THE ROLE OF PARENTING INTERVENTIONS IN PROMOTING TREATMENT ADHERENCE IN CYSTIC FIBROSIS

A thesis submitted to The University of Manchester for the degree of

Doctor of Clinical Psychology (ClinPsyD)

in the Faculty of Biology, Medicine and Health

2016

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TOTAL WORD COUNT: 20 557 (excluding abstracts, tables, and references)

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Abstract

The Role of Parenting Interventions in Promoting Treatment Adherence in Cystic Fibrosis

Doctor of Clinical Psychology, the University of Manchester.

Emma Wells. June 2016.

Within the Cystic Fibrosis (CF) literature it is acknowledged that parents play a significant role in supporting children with treatment procedures. Furthermore, a number of parenting variables have been associated with treatment adherence within the paediatric CF population. Interventions that target parenting practices may therefore have the potential to improve CF treatment adherence. Paper one presents a systematic literature review of parenting interventions targeting treatment adherence in children and adolescents with CF. The majority of studies focussed on dietary adherence and overall findings from these studies suggested that combined behavioural and nutritional counselling parenting interventions led to improvements in calorie intake and positive parenting practices. Interventions specifically targeting exercise adherence and interventions targeting multiple aspects of the CF treatment regimen were also shown to improve treatment adherence. The review highlighted that interventions targeting some of the more laborious treatments (i.e. chest physiotherapy) were lacking, as were interventions specifically tailored to the needs of adolescents and their parents.

Over recent years, CF life expectancy has increased substantially due to medical advances. As a result, more children are living into adulthood, therefore needing to adhere to an increasingly complex treatment regime in order to manage increasing symptoms. Adolescence is a particularly challenging time for treatment adherence as children increase their independence and parents begin to allow the child to manage their own disease management. The study described in Paper 2 aimed to explore the acceptability and feasibility of the Self-Directed Teen Triple P parenting intervention within the adolescent CF population. It also explored whether parent-reported treatment adherence, positive parenting practices, parent wellbeing, and child emotional and behavioural functioning were increased as a result of this intervention. Whilst data from two cases indicated increasing trends in treatment adherence and positive parenting practices following the onset of the parenting intervention, uptake and retention to the intervention was poor. Interviews with parents and CF nurses indicated low acceptability and feasibility of the intervention in its current form and a number of adaptations were reported. The study concludes that researchers need to include parents within the design of tailored parenting interventions within this population in order to increase acceptability. Following this, larger scale studies are required to increase the reliability and rigor of research findings in this area.

Paper 3 is a critical reflection and considers both Paper 1 and Paper 2. Within this paper the approaches used, the challenges encountered, and future research are considered.

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Acknowledgements

I would like to offer my sincere gratitude to my supervisor Prof Rachel Calam for her continued guidance and support throughout the thesis process. These thanks are also extended to Dr Clare Murray and Prof Adrian Wells for their support with research design and recruitment. The project would also have not been possible without the continued dedication of the cystic fibrosis nurses and parents who gave their time to take part in the research. Thanks also to my ClinPsyD peers who shared this rewarding and challenging journey with me. Particular thanks are extended to Katie Swift for her support with the quality assessment process.

The biggest thanks are saved for my wonderful family and adored partner, Andy, for their continuing love, kindness, support, and patience throughout the research process.

This thesis is dedicated to my late and dearly beloved father. Your love, laughter, and fatherly pride continues to inspire and encourage me every day.

Paper 1: Systematic Review

A Systematic Review of the Literature Exploring the Use of Parenting Interventions to Improve Treatment Adherence in Children And Adolescents with Cystic Fibrosis

The following paper has been prepared for submission to
'Journal of Clinical Psychology in Medical Settings'
The guidelines for authors can be found in Appendix A.

Word Count: 7378 (excluding tables and references)

Abstract

Objectives. In light of recent advances in the medical care and treatment of children with Cystic Fibrosis (CF) and the need for increasingly complex treatment regimen, the aim of the current review was to provide an up to data synthesis of research studies utilising parenting interventions to improve treatment adherence in children and adolescents with CF. *Methods.* A systematic search of EMBASE, PsycInfo, Medline, Pubmed, and Web of Knowledge databases was conducted. Fifteen studies were included in the review. The Quality Assessment Tool for Studies using Diverse Designs (QUATSDD) was used to guide the assessment of methodological quality. *Findings.* The majority of studies reported outcomes from behaviourally oriented parenting interventions to increase dietary adherence, with other investigations focussing on exercise adherence and more generic treatment adherence. Overall, findings were positive and encouraging. *Conclusions.* Parenting interventions offer potential to improve treatment adherence and associated physical health outcomes as well as parenting practices and parent and child behaviour. Studies were heavily biased towards younger children and were predominantly observational in design, indicating gaps in the literature and the need to interpret these findings with caution. Implications for future research are discussed.

Key words: Parenting Interventions, Cystic Fibrosis, Adherence, Children, Adolescents

Introduction

Cystic fibrosis treatment

Cystic fibrosis (CF) is a progressive multi-system pulmonary disease that occurs in approximately one in 3500 births. The treatment regime is complex, time consuming and laborious, often taking several hours a day to complete (Quittner, Espelage, Ievers-Landis, & Drotar, 2000). Adherence to treatments amongst children with CF is reported to be, on average, below 50% (Modi & Quittner, 2006). A number of factors have been associated with poor treatment adherence in CF, including child age (Ricker, Delamater, & Hsu, 1998), child psychopathology (White, Miller, Smith, & McMahon, 2009), disease severity (Zindani, Streetman, Streetman, & Nasr, 2006), and child illness beliefs (Bucks et al., 2009). Furthermore the treatment regime in cystic fibrosis can be challenging for the wider family and a considerable body of research findings has demonstrated that poorer family functioning is associated with poorer treatment adherence in cystic fibrosis (DeLambo, Ievers-Landis, Drotar, & Quittner, 2004; Everhart, Fiese, Smyth, Borschuk, & Anbar, 2014).

The changing landscape of CF treatment

Although child CF treatment adherence has been reported to be poor for many years, it needs to be considered within the context of medical advances. Since the discovery of the CF gene in 1989 (Davis, 2006), advances in medical research have accelerated therapeutic improvements, and the mean predicted survival rate for individuals living with CF is now between 30-40 years of age. This is a 10-year improvement compared with only a decade ago (Sawicki & Tiddens, 2012). The introduction of new-born screening within the UK in

2007 has led to the adoption of more complex and aggressive therapies earlier in life. As life expectancy increases, more children will continue to live into adulthood, therefore needing to adhere to increasingly complex treatment regimes in order to manage increasing symptoms (Sawicki, Sellers, & Robinson, 2009). These factors are likely to increase treatment burden and associated treatment adherence difficulties in both earlier and later childhood years (Agh, Inotai, & Meszaros, 2011). Therefore, although adherence difficulties have been reported consistently across the past two decades, the social, psychological and medical context surrounding disease coping and adherence behaviours is likely to have changed and such factors are important to bear in mind when reviewing the literature within this area.

The potential role for parenting interventions

Parents play a central role in paediatric CF treatment adherence (Eddy et al., 1998). Studies over the past two decades have demonstrated that parents of children with CF continue to engage in a higher frequency of ineffective parenting strategies, and that parents who are more coercive, negative, and inconsistent are significantly more likely to experience adherence difficulties (Sanders, Patel, Le Grice, & Shepherd, 1993; Stark, Bowen, Tyc, Evans, & Passero, 1990; Stark & Powers, 2005). In contrast, positive parental interactions, positive attention, praise, clear instruction and avoidance of negative and conflictual interactions continue to be associated with greater adherence to CF treatments, such as chest physiotherapy (Butcher & Nasr, 2014).

Across childhood chronic illnesses a number of research studies have investigated the potential effectiveness of parenting interventions in assisting with child medical

adherence, with a meta-analysis showing that interventions combining educational and behavioural elements have the largest effect sizes (Graves, Roberts, Rapoff, & Boyer, 2010). Only one systematic review has been conducted to investigate the usefulness of parenting interventions within the paediatric CF population (Bernard & Cohen, 2004). This review of nine studies demonstrated that behavioural techniques including token economies, contingency management and behaviour modification principles positively influenced adherence to various elements of the paediatric CF treatment regime. However, the majority of these findings were from studies using toddlers and younger children with CF, which may be reflective of the lower life expectancy and less advanced medical management of CF in the years covered by this review.

Given that there have been a number of advances in CF treatment in the last ten years, interventions tailored to support increasing demands may have generated more research in recent years. Furthermore, the literature selection and review process was not fully transparent within this previous review, making it difficult to be confident in the research picture portrayed (Moher, Liberati, Tetzlaff, & Altman, 2009). Therefore, a more up-to-date review of the literature is required.

Aims of the current review

The aims of this literature review were to systematically search the literature in order to provide an updated and detailed narrative account of the range, format and delivery of the full spectrum of parent-directed psychosocial interventions designed to improve treatment adherence in children and adolescents with CF. A further aim was to explore any changes in the types and focus of parenting interventions over the past two decades,

in line with medical advances and increased life expectancy. Finally, the current review aimed to highlight any outstanding areas for further research. A narrative synthesis was used because it was anticipated that an array of research designs would be incorporated, therefore making statistical comparisons difficult.

Method

Literature Search

The literature review included publications from 1990-2015 (last search completed on 27th September 2015), in order to encompass the changes in life expectancy and medical advances within this period. Computerised searches were conducted using EMBASE, PsycInfo, MedLine, PubMed and Web of Knowledge databases. Reference lists of included papers were examined for additional relevant articles, and citation searches were completed on included papers. Databases were searched using Boolean logic using the following search terms: 'Cystic fibrosis', AND 'adherence' OR 'compliance' OR 'concordance', AND 'parent*' OR 'family' OR 'caregiver', AND 'training' OR 'program*' OR 'intervention' OR 'support' OR 'education' OR 'psychoeducation' OR 'therapy'.

