Self-Reported Barriers to Hemophilia Care in People with Factor VIII Deficiency

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Background: In 1975, a national network of hemophilia treatment centers (HTCs) was created to increase access to healthcare services for individuals with hemophilia. Studies demonstrate that care in HTCs improves outcomes and reduces costs.

Purpose: The objective of the study was to assess the association of demographic, insurance, and clinical characteristics with self-reported barriers to HTC utilization.

Methods: Data were collected from six HTCs from 2005 through 2007. Adult participants and parents of children aged <18 years were interviewed. Barriers were assessed by asking whether it was difficult to obtain care in the past 12 months. Chi-square test and logistic regression were used to assess factors associated with self-reported barriers to care. All analyses were performed in 2010–2011.

Results: Data for 327 participants (50% adult, 64% severe hemophilia) were analyzed in 2010–2011. Most participants/parents did not report barriers to HTC utilization. However, 46 participants/parents (14%) reported one to six barriers, and 23 reported one barrier. Most frequently reported barriers were "distance to the clinic" for children (44%) and "insurance coverage" for adults (40%). Factors significantly associated with self-reported barriers were: lower income (<$20,000; OR=3.11, 95% CI=1.14–8.45), difficulty finding insurance or obtaining full-year coverage (OR=5.71, 95% CI=2.63–12.41), and decreased state Medicaid coverage for low-income, non-elderly individuals (OR=0.93, 95% CI=0.89–0.98).

Conclusions: This study indicates that, although few people with hemophilia have barriers to care at HTCs, those with lower income, difficulty finding or maintaining adequate insurance coverage, or living in states with lower Medicaid generosity are more likely to report barriers. Identifying and resolving such barriers may improve care access and patient-reported outcomes.

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Background

Inherited chronic disorders that require lifelong clinical management are a public health concern. Historically, the U.S. healthcare delivery system was designed to provide acute, episodic, and curative care or to treat injuries and has not until recently focused on providing long-term management for people with chronic conditions.[1,2] For people with rare chronic genetic diseases, such as hemophilia and cystic fibrosis, primary care is usually insufficient to meet their specialized needs, and access to specialty health services is often limited or fragmented. In addition, these individuals and their families often face physical, emotional, social, and financial challenges throughout their lives. Thus, individuals with inherited disorders require care that is disease-specific, comprehensive, and multidisciplinary, and which in-

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cludes both appropriate medical and psychosocial services. The national hemophilia comprehensive care program in the U.S. has been recognized as one of the most successful comprehensive care approaches for the care of people with inherited diseases.[3]

Hemophilia is a rare genetic bleeding disorder that occurs among one in 5000 male births and affects approximately 20,000 people in the U.S., based on the expected births and deaths since 1994.[4,5] People with hemophilia either are deficient in or are missing clotting factor VIII or IX, which places them at high risk of internal, muscular, and joint bleeding, as well as prolonged bleeding following trauma or surgery. Repeated hemorrhages, especially in people with severe hemophilia (factor activity less than 1% of the normal level), can lead to the development of chronic arthropathy. Over time, this condition can cause joint pain, reduction in joint range of motion, crippling musculoskeletal deformity, and disability. Treatment consists of injecting intravenously the missing clotting factor.

The complexity of treatment and the psychosocial aspects of hemophilia make care in a general hematology department or practice less desirable. In the U.S., a network of federally funded hemophilia treatment centers (HTCs) was initiated in 1975 to provide comprehensive care for people with congenital bleeding disorders.[3] Since the 1980s, the CDC has provided additional funding to implement prevention programs, with an emphasis on risk-reduction practices. Over the years, the original network of 22 HTCs has expanded to more than 130 HTCs, which are organized into 12 regional networks. Treatment centers are currently funded by the Health Resources and Services Administration (HRSA) and by the CDC to provide comprehensive care and preventive services to people with hemophilia and other bleeding disorders.[6]

A core HTC team usually consists of a pediatric or adult hematologist who serves as medical director, a nurse coordinator, a physical therapist, and a psychosocial professional. Additional members may include an adult or pediatric hematologist, dentists, orthopedists, genetic counselors, pharmacists, infectious disease specialists, social workers, and research coordinators. HTCs also provide extensive infusion education and treatment plan development. Many of the larger HTCs increase individuals' access to hemophilia care by operating satellite clinics in rural areas and by offering telephone counseling to patients in remote or underserved areas. Many HTCs are covered entities in the federal 340B Drug Pricing Program, allowing them to purchase clotting factor concentrates at discounted prices, which generates program income that is used to maintain and expand HTC services.

