Social Experiences of Adolescents with Cystic Fibrosis: An Interpretative Phenomenological Analysis

by

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OVERVIEW

This thesis is submitted by Jessica Rose Seddon in partial fulfilment for the degree of Doctor of Clinical Psychology (ClinPsyD) at The University of Birmingham and comprises of two volumes. Volume One of the thesis is the research component and is made up of three chapters. Chapter One is a literature review and meta-analysis on the effectiveness of remotely-delivered psychological interventions on health-related quality of life (HR-QoL) in adults with physical health conditions. Chapter Two is a qualitative empirical research paper on the lived social experiences of adolescents with Cystic Fibrosis using Interpretative Phenomenological Analysis (IPA). Chapter Three comprises press releases for both the literature review and empirical research paper.

Volume Two of the thesis comprises five Clinical Practice Reports (CPRs) that were completed on clinical placement as part of the doctorate whilst working in different settings across the lifespan. CPR 1 describes the assessment, formulation and intervention for a male with an intellectual disability who was struggling with anger and bereavement using a combination of Cognitive-Behavioural and Psychodynamic models. CPR 2 is a service evaluation on the Transition Pathway within a community Learning Disability Service. CPR 3 is a case-study outlining the assessment, formulation and intervention for an 80-year-old woman experiencing severe generalised anxiety. CPR 4 is a Single Case Experimental Design (SCED) into the effectiveness of an Acceptance and Commitment Therapy intervention for a 25-year-old woman. CPR 5 comprises the abstract of an oral presentation of a comprehensive neuropsychology assessment conducted with a 14-year-old girl who had experienced a Traumatic Brain Injury (TBI).

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VOLUME ONE

CHAPTER ONE: LITERATURE REVIEW

The Effectiveness of Remotely Delivered Psychological Interventions on Health-Related Quality of Life (HR-QoL) in Adults with Physical Health Conditions: A Meta-Analysis

1.0 Abstract

Background: Remotely delivered psychological interventions have continued to grow in popularity and for those with physical health conditions, they provide alternative means of receiving support when physical symptoms such as pain and impaired movement may affect daily functioning. There is limited evidence for the impact of such interventions on quality of life outcomes for adults with physical health conditions.

Method: A systematic search of the literature yielded 24 studies (N=1701), after duplicates had been removed and articles were screened by title and then abstract to ensure they were focussed upon adults with a physical health condition, were employing a psychological intervention, were delivered remotely and were measuring health-related quality of life (HR-QoL) as an outcome. A quality index was devised to consider the impact of risk of methodological bias and study design.

Results: A small positive overall treatment effect was found for remotely delivered psychological interventions on HR-QoL in adults with physical health conditions when compared to control groups (SMD= 0.3288 95% CI 0.23-0.43). Sub-group analysis found significant differences in pooled treatment effect when studies were separated according to treatment delivery method (X^2= 10.93, p = 0.01). Other differences were noted according to therapeutic model employed and type of HR-QoL measure used although these differences did not reach statistical significance.

Discussion and Conclusion: Overall, this meta-analysis indicates small treatment effects for remote psychological interventions for HR-QoL in adults with physical health conditions. These findings support previous meta-analytic research demonstrating effectiveness for remotely delivered CBT for mental health conditions and face-to-face therapy for physical

health conditions. Methodological bias varies across the literature and further high quality RCTs are recommended.

1.1 Introduction

1.1.1 Psychological Interventions for Mental Health Conditions

Face-to-face psychotherapy has shown positive outcomes for a wide range of conditions, including depression and anxiety (Wampold & Imel, 2015). Partly due to the ease of manualisation and number of randomised controlled trials published, robust treatment effects have been found particularly for the effectiveness of cognitive-behavioural therapy including for anxiety and related disorders (Carpenter et al., 2018) and depression (Zhang et al., 2019). The evidence base has since developed for so called 'third-wave' treatment models that incorporate elements of CBT with additional interpersonal, evolutionary and behavioural components, such acceptance and commitment therapy (ACT) and compassion-focussed therapy (Hayes & Hoffman, 2017; Wilson et al., 2018). Compassion-focussed interventions have shown a moderate treatment effect on both anxiety (g= 0.46) and depression symptoms (g= 0.40), although findings are not superior to active control conditions, such as CBT (Wilson et al., 2018).

1.1.2 The Connection Between Mental and Physical Health

The biopsychosocial model was developed to challenge the historically dominant medical model of health. The model emphasises the importance of considering the links between an individual's physical and mental health with particular focus on the interaction between social, psychological and biological factors (Engel, 1981; Karunamuni et al., 2021). Research has since indicated that specific biological factors such as cortisol levels and inflammation are linked to both physical and mental health conditions (Bennett et al., 2018; Gatchel, 2004). There has also been increased recognition of the importance of such links when providing care and treatment for both mental and physical health conditions. The prevalence of co-morbid conditions has also been highlighted in more recent research and policy developments. A King's Fund report published in 2012, over 15 million people in England were deemed to be living with a long-term health condition. Of these people, it was estimated that over 4 million had a comorbid diagnosable mental health condition (Naylor et al., 2012). The recognition of a need to bridge the gap between mental and physical health was highlighted in further King's Fund report published in 2016 (Naylor et al., 2016). This report called for an increase in integration of physical and mental health care provision due, in part, to the recognition that physical and mental health are intrinsically connected. The report emphasised the reciprocal relationship between mental and physical health, which can impact on overall quality of life (Naylor et al., 2016).

Specific research has also shown correlational links that may affect the trajectory of physical health and mental health conditions. As an example, diabetes has been associated with higher rates of depression when compared with the general population and this relationship is thought to be bi-directional (Holt et al., 2014). It is also thought to affect treatment adherence and long-term physical health outcomes (Bauer et al., 2017). As a result of such findings, The NHS Long Term Plan, published in 2019, called for integrated care teams with the aim of improving the service provision and care for those with comorbid physical and mental health conditions (National Health Service, 2019). The need for digitally-enabled outpatient care was also emphasised within the report which has since seen an exponential rise in use, partly due to the Covid-19 pandemic (Peet et al., 2020).

Qualitative reviews of treatment burden have highlighted several factors that some individuals with physical health conditions experience including loss of independence, feelings of isolation, stigma, medication side effects and resultant negative emotions (Demain et al., 2015). Despite individuals often not receiving a formal mental health diagnosis, physical health conditions have long been associated with heightened risk of low mood, anxiety and reduced quality of life (Demain et al., 2015).

1.1.3 Psychological Interventions for Physical Health Conditions

Alongside growing evidence for the effectiveness for talking therapies in the treatment of mental health conditions, there has been an increasing focus placed upon the possible role of such interventions to improve the lives of those living with physical health conditions. Due to the popularity and widespread use of CBT in the treatment of several mental health conditions, it is perhaps not surprising that the largest portion of the literature base focusses upon the delivery of CBT within both randomised-controlled trials (RCTs) and meta-analyses (e.g. Hesser et al., 2011; Hind et al., 2014; Kalter et al., 2018; Tang et al., 2020; Ye et al., 2018). These above referenced studies have focussed upon a range of physical health conditions including cancer, tinnitus and multiple sclerosis and have found small to medium effect sizes (d=0.2-0.7) as outlined by Cohen (Cohen, 1988).

A meta-analysis of 18 RCTs indicated that cognitive-behavioural stress management may be effective in reducing symptoms of depression and increasing positive affect in breast cancer patients (Tang et al., 2020). However, this review was limited due to the low overall methodological quality of some of the included studies and completer-only analysis that did not consider attrition.

A further systematic review concluded that face-to-face Acceptance and Commitment Therapy (ACT) may be effective in improving psychological flexibility and quality life in people with long-term physical health conditions (Graham et al., 2016). However yet again, the methodological quality of the included studies was generally poor, with a third being clinical case studies and the authors calling for an increase in high quality RCTs (Graham et al., 2016). A further meta-analysis on acceptance-based interventions for chronic pain found small treatment effects of 0.37 on pain outcomes specifically. However, some of the included studies included only one element of ACT, such as mindfulness, and therefore it is difficult to make conclusions about the effectiveness of ACT for chronic pain from this research alone (Veehof et al., 2011). Since then, meta-analytic research has continued to indicate the effectiveness of ACT for physical health conditions and it has recently been recommended in NICE guidelines (A-Tjak et al., 2015).

Outcomes used in such research vary from being mental health focussed, for example reductions in measures for depression and anxiety for those with comorbid mental health diagnoses (e.g. Kellett et al., 2016), to focus on reductions in physical indicators such as a reduction in specific physical symptoms. A way to bridge this gap has been to develop specific health-related quality of life measures (HR-QoL) that aim to cover specific factors related to an individual's life quality in the context of their physical health condition.

1.1.4 Health-Related Quality of Life Measures

Health-Related Quality of Life (HR-QoL) measures are multi-dimensional outcomes often employed within studies examining the effectiveness of psychological interventions on physical health conditions. HR-QoL measures have been recognised as an effective tool to examine the impact of interventions on a wide range of factors including emotional functioning, physical functioning, mental and physical health (Calvert & Freementle, 2003). Researchers have commented on the importance of such measures being responsive and valid, to ensure that changes can be detected and that the outcome measures what it is designed to measure (Guyatt et al., 2007). As well as general HR-QoL measures such as the SF-36 (Ware & Sherbourne, 1992), tools have also been designed for specific physical health conditions such as the EORTC QLQ-C30 for cancer (Fayers & Bottomley, 2003). These measures include items specific to particular medical conditions and therefore may detect changes not captured within more general HR-QoL measures.

A benefit of measuring HR-QoL is that it focusses upon an individual's overall functioning and wellbeing in the context of their physical health (Sitlinger, & Zafar., 2018). Meta-analyses that have included HR-QoL measures have found small to moderate effect sizes for predominantly in-person psychological interventions including CBT for adults with cancer (e.g. Kalter et al., 2018; Osborn et al., 2006). Caution needs to be taken, however, when interpreting such results as sometimes they are only based upon a small number of studies. In addition, generalisability is sometimes limited by focusing on only one type of cancer (e.g., Osborn et al., 2006). For these reasons, this review will focus specifically on health-related quality of life (HR-QoL) measures.

1.1.5 Remotely Delivered Psychological Interventions

In addition to the robust findings regarding the effectiveness of face-to-face talking therapy (e.g. Wampold & Imel., 2015), the evidence base is building for remotely-delivered psychotherapy for mental health conditions such as CBT for anxiety and depression (Andrews et al., 2018; Cuijpers et al., 2010; Olthuis et al., 2016). These reviews have shown superior outcomes when compared to waitlist controls or Treatment as Usual (TAU) control conditions, with moderate effect sizes. Some research has also indicated that internet delivered CBT may be as effective as face-to-face CBT (Andrews et al., 2018). However, the studies included in some reviews have been rated as low to moderate in quality, with some rated as high risk of bias due to unclear treatment fidelity and a lack of clarity regarding therapist role (e.g., Olthuis et al., 2016).

Nevertheless, remotely delivered psychotherapy arguably provides increased flexibility to services and patients whilst being cost-effective (Hedman et al., 2012). In addition, qualitative research with NHS managers and commissioners indicated favourable opinions regarding the addition of remote interventions to mental health services due to Covid-19 (Simon et al., 2021).

1.1.6 Remotely Delivered Psychological Interventions for Physical Health Conditions

Adults with physical health conditions may benefit from having the flexibility to receive remotely delivered therapy, due to the impact of symptoms, treatment burden or possible associated fatigue and pain (Demain et al., 2015). Qualitative research has indicated that long-term physical health conditions are often associated with depression or anxiety and that the impact of certain conditions can sometimes worsen the experience through social isolation or being house-bound (DeJean et al., 2013). Access to psychological support for those with physical health conditions may also be limited if their conditions affect their ability to attend regular face to face appointments in a clinical setting. Significant disease-related distress is also associated with poor disease management and people with long-term conditions are two to three times more likely to experience mental health problems than the general population (King's Fund and Centre for Mental Health, 2012).

Several benefits have been highlighted when considering remotely delivered psychological interventions including saving therapist time, giving patients more flexibility and options for those who may feel uncomfortable about going to see a therapist in person (Cuijpers et al., 2007). A recent meta-analysis found small to moderate effect sizes (g= 0.19-0.36) on anxiety, depression and distress measures respectively for remotely delivered interventions in those with a physical health condition (White et al., 2020). Additional meta-analytic research examining the effectiveness of remotely delivered interventions have included HR-QoL outcome measures. However, previous meta-analyses sometimes focus on one specific type of health condition such as cancer and therefore findings may not generalise to other conditions (Qan'ir and Song, 2019). More recently, meta-analytic research has indicated that internet-delivered CBT (CBT-I) may be similarly effective to face-to-face cognitive behavioural therapy (Carlbring et al., 2018). Such reviews that consider active control as opposed to TAU are strengthening the evidence base for remotely delivered psychological interventions. However, the authors note that the majority of studies do not include active controls.

1.1.7 Summary of Previous Research

As outlined, the evidence base is building for remotely delivered psychological interventions. The Covid-19 pandemic has further accelerated the need and provision of remotely delivered psychological interventions, with several elements likely to remain long after the pandemic is under control (Wind et al., 2020). The additional vulnerability to Covid-19 in those individuals with health conditions such as cancer and diabetes has further highlighted the call for remotely delivered psychotherapy (Gamble et al., 2020; Raymond et al., 2020).

Previous meta-analyses and systematic reviews have focussed upon quality of life outcomes for psychological interventions in physical health conditions. However, the majority of these have focussed upon interventions delivered, at least in part, face-to-face. Previous meta-analyses have explored psychological interventions for health-related quality of life (HR-QoL) for individuals with physical health conditions and comorbid depression. For example, in cardiac patients, a small to moderate effect size was found in mental health related quality of life for those who undertook a face-to-face CBT intervention in comparison to Treatment as Usual (TAU) controls (O'Neill et al., 2011). A further study examining physical health and comorbid depression found small effects (SMD = 0.31) on quality of life measures (Jeyanantham et al., 2017). The heterogeneity of the literature, with some studies examining the effectiveness of psychotherapy on mental health and other studies examining those with comorbid physical health conditions also need to be considered when examining the wider evidence base.

1.1.8 Limitations of Previous Reviews

It is difficult to determine how widely results of previous reviews can be generalised. For example, 9 of the 22 RCTs included in meta-analysis by Kalter and colleagues utilised a 'coping skills training' intervention, as opposed to formal psychological therapy such as CBT (Kalter et al., 2018). Other previous reviews were limited as they included studies with less rigorous designs and small sample sizes, increasing the chance of methodological bias affecting the results (Jeyanantham et al., 2017). Authors also note that QoL measures themselves may be affected by social desirability bias due to their self-report nature and therefore this needs to be considered when drawing conclusions on quality of studies within this review.

The evidence is building for remotely delivered psychological interventions including possible evidence for equally efficacious results when comparing face-to-face with internet delivered CBT (Carlbring et al., 2018). Up until this point, there has not been a meta-analytic review that focusses specifically on QoL measures in adults with physical health conditions. Both general and disease-specific HR-QoL outcomes can indicate an individual's functioning and wellbeing in the context of their condition (Guyatt et al., 2007).

1.1.9 Aims of the Meta-Analysis

The evidence base has been building for the possible use of psychological therapies in individuals with physical health conditions. This is a promising area of literature that may improve patient quality of life whilst considering the ever-changing landscape of digital healthcare provision. As opposed to focussing upon outcomes directly linked to mental health, such as depression or anxiety symptoms, focusing upon health-related quality of life may give a better indicator of perceived burden and impact of the physical condition of an individual's perceived wellbeing (Guyatt et al., 2007). This meta-analysis therefore aims to fill this gap in the literature, by examining the effectiveness of remotely delivered psychological interventions on quality of life for adults with physical health conditions.

1.2 Method

1.2.1 Search Strategy

A systematic search of the literature was conducted up until 24th July 2020 using the following databases: Embase, Medline, Cochrane, Web of Science and PsychINFO. The search terms utilised can be seen in *Table 1*. The search aimed to give a systematic overview of the literature examining the effectiveness of remotely delivered psychological interventions on quality of life in adults with physical health conditions. Additional studies were identified through examination of the references of included studies, such as a meta-analysis of interventions for gastrointestinal disorders (Hanlon et al., 2018). Previous systematic reviews and meta-analyses were reviewed to capture search terms used within the literature for a broad range of physical health conditions (e.g. Forbes et al., 2017). With regards to therapy type, emphasis was placed upon therapeutic modalities that had been adapted for remote delivery. Further meta-analytic reviews were consulted when deciding upon the search terms outlined below (e.g. Norwood et al., 2018). The inclusion of the term psycho* was also included to capture other types of psychotherapy including psychodynamic psychotherapy.

Table 1 - Search Strategy

Construct	Search Terms	Method of Search	Limits
Physical health condition	'health condition' 'physical health' 'long-term health' 'chronic disease' 'cystic fibrosis' 'diabetes' 'cancer' 'heart disease' 'COPD' 'cardiac' 'coronary' 'asthma' 'arthritis' 'epilepsy' 'bowel disease' 'multiple sclerosis' 'Parkinson's'	Free search terms All search terms combined with *OR*	Peer-reviewed journal articles and dissertations published 1974-July 2020

AND	'fibromyalgia'
Psychological intervention **AND**	'psycho*' 'cognitive behav*' 'CB 'third-wave' 'mindful* 'acceptance' 'commitment therapy' 'ACT' 'CFT' 'compassion focussed therapy' 'behavioural therapy'
Remotely delivered	'internet' 'tele*' 'video*' 'computer* 'web*' 'distance' 'remote* 'online'

1.2.3 Inclusion/Exclusion Criteria

It was necessary for the inclusion criteria to be kept quite broad to capture the scope of remote psychological interventions and physical health conditions. Despite the focus being placed upon remotely delivered psychological interventions, the nature of these varied; some were delivered by therapists, others were self-directed, some were delivered online and some by telephone. As QoL was often measured as a secondary outcome, it was not appropriate to limit titles or abstracts to include the term 'quality of life' and therefore this phrase was not included in the search strategy. Additional screening of abstracts and full papers was required for a portion of the literature as outlined in the PRISMA flow diagram in *Figure 1*.

It was important for the purposes of the review that specific psychological interventions were included such as CBT and ACT. Papers were examined to determine if interventions followed a specific treatment protocol and a breakdown of each module or sessions was given (e.g. 12

sessions of CBT with a brief outline given for each). Treatment fidelity was also included as a type of methodological bias in the quality effects ratings to determine how well interventions were described and whether protocols were outlined in detail for the purposes of replication. Interventions that did not have elements of formal psychological theory, such as exercise, telehealth monitoring or physical health interventions were excluded. Those that included psycho-education without formal elements of therapy such as CBT were identified as such in the sub-group analysis. Full inclusion and exclusion criteria with justification are described in

Table 2 - Inclusion/Exclusion criteria and justification

Inclusion criteria	**Justification**
Participants are adults with a physical health condition	Remotely delivered psychological interventions individuals with physical health conditions.
Remotely delivered Psychological intervention	The meta-analysis is focussed upon remotely delivered psychological interventions as opposed to those delivered face-to-face.
Primary or secondary Health-Related Quality of Life (HR-QoL) outcome measure included.	Meta-analysis is focussed upon effectiveness on an individual's HR-QoL.
Exclusion Criteria	**Justification**
Meta-analyses or systematic reviews, qualitative research, feasibility/acceptability, theoretical/prevalence studies/psychometric studies.	These articles do not provide HR-QOL outcome data required for the meta-analysis.
Insufficient data reported to calculate effect size for QoL outcome	Cannot be included in meta-analysis if insufficient data is reported.

The results of the systematic search are presented in the PRISMA flow diagram in *Figure 1*. The search yielded 1701 results and 595 articles remained once duplicates had been removed. Titles were then screened using the inclusion and exclusion criteria; abstracts were reviewed when the content of the article was not clear from the title.

The most common reason for an article to be excluded was for being an acceptability, feasibility, conference or pilot trial with insufficient data to analyse (n=173). Several articles were excluded for describing purely medical interventions or telemedicine (n=122). Further articles were excluded for covering theoretical concepts, prevalence studies or psychometric properties of outcome measures (n=65). Studies were also excluded if the sample of participants were children, caregivers or partners of those with a physical health condition (n=65). Meta-analyses and systematic reviews were also excluded (n=27) but their reference sections were later reviewed to find additional studies that were not identified in the electronic search. Articles examining the effectiveness of online support groups (n=20), qualitative or narrative analysis (n=20) were also excluded. Further references were excluded as participants had a mental health condition but no known physical health condition (n= 13).

Of the remaining 90 articles, abstracts were screened to determine whether HR-QoL was included as an outcome measure. As this was not often included in the article title, abstracts were screened of the 90 remaining articles. If it was still remained unclear from reading the abstracts, full article reviews were conducted. A further 65 studies were excluded as they did not include a HR-QoL measure. Five studies were excluded as they did not report sufficient data to calculate standardised mean difference or effect sizes.

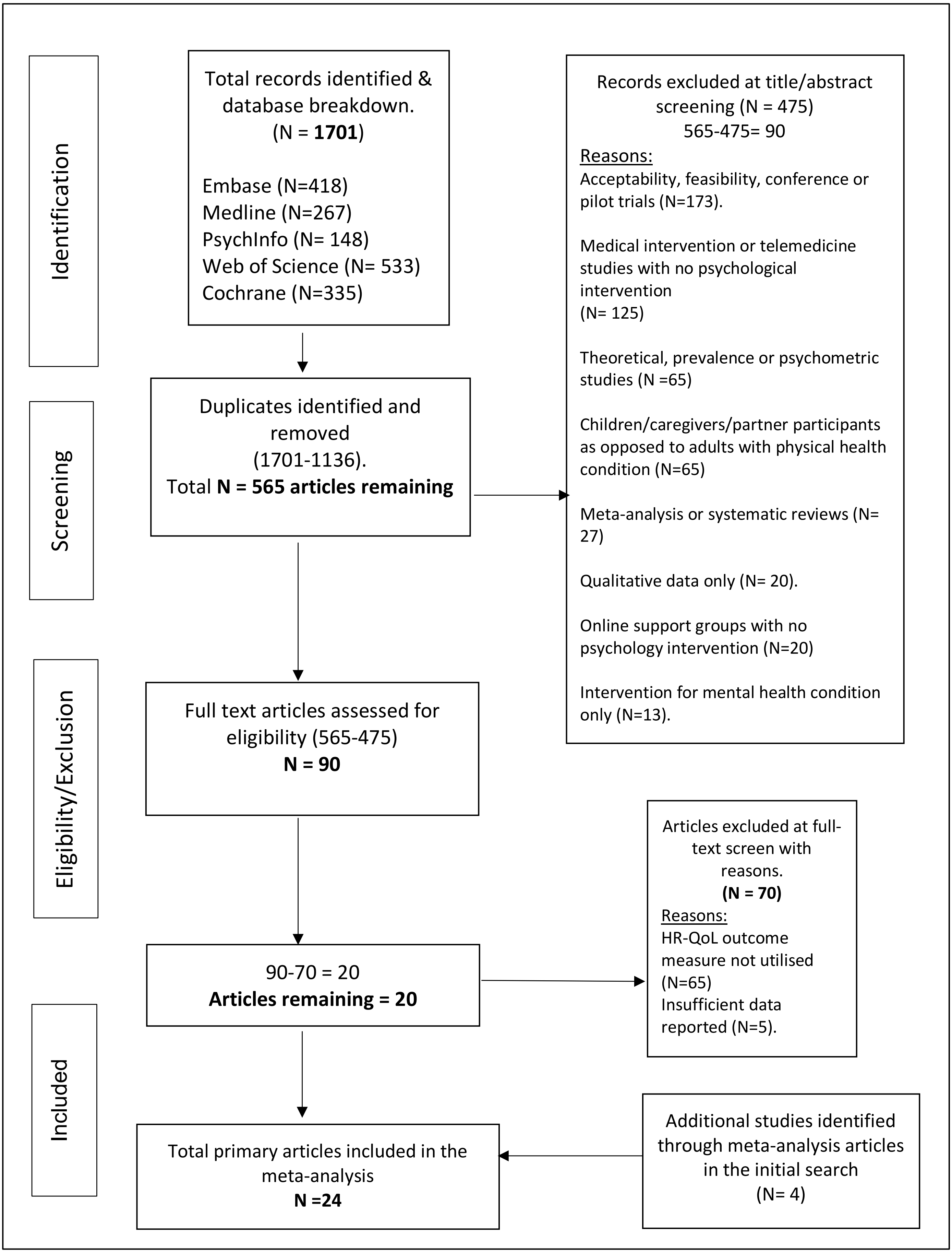

Figure 1 - A PRISMA flow diagram for the meta-analysis (Adapted from Moher et al., 2010).

A total of 20 papers met the full inclusion criteria. An additional 4 articles were identified through references of meta-analyses and systematic reviews from the original search (e.g. Hanlon et al., 2018). Of the 24 included studies, six articles included separate mental and physical HR-QoL subscale scores and therefore were included in the analysis twice. One article examined mental and physical quality of life as well as a disorder specific HR-QoL measure and was therefore included in the analysis three times (Newby et al., 2017).

1.2.4 Quality Criteria

It was important to assess risk of bias in each of the included articles due to the possible impact on estimated treatment effect. The quality criteria utilised within this meta-analysis were adapted from 'The Cochrane Collaboration Risk of Bias Tool' for assessing risk of bias in randomised-controlled trials (Higgins et al., 2011). Seven types of methodological bias were assessed, with descriptions for 'low', 'unclear' and 'high' outlined in *Table 3*. The tool was adapted to include elements specifically relevant to the research question such as treatment fidelity, reporting bias and generalisability. In addition, "Attrition bias" named within the original tool was incorporated into the sections of detection bias and statistical bias.

Each study was given a rating of 'low', 'unclear' or 'high' risk of bias for each of the seven types of methodological bias. *Table 3* below outlines the seven types of methodological quality as well as descriptions for a low', 'unclear' or 'high' rating. See *Figure 1* for study design, risk of bias and quality index ratings for each of the included studies. The quality index rating is then used to weight studies within the analysis according to design and methodological quality to ensure that those of lower risk of bias are given more weight (Doi et al., 2015).