Inclusion and exclusion criteria

Studies were required to report a parenting intervention that was informed by psychological theory and/ or principles aiming to alter CF treatment adherence. A quantifiable measure of treatment adherence (e.g., validated questionnaire, self-report diary, coded observations) was required as either a primary or secondary outcome variable. The review included a diverse range of study designs, such as randomised controlled trials, quasi-experimental designs, and observational studies given the limited

literature and quality of evidence available. Articles were excluded if they were not written in English and if they were unpublished because such studies may not have undergone peer reviews to establish research quality. Studies reporting on outcomes for parents of children with comorbid intellectual disabilities were excluded because this would likely require more specifically tailored and adapted interventions.

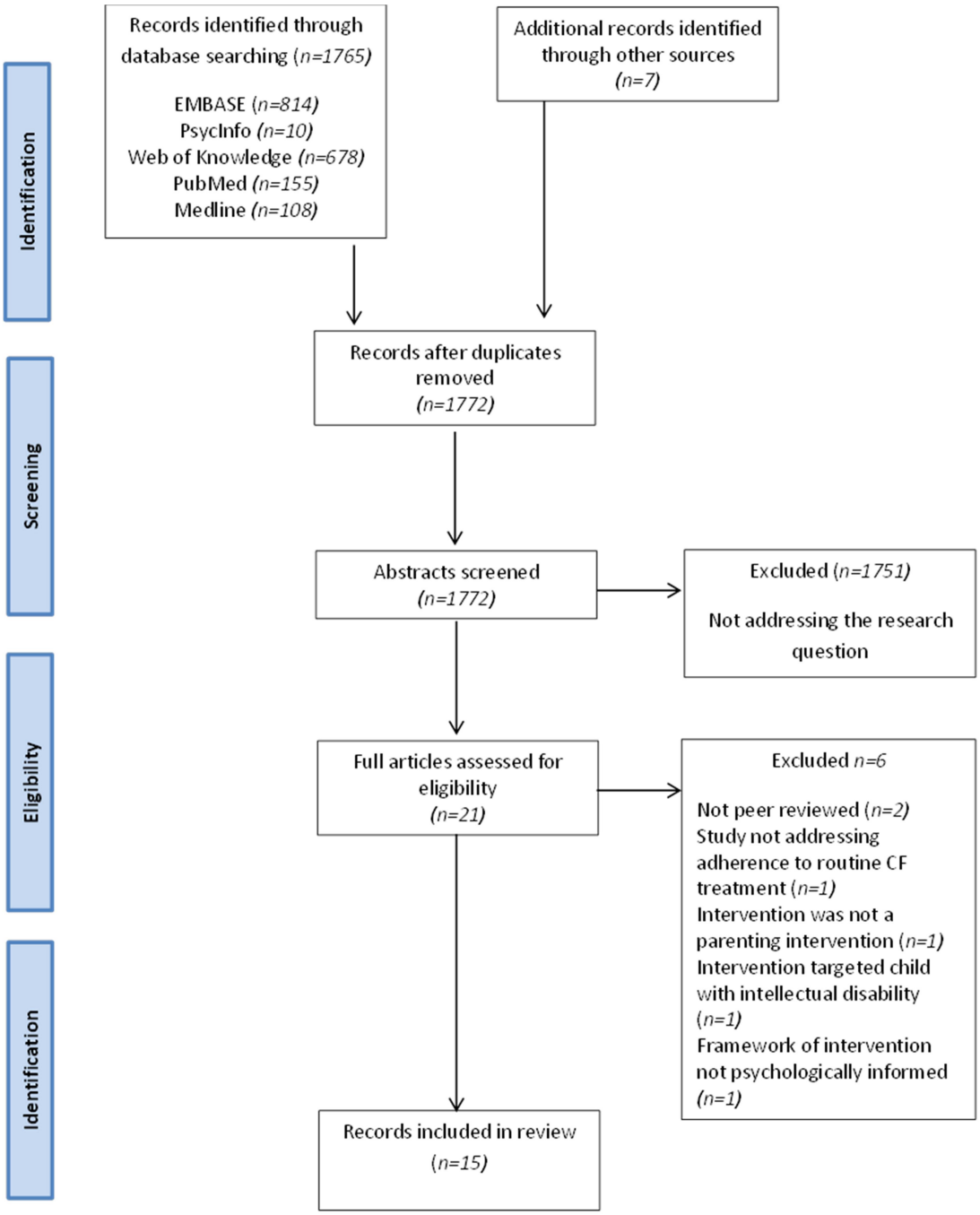
Screening

The abstracts of identified articles were screened by the first author (EW) to determine whether they were relevant to the review. Full-text articles were then retrieved for those articles deemed relevant and were scrutinised in further detail. Seven of the full text articles obtained (47%) underwent a second screening process by an independent researcher (KS) to increase the reliability of studies included in the review. Disagreements regarding inclusion were discussed and resolved by consensus after referring to the inclusion and exclusion criteria, leading to 100% agreement over the papers included. A flowchart outlining the screening and selection process for inclusion in this review is outlined in Figure 1.

Data extraction and synthesis

Information of relevance to the research question was extracted from all included articles via a tabulated proforma (Appendix B). Extracted data were then collated and reviewed, and the findings from each article were synthesised to provide a comprehensive overall review of the literature. The findings of randomised and non-randomised designs were synthesised separately in line with Cochrane guidance on the synthesis of systematic reviews (Reeves, Deeks, Higgins, & Wells, 2008).

Figure 1. Search results (Figure adapted from PRISMA group guidelines, Moher et al., 2009)



Quality Assessment

The methodological quality of studies was assessed using The Quality Assessment Tool for Studies with Diverse Designs (QATSDD; Appendix C) (Sirriyeh, Lawton, Gardner, & Armitage, 2012). The 14 items of the QATSDD applicable to quantitative research studies were rated on a 4-point scale from "not at all" (0) to "complete" (3). Percentage scores were calculated using the actual score and the maximum total score of 42. Appendix D details individual quality ratings for each paper. Papers scoring over 75% were considered "high" quality, those between 50% and 75% "good", 25%–50% "moderate", and below 25% "poor". An independent researcher rated 7 of the 15 included papers (47%) and any discrepancies were resolved through discussion. Cohen's Kappa was 0.71 indicating 'substantial' interrater agreement. See Table 1 for quality ratings.

Results

Study Characteristics

Fifteen studies were included in the review. Study characteristics are presented in Table 1, in chronological order, and full references are provided in the reference list. All studies were carried out in the USA and sample sizes varied from single case illustrations to larger controlled trials, with the largest sample size being 199 parent-child dyads. Child age ranged from 10 months to 18 years of age, although only one study included children aged above 12 years. Gender prevalence averaged at 66% female versus 34% male children across studies that reported this information, although there was substantial variation in representation of genders across these studies. Six studies reported the ethnic origin of participants, with the majority coming from White backgrounds.

Eleven studies reported multicomponent interventions using a behavioural intervention combined with counselling about nutrition. Three studies utilised a pure behavioural approach and one study utilised a multi-faceted self-directed educational approach. The majority of intervention studies were carried out in outpatient clinics, with remaining studies taking place via telehealth, or in family homes and paediatric inpatient wards. The modality of interventions included a majority of either face-to-face group or individual sessions, with one study being self-directed by parents. Different domains of treatment adherence were examined, with the majority of studies addressing energy intake, two studies addressing general treatment adherence, and one examining exercise adherence. The majority of researchers used non-randomised observational research designs (*n*=9) with a small number of researchers using randomised controlled and pre-post comparison group designs (*n*=6).

Main Findings

Interventions addressing energy intake

Disease factors, such as frequent respiratory infections (leading to an increased metabolic rate) and malabsorption of dietary fat, place individuals with CF at high risk of inadequate nutritional intake to provide satisfactory weight gain and growth trajectories (Durie & Pencharz, 1989). Individuals with CF are required to consume between 120-150% of the recommended daily energy intake in order to optimise their physical wellbeing. Unfortunately, adherence to dietary recommendations has been reported to range between 12% and 16% (Mackner, McGrath, & Stark, 2001) and research has shown that individuals can find it difficult to meet these increased energy intake demands. Negative mealtime behaviours can further impede adherence in this domain (Powers et al., 2005).

Table 1. Characteristics of Included Studies

Authors	Number of participants	Characteristics of participants	Design	Description of intervention	Setting	Measures	Key findings	Quality rating (QATSDD)
Randomised Controlled Trials								
Powers et al. (2015) USA	Parents of 78 children (36 experimental, 42 control)	**Mean age:** Intervention group =3.8 yrs .Control group =3.7 yrs **Gender:** Intervention group = 55% female. Control group = 56% female **Ethnicity:** Intervention group =100% White. Control group = 97% White, 3% Hispanic **Socioeconomic status:** Not specified	RCT	Nutrition counselling + behavioural intervention: 7x1 weekly sessions followed by 4x1 monthly sessions	Face to face in CF outpatient clinic (group or individual nature not specified) or via telehealth	Energy intake via weighted food diaries, height, weight	From pre-treatment to post-treatment, the intervention increased daily energy intake by 485 calories vs 58 calories for the control group. Mean weight change (Weight *z* scores) in intervention group from baseline to post-treatment was 0.12, compared to 0.06 in control group. Mean change in height (height *z* scores) from baseline to follow up in intervention group was 0.09, compared to 0.02 for control group.	High
Stark et al. (2009) USA	Parents of 67 children (Behavioural + nutrition group=33, nutrition control group=34)	**Mean age:** Behavioural group = 7.5 yrs. Control group = 7.4 yrs **Gender**: 1Behavioural group = 55% female. Control group = 45% female **Ethnicity:** Behavioural group = 100% White. Control group = 94% White **Socioeconomic status**: Not specified	RCT	Nutrition counselling + behavioural intervention: 5x1 weekly sessions	Face to face groups in CF outpatient clinic	Energy intake via parent-rated weighted food diaries, standardised body mass index and height scores, pulmonary function, Parent Satisfaction Questionnaire	Behavioural group achieved significantly greater increase in daily caloric intake than children in control group. At post-treatment, children receiving behavioural plus nutrition education averaged 383 more calories per day than children in nutrition group. Significantly greater improvement in BMI at post-treatment in behavioural intervention group.	High
Powers et al. (2005) USA	Parents of 10 children (behavioural group= 4, control group=6).	**Mean age:** Behavioural group = 35.2 mths. Control group = 28.1 mths **Gender:** 1Behavioural group = 25% female. Control group = 50% female **Ethnicity:** Behavioural group = 100% White. Control group = 100% White **Socioeconomic status:** Not specified	RCT	Nutrition counselling + behavioural intervention: 6x1 weekly sessions	Individual face-to-face session in outpatient CF clinic	Energy intake via weighted food diaries, height, weight.	At post-treatment, behavioural group reported significantly higher mean energy intake per day compared with control group. At post-treatment all 4 behavioural group participants met 120% RDA goal. Only 1 participant from control group met this goal. Children who received behavioural and nutrition intervention continued to maintain clinically significant increase in energy intake at 3 and 12 months after treatment	High

