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Trisomy 18: disparities of care and outcomes in the State of Texas between 2009 and 2019

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Abstract

Objective: To perform a statewide characteristics and outcomes analysis of the Trisomy 18 (T18) population and explore the potential impact of associated congenital heart disease (CHD) and congenital heart surgery. *Study Design:* Retrospective review of the Texas Hospital Inpatient Discharge Public Use Data File between 2009 and 2019, analysing discharges of patients with T18 identified using ICD-9/10 codes. Discharges were linked to analyse patients. Demographic characteristics and available outcomes were evaluated. The population was divided into groups for comparison: patients with no documentation of CHD (*T18NoCHD*), patients with CHD without congenital heart surgery (*T18CHD*), and patients who underwent congenital heart surgery (*T18CHS*). *Results:* One thousand one hundred fifty-six eligible patients were identified: 443 (38%) *T18NoCHD*, 669 (58%) *T18CHD*, and 44 (4%) *T18CHS*. *T18CHS* had a lower proportion of Hispanic patients (n = 9 (20.45%)) compared to *T18CHD* (n = 315 (47.09%)), and *T18NoCHD* (n = 219 (49.44%)) ($p < 0.001$ for both). Patients with Medicare/Medicaid insurance had a 0.42 odds ratio (95%CI: 0.20–0.86, $p = 0.020$) of undergoing congenital heart surgery compared to private insurance. *T18CHS* had a higher median total days in-hospital (47.5 [IQR: 12.25–113.25] vs. 9 [IQR: 3–24] and 2 [IQR: 1–5], $p < 0.001$); and a higher median number of admissions (n = 2 [IQR: 1–4]) vs. 1 [IQR: 1–2] and 1 [IQR: 1–1], ($p < 0.001$ for both). However, the post-operative median number of admissions for *T18CHS* was 0 [IQR: 0–2]. After the first month of life, *T18CHS* had freedom from in-hospital mortality similar to *T18NoCHD* and superior to *T18CHD*. *Conclusions:* Short-term outcomes for *T18CHS* patients are encouraging, suggesting a freedom from in-hospital mortality that resembles the *T18NoCHD*. The highlighted socio-economic differences between the groups warrant further investigation. Development of a prospective registry for T18 patients should be a priority for better understanding of longer-term outcomes.

Trisomy 18 (T18) or Edwards syndrome is the second most common autosomal trisomy in live births, only preceded by trisomy 21 (T21).[1–3] Its reported prevalence ranges between 0.96 and 1.12 per 10,000 live births.[3] T18 is associated with several anomalies and neurodevelopmental abnormalities that frequently lead to a poor prognosis with a reported median survival of 6–15 days.[1,3,4] Up to 85% of T18 patients have an associated congenital heart disease (CHD), with the most common cardiac anomalies being atrial septal defect (ASD), ventricular septal defect (VSD), patent ductus arteriosus (PDA), and multivalvular disease.[5–7] Nevertheless, the role of these malformations on early mortality remains controversial.[5,6]

Because of a predicted poor prognosis, the most frequent care paradigm in the past was to withhold aggressive management measures, such as cardiac surgery.[8] Recently, this approach has been challenged.[9] It has been suggested that aggressive management of associated comorbidities is not always futile, warranting individualised evaluation and consideration of different treatment modalities in selected cases.[2]

Recent publications report the survival of T18 children beyond the first decade of life, even in the presence of significant comorbidities requiring major interventions.[2,10] However, management practices of T18 patients vary significantly between centres, and there is no consensus regarding the decision to pursue interventions in this patient population.[6,8]

This study aims to utilise real-world data of patients with T18 in the state of Texas during the last decade, with special attention to one of the most frequently associated comorbidities, CHD.

Methodology

This is a retrospective analysis of the Texas Hospital Inpatient Discharge Public Use Data File (TIDD) from January 2009 through December 2019.[11] The TIDD is an administrative dataset that

reports de-identified information of hospital discharges in the state of Texas. It is maintained by the Texas Department of State Health Services Center for Health Statistics and collects information from most hospital discharges in Texas. Exempt from reporting are hospitals located in a county with a population less than 35,000, or those located in a county with a population more than 35,000 and with fewer than 100 licenced hospital beds and not located in an area that is delineated as an urbanised area by the United States Bureau of the Census. The reliability of the data has been assessed and confirmed by other researchers in the past.[12]

Patient race, ethnicity, insurance status, age group, length of stay, discharge location, and discharge status were extracted.

