



STUDY PROTOCOL

REVISED

Steps Ahead: optimising physical activity in adults with cystic fibrosis: Study Protocol for a pilot randomised trial using wearable technology, goal setting and text message feedback. [version 3; peer review: 2 approved]

Previously titled: Steps Ahead: optimising physical activity in adults with cystic fibrosis: Study Protocol for a pilot randomised trial using wearable technology

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Abstract

Background: Physical activity (PA) and exercise are widely documented as key components in the management of cystic fibrosis (CF). In recent years there have been significant improvements in telehealth, in particular; wearable technology, smartphone use and remote monitoring, all of which may have potential to impact on PA in adults with CF. The objective of this pilot randomised trial is to explore the effect of wearable technology, which is remotely monitored, combined with personalised text message feedback and goal setting, on PA in adults with CF. Secondary endpoints include lung function, aerobic capacity, quality of life, body composition, wellbeing and sleep.

Methods: This is a pilot randomised trial which will be conducted at the University Hospital Limerick, Ireland. Participants will be randomised to the intervention or active comparator after their

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baseline assessment. The 12-week intervention will consist of wearable technology (Fitbit Charge 2) which is linked to an online monitoring system (Fitabase) that enables the physiotherapist to remotely monitor participant data. The CF physiotherapist will set individualised PA goals with each participant at baseline and will send text message feedback each week. The text messages will be personalised, one-way texts with positive reinforcement on step count attained by the participant. The active comparator group will receive this wearable technology which is also linked to Fitabase; however, no feedback will be provided to participants in this group. Both groups will be re-assessed at 12 weeks. After this point, both groups will continue with the Fitbit alone for a further 12 weeks. Both groups will be re-assessed at 24 weeks. A semi structured interview will assess satisfaction and acceptability of the intervention.

Discussion: This is a novel concept which utilises modern technology, remote monitoring and personalised feedback to investigate the effect on PA in adults with CF.

Trial registration: ClinicalTrials.gov [NCT03672058](https://clinicaltrials.gov/ct2/show/study/NCT03672058) (14/09/2018)

Keywords

Physical activity, Randomised trial, Cystic fibrosis, fitness tracker, telehealth

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REVISED Amendments from Version 1

The title of this article has been amended.

Any further responses from the reviewers can be found at the end of the article

Background

Cystic fibrosis (CF) is a life limiting, progressive disease which requires lifelong management. The prevalence of CF is 7 per 100,000 in the European Union, with Ireland reporting the highest incidence of CF in the world (Farrell, 2008). CF is a multisystem disease, primarily affecting the respiratory system which leads to recurrent pulmonary infections with retained secretions, airway obstruction and hyperinflation (Chmiel & Davis, 2003). There is significant burden associated with CF for the individual with CF, their family and wider society. Issues related to treatment adherence (Sawicki *et al.*, 2009) and psychological wellbeing (Quittner *et al.*, 2016), are frequently reported in the literature, especially in adults that are balancing family, work and education, as well as managing their chronic disease (Boyle, 2003). Despite the complexity in the management of CF, much of the regime can be completed from home, enabling the individual with CF to integrate treatment into everyday routines with monitoring from the CF team (Calthorpe *et al.*, 2020).

Physical activity (PA) can be described as any bodily movement that causes an increase in energy expenditure above that of resting energy expenditure including leisure-time PA, occupational PA and exercise (Caspersen *et al.*, 1985). Exercise and PA are widely documented in consensus statements as key components in the management of CF (Castellani *et al.*, 2018). PA has several positive benefits in this population as it has been shown to improve sputum clearance (Dwyer *et al.*, 2009), bone mineral density (Gupta *et al.*, 2019) and muscle strength (Burtin *et al.*, 2013). More importantly, PA can improve aerobic capacity (Hebestreit *et al.*, 2010) and it can slow the rate of decline in lung function (Schneiderman *et al.*, 2014), both of which are linked to increased survival in CF (Nixon *et al.*, 1992). As a result, the optimisation of PA among adults with CF is important, however, a Cochrane review found that there was a lack of evidence regarding strategies to promote PA in this population and consideration should be given towards telemedicine applications and health coaching (Cox *et al.*, 2013b).

Numerous subjective and objective methods are reported in the literature for the assessment of PA in adults with CF. Step count measurement can be used to quantify and monitor PA behaviours (Tudor-Locke & Bassett, 2004). Despite the clear benefits of PA in CF, a recent systematic review found that adults with CF fail to meet recommended PA and step count guidelines (Shelley *et al.*, 2019). Pedometers can increase PA in other populations (Bravata *et al.*, 2007; Hospes *et al.*, 2009; Lubans *et al.*, 2009). Therefore, interventions which aim to increase PA in adults with CF are of considerable interest.

The evolution of telehealth in CF management is significant in recent years. Previous studies have investigated the effect of telehealth on monitoring health status (Grzincich *et al.*, 2010), detecting exacerbations (Lechtzin *et al.*, 2017; Wood *et al.*, 2017), assessing exercise capacity (Cox *et al.*, 2013a) and providing outpatient appointments (Wood *et al.*, 2017). Telehealth is well accepted by adults with CF (Cox *et al.*, 2012). While home monitoring via telehealth seems to be a progressing area of CF management, to date, no studies have evaluated the effect of telehealth on PA and health outcomes in adults with CF. Smartphones and wearable technology may assist CF physiotherapists as they can access PA data remotely (Tagliente *et al.*, 2016). Remote monitoring and home-based PA interventions in adults with CF are advantageous for several reasons. It enables the intervention to be easily implemented, participants' personal preferences can be considered, it is more accessible to all participants and it can involve family/friends (Hebestreit *et al.*, 2010). As a result, these benefits may increase adherence to the intervention. Wearable technology and text message feedback has had positive health outcomes amongst other study populations (Cadmus-Bertram *et al.*, 2015; Cook *et al.*, 2013; Moy *et al.*, 2012; Nguyen *et al.*, 2009; Vaes *et al.*, 2013), however limited research has been conducted among adults with CF to date.

In addition to telehealth, goal setting and reviewing goals regularly should be considered to support patients with chronic illness (Coleman & Newton, 2005). In order for goals to be achieved, feedback should be provided that reveals progress in relation to the goal (Locke & Latham, 2002). Furthermore, setting specific goals should offer a plan to break PA goals into more practical, manageable steps (Shilts *et al.*, 2004) which should increase self-efficacy and hence promote continued regular PA levels (Bandura, 2004). Previous literature from patient preference research and effective PA promotion approaches reports that successful strategies include specifically targeting PA (Conn *et al.*, 2008), the use of behavioural strategies such as feedback and goal setting (Conn *et al.*, 2008; Kosma *et al.*, 2005) (George *et al.*, 2012) and the ability to self-monitor (Conn *et al.*, 2008). The Irish national framework for self-management includes goal setting and action planning in chronic disease management (Chronic Conditions Working Group, 2017). PA interventions using pedometers have been shown to be more effective at increasing PA if they include step goals (Bravata *et al.*, 2007). However, goal setting to increase PA in adults with CF is poorly investigated.

This pilot randomised trial aims to explore the impact of wearable technology, text message personalised feedback and goal setting on PA and health outcomes in adults with CF. For the purpose of this study these specific health outcomes include lung function, aerobic capacity, body composition, quality of life, well-being and sleep. Poor sleep quality has been previously highlighted as an issue in adults with CF (Milross *et al.*, 2002) and PA interventions can improve sleep quality in other populations (Lan *et al.*, 2014; Yang *et al.*, 2012).

In addition, it is important to conduct a qualitative analysis to evaluate the implementation, delivery and acceptability of

the intervention as this is the key to the development of future research which this pilot study aims to inform. Therefore, semi structured interviews will be conducted with the study participants and with the healthcare professionals providing the intervention. This will aim to inform future studies in this evolving research area. The current protocol serves to:

- Describe the methodology that will be implemented to evaluate a 12-week intervention consisting of wearable technology with remote monitoring, personalised text message feedback and goal setting amongst adults with CF attending University Hospital Limerick
- Describe the health outcomes in adults with CF that will be evaluated in the trial including PA levels, lung function, aerobic capacity, body composition, quality of life, wellbeing and sleep
- Describe the nested process evaluation through the conduct of semi structured interviews amongst adults with CF and healthcare practitioners regarding the implementation, delivery and acceptability of the intervention.

Methods

Study design

This study represents a single centre pilot randomised trial which will compare the effect of wearable technology with personalised text message feedback and goal setting to wearable technology alone in adults with CF. The CONSORT standardised reporting guidelines will be followed to ensure the standardised conduct and reporting of the research (Schulz *et al.*, 2010). This protocol was registered on ClinicalTrials.gov (NCT03672058) on 14th September 2018 and prepared in accordance with the Standard Protocol Items: Recommendations for Interventional Trials (SPIRIT) guidelines (see Figure 1 and Reporting guidelines).

Setting

The study will take place in the Adult CF Unit, University Hospital Limerick, Ireland. Baseline and follow-up assessments will take place via routine clinic outpatient appointments. Each adult with CF attends their routine clinic appointment every three months. Therefore, we chose this duration for our intervention to dovetail with routine CF care and limit the need for the adult with CF to attend on a more regular basis for testing appointments. This was decided through public and patient involvement (PPI) (completed with adults with CF) and through clinician experience. If the participant cannot attend on a particular clinic day, then they will be offered an additional appointment to suit their schedule. Due to the nature of the intervention, blinding of participants is not possible.

Ethical approval

Ethical Approval was obtained from the University Hospital Limerick Research Ethics Committee (Approval number 054/18).

Population and recruitment

Recruitment strategy. Participants deemed eligible for inclusion to the study based on the inclusion/exclusion criteria

will be approached by the study gatekeeper (CF Physiotherapist – LC) and provided with an outline of the study during their routine clinic outpatient appointment. Participants will be provided with an information leaflet and will be offered an opportunity to ask questions about participation in the study. Prospective participants will then be asked to sign a consent form. Consent and mechanisms relating to data controlling and processing will be compliant with the EU General Data Protection Regulation 2016/679 and in compliance with the Data Protection Act 2018 [(Section 36(2)) (Health Research) Regulations 2018].

Sample size

As this is the first study of its kind a sample size cannot be determined. There are 80 adults with CF attending this CF clinic. Based on inclusion/exclusion criteria at this CF centre in University Hospital Limerick, it is intended to recruit up to 50 participants.

