HHS Public Access
Author manuscript
J Telemed Telecare. Author manuscript; available in PMC 2022 March 20.

Published in final edited form as:
J Telemed Telecare. 2021 August ; 27(7): 431–435. doi:10.1177/1357633X19883558.

A feasibility study of telemedicine for paediatric sickle cell patients living in a rural medically underserved area

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Abstract

Introduction: Sickle cell disease (SCD) is the most common inherited haematological disease, with potentially devastating complications. Improvements in therapies have increased the life span of patients with SCD, but this is contingent on receiving timely evidence-based medical care, including regular evaluations with haematologists, disease-specific education and psychosocial care. Our objective was to evaluate the feasibility of utilizing telemedicine for the provision of subspecialty paediatric SCD care in a rural medically underserved area.

Methods: This was a cross-sectional, observational, feasibility study. All patients 0–21 years old with SCD seen at Riley Hospital for Children Comprehensive Pediatric Sickle Cell Clinic who lived within 30 miles of the spoke telemedicine facility were eligible for recruitment. The Telehealth Satisfaction Scale (TeSS) was adapted for the SCD population and administered at each visit.

Results: Ten SCD patients, ranging in age from 10 months to 18 years old, initiated telemedicine visits during this timeframe. Some 60% were lost to follow-up or did not attend >50% of scheduled visits prior to beginning telemedicine visits. Following initiation of telemedicine, all Hb SS patients were started and/or maintained on hydroxyurea. Nine out of 10 patients who participated during this timeframe had a 100% follow-up rate. All who participated rated the comfort and ease of using the telehealth system as good or excellent and would do a telemedicine visit again.

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Declaration of Conflicting Interests
The author(s) declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

Discussion: This study provides critical information to determine the feasibility and acceptability of a telemedicine intervention to aid in SCD care. To our knowledge, this is the first study to examine the effectiveness of telemedicine to deliver comprehensive paediatric SCD care. Future research with a larger sample size is needed to confirm findings of our study, including expansion of telemedicine sites to include more urban areas.

Keywords

Telemedicine; telehealth; sickle cell disease; paediatric; medically underserved

Introduction

Sickle cell disease (SCD) is the most common inherited haematological disease, affecting approximately 1 in every 400 African American newborns.[1,2] The complications of SCD, which include severe pain, acute chest syndrome and stroke, can be devastating. However, improvements in preventative therapies, as well as the utilization of treatments such as hydroxyurea, have increased the life span for patients with SCD, with >93% of children with SCD surviving to adulthood.[3] Improved patient outcomes, however, are contingent on SCD patients receiving timely evidence-based medical care, including regular patient evaluations with haematologists, disease-specific education, psychosocial care, and genetic counselling.

Previous studies have shown that comprehensive medical care can decrease the adverse outcomes associated with SCD, but this type of comprehensive medical care is not easily accessed.[4–6] Results from a large survey estimated that less than 70% of children with SCD receive comprehensive care, and only 20% experience effective care coordination between primary and subspecialty providers.[7] Children with SCD have also been shown to experience difficulty with transportation, extended wait times, and an inability to access subspecialty providers easily.[8] Research suggests that this limited access may contribute to more severe SCD complications and subsequent hospitalizations.[7,9–12] Further complicating issues is the fact that comprehensive sickle cell centres are most commonly located in major metropolitan areas, making it difficult for families in rural or medically underserved areas to access this type of comprehensive care.[6]

Telemedicine, the remote provision of medical care using real-time audio-visual consultation, is a potentially valuable strategy that can address the barriers experienced by sickle cell patients living in rural and medically underserved areas.[13,14] Studies have demonstrated that telemedicine is safe and efficacious and can increase local access to subspecialty care.[13] With other chronic health conditions, such as diabetes, telemedicine has been shown not only to increase adherence/compliance, but also to decrease significant patient outcomes such as stress, anxiety, travel time, and the burden of lost work.[13,15–18] Telemedicine has been used successfully in various adult populations, such as in diabetes, stroke, and critical care. More recently, data regarding the feasibility of telemedicine in paediatric populations has become available.[13,16] Yet, there are still several gaps in the existing literature; the feasibility and acceptability of telemedicine to provide subspecialty care for paediatric patients is not well established, and there are no published data for children with SCD to date. In addition, the barriers to accessing care specifically for

paediatric patients with SCD have not been widely published, and little is known about how caregivers and patients with SCD feel regarding the use of technology like telemedicine for addressing these barriers.[33]