Older studies report 70% of people with hemophilia in the U.S. receive at least some of their medical care from one of these HTCs.[7,8] The benefits of comprehensive HTC care to reduce hospitalization and unemployment rates, and as a consequence, lower the overall cost of hemophilia are known.[9–11] Based on surveillance data gathered by the CDC, people who receive care at HTCs have a 40% deceased risk of death and a 40% decreased hospitalization rate for bleeding complications compared with people who receive care from non-HTC providers, despite the fact that HTCs provide health care to a disproportionately larger share of individuals with severe complications.[7,12] About 30% of all people with hemophilia in the U.S. do not receive care from HTCs. However, this population is difficult to study because each non-HTC care site has few individuals with hemophilia.

Although at least 70% of the population receives care at HTCs, the barriers to HTC utilization have not been formally studied. Identifying barriers to HTC utilization and implementing strategies to increase access to these services are critical to the improvement of outcomes and the reduction of long-term disabilities. The objective is to identify (1) barriers that may prevent individuals from using HTC services; and (2) patient sociodemographic and clinical characteristics associated with barriers to this care.

Methods

Participants and Procedures

This analysis used data from the Hemophilia Utilization Group Study part Va (HUGS Va), a multicenter, observational study designed to examine the cost and burden of illness in people with hemophilia in the U.S. The details of study design, methodology, and baseline data have been described elsewhere.[13] Briefly, data were collected prospectively among people with hemophilia A who received care at one of six HTCs located in California (2 centers), Colorado, Indiana, Massachusetts, and Texas. These sites were selected because they are geographically diverse and representative of HTCs in the U.S., located in populous states, thus serving relatively large hemophilia populations allowing for adequate subject enrollment, and had research personnel willing to participate and conduct IRB review. Participant selection was stratified by level of factor VIII deficiency based on the CDC surveillance report to obtain a closely representative sample of individuals with mild, moderate, and severe hemophilia in each state where the HTCs are located. Eligibility criteria included: (1) aged between 2 and 64 years; (2) factor VIII level $\leq$30%, with or without a history of inhibitor; (3) receiving at least 90% of hemophilia care at the participating HTC; (4) obtaining care at the HTC within 2 years prior to enrollment in the study; and (5) English speaking. Individuals were excluded from participation in the study if they were determined to be cognitively impaired or had an additional bleeding disorder.

From 2005 through 2007, some 329 individuals with hemophilia A were enrolled in the HUGS Va study. After signing informed

consent, adult participants or parents of children aged <18 years completed an initial interview. Individuals completed the survey by self-administration or were interviewed by research staff, so there would be no potential bias in response to the barriers to care questions in the survey. The interview gathered sociodemographic information, health insurance status, perceived barriers to hemophilia care, self-rated joint pain and motion limitation, infusion method, clotting factor utilization, and comorbidities. Clinical data abstracted through chart review included factor VIII activity level, body weight and height, current and past inhibitor levels, history of immune tolerance therapy, hepatitis virus serology, infusion method, and treatment regimen.

Measurement of Barriers to Care

The perception of barriers to HTC utilization among adult participants and parents seen at HTCs was assessed by examining responses to a single question: *In the past 12 months, has there ever been a time that you needed hemophilia care, but it was difficult to get?* Ten specific barriers and one open-ended question were assessed for study participants who reported difficulty receiving care (Table 1).

