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ORIGINAL ARTICLE

Regional Differences in Cost and Length of Stay in Neonates with Hypoplastic Left Heart Syndrome

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Abstract

Hypoplastic left heart syndrome (HLHS) is a highly resource-intensive diagnosis. Geographic variation in cost and length of stay (LOS) in HLHS is not well described. Neonates diagnosed with HLHS between 2000 and 2012 were identified using the Kids' Inpatient Database. Hospitalizations were stratified into two groups: (1) birth and (2) secondary. United States regional differences in hospital charges and LOS were compared using adjusted linear regression. Of 2431 birth hospitalizations, 449 neonates (18.5%) died while inpatient and mortality rates differed by region ($p=0.02$). After birth, 40.5% ($n=985$) of neonates were transferred; transfers were most common in the Midwest ($p<0.0001$). Adjusted average LOS was shortest in the West and longest in the South (26.1 days; 95% CI 24.0, 35.1 vs. 34.9 days; 95% CI 31.8, 38.1). Average adjusted charges were lowest in the Northeast ($324,600; 95% CI $271,400, $377,900) and highest in the West ($400,500; 95% CI $346,700, $454,300, $p=0.05$). Among 1895 secondary hospitalizations, 24.9% of neonates died as inpatients, and the average adjusted LOS was shortest in the West (26.8 days; 95% CI 23.9, 29.7) and longest in the South (38.5 days; 95% CI 34.4, 42.4). Average adjusted charges were lowest in the Northeast ($326,900; 95% CI $270,700, $383,100) and highest in the South ($505,900; 95% CI $450,200, $561,500, $p<0.0001$). Significant geographic variations in mortality, LOS, and hospital charges exist in care of US HLHS neonates. Reducing variation in care should remain a priority in national quality efforts in congenital heart disease.

Keywords Hypoplastic left heart syndrome · Congenital heart disease · Outcomes · Cost analysis

Abbreviations

CHD	Congenital heart defects
HCUP	Healthcare Cost and Utilization Project
HLHS	Hypoplastic left heart syndrome
ICD-9-CM	International Classification of Disease, 9th revision, Clinical Modification
KID	Kids' Inpatient Database
LOS	Length of stay
US	United States

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Introduction

Hypoplastic left heart syndrome (HLHS) is a rare but serious heart defect accounting for 2–3% of all congenital heart defects (CHD), with a prevalence of 2–3 cases per 10,000 live births in the United States [1–3]. Despite tremendous efforts in surgery and critical care, HLHS remains one of the most high-risk congenital heart diseases, with no curative surgical option. Advancements in the surgical and medical management of HLHS have been transformative. Once uniformly fatal, HLHS is now associated with a 5-year survival rate of approximately 70% for infants undergoing treatment in the modern era [3, 4]. However, long-term morbidity and mortality remains uncertain despite multi-stage palliation [5–7].

Improved neonatal survival has led to higher utilization of hospital resources. For the past three decades, staged palliative repair, involving three sequential cardiac operations, has become the mainstay treatment option. As such, primary cardiac transplantation for neonates with HLHS has become less common, and transplantation for end-stage heart failure

in older patients who have undergone palliation has become more common. With improvements in surgical outcomes, more patients are undergoing palliation, resulting in a net increase in total costs and longer hospital stays for treatment of HLHS [8]. Recent investigations suggest that investment of resources in inpatient care of HLHS is high compared to other congenital heart defects, and disproportionately so among those who do not survive their hospitalization [9].

A better understanding of cost and length of stay (LOS) among neonates diagnosed with HLHS will provide greater information on the overall health care burden of HLHS and facilitate better regional and national programmatic planning and resource allocation. Significant differences in these outcomes between regions would highlight the need for focused multi-center, collaborative quality improvement projects and would inform policy decisions related to regionalization of congenital heart services. Thus, the purpose of this study was to describe United States (US) regional differences in hospital charges and LOS for neonates with HLHS.