Inclusion criteria

- Age \geq 18 years
- Confirmed diagnosis of CF (based on CF-causing mutations and/or a sweat chloride concentration during two tests of > 60 mmol/l)
- Clinically stable patients with CF attending University Hospital Limerick, determined by those who are not experiencing a pulmonary exacerbation. For the purpose of this study pulmonary exacerbation will be defined as acute or subacute worsening of respiratory symptoms which warrant change in treatment (i.e., new oral or intravenous antibiotics), as per previous research (Savi *et al.*, 2015).
- Access to a smartphone/tablet to access and ability to upload to Fitbit Application
- Capacity and willingness to give explicit informed consent

Exclusion criteria

- $FEV_1 < 25\%$. Individuals with $FEV_1 < 25\%$ would typically require supplemental oxygen for exercise and at this point may require transplant assessment. Furthermore, they are more likely to experience exacerbations and would not be suitable for a study over 24 weeks. This is a lower cutoff FEV_1 than most studies would include (Hebestreit *et al.*, 2010; Klijn *et al.*, 2004; Kriemler *et al.*, 2013).
- Patients on the waiting list for lung transplantation and those who have undergone lung transplantation.
- Patients with an exacerbation in the four weeks prior to the study. Patients can undergo testing once they are finished their antibiotics and deemed clinically stable by the Respiratory Consultant (BC).
- Patients dependent on supplemental oxygen for exercise.
- Adults with CF who are pregnant

TIMEPOINT	Study Period			
	Enrolment	Allocation	Post Allocation	
			12 weeks	24 weeks
ENROLMENT:				
Eligibility screen	X			
Informed consent	X			
Baseline Assessment	X			
Allocation		X		
INTERVENTIONS:				
Fitbit plus Text Messaging Feedback & Goal Setting, progressing to Fitbit Only after week 12			←————→	←————→
Fitbit Only			←————→	
ASSESSMENTS:				
Descriptive Data		X		
Lung Function		X	X	X
Aerobic Capacity		X	X	X
Number of steps/day		X	X	X
Quality of Life (CFQR)		X	X	X
Grip Strength		X	X	X
Body Composition		X	X	X
International Physical Activity Questionnaire		X	X	X
Pittsburgh Sleep Quality Index		X	X	X
UCSD Shortness of Breath Questionnaire		X	X	X
Wellbeing (AWE score)		X	X	X
Exacerbation Rates		X	X	X

Figure 1. Study Schedule.

- Patients with any cardiac, neurological or musculoskeletal impairment that may impact on their ability to participate in the PA intervention will not be eligible to take part in this study
- Participation in another clinical trial up to 4 weeks prior to the first baseline visit

Randomisation

Should participants explicitly consent to participate in the study, they will undergo baseline testing. To minimise the

possibility of selection bias, a researcher independent of the recruitment process (MC) will complete the first random allocation using a sealed opaque envelope. Following this a minimisation randomisation procedure will be completed based on lung function, where FEV₁ of >80% predicted lung function will be classified as having mild lung disease. While those with an FEV₁ of 50–79% predicted lung function will be classified as having moderate lung disease, 30–49% as severe lung disease and <30% indicating very severe lung disease (Ranu *et al.*, 2011). Allocation will be revealed after recruitment and baseline assessments have occurred. Participants will then either

receive the intervention (Fitbit with text messaging feedback and goal setting) or the active comparator (Fitbit only).

Experimental and active comparator intervention Intervention

The intervention consists of wearable technology, text message feedback and goal setting.

Wearable technology:

If the participant is allocated to the intervention, they will be provided with wearable technology (Fitbit Charge 2), educated on how to use it, and this will also be linked to an online monitoring system (Fitabase). Participants will be encouraged to enable Bluetooth and upload data regularly. Fitabase, the online monitoring system enables the physiotherapists to access step count data remotely. When a participant enables Bluetooth and syncs their Fitbit through the Fitbit app, this automatically updates participant data on the Fitabase website. Engagement with the technology will be monitored through systematic review of online data. Participants in both groups will receive a “reminder” message on his or her mobile phone if their data has not been synced via the Fitbit App in the previous seven days. This ensures that data is continuously collected over the full study duration. The data collected through the Fitbit Charge 2 and recorded on the Fitabase system are step count, activity minutes and sleep. However, for the purpose of this study, the researchers are only investigating step count alone. Any further research would require the Fitbit to be assessed under each of these conditions to ensure validity and reliability. This is outside the scope of this research.

Goal setting:

The physiotherapist will discuss the participant’s PA levels (as measured at baseline by an accelerometer) and individual patient centred PA goals will be set with each participant. These will be discussed with their physiotherapist to ensure they are specific, measurable, achievable, realistic and timed (SMART). They will be encouraged to write a minimum of three goals. Participants will be asked to set a step count target for week four, eight and 12 and consider ways to achieve these goals, both of which will be discussed with the physiotherapist during the baseline appointment. Goals will be individualised to the participant taking into account their preferences. Participants may also set goals related to health outcomes being assessed – for example, a participant may have a goal to improve lung function, aerobic capacity, sleep etc. during the study period. The text message feedback will refer to step goals only. The participant will be given a copy of their goals.

Text message feedback:

Every week participants will be sent a one-way personalised text message by their CF physiotherapist for 12 weeks overall. The text messages in this study will be personalised, one-way texts with positive reinforcement on step count attained by the participant.

A meta-analysis found personalisation strategies, such as using a participant name has been proven to increase intervention

efficacy (Head *et al.*, 2013) and tailoring messages is a key ingredient in text messaging interventions (Noar *et al.*, 2007). Furthermore, one way communication is as effective as two-way dialogue (Head *et al.*, 2013). Therefore, this study investigated one-way messages only.

This individualised message will be sent to each participant once a week, at a day and time chosen by the participant at the start of the intervention. The authors will follow a standardised approach. The message will address the participant by name and will have positive feedback in the form of encouragement and praise messages (Adams *et al.*, 2013) in relation to their step count attained in the previous week. It will then focus on a PA related goal set by the subject and will sign off with a further positive comment. Research has indicated that it is crucial to reinforce improvements to develop new behaviour or to strengthen a habit (Bandura, 2004; Glanz *et al.*, 2008; Hovell *et al.*, 2009). Each time a participant meets his/her goal they will receive positive feedback. Participants who do not meet the goal will be provided with a positive comment in relation to their step count achieved to date and provided their next step goal. This is to avoid negative messages that could be discouraging (Adams *et al.*, 2013). If the participant does not achieve their target step count goal then the physiotherapist will set a target increase of 10% from the mean step count achieved in the previous week.

A sample text message is as follows:

Hi Paul, Well done on achieving an average daily step count of 7,600 steps this week. Next week aim to hit your goal of 8,000 steps. Keep up the good work!

Comparison

If the participant is allocated to the active comparator, they will be provided with a Fitbit Charge 2 and educated on how to use it and this will also be linked to “Fitabase” for data collection purposes. However, no feedback will be provided to the participants on their PA levels throughout the study period.

Follow Up

At week 12 both groups will have outcome measures re-assessed. Both groups will continue with the Fitbit Charge 2 only for the following 12 weeks. At the end of the 24 weeks participants will have all outcome measures repeated.

Subsequently, a qualitative assessment will be conducted through semi structured individual interviews to determine participants’ satisfaction and feedback on the intervention and their suggestions going forward. The interview guide is available as extended data to this manuscript (Curran, 2020). These interviews will be completed by a CF Physiotherapist involved in the study (LK) using a digital voice recorder and will be transcribed verbatim. The transcribed texts will be coded independently by two investigators who will develop code books. These will be compared and agreed upon. Data will be analysed using NVIVO V.11 Plus (QSR International Pty Ltd) and using the six steps for thematic analysis, in order to highlight the central themes to this study (Braun & Clarke, 2006).

Exacerbations

If the participant has an exacerbation of CF during the study period, their involvement in the study will be paused, and re-started 1 month later.

Baseline Assessment of step count

The ActivPAL accelerometer (PAL technologies) will be used to assess step count at baseline. This will be used to inform goal setting only. This forms part of another study which assessed PA behaviour in adults with CF. Participants will wear the accelerometer for seven days. Based on PPI it was reported by the participants that they did not want to wear an ActivPAL other than at baseline as it is a thigh worn device which is not 'fashionable'. Similar concerns have been cited previously in the research (Dias *et al.*, 2012). Data will be included if at least four full days (at least three weekdays and one weekend day) of measurements with a minimum of 10 hours (h) for the weekdays and 10 h for the weekends are measured (Freedson *et al.*, 2005).

Usual Care

Usual care will be provided to all participants in both groups. This would typically include PA and exercise advice as per Australian and New Zealand Cystic Fibrosis guidelines. People with CF are advised to exercise for at least 30 minutes, five days a week. They are advised to include a combination of aerobic and resistance training. It is personalised depending on patient preference, as per recommendations (Button *et al.*, 2016).

Instrumentation/outcome measures

A range of outcome measures will be employed to identify the potential impact of this study on health outcomes in adults with CF. Each of these outcomes will be assessed at baseline, at 12 weeks and 24 weeks.

Primary

Fitbit step count data

The Fitbit Charge 2 (Fitbit Inc.) will be used to record step count during this study with data uploaded to Fitabase (Small Steps Labs LLC). Prior to this intervention, the validity and reliability of the Fitbit Charge 2 to measure step count in CF was assessed by the same research group. Twenty-one participants were recruited from an adult CF Centre for a single session of testing. Participants walked for five minutes at five pre-determined speeds in a controlled testing environment (2, 2.5, 3, 3.5 and 4 miles per hour on a treadmill) and at three self-selected speeds on a corridor (slow, medium and fast). The Fitbit Charge 2 was compared to visual observation. It was found that the Fitbit Charge 2 ranged from weak to very strong correlations when compared to visual observation (0.34–0.84). The Fitbit Charge 2 underestimated step count by 2.8%–9.2%. This is within acceptable limits of variability based on previous research (Schneider *et al.*, 2004). Therefore, the Fitbit Charge 2 is a valid and reliable measure to assess step count in adults with CF (Curran *et al.*, 2020).

