ORIGINAL RESEARCH

Resource Utilization in the First 2 Years Following Operative Correction for Tetralogy of Fallot: Study Using Data From the Optum's De-Identified Clinformatics Data Mart Insurance Claims Database

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BACKGROUND: Despite excellent operative survival, correction of tetralogy of Fallot frequently is accompanied by residual lesions that may affect health beyond the incident hospitalization. Measuring resource utilization, specifically cost and length of stay, provides an integrated measure of morbidity not appreciable in traditional outcomes.

METHODS AND RESULTS: We conducted a retrospective cohort study, using de-identified commercial insurance claims data, of 269 children who underwent operative correction of tetralogy of Fallot from January 2004 to September 2015 with ≥2 years of continuous follow-up (1) to describe resource utilization for the incident hospitalization and subsequent 2 years, (2) to determine whether prolonged length of stay (>7 days) in the incident hospitalization was associated with increased subsequent resource utilization, and (3) to explore whether there was regional variation in resource utilization with both direct comparisons and multivariable models adjusting for known covariates. Subjects with prolonged incident hospitalization length of stay demonstrated greater resource utilization (total cost as well as counts of outpatient visits, hospitalizations, and catheterizations) after hospital discharge (*P*<0.0001 for each), though the number of subsequent operative and transcatheter interventions were not significantly different. Regional differences were observed in the cost of incident hospitalization as well as subsequent hospitalizations, outpatient visits, and the costs associated with each.

CONCLUSIONS: This study is the first to report short- and medium-term resource utilization following tetralogy of Fallot operative correction. It also demonstrates that prolonged length of stay in the initial hospitalization is associated with increased subsequent resource utilization. This should motivate research to determine whether these differences are because of modifiable factors.

Key Words: congenital heart disease ■ health services research ■ outcomes ■ pediatrics

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In contemporary series, early mortality after operative correction of tetralogy of Fallot (TOF) is <2% across a wide range of hospitals.[1–3] However, this may overestimate the health and well-being of children after this operation. Residual anatomic lesions such as branch pulmonary artery stenosis and recurrent right ventricular outflow tract obstruction are common.[1] These lesions can have significant hemodynamic consequences that

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CLINICAL PERSPECTIVE

What Is New?

- Morbidity and prolonged length of stay during the incident hospitalization after operative correction of tetralogy of Fallot is associated with increased resource utilization in the following 2 years.

What Are the Clinical Implications?

- Though further research is necessary, the data presented suggest that perioperative mortality insufficiently describes the variability in health and well-being of patients after operative correction of tetralogy of Fallot, and that strategies of care that reduce the risk of early morbidity may have durable benefits in this patient population.

Nonstandard Abbreviations and Acronyms

ICD	International Classification of Diseases
IQR	interquartile range
LOS	length of stay
TOF	tetralogy of Fallot
US$	2016 United States dollars

may eventually require transcatheter and/or surgical interventions,[4–9] as well as increased utilization of both outpatient and inpatient care. The magnitude of this morbidity and its variability are not described in case series using traditional outcome metrics. The coincidence of residual lesions and intensive medical care both in terms of re-intervention and medical follow-up in TOF and other conotruncal anomalies are associated with diminished health-related quality of life and functional status.[10–12]

Previously, the cumulative impact of postoperative morbidity on children's health has been challenging to measure, especially in observational data sets, because it is manifest in multiple events (not only increased re-interventions but also inpatient, emergency department, and outpatient encounters) whose relative importance is hard to compare. Cost data have been used to overcome this obstacle. They not only measure the accumulated economic impact of medical care but also integrate intensity and frequency of medical care and morbidity, which provides a surrogate for cumulative morbidity.[13–16] This provides a statistically expedient and potentially more comprehensible measure than counts of individual

encounters and/or interventions. Previous studies have demonstrated that the costs during the hospitalization increase in patients with postoperative complications and longer total length of stay (LOS).[17–19] We hypothesize that resource utilization over the first 2 years after operative correction is a potentially valuable proxy for the health and well-being of these patients and may help inform decisions about how best to care for these patients. To our knowledge, this has not been studied in patients after operative correction of TOF.

To that end, we performed a retrospective cohort study using commercial insurance claims data to generate a national sample and characterize resource utilization over the first 2 years after operative correction of TOF. Second, we sought to determine whether early morbidity (reflected in prolonged LOS in the incident hospitalization) was associated with higher costs of subsequent care. We hypothesized that early morbidity is associated with persistent increases in morbidity and cost over the follow-up period. Finally, we sought to explore whether these costs varied between geographic regions.