The purpose of this study was to evaluate the feasibility of utilizing telemedicine for the provision of subspecialty paediatric sickle cell care in a rural and medically underserved area.

Methods

This cross-sectional, observational, feasibility study was approved by the Indiana University Institutional Review Board with exempt status. All caregivers of eligible patients were contacted by the sickle cell team coordinator by phone regarding the option of participating in telemedicine clinic visits. A hub and spoke telemedicine model was used, with Riley Hospital for Children serving as the tertiary care centre, or 'hub', with the subspecialty sickle cell team present, and an outpatient clinic in Evansville, Indiana serving as the 'spoke', or remote, rural establishment where patients/caregivers would arrive for their visit.

Participants and sites

The Pediatric Sickle Cell Clinic at Riley Hospital for Children cares for approximately 260 patients with SCD. Nearly 16% of these established patients travel greater than 60 miles to access comprehensive SCD care at Riley, of which close to 5% live near the spoke facility in this study. All patients who had been seen at the Riley Hospital for Children Sickle Cell Clinic in Indianapolis, Indiana over the last 10 years and lived within 25 miles or approximately 30 minutes from the spoke facility in Evansville, Indiana were eligible to participate in our telemedicine clinic. Patients requiring chronic red cell transfusions were not eligible, as they would require regular visits at the hub site for infusion. This resulted in caregivers for 12 patients being contacted.

Tele-consultation equipment

Video consultations were done via a secure software connection using Cisco Jabber and a fixed Cisco end-point (https://www.cisco.com/c/en/us/products/unified-communications/jabber/index.html). This provided two-way videoconferencing between a paediatric haematologist located at our clinic (hub) and a remote clinic office (spoke) where the patient and caregiver and a staff medical assistant were located. The sites were connected via a dedicated secure broadband Internet connection with encrypted data exchange that requires a username and password at both the hub and spoke sites to protect confidentiality. The equipment was operated by the paediatric haematologist and the staff nurse/nurse practitioner, not the patient.

Procedure

All patients were scheduled for a telemedicine visit at the Evansville office at least 2 weeks in advance. Once the patient and caregiver dyad arrived at the spoke office, a standard consent for treatment was obtained. The patient and caregiver(s) were then escorted into a clinic room where the telemedicine equipment was available and functioning. The patient

and caregiver(s) were oriented to the equipment and how the visit would proceed. A physical exam was performed by a consistent licensed Pediatric Nurse Practitioner, who would remain in the room while the sickle cell provider from Indianapolis performed the remainder of the telemedicine visit via video. The visit consisted of reviewing interim history, past medical, family, and social histories, medications, and laboratory results. If specific school, financial, or leave concerns were expressed, the sickle cell team's school educator, nurse coordinator, and/or social worker were able to join the telemedicine visit remotely to address concerns in a timely fashion.

Data collection

Visit information, including who attended the visit, was documented by the sickle cell provider. Patient demographics were collected through medical record extraction, including gender, race, age, sickle cell genotype, zip code, payer information, hydroxyurea use, and recent hospitalizations. The validated Telehealth Satisfaction Scale (TeSS) was adapted for the SCD population and administered at the end of each visit to caregivers.[19]

Data analysis

Given the exploratory nature of this observational feasibility study and the small sample size, only simple descriptive statistics were calculated. For nominal and ordinal data, frequencies and percentages were reported. In the case of ratio data, mean and ranges were reported.

