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Costs, Mortality and Hospital Utilization in Relation to Prenatal Diagnosis in d-Transposition of the Great Arteries

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Abstract

Background—The impact of prenatal diagnosis of d-transposition of the great arteries (dTGA) on health care utilization is largely unknown. We evaluated a population based cohort to assess costs, mortality and inpatient encounters by whether dTGA was prenatally diagnosed or not.

Methods—dTGA cases (born 1997–2011) identified at the Utah Birth Defect Network (UBDN), which includes data on timing of diagnosis, were linked to statewide inpatient discharge data. We excluded preterm cases or with additional major heart defects. We evaluated hospitalizations and costs for infants (1st year of life) and mothers (10 months prior to birth) using multivariable models adjusted for demographic and clinical risk factors

Results—Of 119 cases, 14 (12%) were prenatally diagnosed. Birth weight, surgical complexity and extracardiac defects/syndromes were similar between groups. Of 7 deaths (6%), two occurred pre-intervention in postnatally diagnosed infants. Prenatal diagnosis was associated with more in-hospital days (estimate 13 additional days, $p=0.03$) and higher mean costs for mothers (\$4,141 vs \$12,148) and infants (90,419 vs \$49,576). Prenatal diagnosis independently predicted higher adjusted costs for the overall cohort (\$22,570, $p=0.045$). After excluding deaths, total costs were no longer significantly different.

Conclusions—Mothers of prenatally diagnosed infants with dTGA had higher inpatient costs compared to those postnatally diagnosed. Costs trended towards higher for their infants though were not significantly different. Linkage of population-based surveillance systems and outcome databases can be a powerful tool to further explore the complex relationship of prenatal diagnosis to costs and outcomes in other types of CHDs.

Keywords

congenital heart disease; prenatal diagnosis; costs

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Introduction

With advances in cardiac care, survival of infants with complex heart defects has improved significantly over the last decades.(Nieminen and others, 2001) However, many infants with complex lesions still require lifelong care, resulting in high health care expenditures. (Waitzman and others, 1996) The Health Care Cost and Utilization Project estimated that in 2004 alone, congenital heart disease (CHD) was responsible for 1.4 billion dollars in health care costs in the United States, over half of costs attributed to all birth defects combined. (Russo and Elixhauser, 2007) Costs often begin with prenatal screening and diagnosis. However, the impact of prenatal diagnosis of CHD on health care costs is still unclear: healthcare costs may decrease, for example if prenatal diagnosis leads to fewer emergent transports (Jegatheeswaran and others, 2011) and decreases morbidity associated with later diagnosis.(Bonnet and others, 1999; Mahle and others, 2001; Sivarajan and others, 2009; Tworetzky and others, 2001) However, overall costs could also increase because of the additional prenatal procedures and planned deliveries at tertiary centers.

Evaluating the magnitude and determinants of such costs can help to identify ways in which to deliver care more efficiently while optimizing outcomes. Evaluating costs related to timing of diagnosis, and specifically to prenatal diagnosis, is challenging, for several reasons, including the interplay of factors such as anatomic complexity (severity of the CHD and presence of extra-cardiac anomalies) and other unidentified characteristics that bias towards prenatal diagnosis. Additional challenges in such studies include censoring due to losses to follow-up or death, and access to longitudinal, population-based information. The integrated datasets available in Utah provide a valuable opportunity to comprehensively assess the relation between prenatal detection of CHD and health care utilizations, specifically in-patient costs, from the prenatal period and extending beyond the surgical hospitalization. A similar methodology for linking birth registries with state hospital data have been described by Salemi et al (Salemi and others, 2013b) and have provided valuable information on population costs and outcomes in other birth defects such as oral clefts (Bell and others, 2016a; Bell and others, 2016b; Cassell and others, 2008; Razzaghi and others, 2015). However, similar information on CHDs is still lacking, particularly in relation to prenatal diagnosis and incorporating maternal as well as infant costs.