METHODS

Study Design and Data Source

We conducted a retrospective cohort analysis using the Optum de-identified Clinformatics DataMart (OptumInsight, Eden Prairie, MN), a de-identified administrative health database, including claims data from recipients of commercial health insurance and Medicare Advantage (C and D). This database, updated annually, comprises 63 million unique individuals, spanning all 50 states, providing a geographically representative sample of children and families. It does not include recipients of Medicaid who represent a significant minority (30%–40%) of children with congenital heart disease seen in other studies using observational data sets.[14,15,20–24] Therefore, the resulting study population has a higher socioeconomic status than the total population with TOF. The DataMart comprises detailed enrollment information, medical records, prescription drug use, and inpatient hospitalization data. The study team had full access to the data. Data cannot be made publicly available because that runs contrary to our data use agreement. The study was reviewed by the Institutional Review Board at The Children's Hospital of Philadelphia and was judged as not constituting human subjects research in accordance with the Common Rule (45 CFR 46.102(f)).

Study Population

We identified subjects who underwent operative correction for TOF from January 1, 2004 and September

30, 2015 as defined by the (*ICD-9*) procedure code for complete TOF operative correction (35.81). To ensure data completeness and avoid bias, we restricted analysis to subjects with continuous enrollment in the database for at least 2 years after the admission where operative correction was performed, the incident hospitalization. This choice prioritizes generating a complete data set over generating the largest possible sample. To evaluate for potential selection bias, the demographics and LOS following operative correction were compared between study subjects (those with >2 years of continuous data) and those with <2 years of data. There were no significant differences between the 2 groups (Table S1). These inclusion criteria also exclude children who died during their initial hospitalization. By definition there is no resource utilization after discharge for subjects who die during their initial hospitalization; therefore their omission does not bias or influence our primary outcome. Two years of follow-up was chosen because previous studies have demonstrated that the greatest intensity (frequency and severity) of resource utilization occurs in the first 2 years after operative correction for TOF[8,9,25] and other conotruncal anomalies.[10,11]

Study Measures and Variables

Data were extracted directly from the Optum de-identified Clinformatics DataMart and included age (measured in integer years), sex, race (white versus nonwhite), total household income, and highest household education level. Geographic regions were divided into the 4 United States Census Bureau regions: (Midwest, Northeast, South, and West). The following subject level covariates were also extracted (using [*ICD-9* and *ICD-10*] codes): prematurity (gestational age <37 weeks), genetic syndromes (including Alagille syndrome, CHARGE association, 22q11.2 microdeletion syndrome, Goldenhar syndrome, Kabuki syndrome, Noonan syndrome, Williams syndrome, Turner syndrome, Trisomy 21, Trisomy 13, Trisomy 18, cri du chat, VATER association, and Holt-Oram syndrome), and extracardiac malformations (including cleft palate, cleft lip, esophageal atresia, tracheoesophageal fistula, duodenal atresia, anal atresia, biliary atresia, congenital diaphragmatic hernia, intestinal malrotation, gastroschisis, omphalocele, and renal agenesis). *ICD-10* codes were used to identify these covariates starting in October 1, 2015.

Data for the incident hospitalization and for subsequent health care were collected. Total standardized costs were calculated for each inpatient and outpatient visit. Adjustments for inflation were performed by correcting all costs to 2016 United States dollars (US$2016) using the consumer price index for medical care. To account for differences in pricing across

health plans and provider contracts, Optum applies standard pricing algorithms to the claims data in the de-identified Clinformatics DataMart. These algorithms are designed to create standard prices that reflect allowed payments for all provider services across regions. For example, professional service rates are standardized using a resource-based relative value scale approach with prices standardized at ≈130% of Medicare Fee for Service amounts. Cases with costs listed as ≤US$0 were excluded. Other component measures of resource utilization that were collected were total hospital LOS for the incident and subsequent hospitalizations, number of hospitalizations and number of cardiac surgeries following initial TOF operative correction, number of cardiac catheterizations, outpatient visits, emergency room visits, ECGs, and echocardiograms. The number of outpatient visits was defined as the sum of cardiac-specific (defined as primary diagnosis of a cardiology-related code) and noncardiac visits. Segregating costs during initial and subsequent inpatient admissions between cardiac and noncardiac admissions/services was not possible.

Statistical Analysis

Descriptive statistics were calculated for the baseline characteristics, including demographics and comorbidities. Categorical data are presented as frequencies and percentages, and continuous variables are presented as medians and interquartile range (IQR).