Authors	Number of participants		Design	Description of intervention	Setting	Measures		Quality rating (QATSDD)
Powers et al. (2003) USA	Parents of 8 children	**Mean age:** Range = 12-36 mths (means not stated) **Gender:** Not specified **Ethnicity:** Not specified **Socioeconomic status:** Not specified	Small Scale RCT	Nutrition counselling + behavioural intervention: 4x2-3 weekly sessions + 4 sessions spread regularly through rest of year	Individual face-to-face session in outpatient CF clinic	Energy intake via weighed food diaries, weight, height, Behavioural Paediatrics Feeding Assessment Scale.	Paired samples t-tests indicated that behavioural intervention did not lead to significantly greater improvements in calorie intake than the control group. Children in behavioural group demonstrated an increase of 406 calories per day from pre- to post-intervention, compared to 285 calorie intake in nutrition alone group. Children in the BEH group showed an increase of 31% in RDA energy per day, and children in the NTR group showed a 22% increase.	Good
Non-RCT studies								
Hourigan et al. (2013) USA	Parents of 4 children	**Mean age:** 25.75 mths **Gender:** 75% female **Ethnicity:** 100% White **Socioeconomic status:** 75% SES status IV, 25% SES status III (Hollingshead index)	Case series (AB design)	Nutrition counselling + behavioural intervention: 6x1 weekly sessions	Face to face groups in CF outpatient clinic	Energy intake via weighted food diaries, video recordings of 1 meal per week coded using Dyadic Interaction Nomenclature for Eating system, height, weight	For the 2 children initially malnourished: Increased Body Mass Index (BMI), Energy intake, and weight from post-intervention (gains maintained at follow-up) For the 2 children of adequate weight : Increased diet quality and age appropriate food choices for one child. No change reported for other child.	High
McClellan et al. (2009) USA	Parents of 2 children	**Mean age:** 4.75 yrs **Gender:** 100% female **Ethnicity:** Not specified **Socioeconomic status:** Not specified	Case series (ABAB design)	Behavioural intervention: length dependent upon time taken to achieve stability in child compliance levels.	Telehealth	CF problem checklist, observational assessment of child's treatment compliance coded at 5 second intervals	Increase in observed treatment compliance in all domains for one child, with other child showing increases in all domains except chest physiotherapy. Decline in parent rated CF problem intensity and reduced desire for professional support. Parents preferred time out to typically used strategies.	Good
Bernard et al. (2009) USA	Parents of 3 children	**Mean age:** 11 yrs **Gender:** 100% female **Ethnicity:** 100% White **Socioeconomic status:** 5% parental income $25000-$40000, 25% parental income $40000-$60000	Case series (ABAB design)	Behavioural intervention: 3x2hr sessions	Face to face in patient home	Children's OMNI scale of perceived exertion, pedometer ratings, parent and child self-report exercise diaries (minutes of exercise per day)	Higher pedometer readings for all children during intervention phases when compared to baseline phases. Higher frequency of days exercised during intervention phases, when compared to baseline. All participants showing consistent gains in exercise amount at 1 and 3 mth follow-up when compared to baseline	High

Authors	Number of participants		Design	Description of intervention	Setting	Measures		Quality rating (QATSDD)
Piazza-Waggoner et al. (2006) USA	Parent of 1 child	**Mean age:** 21 mths **Gender:** 100% female **Ethnicity:** Not specified **Socioeconomic status:** Not specified	Case series (AB design)	Nutrition counselling + behavioural intervention: 7x 1 weekly phone calls + handouts posted to parents	Telehealth	Energy intake via weighted food diaries, weight and height.	From pre- to post-treatment, energy intake increased (93% RDA to 132% RDA). Across follow-up assessments, energy intake continued to increase to 164% RDA at most recent assessment. Child exceeded the growth velocity for height at all three time points post-intervention.	Moderate
Stark et al. (2003) USA	Parents of 7 children (Behavioural group=3, control group=4)	**Mean age:** 10 yrs **Gender:** Not specified **Ethnicity:** Not specified **Socioeconomic status:** 57% business/ professional. 29% skilled craftsmen/ clerical/ sales. 14% semi-skilled (Hollingshead scale)	Case series	Nutrition counselling + behavioural intervention: 5x 1 weekly sessions	Face to face groups in CF outpatient clinic	Energy intake via weighted diet diaries, weight and height, Global Rating Scale for Feeding situations , Family Stress Scales, Role play Inventory of Situations and Coping Strategies	Children in behavioural intervention demonstrated greater increase in daily caloric intake and weight gain than the nutritional intervention. Parent and child feeding behaviours remained stable across assessment points, with no differences between groups. Maternal mood during mealtimes improved in both conditions. Mean family stress scores decreased from baseline to posttreatment in both conditions. Children in behavioural intervention demonstrated a decrease in frequency and difficulty of eating, and weight concerns. Children in nutrition group demonstrated reduced frequency of eating concerns but no change in difficulty ratings, and a decrease in competency scores.	Good
Bartholomew et al (1997) USA	199 parent-child dyads included (experimental group=104, control group= 95)	**Mean age:** 8.6 yrs **Gender:** 53% female **Ethnicity:** Not specified **Socioeconomic status:** All parents rated as 'middle class' (Hollingshead scale)	Quasi-experimental pre-post non-equivalent group design	Educational programme: Self-paced independent learning for parents	At home (parent directed)	Test of knowledge of CF questionnaire, Self-efficacy Expectation Scales, Self-Management Questionnaire of CF, Interpersonal Coping and Problem Solving scale, Means-Ends Problem Solving scale, pulmonary function, weight, height, Child Behaviour Checklist, Quality of Wellbeing Scale, Impact on Family Scale, Parenting Stress Index	At post-intervention, parents in educational group demonstrated significantly higher scores than control group on measures of caregiver, adolescent and child disease knowledge, caregiver and child self-efficacy, adolescent and parent self-management, parental problem solving, child behaviour scores, and pulmonary functioning.	Good

Authors	Number of participants		Design	Description of intervention	Setting	Measures		Quality rating (QATSDD)
Stark et al. (1996) USA	Parents of 9 children (behavioural group=5, control group=4).	**Mean age:** Behavioural group = 7yrs 3 mths. Control group = 6 yrs 3 mths **Gender:** Not specified **Ethnicity:** Not specified **Socioeconomic status:** Behavioural group = mean SES category III. Control group = mean SES category II (Hollingshead Scale)	Quasi-experimental pre-post non-equivalent group design	Nutrition counselling + behavioural intervention: 6x 1 weekly sessions	Face to face groups in CF outpatient clinic	Energy intake via weighed food diaries, weight, height, pulmonary functioning, resting energy expenditure, physical activity via Caltrac electronic accelerometer	Pre- to post-intervention calorie intake increases were significantly greater in the behavioural intervention group. Children in the behavioural group showed significantly more weight gain than children in control group. Children in both groups showed increases in absolute height. No changes were found on any of the physiological measures or body fat.	Good
Stark et al. (1994) USA	Parents of 2 children	**Mean age:** 2yrs 11 mths, 5 yrs 10 mths **Gender:** 100% male **Ethnicity:** Not specified **Socioeconomic status:** Both parents rated category II (Hollingshead scale)	Case series (AB design)	Behavioural intervention: 9x90minute weekly sessions	Individual face-to-face session in outpatient CF clinic	Global Rating Scale of Feeding Situations, energy intake via daily weighted food diaries, weight	Parental attention to disruptive mealtime behaviours reduced immediately following implementation of intervention. Parental mealtime control increased, and was maintained at follow-up. Appropriate mealtime behaviours increased, and disruptive mealtime behaviours decreases after intervention implementation in both cases. One child increased calorie intake substantially from pre- to post-intervention. The other child showed slight decrease in calorie intake and an increase in calories taken from developmentally appropriate foods.	Good
Stark et al. (1993) USA	Parents of 3 children	**Mean age:** 3 yrs 11 mths, 6 yrs 5 mths, 8 yrs 5 mths **Gender:** 66.6% female **Ethnicity:** Not specified **Socioeconomic status:** All parents rated category II (Hollingshead scale)	Case series (AB design)	Nutrition counselling + behavioural :intervention: 6 x 1 weekly sessions	Face to face groups in CF outpatient clinic	Energy intake via weighted food diaries, height, weight, pulmonary functioning, pace of eating during recorded meal time observation	Paired t-tests demonstrated significant improvements in calorie intake from pre-to post-intervention, with gains being maintained at follow-up. All children were below RDA energy intake before intervention, with two exceeding RDA energy intake at post-intervention. Increases in weight were observed (mean increase = 0.66kg) between pre and post intervention across all children. Lung function remained relatively stable across treatment.	Good

