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Thoracic Conditions

Sources of regional and center-level variability in survival and cost of care for congenital diaphragmatic hernia (CDH)

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ABSTRACT

Purpose: Enormous variability in management and cost occurs in CDH care. The purpose of this study was to identify regional mortality and cost patterns underlying this variability.
Methods: This is a retrospective study of neonatal CDH patients at U.S. hospitals using data from the Pediatric Health Information System (PHIS) database (2015–2018). Patients were risk-stratified using CDH Study Group predicted survival (CDHSG-PS), and mortality and costs were assessed by region (East, West, Mid-West, and South) and center.
Results: Higher mortality and extracorporeal life support (ECLS) rates were found in the Mid-West and South ($p<0.0001$). Higher mortality was seen with ECLS among low-volume centers in the South ($p=0.007$). When broken down by CHDSG-PS, higher severity patients had higher mortality in the Mid-West and South ($p=0.038$). Cost was significantly lower for high severity nonsurvivors than survivors ($244,005 vs $565,487, $p=0.0008$). The East spent more on high-severity patients with lower mortality compared to other regions, but also spent 3.5 times more on low severity nonsurvivors than survivors. Costs were higher at high-volume centers for low- and medium-severity patients, but all centers spent the same on high-severity patients.
Conclusion: Center volume, region, and patient severity all contribute to the complex survival and cost disparities that exist in CDH care. Standardization of care may improve survival and reduce cost variability.
Type of study: Retrospective database study.
Level of evidence: Level II

Congenital diaphragmatic hernia (CDH) is a common neonatal surgical condition seen in approximately 1 in 3,000 live births. It is associated with high extracorporeal life support (ECLS) rates and high mortality rates. There is a broad range of severity of disease, and highly variable management strategies and practices. Little consensus exists in the literature regarding standardization of care [1], and only a few studies have focused on cost of care in the treatment of CDH [2–4]. A recent study by Cameron *et al.* found that CDH had the highest median cost of all pediatric surgical conditions, with an average cost per case of $158,113 [5]. Moreover, they found that CDH has the second highest interhospital cost variation, suggesting that costs are highly variable at different hospitals. One retrospective study of the Kids' Inpatient Database (KID) demonstrated that high-volume centers have higher associated costs compared to lower volume centers [6], and several studies have shown significant variations between those that require ECLS and those that do not [2,4], both contributing to some of the cost variations.

While lack of practice guidelines contributes to cost variations, it does not fully explain the high variability of CDH costs. This study sought to investigate regional patterns, trends, and underlying sources of cost variations in the treatment of CDH, taking into consideration severity of disease and center CDH volume. It is hypothesized that CDH cost patterns will be highly variable, that the origins of such variability will be multifactorial and complex, and that higher survival will be associated with increasing costs and high center volume.

1. Material and methods

1.1. Data source

This was a retrospective study of neonatal CDH patients at U.S. hospitals using data from the Pediatric Health Information System (PHIS) [7] from 2015 to 2018. PHIS is an administrative database affiliated with the Children's Hospital Association (Lenexa, KS) that contains inpatient, emergency department, ambulatory surgery, and observation encounter-level data from 51 not-for-profit tertiary care pediatric hospitals in the U.S. Data quality and reliability are assured through a joint effort between the Children's Hospital Association and participating hospitals. Portions of the data submission and data quality processes for the PHIS database are managed by IBM Watson Health (Ann Arbor, MI). For the purposes of external benchmarking, participating hospitals provide discharge/encounter data including demographics, diagnoses,

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and procedures. Nearly all of these hospitals also submit resource utilization data (e.g. pharmaceuticals, imaging, and laboratory) into PHIS. Data are deidentified at the time of data submission, and data are subjected to a number of reliability and validity checks before being included in the database. Furthermore, costs are standardized to account for regional price differences; charges listed in the PHIS database are adjusted for the wage and price index (published annually in the Federal Register). Institutional Review Board approval was obtained.