Secondary

Cardiopulmonary exercise testing

The reference standard exercise test is an incremental cardiopulmonary exercise test (CPET), utilising a ramp protocol. The ramp protocol ensures that work rate is progressively and linearly incremented until maximal effort is achieved (Herdy *et al.*, 2016). All standard CPET outcomes will be considered including VO_2 max, duration, load, pulmonary ventilation (VE), Respiratory Exchange Ratio (RER), ventilatory equivalents for oxygen (VE/VO_2) and for carbon dioxide (VE/VCO_2). This will be conducted using the Medisoft Ergocard Professional CPET equipment and analysed by ExpAir, the Medisoft software. Maximal exercise testing is an independent predictor of mortality in CF (Nixon *et al.*, 1992). Breath-by-breath ventilatory gas analysis allows the accurate measurement of maximal oxygen uptake ($\text{VO}_{2\text{max}}$) – the gold standard measure of exercise capacity (Urquhart, 2011). Supramaximal verification be conducted to ensure a maximal effort during the CPET. After an incremental CPET, participants will be provided with a ten-minute break. They will then be asked to complete a supramaximal exercise test (Causser *et al.*, 2018; Saynor *et al.*, 2013) whereby participants will exercise at 110% of their power output (as determined by CPET test previously). This test has been conducted amongst adults with CF previously and has been deemed safe and ensures validity of the results which are obtained in a CPET test.

Spirometry

Spirometry will be performed according to American Thoracic Society (ATS) standard techniques (Miller *et al.*, 2005) using the Carefusion Microlab spirometer. Values will be expressed as a percentage of the predicted value for height, sex and age for adults (Hankinson *et al.*, 1999). Forced expiratory volume in one second (FEV_1) will be used to classify the severity of CF lung disease for each participant. It will also be used to determine the effect the intervention may have on pulmonary function. Other pulmonary function measures such as % forced vital capacity (FVC) and forced expiratory flow (FEF_{25-75}) will also be considered.

International Physical Activity Questionnaire (IPAQ)

The IPAQ is a self-reported measure of PA and relies on user recall over the previous seven days. This tool was developed to assess PA levels using a questionnaire, has very good repeatability and is as reliable as other measures for self-reported PA (Craig *et al.*, 2003). This nine-item questionnaire relies on participant recall and records PA at four levels: vigorous (e.g. aerobics), moderate (e.g. leisure cycling), walking and sitting.

Hand dynamometry

Grip strength will be recorded as a measure of general musculoskeletal strength, independent of lower limb strength. Previous research has shown that grip strength is correlated to lung function and VO_2 peak (Wells *et al.*, 2014). This will be conducted using a Jamar Hydraulic Hand Dynamometer. Test – retest reliability has been proven in respiratory patients

(Dowman *et al.*, 2016). The participant will be asked to stand and hold the dynamometer in by their side, with their elbow at 90 degrees and forearm in neutral. They will be instructed to squeeze the device as hard as possible. They will be provided with a 30 second rest period between each trial. The greater of two trials from each hand will be used and added together to give overall handgrip strength. This will be measured in kilograms (Martínez-García *et al.*, 2020).

Bioelectrical Impedance Analysis (BIA)

BIA is an easily available, quick method to assess body composition. A low voltage current will be passed through the body, whereby impedance (tissue resistance and reactance) is measured. This will be conducted using the Seca 515 Medical Body Composition Analyser. BIA will be performed with the participant standing barefoot on the instrument platform as per manufacturers guidelines (Seca, Birmingham, United Kingdom). The device has an integrated scale and uses four pairs of electrodes of stainless steel that are positioned at each hand and foot, through which the current enters the limbs.

CF Quality of Life Questionnaire Revised (CFQR)

The CFQR questionnaire is a fully validated disease specific measure consisting of 52 items across nine domains of functioning which have been identified by, and are of importance to, adolescents and adults with CF. This questionnaire is valid, sensitive and has strong test-retest reliability (Gee *et al.*, 2000). The minimum clinically important difference for the CFQR – respiratory score is 4 points (Quittner *et al.*, 2009).

Pittsburgh Sleep Quality Index (PSQI)

The PSQI is a self-rated questionnaire used to measure the quality and patterns of sleep in adults. It differentiates “poor” from “good” sleep quality by measuring seven areas (components): subjective sleep quality, sleep latency, sleep duration, habitual sleep efficiency, sleep disturbances, use of sleeping medications, and daytime dysfunction over the last month. The PSQI is effective in assessing sleep quality in CF (Milross *et al.*, 2002).

The University of California San Diego (UCSD) Shortness of Breath Questionnaire

This questionnaire assesses dyspnoea associated with activities of daily living (ADLs). There are 24 items on this questionnaire. Each item is assessed on a 6-point scale (0 = “not at all” to 5 = “maximal or unable to do because of breathlessness”). Scores range from 0 to 120 with higher scores indicating activities of daily living are extremely limited by shortness of breath. It has been validated to assess dyspnoea over time in respiratory patients (Swigris *et al.*, 2012)

Awescore

This questionnaire assesses state of wellness to assist in providing best health care (Button *et al.*, 2014). There are ten questions, which are scored from 0–10. 10 reflects most well state of being possible while zero reflects least well state. Scores range from 0–100 with higher scores indicating good state of wellness. This is a reliable tool for measurement of

multidimensional wellness in adults with CF that is appealing to patients (Button *et al.*, 2014).

Data collection & management

Outcome assessment will be conducted by qualified physiotherapists in the Adult CF Unit. Assessors cannot be blinded as the intervention involves the CF physiotherapists for the delivery of weekly text messages, who will also be assisting with repeating objective outcome measures. All self-report measures will be completed by the participants independently, however a research assistant (RA) (AJ) will be available if there are any queries or concerns. These subjective assessments will be analysed by the RA who is blinded to group allocation.

Each participant in the study will be assigned a numerical identifier code. Aggregate data will be anonymised. Paper copies of all measures will be identifiable only by the identifier code. All data will be stored in locked filing cabinets in locked offices in the Adult CF Unit at University Hospital Limerick. All identifying paper data (e.g. signed consent forms and forms listing participant codes) will be stored in a separate locked filing cabinet to all other anonymised data and will be accessible only to the research team. All interviews will be coded and entered to a study data file. These computer files will be stored on the hard drive of a password protected desktop computer by the study lead investigator (MC). All audio and electronic data will be stored on encrypted hard drives.

Data analysis

Step count data will be collected over seven days and an average weekly step count will be obtained. It is anticipated that this step count data will be analysed to assess for a novelty effect in the first two weeks, at week six and 12 during the intervention and at week 18 and Week 24 for the follow up. Appropriate descriptive statistics will be used to describe the baseline characteristics of study participants. These will include proportions, percentages, ranges, means and standard deviations and medians and interquartile ranges (where data are not normally distributed). Data will be assessed for normality. Parametric tests will be used to compare differences across groups where data are normally distributed, and the non-parametric equivalent will be applied in cases where data are not normally distributed. A p-value <0.05 will be considered significant. Primary analyses will be performed according to intention-to-treat (ITT) principles with all participants who were originally allocated by randomisation and those who dropped out from the study. In case of missing values, a multiple imputation method, inverse probability weighing or mixed models will be used to handle the missing data. Data will be presented as means and SDs for data that is normalised and medians (IQR) for data that is skewed. A repeated measures ANOVA, controlling for baseline values and other potential confounders will be conducted. Bonferroni correction will be applied for multiple testing.

Dissemination of information

The results of this study will be disseminated via peer-reviewed publications and conference presentations.

Study status

Recruitment began in January 2019. It is anticipated this will be completed by June 2020.

Discussion

This is the first study to assess the effect of wearable technology with personalised text message feedback and goal setting on PA and key health outcomes in adults with CF. Previous research has investigated the feasibility and acceptability of a telehealth intervention in adults with CF with positive results reported for such an intervention (Cox *et al.*, 2015). The use of wearable technology with remote monitoring is a novel concept in CF.

Regular PA is a well-accepted and valued part of CF care (Dwyer *et al.*, 2011; Rand & Prasad, 2012). The primary aim of this research is to increase PA in adults with CF. Adherence to any PA intervention is a vital component of any study, particularly in chronic disease. The use of pedometers to provide feedback on PA levels can increase awareness of daily PA, provide motivation and visual feedback (Lauzon *et al.*, 2008). Pedometers can increase PA in an adult population (Bravata *et al.*, 2007), in a young population (Lubans *et al.*, 2009) and in chronic obstructive pulmonary disease (Hospes *et al.*, 2009). To date, there is limited research conducted assessing wearable technology such as Fitbits in this population.

Previous literature has demonstrated that a partially supervised programme in CF can improve health outcomes and is easily implemented (Hebestreit *et al.*, 2010). Other partially supervised interventions have investigated the effect of providing personalised text message feedback on PA levels which has enhanced adherence to home based programmes in other populations (Blaauwbroek *et al.*, 2009; Strath *et al.*, 2011). Furthermore, a systematic review found that having a step goal was the key predictor of increasing PA. Studies which did not include a step goal as part of their intervention showed no significant improvement in PA in comparison to those that set step goals (Bravata *et al.*, 2007).

The secondary aims of this research is to consider the effect of this intervention on other important endpoints such as lung function, aerobic capacity, quality of life, wellbeing and sleep. The broad range of outcomes assessed in this study will facilitate a comprehensive review on the impact of this research on participants with CF, both subjectively and objectively. A strength of this study is the use of the Fitbit Charge 2 which will allow participants to update their PA levels through the Fitbit App which will enable the assessors to review data remotely and this will also provide feedback on compliance to the intervention.

There is no true control group in this study for a couple of reasons. Firstly, the primary aim of this study was to determine if a partially supervised programme would be effective in improving PA and health, and it is unknown if any of these key outcomes could be improved with the provision of a Fitbit alone. Therefore, it was deemed more appropriate to compare the intervention with the Fitbit alone rather than a true

control group. Secondly, based on clinical experience and PPI, both the CF Physiotherapists and adults with CF felt that this would be a limitation to recruitment and retention if only one group received the Fitbit.

Gene modifiers and potentiators may be a confounder in this study. This will be monitored throughout the study duration and will be controlled for during statistical analysis, if necessary. The research team will also monitor exacerbation rates (participants requiring oral or IV antibiotics during the study period), hospital admission rate and colonisation status.

Limitations

It is not possible to blind participants in PA intervention research. Due to lack of resources it is not possible to blind the CF Physiotherapists who will be assisting with text messaging feedback and repeating objective outcome measures, however self-report measures will be analysed by a research assistant blinded to group allocation. There may be a selection bias towards more physically active adults with CF as those who are more active may be more likely to partake in this study.

The results of this study may provide valuable insights into the development of a larger definitive trial exploring the use of wearable technology for optimising PA and health outcomes in adults with CF.

Ethics approval and consent to participate

The study received ethical approval from the University Hospital Limerick Research Ethics Committee (Ref: 054/18). Written informed consent will be obtained from all study participants.