Authors	Number of participants		Design	Description of intervention	Setting	Measures		Quality rating (QATSDD)
Singer et al. (1991) USA	Parents of 4 children	**Mean age:** 10 mths, 3 yrs 6 mths, 13mths **Gender:** 75% female **Ethnicity:** 75% White. 25% Black. **Socioeconomic status:** Not specified	Case series (AB design)	Behavioural intervention: 8-33 days (depending on length of stay)	Individual face to face sessions in inpatient paediatric ward	Energy intake via nurse completed menu records, height, weight	Mean percentage energy intake increased from 54% to 93% for the four participants. 75% of children demonstrated continued catch up in weight and growth post discharge, ranging from 10th to 5th percentile.	Moderate
Stark et al. (1990) USA	Parents of 5 children (2 Siblings)	**Mean age:** 8 yrs3 mths, 8 yrs 7 mths, 10 yrs 1 mth, 12 yrs 1 mth, 5 yrs 10 mth. **Gender:** 60% female **Ethnicity:** Not specified **Socioeconomic status:** 60%category II, 20% category III, 20% category IV (Hollingshead Scale)	Case series (AB design)	Nutrition counselling + behavioural :intervention: 6 x 1 weekly sessions	Face to face groups in CF outpatient clinic	Energy intake via weighted food diaries, height, weight, pulmonary functioning	A significant increase in daily calorie intake from baseline to posttreatment, and baseline to nine month follow-up was demonstrated. Significant increases in weight from pre-to post intervention were also demonstrated Average pre-intervention weight = 24.7 kg. Significant increases in child growth from pre-to post-intervention were also demonstrated.	Good

Randomised controlled trials (RCTs)

Four clinical trials examined the role of parent-based interventions to improve adherence to dietary recommendations (Powers et al., 2015; Powers et al., 2005; Powers et al., 2003; Stark et al., 2009). A combination of nutritional counselling and behavioural parenting approaches were used. Across these studies nutritional counselling broadly covered the following areas: the interplay between nutrition, lung function and physical health status, strategies to boost nutritional intake, education about the effects of enzymes and vitamin deficiency on the body, strategies for introducing new foods into their child's diet, and strategies for maintaining nutritional intake during periods of illness. The behavioural training provided in these four trials was based upon social learning theory principles and taught parents to give clear and direct commands to their child, the use of contingent and differential attention to increase food variety, the use of consequences, such as ignoring and time out for non-adherent behaviours, and problem solving skills. Across these four studies, children were set individualised calorie goals for each meal, and main outcome measures consisted of calorie intake (as measured by weighted food diaries) and changes in height and weight.

Two of these RCTs targeted younger children aged 12 to 18 months. The first of these was a pilot randomised controlled trial conducted by Powers et al. (2003). Eight parents were randomly allocated to an eight session nutritional counselling intervention or a combined nutrition plus behavioural intervention. At post-intervention children in the behavioural group demonstrated an average increase of 406 calories per day and children in the nutrition group exhibited an increase of 285 calories per day. Furthermore, children in the behavioural group showed an increase of 31% in their recommended daily allowance

(RDA) of energy per day, and children in the nutrition counselling group showed a 22% increase. These results suggest that more substantial improvements were made following the behavioural intervention. A statistical comparison of calorie intake from pre to post-intervention in the behavioural group approached significance. A similar comparison for the nutrition intervention was reported as not significant; however, the authors did not explicitly state the statistical results from this test. The small sample sizes used in these tests may lack sufficient power to detect statistically significant change.

An increase in weight from pre-intervention (nutrition group *M*= 10.1kg; Behavioural group M=11.6kg) to post-intervention (nutrition group *M*= 12.8kg; Intervention group *M*=14.1kg*)* was demonstrated in both groups. The authors reported that weight trajectories did not differ between groups, although statistical comparisons were not reported. There was substantial variability in height trajectories in both groups from pre- to post-intervention. Post hoc analyses using paired *t*- tests for the combined sample's pre- and post-intervention data indicated that the nutritional component common to both groups was effective, demonstrating significant increases in calorie intake and weight gain pre- to post-treatment. This suggested that increasing the comprehensiveness and frequency of nutritional support may in itself be beneficial in improving nutritional outcomes for young children with CF. However, the lack of a standard care control group makes it difficult to draw firm conclusions about which components of the behavioural and nutritional protocols were effective.

A second study by Powers et al. (2005) replicated this study by comparing a combined nutritional counselling and behavioural intervention with a standard care control

condition; the latter involving scheduled clinic visits every three months. Ten parents of children aged 18-24 months were randomly allocated to one of the two treatment conditions. At post-treatment, the behavioural group reported a significantly higher mean daily energy intake than the control group. The change in energy intake was 842 kcal/day for the behavioural intervention and 131 kcal/day for the control group. No individuals in either group were achieving the recommended 120% RDA energy per day prior to intervention; however, all four participants in the behavioural condition had achieved this at post-intervention, compared to only one of the participants in the control group. Treatment gains were maintained at three and 12 months, with 89% of participants sustaining 120% RDA energy per day at 3 months and 100% at 12 months.

The small sample sizes used in these pilot randomised controlled trials (Powers et al., 2003; Powers et al. 2005) limits the reliability and generalisability of the conclusions drawn. It is also possible that the significant findings of Powers et al. (2005) could be due to increased monitoring of calorie intake or contact with services, rather than the intervention characteristics, as contact with services was not controlled for between the two interventions.

Two further RCTs have been conducted which include larger sample sizes. The most recent study (Powers et al., 2015) included parents of 78 children with CF and pancreatic insufficiency aged 2-6 years. In this study parents were randomised to either a behavioural plus nutrition counselling intervention or an education and attention control group. From pre-treatment to post-treatment, the behavioural and nutrition counselling intervention increased daily energy intake by 485 calories, whereas the control group

demonstrated an increase of 58 calories per day. The pre to post intervention difference between the two groups was statistically significant and was maintained at 12-month-follow-up. Although no significant differences were found between the two groups in terms of weight change, the behavioural group showed significantly greater increases in height following the intervention. However, the broad range of nutritional statuses used in this study may have impacted the magnitude of change possible at group mean level, thereby potentially distorting the clinical picture.

Methodological Quality of RCTs

Three out of the four RCTs (Powers et al., 2015; 2005; Stark et al., 2009) obtained a 'high' quality rating on the QATSDD (Sirriyeh et al., 2012). All of these studies reported clear, detailed and explicit information about the research aims and objectives. They reported clear recruitment and data collection information, and gave sound reasoning for the research methodologies and analyses used. The fourth RCT (Powers et al., 2003) obtained a 'good' quality rating. This slightly lower rating was predominantly due to the inappropriate use of statistical comparisons on a very small sample of participants, which may have biased the interpretation of research findings. Furthermore, justifications for the use of statistical analysis and its associated weaknesses were limited within this paper.

Non-randomised studies

A number of case series designs have also been used to assess the influence of behavioural parenting programmes on dietary adherence. Stark et al. (1990) evaluated the use of a six-week group-based behavioural and nutrition program to increase calorie

consumption in five mildly malnourished children aged 5-12 with CF. A multiple baseline AB case series design was used to systematically target calorie increase across snack, breakfast, lunch and dinner. A changing criterion design evaluated the increase in total daily calorie intake to ensure that children did not compensate for increasing calories during the target meal by decreasing calories during other meals. Children attended group sessions in parallel with their parents and were provided with child-friendly adapted information to complement parent sessions. Stark et al. (1993) conducted a systematic replication of this study with a few modifications, such as adding a relaxation component to address abdominal discomfort associated with eating and using a longer two-year follow-up period.

Results from both studies showed an increase in calorie consumption following the behavioural intervention. Children in Stark et al.'s (1990) study showed an average increase of 1050 calories per day (range: 527 to 1,475 kcal/day), and children in Stark et al.'s (1993) study demonstrated an average calorie increase of 46.9%. In both studies, these calorie gains were maintained at nine-month and two-year follow ups. Stark et al. (1990) conducted a paired samples t-test that demonstrated a significant increase in weight from pre-intervention (*M*=24.7kg) to post-intervention (*M*= 26.8kg), with improvements in weight also being demonstrated by Stark et al. (1993). Stark et al. (1990) also found a small but non-significant increase in pulmonary functioning, whilst Stark et al. (1993) found an increase in the pace of eating at mealtimes.

In a similar study using a quasi-experimental pre-post non-equivalent groups design, Stark et al. (1996) randomly assigned a small group of parents of children aged 5-10 years to

either a behavioural plus nutrition education programme or a waiting list control group and found that increases in calorie intake and weight were significantly greater in the behavioural intervention group compared with the control group. During the intervention period, children in the behavioural group increased their average energy intake by approximately 1000 calories per day, which the control group increased their intake by 244 calories. The more systematic nature of this design, alongside the addition of a control group, adds further weight to the claims made by previous studies conducted by Stark et al. (1990, 1993); however, intervention and control groups used in Stark et al.'s (1996) study were not equal on measures of calorie intake and weight percentile at baseline, with the treatment group consuming more calories and attaining greater weight percentiles than the control group before intervention. These differences may indicate mealtime behavioural differences between the two groups that led the treatment group to be more responsive to the intervention.

Although some of the above case series designs provided anecdotal evidence that behavioural interventions had positive impacts upon parent-child mealtime behaviours, none measured this quantitatively. Stark, Powers, Jelalian, Rape and Miller (1994) conducted a further replication of their previous studies, adding a quantifiable measure of parent and child mealtime interactions, the Global Rating Scale for Feeding Situations (GRSFS). To separate the effects of the nutritional and behavioural elements used in previous interventions, the authors solely examined the effectiveness of the behavioural elements of treatment using a case series of two parents. Following the implementation of the intervention, parents showed lower ratings of attention towards negative mealtime behaviours, which occurred in a multiple baseline fashion that was maintained

at follow-up. Positive child mealtime behaviour increased following the implementation of the intervention, with both children showing a decrease in disruptive behaviours, although these behaviours were still variable throughout. One child's calorie intake and weight increased and was maintained at follow-up. The other child showed slight decrease in calorie intake between pre-post measurement points, but demonstrated an increase in more developmentally appropriate feeding behaviours. Considering the very small sample size, these findings provide tentative internal validity for the behavioural intervention, demonstrating that the targeted child and parent behaviours were likely to have been altered as a result of the intervention.