We then described resource utilization in the incident hospitalization and subsequent 2 years of follow-up after TOF operative correction using total cost as the primary outcome and number of hospitalizations and other services as secondary outcomes. Second, we studied the association between postoperative morbidity (represented by increased LOS during the incident hospitalization) and persistently increased resource utilization (indicative of ongoing morbidity). For this analysis, we divided the sample into 2 groups based on the median LOS for the entire cohort (7 days) with a shorter LOS (ie, LOS ≤7 days) and prolonged LOS Group (LOS >7 days). Though evaluating for the specific cut point for LOS would be important and useful, this is not feasible in the relatively small data set we have available, so no analyses using splines or other techniques were performed. We used all available hospitalizations (ie, not restricted to cardiology visits) for each subject after the incident hospitalization within the 2-year period and aggregated the outcomes. Univariable and multivariable generalized linear models were used to test whether prolonged LOS in the incident hospitalization was associated with greater resource utilization in subsequent follow-up. We modeled count outcomes (number of outpatient visits, catheterizations,

subsequent hospitalization(s)) using a Poisson distribution. Likelihood of dichotomous outcomes (eg, having at least 1 subsequent hospitalization) were modeled with a binomial frequency distribution (with a logit link). For cost data, gamma distributions were used as described previously. Covariates included sex, parental education, age at surgery, prematurity, genetic syndrome, extracardiac malformation, and census region. Because ages are rounded to the nearest whole year, it is not possible to determine which subjects underwent neonatal operative correction, a group at higher risk for adverse outcome and reintervention.[26] Also, since the analysis focused on the association between hospital course after correction and subsequent resource utilization, palliation with Blalock-Taussig shunt or other means was not included as a covariate. Since costs were adjusted for regional wage–price indices, census region was not included as a covariate in multivariable models of cost.

Next we studied regional variation in resource utilization. The Kruskall–Wallis test was used to test whether the LOS of the incident hospitalization differed between census regions and to evaluate the association between census regions and the likelihood of subsequent hospitalizations. Before analysis, there was no suspicion that patient clinical characteristics would vary between census regions, but we did suspect that race and family socioeconomic status might vary. To address this, we calculated estimates of LOS and cost adjusted for race and maternal educational level using generalized linear models with a gamma distribution. There is no criterion standard modeling strategy for costs and LOS, which are inherently ≥0 and skewed. Gamma frequency distributions were used to model total cost because they have demonstrated benefits for analysis of cost data in simulations[27] and have been used extensively in previous studies in young patients with cardiac disease.[14,15,20,22,28]

All analyses were performed using SAS version 9.3 (SAS Institute, Inc., Cary, NC). A threshold for significance was set at $P<0.05$.

RESULTS

Study Population

In total, 543 subjects met initial criteria for inclusion. Of these, 50% (n=269) had continuous enrollment data for at least 2 years following operative correction and were evaluated. Baseline demographic and socioeconomic characteristics are presented in Table 1. Overall, 53% of subjects were male, and 65% were white. South (49%) and Midwest (25%) census regions accounted for a higher proportion of cases. Total annual

Table 1. Demographic Characteristics (n=269)

Male sex	143 (53)
White race (vs other races)	176 (65)
Age at operative correction, y	
0	133 (49)
1	121 (45)
2	15 (6)
Regions	
Northeast	28 (10)
Midwest	68 (25)
South	131 (49)
West	42 (16)
Parental highest education level	
<12th grade	3 (1)
High school diploma	48 (18)
<Bachelor's degree	125 (46)
Bachelor's degree plus	89 (33)
Unknown/missing	4 (1)
Family annual income	
<$40K	15 (6)
$40K–$49K	13 (5)
$50K–$59K	13 (5)
$60K–$74K	16 (6)
$75K–$99K	35 (13)
>$100K	112 (42)
Unknown/missing	65 (24)
Insurance type	
Point of service	187 (70)
Exclusive provider organizations	36 (13)
Health maintenance organizations	33 (12)
Preferred provider organization	12 (4)
Other	1 (0.4)

Data are presented as N (%).

household income was >US$100 000 for 42% of subjects for whom income was recorded.