1.2. Patient population

The PHIS database was queried for neonates with a diagnosis of CDH, using an ICD10 code of Q790, who were discharged between October, 2015 and December, 2018. The following data were obtained: demographics, hospital, length of stay, ICU/NICU status, birthweight, gestational age, procedure codes, Apgar (Appearance, Pulse, Grimace, Activity, and Respiration) scores, diagnosis codes including comorbidities, date of birth, admission date, discharge date, disposition, and total cost of hospital admission. Patients were excluded for late-diagnosed CDH, defined as admission greater than 30 days after birth. Subsequent admissions after the initial admission were also excluded. If a patient was transferred from one facility to another, their data were consolidated into a single entry.

1.3. Statistical analysis

Centers were considered high-volume if they had ≥ 10 patients with CDH for ≥ two years. This was based on previous work that identified high-volume centers as those with 10 or more patients per year [8,9]. Patients were risk-stratified using CDH Study Group predicted survival (CDHSG-PS) [10]. The CHDHS-PS is based on 5-min Apgar score and birthweight, and was chosen because birthweight and Apgarwere variables included in the database allowing for calculation of the score. Furthermore, CDHSG-PS has been validated in multiple studies, has shown to be indicative of the initial response to resuscitation, has remained in use for several decades [11], and is at least as accurate as some other models when tested [11,12]. Based *a priori* on the CDHSG-PS, survivability of 0%–20% was considered high-risk or high-severity, 20%–80% was considered medium-severity, and 80%–100% was consider low-severity. Patients were also grouped by region: East, West, Mid-West, and South (Appendix A, Table 1). All costs were discounted to 2019 dollars using standard 3% discounting [14]. Finally, to account for survivorship bias, cost per day (cost intensity) was calculated as total estimated cost divided by length of stay in days. Student's t-test, Kruskal–Wallis, Chi-Squared, univariate, and multivariable regression analyses were performed when appropriate. All statistics were performed using R software [15]. Significance was defined as $p < 0.05$.

2. Results

Fifty-one centers provided data on 1,687 patients who met inclusion criteria. Overall patient characteristics are shown in Table 1. More than half of patients were male (59%), more than half were Caucasian (59%), and patients were roughly evenly distributed between regions. Overall mortality was 24.4%. Approximately 30% required ECLS support during their hospitalization.

2.1. Outcomes analysis

Univariate analysis was performed examining mortality (Table 1). Lower birthweight, earlier gestational age, and shorter length of stay were significantly associated with higher mortality ($p<0.0001$). Those with cardiovascular and neurological comorbidities were less likely to survive. Gender did not influence outcome, nor did the center volume. African Americans had higher mortality (32%) compared to Caucasians (21%, $p=0.0035$). ECLS use was associated with higher mortality rates (45% mortality in those that required ECLS vs 16% mortality in those that did not). Mortality was higher in the Mid-West and South ($p<0.0001$).

On multivariate analysis, race and region were no longer significant factors in predicting outcomes. ECLS use, cardiovascular, neurologic comorbidities, length of stay, gestational age, and birthweight were all independent predictors of mortality (Table 2). High center volume was not associated with a decrease in mortality (OR 1.06, CI 0.74–1.51, $p=0.76$).

Table 1
Overall patient demographics and univariate analysis of outcomes.