Covariates

Several factors hypothesized to be associated with barriers to HTC utilization were included in the analysis. For some variables, such as marital status, education level, and employment status, parents' status for participants aged <18 years were combined with adult participants' data. Sociodemographic characteristics included: age; education (high school or less versus beyond high school); marital status (married or with partner versus "other" status); employment (part-time or full-time employed versus unemployed); income (household income <$20,000 per year versus ≥$20,000 per year); and race (white versus nonwhite). To adjust for the effect of comorbidities and health status, clinical characteristics (such as hemophilic severity, history of inhibitors, HIV/AIDS infection, and liver disease/hepatitis) were included in the analysis. Participants in rural areas often face issues of geographic distance and availability of transportation when seeking health care. Therefore, "distance to HTC," which is the distance from the participants' home ZIP code to their regular HTC clinic and HTC outreach clinic, was also included as a variable in the analysis.

Insurance status was assessed using both individual- and state-level data. Two individual-level insurance variables were collected: length of insurance coverage (none or less than 12 months coverage versus full-year coverage) and difficulty finding insurance. Because these two variables measure similar aspects of insurance problems, they were combined into a new variable, health insurance issues, in the multivariable analysis. Studies indicate that the relative generosity of state Medicaid eligibility can influence access to health care, not only for those eligible for Medicaid, but also for all low-income, non-elderly adults who are affected by Medicaid coverage decisions.[14] To account for state variation in Medicaid generosity, state-level Medicaid coverage rates were examined in the states where centers are located. Using the methodology developed by Weissman et al.,[14] the Medicaid coverage rate for each state was calculated as the actual number of low-income non-elderly individuals (<200% of poverty) covered by Medicaid divided by the number of all low-income non-elderly individuals without private insurance. State data provided by the Kaiser Commission on Medicaid and the Uninsured, 2005–2006, were used in the calculation.[15]

Table 1. Types of barriers to HTC utilization[a]

Barriers
1. Distance to the center
2. Transportation to center
3. Insurance does not pay for comprehensive care at HTC
4. Difficulty getting off of work
5. The clinic hours were not convenient
6. You needed someone to take care of your children
7. You would lose pay from work
8. You had a conflict with the staff at HTC
9. HTC staff is not responsive or receptive to your needs
10. Language barrier
11. Other barrier, specify

[a]Options as listed in study questionnaire
HTC, hemophilia treatment center

Statistical Analysis

The proportion of participants who reported any barrier to HTC utilization was calculated, and specific barriers were also identified. Specific barriers reported by adults and those reported by the parents of children aged <18 years were compared. Due to the small number of participants who reported each specific barrier, the subsequent analyses focused on the factors associated with any barrier to HTC utilization. A series of bivariate analyses (chi-square or Fisher's exact test and Student's *t*-test) was run to evaluate the association between overall barriers and each covariate. To identify characteristics associated with any barrier to care, a series of logistic regression models was developed. Univariate logistic analysis was conducted to examine each independent variable separately, and then a multivariable logistic regression containing all independent variables was conducted to adjust for variation in characteristics among the participants.

Because comorbidities occur predominantly in adults, the association of patient characteristics with barriers in children and in adult participants was examined separately. Comorbidities, including AIDS/HIV infection and liver disease, were considered in the model for adults. Participants' history of inhibitor development was not included in the multivariable model because of the high association with hemophilic severity. For categorical variables, the group with fewer barriers to HTC utilization was designated as the reference group. The model's overall significance was tested by likelihood ratio test (χ^2_{Model}). Variables in the model were checked for multicollinearity by using correlation, tolerance, and variance inflation factors. Finally, the Hosmer-Lemeshow chi-square test (χ^2_{HL}) was used to test goodness of fit of the model to the data. All analyses were performed in 2010–2011 using SAS statistical software version 9.2.

Results

Of 329 HUGS Va participants, 327 (99.4%) adult participants or parents of children >18 years who completed the barrier to HTC utilization questionnaire were included in the analysis. About half (50.2%) of participants were adults, and nearly two thirds (64.2%) had severe hemophilia. Mean age was 9.7 years for children and 33.7 years for adults. A total of 32 (9.7%) participants received their usual care at local outreach clinics that are affiliated with two HTCs. For these participants, the average distance to the outreach clinic was 47.8 miles, compared to 271.2 miles if they attended the primary clinic ($p<0.0001$).