Data availability

Underlying data

No data are associated with this article

Extended data

Open Science Framework: Steps Ahead: optimising physical activity and health in people with cystic fibrosis. Study protocol for a pilot randomised trial <https://doi.org/10.17605/OSF.IO/DEGHT> (Curran, 2020)

This project contains the following extended data:

- Participant Consent Form
- Participant Information leaflet
- Interview Guide

Reporting guidelines

Spirit Checklist for Steps Ahead: optimising physical activity and health in people with cystic fibrosis. Study protocol for a pilot randomised trial <https://doi.org/10.17605/OSF.IO/DEGHT> (Curran, 2020)

Data are available under the terms of the [Creative Commons Zero "No rights reserved" data waiver](https://creativecommons.org/licenses/by/4.0/) (CC0 1.0 Public domain dedication).

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Open Peer Review

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This title is better able to reflect the study objectives.

Competing Interests: No competing interests were disclosed.

Reviewer Expertise: Limb muscles in adults with cystic fibrosis.

I confirm that I have read this submission and believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard.

Version 2

Reviewer Report 12 November 2020

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This is an improved version of the document. I still have some concerns on how the study design would be able to achieve what the objective of this study set out.

My understanding is the experimental group (Exp) receives wearable technology (WT) + goal setting (GT) + text message feedback (TF), while the "active comparators" (Control) will only be given WT, i.e.

Exp: WT + GT + TF

Control: WT

I'm curious how this study design would be able to tell if wearable technology can change the physical activity level, if both groups receive WT intervention. Perhaps you did mean wearable technology = Fitbit Charge 2 + GT + TF, while the control group only uses Fitbit and thus not considered wearable technology. Or you need to change the wording of the objective (and the title), by replacing "wearable technology" by goal setting and feedback.

Also, can the subjects be able to see their step count on the Fitbit Charge 2, if so, do you think the goal setting and text message are "external" feedback that would be more helpful than what the patients can do for themselves.

Competing Interests: No competing interests were disclosed.

Reviewer Expertise: Limb muscles in adults with cystic fibrosis.

I confirm that I have read this submission and believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard, however I have significant reservations, as outlined above.

Author Response 13 Nov 2020

Maire Curran, University of Limerick, Limerick, Ireland

Dear Dr Wu,

Thank you for your feedback.

The intervention group will receive the wearable technology, goal setting and text message feedback. Therefore, we have added "goal setting and text message feedback" to the title: **Steps Ahead: optimising physical activity in adults with cystic fibrosis: Study Protocol for a pilot randomised trial using wearable technology, goal setting and text message feedback.**

These objectives have already been stated throughout the introduction and methods. We do not feel that we should remove the words "wearable technology" as it is through the use of wearable technology that physiotherapy staff will see PA data remotely and send text

message feedback on same and it is an important component of our study. Furthermore, the wearable technology will allow the participants to monitor their daily PA, as well as hopefully encourage further PA in order to attain their goals.

The objective of this study is to determine if the intervention (wearable technology, goal setting and text message feedback) will increase step count more than wearable technology alone. Subjects in both groups will be able to see their step count on the Fitbit Charge 2. We think that goal setting and text message feedback will positively impact on PA levels rather than just providing participants with the Fitbit alone. We feel this external feedback may be more beneficial than what the patient can do themselves. The provision of a Fitbit to the active comparator group should enable us to determine if this is the case.

Competing Interests: No competing interests were disclosed.

Reviewer Report 05 November 2020

<https://doi.org/10.21956/hrbopenres.14198.r28127>

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Sarah Rand 

UCL Great Ormond Street Institute of Child Health, University College London, London, UK

Thank for asking me to review this updated version. The authors have obviously taken all comments on board and have made many useful revisions and additions to the article based on the feedback provided. The article in my opinion is now a much clearer and more comprehensive outline of the protocol.

I have no further comments or suggestions to make at this stage.

Competing Interests: No competing interests were disclosed.

Reviewer Expertise: Cystic Fibrosis, Physical activity, exercise, health technology.

I confirm that I have read this submission and believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard.

Version 1

Reviewer Report 24 August 2020

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Kenneth Wu

¹ Toronto Adult Cystic Fibrosis Centre, St. Michael's Hospital, Unity Health Toronto, Toronto, ON, Canada

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This is an interesting study protocol. The title sounds catching. Nevertheless, I wonder the wording should be more specific, e.g. the word "health" is rather vague. The primary outcomes of this study are exercise capacity (with CPET) and lung function (spirometry), perhaps it can be more specific. Also, the study participants are all adults. The word "adults" should be used instead of "people", as the study design, hence the implications, may be different if it was with children.

For the concept of physical activity, the authors talked about exercise and physical activity as two different entities in the Introduction, then the focus switched to only physical activity. I wonder how the authors conceptualize physical activity in this study. Does physical activity include exercise for this study? Many studies separated physical activity to various intensities - mild, moderate, and vigorous intensities (i.e. including exercise), e.g. Trooster *et al.* (2009)¹.

Study Design

In terms of intervention, I am curious how the physiotherapist re-assess participant's goals while a one-way text message was used. Should goal-setting involve a discussion between the patient and health-care providers? Or was it more a physical activity prescription?

I don't think the study design can achieve the study objectives when the study participants in the control group also wear the Fitbit (while the study objectives were to explore the impact of a fitness tracker, text message personalised feedback and goal setting on physical activity and health outcomes).

Since the sample size was not calculated based on previous studies, it would be helpful to provide information on the size of the Adult CF clinic.

It is unclear in the Methods how study participants were recruited. Were they approached when they were going for an outpatient CF appointment? Or was it done over the phone or via Email? Also, related to this, was study conducted using convenience sampling?

In terms of exclusion criteria, should patients having comorbidities that might affect their participation in physical activity (e.g. people with cardiac, neuromuscular issues, if they had injuries to their ligaments/tendons etc, or mental health issue). Also, what if the study participants do lots of "physical activity" or exercise already, before they were enrolled in the study, would they be expected to have any effects from the intervention?

It is unclear why the group that also wore the fitness tracker (only) and why having people wearing the fitness tracker for 12 more weeks. Were the extra 12 weeks function like a control

period? If so, would there be any carry-over effect in your experimental group if that's the case?

For the handgrip strength assessment, I am not sure if it really indicates "physical function." In the CF literature, handgrip strength was used as a surrogate measure for general muscle strength (Martinez-Garcia *et al.*, 2020²; Rietschel *et al.*, 2008³; Ward *et al.*, 2013⁴). Physical function is a broader concept than muscle strength. I am curious why a study in ILD should be cited here.

Since your outcome measures were mostly self-report, or else they are easy to perform (and to learn), and there were many people working on this study, would it enhance the quality of the study by blinding the assessor (i.e. not using the same physiotherapist providing the feedback to the study participants).

Statistical Analysis

There are 10 outcome measures in this study. A p -value of $p = 0.05/10 = 0.005$ should be used to determine statistical significance. Perhaps the authors should consider limiting the number of outcome measures. For example, can IPAQ be eliminated since the fitness tracker was keeping track of the same/similar information?

Even if only the primary outcomes are considered, the p -value of $p = 0.05/2 = 0.025$ should be used.

Minor Correction Recommendation

In the Introduction and Abstract section, "pilot randomised trial" was written in this order, but in the Study Design section and perhaps some other places, it was put as "randomised pilot trial". To me, the former order seems correct.

References

1. Troosters T, Langer D, Vrijnsen B, Segers J, et al.: Skeletal muscle weakness, exercise tolerance and physical activity in adults with cystic fibrosis. *Eur Respir J.* 2009; **33** (1): 99-106 [PubMed Abstract](#) | [Publisher Full Text](#)
2. Martínez-García MDM, Rodríguez-Juan JJ, Ruiz-Cárdenas JD: Influence of sex gap on muscle strength and functional mobility in patients with cystic fibrosis. *Appl Physiol Nutr Metab.* 2020; **45** (4): 387-392 [PubMed Abstract](#) | [Publisher Full Text](#)
3. Rietschel E, van Koningsbruggen S, Fricke O, Semler O, et al.: Whole body vibration: a new therapeutic approach to improve muscle function in cystic fibrosis?. *Int J Rehabil Res.* 2008; **31** (3): 253-6 [PubMed Abstract](#) | [Publisher Full Text](#)
4. Ward N, White D, Rowe H, Stiller K, et al.: Physical activity levels of patients with cystic fibrosis hospitalised with an acute respiratory exacerbation. *Respir Med.* 2013; **107** (7): 1014-20 [PubMed Abstract](#) | [Publisher Full Text](#)

Is the rationale for, and objectives of, the study clearly described?

Yes

Is the study design appropriate for the research question?

Partly

Are sufficient details of the methods provided to allow replication by others?

No

Are the datasets clearly presented in a useable and accessible format?

Not applicable

Competing Interests: No competing interests were disclosed.

Reviewer Expertise: Limb muscles in adults with cystic fibrosis.

I confirm that I have read this submission and believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard, however I have significant reservations, as outlined above.

Author Response 30 Sep 2020

Maire Curran, University of Limerick, Limerick, Ireland

Dear Dr Wu,

Re: Steps Ahead: optimising physical activity and health in people with cystic fibrosis: Study Protocol for a pilot randomised trial

This is an interesting study protocol. The title sounds catching. Nevertheless, I wonder the wording should be more specific, e.g. the word "health" is rather vague. The primary outcomes of this study are exercise capacity (with CPET) and lung function (spirometry), perhaps it can be more specific. Also, the study participants are all adults. The word "adults" should be used instead of "people", as the study design, hence the implications, may be different if it was with children.

We sincerely thank you for your detailed and critical analysis of our article. Please find our responses below to your feedback (in bold). All changes have been made and are illustrated through italics and will be submitted in the second version of this article.

We agree the title should be more specific and given reviewers feedback we have changed the title to *"Steps Ahead: optimising physical activity in adults with cystic fibrosis: Study Protocol for a pilot randomised trial using wearable technology"*.

For the concept of physical activity, the authors talked about exercise and physical activity as two different entities in the Introduction, then the focus switched to only physical activity. I wonder how the authors conceptualize physical activity in this study. Does physical activity include exercise for this study? Many studies separated physical activity to various intensities - mild, moderate, and vigorous intensities (i.e. including exercise), e.g. Trooster *et al.* (2009)¹.

Thank you for this feedback. We have clarified PA in the introduction. This study is focusing on step count only as a proxy measure for PA.