Factor	All ($n=1687$)	Survivors ($n=1269$)	Nonsurvivors ($n=412$)	p-value
Birthweight (g)	2932 ± 665	3030 ± 598	2643 ± 770	<0.0001*
Length of stay (days)	59.75 ± 72.3	66.32 ± 73.6	37.47 ± 73.4	<0.0001*
Gestational age (wks)	37.35 ± 2.38	37.7 ± 2.01	36.16 ± 3.07	<0.0001*
Cardiovascular comorbidity, *n* (%)	793 (47%)	564 (44%)	229 (55%)	0.0001*
Neurological comorbidity, *n* (%)	180 (11%)	119 (9%)	61 (15%)	0.003*
Estimated hospital costs per patient ($)	$359,717 ± $469,781	$348,738 ± $426,847	$382,632 ± $569,803	0.20
Volume, *n* (%)				
High	1259 (75%)	956 (76%)	301 (24%)	0.25
Low	428 (25%)	313 (74%)	111 (26%)	
ECLS use, *n* (%)				
Yes	503 (30%)	278 (55%)	224 (44%)	<0.0001*
No	1184 (70%)	991 (84%)	188 (16%)	
Gender, *n* (%)				
Female	680 (40%)	504 (74%)	174 (26%)	0.65
Male	1002 (59%)	761 (76%)	237 (24%)	
Race, *n* (%)				
Caucasian	1002 (59%)	788 (79%)	211 (21%)	0.0004*
African American	189 (11%)	128 (68%)	60 (32%)	
Pacific Islander	14 (0.8%)	11 (79%)	3 (21%)	
American Indian	4 (0.2%)	4 (100%)	0 (0%)	
Mixed	10 (0.6%)	10 (100%)	0 (0%)	
Unknown/other	468 (28%)	328 (70%)	138 (30%)	
Region, *n* (%)				
East	317 (19%)	255 (81%)	61 (19%)	<0.0001*
West	349 (21%)	273 (78%)	76 (22%)	
Mid-West	423 (25%)	299 (71%)	121 (29%)	
South	598 (25%)	442 (76%)	154 (26%)	

* $p<0.05$.

2.2. Regional variations

Regional variations were then examined. Overall patient characteris-

Table 2
Multivariate analysis of outcomes.

Variable	Odds ratio	95% CI	p-value
Volume	1.06	0.739–1.51	0.76
ECLS use	10.1	7.03–14.8	<0.0001*
Length of stay	0.981	0.976–0.985	<0.0001*
Cardiovascular comorbidity	1.82	1.31–2.54	0.0004*
Gestational age	0.796	0.725–0.871	<0.0001*
Birthweight	0.999	0.999–1	<0.0001*
Neurological comorbidity	2.15	1.33–3.47	0.002*
Region			
East	Reference		
West	1.31	0.739–2.35	0.36
Mid-West	1.57	0.92–2.72	0.10
South	0.781	0.451–1.37	0.38
Race			
Caucasian	0.505	0.35–7.28	0.0003
African American	0.747	0.427–1.29	0.30
Pacific Islander	0.499	0.072–2.67	0.45

* $p<0.05$.

tics are seen in Table 3. ECLS use, 5 min Apgar score, cardiovascular, and gastrointestinal comorbidities differed significantly between regions. Higher rates of ECLS use were seen in the Mid-West and South (31% and 36%, $p<0.0001$), as were cardiovascular comorbidities (48% and 51% respectively, $p=0.011$). Higher rates of gastrointestinal comorbidities were seen in the West (25%, $p=0.014$). As seen above, higher mortality was seen in the Mid-West and South. Lastly, there was no difference in the proportion of high- vs low-volume centers between regions.

2.3. Severity

Severity did not differ between high- and low-volume centers ($p=0.12$). When looking at severity by region, slight differences existed ($p<0.001$) (Fig. 1a). The Mid-West had higher rates of high-severity patients (13.7%) and the South had higher rates of medium-severity patients (65%). The East had the largest proportion of low-severity patients (42%) while the South had lower rates of low-severity patients (30%). Mortality was roughly the same in all regions for the low- and medium-severity patients (3%–12%, $p=0.06$ and 24%–29%, $p=0.8$, respectively). However, within the high-severity patients, the Mid-West and South had higher mortality rates (77%, 55%) compared to the East and West (33%, 46%, $p=0.009$).

2.4. ECLS use

ECLS use was compared by region (Fig. 1b). While ECLS use was found to be higher in the Mid-West and South, survival following ECLS use did not differ significantly between regions ($p=0.8$). However, when further stratified by high- vs. low-volume centers, low-volume centers in the South had significantly higher mortality (62%) compared to high-volume centers in the South (38%) with ECLS use ($p=0.007$).