Most of the adult participants or parents of children (86%) reported no barriers to HTC utilization. However, 46 participants/parents (14%) reported at least one barrier to HTC utilization (Table 2). Adults were more likely to report having barriers to care than were the parents of children with hemophilia ($p=0.03$). Among those with perceived barriers, 23 participants/parents (50%) reported only one barrier, whereas one participant (2%) reported six barriers. The most frequently reported barrier to care for parents was "distance to the clinic," which was cited by 44% of parents who perceived barriers. Among adult participants, "insurance coverage" was cited as a barrier to care by 40% of adults reporting barriers. Adult participants were more likely than parents to report "insurance coverage" as a barrier to HTC care.

The demographic and clinical characteristics of the study population by overall barriers to HTC utilization are summarized in Table 3. Compared with participants who reported no barriers, those who reported barriers to HTC utilization were more often unemployed (45.7% vs 29.9% among participants with no barriers, $p=0.03$), to be from a family with income <$20,000 (35.7% vs 14.7%, $p=0.0009$), to have no or less than 12 months' health insurance coverage (31.1% vs 6.2%, $p<0.0001$), and to have had difficulty finding insurance (68.9% vs 21.1%, $p<0.0001$). Geographic variations were also found among individuals who reported barriers. Participants from some states were more likely to report barriers to care than those from other states ($p=0.005$) (Table 3). Distance to HTC was not significantly different among participants who reported barriers to care compared with those with no barriers ($p=0.15$). Hemophilic severity also was not associated with participant/parent report of barriers to care. Individuals with liver disease or hepatitis more frequently reported barriers than

Table 2. Comparisons of participant/parent-reported barriers to HTC utilization for children versus adults with hemophilia A (*n*, %)

Barriers	Total sample	Children	Adults
Overall[a]	(*n*=327)	(*n*=163)	(*n*=164)
	46 (14.1)	16 (9.8)	30 (18.3)*
Specific[b]	(*n*=46)	(*n*=16)	(*n*=30)
Distance to the center	16 (34.8)	7 (43.8)	9 (30.0)
Clinic hours were not convenient	14 (30.4)	6 (37.5)	8 (26.7)
Insurance does not pay for comprehensive care at HTC	13 (28.3)	1 (6.3)	12 (40.0)*
Transportation to center	11 (23.9)	5 (31.3)	6 (20.0)
You would lose pay from work	11 (23.9)	3 (18.8)	8 (26.7)
Difficulty getting off of work	8 (17.4)	2 (12.5)	6 (20.0)
You needed someone to take care of your children	4 (8.7)	2 (12.5)	2 (6.7)
You had a conflict with the staff at HTC	4 (8.7)	2 (12.5)	2 (6.7)
HTC staff is not responsive or receptive to your needs	4 (8.7)	2 (12.5)	2 (6.7)
Language barrier[c]	—	—	—
Other barriers	4 (8.7)	2 (12.5)	2 (6.7)

Note: Data are presented as frequency (column %). Barriers for parents of children aged <18 years and those for adults differ significantly.
[a]Response to question: *In the past 12 months, has there ever been a time that you needed hemophilia care but it was difficult to get it?*
[b]The percentages for specific barriers are based on those who reported a barrier to HTC utilization (*n*=46).
[c]No participants reported a language barrier.
*$p<0.05$
HTC, hemophilia treatment center

Table 3. Distribution of characteristics of study population, overall and by self-reported barriers to HTC utilization, *n* (%)

