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Original Article

Patient and family perceptions of telehealth as part of the cystic fibrosis care model during COVID-19



J. Davis^{a,1}, A. NeSmith^{b,1}, R. Perkins^a, J. Bailey^b, C. Siracusa^{c,d}, N. Chaudary^e, M. Powers^f, G.S. Sawicki^a, G.M. Solomon^{b,*}

^a Boston Children's Hospital, Boston, MA, USA

^b University of Alabama at Birmingham, Birmingham, AL, USA

^c Cincinnati Children's Hospital, Cincinnati, OH, USA

^d University of Cincinnati College of Medicine, Cincinnati, OH, USA

^e Virginia Commonwealth University, Richmond, VA, USA

^f Oregon Health & Science University Doernbecher Children's Hospital, Portland, OR, USA

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ABSTRACT

Background: Cystic Fibrosis (CF) is a chronic multi-system disease best cared for at Care centers with routine monitoring by interdisciplinary teams. Previously, remote home monitoring technology has been explored to augment in-person care. During the COVID-19 pandemic, traditional in-person care was limited and CF centers rapidly adapted to a telehealth delivery model. The purpose of this study was to understand how people with CF (PwCF) and families of PwCF experienced the shift to telehealthcare delivery.

Methods: This was a cross-sectional survey-based study conducted in 11 CF Centers. Two surveys were designed (one for adult PwCF and one for parents/guardians of PwCF) by participating CF center members with patient and family partner input. Surveys were disseminated electronically via email/text to all patients who completed a telehealth visit, and data were collected on secure Google Forms.

Results: Respondents rated their telehealth experiences as positive. Most were highly satisfied with their telehealth visit (77% adult, 72% pediatric) and found the visits to be highly convenient (85% for all surveyed). A majority of patients reported they had adequate time during the visit and had all questions and concerns addressed. Importantly, we also identified concerns regarding lack of in-person assessments including pulmonary function testing (PFT) and throat/sputum culture.

Conclusion: Telehealth was a feasible and well-accepted mechanism for delivering care in a chronic CF care model during the COVID-19 pandemic and may be useful in the post-pandemic era. Further work is needed to understand the impact of telehealth on patient outcomes, healthcare utilization and associated cost.

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Background

Cystic fibrosis (CF) is a chronic multisystem disease that necessitates frequent clinical care and regular follow up. Survival in CF has improved, in part due to the consolidation of chronic CF care amongst multidisciplinary specialist teams and age-specific care centers [1]. National and international CF care guidelines recommend regular visits and frequent communication with providers at

an accredited CF Care center, and this care model has been shown to improve clinical and respiratory outcomes when compared to those managed regionally [2,3]. During the COVID-19 pandemic, as access to routine in person care was restricted due to the need for social distancing, telehealth – the use of telecommunications technologies to support real-time clinical health care – was rapidly implemented to prevent gaps in care. Understanding patient willingness to participate is a first step to garnering an appreciation for its longer-term role beyond the pandemic and the proper method of delivery of telehealth services.

Before the pandemic, telehealth had been utilized to care for people with CF (PwCF) in limited settings, including to reach those living far from regional CF centers [4–6]. Telehealth has multiple advantages, including minimizing the burden of taking time off

Abbreviations: CF, Cystic Fibrosis; PwCF, People with CF; COVID-19, Novel SARS-CoV-2 coronavirus; PFT, Pulmonary Function Testing.

* Corresponding author.

E-mail address: gsolomon@uabmc.edu (G.M. Solomon).

¹ Indicates Authors Contributed equally.

work, children missing school and costs associated with travel [7], all of which can be especially taxing in larger US states where distance between CF centers can be considerable. Despite interest in telehealth, and small studies supporting the use of telemonitoring as an adjunct to in person care [8–13], regulatory, technological, and other perceived barriers had hampered broad implementation of a virtual care model. In response to the COVID-19 pandemic, unprecedented use of telehealth services was quickly enacted at multiple CF centers to ensure continued access to care [14]. To our knowledge, prior to the current COVID-19 pandemic, there has been no description in published literature of comprehensive clinical telehealth programs widely offered to a CF center's patient panel.

