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Short Communication

The impact of telehealth based care on paediatric cystic fibrosis outcomes

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ABSTRACT

In response to the COVID-19 pandemic telehealth utilisation amongst the Cystic Fibrosis (CF) population increased. Our aim was to assess the impact of CF telehealth clinics on CF outcomes. We conducted a retrospective chart review of patients seen in the CF clinic at the Royal Children's Hospital (Victoria, Australia). In this review we compared spirometry, microbiology and anthropometry in the year preceding the pandemic to during the pandemic, and to the first in-person appointment in 2021. 214 patients were included. First in-person FEV_1 was median 5.4% below individuals' best FEV_1 in 12 months prior to lockdown and decreased by >10% in 46 (31.9%) patients. There were no significant findings with regards to microbiology or anthropometry. The reduction in FEV_1 observed on return to in-person appointments highlights the importance of ongoing improvement of telehealth-based care along with continued face-to-face review for the paediatric CF population.

1. Introduction

The care and outcomes of patients with cystic fibrosis (CF) has benefitted from multidisciplinary CF clinics [1], and the COVID-19 pandemic accelerated the adoption of telehealth to deliver these clinics. Like many institutes around the world, the CF clinic at the Royal Children's Hospital (RCH), Melbourne, Australia moved to an exclusive telehealth model for ambulatory care during the COVID-19 pandemic. Previous research has shown feasibility [2,3] and patient/parental preference [4,5] for telehealth, however data regarding the impact on health outcomes are limited. This study aimed to assess the impact of CF telehealth clinics on clinical outcomes, specifically lung function, microbiology, nutritional status and admissions.

2. Methods

2.1. Study design, setting, outcome

We conducted a retrospective chart review of patients attending the CF clinic at RCH, a large tertiary care hospital and one of two specialist paediatric CF centres in Victoria, caring for approximately 230 patients aged 0–19 years. As per standard guidelines [6], patients are reviewed at minimum four times per year in the CF multidisciplinary clinic. In response to the COVID-19 pandemic, on the 19/3/20 the CF clinic transitioned to a telehealth model. During this time all clinics were performed over telehealth unless there was an urgent clinical indication for in-person review. During telehealth visits the patient and family were able to see the entire CF multidisciplinary team. As per usual care during the visit, the team would address all relevant areas of CF care including pulmonary, nutritional, and mental health as well as treatment adherence. Portable spirometers were distributed to 90 patients deemed highest risk of deterioration based on FEV_1 percent predicted (FEV_1) results or clinician discretion. All home spirometry assessments were completed via telehealth using the NDD Easy-On PC Spirometer. Each testing session was performed under the supervision of a qualified respiratory scientist and conducted in accordance with the 2019 ATS/ERS Spirometry guidelines [7]. Data was considered acceptable if the test session achieved a quality of A or B grade. A minimum of three attempts and a maximum of eight were performed. Families were provided with equipment, written instructions and instructional videos for microbiological sampling (cough swabs and sputum samples) and asked to collect the sample in advance of the clinic and return to hospital or local pathology collection centre. During the pandemic period, if an exacerbation was detected (either using symptoms or change in FEV_1 for those with spirometers) treatment was initiated in the home with oral antibiotics, with admission to hospital remaining

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Table 1
Patient demographics, baseline lung function and admission number in all patients ($N = 144$), patients with home spirometer ($N = 80$) and patient with no home spirometer ($N = 64$).

	All patients ($n = 144$)	Home spirometer ($n = 80$)	No home spirometer ($n = 64$)
Age (mean, SD)	12.3 ± 3.6	13.2 ± 2.94	11.1 ± 4.05
Sex, Male (n,%)	66, 45.8%	41, 51.3%	37, 57.8%
Baseline BMI z score (median)	0.11 (−0.50–0.66)	0.06 (−0.55–0.56)	0.21 (−0.4–0.87)
Baseline%FEV1 (median)			
Pre lockdown	95 (84–105)	92 (84–103)	98 (85–110)
During lockdown	92 (82–103)	93 (82–103)	92 (82–99)
Post lockdown	89 (77–100)	86 (74–96)	94 (83–106)
Number of spirometry during pandemic (median)	4 (3–6.0)	5 (4–7)	2 (10–3)
1 or more admission during lockdown n (%)	50, 34.7%	37, 46.3%	13, 20.3%
Admissions due to reduction in FEV1 n (%)	8, 5.6%	7, 8.7%%	1, 1.6%

Table 2
Percentage change in FEV1 from pre to post lockdown.

