economic outcomes for patients with congenital heart diseases. We have identified parameters that could predict expenditure, but which will have to be examined in future research. This is needed to develop guidelines for health insurance for those with congenital heart diseases.

Keywords: grown-up congenital heart disease; health services research; adolescents

Due to improved medical, surgical, and technological evolutions, approximately 90% of children with congenital heart disease survive to adulthood,[1] resulting in a growing population of patients. Although the majority of these patients do not present with hemodynamic problems, and achieve a good quality of life, a substantial proportion of this population require life-long follow-up to avoid complications, such as atrial arrhythmias, bacterial endocarditis, congestive heart failure or pulmonary vascular disease.[2,3] Because of the regular visits to physicians, and the prevalence of complications, patients with congenital heart disease are presumed to be high users of resources for health care. Consequently, companies are often reluctant to provide health insurance to adults with congenital heart disease,[2,4,5] even in those with successfully corrected or benign malformations, resulting in associated psychosocial problems and compromised social integration of the population.

Correspondence to: Philip Moons, Center for Health Services and Nursing Research, School of Public Health, Katholieke Universiteit Leuven, Kapucijnenvoer 35, B-3000 Leuven, Belgium. Tel: + 32 16 336975, Fax: + 32 16 336970; E-mail: Philip.Moons@ med.kuleuven.ac.be

This study was supported by a grant from the Belgian National Foundation for Research in Pediatric Cardiology

Accepted for publication 13 December 2000

The issue of insurability is most prevalent in countries with a private health insurance system, such as the United States of America.[2,4,5] In most European countries, health insurance for congenital heart patients or other disabled persons is not that problematic,[6,7] because the Health Care system is based on a National Health Service, as in the United Kingdom, Greece, Ireland, Denmark, Italy, Portugal, and Spain, or compulsory health insurance as in Belgium, Germany, France, and The Netherlands. Both systems emphasize solidarity. Reforms of health care in some European countries, nonetheless, have introduced private health insurance in addition to the coverage provided by government or social security. The profit nature of insurance companies might now increase the number of patients with congenital heart diseases denied additional health insurance in Europe, or mean that they are insured at higher premiums, based on presumed risks. Data on utilization of resources, and expenditures in health care for patients with congenital heart disease are needed for evidence-based decision-making regarding the insurance of this population of patients.

Despite the extensive body of evidence concerning cost-effectiveness of selected surgical or catheter interventions, empirical studies on expenditures and utilization of services are relatively rare in this population. For example, a recent book on 'Resource utilization in cardiac disease',[8] did not consider congenital heart patients. Previous research has focussed predominantly on hospital expenditures in the first year of life,[9] or on hospital charges or costs of cardiac surgery, in particular costs incurred in the perioperative period.[10–16] These studies included only children. To date, only one study has examined adults with congenital heart disease and estimated the expenditures per patient for different classes of congenital cardiac malformations.[1] According to this study, annual expenditures, defined as hospital charges, professional fees and prices of medication, for such patients averaged $2851 per year, calculated in 1992 US dollars, between birth and 21 years of age, and $1120 per year between the ages of 22 and 40 years. It is estimated that three-quarters of the charges from birth to the age of 40 are spent in the first 21 years of life,[1] indicating that most expenditures are made in childhood. With respect to hospitalizations, only about one-twentieth of adults with congenital heart disease require one or more hospital admissions per year, although hospital care for adults and adolescents with congenital heart disease is apparently more costly than hospital care for other adults requiring cardiac medicine or surgery.[3] Compared with other chronic diseases occurring in children, such as hemophilia, cystic fibrosis, cancer and muscular dystrophy, treatment and follow-up of congenital cardiac defects is estimated to be less expensive.[1] The contribution of primary care in the expenditures on adults with congenital heart disease, and the extent to which the expenditures are comparable with those for the general population, is not yet known.

Utilization of resources by patients with congenital heart disease may vary widely. Clinical characteristics, such as cyanosis, type of operation, or occurrence of left heart failure, are associated with higher utilization, and consequently increased expenditures.[1,3,11]

In the early 70s, Andersen & Newman[17] developed a theoretical framework analyzing the societal and individual determinants of utilization of resources for health care. This model established three levels of determinants: predisposing, enabling and need factors, the latter representing the level of illness. Predisposing factors include demographic and socio-cultural variables, for example gender, age, education, race, attitudes toward health services, or knowledge about diseases. Enabling factors include family and community-related variables, for instance income, health insurance, price of health services or urban-rural character of the region. The need factors are considered as the strongest predictors of utilization. They consist of objective or evaluated needs, such as diagnosis, comorbidity or functional status; and subjective or perceived needs, the latter being psychological distress, depressive feelings, or experience of symptoms.[17] The validity of this model in the prediction of utilization of resources has been studied extensively in several populations. A recent review of the literature addressing predictors of utilization of resources in the chronically ill[18] revealed that the explained variance in hospital utilization by the 3 identified factors varied from 6.3% to 54%. A systematic identification of determinants of such utilization and expenditures in adults with congenital heart disease has not been undertaken to date.

In addition to mortality, morbidity, quality of life and patient satisfaction, data on expenditures are considered to be important parameters of outcome to evaluate the provision of health care.[19] Research on expenditures in adults with congenital heart disease may offer a detailed picture of the financial consequences of congenital heart disease, which can then provide a basis for accurate allocation of resources in programs designed to cater for this population. Knowledge of determinants of utilization of resources in these patients is critical in developing guidelines for health insurance, in order

https://doi.org/10.1017/S1047951101000336 Published online by Cambridge University Press

to avoid inappropriate denial of insurance or higher insurance fees to patients.

The aims of this pilot study were to examine the annual expenditures on, and utilization of, resources for adults with congenital heart disease in 1997, to compare the expenditures with age and gender corrected data from the general Belgian population, and to explore whether they could be predicted by 1996 clinical and demographic factors.