"PA can be described as any bodily movement that causes an increase in energy expenditure

above that of resting energy expenditure including leisure-time PA, occupational PA and exercise (Caspersen et al., 1985). Exercise and physical activity (PA) are widely documented in consensus statements as key components in the management of CF (Castellani et al., 2018). PA has several positive benefits in this population as it has been shown to improve sputum clearance (Dwyer et al., 2009), bone mineral density (Gupta et al., 2019) and muscle strength (Burtin et al., 2013). More importantly, PA can improve aerobic capacity (Hebestreit et al., 2010) and it can slow the rate of decline in lung function (Schneiderman et al., 2014), both of which are linked to increased survival in CF (Nixon et al., 1992). As a result, the optimisation of exercise and PA among PWCF is important, however, a Cochrane review found that there was a lack of evidence regarding strategies to promote PA in this population and consideration should be given towards telemedicine applications and health coaching (Cox et al., 2013b).

Numerous subjective and objective methods are reported in the literature for the assessment of PA in adults with CF. Step count measurement can be used to quantify and monitor PA behaviours (Tudor-Locke and Bassett, 2004). Despite the clear benefits of PA in CF, a recent systematic review found that adults with CF fail to meet recommended PA and step count guidelines (Shelley et al., 2019). Therefore, interventions which aim to increase PA in this population are of considerable interest.

Study Design

In terms of intervention, I am curious how the physiotherapist re-assess participant's goals while a one-way text message was used. Should goal-setting involve a discussion between the patient and health-care providers? Or was it more a physical activity prescription?

Thank you for this feedback. The following text has been added to the intervention section to ensure clarity around text messaging and goal setting:

“The text messages in this study will be personalised, one-way texts with positive reinforcement on step count attained by the participant. A meta-analysis found personalisation strategies such as using a participant name has been proven to increase intervention efficacy (Head et al., 2013) and tailoring messages is a key ingredient in text messaging interventions (Noar et al., 2007). Furthermore, one way communication is as effective as two-way dialogue (Head et al., 2013). Therefore, this study investigated one-way messages only.

An individualised message will be sent to each participant once a week, at a day and time chosen by the participant at the start of the intervention. The authors will follow a standardised approach. The message will address the participant by name and will have positive feedback in the form of encouragement and praise messages (Adams et al., 2013) in relation to their step count attained in the previous week. It will then focus on a physical activity related goal set by the subject and will sign off with a further positive comment. Research has indicated that it is crucial to reinforce improvements to develop new behaviour or to strengthen a habit (Hovell et al., 2009, Glanz et al., 2008, Bandura, 2004). Each time a participant meets his/her goal they will receive positive feedback. Participants who do not meet the goal will be provided with a positive comment in relation to their step count achieved to date and provided their next step goal. This is to avoid negative messages that could be discouraging (Adams et al., 2013).”

I don't think the study design can achieve the study objectives when the study

participants in the control group also wear the Fitbit (while the study objectives were to explore the impact of a fitness tracker, text message personalised feedback and goal setting on physical activity and health outcomes).

Thank you for this comment. However, we feel that the study design can achieve the study objectives. The control group will wear the Fitbit also, however we are keen to investigate if using the wearable technology with text message feedback combined with goals will impact on physical activity and health outcomes. We felt it would be important to consider whether the Fitbit alone could achieve increases in PA.

The control group is not 'usual care' so essentially, we are assessing the use of text messaging feedback and the Fitbit is to inform this process.

Furthermore, based on PPI, we found that adults with CF reported they would be less likely to comply if they didn't also receive a Fitbit.

We considered other study designs such as a crossover study. However, we felt this was too complex for a small pilot study and taking into account overall patient burden.

Since the sample size was not calculated based on previous studies, it would be helpful to provide information on the size of the Adult CF clinic.

Thank you for this comment. We have provided further information under sample size.

"There are 80 adults with CF attending this CF clinic. Based on inclusion/exclusion criteria at this CF centre in University Hospital Limerick, it is intended to recruit up to 50 participants."

It is unclear in the Methods how study participants were recruited. Were they approached when they were going for an outpatient CF appointment? Or was it done over the phone or via Email? Also, related to this, was study conducted using convenience sampling?

Participants were approached during their outpatient CF appointment which they attend every three months. Convenience sampling was not used as all people with CF attending this CF clinic, who met the inclusion/exclusion criteria were asked if they would like to participate in this study.

"Participants deemed eligible for inclusion to the study based on the inclusion/exclusion criteria will be approached by the study gatekeeper (CF Physiotherapist – LC) and provided with an outline of the study during their routine clinic outpatient appointment. Participants will be provided with an information leaflet and will be offered an opportunity to ask questions about participation in the study. Prospective participants will then be asked to sign a consent form."

In terms of exclusion criteria, should patients having comorbidities that might affect their participation in physical activity (e.g. people with cardiac, neuromuscular issues, if they had injuries to their ligaments/tendons etc, or mental health issue).

Thank you for this suggestion. We have added the following to the exclusion criteria:

"Patients with any cardiac, neurological or musculoskeletal impairment that may impact on their

ability to participate in the PA intervention will not be eligible to take part in this study."

Also, what if the study participants do lots of "physical activity" or exercise already, before they were enrolled in the study, would they be expected to have any effects from the intervention?

Thank you for this feedback. We agree that those who are already engaged in a high level of physical activity would be less likely to see the benefits of the intervention. However, it is hoped that the intervention may lead to continued adherence to the PA/exercise guidelines in CF. The Fitbit will also provide the participants with the ability to self-monitor their progress. Furthermore, previous literature suggests that the majority of people with CF fail to meet their step count targets (Shelley et al., 2019).

It is unclear why the group that also wore the fitness tracker (only) and why having people wearing the fitness tracker for 12 more weeks. Were the extra 12 weeks function like a control period? If so, would there be any carry-over effect in your experimental group if that's the case?

The extra 12 weeks was to serve as a control period for both groups. As this was conducted as a pilot randomised study, the authors were unsure if the Fitbit alone may lead to an increase in any of the clinical outcome measures. And if an increase might be seen at 12 weeks (in for example step count/aerobic capacity etc.), would these be sustained at 24 weeks.

For the handgrip strength assessment, I am not sure if it really indicates "physical function." In the CF literature, handgrip strength was used as a surrogate measure for general muscle strength (Martinez-Garcia et al., 2020; Rietschel et al., 2008; Ward et al., 2013). Physical function is a broader concept than muscle strength. I am curious why a study in ILD should be cited here.

Thank you for this comment. Grip strength is a surrogate measure for general musculoskeletal strength but furthermore is correlated to lung function and VO₂ peak, both of which are key indicators of prognosis in CF. The ILD study was referenced as this study looked at the test re-test reliability of a handheld dynamometer. Test re-test reliability has not been conducted in adults with CF, however there would be some similarities across cohorts in this regard.

The following has been changed in the methods section for grip strength:

"Grip strength will be recorded as a measure of general musculoskeletal strength, independent of lower limb strength. Previous research has shown that grip strength is correlated to lung function and VO₂ peak (Wells et al., 2014). This will be conducted using a Jamar Hydraulic Hand Dynamometer. Test – retest reliability has been proven in respiratory patients (Dowman et al., 2016). The participant will be asked to stand and hold the dynamometer in by their side, with their elbow at 90 degrees and forearm in neutral. They will be instructed to squeeze the device as hard as possible. They will be provided with a 30 second rest period between each trial. The greater of two trials from each hand will be used and added together to give overall handgrip strength. This will be measured in kilograms (Martínez-García et al., 2020).

Since your outcome measures were mostly self-report, or else they are easy to perform (and to learn), and there were many people working on this study, would it enhance the quality of the study by blinding the assessor (i.e. not using the same physiotherapist providing the feedback to the study participants).

Thank you for this comment. We agree that ideally, we would prefer to use a blinded assessor, however this is not feasible in our current setting due to lack of resources.

"Assessors cannot be blinded as the intervention involves the CF physiotherapists for the delivery of weekly text messages, who will also be assisting with repeating objective outcome measures. However, all self-report measures will be completed by the participants and these subjective assessments will be analysed by a research assistant (RA) (AJ) who is blinded to group allocation."

Statistical Analysis

There are 10 outcome measures in this study. A p-value of $p = 0.05/10 = 0.005$ should be used to determine statistical significance. Perhaps the authors should consider limiting the number of outcome measures. For example, can IPAQ be eliminated since the fitness tracker was keeping track of the same/similar information? Even if only the primary outcomes are considered, the p-value of $p = 0.05/2 = 0.025$ should be used.

Thank you for highlighting this. The primary outcome measure is step count and this has been adjusted in the methods section of the article. Bonferroni correction will be applied for multiple testing.

Minor Correction Recommendation

In the Introduction and Abstract section, "pilot randomised trial" was written in this order, but in the Study Design section and perhaps some other places, it was put as "randomised pilot trial". To me, the former order seems correct.

Thank you for this feedback. We agree this wording was inconsistent. We have written it as "pilot randomised trial" throughout the document.

References:

Caspersen, C. J., Powell, K. E. and Christenson, G. M. (1985) 'Physical activity, exercise, and physical fitness: definitions and distinctions for health-related research', *Public health rep*, 100(2), pp. 126-31.

Martínez-García, M. d. M., Rodríguez-Juan, J. J. and Ruiz-Cárdenas, J. D. (2020) 'Influence of sex gap on muscle strength and functional mobility in patients with cystic fibrosis', *Applied Physiology, Nutrition, and Metabolism*, 45(4), pp. 387-392.

Shelley, J., Boddy, L. M., Knowles, Z. R., Stewart, C. E. and Dawson, E. A. (2019) 'Physical activity and associations with clinical outcome measures in adults with cystic fibrosis; a systematic review', *Journal of Cystic Fibrosis*.

Tudor-Locke, C. and Bassett, D. R. (2004) 'How many steps/day are enough?', *Sports medicine*, 34(1), pp. 1-8.

Wells, G. D., Wilkes, D. L., Schneiderman, J. E., Thompson, S., Coates, A. L. and Ratjen, F. (2014) 'Physiological correlates of pulmonary function in children with cystic fibrosis', *Pediatric pulmonology*, 49(9), pp. 878-884.

Competing Interests: No competing interests were disclosed.

Reviewer Report 25 June 2020

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Sarah Rand 

UCL Great Ormond Street Institute of Child Health, University College London, London, UK

Thank you for asking me to review this interesting and very topical protocol.

The authors have made a very good effort at identifying the need for undertaking this study (which is even more relevant given the current circumstances around the world) for PWCF. As the authors correctly say there is an ever increasing research base investigating telehealth for individuals with long term conditions such as CF so this is a pertinent and relevant study protocol.