%Change in FEV1	≥ −10%	−10% - −5%	−5% - 0%	0% - 5%	5% - 10%	≥10%
All patients ($n = 144$)	46 (31.9%)	28 (19.4%)	37 (25.7%)	19 (13.2%)	6 (4.2%)	8 (5.6%)
Home spirometer ($n = 80$)	33 (41.3%)	14 (17.5%)	19 (23.8%)	8 (10.0%)	3 (3.8%)	3 (3.8%)
No Home spirometer ($n = 64$)	13 (20.3%)	14 (21.9%)	20 (31.2%)	11 (17.2%)	3 (4.7%)	5 (7.8%)

an option for those who did not improve. From 7/1/2021, there was a gradual return to face-to-face clinics.

All patients aged >12-months attending the RCH CF Clinic in January 2021 were included in the analysis. We compared spirometry (if age ≥ 6-years), microbiology, anthropometry and admissions in the year preceding the pandemic (19/3/2019–18/3/2020), to the 9-month period of the state's major lockdown (19/3/2020–31/12/2020) and to the first in-person appointment in 2021. Pre lockdown FEV_1 was based on the best FEV_1 in the preceding 12 months. Patients who did not perform an in-person spirometry by 30/06/2021 were excluded from the spirometry analysis,

2.2. Data collection, analysis and ethics

Data was collected from the institution's electronic medical records by a single investigator (KR) and managed using REDCap electronic data capture tool [8]. Simple descriptive analysis was performed on multiple variables. This study approved by the Royal Children's Hospital Melbourne Human Research Ethics Committee, reference number 65,651.

3. Results

3.1. Participants

A total of 214 patients were included in the study. The median age was 11 (range 1–19) years, and 58% ($N = 124$) were male.

3.2. Spirometry

Out of the 164 patients with spirometry data available pre-pandemic, 144 patients were included in the analysis, with $n = 20$ patients excluded from the analysis as post-lockdown spirometry had not been performed. Baseline demographics and baseline FEV_1 results are outlined in table 1. All patients with a home spirometer completed at least one lung function test during the 9-month lockdown compared to 50% ($n = 32$) of those without. Overall, the median change in FEV_1 from pre to post-lockdown was a decline of 5.4% (Fig. 1) with 31.9% of patients declining by >10% (table 2). The same trend was observed in those with and without a home spirometer however, the median change was greater in those with a home spirometer (6.5% vs 3.5%). Out of the 50 patients with at least one admission during the lockdown period, the majority

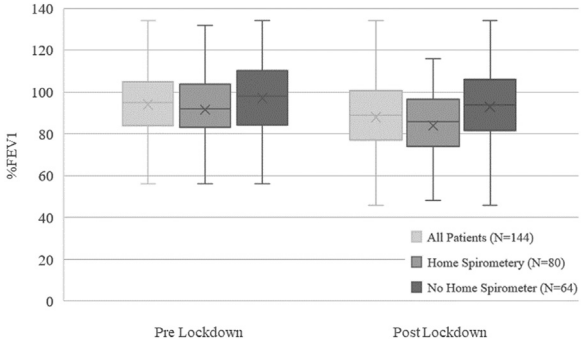

Fig. 1. FEV1 pre and post lockdown - All patients ($N = 144$) vs patients with home spirometer ($N = 80$) vs patients without home spirometer ($N = 64$).

(74%) had a home spirometer, with 19% of admissions due to a reduction in FEV_1. The number of patients requiring admission was less compared to the year preceding the lockdown ($n = 64$, 44.4%).

3.3. Microbiology

There was a reduction in the number of microbiology samples, from a median of 5 per patient per year pre-pandemic to a median of 3 during the lockdown period. Seven percent of patients did not obtain a microbiology sample during lockdown. Forty-seven (22%) patients grew *P. aeruginosa* in the year pre-pandemic compared to 36 (16.8%) during the lockdown. Only 1 of the 16 patients positive for *P.aeruginosa* in the first in-person airway sample post lockdown were positive for the first time during the study.

3.4. Anthropometry & admissions

Weight and BMI remained stable during lockdown, with a median change in z-score of 0.1 (IQR −0.2 - 0.32, $p = 0.4$) and 0.01 (IQR −0.29 - 0.39, $p = 0.7$) respectively. There was a reduction in the number of patients who had admissions during lockdown (pre $n = 82$, 38% vs during $n = 63$, 30%).