Materials and methods

Population studied

For our study, we enrolled all patients with congenital heart disease of 18 years or older, who were examined during an in- or out-patient visit, including an echocardiogram, between July 1, 1996 and December 31, 1996 at the University Hospitals of Leuven, Belgium. In all, we identified retrospectively a total of 212 potential patients. Of the group, 9 could not be contacted by mail or by telephone to obtain informed consent, six patients refused to participate, and no data on expenditures were available for five patients, resulting in a residual sample of 192 subjects. All patients were Caucasian, which is representative of the racial distribution in the Flemish region where this study was performed. Table 1 summarizes demographic and clinical characteristics of the sample. Of the total, 110 were male (57.3%), and 82 were female (42.7%). The median age was 21.8 (Q_1 = 19.3;Q_3 = 26.9) years. The majority of the patients were employed (47.1%) or were students (40.6%). Only 6% was unable to work, and approximately 10% were in poor functional status, in grades III or IV of the Ability Index.[20] Cardiac surgical and therapeutic catheter interventions had been performed in 65.8%, and 12.6%, of the patients, respectively (Table 1). The most prevalent primary diagnoses, based on the 9th revision of the International Classification of Disease,[21] were Tetralogy of Fallot (20.1%), ventricular septal defect or Eisenmenger complex (18.8%), coarctation of the aorta (10%), and

Table 1. Demographic and clinical characteristics of 192 adults with congenital heart disease.

Variable		
Gender:	Male	110 (57.3%)
	Female	82 (42.7%)
Median age (in years):		21.8 (Q_1 = 19.3;Q_3 = 26.9)
Educational level:		
Junior High School		16.4%
High school graduate		45.4%
University/College		30.1%
Education for people with learning disabilities		8.2%
Employment status		
Student		40.6%
Working		47.1%
In search of work		6.4%
Not able to work		5.9%
Median home to hospital distance (in kilometers)		46 (Q_1 = 23;Q_3 = 55)
Median age at diagnosis (in months):		4.5 (Q_1 = 0.25;Q_3 = 42)
Ability Index[20]:	I	46.4%
	II	43.2%
	III	9.9%
	IV	0.5%
Former cardiac surgery		
1 operation		36.5%
2 operations		21.4%
3 operations		6.3%
4 operations		1.6%
Former therapeutic catheter interventions		
1 intervention		8.4%
2 interventions		2.1%
3 interventions		2.1%

https://doi.org/10.1017/S1047951101000336 Published online by Cambridge University Press

congenital stenosis of the aortic valve (10%). Other diagnoses occurred in less then 10% of the patients (Table 2).

Setting

Belgium has a compulsory system for health insurance, covering almost the entire population. Insurance is embedded in the national social security system, although management and administration is assigned to 6 non-governmental associations known as sickness funds.[22] The associations, in principle, are non-commercial and nonprofit-making, but are ideologically aligned. Contributions of employers, employees, and the self-employed, coupled with government subsidies, are distributed over the 6 associations. Each citizen is obliged to hold membership in one of the associations, which are responsible for the reimbursements of charges for health care.

Primary and hospital care are paid on a fee-for-service base. Professional fees are negotiated between the professional syndicates and the health insurance associations, and are formalized nationally by the government, resulting in the absence of competition among the providers of health care. For hospital stays, the government calculates annually a standard cost per day for each individual hospital, based on its structural characteristics such as the number of beds and specialization, its nursing requirements, and its medical activities.[23] The average length of stay in Belgian hospitals in 1997 was 8.79 days.[24] There are strong financial incentives to reduce length of stay. Approximately 75% of the professional fees, and 95% of the charges for hospital stay and nursing care, are reimbursed, resulting in relatively small out-of-pocket costs for patients.

Pharmaceuticals are classified in several groups. Rates of reimbursement are determined according to therapeutic and social utility, ranging from 100% for life-saving drugs to 0% for over-the-counter drugs. The provision of primary care in Belgium is extensive, probably due to the high number of general practitioners, even though there is open access to specialty and hospital care. The percentage of gross domestic product spent on health care in 1997 in Belgium was 7.6%.[25]

For our purposes, we have divided expenditures into reimbursements made by the health insurance associations, and out-of-pocket expenses, the latter being the portion paid by the patients.

Sources and collection of data

Patients eligible for the study were recruited from the computerized patient files of the department of

Table 2. Prevalence of primary medical diagnoses in the sample of 192 adults with congenital heart disease

Primary medical diagnosis	ICD-9-CM code[21]	Prevalence
Tetralogy of Fallot	745.2	20.1%
Ventricular Septal Defect or Eisenmenger complex	745.4	18.8%
Coarctation of the aorta	747.1	10%
Congenital stenosis of aortic valve	746.3	10%
Complete transposition	745.10	7.8%
Congenital mitral insufficiency	746.6	6.7%
Atrial septal defect in the oval fossa	745.5	5.7%
Pulmonary valvar stenosis (congenital)	746.02	5.7%
Stenosis of pulmonary artery	747.3	3.6%
Infundibular or subvalvar pulmonary stenosis	746.83	3.1%
Subaortic stenosis	746.81	2.6%
Supra(valvar)-aortic stenosis	747.22	2.6%
Corrected transposition	745.12	2.1%
Double Inlet Left Ventricle, Double Inlet Right Ventricle, Double Outlet Left Ventricle or Univentricular heart	745.3	2.1%
Ostium primum defect	745.61	1.6%
Congenital insufficiency of aortic valve	746.4	1.6%
Atrio-Ventricular Septal Defect – Endocardial Cushion defect	745.60	1.6%
Hypertrophic obstructive cardiomyopathy	425.1	1%
Congestive cardiomyopathy	425.4	1%
Paroxysmal supraventricular tachycardia	427.0	1%
Pulmonary hypertension	416.8	0.5%
Tricuspid atresia and stenosis (congenital)	746.1	0.5%
Ebstein's malformation	746.2	0.5%
Coronary arterial anomaly	746.85	0.5%
Patency of arterial duct	747.0	0.5%
Partially anomalous pulmonary venous connection	747.42	0.5%

https://doi.org/10.1017/S1047951101000336 Published online by Cambridge University Press

congenital cardiology in our center. A letter with information was sent to the patients, after they were contacted by telephone to inform them about the purpose of the study. Patients were asked to give written permission to retrieve data regarding their use of the resources for health care.