Materials and Methods

Study Design and Population

The study was conducted using the Healthcare Cost and Utilization Project (HCUP) Kids' Inpatient Database (KID) and was deemed exempt from further review by the University of North Carolina Institutional Review Board [10]. The KID is the largest publically available all-payer health pediatric inpatient care database in the United States, including over 3 million pediatric discharges each year. The KID is currently available every 3 years from 1997 to 2012.

Eligible infants were identified using International Classification of Disease, 9th revision, Clinical Modification (ICD-9-CM) diagnostic codes. Neonates with HLHS (ICD-9-CM 746.7) were eligible for inclusion. Records with either age at admission > 28 days old (i.e., outside of the neonatal period) or missing age (in days) and without a live birth diagnosis code (ICD-9-CM 765.20, 765.29, and V30.0–V39.2) were excluded. Neonate records were then stratified into two groups: (1) birth hospitalizations (identified through live birth diagnosis codes), and (2) secondary hospitalizations (hospitalizations occurring after birth hospitalization discharge but before the infant is > 28 days old). Birth hospitalizations were further classified as either primary treatment hospitalizations or transfers. Within a birth hospitalization, neonates who were (1) hospitalized for ≤ 2 days, (2) discharged alive, and (3) classified as transfers on discharge disposition were assumed to have been transferred for cardiac critical care and/or intervention such as surgical palliation. Secondary hospitalizations could include those transfers or readmissions within 28 days after birth hospitalization discharge.

Statistical Analyses

Birth hospitalizations and secondary hospitalization records were analyzed separately. Neonate demographics, comorbidities, and hospital characteristics were compared across geographical regions using Fisher's exact test. A p value < 0.05 was considered statistically significant. Regions were categorized using US Census Bureau definitions (Northeast, Midwest, South, and West) [11]. Comorbidities were classified into other cardiac anomalies (745–747.4), congenital structural anomalies (740–744.9 and 747.5–757.9), chromosomal anomalies or syndromes (758–758.9), other congenital anomalies (759–759.9), and other complications (427–427.9, 432–432.9, 516.64, 770.4, and 901.4–901.42).

Differences in average LOS and hospital charges (in thousands of U.S. dollars) between regions were analyzed using linear regression. Multivariable linear regression, adjusting for admit year, gender, race/ethnicity, insurance status, household income, comorbidities, operations, and procedures performed during the hospitalization, inpatient mortality, hospital size, and teaching status, was also performed. Operations and procedures were classified by body system: nervous system (01–05.9), endocrine (06–07.99), eye (08–016.99), nose/mouth/pharynx (21–29.99), respiratory (30–34.99), cardiovascular (35–39.99), hemic and lymphatic (40–41.99), digestive (42–54.99), urinary (55–59.99), male genital (60–64.99), female genital (65–71.9), musculoskeletal (76–84.99), integumentary (85–86.99), other interventions (00–00.96), and other diagnostic and therapeutic procedures (87–99.99).

All analyses were performed using SAS 9.4 (SAS Inc., Cary, NC).

Results

Birth Hospitalizations—Study Population

Overall, 2431 neonatal birth hospitalizations were included in this study. Table 1 describes neonate demographics, insurance and socioeconomic status, comorbidities, and hospital characteristics, stratified by region. Significant demographic differences were found between regions. Neonates born in the South were more likely to be from low-income households ($p < 0.0001$), be non-Hispanic Blacks ($p < 0.0001$), and have public insurance ($p < 0.0001$), compared to the other regions. In contrast, the Northeast had the highest proportion of neonates born with private insurance and the largest proportion of neonates from the highest income households (Table 1). Neonates born in the South had proportionally

Table 1 Demographics and hospital characteristics of neonates with HLHS during birth hospitalizations, stratified by region, $n = 2431$