As the behavioural intervention used in previous studies by Stark and colleagues (Stark et al., 1990; Stark et al., 1993; Stark et al., 1996; Stark et al., 1994) was primarily designed to target school aged children, a further study by Hourigan, Helms, Christon and Southam-Gerow (2013) assessed the feasibility of a developmentally adapted version of this intervention in order to address nutritional adherence in children aged 18-36 months. Adaptations were made to account for normative toddler development and toddler feeding information as well as developmentally appropriate adaptations to evidence based behavioural principles. As well as the use of weight, height and food diaries, the researchers coded video recordings of one meal per week using the Dyadic Interaction Nomenclature for Eating system (Stark et al., 2000). This allowed the researchers to further explore the changes in parent and child feeding behaviours.

For two initially underweight participants, nutritional goals focussed around increasing energy intake and weight. Results showed that both of these children increased their

calorie intake and Body Mass Index's (BMI) throughout the intervention, with one child increasing from the 26th percentile to the 51st percentile and the second child increasing from the 10th to 61st percentile for BMI. Increases in RDA energy intake for these two children were also demonstrated and maintained at follow-up. Observable behavioural outcomes were only reported for one of these children who showed a notable downward trend in negative mealtime behaviours, such as food refusal, crying and away from the table behaviours throughout the intervention and follow-up. The other two children involved in the study had adequate to overweight body compositions but consumed diets that were considered to be of poor nutritional quality or developmentally inappropriate. The scope of the intervention was therefore extended to address a broader range of paediatric feeding problems, including those associated with paediatric obesity and food texture acceptance. Progress was made by one of these children who increased the level of non-pureed food consumed from 0-30% pre-intervention to 90-100% post-intervention. The other child showed no improvements in the level of nutritious foods consumed following the intervention.

Although it is acknowledged that these researchers were attempting to be flexible in how to apply behavioural principles to individual family needs, in doing so the initial focus of the intervention to improve calorie intake was diluted. It is probable that the influences and needs of families with a child who is underweight versus overweight are quite different, thus requiring separate investigations. The lack of parent and child mealtime behaviours reported for three out of the four children makes it difficult to ascertain how the behavioural intervention impacted upon these factors.

A further two case series studies have adapted the use of behavioural parenting interventions for parents unable to attend regular outpatient visits (Piazza-Waggoner, Ferguson, Daines, Acton, & Powers, 2006) and for parents of children who required in-patient admission for malnutrition (Singer, Nofer, Benson-Szekely, & Brooks, 1991). Such studies are informative because they allow for the examination of clinical sub-groups who may be behaviourally distinct from those groups who are motivated and able to attend regular out-patient appointments. Singer et al. (1991) recruited parents of children (all aged under five years) residing on a medical-behavioural ward for complex medical difficulties due to feeding and growth problems. For each child a minimum of three observed meals established baseline child and parent mealtime behaviours. A purely behavioural treatment took place over a period of 8-33 days, depending on length of admission. There was an increase in daily calorie intake from a mean of 54% at enrolment to 94% at discharge. Furthermore, follow-up data collected between 7 and 24 months post-discharge showed that three out of the four children demonstrated continued catch up in weight and growth post-discharge, ranging from 10th to 15th percentile. Unfortunately, sufficient detail regarding the behavioural intervention protocol and the dosage of the intervention were not provided making it difficult to ascertain the mechanisms by which this intervention may have been helpful. It is also possible that other factors related to residing on a medical ward may have influenced greater adherence, such as additional support from the medical team.

Piazza-Waggoner et al. (2006) utilised a telehealth approach to address nutrition adherence difficulties in a 21-month-old child with a number of food allergies. These additional factors made dietary adherence a particularly challenging aspect of the

treatment. A pure behavioural intervention using differential attention, contingency management, limit setting and problem solving was delivered via seven weekly telephone calls and additional handouts were posted to parents each week. From pre- to post-treatment, energy intake increased by on average 503 calories per day with an increase from consuming 93% RDA energy intake pre-treatment to 132% post-treatment, which was maintained at follow-up. As this study reports a single case, the results provide very tentative support that evidence-based behavioural treatment for toddlers and pre-schoolers with CF can be modified to address individual barriers to the delivery of optimal nutrition care. However, the authors note that the participating parent was highly motivated and intelligent, with good family support. The inclusion of parents from an array of differing socio-cultural backgrounds would allow for more careful examination of the wider feasibility and acceptability of this intervention.

Interventions addressing exercise adherence

Non-randomised studies

Only one study examined the effectiveness of a parenting intervention to assist with exercise adherence; a treatment domain that is often used to supplement other airway clearance methods, such as chest physiotherapy (Bernard et al., 2009). This reflects a continuing trend in the lack of studies found in this area, because very few studies were also found in the previous review (Bernard & Cohen, 2004). Bernard, Cohen and Moffett (2009) used an ABAB case series reversal study using three parents of school aged children who were not adhering to exercise regimes despite standard efforts, all of whom also had mildly impaired lung function. During the intervention phases, a psychologist visited parents at home for three weekly two-hour-sessions and taught them how to

implement a token economy system. Parents were observed implementing this token economy and were corrected and advised as necessary. During the reversal phases parents were instructed not to use the token economy and to refer back to previous parenting practices.

Pedometer readings and parent and child diaries indicated a higher frequency of exercise during the intervention phases of the study. Two participants returned to baseline levels of exercise during the reversal phases, whilst a third showed more exercise than baseline but less than during the intervention, suggesting that the token economy was likely to be effective in reinforcing adherent exercise behaviour. All three participants were exercising above baseline levels during the one and three month follow-ups, indicating some long term maintenance of the intervention. However, exercise duration and frequency was much more variable during follow-up indicating that 'top-up' or 'booster' sessions may be required to sustain treatment gains.

A methodological advantage of this study was the use of an ABAB reversal design, which increases the reliability of any conclusions drawn about the effects of the intervention. However, the consistency of parent behaviour across treatment and reversal phases could not be determined, making it difficult to systematically compare the two phases of treatment. Furthermore, the lack of a longer term follow up period with measurement of physical health parameters means that it is not possible to draw conclusions regarding physical health in the longer term.

Interventions assessing generic adherence issues

Non-randomised designs

McClellan, Cohen and Moffett (2009) did not target a specific domain of treatment adherence within their intervention. Instead they focussed on examining the role of a time-out based strategy in decreasing treatment avoidance in children with CF who demonstrated non-compliance to parents' treatment request across the span of their treatment regimen. A case series reversal design was used whereby parents of two children (aged 4 and 5) deemed to be at least 50% non-compliant with at least one parent-initiated daily CF treatment component were recruited.

During a baseline period, any attempts that parents made to support the targeted treatment components were video recorded. Following this, a single two-hour session was undertaken during which parents were instructed how to use the time out programme, which was based upon an empirically supported parenting program (Forehand & Long, 2002). Parents were taught to praise their child, when compliance occurred, to provide clear choices between complying with command or going to time out, as well as how to place the child in appropriate time-out location, and how to deal with refusal. Parents were also given handouts providing a flow chart of time out procedures. Video-tapes of treatment interactions were assessed during the intervention in order to code the child's level of treatment compliance via an adapted version of the Dyadic Parent-Child Interaction Coding System (Eyberg & Robinson, 1981). Following a period of stability whereby the parent was at least 75% adherent to the time-out based discipline strategy, the reversal condition was put into place whereby parents were instructed to stop using the time out strategy.

A decline in parent-rated CF problem intensity, as measured by the Cystic Fibrosis Problem Checklist (Sanders, Gravestock, Wanstall & Dunne, 1991) was reported by parents following the intervention. In one case clear differences in the percentage of observed treatment compliance were demonstrated during baseline and intervention phases (37%, 65%, 49%, and 66% respectively). The second child demonstrated an increase in compliance during intervention implementation, which remained throughout the remainder of participation in the study. However, this child was not sent to time-out throughout all phases of the intervention and so it is possible that other factors may have been involved in increased compliance. McClellan et al. (2009) suggest that other behavioural elements of the intervention, such as reducing the availability of negative reinforcement for non-compliance, may have improved adherence in this particular case. The lack of a reversal, however, makes this difficult to confirm. Case series reversal designs can be difficult to implement when using behavioural interventions due to the possibility of carry-over effects which may help to explain these current findings. It is therefore difficult to determine whether it was the intervention that led to the initial positive behavioural change.

Multi-faceted educational programmes

Non-randomised studies

Bartholomew et al. (1997) conducted a quasi-experimental study evaluating the effectiveness of a home-based family education program to increase CF disease knowledge, self-efficacy and disease management behaviours in children with CF and their parents. This was a multi-faceted programme whereby treatment related

behaviours/adherence were only part of the overall empirical focus. One hundred and ninety-nine parent-child dyads took part in this intervention, which was based upon social cognitive constructs of self-efficacy, social reinforcement and behavioural capability. It was self-paced and involved parents being posted paper based resources and strategies to try at home. The curriculum included respiratory care and nutrition information, and coping and communication skills. Social learning theory principles, such as contingency management, reinforcement, goal setting, modelling and self-monitoring skills, were also included. As the children varied in age from 1-18 years of age, strategies were tailored to three age groups: early childhood, middle childhood, adolescence. A control group of families were recruited from a separate CF clinic and received treatment as usual.

Results indicated that children and parents who took part in the family education programme benefitted in terms of increasing disease self-management behaviours, as measured by the Cystic Fibrosis Self-Management Questionnaire (Sockrider, Swank, Mariotto, Bartholomew & Seilheimer, 1991). Furthermore, the intervention group showed significant increases in caregiver, child and adolescent CF knowledge, parental problem solving, parent and child self-efficacy, child behavioural functioning, as well as measures of physical health status and pulmonary function. Whilst this study demonstrates the potential for a multi-faceted intervention to address multiple areas of difficulty amongst families with a child with CF, the limitation of using such a rich and comprehensively covered intervention is that it makes it difficult to determine which elements of the intervention are effective and for what purpose (Bernard & Cohen, 2004).

