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It's a Bleed: Pediatric Hemophilia and Length of Stay, Rural vs Urban Hospitals

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ABSTRACT

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Hemophilia is a rare genetic disorder that requires specialty care and treatment. Pediatric patients with hemophilia have unique medical issues that may lead to permanent disability or death if not properly diagnosed and treated in a timely manner. Due to lack of resources and proper training of staff, rural hospitals are not equipped to properly treat pediatric hemophilia patients. Utilizing the Healthcare Cost and Utilization Project (HCUP) Kids´ Inpatient Database (KID) of the Agency for Healthcare Research and Quality. I have found, across all hospital types, pediatric hemophilia patients have longer lengths of stay, 2.7 days for rural hospitals, 4.6 days for urban hospitals, and 5.1 days for teaching hospitals, compared to the national mean for pediatric LOS of 1.8 days (Heys et al. 2017).

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Introduction

Hemophilia is a rare genetic disorder that requires specialty medical treatment and care. This specialty care and treatment requires special training for physicians, hospital staff and patients, to properly treat the hemophilia. Healthcare providers who are not properly trained may misdiagnose or not understand how to treat hemophilia, causing a delay of a necessary treatment. A delay in treatment can be catastrophic for a person with hemophilia. They can suffer long term pain, disability, and even death with delayed or improper treatment.

The NHF (2019), recommends "Individuals with bleeding disorders should be triaged urgently as delays in administering appropriate therapy, such as infusion of factor concentrate, can significantly affect morbidity and mortality" and "administration of clotting factor replacement to the patient should not be delayed waiting for a consultation."

Inequality between rural and urban hospitals, in resources and specialty staff, may lead to delays in treatments and diagnosis for pediatric patients with hemophilia. These delays can lead to increased length of stays or transfer to another hospital for better treatment. This may cause pain, suffering, and possibly death while the patient waits for proper treatment.

Although many studies considered length of stay, very few looked at pediatric hemophilia patients. Yeung et al. (2016) stated, "There were a paucity of data from the studies to group by disease severity [… and] age (in particular paediatric and geriatric groups" (39). And Soucie et al. (2000) pointed out, "No studies have directly examined the associations between mortality and factors related to medical management, such as care received in HTCs [Hemophilia Treatment Center]" (437). This study adds to the knowledge on pediatric hemophilia.

Due to the unique nature of hemophilia and how it is diagnosed, I posit pediatric patients in rural hospitals, with hemophilia, will have longer length of stays due to lack of resources and staff unfamiliar with hemophilia. Utilizing the Healthcare Cost and Utilization Project (HCUP) Kids´ Inpatient Database (KID) of the Agency for Healthcare Research and Quality, this study examines variables associated with inpatient admissions for pediatric hemophilia patients in rural, urban, non-teaching and urban, teaching hospitals for 2009, 2012, and 2016. I test whether type of hospital is associated with length of stay.

Literature Review

Hemophilia:

Hemophilia is a rare genetic bleeding disorder found on a recessive x chromosome that leads to deficiencies of clotting factors, specifically hemophilia A (factor VIII deficiency) and hemophilia B (factor IX deficiency) for this study (Mirchandani et al. 2011; Soucie et al. 2000; Yan and Kung 2013). Since it is a recessive x chromosome it most often passed from the mother to her child and usually expresses in male children due to hemizygosity where the male has one x chromosome, from the mother, and one y chromosome, from the father. Females, who have two x chromosomes, may have hemophilia but only if both parents passed on a recessive x chromosome. This would mean the father had to have hemophilia and the mother either had hemophilia or was a carrier (Flantzer 2021; Pemberton 2011). See Figure 1. This has led to many in the medical field who dismiss the idea of a female who has hemophilia and lack of diagnoses or proper treatment for females.

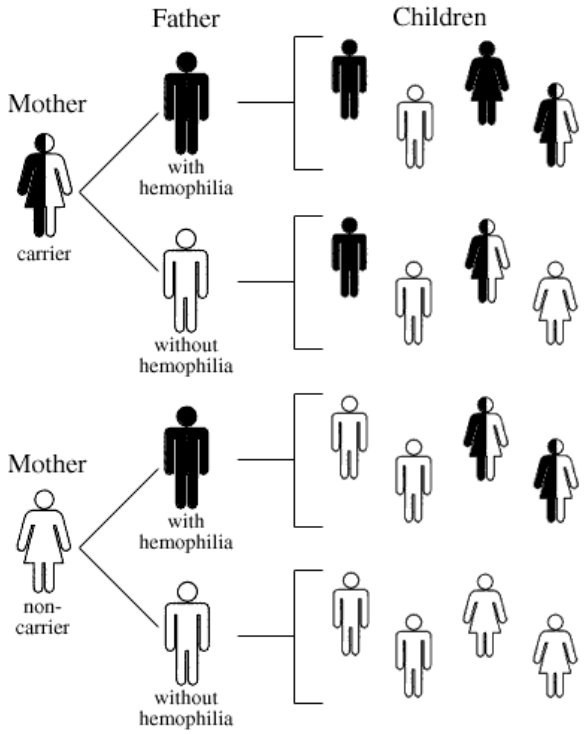

Figure 1

How Hemophilia is Inherited

(Source: http://www.unofficialroyalty.com/royal-illnesses-and-deaths/hemophilia/what-is-hemophilia/)