Below are some specific section comments:

Title:

The title is a little confusing as health is all encompassing and can include many factors from physical to mental health. Physical activity itself also includes exercise so I think that the authors perhaps need to consider making the title a little more specific to the study aims and objectives. For example is exercise going to be assessed separately to physical activity from the steps data using perhaps HR data from the FitBits? In the abstract introduction quality and wellbeing have been used as secondary 'endpoints' - is this what the authors are referring to when they say health in the title. Perhaps the title should be more specific to reflect this? Also is the aim of the study not more focussed on investigating the ability of telehealth to impact on health behaviours in PWCF? There is no mention of telehealth or technology in the title - perhaps this should be considered?

Abstract:

The abstract is relatively clear however consistency with the title as suggested above is needed in terms of the aims of the study.

The manufacturer details are needed for the Fitbit and the Fitabase to ensure reproducibility. What does the online monitoring system actually do - there are limited details pertaining to this?

What type of data is collected and how often and therefore what type of information is available to the healthcare professional.

I think the authors need to be clear about what the text messages will include - is it only positive reinforcement plus changes to targets based on the weekly step counts etc (how will this be progressed? Is a standardised method going to be used for all participants? References perhaps needed for this. Or will it also include 'feedback'? What will the 'feedback' include?

Is there a need for a wash-in period (to allow for the novelty effect) at the beginning of the study and/or a washout period between the 12 week time period to allow for the positive or negative impact of familiarisation of using a fitness tracker?

A point of note: wearable technology is a more appropriate term rather than fitness tracker.

Background:

There is a lack of data to support the statements being made throughout this section e.g. 'higher levels of exercise' what do the authors mean by higher and using what outcome measures? More specific detail is required in terms of the evidence base being used to support the aims, objectives and methodology of this protocol under the 4 sub sections - CF, PA, telehealth and goal setting.

A clear definition of physical activity is needed. Exercise is a sub section of physical activity and it is not clear if an specific 'exercise' intervention is planned via the SMS 'feedback' which should be outlined.

In addition in the background reference to the need for an assessment of delivery and acceptability is made but this has not been mentioned in the abstract or introduction.

Methods:

What do the authors mean by efficacy? Is engagement with the technology enough for the study to be deemed efficacious? Will engagement with the technology be recorded?

Will the outcome measures be measured during routine outpatient appointments - or will additional appointments need to be made? What will the additional burden (physical and time) be for the participants? Has there been any PPI used in the planning of this study to take this into account?

Exclusion criteria - why have individuals with a FEV1 <25% been excluded?

The term PWCF who are pregnant rather than pregnancy should be used.

No mention of 'usual care' has been given and would be useful for the reader to understand with respect to physical activity advice and interventions that the participants normally receive.

Randomisation: the lung function categories are confusing - <70% mild - should this not be 70% and above? 30-50% moderate - what about those with 51-69%.....?

Intervention - more detail is needed to clearly explain and outline all of the details pertaining to the initial information - how will this be provided - verbal or written form? What will it include? Etc.

How will baseline physical activity be measured? Using the Fitbits provided or another accelerometer? How long will this be measured for? Specific details are needed here - number of hours and on which days - weekdays and/or weekends etc.

Goal setting intervention - what strategies and approach will be used for this? Will a specific standardised format be used for each participant? How many goals will be used? Will this be standardised?

As previously mentioned are wash-in and washout periods needed for data collection? Would a randomised crossover study design be more useful to account for this?

Outcome measures:

Primary: In the abstract PA is included as a primary endpoint but listed as a secondary outcome here.

Is CPET an appropriate primary outcome measure - an exercise intervention has not been included or described in this protocol. Is the aim for fitness to be impacted or PA only? This needs to be clarified.

Has the well validated supramaximal verification CPET protocol for PWCF been considered as an option?

More details are required regarding the 'ramp protocol' that has been referred to and a reference added. What outcomes will be used from the CPET - VO2 max only or will others be considered e.g. VE/VO2, RER as well as standard CPET outcomes in terms of duration, load etc. More details are needed.

Is FEV1 only going to be used as a classification measure? If this is the case then it is not a primary outcome measure but purely a method of categorising the participants rather than a measure of change or a measure of the impact of the technology.

Are other PFTs measures going to be considered?

Secondary

More detail regarding the step count data collection needed. How frequently will data be collected? How will this be analysed - how many timepoints? Will all 7 days be used or only those with a pre-specified number of hours data? Etc.

More detail is needed regarding the protocols for each of the outcome measures e.g. hand dynamometry - will it be a best of three measure, what unit of measurement will be used, will a standardised protocol be used - what does this include?

Will the participants complete the questionnaires independently or in the presence of a study team member?

Why is sleep being used as an outcome measure? What is the relevance or need for this? Its not clear. This perhaps comes back to my comment regarding a need for a clear definition from the outset for 'physical activity and health'.

There are a lot of outcome measures - again the burden to the participant needs to be considered? Has participant fatigue been considered? Are all measures necessary to answer the

research question?

Data analysis: limited statistical analysis plans have been provided. Further details are needed and some reference to intention to treat analysis, missing data and drop outs is required. In addition it would be useful to have an idea as to the possible options for data display.

Discussion:

The discussion is quite limited and a number of broad generalised statements have been made which require further detail and explanation to fully convince the reader of the rationale and need for this study.

A clear and evidence based discussion as to the validity and reliability of the chosen wearable technology is also needed.

A brief reference to the fact that gene therapy will be monitored during the study has been made - this is a very important point and needs to be considered carefully. A clear outline of what 'other' data will be monitored during the study is also needed - e.g. hospital admission, clinical appointments, colonisation status etc.

Is the rationale for, and objectives of, the study clearly described?

Partly

Is the study design appropriate for the research question?

Partly

Are sufficient details of the methods provided to allow replication by others?

No

Are the datasets clearly presented in a useable and accessible format?

Not applicable

Competing Interests: No competing interests were disclosed.

Reviewer Expertise: Cystic Fibrosis, Physical activity, exercise, health technology.

I confirm that I have read this submission and believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard, however I have significant reservations, as outlined above.

Author Response 30 Sep 2020

Maire Curran, University of Limerick, Limerick, Ireland

Dear Dr Sarah Rand,

Re: Steps Ahead: Optimising physical activity in adults with cystic fibrosis: Study Protocol for a pilot randomised trial

We sincerely thank you for your detailed and critical analysis of our article. Please find our responses below after your feedback (in bold). All changes have been made and are illustrated through italics and will be submitted in the second version of this article:

Thank you for asking me to review this interesting and very topical protocol. The authors have made a very good effort at identifying the need for undertaking this study (which is even more relevant given the current circumstances around the world) for PWCF. As the authors correctly say there is an ever-increasing research base investigating telehealth for individuals with long term conditions such as CF so this is a pertinent and relevant study protocol.

Thank you for your positive response. We believe this is very timely research, particularly considering current circumstances and the current landscape for PWCF. We have attempted to address your concerns below to enhance the quality and transparency of our research.

Below are some specific section comments:

Title:

The title is a little confusing as health is all encompassing and can include many factors from physical to mental health. Physical activity itself also includes exercise so I think that the authors perhaps need to consider making the title a little more specific to the study aims and objectives. For example is exercise going to be assessed separately to physical activity from the steps data using perhaps HR data from the FitBits? In the abstract introduction quality and wellbeing have been used as secondary 'endpoints' - is this what the authors are referring to when they say health in the title. Perhaps the title should be more specific to reflect this? Also is the aim of the study not more focussed on investigating the ability of telehealth to impact on health behaviours in PWCF? There is no mention of telehealth or technology in the title - perhaps this should be considered?

Thank you for highlighting this. We agree the title should be more specific and so this has been changed to *"Steps Ahead: optimising physical activity in Adults with Cystic Fibrosis: Study protocol for a pilot randomised trial using wearable technology"*.

Exercise is not being assessed separately to physical activity (step count data) in this present study. The Fitbit Charge 2 can measure 'activity minutes', through the use of heart rate (HR) data, however, this has not yet been evaluated as a valid and reliable measure for HR in people with CF. In fact, a recent study assessed HR data amongst 15 healthy participants and found that the Fitbit Charge 2 could underestimate HR by almost 30 beats per minute (Benedetto et al., 2018). Therefore, this pilot study is focusing on step count alone as a proxy measure for physical activity.

Thank you for this important observation. The use of the word health is broad and initially was used to refer to a number of variables described in the methods. As this is a pilot study, we are keen to investigate if this intervention will improve a range of clinical outcomes (including step count itself, lung function, exercise capacity, quality of life, wellbeing, and sleep). The word health in our study was used to broadly encompass these key components.

However, our primary aim is to improve PA and so we have removed the word health from the title. We have provided further description of the health behaviours in the introduction and methods.

Abstract:

The abstract is relatively clear however consistency with the title as suggested above is needed in terms of the aims of the study.

The manufacturer details are needed for the Fitbit and the Fitabase to ensure reproducibility.

The title has changed to reflect your comments above.

Manufacturer details have been added under the title primary outcome measure – Fitbit Charge 2, as follows:

“The Fitbit Charge 2 (Fitbit Inc.) will be used to record step count during this study with data uploaded to Fitabase (Small Steps Labs LLC).”

What does the online monitoring system actually do - there are limited details pertaining to this? What type of data is collected and how often and therefore what type of information is available to the healthcare professional.

Thank you for this comment which has been addressed in the script in the intervention “wearable technology section” as follows:

“Fitabase, the online monitoring system enables the physiotherapists to access step count data remotely. When a participant enables Bluetooth and syncs their Fitbit through the use of the Fitbit app, this automatically updates on the Fitabase website. This ensures that data is continuously collected over the full study duration. The data collected through the Fitbit Charge 2 and recorded on the Fitabase system are step count, activity minutes and sleep. However, for the purpose of this study, the researchers are only investigating step count alone. Any further research would require the Fitbit to be assessed under each of these conditions to ensure validity and reliability. This is outside the scope of this research.”

I think the authors need to be clear about what the text messages will include - is it only positive reinforcement plus changes to targets based on the weekly step counts etc (how will this be progressed? Is a standardised method going to be used for all participants? References perhaps needed for this. Or will it also include 'feedback'? What will the 'feedback' include?

Thank you for this feedback. The following text has been added to the intervention section to ensure clarity around text messaging.

“The text messages in this study will be personalised, one-way texts with positive reinforcement on step count attained by the participant.

A meta-analysis found personalisation strategies such as using a participant name has been proven to increase intervention efficacy (Head et al., 2013) and tailoring messages is a key ingredient in text messaging interventions (Noar et al., 2007). Furthermore, one way communication is as effective as two-way dialogue (Head et al., 2013). Therefore, this study

investigated one-way messages only.