2.5. Costs

The average cost was \$348,738 ± \$426,847 for survivors and \$382,632 ± \$569,803 for nonsurvivors ($p=0.2$). However, when adjusted for length of stay, average cost per day was higher in nonsurvivors than survivors (\$13,243 ± \$7,405 vs \$5,095 ± \$2,022, $p<0.0001$). There were no cost differences between regions ($p=0.96$). ECLS was found to be associated with significantly higher costs (\$627,416 vs \$245,990 without ECLS, $p<0.0001$). High-volume centers also had higher costs (\$395,291 vs \$255,074 for low-volume, $p<0.0001$). Stratified by severity, the average cost of a high-severity patient was \$365,892, a medium-severity patient was \$446,629, and a low-severity patient was \$236,668 ($p=0.48$). However, costs were higher in high-severity survivors than high-severity nonsurvivors (\$565,487 vs \$244,005, $p=0.0009$), and higher in low-severity nonsurvivors than low-severity survivors (\$566,116 vs \$214,153, $p=0.048$). There was no difference in costs between survivors and nonsurvivors in the medium-severity group ($p=0.9$).

Next, costs were examined by region stratified by severity (Fig. 2). For high-severity patients, the East spent significantly more on survivors (\$917,685) compared to nonsurvivors (\$188,631, $p=0.02$), and more compared to other regions ($p=0.04$). All regions spent roughly the same on medium-severity patients ($p=0.83$). For low-severity patients, the East spent significantly more on nonsurvivors (\$949,832, $n=2$) compared to survivors (\$267,435, $n=74$, $p=0.04$). These two nonsurvivors were both treated at the same center and had lengths of stay greater than 80 days, and neither developed a surgical complication. Similar trends are seen in other regions with higher expenditures and high variation in low-severity nonsurvivors, as were seen overall, but do not reach statistical significance in other regions.

Costs were also stratified by center volume and severity (Fig. 3). For high-severity patients, both high- and low-volume centers spent significantly more on survivors than nonsurvivors, but there was no difference in costs between high- and low-volume centers. However, in the medium- and low-severity patients, high-volume centers spent more on survivors (\$481,634 for medium, and \$239,937 on low severity)

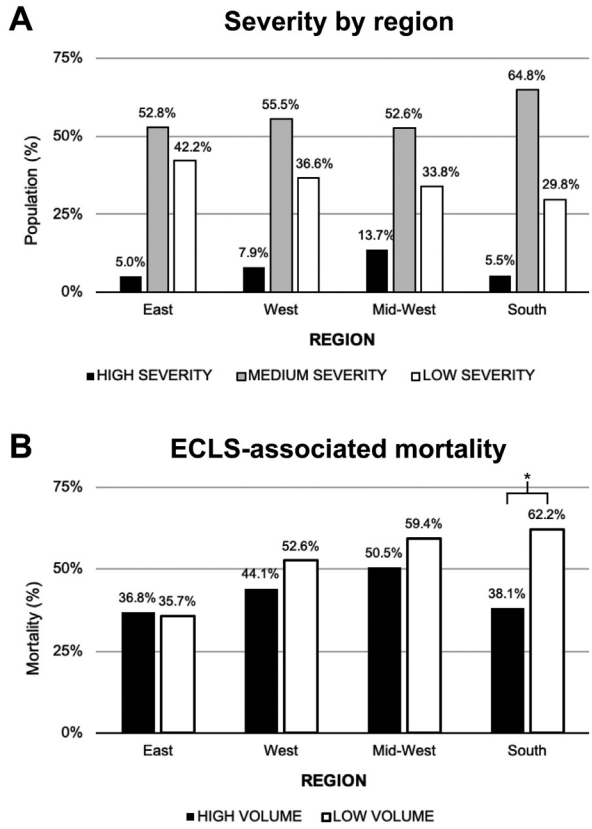

Fig. 1. a) Severity by region: Percent of the population in each severity group, high, medium and low, stratified by region. b) Mortality with ECLS by region and volume: ECLS-associated mortality stratified by region, between high- and low-volume centers. Low-volume centers have significantly higher ECLS associated mortality. * $p<0.01$

Table 3
Regional differences in patient demographics, outcomes and costs.

