Different Levels of Care for Follow-up of Adults with Congenital Heart Disease

A Cost Analysis Scrutinizing the Impact on Medical Costs, Hospitalizations, and Emergency Department Visits

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Competing interests

None to declare.

Availability of data and material

Not available.

Code availability

Not applicable.

Ethics approval

The study was approved by the privacy commission (SCSZG/17/184) and the ethical committees of the participating hospitals (S59858, B670201731994, 2017/26JUI/332).

Contribution

RW: conceptualization, methodology, formal analysis, data curation, writing – original draft, visualization, project administration; FO: conceptualization, methodology, data curation, writing – review & editing, project administration; EG: conceptualization, methodology, writing – review & editing; KDG: resources, writing – review & editing; WB: funding acquisition, resources, writing – review & editing; SM: funding acquisition, resources, writing – review & editing; MDH: resources, writing – review & editing; LVB: project administration, writing – review & editing; AM: funding acquisition, writing – review & editing; PM: writing – review & editing, funding acquisition; JDB: supervision, resources, writing – review & editing, funding acquisition; LA: supervision, writing – review & editing, funding acquisition.

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ABSTRACT

Aim

To scrutinize the economic impact of different care levels, such as shared care, in the follow-up of adult congenital heart disease (ACHD) patients.

Methods

The BELgian COngenital heart disease Database combining Administrative and Clinical data (BELCODAC) was analyzed. Patients (N=6,579) were categorized into five care levels based on their cardiac follow-up pattern between 2006 and 2010. Medical costs, hospitalizations, and emergency department visits were measured between 2011 and 2015.

Results

In patients with moderate lesions, highly specialized cardiac care (HSC; exclusive follow-up by ACHD specialists) and shared care with predominantly specialized cardiac care (SC+) were associated with significantly lower medical costs and resource use compared to shared care with predominantly general cardiac care (SC-) and general cardiac care (GCC). In the patient population with mild lesions, HSC was associated with better economic outcomes than SC- and GCC, but SC+ was not. HSC was associated with fewer hospitalizations (-33%) and less pharmaceutical costs (-46.3%) compared to SC+. Patients with mild and moderate lesions in the no cardiac care (NCC) group had better economic outcomes than those in the GCC and SC- groups, but post-hoc analysis revealed that they had a different patient profile than patients under cardiac care.

Conclusion

More specialized care levels are associated with better economic outcomes in patients with mild or moderate lesions in cardiac follow-up. Shared care with strong involvement of ACHD specialists might be a management option to consider. Characteristics of patients without cardiac follow-up but good medium-term economic prospects should be further scrutinized.

Keywords

Healthcare economics; congenital heart disease; health services; follow-up care

JEL classification codes

D610, I110

INTRODUCTION

The adult population with congenital heart disease (CHD) expanded[1,2] over the past decades and the care needs per patient increased. Together this could impose a pressing burden on available medical and financial resources[3,4]. Adult congenital heart disease (ACHD) specialized centers have been established to deliver appropriate care to these patients with lifelong care needs[5,6], but the existing ACHD specialized centers may eventually face saturation as the patient population is expected to keep on growing and ageing. To ensure and to improve the quality and sustainability of the long-term care for adults with CHD, international expert-opinion based recommendations advocated a three-level care model[7-9]. Adults with CHD should be seen at least once at an ACHD specialized center to determine the most appropriate follow-up level based on anatomy and physiological stage: (i) exclusive care at an ACHD specialized center, (ii) shared care between the specialized center and general cardiac care providers, and (iii) non-specialist cardiac care provided by general cardiologists with direct access to specialized care if needed.

The limited research available on the topic of the dominant level of care demonstrated better patient outcomes following specialized cardiac care compared to general cardiac care, particularly in patients with severe lesions[10,11]. Mylotte et al. showed a significant increase of patients referred to ACHD centers after national consensus guideline publication, resulting in a significant decrease in mortality a few years later. The analysis of the impact of shared care on mortality was hampered by the small number of patients categorized in this care group[10]. Cordina et al. demonstrated that general cardiologists deviated significantly more from clinical guidelines than ACHD specialists. Patients followed by general cardiologists subsequently suffered more from negative outcomes (e.g., death, permanent disability) or inadequate management (e.g., not recommending a necessary intervention)[11]. The impact of shared care is ought to be investigated.

The aim of this study was to explore the association between different care levels and future economic outcomes such as medical costs and resource use (i.e., emergency department (ED) visits and hospitalizations). Although a three-level care model was advocated in the above mentioned guidelines, we explored five different care levels. Shared care was dichotomized as the grade of collaboration between general cardiologists and ACHD specialists can vary widely. Furthermore, we aimed to explore the association between no cardiac care and economic outcomes.

METHODS

Database and patient population

The Belgian Congenital Heart Disease Database combining Administrative and Clinical data (BELCODAC) has been used to conduct this retrospective study. A detailed description of the linking process and the included variables have been described in a separate paper[12]. Briefly, the BELCODAC comprises healthcare utilization data from ten consecutive years (i.e., 2006-2015), and clinical data from the same period and before (e.g., information about interventions). Particularly, ten databases from five organizations were merged:

- The Intermutualistic Agency (IMA) is the umbrella organization of the seven Belgian sickness funds. The IMA delivered three population-level databases: (i) the population database (socio-demographic information), (ii) the pharmanet database (medication supply information), and (iii) the medical claims database (medical care information).
- Statistics Belgium collects, processes and distributes data about the Belgian society, and they delivered four population-level databases: (i) the death certificate database, (ii) the socio-demographic database, (iii) the socio-economic database, and (iv) the IPCAL database (income information).
- Ghent University Hospital, University Hospitals Leuven and St-Luc University Hospitals each provided part of the study population and delivered clinical information.

In 2004, Belgian law designated four specialized hospitals to provide the full spectrum of CHD care including congenital cardiac surgery and ACHD care[13], and outpatient clinics have been established in affiliated satellite centers as well. The BELCODAC was populated with all CHD patients from the two specialized Flemish hospitals (Ghent University Hospital and University Hospitals Leuven). Additionally, patients with severe lesions of St-Luc University Hospital

(Brussels Capital region) were included in the BELCODAC to get a reasonable number of patients in all lesion complexity groups[14]. Included patients attended the specialized clinic at least once throughout their life. This was not necessarily between 2006 and 2015 as the visit could have taken place earlier in life. Patients who have never visited a specialized center were not included in the database. For the present study's research aim, a closed cohort was applied with the inclusion of all patients alive and 18 years or older on 01/01/2006, and still alive on 31/12/2010.

Statistical framework

Patients were categorized based on their primary diagnosis into lesion complexity groups (i.e., mild, moderate, severe) as defined by Task Force 1 of the 32nd Bethesda Conference[15], but the presence of a secondary diagnosis could have resulted in getting categorized into a more severe lesion complexity group. Patients per lesion complexity were then categorized in five care levels based on the frequency and proportion of their outpatient cardiology visits with general cardiologists and ACHD specialists between 2006 and 2010. Five care levels were defined: (i) highly specialized cardiac care (HSC), (ii) shared care with predominantly specialized cardiac care (SC+), (iii) shared care with predominantly general cardiac care (SC-), (iv) exclusive general cardiac care (GCC), and (v) no cardiac care (NCC) (see Table 1). Visits with general cardiologists in the ED context were not taken into account while determining the care level.

- Please insert Table 1 around here -

Since there are multiple care levels for each complexity group, we applied the Propensity Score Subclassification method for multiple treatment options to evaluate the impact of care level on outcomes. This method has been found useful in non-randomized study designs to approach randomization. Particularly, this work followed the statistical methodology as described by Zanutto et al.[16]. A detailed description of the statistical procedures (e.g., how we dealt with covariate imbalance) can be found in supplement S1.

To start, for patients with mild, moderate and severe lesions separately, Ordinal Logit Models were used to calculate patient-specific propensity scores. A propensity score can be defined as the chance of receiving a given level of care based on a set of baseline characteristics. In this study, 12 demographic, socio-economic and clinical covariates were selected (i.e., self-

employment, marital status, household type, household income, eligibility for increased reimbursement, an indicator of chronic illness, number of available informal caregivers, number of interventions, age, sex, syndrome, and primary CHD diagnosis).

