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Telemedicine in cystic fibrosis

Marisa E. Desimone^{a,*}, Jordan Sherwood^b, Sarah C. Soltman^c, Antoinette Moran^d^a Division of Endocrinology, Diabetes and Metabolism, State University of New York at Upstate Medical University 750 East Adams Street, Syracuse, NY 13210, USA^b Division of Pediatric Endocrinology, Massachusetts General Hospital, 55 Fruit Street, Boston, MA 02114, USA^c Division of Endocrinology, Diabetes and Clinical Nutrition, Oregon Health and Science University, 3270 SW Pavilion Loop, Portland, OR 97239, USA^d Division of Pediatric Endocrinology, University of Minnesota, 2512 S 7th Street, Minneapolis, MN 55454, USA

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ABSTRACT

Cystic Fibrosis (CF) requires lifetime multidisciplinary care to manage both pulmonary and extra pulmonary manifestations. The median age of survival for people with CF is rising and the number of adults with CF is expected to increase dramatically over the coming years. People with CF have better outcomes when managed in specialty centers, however access can be limited. Telemedicine and technology-based care solutions may help to overcome barriers to availability and improve access. This review outlines the use of telehealth for CF management. Telehealth has been utilized for CF across a broad variety of indications, even prior to the COVID-19 pandemic, and in general has been well accepted by patients and providers. There are a paucity of data, however, related to health outcomes, and the healthcare utilization specific to CF and its related comorbidities. Future studies are needed to address the questions of health outcomes, cost, burdens of telehealth and barriers to implementation.

Introduction

Cystic Fibrosis (CF), due to its progressive nature and multi-system manifestations, requires lifetime multidisciplinary care. People with CF have better outcomes when managed in specialty centers, however access can be limited by distance and availability [1–2]. In addition, due to the changing demography of CF there has been an increase in the number of adults living with CF and its complications, thus increasing the need for specialist availability [3–4]. The current and future challenges of meeting patient care needs by specialist centers will require new models of care delivery; technology-based solutions to bridge gaps in access and availability have been evaluated as possible solutions to these challenges.

Telemedicine is the remote delivery of health care services using telecommunications technology. Telemedicine can be delivered in a variety of ways including: (A) Virtual visit: a live (synchronous) interactive encounter between patient and provider, (B) Chat-based interactions: an asynchronous online or mobile app communication to transmit health data for review, diagnosis or treatment plan at a later time, (C) Remote patient monitoring: the collection and transmission of data to a provider or healthcare team for synchronous or asynchronous chronic disease management, (D) Store and forward: the

asynchronous transmission of patient health data for later review and diagnosis [5].

There has been significant interest in utilizing telehealth for disease management even prior to the coronavirus 2019 (COVID-19) pandemic, and there is a robust and growing body of literature on this topic. Studies have evaluated telehealth use in both adult and pediatric populations. Because studies on this topic are heterogeneous in the population studied, the method of telehealth used and the outcomes evaluated, it can make drawing conclusions and generalizations difficult. However, several themes have emerged that support the use of this mode of care. Telehealth may offer benefits compared to in-person visits (Table 1). Telemedicine may allow for more frequent contact with the healthcare team, has the potential to improve access to specialist care, can reduce the cost of obtaining care by removing the need for patients and their families to travel, may reduce the exposure of high-risk patients to infection in the clinic, and may help to improve patient engagement and adherence, thus improving outcomes [6–21].

* Corresponding author.

E-mail address: desimoma@upstate.edu (M.E. Desimone).<https://doi.org/10.1016/j.jcte.2021.100270>

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Table 1

General benefits of and barriers to telemedicine.