Resource Utilization

The median LOS for the incident hospitalization was 7 days (IQR 5, 11), with a median cost of US$58 362 (IQR: 43 672, 87 680). Of 269 subjects, 96 (36%) had at least 1 subsequent hospitalization with a total of 215 repeat hospitalizations. Subsequent hospitalizations had a median LOS of 6 days per admission (IQR 3–12), with corresponding median cost of US$26 280 (IQR 11 712, 78 457). Subsequent cardiac surgeries (within 2 years of operation correction) occurred in 40 subjects (15%), and 32 (12% of the total population) underwent >1 subsequent operation. Forty-nine subjects (18%) had subsequent cardiac catheterizations, 32 (12%) of whom had >1 catheterization. The median number of outpatient cardiology visits was 6

(IQR 4, 8) and of emergency room visits was 1 (IQR 0, 2).

Association of Prolonged LOS After TOF Operative Correction and Later Resource Utilization

To determine whether morbidity after the incident operation was associated with increased resource utilization after discharge, the population was divided between subjects with greater than median LOS (>7 days) during their incident hospitalization for operative correction and those with shorter LOS. The baseline characteristics of subjects with prolonged initial LOS differed significantly from those with shorter LOS (Table 2). Subjects with prolonged LOS had higher likelihood of noncardiac malformations (P=0.04), genetic syndromes (P=0.0003), and younger age at initial TOF operative correction (P=0.01). The proportion with a history of prematurity was higher (23% versus 14%) but this association was not significant (P=0.06).

As expected, prolonged initial LOS was associated with more than a 2-fold increase in inpatient cost (P<0.0001) for the incident hospitalization (Table 3). It was also associated with a larger number of subsequent outpatient visits (median 29 versus 18, P<0.0001) as well as a higher total cost (median US$29 219 vs US$17 039, P=0.009) for those visits. Prolonged LOS during incident hospitalization was also associated with an increased likelihood of undergoing subsequent

inpatient hospitalization(s) (P<0.0001). The median LOS for subsequent hospitalizations was also longer for those with prolonged initial LOS (7 days, IQR: 3, 13) compared with those with shorter LOS (4 days, IQR: 2, 9), but this difference was not statistically significant (P=0.06). The proportion of subjects undergoing catheterization appeared higher in the prolonged LOS group compared with those with shorter length of stay, but this also was not statistically significant (P=0.06). No differences were identified in number of repeat surgeries, ECGs, or echocardiograms between the 2 groups.

To adjust for measurable confounders, we conducted multivariable regression models to test whether prolonged LOS was associated with greater resource utilization after discharge (Figure). Prolonged initial LOS was independently associated with increased total cumulative costs (cost ratio 2.24, P<0.0001). Prolonged LOS was also associated with a greater number of subsequent hospitalizations (P<0.0001), increased risk of ≥1 hospitalization (P<0.0001), and greater total outpatient visits (P<0.0001). There was no significant association between prolonged LOS and the number of subsequent cardiac operations (P=0.94), risk of ≥1 subsequent cardiac operation (P=0.65), and number of cardiac catheterizations (P=0.15).

Regional Variability in LOS and Resource Utilization

Observed differences in resource utilization were calculated (Table S2). Estimates adjusted for race and maternal education are summarized in Table 4. The LOS for the incident hospitalization, during which TOF operative correction was performed, was not significantly different across regions (P=0.19). However, costs were significantly different (P=0.002), with the lowest cost in the Northeast (US$55 684; 95% CI, 38 911–79 689) compared with ≈150% higher costs in the South (US$82 155; 95% CI, 64 100–105 296) and ≈180% higher costs in the West (US$98 679; 95% CI, 71 944–135 349).

After discharge, there were also regional differences in resource utilization. The number of subsequent hospitalizations (P=0.0002) and their associated costs (P=0.05) were highest in the South relative to other regions, as were subsequent outpatient visits (P=0.01) and their associated costs (P=0.008). The number of emergency department visits was significantly higher in the Midwest and Northeast relative to the South and West (P=0.03). Costs associated with emergency department visits were highest in the South compared with other regions (P=0.01). This difference did not result in a significant difference in cumulative total costs for emergency department visits during follow-up (P=0.28).

Table 2. Comparison of Baseline Characteristics of Subjects With Prolonged and Shorter Total Hospital Length of Stay During Incident Hospitalization

	LOS ≤7 d (n=151)	LOS >7 d (n=118)	P Value
Male sex	83 (55)	60 (51)	0.50
White race	97 (64)	79 (67)	0.64
Age at surgery, y			0.01
<1 y	67 (44)	66 (56)	
1–2 y	79 (52)	42 (36)	
≥2	5 (3)	10 (9)	
Regions			0.38
Northeast	20 (13)	8 (7)	
Midwest	37 (25)	31 (26)	
South	72 (48)	59 (50)	
West	22 (15)	20 (17)	
Premature (<37 wk)	21 (14)	27 (23)	0.06
Noncardiac malformation	11 (7)	18 (15)	0.04
Genetic syndrome	30 (20)	47 (40)	0.0003

Data are presented as N (%). LOS indicates length of stay.