In this study we linked the population-based birth defect surveillance registry with administrative databases in Utah to evaluate the relationship of prenatal diagnosis with inpatient costs and length of stay among a homogeneous, population-based cohort of children with d-transposition of the great arteries (dTGA).

Methods

We conducted a population-based retrospective cohort study of children with dTGA, live born from 1997 through 2011 to mothers who were Utah residents at the time of the child's birth. dTGA was selected because of its requirement for early intervention and its relative clinical homogeneity (low rate of associated extra-cardiac defects and syndromes and low rate of preterm birth).

The cohort was identified through the Utah Birth Defect Network (UBDN), a state-wide population-based program that actively monitors major birth defects among all pregnancy outcomes (live births, stillbirths and pregnancy terminations) in Utah residents. dTGA has been an eligible defect in the UBDN since 1997. Trained abstractors from the UBDN actively ascertain cases through multiple sources (prenatal and postnatal); the program also uses additional (passive) reporting sources.(Feldkamp and others, 2005) State law mandates hospitals and laboratories to report cases. The combination of active case ascertainment with the use of multiple sources result in a high level of case ascertainment. Data are abstracted from medical records on cases diagnosed from 20 weeks gestation to 2 years of age. For the study cohort, potential cases were reviewed within the UBDN by clinicians with training in medical genetics and pediatric cardiology to ensure accurate coding and classification. Data collected by the UBDN include maternal and infant demographic data and details on diagnoses and outcomes, including timing of diagnosis.

For this study we selected term (>36 weeks gestation) live born infants with simple dTGA (Center for Disease Control/British Pediatric Association (BPA) codes 745100, 745110 or 745190). To minimize the effects of comorbid conditions and decrease the likelihood of differences in severity of cases between pre and postnatal diagnoses, all eligible cases were reviewed by a single pediatric cardiologist (NP) to exclude those with additional major heart disease (e.g. double outlet right ventricle, tricuspid atresia, atrioventricular canal, single ventricle, and total anomalous pulmonary venous return).

Data on all longitudinal inpatient hospitalizations were obtained through linkage with the Utah Population Database (UPDB). The UPDB is a unique resource that contains longitudinal vital statistics as well as administrative records and health records at the individual and family levels for residents of the state.(DuVall and others, 2012; Kirchhoff and others, 2014) The health records include inpatient hospital discharge abstracts for all hospitals in the state. A deterministic linkage (by birth certificate number or demographic data), maintained by the UPDB, links mothers and infants to the UPDB. Data for all inpatient hospitalizations were retrieved to obtain lengths of stay, associated diagnoses and procedures, and total charges. Survival status was also obtained from vital records.

The primary study outcome were inpatient costs (total as well as separately for mother and child) accrued statewide. Secondary study outcomes were mortality and inpatient hospital utilization (total hospital days and total hospital encounters). Mortality was examined in the entire cohort. In analyses of hospital utilization, we excluded patients who did not receive care at Primary Children's Hospital (the only tertiary hospital in the state performing pediatric heart surgery) or those who had more than a standard arterial switch operation with or without VSD closure and/or coarctation repair (such as patients who required more complex intervention for left ventricular outflow tract obstruction (LVOT)). Complexity of surgical repair was graded as follows: arterial switch operation (ASO) only - 0, ASO with VSD repair - 1, ASO with aortic arch repair - 2, and ASO with VSD and arch repair - 3. Postnatal hospital utilization and costs were initially collected through December 2011 but for the purposes of statistical analysis were truncated to the initial 365 days following birth as inpatient hospital encounters beyond this period were rare. To capture prenatal events, maternal hospitalizations and inpatient costs were also determined for the 10 months prior to

the date of the child's birth. The study was approved by the Institutional Review Boards of the University of Utah, UPDB and Utah Department of Health with a waiver of consent.