Table 3 - Methodological bias descriptions and criteria for ratings

Type of Methodological Bias	Description	Rating		
		Low	**Unclear**	**High**
Selection Bias	Considers extent to which there are differences between those selected to receive treatment and those who are allocated to a control condition.	The characteristics of the study population are clearly described. Non-response rate is reported and of an acceptable level (max 50%). Source of sample described and the study reports participant demographics e.g., gender, specific physical health condition etc. The recruitment method is clearly reported and well defined.	Non-response rate is not reported. The characteristics of the study population are not clearly reported. For example, the country, setting, location, population demographics were not adequately reported. The recruitment process/ sampling method of individuals are unclear or has not been reported. Convenience sampling used.	Includes an unacceptable (reporting less than 30% of the data) level of non-response rate. Target sampling was used. The characteristics of the study population are not reported.
Performance Bias	Refers to possible systematic differences in participant motivation between the treatment and control groups, e.g. through different incentives.	Study reports level of confidentiality and anonymity. Participants were not rewarded for their participation in the study.	The study does not report levels of confidentiality and anonymity. It is not clear if participants were rewarded for their participation (e.g. motivation to respond in a certain way).	Participants were rewarded for their participation in the study. Participants were told which condition/ what questionnaires they were completing and why and any proposed hypotheses.
		Information and procedures are provided in a way that does not differentially motivated participants	Unclear whether participants were blinded to allocated condition.	
Treatment Fidelity	The extent to which treatment interventions are adequately described, replicable and adhered to consistently.	Well defined, replicable treatment intervention. Treatment includes elements specifically targeting Quality of Life. Information including protocol, number of sessions/outlines. Clear role of therapist (e.g. how much time/input each participant has). Fidelity measures in place (e.g. blindly rated by someone external). Those delivering treatment trained adequately	Unclear treatment protocol, not included details re session structure. Not stated whether any fidelity tests were done. Unclear description of treatment. Unclear whether treatment aims to improve Quality of Life despite this being an outcome.	Multi-component treatment, not controlled, not described. Treatment clearly not aiming to target Quality of Life
Detection Bias	Assesses the extent to which the design of the study allows for control and detection of the affect in question (Quality of life outcomes).	The outcome measures are clearly defined, valid and reliable, and are implemented consistently across all participants. Cronbach's alpha above 0.7. Outcome measures are blindly rated.	Information regarding the outcome measures are either not reported or not clearly reported e.g. definition, validity, reliability. Cronbach's Alpha for outcome measures is between 0.6 and 0.7. Test retest reliability for outcome measures is between .6 and .7 It is not clear if the measure was	The outcome measures were implemented differently across participants. The outcome measures used had poor reliability and validity reported e.g. Cronbach's Alpha < 0.6. and/or test/retest reliability < 0.6 Only using one dimension/ subscale of the QoL measure, omitting total or subscales scores.

			implemented consistently across all participants.	
Statistical Bias	The extent to which the appropriate statistical analysis is undertaken on the data. Includes consideration of impact of attrition through completer vs. intent-to-treat analysis.	Appropriate statistical testing was used. Intention-to-treat calculations included to account for attrition The study has reported a SMD (e.g. Cohen's) or an effect size can be calculated from raw data provided. Attrition rate – data loss is reported at analysis at an acceptable level (50%)	Unclear what statistical test was used. Confidence intervals or exact p-values for effect estimates were not reported and could not be calculated. Attrition – Completer-only analysis reported.	Statistics were not reported. Inappropriate statistical test was used. Attrition rate – data loss is reported at analysis at an unacceptable level (30%). Attrition rates not reported.
Reporting Bias	The extent to which the results section reports all analysis, statistical data for all outcomes outlined in the method section. Specific focus on those related to QoL or HR-QoL.	Reported all results of all measures as outlined in the method, regardless of significance. Specifically, those related to QoL and sub-scales if relevant. If Cohen's d or Hedges g not reported, enough alternative raw data provided so that SMD/effect size can be calculated.	Not all descriptive and/or summary statistics are presented. There is a description (narrative) in the results but do not record statistics. Reported more than one SMD.	Not reported full outcome measures that are stated in the method section/ reported only a subsample of results/only reported SMD that reached significance.
Generalisability	The extent to which research findings can be applied to settings other than that in which they were originally tested. This includes any differences between the study participants and those persons to whom the review is applicable	Sufficient sample for generalisation and representative of target population. A sample size justification, estimate and power analysis was provided. The sample size is adequate to detect an effect in line with the power calculation.	Sufficient sample for generalisation but with some idiosyncratic features. A sample size justification, estimate and power analysis were not provided. Demographics of participants do not appear to be representative of clinical population the study aims to generalise to.	Small sample with or without idiosyncratic feature. High percentage (over 80%) of sample is represented by one demographic and cannot be generalised to other demographics (e.g. A specific type of cancer only, only one gender unless for something with much higher prevalence e.g. breast cancer in females). The sample size is not adequate to detect an effect.

1.2.5 Quality Index Ratings

All included studies were RCTs with the exception of two that utilised pre-post designs without a control group (Luyster et al., 2020; McCarthy., 2016). The overall quality index rating was made up of a base rating out of 30 to indicate study design quality. The two 'studies that employed a 'pre-post- design were given a base rating of 10 and the 22 RCTs given a base quality index rating of 30. This was based upon the assumption that RCTs are of higher methodological quality due to their use of a control group and random allocation to treatment condition. The base ratings were added to the scores out of 14 for the seven types of methodological bias with each study given a 'low' (green = 2), 'unclear' (Yellow = 1) or 'high' (red = 0). Total scores were then converted to an overall percentage quality index rating. Overall quality index ratings can be seen below in *Figure 2.*

Figure 2 - Study design, risk of bias and quality index ratings

Study Name (Lead Author and Publication Year)	Study Design	Type of bias							Total Quality Index Rating (/100%)
		Selection Bias	Performance Bias	Treatment Fidelity	Detection Bias	Statistical Bias	Reporting Bias	Generalisability	
Dirkse 2020	RCT								93
Yun 2012	RCT								95
Hunt 2009	RCT								86
Ljótsson 2010	RCT								86
McCombie 2016	RCT								80
Hummel 2017	RCT								86
Luyster 2020	Pre-Post								32
McCarthy 2016	Pre-Post								27
Murphy 2019	RCT								86
Piette 2011	RCT								89
Proctor 2018	RCT								77
Compen 2018	RCT								95
Schneider 2020	RCT								77
Schröder 2014	RCT								77
Wenzel 2015	RCT								89
Beatty 2016	RCT								84
Hesser 2012	RCT								89
Willems 2017	RCT								80
Gotink 2017	RCT								86
Bond 2010	RCT								75
Abrahams 2017	RCT								75
Admiraal 2017	RCT								84
Chambers 2018	RCT								82
Newby 2017	RCT								80

Selection Bias

Eleven studies were rated as having a high risk of selection bias for targeted or opportunity sampling via the internet (e.g. Hunt et al., 2009). At times, non-response rate was unclear or not -reported and therefore, 10 studies were rated as having an unclear risk of selection bias. Low rated studies (n = 3) reported full selection characteristics including demographics.

Performance Bias

Sixteen of the included studies were rated as 'unclear' risk, five were rated as low and five as high risk of performance bias. Patients were sometimes rewarded financially for their participation (e.g. Wenzel et al., 2015) and often it was not made clear if participants were made aware of their allocated condition.

Treatment Fidelity

Due to the heterogeneity of treatment models, therapeutic delivery method and duration, treatment fidelity was often rated as high risk of bias. Seven of the included studies were rated at high risk of bias, with some not giving detailed information regarding treatment protocol. As some models are easier to manualise and assess for protocol adherence (e.g. CBT), it is perhaps not surprising that some studies that used integrated models (e.g. Yun et al., 2012) or ACT (e.g. Proctor et al., 2018) were rated as high risk in terms of treatment fidelity in comparison.

Detection Bias

Eight studies were rated as high risk and eight studies were rated as unclear risk of detection bias, with several not reporting reliability indicators or indicating whether assessors were blinded to participant treatment condition. Low rated studies (e.g. Dirkse et al., 2020) included reliable change index calculations alongside validity indicators for QoL outcome measure utilised.

Statistical Bias

Nine of the included studies were rated as high and nine as unclear risk of statistical bias. This was often due to studies not including detailed information regarding attrition or providing completer-only analysis. High risk of bias in this area indicates that a study has not included cases that drop-out of treatment in the analysis through intent-to-treat calculations (e.g. Luyster et al. 2020). Those rated as low risk of bias (e.g. Hummel et al., 2017) accounted for participation attrition through imputation of missing data.

Reporting Bias

Sixteen of the included studies were rated as low in risk of reporting bias due to high levels of reporting of outcome measures outlined in the assessment. Of particular focus for this meta-analysis was the reporting of QoL outcome measures which were often employed as secondary outcomes. One study was rated as high risk of bias in the reporting domain (Chambers et al., 2018) as the study did not include standardised mean difference data for QoL, instead reporting non-significant raw data in the appendices.

Generalisability

The majority of the studies, eighteen, were rated as high risk of generalisability bias. Despite the studies focussing upon one long-term health condition (e.g. women with breast cancer), sampling methods often only focussed upon one clinic, region or practice. In addition, several of the studies did not report ethnicity (e.g. Ljótsson et al., 2010).

Summary

Levels of bias varied across studies, with several being rated as unclear due to details not being specified. There were three studies that were not reported as a high risk of bias in any domain (Compen et al., 2018; Dirkse et al., 2020; Piette et al., 2011).

1.2.6 Data extraction and analysis

Where a standardised mean difference (SMD) was not reported through either Cohen's *d* (Cohen, 1988) or Hedges' *g* (Hedges, 1971), raw means and standard deviations were extracted and transformed using an Effect Size Calculator programme developed for R Studio (Jones, 2020). Some have argued that Cohen's d may slightly overestimate the true standardised mean difference, especially in studies with small sample sizes and therefore a correction was made control for this bias (Borenstein et al., 2009). For ease of interpretation, the corrected effect sizes (Hedges' *g)* were transformed back to the more familiar Cohen's *d* for overall summary tables and plots.

Following initial meta-analysis using the random effects model, a quality effects model was calculated to further examine the impact of methodological bias. Whereas the random effects model weighted studies by sample size and deviation from the meta analytic average, the quality effect model utilises the overall risk of bias ratings reported in *Figure 2.* The quality effects model estimates the pooled effect size if all studies were the same methodological quality as the study rated highest in the review. The quality effects model utilises an overall quality rating out of 100%, combining methodological bias ratings with study design. All included RCTs were Given a base score of 30) and pre-post designs were given a base score of 10. The remaining 70% of the quality index percentage was determined by the seven types of methodological bias ratings of 'low', 'unclear' or 'high'. Methodological bias ratings and quality index ratings are shown in *Figure 2* above. All omnibus effects, quality effects model and sub-group analysis was conducted using a specific programme developed for use in 'R Studio' (Balduzzi et al., 2019; Jones, 2020*b*).

1.3 Results

1.3.1 Participant characteristics

This meta-analysis comprises of 24 studies reporting a total of 3485 participants. Six of the studies reported mental health related QOL and physical QoL separately and are therefore duplicated in the analysis. One study reported Quality of Life across three domains, mental, physical and a specific diabetes QoL measure (Newby et al., 2017). One study three-arm randomised controlled trial (RCT) examined the effectiveness of remotely delivered Acceptance and Commitment Therapy (ACT) and remotely-delivered Cognitive Behavioural Therapy (CBT) and therefore these effect sizes were also included separately to allow for subsequent sub-group analysis.

Participants were adults recruited from a variety of locations including healthcare settings, outpatient clinics, advertisements and online. The studies covered a range of physical health conditions including cancer (Abrahams et al., 2017; Admiraal et al., 2017; Beatty et al., 2016; Chambers et al., 2018; Compen et al., 2017; Dirkse et al., 2020; Hummel et al., 2017; McCarthy., 2016; Murphy et al., 2019; Wenzel et al., 2015; Willems et al., 2017; Yun et al., 2012;), irritable bowel syndrome (Hunt et al., 2009; Ljótsson et al., 2010), asthma (Luyster et al., 2020), inflammatory bowel disease (McCombie et al., 2016), epilepsy (Schröder et al., 2014), diabetes (Bond et al., 2010; Newby et al., 2017; Piette et al., 2011), multiple sclerosis (Proctor et al., 2018), heart disease (Gotink et al., 2017; Schneider et al., 2020) and tinnitus (Hesser et al., 2012).

The primary studies covered a range of remotely delivered interventions including CBT (16 studies), mindfulness-based interventions (2 studies), ACT (2 studies), integrated treatment (4 studies). The integrated studies typically included some elements of CBT with additional models such as transactional analysis or problem-solving therapy. Although all were delivered remotely, the delivery method varied across the studies, including self-guided interventions, those with brief or minimal therapist input to those delivered directly via telephone or video by a therapist.

Sample sizes varied from 19 (McCombie et al., 2016) to 409 (Willems et al., 2017) with a mean average of 134 participants per study. *Table 4* below includes key study information and demographics for each of the 24 primary articles.

Table 4 - Relevant Descriptive Characteristics of the Included Articles

Study	*N*	Country	Physical Health Condition	% Female	Intervention Delivery	Therapy Model	QoL Measure	Control Type	Intervention length
Dirkse et al., 2020 (MQoL & PQoL	86	Canada	Cancer (Mixed)	83	Online, therapist guided	CBT	SF-12 (Mental & Physical)	Active control – self guided	5 modules, 8 weeks
Yun et al., 2012	273	Korea	Cancer (Mixed)	73	Online self-guided	Integrated (CBT for fatigue &, Psychoed	EORTC-QLQ-C30 (Cancer)	Waitlist control	6 modules, 12 weeks
Hunt et al., 2009	54	USA	Irritable Bowel Syndrome (IBS)	81	Online self-guided, limited therapist input by email	CBT	IBS-QoL (IBS)	Waitlist control	5 modules, 5 weeks
Ljótsson et al., 2010	86	Sweden	IBS,	72	Online self-guided, weekly message from therapist	CBT	IBS-QoL (IBS)	Waitlist control	10 week
McCombie et al., 2016	19	New Zealand	Inflammatory Bowel Disease (IBD), Crohn's Disease, Colitis	64	Online self-guided	CBT	SF-12 (Mental), SF-12 (Physical) , IBD-	(Treatment as Usual) TAU	8 week, online
Hummel et al., 2017	169	The Netherlands	Cancer (Breast)	100 (Breast cancer)	Online therapist guided, weekly input	CBT	SF-36 (General)	Waitlist control	Weekly- 20-24 weeks, 4-5 modules
Luyster et al.,2020	23	USA	Asthma	63	Self-guided	CBT	AQLQ-Marks (Asthma QoL)	None	6 modules over 6 weeks
McCarthy, 2016	58	USA	Cancer	100 (Breast Cancer)	Video	CBT	EORTC-QLQ-C30 (Cancer)	Placebo and active controls	6 session, 6 weeks, 30-45 mins each
Murphy et al., 2019	114	Australia	Cancer	89 (Breast cancer)	Online, Therapist-guided	CBT	FACT- G (Cancer QoL)	TAU	8 lessons over 16 weeks
Piette et al., 2011 (MQoL & PQoL)	291	USA	Diabetes	51	Telephone	CBT	SF-12 (Mental & Physical)	"Enhanced" usual care - CBT self-help book.	12 weeks intensive + 9 monthly boosters
Proctor et al., 2018	27	UK	Multiple Sclerosis	88	Telephone supported self help	ACT	EQ-5D-5L (General)	TAU	8 weekly telephone calls + self help

Study	*N*	Country	Physical Health Condition	% Female	Intervention Delivery	Therapy Model	QoL Measure	Control Type	Intervention length
Compen et al., 2018 (MQoL & PQoL)	245	The Netherlands	Cancer	86	Online, Therapist guided	MBCT	SF-12 (Mental & Physical)	Active & TAU (face to face MBCT)	8 weeks
Schneider et al., 2020 (MQoL & PQoL)	62	Canada	Heart disease	59	Online, therapist brief support	CBT	SF-12 (Mental & Physical)	Wait list control	5 lessons, 8 weeks
Schröder et al., 2014	78	Germany	Epilepsy	75	Online without therapist	CBT	QOLIE-31 (Epilepsy)	Waitlist control	9 weeks
Wenzel et al., 2015	204	USA	Cancer	100 (Cervical cancer)	Telephone	Integrated (CBT/Transactional analysis)	FACT-Cx (Cancer)	TAU	5 weekly + 1 month booster
Beatty et al., 2016	60	Australia	Cancer	95 (76% breast cancer)	Online, self-guided. Interactive, worksheets etc.	CBT	EORTC-QLQ-C30 (Cancer)	Information only web control (active)	6 modules
Hesser et al., 2012	99	Sweden	Tinnitus	43	Online, guided therapist brief support	CBT and ACT separately	QOL-I (General)	Internet forum waitlist Control (CBT also compared to ACT)	8 modules
Willems et al., 2017	409	The Netherlands	Cancer	81 (71% breast cancer)	Online, self-guided	Integrated CBT and PST (Problem solving therapy)	EORTC-QLQ-C30 (Cancer)	Waitlist control	8 modules
Gotink et al., 2017 (MQoL & PQoL)	324	The Netherlands	Heart Disease	47	Online, self-guided	MBSR	SF-36 (Mental & Physical)	TAU	12 weeks
Bond et al., 2010	62	USA	Diabetes	45	Online, self guided, some interaction with nurse	Integrated (CBT elements/Psycho-ed).	PAID (Diabetes)	TAU	6 months
Abrahams et al., 2017	132	The Netherlands	Cancer	100 (Breast cancer)	Online, therapist guided	CBT	EORTC-QLQ-C30 (Cancer)	TAU	8 modules
Admiraal et al., 2017	138	The Netherlands	Cancer	100 (Breast cancer)	Online, self-guided	Psycho-Ed	EORTC-QLQ-C30 (Cancer	TAU	12 week
Chambers et al., 2018	106	Australia	Cancer	68 (25% breast cancer)	Online, self-guided	CBT	AQoL-8D (General)	Information only web control (active	6 modules, 6 weeks
Newby et al., 2017 (MQoL & PQoL & PAID)	91	Australia	Diabetes	64	Online, self guided	CBT	PAID (Diabetes)	TAU	6 lessons, 10 weeks

1.3.2 Meta-analytic model

The distribution of primary study effects is shown in *Figure 3* below. The variance of the true effect (tau^2) was calculated using the DerSimonian and Laird method (DerSimonian & Laird, 1986).

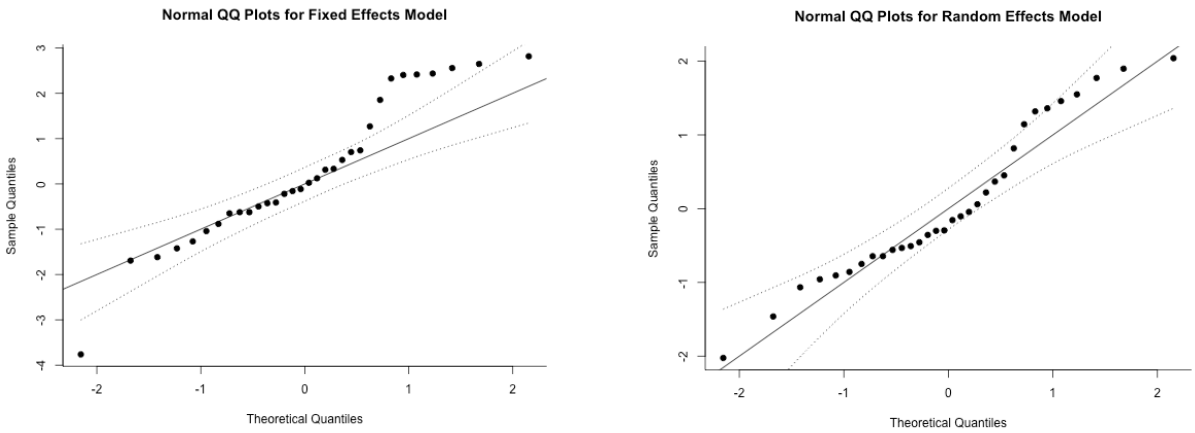

Figure 3 - QQ plots of the distribution of standardised mean difference for fixed and random effects models, plotted using the DL model (DerSimonian & Laird, 1986).

As can be seen from *Figure 3*, the distribution of the standardised mean differences using the fixed effects model is clearly and markedly non-normal. In the random effects model there is some evidence of non-normality in the distribution of standardised mean difference within the primary studies, however, only two study effects fall outside of the 95% confidence interval for the expected normal values. Therefore, the random effects model, using the DerSimonian and Laird estimate of between groups variation (DerSimonian & Laird, 1986) was deemed an appropriate method for the calculation of this meta-analysis.

1.3.4 The omnibus test

A random effects model was calculated using the generic inverse variance method. The random effects model suggested a weighted average standardised mean difference (SMD) g=0.3288 ($z = 6.52$, $p < 0.01$) and a 95% CI of 0.23 to 0.43. A treatment effect of this magnitude would be considered small according to Cohen's suggested effect size categories of small (0.2) medium

(0.5) and large (0.8) respectively (Cohen,1988). The treatment effects described in the primary studies can be seen in *Table 5* and are displayed in a forest plot in *Figure 4*.

Table 5 - Treatment Effects for Primary Studies

Study Name	SMD	Lower 95%-CI	Upper 95%-CI	%W(fixed)	%W(random)
Dirkse 2020 (MQoL)	0.30	-0.12	0.72	2.0	2.9
Dirkse 2020 (PQoL)	0.14	-0.23	0.56	2.0	2.9
Yun 2012	0.26	0.23	0.50	6.4	4.4
Hunt 2009	1.08	0.48	1.68	1.0	1.9
Ljótsson 2010	0.91	0.46	1.36	1.8	2.7
McCombie 2016	0.32	0.04	0.60	4.6	4.0
Hummel 2017	0.06	-0.24	0.36	4.0	3.9
Luyster 2020	0.49	-0.09	1.08	1.0	1.9
McCarthy 2016	0.98	0.38	1.58	1.0	1.9
Murphy 2019	0.74	0.36	1.12	2.5	3.2
Piette 2011 (MQoL)	0.20	-0.03	0.43	6.8	4.5
Piette 2011 (PQoL	0.13	-0.10	0.36	6.8	4.5
Proctor 2018	0.11	-0.64	0.86	0.6	1.3
Compen 2018 (MQoL)	0.67	0.36	0.98	3.7	3.8
Compen 2018 (PQoL)	0.24	-0.64	0.86	3.9	3.8
Schneider 2020 (MQoL	0.23	-0.31	0.77	1.2	2.2
Schneider 2020 (PQoL)	-0.17	-0.71	0.37	1.2	2.2
Schröder 2014	0.04	-0.40	0.48	1.8	2.7
Wenzel 2015	0.14	-0.16	0.44	3.9	3.8
Beatty 2016	0.28	-0.23	0.79	1.4	2.3
Hesser 2012 (CBT)	0.45	-0.05	0.95	1.5	2.4
Hesser 2012 (ACT)	0.35	-0.13	0.83	1.5	2.5
Willems 2017	0.13	0.05	0.31	11.6	5.0
Gotink 2017 (MQoL)	0.22	-0.04	0.48	5.2	4.2
Gotink 2017 (PQoL)	-0.14	-0.36	0.08	7.2	4.6
Bond 2010	0.60	0.09	1.11	1.4	2.3
Abrahams 2017	0.70	0.35	1.05	2.9	3.4
Admiraal 2017	0.19	-0.14	0.52	3.2	3.6
Chambers 2018	0.40	-0.07	0.87	1.6	2.5
Newby 2017 (MQol)	0.67	0.25	1.09	2.0	2.9
Newby 2017 (PQol)	0.14	-0.28	0.56	2.0	2.9
Newby 2017 (PAID)	0.79	0.37	1.21	2.0	2.9

Note. SMD = Standardised Mean Difference - (Cohen's d).

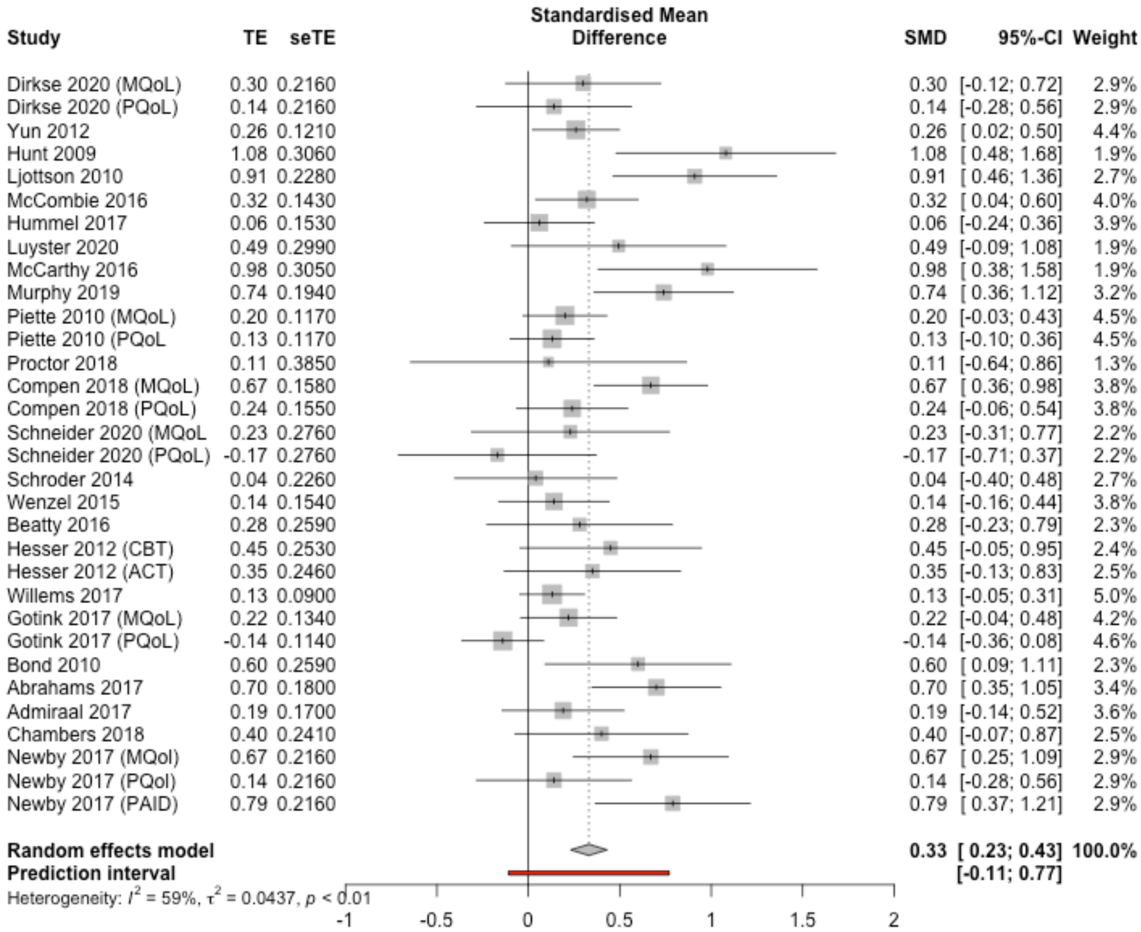

Figure 4 - Forest plot showing a random effects model analysis of included studies, with confidence intervals and prediction interval.