A diagnosis of hemophilia or other type of bleeding disorder happens when there is a deficiency of one of 13 blood proteins known as factors. These factors affect blood clotting and hemostasis within the human body. The most common type of factor deficiency is hemophilia A or Factor VIII deficiency, with approximately 80 - 85% of cases, followed by hemophilia B or Factor IX deficiency (NHF 2020; Yan 2013).

Hemophilia is treated with an antihemophilic factor products, known as “Factor,” in the community. This Factor product has, in the past, been produced through a pooled blood procedure made by using tens of thousands of units of donated blood. Factor is isolated from the

pooled blood and made into a concentrate that is used for treatment. Synthetic products have been produced in vitro, using cell culture lines that do not contain human blood since 1992. The Factor concentrate used as an intravenous infusion to treat hemophilia, either by the person with hemophilia or a trained individual (DePrince 2017; NHF 2020; Resnik 1999; Weinberg and Shaw 2017).

There are conflicting data concerning the number of people with hemophilia in the United States. Soucie et al. (1998), collected data from Hemophilia Treatment Centers in six states and determined, through extrapolation, there were approximately 13,000 people with hemophilia in the United States. The CDC (2019) reports in 1994 there were around 17,000 and in 2019, it is estimated that around 20,000 people have Hemophilia in the United States. Another study by Iorio et al. (2019), using data from six countries with universal healthcare, estimated the numbers to be 17.1/100,000 males worldwide which extrapolates to ~ 20,000 males with hemophilia in the United States. Lastly, the CDCa (2019) provides numbers of people with all types of hemophilia being treated at all the Hemophilia Treatment Centers in the United States, from the years 2012 through 2018 ranging from the lowest in 2018 of 21,861 to the highest in 2017 of 34,346.

In 1975 Congress passed the Public Health Service Act which contained provisions that, "established and funded a network of Hemophilia Diagnostic and Treatment Centers throughout the United States" (Smith et al. 1984:616). These centers became Hemophilia Treatment Centers or HTCs. Mirchandani et al. (2011), referred to HTC's when they stated, "HTCs effectively function as sentinel sites for passive surveillance of hemophilia and other bleeding disorders" (s355). And McCavit et al. (2011) pointed out, "hemophilia treatment centers, … dramatically improve costs, service utilization, and functional outcomes" (377).

Hemophilia Treatment Centers provide comprehensive treatment for anyone with a diagnosed bleeding disorder. The staff at these HTCs are highly trained in the care and treatment of hemophilia and bleeding disorders (Mirchandani et al. 2011; Soucie et al. 2000; Yeung et al. 2016; Yan and Kung 2013). Smith et al. (1984:616) states,

> The minimum services provided by each center were:
> - A coagulation laboratory of recognized high standards;
> - A blood bank providing all of the blood components needed by hemophiliacs;
> - A multidisciplinary hemophilia care team including a hematologist, an internist, a pediatrician, an orthopedic surgeon, a physical therapist, a dentist, a social worker, and a registered nurse;
> - Formal linkages with mental health, genetic counseling, and rehabilitative services;
> - A training course in self-therapy (home care) and updated hemophilia concepts for patients and family members;
> - An outreach program to enable every hemophiliac within the area served to receive services of the program

Much of the literature estimates approximately two thirds of hemophilia patients receive at least some of their treatment at HTCs (Mirchandani et al. 2011; Soucie et al. 2000; Yeung et al. 2016). Common issues faced by the person with hemophilia include: associated costs of medicines and lost productivity, treatment side effects, arthropathy of joints leading to physical disabilities and mobility problems, chronic pain, and a shorter projected lifespan (Barkdull 2014; DePrince 2017; Resnik 1999; Weinberg and Shaw 2017; White and Cunningham 1991). According to Soucie et al. (2000), "survival is significantly greater among hemophiliacs who receive medical care in HTCs" (437).

Hemophilia Diagnosis and Treatment:

A reliable method of diagnosing hemophilia was not available until the 1950's when scientists developed clotting factor assays, specifically a "thromboplastin generation test" in 1953 and at the same time frame the "partial thromboplastin time" test was also developed (Pemberton 2011:92). According to the Center for Disease Control (CDC b 2020) and Dr.