Given the infrequent use of telehealth in the CF chronic care model prior to this time, patient experience in this virtual care paradigm is unknown. In order to better understand patient and family perceptions of telehealth in the CF population during the COVID-19 pandemic, several CF centers across the United States collaborated to design and disseminate an experience of care survey following telehealth visits. In general, we hoped to characterize the viewpoints of adult and pediatric PwCF and families of PwCF regarding integrating telehealth into their usual care. The objectives of the survey were to assess patient and family's attitudes about convenience of visit, level of concern about absence of usual in-person assessments (pulmonary function testing (PFT), weight, sputum culture, vital signs and physical exam), and their desired role of telehealth in future care. We previously assessed the viewpoints of CF providers surrounding telehealth [15].

Methods

Survey development

To rapidly produce a survey focused on patient experience with telehealth, the CF Centers at the University of Alabama and Boston Children's Hospital formed a multi-center working group that met regularly via Zoom and received approval by patient/family partners. Survey development methods included cognitive mapping and brainstorming, pilot testing with patient and family partners and subsequent adjustment. Two surveys were ultimately developed in parallel, one for adults with CF receiving care at an adult CF program and a second for parents of children and young adults with CF receiving care at a pediatric CF program. The adult and pediatric surveys each consisted of 21 questions (5 multiple choice, 7 Likert scale, 5 yes/no, 3 short answer, and 1 checkbox question). Demographic data captured general age range and primary CF center. We asked participants to indicate which device (smartphone, computer or tablet) they used to access their telehealth visit and whether they used audio only. The survey focused on 3 themes to assess telehealth experience: understanding 1) the structure of and satisfaction with the visit, 2) the level of concern with absence of usual in-person assessments (PFT, weight, sputum culture, vital signs and physical exam), and 3) the desired role of telehealth in their future care.

Five-point Likert scales were used to assess overall satisfaction and convenience of visits: 5 was defined as "very satisfied" or "very convenient," 1 was defined as "not satisfied" and "not convenient," and 3 was "neutral." In addition, a 5-point Likert scale was used to assess concern with lack of usual in-person assessments, where "1" was defined as "low concern" and 5 as "very concerned." We defined a score of "2" or less as a lower level of concern and "3" as moderate concern. Full surveys questionnaires are available in the online supplement.

Data acquisition

We administered the final survey to PwCF who engaged in a telehealth visit at their CF center between April 2020 and June 2020. A telehealth visit was defined as a distinct healthcare encounter with at least one medical provider, conducted at a distance by telecommunications. The survey was deployed across 11 CF programs (4 adult and 7 pediatric): The University of Alabama at Birmingham Adult Program, Boston Children's Hospital/Brigham and Women's Hospital Adult Program, Virginia Commonwealth University Adult Program, University of West Virginia Adult Program, Boston Children's Hospital Pediatric Program, Cincinnati Children's Hospital Pediatric Program, OHSU Doernbecher Children's Hospital Pediatric Program, UVA Children's Hospital Pediatric Program, WVU Children's Hospital Pediatric Program, Primary Children's Cystic Fibrosis Center Pediatric Program, CHOR at Virginia Commonwealth University Pediatric Program, and Children's Medical Center of Dallas Pediatric Program. Adults with CF or parents/guardians of those with CF seen in a pediatric center were invited to participate in the online survey via a link sent by email, text or electronic medical record within one week of the patient's first telehealth visit. Parents or guardians of patients 18 years and older who were seen in a pediatric center were sent the pediatric survey. There were no repeated measures done during this study's timeframe. Survey data were collected in an anonymized fashion from April to July 2020 and analyzed using Google Forms in weekly intervals. This study was conducted under IRB approval of the parent institutions of the participating centers. GraphPad Prism 8.4 was used to create the figures and produce descriptive statistics of the survey data.

Results

Surveys were completed by 120 adult patients across 4 adult CF centers and 141 parents/guardians of patients seen in 7 pediatric centers during the study period. Total response rate was 25%. Respondents had an even distribution across the pediatric and adult age spans (Supplementary table). Patients accessed their telehealth visits via a variety of devices, most commonly by cell-phone/smartphone, and computer/laptop and less commonly by tablet/iPad (Fig. 1).