Demographic and clinical data were collected from the medical records. Demographic variables included age, gender, educational level, employment status and distance from the patients' home to the University Hospitals. Clinical variables referred to the condition at the last examination in 1996, and comprised medical diagnosis, age at diagnosis, functional status, comorbidity, mental retardation, syndromal associations, chronic disease score, medical interventions, occurrence of symptoms, hemodynamic and electrophysiological parameters, and cardio-thoracic index. Operationalisation, and measurement, of these variables are described in Table 3. Note that in this study we measured only the objective needs of the model created by Andersen & Newman, these being the variables which are systematically collected by physicians in their assessment of the condition of their patient. It is known, however, that these variables explain only a portion of the variance in uses of resources for health care. Some variables were previously identified as determinants of either uses of resources, or expenditure on, health care,[1,3,9–16,18,20] whereas the impact of other variables has not yet been explored.

Individualized data on expenditures and utilization for 1997 for patients included in this study was provided by three participating health insurance associations, which together insure more than 75% of the Belgian population. The use of archival data of these associations is the most complete and reliable method obtaining information on expenditures on health care in Belgium, because records are maintained of expenses paid by both the associations and patients themselves. For this study, reimbursements and out-of-pocket expenses for primary care, hospital care, and overall expenses for 1997, were retrieved, including all charges for hospitalization, radiographic and other technical examinations, laboratory tests, primary care and professional fees, not limited to the cardiac anomaly. Hospital expenses in this study refer not only to charges incurred for hospitalizations, but also for outpatient visits and technical examinations. Data on expenses for medication were not available from the associations, but reimbursement and out-of-pocket expenses could be estimated based on the prescribed medication documented in the patient record. For the calculation of expenditures for medication, we included only drugs which are taken continuously. Drugs excluded from analysis were antibiotics, cough syrup, medication against migraine, throat spray, vitamins, antifungal drugs, as well as contraceptives. Computing the expenditures, it is assumed that patients take the prescribed medication for at least 1year, and that they were fully compliant. The overall expenditures are not merely a summation of hospital and primary care. Some health services, for instance institutional care, are not included in primary or hospital care, but remain reflected in the overall expenses. We have expressed expenditures in 1997 ECU*, to allow comparisons across European countries. Use of resources was calculated in terms of number of hospitalizations, length of stay, and number of cardiac and non-cardiac operations and cardiac catheterizations. In accordance with Belgian law, both data sets with clinical and expenditure information were merged and made anonymous by the Crossroads Bank for Social Security (Kruispuntbank van de Sociale Zekerheid), an independent governmental organization, to guarantee anonymity of the patients.

Data for expenditure of the general population, corrected for age and gender, could be provided by only one health insurance association, insuring 75% of the patients in this sample. Although only data from 1998 were available, expenditures for 1997 could be estimated based on the average increase of overall reimbursements.[29]

Analysis of data

Descriptive statistics of demographic and clinical variables, after having been checked for normality, were expressed in mean values and standard error of the mean for normally distributed continuous variables; in medians and quartiles (25th percentile; 75th percentile) for continuous variables with a non-normal distribution; and in frequencies for nominal variables. Ordinal data were expressed in medians or proportions. Though expenditure data are often skewed, both means and medians were calculated to illustrate the skewness of the distribution, to compute the total expenditures, and to compare the results with other studies, which report mostly mean expenditures. Linear relations between ordinal and continuous variables were determined by Kendall's Tau-b correlation coefficient. The Mann-Whitney U test and Kruskal-Wallis test were used for univariate group comparison. Multiple regression analysis was

* Average exchange rate in 1997 for 1 ECU was 40.4109 Belgian Francs

https://doi.org/10.1017/S1047951101000336 Published online by Cambridge University Press

Table 3. Operationalization and measurement of clinical variables.

Variable	Level of measurement	Scores
Primary medical diagnosis using ICD-9-CM[21]	nominal	Codes from 415 to 429 and from 745 to 747
Age at diagnosis	continuous	
Functional status using:		
NYHA-classification[26]	ordinal	Class I to IV
Ability Index[20]	ordinal	Class I to IV
Adapted categorization used in Garson et al[1]	ordinal	Class I to V
Classification constructed for this study	ordinal	Class I to V
Comorbidity using Charlson's comorbidity index[27]	continuous	
Mental retardation	dichotomous	Yes No
Syndromal associations	dichotomous	Yes No
Chronic Disease Score[28]	continuous	
Former medical interventions:	continuous	
Number of diagnostic catheter interventions		
Number of therapeutic catheter interventions		
Number of cardiac surgery		
Number of non cardiac surgery		
Types of former cardiac surgery	nominal	No surgery Palliative surgery Reparative surgery Heart or heart-lung transplantion
Symptomatology:	continuous	
Number of symptoms of arrhythmia's		
Number of precordialgias		
Number of symptoms of right heart failure		
Number of symptoms of left heart failure (referring to the systemic ventricle)		
Number of atypical symptoms		
Cyanosis	dichotomous	Yes No
Central venous pressure	dichotomous	Normal Abnormal (≥7 cm H20)
Left ventricular ejection fraction	dichotomous	Normal Abnormal (≥30%)
Left ventricular end diastolic diameter	dichotomous	Normal Abnormal (≥54 mm)
Aortic valvar insufficiency	ordinal	0/4 1/4 2/4 3/4 4/4
Aortic valvar stenosis	dichotomous	Yes No
Pulmonary valvar insufficiency	ordinal	1/4 2/4 3/4 4/4 atresia
Pulmonary valvar stenosis	dichotomous	Yes No
Tricuspid valvar insufficiency	ordinal	1/4 2/4 3/4 4/4 atresia
Tricuspid valvar stenosis	dichotomous	Yes No

https://doi.org/10.1017/S1047951101000336 Published online by Cambridge University Press