Samuel Merrill (pers. comm.) currently, several methods are utilized for hemophilia diagnosis, they include complete blood count (CBC), activated partial thromboplastin time (aPTT), prothrombin time (PT), fibrinogen, and specialized clotting factor tests and factor assays.

The CBC can tell how many red blood cells are in the blood and if the number is low, it may indicate excess bleeding. An abnormal aPTT could indicate a deficiency of a clotting factor such as Factor VIII or IX. A PT tests for different clotting factor deficiencies. And a Fibrinogen test for the ability to form clots (CDC b 2020; Dr. Merrill (pers. comm); Pemberton 2011). According to Dr. Samuel Merrill (pers. Comm), there are currently more than 50 different recognized bleeding disorders, hemophilia A and hemophilia B are two of the more prevalent.

The clotting factor assays will provide the type of specific factor deficiency, for example factor VIII (hemophilia A) or factor IX (hemophilia B) and the severity of the hemophilia (CDC b 2020; Dr. Merrill (pers. comm); Pemberton 2011). Severity indicates the level of clotting factor deficiency. The levels include, severe, which is less than 1% of normal, moderate, which is 1-5% of normal, and mild, which is 6- 30% of normal (Soucie 2000). According to Kulkarni and Souci (2011), “Approximately two-thirds of persons with hemophilia have severe disease; 15% have moderate disease; and 20% have mild disease” (737).

Hemophilia treatment has come a long way in the last 100 years. From no special treatment other than poultices, rest, heat, and ice to whole blood transfusions, to cryoprecipitate (the sludge scraped off the bottom of thawed plasma), to “Factor” (a concentrated form of the specific clotting factor, such as factor VIII or IX, made from pooled donated blood or *in vitro*), to the newest gene therapy (Barkdull 2014; DePrince 2017; NHF 2020; and Pemberton 2011).

Today most people with severe hemophilia treat prophylactically with recombinant factor concentrates. The individual, parent or someone trusted are taught to infuse through venipuncture

in a vein or a central line. Two regularly used central lines include a peripherally inserted central line (PICC line) which, according to the Mayo Clinic (2021), is a thin tube inserted into a vein in the arm, and rarely in the leg or neck, and threaded through larger veins close to the heart. The PICC makes it easier to infuse and saves the individual from numerous venipunctures. And a Port, which, according to The National Cancer Institute (2022), is a device used for infusions and drawing blood that is inserted under the skin in the upper chest and is attached to a thin tube that is inserted into a large vein and placed near the heart. Once they learn how to self-infuse, they are then provided with the product and essential accessories to perform this on a regular basis (Kulkarni and Soucie 2011; NHF 2020; Pemberton 2011).

Prophylactic treatment is done at home by the individual or a trusted person, two or more times a week to maintain factor levels in the normal range and is meant to be proactive in the prevention and treatment of bleeds. The process of prophylactic treatment not only requires special training but also maintaining sterile procedures and making arrangements for proper disposal of infusion equipment, such a needles and syringes. As Bertamino et al. (2017) report, "prophylactic treatment that is started early with clotting-factor concentrates has been shown to prevent hemophilic arthropathy and is, therefore, the gold standard of care for hemophilia A and B in most countries with adequate resources" (1).

Pediatric hemophilia:

Kulkarni and Soucie (2011) sum up pediatric hemophilia treatment when they state, "hemophilia is a serious congenital bleeding disorder that requires early diagnosis, intensive family and patient education, and regular comprehensive care to prevent life-threatening complications and potentially lifelong disability" (743). Pediatric hemophilia has different

treatment requirements than adults and requires closer personal care to ensure continuity of therapy as they grow up.

Neonatal patients may have issues with intracranial hemorrhages due to delivery method or just regular head bumps from everyday activity. They also may manifest bleeds during circumcision, teething, heel pricks at birth or even venipuncture; older pediatric patients with hemophilia manifest with injuries to joints or soft tissue due to falls, bumps, scrapes from being children (Bertamino et al. 2017; Ettingshausen et al. 2001; Kulkarni and Souci 2011).

It is imperative for pediatric bleeds to be treated immediately or the bleed may cause irreparable damage to the brain, joints, and or soft tissue and could even cause death, if left untreated. Kulkarni and Soucie (2011) state,

> "bleeding episodes in hemophilia often result in physical (pain, hemophilic arthropathy, organ dysfunction) or mental impairments leading to restriction in activities including school and social participation that affect education and QOL. The major functional impairment in mobility is secondary to limited joint ROM with BMI as a significant predictor" (742).