Characteristics	Total sample (*n*=327)	Barriers to care[a]		*p*-value
		Yes (*n*=46)	No (*n*=281)	
Age, years (M [SD])	21.6 (15.2)	23.5 (14.1)	21.3 (15.3)	0.3582
Age group, years				**0.0275**
Child, 2–17	163 (49.8)	16 (34.8)	147 (52.3)	
Adult, ≥18	164 (50.2)	30 (65.2)	134 (47.7)	
Education[b]				0.9424
≤ high school	99 (30.7)	14 (31.1)	85 (30.5)	
> high school	224 (69.3)	31 (68.9)	193 (69.4)	
Marital status[b]				0.7444
Married/with partner	192 (58.7)	26 (56.5)	166 (59.1)	
Not married	135 (41.3)	20 (43.5)	115 (40.9)	
Employment[b]				**0.0338**
Unemployed	105 (32.1)	21 (45.7)	84 (29.9)	
Full-time or part-time	222 (67.9)	25 (54.3)	197 (70.1)	
Income ($)[b]				**0.0009**
<20,000	54 (17.5)	15 (35.7)	39 (14.7)	
≥20,000	254 (82.5)	27 (64.3)	227 (85.3)	
Race				0.9525
White	212 (64.8)	30 (65.2)	182 (64.8)	
Non-white	115 (35.2)	16 (34.8)	99 (35.2)	
Insurance coverage[c]				**<0.0001**
None or <12 months	31 (9.7)	14 (31.1)	17 (6.2)	
Full year	288 (90.3)	31 (68.9)	257 (93.8)	
Difficulty finding insurance[c]	88 (27.9)	31 (68.9)	57 (21.1)	**<0.0001**
Location of HTCs				**0.0045**
California	99 (30.3)	8 (17.4)	91 (32.4)	
Colorado	61 (18.7)	14 (30.4)	47 (16.7)	
Indiana	56 (17.1)	9 (19.6)	47 (16.7)	
Massachusetts	53 (16.2)	2 (4.4)	51 (18.1)	
Texas	58 (17.7)	13 (28.3)	45 (16.0)	
Distance to HTC, miles (M [SD])[d]	45.2 (63.6)	62.8 (94.0)	42.2 (56.8)	0.1530
Hemophilia severity				0.6284
Mild/moderate	107 (35.8)	15 (32.6)	102 (36.3)	
Severe	210 (64.2)	31 (67.4)	179 (63.7)	
History of inhibitors	51 (15.6)	7 (15.2)	44 (15.7)	0.9391
HIV/AIDS	44 (13.5)	9 (19.6)	35 (12.5)	0.1902
Liver disease/hepatitis	110 (33.6)	22 (47.8)	88 (31.3)	**0.0280**

Note: Data are presented as frequency (column %). *p*-values in bold are significant.
[a]Response to question: *In the past 12 months, has there ever been a time that you needed hemophilia care, but it was difficult to get it?*
[b]For participants or parents of child aged <18 years
[c]Data do not add up to *n*=327 due to nonresponse.
[d]Distance to regular clinic or outreach if outreach clinic is available in the area
HTC, hemophilia treatment center

Table 4. Multivariable logistic regression analysis of participant characteristics associated with barriers to HTC utilization

	Model 1 overall (n=302)	Model 2 children (n=149)	Model 3 adults (n=153)
	OR (95% CI)	OR (95% CI)	OR (95% CI)
Aged ≥18 years	1.59 (0.64–3.95)	—	—
Married/with partner	1.41 (0.59–3.38)	0.82 (0.14–4.69)	1.93 (0.66–5.65)
Unemployed	1.15 (0.48–2.76)	1.66 (0.41–6.78)	0.85 (0.25–2.96)
Income <$20,000	**3.11 (1.14–8.45)**	7.55 (0.99–57.77)	2.82 (0.76–10.51)
White	0.92 (0.47–2.76)	2.10 (0.46–9.55)	0.68 (0.23–2.01)
Insurance issues: yes[a]	**5.71 (2.63–12.41)**	2.19 (0.59–8.08)	**10.38 (3.36–32.05)**
State Medicaid coverage, %[b]	**0.93 (0.89–0.98)**	**0.88 (0.80–0.97)**	0.95 (0.89–1.00)
Distance to HTC, miles[c]	1.00 (1.00–1.01)	0.99 (0.98–1.01)	1.00 (1.00–1.01)
Hemophelia severity: severe	1.27 (0.57–2.88)	0.82 (0.21–3.17)	1.80 (0.55–5.89)
Liver disease/hepatitis: yes	—	—	1.54 (0.47–4.97)
HIV/AIDS: yes	—	—	0.62 (0.17–2.24)