	East	West	Mid-West	South	p-value
Birthweight (g)	2955 ± 674	2924 ± 669	2934 ± 702	2924 ± 630	0.55
Length of stay (d)	66.9 ± 82.4	56.4 ± 62.9	59.9 ± 84.3	57.8 ± 70.9	0.06
Gestational age (wks)	37.3 ± 2.6	37.3 ± 2.6	37.3 ± 2.4	37.5 ± 2.1	0.72
Costs (\$)	364,752 ± 489,716	348,066 ± 399,425	365,715 ± 537,774	359,607 ± 445,852	0.96
Mortality, *n* (%)	61 (19%)	76 (22%)	121 (29%)	154 (26%)	<0.0001*
5 min Apgar	7.09	6.79	6.26	6.69	0.004*
ECMO use, *n* (%)	83 (26%)	78 (22%)	129 (31%)	213 (36%)	<0.0001*
Comorbidities					
Cardiovascular, *n* (%)	138 (44%)	152 (44%)	203 (48%)	304 (51%)	0.011*
Neurological, *n* (%)	29 (9%)	42 (12%)	52 (12%)	58 (10%)	0.83
Gastrointestinal, *n* (%)	71 (22%)	86 (25%)	94 (22%)	117 (22%)	0.014*
Infections, *n* (%)	135 (43%)	178 (51%)	172 (41%)	270 (45%)	0.81
Surgical complications, *n* (%)	65 (21%)	100 (29%)	117 (28%)	146 (24%)	0.65

* $p<0.05$.

compared to low-volume centers ($329,514, p = 0.0001, and $167,108, p = 0.008, respectively), and spent more on nonsurvivors ($513,939 and $867,782) compared to low-volume centers ($311,615, p = 0.04 and $130,376, p = 0.01). Furthermore, high-volume centers also spent significantly more on low-severity nonsurvivors ($867,782) compared to low-severity survivors ($239,937, p = 0.03).

3. Discussion

Using the PHIS database, outcomes and costs in CDH care were found to be highly variable. Lower birthweight, earlier gestational age, cardiovascular comorbidities, neurological comorbidities, African American race, and ECLS use were all associated with higher mortality. High-volume centers did not have lower mortality. The South and Mid-West were also found to have higher mortality rates, stemming from higher rates of cardiovascular comorbidities, higher ECLS rates, and higher mortality in high-severity patients. ECLS-associated mortality at low-volume centers was significantly higher in the South.

Higher mortality in African Americans compared to Caucasians was previously reported [16] and again seen here. However, the same study found better survival at high-volume centers, something not seen in the PHIS data set. Higher mortality rates in the South and Mid-West are partially explained by the findings that the same regions have higher rates of cardiovascular comorbidities, which are independently associated with higher mortality. While more investigation is needed to understand why there are higher rates of cardiovascular comorbidities in the South and Mid-West, it was also seen that the South and Mid-West have higher mortality in high-severity patients and that the South in particular had higher ECLS-associated mortality in low-volume centers. These are potential areas where improvements can be made.

Overall costs of CDH care were on average greater than $350,000 per patient, and extrapolating from these data, CDH has an annual national cost burden of upwards of $390,000,000. No overall difference in costs was found between survivors and nonsurvivors, despite increased length of stay in survivors, or between regions, and cost-per-day was higher in nonsurvivors. ECLS use and high-volume centers were also associated with significantly higher costs. High-severity survivors cost more than nonsurvivors, regardless of center volume, while low-severity nonsurvivors had higher costs than survivors in high-volume centers. These trends were specifically seen only in the East once divided by region. The trend that high-volume centers in the East are spending substantially more on low-severity nonsurvivors is difficult to explain, but because of low sample size, these may be outliers.