Research has shown the earlier a pediatric patient with hemophilia is treated the better the long-term outcomes. By treating pediatric hemophilia patients in a timely manner, the more likely they are to avoid long-term damage and disability: "The goal must be to avoid bleeding complications and joint damage in the pediatric age in order to enable the hemophiliac patient to reach adulthood as healthy as possible" (Bertamino et al. 2017:8).

Rural vs non-Teaching, Urban vs Urban Teaching Hospitals

The literature shows a discrepancy in treatment options concerning rare disorders, including hemophilia, between rural and urban hospitals (teaching and non-teaching). Due to these discrepancies, treatment is delayed, and complications arise requiring longer length of stays (LOS) and/or transfer to urban hospitals (Akintoye et al. 2017; Robinson and Luft 1985; Vohra

et al. 2020). As Vohra et al. (2020) points out, “rural-urban disparities also present in children’s health outcomes” (493) and “higher mortality incidence exists in rural counties compared to urban counties for infants, children, and young adults” (493)

Research further shows LOS is affected by various conditions, including diagnosis, regulations, bed size (number of beds in the hospital), costs, and physician/patient choice. Studies have shown urban hospitals usually have longer average length of stays than rural hospitals, with urban teaching hospitals regularly having the longest average length of stay. While LOS appears to be longer in non-teaching and teaching urban hospitals, rare diseases being treated in rural hospitals seem to show longer LOS or transfer to urban hospitals when the rural hospital is unable to provide the necessary care (Jatwani et al. 2019; Lorch et al. 2004; Singh and Ladusingh 2010): “This suggests that 2 distinct processes are at work, i.e., routine treatment of patients with uncomplicated conditions who are discharged promptly and treatment of patients with special complications that delay discharge” (Lorch et al. 2004:e401).

Lee et al. (2003) states, “length of stay (LOS) is an important measure of hospital activity and health care utilization” (681) and “length of stay (LOS) is an important measure of health care utilization and determinant of hospitalization cost” (681). Hospital administration, physicians, and costs factor into services available at hospitals. Services and amenities factor into patient satisfaction and hospitals in large markets usually have more services and amenities due to competition while smaller hospitals are unable to compete because of costs of equipment and other resources (Robinson and Luft 1985).

As a result of hospitals trying to control rising costs some services and overall quality of healthcare may have suffered: “Systematic clinical cost-containment efforts have increased in response to the need to control health care expenditures” (Weingarten et al. 1998:33). This may

have led to rural healthcare lacking proper resources and lacking specialist providers. Which leads to, “higher mortality incidence exist[ing] in rural counties compared to urban counties for infants, children, and young adults” (Vohra et al. 493)

Because the “potential differences between rural and urban hospitals may influence the efficiency of care received by hospitalized children” (Lorch et al. 2004:e400), pediatric patients with hemophilia will suffer. Leyland and Andrew (1997) also point out that there is “a negative correlation between residuals for length of hospital stay and those for readmission rates suggesting that pressures to reduce lengths of stay may have the effect of increasing rates of readmission” (141) and “for fixed bed numbers - the length of stay will affect and be affected by admission rates and that readmission rates will have a similar relationship” (141).

Due to the complex nature of hemophilia, hospitals must possess proper resources and trained staff. In most instances, rural hospitals do not possess these resources or training. This should lead to longer length of stays for pediatric patients with hemophilia. With longer lengths of stay, pediatric patients with hemophilia suffer for, first, lack of or delay in proper treatment and second, quality of life.

Hypotheses

H1 - Pediatric patients with hemophilia in rural hospitals will have longer length of stays compared to urban hospitals.

Methods

Data – This study utilized the Healthcare Cost and Utilization Project (HCUP) Kids´ Inpatient Database (KID) of the Agency for Healthcare Research and Quality. The HCUP Kids dataset contains over 4 million pediatric discharges and is part of HCUP which is one of the largest data collections of longitudinal hospital care in the United States, developed by a

partnership consisting of Federal, State and Industry. The Federal government along with State data organizations, hospital organizations, and private data organizations created the dataset as a "national information resource of encounter-level healthcare data." HCUP has been compiling data since 1988 with "The Kids' Inpatient Database" being collected since 1997 and contains a sample of nationwide pediatric inpatient discharges.

This study examines inpatient discharges for pediatric hemophilia patients in 2009, 2012, and 2016. For this study, I only consider length of stays with a diagnosis of either hemophilia A or hemophilia B.

Cases – Cases were identified using ICD-10 diagnosis code 286.0 Congenital factor VIII deficiency (hemophilia A) and 286.1 Congenital factor IX deficiency (hemophilia B). All variable descriptions are copied directly from the HCUP KID dataset codebook (https://www.hcup-us.ahrq.gov/db/nation/kid/kiddde.jsp).

Dependent Variable

LOS (length of hospital stay) is calculated by subtracting the admission date from the discharge date. Same-day stays are coded as 0. Leave days are not subtracted. Ranges from 0 up to 228 days.