For the univariate analysis of descriptive data we used the Wilcoxon rank sum (for continuous non-normal data) or Fisher's exact test (for dichotomous data). We evaluated mortality with Kaplan-Meier curves. We evaluated total hospital days and encounters with linear regression models adjusted for child weight, gestational age, maternal age at delivery, maternal race, year of birth, complexity of surgery as reflective of anatomic complexity (collapsed into simple ASO vs all others), presence of additional non-cardiac defects/syndromes, type of insurance, population education and income. Maternal address at delivery was used to estimate socioeconomic status, based on variables from the 2010 Census. The University of Utah's Department of Geography's Digitally Integrated Geographic Information Technologies (DIGIT) lab provided these measures by census tract from which we used geographic median income or % of census tract below poverty level. These variables were chosen apriori as potential confounders previously associated with higher resource utilization.(Connor and others, 2005; Ungerleider and others, 1997) Co-morbidities that occur after delivery were not used in models as they may be influenced by the timing of diagnosis. The distribution of fit was examined and models were transformed as necessary.

Cost data was obtained by applying hospital specific cost to charge ratios reported to the Center for Medicare and Medicaid Services (CMS) and were adjusted to 2012 dollars using the Consumer Price Index. Costs were modeled using a generalized linear regression with gamma distribution and a log link function, adjusting for the same covariates listed above. To assess the robustness of the analyses of costs and total LOS we performed sensitivity analyses looking at the following three groups: 1) only patients who had cardiac surgery since their resource use would be expected to be different from those who never underwent surgery, 2) only patients without additional major congenital defects to alleviate any differences in cohorts that may be related to this and 3) only patients who survived through the first 365 days after birth (since deaths many have a varying influence on costs that be can be difficult to interpret collectively). For all the analyses, we used Stata 12 (Stata Corp, College Station, Texas).

Results

From the initial cohort of 130 live born infants with dTGA born from 1999 through 2011 ascertained using BPA codes, we excluded 11 who were found to have associated major CHD, for a final study sample of 119 cases. The livebirth prevalence of dTGA for this time period was 25 per 100,000 live births. There were no terminations of pregnancies prenatally diagnosed with this condition. All infants were successfully linked to inpatient discharge abstracts, though one did not link beyond the birth admission (linkage success 99%). In 14 infants (11.7%) dTGA was prenatally diagnosed. Prenatal and postnatal diagnosis groups were similar in demographic and socioeconomic factors (Table 1).

Seven infants with dTGA died (5.8%), all within the first 120 days of life (DOL). Mortality did not differ between the prenatal and postnatal diagnosis groups (7.1% (1/14) vs. 5.7%,

(6/105) respectively, $p=0.86$). However, 2 of the 5 deaths in the postnatal diagnosis group occurred prior to any intervention. The other 4 deaths occurred after ASO, 3 from postoperative complications (DOL 7 to DOL 60) and 1 sudden unexplained death at home. The single death in the prenatally diagnosed group occurred after ASO at DOL 120 after a lengthy hospitalization secondary heart failure from a coronary complication and being listed for heart transplant.

The analyses of morbidity included the 110 patients (92% of the cohort) who were hospitalized and had an ASO in Utah after excluding the 2 pre-intervention deaths, 3 patients who had surgery out of state and 3 patients who required more complex surgery for LVOT obstruction. An additional 2 patients did not have cost to charge data available for their birth facility. Almost all hospitalizations (82%) and hospital days (>90%) occurred in the first year of life, so subsequent analyses focused on this time period. Compared to the postnatal diagnosis group, the prenatal diagnosis group experienced more hospital days (mean 29.7 vs. 22.8 days) though this was not statistically significant on univariate comparison ($p=0.44$). In the multivariable analysis, prenatal diagnosis was independently associated with a longer surgical and total LOS (estimate of 12.0 and 12.7 additional days respectively) (Table 2). Less complex surgical intervention, private insurance and older maternal age were independently associated with fewer hospitalizations. Birth weight, gestational age and the presence of additional defects were not associated with hospitalizations or hospital days. There was no significant difference in preoperative LOS.