Benefits of Telemedicine:
More frequent contact between patients and providers
Improved healthcare access
Reduced travel costs
Reduced time away from work/school
Reduced infection risk
Improved patient engagement
Improved patient adherence
Barriers to Telemedicine:
Limited evaluation due to lack of physical exam
Security of data transfer
Provider licensure requirements
Patient location requirements
Disparities in high speed internet access
Disparities in technology adoption
Availability of interpreter services
Ability to teach patients & caregivers proper device use

Telemedicine in general

Telehealth logistics and practical considerations

Effective delivery of telehealth requires both patients and clinics to have the necessary digital infrastructure, including hardware and software, support personnel and dedicated clinician space (Table 2). Data from self-monitoring devices must be uploaded by patients prior to the visit so that these measures can be available to the care team. Often these require proprietary software, although third party platforms are available which can integrate and upload multiple device types and brands [22]. In some cases, self-monitoring devices upload data directly to cloud-based storage systems in real time, and can be visualized by clinicians [23]. Video visits require high-speed internet as well as a device for connection, such as a smart-phone, tablet, laptop, or desktop computer. Clinicians require secure dedicated space in the clinic or at a secure home office in order to conduct a visit as well as an electronic health system with an embedded, HIPPA compliant platform or third-party system to conduct a virtual visit. Support staff are needed to ensure patients are prepared for the visit and remotely checked in to the encounter to ensure necessary device data are available prior to the visit start. Information technology support staff are required to troubleshoot any issues that may arise in real time to ensure a successful visit. Billing systems must be established with payors to ensure reimbursement.

Barriers to Telehealth

Although telehealth may serve as a useful modality to deliver effective healthcare there are several medical-legal considerations that must be considered (Table 1). Remote visits lack a physical exam, thus diagnosing and treating new conditions may be limited. HIPPA and patient privacy concerns must also be taken into account and the use of secure modes of data transfer are needed. Location of the patient at the time of telehealth visit and state of licensure of the treating physician need to be considered to ensure the visit adheres to licensure

Table 2

Necessary components for both patients and providers for a successful telemedicine visit.

Necessary Components for Successful Telemedicine:
Ability for patient to upload self-monitoring devices for clinician review
High speed internet connection
Device to connect to the visit (smartphone, tablet, laptop or desktop computer)
Secure and dedicated clinician space
HIPPA compliant platform to conduct virtual visit
Support staff to prepare patient
IT support
Billing systems to ensure reimbursement

requirements. Prior to the COVID-19 pandemic telehealth visits were not available to most patients and the U.S Centers for Medicare and Medicaid Services had strict requirements for eligibility for reimbursement. Reimbursement was largely only available to those in designated rural areas [24]. With the emergence of COVID-19 pandemic and the need for physical distancing to decrease rates of transmission, many policy barriers have been lifted [25]. In many states virtual visits are being reimbursed at parity with in-person visits and restrictions on the physical location of patient and physician have been temporarily modified. Long-term changes to telehealth billing and licensure remain to be determined.

Inequities in healthcare access are multiple, and those who might stand to benefit from telehealth may have the most barriers. There is evidence for less adoption of technology and high-speed internet access amongst older patients, minority groups and those with lower socioeconomic status [26–27]. This digital divide may further exacerbate inequities in the post-COVID –19 landscape of widespread telemedicine use. A recent study of 148,402 unique patient telemedicine visits for primary and subspecialty ambulatory care during the early phase of COVID-19 found that older, Asian and non-English speaking patients had lower rates of telemedicine use in general, while older, female, Black, Latinx and poorer patients had less video use [27]. A separate study in patients with type 1 diabetes in the early phase of the COVID-19 pandemic found lower telemedicine use among non-English speaking, and Medicaid-insured pediatric patients [28]. Visits that require a language interpreter may be particularly challenging when they are not conducted in person, as are visits to teach patients how to use a device. Disparities in telemedicine utilization warrant further study.

Outcomes and economics of Telehealth

Telehealth may offer several benefits compared to in person visits. Regionally, access to specialists is limited in rural regions. Remote visits provide patients access to needed specialty care, can decrease transportation costs and reduce lost wages due to time away from work [8–9]. Telehealth modalities including phone calls, texting, and web-based interactions may allow for more frequent patient contact and review of patient data allowing patients to be more engaged with their care team, allowing for increased education and points of intervention [6–7]. Multiple randomized-controlled trials (RCTs) have examined clinical outcomes of telehealth interventions compared to in-person care and the comparisons have generally been favorable [29–30]. Broadly speaking, studies conducted to date have analyzed various telehealth modalities including synchronous and asynchronous interventions, remote monitoring, as well as remote screening for complications. Studies comparing telehealth interventions are lacking and further data are warranted.