Heterogeneity refers to the difference in the reported effect sizes that cannot be attributed to true variation. It is often interpreted as an indication of methodological bias or participant level effects that may affect treatment efficacy. Higgins I^2 is a useful measure of heterogeneity, in that it reports the proportion of total variation (Higgins et al., 2003). Categories for Higgins I^2 have been suggested as 0 to 50% = low, 50 to 75% = moderate and 75% to 100% = high heterogeneity (Higgins et al., 2003). As heterogeneity increases, it becomes more likely that methodological bias or uncontrolled factors may be present in the literature and may confound the treatment effect. As heterogeneity increases beyond a given threshold (i.e.75%) then the focus of the meta-analysis shifts from synthesis of the studies to the exploration of unexplained variation between study estimates.

A low to moderate level of heterogeneity was found in the primary studies included in this meta-analysis ($tau^2 = 0.0437$, Higgin's $I^2 = 58.9\%$, $Q = 75.38$, $p < 0.01$). In other words, it is estimated that 58.9% of the variability in treatment effect estimates was due to real study differences and 41.1% was due to chance (Riley et al., 2011). Therefore, the 24 included studies reported a fairly consistent effect size for the effectiveness of remotely delivered psychological interventions on QoL for adults with physical health conditions. Nevertheless, it was deemed important to further examine the individual study-level differences through additional sub-group analysis.

1.3.5 The impact of influential primary studies

To determine whether any single studies had a disproportionate effect on the overall treatment effect, a "leave-one-out" analysis was calculated. Separate random effects models were calculated with each of the primary studies removed in turn. Changes in weighted average effect size and heterogeneity were recorded. The result of this "leave-one-out" analysis is presented in the Baujat plot (Baujat et al., 2002) in *Figure 5.*

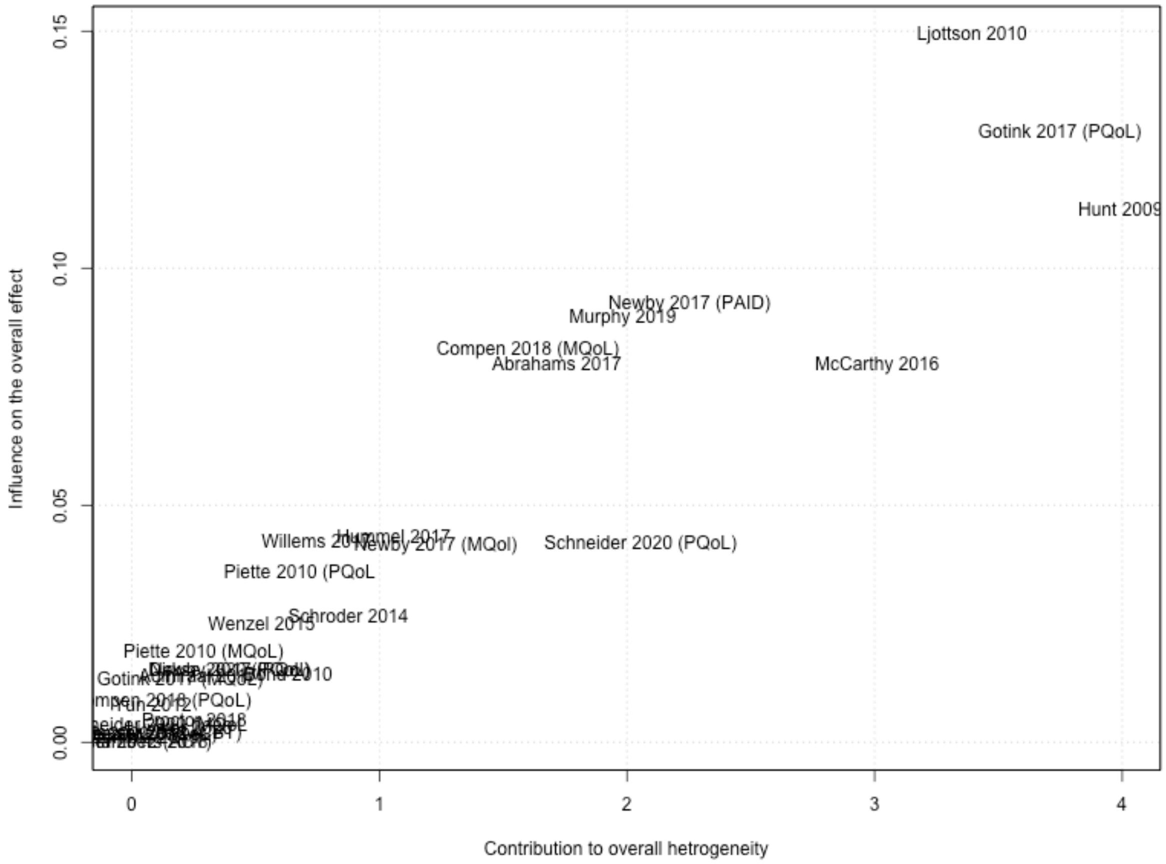

Figure 5 - Baujat plot of heterogeneity. Note. The vertical axis reports the influence of each study on the overall effect. The horizontal axis reports the discrepancy of each study from the rest of the literature (Baujat et al., 2002).

The overall contribution of each study to the heterogeneity rating is plotted on the x-axis, whilst the y-axis depicts the contribution of each study to the overall weighted effect. As can be seen in the Baujat plot in *Figure 5*, three studies (Gotink et al., 2017; Hunt et al., 2009; Ljótsson et al., 2010) appear in the upper right corner of the Baujat plot, suggesting that these three studies appear to be disproportionately influencing the overall effect whilst being discrepant from the rest of the literature. Despite this discrepancy, the removal of these studies did not result in an overall meta-analytic effect that falls outside of the 95% confidence interval (0.2300 to 0.4276) of the omnibus meta-analysis. In fact, re-analysis with all three discrepant and influential studies removed reported a SMD of 0.31, which represents a decrease of less than 5% of the original estimate.

Upon closer inspection, it became apparent that two of the discrepant studies utilised a specific QoL measure for irritable bowel syndrome, the IBS-QOL (Lea & Whorwell, 2001). The difference in type of QoL measure used may partly explain their discrepancy and was therefore examined through additional sub-group analysis.

1.3.6 The effect of risk of bias

The quality effects model assumes that risk of bias and study design will have an impact on estimated treatment effects. This model ensures that studies rated as lower risk of bias are given more weight than those considered to be at high risk of bias. The quality effects model reported a synthesis of SMD=0.3173 (95% CI 0.2175 to 0.4172). The quality effects model evidences an approximately 3.5% decrease relative to the uncorrected random effects estimate SMD=0.3288. Accordingly, when the synthesis includes information about the methodological quality of the studies there is a small change in the weighted average of the included studies.

To further explore heterogeneity, it was important to assess the impact of study level risk of risk of bias by completing additional subgroup analysis. The quality effects model utilises the risk ratings to calculate the meta-analytic effect if all included studies were of the same methodological quality as the best in the literature review. These were undertaken for ratings deemed 'Low risk' and 'Any risk', with the latter combining the 'Unclear' and 'High' risk of bias ratings for all seven types of methodological bias.

Table 6 - Sub-group Analysis to Determine Impact of Methodological Bias

	Low Risk			Any Risk				
	SMD	**95% CI**	**k**	**SMD**	**95% CI**	**k**	**X^2**	**P**
Selection bias	0.14	-0.04 to 0.32	5	0.37	0.26 to 0.48	27	4.81	0.03*
Performance bias	0.37	0.23 to 0.50	7	0.32	0.20 to 0.44	25	0.27	0.60
Treatment fidelity	0.38	0.17 to 0.58	7	0.32	0.20 to 0.43	25	0.27	0.60
Detection bias	0.44	0.27 to 0.61	11	0.27	0.15 to 0.38	21	2.77	0.10
Statistical bias	0.35	0.19 to 0.50	11	0.32	0.19 to 0.45	21	0.07	0.79
Reporting bias	0.33	0.21 to 0.46	9	0.32	0.16 to 0.48	23	0.02	0.89
Generalisability	0.19	0.06 to 0.33	3	0.35	0.24 to 0.47	29	3.17	0.08

**= Statistical significance at the p <0.05 level*

As can be seen above in *Table 6*, selection bias evidenced a statistically significant difference in estimates of treatment efficacy, with studies rated as low risk of selection bias being associated with lower estimates of the impact of remote psychological interventions on quality of life. The Higgins I^2 value for the 5 studies rated as low risk for selection bias was $I^2 = 46\%$.

1.3.7 The impact of publication and small study bias

Publication bias refers to the systematic bias present in the literature due to the fact that studies reporting a statistically significant positive effects are more likely to be published than those reporting null effects (Duval & Tweedie., 2000; Sterne et al., 2011). Due to the lower power and precision often present within small *n* studies, small study bias refers to the tendency of greater variation being found in the study level effects when compared to larger studies (Sterne et al., 2011).

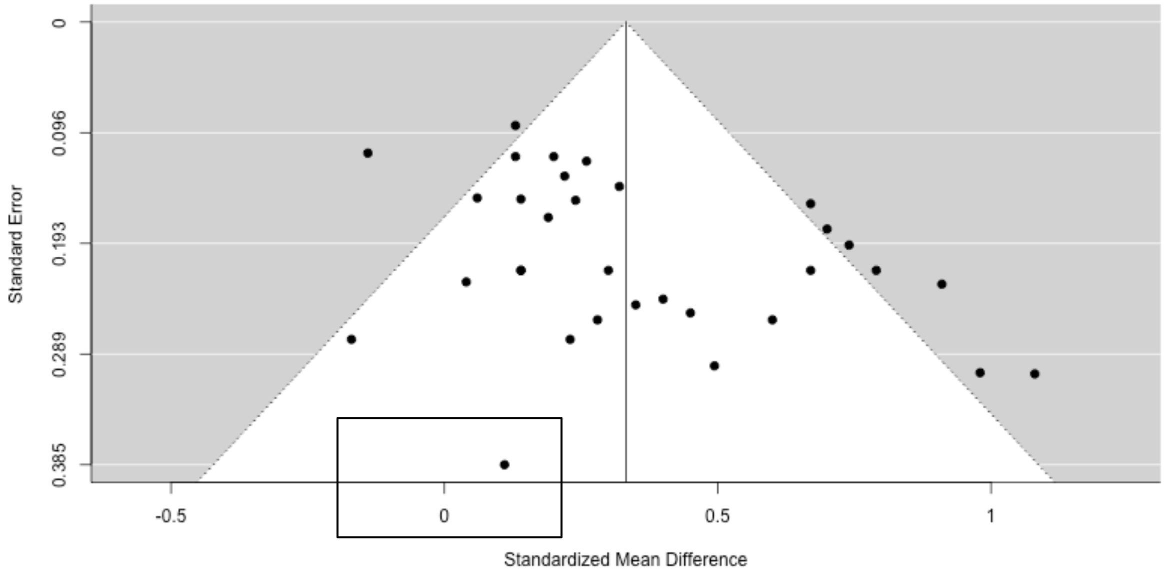

Figure 6 - Funnel plot of publication bias.

Note: the standardised mean difference plotted against the standard error for each study effect. The 95% confidence interval of the expected distribution of the treatment effect is shown as an inverted "funnel". The area marked with a rectangle is that associated with publication bias

Funnel plots of publication bias such as that depicted in *Figure 6* are predominantly used as visual aids. The funnel plot depicts a scatterplot with each study's SMD and precision plotted in relation to their proximity to the average SMD indicated by the middle vertical line. In the absence of any publication bias, scores with higher precision are expected to be plotted nearer to the average SMD and studies with lower precision are expected fall evenly on both sides of the average, creating an inverted funnel (Sterne et al., 2011).

Within this meta-analysis, the area of the funnel plot associated with small sample sizes and non-significant results is marked with a rectangle at the bottom left hand corner of the funnel. A visual inspection of the funnel plot in *Figure 5* indicates that there may be some evidence of publication bias in the distribution of effect. To examine this further, the effect of publication bias was simulated using a trim and fill method (Duval & Tweedie, 2000). This method removes some of the most extreme small studies on the side of the funnel associated with positive treatment effects (i.e., the right-hand side) and recalculates the effect size at each iteration until the plot is symmetrical around the corrected SMD.

In the 'trim and fill' funnel plot in *Figure 6*, the dark circles represent the included studies. The white circles depict the imputed studies (n=8) following the procedure. The trim and fill method (Duval & Tweedie., 2000) led to a corrected random effects model of 0.1911. The adjusted estimate would represent a 42% decrease, relative to the original omnibus analysis.

However, as can be seen in *Figure 7*, several of the 'imputed' studies have been added to the area of the plot associated with at least moderate sample sizes. Therefore, they are not reflective of the unpublished, small studies that the 'trim-and-fill' procedure is designed to add. In addition, the method appeared to have resulted in over-correction, as several of the imputed studies indicated that interventions may decrease health-related quality of life, which is not present or reflective of the existent literature.

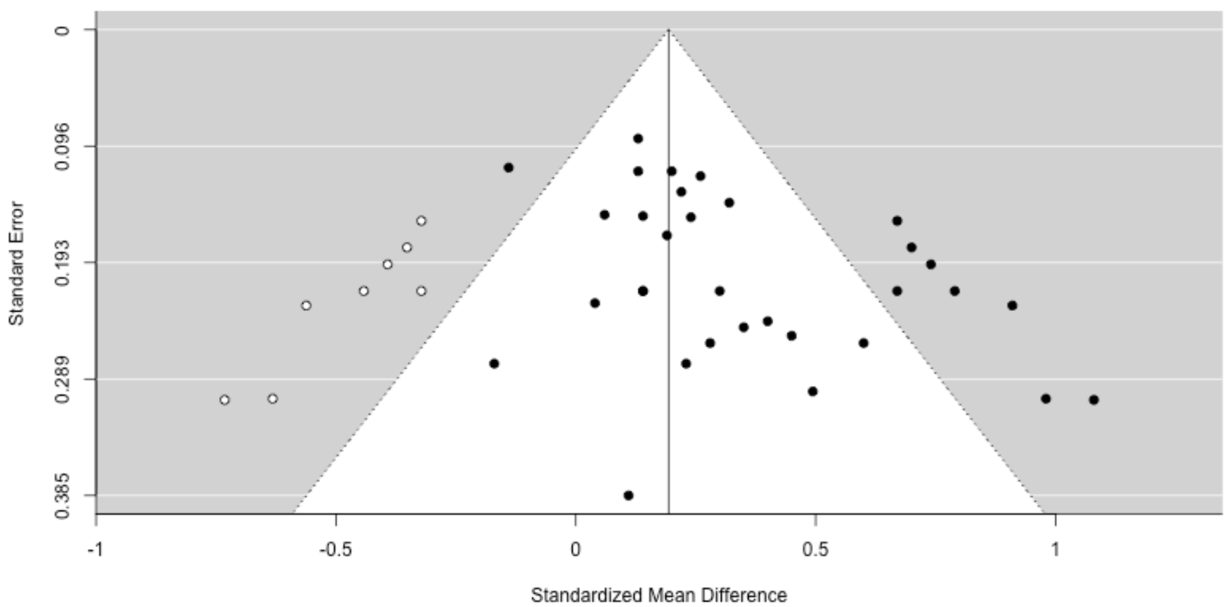

Figure 7 - Funnel plot for the 'trim-and-fill method with imputed studies as white circles (Duval & Tweedie, 2000).

As an alternative to the trim-and-fill procedure, the Rosenthal method (Rosenthal, 1979) can be used to calculate the number of missing null effect studies that would be required for the observed effect to no longer reach significance. Using the Rosenthal algorithm, based on the included studies, 1116 unpublished null studies would be required to reduce the meta-analytic effect to non-significance (Rosenthal, 1979). This indicates that despite an indication of possible publication bias present within the literature, the observed results are robust enough to withstand such a bias.

1.3.8 Subgroup analyses

To further examine possible heterogeneity of the included articles, additional sub-group analyses were calculated to further examine the impact of study level covariates such as type of QoL measure used, therapeutic approach and method of therapy delivery.

Type of Health-Related Quality of Life Outcome Measure

Initial subgroup-analysis was conducted to determine whether the type of QoL outcome measure employed made a difference to the observed SMD. Studies were separated by HR-QoL outcome measure type to compare general HR-QoL, disease/disorder specific HR-QoL and those that reported mental and physical health subscale scores, such as the SF-36 (Ware & Sherbourne, 1992).

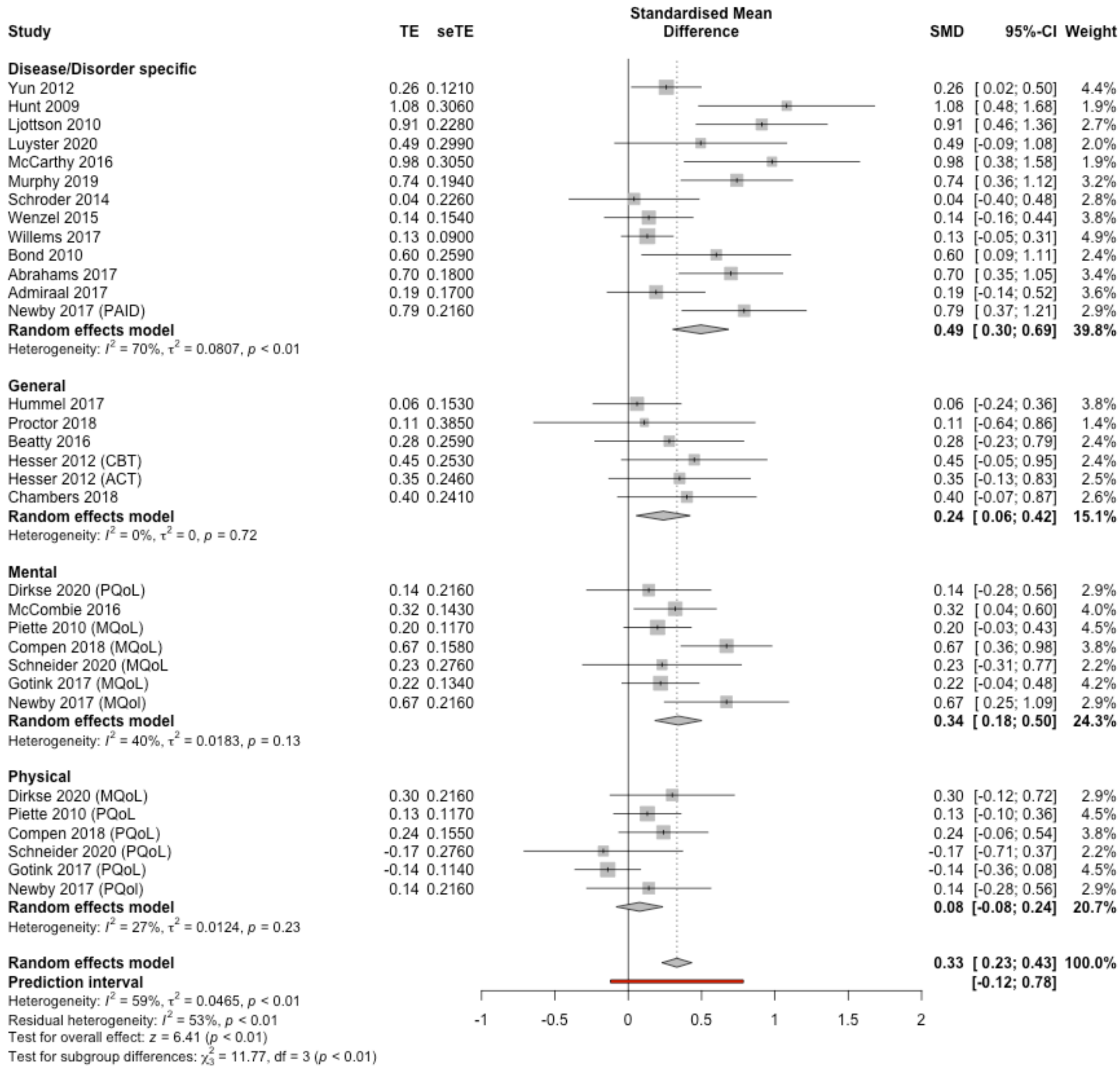

Figure 8 - Forest plot showing sub-group analysis according to type of HR-QoL outcome measure used

As can be seen in *Figure 8,* the estimate of treatment efficacy for disease/disorder specific measure of HR-QoL was SMD = 0.49 (95% Confidence Interval (CI) 0.30-0.69), SMD= 0.24 (95% CI 0.06-0.42) for General HR-QoL, SMD = 0.34 (95% CI 0.18-0.50 for 'Mental' QoL subscales and a SMD = 0.08 (95% CI -0.08-0.24 for 'Physical' HR-QoL subscales. The difference between the types of Quality of Life outcome measure was statistically significant (X^2= 11.77, p< 0.01). This indicates that those studies that measured HR-QoL using a disease/disorder specific measure report better results with regards to HR-QoL.

Therapeutic modality

Further sub-group analysis was undertaken to estimate whether the pooled effect varied according to therapeutic model employed. As shown in the forest plot in *Figure 9* below, treatment efficacy for studies that employed CBT was SMD=0.40 (95% CI 0.26 – 0.54) compared to overall treatment effect of SMD= 0.3288 (95% CI 0.23-0.43). However, due to the small number of studies that employed alternative treatment models, it is difficult to determine whether these represent true differences. Overall, the difference in pooled effect size according to therapeutic model employed did not reach statistical significance (X^2=4.33, p=0.36).

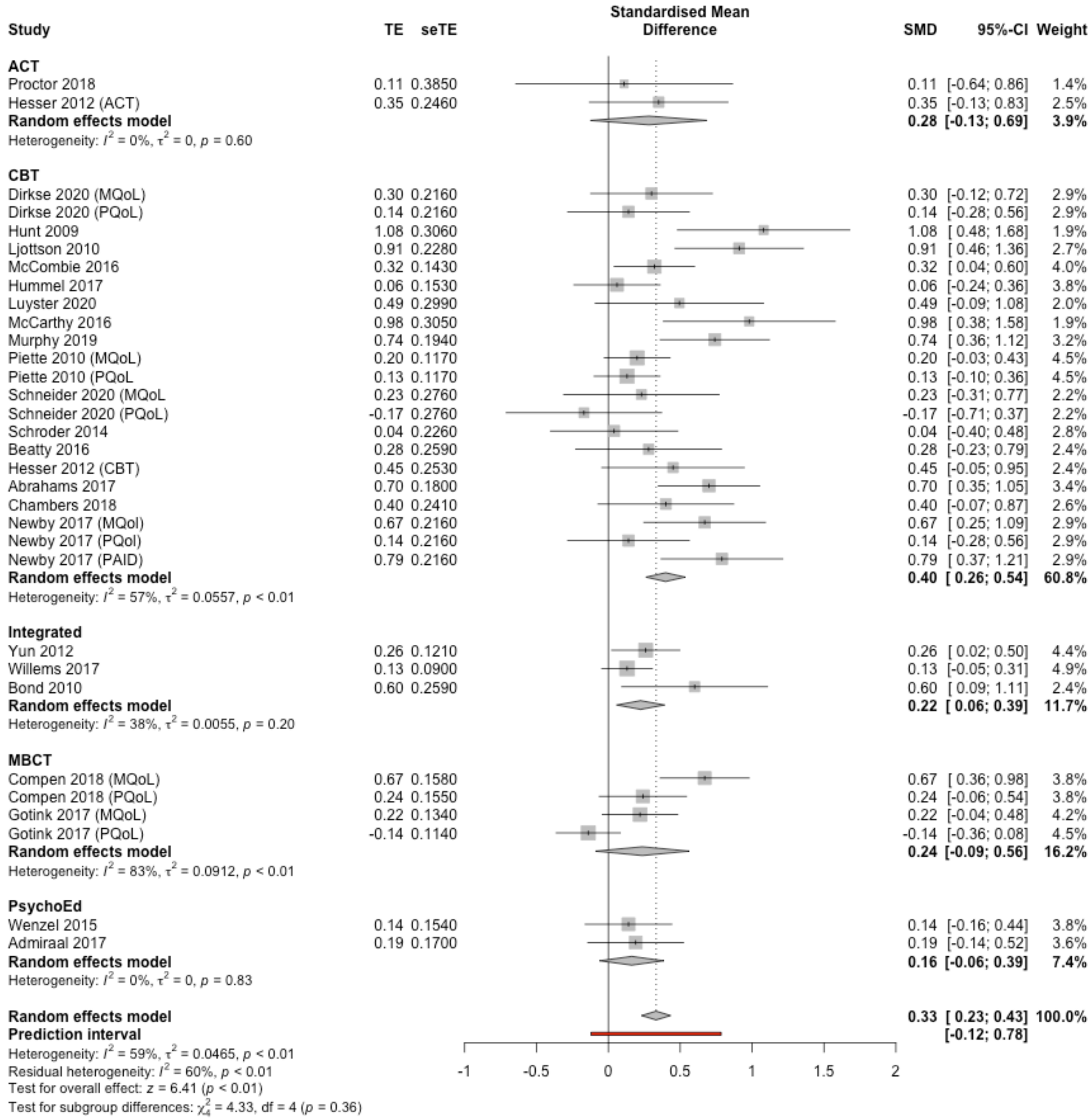

Figure 9 - Forest plot showing sub-group analysis according to therapeutic modality

Delivery Method

It was deemed important to explore differences in pooled treatment effects according to chosen delivery method. Overall, a statistically significant difference in pooled treatment effect was found (X^2= 10.93, p = 0.01), with those delivered online and involving direct therapist contact showing increased treatment effects on HR-QoL outcomes. As can be seen in *Figure 10*, treatment efficacy for studies that were conducted online with therapist input reported SMD=0.43 (95% CI 0.25 – 0.62) in comparison to SMD=0.27 (95% CI 0.14-0.40) for those studies that were self-directed online. It is difficult to directly compare studies that utilised telephone and video delivery due to the small number of studies in each of these sub-groups.