	Northeast 434 (17.9%)	Midwest 534 (22.0%)	South 881 (36.2%)	West 582 (23.9%)	*p* value
Gender, *n* (%)					
Male	261 (60.1)	317 (59.4)	515 (58.5)	344 (59.4)	0.95
Female	173 (39.9)	217 (40.6)	366 (41.5)	235 (40.6)	–
Race/ethnicity, *n* (%)					
Non-Hispanic White	249 (62.4)	241 (72.6)	363 (48.1)	197 (42.6)	**< 0.0001**
Non-Hispanic Black	45 (11.3)	45 (13.6)	170 (22.6)	35 (7.6)	**< 0.0001**
Hispanic	50 (12.5)	23 (6.9)	172 (22.8)	178 (38.4)	**< 0.0001**
Other	55 (13.8)	23 (6.9)	49 (6.5)	53 (11.5)	**0.0001**
Primary insurance, *n* (%)					
Public	163 (37.6)	227 (42.5)	461 (52.9)	242 (41.6)	**< 0.0001**
Private	241 (55.5)	261 (48.9)	351 (40.3)	286 (49.1)	**< 0.0001**
Other/self-pay	30 (6.9)	46 (8.6)	60 (6.9)	54 (9.3)	0.29
Household income[a], *n* (%)					
Low	87 (20.4)	126 (23.8)	282 (32.5)	112 (19.6)	**< 0.0001**
Medium	109 (25.6)	143 (27.0)	237 (27.3)	151 (26.4)	0.93
High	100 (23.5)	173 (32.6)	191 (22.0)	178 (31.2)	**< 0.0001**
Highest	130 (30.5)	88 (16.6)	157 (18.1)	130 (22.8)	**< 0.0001**
Comorbidities					
Other cardiac anomalies	262 (60.4)	270 (50.6)	524 (59.5)	303 (52.1)	**0.0005**
Structural anomalies	71 (16.4)	102 (19.1)	198 (22.5)	128 (22.0)	**0.04**
Chromosomal anomalies	26 (6.0)	26 (4.9)	81 (9.2)	45 (7.7)	**0.01**
Other anomalies	15 (3.5)	20 (3.8)	50 (5.7)	24 (4.1)	0.21
Other complications[b]	29 (6.7)	42 (7.9)	78 (8.9)	41 (7.0)	0.48
Hospital size[c], *n* (%)					
Small	13 (3.0)	11 (2.2)	12 (1.4)	12 (2.1)	0.27
Medium	64 (14.8)	58 (11.5)	159 (18.7)	83 (14.3)	**0.004**
Large	357 (82.3)	437 (86.4)	681 (79.9)	484 (83.6)	**0.02**
Hospital type, *n* (%)					
Non-teaching	27 (6.2)	53 (10.5)	201 (23.6)	133 (23.0)	**< 0.0001**
Teaching	407 (93.8)	453 (89.5)	651 (76.4)	446 (77.0)	–

Statistically significant values ($p < 0.05$) are given in bold

[a]In 2000, household income was characterized by the following quartiles: \$1–\$24,999 (low), \$25,000–\$34,999 (medium), \$35,000–\$44,999 (high), and \$45,000 and above (highest); from 2003 onward, income was characterized into quartiles within each ZIP code

[b]Other complications includes cardiac dysrhythmias, intracranial hemorrhage, alveolar capillary dysplasia, primary atelectasis, and pulmonary vessel injury

[c]Hospital size categories are based on the number of hospital beds; cut points were chosen for each region and location (rural, non-teaching, urban non-teaching, and urban teaching) combination so that approximately 1/3 of hospitals would appear in each size category

more chromosomal anomalies ($p = 0.01$), and neonates born in the Northeast had proportionally fewer structural anomalies ($p = 0.04$). Of the babies that were born in their primary treatment hospital (not transferred), 18.5% ($n = 449$) neonates died during hospitalization. Significant differences in mortality were seen across regions, as 21.6% ($n = 190$) of neonates born in the South died during the birth hospitalization, compared to 15.2% ($n = 66$) in the Northeast, 16.5% ($n = 88$) in the Midwest, and 18.0% ($n = 105$) in the West ($p = 0.02$). Additionally, 40.5% of neonates ($n = 985$) were transferred after being born. Neonates in the Midwest were significantly more likely to be transferred, compared to the West, Northeast, and South (48.7, 43.6, 36.9, 35.3%, respectively), $p < 0.0001$.

