



Impact of anatomical subtype and medical comorbidities on hospitalizations in adults with single ventricle congenital heart disease



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ARTICLE INFO

Article history:

Received 19 April 2013

Received in revised form 14 July 2013

Accepted 19 July 2013

Available online 25 July 2013

Keywords:

Adult congenital heart disease

Comorbid medical conditions

Hypoplastic left heart syndrome

Single ventricle

Protein-losing enteropathy

ABSTRACT

Background: Most patients with single ventricle congenital heart disease (SV) are now expected to survive to adulthood. Medical comorbidities are common in SV.

Methods: We used data from 43 pediatric hospitals in the 2004 to 2011 Pediatric Health Information System database to identify patients ≥ 18 years of age admitted with International Classification of Diseases-9th Revision codes for a diagnosis of either hypoplastic left heart syndrome (HLHS), tricuspid atresia (TA) or common ventricle (CV). Primary (PD) and secondary diagnoses (SD), length of stay (LOS) and hospital charges were determined. Multilevel models were used to evaluate differences in demographics, diagnoses, and admission outcomes among the three subgroups (HLHS, TA, and CV). Interactions of charges with PD and admission year were examined using ANOVA.

Results: There were 801 SV patients with 1330 admissions during the study period. Mean age was 24.8 ± 6.2 years (55% male) and mean LOS was 6.8 ± 11.3 days. Total hospital charges were \$135 million with mean charge per admission of $\$101,131 \pm 205,808$. The mean charge per day was $\$15,407 \pm 16,437$. Hospital charges correlated with PD group ($p < 0.001$). Admission rate remained stable ($\sim 180/\text{year}$) from 2006 to 2011. LOS decreased ($p = 0.0308$) and hospital charges per day increased across the study period ($p < 0.001$). PD was non-cardiac in 28% of admissions. Liver-related conditions were more common in patients with HLHS ($p < 0.001$).

Conclusions: Hospitalization costs in adults with SV are significant and are impacted by comorbid medical conditions. Hospitalization rates for adults with SV are not increasing. Gastroenterologic comorbidities including protein-losing enteropathy (PLE) are common in HLHS.

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1. Introduction

Congenital heart disease (CHD) is the most common birth defect [1]. Moderate or severe forms necessitating ongoing care occur in 6/1000 live-births [1]. Advancements in care have led to improved patient survival in even the most severe forms of CHD [2], and survival to adulthood can be expected in 90% of patients with CHD [3].

Comorbid medical conditions such as hypertension, obesity, chronic renal disease, hyperlipidemia, atrial fibrillation, and heart failure are more common in adults with CHD (ACHD) than the general population [4]. The prevalence of comorbid conditions is associated with increasing age in ACHD, and is associated with increased healthcare utilization [4].

Congenital heart defects palliated with a single ventricle (SV) strategy account for approximately 8% of all CHD births [1]. The era of improved survival of patients with SV CHD was born when Fontan and

Baudet introduced a successful surgical strategy for palliation of tricuspid atresia (TA) [5]. Subsequent work by Norwood et al. with hypoplastic left heart syndrome (HLHS) [6], has resulted in a change of survival from nearly zero to a degree that 70% are now expected to survive to adulthood [7]. Patients with SV who have undergone three-stage palliation including the Fontan operation have significant medical comorbidities [7], an increasingly limited functional status [8], and may develop protein-losing enteropathy (PLE), and a poor prognosis for survival [9].

To our knowledge, no studies have sought to look at the impact of adults with SV on the healthcare system. The aims of the study were to characterize trends in pediatric hospital utilization by adults with SV; to determine if the anatomic subtypes of SV affect the distribution of comorbid medical conditions; and to determine the impact of the comorbid conditions on healthcare costs.

2. Methods

With the approval of the Institutional Review Board of the University of Arkansas for Medical Sciences, data were obtained from the Pediatric Health Information System (PHIS), a large inpatient administrative database of 43 participating children's hospitals in the Child Health Corporation of America (CHCA), a children's hospital consortium. The PHIS data include detailed, de-identified information on each inpatient's demographics, diagnoses,

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procedures, medications, and outcomes. Data quality assurance is ongoing and data from individual hospitals are accepted when classified errors for a given quarter occur less frequently than a criterion threshold of 2%. The study design was a multi-centered, retrospective cohort investigation of all adults cared for in the children's hospitals within the CHCA network with HLHS, TA, or CV. Data were limited to the time period between January 1, 2004 to December 31, 2011 in the PHIS database for patients ≥ 18 years of age admitted to the hospital with an International Classification of Diseases–9th revision code, whether principal or secondary, for either HLHS (ICD-9 746.7), TA (746.1), or CV (745.3). To provide data relevant to adults with SV physiology, inclusion criteria were limited to birth from January 1, 1965 to January 1, 1993. Collected data included demographic data, principal and secondary diagnosis codes, admission to the intensive care unit, mechanical ventilation, procedures performed, duration of hospital length of stay (LOS), survival to hospital discharge, total hospital charges, and payor status. Principal diagnoses were grouped into categories based on organ systems. All charge data were adjusted for inflation to 2011 dollars using Consumer Pricing Indexes data.