4. Discussion

Melbourne endured some of the toughest restrictions in the world during the COVID-19 pandemic with 173 days of hard lockdown in 2020 [9]. The ongoing care to our CF cohort was en-

abled with the implementation of telehealth clinics. Previous research has reported patient and clinician preference for telehealth for reasons including convenience, accessibility and limiting absence from school/work [4,5,10]. In our study a change to an almost exclusive telehealth service, in the midst of a pandemic, was associated with a decline in FEV_1 on first in person post-lockdown spirometry compared to pre-pandemic results. While this does not demonstrate a causal relationship between telehealth-based care, and poorer CF outcomes, it does highlight the need for further evaluation of monitoring of telehealth especially given patient and clinician preference for its ongoing use.

A few European studies have investigated the impact of lockdown on FEV_1 in paediatric patients. Thee et al. [11] reported a decline in FEV_1 in a cohort of 42 paediatric patients over a 2-month lockdown period, whereas three studies demonstrated an improvement in FEV_1 [12–14]. These studies were not exclusive to a paediatric population, were of shorter duration (2–6 months), and in one, patients had access to elexacaftor/tezecaftor/ivacaftor therapy. These studies did not utilise telehealth clinics or home spirometry.

In our cohort, home and in person spirometry was performed during the lockdown period. Pre-pandemic we had an established telehealth service for patients living in rural and remote areas [3], thus widespread implementation of an entirely telehealth service involving all members of the multi-disciplinary team was simplified. To limit inaccuracy [15] and overestimation of FEV_1 [16]; all home spirometry was supervised by respiratory technicians with anthropometric parameters performed each clinic by a family member educated on performing this correctly. Despite this, FEV_1 decreased similarly in those with and without home spirometers, raising questions about the reliability of home spirometry over prolonged periods in the paediatric population. Even with supervised testing, it relies on accurate home measurements of height. Under-estimation of linear height (or reliance on an earlier measurements) in a growing child/adolescent will result in overestimation of FEV_1. As such, inaccurate height measurement may have been a significant barrier to home spirometry detecting deterioration in FEV_1. Despite the limitations of home spirometry, all patients with a spirometer underwent lung function surveillance during the lockdown period compared to only half of those without. Therefore, early detection of declining lung function was potentially missed in the group without home spirometers which may contribute to the reduction in FEV_1 in post lockdown testing.

The decline in FEV_1 may also reflect the impacts of the 9-month lockdown, including reduced physical activity, poor mental health and non-adherence to airway clearance, rather than a telehealth related phenomenon. Whilst psychological and adherence support was available to all patients in our service, access to care does not necessarily ameliorate the potential impact of poor mental health on adherence and CF outcomes.

While hospitalisation rates decreased during the lockdown, likely, at least in part, due to a reduction in viral infections, spirometry results suggest there were additional patients who may have benefited from admissions, and that lung function decline may have been recognised earlier had routine in-person reviews continued.

Despite an overall reduction in microbiology samples, our results suggest home samples were sufficient in detecting *P. aeruginosa*. This is contrary to a recent study, which reported that home samples during the COVID-19 pandemic was associated with missed or delayed opportunities to detect new pathogens, including *P. aeruginosa* [17].

There are some limitations that must be acknowledged. This study was necessarily retrospective. Data from first in-person clinic visit was used to represent post-lockdown health, which does not account for variation in FEV_1 across a 12 month period. The decision was made to use the first in-person clinic visit, rather than best value in the subsequent 12 months, as it was likely that with further pandemic waves CF clinic would return to a telehealth model, which was indeed what occurred. Further, pre-telehealth rate of decline in FEV_1 was not calculated, which may have added further data regarding lung function trends. Data was not adjusted for treatment with highly effective modulator therapy, as ivacaftor was the only medication in this class available in Australia during the study period and only 8.7% of patients in our clinic received this medication. Further, we did not adjust or account for other CF related comorbidities, which is a limitation given the influence of comorbidities on lung function.

Despite these limitations our single-centre study identified an association between an exclusive telehealth-based model of care during a pandemic and reduced FEV_1. Given patient and clinician preference for telehealth, future work should evaluate and identify methods for safely incorporating telehealth into routine CF care. Methods for home and/or in-person spirometry, anthropometric measurements, and microbiological sample collection need to be easily accessible, with correct training and supervision. It is likely that telehealth based care should complement, rather than replace in person assessment, and much further work is needed to define the optimal use of this technology.

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CRediT authorship contribution statement

Kristene Rimbaldo: Conceptualization, Data curation, Methodology, Investigation, Supervision, Writing – original draft. **Katherine B Frayman:** Methodology, Investigation, Writing – review & editing. **Shivanthan Shanthikumar:** Conceptualization, Methodology, Investigation, Supervision, Writing – review & editing.