Then, a region of overlap had to be determined based on the propensity scores. The boundaries of the region of overlap can be defined as the highest minimum score and the lowest maximum score across the five care levels. Patients with a propensity score outside the region of overlap were excluded from further analysis. Summary statistics of the included and excluded patients were checked as the obtained results could only be applied on the subpopulation represented by the region of overlap. Patients included for further analysis were classified in ten equally sized deciles based on their propensity score (five equally sized quintiles for patients with severe lesions because of the smaller sample size).

Outcomes were estimated using generalized linear models (gamma distribution, log-link function) with the care level as independent variable. The impact of care level was adjusted for the propensity score quintile/decile and, if covariate imbalance could not be avoided, for the covariates with remaining imbalance.

The impact of care level between 2006 and 2010 was measured on future economic outcomes between 2011 and 2015, taking a payer's perspective. The primary outcome was total medical costs (cardiac and non-cardiac aggregated), which consisted of hospitalization, outpatient (e.g. honoraria, technical medical provisions), and pharmaceutical costs. All costs between 2011 and 2015 were inflated to the same base year applying the Consumer Price Index[17]. Two types of resource use were considered as secondary outcomes (i.e., ED visits and all-type hospitalizations). All outcomes were recalculated to a measurement per patient year to account for mortality between 2011 and 2015. Results are presented as the percentage difference between care levels. P-values of ≤ 0.05 were considered significant with two-sided tests. We did not adjust for multiple testing: this study should be seen as exploratory as there is currently little literature available on the topic.

Sensitivity analysis: exclusion of deceased patients

Although the results were calculated per patient year, we should acknowledge that the economic outcomes of this study remained subject to the competing risk of death. For example, hospital admissions costs are known to be higher when a patient dies during

admission[18]. Therefore, the robustness of our results was checked by redoing our analyses, but with the exclusion of patients who died during the study period.

Analyses were performed with SAS Enterprise Guide V.7.1 (SAS Institute Inc., Cary, NC, USA).

RESULTS

Demographics

The initial sample consisted of 6,959 patients of which 111, 162 and 107 patients with respectively mild, moderate and severe lesions were excluded because their propensity score was outside the region of overlap. Therefore, the final sample size yielded 3,633 (97.0% of the initial sample size), 2,727 (94.4%), and 219 (67.2%) patients with mild, moderate, and severe lesions respectively, totaling 6,579 patients. Atrial septal defect (ASD) type 2 and ventricular septal defect (VSD) were the most prevalent primary diagnoses in patients with mild lesions. A large majority of patients with mild lesions (80%) had no contact with an ACHD specialist between 2006 and 2010 and were therefore categorized in either care level NCC (42.2%) or GCC (37.8%). Pulmonary valve abnormality, coarctation of the aorta, and tetralogy of Fallot were the most prevalent primary diagnoses in patients with moderate lesions. Nearly half of these patients were classified in care levels representing shared care (care level SC- or SC+) or highly specialized care (care level HSC). Transposition of the great arteries, univentricular physiology, and double outlet right ventricle were the most prevalent primary diagnoses in patients with severe lesions. A large majority of patients in this group (82.2%) were classified in care levels representing shared care or HSC (Table 2).

- Please insert Table 2 around here -

Figure 1 shows the total cost differences between the five care levels for patients with mild and moderate lesions. Figure 2 shows the ED visits and hospitalization rate differences. Full results can be found in Supplement S1. Results for patients with severe lesions were hampered by a small sample size (see also the discussion section).

Patients with moderate lesions

Medical costs

HSC and SC+ were associated with significantly less total costs compared to SC- and GCC. The HSC and SC+ groups incurred significantly less hospitalization costs than the SC- (-41.8% and -42.2% respectively), and the GCC (-34.8% and -35.3% respectively) groups. Pharmaceutical

costs were significantly lower in the HSC and SC+ groups compared to the SC- (-44.6% and -36.3% respectively) and GCC (-27.5% and non-significant respectively) groups. The HSC group also incurred significantly less outpatient costs than the SC- (-13.9%) and GCC (-14.8%) groups. A trend was observed on the outpatient cost difference between on the one hand the SC+ group and on the other hand the SC- and GCC groups.

The NCC group was associated with significantly lower total costs compared to the SC- and GCC groups, caused by higher hospitalization, outpatient, and pharmaceutical costs in the SC- group (+43.8%, +26.3%, and +99.1% respectively), and higher outpatient and pharmaceutical costs in the GCC group (+27.6% and +52.0% respectively).

- Please insert Figure 1 around here -

ED visits and hospitalizations

Patients in the HSC and SC+ groups were significantly less hospitalized than those in the SC- (-38.5% and -24.4% respectively), and the GCC groups (-34.3% and non-significant respectively). The number of ED visits did not differ significantly.

Hospitalizations and ED visits were significantly more frequent in the SC- (+37.7% and +33.2% respectively) and GCC groups (+28.9% and +32.2% respectively) than in the NCC group.

- Please insert Figure 2 around here -

Patients with mild lesions

Medical costs

The HSC group incurred significantly less total costs up to 5 years later than the GCC group, and a trend towards less medical costs was observed compared to the SC- group. Total costs between the SC+ group and SC-, and GCC groups did not differ. Specifically, the HSC group was associated with significantly lower hospitalization costs than the GCC group (-48.2%) and a trend towards lower hospitalization costs than the SC- group (-33.3%). Pharmaceutical costs were significantly lower for the HSC group compared to the SC+ (-46.3%), SC- (-35.1%) and GCC groups (-26.1%). The HSC and SC- groups were also associated with less outpatient costs compared to the GCC group (-14.1% and -15.6% respectively).

The NCC group was associated with less total costs than the GCC group and a trend towards less total costs compared to the SC- group, as a result of higher pharmaceutical costs in the

SC- (+83.3%) and GCC groups (+60.9%), and more hospitalization (+21.5%) and outpatient costs (+23.5%) in the GCC group.

ED visits and hospitalizations

Patients in the HSC group were significantly less hospitalized than those in the SC- group (-30.8%), and the GCC group (-35.2%), and there was a trend towards fewer hospitalizations compared to the SC+ group (-33.0%, p-value=0.07). The HSC group was also associated with significantly fewer ED visits than the SC- group (-29.3%) and a trend towards fewer ED visits than the GCC group (-24.7%, p-value=0.052). No other estimates yielded significant differences.

Patients in the GCC group were significantly more hospitalized (+22.0%) than those in the NCC group, and ED visits occurred significantly more frequently in the SC- (+32.3%) and GCC groups (+24.3%) than in the NCC group.

Differences between patients with and without cardiac follow-up

Although the statistical analyses tried to account for the care levels so that patients in different levels had similar baseline characteristics, some imbalance remained (Supplement S1). Indeed, post-hoc analysis showed how patients without cardiac follow-up differed from all other patients: patients with mild lesions not under cardiac follow-up were younger, had fewer comorbid syndromes, less chronic illness, and more often had a ventricular septal defect as a primary diagnosis. Patients with moderate lesions not under cardiac follow-up had undergone fewer interventions, had less chronic illness, more often had a pulmonary valve or aortic abnormality as a primary diagnosis, and less often a tetralogy of Fallot or coarctation of the aorta.

Sensitivity analysis

Full results of the sensitivity analysis can be found in Supplement S2. The exclusion of patients who died during the study period led to a few statistically significant results becoming insignificant or just a trend towards significance (and vice versa as well). However, this did not lead to substantial differences or conclusion-changing results, which reinforced the initial results.

DISCUSSION

This multi-center study used a retrospective database research design linking administrative and clinical data to determine the impact of care levels on medical costs and resource use in the medium term. Patients with mild or moderate lesions who received cardiac care appeared to incur less medical costs in the medium term (i.e., up to 5 years later) when being followed at more specialized cardiac care levels.