The TIDD uses the International Classification of Disease (ICD) to report admitting diagnosis, principal diagnosis and up to 24 additional diagnoses for each discharge. The 9th edition of the ICD (ICD-9) was used from 2009 to through the third quarter of 2015, while the 10th edition (ICD-10) was used from the last quarter of 2015 through 2019. Discharges of T18 patients were identified using the ICD-9/10 diagnostic codes (758.2, Q91.0–Q91.3). Discharges of patients with other coded autosomal trisomy diagnoses (i.e. Trisomy 13, T21) were excluded.

Since the TIDD offers de-identified hospital discharge records, it allows for the possibility of a single person having multiple entries. Attempting to evaluate patients instead of discharges, patient tracking, based on multiple variables, was used for linking discharges. For this, the year and quarter of discharge, age group, zip code, race, ethnicity, insurance information, and diagnostic characteristics were evaluated for consistencies. Two researchers independently evaluated the discharges to determine linkages. The findings were then compared, and discrepancies were discussed, reaching a concordance rate of 98.3%. The discharges in which discrepancies remained were assessed by a third independent reviewer. Direct patient re-identification was never attempted nor intended.

The TIDD divides patient ages by groups (1–28 days of life; 29–365 days of life; 1–4 years; 5–9 years; 10–14 years; 15–17 years, 18–19 years; 20–24 years; and 25–29 years). Although recent publications have identified patients with T18 surviving into adulthood, they remain limited, so all linked records registering an age group of ≥25 years were excluded from this analysis. Patients who had evidence of T18 mosaicism (by the presence of the ICD-10 code Q91.1) at any of their hospital discharges were considered as such and excluded from this study. The presence of CHD was identified using ICD codes.

The TIDD uses ICD procedure codes to report the principal procedure and a maximum of 24 additional procedures for each discharge. Cardiac interventions were extracted using ICD-9/10 procedural codes. Both diagnostic and procedural codes were used to identify the presence of gastrostomy tubes and tracheostomies.

The number of hospital admissions per individual was noted. Length of stay was recorded as total days inpatient, mean duration of each patient's hospitalisations (mean length of stay) and single longest hospitalisation.

Patients were divided into three groups: T18 patients without a CHD diagnosis (*T18NoCHD*); T18 patients with a CHD diagnosis without a record of congenital heart surgery (*T18CHD)*; and T18 patients with a CHD diagnosis who underwent congenital heart surgery (CHS) at any point across their hospitalisation history (*T18CHS*). Patients with T18 and a CHD diagnosis who underwent a percutaneous congenital cardiac intervention could not be placed in either of the groups and were excluded from this analysis. Further, the most common congenital heart diagnosis among the *T18CHS* group was selected for a sub-group analysis between surgical and non-surgical CHD T18 patients. Surgical hospitals were defined as any unique centre in which a patient in this analysis underwent a CHS.

The Institutional Review Board for the University of Texas at Austin Dell Medical School waived the need for review of this study due to the use of publicly available and de-identified data.

Descriptive statistics were used to report demographics and outcomes across groups. Comparisons of non-continuous variables were made using Chi-square and Fisher exact tests. For non-parametric continuous variables, the Kruskal–Wallis and Wilcoxon signed-rank tests were used. The Kaplan–Meier method was used to evaluate survival across groups. A multivariable regression model was used to evaluate the odds of undergoing CHS. All statistical analyses were two-tailed and a p-value of <0.05 was considered significant. All statistical analyses were performed using R and RStudio.[13]

Results

Study population

A total of 2578 hospital discharges with the diagnosis of T18 were identified. Of these, 396 discharges reported an additional autosomal trisomy diagnosis, and thus, were excluded. After having linked the 2182 remaining discharges, patients with a recorded age of ≥25 years (64 linked patients) were excluded; as well as all discharges corresponding to patients with evidence of T18 mosaicism (159 linked discharges). The final dataset included 1159 patients with 1959 total discharges.