General cost effectiveness

Many studies have attempted to quantify the cost-effectiveness of telehealth interventions. Methods to analyze the economic benefit vary and are often based on short time horizons. Depending on the modality of the intervention and the patient population, cost-savings varied with some interventions resulting in cost savings and other resulting in increased healthcare expenditures [10,31–33]. From the patient perspective, telehealth reduces travel-time and lost time from work and these considerations should be considered when evaluating the overall economic costs [10–11]. Real-world long-term economic data on telehealth modalities is lacking and further data are needed.

General patient and provider perspectives

Patient reported outcomes and satisfaction have been a focus of healthcare systems outcomes. Uptake and use of telehealth formats require buy-in from both patients and healthcare systems for its successful implementation. Patients and their caregivers have reported

satisfaction with telehealth noting increased communications, ease of use and reduction of travel time [12–15]. Healthcare organizations and physicians have embraced technology given its potential decreased wait times, improved no-show rates, increased medication adherence and decreased rates of admission [18–21]. Successful implementation of telehealth requires investments from hospital systems and administration.

Overview of telehealth for the management of diabetes

The use of telehealth in the field of diabetes management has been utilized for care delivery for decades and is in many respects a paradigm for telehealth delivery [34]. Effective diabetes self-management requires a combination of factors including blood glucose monitoring, medication adherence, as well as incorporating healthy lifestyle factors such as dietary habits and regular physical activity [35]. Adequate glycemic control is critical to mitigate the risk of long-term macro- and microvascular complications [36]. Diabetes care involves a team-based approach with the integration of healthcare professionals including endocrinology, diabetes nurse educator, nutrition, podiatry, psychology, and ophthalmology, among others. Telehealth is one potential modality that can allow patients to remotely access multiple specialists for co-management of this chronic disease.

Diabetes self-management requires patients to capture high density data including daily self-monitoring of blood glucose levels and/or continuous glucose monitoring, as well as insulin dosing via multiple daily injections and/or continuous subcutaneous insulin infusion (pump therapy). The data requires frequent interpretation by the care team for medication titration to ensure patients are maximizing time in target glucose ranges. Medication needs frequently change over time due to changes in body mass, puberty, activity level, dietary factors, concomitant medication usage and intercurrent illness. Telehealth lends itself as a potential modality to facilitate care in diabetes by increasing contact between patients and practitioners to improve overall care. Increased patient-provider contact in patients with diabetes is associated with improved outcomes [37]. Many patients in remote and rural areas may lack access to specialty care, and travel and time lost from work can be minimized with virtual visits [38]. Telehealth interventions represent a potential modality to increase patient contact and remove barriers to care.

Several *meta*-analyses have been published reviewing diabetes clinical outcomes and, in general, telehealth interventions have been associated with reduced hemoglobin A1c in patients with all forms of diabetes and age ranges [39–51]. A recent umbrella review by Timpel et al. included 46 systematic reviews and *meta*-analyses of randomized controlled trials, most published since 2015. They reported that telemedicine interventions lead to a statistically significant and clinically relevant reduction rate for hemoglobin A1c (HbA1c) of $\leq -0.5\%$. Higher reduction rates were found for interventions that were frequent, intense and of shorter duration (≤ 6 months). Higher reduction rates were also found in patients who were younger, more recently diagnosed with diabetes and with higher baseline HbA1c ($>8\%$). Of note, they assessed that the overall and subgroup-specific certainty of evidence was low to very low [46]. Thus, telehealth has the potential to improve clinical outcomes in patients with diabetes, though certain intervention types and populations may produce more positive results and should therefore be preferentially targeted.

There may be a role for telehealth in screening for and monitoring of diabetic microvascular complications. In particular, many studies have shown that the use of telemedicine for retinal screening using digital retinal photos is beneficial and cost-effective [52–56]. A study by Smith-Strom et al. found that the adjunctive use of telehealth for monitoring of patients with diabetic foot ulcers was noninferior to standard care [57].