Our economic findings are in line with the clinical results of Cordina et al.[11] and Mylotte et al.[10] who found that, regardless of lesion's complexity grade, patients with CHD benefit from specialized cardiac care over general cardiac care. Our study showed that both patients with mild and moderate lesions under highly specialized cardiac care incurred less hospitalization, outpatient and pharmaceutical costs in the medium term compared to patients under general cardiac care. It is therefore important to keep working on referral patterns from general to specialized cardiac care by improving communication, feedback, and knowledge distribution between care levels[19].

A particular interest of this paper was to investigate the potential role of shared care as suggested in existing clinical guidelines[7-9]. Shared care in this paper was dichotomized in shared care with a high share of specialized cardiac care (SC+) and a low share of specialized cardiac care (SC-). In patients with moderate lesions, the economic outcome in SC+ and HSC did not differ and those two levels of care were associated with fewer hospitalizations and less medical costs compared to SC-. Similarly, in patients with mild lesions, SC- was associated with higher costs, more hospitalizations and ED visits compared to highly specialized care, but not to SC+. Moreover, SC+ was associated with higher pharmaceutical costs and a trend towards more hospitalizations compared to highly specialized cardiac care. On first sight, it may seem contra-intuitive to find statistically significant differences between the SC+ group and the highly specialized cardiac care group in patients with mild lesions while there were no such differences found in patients with moderate lesions. One would expect it to be the other way around. One explanation could be that patients with mild lesions in this study were considerably older on average and previous research demonstrated increasing medical needs with higher age[4,20,21]. For example, CHD patients ≥ 40 years had an 87% higher risk of being hospitalized compared to CHD patients under 30[4]. Following this reasoning, exclusive specialized cardiac care may result in lower costs in older adults with CHD while shared care

may be sufficient for young adult patients with CHD if the ACHD specialist is strongly involved in the follow-up process. The specificity of CHD in conjunction with acquired heart disease[20,22] and non-cardiac comorbidities[23,24,22], and the commonly faced need for (re)operation[25] with advancing age may require exclusive specialized cardiac care.

The economic outcomes of patients who did not receive any cardiac care between 2006 and 2010 were not in line with the expected (and otherwise also observed) relation that higher care levels result in less medical costs and less resource use. Patients without cardiac follow-up had rather good economic prospects, suggesting that things do not go wrong in the medium term. It may be that particular patient groups would not immediately benefit from specialized cardiac care. Our post-hoc analyses provide important information on which patients are less prone to be under cardiac follow-up. These results should be seen as an important first step towards determining which patients do not necessarily benefit from cardiac follow-up. Such research is of particular importance as human and financial resources are limited, and overuse should be avoided so that more resources become available to properly care for those patients in need. These results thus support the recommendation that every patient should be seen at a specialized center at least once[7-9] to determine the appropriate level of care, which may be no cardiac care. However, future research on the specific profile of these patients is warranted as this study cannot make inferences on long-term prospects. It is possible that patients without cardiac follow-up have a subclinical cardiac problem that would already have been noticed and treated to prevent worse outcomes on the long-term if they had had regular cardiac follow-up[26]. The question remains if an infrequent but periodical cardiac check-up would still be relevant for all patients because safe might be better than sorry.

Limitations

Several limitations inherent to the analyses were identified, next to known limitations of retrospective database research such as the fact that data were not primarily collected to answer the research question[27].

First, for methodological reasons, analyses were restricted to the propensity score region of overlap, which is methodologically sound, but hampers the generalization to the entire ACHD population. Patient population characteristics of the excluded patients deviating from the included patient population can be found in Supplement S1.

Second, the small number of patients with severe lesions in some care levels hampered the analysis. Robust inferences could not be drawn from this subpopulation. Moreover, a similar remark could be made concerning the results for patients with mild and moderate lesions. Although the sample sizes were quite large, there was a skewed distribution of patients across the five care levels. Together with strong within-group variance, which is typical for economic data such as cost data, statistically significant results could not always be obtained for substantial numerical differences between care levels.

Third, the categorization of patients led inevitably to a loss of information. The definition we applied for categorization patients into care levels has its limitations because, for example, a patient with only one outpatient contact with an ACHD specialist was categorized in the HSC group while at the same time the recommended follow-up frequency might be higher. Can the patient then be considered as someone in the highest care level? Moreover, the BELCODAC does not allow to study the quality of the collaboration between general cardiologists and ACHD specialists, which may have an important impact on the quality of the shared care, and thus on patient outcomes.

Fourth, we did not adjust for multiple testing because this study should be interpreted as an exploratory study as available literature on the topic is rare. Type 1 errors are less harmful than type 2 errors at this stage. Future confirmatory studies with enough statistical power should adjust for multiple testing as found appropriate.

Last but not least, the analysis of the impact of care level on medical costs and resource use in patients with severe lesions was hampered by the small sample size resulting in wide confidence intervals and a high probability of type 2 errors. Results were therefore not explored in detail. Given previous findings which compared specialized with general cardiac care in patients with severe lesions[10,11], we can expect that with a larger sample size, we would have obtained results in favor of specialized care levels in line with the results that were obtained in patients with mild and moderate lesions, but even more pronounced. However, we cannot make inferences about the potential of shared care with a strong component of specialized care (i.e., care level SC+) next to highly specialized care for patients with severe lesions.

Conclusion

This study indicates that patients with mild and moderate lesions followed exclusively at ACHD specialized centers incurred less medical costs than patients followed by general cardiologists. Shared care with a strong involvement of ACHD specialists in follow-up care might result in lower future medical costs as well, but future research should scrutinize the impact of moderating variables such as patients' age. Finally, research to scrutinize characteristics of patients without cardiac follow-up, but seemingly good economic prospects, is warranted.

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FIGURES

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Fig 1 Total medical costs differences between the five care levels for patients with mild and moderate lesions. HSC: highly specialized cardiac care; SC+: shared care with predominantly specialized cardiac care; SC-: shared care with predominantly general cardiac care; GCC: general cardiac care; NCC: no cardiac care. If the confidence interval crosses the orange line, then there is no statistically significant difference on a p-value threshold of 0.05. A negative percentage means that the cost is lower for patients categorized in the former care level, a positive percentage means that the cost is lower for patients categorized in the latter care level.

Fig 2 Emergency department visits and hospitalization rate differences between the five care levels for patients with mild and moderate lesions. HSC: highly specialized cardiac care; SC+: shared care with predominantly specialized cardiac care; SC-: shared care with predominantly general cardiac care; GCC: general cardiac care; NCC: no cardiac care. If the confidence interval crosses the orange line, then there is no statistically significant difference on a p-value threshold of 0.05. A negative percentage means that the cost is lower for patients categorized in the former care level, a positive percentage means that the cost is lower for patients categorized in the latter care level.

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TABLES

453 **Table 1.** *Definitions of the five care levels.*

Care level	Abbreviation	Definition
Highly specialized cardiac care	HSC	≥ 81% of cardiology visits at the specialized care level
Shared care – predominantly specialized cardiac care	SC+	51 – 80% of cardiology visits at the specialized care level
Shared care – predominantly general cardiac care	SC-	1 – 50% of cardiology visits at the specialized care level
General cardiac care	GCC	All cardiology visits at the general cardiology care level
No cardiac care	NCC	No cardiology visits between 2006 and 2010

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455 **Table 2.** Characteristics of the patient population included for final analysis after propensity score matching