Methodological Quality of Non-Randomised Studies

Overall the methodological quality ratings for non-randomised studies were good but slightly lower than the quality ratings reported for RCTs. This may be because many quality assessment tools remain biased towards RCTs because they are regarded as the gold standard research methodology. Two studies received a 'high' quality rating (Bernard et al., 1999; Hourigan et al., 2013), seven studies obtained a 'good' overall quality rating (Bartholomew et al., 1997; McClellan et al., 2009; Stark et al., 1990; 1993; 1994; 1996; 2003), and a further two studies obtained a 'moderate' rating (Piazza-Waggoner et al., 2005; Singer et al., 1991). Due to the nature of case series designs using small sample sizes, many of these studies received lower ratings due to limitations regarding the generalisability of findings. Studies obtaining a 'moderate' quality rating did so because they did not give a clear rationale for the use of a case series design. They also gave limited justifications for chosen data analysis methods, and lack of clear recruitment information.

Discussion

The findings of this review provide an encouraging picture for the role of parent-based interventions to promote treatment adherence in children with CF. Although the search strategy allowed for the inclusion of interventions from a variety of psychosocially informed backgrounds, the majority of studies continue to utilise behaviourally informed interventions, indicating little change in intervention focus since the previous review by Bernard and Cohen (2004). A large proportion of studies combined behavioural and nutritional counselling approaches for dietary adherence in CF, with a number of RCTs and small scale RCTs showing that these types of interventions increased children's

calorie intake and RDA energy per day both immediately after intervention and at longer term follow-up. Results also showed that these interventions led to improved physical health outcomes; for example, significant increases in weight (Powers et al., 2005), Body Mass Index (Stark et al., 2009) and height (Powers et al., 2015).

A number of observational studies have provided further evidence for the effectiveness of pure behavioural and combined behavioural and nutritional counselling interventions, demonstrating increases in caloric intake and weight from baseline to post-intervention in both group and individualised formats (Hourigan, Helms, Christon, & Southam-Gerow, 2013; Stark et al., 1990; 1993; 1996). Similar observational outcomes have been found from telehealth behavioural interventions (Piazza-Waggoner et al., 2006), indicating that there is scope for behavioural interventions to be adapted in ways that increase accessibility. Positive changes in both parent and child behaviours during treatment procedures, such as during meal times, have also been demonstrated by a small number of studies (Hourigan et al., 2013; Stark et al., 1994), providing evidence for the internal validity of parenting interventions alongside preliminary evidence that behavioural interventions may be effective in improving adherence via a mechanism that addresses unhelpful parent and child interactions and ineffective parenting strategies.

This review also provided preliminary evidence that behavioural parenting interventions may be a cost-effective way to simultaneously promote adherence to different domains of CF treatment (McClellan et al., 2009), increasing children's compliance to parental treatment requests across the spectrum of CF treatments. The array of intervention delivery formats used in the studies located in this review demonstrates the flexibility of

behavioural parenting approaches, which is important when considering the increased demands placed upon parents of children living with a chronic illness.

Consistent with a previous literature review (Bernard & Cohen, 2004) no papers were located that examined the role of behavioural interventions for improving adherence to airway clearance techniques. Given that treatment adherence is reported to be particularly poor within this domain (Kettler, Sawyer, Winefield, & Greville, 2002), this finding is surprising. Poor adherence to airway clearance methods is associated with high mortality, progressive lung disease and infection (Flume & Stenbit, 2008) and should therefore be an area where supportive interventions are targeted. One observational study examined the role of a token economy intervention to increase adherence to exercise, something that has been proposed to be an effective adjunct to chest airway clearance methods (Baldwin, Hill, Peckham, & Knox, 1994; Thomas, Cook, & Brooks, 1995). This study showed positive outcomes including increases in exercise levels during the intervention, which were maintained at follow-up. Further studies examining parenting interventions within this domain are therefore required to support children with more laborious treatments, such as airway clearance.

A further observation from the results of this review was that the majority of studies continue to be heavily biased towards younger children with CF. This echoes findings from a previous systematic review (Bernard & Cohen, 2004) and was unforeseen, given the significant increase in life expectancy within CF over the past ten years since this previous review. Given that adolescence is a period associated with increased treatment complexity and reduced treatment adherence (Sawicki et al., 2009), one would expect

parenting research within this age group to have increased in recent years, particularly because parents continue to be one of the most important positive influences on treatment adherence during this age (Taylor, Gibson, & Franck, 2008). The lack of studies found in this age group may reflect the increasing independence of adolescents with CF, because there seems to be an increasing evidence base for individual therapeutic interventions to support treatment adherence in adolescents; for example, motivational interviewing approaches (Duff & Latchford, 2010; Erickson, Gerstle, & Feldstein, 2005). Furthermore, in line with the addition of new-born screening in more recent years, the focus on more rigorous early interventions for children and families may be another driving force behind the continuing bias towards parenting interventions for younger children with CF. Whilst early years interventions are important, the increasing autonomy and independence that characterises adolescence suggest that this is an important time to intervene in order to ensure a smooth transition of treatment responsibility from the parent to the child.

Limitations of included studies and the review process

The studies included in this review had significant methodological weaknesses, precluding firm conclusions about the unique effects of parenting interventions on treatment adherence in children with CF; these will be considered in turn. Firstly, a large proportion of the included studies were observational in nature. Whilst case series designs are useful when developing preliminary treatment techniques/protocols (Barlow & Hersen, 1984), their use of small sample sizes restricts the confidence and generalisability of the conclusions that can be made. Furthermore, the lack of control subjects makes this type of research more prone to bias. Although a small number of randomised controlled trials

demonstrated similarly positive results for the role of behavioural parenting interventions, two of these also used very small sample sizes. Overall, the results of this review therefore need to be interpreted with caution.

Additionally, whilst some studies made attempts to record and rate the facilitator's adherence to the intervention protocol being used (Powers et al., 2003; Stark et al., 2009), very few studies utilised treatment integrity assessments to track how well parents implemented the strategies taught. It is therefore difficult to ascertain to what extent improvements in adherence reflect the use of these strategies taught. It is also notable that there was substantial variation in the delivery of interventions in respect of the frequency and duration of sessions, and the format and length of the treatment programmes, making it difficult to determine the most effective dosage and format of interventions.

Furthermore, some studies reported that high percentages of families who were approached were unable to take part or withdrew during the intervention. For example, 40% attrition was reported by Powers et al. (2003, 2015), 52% by Stark et al. (2009), and 65% by Stark et al. (2003). It is possible that parents who consented to take part demonstrated more insight and motivation to engage in different parenting approaches, thereby making positive results more likely. Additionally, none of the studies investigated reasons why so many families were unable to take part, which makes it difficult to ascertain the feasibility of these types of parenting interventions.

As all included studies utilised samples from predominantly White backgrounds it is difficult to generalise these research findings to other ethnic and socioeconomic backgrounds. Whilst the prevalence of CF is reported to be much lower in non-White groups (Phillips, Bishop, Woods, & Elias, 1995), issues such as language barriers and cultural belief systems may impact upon disease understanding, access to treatment, and treatment behaviours in families from ethnic minority backgrounds (Duff, 2003). Additionally, all included studies were carried out in the USA meaning that the results may not be generalisable to the UK CF population. Health care systems and funding in the UK and USA differ significantly. In the UK, universal access to CF care is available while in the US that is not the case and the system operates around health insurance. Therefore there may be different factors influencing not only treatment adherence but also parents' motivation to seek supportive parenting interventions for such difficulties. This means that the characteristics of parents participating in studies within the USA may be quite different to parents participating in studies within countries offering a free health care system. Further research is required to explore these factors.

Finally, the search strategy used in this review was limited to papers written in English, therefore meaning that studies from other cultures may have been missed. Furthermore, given that the current review limited the search to studies published in peer reviewed journals for quality assessment purposes it is possible that a number of service development projects and routine clinical evaluations utilising parenting interventions may have been missed, potentially biasing the findings reported.

Future research

Larger scale studies which allow for more sophisticated and well powered statistical analyses are required to increase the confidence and generalisability of findings regarding parenting interventions within the paediatric CF population. However, given the reported low uptake of the interventions offered in the studies reported, future studies will first of all need to address how acceptable and feasible different interventions are and what types of interventions families themselves think they may benefit from. It is important that researchers gain sufficient knowledge about this before investing in further larger scale studies to ensure that future research resources and participant time is utilised appropriately and ethically (Craig et al., 2008). Future research should focus on conducting studies that dismantle behavioural and multi-component interventions in order to clarify the effective components of implemented interventions (Gardner, Hutchings, Bywater, & Whitaker, 2010). They should include more measures of psychosocial variables, such as family functioning, wellbeing, and parent-child relationships, in order to understand the underlying mechanisms through which these interventions are likely to work as well as to understand the impact that such interventions might have on wider child and family wellbeing.

More research is also needed to investigate the potential usefulness of parent based interventions for adolescents with CF who have adherence difficulties. As medical research continues to advance treatments, it is likely that more children will live through adolescence and adulthood, whilst being required to maintain an increasingly complex treatment regime (Sawicki et al., 2009). These factors alongside the transition of CF care responsibilities during adolescence makes it a critical time to intervene to ensure that

young people can continue to care for themselves appropriately and therefore maintain good health into adulthood.

Conclusions

The findings of this review provide tentative evidence that parenting interventions, mainly those that have their roots in behavioural psychological principles, may improve treatment adherence in children with CF. However, future work is needed, incorporating feasibility and acceptability data alongside larger sample sizes, to establish clarity about the impact of parenting interventions on treatment adherence in children and adolescents with CF. The increasing life expectancy and treatment complexity for children and adolescents with CF is likely to pose continuing challenges to treatment adherence, meaning that further research in this field will be of increasing priority.