Table 3. Comparison of Postoperative and Short-Term Outcomes of Subjects With Prolonged and Shorter Total Hospital Length of Stay During Incident Hospitalization

	LOS ≤7 d (n=151)	LOS >7 d (n=118)	*P* Value
Inpatient costs for incident hospitalization ($)	43 672 (38 480, 58 362)	93 575 (71 738, 14 927)	<0.0001
Total outpatient visits			
Number	18 (12, 29)	29 (19, 48)	<0.0001
Cost ($)	17 039 (7496, 345 112)	29 219 (12 469, 66 081)	0.009
Subsequent hospitalizations			<0.0001
0	118 (78)	55 (46.61)	
1	16 (11)	29 (24.58)	
≥2	17 (11)	34 (28.81)	
Cumulative LOS, d	4 (2, 9)	7 (3, 13)	0.06
Cumulative cost ($)	24 004 (9513, 81 649)	27 852 (13 438, 79 648)	0.60
Subsequent cardiac operations			0.20
0	115 (76)	100 (83)	
1	14 (9)	9 (8)	
≥2	23 (15)	9 (8)	
Catheterizations			0.13
0	131 (87)	89 (75)	
1	7 (5)	13 (11)	
≥2	13 (9)	16 (14)	
ECGs	3 (2, 5)	4 (2, 6)	0.07
Echocardiograms	4 (2, 5)	4 (3, 6)	0.13

Data are presented as N (%) or median (interquartile range). LOS indicates length of stay.

DISCUSSION

The current multicenter cohort study demonstrates that there is significant resource utilization in the 2 years following operative correction of TOF. It also demonstrates that prolonged LOS after operative correction is associated with increased resource utilization even after discharge. It appears that morbidity in the perioperative period persists after hospital discharge. This highlights that conventional outcome measures following operative correction (ie, operative mortality) do not provide a complete depiction of the health of these patients. More optimistically, this suggests that strategies that reduce morbidity and LOS after operative correction may have durable benefits extending beyond hospital discharge. To our knowledge this is the first study to evaluate resource utilization and economic impact after hospital discharge in this population.

The low rate of mortality after operative correction of TOF[1–3] may overestimate the health and well-being of infants after discharge. Residual or iatrogenic anatomic lesions are relatively common as are postoperative adverse events.[7–9] Data from the Society for Thoracic Surgeons Congenital Heart Surgeons Database (reflecting the experience of the vast majority of US congenital cardiac surgery centers) demonstrate a discrepancy between the calculated standardized risk of mortality (STAT category 2/5) and standardized risk of morbidity (Morbidity category 3/5).[2,29] Tremendous variability in the cost of cardiac surgeries (including operative correction of TOF) has been demonstrated, which is driven, in part, by variation in the incidence of major adverse events.[16,30,31] The risk of residual anatomic lesions and perioperative complications may vary more between hospitals than the uniform mortality rates suggest. This variability also represents an opportunity to improve both short- and long-term outcomes in these patients.