Independent Variable

Hospital Location indicates the location and teaching status of hospital. A hospital is considered a teaching hospital if it has one or more Accreditation Council for Graduate Medical Education (ACGME) approved residency programs, is a member of the Council of Teaching Hospitals (COTH) or has a ratio of full-time equivalent interns and residents to beds of .25 or higher. Rural hospitals were not split according to teaching status,

because rural teaching hospitals were rare. 1 = rural, 2 = urban nonteaching, and 3= urban teaching.

Control Variables

AGE (age at admission) is calculated from the patient's date of birth and the admission date, specifically ages 0 - 20.

FEMALE (sex of patient) is provided by the data source. All non-male, non-female (e.g., "other") values were set to missing by the data source (0 = Male and 1 = Female).

RACE (race of patient), 1 = White, 2 = Black, 3 = Hispanic, 4 = Asian or Pacific Islander, 5 = Native American, or 6 = Other.

Median Household Income provides a quartile classification of the estimated median household income of residents in the patient's ZIP Code, 1= 0 – 25th quartile, 2 = 26th – 50th quartile, 3 = 51st – 75th quartile, and 4 = 76th – 100th quartile.

Number of Hospital Beds (see Table 1) are based on the number of hospital beds, the hospital's location, and teaching status. Bedsize assesses the number of short-term acute care beds set up and staffed in a hospital. Hospital information was obtained from the American Hospital Association Annual Survey of Hospitals. The hospital's bedsize categories are defined using region of the U.S., the urban-rural designation of the hospital, and the teaching status of the hospital. 1 = small, 2 = medium and 3 = large based on the following table from the HCUP KIDS code book (https://www.hcup-us.ahrq.gov/db/vars/hosp_bedsize/kidnote.jsp).

Table 1. HCUP KIDS Hospital Bedside Definition

Location and Teaching Status	**Hospital Bedsize**		
	Small	Medium	Large

NORTHEAST REGION			
Rural	1-49	50-99	100+
Urban, nonteaching	1-124	125-199	200+
Urban, teaching	1-249	250-424	425+
MIDWEST REGION			
Rural	1-29	30-49	50+
Urban, nonteaching	1-74	75-174	175+
Urban, teaching	1-249	250-374	375+
SOUTHERN REGION			
Rural	1-39	40-74	75+
Urban, nonteaching	1-99	100-199	200+
Urban, teaching	1-249	250-449	450+
WESTERN REGION			
Rural	1-24	25-44	45+
Urban, nonteaching	1-99	100-174	175+
Urban, teaching	1-199	200-324	325+

Hospital Control The control/ownership of the hospital is operationalized as: 1 = government, non-federal, 2 = private, non-profit, and 3 = private, investor owned. *Hospital Region* indicates the hospital's census region (1 = Northeast, 2 = Midwest, 3 = South, and 4 = West).

Patient location represents a six-category urban-rural classification scheme for U.S. counties developed by the National Center for Health Statistics (NCHS) especially for use in health care research. The classification emphasizes urban distinctions and is unique in differentiating between central and fringe counties of large metropolitan areas. Smaller metropolitan counties are subdivided by population. Non-metropolitan counties are divided simply into micropolitan and non-core categories. The county classifications are based on the Office of Management and Budget (OMB) metropolitan/micropolitan assignments. (1 = Central counties of metro areas of >= 1 million population, 2 = Fringe counties of metro areas of >= 1 million population, 3 Counties in metro areas of 250,000-999,999 population, 4 = Counties in metro areas of 50,000-249,999 population, 5 = Micropolitan counties, and 6 = Not metropolitan or micropolitan counties).

Hospital STRATUM (Stratum used to post-stratify hospitals) identifies rural, urban non-teaching, and urban teaching hospitals.

Patient Disposition indicates the disposition of the patient at discharge, 1 = routine, 2 = transfer to short-term hospital, 5 = transfer to other (skilled nursing, intermediate care, another type of facility), 6 = home health care, 7 = against medical advice (AMA), codes 20 = died and 99 = discharged alive destination unknown were recoded into one category under 99 due to only 3 cases having died.

TRAN_IN indicates that the non-newborn patient was transferred into the hospital (0 = Not transferred in or newborn admission indicated, 1 = patient transferred in from different acute care hospital, 2 = Transferred in from another type of health facility).

Payer indicates the expected primary payer (1 = Medicare and 2 = Medicaid recoded into one category under 1, 2 = private insurance, 4 = self-pay, codes 5 = no charge and other).

Statistical Analyses – I present descriptive statistics first followed by multivariate Poisson regression models since my outcome is a count variable (LOS). Missing cases were deleted listwise resulting in a sample size of 4,645 in-patient stays for pediatric hemophilia patients. Data is weighted to be representative of pediatric discharges from U.S. community, non-rehabilitation hospitals.