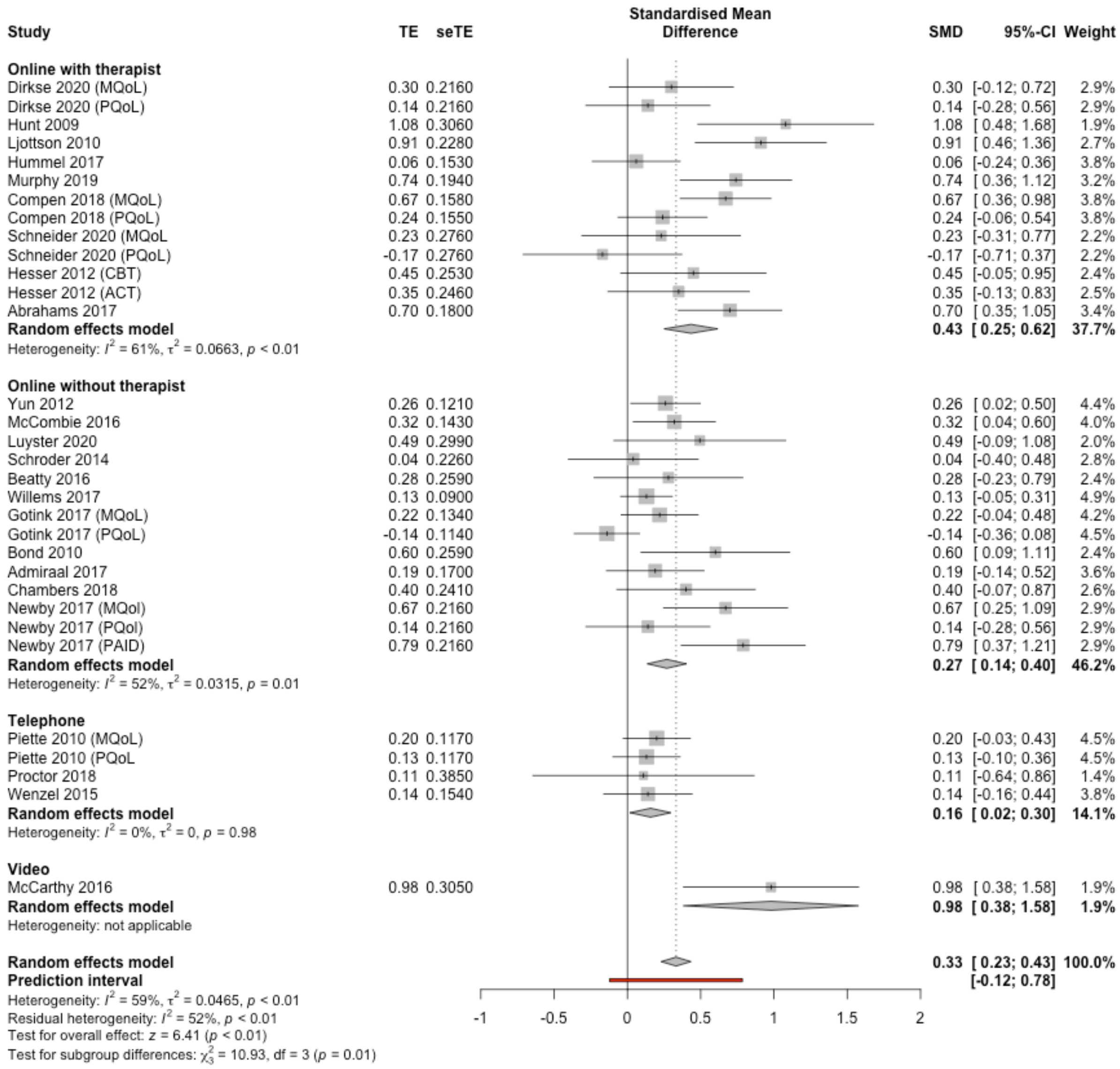

Figure 10 - Forest plot showing sub-group analysis according to therapy delivery method

1.5 Discussion

1.5.1 Summary of Findings

The aim of this meta-analysis was to determine the effectiveness of remotely delivered psychological interventions on HR-QoL in adults with physical health conditions. The evidence base exploring the effectiveness of such interventions is limited, especially those designed to measure health-related quality of life (HR-QoL) outcomes. Twenty-four studies were included in the review, with 6 studies measuring HR-QoL using multiple outcome measures (e.g. Newby et al., 2017). Overall, this systematic review of the literature provides indication that remotely-delivered psychological interventions can have small to moderate treatment effects on QoL outcomes when compared to controls.

Sub-group analyses indicated that those studies that employed remotely delivered CBT led to slightly increased effect sizes when compared to those that employed MBCT, ACT or integrated treatment interventions. However, this difference did not reach statistical significance and should be interpreted with caution, especially as the majority of studies (60%) did employ remotely-delivered CBT. Additional variation was noted through further sub-group analysis according to method of delivery, with larger effect sizes noted in studies that involved direct therapist input. However, this difference did not reach statistical significance and therefore firm conclusions cannot be drawn. In addition, the duration and nature of therapist input varied across the included studies with some that used video-conferencing (McCarthy et al., 2016) and other studies that involved weekly check-in emails with a therapist (e.g. Hunt et al., 2009).

1.5.2 Quality of the Evidence

It was important to explore study quality and the impact of methodological bias and study design when undertaking the present research. This allowed the researcher to give increased weight during the analysis process to those studies with higher quality study designs (e.g. RCTs) and those deemed to have a lower risk of methodological bias. The quality of included studies varied, with several deemed to be at high risk of methodological bias due to factors such as non-blinding of participants or researchers to treatment condition. In addition, only two of the included studies were RCTs with active control conditions (Compen et al., 2018; Dirke et al., 2020) and therefore it is not possible to determine how remotely delivered interventions differ in comparison to face-to-face psychological therapy.

1.5.3 Findings in the Context of the Existing Literature

The overall findings indicate a small treatment effect for the effectiveness of remotely-delivered psychological interventions for HR-QoL outcomes in adults with physical health conditions (*SMD g*=0.3288). These findings are comparable to recent meta-analytic research of internet-delivered CBT for anxiety and depression (SMD *g*=0.42-0.80; Andrews et al., 2018; Etzelmueller et al., 2020) and remote psychological interventions for mental health outcomes in long-term physical health conditions (SMD=0.13-0.63; Mehta et al., 2019; White et al., 2020).

As with the current review, moderate levels of heterogeneity within the previous literature was prevalent, due to the wide variety of methods and study designs employed. Nevertheless, the findings add to the evidence base for the effectiveness of remotely delivered psychological interventions on HR-QoL in adults with physical health conditions.

With regards to sub-group analysis, the findings raise additional questions regarding the use of both generic and disease-specific HR-QoL outcomes (Engström et al., 2001; Taft, 2021). There is some ongoing debate, with some calling for a unified definition of HR-QoL to allow for measurement across conditions. Others argue that nuanced disease-specific factors necessitate the use disease-specific HR-QoL outcomes (Engström et al., 2001). An extension to CONSORT, the widely used reporting standards for RCTs now recommends that patient-reported outcomes such as HR-QoL be identified in study abstracts if employed as primary or secondary outcomes (Calvert et al., 2013).

1.5.4 Clinical Implications

The findings of this meta-analysis support the growing evidence base for the potential effectiveness of remotely delivered psychological interventions for those with physical health conditions. This client group in particular may face additional barriers with regards to accessing treatment, due to physical symptoms, fatigue or medical treatments (Allerton & Emerson., 2012). This review also builds upon the evidence base for the consideration of tiered treatments for those who may not require 1:1 in-person therapy but who may benefit from remotely delivered, less intensive psychological support.

Improving Access to Psychological Therapies (IAPT) have established pathways for those with long-term conditions in several areas and qualitative research has highlighted the importance of providing flexible and individually-tailored interventions (Carroll et al., 2020; National Collaborating Centre for Mental Health., 2018). This meta-analysis provides support for remotely-delivered psychology interventions on HR-QoL outcomes which has clinical implications for service provision. Remotely delivered interventions are likely to be more cost-effective when compared to face-to-face interventions (Hedman et al., 2012; Lunkenheimer et al., 2021). In addition, the results of the present review provide some support for the effectiveness of self-directed therapy on HR-QoL outcomes. However, overall, those interventions that included therapist input showed increased treatment effects. Therefore, it may be that such interventions could be employed as part of a tiered service dependent on an individual's baseline HR-QoL and symptoms.

These results provide indication that it may be helpful for Clinical Psychologists working within health psychology settings or in the community with adults with physical health conditions to consider remotely delivered interventions. Prior to the Covid-19 pandemic, remote interventions for this client group had not been routinely offered which raises questions for access options for those who may struggle to attend clinics in person.

The current meta-analysis also highlights the importance of considering the type of HR-QoL measure utilised in both assessment and intervention. Increased treatment efficacy was found in disease-specific HR-QoL measures when compared to general HR-QoL measures and this has implications for those that should be utilised routinely in clinical practice, both as part of assessment and intervention. Clinicians should consider carefully the appropriate HR-QoL outcome measure when working with adults with physical health conditions. The present results suggest that disease-specific HR-QoL may be more sensitive to change and therefore may be more appropriate when measuring intervention effectiveness or change in outcomes over time.

The findings highlight the potential effectiveness of remotely delivered interventions but do not provide evidence for effectiveness in comparison to face-to-face therapy. This is due to the limited number of included studies that employed an active control condition. Nevertheless, this review builds upon the existent literature at a time when remotely delivered psychological interventions are being used extensively in part due to the Covid-19 pandemic. This shift is likely to remain, at least in some part, in those services and patients who which benefit from increased flexibility and capacity (Wind et al., 2020).

1.5.6 Limitations

The current meta-analysis had several limitations. Firstly, when sub-group analyses were employed, some of the groups (e.g. video delivery) only had one or two studies in and therefore due to reduced power it was difficult to meaningfully interpret the findings. In addition, the included studies varied with regards to methodological quality although the quality effects model allowed for weighting and further examination of the impact of individual study bias. Thirdly, the majority of the included studies did not include an active control condition and therefore it is difficult to determine whether the review supports previous literature that has found internet delivered CBT may be as effective as face-to-face CBT for mental health conditions (e.g. Andrews et al., 2018).

A fourth consideration with regards to potential limitations of the current review is that the present findings should be viewed with some caution and an awareness of factors that may affect reliability of self-report measures such as social desirability bias (Caputo, 2017; Van de Mortel, 2008). A review into the use of HR-QoL measures highlighted the methodological difficulties that arise with subjective, as opposed to objective outcomes (Lin et al., 2013). The measures of HR-QoL utilised within the included studies tended to rely upon self-report (e.g. the SF-36; Ware & Sherbourne; 1992).

A further consideration, more widely, is that health-related QoL is somewhat poorly defined across the literature with some outcome measures encompassing areas that would indicate self-perceived health status as opposed to quality of life (Karimi & Brazier, 2016). However, often these are both captured under the the umbrella of HR-QoL.

This is evident in the range of outcome measures used across the RCTs included within this meta-analysis. As the present results found larger treatment effects for studies that employed disease or disorder specific HR-QoL outcomes, this supports the notion for the importance of utilising the most appropriate outcome measure for each patient group.

1.5.7 Recommendations for Future Research

The evidence base would be further strengthened by further high quality RCTs that focus upon treatment fidelity, blinding of condition and active control conditions to determine how remotely delivered interventions compare to those delivered face-to-face. Further research into the effectiveness of remotely delivered interventions that utilise alternative models to CBT would also be advantageous. For example, online interventions utilising mindfulness have shown small to moderate treatment effects for mental health outcomes (Spijkerman et al., 2016) and small treatment effects have been found for remotely delivered ACT for depression and anxiety (Thompson et al., 2021).

Meta-analytic research has found small treatment effects for ACT in patients with chronic pain (Öst, 2014) and that face-to-face ACT may be as effective as CBT in treating somatic health problems (A-Tjak et al., 2015). High-quality RCTs that employ alternative treatment models such as ACT, compassion-focussed-therapy or mindfulness-based interventions would give services and patients the choice if they are deemed to be as effective as CBT.

Recent reviews have also highlighted that delivery of transdiagnostic psychological interventions may be advantageous to the development of those for specific physical health conditions (Thompson et al., 2021). Although given the poorly defined concept of HR-QoL more generally and the current findings that disease-specific measures may lead to better treatment outcomes, transdiagnostic approaches may not always be appropriate.

1.5.9 Conclusion

The effectiveness of remotely-delivered psychological interventions for HR-QoL in adults with physical health conditions shows some promise. This review indicates small treatment effects across a number of health conditions such as cancer, tinnitus, diabetes and irritable bowel syndrome. This treatment effect may reduce with increased numbers of high quality RCTs. This review provides an overview of the current literature into outcomes of HR-QoL specifically and further research examining alternative models and with greater methodological quality would strengthen these findings.

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** indicates a primary included study*

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VOLUME ONE

CHAPTER TWO: LITERATURE REVIEW

The Effectiveness of Remotely Delivered Psychological Interventions on Health-Related Quality of Life (HR-QoL) in Adults with Physical Health Conditions: A Meta-Analysis

2.0 Abstract

Introduction: Qualitative social experiences of adolescents with Cystic Fibrosis (CF) in the U.K. is lacking despite previous research focussed upon cross-infection control guidelines, disclosure and transition. The current research aimed to explore the social experiences of adolescents with CF in an outpatient setting.

Method: Five adolescents, three males and two females were interviewed via video to explore their social experiences of CF. Interviews were transcribed and analysed using Interpretative Phenomenological Analysis, a method that aimed to capture the lived experiences of individuals through the interpretation of the researcher.

Results: Two superordinate and six subordinate themes were identified to represent the social experiences of the recruited participants. A superordinate theme of 'A desire to belong and be understood in the context of CF' was identified with the following four subthemes: Not wanting to be Treated Differently because of CF, The Burden of Disclosure and the Importance of Trust, Finding Common Ground and Connecting in Other Ways. A second superordinate theme 'The relationship between increasing independence and social relationships in the context of CF' was organised into the subthemes: Experiences of Defiance and Conflict with Others (e.g. family) and Independence as a Process - Reflections and Mixed Emotions.

Discussion and Conclusion: Increasing independence is experienced differently for adolescents with CF and sometimes involves conflict, with others or within the self. The importance of trust and social support from others socially was considered important across both superordinate themes. The social experiences of adolescents with CF bear some similarity to the experiences of their peers and to those with other chronic health conditions. An awareness of such experiences can help clinicians who work with adolescents during this stage, as they prepare to transition to adult services.

2.1 Introduction

2.1.1 Cystic Fibrosis

Cystic Fibrosis (CF) is a life-shortening hereditary condition caused by a gene mutation which leads to the build-up of thick mucus in the respiratory system. CF causes chronic respiratory symptoms, digestive difficulties and increased susceptibility to lung infections that can affect overall lung functioning. Newborn screening is now widely undertaken in developed countries and diagnosis of CF occurs in approximately 1 in every 2500 births in the United Kingdom (Cystic Fibrosis Trust, 2020). Approximately 10,700 individuals are currently listed on the UK CF registry (Cystic Fibrosis Trust, 2020). Once a condition associated with child mortality, CF treatment advances in the past few decades have increased the average life expectancy to 45-50 years of age (Bell et al., 2020; Cystic Fibrosis Trust., 2020). Nevertheless, CF remains characterised by high levels of treatment burden (e.g. Davies et al., 2020) and meta-analytic reviews have found a higher prevalence of anxiety and depression in those with CF, when compared to the general population (Barker et al., 2019; Kimball et al., 2021; Quittner et al., 2014). In addition, due to the risk of passing on infections, cross-infection control guidelines stipulate that individuals with CF cannot meet each other face-to-face. This means that socially, they are unable to form in-person relationships with others who have the same condition (Cystic Fibrosis Trust, 2021).

2.2.2 The Focus on Adolescence

According to Erikson's theory of psychosocial development, the period between 12 and 18 years is characterised by identity versus role confusion and associated conflict (Erikson, 1968). Adolescence is thought to involve the development of a sense of self through a detailed exploration of values, goals and beliefs in relation to peers (Ragelienė, 2016). This period often also involves educational transition and a shift in emphasis from parent to peer relationships (Albarello et al., 2017; Ragelienė, 2016;). Neurodevelopmentally, the frontal cortex associated with emotion, impulse control and decision making continues to develop throughout adolescence and into young adulthood (Blakemore & Robbins, 2012; Pfeifer & Berkman, 2018). Given the importance of adolescence in terms of social and neurodevelopment, it is often an area of focus when exploring experiences of those with physical health conditions.

2.2.3 Adolescence and Physical Health Conditions

The period of adolescence is often associated with increased levels of treatment autonomy and health-related decision making for those living with physical health conditions. Peer support has also been identified as a social determinant in improving the health outcomes of those with physical health conditions, alongside supportive family relationships and access to education (Kirk & Milnes., 2021; Viner et al., 2012).

For adolescents with CF, daily treatment can involve a complex and ever-changing combination of inhaled medications, oral antibiotics and physiotherapy (Sawicki et al., 2013). Occasionally, patients are admitted to a hospital for additional treatment which may have social implications for adolescents who are physically absent from education settings and peers (Lum et al., 2017; Singh et al., 2020). A meta-synthesis of seven studies indicated that social support may be a factor in facilitating physical activity treatment adherence in CF specifically (Denford et al., 2020) and as such, is an important factor in understanding treatment of CF. In addition, the cross-infection control guidelines that prevent CF patients from meeting each other which is different to those with other long-term physical health conditions (Cystic Fibrosis Trust, 2021).

2.2.4 Qualitative Experiences of those with Physical Health Conditions

Patient involvement and collaboration have been identified as important ways to help improve the health-related quality of life of those who live with physical health conditions. There has been an increase in the development of research in this area with a view to improve service provision and overall health outcomes (Bourgeault et al., 2013). The perceived value of qualitative research continues to grow amid recognition of the rich and detailed insight it can provide of the lived experiences of patients (Braun & Clarke, 2019).

Qualitative approaches, such as Interpretative Phenomenological Analysis, have been utilised across the field of health psychology to gain a deeper understanding of the lived experiences of patients (Brocki & Wearden, 2006; Peat et al., 2018; Tang & Dos Santos, 2017). Such approaches have also focussed on adolescents with health conditions and have focussed upon specific areas such as transition (Rasalingham et al., 2021) and disclosure (Kaushansky et al., 2017).

With regards to CF specifically, a systematic review of qualitative research undertaken with children highlighted six common themes including gaining resilience, lifestyle restriction, treatment resentment and emotional vulnerability (Jamieson et al., 2014). However, this review summarised the research with children as opposed to adolescents and did not focus on social experiences, meaning that the impact of CF on social development remains unclear.

Internationally, qualitative research with adolescents who have CF has indicated that individuals may experience social isolation from peers (Moola et al., 2018); Toth et al. 2016). A thematic analysis of nine interviews grounded in social theories of risk explored the experience of cross-infection control guidelines that prevent individuals with CF from meeting each other face-to-face (Moola et al., 2018). This Canadian research found that some young people with CF experience feelings of loneliness as a result of social isolation and that social media was an important tool for connecting with others. Similarly, a grounded theory analysis of interviews with ten young adults in the United States found similar themes alongside supported autonomy and relationships with siblings (Toth et al., 2016).

2.2.5 Social Experiences of Young People with Physical Health Conditions

Previous research has explored the role of social support and friendships for children and adolescents with physical health conditions. For example, for those with diabetes, the importance of close friendships has been recognised and has led to the development of group meetings and summer camps for those with diabetes to socialise together (Bultas et al., 2015; La Greca et al., 1995). Similar findings have been noted when exploring the effectiveness of summer camps for young people with various physical conditions including cancer and asthma (Meltzer et al., 2018). However, due to cross-infection control guidelines, such camps are not possible for adolescents with CF (Cystic Fibrosis Trust, 2021). Peer support has also been found to negatively predict diabetes related distress in longitudinal research (Raymaekers et al., 2017). In addition, qualitative research has found similar themes in samples of children who have been hospitalised for cancer treatment, with them benefitting from peer support when reintegrating back into school (Ingersgaard et al., 2021).

2.2.6 Research Aims

Healthcare providers increasingly recognise the value of qualitative research when seeking to improve service provision and overall health outcomes for those with physical conditions. A recent report outlining future directions for global CF care highlighted the importance of patient engagement (Bell et al., 2020).

Qualitative research to date has illuminated the overall experiences of children and young adults with CF (e.g. Jamieson et al., 2014) with some focus on the general social experiences of adolescents with CF conducted internationally (Moola et al., 2013; Toth et al., 2016). As cross-infection control guidelines limit face-to-face contact, some previous research has highlighted this as a potential factor for consideration (Moola et al., 2018). Resultant themes have explored specific experiences such as the impact of cross-infection control guidelines alongside more general social experiences of connection and isolation (Moola et al., 2018; Toth et al., 2016). Such qualitative research has yet to be undertaken in the United Kingdom with an outpatient adolescent sample. This group of CF patients has been chosen for the current research as they encompass the important social developmental stage of adolescence whilst living within the community. In addition, adolescents with CF are unable to access disease-specific residential camps and in-person support groups that have been associated with increased self-efficacy and perceived social support in those with other physical health conditions (Meltzer et al., 2018).

Previous qualitative research on the experiences of CF has often employed deductive methodologies that are grounded in the existent literature and seek a common truth (e.g. Moola et al., 2018). In contrast, the present research will employ an idiographic and interpretative methodology (IPA) with a view to explore the individual and lived social experiences of each participant, whilst holding on to the impact of the researcher in this process (Smith et al., 2009). In summary, this research aims to explore the lived social experiences of adolescents with Cystic Fibrosis in a U.K. outpatient setting, in the context of cross-infection control guidelines that prevent them from meeting each other face-to-face.

2.2 Method

2.2.1 Design and Rationale

The aim of this research was to gain a sense of the lived social experiences of adolescents with CF and therefore it was important to consider the most appropriate qualitative approach. Phenomenology is a term used to describe the philosophical study of individual conscious experiences in relation to specific events or phenomena (Shinebourne, 2011). Interpretative Phenomenological Analysis (IPA) is a qualitative approach that uses phenomenology to examine the experiences and sense-making of individual participants through the interpretation of the researcher (Smith et al., 2009). Given the research aims, IPA was deemed to be the most appropriate research methodology.

The *hermeneutic cycle* within IPA refers to consideration of the individual parts in the context of the whole. This cycle may be present when analysing the meaning of words within sentences, sections of text within a transcribed interview or individual participants within whole samples. In practice, this may involve attention being paid to a single word, broadening out to whole sentences or portions of text and then considering the wider context of the entire interview and sample (Smith et al., 2009; Smith, 2017).

The hermeneutic cycle is also part of the iterative nature of IPA, meaning that the process of analysis tends to evolve over time and may go back and forth between participants as opposed to being linear (Smith et al., 2009). In this sense, IPA integrates the researcher's sense-making with that of each individual participant, sometimes known as the *double hermeneutic* (Smith et al., 2009). The aim of the IPA approach therefore is to ensure that throughout the analytic process the researcher's own interpretations are explored as an integral part of the analysis (Smith et al., 2009).

2.2.2 Recruitment, sampling and ethical approval

As the research question aimed to examine the lived experiences and sense-making of adolescents with CF, the sampling method utilised was convenience and purposive to ensure that a homogenous sample of this nature could be recruited. The Principle Investigator assisted with initial identification of potential participants from a pool of patients who were receiving outpatient clinical care for CF. Those approached were receiving outpatient clinical care for CF from one large regional healthcare site. This level of specificity in sampling method would ensure that the research question could be answered meaningfully for a particular set of individual participants. As the research involved recruitment of patients via the National Health Service (NHS), formal ethical approval was granted through the Health Research Authority, Research Ethics Committee (REC) and the local Research & Development team. Ethical approval letters and emails can be found in Appendices A and B. As the research sought to gather data from a homogenous and purposive sample of adolescents with CF, some inclusion and exclusion criteria are shown in *Table 1*.

Table 7 - Inclusion Criteria and Rationale

Inclusion Criteria	Rationale
Children receiving outpatient clinical care for Cystic Fibrosis.	Purposive sampling – To ensure homogeneity of sample. Convenience Principle Investigator a clinician at the recruitment site required to have access to patients to enable initial contact/identification of possible participants.
Participants must be between 13 and 16 years of age at the time of interview.	Research examining lived experiences of adolescents with CF receiving outpatient care for CF, those 17 years and older would be transitioning to adult services.
Participants must be able to speak English fluently	IPA methodology is not suitable through use of interpreters due to the focus on meaning and sense-making as conveyed through the use of language Analysis process involves consideration of specific language used and is limited through researcher's knowledge of the English language.

The age range for inclusion in the research was between 13 and 16 years of age. Those under 13 years or over 16 years at the time of interview were not approached for participation by the Principle Investigator. Those aged 17 years and over would be in the process of transitioning to adult CF care. Participants were recruited through an outpatient CF clinic.

2.2.3 Schedule of Events for Participant Recruitment

For ethical reasons, the researcher/Chief Investigator (CI) was not involved in the initial identification of participants. Potential participants were approached by a member of the clinical team (Principal Investigator) following examination of the clinical database to identify those who met the inclusion criteria. The full schedule of participant recruitment is outlined in *Figure 1* below. All participants were receiving outpatient clinic care for CF within one clinical setting within which the Principal Investigator worked. Local ethical approval was also sought through the specific healthcare setting and for this reason, the potential participant recruitment pool was limited to one healthcare setting.