Most patients surveyed (72% adult, 77% pediatric) had no prior experience with telehealth and despite this, most (87% of all surveyed) denied any difficulty accessing their telehealth visit (Fig. 2). Many visits were multidisciplinary, with physicians seen most frequently. Other providers most commonly seen by pediatric patients included a dietitian, social worker and registered nurse. Adults had lower rates of seeing their non-physician care team members including an advanced practice practitioner, a dietitian, and a registered nurse (Fig. 3). Most patients rated their telehealth visits the highest level of convenience (85% for all surveyed) and satisfaction (77% adult, 72% pediatric) (Fig. 2). Most indicated they saw all desired disciplines, had adequate time with their team and all concerns were addressed (Fig. 2).

In terms of missing in-person assessments, significant proportions of patients reported at least moderate concern regarding lack of usual measures and testing offered in an ambulatory clinic (Fig. 4). The highest level of concern expressed was regarding lack of throat/sputum cultures in both the adult population (54% expressing at least moderate concern) and pediatric population (64% expressing at least moderate concern). Fifty-two percent of adult respondents and respondents on behalf of the pediatric population expressed concern regarding lack of pulmonary function testing. A majority of surveyed patients and parents/guardians reported low concern regarding the lack of other measures: lack of weight and lack of vital signs with physical exam (Fig. 4).

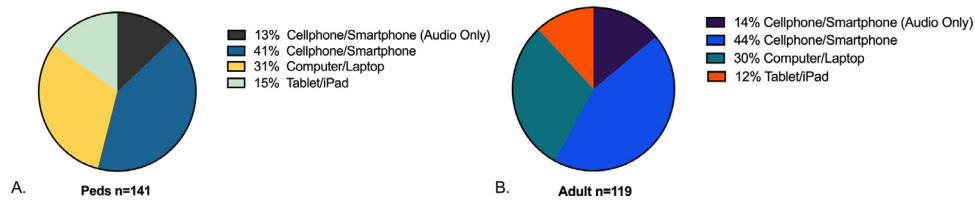


Fig. 1. Method used to access telehealth visit for (A) Adult (n=119) and (B) Pediatric respondents (n=141).

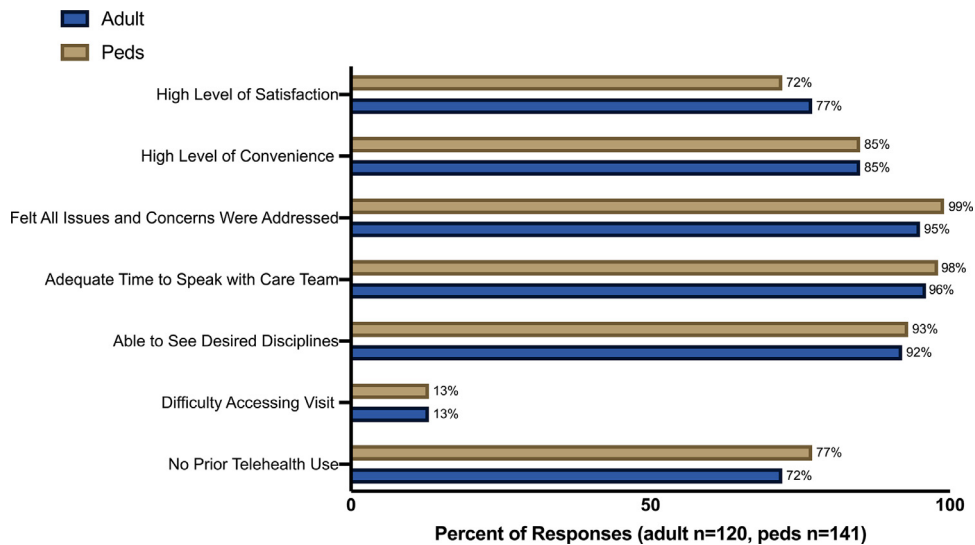


Fig. 2. Summary of survey results from pediatric and adult cohorts capturing prior experience with telehealth and perceptions of their recent CF care center visit. Total respondents (n=261).