Table 3. continued

Variable	Level of measurement	Scores
Mitral valvar stenosis	dichotomous	Yes No
Heart rhythm	dichotomous	Normal (sinus rhythm) Abnormal
Heart rate	continuous	
Axis of the heart	dichotomous	Normal Abnormal (right, left or extreme deviation)
PR distance	dichotomous	Normal Abnormal (> 5 mm)
Conduction defects	dichotomous	Yes No
Cardio-Thoracic Index	dichotomous	Normal Abnormal (≥53)

performed after logarithmic transformation of the dependent variables, in order to satisfy the assumptions. A backward stepwise procedure was used, starting with all covariates in the model. After this procedure, factors being associated at $p \leq 0.1$ level in the univariate analysis were re-entered into the model. Based on the data from the general population, patients were assigned to groups of low or high health consumption, where the mean expenditures of the former group are equal to the general population, whereas the latter group is responsible for expenses in our sample higher than the reference population. The cut-off point was established by a stepwise exclusion of the patients with the highest overall reimbursement, until the reimbursement was equal to the overall reimbursement for the general population. Variables predicting high or low health consumption were determined using logistic regression. The level of significance was set at $p \leq 0.05$.

Results

Use of resources for health care

In 1997, 20.3% of the patients included in this study required a hospital admission. For 14.6%, 4.7%, 0.5% and 0.5% of the patients, 1, 2, 3 and 4 hospitalizations were needed, respectively, resulting in an average of 0.23 hospitalizations per patient. The median length of stay for hospitalized patients was 5 (Q_1 = 2; Q_3 = 13; range 1–84) days. Ten cardiosurgical procedures were performed in 7 patients (3.7%; 0.052 operations/patient), of which 2 heart and 1 heart-lung transplantation, and 33 non-cardiac operations were documented in 18 patients (9.4%; 0.172 operations/patient). In 9 patients (4.7%), at least 1 diagnostic cardiac catheterization was performed in 1997, of which 2 patients – both being transplanted – underwent 15 and 31 catheterizations including myocardial biopsies, respectively. Only 1 patient (0.5%) included in this study needed a catheter intervention. Use of medications was documented in 27% of the patients. Drugs most frequently used were glycosides (10.1%), diuretics (6.3%), beta-blockers (5.8%) and inhibitors of angiotensin converting enzyme (5.3%).

Expenditures on health care in patients with congenital heart disease

The average overall reimbursement for the 192 patients in 1997 was 1794.5 ECU (SEM: 471.2; median: 256) and ranged from 2 to 66000.2 ECU. Patients paid, on average, 189.5 ECU (SEM: 32.6; median: 54; range 0 – 4191) out-of-pocket. The average reimbursement and out-of-pocket expenses were 1079 ECU and 48.8 ECU for hospital care, and 137.8 ECU and 37.8 ECU for primary care, respectively (Table 4). These results were dominated by three outliers for whom the reimbursement was 36119.6 ECU, 44331.2 ECU and 66000.2 ECU, respectively, due to a heart or heart-lung transplantation. Excluding these outliers in this analysis, average reimbursement decreased to 1048.1 ECU (median: 252 ECU; range 2 – 13434 ECU) and out-of-pocket expenses declined to 163.8 ECU (median: 52; range 0 – 4190.8 ECU). The medians, however, barely changed. The mean expenditures were considerably higher than the medians, due to a small proportion of expensive users, skewing the distribution of expenditures. These data include all charges for hospital, primary and institutional care, as well as professional fees, and hospital costs include also outpatient visits and technical examinations. Reimbursement and out-of-pocket expenses for medication were estimated to be 78.0 ECU and 20.0 ECU, respectively, including the

https://doi.org/10.1017/S1047951101000336 Published online by Cambridge University Press

immunosuppressive therapy in the 3 transplant recipients. Excluding these three patients would result in a reimbursement of 38 ECU, and out-of-pocket expenses of 15 ECU, for medication.

For the most prevalent diagnoses occurring in this sample, the expenditures for 1997 were calculated. Treatment and follow-up of the Eisenmenger ventricular septal defect was found to be the most expensive in terms of reimbursement (Figure 1). Out-of-pocket expenses were highest in patients with pulmonary valvar stenosis. Even within groups with the same primary diagnosis, a wide variance in expenditures could be observed, as illustrated by the large SEM for reimbursement for Tetralogy of Fallot (SEM= 911.6 ECU), ventricular septal defect or Eisenmenger complex (SEM= 2064.5 ECU), coarctation of the aorta (SEM= 608.9 ECU) and congenital stenosis of the aortic valve (SEM= 207.8 ECU). This finding is related to the expenditures of the patients undergoing transplantation (one with Tetralogy of Fallot; one with ventricular septal defect, and one with the Eisenmenger complex) as well as to the small sizes of the samples. Exclusion of the three transplant recipients would result in a reimbursement of 884.4 ECU for Tetralogy of Fallot, and 997.4 ECU for ventricular septal defect and the Eisenmenger complex, giving a more balanced figure of the expenditures per diagnostic category. Having excluded these patients, coarctation of the aorta, would become the most expensive defect in terms of reimbursement.

Comparison with the general population

Gender and age corrected data from the general population revealed that, in 1998, on average 436.4 ECU was reimbursed per patient for health care. Estimations for 1997, based on an increase of 5.99% in overall reimbursements from 1997 to 1998,[29] suggested an average overall reimbursement of 411.7 ECU, which is considerably lower than the amount of 1794.5 ECU required for patients with congenital heart disease. When patients with congenital heart disease having an overall reimbursement of 2500 ECU or higher (n= 24; 12.5%) were excluded from analysis, the mean reimbursement decreased to 413.4 ECU, which was comparable with the general population. This indicated that only 12.5% of patients with congenital heart disease were responsible for higher average costs of health care in this population, and can consequently be regarded as high users of resources. Of the patients, 61% required less overall reimbursement in 1997 than the mean reimbursement for their counterparts in the general population.