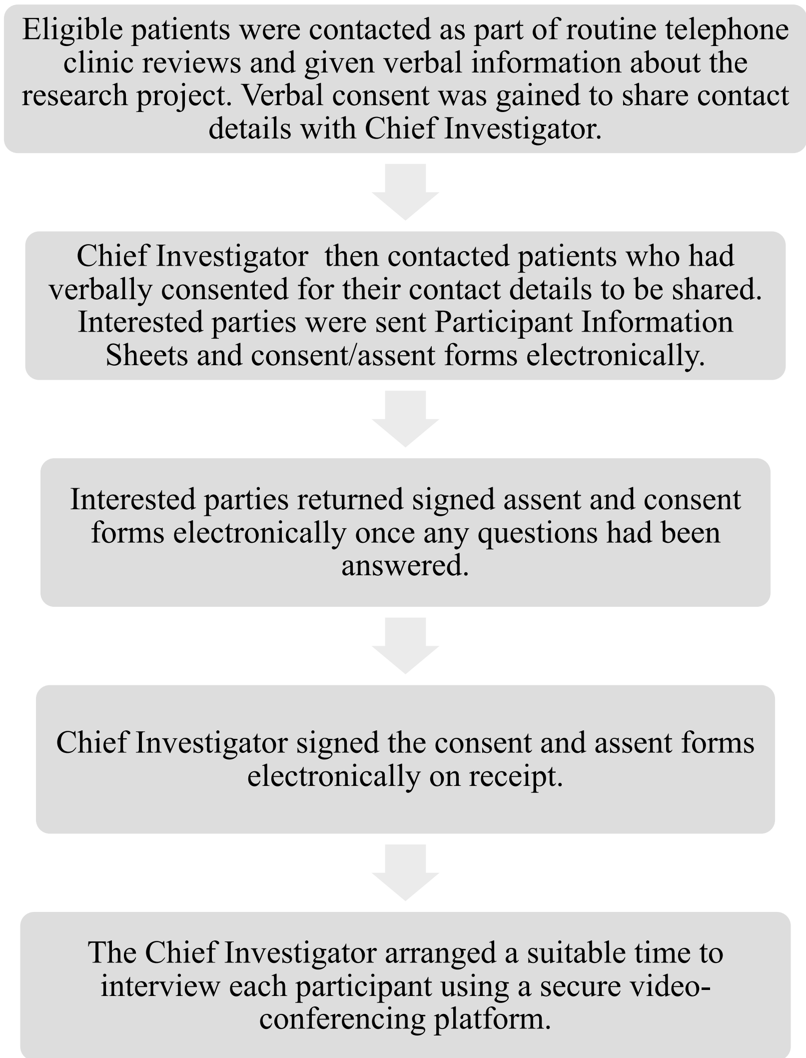

Figure 1 - A flow chart of participant recruitment

Once a potential participant had been identified, the clinician sought verbal consent to share their contact details with the CI. The CI then contacted the identified individuals and their parents (if under 16 years) by telephone to discuss the research further, answer any questions and talk through the Participant and Parent Information Sheets (Appendices C, D and E) that were sent to them electronically via email, Participant/Parent consent forms (Appendices F and G) and Assent forms (Appendix H). Parental consent and participant assent were required for participants under 16 years of age. Electronic consent was sought directly for participants who had reached 16 years of age. To allow time for potential participants to withdraw from the study, qualitative interviews were then arranged at least 48 hours after consent was received. At the point of interview, participants were assigned a pseudonym to maintain their anonymity and these are used throughout this report. Participants were informed they would have two weeks following the interview to withdraw consent and have their data removed. After this point, the analysis process would have commenced and it would not be possible to remove their data.

2.2.4 Sample

The overall sample of participants that were met the inclusion criteria and were approached was twenty-two. Of those approached by the Principal Investigator, fifteen agreed to be contacted by the Chief Investigator. A total of five participants (three males and two females) were recruited for participation in the research, aged between 13 and 16 years of age at the time of interview. Reasons for not wanting to take part included focus on academic work, exams, illness and family circumstances. Recruitment stopped at five as all eligible participants had been contacted and had either agreed or declined to take part in the research at the time they were approached. A sample size of five is deemed an appropriate sample size for an IPA project undertaken as a student, with between 3-6 suggested as sufficient for IPA research as part of training (Smith et al., 2009). In addition, IPA has been deemed appropriate for single case analysis and in small samples (4-8 participants) as long as extracts are included from at least half the participants (Smith, 2011). In the present research, care was taken to ensure that several participant quotes were included as evidence for each sub-theme to ensure that this standard was met. All five participants identified as White British and lived in the community whilst they received ongoing outpatient CF care. Routine care included clinic reviews with members of the CF team several times a year. See Table 8 below for sample demographics.

Table 8 - Participant Demographics

Participant Pseudonym	Gender	Age	Ethnicity	Age at diagnosis
Sarah	Female	15	White British	Newborn
Natalie	Female	16	White British	Newborn
Harry	Male	13	White British	Newborn
Aiden	Male	16	White British	Newborn
Daniel	Male	13	White British	Newborn

2.2.5 Data Collection and Interview Schedule

The data was qualitative and was collected through semi-structured interviews conducted by the researcher/CI. Semi-structured interviews were chosen as the method of data collection due to the fact that they allow the researcher to collect a rich source of data whilst maintaining confidentiality that would not be possible through a focus group (Smith, 2004). In addition, due to cross-infection control guidelines and the Covid-19 pandemic, all research activities including the interviews were conducted remotely via secure videoconferencing software.

The interview schedule was developed by the researcher/CI and reviewed by the Principle Investigator and academic supervisor, with the chosen methodology of IPA in mind. A semi-structured interview would ensure structure whilst being flexible to the chosen topics and areas of discussion dependent on each individual participant. It was important to ensure that questions were open ended to encourage participants to talk openly and in detail. The semi-structured interview schedule (Appendix I) was also reviewed by three individuals with CF who had received clinic outpatient care for CF but were unable to participate due to their age (e.g. being under 13 years or over 16 years at the time of recruitment).

The semi-structured interview schedule commenced with an open-ended question on current social circumstances, family and friendships. Specific social experiences of CF are then explored, such as cross-infection control guidelines advising against face-to-face contact with others with CF. An additional question was also added to explore the social experiences of adolescents with CF in the context of the Covid-19 pandemic. The structure of the interview was open and flexible to ensure that participants could share their overall social experiences of CF. Due to the pandemic, all interviews were conducted online using secure video-conferencing software and recorded onto an encrypted dictaphone prior to being transcribed verbatim by the researcher.

2.2.6 Data Analysis

The steps taken as part of the IPA analysis were based upon those outlined by Smith and colleagues (Smith et al. 2009).

1. Anonymised transcripts were read through on a number of occasions to enable the researcher to familiarise herself with the qualitative data in written form.
2. **Coding -** Initial exploratory comments were made and distinguished as being descriptive, *linguistic* or conceptual in nature through use of a key (Example shown in Appendix J)
3. **Emergent themes** were identified through examination of transcripts and patterns within the exploratory comments at the individual participant level.
4. **Emergent themes were organised into superordinate** and **subordinate themes** at the individual level (e.g. Appendix K). A summary paragraph was developed for each participant which gave an overview of their qualitative contribution to the dataset.
5. **Patterns and connections** across participant superordinate and subthemes were examined in the context of the dataset as a whole.
6. Similar superordinate and subthemes were merged to create overall superordinate and subthemes and corresponding quotes for the whole sample (Appendix L).
7. Themes were discussed and refined within the academic IPA support group and with the Principal Investigator, a clinician within the CF team.

Figure 11 - Summary of the Data Analysis Strategy (Based upon Smith et al., 2009)

The IPA analysis procedure outlined above allowed the researcher to make sense of the experiences of the adolescents interviewed and the emergent themes to become apparent over time. These themes were then reviewed in light of each individual transcripts and in the context of the whole dataset when final superordinate and sub-themes were determined. Firstly, the researcher familiarised herself with the individual transcripts by reading and re-reading prior to coding them with initial comments, separated at the descriptive, linguistic and conceptual level. Emergent superordinate and subordinate themes were then identified at the individual level and a summary paragraph was written for each participant. Patterns, connections and divergences were then explored across transcripts and emergent themes to develop superordinate and subordinate themes for the sample as a whole (Smith et al., 2009). As part of the analysis process, emergent themes were shared and discussed within an IPA support group with fellow researchers and academic supervisors as well as with the Principle Investigator, a Clinical Psychologist working within the CF team. This process allowed for themes to be shared and discussed, resulting in some slight amendments to theme titles. This discussion also allowed for exploration of the findings in the context of wider clinical implications explored within the discussion.

2.2.7 Quality Standards and Reflexivity

It was important to consider quality throughout this process to ensure that the resultant themes were reflective of the individual and group participant narratives. IPA was the chosen methodology as it focussed on the idiographic experiences of a purposive sample (Smith et al., 2009). For this reason, it is perhaps less relevant to consider reliability in the context that it would be applied to quantitative methods. Techniques such as member checking and inter-rater reliability assume that there is one common truth to be sought, regardless of researcher (Smith & McGannon, 2018). For qualitative research and in particular IPA, quality is arguably more appropriately appraised through consideration of general quality standards such as transparency and sensitivity to context (Smith and McGannon, 2018; Yardley, 2000).

As this research aimed to gain a sense of the social experiences of adolescents with CF, the approach taken demonstrates '*sensitivity to context'* which is one of the four principles outlined for assessing the quality of qualitative research (Yardley, 2000). A second principle, *'commitment and rigor'* was demonstrated through the collection of data from a homogenous sample of adolescents with CF. In addition, the process of analysis aimed to ensure that individual participant data was analysed in a systematic way throughout.

It was important for me, as the researcher, to engage in ongoing supervision both individually and through an IPA support group facilitated by academic staff at the University. This process is sometimes referred to as use of a '*critical friend'* within the literature and it encouraged ongoing reflexivity, through feedback and refinement of initial themes following discussion with fellow researchers (Smith & Macgannon, 2018). This allowed me, as researcher, to reflect upon the process and receive feedback on my initial exploratory comments and emergent themes through consultation with peers and research supervisors. This process allowed me to share transcripts with other researchers and peers, to discuss their thoughts and interpretations of the data in the context of my initial exploratory comments and themes. Linked to this is the third quality principle of qualitative research described by Yardley of '*Transparency and Coherence*'. This principle has been demonstrated thorough the use of participant quotes when discussing themes, alongside examples from stages of the analysis process included in the appendices (Yardley, 2000).

Due to the *double hermeneutic*, the role of the researcher within IPA is an integral part of the data analysis process and therefore it is important to briefly outline my earlier experiences and subjective position. Prior to undertaking this research, I had not previously known or worked with anyone with CF. However, I have previously worked in a health psychology setting with children who have various physical health conditions and I am interested in child and adolescent mental health. I am interested in empowering others to share their lived experience and for this reason, phenomenological methodologies suit my subjective position as researcher. I am interested more generally in the social factors involved in physical and mental health and wellbeing. As part of the research process, I undertook some wider reading around CF, more generally to familiarise myself with the condition. However, I was mindful not to seek out personal experiences of CF prior to the undertaking of this research as I wanted to ensure that the analysis process was grounded within my interpretation of the participant interviews. I am interested more widely in idiographic approaches, engagement and collaboration with patients and service users in the context of improving the patient experience.

IPA as an approach does not seek to analyse data objectively and independently, instead it involves the researcher within the interpretative process (Smith et al., 2009). Throughout the process, I kept a reflective research diary to record initial thoughts, sense-making and reflections which I later consulted during the data-analysis phase. Referring back to the diary at different points allowed me, as researcher, to explore how my own experiences may have impacted upon the interpretations made. Use of the reflective research diary allowed me to bracket off my initial thoughts and feelings whilst undertaking the qualitative interviews and it helped me to make sense of the participants' experiences within my own subjective position. As part of this process, it was important to reflect on some processes and elements of the research that I found challenging, such as the shift in role, from therapist to researcher. I found supervision and my reflective diary useful when exploring this shift, especially when participants disclosed difficult experiences, such as being bullied.

2.3 Analysis and Results

2.3.1 Summary of Results

The five participants described their experiences of Cystic Fibrosis (CF) in relation to their social world, including relationships with family, friends and wider systems such as healthcare settings and school. They shared experiences of similarity and difference in the context of evolving peer relationships and a strive to be understood and belong. Within these discussions was a common experience of the burden of disclosure of CF and a preference for familiarity, comfort and safety within social networks. Finding common ground with others was seen as important for the adolescents, and for some, there was a preference to be treated in the same way as their peers or connect through shared interests. For others, they shared a preference of building relationships with peers who have CF or another health condition with shared experiences.

A further key theme focussed upon the process of increased independence, reflections and the mixed emotions experienced of increased CF treatment autonomy. During adolescence, CF patients are encouraged to increase their independence with treatment adherence and more generally in preparation for transition into adult clinical care (Lapp & Chase., 2018).

Development of autonomy in the context of CF treatment and associated feelings were explored in the context of family relationships and the development of sense of self. A period of defiance and conflict followed by acceptance and gratitude was experienced by some of the participants. The experience of increased independence was associated with varied emotions, with some sharing feelings of anxiety and fear and others explaining that they felt happier due to a perceived reduction in burden on family members.

The importance of practical and emotional support, trust and understanding were noted throughout, in the context of both being understood and in light of increased independence. Reflections were made considering the Covid-19 pandemic and the overall value placed on human connection, including the navigation of cross-infection control guidelines. Participants shared how they often connect to others in alternative ways and this has increased further due to the pandemic. *Table* 9 below summarises the two superordinate and six subordinate themes alongside pseudonyms of participants that contributed to each subtheme. A full breakdown of superordinate themes, subthemes and associated quotes can be found in Appendix K.

Table 9 - Summary of Superordinate and Subordinate Themes with Descriptions and Participant Pseudonyms

Superordinate Theme	**Sub-Themes**	**Participants who contributed (Pseudonyms)**
A desire to belong and be understood as a young person and as someone with Cystic Fibrosis	**Not wanting to be treated differently because of CF** - *Desire to be treated like everyone else* - *Not wanting to be seen or treated as "different"* - *Associated emotions such as embarrassment, sadness and fear.*	Sarah Aiden Daniel Harry Natalie
	The Burden of disclosure and the importance of trust - *Difficulty explaining CF to those that are unfamiliar* - *Reluctance to disclose for fear of reaction* - *Avoidance of making new friends, keeping familiar* - *Importance of trust and trust being broken following disclosure*	Sarah Aiden Daniel Harry Natalie
	Finding common ground - Connecting with others through shared experiences - e.g. those with other health conditions. - Connecting to others through shared interests e.g. Gaming, YouTube, sports. - Connecting with others with CF who understand what it is like to live with CF. - Sharing experiences and desire to support others with CF.	Sarah Aiden Daniel Harry Natalie
	Connecting in Other Ways - Finding alternative ways to connect with others (e.g. online, social media, video calls) - Merging shared interests with connection, e.g. talking to friends whilst playing computer games) - Impact of cross-infection control guidelines and the Covid-19 pandemic.	Sarah Aiden Daniel Harry Natalie
The relationship between increasing independence and social relationships in the context of CF.	**Experiences of Defiance and Conflict with Others (e.g. family)** - *Conflict, disagreements within family* - *Defiance, 'fight' with CF* - *Refusal to adhere to treatment, cross-infection control guidelines* - *Conflict within self – links to life expectancy and motivation*	Sarah Natalie Aiden

Independence as a process - Reflections and mixed emotions.	
- *Acceptance. Being 'at peace' and not resisting any longer, pressure off.* - *Awareness that certain things cannot be changed* - *Happy as feel less of a burden on family* - *Gratitude for the support of others*	Sarah Harry
In contrast: Alternative emotions of fear and anxiety about increased independence and transition to adult CF service	Natalie

2.3.2 Superordinate Theme: A desire to belong and be understood as a young person and as someone with Cystic Fibrosis

All five participants discussed peer relationships within the context of CF, including comparisons with peers and an overall desire to belong. Daniel, for example, spoke of not wanting to be treated differently to his peers: *"I dunno.. it's just kind of like… unnecessary if you're kind of treated different… cause you don't need any more help"* (Line 114-115*).* Here, Daniel describes wanting to ensure that his CF does not indicate to others that he requires additional support from others. Throughout the discussions, participants referenced social experiences whereby others would hold different levels of understanding about their CF.

There also appeared some hesitancy and emphasis of language that perhaps reflected the participants desire to be understood within the interviews themselves. Evidence from the transcripts were noted when participants were describing their experiences socially and of CF. Sarah, for example, when asked about her friendship network, said *"… it's really hard to describe my friends…"* (Line 28*),* later saying:

"*I sort of… [sigh] this is really hard to put into words. Umm, sort of living with CF, it's not been an enjoyable experience [laughs], I won't say that, but it's been… OK.*" (Lines 443-444).

Overall, this superordinate theme describes the participants' social experiences with focus on understanding and belonging. These experiences have been grouped within the following four sub-themes: *Not wanting to be treated differently because of CF*, *The Burden of Disclosure and the Importance of Trust in the context of CF*, *Finding Common Ground* and *Connecting in Other Ways*.

2.3.3 "You just don't really want to stand out" - Not wanting to be treated differently because of CF.

All five participants noted a desire or preference for being treated the same as their peers and not wanting others to treat them differently or judge them because of their CF. Natalie reflected upon her experiences of peer relationships at school:

"Some people treat me differently but then others didn't so it was ok… So I just left it… and then people just treat me the same… how everyone else would be treated." (Line 207-210).

Participants described experiences of wanting to fit in with peers in the context of their CF and explained why they preferred to be treated the same. Daniel elaborated on why this is important for him and what it may feel like to be treated differently because of his CF:

"…it would just make me kind of stand out more and being a bit embarrassing… you just don't really want to stand out." (Line 121-124).

Natalie emphasised the importance of this preference through sharing specific experiences where she felt she had been treated differently by peers because of her CF:

*"The adults didn't really care… but like when I went for a sleepover, I would have to take all my meds and stuff… [long pause] and they'd just look at me really weirdly… for everything I did" (*Line 219-221).

Here, Natalie described a time where she felt her peers were treating her differently because they had seen her adhere to her daily CF medication outside of school at a sleepover. Similarly, when discussing family dynamics and living with CF, Aiden spoke of a preference for his parents to treat him in the same way as his siblings, who do not have CF:

"…they [family] don't like sympathise me…they just treat me like the others... like a normal person…". (Line 95-96).

Here, Aiden talks about his experiences of normalcy within his family, suggesting that being treated different to others because of his CF may contribute to feelings of not being normal for Aiden. This is something he later elaborated on when discussing his experiences of being bullied at school. This is elaborated on in the subtheme '*The Burden of Disclosing CF and the Importance of Trust'.* All five participants shared experiences of their CF in relation to the development of peer relationships and family dynamics and noted a preference to being treated the same as their peers to avoid emotions such as embarrassment or experiences of judgement. When Harry discussed his friendship group at school he noted:

"I do feel more comfortable around my close friend group because I know that they're not going to judge me over it [CF]… And it's much nicer to know that they're not kind of thinking different of me and not like kind of trying to like push me out of the group just because of the symptoms and stuff..." (Lines 332-337).

Here, Harry spoke about the importance of knowing that he is not going to be judged by others and hints at fears he held around being left pushed out of social groups because of visible CF symptoms. Similarly, when Sarah discussed her experience of the relatively new gene therapy, she described the impact on her visible symptoms:

"…like I don't get afraid of shaking people's hands because they're not sweaty anymore, so I can sort of, everything's difficult, but it used to show and it doesn't…"

(Lines 415-418).

In the quote above, Sarah reflects upon the differences she has noticed socially since she started gene therapy treatment and this further highlights the considerations those with CF have to make in social situations for fear of being seen as different.

In contrast to the other four participants, Aiden shared an experience of being bullied at school and he believed this was directly linked to his CF and being perceived as different to his peers. He explained that the bullying was focussed on elements of CF in particular, such as the associated reduction in life expectancy:

"... cause it was sort of a… it was sort of a thing that they attacked every single time… like for instance one of the things they'd say… is "remind me to come to your funeral when you're thirty"... so when you explain they know and that's... that is one of the easiest things to attack with it... is the age that you die at... so yeah it's... it was erm… for an unmatured mind it was very hard to deal with." (Line 159-170).

Here, Aiden described how he has made sense of this period of his adolescence and reflected upon the shift in his maturity level, as well as the difficulty of the experience. He later explained that following this experience, he sought human connection and belonging elsewhere by joining a gang for a short period of time:

"*I'd latch on to any bit of human connection that actually cared about me that I could…and then I was like "Oh, can I join your gang?'"* (Line 324-326).

We can understand how important belonging, human connection and the feeling of being understood were for Aiden and how a gang could be perceived to offer this to him. More broadly, we can see how the importance of belonging appears to be for participants, and the concern that standing out, or not being seen as 'normal' may threaten this. The second linked sub-theme covers the difficulties that participants described when they felt the need to tell others about their CF. Bearing in mind the present theme, we can begin to understand how important and potentially risky such disclosures may be for young people.

2.3.4 The Burden of Disclosure and the Importance of Trust in the Context of CF

Linked to the sub-theme of not wanting to be seen as different, several participants spoke about the difficulty that they had experienced during CF disclosure. This difficulty is heightened at times as some symptoms of CF are not acutely visible to others. The challenges of people knowing very little about the condition seemed to be prominent in this experience alongside the importance of trust. Harry spoke about the considerations and feelings he experienced prior to disclosing CF to his friendship group at secondary school:

"I was pretty scared about it 'cause I didn't know how they'd react but, erm, but as soon as I did it they were all really accepting about it and they, err, were like 'oh yeah that's fine, I don't mind about this kind of stuff that you have and it doesn't really matter'".
(Lines 404-406).

This experience provides insight into some of the considerations and concerns that the participants contemplate when forming new friendships. These considerations may include how they think others may respond to disclosure, fear of judgement or misunderstanding and how and when it is best to disclose. Harry went on to describe the emotional and physical sense of relief following disclosure of his CF to his peers, once he realised they were going to be supportive:

"… kind of all of that worry and stuff.. kind of left my body because I was realised that these are really nice people that I'm hanging out with stuff… so yeah, it's pretty nice" (Line 412-414).

Similarly, Natalie explained "*It was hard to like, describe it [CF] to people…'cause they wouldn't really understand..." (Line. 154-155).* For Natalie, it was important to have a close friend who understood and supported her through the process of disclosing CF to others:

"So when I was young young… erm… I used to have a friend, I'm still friends with her, we've like, we've been best friends for ages, but umm… she like straight away even though we were young figured it [CF] out … and then once we got older she like knew what I went through…if I didn't want to explain, she would have basically done it for me." (Lines 163-168).

Here, Natalie described how her friend helped her to explain CF to others, suggesting that she realised her friend was aware of the difficulties of disclosure. In a similar way, Sarah described experiences of making new friends at school and the experience of feeling as though she needed to explain herself:

"When I made new friends I sort of had to explain it…… like the little things, like why I can skip the queue at lunch, why I'm taking these meds now and then…so it was quite... weird… I just sort of avoided making new friends just for the ease of keeping the old ones" (Lines 126-130)

Sarah begins to elaborate how the burden of disclosure has affected her social network as she spoke of the conscious decision to keep friends who already know about her CF to avoid new disclosures. This appeared to have had an impact on the friendships the participants made and maintained through childhood and adolescence including the transition to secondary school. Sarah reflected upon her friendship network at school and made sense of this in the context of a preference for familiarity and avoidance of new disclosure:

"I think it was easier to keep my friends that I've known forever than make new ones because they of course knew about it [CF] because I went to primary school with them." (Line 132-134).

Here we can see how explaining elements of CF to new friends felt strange for Sarah, and as such, she preferred to keep her established and familiar friendship group. Similarly, when talking about his closer group of friends Daniel described *"…they've kind of just known [about his CF] for a really long time..."* (Line 55). Linked to considerations made linked to CF disclosure, Harry emphasised the importance of trust in his family and friends:

"… the trust is like very important…for like Cystic fibrosis because if you don't trust people then you wouldn't be able to do any of the stuff you need to do with cystic fibrosis… because

like you need help to get your meds and stuff… and make them understand what you're going through and… for them to help you.." (Lines 165-173).

Here, Harry shared his thoughts around the importance of placing trust in others as he has had to rely on them for support with his CF. Aiden also spoke about the importance of trust in the context of regrets he held around disclosing his CF to others at school and the subsequent experience of bullying:

"At school I didn't really tell anyone that I had CF because… looking back on things now I wish I hadn't told nobody but I told a few people… and then it was sort of used against me in a sly way and then I ended up coming out of school because of it..." (Lines 111-116).

We can see here how Aiden reflects upon negative experiences he has had in relation to disclosure and how this may relate to the trust he places in others. He shared how he initially told one friend, who then told others:

"I told them in… year 7. But I only told my best mate and then he told them…and I was like 'oh yeah.. 'cheers for that thanks'… (Lines 265-267).

Aiden made sense of the bullying that he experienced as the trust being broken following disclosure to a peer. Although the other participants also shared a desire to fit in and worries around disclosure, Aiden was the only participant to describe direct experiences of being victimised due to his CF following disclosure. He later described how this experience was linked to the trust he now places in his social network:

"…it puts you in a bit of an untrustworthy situation with everyone so you don't want to tell them anything because it could... it's that easy to throw it back in your face."
(Lines 117-119).

Overall, this subtheme covers the considerations that the adolescents have to make when developing new friendships, how much information they disclose about their CF and the possible consequences of disclosure. This is intricately linked to the previous subtheme of a desire to not be seen as 'different' to peers. Several of the participants shared experiences of the importance of finding common ground with others which is explored in the following sub-theme.

2.3.5 Finding Common Ground

The third sub-theme explores the sense-making and experiences that related to finding common ground with others. All five participants described how they connected with others through shared interests, hobbies or experiences. For some participants, such as Natalie, she described her connections as linked to her interests in streaming platforms such as YouTube and Twitch. Sarah spoke of finding common ground with peers in school who had experience of other health conditions, such as allergies:

> *One [close friend] has a nut allergy… So we've always sort of bonded over being close and knowing somebody who is slightly… not different but has an addition, like something that is additional alongside it. I think it sort of… it sounds stupid but it sort of helped our friendship."* (Lines 48-52).

Similarly, Harry described friendships that he had made through a sports club for those with long-term conditions and described the sense of safety he experienced when in attendance:

> *"…it makes me feel more safe around everyone because I know that I'm not the only one going through something and I'm not the only one that has… that I'm not perfect and everyone else there is very accepting of all that kind of stuff and it makes me… it's like a very safe environment for me to like talk about stuff".* (Line 450-454).

Here, Harry describes feeling accepted and safe in the company of others who have some shared experience with him through other long-term health conditions. For Harry, this shared experience is associated with feelings of comfort and an ability to talk openly with others. For both Harry and Sarah, it was not always a desire to connect with someone else who has CF, rather it was an element of shared experience in relation to a different health condition. Aiden described how meeting others with CF made a difference due to the common experiences, ability to relate and understand:

"... talking to other people is hard… like when they haven't been through that sort of stuff… they don't know about it… so speaking to someone who goes through the same as you... it's almost like... yeah, you're gonna relate to everything I'm going to tell you."
(Lines 774-778).

Similarly, Sarah spoke in detail about the close bond she has with a friend who has CF, despite not being able to meet in person due to cross-infection control guidelines:

"I think having someone there who completely gets it, is so worth it… because having somebody you can speak to, who will 100% get it, is so rare."
(Lines 266-268).