Data were available for 115 mothers for their delivery hospitalization in the UPDB (111 in postnatal diagnosis and 14 in prenatal diagnosis groups). No mother had >1 inpatient hospitalization during their pregnancy. Among those without a record of a delivery hospitalization, 3 were home births and 1 had no record related to delivery found by linkage. In the prenatal diagnosis group, caesarean section was more frequent (27% vs 17%, $p=0.41$) and maternal LOS for delivery was longer (mean 2.3 vs. 1.2 days). In multivariable analysis prenatal diagnosis was the only independent predictor of longer maternal LOS by 1.1 days (Table 2).

Compared to postnatal diagnosis, prenatal diagnosis was associated with an 86% increase in adjusted mean total inpatient hospital costs (Figure 1) (median \$90,419 vs \$49,576). In the multivariable analysis, prenatal diagnosis was an independent predictor of higher total costs (increasing costs by \$22,570, Table 3) and maternal costs ($\uparrow$ \$1,691). Older maternal age and increased severity also predicted higher costs for the overall cohort. When considering child costs alone, prenatal diagnosis had higher costs, but this did not reach statistical significance. The only factor associated was maternal age.

In the sensitivity analyses, prenatal diagnosis remained the only independent predictor of total costs when considering only those who underwent surgical intervention, but was no longer associated when considering only the subset without extracardiac anomalies/syndromes (data not shown). After excluding deaths (Table 3), maternal costs remained significantly higher (\$1,673, $p=0.001$), but total and child costs no longer differed between the groups. The only factors associated with increased costs were identified in survivors for total costs were severity and older maternal age. Child LOS also no longer was significantly

different between groups in survivors in multivariable modeling (prenatal diagnosis increase of 4.5 days, $p=0.37$).

Discussion

In this population-based study of infants with well-characterized, simple dTGA we found that prenatal diagnosis was associated higher total health care costs but this excess cost did not persist once deaths were removed. While mean child inpatient costs were higher in the prenatally diagnosed group, prenatal diagnosis was not significantly associated with cost in after adjusting for risk factors. Finally, prenatal diagnosis was associated (not unexpectedly) with higher prenatal maternal inpatient costs and length of hospital stay; such maternal costs were but a small fraction of the combined costs (maternal and child costs).

These findings add to the developing body of evidence on the relation between prenatal diagnosis and health and economic outcomes of children with congenital heart disease. Unfortunately, the findings are inconsistent across different studies, possibly due to differences in study populations (e.g., types of CHDs included), methods (single institution vs. population-based) and data sources. For example, in a single-center study, prenatally diagnosed patients had higher associated costs compared to postnatally diagnosed patients, but only in those who required univentricular palliation. (Copel and others, 1997) The lack of clear evidence of lower costs with prenatal diagnosis may seem surprising especially in light of the reported lower morbidity in prenatally diagnosed infants with dTGA, (Bonnet and others, 1999; Fuchs and others, 2007). However, other studies were not unable to document such lower morbidity (Kumar and others, 1999; Peterson and others, 2013), and perhaps such differences in morbidity may be insufficient to drive detectable changes in gross measures of resource utilization such as LOS and total hospital costs.

The impact of timing of diagnosis on the measured outcomes in dTGA may be more challenging to assess than in other complex CHD. In dTGA patients who are undiagnosed prior to delivery, the diagnosis is nearly always made (90% or higher) in the first 24-48 hours after birth and prior to discharge from the birthing center, (Peterson and others, 2013), thus likely limiting the adverse effects of delayed diagnosis. In the era of newborn screening for critical congenital heart defects, the marginal advantage of prenatal diagnosis specifically for dTGA may be even smaller in some settings such as urban, well equipped hospitals, but could be significant in more rural settings and smaller birthing centers located away from advanced medical resources. Further study of how these findings may differ in other types and severity of CHD is necessary.

In this study we evaluated both maternal and child hospitalizations and costs. The increased maternal delivery LOS and costs in the prenatally diagnosed cohort may be related to the higher number of planned deliveries. Such deliveries are more often induced than spontaneous. (Raboisson and others, 2009) Whereas this may be expected, understanding how costs are allocated may help direct efforts to reduce resource use, while optimizing perinatal care and outcomes. Nonetheless, inpatient costs associated with maternal care are dwarfed by those expended on the postnatal care of affected infants which did not differ

significantly related to timing of diagnosis. Thus, if prenatal diagnosis led to even small improvements in outcomes for children, the overall benefit would likely be considerable.