Birth Hospitalizations—Length of Stay and Hospital Charges

Among infants that were not transferred ($n = 1446$), the median LOS was 18 days (interquartile range [IQR] 4–40 days). After adjustment, compared to the Northeast's average LOS of 30.9 days (95% confidence interval [CI] 26.7, 35.1), the Midwest's average LOS was 4.3 days shorter (95% CI − 10.8, 2.3, $p = 0.20$), the average LOS in the South was

4.1 days longer (95% CI − 1.3, 9.4, $p = 0.14$), and the average LOS in the West was 2.7 days shorter (95% CI − 8.8, 3.3, $p = 0.37$) (Table 2). The Northeast charged on average, $324,600 (95% CI $271,400, 377,900) for a birth hospitalization for a neonate with HLHS. Compared to the Northeast, the Midwest charged, on average, $28,111 more (95% CI − $54,400, 110,700, $p = 0.50$), the South charged, on average, $37,500 more (95% CI − $30,000, 105,000, $p = 0.28$), and the West charged, on average, $75,900 more (95% CI − $1,500, 153,200, $p = 0.05$), after adjusting for neonate and hospital characteristics (Table 2).

Secondary hospitalizations—Study Population

In total, 1895 neonates were re-hospitalized or admitted by transfer shortly after birth. Table 3 describes this specific population, including neonate demographics, insurance and socioeconomic status, comorbidities, and hospital characteristics, stratified by region. Neonates in the South were more likely to be Non-Hispanic Black ($p < 0.0001$), and neonates in the West were more likely to be Hispanic ($p < 0.0001$), compared to other regions. Proportionally more neonates in the South had public insurance ($p < 0.0001$). Significant differences were also seen in income ($p < 0.0001$). Infants in the Northeast were most likely to come from the highest income quartile (31.6%), compared to the West (23.9%), Midwest (16.2%), and South (10.3%). The South was most likely to have infants from low-income households (40.1%), compared to the Northeast, Midwest, and West (21.5, 24.5, and 19.9%, respectively). Neonates in the Northeast were the least likely to present with other cardiac anomalies during secondary hospitalizations ($p < 0.0001$). Of the neonates that were transferred in or readmitted within 28 days of being born, 24.9% ($n = 472$) died during this secondary hospitalization. Significant differences in mortality were seen across regions ($p = 0.02$); in the Northeast 18.9% ($n = 57$) of neonates died, compared to 28.4% ($n = 183$) in the West, 34.5% ($n = 139$) in the Midwest, and 24.4% ($n = 93$) in the South. More than 95% of secondary hospitalizations in each region took place in teaching hospitals.

Table 2 Crude and adjusted average length of stay (LOS) and hospital charges (in thousands of dollars) among neonates not transferred for surgery during birth hospitalizations, stratified by region, $n = 1,446$

	Average LOS (95% CI)	Average hospital charges[a] (95% CI)
Crude		
Northeast	24.6 (20.1, 29.1)	243.1 (186.9, 299.2)
Midwest	30.5 (26.0, 35.0)	365.0 (308.9, 421.1)
South	33.7 (30.6, 36.8)	340.1 (300.7, 379.5)
West	25.6 (21.5, 29.7)	366.5 (313.2, 419.8)
Adjusted[b]		
Northeast	30.9 (26.7, 35.1)	324.6 (271.4, 377.9)
Midwest	26.6 (21.6, 31.6)	352.8 (290.5, 415.0)
South	34.9 (31.8, 38.1)	362.1 (322.2, 402.0)
West	26.1 (24.0, 35.1)	400.5 (346.7, 454.3)

[a]In thousands of dollars

[b]Adjusted for admit year, gender, race/ethnicity, insurance status, household income, comorbidities, operations and procedures, inpatient mortality, hospital size, and hospital teaching status