Sarah later referred to the bond with her friend who also has CF as *"irreplaceable"* (Line 297) and initially explained how unusual it was to have met someone else with CF:

"*It's really weird to actually talk to somebody who completely gets it because when I found out about the gene therapy stuff was coming through like it was legalised... I didn't text my friends, I didn't tell any of my family, the first person I texted was him."* (Lines 87-92).

This illustrates the contrast that participants make when reflecting upon their relationships with others and the different levels and types of support and understanding that others can provide. For Sarah, the level of understanding her friend with CF provides is different to the friends and family who do not have lived experience of CF.

For each participant, the importance of finding common ground was reflected in different ways. For Sarah, she spoke of friendships with others who had direct experience of CF or other health conditions. Harry connected with others who had also had long-term conditions and Aiden and Natalie spoke of connecting with others who have CF alongside connections to peers through online gaming and streaming platforms. For the youngest participant, Daniel, he spoke of his friends tending to be those he had met through school and a preference to being treated similarly to his peers. As Daniel had recently turned 13 at the time of interview, he had yet to experience meeting others with CF although shared a desire to be seen of as similar to his peers in school. When discussing the cross-infection control guidelines that prevent CF patients from meeting each other, Daniel explained that they had not affected him personally:

"I dunno… It's not really bothered me much because I've not really known anyone much that I've kind of liked... that I've wanted to meet in person."
(Line 201-202).

For Daniel, he described a preference for not wanting to be seen as different to peers and described finding common ground in other ways, such as through playing computer games and sports. Within the sub-theme of *Finding Common Ground*, it was also noted that several of the participants shared a desire to support others with CF and for some this appeared to be an important factor when deciding to participate in the research. For example, towards the end of her interview, Sarah said:

***"**Yeah… well thank you for the offer, because it's been very, very nice to know I'm sort of helping out with something…" (Lines* 868-870)

Overall, this subtheme highlights the importance of finding common ground with others and this links to the next subtheme that covers the experiences of connecting to others in alternative ways.

2.3.7 Connecting in Other Ways

All five of the participants shared experiences of connecting to others, extended family and friends in alternative ways to traditional face-to-face communication. This appears to have been heightened due to the Covid-19 lockdown and subsequent 'shielding' status given to the participants during the first lockdown in March 2020. This is in addition to the already in place cross-infection control guidelines that advise against individuals with CF from meeting each other in person. For Natalie, she shared experiences of connecting to friends through streaming platforms:

"...one of my friends loves Minecraft and all the same YouTubers such as [name of Youtube streamer]... and she got me into it and now we just discuss it every time they go live."
(Lines 494-496)

Similarly, Daniel shared that he connects with others using other means and how the Covid-19 pandemic has led to these methods of communication being used more regularly:

"...sometimes I find myself playing games with them a lot more since the pandemic but obviously you can't go round their houses and stuff."
(Lines 268-269)

All five adolescents discussed how social networking applications had helped them to connect with others. Harry reflected on the shift that had to be made to the increased reliance online communication with peers in the past year:

"...*we've found other ways to speak to each other and we all have group chats... on WhatsApp, Xbox and stuff like that.." (Lines 574-578)*

Similarly, Aiden spoke about the use of social media and videocalls to maintain human connection with peers and his experience of spending lots of time at home:

"I didn't go out at all but like now I don't go out... erm and like we message a lot but like being in lockdown we've been talking on Xbox a lot more..."
(Line 562-565).

Aiden also shared that he prefers to connect with specific friends in different ways and how these alternative ways to connect helped him through a period of depression which he referred to as the "*black hole*":

- "... like FaceTime and that I don't do it with my boy mates... I tend to do it with my girl mates 'cause they do it more... so sometimes of a night time I'll just go on FaceTime to someone...and then just talk for a bit... sometimes fall asleep on the phone... sometimes not [smiles]. But yeah... stuff like that helps me to keep in touch like... with not slipping back into that black hole..."
(Lines 650-656)

As well as connecting with peers in school and family, social media appeared to have helped the adolescents to navigate the cross-infection control guidelines and build relationships with others who have CF. Natalie described the experience of joining and developing a group online with other adolescents who have CF:

> *"I was a bit scared at first but we started off erm... we started off on Facebook and then it was on Messenger and then me and this girl I used to be friends with said "oh my god we should put it on Snapchat!" 'cause everyone uses Snapchat a lot…so me and her discussed and then put it on Snapchat…and now we're all talking on there."*
> (Lines 320-326).

Here, Natalie describes her initial apprehension about joining a group for adolescents with CF who could not meet in person due to infection control guidelines. Despite this initial fear, she spoke with a friend that she had already met and together they made the decision to share the CF group to an alternative platform that "*everyone uses*". As a result, the group has grown and everyone has been able to connect online despite the face-to-face contact restrictions.

In summary, this superordinate theme has focussed upon on a desire to belong and be understood in the context of CF. Participants shared similar experiences regarding the burden of disclosure, the importance of trusting others and finding common ground with others in various ways. For some, this was a close bond with someone CF who, as Sarah described: *"completely gets it*" (Line 266). For others, finding common ground in alternative ways has been preferred.

2.3.8 Superordinate Theme: The Relationship Between Increasing Independence and Social Relationships in the Context of CF.

When discussing social relationships, all five participants discussed increased independence and reliance on others for support with regards to their CF treatment. At the time of interview, the participants' ages ranged from 13 years (Daniel) to 16 and a half years (Aiden). Harry, who was almost 14 years at the time of his interview, described how he had wanted to gain more treatment autonomy due to the impact he felt it would have upon his family:

"I can get it all set up and do it all by myself and put it all away and wash it up… so it's much easier on my parents to kind of… they don't need to be there all the time to get me to do my physio properly 'cause I can just do it by myself now."
(Lines 268-272).

The three participants who were due to transition to adult services within the next eighteen months, Sarah, Natalie and Aiden, described different stages of this process in relation to gaining increased levels of independence and subsequent treatment autonomy. The social impact of increased independence will be discussed within the following two sub-themes: '*Experiences of defiance and conflict with others in relation to increased independence'* and '*Independence as a process - Reflections and mixed emotions'*.. For some, like Sarah, conflict was with her parents and for other participants such as Aiden, this conflict was with himself as he struggled to motivate himself to engage in treatment independently.

2.3.9 Experiences of Defiance and Conflict in Relation to Increased Independence

Three participants, Natalie, Sarah and Aiden, reflected upon experiences of defiance as linked to increased independence in relation to treatment adherence, motivation and the family relationships. Sarah reflected upon the conflict within the family as she initially refused to adhere to her treatment when she first reached adolescence:

"...yeah I just used to hate everything… I'd sort of gone to sort of rejecting it… instead of sort of accepting everything I just have to do. I was just like… I wouldn't do it properly… I'd just sort of be very defiant… and just like 'I'm not gonna do this'. I used to have so many arguments with my parents about it… I used to sort of scream at them like 'This is your fault!', 'I'm not doing it, I don't care'."
(Lines 340-349).

Sarah later described how difficult she had initially found the cross-infection control guidelines that prevent face to face contact with others who have CF:

"*… erm I remember really detesting it [cross infection control guidelines]. I remember being like… eight or nine and hating the idea that I couldn't meet somebody like me or get on with somebody who has the same things as me."*
(Lines 503-506).

This experience was prior to Sarah having the ability to connect with others using alternative means, such as social media. Natalie similarly shared experiences of defiance when faced with the cross-infection control guidelines during her hospital admissions:

"So we used to always... we wasn't meant to but we used to always say hi to each other…we wasn't actually meant to go by each other but we just broke the rules a lot… because we didn't actually care at the time… but then we [laughs] we just kept getting told off"
(Lines 263-267).

Here, Natalie describes how this defiance was linked to her desire to connect with others who have CF, despite the potential consequences to her health. In the context of CF, such decisions could lead to increased susceptibility to lung infections which can affect overall lung functioning (Rowbotham et al., 2019; Saiman & Siegel, 2003). However, the period of adolescence is also likely to be the first time that patients begin to make independent decisions regarding medication and treatment adherence. Aiden spoke about coming to terms with the fact he could now choose to not adhere to his treatment plan:

"...it can be very easy to just open to top of the meds, tip the liquid away, not do that. And crack the tablets open and not inhale them, with the nebuliser and the physio…"
(Lines 639-642).

For Aiden, the conflict that he experienced, was not with parents or healthcare staff but within himself. He shared difficulties around motivating himself to adhere to treatment and appeared to make sense of this difficulty through consideration of the shortened life expectancy associated with CF:

"You have to look at the... it can be quite a vicious cycle because you have to look at the bigger picture without looking too far into it because it's like… I aint gonna live that long anyway so what's the point?... There has to be like… limits to where you see… but then to wanna get the motivation to do it, you have to look into the future…"
(Lines 687-698)

In summary, gaining increased treatment autonomy is a reality for adolescents with CF as they are encouraged to prepare for the transition to adult services during this period. For some, they experience conflict with their caregivers and/or clinical team when they begin to make independent decisions about their health. For others, such as Aiden, this is experienced as inner conflict and linked to difficulties with motivation and the awareness of having a shortened life expectancy.

2.3.9b Independence as a Process - Reflections and Mixed Emotions.

Although initially, increasing independence is sometimes associated with periods of conflict, several of the participants reflected upon subsequent experiences of acceptance and feelings of gratitude for others. Sarah described the shift from resistance to acceptance:

"*…But I just sort of realised that there's no point resisting it, 'cause I'm gonna have to do it at some point, I feel like I've just lost the energy to fight with it.*"
(Lines 381-384)

"…its not a fun thing to live with, I just sort of, I've just stopped resisting it. I'm like Ok, it's just me. It's just something I have to just deal with. The less that I think about the impacts, the smaller it feels…. I sort of.. [sigh] this is really hard to put into words. Umm, sort of living with CF, it's not been an enjoyable experience [laughs], I won't say that, but it's been… Ok." (Lines 439-444)

Here, Sarah refers to an earlier period of defiance and fighting with CF when in early adolescence, before coming to the realisation that CF is something she cannot change. In light of increased independence and acceptance of CF, participants spoke of their gratitude for the support of others. The youngest of the participants, Daniel spoke about the close bond he had developed with his nan and the support she had given with his CF:

Erm... I'm probably closer with my nan that lives by the school… she looked after me after school and stuff… 'cause like mum was the oldest on that side and ever since I was a baby I've just always gone round to her house and that…She's like been looking after me for years so she's been doing all my meds and stuff..."
(Lines 277-287)

Harry spoke about the positive impact that he feels his increased autonomy and independence has had on his family:

"… It's felt pretty good… because I'm more independent, everyone's less stressed about… kind of the interactions and like the stuff that I'm needing to do and everyone's getting more time to themselves…It makes me feel much happier knowing that everyone's happier..."
(Line 281-285).

Here, Harry linked the happiness he felt with increased independence to the positive impact he felt it would have upon his family, including his parents and brother:

"…I can do my own physio by myself and make up my [medication name] and I can get it all set up and do it all by myself and put it all away and wash it up… so it's much easier on my parents… they don't need to be there all the time to get me to do my physio properly cause I can just do it by myself now… and it's much easier on [brother's name] as well… he can have like more time with my parents and stuff because… they're not needing to be there next to me…" (Lines 268-275).

He also reflected upon a sense of gratitude for his circumstances and the support he has had from family in the context of his CF:

"…the mental side of things… it's just made it much easier for me… having a nice family and… having a nice house… and a nice environment… with people that I can trust…"
(Lines 159-161).

In contrast to Harry, Natalie spoke about feelings of unfamiliarity and anxiety when she has been encouraged to prepare for the transition to adult CF care. When asked how Natalie felt to be sixteen she said:

"Weird… cause I've got to do everything myself… I've gotta call like... nurses on my own.... well I was gonna say book appointments but my mum does that because I'm too scared to even do that [laughs]. Erm... the other day she [mum] made me do my prescription…"
(Lined 12-16)

Here, Natalie described the elements of her CF care that she is now expected to do herself in preparation for the transition to adult services. She shared experiences of her mother and the CF team encouraging her throughout this process and the anxiety that she has experienced with these unfamiliar and new experiences. Later, Natalie continued to discuss this reluctance and shared that she felt the adult CF team also did not want her to move on:

"I'm always on that ward but they like me too much to want me to actually… so I just go there…they don't want me... umm because I'd have to go to adults and they don't want me to leave…'cause I've been there for ages…"
(Line 356-361)

Natalie appeared to have made sense of the situation as being something outside of her control although also she has considered that the CF team may also find it difficult for her to transition to adult services.

In summary, this theme covers the varied experiences that participants have had with increased treatment autonomy and independence in adolescence and the relationships with family and friends. These experiences involved mixed emotions such as hesitancy, anxiety and defiance to later feelings of acceptance and gratitude for others. Some participants shared an awareness of the impact that increased independence would likely have upon their family and for this reason, they felt that it was important to work towards treatment autonomy. For others, change and unfamiliarity were to be avoided. These discussions perhaps also reflected the participants' overall shift in social identity and their development of sense of self in the context of adolescence and CF.

2.4 Discussion

2.4.1 Summary of Findings

The aim of this research was to explore the social experiences and sense-making of adolescents with Cystic Fibrosis. Semi-structured interviews with five adolescents, three males and two females, explored their social experiences in the context of their CF. Participant ages ranged from 13 to 16 years and all five participants identified as White British. The two superordinate themes that encompassed the experiences of the five participants related to a desire for belonging and understanding in the context of CF and the emotional and social impact of gaining increased levels of independence with CF. Of particular focus is the desire to belong and be understood in the context of CF and this can be understood more widely within the adolescent social identity development literature.

2.4.2 Findings in Context of the Wider Literature

The stage of adolescence is often considered to be of key importance with regards to developing peer relationships alongside identity development in relation to others (Albarello et al., 2017; Ragelienė, 2016; Tanti et al., 2011). As Erikson described, adolescence is often a period of preoccupation with the question *"Who am I?",* with some resolution sought throughout this stage as individuals develop their sense of self in relation to other and wider society (Erikson, 1968; Sharma & Sharma, 2010). Tajfel and Turner's 'Social Identity Theory', suggests that of key importance during the development of sense of self is the perceived belonging to a social group (e.g. Tajfel, 1982). Alongside these developments is ongoing physical, cognitive and emotional maturation associated with the period of adolescence and early adulthood.

The present research explored the increased independence and experiences of conflict and defiance within families and within the self in adolescents with CF. The identification of this common theme across participants bears similarity to a literature review exploring parent-adolescent relationships. This review was not focussed upon physical health but highlighted that such conflicts may serve as a means to negotiate relational changes as such relationships become increasingly egalitarian (Branje, 2018). The review also noted that adolescents may determine that certain issues can be resolved independently as opposed to being under the direction of parents. In the context of this research and adolescents with CF, this theory could be applied to some of the conflict initially experienced within the family as adolescents make their first treatment decisions independently.

Alongside a desire to belong, the period of adolescence is often associated with increased autonomy and the present findings are comparable to qualitative explorations of adolescents with other health conditions, such as epilepsy (Chew et al., 2019). Such research has noted that condition specific impairments may serve as a barrier to social participation. Authors also highlighted the protective factors of social support from family and friends which was also noted within the present research (Chew et al., 2019).

Further qualitative research with young adults with various chronic health conditions found that disclosure in particular may be influenced by several factors including condition visibility, anticipated reactions in others and trust (Kaushansky et al., 2017). These findings are similar to the present research, with participants emphasising the importance of trust through disclosure. Several participants also expressed a desire to be seen and treated similarly to peers and discussed worries about being seen as “different” because of symptoms such as sweaty palms.

For adolescents with a chronic and life-shortening condition, such as CF, they additionally have to consider how the condition fits within their sense of self and in relation to their peers who do not have CF. Research has indicated that social identification with others may partly predict health outcomes via moderators such as self-efficacy (Cameron et al., 2018). The experience of CF may be somewhat different to other physical health conditions, in that it is often diagnosed soon after birth. During her interview, Sarah shared that she was grateful for this element of CF and compared it to other conditions that may be diagnosed later in life:

"…the thing that I'm probably most grateful for, that is a kind of weird thing to be grateful for, is that I've always known it was there, I've always had it…"
(Lines 510-512).

The present findings also add to the literature base of previous, more general, qualitative explorations of young people with CF. An earlier IPA of 31 interviews with young people exploring overall subjective experiences highlighted that young people wanted to be seen of as 'normal' in relation to their peers. The authors also noted that this sometimes was in direct conflict with elements of their recommenced treatment which was also noted in the present research (Badlan, 2006). For example, Natalie described how peers would "*look at me really weirdly*" (Line 221*)* when taking her CF medication at a sleepover.

International qualitative research undertaken in Canada that employed thematic analysis highlighted feelings of loneliness and social isolation alongside use of social media to connect with others (Moola et al., 2018). In the current research, feelings of loneliness and isolation were only discussed by one participant (Aiden) and the majority of participants felt connected to others and had established close friendships both in education settings and online. The current findings also support a grounded theory analysis undertaken in the United States (Toth et al., 2016). In this study, researchers found common experiences of supported autonomy and impact on relationships with siblings, which bear a resemblance to the superordinate theme in the current research related to the social impact of increased independence.

The current findings can also be viewed in context of the wider CF literature, including a systematic review that focussed on the importance of the transition from Child to Adult CF care (Singh et al., 2020). This review described the transition as the "*perfect storm*" due to the concurrent neurodevelopmental and social shifts that are associated with the stage of adolescence.

2.4.3 Clinical Implications and Dissemination

The present research aimed to explore the adolescent social experiences of individuals with CF who were receiving care through an outpatient clinic. Such research had yet to be undertaken and it is hoped that the findings will be helpful for clinicians working within such settings when supporting adolescents and preparing them for the transition to adult CF care.

As part of the analysis, discussion with the Principle Investigator, a clinician in the CF clinic, provided the opportunity to discuss, review and refine the themes in the specific clinical context. Corroboration with the Principle Investigator also allowed for discussion of the results in terms of possible clinical implications for patients and CF clinical teams. Such corroboration is deemed an important part of the IPA process with regards to transparency, coherence and rigor (Smith et al., 2009; Smith & Macgannon, 2018).

It is hoped that the identified themes, wider results and clinical implications can be disseminated to the CF team to ensure that positive changes can be made in light of the findings to improve overall experiences of adolescents with CF. There are possible clinical implications with regards to transition planning for adolescents with CF. Specifically, the finding that adolescents often have different views of increased independence in the context of their CF. It is important for individualised care plans to be developed, taking these individual experiences into consideration. For example, some CF patients may require more intense support and guidance when preparing to take their own medication. Others may be keen to increase their independence and such factors also need to be considered within the context of each patient's family and systemic factors such as the complexity of their treatment regime.

The present findings have highlighted that some adolescents may experience challenging social experiences such as alienation from peers or bullying directly linked to their CF. It is hoped that increased awareness of such social experiences of adolescents with CF will help CF teams provide individualised care and additional support for those identified as affected. Specifically, clinicians may wish to provide support for those when disclosing their CF to peers as the present findings have highlighted that this can be a difficult experience. Additionally, it will likely be important for clinicians to liaise with others systems such as schools to offer support to young people across these areas.

The CF team involved within this research were already considering the usefulness of online interventions to connect adolescents with CF prior to this research and the present findings can be taken forwards to ensure that individual adolescent preferences are considered. These results have highlighted that such psychosocial support groups could be offered that connect adolescents socially who may have shared interests and experiences, as opposed to the same specific medical condition.

The importance of taking into consideration the individual subjective experiences of adolescents with CF was further highlighted through this research. Although some social experiences were shared across participants, there were some divergences and this highlights the need for ongoing individualised care planning and treatment (Bell et al., 2020).

2.4.5 Strengths and Limitations

The present research used IPA to allow for exploration of the lived social experiences of adolescents with CF, without seeking to confirm or deny convergence with a specific theory or body of knowledge. Use of IPA allowed the researcher to use reflexivity throughout the process to be directly involved with the interpretation of the qualitative data (Smith et al., 2009; Smith & Macgannon et al., 2018). Elements of strength of the present research include demonstrations of *transparency* and *rigor*, through use of illustrative quotes, examples of the analysis process in the appendices and critical discussion with supervisors and peers throughout the analysis process (Smith and Macginnon et al., 2018; Yardley, 2000).

When considering limitations, it is important to note the impact of the Covid-19 pandemic as all research activities including recruitment and semi-structured interviews were undertaken online. This may have had a direct impact on recruitment as the conversations that involved initial identification of participants undertaken by CF clinicians could not take place in person. Fortunately, the participants that agreed to be interviewed were comfortable using video-conferencing software for the interviews and were able to share their experiences openly in this way. It has been suggested that rapport building with participants may be more difficult when conducting qualitative research remotely (e.g. Weller, 2017). However, qualitative research examining the experiences of nurses and participants found that video-conferencing is often preferable to non-visual methods such as telephone or e-mail (Archibald et al., 2019).

Since the Covid-19 pandemic, research has also been conducted into the possible impact of restrictions, noting that some individuals may be less likely to participate in research, especially if reporting heightened levels of distress (Cardel et al., 2020). However, others have suggested that high quality qualitative research can be undertaken virtually, providing that methodological and ethical factors are taken into consideration (Roberts et al., 2021).

Unlike considerations that may be made regarding generalisability of findings in quantitative research, the present findings do not aim to be representative of the entire CF adolescent population. Rather, the current research findings represent an interpretation of the lived social experiences of adolescents with CF in a specific, homogenous sample. It is hoped that the present research will build upon previous qualitative research undertaken internationally and can be shared with the CF team.

2.4.6 Recommendations for Future Research

Although all five participants shared some common social experiences of living as an adolescent with CF, the awareness of self within the context of reduced life-expectancy was discussed by one participant, Aiden. Future research into the experiences of those reaching late adolescence and early adulthood may provide further insight into the impact of reduced life expectancy.

Newer treatment advances for CF, such as gene therapy, are now being offered to some patients and therefore life expectancy remains ever-changing (Yan et al., 2019). Some participants, such as Sarah, shared feeling of elation within herself and her family when she was informed that she was eligible. Future qualitative research into the experiences of those offered such therapies and the impact that this had had in relation to sense of self and future planning may help support young adults going through this process. As life-expectancy continues to rise due to ongoing advances in treatment, more young adults with CF are choosing to become mother and fathers. Qualitative research examining such experiences may be useful, in the context of CF being a hereditary condition. It may also have been useful to include an accompanying psychometric measure to focus on specific factors such as a Transition Readiness Scale (Dudman, Rapley & Wilson, 2011) or disease-specific quality of life (e.g. Taft, 2021).

2.4.7 Conclusion

Adolescents with CF experience their social worlds in similar ways to their peers without physical health conditions. However, ongoing treatment and increased autonomy and considerations related to disclosure mean that adolescents with CF have additional and unique challenges to overcome throughout this important developmental stage. This research highlights the critical role of ongoing, individualised care planning as patients transition from child to adult services.

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CHAPTER THREE: PRESS RELEASES

3.1 Literature Review: Meta-Analysis

Remotely delivered therapy can improve quality of life of adults with physical health conditions.

Covid-19 has rapidly increased the use of remotely delivered therapy but effectiveness has remained unclear for those with physical health conditions. However, new research conducted at The University of Birmingham has shown that adults with a range of physical health conditions such as cancer, tinnitus and irritable bowel syndrome may benefit from remotely delivered therapy. The review combined data from 24 studies and found that therapy delivered remotely can improve a patient's health-related quality of life.

The author, Jessica Seddon, based in the Centre for Applied Psychology at the University of Birmingham, hopes that the findings may increase the options for those with physical health conditions. Due to pain, mobility issues or intensive medical treatment, adults with physical health conditions may particularly benefit from the option of receiving therapy from the comfort of home.

The included studies covered a range of different therapies across several different health conditions. An approach called Cognitive-Behavioural-Therapy (CBT) was the most widely used remote therapy and the strongest findings were found for the studies that used this model. The review also found that studies that included direct therapist involvement appeared to be more effective than those than those without which suggests that this may be an important factor to consider if services decide to use remotely delivered therapy in the long-term. The quality of the included studies varied and this was taken into consideration as part of the research process. The findings support earlier research that found online therapy can work for common mental health difficulties and therefore it is likely remote therapy may be seen in services long after the pandemic is over.

3.2 Empirical Paper

Teenagers with Cystic Fibrosis have both similar and different social experiences to their peers as they navigate their social lives and increasing independence.

The social experiences of teenagers with physical health conditions such as Cystic Fibrosis could be better understood through findings of Psychology research undertaken at the University of Birmingham.

Interviews with teenagers who have Cystic Fibrosis revealed that a desire to belong and be more independent was important to them. Teens with Cystic Fibrosis also have to consider when to tell others about their condition and possible reactions of others, their long-term health and changing social lives. The author, Jessica Seddon, of the University of Birmingham's Centre for Applied Psychology, hopes that the findings can be shared to help improve the experiences of teenagers with Cystic Fibrosis who receive health care in the area.

Five teenagers with Cystic Fibrosis were interviewed to explore their social experiences and how they make sense of being a teen with long-term physical health condition. The study found several themes of importance for teenagers with Cystic Fibrosis. Although some social experiences were similar to those of peers, such as changing family relationships and increased decision making, teens with Cystic Fibrosis have other factors to consider. Deciding when and how to tell peers about their condition and navigating cross infection control guidelines that prevent teens with Cystic Fibrosis from meeting each other in person. The research also showed how the teens use different ways to connect with others, such as social media, gaming, streaming and video calls.

The study involved looking at interviews with teens who have Cystic Fibrosis to explore their social experiences. The findings suggested that teens with Cystic Fibrosis have both similar and different social experiences to their peers without Cystic Fibrosis. As they seek to belong in social groups, the adolescents consider how visible their condition is and when best to tell their friends.

The research is important as it highlights the social experiences of teenagers with Cystic Fibrosis and how these experiences are similar and different to their peers. The findings are supported by previous research looking at interviews with teenagers who have other health conditions and international research in the U.S.A and Canada. It is hoped that the findings will help improve the experiences of teenagers with Cystic Fibrosis who are prevented from meeting each other face-to-face due to the danger of infection.

All of the research activities, including the interviews with the teenagers were conducted online due to the coronavirus pandemic.