From the 1159 patients, 443 (38%) did not have a CHD diagnosis, 669 (58%) had a CHD diagnosis and no record of congenital heart surgery, and 44 (4%) had a CHD diagnosis and underwent congenital heart surgery. Three additional patients with T18 and a CHD diagnosis underwent an isolated percutaneous cardiac intervention (two underwent a percutaneous balloon valvuloplasty and one had a percutaneous insertion of an aortic device) and were excluded from this analysis.

Among the *T18CHS* group, multiple cardiac procedures were observed, the majority of which were patent ductus arteriosus closures, atrial septal defect and/or ventricular septal defect repairs (*Supplementary Table S1*). The 44 congenital heart surgeries were performed among 12 hospitals, including all seven Society of Thoracic Surgeons Congenital Heart Surgery Database reporting congenital heart centres in Texas (https://publicreporting.sts.org/chsd?title=&location_depth=2893).

Demographic characteristics

Of the cohort, 734 (64%) were female, 637 (58%) were white and 543 (47%) were Hispanic, with a higher proportion of female patients ($n = 33$ (75%), $p = 0.024$) and a lower proportion of Hispanic patients ($n = 9$ (21%), $p = 0.001$) in the *T18CHS* group. A lower proportion of patients with Medicare/Medicaid insurance (MC/MA) was found among the *T18CHS* group ($n = 14$ (32%), $p < 0.001$) (Table 1a).

After adjusting for ethnicity, race, and admission to a surgical centre, MC/MA had a 0.42 (95%CI: 0.20–0.86, $p = 0.020$) odds of undergoing congenital heart surgery compared to private insurance (*Supplementary Table S2*). Patients with MC/MA had a similar distribution of CHD diagnoses compared to other insurance types (*Supplementary Table S3*).

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Table 1. a. Demographic characteristics of Trisomy 18 (T18) patients in the state of Texas 2009–2019. **b.** Outcomes among the different T18 groups

	Total, n = 1156	*T18NoCHD*, n = 443	*T18CHD*, n = 669	*T18CHS*, n = 44	Overall sig.
a. Demographics, n (%)					
Female, n (%)	734 (63.49)	262 (59.14)	439 (65.62)	33 (75.00)	**p = 0.024**
Race, n (%)					
Am. Indian/Eskimo/Aleut	6 (0.52)	3 (0.68)	2 (0.3)	1 (2.27)	**p = 0.043**
Asian or Pacific Islander	23 (1.99)	6 (1.35)	17 (2.54)	0 (0)	
Black	146 (12.63)	45 (10.16)	94 (14.05)	7 (15.91)	
White	673 (58.22)	262 (59.14)	380 (56.80)	31 (70.45)	
Other	308 (26.64)	127 (28.67)	176 (26.31)	5 (11.36)	
Hispanic, n (%)	543 (46.97)	219 (49.44)	315 (47.09)	9 (20.45)	**p = 0.001**
Insurance, n (%)					
Private	407 (35.21)	146 (32.96)	235 (35.13)	26 (59.09)	**p < 0.001**
Uninsured	50 (4.33)	29 (6.55)	20 (2.99)	1 (2.27)	
Medicaid/Medicare	668 (57.79)	259 (58.47)	395 (59.04)	14 (31.82)	
Other/unknown	31 (2.68)	9 (2.03)	19 (2.84)	3 (6.82)	
b. Clinical characteristics and outcomes					
Median Admissions Records (n [IQR])					
Excluding <2 admissions	1 [1–1]	1 [1–1]	1 [1–2]	2 [1–4]	**p < 0.001**
	3 [2–4]	2 [2–3]	3 [2–4]	4 [2.75–6.25]	**p = 0.016**
Median LOS (days [IQR])					
Total	5 [1–18]	2 [1–5]	9 [3–24]	47.5 [12.25–113.25]	**p < 0.001**
Mean	4 [1–11]	2 [1–4]	6.5 [2.6–15]	14.38 [6.94–41.56]	**p < 0.001**
Longest stay	4 [1–15]	2 [1–5]	8 [3–20]	30.5 [9.75–68.25]	**p < 0.001**
Gastrostomy status, n (%)	247 (21.37)	72 (16.25)	147 (21.97)	28 (63.64)	**p < 0.001**
Tracheostomy status, n (%)	55 (4.76)	15 (3.39)	31 (4.63)	9 (20.45)	**p < 0.001**