Results

Demographic data/patient characteristics

Of the caregivers approached regarding participation in telemedicine visits, only one refused due to lack of availability of a specific hub provider for telemedicine visits, and one was not able to be reached by phone. Ten patients with SCD, ranging in age from 10 months to 18 years old, initiated telemedicine visits during this timeframe. Four of the 10 patients who participated in telemedicine visits had Hb SS disease, five had Hb SC disease, and one had Hb S Trait (Table 1); 50% of patients were female. All lived greater than 160 miles from our main hospital in Indianapolis. Some 60% had been lost to follow-up or did not attend >50% of scheduled sickle cell visits prior to beginning telemedicine visits. Table 1 illustrates characteristics of all telemedicine patients seen at our centre during this time period.

Nine out of 10 patients who participated in telemedicine visits during this timeframe had a 100% follow-up rate, and 100% of patients who participated missed no more than one visit. Some 90% of patients had more than two telemedicine visits during this time period. All patients under the age of 5 years old were prescribed penicillin and had caregiver-reported compliance with it. All Hb SS patients who participated in telemedicine were started and maintained on hydroxyurea, with three of the four starting hydroxyurea during this time. All patients were up to date on vaccinations, including all receiving influenza vaccines.

Patient satisfaction

All caregivers participating in telemedicine had the opportunity to complete the 10-question satisfaction survey (TeSS) (Figure 1). Each response was rated as Excellent, Good, Fair, or Poor. 100% of respondents stated the quality of the equipment as good or excellent (Table 1). 100% of respondents rated the comfort and ease of using the telehealth system as good or excellent. 100% of caregivers responded good or excellent to questions regarding the courtesy, thoroughness, and communication of the sickle cell team. In addition to answering the modified-TeSS, caregivers were asked if they would do a telemedicine visit again, all of whom answered 'yes'.

Discussion

Our study demonstrates the feasibility of a telemedicine intervention to aid in SCD care. In addition, to our knowledge, this is the first study to examine the feasibility and acceptability of telemedicine to deliver comprehensive paediatric SCD care.

Since its first use in the 1960s by NASA as a means of monitoring astronauts, telemedicine has been a rapidly growing facet of healthcare in the US.[35] Studies have demonstrated that telemedicine is safe and efficacious, and increases local access to subspecialty care.[13] Marcin et al. demonstrated that telemedicine was a viable method of providing care to children with special healthcare needs, as well as a method to address health disparities in rural communities.[13,14] Telemedicine has also been used in other avenues, such as adult diabetes care, to provide improved availability of subspecialty care to remote areas.[13,18] Telemedicine has been shown to improve satisfaction for patients and primary care providers; it also has been demonstrated to increase the availability of care, decrease stress, decrease lost wages/cost, and increase adherence with medications such as insulin.[13,18,20,36]

Children with chronic diseases who live in rural and medically underserved communities have been shown to experience a greater disparity in accessing comprehensive care, which can cause caregiver and primary care provider stress.[14,33,34] This is particularly true for patients with SCD, as a large proportion of these patients are from rural and urban medically underserved communities, and therefore do not have easy access to comprehensive sickle cell centres.[6] In the State of Indiana, for example, the only comprehensive children's hospital in the state is in Indianapolis, the state's largest metropolitan area, which is approximately 145 miles from the northern and southern cities of the state. This comprehensive children's hospital, Riley Hospital for Children at IU Health, cares for more than 260 families with SCD, and sees approximately 30 newborns in the state each year with the disease. These patients come from all over the state, as well as parts of Illinois, Kentucky, and Ohio. Well over 16% of families travel more than 60 miles in order to access comprehensive SCD care. For many of these families, an appointment requires a full day off work, half a day of travel, and transportation costs. Therefore, initiating therapies such as hydroxyurea and ensuring compliance with its frequent laboratory testing and visits is often not feasible for these families. Telemedicine is a potentially valuable strategy that can address the access barriers experienced by sickle cell patients living in rural and medically underserved areas.[13,14]