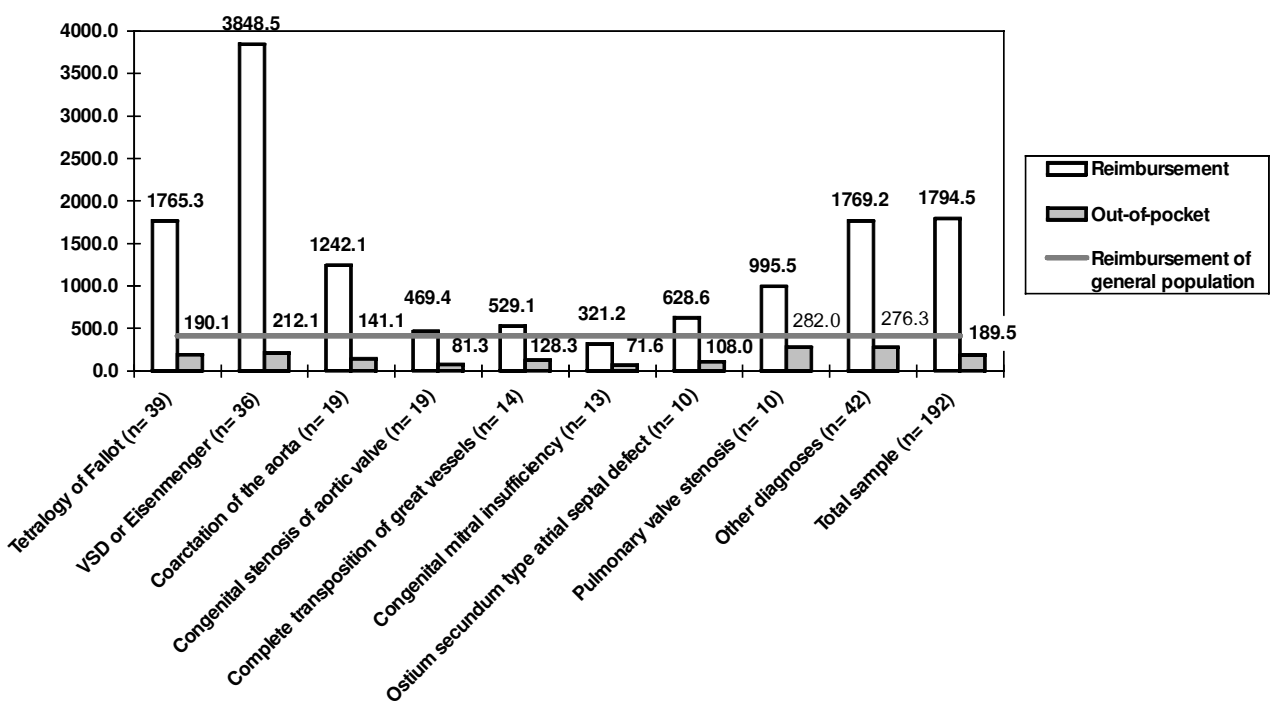

Figure 1:
Mean health care expenditures for the most prevalent diagnoses in this sample.

https://doi.org/10.1017/S1047951101000336 Published online by Cambridge University Press

Predictors of expenditure

For the regression analyses to determine predictors of expenditure, we excluded the 3 recipients of heart and heart-lung transplantation, as their expenditures were extremely outlying, and because three patients are insufficient to draw conclusions regarding predictors of expenses in recipients of transplantation. Univariate analysis revealed that an abnormal left ventricular end-diastolic diameter, female gender, functional impairment – as measured by the classification developed for this study – and higher age were associated with both increased overall reimbursement and overall out-of-pocket expenses in 1997 (Table 5). These four variables, in addition to the number of symptoms of left heart failure, were also determining higher reimbursement for hospital care. Out-of-pocket expenses for hospital care were determined by an abnormal left ventricular end-diastolic diameter, female gender and higher age. For primary care, increased reimbursement was associated with female gender, functional impairment, tricuspid valvar insufficiency, and cyanosis; and higher out-of-pocket expenses with functional impairment, higher number of symptoms of arrhythmia, and increased heart rate (Table 5). Consumers of increased health care (≥2500 ECU for overall reimbursement) differed from low consumers in that they had a higher age, more symptoms of left heart failure, higher degrees of mitral valvar insufficiency, and more often an abnormal left ventricular end-diastolic diameter (Table 5). Though these variables are significantly associated with expenditures, correlations are very low.

After logarithmic transformation of the dependent variables, multiple regression analysis was performed to determine predicting variables for reimbursement and out-of-pocket expenses for hospital care, primary care and overall expenditures, respectively. These results showed that a

Table 4. Reimbursements and out-of-pocket expenses in 1997.

Expenditures	Mean (SEM)	Median (Quartiles)	Range
Overall reimbursement 1997	1794.5 (471.2)	256.0 (Q_1= 96.9; Q_3= 890.1)	2.0 – 66000.2
Overall out-of-pocket 1997	189.5 (32.6)	54.0 (Q_1= 25.4; Q_3= 138.3)	0 – 4191.0
Reimbursement hospital care 1997	1079.0 (252.5)	147.1 (Q_1= 26.6; Q_3= 577.5)	0 – 28999.2
Out-of-pocket hospital care 1997	48.8 (6.1)	22.6 (Q_1= 6.7; Q_3= 54.9)	0 – 878.2
Reimbursement primary care 1997	137.8 (15.0)	71.8 (Q_1= 32.4; Q_3= 168.7)	0 – 2018.8
Out-of-pocket primary care 1997	37.8 (3.3)	21.8 (Q1= 8.5; Q3= 50.9)	0 – 347.2

Table 5. Predictors of health care expenses based on univariate analysis.