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Paper 2: Empirical paper

A Case Series Examining the Acceptability and Feasibility of the Self-Directed Teen Triple P Parenting Programme for Adolescents with Cystic Fibrosis

The following paper has been prepared for submission to
'Journal of Clinical Psychology in Medical Settings'.
The guidelines for authors can be found in Appendix A**.**

Word Count: 8213 (excluding abstract and references)

Abstract

Objectives: There is a paucity of research examining the role of developmentally appropriate parenting interventions in promoting adolescent Cystic Fibrosis (CF) treatment adherence despite research suggesting that adherence is poor during adolescence. *Methods:* A mixed methods case series methodology examined the feasibility and acceptability of a self-directed parenting intervention for this purpose. Parents of 11-16 year olds were recruited via CF clinics and a CF charity website. The 10-week intervention comprised the Self-directed Teen Triple P workbook plus chronic illness tip sheet. Semi-structured interview data regarding the acceptability and feasibility of the intervention were collected from parents and CF professionals. *Results:* Six parents gave consent to participate in the intervention; however, only two parents completed the intervention. In both cases increases in parent-reported treatment adherence and positive parenting strategies were observed following the onset of the intervention. Feasibility interviews were conducted with seven parents and two CF nurses. Issues, such as competing illness-related time demands, were identified as key barriers to the intervention. A number of adaptations were reported by parents and staff in order to increase acceptability. *Conclusions:* Although parents demonstrate a desire for support in transitioning treatment responsibility to their adolescent, further work is needed to refine parenting interventions in order to increase uptake.

Key words: Cystic Fibrosis, Adolescents, Adherence, Parenting Intervention

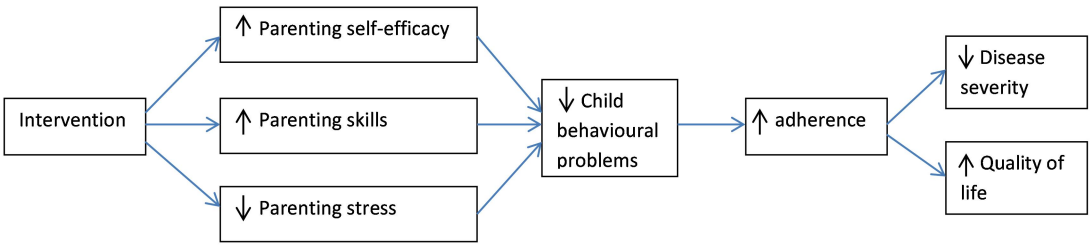
Introduction

Cystic fibrosis (CF) involves a complex and time-intensive treatment regime that can take several hours a day to complete. It is reported to be one of the most challenging chronic illnesses for children and their parents to manage (Mitchell, Powers, Byars, Dickstein, & Stark, 2004). Research has suggested that CF treatment adherence reduces with child age (White et al., 2009). Adolescence is a particularly challenging time for disease management as children show increased desire for more independence and autonomy and parents need to help their child take charge of their own health. However, autonomy is often juxtaposed with the lifestyle changes enforced by the physical symptoms of CF and by parents' continuing need to oversee and remain legally responsible for their child's health and their CF treatments (Field & Duchoslav, 2009). Subsequently, families of adolescents with CF experience new challenges that may contribute towards an understanding of these exacerbated adherence difficulties.

Parents have been found to be the best allies in helping adolescents with the disease and guiding them through the demands of treatment (Laursen & Collins, 2009; Taylor et al., 2008). Parental monitoring, support and supervision remain important for increasing the likelihood of positive health outcomes and disease management (Modi, Marciel, Slater, Drotar, & Quittner, 2008). The use of positive, consistent and cooperative parental interactions have been demonstrated to lead to increases in treatment adherence (Butcher & Nasr, 2014). However, the use of ineffective parenting practices has been reported to be more common in chronic illnesses, which may result from differing expectations of the child's behaviour and/ or reduced parental wellbeing and parental stress (Fiese & Schwartz, 2008; Mullins et al., 2007). These negative parenting practices

have been associated with poorer treatment adherence. Additionally, the increased demands and stresses that are placed on the adolescent and parent as a result of poor treatment adherence are likely to influence the efficacy of different coping strategies for parents and adolescents, thereby influencing parent and child emotional wellbeing and subsequent disease management behaviours (Field & Duchoslav, 2009). Furthermore, parents who struggle to support their adolescent with their treatments are likely to feel less capable and competent in their parenting role (Kedesdy & Budd, 1998), and perceived parental self-efficacy is proposed to be central in promoting child disease management behaviours (Bandura, 2004; Morawska, Calam, & Fraser, 2015; Rogers & Matthews, 2004). Parenting interventions therefore have the potential to improve treatment adherence via their mediating effects on some of these factors, including increasing positive parenting practices, increasing parent self-efficacy, and reducing parental stress (Figure 1). Investigating such interventions is particularly important within the adolescent CF population in order to sustain quality of life and wellbeing within this age group.

Figure 1. A conceptual framework demonstrating the mechanisms by which parenting interventions may improve treatment adherence (Morawska, 2015)



Prior to the development of new health care interventions, it is important to explore the acceptability and usefulness of existing resources that may be readily adaptable (Sanders

& Kirby, 2012). The Triple P Positive Parenting Programme (Sanders, 1999) is one approach that may offer enough flexibility to cater for the demands of supporting an adolescent with CF. This intervention has an extensive international research base that draws upon social learning theory, cognitive behavioural and developmental psychology principles (Thomas & Zimmer-Gembeck, 2007). It uses a public health perspective, offering five different tiers of support including individual, group, telephone assisted and self-directed programs (Sanders, Markie-Dadds, & Turner, 2012). A recent meta-analysis demonstrated that Triple P has positive influences on parenting skills, parental wellbeing, and child behaviour, with comparable outcomes being found for individual, group, and self-directed variants of the programme (Nowak & Heinrichs, 2008). Parental self-efficacy and parental stress has also been shown to improve as a result of these interventions (Markie-Dadds & Sanders, 2006).

The increased accessibility and cost effectiveness of self-directed Triple P offers a potentially beneficial parenting intervention for parents of adolescents with CF who are likely to find it difficult to attend regular face-to-face appointments. The Teen Triple P workbook (Ralph & Sanders, 2001) is a self-directed 10-week parenting intervention that is based upon a self-regulatory model designed to promote healthy teenage development and support parents to transition responsibility to their child. The programme also has a series of accompanying tip sheets, including one about chronic illness (Morawska & Sanders, 2010), which addresses common issues facing families living with a chronic illness.

A recent randomized controlled trial within adolescent diabetes found that parents who completed this intervention reported significantly lower levels of family conflict,

increased parenting competence, improvements in the use of adaptive parenting strategies, and improvements in child emotional and behavioral wellbeing (Doherty, Calam, & Sanders, 2013). Additionally, some of the key positive behavioural principles that are central to the Triple P programme, such as limit setting, contingent reinforcement and problem solving, have also been shown to improve medication adherence in younger children with CF (Hourigan et al., 2013; Stark et al., 2003; Stark et al., 2009). This provides promise for the role of this intervention in promoting treatment adherence in adolescents with CF.

The current study

As self-directed Teen Triple P has not yet been used with parents of adolescents with CF the current research had two over-arching aims. The first aim was to utilize a case series design to test the hypotheses that self-directed Teen Triple P has the potential to increase (1) adolescent CF treatment adherence, (2) the use of adaptive parenting strategies, (3) parenting competence, (4) parent and child wellbeing, as well as a reduction in caregiver stress. A case series design was used because it allows researchers to undertake preliminary explorations investigating whether an intervention may be clinically useful within a new population before investing time in potentially costly large RCTs (Kazdin, 2011).

The second over-arching aim was to gain quantitative and qualitative information from parents and CF professionals, via semi-structured interviews, regarding the acceptability and feasibility of the self-directed Teen Triple P intervention. Consideration of service user and consumer feedback is important in order to ensure that intervention resources

are accessible, meaningful and attractive for parents (Metzler, Sanders, Rusby, & Crowley, 2012).

Method

Design

In order to investigate Aim 1, a within subjects A-B case replication series design was used (Barlow & Hersen, 1984). Participating parents were initially randomly assigned to one of several predetermined baseline lengths ranging from 2-8 weeks. During baseline, outcome measures were collected from parents via weekly telephone calls from the researcher. The Triple P intervention was initiated at the end of the designated baseline period if baseline stability was determined. Baseline stability was defined as the absence of a decreasing trend for at least two consecutive data points prior to the introduction of the intervention (Wells et al., 2009). Parents then completed the Triple P intervention at home. This involved working through a series of ten weekly manualised modules that took approximately one hour to complete. Outcome measures continued to be collected from parents on a weekly basis. Parents were followed up four weeks post-intervention and all outcome measures re-assessed.

In order to investigate aim 2, feasibility and acceptability interviews were simultaneously coordinated with recruitment to the case series. Parents who declined participation in the Triple P intervention, alongside parents who dropped out or completed the intervention were asked to complete a semi-structured telephone interview at the point of drop-out or completion. This allowed for the examination of possible barriers to participation, as

well as strengths, weaknesses, and improvements to the intervention and research methodology.

Participants and recruitment

Inclusion Criteria: Parents of children aged 11-16 were eligible to participate. Parents of 17 and 18 year old adolescents were not included due to issues surrounding transition to adult CF services. The sample was self-selecting and parents were eligible to take part if they qualitatively reported that their child was not adhering to their prescribed treatment plans, or when parents' attempts to support their child to adhere to their treatments were creating significant challenges for parents.

Exclusion Criteria: Parents were excluded from the study if their child was currently accessing psychological intervention for treatment adherence difficulties. Parents were also excluded if they had difficulties with reading and understanding English and had no one to support them with this.

Staff were eligible to participate in feasibility interviews if they were currently working in NHS CF teams within which appropriate local ethical approvals had been granted.