The overall event rate for mortality in our study was quite low, consistent with excellent overall survival of infants with dTGA in the current era.(Hutter and others, 2002; Wong and others, 2008). The relation of prenatal diagnosis with mortality is unclear in published studies. In general, some studies suggest improved survival with prenatal diagnosis, whereas others find no difference.(Blyth and others, 2008; Bonnet and others, 1999; Khoshnood and others, 2005; van Velzen and others, 2014) A recent meta-analysis found that compared to postnatal diagnosis, prenatal diagnosis was associated with lower mortality when specifically examined prior to any intervention.(Holland and others, 2015). In our study, of the 6 deaths in the postnatal diagnosis group, two were possibly preventable (child undiagnosed, one at home, with deaths prior to intervention). Evidence for potentially preventable deaths in babies with dTGA after birth has been documented in some studies. (Blyth and others, 2008; van Velzen and others, 2014) Additionally, the single infant who was prenatally diagnosed and ultimately died was the longest survivor amongst those who eventually died (120 days).

Our study has several strengths and limitations. Focusing on dTGA allowed us to minimize the effect of major confounders such as concurrent comorbidities, prematurity, and the presence of extracardiac defects or syndromes, which are all relatively uncommon in dTGA. Babies with dTGA also typically require a single major surgical hospitalization, unlike the more complex critical CHDs, further improving our ability to examine the impact of timing of diagnosis in a clinically important and severe congenital heart disease. However, the low rate of prenatal diagnosis in DTGA (similar to that previously reported by others) (Friedberg and others, 2009; Khoo and others, 2008; Peterson and others, 2013) impaired the statistical power of the study (few infants in the prenatally diagnosed cohort), and may have increased the influence of outliers. Although we controlled for several potential confounders (including severity), unmeasured confounding may have reduced the differences in outcomes (morbidity, mortality, costs). Additionally, exclusions of additional heart disease, prematurity, and surgeries that were more complex than an arterial switch, as well as adjustment for anatomic variants that required intervention such as a VSD or coarctation, minimized that likelihood of severity confounding the association of timing of diagnosis with the outcomes of interest.

Another factor that may have made it more difficult to identify differences in severity by timing of diagnosis was the exclusion of preterm babies; however, preterm birth is rare in babies with dTGA (<10% in our data), so the effect of this restriction is likely small. With respect to cost outcomes, we were able to access data on total inpatient costs only, and the cost to charge ratios were limited to facility rather than department level which may be less precise.(Salemi and others, 2013a) Additionally, we limited comparison of resource use to only children that had surgery in our state. Since such surgeries occur at a single institution in Utah and associated surgical costs far exceed newborn hospitalization costs, this may limit generalizability. Some costs, such as transport costs, were not included as they typically are not part of the hospital charge. We elected to disregard transport costs as these would be based on estimates rather than actual charges and would require different

methodology; also, most CHD-related health care costs are related to inpatient costs, (Boulet and others, 2010). Future studies can provide more comprehensive estimates by including these additional cost centers.