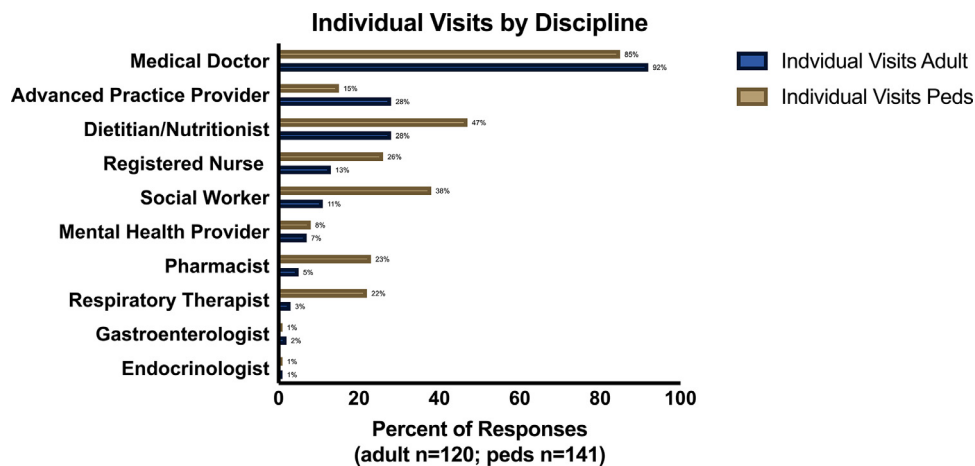


Fig. 3. Visits by discipline. Patients were asked to indicate which providers they met with as part of their telehealth visit. Total respondents (n=261).

Despite some concern with lack of in-person assessments, there was striking interest in using telehealth as part of future care. Most patients (53% adult, 69% pediatric) expressed an interest in having “some” visits be conducted virtually, with an additional smaller group (34% adult, 21% pediatric) preferring for “most” future visits to take place virtually (Fig. 5). Both groups of respondents (94% adult, 97% pediatric) were interested in technology that would allow for at-home assessment of pulmonary function (Fig. 5) to allay concerns discussed above.

Discussion

This study describes the experiences of over 250 PwCF across the pediatric and adult age-span who participated in telehealth

services at their CF centers across the United States during the COVID-19 pandemic. Our findings highlight the overwhelmingly positive experience reported by participants and suggest a more permanent role for telehealth in the chronic care delivery model for CF may exist. Prior studies have determined that telemonitoring as an adjunct to routine in-person care is feasible with at-home technologies, though variable effects on clinical outcomes were seen. In the present study, we had the opportunity to evaluate the experience of a near-complete transformation to virtual healthcare delivery in the setting of the COVID-19 pandemic.

Our group also recently published similar positive findings of the experience of providing telehealth by providers in the same multi-center group. We found similar concerns about access to remote monitoring by multi-disciplinary providers but noted agree-