The complexity of CDH patients clearly contributes to outcome and cost variations. ECLS has previously been cited as a significant driver of costs, which was seen again in this study [4], as well as being an indicator of severity on its own. There was an almost $400,000 difference in cost between those receiving ECLS and those that did not, which would contribute to such large cost variations that were previously reported by Cameron et al. [5]. Higher volume centers also had higher expenditures, almost $150,000 more than low-volume centers, and high-volume centers did not have improved survival. Higher volume centers have been previously reported to have higher costs [6], which may be attributable to high-volume centers having a higher proportion of sicker patients. This was not found to be the case in the present study, though, as no difference in severity was found between high and low-volume centers. Furthermore, no statistically significant difference in expenses was found between high-, medium-, and low-severity patients. So, the presence of higher severity patients at high-volume centers does not explain the higher costs. Further research is indicated to deter-

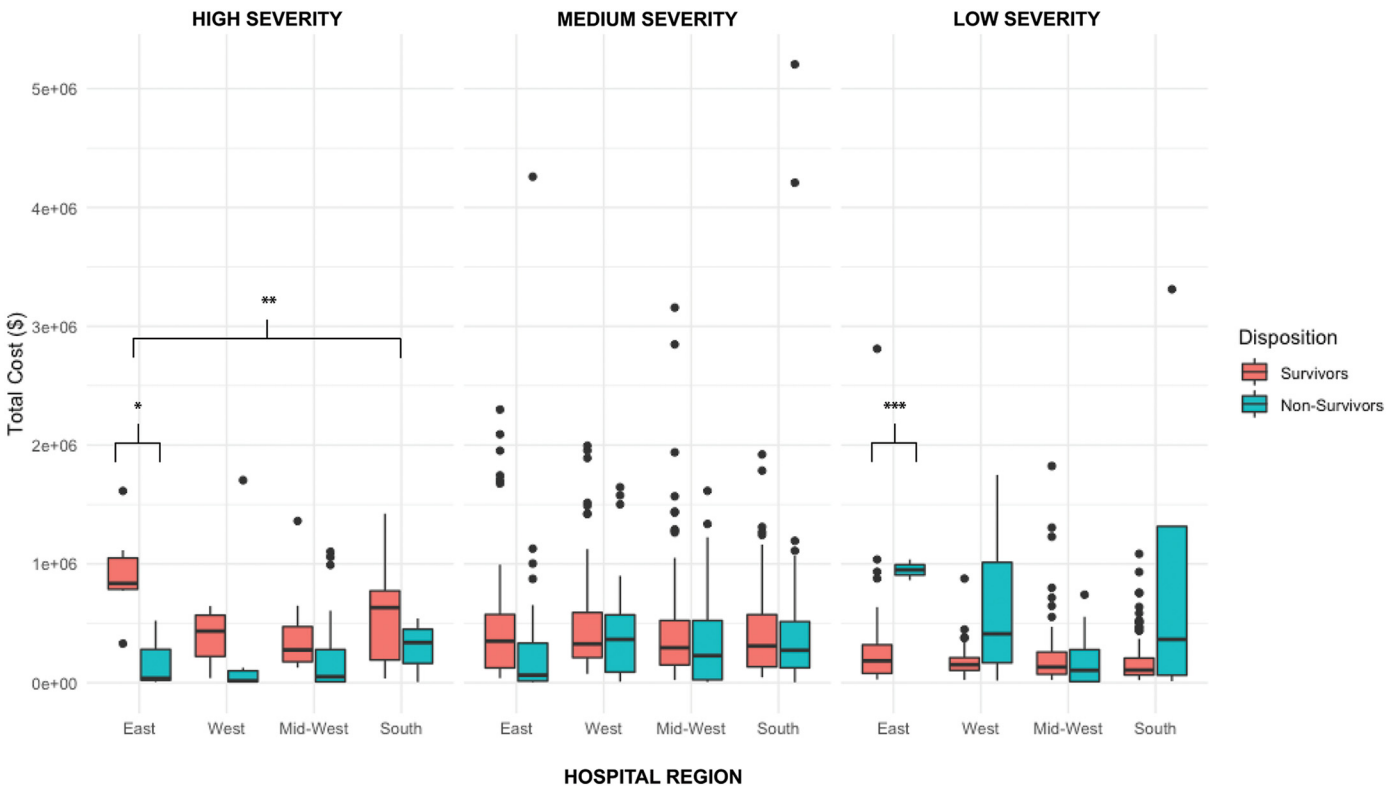

Fig. 2. Average costs by region, stratified by severity: Box and whisker blot of average hospital costs in 2019 dollars, stratified by region and survival status. The East spent significantly more on survivors compared to nonsurvivors in the high severity group (*, p = 0.02), and more on survivors and less on nonsurvivors in the high severity group compared to other regions (**, p = 0.04). In the low severity group, the East spent significantly more on nonsurvivors compared to survivors (***, p = 0.04).