An individualised message will be sent to each participant once a week, at a day and time chosen by the participant at the start of the intervention. The authors will follow a standardised approach. The message will address the participant by name and will have positive feedback in the form of encouragement and praise messages (Adams et al., 2013) in relation to their step count attained in the previous week. It will then focus on a physical activity related goal set by the subject and will sign off with a further positive comment. Research has indicated that it is crucial to reinforce improvements to develop new behaviour or to strengthen a habit (Hovell et al., 2009, Glanz et al., 2008, Bandura, 2004). Each time a participant meets his/her goal they will receive positive feedback. Participants who do not meet the goal will be provided with a positive comment in relation to their step count achieved to date and provided their next step goal. This is to avoid negative messages that could be discouraging (Adams et al., 2013)."

Is there a need for a wash-in period (to allow for the novelty effect) at the beginning of the study and/or a washout period between the 12 week time period to allow for the positive or negative impact of familiarisation of using a fitness tracker?

The authors acknowledge that no wash-in or wash-out period was applied and accept that there may be a novelty effect at the start of the study. This study included public and patient involvement (PPI) at the methodological development stage and both participants and clinicians felt that a 12-week intervention would be more readily accepted by this cohort as the study appointments could coincide with their routine clinic appointments. However, data will be regularly uploaded to Fitabase and therefore we can analyse and determine if there is a significant increase in steps in the first two weeks in comparison to subsequent weeks per participant and as a group.

A point of note: wearable technology is a more appropriate term rather than fitness tracker.

Thank you for this comment. We agree with this and the term wearable technology has been used throughout the article.

Background:

There is a lack of data to support the statements being made throughout this section e.g. 'higher levels of exercise' what do the authors mean by higher and using what outcome measures?

More specific detail is required in terms of the evidence base being used to support the aims, objectives and methodology of this protocol under the 4 sub sections - CF, PA, telehealth and goal setting. A clear definition of physical activity is needed.

Exercise is a sub section of physical activity and it is not clear if an specific 'exercise' intervention is planned via the SMS 'feedback' which should be outlined.

The authors thank the reviewer for this comprehensive feedback. A definition of PA has been added to the introduction along with further evidence to support PA in CF. There is no specific 'exercise' intervention planned via the feedback provided through the text messages. The study aims to look at step count behaviour only. The statements around 'higher levels of exercise' has been re-phrased. The following has been added/modified to the introduction:

“PA can be described as any bodily movement that causes an increase in energy expenditure above that of resting energy expenditure including leisure-time PA, occupational PA and exercise (Caspersen et al., 1985). Exercise and physical activity (PA) are widely documented in consensus statements as key components in the management of CF (Castellani et al., 2018). PA has several positive benefits in this population as it has been shown to improve sputum clearance (Dwyer et al., 2009), bone mineral density (Gupta et al., 2019) and muscle strength (Burtin et al., 2013). More importantly, PA can improve aerobic capacity (Hebestreit et al., 2010) and it can slow the rate of decline in lung function (Schneiderman et al., 2014), both of which are linked to increased survival in CF (Nixon et al., 1992). As a result, the optimisation of exercise and PA among PWCF is important, however, a Cochrane review found that there was a lack of evidence regarding strategies to promote PA in this population and consideration should be given towards telemedicine applications and health coaching (Cox et al., 2013b).

Numerous subjective and objective methods are reported in the literature for the assessment of PA in adults with CF. Step count measurement can be used to quantify and monitor PA behaviours (Tudor-Locke and Bassett, 2004). Despite the clear benefits of PA in CF, a recent systematic review found that adults with CF fail to meet recommended PA and step count guidelines (Shelley et al., 2019). Therefore, interventions which aim to increase PA in this population are of considerable interest.

In addition to telehealth, goal setting and reviewing goals regularly should be considered to support patients with chronic illness (Coleman & Newton, 2005). In order for goals to be achieved, feedback should be provided that reveals progress in relation to the goal (Locke & Latham, 2002). Furthermore, setting specific goals should offer a plan to break PA goals into more practical, manageable steps (Shilts et al., 2004) which should increase self-efficacy and hence promote continued regular PA levels (Bandura, 2004). Previous literature from patient preference research and effective PA promotion approaches reports that successful strategies include specifically targeting PA (Conn et al., 2008), the use of behavioural strategies such as feedback and goal setting (Conn et al., 2008, Kosma et al., 2005) (George et al., 2012) and the ability to self-monitor (Conn et al., 2008). The Irish national framework for self-management includes goal setting and action planning in chronic disease management (Chronic Conditions Working Group, 2017). PA interventions using pedometers have been shown to be more effective at increasing PA if they include step goals (Bravata et al., 2007). However, goal setting to increase PA in adults with CF is poorly investigated.”

In addition in the background reference to the need for an assessment of delivery and acceptability is made but this has not been mentioned in the abstract or introduction.

Thank you for highlighting this. The following has now been added to the introduction:

“This pilot randomised trial aims to explore the impact of wearable technology, text message personalised feedback and goal setting on PA and health outcomes in adults with CF. For the purpose of this study these specific health outcomes include lung function, aerobic capacity, body composition, quality of life, well-being and sleep. Poor sleep quality has been previously highlighted as an issue in adults with CF (Milross et al., 2002) and PA interventions can improve sleep quality in other populations (Yang et al., 2012, Lan et al., 2014).

In addition, it is important to conduct a qualitative analysis to evaluate the implementation, delivery and acceptability of the intervention as this is the key to the development of future research which this pilot study aims to inform. Therefore, semi structured interviews will be conducted with the study participants and with the healthcare professionals providing the intervention. This will aim to inform future studies in this evolving research area."

Methods:

What do the authors mean by efficacy? Is engagement with the technology enough for the study to be deemed efficacious? Will engagement with the technology be recorded?

The authors are referring to the ability of the tracker to change PA levels, FEV1 or exercise capacity (being the primary outcome measures).

Thank you for your feedback on this. The term efficacy is nebulous and so this has been changed to more concrete term reflecting what we aim to do in this study -

"This study represents a single centre pilot randomised trial which will compare the effect of wearable technology with personalised text message feedback and goal setting to wearable technology alone in adults CF."

Efficacy refers to the ability of the wearable technology, text message feedback and goal setting to increase PA levels (being the primary measure). The following has been added in relation to engagement with the technology:

"Engagement with the technology will be monitored through systematic review of online data. Participants in both groups will receive a "reminder" message on his or her mobile phone if their data has not been synced via the Fitbit App in the previous seven days. This ensures that data is continuously collected over the full study duration."

Will the outcome measures be measured during routine outpatient appointments - or will additional appointments need to be made? What will the additional burden (physical and time) be for the participants? Has there been any PPI used in the planning of this study to take this into account?

Thank you for highlighting this. We have added the following for clarification on appointments -

"Each adult with CF attends their routine clinic appointments every three months. Therefore, we chose this duration for our intervention to dovetail with routine CF care and limit the need for participants to attend on a more regular basis for testing appointments. This was decided through PPI (completed with adults with CF) and with clinician input. If the participant cannot attend on a particular clinic day, then they will be offered an additional appointment to suit their schedule."

The cardiopulmonary exercise test (CPET) is only completed at annual assessment typically, and so, this will require additional time on their routine clinic appointment. It is aimed to conduct this CPET either pre or post clinic review. The anticipated time to complete the CPET is between eight

to 12 minutes.

Exclusion criteria - why have individuals with a FEV₁<25% been excluded?

The following has been added to the exclusion section:

"Individuals with FEV₁ <25% would typically require supplemental oxygen for exercise and at this point may require transplant assessment. Furthermore, they are more likely to experience exacerbations and would not be suitable for a study over 24 weeks. This is a lower cutoff FEV₁ than most studies would include (Kriemler et al., 2013, Hebestreit et al., 2010, Klijn et al., 2004)"

The term PWCF who are pregnant rather than pregnancy should be used.

Thank you, this has been changed in the manuscript.

No mention of 'usual care' has been given and would be useful for the reader to understand with respect to physical activity advice and interventions that the participants normally receive.

Thank you for this suggestion which the authors agree would be helpful to include in the manuscript. We have added the following to the methods section of the article:

"Usual care would typically include PA and exercise advice as per Australian and New Zealand Cystic Fibrosis guidelines (Button et al., 2016). People with CF are advised to exercise for at least 30 minutes, five days a week. They are advised to include a combination of aerobic and resistance training. It is personalised depending on patient preference (Button et al., 2016)."

Randomisation: the lung function categories are confusing - <70% mild - should this not be 70% and above? 30-50% moderate - what about those with 51-69%.....?

Thank you for this comment. This was an oversight and has been adjusted as per below.

"Following this a minimisation randomisation procedure will be completed based on lung function, where FEV₁ of >80% predicted lung function will be classified as having mild lung disease. While those with an FEV₁ of 50-79% predicted lung function will be classified as having moderate lung disease, 30-49% as severe lung disease and <30% indicating severe lung disease (Ranu et al., 2011)"

Intervention - more detail is needed to clearly explain and outline all of the details pertaining to the initial information - how will this be provided - verbal or written form? What will it include? Etc.

We have endeavoured to provide more detail on the intervention in the manuscript as follows:

"Baseline activity levels will be obtained using an ActivPAL accelerometer as part of a parallel

study which assesses PA levels in this group. This will be used to inform goal setting only. The data obtained from the ActivPAL will be discussed with the participant at baseline. Data will be included if at least four full days (at least three weekdays and one weekend day) of measurements with a minimum of 10 hours (h) for the weekdays and 10 h for the weekends are measured (Freedson et al., 2005). Based on PPI it was reported by the participants that they did not want to wear an ActivPAL other than at baseline as it is a thigh worn device which is not 'fashionable'. Similar concerns have been cited previously in the research (Dias et al., 2012).

"The intervention consists of wearable technology, text message feedback and goal setting.

Wearable technology:

If the participant is allocated to the intervention, they will be provided with wearable technology (Fitbit Charge 2), educated on how to use it, and this will also be linked to an online monitoring system (Fitabase). Participants will be encouraged to enable Bluetooth and upload data regularly.