T18NoCHD: T18 patients with no record of a CHD.
T18CHD: T18 patients with a record of a CHD but no record of congenital heart surgery.
T18CHS: T18 patients with a CHD and a record of a congenital heart surgery.
Statistical significance (p<0.05) values are in bold.

Outcomes

The median number of admissions per patient was higher among the *T18CHS* group (2 [IQR: 1–4]), compared to 1 [IQR: 1–2] in *T18CHD* and 1 [IQR: 1–1] in *T18NoCHD* ($p < 0.001$). After excluding patients who were only admitted once, the number of admissions remained higher among the *T18CHS* group (4 [IQR: 2.75–6.25]), compared to the *T18CHD* (3 [IQR: 2–4], $p = 0.025$) and *T18NoCHD* groups (2 [IQR: 2–3], $p = 0.04$). The median number of postoperative admissions in the *T18CHS* group was 0 [IQR: 0–2], with 26 (59%) patients having no post-operative admissions. Measures of length of stay were longer among the *T18CHS* group with a median total days inpatient, mean length of stay, and longest length of stay of 48, 14, and 31 days, respectively ($p < 0.001$ for all) (Table 1b).

The *T18CHS* group had a higher proportion of patients with evidence of a gastrostomy tube (n = 28, 64%), compared to the *T18CHD* (n = 147, 22%) and *T18NoCHD* groups (n = 72, 16%) ($p < 0.001$); as well as a higher proportion of tracheostomies (n = 9, 21%), compared to the *T18CHD* (n = 31, 5%) and *T18NoCHD* (n = 15, 3%) ($p < 0.001$) groups.

From the 28 *T18CHS* patients with a gastrostomy, 12 (43%) were placed before the congenital heart surgery, with 2 of the 12 being placed during the same hospitalisation as the congenital heart surgery. Of the 16 *T18CHS* patients with post-congenital heart surgery gastrostomies, 11 (68.8%) were placed during the congenital heart surgery hospitalisation at a median of 36 days [IQR: 14–48] after congenital heart surgery. Of the 9 *T18CHS* patients with tracheostomies, 3 (33%) were placed before the CHS. Of the six patients with post-congenital heart surgery tracheostomy, 4 (66.7%) were during the congenital heart surgery hospitalisation at a median of 61.5 days [IQR: 42.75–74.5] after the congenital heart surgery.

A significantly lower in-hospital mortality was observed in the *T18CHS* group, largely driven by the mortality difference during the first month of life. Figure 1 presents a Kaplan–Meier curve (KM) including only the patients who could be tracked from birth. A graphical representation of the oldest known age group distribution for the entire studied population can be observed in Figure 2.

A separate analysis performed on the patients whose records indicated any kind of non-cardiac surgical intervention, utilised

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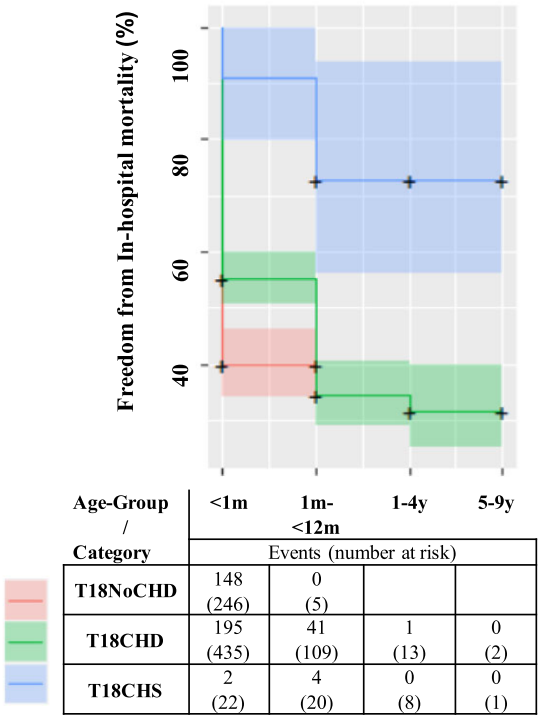