Beyond these outcomes, telehealth has the potential to improve psychosocial outcomes in diabetic patients. Nobis et al. reported that a guided, web-based intervention to reduce depression in adults with

diabetes reduced both depressive symptoms and diabetes-specific emotional distress [58]. In another study on young adults with type 1 diabetes by Bakhach et al., group home telemedicine reduced diabetes distress and improved diabetes self-efficacy and diabetes-specific communication [59].

Studies have shown that there is measurable patient time savings using telehealth for diabetes care, especially in rural areas. A study in the VA system by Xu et al. implementing telemedicine for 32 type 1 diabetes patients in rural Alabama and Georgia reported a median time savings of 78 min travel time each way [60]. In a study by Raymond et al. young adults with type 1 diabetes reported saving over 6 h from their work or school day when completing their diabetes clinic visit virtually instead of in-person [61].

In general, patients express satisfaction with telehealth [51,60–66]. A recent systematic review of patients with type 2 diabetes identified time-saving, access and support as the key identifiable factors which led to higher patient satisfaction with telemedicine [62]. A study by Scott et al. surveyed 7477 patients globally with type 1 diabetes during the COVID-19 pandemic and reported that 86% of patients found remote appointments useful and that they would agree to use telehealth again in the future [65]. Lee et al. performed in-depth and focus group interviews with patients who had used telemedicine for type 2 diabetes. Patients were primarily positive about the benefits of telemedicine, though reported obstacles related to cost, internet connectivity and difficulties experienced with system interface [63].

It appears that providers also find telemedicine to be a satisfactory method of care for patients with diabetes [66–67]. A survey of predominantly rural primary care providers involved in the use of telemedicine for Medicare patients with diabetes in medically underserved areas of New York found that the primary care providers (PCPs) had a positive experience with the program [63]. Other studies have also demonstrated PCP satisfaction with and acceptance of telemedicine consultations [68]. Studies are needed to assess endocrinologist satisfaction with providing telemedicine consultations as well as provider satisfaction with the use of telehealth for diabetes care during the COVID-19 pandemic.

Cystic fibrosis and telemedicine

The use of telemedicine in CF has been evaluated in numerous studies. Most are small feasibility studies, with heterogeneous participants and outcomes evaluated. One review by Cox et al. evaluated eight studies of telemedicine specific to CF and found that telemedicine was feasible, patients were willing to utilize telemedicine and there were no concerns regarding data transmission or adverse outcomes [69]. While this is encouraging, the studies included clinically stable patients, with many failing to provide data at required time points, thus limiting generalizability [69].

Exercise interventions

Routine physical activity is recommended for people with CF in order to improve quality of life and outcomes [70–72]. Adherence with exercise regimens is often poor, however, and optimal strategies to overcome barriers have not been established [73]. Several small studies have demonstrated feasibility of and favorable patient perspectives on telemedicine exercise interventions [74–77]. Benz et al. showed that home-based physiotherapy resulted in a more frequent return to baseline lung function in pediatric patients recovering from an exacerbation (49% hybrid care model vs. 32% standard care model) and had considerable savings regarding travel time and travel distance [78]. A review of four of RCTs of exercise interventions in people with CF found that counselling over six months improved participation in physical activity, however, no training program showed improved quality of life [73]. Active video game (AVG) -based interventions have also been evaluated, as they may be more appealing and engaging way for people

to participate in physical activity. A review of twelve AVG-based interventions found similar or improved exercise capacity compared to traditional exercise, and the interventions were significantly more enjoyable compared to traditional programs; however, there was limited evidence for long term effectiveness and adherence [79].