Goal setting:

The physiotherapist will discuss the participant's PA levels (as measured at baseline by an accelerometer) and individual patient centred PA goals will be set with each participant. These will be discussed with their physiotherapist to ensure they are specific, measurable, achievable, realistic and timed (SMART). They will be encouraged to write a minimum of three goals. Participants will be asked to set a step count target for week four, eight and 12 and consider ways to achieve these goals, both of which will be discussed with the physiotherapist during the baseline appointment. Goals will be individualised to the participant taking into account their preferences. Participants may also set goals related to health outcomes being assessed – for example, a participant may have a goal to improve lung function, aerobic capacity, sleep etc. during the study period. The text message feedback will refer to step goals only. The participant will be given a copy of their goals.

Text message feedback:

The text messages in this study will be personalised, one-way texts with positive reinforcement on step count attained by the participant. A meta-analysis found personalisation strategies such as using a participant name has been proven to increase intervention efficacy (Head et al., 2013) and tailoring messages is a key ingredient in text messaging interventions (Noar et al., 2007). Furthermore, one way communication is as effective as two-way dialogue (Head et al., 2013). Therefore, this study investigated one-way messages only.

An individualised message will be sent to each participant once a week, at a day and time chosen by the participant at the start of the intervention. The authors will follow a standardised approach. The message will address the participant by name and will have positive feedback in the form of encouragement and praise messages (Adams et al., 2013) in relation to their step count attained in the previous week. It will then focus on a physical activity related goal set by the subject and will sign off with a further positive comment. Research has indicated that it is crucial to reinforce improvements to develop new behaviour or to strengthen a habit (Hovell et al., 2009, Glanz et al., 2008, Bandura, 2004). Each time a participant meets his/her goal they will receive positive feedback. Participants who do not meet the goal will be provided with a positive comment in relation to their step count achieved to date and provided their next step goal. This is to avoid negative messages that could be discouraging (Adams et al., 2013). If the participant

does not achieve their target step count goal then the physiotherapist will set a target increase of 10% from the mean step count achieved in the previous week.

A sample text message is as follows:

Hi Paul, Well done on achieving an average daily step count of 7,600 steps this week. Next week aim to hit your goal of 8,000 steps. Keep up the good work!

How will baseline physical activity be measured? Using the Fitbits provided or another accelerometer? How long will this be measured for? Specific details are needed here - number of hours and on which days - weekdays and/or weekends etc.

As per above previous comment above, baseline PA levels will be measured by an ActivPAL accelerometer (PAL technologies). This will not be further used in the study as based on PPI adults with CF will not comply to wear this device on subsequent testing. This has been added to the methods section of the article.

Goal setting intervention - what strategies and approach will be used for this? Will a specific standardised format be used for each participant? How many goals will be used? Will this be standardised?

Please see previous paragraph on goal setting. This will be illustrated under the Intervention section of the article.

As previously mentioned are wash-in and washout periods needed for data collection? Would a randomised crossover study design be more useful to account for this?

As mentioned above we felt that a wash-in and wash out period would not be necessary in a small pilot study that is 12 weeks in duration with a further 12 week follow up. A crossover study would increase the need for a washout period and would prolong the length of the study. We had considered a crossover design but we felt this was too complex for a small pilot study and taking into account overall patient burden.

Outcome measures:

Primary: In the abstract PA is included as a primary endpoint but listed as a secondary outcome here

Thank you for highlighting this. This has been changed. It is a primary outcome measure.

Is CPET an appropriate primary outcome measure - an exercise intervention has not been included or described in this protocol. Is the aim for fitness to be impacted or PA only? This needs to be clarified.

Thank you for this feedback. CPET is a secondary outcome measure.

Has the well validated supramaximal verification CPET protocol for PWCF been considered as an option?

Thank you for this suggestion. The supramaximal verification protocol will be used. This has been added to the methods section of the study.

“Supramaximal verification be conducted to ensure a maximal effort during the CPET. After an incremental CPET, participants will be provided with a ten-minute break. They will then be asked to complete a supramaximal exercise test (Causer et al., 2018, Saynor et al., 2013) whereby participants will exercise at 110% of their power output (as determined by CPET test previously). This test has been conducted amongst PWCF previously and has been deemed safe and ensures validity of the results which are obtained in a CPET test.”

More details are required regarding the 'ramp protocol' that has been referred to and a reference added. What outcomes will be used from the CPET - VO₂ max only or will others be considered e.g. VE/VO₂, RER as well as standard CPET outcomes in terms of duration, load etc. More details are needed.

Thank you for this comment. The following text has been added to the methods section under secondary outcome measures and “CPET”.

“The reference standard exercise test is an incremental cardiopulmonary exercise test (CPET), utilising a ramp protocol. The ramp protocol ensures that work rate is progressively and linearly incremented until maximal effort is achieved (Herdy et al., 2016). All standard CPET outcomes will be considered including duration, load, pulmonary ventilation (VE), Respiratory Exchange Ratio (RER), ventilatory equivalents for oxygen (VE/VO₂) and for carbon dioxide (VE/VCO₂).”

Is FEV₁ only going to be used as a classification measure? If this is the case then it is not a primary outcome measure but purely a method of categorising the participants rather than a measure of change or a measure of the impact of the technology. Are other PFTs measures going to be considered?

Thank you for highlighting this. FEV₁ will be used as a classification measure but will also be used to determine the effect that this intervention may have on FEV₁ as a secondary outcome measure. Furthermore, we will also consider other PFT's such as FVC and FEF₂₅₋₇₅. The following has been added to the methods section:

“Forced expiratory volume in one second (FEV₁) will be used to classify the severity of CF lung disease for each participant. It will also be used to determine the effect the intervention may have on pulmonary function. Other pulmonary function measures such as % forced vital capacity (FVC) and forced expiratory flow (FEF₂₅₋₇₅) will also be considered.”

Secondary

More detail regarding the step count data collection needed. How frequently will data be collected? How will this be analysed - how many timepoints? Will all 7 days be used or only those with a pre-specified number of hours data? Etc.

The data will be continuously collected throughout the study time period. Every time the participant syncs their Fitbit, the data will automatically upload on to Fitabase.

The following has been added to the data analysis section:

“Data will be collected over seven days and an average weekly step count will be obtained. It is anticipated that step count data will be analysed to assess for a novelty effect in the first two weeks, at week six and 12 during the intervention and at week 18 and Week 24 for the follow up. A repeated measures ANOVA, controlling for baseline values and other potential confounders will be conducted.”

More detail is needed regarding the protocols for each of the outcome measures e.g. hand dynamometry - will it be a best of three measure, what unit of measurement will be used, will a standardised protocol be used - what does this include?

Thank you for this feedback. We have added more detail to the protocol in the methods section.

“The participant will be asked to stand and hold the dynamometer in by their side, with their elbow at 90 degrees and forearm in neutral. They will be instructed to squeeze the device as hard as possible. They will be provided with a 30 second rest period between each trial. The greater of two trials from each hand will be used and added together to give overall handgrip strength. This will be measured in kilograms (Martínez-García et al., 2020).

Will the participants complete the questionnaires independently or in the presence of a study team member?

Thank you for highlighting this. The following has been added to the Data Collection and Management section:

“All self-report measures will be completed by the participants independently, however a research assistant (RA) (AJ) will be available if there are any queries or concerns. These subjective assessments will be analysed by the RA who is blinded to group allocation.”

Why is sleep being used as an outcome measure? What is the relevance or need for this? It's not clear. This perhaps comes back to my comment regarding a need for a clear definition from the outset for 'physical activity and health'.

Thank you for this suggestion. We have added the following to clarify these specific health outcomes in the introduction section:

“This pilot randomised trial aims to explore the impact of wearable technology, text message personalised feedback and goal setting on PA and health outcomes in adults with CF. For the purpose of this study these specific health behaviours include lung function, aerobic capacity, quality of life, well-being and sleep. Poor sleep quality has been previously highlighted as an issue in adults with CF (Milross et al., 2002) and PA interventions can improve sleep quality in other populations (Yang et al., 2012, Lan et al., 2014).”

There are a lot of outcome measures - again the burden to the participant needs to be considered? Has participant fatigue been considered? Are all measures necessary to

answer the research question?

Participant fatigue has been considered. The authors have used a PPI approach in the study design and set up and based on feedback participants were satisfied with timing and breaks between each outcome assessment. Furthermore, the adults with CF attending this CF centre would be familiar with most of these outcome measures which would be conducted as part of their annual assessment. As this is a pilot study the authors incorporated a range of outcome measures to answer the key research questions and to inform future research.

Data analysis: limited statistical analysis plans have been provided. Further details are needed and some reference to intention to treat analysis, missing data and drop outs is required. In addition it would be useful to have an idea as to the possible options for data display.

The following has been added to the data analysis section:

“Primary analyses will be performed according to intention-to-treat (ITT) principles with all participants who were originally allocated by randomisation and those who dropped out from the study. In case of missing values, a multiple imputation method, inverse probability weighing or mixed models will be used to handle the missing data. Data will be presented as means and SDs for data that is normalised and medians (IQR) for data that is skewed.”

Discussion:

The discussion is quite limited and a number of broad generalised statements have been made which require further detail and explanation to fully convince the reader of the rationale and need for this study.

The discussion has been revised with further detail added on the rationale and need for the study.

A clear and evidence based discussion as to the validity and reliability of the chosen wearable technology is also needed.

Thank you for highlighting this. This research has been conducted and has been submitted for publication. This research concluded that Fitbit Charge 2 ranged from weak to very strong correlations when compared to visual observation (0.34-0.84). The Fitbit Charge 2 underestimated step count by 2.8%-9.2%. This is within acceptable limits of variability based on previous research (Schneider et al., 2004). The following has been added to the methods:

“Prior to this intervention, the validity and reliability of the Fitbit Charge 2 to measure step count in CF was assessed by the same research group (Curran et al., 2020). Twenty-one participants were recruited from an adult CF Centre for a single session of testing. Participants walked for five minutes at five pre-determined speeds in a controlled testing environment (2, 2.5, 3, 3.5 and 4 miles per hour on a treadmill) and at three self-selected speeds on a corridor (slow, medium and fast). The Fitbit Charge 2 was compared to visual observation. It was found that the Fitbit Charge 2 ranged from weak to very strong correlations when compared to visual observation (0.34-0.84). The Fitbit Charge 2 underestimated step count by 2.8%-9.2%. This is within acceptable limits of variability based on previous research (Schneider et al., 2004).”

A brief reference to the fact that gene therapy will be monitored during the study has been made - this is a very important point and needs to be considered carefully. A clear outline of what 'other' data will be monitored during the study is also needed - e.g. hospital admission, clinical appointments, colonisation status etc.

Thank you for this comment.

"The research team will also monitor exacerbation rates (participants requiring oral or IV antibiotics during the study period), hospital admission rate and colonisation status."

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