LOS after operative correction can be prolonged by a multitude of complications, including post–cardiopulmonary bypass systemic inflammation, kidney injury, prolonged ventilation, cyanosis, residual lesions, and other postoperative cardiac and noncardiac complications.[32] The current study cannot demonstrate that the events that affect LOS are the same as those influencing the observed increase in resource utilization, but the association is intriguing, especially since there is mounting evidence that the risk of prolonged LOS after operative correction for TOF may be modifiable. For example, patients with TOF undergoing neonatal intervention have a higher risk of residual anatomic lesion and longer LOS. Palliation with an operative shunt (instead of neonatal repair) is associated with reduced risk of operative mortality and major adverse events compared with single-stage operative correction.[25] Transcatheter (stent angioplasty of either the right ventricular outflow tract or patent ductus arteriosus) options for
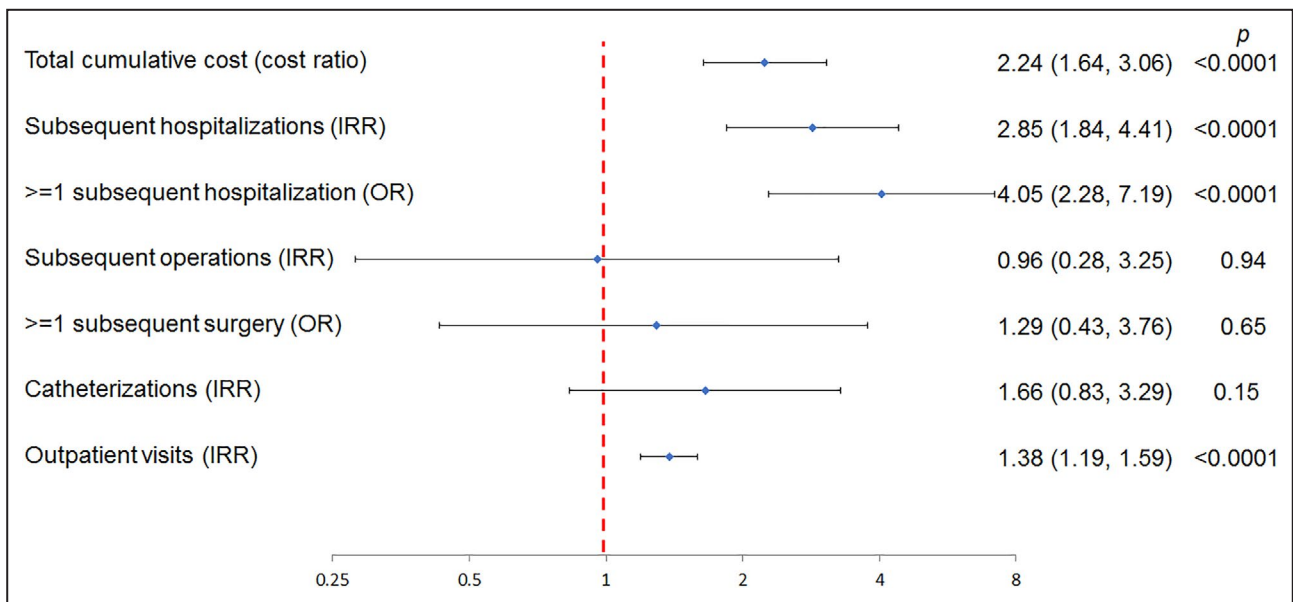
Figure. **Association of prolonged LOS after operative correction and subsequent resource utilization in multivariable models.**
This Forest plot depicts the results of a series of multivariable models evaluating the association between prolonged LOS after initial operative correction and subsequent measures of resource utilization. Point estimates for each model are depicted with a light blue diamond. Brackets represent 95% CIs. Point estimates and confidence intervals that are completely to the right of 1 (red dashed line) reflect increased resource utilization. Those that are completely to the left reflect decreased resource utilization. Each model is adjusted for sex, parental education, age at surgery, genetic syndrome, noncardiac congenital malformations, prematurity, and census region. The exception to this is the model of cost, which was not adjusted for census region. IRR indicates incidence rate ratio; LOS, length of stay; and OR, odds ratio.

palliation are also available with at least equivalent risk of short-term mortality[33,34] and some evidence of superior pulmonary artery growth than is the case with operative shunts.[35,36] The current data raise the possibility that reductions in short-term morbidity that may be achieved with initial palliation might also

Table 4.   **Association Between Census Region and Resource Utilization Adjusted for Race and Maternal Education**

	Northeast (N=28)	Midwest (N=68)	South (N=131)	West (N=42)	P Value
Hospitalizations for operative correction					
Total length of stay	6.9 (4.8–9.8)	8.9 (6.6–11.9)	9.5 (7.5–12.1)	8.1 (6.0–11.1)	0.19
Cost ($)	55 684 (38 911–79 689)	64 645 (47 990–87 081)	82 155 (64 100–105 296)	98 679 (71 944–135 349)	0.002
Subsequent hospitalizations					
Total length of stay	3.8 (1.26–11.2)	3.4 (1.5–7.4)	10.8 (5.6–20.7)	3.5 (1.5–7.8)	0.0002
Cost ($)	15 116 (4719–48 417)	16 648 (7053–39 300)	37 086 (18 494–74 368)	19 369 (8153–46 018)	0.05
Total outpatient visits					
N	19.8 (14.6–26.8)	16.7 (13.0–21.5)	21.9 (17.7–27.1)	17.0 (13.1–22.0)	0.01
Cost ($)	7143 (5063–10 075)	4453 (3353–5914)	5834 (4615–7374)	6122 (4556–8226)	0.008
Cardiac outpatient visits					
N	6.7 (4.8–9.2)	5.0 (3.8–6.6)	6.3 (5.0–7.9)	5.6 (4.2–7.5)	0.10
Cost ($)	332 (197–589)	287 (181–454)	308 (214–561)	340 (206–561)	0.85
Noncardiac outpatient visits					
N	12.3 (8.3–18.1)	10.7 (7.8–14.8)	14.2 (10.9–18.5)	10.5 (7.6–14.6)	0.05
Cost ($)	6820 (4791–9709)	4100 (3063–5489)	5482 (4306–6979)	5696 (4207–7712)	0.005
Emergency department visits					
N	3.1 (1.6–5.9)	3.0 (1.8–5.1)	2.1 (1.3–3.2)	1.4 (0.8–2.5)	0.03
Cost ($)	1159 (543–2477)	1331 (707–2506)	2402 (1332–4332)	1102 (537–2264)	0.01