References

[1] Dodge JA. Management in paediatric and adult cystic fibrosis centres improves clinical outcome. Br Med J 1998;316(7147):1775.
[2] Vagg T, Shanthikumar S, Morrissy D, Chapman WW, Plant BJ, Ranganathan S. Telehealth and virtual health monitoring in cystic fibrosis. Curr Opin Pulm Med 2021;27(6):544–53. doi:10.1097/MCP.0000000000000821.
[3] Logie K, Welsh L, Ranganathan SC. Telehealth spirometry for children with cystic fibrosis. Arch Dis Child 2020;105(12):1203–5. doi:10.1136/archdischild-2019-317147.
[4] Shanthikumar S, Moore E, Corda J, Reardon N, Louey S, Frayman K, et al. Patient and family perspectives regarding the use of telehealth for cystic fibrosis care. Pediatr Pulmonol 2021;56(5):811–13. doi:10.1002/ppul.25262.
[5] Hendra K, Neemuchwala F, Chan M, Ly NP, Gibb ER. Patient and provider experience with cystic fibrosis telemedicine clinic. Front Pediatr 2021;9(1376). doi:10.3389/fped.2021.784692.
[6] Bell S., Robinson P., Fitzgerald D. Cystic fibrosis standards of care, 2008 Australia2008 01/01. Available from: https://shop.cysticfibrosis.org.au/standards-of-care.
[7] Graham BL, Steenbruggen I, Miller MR, Barjaktarevic IZ, Cooper BG, Hall GL, et al. Standardization of spirometry 2019 update. An official American thoracic society and european respiratory society technical statement. Am J Respir Crit Care Med 2019;200(8):e70–88. doi:10.1164/rccm.201908-1590ST.
[8] Harris PA, Taylor R, Thielke R, Payne J, Gonzalez N, Conde JG. Research electronic data capture (REDCap)–a metadata-driven methodology and workflow process for providing translational research informatics support. J Biomed Inform 2009;42(2):377–81. doi:10.1016/j.jbi.2008.08.010.
[9] Stobart A, Duckett S. Australia's response to COVID-19. Health Econ Pol Law 2022;17(1):95–106. doi:10.1017/S1744133121000244.
[10] Perkins RC, Davis J, NeSmith A, Bailey J, Powers MR, Chaudary N, et al. Favorable clinician acceptability of telehealth as part of the cystic fibrosis care model during the COVID-19 pandemic. Ann Am Thorac Soc 2021;18(9):1588–92. doi:10.1513/AnnalsATS.202012-1484RL.
[11] Thee S, Busack LM, Mall MA, Stahl M. Impact of lockdown during the COVID-19 pandemic on health status in patients with cystic fibrosis: a mono-centre observational study. ERJ Open Res 2022;8(1). doi:10.1183/23120541.00588-2021.

[12] Servidio AG, Capata G, Levantino L, Riccio G, Contorno S, Barbi E, et al. COVID-19 lockdown beneficial effects on lung function in a cohort of cystic fibrosis patients. Ital J Pediatr 2021;47(1):12. doi:10.1186/s13052-021-00970-4.
[13] Loukou I, Moustaki M, Petrocheilou A, Zarkada I, Douros K. Impact of COVID-19 lockdown on pulmonary and nutritional status in children and young adults with cystic fibrosis, in Greece. J Patient Exp 2021;8:23743735211008295. doi:10.1177/23743735211008295.
[14] Hatziagorou E, Toulia I, Avramidou V, Kampouras A, Tsara V, Tsanakas J. Change in CF care during COVID-19 pandemic: single-center experience in a middle-income setting. Pediatr Pulmonol 2021;56(9):3065–7. doi:10.1002/ppul.25560.
[15] Muhlebach MS, Cunningham F, LiPuma JJ, Zhou JJ, Saiman L. Impact of the SARS-CoV-2 pandemic on cystic fibrosis centres and care: survey results from US centres. ERJ Open Res 2021;7(4). doi:10.1183/23120541.00249-2021.
[16] Quanjer PH, Stanojevic S, Cole TJ, Baur X, Hall GL, Culver BH, et al. Multi-ethnic reference values for spirometry for the 3-95-yr age range: the global lung function 2012 equations. Eur Respir J 2012;40(6):1324–43. doi:10.1183/09031936.00080312.
[17] Franciosi AN, Wilcox PG, Quon BS. Cystic fibrosis respiratory microbiology monitoring during a global pandemic: lessons learned from a shift to telehealth. Ann Am Thorac Soc 2022;19(3):498–500. doi:10.1513/AnnalsATS.202101-087RL.