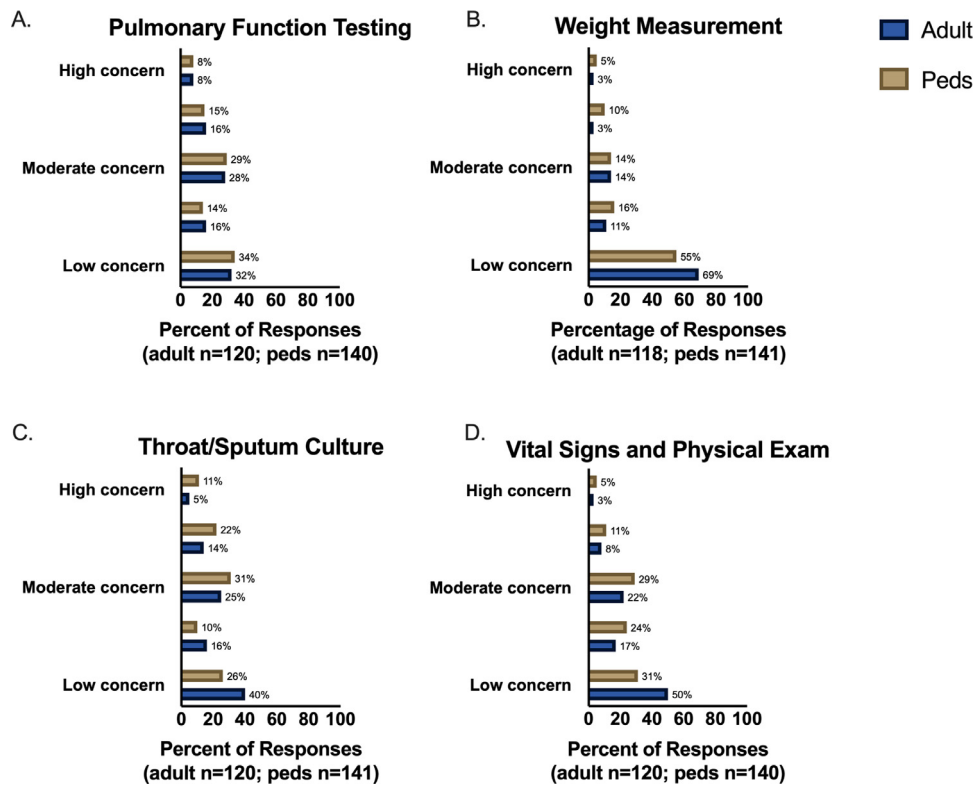


Fig. 4. Degree of concern regarding absence of (A) Pulmonary Function Testing (Adult n=120, Peds n=140); (B) Weight Measurement (Adult n=118, Peds n=141); (C) Throat/Sputum Culture (Adult n=120, Peds n=141); (D) Vital Signs and Physical Exam for Adult (Adult n=120, Peds n=140).

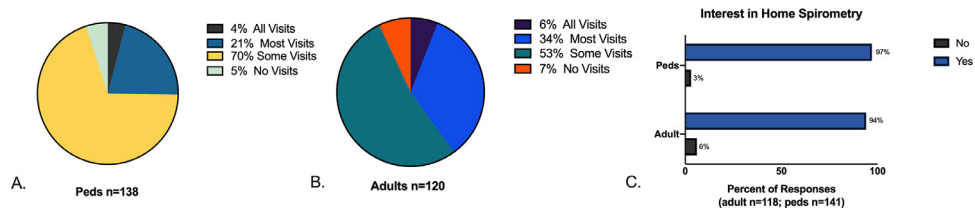


Fig. 5. Desired frequency of future virtual visits for (A) Adult (n=120) and (C) Pediatric respondents (n=138) and (C) Interest in technology allowing for home pulmonary function assessment.

ment around access and convenience for patients resulting in a desire to integrate telehealth services into the CF chronic care model of C after the pandemic [15].

We recognize the uniqueness of the context allowing for this experiment, and even while interpreting our findings through this lens, we argue there are lessons to be learned about how to integrate tele-healthcare delivery into our existing care paradigms. First, this study demonstrates PwCF are willing and able to engage with care virtually. Though roughly three quarters of all respondents surveyed had never used telehealth before, almost 90% denied any technological difficulty. This finding supports the idea that existing technology allows for seamless adoption of telehealth in the CF chronic care model. Second, PwCF were satisfied with their experiences, many reported all their concerns were addressed, and could meet with all the members of their healthcare team they wished to. These findings are important because they show not only that telehealth was well-received, but respondents felt they were given adequate time to discuss their concerns and meet with members of the multidisciplinary care team, as they do during a typical in-person visit. Third, there are limitations, specifically usual in-person assessments, to what can be accomplished by telehealth and lack of sputum cultures and pulmonary function testing were causes of concern for those in this study. Despite

these limitations, a majority of patients still expressed interest in continued use of telehealth in the future.

Home spirometers and other remote monitoring technologies may alleviate concerns of PwCF regarding lack of in-person testing. In response to this concern, the Cystic Fibrosis Foundation has begun an initiative to provide PwCF at-home spirometry technology, which has the potential to improve the quality of remote assessment and accuracy of clinical telehealth. In the present study, an overwhelming number of both adult and pediatric respondents expressed interest in technology that would allow for pulmonary function assessment at home. Other at home assessment tools, such as scales and culture-kits, will need to be developed to allow for effective care in the telehealth arena.