Results

Table 2. Descriptive Statistics for Patient-Level Variables (N=4,645)

	Mean or %	SD	min/max
LOS	5.00	9.12	0/228
Age	7.90	6.80	0/20
Sex			
Male	95%		
Female	5%		
Race			
White	49%		
Black	17%		
Hispanic	25%		
Asian/Pacific Islander	3%		
Native American	1%		
Other	5%		
Median Household Income			
0-25%	31%		
26-50%	25%		
51-75%	22%		
76-100%	22%		
Patient Location, Rural/Urban			
Central Metro >= 1 million	37%		
Fringe Metro >= 1 million	22%		
Metro 250K-999,999K	20%		
Metro 50K-249,999K	7%		
Micropolitan	9%		
Non-Metro/Micro	5%		

Primary Payer		
	Medicare/Medicaid	55%
	Private Insurance	37%
	Self-Pay	2%
	No Charge/Other	6%

Table 2 presents the descriptive statistics for the patient-level variables. The mean for length of stay is 5 days with a standard deviation of 9.12 days. Length of stay ranged from zero days up to 228 days. This is much longer than the national mean for pediatric LOS of 1.8 days (Heys et al. 2017). The mean age of pediatric patients was 7.9 years with a standard deviation of 6.8 years. Age range was newborn through 20 years of age. The sex of pediatric patients was overwhelmingly male at 95% with female at 5%. This is not out of the ordinary since hemophilia is a recessive x chromosome disorder and usually presents in males. Racial makeup of pediatric hemophilia patients was 49% White, 17% Black, 25% Hispanic, 3% Asian/Pacific Islander, 1% Native American, and 5% reported as other. This racial makeup is different from the 2019 Census Bureau data which show the population 76% White, 13% Black, 19% Hispanic, 6% Asian/Pacific Islander, and 1% Native American. Soucie et al. (1998) found that the incident rates of hemophilia were similar among whites, Blacks, and Hispanics. Yet these findings point to a difference in hospitalizations based on ethnicity. The difference cannot be just due to the 5% reported other in these findings. The income quartiles ranging from 31% in the 0-25 quartile, 25% in the 26-50 quartile, 22% in the 51-75 quartile, and 22% in the 76-100 quartile. With the US Census reporting the 26-50 quartile being the median for the country.

Looking at patient location, 86% of inpatient stays were from patients who live in metropolitan areas with populations over 50,000, 9% were from micropolitans, and 5% were from non-metropolitan/micropolitan, which for this study is considered rural. The other variable

was type of payee. Medicare/Medicaid accounted for 55% of coverage, private insurance was 37%, 2% were self-pay, and 6% had no charge or other. Insurance or type of payee may contribute to difference in length of stay or type of hospital used.

Table 3 presents the descriptive statistics for the hospital-level variables. Bedsize of hospitals in the study ranged from the smallest with 10% of pediatric hemophilia patients, 20% in the medium category, and the majority, 70%, in the large category. This finding may show the majority of hemophilia patients are seen or transferred to large urban teaching hospitals. In terms of type of hospital, 2% of inpatient stays were at rural hospitals, 9% were at urban non-teaching hospitals and the majority, 89%, were at urban teaching hospitals. Again, this may point to pediatric hemophilia patients being transferred to these large urban teaching hospitals.

When looking at hospitals by region, 17% of pediatric hemophilia in-patient stays were in the Northeast, 20% were in the Midwest, 37% in the South, and 26% in the West. In terms of transfer between hospitals, on discharge, 89% of pediatric hemophilia patients were routinely discharged, 2% were transferred to another short-term hospital, 1% were transferred to other facilities like skilled nursing or rehabilitation hospitals, and 8% to home healthcare. The findings for pediatric hemophilia patients being transferred into another hospital, show 91% with no transfer, 8% were transferred in from another acute care hospital, and 1% were transferred in from another type of facility. In terms of hospital control/ownership, 13% of hospitals were public hospitals, 80% were private non-profit hospitals, and 7% were private for-profit hospitals.