Psychological interventions

The psychological burden of cystic fibrosis is well described and can include depression, anxiety and decreased quality of life [80–81]. Intensive treatment regimens and multisystem complications, as well as shortened life expectancy are all contributing factors. One small pilot study found that self-efficacy could be improved with use of a mentoring program and mobile phone app, compared to mentoring alone or control group [82]. O'Hayer et al. evaluated delivery of acceptance and commitment therapy (ACT) via telehealth and found that it was feasible and potentially effective for treatment of anxiety, depression and improving psychological flexibility [83]. A review by Goldbeck et al. of 16 studies of psychological interventions for people with CF and their families found that targeting specific illness-related challenges can be effective, however, there was heterogeneity between studies regarding type of intervention and the specific issue targeted [84]. E-health interventions for treatment of anxiety and depression has also been an area of evaluation. A review of e-health in children and adolescents found that overall there is low quality evidence for treatment of depression, anxiety or change in quality of life and concluded that while telemedicine in patients with CF holds promise, there is insufficient evidence at this time to conclude on its efficacy [85].

Improved care access

A particular appeal of telemedicine is its ability to expand access to subspecialty care by preventing the need to travel long distances. One study evaluating the uptake of telehealth for individuals with CF from rural areas found that access to care improved, with over 90% of participants attending clinic visits and 2/3 of participants choosing telehealth over in-person clinic visits [86]. There was increase in healthcare utilization in this study, with increases in intravenous antibiotic days, hospital admissions and admission days per participant [86]. While this may be adverse from a cost standpoint, early detection and treatment of exacerbations may improve long term outcomes by helping to preserve lung function [87].

Improved communication

Patients with CF value a close relationship with their clinical care team and are used to frequent contact [88]. Telemedicine and mobile technologies may assist with improving patient education and communication with the specialty team. Video chats and text messaging have been shown to be feasible and acceptable for communication [89]. Smartphone applications have been used to report symptoms of pulmonary exacerbations, and showed a decrease in median time to detection of exacerbations [90]. Web-based symptom screening has also been utilized with success for monitoring of symptom distress and advance care planning for those enrolled in palliative care [91]. A study by Dhingra et al. used an online screening system to assess physical and psychological distress symptoms, and found that it could identify patients who may benefit from immediate medical or psychological attention [92]. This model carried a high burden of time and effort investment by CF center staff, and the authors suggested that modifications to improve efficiency could be considered.

Improved medication adherence

Patient adherence to medication and treatment regimens is often suboptimal; estimations of medication usage indicate that 30–50% of

medicines are not taken as prescribed, and adherence declines over time [93–94]. Average patients with CF are prescribed seven or more medications per day with over 100 min per day spent on pulmonary treatment [95]. The complexity and time-consuming nature of CF care makes adherence even more challenging. One study of over 3,000 people with CF found that adherence was 48% and decreased with age, especially during the adolescent time period [96]. Lower treatment adherence is associated with worse health outcomes, making this an important target for advancing health in the CF population [96]. Text message reminders have been shown to improve adherence in children and adolescents with moderate baseline medication usage [97]. A pilot study of a mobile health app in adolescents found that the medication reminder function was used the most and perceived most useful, however there was significant reduction of use over time and most did not want to continue using the app [98]. One study of home spirometry with weekly telephone calls and text messaging showed improved medication adherence in adolescents with CF [99]. However, another study of home spirometry in adults resulted in poor adherence and reports of high treatment burden [100]. A pilot study of electronic nebulizer adherence monitoring paired with interventionist support tailored to patient need showed improved adherence to nebulizers in adults with CF [101]. These differences in outcomes highlight the importance of personalized empowerment interventions that are carefully considered, in order to avoid additional burdens on individuals with already complex treatment regimens.

Cost savings

Telemedicine may help to reduce the cost of CF care. Pulmonary consultations via telemedicine to patients in underserved rural areas resulted in cost savings of \$US 1000/patient compared to in-person visits [102]. An Italian cost analysis showed benefits over time when telemonitoring was used to follow patients with CF at home [103]. Additional long-term data on the cost savings of telemedicine in CF across different healthcare systems and with use of multidisciplinary teams is needed.