		Mild (N=3,633)		Moderate (N=2,727)		Severe (N=219)
		Mean or %		Mean or %		Mean or %
Age beginning 2006		38.9y		34.1y		28.1y
Age Category beginning 2006	18-29y	34.9%		48.6%		65.8%
	30-39y	20.5%		22.3%		29.7%
	40+y	44.5%		29.1%		4.6%
Sex	Female	56.8%		48.7%		42.0%
Mortality 2011-2015		2.7%		3.5%		5.5%
Care level	NCC	42.2%		20.8%		6.4%
	GCC	37.8%		30.7%		11.4%
	SC-	12.1%		27.8%		47.5%
	SC+	3.1%		10.7%		23.7%
Primary Diagnostic Code	HSC	4.8%		10.1%		11.0%
	AV abnormality	9.7%	TOF	16.0%	UV physiology	21.9%
	ASD type 2	26.5%	CoA	16.5%	Tricuspid atresia	1.8%
	VSD	28.9%	AVSD	3.8%	TOF**	2.7%
			ASD type 1	3.7%	PA with VSD	0.5%
	MV abnormality	8.2%	Ebstein anomaly	1.8%	PA with intact ventricular septum	1.8%
	Other	26.6%	PV abnormality	20.0%	DORV	11.4%
			AV abnormality*	8.5%	DILV	0.5%
			Aortic abnormality	11.6%	TA	1.8%
			LVOT obstruction	1.0%	TGA	48.9%
			ASD type 2*	6.3%	cc-TGA	8.7%
			VSD*	4.6%		
			MV abnormality*	1.4%		
			Pulmonary vein abnormality	4.6%		
			Other*	0.4%		

456 ASD: atrial septal defect; AV: aortic valve; AVSD: atrioventricular septal defect; CoA: coarctation of the aorta;
457 DILV: double inlet left ventricle; DORV: double outlet right ventricle; LVOT: left ventricular outflow tract; MV:
458 mitral valve; PA: pulmonary atresia PV: pulmonary valve; TGA: transposition of the great arteries; cc-TGA:
459 congenitally corrected transposition of the great arteries; TOF: tetralogy of Fallot; UV: univentricular; VSD:
460 ventricular septal defect.

461 GCC: general cardiac care; HSC: highly specialized cardiac care; NCC: no cardiac care; SC-: shared care
462 predominantly general cardiac care; SC+: shared care predominantly specialized cardiac care.

463 * These diagnoses are moderate defects if they are associated with complications (e.g. pulmonary arterial
464 hypertension with ASD/VSD) or with significant functional abnormalities (valvular dysfunction).

465 ** Uncorrected Tetralogy of Fallot with pulmonary arterial hypertension
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Supplement S1 Step-by-step statistical analysis and full results

of the paper

Different Levels of Care for Follow-up of Adults with Congenital Heart Disease; Impact on Medical Costs, Hospitalizations, and Emergency Department Visits

Running title: Different Care Levels for ACHD Follow-up

The analysis conducted in this paper was inspired by a statistical paper from Zanutto et al.¹ on how to use propensity score subclassification for multiple treatment arms. We followed their lead to a great extent but specific data issues urged for a tailored approach (e.g. how we dealt with missing data, the choice of the number of subclassification groups).

Preparation phase

Selection of covariates. For every single patient, a propensity score was calculated based on a set of 12 demographic, socio-economic and clinical covariates. Covariates were either categorical or continuous (Table 1).

Table 1. Covariates to calculate the propensity score.

Variables	Description	Coding
Self-employment	Job status: was the patient self-employed in 2005?	0: No 1: Yes
Marital status	Marital status in 2005.	1: Unmarried 2: Married 3: Widow(er) 4: Divorced
Household type	Household type 2005	1: Single, children 2: Single, no children 3: Partner, children 4: Partner, no children 5: Collective household 6: Other
Increased reimbursement	The patient exceeded in 2005 a certain threshold of out-of-pocket medical costs.	0: No 1: Yes
Household income	Patients divided in quartiles based on household income in 2005.	0: lower 25% 1: Average lower 25% 2: Average higher 25% 3: upper 25%
Chronical ill	The patient had a status of chronically ill and/or had a disability allowance in 2005.	0: No 1: Yes
Available caregivers		Continuous variable

Interventions	Caregiver: someone who's 25 or older; at least partially unemployed or retired; not disabled; not chronically sick. (minimum number available in 2005).	Continuous variable
Age	Number of interventions pre 2006 Stratified in three categories	1: 18-29 years old 2: 30-39 years old 3: 40-... years old
Syndrome		0: absent
Sex	Syndrome (catch 22, trisomy 21, Noonan, Marfan, Turner, Williams) Man or woman	1: present 0: woman 1: man
Primary diagnose	Primary diagnostic code	1: Hypoplastic left heart syndrome 2: Univentricular physiology 3: Tricuspid atresia 4: Tetralogy of Fallot 5: Pulmonary atresia with ventricular septal defect 6: Pulmonary atresia without ventricular septal defect 7: Double outlet right ventricle 8: Double inlet left ventricle 9: Truncus arteriosus 10: Transposition of the great arteries 11: Congenitally corrected transposition of the great arteries 12: Coarctation of the aorta 13: Atrioventricular septal defect 14: Atrial septal defect type 1 15: Ebstein malformation 16: Pulmonary valve abnormality 17: Aortic valve abnormality 18: Aortic abnormality 19: Left ventricular outflow tract obstruction 20: Atrial septal defect type 2 21: Ventricular septal defect 22: Mitral valve abnormality

		23: Pulmonary vein abnormality 24: Other
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Dealing with missing values. Several covariates had a limited number of missing values. These missing values were found to exist in a limited number of patients (n=221, 3.08%). Patients with missing values were excluded from further analyses, resulting in an initial sample size of 6,959 patients.

Dealing with skewed distribution of covariates. The next step was to look at the covariates' distributions. The distributions of the two continuous variables (number of available caregivers and number of interventions) were characterized by an extreme skewness. Therefore, the square roots of these covariates were calculated and used as covariates to calculate the propensity score.

Further analyses were stratified for adult patients with either mild, moderate or severe lesions. The respective sample size before propensity score subclassification was 3,744 (53.8%), 2,889 (41.5%), and 326 (4.7%) patients.

Analysis phase

Step 1. An Ordinal Logit Model was constructed with care level as the dependent variable and the main effects of the 12 covariates (Table 1) as independent variables. A propensity score was calculated per patient. The propensity score can be interpreted as the theoretical chance of being treated in the highest of five care level (Table 2).

Table 2. Definitions of the five care levels. Cardiology visits between 2006 and 2010 were taken into account.

Care level	Abbreviation	Definition
4: Highly specialized care	HSC	≥ 81% of cardiology visits at the specialized care level
3: Shared care – predominantly specialized care	SC+	51 – 80% of cardiology visits at the specialized care level
2: Shared care – predominantly general care	SC-	1 – 50% of cardiology visits at the specialized care level
1: General cardiac care	GCC	All cardiology visits at the general cardiology care level
0: No cardiac care	NCC	No cardiology visits between 2006 and 2010

Step 2. The minimum and maximum propensity score per exposure level was checked. The highest minimum score and the lowest maximum score were taken to define the boundaries (=the region of overlap). Patients with a propensity score lower than the minimum score or higher than the maximum score were deleted from further analysis. For example:

	Range	
	Minimum	Maximum
Care level 0	0.05	0.40
Care level 1	0.02	0.55
Care level 2	0.06	0.45
Care level 3	0.15	0.50
Care level 4	0.10	0.56

In this example the region of overlap is 0.15 – 0.40. Patients with a propensity score below 0.15 or above 0.40 were excluded from further analysis, and therefore called the ‘excluded low group’ and ‘the excluded high group’ respectively.

Step 3. Included patients with mild or moderate lesions were categorized into ten equally sized deciles. Patients with severe lesions were categorized into five equally sized quintiles because of the lower sample size. More groups lead to less imbalance, but it has been demonstrated that the use of quintiles already reduces the existing imbalance with 90%¹⁻³. This resulted in patients being categorized in either a 10x5 or 5x5 matrix:

	Care level 1-5				
Quintiles 1-5 or deciles 1-5					
Deciles 6-10					

Step 4. We checked if any imbalance existed between the five care levels. A Two-Way Anova for continuous variables or a logistic regression for non-continuous variables was modelled with the covariate as dependent variable. Independent variables were the quintile rank, the

care level, and the interaction variable. If the care level or the interaction was significant on a p-value threshold of 0.05, imbalance between care levels existed.