Figure 1. Kaplan–Meier survival curve for in-hospital mortalities across the three patient groups among individuals with a birth record. All differences were statistically significant ($p \leq 0.002$).

as a proxy for patients who were not initially considered for comfort care only, revealed similar outcomes (*Supplementary Table S4*).

Sub-group analysis: patients with ASD and VSD

The most frequent *T18CHS* cardiac diagnostic combination was atrial septal defect and ventricular septal defect, with or without patent ductus arteriosus, found in 16 (36%) patients in the *T18CHS* group ($T18CHS_a$), and in 128 (19%) patients in the *T18CHD* group ($T18CHD_a$).

The atrial septal defect and ventricular septal defect with or without patent ductus arteriosus ($T18CHS_a$+ $T18CHD_a$) subgroup had demographic findings consistent with the overall cohort, with the majority of the patients being female (n = 95 (66%)), white (n = 80 (56%)), and non-Hispanic (n = 79 (55%)), with no statistical difference between the two groups. More $T18CHD_a$ patients had MC/MA than $T18CHS_a$ (63% vs. 25%, p = 0.013) (Table 2a), resulting in a 0.167 (95%CI: 0.044–0.512, p = 0.003) OR of undergoing congenital heart surgery when insured by MC/MA compared to private insurance.

The median number of admissions was higher for $T18CHS_a$ patients (2.5 [IQR: 1–4.75]) compared to $T18CHD_a$ (1 [IQR: 1–2], p = 0.038). However, after the exclusion of patients with a single discharge, the difference was no longer significant (3 [IQR: 2.75–7] vs. 3 [IQR: 2–5], p = 0.108). The median number of post-operative admissions for the $T18CHS_a$ group was 0 [IQR: 0–2].

The $T18CHS_a$ group had more gastrostomies (56% vs. 26%, p = 0.011) and tracheostomies (31% vs. 8%, p = 0.014) compared to the $T18CHD_a$ cohort (Table 2b).

Discussion

This analysis of more than a decade of real-world data from a statewide dataset describes one of the largest T18 populations reported in the literature. It presents a medium-to-long term follow-up of patients born with T18 with or without associated CHD and compares their demographics and outcomes among the different patient groups.

In an analysis of the Society of Thoracic Surgeons (STS) Congenital Heart Surgery Database of CHD surgeries performed on patients with T18 and Trisomy 13 between 2010 and 2017, Cooper et al found that 70% of the STS reporting centres operated on T18 patients, despite the common perception of poor prognosis among medical providers.[5] In agreement with these findings, this analysis identified CHD surgeries performed at all STS reporting congenital heart centres in Texas.

The administrative character of the data used relies on accurate coding by reporting institutions, likely underestimating the true incidence of CHD in patients with T18, in particular, patients who did not undergo extensive diagnostic work-up before discharge. This might explain the lower number of T18 with CHD observed in this study (62%), compared to what is currently reported in the literature (>80%).[6] Besides, the number of procedures performed on T18 patients likely underestimates the totality of patients considered candidates for surgery, since it does not reflect the proportion of patients who were offered surgery and whose families declined. The decision of whether to proceed with congenital heart surgery or not is a challenging choice for families and providers. As Cooper et al signals, this question is assessed depending on patient clinical status, goals of treatment, and parental choice.[5] And as suggested by Sullivan et al in their historic review of clinical decision-making in complex neonatal cases, a shared decision-making approach between the medical team and families is encouraged.[14]

Our findings suggest the potential for an important disparity in care among patients with T18 and CHD, as insurance-type differences were observed between patients undergoing surgical interventions and those who did not. When evaluating the entire T18 population with CHD (*T18CHD* and *T18CHS*), and after adjusting for potential confounding variables, we identified significantly fewer patients with MC/MA in the *T18CHS* group compared to the *T18CHD* group. The lower proportion of CHS cases recorded in patients with MC/MA may derive from multiple factors including lower socio-economic status, unequal access to health services, as well as parental preference.[15] Further investigation and more clinical patient data are necessary to better understand this complex relationship of insurance type and undergoing congenital heart surgery in T18.