Predicting variables	Overall reimbursement	Overall out-of-pocket	Reimbursement hospital care	Out of pocket hospital care	Reimbursement primary care	Out of pocket primary care	High health care consumers (≤2500ECU)
left ventricle end-diastolic diameter	U= 135*	U= 1321*	U= 1282*	U= 1297*			χ^2= 3.86*
female gender	U= 354#	U= 3308#	U= 3612*	U= 3524#	U= 3602#		
functional impairment	τ= 0.13*	τ= 0.13*	τ= 0.12*		τ= 0.16*	τ= 0.16#	
higher age	τ= 0.12*	τ= 0.10*	τ= 0.12*	τ= 0.10*			U= 1285#
number of symptoms of left heart			τ= 0.13*				χ^2= 4.79*
tricuspid valvar insufficiency					τ= 0.14*		
cyanosis					U= 492*		
number of symptoms of arrhythmias						τ= 0.16*	
higher heart rate						τ= 0.14*	
mitral valvar insufficiency							U= 1492*

U= Mann Whitney U Test statistic; τ= Kendall's Tau-b correlation coefficient; χ^2= Chi-square
*p≤0.05; # p≤0.01

https://doi.org/10.1017/S1047951101000336 Published online by Cambridge University Press

higher overall reimbursement for patients with congenital heart disease in 1997 could be predicted from higher age and an abnormal left ventricular end-diastolic diameter, while an increase in overall out-of-pocket expenses was predicted by higher age, abnormal left ventricular end-diastolic diameter, and female gender (Table 6). For higher reimbursement of hospital care, predictors were abnormal left ventricular end-diastolic diameter and female gender, whereas higher out-of-pocket expenses for hospital care were determined by a lower number of diagnostic cardiac catheterizations, functional impairment, abnormal left ventricular end-diastolic diameter and female gender. Increased reimbursement of primary care could be predicted by a variety of variables: a lower degree of aortic valvar insufficiency, a lower number of non-cardiac operations, a lower extent of precordialgia, a higher number of symptoms of arrhythmia, lower comorbidity, functional impairment, higher age, left ventricular end-diastolic diameter, and a normal left ventricular ejection fraction. For the prediction of out-of-pocket expenses for primary care, higher age, functional impairment, normal left ventricular ejection fraction and the absence of tricuspid valvar stenosis could be used. Multiple logistic regression revealed that only the number of symptoms of left heart failure was of value in determining the high or low use of resources for health care (area under the curve = 0.587; 95% CI = 0.444–0.730). The explained variance for the seven models varied from 4.5% to 20.6% (Table 6).

Discussion

Data on expenditure

Our pilot study has shown that a limited proportion of one-eighth of adults with congenital heart disease generate high to very high health care

Table 6. Predictors of health care expenditures based on multivariate analysis

Dependent variables	Trans-formation	Covariate	Esti-mation	Standard Error	p-value	R^2
Overall reimbursement 1997	In(x)	constant	7.857	0.491		0.085
		age	0.059	0.020	0.004	
		left ventricle end-diastolic diameter	0.883	0.340	0.0103	
Overall out-of-pocket 1997	In(x+ 100)	constant	6.685	0.0411		0.106
		age	0.035	0.017	0.0411	
		left ventricle end-diastolic diameter	0.856	0.282	0.0028	
		gender	0.461	0.203	0.0242	
Reinbursement hospital care 1997	In(x+ 100)	constant	7.902	0.229		0.080
		left ventricle end-diastolic diameter	1.404	0.461	0.0027	
		gender	0.789	0.328	0.0173	
Out-of-pocket hospital care 1997	In(x+ 100)	constant	5.936	0.338		0.111
		number of diagnostic catheterizations	–0.233	0.091	0.0110	
		functional impairment	0.331	0.132	0.0132	
		left ventricle end-diastolic diameter	0.602	0.281	0.0377	
		gender	0.529	0.198	0.0083	
Reimbursement primary care 1997	In(x+ 100)	constant	7.057	0.493		0.206
		aortic valve insufficiency	–0.357	0.124	0.0045	
		number of non cardiac operations	–0.435	0.198	0.0297	
		number of precordialgia's	–0.762	0.351	0.0314	
		number of arrhythmia symptoms	0.458	0.186	0.0149	
		comorbidity	–0.470	0.226	0.0392	
		functional impairment	0.247	0.112	0.0290	
		age	0.034	0.016	0.0308	
		left ventricle end-diastolic diameter	0.583	0.276	0.0364	
		left ventricular ejection fraction	–0.749	0.317	0.0194	
Out-of-pocket primary care 1997	In(x+ 100)	constant	5.668	0.372		0.090
		age	0.030	0.013	0.0166	
		functional impairment	0.209	0.079	0.0089	
		left ventricular ejection fraction	–0.501	0.248	0.0447	
		tricuspid valve stenosis	–1.175	0.572	0.0416	
High health care consumers (≥2500 ECU)	n.a.	constant	–2.365	0.278		0.045*
		number of symptoms of left heart failure	1.101	0.465	0.0297	

n.a. = not applicable; * Nagelkerke R^2

https://doi.org/10.1017/S1047951101000336 Published online by Cambridge University Press

expenses, while the expenditures of the majority of these patients are comparable to the general population. This may be explained by the small number of patients undergoing cardiac surgery, because these procedures are considered as highly expensive. We had not expected the expenditures for the majority of adults to be very high, as arguably most of the costs for health care in this population are generated in the first 21 years of life.[1] Most medical expenses for common cardiac malformations are associated with the initial surgical repair in childhood; the ongoing costs of treatment are relatively small,[9] except for patients needing transplantation of the heart or the heart and lungs. Hospitalization of one-fifth of the sample was substantially higher than the 5 to 7% previously reported.[3] Our results, however, included non-cardiac admissions, as we could not determine from the database whether the hospitalization was related directly to the cardiac defect.