Step 5. An iterative process started with the replication of Step 1 to Step 4, with the difference that in Step 1 an Ordinal Logit Model was constructed with care level as the dependent variable and the main effects of the 12 covariates as independent variables, plus the addition of one or more independent variables: the quadratic or interaction of variables with imbalance were added. The aim was not to have a parsimonious model but to reach as much balance as possible.

Step 6. Summary statistics of the included and excluded patients were checked using the Kruskal Wallis Test with Dwass, Steel, Critchlow-Fligner (DSCF) test for multiple comparisons (P-value ≤ 0.05). As the analysis was restricted to the region of overlap, obtained results can only be applied on the subpopulation represented by this region of overlap.

Step 7. Outcomes were calculated with following formulas:

$$Y_j = \sum_i^{5 \text{ or } 10} \frac{1}{5 \text{ or } 10} Y_{ij}$$

with Y_j = the weighted outcome of patients in care level j ; Y_{ij} = the average outcome of patients in care level j and quintile/decile i . In other words: each quintile/decile had an equal weight in the calculation of the average outcome per care level.

$$SE(Y_j) = \frac{1}{5 \text{ or } 10} \sqrt{\sum_i^{5 \text{ or } 10} \frac{S_{ij}^2}{N_{ij}}}$$

with $SE(Y_j)$ = the standard error of the outcome in care level j ; S_{ij}^2 = the variance in quintile/decile i of care level j ; N_{ij} = the number of patients in quintile/decile i of care level j .

Results were calculated using regression models with the outcome as dependent variable and the care level as independent variable. The regression models were adjusted for the propensity score quintile/decile and, if covariate imbalance could not be avoided, for the covariates with remaining imbalance⁴.

Outcomes were measured in a five year period from 2011 to 2015: total medical costs, total hospitalization costs, total outpatient costs, total pharmaceutical costs, emergency department (ED) visits, and the number of hospitalizations (all calculated per patient year). In particular, we calculated the difference (in percentages) between the five different care levels.

Adult patients with mild lesions

It appeared to be infeasible to delete all imbalance by adding quadratics and interactions. The final Ordinal Logit Model with 12 main effects, one quadratic effect (i.e. number of interventions), and nine interaction effects (i.e. number of interventions*age category, number of interventions*primary diagnose, marital status*household type, household type*household income, age category*household income, age category*marital status, age category*household type, marital status*available caregivers, household type*available caregivers) resulted in remaining imbalance in 6 out of the 12 covariates (i.e. marital status, household type, household income, primary diagnose, age category, and number of interventions). These covariates were added to the final regression model to adjust the impact of care level on the respective outcomes.

Table 3 shows how the excluded patients (N=111, 3.0%) differed from the included patients (N=3,633, 97%). The results did not apply on (i) a small group of patients who can be characterized by having had less available informal caregivers and less interventions, a lower household income, a lower age, with more widow(er)s, a smaller proportion of patients with a partner but no children, and more ventricular septal defect (VSD) as a primary diagnose. And (ii) a small group of patients who can be characterized by having had more interventions, be chronic ill, a greater proportion of males, being unmarried, and who were primarily diagnosed with an aortic valve (AV) abnormality.

Table 3. Covariates, care level, and outcomes of included and excluded patients with mild lesions.

		Excluded low (N=51) Mean/%	Included (N=3,633) Mean/%	Excluded High (N=60) Mean/%
√(number of available informal caregivers)		0.17*	0.40	0.47
√(number of interventions)		0.13*	0.34	1.52*
Self-employment		10%	9%	5%
Increased reimbursement		4%	10%	17%
Chronical ill		2%	7%	20%*
Syndrome		0%	3%	5%
Income rank (quartiles)		0.78*	1.56	1.60
Age category		1.82*	2.10	1.88
Gender (male)		43%	43%	65%*
Marital status	Unmarried	59%	45%	65%*
	Married	27%*	47%	33%
	Widow(er)	10%*	2%	0%
	Divorced	4%	6%	2%
Household type	Single, children	29%*	8%	12%
	Single, no children	24%*	9%	2%
	Partner, children	16%	21%	22%
	Partner, no children	20%*	61%	57%
	Collective household	2%*	0%	5%*
	Other	10%*	1%	3%
Primary diagnostic code	AV abnormality	0%*	10%	88%*
	ASD 2	12%*	27%	2%*

	VSD	88%*	29%	0%*
	MV abnormality	0%	8%	5%
	Other	0%*	27%	5%*
Care level		0.37*	0.90	2.05*
Emergency department visits (per PY)		0.34	0.23	0.24
Hospitalizations (per PY)		0.24	0.29	0.33
Hospitalization cost (per PY)		1 424 €	2 082 €	2 092 €
Pharmaceutical costs (per PY)		488 €	823 €	1 179 €
Ambulant costs (per PY)		1 552 €	1 763 €	2 492 €
Total medical costs (per PY)		3 464 €	4 668 €	5 763 €

**=significantly different from the included group. ASD 2: atrial septal defect type 2; AV: aortic valve; MV: mitral valve; PH: pulmonary hypertension; PY: patient year; VSD: ventricular septal defect. Care level: from 0 (general care) to 4 (highly specialized care) Income rank: 0 (lowest quartile) to 3 (highest quartile). Age category: 1=18-29 years old, 2=30-39 years old, 3=40+ years old.*

Total costs difference

	z Value	Pr > z	Difference	95% Low	95% High
HSC vs SC+	1.25	0.21	-16.1%	-36.2%	10.5%
vs SC-	1.72	0.09	-16.5%	-32.0%	2.5%
vs GCC	3.34	0.00	-27.1%	-39.4%	-12.2%
vs NCC	-0.69	0.49	-6.2%	-21.9%	12.6%
SC+ vs SC-	0.04	0.97	-0.5%	-21.6%	26.3%
vs GCC	1.24	0.21	-13.1%	-30.4%	8.5%
vs NCC	0.98	0.33	11.7%	-10.5%	39.4%
SC- vs GCC	2.14	0.03	-12.7%	-22.9%	-1.1%
vs NCC	1.81	0.07	12.3%	-0.9%	27.2%
GCC vs NCC	5.7	<.0001	28.6%	17.9%	40.2%

Hospitalization costs difference

	z Value	Pr > z	Difference	95% Low	95% High
HSC vs SC+	0.97	0.33	-25.0%	-58.1%	34.3%
vs SC-	1.81	0.07	-33.3%	-57.0%	3.4%
vs GCC	3.27	0.00	-48.2%	-65.1%	-23.2%
vs NCC	-2.31	0.02	-37.1%	-57.6%	-6.9%
SC+ vs SC-	0.46	0.65	-11.1%	-46.4%	47.2%
vs GCC	1.55	0.12	-31.0%	-57.0%	10.5%
vs NCC	-0.73	0.46	-16.2%	-47.9%	34.6%
SC- vs GCC	1.85	0.06	-22.4%	-40.7%	1.5%
vs NCC	-0.43	0.67	-5.7%	-28.2%	23.7%
GCC vs NCC	2.06	0.04	21.5%	1.0%	46.1%

Outpatient costs difference

	z Value	Pr > z	Difference	95% Low	95% High
HSC vs SC+	0.2	0.84	-2.3%	-21.9%	22.2%
vs SC-	-0.2	0.84	1.7%	-13.9%	20.2%
vs GCC	1.98	0.05	-14.1%	-26.1%	-0.1%
vs NCC	0.78	0.44	6.1%	-8.6%	23.2%
SC+ vs SC-	-0.41	0.68	4.1%	-14.2%	26.3%
vs GCC	1.41	0.16	-12.1%	-26.6%	5.2%
vs NCC	0.9	0.37	8.6%	-9.3%	30.0%
SC- vs GCC	3.3	0.00	-15.6%	-23.6%	-6.6%
vs NCC	0.81	0.42	4.3%	-5.8%	15.5%
GCC vs NCC	5.86	<.0001	23.5%	15.1%	32.6%