The survival to discharge after congenital heart surgery in our study was 86%, similar to an analysis performed by Ma et al of the Kid's Inpatient Database, which showed an 88% survival to hospital discharge among T18 patients undergoing congenital heart surgery.[6] These data suggest patient selection for congenital heart surgery was overall appropriate.

After the first month of life, patients with T18 and CHD who underwent congenital heart surgery had a similar freedom from in-hospital mortality compared to patients with T18 and no CHD and superior to those with CHD who did not undergo congenital heart surgery. Our findings are consistent with those reported by Peterson et al, who showed a significantly higher 15-year survival for patients with T18 and CHD who underwent congenital heart surgery, compared to patients with T18 and CHD receiving only palliative measures.[15] This further points to the appropriateness

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Table 2. a. Demographics of T18 patients with ASD + VSD ± PDA who did not undergo surgical intervention ($T18CHD_a$) and T18 patients with ASD + VSD ± PDA who underwent a congenital heart surgery ($T18CHS_a$). **b.** Outcomes of $T18CHD_a$ and $T18CHS_a$ patients.

	Total, n = 144	$T18CHD_a$, n = 128	$T18CHS_a$, n = 16	Overall sig.
a. Demographics n (%)				
Female, n (%)	95 (65.97)	82 (64.06)	13 (81.25)	p = 0.263
Race, n (%)				
Asian/Pacific Islander	5 (3.47)	5 (3.91)	0	p = 0.676
Black	26 (18.06)	23 (17.97)	3 (18.75)	
White	80 (55.56)	69 (53.91)	11 (68.75)	
Other	33 (22.92)	31 (24.22)	2 (12.5)	
Hispanic, n (%)	65 (45.14)	61 (47.66)	4 (25.00)	p = 0.112
Insurance, n (%)				
Private	52 (36.11)	40 (31.25)	12 (75.00)	**p = 0.013**
Uninsured	3 (2.08)	3 (2.34)	0 (0)	
Medicaid/Medicare	84 (58.33)	80 (62.50)	4 (25.00)	
Other/unknown	5 (3.47)	5 (3.91)	0 (0)	
b. Clinical characteristics and outcomes				
Median Admissions Records (n [IQR])				
Excluding <2 admissions	1 [1–3]	1 [1–2]	2.5 [1–4.75]	**p = 0.038**
	3 [2–6.75]	3 [2–5]	3 [2.75–7]	p = 0.108
Median LOS (days [IQR])				
Total	5 [1–18]	9 [3–24]	47.5 [12.25–113.25]	**p < 0.001**
Mean	4 [1–11]	6.5 [2.6–15]	14.375 [6.942–41.562]	**p < 0.001**
Longest stay	4 [1–15]	8 [3–20]	30.5 [9.75–68.25]	**p < 0.001**
Gastrostomy status, n (%)	42 (29.17)	33 (25.78)	9 (56.25)	**p = 0.011**
Tracheostomy status, n (%)	15 (10.42)	10 (7.81)	5 (31.25)	**p = 0.014**

Statistical significance (p<0.05) values are in bold.