Estimate (95% CI).

be associated with reduced resource utilization in the medium term. The relative merits of these strategies could not be evaluated in this study, nor could their effect(s) on resource utilization. Future studies should evaluate the effects of these choices not only on perioperative outcomes, but also on medium-term morbidity and resource utilization. It is important to measure the impact of these decisions beyond operative mortality, since early morbidity appears to persist into childhood.

It is not possible in this study to connect prolonged LOS and subsequent resource utilization with deficits in health and well-being from the perspective of the patient. These measurements are not available in medical records, much less insurance claims data. Measurement of health-related quality of life, functional status, and other patient-reported outcomes would be helpful in understanding the degree to which differences in resource utilization reflect deficits in patient experience or reflect subclinical disease. This highlights the importance of including patient-reported outcomes in future prospective studies.

Variation in resource utilization and cost provide evidence of potential to improve the value of care delivery (by reducing costs and improving outcomes). Understanding at what level these differences are manifest is important to direct efforts to improve the value of care delivered. Systematic geographic differences in the cost of congenital cardiac surgery have been demonstrated.[37,38] In hypoplastic left heart syndrome, specifically, significant regional variation in both LOS and costs have been demonstrated, though the explanation for these differences was not clear.[39] There is also some evidence that practice patterns in other areas of congenital cardiology vary by geographic region.[40–42] To our knowledge, regional variation in cost of care of children with TOF has not been evaluated previously. The current study did not demonstrate differences in LOS after operative correction, but did demonstrate significant differences in cost for that hospitalization and in both the amount and cost of subsequent inpatient and outpatient care. These associations were noted despite limitations, specifically, a relatively small sample size and unmeasured confounding because of data that are not recorded in the current database. Census regions are broad, including multiple regions and hospital settings. Further research using more granular geographic distinctions and hospital characteristics will be important to study this question. Some possible explanations include broad regional differences in wages and prices. Importantly, however, it is critical to disentangle differences in resource utilization caused by these changes (as well as those caused by differences in case-mix), both of which are not generally modifiable, and those caused by differences in patterns of practice or quality of care/outcomes, which are more modifiable. Previous studies using hospital-based administrative data have shown how regional wage–price indices can be used to "control" for these local variations to isolate variation in cost,[14–16,22,28,43,44] but further work is necessary to effectively control for case-mix and identify specific modifiable factors that influence resource utilization.

As noted, the study population has higher socioeconomic status than the population at large. It is not possible in this study to evaluate whether this has an effect on resource utilization. Though several studies have evaluated the association between insurance status and the risk of mortality after cardiac surgery, the results have been equivocal.[14,22,38,45–48] There is evidence that lack of insurance is associated with increased risk of adverse outcome for neonates undergoing heart surgery during and after initial hospitalization.[45] However, to our knowledge, there is no evidence that there is an analogous association between receipt of Medicaid and the risk of adverse events. Further studies incorporating recipients of both Medicaid and commercial insurances are necessary to address this question. At the same time, a critical part of any such study is to effectively evaluate how interconnected factors (insurance, race, ethnicity, income, and education) may contribute to these issues.

There were several other limitations to this study. Claims data provide a novel means of accessing longitudinal resource utilization data from a geographically representative cohort of children undergoing operative correction of TOF. However, not all clinically relevant data are available (eg, details of preoperative anatomy and any residual anatomic lesions postoperatively), which raise the possibility of unmeasured confounding. A specific concern noted previously is that mortality data are not available. As a result, bias because of the omission of the small percentage of patients who die within 2 years of their incident hospitalization could not be addressed. The data presented provide a best estimate of resource of patients surviving at least 2 years. For inpatient admissions, attributing costs to cardiac or noncardiac issues is also not possible, limiting the inferences possible in this study. Finally, over time as care of TOF continues to advance, differences in resource utilization may continue to evolve and deserve continued attention.