2.1. Statistical methods

Summary statistics were expressed as mean \pm standard deviation for continuous variables, and frequency and percentage for categorical variables. Because of the nested clustering (admission–patient–hospital), multilevel models were used to evaluate the difference in the demographics, diagnoses, and outcomes of admissions among the three anatomic subgroups (HLHS, TA, and CV). The means of continuous variables were compared using the mixed model by taking into account the correlated admissions from the same patient, and the correlated patients from the same hospital unit. The proportions of binary variables were compared using the generalized estimating equations (GEEs) after taking into account the nested clustering. In the case when there existed zero cells for some diagnosis groups, the comparisons of three groups were performed using the logistic regression model with the Firth penalized likelihood approach to obtain the bias-corrected standard error estimates. The length of stay (LOS) was considered to be time-to-event outcome, while discharge from the hospital was treated as the event, and the patients who died during the hospital stay were treated as censored. The frailty models were fitted for the LOS after taking into account the admissions from the same hospital unit. Similar analyses were carried out to assess the difference across years (2004–2011). A mixed model was fitted for the hospital charges as a function of anatomic subtype (HLHS, TA or CV), hypertension, obesity, chronic renal failure, hyperlipidemia, atrial fibrillation, and heart failure to evaluate the effects of selected diagnoses on hospital charges simultaneously. p -Values < 0.05 were considered to be statistically significant.

3. Results

During the study period, there were a total of 1333 hospital admissions in 803 patients from 43 participating hospitals. Three admissions involving two patients were excluded due to overlapping diagnoses of TA and HLHS. Multiple admissions occurred in 256 patients with a median number of admissions of 3.0 (range 2–15). Demographic and clinical outcome data for the three groups are shown in Table 1. The total hospital charges for the study period were \$135 million with mean charge per admission of \$101,131 \pm 205,808. The mean charge per hospital day was \$15,407 \pm 16,437. The number of admissions per year did not change from 2006 to 2011, and are shown in Fig. 1A. As shown in Fig. 1B, the mean age at admission increased across the study period ($p < 0.001$).

The mean LOS, shown in Fig. 2, decreased from 2004 to 2008 ($p = 0.0308$), and remained stable from 2008 to 2011 ($p = 0.9138$). The mean charges per hospital day, also shown in Fig. 2, increased across the study period ($p < 0.001$), though the total hospital charges per

hospitalization did not change significantly ($p = 0.62$). As shown in Table 2, mean charges per hospital day were significantly different among principal diagnosis categories ($p < 0.001$). Among principal and secondary diagnosis categories, gastroenterologic diagnoses were more common in HLHS admissions than in either TA or CV ($p < 0.001$), otherwise there were no differences among the three groups. Differences among individual diagnoses between the study groups occurred and are demonstrated in Table 3. As shown in Table 4, heart failure, hypertension, atrial fibrillation, and chronic kidney disease increased hospital charges significantly.

Surgical procedures were performed in 457 (34%) admissions. Table 5 demonstrates the most common surgical procedures performed in the study cohort. Interestingly, extracorporeal membranous oxygenation not associated with surgery was performed in only 11 (2%) admissions. Subanalysis was conducted on hospitalizations during which Fontan, cavopulmonary anastomosis (Glenn), and/or heart transplantation procedures were performed, and demonstrated significant differences in LOS and hospital charges depending on the anatomic diagnosis group and the surgical intervention performed (Table 6). A Glenn and/or Fontan procedure was performed in 12.9% of admissions. Further analysis of this subgroup, shown in Table 7, demonstrated significant increases in hospital LOS and charges for patients who underwent Fontan, Glenn, or heart transplantation.

4. Discussion

The present study provides a number of important insights into the hospital care and medical conditions of adults with SV. Comorbid medical conditions are common in adults with SV, and are impacted by SV anatomic subtype. Hospitalizations of adults with SV have significant impacts on the healthcare system, and are significantly affected by comorbid medical conditions. The trends in hospital utilization of adults with SV are changing significantly with increasing costs per patient-day in the face of decreasing LOS.

In 2010, the average cost per hospitalization for adults with non-hypertensive, congestive heart failure was \$10,200, while the average cost for hospitalization for acute myocardial infarction was \$18,200 [10]. In the same year, the average cost per hospitalization was \$87,900 for adults with SV in the present study. Cardiovascular procedures are common during hospitalizations in adults with SV. These and other procedures contributed to the significant increase in hospital costs in these patients. A significant number of patients in the present study underwent Glenn and/or Fontan operations. The majority of the patients who underwent those surgeries had TA, which contributed to the increased rates of ICU admission, mechanical ventilation and hospital charges in those patients. The database used for the study does not allow for the determination of what proportion of those surgeries were for revision from an atriopulmonary Fontan to a lateral tunnel or extracardiac Fontan. Given that the majority of patients who underwent a Fontan operation carried the diagnosis of TA, it is likely that some of

Table 1
Demographic and hospital clinical outcomes comparisons of adults with single ventricle congenital heart disease.

Variable	HLHS	TA	CV	Composite	p-Value
Admissions	198	520	612	1330	
Patients	122	348	363	801	
Demographics					
Age (years)	21.9 \pm 4.2	25.9 \pm 6.7	24.7 \pm 6.1	24.8 \pm 6.2	<0.001
Male gender	126 (64%)	291 (56%)	312 (56%)	729 (55%)	0.16
Clinical outcomes					
Length of stay (days)	7.3 \pm 12.2	7.3 \pm 8.9	6.3 \pm 12.6	6.8 \pm 11.2	0.01
Hospital charges	\$110,228 \pm 313,589	\$121,419 \pm 204,289	\$80,915 \pm 154,973	\$101,131 \pm 205,808	0.02
Hospital charges per day	\$14,113 \pm 11,321	\$16,812 \pm 18,654	\$14,636 \pm 15,759	\$15,407 \pm 16,437	0.14
ICU admission	78 (39%)	279 (54%)	286 (47%)	643 (48%)	0.001
Mechanical ventilation	36 (18%)	158 (30%)	97 (16%)	291 (22%)	<0.001
In-hospital death	4 (7%)	11 (4%)	6 (2%)	21 (3%)	

HLHS indicates hypoplastic left heart syndrome; TA, tricuspid atresia; CV, common ventricle without further anatomic delineation; composite, combination of all study groups.

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