Going forward, it is reasonable to plan for telehealth to play some role in the care model for PwCF. There are a number of different ways to incorporate telehealth into the current care paradigm, including replacing one or more routine in-person visit with a telehealth appointment in clinically stable PwCF, conducting some part of the care team’s assessment virtually (i.e. nutrition assessment, nursing coordination or social work check-in), providing hospital or home IV follow-up, or possibly triaging sick calls from the nursing line. A reduction in the number of in-person visits could decrease the travel burden experienced by some PwCF,

possibly reduce missed clinic appointments and minimize infection risks. Future care plans integrating telehealth will need to remain flexible and considered individually for each patient, and their results warrant continued study.

Our study has noted strengths and some limitations. Strengths to this study include its novel and timely description of perceptions of telehealth in the CF population. We surveyed an array of PwCF and their family members, across both the pediatric and adult age span, and across multiple CF centers in the United States, including the opinions of over 250 respondents. As with any survey design study, our data depends upon those who choose to respond, rather than a random sample of participants. We minimized recall bias by sending the surveys shortly after the visit was completed. Given the perceptions elicited in this study are colored by the specific situation of using telehealth during the early months of the COVID-19 pandemic, these results may not necessarily be generalizable to a time when social distancing is no longer mandated. Further, our results capture one point in time per respondent and recognize that as the programs developed during these early months, experiences could also have changed. We do not have demographic data beyond age, limiting our ability to study factors related to social determinants of health, including the potential digital divide based on socioeconomic status that limits internet availability. Telehealth visits with interpreter services were offered to those who do not use English as a primary language; as our survey was offered only in English, we were unable to capture responses from the small proportion of CF patients who are non-English speakers. Future studies should be designed to understand and optimize the experience for this subpopulation. Respondents on behalf of patients at pediatric centers aged 18 years and greater were caregivers, whose contact information is primary; this focuses on the patient family experience, rather than that of these adult-aged patients. Finally, as the survey period spanned just several months, our study cannot answer whether telehealth meets the standards of CFF recommended care. While at this time, PwCF show interest in continuing telehealth visits in the future, it will be important to reassess attitudes and study outcomes in a longitudinal fashion and in the post-pandemic era.

Telehealth is a powerful tool that has enabled continued access to care for chronically ill patients during an unprecedented time for our healthcare system. It will remain an important aspect of care for PwCF as long as social distancing recommendations remain in place and we should consider how to integrate this promising technology into standards of care going forward. By capitalizing on its merits of convenience and ability to connect entire care teams to patients remotely, we can more easily reach those geographically or logistically distanced from major care centers. By reducing the burden on clinical space, we may reduce health resource utilization and its associated costs. When implementing more permanent programs, we must ensure all have access to telehealth in the CF community and should seek solutions for issues of limited internet access, digital literacy, or language barriers. Future studies should aim to both optimize effectiveness of telehealth programs and evaluate the effects on CF outcomes including rates of pulmonary exacerbations, need for inpatient admissions and other patient-reported outcomes such as quality of life. Continued collaboration across centers will be essential to achieve these goals. Telehealth, in combination with remote monitoring devices, has the promise to drastically impact our current approach to healthcare delivery for those with CF, bringing convenience, patient satisfaction and possibly even improved value.

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CRediT authorship contribution statement

J. Davis: Conceptualization, Methodology, Investigation, Data curation, Visualization, Writing - original draft, Writing - review & editing. **A. NeSmith:** Conceptualization, Methodology, Investigation, Data curation, Visualization, Writing - original draft, Writing - review & editing. **R. Perkins:** Conceptualization, Methodology, Investigation, Data curation, Visualization. **J. Bailey:** Conceptualization, Methodology, Investigation. **M. Powers:** Conceptualization, Methodology, Investigation. **G.S. Sawicki:** Conceptualization, Methodology, Investigation, Supervision, Writing - review & editing. **G.M. Solomon:** Conceptualization, Methodology, Investigation, Data curation, Visualization, Supervision, Writing - review & editing.

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OHSU: Ben McCullar, RN

WVU: Ryan Juel, MD

Supplementary materials

Supplementary material associated with this article can be found, in the online version, at doi:[10.1016/j.jcf.2021.03.009](https://doi.org/10.1016/j.jcf.2021.03.009).

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