The overall charges in 1997 for our patients with congenital heart disease was 2082 ECU*, comprising reimbursement along with out-of-pocket expenses and medication, was double the estimates of Garson et al.[1] who calculated the annual charges for adults with congenital heart disease at $1120 per year in 1992 dollars.† Yet, there are some issues to consider in comparing these results. First, the estimates of Garson et al.[1] included only hospital charges, professional fees and medication prices related to treatment and follow-up of the congenital cardiac malformation, while our study included all charges and fees for both hospital and primary care, regardless of the association with the cardiac defect. From the perspective of the health insurer, the second approach is preferred in order to allow computation of the overall financial impact of reimbursement for this population. Second, based on the categorization used by Garson et al.,[1] it is suggested that our sample included substantially more patients with benign heart defects, and less patients with Mustard or Fontan operations or pulmonary vascular obstructive disease, implying that the profile of patients in the study of Garson et al. was more severe than in ours. This would suggest higher expenses in Garson et al.[1] compared to our study. It is not clear, however, whether they included patients in need for a transplantation, this having a tremendous impact on the expenditures. Third, the longitudinal perspective of Garson et al.[1] resulted in an estimation of charges in the period from birth to 40 years of age, while our study had a time-frame of 1 year. Indeed, a cost-of-illness study would provide more accurate information on the costs of health care in these patients, particularly because our sample comprised predominantly patients in the second decade of their life. Moreover, our study indicated that a higher age was associated with increased costs. Fourth, our study was conducted in Belgium, which has a system for health care characterized by solidarity and nonprofit-making hospitals, contrasting with the health care system of the United States of America. Furthermore, the variation in prices in Belgium is limited to the charges for hospital stay, since professional fees are predominantly established nationally.

These pilot data offered valuable information on costs of health care in patients with congenital heart disease in the setting of Belgium. Precise estimates of the costs were not possible, because the sample is too small for estimations of costs that are driven by small-probability, but high-cost events such as transplantation. Though only few patients with congenital cardiac disease are in need of transplantation, the expenditures for these patients are of particular importance to insurers, as those costs determine their expected expenditures. In addition, it is known that congenital heart diseases constitute a heterogeneous group, which makes the costs of health care in this population even more difficult to estimate.

Predictors

Our study identified some crucial demographic and clinical parameters to detect patients at risk for higher costs in health care, excluding patients requiring transplantation. In general, an abnormal left ventricular end-diastolic diameter, female gender, functional impairment, and higher age were both univariately and multivariately associated with higher expenses. The association of expenditures with abnormal left ventricular end-diastolic diameter and functional impairment can be explained by the more intensive follow-up which is needed in those patients. The finding that female gender and higher age are associated with higher expenditures is also found in studies performed in the general population.[30] It is in contrast with data from different populations of patients with chronic disease, as age and gender are not demonstrated to be predictors for use of resources in health

* = 84136 Belgian francs

† In 1997 Belgian francs, this would be 42000 BF at Purchasing Power Parity.

https://doi.org/10.1017/S1047951101000336 Published online by Cambridge University Press

care.[18] The direction of the association of expenditures in primary care with aortic valvar stenosis, comorbidity, left ventricular ejection fraction, tricuspid valvar stenosis, non-cardiac operations and precordialgia was unexpected. We hypothesized that patients with those problems are more often followed-up by the congenital cardiologist instead of the general practitioner. Therefore, the expenditures for primary care in those patients might be lower.

The explained variance of the multiple regression models was limited. Yet, this finding was expected, because only the objective need component of Andersen & Newman's model[17] was measured in this study. Other variables, in addition to the objective needs, have to be assessed to explain more of the variance of health care expenditures in patients with congenital heart disease, such as quality of life, perceived health, and psychological distress. Though prediction of costs by means of the determinants used in this study seems to be insufficient, our study has highlighted some clinical and demographic factors that must be scrutinized in future prospective studies. Refinement of the predictive models is needed in order to allow accurate identification of patients with expenditures equal to the general population. This will allow parity status for these patients with the general population, and their integration into plans for general insurance, without higher fees or exclusion of the cardiac defect from coverage. These measures should be exclusively reserved for patients demonstrated to have higher costs for health care when compared to the general population.

Methodological issues

The sample included in this study may not be fully representative for the population of patients with congenital heart disease. Subjects were selected if they were examined by a congenital cardiologist during the last 6 months of 1996. A substantial proportion of patients born with a cardiac defect is treated in the first years of life, and does not need regular follow-up in adulthood. These patients, who may be assumed to have limited expenditures, were not included in this study. This may have resulted in an overestimation of the expenses in health care for the population with congenital cardiac defects at large. Furthermore, selecting patients in the last 6 months of 1996 for the calculation of expenditures in 1997 may also have introduced some bias. If the cardiologist had decided that a catheter intervention was needed, this procedure would have been planned shortly after the examination in 1996, and not postponed for several months. This explains the very low number of subjects in this study undergoing a catheter intervention in 1997. The primary aim of the study, nonetheless, was to provide pilot data and not to furnish generalizable results.

Due to the retrospective design of this study, medical records of the patients had to be reviewed. As some variables used in this study were not documented systematically, a considerable number of missing values was observed. On the other hand, since most adults with congenital heart disease are followed-up by the same physician, inter-rater consistency of the medical reports is not an issue.

Medication expenditures were based on the prescribed medication documented in the patient records, as these data were not available from the health insurance associations. For each drug, the price and reimbursement in 1997 was determined. It is possible that the drugs prescribed by a general practitioner were not known by the cardiologist, possibly resulting in an underestimation of the expenditures for medication.

This study reported the charges for health care for patients with congenital heart disease and not the true costs. It is noted in the Belgian system for health care that the ratios of costs to charges are not known.

Conclusion

Our pilot study provides crucial information for health care professionals, administrators, makers of health policies, and insurance companies better to understand the economic consequences of the care for patients with congenital heart disease. It demonstrated that expenses vary significantly among the patients, and that only a limited proportion of patients generated expenditures higher than the general population. Based on these preliminary results, it is not defensible to consider congenital heart diseases as a homogeneous group when evaluating suitability for health care expenditures. Differentiation based on scientific evidence is needed. Accurate detection of those patients at risk for high expenditures, and increased use of health services, however, is not yet feasible. Prospective, longitudinal cost-of-illness studies in larger study samples would provide a more balanced view on expenditures, and could offer a basis for developing guidelines for health insurance in patients with congenital heart disease.

Acknowledgement

We express our grateful thanks to Mr. G. Tormans (National Alliance of Christian Mutualities), Dr V.

Werbrouck (National Alliance of Socialistic Mutualities), and Dr. J.P. Bronckaers (National Alliance of Liberal Mutualities) for their collaboration in the execution of this project.