Secondary Hospitalizations—Length of Stay and Hospital Charges

The median LOS was 10 days (IQR 4–29), range 0–315 days. After adjustment, compared to the Northeast's average LOS of 31.9 days (95% CI 27.9, 36.0), LOS in the Midwest was 6.5 days longer (95% CI 0.8, 12.3, $p = 0.03$), LOS in the South was 6.5 days longer (95% CI 0.8, 12.2, $p = 0.02$), and LOS in the West was 5.1 days shorter (95% CI − 10.3, 0.0, $p = 0.05$) (Table 4). Additionally, after adjustment, the Northeast was found to have the lowest average hospital charges. The Northeast charged, on average, $326,900 (95% CI $270,700, 383,100) per secondary hospitalization (Table 4). Compared to the Northeast, the Midwest charged, on average, $100,114 more (95% CI 20,000, 180,200, $p = 0.01$), the South charged, on average, $179,000 more (95% CI $100,400, $257,5000, $p < 0.0001$), and the West charged, on average, $12,300 more (95% CI − $60,000, $84,500, $p = 0.74$).

Discussion

The current study demonstrates that LOS and cost related to the care of neonates with HLHS varies widely across regions. Among birth hospitalizations, neonates in the West and Midwest experienced the shortest inpatient stays, and neonates from the South experienced the longest hospital stays, after adjustment. Interestingly, hospital charges were lowest in the Midwest and highest in the West. Among secondary hospitalizations, hospital stays were shortest in the West and longest in the South, after adjustment, and hospital charges were lowest in the Northeast and highest in the South.

There is a knowledge gap related to cost of care and length of stay for neonates diagnosed with HLHS, particularly for neonates who do not undergo surgical palliation. This study is among the first to compare length of stay and cost among all inpatient neonates diagnosed with HLHS in the United States. While previous studies have described the prevalence as well as expected costs and lengths of stay associated with the surgical treatment of HLHS, our analysis includes neonates not treated with surgery, and

Table 3 Demographics and hospital characteristics among neonates with HLHS during secondary hospitalizations, stratified by region, $n = 1,895$

	Northeast 301 (15.9%)	Midwest 568 (30.0%)	South 382 (20.2%)	West 644 (34.0%)	*p* value
Gender, *n* (%)					
Male	200 (66.5)	337 (59.3)	219 (57.3)	373 (57.9)	0.06
Female	101 (33.6)	231 (40.7)	163 (42.7)	271 (42.1)	–
Race/ethnicity, *n* (%)					
Non-Hispanic White	150 (55.6)	245 (73.1)	156 (57.6)	201 (36.6)	**< 0.0001**
Non-Hispanic Black	26 (9.6)	46 (13.7)	51 (18.8)	24 (4.4)	**< 0.0001**
Hispanic	28 (10.4)	17 (5.1)	39 (14.4)	246 (44.8)	**< 0.0001**
Other	66 (24.4)	27 (8.1)	25 (9.2)	78 (14.2)	**< 0.0001**
Primary insurance, *n* (%)					
Public	117 (39.4)	263 (46.4)	224 (58.8)	306 (47.5)	**< 0.0001**
Private	156 (52.5)	266 (46.9)	131 (34.4)	275 (42.7)	**< 0.0001**
Other/self-pay	24 (8.1)	38 (6.7)	26 (6.8)	63 (9.8)	0.20
Household income[a], *n* (%)					
Low	62 (21.5)	136 (24.5)	151 (40.1)	126 (19.9)	**< 0.0001**
Medium	79 (27.4)	181 (32.6)	119 (31.6)	166 (26.2)	0.07
High	56 (19.4)	149 (26.8)	68 (18.0)	190 (30.0)	**< 0.0001**
Highest	91 (31.6)	90 (16.2)	39 (10.3)	151 (23.9)	**< 0.0001**
Comorbidities					
Other cardiac anomalies	194 (64.5)	446 (78.5)	293 (76.7)	478 (74.2)	**0.0001**
Structural anomalies	56 (18.6)	133 (23.4)	94 (24.6)	141 (21.9)	0.26
Chromosomal anomalies	< 11	24 (4.2)	19 (5.0)	27 (4.2)	0.65
Other anomalies	14 (4.7)	22 (3.9)	17 (4.5)	25 (3.9)	0.90
Other complications[b]	46 (15.3)	121 (21.3)	59 (15.5)	132 (20.5)	**0.03**
Hospital size[c], *n* (%)					
Small	15 (5.0)	186 (36.3)	22 (5.9)	44 (6.9)	**< 0.0001**
Medium	114 (37.9)	88 (17.2)	97 (26.2)	357 (55.6)	**< 0.0001**
Large	172 (57.1)	239 (46.6)	252 (67.9)	241 (37.5)	**< 0.0001**
Hospital type, *n* (%)					
Non-teaching	13 (4.3)	< 11	17 (4.6)	27 (4.2)	**0.04**
Teaching	288 (95.7)	504 (98.33)	354 (95.4)	615 (95.8)	–