VOLUME ONE: LIST OF APPENDICES

Appendix A – HRA/REC Ethical Approval Letter

Ymchwil Iechyd a Gofal Cymru
Health and Care Research Wales

NHS
Health Research Authority

Miss Jessica Seddon
[redacted]

Email: [redacted]

19 June 2020

Dear Miss Seddon

HRA and Health and Care Research Wales (HCRW) Approval Letter

Study title:	**A qualitative exploration of the social experiences of young people with Cystic Fibrosis (CF)** [redacted]
IRAS project ID:	**275378**
Protocol number:	**RG_19-217**
REC reference:	**20/WA/0163**
Sponsor	**University of Birmingham**

I am pleased to confirm that HRA and Health and Care Research Wales (HCRW) Approval has been given for the above referenced study, on the basis described in the application form, protocol, supporting documentation and any clarifications received. You should not expect to receive anything further relating to this application.

Please now work with participating NHS organisations to confirm capacity and capability, in line with the instructions provided in the "Information to support study set up" section towards the end of this letter.

Appendix A2 - Minor amendment - Covid-19

Amendment Tool v1.2 11 Jun 2020	For office use QC: No

Section 1: Project information

Short project title*:	Qualitative social experiences of young people with cystic fibrosis
IRAS project ID* (or REC reference if no IRAS project ID is available):	275378
Sponsor amendment reference number*:	Amendment 01
Sponsor amendment date* (enter as DD/MM/YY):	14 July 2020
Summary of amendment including justification*:	Due to Covid-19 restrictions in place at the research site [redacted]ess cannot currently be granted to visit the site in person or for direct [redacted]. Therefore, all research activity is to be completed remotely, with audio recordings of interviews being captured directly via secure video-confernecing software or through use of an encrypted dictaphone. [redacted]
Project type:	(•) Specific study ( ) Research tissue bank ( ) Research database
Has the study been reviewed by a UKECA-recognised Research Ethics Committee (REC) prior to this amendment?:	(•) Yes ( ) No

Appendix B - Local R&D approval

[redacted]

[redacted]

14 October 2020

[redacted]

[redacted] **R&D Confirmation of Capacity & Capability**

Project Title: Qualitative social experiences of young people with cystic fibrosis
IRAS: 275378
REC Reference: 20/WA/0163
R&D No : 20_BC_DTS_NO_438
Protocol : RG_19-217

Thank you for informing [redacted] R&D office of the above project.

I can confirm that [redacted] the capacity and capability to deliver the above referenced study and therefore is issuing R&D Confirmation of Capacity & Capability for this project.

Please find attached our Organisation Information Document as confirmation.

Appendix C - Participant Information Sheet (13-14 yrs.)

UNIVERSITY OF BIRMINGHAM

[redacted]

[redacted]

Participant Information Sheet 13-14yrs

A qualitative exploration of the social experiences of young people with Cystic Fibrosis (CF) [redacted]

Researchers:

My name is Jessica Seddon and I am a Trainee Psychologist studying at the University of Birmingham.

What is this research about? The project is looking at how young people with CF interact. We are hoping to interview young people with CF who receive clinical care [redacted]
We are looking at the impact of infection control guidelines and the overall social experiences of young people.

Why have I been asked to take part?

You have been asked to take part as you have cystic fibrosis and can help us to understand more about your experiences. We are looking to speak to individuals with CF who are 13-16 years of age [redacted]

Do I have to take part?

No, taking part is entirely up to you. If you decide not to take part this will not affect your treatment in any way.

[redacted]

What will happen to me if I agree to take part?

1. You will have the opportunity to ask any questions before agreeing to take part. A detailed Parent Information Sheet will also be given to your parent or guardian.
2. A suitable time and date will be arranged for the interviews to take place. Participants will have at least 48 hours to consider whether they want to take part.
3. You will be asked to complete a form confirming that you are willing to take part. We will also ask your parent or guardian for their consent. You will only be allowed to take part with your parent's consent.
4. Interviews will take place remotely via video call or telephone if video is not possible, with a choice of dates and times given.
5. Interviews will last for around 1-1.5 hours. Questions asked will be related to your experience of living with Cystic Fibrosis and your wider social experiences.
6. At the start of the interview, the Chief Investigator (Jessica Seddon) will ensure you still want to take part and will remind you of your right to withdraw and limits to confidentiality if risk is shared.
7. Interviews will be audio recorded and then typed up. Your name will be removed.
8. The researcher will then look at the data from the interviews for common themes.

What will happen if I do not want to carry on with the study?
It is up to you if you want to take part in the study. You are free to stop taking part at any time without giving a reason. If you decide to stop taking part this will not affect your care whilst in hospital.

What happens to my data and information about me?

If you take part in the study then you will be assigned a random number and your data (Name, NHS number and interview) will be given a code by the Chief Investigator. No one other than the research team will know who you are. All data will be stored on a password protected file, data will be transferred using a secure USB memory stick. Only the research team will only have access to the anonymised data.

People who do not need to know who you are won't be able to see your name or contact details. Your data will be given a code instead.

Who should I contact if I have any questions about the study?

If you would like more information, have any questions or are worried about anything to do with this study please do contact the research team and we will do our best to answer your questions. Your parents will have our contact information and can contact us on your behalf or you can email us on:

Jessica Seddon [redacted]

General support and advice can also be found on the Cystic Fibrosis Trust website: www.cysticfibrosis.org.uk

What will happen to the results of the research study?

Information collected about you during the study will be shared with your clinical team. Your data will be coded and anyone who does not need to know who you are will not be able to identify you. The findings from this study will be published in a research journal and included in a thesis as part of a post graduate degree known as a doctorate.

Summaries of results can also be shared with you and your parents, if you agree to being contacted.

Appendix D – Participant Information Sheet (15-16 yrs.)

UNIVERSITY OF BIRMINGHAM

[redacted]

Participant Information Sheet 15-16 yrs.

A qualitative exploration of the social experiences of young people with Cystic Fibrosis (CF) [redacted]

Researchers:

My name is Jessica Seddon and I am a Trainee Psychologist studying at the University of Birmingham.

What is the purpose of this research?

The project is looking at social experiences of young people with CF. We are hoping to interview young people with CF who receive clinical care [redacted] are looking at the impact of infection control guidelines and the overall social experiences of young people.

Why have I been invited to take part?

We are keen to hear from young people, like yourself, experiencing CF directly, as we feel this will be the best way to capture experiences. We are looking to speak to individuals with CF who are 13-16 years of age who are receiving [redacted]

Do I have to take part?

No, taking part is entirely up to you. If you decide not to take part this will not affect your treatment in any way.

[redacted]

What will happen to me if I agree to take part?

1. You will have the opportunity to ask any questions before agreeing to take part. A suitable time and date will be arranged for the interviews to take place.
2. Written informed consent, if you are 16 years old, or assent if you are under the age of 16 will be sought first. We will ask your parent or guardian for their consent first, if you are below the age of 16.
3. Following consent an interview will be arranged by video call or telephone. A choice of dates and times will be given for the interviews to take place.
4. At the start of the interview, the Chief Investigator (Jess Seddon) will ensure you still want to proceed and be reminded of their right to withdraw and limits to confidentiality if risk is disclosed.
5. Interviews will be audio recorded through secure video-conferencing software or onto a secure device known as a Dictaphone and then later transcribed. During this process, all patient identifiable data will be removed.
6. Audio files will then be anonymised using a code and the researcher will look for common themes within the data.

What are the benefits to taking part in this study?

There are no direct benefits to your as a result of taking part in this study but we hope that the information that [redacted]

Are there any risks to participating in this study?

While the interview is unlikely to deviate from areas covered within routine clinic reviews, it is possible that you might become upset, if that happens we can stop the interview until you feel ready to continue. Should you become distressed we can stop the interview and refer you [redacted]

In the event that you disclose information that may indicate you are at risk of harm then local safeguarding procedures will be followed to ensure your safety.

The study participation will take approximately 2-3 hrs in total, with the interview being 1-1.5hrs in duration.

How will we use information about you?

We will need to use information from your medical record for this research project. This information will include your:

- Name
- Age - Specifically, we are looking to recruit individuals who are aged 13-16 years of age who are receiving outpatient care for their [redacted]

[redacted]

- Contact Details [redacted]

You will only be contacted by the Chief Investigator of the study if you want to hear more or if you are interested in taking part. People who do not need to know who you are will not be able to see your name or contact details as your data will have an ID code instead. We will keep all information about you safe and secure. Some of your information, such as the key linking your data with the ID code will be stored at University of Birmingham on secure servers. They also have to follow rules to ensure this data is kept secure.

Once we have finished the study, we will keep some of the data securely, including signed paper consent forms and anonymised interview transcripts for ten years. Other patient identifiable data, including the audio recordings, will be securely destroyed once interviews have been transcribed (Up to 4 weeks after interviews have taken place). Reports will be written in a way that no-one can work out that you took part in the study.

What are your choices about how your information is used?

- You can stop being part of the study at any time, without giving a reason, but we will keep information about you that we already have. This will not affect your care in any way.
- We need to manage your records in specific ways for the research to be reliable. This means that we won't be able to let you see or change the data we hold about you. Two weeks after interviews have taken place, it will not be possible to remove this data as the transcription and analysis process will have begun. The key linking Study ID code and patient names will be securely destroyed following interview transcription completion (Up to four weeks after the interviews have taken place).
- All investigators and study site staff will comply with the requirements of the Data Protection Act 2018 with regards to the collection, storage, processing and disclosure of personal information and will uphold the Act's core principles. Access to the anonymised transcripts of audio recorded interviews will be limited to the Chief Investigator, Local Collaborator and Academic/Research Supervisor at the University or Birmingham.

What if I want to withdraw from the study?

You are free to withdraw from the study at any time without reason, your decision to withdraw will not affect your treatment in any way. You have up to two weeks after the interviews have taken place to withdraw your data, after this while you can still withdraw it will not be possible to withdraw your data and any anonymised data collected up to the point of withdrawal will be retained in the study.

Where can you find out more about how your information is used?

- at www.hra.nhs.uk/information-about-patients/
- By emailing the Chief Investigator: [redacted]

[redacted]

- By contacting the research governance and sponsor representative at University of Birmingham: [redacted]

Who to contact if you have any concern or want to make a complaint?

- Chief Investigator: [redacted]
- For complaints regarding how your data has been handled by the University please contact the Data Protection Officer [redacted]
- If you are not happy with their response or believe they are processing your data in a way that is not right or lawful you can complain to the Information Commissioner's Office (ICO) via their website [redacted]
- If your questions are still not answered and you have concerns regarding the way the study has been conducted or wish to make a complaint then please contact: [redacted]
- More general advice and support specifically related to CF can be found at the Cystic Fibrosis Trust website: https://www.cysticfibrosis.org.uk

Who has insured and funded this study?

The study sponsor has in place a Public Liability Policy which provides cover for negligent harm, activities conducted at the NHS sites are covered by the NHS indemnity for that site.

There is no external funding for this study.

Who has reviewed the study?

This study has been reviewed and approved by the following Research Ethics Committee: Wales REC 6.

What will happen to the results of the research study?

Research findings including anonymised quotes, will be shared with the clinical team and potentially for publication as well as written up as part of a doctoral thesis. Summaries of results can also be shared with participants, if they consent to being contacted.

Appendix E – Parent/Guardian Information Sheet

Parent/Guardian Information Sheet V8 10/07/2020 IRAS: 275378

UNIVERSITY OF BIRMINGHAM

[redacted]

Parent/Guardian Information Sheet

A qualitative exploration of the social experiences of young people with Cystic Fibrosis (CF) [redacted]

What is the purpose of this study?

This is a study sponsored by the University of Birmingham looking to explore the social experiences of young people with Cystic Fibrosis (CF) [redacted]

Infection control guidelines stipulate that individuals with CF are prevented from face-to-face contact. It is unclear whether this has an impact on the lived experiences and sense making of young people with CF.

This study is looking to explore social experiences through semi-structured interview discussions with young people. It is hoped that gaining a better understanding of the lived social experiences of young people with CF will provide teams with useful information to improve experiences.

What does this study involve?

Young people and their parents or carers will have the opportunity to ask further questions prior to informed written consent being gained. Following informed consent and/or assent (Parental consent required for young people under the age of 16), a date and time will be arranged for an interview with the young person.

Interviews will take place virtually via video-conferencing software or telephone if video is not possible. The semi-structured interviews should take between 1 and 1.5 hrs.

The interviews will be audio recorded via the secure video-conferencing software or onto an encrypted Dictaphone and qualitative data collected will then be analysed to make sense of the young person's experiences.

[redacted]

Why has my child been invited to take part?

Your child is receiving [redacted] and is eligible to take part if they are aged 13-16 years. This is because the research study is focussed specifically on adolescent social experiences of CF.

Does my child have to take part?

It is entirely up to you whether you and your child choose take part in this study. If you do decide to take part you will be asked to sign a consent form and your child will be asked to sign an assent form so that we can take their views about the study into account. You are free to withdraw at any point from the study without giving a reason, even when you have previously given consent to be involved.

Participating or choosing not to participate will not impact on the usual care your child receives.

What are the benefits to taking part in this study?

There are no direct benefits to your as a result of taking part in this study but we hope that the information that we gain from the study will help us support other young people and adolescents [redacted] in future.

Are there any risks to participating in this study?

Discussion points are unlikely to deviate from areas covered within routine clinic reviews. It is possible that your child may become upset during the interviews, if that happens we can stop the interview until your child feels ready to continue. In the event that your child becomes distressed we can stop the interview and you and your child will be referred to [redacted] website for support.

In the event that your child discloses information about themselves that may indicate they are at risk of harm to themselves or others then local safeguarding procedures will be followed to ensure yours and your child's safety including the possibility of a Psychology referral. This will be in collaboration with your child.

The study participation will take approximately 2-3 hrs in total, with the interview being 1-1.5hrs in duration.

How will we use information about your child?

We will need to use information from your child's medical records for this research project. This information will include your child's:

- Name
- Age - Specifically, we are looking to recruit individuals who are aged 13-16 years of age who are receiving [redacted]
- [redacted]
- Contact Details – [redacted]

You will only be contacted by the Chief Investigator of the study if you want to hear more or if you are interested in taking part. People who do not need to know who you child is will not be able to see your name or contact [redacted] secure. Some of your child's information, such as the key linking your child's data with the ID code will be stored at University of Birmingham on secure servers. They also have to follow rules to ensure this data is kept secure.

Once we have finished the study, we will keep some of the data securely, including signed paper consent forms and anonymised interview transcripts for ten years. Other patient identifiable data, including the audio recordings, will be securely destroyed once interviews have been transcribed (Up to 4 weeks after interviews have taken place). Reports will be written in a way that no-one can work out that your child took part in the study.

What are your choices about how your child's information is used?

- Your child can stop being part of the study at any time, without giving a reason, but we will keep information about them that we already have. This will not affect your child's care in any way.
- We need to manage your child's records in specific ways for the research to be reliable. This means that we won't be able to let you see or change the data we hold about you. Two weeks after interviews have taken place, it will not be possible to remove this data as the transcription and analysis process will have begun. The key linking Study ID code and patient names will be securely destroyed following interview transcription completion (Up to 4 weeks after the interviews have taken place).
- All investigators and study site staff will comply with the requirements of the Data Protection Act 2018 with regards to the collection, storage, processing and disclosure of personal information and will uphold the Act's core principles. Access to the anonymised transcripts of audio recorded interviews will be limited to the Chief Investigator, Local Collaborator and Academic/Research Supervisor at the University or Birmingham.

What if I want to withdraw from the study?

You are free to withdraw your child from the study at any time without reason, your decision to withdraw will not affect your child's treatment in any way. You have up to two weeks after the interviews have taken place to withdraw your child's data, after this while you can still withdraw your child it may not be possible to withdraw their data and any anonymised data collected up to the point of withdrawal will be retained in the study.

Where can you find out more about how your child's information is used?

[redacted]

- at www.hra.nhs.uk/information-about-patients/

[redacted]

- For complaints regarding how your data has been handled by the University please contact the Data Protection Officer via [redacted]
- If you are not happy with their response or believe they are processing your data in a way that is not right or lawful you can complain to the Information Commissioner's Office (ICO) via their website [redacted]
- If your questions are still not answered and you have concerns regarding the way the study has been conducted or wish to make a complaint then please contact: [redacted]
- More general advice and support specifically related to CF can be found at the Cystic Fibrosis Trust website: https://www.cysticfibrosis.org.uk

[redacted]

Who has insured and funded this study?

The study sponsor has in place a Public Liability Policy which provides cover for negligent harm, activities conducted at the [redacted]

There is no external funding for this study.

Who has reviewed the study?

This study has been reviewed and approved by the following Research Ethics Committee: Wales REC 6.

What will happen to the results of the research study?

Research findings including anonymised quotes, will be shared with the clinical team and potentially for publication as well as written up as part of a doctoral thesis. Summaries of results can also be shared with participants, if they consent to being contacted.

Appendix F - Participant Consent Form (16yrs)

UNIVERSITY OF BIRMINGHAM

[redacted]

Consent form - 16 years

A qualitative exploration of the social experiences of young people with Cystic Fibrosis (CF) [redacted]

[redacted]

Study ID code:

Please initial to confirm

I confirm that I have read and understood the information sheet for this study and any questions that I had have been answered by the research team.	☐
I confirm that am aware of what the study will involve, including a video call and an audio-recorded interview. I agree to being interviewed by video call and consent to my audio recordings being held by the researchers at the University of Birmingham for this study	☐
I can confirm that I understand that my participation is voluntary and that I am free to withdraw from any part of the study without giving a reason.	☐
I understand that if I withdraw from the study two weeks after the interviews have taken place that data collected about me up to that point will remain in the study.	☐
I understand that a referral for additional support may be made on my behalf if deemed necessary by the researchers, for example if risk of harm is disclosed. This will be discussed openly with you beforehand.	☐
I understand that my identifiable data will be linked to transcribed interviews held on password-protected secure servers at the University of Birmingham for the purpose of this study, and I consent to this.	☐

I understand that data collected during the study may be looked at by individuals from [redacted] and the research team at the University of Birmingham, where it is relevant to my taking part in this research. I give permission for these individuals to have access to my relevant clinical data. ☐

I agree to take part in this study ☐

________________	__________________	______________
Participants' name	**Participants' signature**	**Date of signature**
________________	__________________	______________
Researchers' name	**Researchers' signature**	**Date of signature**

If completing electronically: Once signed, please scan or take a photograph of both pages [redacted]

Researcher to sign electronically on date of receipt.

One copy for the parent/guardian, one for the patients' medical record and one copy for the study file

Appendix G – Parent/Guardian Consent Form

UNIVERSITY OF BIRMINGHAM

[redacted]

Consent form for Parents/ Guardians

Study title: **A qualitative exploration of the social experiences of young people with Cystic Fibrosis (CF)** [redacted]

Participant/ Patient's Study ID:

	Please initial to confirm
I confirm that I have read and understood the information sheet version ___ dated ___ for this study and any questions that I had have been answered by the research team	☐
I can confirm that I have understood what is involved with the study and agree to my child's participation in an interview via video conference call or telephone about their social experiences.	☐
I understand that my child's interview will be audio recorded and I consent to my child's audio recordings being held on secure servers by the researchers at the University of Birmingham for this study.	☐
I can confirm that I understand that my participation, and that of my child is voluntary and that either of us is free to withdraw at any time, without giving a reason. This will not affect any aspect of my child's care.	☐
I understand that if I withdraw my child two weeks after the interviews have taken place that data collected up to that point will remain in the study.	☐
I understand that a referral for additional support may be made on my child's behalf according to local procedures if deemed necessary by the researchers.	☐

I understand that my child's identifiable data will be linked to transcribed interviews with a code, held on password-protected secure servers at the University of Birmingham for the purpose of this study, and I consent to this. ☐

I understand that data collected during the study may be looked at by authorised individuals from [redacted] and the research team at the University of Birmingham, where it is relevant to my child taking part in this research. I give permission for these individuals to have access to my child's relevant clinical data. ☐

I agree for my child to take part in this study. ☐

Name of Child:

Relationship to the Child: (Parent/Guardian – delete as applicable)

By signing this form I confirm that I have parental responsibility for this child.

___________________________	______________________	____________
Print Name (Parent/ Guardian)	**Parent/ Guardian signature**	**Date**
___________________________	______________________	____________
Print Name (Researcher)	**Researcher signature**	**Date**

If completing electronically: Once signed, please scan or take a photograph of both pages email this back to the Chief Investigator: [redacted] **If not possible to sign, please ensure your full name is included.**

Researcher to sign electronically on date of receipt.

One copy for the parent/guardian, one for the patients' medical record and one copy for the study file.

Appendix H – Participant Assent Form (13-15 yrs.)

UNIVERSITY OF BIRMINGHAM

[redacted]

Assent form for children aged 13-15 years

A qualitative exploration of the social experiences of young people with Cystic Fibrosis (CF) [redacted]

Study ID code:

	Please initial if you agree
I can confirm I have read and understood the Participant Information Sheet for this study.	☐
I have asked all the questions that I want to about the study and my questions have been answered by the research team.	☐
I'm happy that I know everything I need to about this study, including the interview taking place by video call or telephone and that my interview will be audio-recorded.	☐
I understand that I am free to stop taking part in the study at any time, without giving a reason and this will not affect my treatment at the hospital.	☐
I understand that data collected about me during the study may be looked at by people who have permission from [redacted] and the research team at the University of Birmingham to make sure the research is being done properly. I agree to them having access to my data.	☐

[SERVICE LOGO REDACTED]

I agree to take part in this study ☐

Name of Participant: ___________________

________________	________________	________________
Researchers' name	**Researchers' signature**	**Date of signature**

<u>If completing electronically: Once initials have been inserted into the boxes and a name confirmed, please email this back to the Chief Investigator:</u> [redacted]

Researcher to sign forms electronically on date of receipt.

One copy for the parent or guardian, one original for the study file and one copy for the patient's medical record

Appendix I - Semi-Structured Interview Schedule

Introduction: Thanks for agreeing to take part in this research. I'm really interested to hear about your life and who is around and how they make you feel. We will also chat about your experiences of CF.

1. First, I would like to understand who is important to you in your life. I'm interested in who is around you, and would like us to draw this out on a bit of a map – including all the people who are important for you, even if you don't see everyone often or in person.

2. ** Draw out with participant an ecomap of important people and systems (e.g. school) in YP's social network to help facilitate the discussion. Record on Dictaphone while doing ecomap**

3. So you mentioned… (*This friend, school,* [redacted] why is that an important part or not so important to you?

4. Has this changed over time? How?

5. So tell me about living with CF? What has that been like for you?
PROMPTS: *what has been difficult? What things have been harder? What has been easier?*

6. How do you feel that CF has affected these? (Refer back to ecomap)

7. The cross-infection control guidelines [redacted] people with CF do not meet face to face. Has this affected who you see? In what way? *Refer to ecomap.*

8. What has you experience been of the social distancing measures brought in as a result of the Covid-19 pandemic?

9. Do you speak to others with CF? How? Where? How do you find this compared to people without CF?
PROMPT: Online, social media?

10. [redacted]
PROMPT: Would this be useful for you?
If yes… why?
If no… why not?

Interview schedule will remain fairly open depending on participants chosen discussion points, so although guided, the schedule aims to allow participants to set the parameters of discussion points.

Appendix J – Example of initial descriptive, linguistic and conceptual comments for "Natalie"

Natalie: Yup	Initial descriptive, *linguistic* and conceptual comments
Interviewer: And are there other things that you'll be doing more of, related to your Cystic Fibrosis?	
Natalie: Errrr, I dunno… I don't really know.	*Finding it difficult to articulate experience*
Interviewer: Mmhmm, so in terms of I guess having CF overall, so for someone that's never had CF themselves, whats that actually been like as a child to grow up with CF?	Looking into the experiences of having CF more generally.
Natalie: Errr.. it was hard but not very hard, but now cause I'm like older… it's Ok to cope with.	*Hesitancy.* Relied on parents in childhood – Increased independence. Maturity. acceptance
Interviewer: mmhmm	
Natalie: But now… when I was younger, you had to like.. erm, rely on your parents to do everything for you	Reflecting on childhood and increased independence, increased responsibility as *'weird'* (unfamiliar?)
Interviewer: Mmm. Yeah, and what kind of things we you relying on your parents to do?	
Natalie: The would always do.. my physio for me.	
Interviewer: Mmmhmm	Use of the emphasising language '*always*' - emphasising it is not something she is used to. *Use of humour*
And they'd set everything up and… [laughs] do the talking… whenever the doctor and that asked like.. … cause I can do my nebs… cause they're just easy	. Familiarity, reliance, consistent support - Relationship with independence
Interviewer: yeah so it sounds like some parts are a bit easier to get used to?	
Natalie: Mmmhmm	
Interviewer: And you mentioned some part of having Cystic Fibrosis was hard when you were younger?	Difficult to describe/explain CF to others, worried about being misunderstood.
Natalie: It was hard to like, describe it to people… cause they wouldn't, they wouldn't really understand..	Disclosure Impact of CF – *hard*
Interviewer: Mmm	Experience of people being confused with other conditions?
Natalie: And they'd like, think it was something else	
Interviewer: Mmm	Desire to be understood – difficult to explain CF to others.
Natalie: So they could say… is it like this, this and I'd be like… umm it's not like that at all.	Experience of being repeatedly misunderstood by peers.
Interviewer: Yeah	
Natalie: So it had been harder for them to understand what it was.	Retelling experiences of being misunderstood.
Interviewer: Mmm.. and who.. was there any people or groups of people that found it harder to understand?	