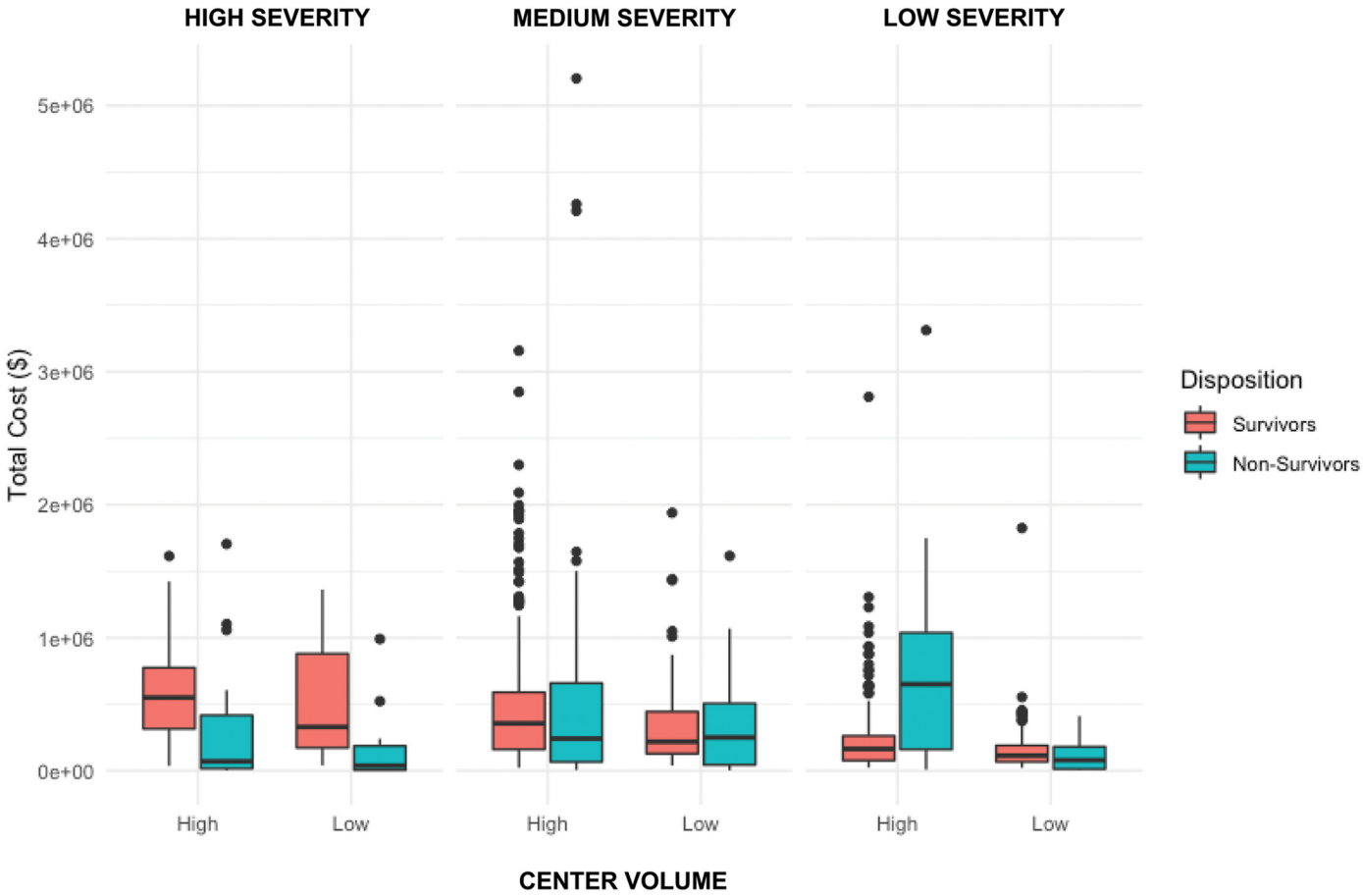

Fig. 3. Average costs ($) by center volume and severity: Box and whisker blot showing average hospital costs in 2019 dollars, stratified by center volume status and severity. Both high- and low-volume centers spent more on high severity survivors than nonsurvivors ($p = 0.01$, $p = 0.03$ respectively). However, in the medium- and low-severity patients, high-volume centers spent more on survivors compared to low-volume centers ($p = 0.008$) and spent more on nonsurvivors compared to low-volume centers ($p = 0.01$). Furthermore, high-volume centers also spent significantly more on low severity nonsurvivors compared to low severity survivors ($p = 0.03$).

mine the ultimate contributor to higher costs in high-volume centers and will be the next steps in this research.

While this study is unable to identify the source of much of this variation, it does provide some specific areas of focus. Much of the variability seen in outcomes and costs may benefit from standardization of care, specifically areas like low-volume centers using ECLS in the South. Previous attempts have found standardization of care for CDH difficult owing to highly variable strategies, multidisciplinary teams, and medically complex patients [1]. While challenging, a move towards standardization of care may not only improve outcomes, but also address variations in outcomes and costs.

This study had several limitations. As a database study, information was limited to what was collected by the database and missing information or unknown information was a noteworthy barrier. The PHIS database did not collect information related to transfers, nor did it provide a unique identifier for each unique patient that can be tracked when a patient was transferred between two facilities. It was also difficult to determine if patients were transferred from non-PHIS hospitals to a PHIS hospital, especially if transfer occurred the same day as birth. Long-term or delayed mortality was unable to be assessed. Final disposition was unknown if a patient was discharged to a rehab facility and this study was thus limited to survival to discharge. PHIS also did not include data