Statistically significant values ($p < 0.05$) are given in bold

[a]In 2000, household income was characterized by the following quartiles: $1–$24,999 (low), $25,000–$34,999 (medium), $35,000–$44,999 (high), and $45,000 and above (highest); from 2003 onward, income was characterized into quartiles within each ZIP code

[b]Other complications include cardiac dysrhythmias, intracranial hemorrhage, alveolar capillary dysplasia, primary atelectasis, and pulmonary vessel injury

[c]Hospital size categories are based on the number of hospital beds; cut points were chosen for each region and location (rural, non-teaching, urban non-teaching, and urban teaching) combination so that approximately 1/3 of hospitals would appear in each size category

contributes the dimension of comparison across the Northeast, South, Midwest, and West.

Several groups have demonstrated geographic variation in surgical intervention for CHD, including HLHS within the first year of life [12–15]. While Husain et al. reported differences in the rate of surgical interventions across regions, limitations include the inability to accurately describe the background prevalence of CHDs. Other groups have demonstrated significant practice-pattern variation across North American congenital heart centers caring for infants with HLHS [6, 16]. The Single Ventricle Reconstruction (SVR) trial highlighted how standardization of care might improve outcomes for infants with HLHS [6].

Previous studies have suggested that LOS and postoperative complications are important drivers of high cost [17–19]. Postoperative complications are especially common following stage one palliation with the Norwood operation [6]. Eighty-two percent of patients experienced at least one complication captured in the SVR trial, and more than twenty-five percent experienced four or more complications [19]. These complications were associated with worse clinical outcomes, including prolonged LOS. Of note, cost did not increase significantly until at least three complications

Table 4 Crude and adjusted average length of stay (LOS) and hospital charges (in thousands of dollars) among neonates during secondary hospitalizations, stratified by region, $n = 1,895$

	Average LOS (95% CI)	Average hospital charges[a] (95% CI)
Crude		
Northeast	24.4 (20.1, 28.7)	223.2 (168.9, 277.6)
Midwest	38.2 (35.0, 41.3)	374.7 (334.2, 415.4)
South	38.6 (34.7, 42.4)	470.1 (421.5, 518.8)
West	28.0 (25.0, 30.9)	358.0 (319.7, 396.3)
Adjusted[b]		
Northeast	31.9 (27.9, 36.0)	326.9 (270.7, 383.1)
Midwest	38.5 (34.6, 42.4)	427.0 (372.3, 481.7)
South	38.4 (34.4, 42.4)	505.9 (450.2, 561.5)
West	26.8 (23.9, 29.7)	339.1 (297.3, 381.0)

[a]In thousands of dollars

[b]Adjusted for admit year, gender, race/ethnicity, insurance status, household income, comorbidities, operations and procedures, transfer status, inpatient mortality, hospital size, and hospital teaching status

were present [19]. However, in our analyses, while LOS was correlated with total hospital charges, $p = 0.77$ (data not shown), LOS did not appear to be the sole driver of cost. For example, among birth hospitalizations, the West was associated with both the lowest LOS and the highest hospital charges, even after adjustment. Further studies are needed to examine the frequency as well as types of postoperative complications, and their influence on driving costs throughout the US, as efforts to reduce complications can lead to improved outcomes, decreased LOS, and lower total hospital charges.