Pharmaceuticals costs difference

	z Value	Pr > z	Difference	95% Low	95% High
HSC vs SC+	3.36	0.00	-46.3%	-62.6%	-22.8%
vs SC-	3.07	0.00	-35.1%	-50.7%	-14.5%
vs GCC	2.37	0.02	-26.1%	-42.4%	-5.1%
vs NCC	1.41	0.16	19.0%	-6.6%	51.6%
SC+ vs SC-	-1.16	0.25	20.8%	-12.3%	66.3%
vs GCC	-2.09	0.04	37.6%	2.0%	85.6%
vs NCC	5.28	<.0001	121.4%	64.8%	197.5%
SC- vs GCC	-1.56	0.12	14.0%	-3.3%	34.3%
vs NCC	7.21	<.0001	83.3%	55.5%	116.2%
GCC vs NCC	8.26	<.0001	60.9%	43.7%	80.1%

Emergency department visits difference

	z Value	Pr > z	Difference	95% Low	95% High
HSC vs SC+	1.56	0.12	-28.7%	-53.3%	8.9%
vs SC-	2.14	0.03	-29.3%	-48.5%	-2.9%
vs GCC	1.94	0.05	-24.7%	-43.5%	0.3%
vs NCC	-0.46	0.65	-6.4%	-29.6%	24.4%
SC+ vs SC-	0.05	0.96	-0.9%	-31.7%	43.9%
vs GCC	-0.3	0.76	5.5%	-25.4%	49.3%
vs NCC	1.52	0.13	31.1%	-7.4%	85.7%
SC- vs GCC	-0.63	0.53	6.4%	-12.4%	29.3%
vs NCC	2.82	0.00	32.3%	8.9%	60.7%
GCC vs NCC	3.15	0.00	24.3%	8.6%	42.3%

Hospitalizations difference

	z Value	Pr > z	Difference	95% Low	95% High
HSC vs SC+	1.84	0.07	-33.0%	-56.3%	2.6%
vs SC-	2.26	0.02	-30.8%	-49.7%	-4.8%
vs GCC	2.96	0.00	-35.2%	-51.4%	-13.7%
vs NCC	-1.62	0.11	-21.0%	-40.5%	5.1%
SC+ vs SC-	-0.17	0.86	3.4%	-28.6%	49.7%
vs GCC	0.19	0.85	-3.2%	-31.5%	36.7%
vs NCC	0.93	0.35	18.0%	-16.6%	67.1%
SC- vs GCC	0.66	0.51	-6.4%	-23.0%	13.8%
vs NCC	1.33	0.18	14.2%	-6.1%	38.8%
GCC vs NCC	2.86	0.00	22.0%	6.5%	39.8%

Adult patients with moderate lesions

It appeared to be infeasible to delete all imbalance by adding quadratics and interactions. The final Ordinal Logit Model with 12 main effects, and six interaction effects (i.e. number of interventions*age category, age category*household type, age category*primary diagnostic code, number of interventions*primary diagnose, household type*household income, household type*available caregivers, available caregivers*household income) resulted in remaining imbalance in 5 out of the 12 covariates (i.e. household type, household income, primary diagnose, syndrome, and number of interventions). These covariates were added to the final regression model to adjust the impact of care level on the respective outcomes.

Table 4 shows how the excluded patients (N=162, 5.6%) differed from the included patients (N=2,727, 94.4%). The results does not apply on (i) a small group of patients who can be characterized by having had less interventions, having no syndrome, being older, with a different marital status, living more in a collective household, and having a pulmonary valve abnormality or a pulmonary vein abnormality as a primary diagnose. These patients experienced a higher care level on average. And (ii) a small group of patients who could have been characterized by having had more interventions, being more self-employed, being more entitled for an increased reimbursement, suffering more from a syndrome, being younger, being unmarried, living more in a collective household, and who were relatively more diagnosed primarily with an atrial septal defect type 1, an Ebstein malformation, or a left ventricular outflow tract obstruction. These patients experienced a higher care level on average.

Table 4. Covariates, care level, and outcomes of included and excluded patients with moderate lesions.

		Excluded low (N=147)	Included (N=2,727)	Excluded High (N=15)
		Mean/%	Mean/%	Mean/%
√(number of available informal caregivers)		0.39	0.41	0.63
√(number of interventions)		0.05*	0.80	1.84*
Self-employment		3%	8%	27%*
Increased reimbursement		15%	16%	47%*
Chronical ill		7%	14%	33%
Syndrome		0%*	16%	53%*
Income rank (quartiles)		1.54	1.53	1.13
Age category		2.37*	1.80	1.07*
Sex (men)		46%	51%	60%
Marital status	Unmarried	33%*	62%	100%*
	Married	53%*	32%	0%*
	Widow(er)	1%	1%	0%
	Divorced	14%*	4%	0%
Household type	Single, children	12%	10%	7%
	Single, no children	12%	10%	0%
	Partner, children	14%	18%	0%
	Partner, no children	57%	61%	67%
	Collective household	2%*	0%	13%*
	Other	3%	1%	13%*
Primary diagnostic code	TOF	1%*	16%	20%

CoA	1%*	16%	7%
AVSD	0%*	4%	0%
ASD 1	0%*	4%	27%*
Ebstein malformation	0%	2%	13%*
PV abnormality	76%*	20%	13%
AV abnormality	0%*	9%	0%
Aortic abnormality	2%*	12%	7%
LVOT obstruction	1%	1%	13%*
ASD 2	1%*	6%	0%
VSD	1%	5%	0%
MV abnormality	0%	1%	0%
Pulmonary vein abnormality	17%*	5%	0%
Other	1%	0%	0%
Care level	0.63*	0.49	0.77*
Emergency department visits (per PY)	0.20	0.14	0.25
Hospitalizations (per PY)	0.24	0.17	0.30
Hospitalization cost (per PY)	1 593 €	1 006 €	2 179 €
Pharmaceutical costs (per PY)	552 €	327 €	778 €
Outpatient costs (per PY)	1 417 €	1 078 €	1 755 €
Total medical costs (per PY)	3 562 €	2 618 €	4 506 €

**=significantly different from the included group. ASD 1: atrial septal defect type 1; ASD 2: atrial septal defect 2; AV: aortic valve; AVSD: atrioventricular septal defect; CoA: coarctation of the aorta; LVOT left ventricular outflow track; MV: mitral valve; PV: pulmonary valve; PY: patient year TOF: tetralogy of Fallot; VSD: ventricular septal defect. Care level: from 0 (general care) to 4 (highly specialized care) Income rank: 0 (lowest quartile) to 3 (highest quartile). Age category: 1=18-29 years old, 2=30-39 years old, 3=40+ years old.*

Total costs difference

	z Value	Pr > z	Difference	95% Low	95% High
HSC vs SC+	0.17	0.86	-1.9%	-21.0%	21.8%
vs SC-	4.69	<.0001	-35.1%	-45.8%	-22.2%
vs GCC	3.15	0.00	-25.3%	-37.7%	-10.4%
vs NCC	-0.34	0.73	-3.3%	-20.3%	17.2%
SC+ vs SC-	4.73	<.0001	-33.8%	-44.2%	-21.5%
vs GCC	2.99	0.00	-23.9%	-36.3%	-9.0%
vs NCC	-0.15	0.88	-1.4%	-18.9%	19.7%
SC- vs GCC	-2.07	0.04	15.1%	0.7%	31.4%
vs NCC	5.07	<.0001	49.0%	27.7%	73.7%
GCC vs NCC	3.61	0.00	29.5%	12.5%	48.9%

Hospitalization costs difference

	z Value	Pr > z	Difference	95% Low	95% High
HSC vs SC+	-0.03	0.98	0.7%	-35.6%	57.4%
vs SC-	2.83	0.00	-41.8%	-60.0%	-15.4%
vs GCC	2.24	0.03	-34.8%	-55.2%	-5.2%
vs NCC	-0.89	0.37	-16.3%	-43.4%	23.7%
SC+ vs SC-	3.12	0.00	-42.2%	-59.1%	-18.5%
vs GCC	2.34	0.02	-35.3%	-55.0%	-6.8%
vs NCC	-0.92	0.36	-16.9%	-44.0%	23.4%
SC- vs GCC	-0.81	0.42	12.0%	-15.0%	47.6%
vs NCC	2.25	0.02	43.8%	4.8%	97.2%
GCC vs NCC	1.72	0.09	28.3%	-3.5%	70.6%