such as nitric oxide use or high frequency oscillatory ventilation, nor did it include information regarding prenatal diagnosis or pulmonary hypertension, and these important variables were therefore not included in the study. Additionally, coding errors remain a possibility, specifically with regard to comorbidities and procedure codes. This analysis was also unable to include information regarding prenatal mortality or fetuses that may have been aborted nor did it include information regarding access to prenatal care. Finally, PHIS hospitals represent a subset of children's hospitals and the results therefore might not be applicable to non-PHIS hospitals.

4. Conclusion

The care and treatment of patients with CDH are highly complex with large amounts of variability. This variability is seen in outcomes difference between regions of the US, with both the Mid-West and South having significantly poorly outcomes. This variability is also seen with expenditures associated with CDH care. Some of the wide variations in costs can be attributable to ECLS use, which comes with a large cost burden, but other variations seen are hard to explain. Further research and a push into standardization of care are needed to address these disparities.

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Author contributions

Study conception and design: TJ
Data acquisition: TJ
Analysis and data interpretation: RL

Drafting of the manuscript: RL
Critical revision: RL, TJ

Appendix A

Table 1
PHIS Hospitals represented 28 different states. They were grouped by region: East, West, Mid-West and South.

East	West	Mid-West	South
Connecticut (CT)	Arizona (AZ)	Illinois (IL)	Alabama (AL)
District of Columbia (DC)	California (CA)	Indiana (IN)	Arkansas (AR)
Massachusetts (MA)	Colorado (CO)	Michigan (MI)	Florida (FL)
New York (NY)	Utah (UT)	Minnesota (MN)	Georgia (GA)
Pennsylvania (PA)	Washington (WA)	Missouri (MO)	Kentucky (KY)
Virginia (VA)		Nebraska (NE)	North Carolina (NC)
		Ohio (OH)	South Carolina (SC)
		Wisconsin (WI)	Tennessee (TN)
			Texas (TX)

Appendix B. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.jpedsurg.2020.09.025.

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