Prior studies have noted the disproportionate investment in inpatient care of HLHS in those who do not survive [9]. The mortality rate of inpatient neonates with HLHS has declined in recent years, but continues to be significant [7]. We found significant mortality variation among neonates with HLHS across U.S. regions in both birth hospitalizations and secondary hospitalizations. The Northeast had the lowest mortality in both groups, whereas the South (birth hospitalizations) and Midwest (secondary hospitalizations) experienced higher mortality rates. Future studies using clinical databases can help determine the types and variation of procedures performed before death, as well as the variation in LOS and total charges among hospital non-survivors.

Pasquali and colleagues have shown that hospital costs in patients undergoing intervention and surgery vary from center to center [13]. Specific hospital characteristics, such as annual congenital heart surgery volume, may also be associated with efficient care, and the number of high-volume heart centers per region is variable [20, 21]. Further analysis is needed to determine the relationship between number of high-volume congenital heart centers in each region, and the regional variation in LOS and cost identified in the current study. Additionally, it is widely known that infants with HLHS that are considered high risk for open surgery may be initially offered a less invasive option for palliation [16]. Because the KID does not include the information needed to stratify patients by risk category, we were unable to comment in detail on the proportion of high-risk infants treated in any particular region. However, the South, which had the highest mortality and longest length of stay among birth hospitalizations, also had the highest proportion of neonates with other structural anomalies and chromosomal anomalies.

Information gained from the current study highlights the need to further explain cost and LOS variation between hospitals within the same region. A useful quality improvement project would lay additional groundwork for regional collaborations of hospitals caring for patients with HLHS. Ideally, hospitals within each region could learn from each other and improve care while decreasing costs for this high-risk cohort of patients.

Limitations

A major limitation of the KID is that like other administrative databases, it relies upon the ICD-9 diagnosis and procedure codes entered during a patient's hospital stay. There is no ICD-9 code for the Norwood operation, and a combination of other codes must be used to identify patients who have undergone this procedure, or a variation of it. The validity of using this method to ascertain the proportion of neonates undergoing particular procedures is unknown.

The KID also has inherent limitations related to risk adjustment; it provides limited information related to severity of illness, which can greatly influence LOS and charges. While this study was useful in determining the outcomes of LOS, total charges, and inpatient mortality, we were unable to analyze other clinically important outcomes, including mortality after discharge such as surgical interstage mortality, and neurologic sequelae after intervention. Also, maternal factors, and hospital costs, as opposed to charges, are not consistently available in the KID.

Lastly, there are many drivers of health care cost. These drivers may be categorized into (1) factors that influence pricing of services and, (2) factors that affect the utilization of services. For example, in any particular region, the market power of both the provider and the health plan and the cost of living affect commercial pricing. Likewise, the health status of the patient, physician practice patterns, patient cost-sharing level, and the number of in-network providers all contribute to differences in utilization. Although the current study is unable to provide a complete regional analysis of these factors, it does provide an essential starting point by describing the differences in inpatient charges for neonates with HLHS in US regions. A key next step will be

to examine reasons for these differences, by a systematic exploration of the aforementioned contributors to cost.

Conclusion

The care of US neonates with HLHS remains resource-intensive. Significant regional variability in LOS and hospital charges exists for this unique population of complex congenital heart patients. A better understanding of variability in US regional LOS and healthcare costs can lead to the development of strategies to alleviate these disparities.

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Compliance with Ethical Standards

Conflict of interest The authors declare that they have no conflict of interest.

Ethical Approval This article does not contain any studies with human participants or animals performed by any of the authors.

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