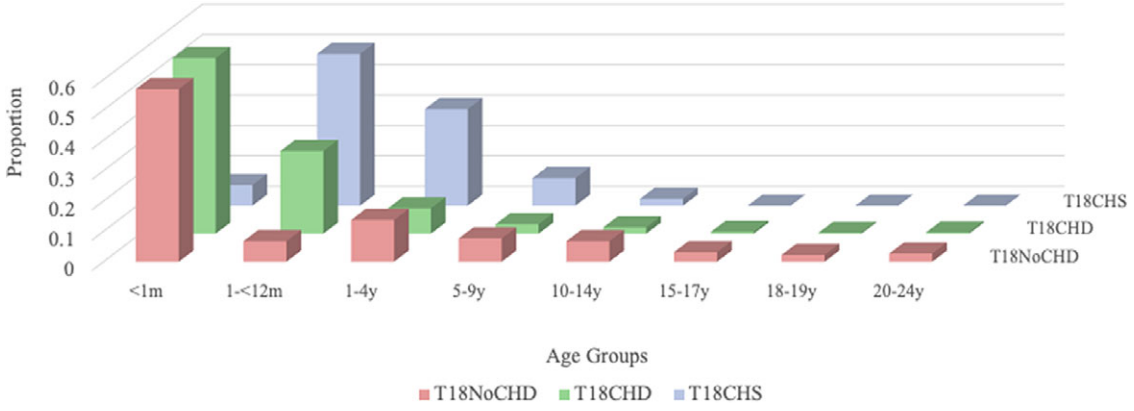

Figure 2. Distribution of oldest known age group in *T18NoCHD*, *T18CHD,* and *T18CHS* patients.

of surgical patient selection and potential survival improvements in these selected cases.

The higher number of admissions prior to the congenital heart surgery hospitalisation and the longer length of stay among the surgical group points to an increased utilisation of health care resources among patients undergoing surgery, particularly before undergoing CHS. It is clear that once the decision of pursuing congenital heart surgery is made, other aggressive measures are accepted in the management of these patients (i.e. gastrostomy and tracheostomy). Kosiv et al also demonstrated a longer length of stay among T18 patients who underwent congenital heart surgery, and a longer length of stay for patients with CHD (without congenital heart surgery), compared to T18 patients without CHD. Similarly, there was a higher rate of gastrostomy tube placement

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among patients with T18 undergoing congenital heart surgery.[1] The median number of post-operative admissions for the *T18CHS* group of 0 [IQR: 0–2] in this analysis reveals that a significant number of hospitalisations for this group happened before their congenital heart surgery. This finding urges further investigation to assess opportunities to streamline the management of patients with T18 and CHD prior to congenital heart surgery.

Limitations

The administrative nature of the data source does not allow for granular clinical evaluation of each record, limiting our analysis to reported codable diagnoses and interventions. There was an inability to accurately and completely identify all mosaic diagnoses, particularly before the fourth quarter of 2015 as ICD-9 did not include a code for T18 mosaicism. Further, the identification of T18, mosaicism, and the rest of diagnoses evaluated in this study was based on the information coded by the reporting institutions and not with diagnostic testing. Introducing the possibility of having missed existing diagnoses among the population examined.

In addition, as a result of working with de-identified data, there is the potential of having erroneously linked multiple discharges to one patient or having missed different discharges corresponding to the same patient. However, matching of records was based on multiple characteristics, and concordance between the two independent reviewers was high.

Since the data used did not allow evaluation of a time domain, the KM does not represent a true survival rate. Further, the survival curve is based only on in-hospital mortalities. This introduces a bias and cannot be compared to a survival curve of the entire T18 population, as it does not capture out-of-hospital mortalities. Besides, the data source does not discern between causes and/or modalities of death; nor it provides reasons for readmission or need for home medical care.

Further, the data are limited to discharges in the state of Texas; thus, hospitalisations outside the state of Texas are not captured.

Conclusions

Given the complex nature of T18, a multidisciplinary approach to the decision for management of CHD is needed. The factors weighing into the decision to pursue congenital heart surgery are not completely described, but these data suggest that insurance-type disparities exist between patients with T18 and CHD who undergo surgery and those who do not.

In addition, the follow-up of patients with T18 and CHD who underwent congenital heart surgery in this study suggests a freedom from in-hospital mortality that resembles that of T18 patients without CHD. Future efforts should be made to form a detailed registry of all T18 patients to assist in better understanding of their needs, comorbidities, outcomes, family experience and burden, and life-long journey.

Supplementary material. To view supplementary material for this article, please visit https://doi.org/10.1017/S1047951123000215

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Conflicts of interest. None.

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