CONCLUSIONS

This study is the first to define medium-term resource utilization in children undergoing operative correction of TOF. It demonstrates that resource utilization is

increased over 2 years among patients who have prolonged LOS during their initial hospitalization. Future research should focus on determining whether higher resource utilization is also associated with decreased health and well-being, and whether strategies to reduce morbidity in the incident hospitalization also reduce resource utilization during subsequent follow-up. Together this would both improve outcomes for patients with TOF and deliver higher value care.

ARTICLE INFORMATION

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Disclosures

None.

Supplementary Materials

Tables S1–S2

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SUPPLEMENTAL MATERIAL

Table S1. Characteristics of survivors after operative correction of tetralogy of Fallot with and without 2 years of continuous insurance claims data.

	Continuous enrollment (n=269)	<2 years of continuous enrollment (n=212)	*p*
Male sex	53% (143)	54% (114)	0.89
White race (vs. other races)	65% (176)	64% (136)	0.99
Age at operative correction (years)			0.68
0	49% (133)	53% (113)	
1	45% (121)	42% (82)	
2	6% (15)	5% (10)	
Census regions			0.95
Midwest	25% (68)	27% (58)	
Northeast	10% (28)	9% (20)	
South	49% (131)	48% (101)	
West	16% (42)	33% (16)	
Highest parental education level			0.73
<12th grade	1% (3)	1% (2)	
High school diploma	18% (48)	16% (33)	
Bachelor's degree	46% (125)	49% (104)	
Graduate degree	33% (89)	29% (62)	
Unknown/missing	1% (4)	5% (11)	
Family annual income			0.75
<$40K	6% (15)	7% (14)	
$40-49,000	5% (13)	5% (11)	
$50-59,000	5% (13)	5% (10)	
$60-74,000	6% (16)	8% (16)	
$75-99,000	13% (35)	11% (24)	
>$100,000	42% (112)	33% (71)	
Unknown/missing	24% (65)	31% (66)	
Insurance type			0.90
Point of service	70% (187)	70% (148)	
Exclusive provider organization	13% (36)	13% (28)	
Health maintenance organization	12% (33)	14% (29)	
Preferred provider organization	4% (12)	3% (6)	
Other	0.4% (1)	0.5% (1)	
Length of stay of hospitalization for operative correction (days)	7 (IQR: 5,11)	7 (IQR: 5,11)	0.76

Table S2. Observed differences in resource utilization between census regions.

	Northeast (N=28)	Midwest (N=68)	South (N=131)	West (N=42)	*p*
Hospitalizations for operative correction					
Total length of stay	6 (4, 9)	7 (5, 11)	7 (4, 11)	7 (6,12)	0.20
Cost ($)	52407 (38480, 73204)	58362 (43672, 75724)	58362 (38480, 91498)	58362 (52407, 95651)	0.16
Subsequent hospitalizations					
Total length of stay	2.5 (1, 11)	4 (2.5 9)	8 (3,18)	4 (2, 11)	0.046
Cost ($)	16,598 (4612, 55654)	19790 (9177, 34312)	36892 (16431, 126160)	34877 (7849, 75892)	0.14
Total outpatient visits					
N	24.5 (15, 35)	18.5 (13, 26)	21 (13, 34)	18 (14, 25)	0.16
Cost ($)	6943 (4587, 11286)	4492 (2635, 7023)	5315 (3618, 8029)	4970 (3454,8198))	0.01
Cardiac outpatient visits					
N	7 (5-8)	5 (3, 8)	6 (4, 8)	6 (4,8)	0.05
Cost ($)	420 (196, 588)	340 (136, 605)	309 (153, 603)	309(153, 603)	0.64
Noncardiac outpatient Visits					
N	14.5 (8.5-28.5)	13 (7.5, 22)	15 (7, 24)	12 (7, 20)	0.28
Cost ($)	6676 (4384, 10654)	3936 (2329, 6338)	4913 (3178, 7321)	4783 (2999, 7674)	0.005
Emergency department visits					
N	1 (0,3)	2 (1,3)	1 (0,2)	0 (0,1)	0.0003
Cost ($)	1029 (656, 3289)	1158 (496, 2031)	1540 (577, 3667)	991 (400, 3573)	0.28

N (IQR)