Note: For categoric variables, the group theorized to have fewer barriers to HTC utilization was used as the reference group. Bold type indicates significant ($p<0.05$) ORs.
Model 1, $\chi^2_{model}=53.70$, $p<0.0001$, $\chi^2_{HL}=7.95$, $p=0.44$.
Model 2, $\chi^2_{model}=17.41$, $p<0.05$, $\chi^2_{HL}=3.56$, $p=0.89$.
Model 3, $\chi^2_{model}=41.27$, $p<0.0001$, $\chi^2_{HL}=5.25$, $p=0.73$.
[a]Coverage <12 months and/or difficulty finding insurance
[b]State Medicaid coverage refers to the percentage of low-income non-elderly covered by Medicaid as a percentage of all low-income non-elderly without private insurance in 2005–2006. California=47.2%; Colorado=33.6%; Indiana =50.4%; Massachusetts=59.5%; Texas=34.6%.
[c]Distance to regular clinic or outreach if outreach clinic is available in the area
HTC, hemophilia treatment center; χ^2_{HL}, Hosmer-Lemeshow chi-square test

those without these comorbidities (47.8% versus 31.3%, $p=0.03$).

An analysis of barriers by state was also conducted. Univariate analysis indicates that overall rates of barriers to care by state ranged from 4% to 23% ($p=0.005$). Ten specific barriers were compared by state between those who reported barriers and those who did not report barriers. "Distance to the clinic" was reported as a barrier in participants from four of the five states analyzed (5% of all participants), with the percentage by state ranging from 0% to 13% ($p=0.01$). Regarding "transportation to clinic," three states had participants who reported this as an issue (overall rate 3%, range from 0% to 9% by state, $p=0.001$). No other specific barriers were found to vary significantly by state.

Table 4 illustrates the multivariable logistic regression analyses of the association between participant sociodemographic and clinical characteristics and having any barrier to HTC utilization. Only individuals with complete data for all variables in the model were included in the analyses ($n=302$). Among the entire sample, individuals with household income <$20,000 were more likely to report a barrier compared with those with higher household income (OR=3.11, 95% CI=1.14–8.45). Compared to those without health insurance issues (no difficulties obtaining insurance and insurance coverage for an entire year), those individuals with insurance issues (less than full-year coverage and/or difficulty finding insurance) had a 470% higher risk of reporting a barrier to care (OR=5.71, 95% CI=2.63–12.41). Participants attending HTCs in states with lower Medicaid coverage rates for the low-income non-elderly were also more likely to report a barrier to HTC care. Each percentage-point increase in the Medicaid coverage rate for the low-income non-elderly resulted in a 7% decrease in the risk of reporting any barrier to care (OR=0.93, 95% CI=0.89–0.98).

Additional analyses were conducted among groups of children and adult participants separately. Although sample size was reduced in these subanalyses, the results indicate that the generosity of state Medicaid programs was significantly associated with barriers to HTC utilization for children (OR=0.88, 95% CI=0.80–0.97). Adults with health insurance issues (less than full-year coverage and/or difficulty finding insurance) were even more likely to report barriers to HTC utilization (OR=10.38, 95% CI=3.36–32.05) compared with the overall sample (OR=5.71).

Discussion

To our knowledge, this analysis is the first prospective cohort study to examine in a comprehensive manner the barriers to HTC utilization among people with hemophilia A and individual characteristics associated with those barriers. It is important to note that although hemophilia is a chronic disease associated with major physical, social, and financial consequences, only one of seven HTC participants or parents who participated in this study reported barriers to hemophilia care. Some of the barriers reported by individuals included those possibly addressed by the individual centers (e.g., clinic hours, child care), and this feedback was provided. However, other participant characteristics such as income, insurance issues (lack of full-year insurance coverage and/or difficulty finding insurance), and the generosity of state Medicaid eligibility requirements were found to be significantly associated with barriers to HTC utilization and are policy issues that need to be addressed more globally.