Appendix K – Example of Emergent Themes at Individual Level for "Sarah"

Emergent Themes	Quotes / line number
Grateful for support from others *Different types of support* Sister practical support (e.g. doing hair, helping with school work) Teachers- Practical and emotional Parents – ensuring she was encouraged to do everything she wanted to.	150 - *..”does make us closer sometimes because like before like hospital admission my sister will sit down with me, she’ll do my hair… she’ll just sit and like, sort of stay with me for a bit I think..”* 160-164 “*when I’m sort of having the days where I’m just so tired and I get loads of homework…, she’ll [sister] like sit me down and talk me through it and she’s like if you don’t have time to do it, then I’ll help you out with it another day, which I think was always nice”* 235-239 “..my form tutor was really good actually. My form tutor would like email me, atleast once or twice in the week and be like, how are you doing do you need any work, it’s ok if you don’t need work, are you tired, do you need this?” 257-260 – *“..so the school was really good with it, my physics teacher earlier, like when we came back to school, he was like this is all, I know that you got the stuff but in case you need any like going over it, all my teachers were like really good with it, which was quite nice.”* *581-591: “.Yeah, I think I’m very lucky because my parents always like pushed for me to be able to do stuff. They always made a way around it…”* *594-600- “…she like phoned the hospital saying ‘Sarah would like to go back, can she?’ So I think having parents, both my mum and dad that really pushed, to make it as normal, whatever normal means, as life for me, it sort of helped that because they never sort of gave up, they sort of said ‘oh well, they said you cant do that so you cant’ and they were like ‘No, here’s all the reasons she can do that’.”* 642-645 – “so I was sort of having to shield and then my dad was, so then my mum and my sister were like, well we’ll stay at home as well, so it was all of us in one house, for like three months, three to four months…” *785-789 – “…… yeah it was nice, cause my cousins a [insert job role], well my cousin and Godmother. So when I had my admission, she was there at the hospital for a couple of months so like erm, in the morning shed like run up and be like ‘I’ve just got here, have a nice day’ I’d be like ‘you too’ and she’d just like visit me during the day, which was really nice… to sort of like see a familiar face.”*
A desire to be understood *Shared experiences. / Keeping familiarity* School friends – Always there, known from beginning, some shared experience (e.g. nut allergy). Not having to explain self.	*50- “…one of them has a nut allergy and [name] is just fine [laughs].So we’ve always sort of bonded over being close and knowing somebody who is slightly… not different but has an addition, like something that is additional alongside it. I think it sort of… it sounds stupid but it sort of helped our friendship…”* *59-65 “*… we both sort of felt a little bit like we were different to everyone else, so we always had that sort of little bond, you know, we both now wat it’s like to be… in the group meetings of the risk assessments for all of the people’s school trips and things like that, we would always be sat there together and be like “Oh God!” so I think that was quite a nice thing 72-79- *“…Yeah, it’s almost like subconsciously. I just get along with people who have allergies or little things like that. I think… they were sort of my first sort of.. taste if you will, of people who knew what it was lik ewhich was really nice because of course I couldn’t really meet anyone else with Cystic Fibrosis, so it was sort of like there was a physical barrier there anyway… so they were like as close as I could get?”*

Being seen as different to peers, disclosure, having to explain condition to others.	123-130 - I've also got my like bigger friendship group full of people that I've like known since I was in primary school…. I think it was easier to keep my friends that I've known forever than make new ones because they of course knew about it because I went to primary school with them. When I made new friends I sort of had to explain it…… like the little things like why I can skip the queue at lunch, why I'm taking these meds now and then…so it was quite.. weird… I just sort of avoided making new friends just the for ease of keeping the old ones. *"…people next to me were like 'oh yeah, I wake up at 8 so I can get to school for 08:30, like what time do you wake up?' and I was like 6 o'clock [laugh] and yeah I'd be like I have to wake up at 6 oclock, cause I have to do this and this and this in the morning. But I think that I used to think that'd I'd have to share that with people but I just don't."*
The missing piece CF friend - Irreplaceable, rare, special, 100% completely gets it. Impact on defiance> acceptance stage. Helped her to process this	*464-467- It was sort of nice to speak with somebody who, cause I'd grew up with her, but like, she didn't like totally get it because she cant, but it was sort of like a bit closer to getting it, with like hospital visits and little things like that.. so it was nice to have someone who like kind of understands, in more of a sympathetic point of view, erm, that sort of helped, especially sort of early on. I wasn't so like, alone in it…"* *283-285- "..we talk quite often and it's usually little things, like oh how are your mocks, how was revising during mocks. Cause I think stuff like that, it is really hard to balance revision, relaxation and physio."* *291-295 - "… but I think having someone there who completely gets it, is so worth it. Like I think that like it should be pushed for, because having somebody you can speak to, who will 100% get it, is so rare. And just in general, especially in these situations it's like, you understand it completely and we can offer advice to one another."* 831-834 – "… I think having a friend with CF, was like so vital, just to me sort of accepting it and not being like 'I'm so different from all my friends' anymore. Because I think it just sort of drew about like actually, at the end of the day, I'm not like… so I think it's a very useful thing…"
Shared experiences with others who have CF *Desire to now support others with shared experiences.*	*307-310- "…so little things like that it's good to sort of give that advice that you can genuinely take as it is, 'cause the hospital give you everything that they can, but it's sort of that first hand oh here's what happened with it, here's what you can look forward to kind of thing, it's sort of very very nice having someone who will completely get it."* *317-320-"… my lung function's up, like it just completely went up, err recently, like the person that I can talk to who completely understands the sort of joy of that is him… like there's nobody else who I can talk to who will completely get it."* *322-225- "…I'll share it with them [Friends without Cystic Fibrosis] of course because they're with me all the time, I can't really escape them! But with him [Friend with CF] I'm sort of like, here's what happened and he'l get it. He'l sort of match that sort of 'Oh my god, that's so exciting'. Its, it's irreplaceable…that sort of bond."* *388-319- "… because the first wave of gene therapy had just came in, and we were just finding out we'd got that. So I remember then we started being like ..'oh my god, do you know when you're getting it?!'."* *395-400 – "Yeah, instead of being like alone in sort of like this is what I have to do, I can't.. I think it was like..I can't talk to anyone that will get it….But then when you find somebody who does, it's like well, it's great now because I don't have to sort of suffer in silence if you know what I mean?"* *747-751- "…and I remember she had an inhaler for like two years and then she didn't use it but she kept that little box and one day she was like 'I'm like you, I have some of these, I'm just like you', 'cause she always used to follow me around everywhere…"* *338-341-" … so it's sort of like, speaking to someone who can completely like get it. I… it's usually, I think she went into hospital, sort of a couple of months after I did, so then I was sort of passing on the information that he gave to me to her."*

	844- "…cause I think even just having one person who you can just speak to is so helpful, like I can't even emphasise how important it was for me.." 868-870 - **:** ***"****Yeah, well thank you for the offer, because it's been very very nice to know I'm sort of helping out with something.."*
Going through adolescence with Cystic Fibrosis – Increasing independence, defiance and then acceptance.	*"…they'd just be like OK, well we'll be here when you do. And of course my guilt would be like 'You have to go and do it now'* *"…yeah I just used to hate everything… I'd sort of gone to sort of rejecting it… instead of sort of accepting everything I just have to do. I was just like… I wouldn't do it properly… I'd just sort of be very defiant… and just like 'I'm not gonna do this'. I used to have so many arguments with my parents about it… I used to sort of scream at them like 'This is your fault!', 'I'm not doing it, I don't care'."* (Line 340-349). 381-384 "..*But I just sort of realised that there's no point resisting it, 'cause I'm gonna have to do it at some point, I feel like I've just lost the energy to fight with it."*
Defiance/Refusal stage – early adolescence *& impact on family*	682-686 – When discussing the Covid-19 pandemic *"…But I completely ignored my mum and did not shield before the country went into lockdown I was still going to school, up until like the Wednesday, when mum was like: 'There is no way you can go into school now' and I ended up missing my sister's last day which was quite harsh…"* *629-633 – "…I don't think CF has made me more worried about it, I don't think its made me more wary of it. It's almost as if I just sort of split up myself, when it got really bad, so it was a bit like, Ok this is the bit with me, this is CF, you're going to have a registered bit of time where youre going to worry about it, but other than that, I'm gonna just do whatever, I'm going to get on with my school work.."*
Process of acceptance and perceived facilitators	503-512 *… erm I remember really detesting it (cross infection control guidelines). I remember being like, I remember being like eight or nine, and hating the idea that I couldn't meet somebody like me or get on with somebody who has the same things as me**.** Whereas I think, 'cause they were very very certain, like they were like we don't think it would be a good idea for you to like get a penpal with it, cause youll want to meet them at some point, youll wanna be able to do that at some point, but I think, if its just like a… I think it would have probably helped a little bit. Because it's just like knowing that you can speak to somebody who entirely understands, who just doesn't have to try to understand, they just do. It's very helpful.*
Gratitude	*558 "..the thing that I'm probably most grateful for, that is a kind of weird thing to be grateful for, is that I've always known it was there, I've always had it."*
Social/systemic impact of gene therapy /treatment advances	*808-816- "I don't know who was more excited, me or my mum, my mum literally started crying when she got the phone call, aggressively. I wasn't there, apparently she picked up the phone in the car, on the way back from picking my sister up from school and my sister was like 'Why are you crying' and my mum was like 'It's just so great' cause she'd pulled over, and then I found out later in the evening, so my sister knew for like, the whole day. I get home and nobody tells me until probably like 7pm. They all sit in my room and I'm like ' Yeah, what's happening?' thinking I was in trouble and they were like 'She's got the new drug' and they all start crying and I'm sat there like 'What?!' [laughs]"* *529-533 – "I think that's probably the maturity of older age, knowing that I can't meet you and that's OK. I think I probably would have struggled knowing that I couldn't meet them in person when I was younger, cause I just think it's a hard thing to wrap your head around."*

	535"…. but I think as I sort of accepted it as like, ok, I know that, you know that I know that, and I just really need to speak to somebody who gets it. So I think that as soon as that happened I was sort of just like you know I'm just, I don't mind not seeing them in person because its worth it.. " 544-548 "Yeah… I think, it was just sort of that one thing on the additional list of things that I couldn't do, that I was just sort of like 'Oh and I cant do that and I cant do that'. But as soon as that sort of taken away, it felt like there was a lot less to be hated about it. There was a lot less that I didn't like about it, but other than that, there's very little that impacts me in the everyday/"
Connecting to people remotely (Due to CF and lockdowns).	*691-695 - we said like every night at one point there were like four oh us [lists four], we were all sort of like on a call and playing random video games, like we'd find a new one each week and be like 'this one looks kind of funny, let's play this one' and then like some of them stuck and we still play them now.* *686-689- I kept up with all my friends and I kept speaking to everyone during the lockdown, because of course we all literally had nothing else to do. But what was quite nice is that we all developed our own ways of speaking with one another.* *746-751- "I FaceTime my little cousin more, in this period that I ever have, because she lives in [Place name]"* *731-745- we usually do a giant call on Whatsapp, 'cause it's the easiest one to have numerous people calling on.. or I actually downloaded Zoom the other day, cause one of my friends was like, 'let's do a Zoom and then we can make presentations, so I'll do one about why there are more than five colours in the rainbow or whatever' and then 'I'll do one on this' so we're all gonna have like a Zoom, almost like we're teachers doing like a little thing like when we were tiny and I was like I would have loved to have done that when I was like five so we were like, let's do it then, let's just do it. 'Cause I think Zoom is quite good, cause one of my friends has like a screen transference, so they can show everything in their camera roll so sometimes we just randomly go through eachothers camera rolls looking at one everyone's stuff we've found. "* 408-415- *"I think.. yeah, I I just feel like I've sort of accepted it as a sort of thing now, especially with gene therapy it feels like I don't have it, because of course I still have to do like physiotherapy and stuff, but like I used to hate that I couldn't breathe through my nose, but now I can and smell all these things and it's so exciting cause I would like be sat and the dinner table and be like 'I can smell your salad' and your friends would be like 'yeah, it's a salad' and I'd be like yeah but I can smell it."* *490-497 - I think, I have to weigh up the good and the bad things of it, like, cause of course the bad things are like the hospital visits, the xrays, sort of everything that's it. But like potential advantages like, I get to skip the lunch queue at break, I get free passes at the front of the queue at any theme park [laughs], things like that, like the little things can be, not make, not make it worth it, but make it doable.* *455-461 - "…I have to work a bit harder to just exist. I think that, for a while, was such a constant reminder. But I think the less that I sort of fight with myself about it, and sort of be in the moment, not in the long term I'm just like, it's just ".. it's just what I have to do. Some people need to take insulin shots, some people need to exercise a lot to maintain the same weight. I just, this is just something I have to do.."* *415-418 "…like I don't get afraid of shaking people's hands because they're not sweaty anymore, so I can sort of, everything's difficult, but it used to show and it doesn't…"* *"… it's not something I would choose for myself, but I would rather me have it than somebody else, if you know what I mean? I mean like, its not a fun thing to live with, I just sort of, I've just stopped resisting it. I'm like Ok, it's just me. It's just something I have to just deal with. The less that I think about the impacts, the smaller it feels. Cause especially of late, there's nothing I really can't, or cannot ever do, with it. I sort of.. [sigh] this is really hard to put into words. Umm, sort of living with CF, it's not been an enjoyable experience [laughs], I won't say that, but it's been… Ok."*

Appendix L - Master Table of Superordinate and Sub-Themes

Superordinate and Sub-Themes	Illustrative Quotes, Line numbers and Participant Pseudonyms
A desire to belong and be understood as a young person and as someone with CF	**Aiden** "*I'd latch on to any bit of human connection that actually cared about me that I could…and then I was like 'Oh, can I join your gang?'*" (Line 324-326).
Not wanting to be treated differently because of CF	**Natalie:** *"Some people treat me differently but then others didn't so it was ok… So I just left it… and then people just treat me the same… how everyone else would be treated."* (Line 207-210). *"The adults didn't really care… but like when I went for a sleepover, I would have to take all my meds and stuff… [long pause] and they'd just look at me really weirdly… for everything I did" (*Line 219-221). **Daniel:** *"…it would just make me kind of stand out more and being a bit embarrassing… you just don't really want to stand out."* (Line 121-124). **Sarah:** "*I sort of… [sigh] this is really hard to put into words. Umm, sort of living with CF, it's not been an enjoyable experience [laughs], I won't say that, but it's been… OK."* (Lines 443-444). **Aiden**: *"…they [family] don't like sympathise me…they just treat me like the others... like a normal person…". (Line 95-96).* *"… cause it was sort of a… it was sort of a thing that they attacked every single time… like for instance one of the things they'd say… is remind me to come to your funeral when you're thirty… so when you explain they know and that's... that is one of the easiest things to attack with it... is the age that you die at... so yeah it's... it was erm… for an unmatured mind it was very hard to deal with." (Line 159-170).* **Harry:** *"I do feel more comfortable around my close friend group because I know that they're not going to judge me over it [CF]… And it's much nicer to know that they're not kind of thinking different of me and not like kind of trying to like push me out of the group just because of the symptoms and stuff..." (Line 332-337)* **Sarah:** *"…like I don't get afraid of shaking people's hands because they're not sweaty anymore, so I can sort of, everything's difficult, but it used to show and it doesn't…"415-418* *"…people next to me were like 'oh yeah, I wake up at 8 so I can get to school for 08:30, like what time do you wake up?' and I was like 6 o'clock [laugh] and yeah I'd be like I have to wake up at 6 oclock, cause I have to do this and this and this in the morning. But I think that I used to think that'd I'd have to share that with people but I just don't."*

The Burden of disclosure and the importance of trust	**Harry**: *"I was pretty scared about it 'cause I didn't know how they'd react but, erm, but as soon as I did it they were all really accepting about it and they, err, were like 'oh yeah that's fine, I don't mind about this kind of stuff that you have and it doesn't really matter'".* (Lines 404-406). *"… kind of all of that worry and stuff.. kind of left my body because I was realised that these are really nice people that I'm hanging out with stuff… so yeah, it's pretty nice" (Line 412-414* *"… the trust is like very important…for like Cystic fibrosis because if you don't trust people then you wouldn't be able to do any of the stuff you need to do with cystic fibrosis… because like you need help to get your meds and stuff… and make them understand what you're going through and… for them to help you*.." (Lines 165-173). **Natalie:** *"So when I was young young… erm… I used to have a friend, I'm still friends with her, we've like, we've been best friends for ages, but umm… she like straight away even though we were young figured it [CF] out … and then once we got older she like knew what I went through…if I didn't want to explain, she would have basically done it for me." (Line 163-168).* **Sarah**: *"When I made new friends I sort of had to explain it…… like the little things, like why I can skip the queue at lunch, why I'm taking these meds now and then…so it was quite... weird… I just sort of avoided making new friends just for the ease of keeping the old ones"* (Line 126-130) *"I think it was easier to keep my friends that I've known forever than make new ones because they of course knew about it [CF] because I went to primary school with them." (Line 132-134).* **Aiden:** *"At school I didn't really tell anyone that I had CF because… looking back on things now I wish I hadn't told nobody but I told a few people… and then it was sort of used against me in a sly way and then I ended up coming out of school because of it..."* (Line 111-116). *" I told them in… year 7. But I only told my best mate and then he told them…and I was like 'oh yeah.. 'cheers for that thanks'…* (Line 265-267).
Finding common ground	**Sarah:** *One [close friend] has a nut allergy… So we've always sort of bonded over being close and knowing somebody who is slightly… not different but has an addition, like something that is additional alongside it. I think it sort of… it sounds stupid but it sort of helped our friendship."* (Lines 48-52). *"It's really weird to actually talk to somebody who completely gets it because when I found out about the gene therapy stuff was coming through like it was legalised... I didn't text my friends, I didn't tell any of my family, the first person I texted was him."* (Lines 87-92). *"I think having someone there who completely gets it, is so worth it… because having somebody you can speak to, who will 100% get it, is so rare."* (Lines 266-268). **Daniel**: *"I dunno, It's not really bothered me much because I've not really known anyone much that I've kind of liked... that I've wanted to meet in person."* (Line 201-202). **Harry:** *"…it makes me feel more safe around everyone because I know that I'm not the only one going through something and I'm not the only one that has… that I'm not perfect and everyone else there is very accepting of all that kind of stuff and it makes me… it's like a very safe environment for me to like talk about stuff".* (Line 450-454). **Aiden**: *"… talking to other people is hard… like when they haven't been through that sort of stuff… they don't know about it… so speaking to someone who goes through the same as you... it's almost like... yeah, you're gonna relate to everything I'm going to tell you."* (Lines 774-778.

Connecting in Other Ways

Natalie: *"…one of my friends loves Minecraft and all the same YouTubers such as [name of Youtuber]… and she got me into it and now we just discuss it every time they go live."*
(Lines 494-496)

Daniel: *"…sometimes I find myself playing games with them a lot more since the pandemic but obviously you can't go round their houses and stuff*."
(Lines 268-269)

Harry: "…*we've found other ways to speak to each other and we all have group chats… on WhatsApp, Xbox and stuff like that.." (Lines 574-578)*

Aiden: *"… I didn't go out at all but like now I don't go out… erm and like we message a lot but like being in lockdown we've been talking on Xbox a lot more..." (Line 562-565).*
"… like FaceTime and that I don't do it with my boy mates... I tend to do it with my girl mates cause they do it more… so sometimes of a night time I'll just go on FaceTime to someone…and then just talk for a bit... sometimes fall asleep on the phone… sometimes not [smiles]. But yeah... stuff like that helps me to keep in touch like... with not slipping back into that black hole…"
(Lines 650-656)
"..I'm in a group chat with about 15 other people with CF.. and I speak to a few more individually who're not in that group chat. (Lines 743-746)

Natalie: *"I was a bit scared at first but we started off erm... we started off on Facebook and then it was on Messenger and then me and this girl I used to be friends with said "oh my god we should put it on Snapchat!" cause everyone uses Snapchat a lot…so me and her discussed and then put it on Snapchat…and now we're all talking on there."* (Line 320-326).
"… My best ever ever friend who has CF who wasn't in it {the online group] but I added her to the group because I knew who she was because we met when we were both [place name redacted]…" (Lines 270-273)

Sarah:

Sarah: 691-695 - we said like every night at one point there were like four oh us [lists four], we were all sort of like on a call and playing random video games, like we'd find a new one each week and be like 'this one looks kind of funny, let's play this one' and then like some of them stuck and we still play them now.

731-745- we usually do a giant call on Whatsapp, 'cause it's the easiest one to have numerous people calling on.. or I actually downloaded Zoom the other day, cause one of my friends was like, 'let's do a Zoom and then we can make presentations, so I'll do one about why there are more than five colours in the rainbow or whatever' and then 'I'll do one on this' so we're all gonna have like a Zoom, almost like we're teachers doing like a little thing like when we were tiny and I was like I would have loved to have done that when I was like five so we were like, let's do it then, let's just do it. 'Cause I think Zoom is quite good, cause one of my friends has like a screen transference, so they can show everything in their camera roll so sometimes we just randomly go through eachothers camera rolls looking at one everyone's stuff we've found. "

686-689- I kept up with all my friends and I kept speaking to everyone during the lockdown, because of course we all literally had nothing else to do. But what was quite nice is that we all developed our own ways of speaking with one another

The relationship between increasing independence and social relationships in the context of CF	**Harry**: *"I can get it all set up and do it all by myself and put it all away and wash it up… so it's much easier on my parents to kind of… they don't need to be there all the time to get me to do my physio properly 'cause I can just do it by myself now."* (Line 268-272).
Experiences of Defiance and Conflict with Others (e.g. family)	**Sarah** *Sarah: "I used to sort of scream at them [parents] like 'This is your fault!', 'I'm not doing it, I don't care'. And they'd just be like OK, well we'll be here when you do. And of course my guilt would be like 'You have to go and do it now'* *Sarah: "Yeah… I think, it was just sort of that one thing on the additional list of things that I couldn't do, that I was just sort of like 'Oh and I cant do that and I cant do that'* *Sarah 503-512: "…erm I remember really detesting it (cross infection control guidelines). I remember being like, I remember being like eight or nine, and hating the idea that I couldn't meet somebody like me or get on with somebody who has the same things as me."* *Sarah:* When discussing the Covid-19 pandemic *"…But I completely ignored my mum and did not shield before the country went into lockdown I was still going to school, up until like the Wednesday, when mum was like: 'There is no way you can go into school now' and I ended up missing my sister's last day which was quite harsh…"* (Lines 682-686) **Aiden** *Aiden: "… erm but its more of the physio side of things that like cause like, they make you have to do everything twice a day like it's just.. it can be very draining.. erm.. because… its not something that any CF person wants to be doing…"* *: "…it can be very easy to just open to top of the meds, tip the liquid away, not do that. And crack the tablets open and not inhale them, with the nebuliser and the physio…" (line 639-642).* *"You have to look at the... it can be quite a vicious cycle because you have to look at the bigger picture without looking too far into it because it's like… I aint gonna live that long anyway so what's the point?... There has to be like… limits to where you see… but then to wanna get the motivation to do it, you have to look into the future…"* (Lines 687-698*)* **Natalie:** *Natalie: "..we wasn't actually meant to go by each other but we just broke the rules a lot… because we didn't actually care at the time… but then we [laughs] we just kept getting told off."*
Independence as a process - Reflections and mixed emotions.	**Harry** *"...yeah, now I've gotten into secondary school its been more independent so…* *I: mmmhmm* *Harry:… ive grown more from secondary school* *I: Mmmhmm* *Harry:… because I havnt had people kind of pestering me about whether I've eaten my lunch, if ive taken the right amounts of [medication name] if I've done all this stuff… so it's made me more independent.. going into secondary school and stuff like that and trips and all of that stuff like that so.. (256-264)*

268-272- "..I can get it all set up and do it all by myself and put it all away and wash it up… so it's much easier on my parents to kind of… they don't need to be there all the time to get me to do my physio properly cause I can just do it by myself now."

282-287 – "…its felt pretty good because I know… I know that because I'm more independent, everyone's less stressed about… kind of the interactions and like the stuff that I'm needing to do and everyone's getting more time to themselves and stuff like that and I'm.. and It makes me feel much happier knowing that everyone's happier that I'm more independent… so that makes kind of me happier about people being happier about me being independent [laughs] so…"

..

Harry: … and it's much easier on [little brother's name] as well, he can have like more time with my parents and stuff because I don't, they don't.. like in the mornings and in the afternoons… because they're not needing to be there next to me …

Harry: "…it was a very pleasant experiences, other than like… having my IVs done…cause everyone was so nice and everyone was so like.. they were generous and stuff like that and it was a very nice experience to be around those kind of people and they'd like only do stuff when I say its ok to.. they wouldn't just take out my needle or put one in without me knowing."

292-297 – Interviewer: And in terms of that so you mentioned the fact that your parents might be a bit less stressed ?

Harry:.. yeah, yeah they'll be less stressed err because theyre not needing to be by my side at all times and they know I can do everything… pretty much everything by myself… and do my own thing at times and yeah.. so they're kind of less worried about me doing stuff right and stuff like that. … its felt pretty good because I know… I know that because I'm more independent, everyone's less stressed about… kind of the interactions and like the stuff that I'm needing to do and everyone's getting more time to themselves and suff like that and I'm.. and It makes me feel much happier knowing that everyone's happier that I'm more independent… so that makes kind of me happier about people being happier about me being independent [laughs] so… yeah, yeah they'll be less stressed err because theyre not needing to be by my side at all times and they know I can do everything… pretty much everything by myself… and do my own thing at times and yeah.. so they're kind of less worried about me doing stuff right and stuff like that.."

303-307 - … like I don't really know how to like… kind of explain it but… it's much easier knowing that i'm more independent that some other people may be and knowing that I am kind of doing.. getting all these stuff my myself but then also getting the help from my parents so it's a pretty good kind of feeling…

422-426- "..it's been Ok… I mean.. cause its had a massive impact on my life and its because of this kind of this kind of symptoms and this illness… it has made my life much better in ways and also not as great in other areas…"

Sarah

"*…But I just sort of realised that there's no point resisting it, 'cause I'm gonna have to do it at some point, I feel like I've just lost the energy to fight with it*." (Line 381-384)

Sarah 529-533 – "I think that's probably the maturity of older age, knowing that I can't meet you and that's OK. I think I probably would have struggled knowing that I couldn't meet them in person when I was younger, cause I just think it's a hard thing to wrap your head around."

Sarah: "Yeah, I think I'm very lucky because my parents always like pushed for me to be able to do stuff. They always made a way around it…"

Sarah: 497-500 "..But as soon as that sort of taken away, it felt like there was a lot less to be hated about it. There was a lot less that I didn't like about it, but other than that, there's very little that impacts me in the everyday."

"…its not a fun thing to live with, I just sort of, I've just stopped resisting it. I'm like Ok, it's just me. It's just something I have to just deal with. The less that I think about the impacts, the smaller it feels…. I sort of.. [sigh] this is really hard to put into words. Umm, sort of living with CF, it's not been an enjoyable experience [laughs], I won't say that, but it's been… Ok." (Lines 439-444)

Natalie

Natalie" "Weird… cause I've got to do everything myself… I've gotta call like... nurses on my own.... well I was gonna say book appointments but my mum does that because I'm too scared to even do that [laughs]. Erm... the other day she [mum] made me do my prescription…"

(Line 12-16)
"Errr.. it was hard but not very hard, but now cause I'm like older… it's Ok to cope with."
(Lines 140-141)
"But now… when I was younger, you had to like.. erm..rely on your parents to do everything for you" (Lines (Lines 142-144)

"I'm always on that ward but they like me too much to want me to actually… so I just go there…they don't want me... umm because I'd have to go to adults and they don't want me to leave…'cause I've been there for ages…"
(Line 356-361)

Daniel

"…theyre helpful… cause theyre not like mean, theyre nice" (Line 189-192)