Outpatient costs difference

	z Value	Pr > z	Difference	95% Low	95% High
HSC vs SC+	0.42	0.67	-3.5%	-18.3%	14.0%
vs SC-	2.1	0.04	-13.9%	-25.1%	-1.0%
vs GCC	2.2	0.03	-14.8%	-26.1%	-1.7%
vs NCC	1.1	0.27	8.8%	-6.3%	26.3%
SC+ vs SC-	1.66	0.10	-10.8%	-22.0%	2.1%
vs GCC	1.74	0.08	-11.7%	-23.2%	1.6%
vs NCC	1.56	0.12	12.7%	-3.0%	31.0%
SC- vs GCC	0.19	0.85	-1.0%	-10.8%	9.9%
vs NCC	3.88	0.00	26.3%	12.3%	42.2%
GCC vs NCC	4.32	<.0001	27.6%	14.2%	42.5%

Pharmaceuticals costs difference

	z Value	Pr > z	Difference	95% Low	95% High
HSC vs SC+	1.06	0.29	-13.1%	-33.0%	12.8%
vs SC-	5.4	<.0001	-44.6%	-55.3%	-31.4%
vs GCC	2.96	0.00	-27.5%	-41.4%	-10.3%
vs NCC	0.82	0.41	10.2%	-12.6%	39.0%
SC+ vs SC-	4.19	<.0001	-36.3%	-48.4%	-21.3%
vs GCC	1.61	0.11	-16.5%	-33.1%	4.1%
vs NCC	1.92	0.05	26.8%	-0.5%	61.6%
SC- vs GCC	-3.3	0.00	31.0%	11.6%	53.7%
vs NCC	7.22	<.0001	99.1%	65.1%	140.0%
GCC vs NCC	4.81	<.0001	52.0%	28.2%	80.3%

Emergency department visits difference

	z Value	Pr > z	Difference	95% Low	95% High
HSC vs SC+	0.62	0.54	-9.4%	-33.6%	23.7%
vs SC-	1.64	0.10	-19.4%	-37.7%	4.3%
vs GCC	1.57	0.12	-18.8%	-37.5%	5.4%
vs NCC	0.49	0.62	7.3%	-19.1%	42.4%
SC+ vs SC-	0.91	0.36	-11.1%	-30.9%	14.5%
vs GCC	0.82	0.41	-10.4%	-31.2%	16.6%
vs NCC	1.17	0.24	18.5%	-10.9%	57.5%
SC- vs GCC	-0.07	0.94	0.7%	-17.3%	22.6%
vs NCC	2.47	0.01	33.2%	6.1%	67.2%
GCC vs NCC	2.7	0.01	32.2%	7.9%	62.0%

Hospitalizations difference

	z Value	Pr > z	Difference	95% Low	95% High
HSC vs SC+	1.29	0.20	-18.6%	-40.4%	11.3%
vs SC-	3.62	0.00	-38.5%	-52.7%	-19.9%
vs GCC	3.11	0.00	-34.3%	-49.5%	-14.4%
vs NCC	-1.17	0.24	-15.3%	-35.8%	11.8%
SC+ vs SC-	2.19	0.03	-24.4%	-41.2%	-2.9%
vs GCC	1.62	0.11	-19.3%	-37.7%	4.7%
vs NCC	0.28	0.78	4.0%	-21.3%	37.6%
SC- vs GCC	-0.66	0.51	6.9%	-12.2%	30.1%
vs NCC	2.8	0.01	37.7%	10.0%	72.3%
GCC vs NCC	2.45	0.01	28.9%	5.2%	57.9%

Adult patients with severe lesions

It appeared to be feasible to delete all imbalance by adding quadratics and interactions. The final Ordinal Logit Model with 12 main effects, two quadratic effect (i.e. number of interventions, primary diagnostic code) and five interaction effects (i.e. number of interventions*age category, number of interventions*primary diagnostic code, number of interventions*chronical ill, number of interventions*increased reimbursement, age category*chronical ill) resulted in total balance in all 12 covariates. However, because the relatively small sample size, we added all covariates to the final regression model to obtain an even more robust estimation of the impact of care level on the respective outcomes.

Table 5 shows how the excluded patients (N=107, 32.8%) differed from the included patients (N=219, 67.2%). The obtained results does not apply on (i) a small group of patients who could be characterized by having had less interventions, had a lower income, older age, a different marital status, and who were relatively more diagnosed primarily with hypoplastic left heart syndrome or transposition of the great arteries, and relatively less with univentricular heart

or congenitally corrected transposition of the great arteries. These patients experienced a lower care level on average. And (ii) a small group of patients who could be characterized by having had more available informal caregivers, being more self-employed, having a syndrome, being younger, and who were relatively more diagnosed primarily with a hypoplastic left heart syndrome, a pulmonary atresia with ventricular septal defect, or a truncus arteriosus. These patients experienced a higher care level on average.

Table 5. Covariates, care level, and outcomes of included and excluded patients with moderate lesions.

		Excluded low (N=65)	Included (N=219)	Excluded High (N=42)
		Mean/%	Mean/%	Mean/%
√(number of available informal caregivers)		0.36	0.45	0.77*
√(number of interventions)		0.67*	1.49	1.64
Self-employment		11%	6%	31%*
Increased reimbursement		25%	24%	21%
Chronical ill		18%	22%	19%
Syndrome		2%	2%	12%*
Income rank (quartiles)		1.15*	1.55	1.14
Age category		2.25*	1.39	1.14*
Sex (men)		60%	58%	52%
Marital status	Unmarried	48%*	79%	86%
	Married	40%*	21%	14%
	Widow(er)	2%	0%	0%
	Divorced	11%*	0%	0%
Household type	Single, children	14%	6%	2%
	Single, no children	11%	14%	7%
	Partner, children	20%	11%	10%
	Partner, no children	54%	68%	81%
	Collective household	0%	0%	0%
	Other	2%	1%	0%
Primary diagnostic code	HLHS	8%*	0%	5%*
	Univentricular physiology	5%*	22%	19%
	Tricuspid atresia	0%	2%	5%
	TOF	11%*	3%	0%
	PA with VSD	0%	0%	19%*
	PA without VSD	2%	2%	5%
	DORV	17%	11%	0%
	DILV	0%	0%	0%
	TA	5%	2%	10%*
	TGA	11%*	49%	38%
	TGA-cc	43%*	9%	0%
Care level		1.09*	2.21	3.02*
Emergency department visits (per PY)		0.60	0.36	0.26
Hospitalizations (per PY)		0.58	0.48	0.41
Hospitalization cost (per PY)		5 147 €	4 056 €	3 199 €
Pharmaceutical costs (per PY)		1 223 €	1 777 €	514 €
Outpatient costs (per PY)		2 992 €	2 087 €	1 646 €
Total medical costs (per PY)		9 362 €	7 920 €	5 359 €

*=significantly different from the included group. DIRV: double inlet left ventricle; DORV: double outlet right ventricle; HLHS: hypoplastic left heart syndrome; PA: pulmonary atresia; PY: patient year; TA: truncus arteriosus; TGA: transposition of the great arteries; TGA-cc: congenitally corrected transposition of the great arteries. Care level: from 0 (general care) to 4 (highly specialized care) Income rank: 0 (lowest quartile) to 3 (highest quartile). Age category: 1=18-29 years old, 2=30-39 years old, 3=40+ years old.