In the preliminary analysis, it was found that participants from some states were more likely to report barriers to care than those from other states. This geographic variation was further explored in terms of generosity of state Medicaid eligibility criteria, distance to HTC, and provision of emergency care. Previous research has demonstrated that the generosity of a state's Medicaid eligibility criteria has an impact on access to health care for all low-income non-elderly adults who are affected by Medicaid coverage decisions.[14] Medicaid coverage rates for the low-income non-elderly were used as an indicator of Medicaid generosity, and it was found that a 1% increase in the Medicaid coverage rate results in a 7% decrease in the likelihood of a participant reporting a barrier to care.

Distance to clinic, particularly for those who live in rural areas, can be a barrier to obtaining care, and is not easily overcome due to time and transportation difficulties involved. However, in the multivariable analysis, measured distance to HTC (regular clinic or outreach) was not significantly associated with barriers to HTC utilization after adjusting for other variables. One explanation is that two HTCs in the study sample provide outreach clinics to individuals who reside in rural areas. The availability of outreach clinics can substantially reduce the travel distance. This issue needs further evaluation. Another potential source of differing barriers to care may be access to emergency care that could differ from center to center. It was found that all HTC sites in this study shared a common procedure for providing emergency care or recommending a same-day appointment, and all had a 24-hour call number. Thus, it did not appear that the emergency care provided by the various HTCs differed in a substantial way. Further research using a larger sample size is needed to evaluate geographic variations among individuals with hemophilia.

The data have limitations, and the study results must be evaluated with these limitations in mind. Because all participants in the study received care at HTCs, the study results are applicable only to individuals receiving care from HTCs, and cannot be generalized to all individuals with hemophilia. Older studies have reported that around 30% of people with hemophilia do not receive any hemophilia care from HTCs, and they are more frequently individuals with mild disease or fewer complications.[7] However, no update to this number has been published in the last 12 years. Still, the overall rate of reported barriers to care in this study may underestimate barriers to care for the entire hemophilia population due to the exclusion of non-HTC–treated individuals. Although including the non-HTC–treated group would be informative, this population is also difficult to identify as the number of individuals seen at non-HTC healthcare sites are small. Future studies should address the barriers to care faced by the non-HTC population. Similarly, because only English-speaking participants were included in this study, these results cannot be generalized to the non–English-speaking population.

Additionally, the study's small sample size (a common limitation when studying rare conditions) may result in biased statistical inference. This also makes it difficult to explore the statistical association of specific barriers with sociodemographic, clinical, and state-specific characteristics. Lastly, the study included 20 households with more than one child participant. Although these siblings are individuals with unique ages and clinical characteristics, they shared the same sociodemographics as a household, and most of them (18 households) reported the same level of barriers. After excluding 20 children with the same characteristics as their included siblings, the results of multivariable analyses remained the same. Therefore, the results from the entire study population are reported in this paper.

Conclusion

In conclusion, this study indicates that lower household income, insurance difficulties, and residence in states with lower Medicaid program generosity are associated with self-reported barriers to HTC utilization. The availability of outreach clinics that serve rural communities may reduce the reported barriers related to distance to HTC care, but more study is needed. Identification of barriers to hemophilia care is the first step in identifying policies that will increase access to care, potentially improving patient outcomes. Interventions that promote adequate insurance opportunities for individuals with hemophilia should be a high priority. The Affordable Care Act legislation may resolve some insurance

barriers (eliminating lifetime caps, expanding insurance coverage, reducing annual spending limits, eliminating preexisting exclusions, and expanding eligibility for Medicaid).[16] It also increases payments to providers in rural areas and provides coverage for preventive services.[16] Because of the wide range of financial and professional resources potentially available to HTCs, these providers are in a unique position to assist people with hemophilia who experience barriers to accessing adequate health care.

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