References

1. Garson A, Allen HD, Gersony WM, Gilette PC, Hohn AR, Pinsky WW, Mikhail O. The cost of congenital heart disease in children and adults: a model for multicenter assessment of price and practice variation. Arch Ped Adol Med 1994; 148: 1039—1045.
2. Gersony WM, Hayes CJ, Driscoll DJ, Keane JF, Kindd L, O'Fallon WM, Pieroni DR, Wolfe RR, Weidman WH. Second Natural History Study of congenital heart defects. Circulation 1993; 87: I-52 – I-65.
3. Somerville J. Management of adults with congenital heart disease: an increasing problem. Ann Rev Med 1997; 48: 283–293.
4. Truesdell SC, Clark EB. Health insurance status in a cohort of children and young adults with congenital cardiac diagnosis (abstract). Circulation 1991;84 (suppl): II-386.
5. Allen HD, Gersony WM, Taubert KA. Insurability of the adolescent and young adult with heart disease. Report from the fifth conference on insurability, October 3–4, 1991, Columbus, Ohio. Circulation 1992; 86: 703–710.
6. Celermajer DS, Deanfield JE. Employment and insurance for young adults with congenital heart disease. British Heart Journal 1993; 69: 539–543.
7. Kaemmerer H, Tintner H, König U, Fritsch J, Sechtem U, Höpp W. Psychosoziale Probleme Jugendlicher und Erwachsener mit angeborenen Herzfehlern. [The psychosocial situation of adult patients with congenital heart disease]. Zeitschrift für Kardiologie 1994; 83: 194–200.
8. Klein LW, Calvin JE. Resource utilization in cardiac disease. Kluwer Academic Publishers, Dordrecht, 1999.
9. Pearson GD, Kidd L, Beittel TM, Neill CA. Cardiac care for infants: Determinants of hospital charges for acute care. AJDC 1991; 145: 1397–1400.
10. Watson DC, Bradley LM, Midgley FM, Scott LP. Costs and results of cardiac operations in infants less than 4 months old. Are they worthwhile? J Thorac Cardiovasc Surg 1986; 91: 667–673.
11. Silberbach M, Shumaker, D, Menashe V, Cobanoglu A, Morris C. Predicting hospital charge and length of stay for congenital heart disease surgery. Am J Cardiol 1993; 72: 958–963.
12. Davis JT, Allen HD, Cohen DM. Fiscal impact of a practice pattern for secundum atrial septal defect repair in children. Am J Cardiol 1994; 74: 512–514.
13. Ungerleider RM, Bengur AR, Kessenich AL, Liekweg RJ, Hart EM, Rice BA, Miller CE, Lockwood NW, Knauss SA, Jaggers J, Sanders SP, Greeley WJ. Risk factors for higher cost in congenital heart operations. Ann Thorac Surg 1997; 64: 44–49.
14. Ungerleider RM, Kanter RJ, O'Laughlin M, Bengur AR, Anderson PA, Herlong JR, Li J, Armstrong BE, Tripp ME, Garson A, Meliones JN, Jaggers J, Sanders SP, Greeley WJ. Effect of repair strategy on hospital cost for infants with Tetralogy of Fallot. Ann Surg 1997; 225: 779–783.
15. Parrish MD, Pollock M, Gauthier N, Park J, Hobde B. Sources of variability in hospital costs of atrial septal defect repair. Am J Cardiol 1998; 82: 252–254.
16. Gajarski RJ, Towbin JA, Garson A Jr. Fontan palliation versus heart transplantation: a comparison of charges. Am Heart J 1996; 131: 1169–1174.
17. Andersen R, Newman JF. Societal and individual determinants of medical care utilization in the United States. Milbank Memorial Fund Quarterly 1973; 51: 95–124.
18. de Boer AGEM, Wijker W, de Haes HCJM. Predictors of health care utilization in the chronically ill: a review of the literature. Health Policy 1997; 42: 101–115.
19. Radosevich DM, Kalambokidis Werni TL. A practical guidebook for implementing, analyzing, and reporting outcomes measurement. Health Outcomes Institute, 1996. pp 1–10.
20. Somerville J. Congenital heart disease in adults: a new cardiovascular subspeciality. Circulation 1986; 84: 1881–1890.
21. International classification of diseases, 9th revision, clinical modification, 5th edition. Practice Management Information Corporation, Los Angeles, 1998.
22. Nonneman WLM. Health care in Belgium. In Casparie AF, Hermans HEGM, Paelinck JHP. (Eds). Competitive health care in Europe. Future prospects. Dartmouth, Aldershot, 1990. pp 51–71.
23. Crainich D, Closon M.C. Cost containment and health care reforms in Belgium. In Mossialos E., Le Grand J (Eds.). Health care and cost containment in the European Union. Ashgate Publishing Limited, Aldershot, 1999. pp 219–266.
24. Ministry of Social Affairs, Public Health and Environment. Statistisch jaarboek van de ziekenhuizen. Deel 3: ziekenhuisactiviteiten, dienstjaar 1997. [Statistical report of hospitals. Part 3: hospital activities, year 1997]. Brussels, 1998.
25. OECD. OECD Health data: a comparative analysis of 29 countries. CD Rom 1998.
26. The Criteria Committee of the New York Heart Association, I. Diseases of the heart and blood vessels: nomenclature and criteria for diagnosis. 6th ed. Little Brown, Boston, 1964.
27. Charlson ME, Pompei P, Ales KL, MacKenzie CR. A new method of classifying prognostic comorbidity in longitudinal studies: development and validation. J Chron Dis 1987; 40; 373–383.
28. Clark DO, Von Korff M, Saunders K, Baluch WM, Simon GE. A chronic disease score with empirically derived weights. Med Care 1995; 33: 783–795.
29. Léonard C. Geneeskundige verzorging in 1998 [Medical care in 1998]. CM-informatie 188, 1999. pp 13–15.
30. Hulka BS, Wheat JR. Patterns of utilization: the patient's perspective. Med Care 1985; 23: 438–460.