Table 3. Descriptive Statistics for Hospital-Level Variables (N=4,645)

Hospital Location	
Rural	2%
Urban, Non-teaching	9%
Urban, Teaching	89%

Hospital Bedsize		
	Small	10%
	Medium	20%
	Large	70%
Hospital Region		
	North East	17%
	Midwest	20%
	South	37%
	West	26%
Patient Disposition		
	Routine	89%
	Transfer to Short-term hospital	2%
	Transfer to other	1%
	Home Health	8%
Transfer Into		
	No Transfer	91%
	Transfer in from Acute Care	8%
	Transfer in Other	1%
Hospital Control		
	Gov	13%
	Private, non-profit	80%
	Private	7%

I'm interested in whether type of hospital is associated with length of stay. To start, I examine mean length of stay by type of hospital (see Table 4). I find that urban hospitals, whether teaching or nonteaching, have longer lengths of stay for pediatric hemophilia patients compared to rural hospitals: mean LOS is 2.7 days for rural hospitals, 4.6 days for urban hospitals, and 5.1 days for teaching hospitals. This is inconsistent with Hypothesis 1. Across all hospital types, pediatric hemophilia patients have longer lengths of stay compared to the national mean for pediatric LOS of 1.8 days (Heys et al. 2017).

Table 4. Mean LOS by type of hospital

Hospital	Mean
Rural	2.696
Urban Non-teaching	4.617
Urban Teaching	5.060

Table 5 presents Poisson regression models predicting length of stay. Incidence Rate Ratios are presented, such that values above one reflect an increased rate of LOS (a longer LOS) and values below one reflect a decreased rate of LOS (a shorter LOS). There is a significant association between age and LOS in hospital as well as sex and LOS. When age increases by one year, the rate ratio for length of stay is expected to decrease by a factor of 0.9908, while holding the other variables constant in the model. Compared to males, females have a rate 1.9042 times greater for LOS net of the other variables.

When looking at race, Hispanic patients had a 1.1521 higher rate of LOS compared to White patients, while holding the other variables constant in the model. Looking at income quartiles, the rate for LOS is 0.8668 times lower for the 2nd quartile income compared to the 1st quartile income. Transfers from other acute care hospitals had a 1.5350 greater rate for LOS compared to no transfers, while holding the other variables constant in the model. There was no significance difference in LOS for transfers from other medical facilities.

I also looked at hospital control and found that private non-profit hospitals had a 0.8188 lower rate of LOS compared to government run hospitals, while holding the other variables constant in the model. Similarly, private for-profit hospitals had a significantly lower rate of LOS compared to government run hospitals, net of the other variables.

When considering LOS, I also looked at the disposition of patients on release. While there was no significant difference for transfers to short term care facilities ($p= 0.886$), I did find a significant association for transfer to other health care facilities ($p=0.000$) such as rehabilitation

hospitals and nursing care facilities and to home health care. Compared to routine discharge, transfers to other healthcare facilities had a 2.3702 times greater rate of LOS and transfers to home healthcare had a 1.8538 times greater rate of LOS (Table 5).

I was also interested in looking at who paid the hospital charges. Patients with private health insurance had a 0.8958 times lower rate of LOS compared to Medicare or Medicaid, while holding the other variables constant in the model. There was no significance difference in rate of LOS found for self-pay or other pay, such as no charge. No significance differences in rates of LOS were found for bedsize of hospitals or hospital region.

When I looked at location of hospital, rural, urban non-teaching, and urban teaching and LOS, I found a significant difference for urban non-teaching and urban teaching hospitals when compared to rural hospitals. Compared to rural hospitals, urban non-teaching hospitals had a 1.7414 greater rate of LOS and urban teaching hospitals had a 1.7680 greater rate of LOS, while holding the other variables constant in the model.

Table 5. Poisson Regression Models Predicting LOS, Incidence Rate Ratios Presented (N=4645)

Variable	Ref Category		IRR	SE
Age			0.9908*	0.0044
Sex	Male		1.9042**	0.3233
Race	White			
		Black	1.0292	0.0705
		Hispanic	1.1521*	0.0756
		Asian/Pacific Islander	1.2675	0.2388
		Native American	1.1469	0.1704
		Other	0.9771	0.1012
Income	1st Quartile			
		2nd Quartile	0.8668*	0.0554
		3rd Quartile	0.9135	0.0644
		4th Quartile	0.9963	0.0796
Bedsize	Small			
		Medium	1.1376	0.1091

		Large	1.0182	0.0717
Region	Northeast			
		Midwest	0.9588	0.0910
		South	0.9668	0.0815
		West	0.9524	0.0870
Patient Location	Central Urban		0.9874	0.0239
Tran In	No Transfer			
		Acute Care Hosp	1.5350**	0.1329
		Other	1.3954	0.4354
Hospital Control	Government			
		Private Non-profit	0.8188*	0.0755
		Private For-profit	0.7321**	0.0823
Disposition	Routine Discharge			
		Transfer Short-term Hosp	1.0412	0.2938
		Transfer Other	2.3702**	0.4807
		Home	1.8538**	0.1556
Payer	Medicare/Medicaid			
		Private	0.8958*	0.0501
		Self	1.0196	0.1569
		Other	0.9816	0.1004
Hospital Location	Rural			
		Urban Non-teaching	1.7414**	0.2403
		Urban Teaching	1.7680**	0.2162