The main strengths of the study were its population-based design and the inclusion of a well-characterized, homogeneous phenotype (simple dTGA). Importantly, cases were identified by active rather than passive ascertainment, providing a more complete and representative cohort of affected children. It has been estimated that passive case ascertainment can miss 15% or more of cases of birth defects. (Salemi and others, 2012) All cases were reviewed by clinicians with expertise in pediatric cardiology, based not only on codes but extensive abstraction of medical records with verbatim descriptions. We were also able to leverage data from multiple data bases connected by established and validated linkages. All these factors were valuable in minimizing the limitations typically associated with the use of administrative or claims databases, which rely exclusively on diagnosis codes with limited or no possibility of independent validation, and with limited ability to track patients longitudinally. (Boulet and others, 2010; Pasquali and others, 2013; Russo and Elixhauser, 2007). Other studies have leveraged robust birth defect surveillance to evaluate population level outcomes in birth defects such as orofacial clefts (Bell and others, 2016b; Cassell and others, 2008; Razzaghi and others, 2015), though this is the first we are aware of that focuses on severe CHDs. We were able to take advantage of existing deterministic linkages (Salemi and others, 2013b) and active rather than a passive birth defect surveillance system. We did not rely on subjects falling under the auspices of one single payer such as Medicaid, so that we could provide more reliable estimates of population costs. Additionally, the UBDN is one of a handful of birth defect registries that includes data on prenatal diagnosis which allowed for investigation of this particular exposure in particular. Finally our linkage allowed for investigation of child as well as maternal costs.

In summary, in this well-defined population-based cohort we found that prenatal diagnosis was not associated with lower costs or mortality in children with dTGA. Maternal inpatient costs were higher, though such costs were small compared to the costs accrued by the affected children. The effects of prenatal diagnosis on health and economic outcomes are less obvious and clearly more complex to assess than anticipated. For more complex CHDs, the findings may well be different. However, longitudinal linkage of population-based surveillance systems of well characterized phenotypes with rich outcome databases will allow us to continue further detailed analysis of the components that contribute to increased health care costs. Such studies should be pursued in order to generate the evidence to direct how we may optimize the diagnosis and treatment of CHD in a manner that also improves the value of the care we deliver.

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Figure 1.

Comparison of inpatient hospital cost between postnatally and prenatally diagnosed cohorts of livebirths with d-transposition of the great arteries and their mothers for a) overall cohort, b) livebirths without extracardiac anomalies/syndromes and c) surviving infants only. Box and whisker plots illustrate total costs (blue), maternal costs (red) and child costs (green). Y axis is in dollars. Solid line is median, box represents interquartile range, whiskers are adjacent values (1.5 times the IQR), and dots are outliers. Y axis is in dollars.

Table 1

Maternal and infant characteristics by timing of diagnosis

	Prenatally diagnosed, n=14 N (%)	Postnatally diagnosed, n=105 N (%)	p value ^c
Individual Level			
Year of Birth			
1997-2002	4 (29)	59 (56)	0.08
2003-2009	10 (71)	46 (44)	
Male	10 (71)	77 (73)	1.0
Gestational Age <37 weeks	1 (7)	3 (3)	0.4
Mean Gestational Age (weeks)	38.6	38.8	0.67
Mean Birth Weight (Kg)	3.2	3.4	0.31
Extracardiac Anomalies/syndromes	1 (7)	10 (10)	1.0
Balloon Atrial Septostomy	6 (43)	67 (64)	0.15
Surgical complexity (n=107)			
Arterial switch operation (ASO) only	8 (67)	68 (72)	0.29
ASO + VSD repair	2 (17)	22 (23)	
ASO + arch repair	1 (8)	1 (1)	
ASO + VSD and arch repair	1 (8)	4 (4)	
Maternal Age (years)	28.1	26.4	0.17
Maternal Race White Non-Hispanic	13 (93)	88 (84)	0.69
Maternal Education (< high school)	0 (0)	11(11)	0.35
Public Payer Insurance	4 (29)	25 (27)	1.0
Follow Up (months)	57	85	0.04
Geographic Level^a			
Median Income	\$91,957	\$54,927 ^b	0.3

^aBased off of U.S. 2010 Census Data for the mother's census tract based on geocoded address at birth.^bOne patient (1%) in the postnatal group could not be geocoded and thus lacked Census Data variables.^cWhen there are less than 5 cases in any category we used Fisher's exact test to compute p-value.

Table 2

Total hospitalizations, surgical length of stay and total hospital days in first year of life for liveborn (Utah 1997-2009) children with dTGA and total maternal hospital days in 10 months prior to giving birth to affected child. Estimates from multivariable linear regression models.