The provision of high-quality, evidence-based care for individuals with SCD has become a global priority, at the forefront of national organizations such as the American Society of Hematology (ASH) and the National Heart, Lung, and Blood Institute of the National Institutes of Health.[26] In fact, one of the four components of the recent *State of Sickle Cell Disease Report* by ASH is improving access to care for patients with SCD to reduce healthcare disparities.[26] Telemedicine is becoming more accepted among healthcare professionals, and the American Academy of Pediatrics has recently recognized telemedicine as an important strategy for increasing access to paediatric subspecialty care.[19]

Limitations of this study include a small sample size, so results may be difficult to generalize to the wider SCD population. However, the variability of telehealth interventions allows for adaptation to the patient population and clinical setting. In addition, this study looked at a single rural medically underserved area with a single, consistent SCD specialist at the hub and nurse practitioner at the spoke. The feasibility of telemedicine may be different in urban versus rural areas, and varied or inconsistent providers, especially as it relates to patient acceptability, provider availability, facilities, and resource utilization.[20] Future research with a larger sample size is needed to confirm findings of our study, including expansion of telemedicine sites to include other rural, as well as urban areas.

Conclusion

Gaps remains in providing comprehensive sickle cell care to children with SCD. This pilot provides a novel and feasible solution to bridging this gap, particularly for patients living in rural and/or medically underserved areas. The patients and caregivers were satisfied with the overall experience of using telemedicine. Additional study is warranted to improve its ease of use and expansion to other regions. Future directions will include an emphasis on adapting the programme with the use of caregiver and patient input, as well as evaluation of patient outcomes.

Acknowledgements

We would like to thank the staff at Deaconess Outpatient Center in Evansville, Indiana and the IU Health Telehealth Department for their support of this project.

Funding

The author(s) disclosed receipt of the following financial support for the research, authorship, and/or publication of this article: Dr. Jacob received funding support from the AHRQ LHS K12 (1K12HS026390-01).

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How satisfied were you with:

1. The voice quality of the equipment
   Excellent Good Fair Poor
2. The visual quality of the equipment
   Excellent Good Fair Poor
3. Your personal comfort in using the Telehealth system
   Excellent Good Fair Poor
4. The ease of getting to the telehealth department
   Excellent Good Fair Poor
5. The length of time with the Sickle Cell team
   Excellent Good Fair Poor
6. The explanation of your treatment by the Sickle Cell team
   Excellent Good Fair Poor
7. The thoroughness, carefulness, and skillfulness of the Sickle Cell team
   Excellent Good Fair Poor
8. The courtesy, respect, sensitivity, and friendliness of the Sickle Cell team
   Excellent Good Fair Poor
9. How well your privacy was respected
   Excellent Good Fair Poor
10. How well the staff answered your questions about the equipment
    Excellent Good Fair Poor

Figure 1.
Modified Telehealth Satisfaction Scale (TeSS).

Table 1

Characteristics of patients participating in telemedicine.

	N (%)
Age (mean)	8.5 years (± 5.7 years)
Female	4 (40%)
Race	
Black	10 (100%)
Genotype	
Hb SS	4 (40%)
Hb SC	5 (50%)
Hb S Trait	1 (10%)
Payer	
Public	8 (80%)
Private	2 (20%)
Hydroxyurea Use	
Prior to Telemedicine	1 (25%)*
After Telemedicine	4 (100%)
Average Distance from Subspecialty Center	167.7 miles
Patient Satisfaction (Good or Excellent)	
Voice Quality of Equipment	9 (90%)
Visual Quality of Equipment	10 (100%)
Comfort with Telemedicine	10 (100%)
Ease of Getting to Telehealth Site	10 (100%)
Length of time with Team	10 (100%)
Communication by Team	10 (100%)
Thoroughness of Team	10 (100%)
Courtesy and Respect of Team	10 (100%)
Privacy Respected	10 (100%)
Questions about Equipment Answered	10 (100%)
Would do Telemedicine Visit Again	10 (100%)

*Percentage of eligible patients.