Note: ** = p< 0.01, * = p<0.05

Discussion

In this study I looked at data from the HCUP Kids' Inpatient Database (KID), Healthcare Cost and Utilization Project (HCUP). Specifically pediatric hemophilia patients' length of stay (LOS) in rural vs urban non-teaching vs urban teaching hospitals. As stated in my hypothesis, I posited pediatric hemophilia patients would have a longer LOS in rural hospitals compared to urban hospitals. My hypothesis was based on research finding that rural hospitals have fewer specialists, less resources, and lack training in rare disorders. However, I found the opposite. These findings may highlight rural hospitals understanding their limitations and quickly transferring pediatric hemophilia patients to urban hospitals better equipped to handle this

disorder. It may also be that pediatric hemophilia patients, aware of the limitations in rural hospitals, will travel to better equipped and knowledgeable hospitals. This raises many questions for further research, such as what are the underlying mechanisms associated with hemophilia patients in rural hospitals having shorter lengths of stay compared to hemophilia patients in urban hospitals.

As pointed out in Table 3 above, there was an increased mean LOS for pediatric hemophilia patients compared to the national mean for pediatric LOS (Heys et al. 2017). Why do pediatric patients with hemophilia spend 3 days more on average in hospital than other pediatric patients? The longer LOS for pediatric hemophilia patients could point to several factors leading to this increase. First, pediatric hemophilia patients may have more complicated medical issues related to hemophilia. Second, pediatric hemophilia patients may require more intensive treatment and monitoring that is not possible as an outpatient. And third, hospitals and staff may not be properly trained or prepared and lack the resources to deal with the specific needs of hemophilia. As has been pointed out, hemophilia is a rare genetic disorder that requires specialty medical treatment and care. This care and treatment require additional training for physicians, hospital staff and patients, to properly treat hemophilia. Healthcare providers who are not properly trained may misdiagnose or not understand how to treat hemophilia, causing a delay of a necessary treatment. The NHF (2019), recommends “Individuals with bleeding disorders should be triaged urgently as delays in administering appropriate therapy, such as infusion of factor concentrate, can significantly affect morbidity and mortality.” A delay in treatment can be catastrophic for a person with hemophilia. They can suffer long term pain, disability, and even death with delayed or improper treatment.

These findings point to the need for more training and resources for hospital staff and hospitals. The National Hemophilia Foundation and the Hemophilia Treatment Centers in the United States can and will provide training when requested. There are programs in place that will visit and train staff. This training consists of proper diagnosis and symptomatic indicators of a hemophilia related condition. The training will also specify the urgency of early treatment as per *MASAC Document 257 - Guidelines for Emergency Department Management of Individuals with Hemophilia and Other Bleeding Disorders* (NHF 2019).

Another area that raised questions was the difference in LOS by sex and age. Is there a difference in diagnosis between male and female hemophilia patients? I would posit there may be a lack of knowledge about hemophilia and the treatment of hemophilia in the hospital system. While it was not possible to ascertain that data from this data set, it may be available elsewhere and should be studied.

Another interesting finding was that Hispanic pediatric hemophilia patients have longer lengths of stay than their White counterparts. This follows what Lasser et al. (2006) found that minorities and immigrants are less likely to have health insurance, seek health care less, and receive improper treatment when sought. Also, the fact that White patients only made up 49% of the sample compared to 76% for the general population is interesting. It is possible that White pediatric patients have better access to preventative care and treatment and thus, require less time in the hospital compared to Hispanic pediatric patients. This would be another area that should be further researched in the pediatric hemophilia community.

Strengths and Limitations

The strength of this study lies in the size and breadth of the data set—it covers 50 states and over 4 million pediatric discharges, including 4,645 pediatric hemophilia discharges. Given

that hemophilia is a rare condition, the large sample size offered by this dataset is an advantage. The study also has several limitations. The data set does not have “transfer out” information for all the years of data I used. There was no way to indicate whether a hospital had a Hemophilia Treatment Center, which may affect LOS. This study was only able to look at rural/urban non-teaching/urban teaching hospital types. Resources, training, specialization of the hospitals were not a part of the data to maintain the anonymity of the hospitals. This information would have added an extra layer of data to understand why LOS was longer for pediatric hemophilia patients in urban hospitals. Another limitation would be the lack of severity of diagnosis on admission which may have a direct effect on LOS and transfer to an urban or teaching hospital. The lack of admission diagnoses also precludes ascertaining the severity of hemophilia and reason for treatment.

Further research is required to examine the differences more fully between male and female pediatric hemophilia patients in their length of stays. It is also important for future research to examine how lack of resources, training, and specialization in rural hospitals affect those with hemophilia or other rare disorders and how race and income affects treatment for hemophilia.

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