Factor	Child						Mother	
	Hospitalizations ^a		Hospital Days ^b		Surgical Hospital Days ^b		Hospital Days ^b	
	Estimate	95% CI	Estimate	95% CI	Estimate	95% CI	Estimate	95% CI
Intercept								
Prenatal Diagnosis	−0.1	−0.6, 0.5	12.7	1.01, 24.5	12.0	1.2, 22.8	1.1	0.5, 1.6
Birth Weight (g)	0.002	−0.002, 0.007	−0.001	−0.01, 0.01	−0.002	−0.01, 0.01	0.0002	−0.0002, 0.001
Gestational Age	0.02	−0.1, 0.2	1.2	−2.5, 4.5	2.1	−1.0, 5.2	−0.1	−0.3, 0.1
Additional Defect	0.01	−0.6, 0.6	−4.9	−18.1, 8.3	−4.2	−16.4, 8.0	−0.3	−1.1, 0.4
Year of birth	−0.04	−0.1, 0.01	−0.1	−1.1, 0.9	0.4	−0.6, 1.3	−0.03	−0.08, 0.01
Severity ^c (complex vs simple)	0.96	0.6, 1.4	5.2	−2.9, 13.4	−0.4	−7.9, 7.1	0.2	−0.2, 0.6
Maternal age	−0.04	−0.08, −0.0041	0.4	−0.4, 1.2	0.6	−0.2, 1.3	0.01	−0.03, 0.04
Private Insurance	−0.7	−1.2, −0.3	−6.5	−15.5, 2.6	−4.5	−12.9, 3.9	0.2	−0.1, 0.6
Median household income (per \$1,000)	0.001	−0.002, 0.004	−0.04	−0.1, 0.03	−0.05	−0.11, 0.02	−0.001	−0.01, 0.01

*Adjusted for gender, maternal education and maternal race in addition to variables presented.

^a Model estimates represent a unit increase in number of hospitalizations

^b Model estimates represent a change in hospital days

^c Complex (categories 1-3) versus simple (category 0) arterial switch surgical repair

Table 3

Multivariable generalized linear regression (log-link gamma function) for total costs, maternal costs and child costs in the first year of life for total cohort. All costs were adjusted to 2011 dollars using the Consumer Price Index.

Variable	Total costs ^c			Maternal costs ^c			Child Costs ^c		
	Estimate (\$)	P value	Survivors Only	Estimate (\$)	P value	Total Cohort	Estimate (\$)	P value	Survivors
Prenatal diagnosis	22,570	0.045	11,893	1,691	0.001	1,673	20,738	0.07	10,349
Birth Weight (grams)	-11	0.21	-6	\$0	0.66	0	-11	0.25	-5
Gestational age (wks)	2,152	0.51	1,264	-188	0.22	-206	2,157	0.52	1,242
Additional defects	-13,212	0.28	-7,238	-188	0.11	813	-14,499	0.24	-8,374
Year of birth	1,585	0.09	1,406	-25	0.54	-31	1,639	0.09	1,475
Severity (complex vs simple) ^b	15,860	0.048	12,870	244	0.49	-236	15,680	0.05	12,736
Private insurance	-2,261	0.78	-4,983	136	0.69	187	-2,274	0.78	-5,049
Maternal age (ref<20 yrs)	476	0.96	4,369	-237	0.52	-215	937	0.92	4,628
20-30	14,808	0.19	18,259	721	0.12	708	14,345	0.21	17,660
>30	49,158	0.02	-3,066	602	0.47	543	48,994	0.02	-3,592
Pseudo R squared	0.006		0.004	0.008		0.008	0.007		0.004

* Adjusted for gender, maternal education, maternal race and geographic poverty level in addition to variables presented. Models were done using tertiary facility as fixed and random effect. Results presented reflect those as a random effect though the results were similar in both cases.

^aPer \$1,000 except for birth weight

^bComplex (categories 1-3) versus simple (category 0) arterial switch surgical repair

^cEstimates represent regression adjusted differences in cost