Total costs difference

	z Value	Pr > z	Difference	95% Low	95% High
HSC vs SC+	1.44	0.15	-32.9%	-61.0%	15.4%
vs SC-	2.19	0.03	-41.6%	-63.9%	-5.6%
vs GCC	2.35	0.02	-51.8%	-73.8%	-11.4%
vs NCC	0.19	0.85	7.7%	-49.3%	129.1%
SC+ vs SC-	0.69	0.49	-12.9%	-41.1%	28.7%
vs GCC	1.26	0.21	-28.2%	-57.2%	20.4%
vs NCC	1.39	0.17	60.6%	-17.8%	214.0%
SC- vs GCC	0.8	0.42	-17.6%	-48.6%	32.2%
vs NCC	1.84	0.07	84.4%	-3.8%	253.5%
GCC vs NCC	2.14	0.03	123.8%	7.0%	367.8%

Hospitalization costs difference

	z Value	Pr > z	Difference	95% Low	95% High
HSC vs SC+	1.07	0.29	-47.6%	-84.1%	72.1%
vs SC-	1.46	0.14	-54.6%	-84.2%	30.7%
vs GCC	1.31	0.19	-57.6%	-88.3%	53.6%
vs NCC	-0.09	0.93	-8.0%	-84.1%	433.2%
SC+ vs SC-	0.3	0.77	-13.3%	-66.2%	122.1%
vs GCC	0.34	0.73	-19.1%	-75.8%	170.2%
vs NCC	0.7	0.48	75.6%	-63.4%	742.8%
SC- vs GCC	0.13	0.90	-6.7%	-67.8%	170.7%
vs NCC	0.89	0.37	102.6%	-57.3%	861.5%
GCC vs NCC	0.86	0.39	117.1%	-63.0%	1172.4%

Outpatient costs difference

	z Value	Pr > z	Difference	95% Low	95% High
HSC vs SC+	-0.15	0.88	3.4%	-32.5%	58.5%
vs SC-	0.77	0.44	-13.9%	-41.3%	26.3%
vs GCC	1.26	0.21	-27.2%	-55.7%	19.5%
vs NCC	1.47	0.14	54.4%	-13.4%	175.1%
SC+ vs SC-	1.24	0.21	-16.7%	-37.7%	11.2%
vs GCC	1.71	0.09	-29.6%	-53.0%	5.4%
vs NCC	1.54	0.12	49.3%	-10.3%	148.5%
SC- vs GCC	0.89	0.37	-15.5%	-41.6%	22.4%
vs NCC	2.39	0.02	79.3%	11.1%	189.5%
GCC vs NCC	2.66	0.01	112.1%	21.9%	269.1%

Pharmaceuticals costs difference

	z Value	Pr > z	Difference	95% Low	95% High
HSC vs SC+	0.74	0.46	-21.8%	-59.2%	49.7%
vs SC-	2.66	0.01	-55.1%	-75.1%	-19.0%
vs GCC	1.63	0.10	-46.7%	-74.9%	13.5%
vs NCC	0.65	0.51	36.6%	-46.4%	248.3%
SC+ vs SC-	2.26	0.02	-42.6%	-64.5%	-7.1%
vs GCC	1.17	0.24	-31.7%	-64.1%	29.6%
vs NCC	1.33	0.18	74.8%	-23.3%	298.4%
SC- vs GCC	-0.58	0.56	18.9%	-33.8%	113.5%
vs NCC	2.73	0.01	204.3%	36.9%	576.6%
GCC vs NCC	2.05	0.04	156.0%	4.3%	528.3%

Emergency department visits difference

Emergency department visits difference - Severe

	z Value	Pr > z	Difference	95% Low	95% High
HSC vs SC+	-0.01	0.99	0.5%	-57.9%	139.8%
vs SC-	-0.27	0.79	10.5%	-47.0%	130.5%
vs GCC	-0.95	0.34	60.8%	-39.6%	328.4%
vs NCC	0.68	0.50	51.0%	-54.1%	396.9%
SC+ vs SC-	-0.3	0.77	9.9%	-41.0%	104.8%
vs GCC	-1.04	0.30	60.0%	-34.1%	288.7%
vs NCC	0.73	0.46	50.3%	-49.4%	346.0%
SC- vs GCC	-0.95	0.34	45.5%	-33.1%	216.8%
vs NCC	0.58	0.56	36.7%	-52.7%	295.4%
GCC vs NCC	-0.1	0.92	-6.1%	-71.8%	213.1%

Hospitalizations difference

	z Value	Pr > z	Difference	95% Low	95% High
HSC vs SC+	0.59	0.56	-23.1%	-67.9%	84.6%
vs SC-	1.28	0.20	-39.8%	-72.3%	30.6%
vs GCC	0.45	0.65	-20.4%	-70.3%	113.4%
vs NCC	0.37	0.71	26.9%	-64.0%	347.2%
SC+ vs SC-	0.74	0.46	-21.8%	-59.2%	50.2%
vs GCC	-0.08	0.94	3.5%	-57.2%	150.1%
vs NCC	0.9	0.37	64.9%	-44.7%	391.3%
SC- vs GCC	-0.7	0.48	32.2%	-39.2%	187.5%
vs NCC	1.32	0.19	110.7%	-30.1%	535.0%
GCC vs NCC	0.72	0.47	59.3%	-54.8%	462.3%

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Supplement S2. Sensitivity analysis: exclusion of patients who died during the study period

of the paper

**Different Levels of Care for Follow-up of Adults with Congenital Heart
Disease; Impact on Medical Costs, Hospitalizations, and Emergency
Department Visits**

Running title: Different Care Levels for ACHD Follow-up

Adult patients with mild lesions

		Excluded low (N=59)	Included (N=3559)	Excluded High (N=25)
		Mean/%	Mean/%	Mean/%
√(number of available informal caregivers)		0.28	0.40	0.43
√(number of interventions)		0.15*	0.35	1.54*
Self-employment		8%	9%	8%
Increased reimbursement		8%	9%	24%*
Chronical sick		3%	7%	28%*
Syndrome		0%	3%	16%*
Income rank (quartiles)		0.63*	1.59	1.40
Age category		1.85	2.08	1.60*
Gender (male)		44%	43%	60%
Marital status	Unmarried	64%*	45%	76%*
	Married	10%*	47%	20%*
	Widow(er)	8%*	2%	0%
	Divorced	17%*	6%	4%
Household type	Single, children	22%*	8%	16%
	Single, no children	37%*	8%	0%
	Partner, children	2%*	21%	16%
	Partner, no children	25%*	62%	56%
	Collective household	2%*	0%	8%*
	Other	12%*	1%	4%
Primary diagnostic code	AV abnormality	0%*	11%	88%*
	ASD 2	15%	26%	4%*
	VSD	83%*	29%	0%*
	MV abnormality	0%	8%	4%
	Other	2%*	26%	4%*
Care level		0.44*	0.91	2.08*
Emergency department visits (per PY)		0.30	0.21	0.14
Hospitalizations (per PY)		0.22	0.26	0.28
Hospitalization cost (per PY)		1 087 €	1 697 €	1 631 €
Pharmaceutical costs (per PY)		353 €	715 €	978 €
Outpatient costs (per PY)		1 374 €*	1 593 €	1 597 €
Total medical costs (per PY)		2 814 €*	4 006 €	4 207 €

ASD 1: atrial septal defect type 1; ASD 2: atrial septal defect 2; AV: aortic valve; AVSD: atrioventricular septal defect; CoA: coarctation of the aorta; LVOT left ventricular outflow track; MV: mitral valve; PV: pulmonary valve; TOF: tetralogy of Fallot; VSD: ventricular septal defect

Total costs difference

	z Value	Pr > z	Difference	95% Low	95% High
HSC vs SC+	1.61	0.11	-19.6%	-38.3%	4.9%
vs SC-	1.62	0.10	-15.1%	-30.3%	3.4%
vs GCC	2.98	0.00	-23.9%	-36.5%	-8.9%
vs NCC	-0.93	0.35	-8.0%	-22.9%	9.7%
SC+ vs SC-	-0.46	0.64	5.6%	-16.1%	32.8%
vs GCC	0.51	0.61	-5.4%	-23.7%	17.2%
vs NCC	1.22	0.22	14.4%	-7.7%	41.8%
SC- vs GCC	1.81	0.07	-10.4%	-20.5%	0.9%
vs NCC	1.31	0.19	8.3%	-3.9%	22.2%
GCC vs NCC	4.35	<.0001	20.9%	11.0%	31.7%