Patient and provider perspectives

Overall, patients with CF have favorable satisfaction with telemedicine across a variety of ages, settings and countries [104]. Pediatric CF patients and their caregivers report high satisfaction with telehealth overall [104]. In one study of home telemonitoring in adults 70% of the participants reported being pleased that they were able to evaluate their own health, and 80% desired continued use in the future [105]. A study of adult patients in rural and remote areas of western Australia showed that most (94%) of participants were satisfied with telemedicine care delivery [86]. CF clinician satisfaction has been reported to be favorable (89%), with the majority reporting a positive impact on clinician patient relationship (57%) and improved efficiency (56%) [106]. Despite these positive perspectives, clinicians were concerned over missing components of routine assessment that are done in-person, such as physical exam, vital signs, pulmonary function testing and cultures, as well as technological limitations of telemedicine [106].

COVID-19 considerations in CF

The COVID-19 pandemic has become a serious world health problem that resulted in many healthcare systems rapidly transitioning to telemedicine in order to allow social distancing and reduction in viral transmission. Reports of swift and successful transition from in-person to telemedicine and hybrid multidisciplinary CF clinics have been published [107–110]. A recent survey of patients with chronic health conditions found that during the COVID-19 pandemic patients with CF had the highest telehealth utilization of all groups, perhaps driven in part by the finding that they reported perceiving a higher risk of severe disease

due to COVID-19 [111]. COVID-19 infection is more likely to be severe in those with underlying comorbidities such as lung disease and diabetes [112]. Patients with CF are more likely to have severe infections due to viral respiratory infection [113]. Despite these risks, the incidence of COVID-19 has been lower in CF than the general population. This may be due to familiarity with infection control measures such as social distancing and use of personal protective equipment [114]. As of July 1, 2021, there have been 1,550 cases of COVID 19 identified in CF patients in the United States, with 14 deaths [115].

CF specific concerns

Cystic fibrosis related diabetes (CFRD) is one of the most common extrapulmonary complications of cystic fibrosis, and its incidence is likely to increase with the increasing longevity of the CF population [116]. Despite the multitude of literature describing the use of telemedicine for diabetes, there are no outcome data for the use of telemedicine for CFRD. While much of the literature for telemedicine in diabetes is positive, extrapolation to CFRD is imprudent. Specific studies evaluating use of telemedicine in CFRD and outcomes are needed.

Part of the routine CF care includes evaluation for possible pulmonary exacerbations. This can be challenging, however, as the definition of a respiratory exacerbation is not standardized [117]. One concern with telemedicine is that the detection of pulmonary exacerbations by daily home spirometry measurements and symptom score may not be accurate [118–120]. Daily monitoring of spirometry can vary up to 13%, and may mask the decline seen with exacerbations [120]. In addition, the evaluation of symptoms is subjective, and the variation in symptom score has not been validated in adults with CF. Lack of physical exam components (lung examination, pulmonary function tests, sputum collection, vitals and weight) could potentially hinder the ability to detect new pathogens or diagnose a subacute exacerbation [104,106]. Nonetheless, telemedicine has been found to be efficient in determining CF clinical status [110]. There is also the concern that telemedicine may lead to less engagement with the CF team [104]. Despite these concerns, telemedicine does offer reduced risk of cross infection by avoiding hospitals, clinics, and close contact with other CF patients [16–17].

Conclusion

The median age of survival for people with CF is well into the 4th and 5th decades of life for developed countries, and the number of adults with CF is expected to increase dramatically over the coming years [121]. New and inventive models of healthcare delivery to accommodate growing numbers of patients with complex medical needs, while maintaining health outcomes will be needed to meet this demand. In the future, more routine care may be delivered remotely, especially as technology for self-monitoring improves in accuracy and availability. Future programs should aim to improve quality of life and reduce burdens of care. Not all patients have access to technology and ways to eliminate this disparity need to be found. The efficacy of telemedicine will need to be more rigorously studied as the effects on health outcomes and healthcare utilization are lacking. As efforts to advance outcomes for people with CF continue and patient-centered care models expand, telemedicine may help to meet the needs of patients and improve engagement and outcomes. While telemedicine holds great promise, the questions of cost, additional patient and provider burdens, and barriers to availability will need to be addressed.

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CRedit authorship contribution statement

Marisa E. Desimone: Conceptualization, Writing – review & editing.
Jordan Sherwood: . **Sarah C. Soltman:** . **Antoinette Moran:** Conceptualization, Writing – review & editing.

Declaration of Competing Interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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