Sample and settings

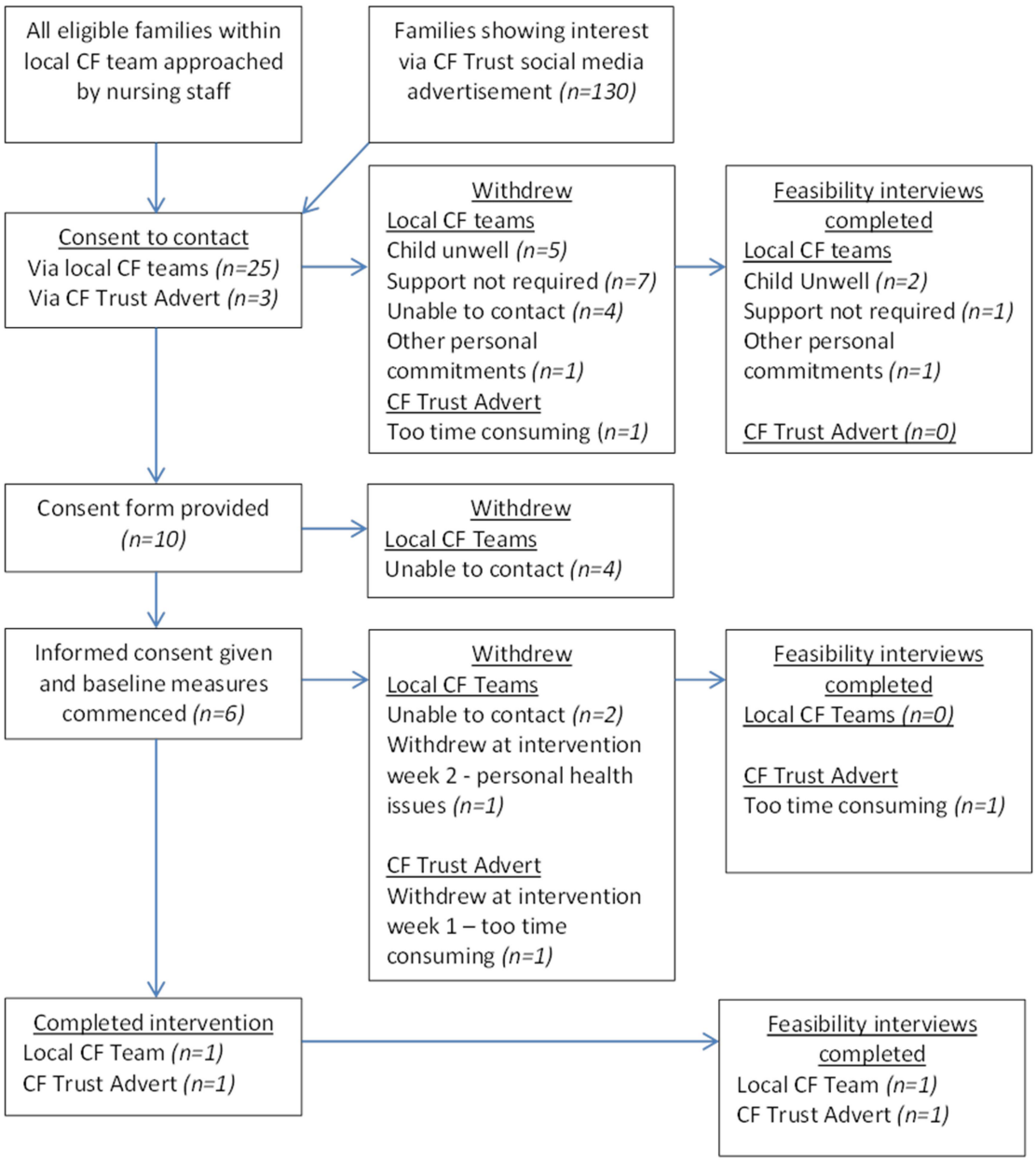
Following review and approval from an NHS research ethics committee (Wales REC 7 ref 15/WA/0096; See Appendix E), parents were recruited from four NHS Cystic Fibrosis clinics located within the North West of England. As the sample was self-selecting, nurses within CF teams handed out participant information sheets to parents (Appendix F) during routine clinic appointments. Nurses then sought verbal consent for the researcher to be able to contact these parents in order to discuss the study in more detail, following

which informed consent was taken by the researcher. Parents were also recruited via an advertisement that was placed on the Cystic Fibrosis Trust website. Interested parents were emailed the participant information sheet, and were posted a consent form (Appendix G) if they demonstrated continued interest in taking part in the intervention. All parents were invited to take part in a feasibility interview regardless of their decision to accept or reject participation in the intervention. This was clearly stated in the information sheet and consent form. Parents who declined participation to the intervention could provide consent to take part in the feasibility interview only, in order to allow exploration of the reasons for non-participation. An adapted participant information sheet was available for adolescents in order to inform them of their parent or carer's involvement in the research (Appendix H) and assent was obtained from children of parents participating in the intervention. A copy of this assent form can be found in Appendix I.

In order to gain feasibility and acceptability interview data from CF professionals, a key nurse from two of the participating CF teams disseminated participant information sheets to the CF team. Interested professionals gave signed consent prior to interview. A copy of the professionals' consent form is provided in Appendix J.

Figure 2 summarises the recruitment and retention to the case series, specifying where each participant was recruited from. It also details the number of feasibility interviews completed alongside their reasons for withdrawal from the study, where applicable. Two parents completed the full Triple P intervention and associated feasibility interviews. A further five families who did not complete the intervention, and two CF nurses completed additional acceptability and feasibility interviews.

Figure 2. CONSORT flow of participants



Measures

Demographic information

The Family Background Questionnaire (FBQ; Appendix K) (Sanders, Markie-Dadds, & Turner, 1999) was completed over the telephone during the first baseline assessment. It

is a 16-item measure of demographic information including gender and age of the parent and the child, socioeconomic status, and level of education.

Treatment adherence

Treatment Adherence Questionnaire–CF (TAQ-CF; Appendix L) (Quittner et al., 2000) is a parent self-report measure assessing the frequency of their child's CF-related treatment compliance. It consists of 14 items asking how often the child completes components of their CF treatment regime, measured via 6-point Likert scales. Parents' responses for actual treatment compliance are subtracted from the prescribed treatment frequency to generate an adherence value for each category of treatments. Prior research has demonstrated adequate 1-year test–retest reliability for the parent rated TAQ (ranging from r=.76 to r=.88) and teen/parent concordance on the TAQ has been rated between r=.69 to r=.88 dependent on the component of treatment (Quittner et al., 2000).

Parenting Scale-Adolescent version

The Parenting Scale- Adolescent version (PSA; Appendix M) (Irvine, Biglan, Smolkowski, & Ary, 1999) is a shortened version of a 30-item scale Parenting Scale (PS) developed for parents of pre-school children (Arnold, O'Leary, Wolff, & Acker, 1993). It is a 14-item parental report measure of parenting style / discipline strategies. It includes two subscales assessing parenting laxness (6 items) and parenting over-reactivity (6 items), alongside a single item assessing parental monitoring. Low scores represent good parenting practices and high scores represent dysfunctional parenting practices. It has high internal consistency (Laxness α =0.82, Over reactivity α =.83, Total score α=.84) and

reliably distinguishes clinical from non-clinical samples (Arnold et al., 1993; Irvine et al., 1999; Prinzie, Onghena, & Hellinckx, 2007).

Parenting Sense of Competence Scale

The Parent Sense of Competence Scale (PSOC; Appendix N) (Johnston & Mash, 1989) involves 16 items related to feelings about being a parent. The scale is scored on two dimensions: satisfaction with the parenting role (9 items) and feelings of efficacy as a parent (7 items). Scores range from 16–96, where high scores indicate higher self-confidence in parenting skills. It shows a satisfactory (α = .79) level of internal consistency (Johnston & Mash, 1989).

Strengths and Difficulties Questionnaire

The Strengths and Difficulties Questionnaire (SDQ; Appendix O) (Goodman, 1997) is a 25-item brief behavioural screening questionnaire that measures parents' perception of prosocial and difficult behaviours in children aged 3 to 16 years. It includes five subscales, each containing five items scored on 3-point Likert scales: emotional difficulties, conduct problems, inattention/ hyperactivity problems, peer relationship problems, and prosocial behaviour. The parent-report version for children aged 4-16 years used in this study has good psychometric properties including high internal consistency (α=.82) (Goodman, 2001), test- retest stability (*r*=.72) (Goodman, 2001) and construct validity (Van Leeuwen, Meerschaert, Bosmans, De Medts, & Braet, 2006).

Depression Anxiety and Stress Scales

Parental depressive symptomatology and anxiety symptomatology were measured using the Depression Anxiety Stress Scales-21 item version (DASS-21; Appendix P) (Lovibond & Lovibond, 1995a). This brief questionnaire assesses symptoms of depression, anxiety, and stress in adults. Individual items are rated on a 4-point Likert scale with higher scores on each subscale indicating more severe symptoms. Good internal consistency has been reported for each subscale (Depression α=.94, Anxiety α=.87) (Antony, Bieling, Cox, Enns, & Swinson, 1998). The scale also has good discriminant and concurrent validity (Lovibond & Lovibond, 1995b).

Pediatric Inventory for Parents

The Pediatric Inventory for Parents (PIP; Appendix Q) (Streisand, Braniecki, Tercyak, & Kazak, 2001) is a 42-item parent-report questionnaire that assesses parental stress in relation to caring for a child with an illness. It uses a 5-point Likert scaling that measures both the frequency and difficulty of illness-related parenting stress across four factors: Communication, Medical Care, Role Functioning and Emotional Distress. Multiplicity effects were minimised by combining the scores into one variable. Scores could therefore range from 84–420, where high scores indicated more frequent and more stressful events. Adequate validity and internal consistency has been previously demonstrated (α = .80 – .96)(Streisand et al., 2001).

The Client Satisfaction Questionnaire (CSQ)

The 13-item Client Satisfaction Questionnaire (CSQ; Appendix R) (Sanders, Markie-Dadds, Tully, & Bor, 2000) addresses the quality of service provided; how well the intervention

met the parents' needs, increased the parents' skills, and decreased the child's problem behaviours; and whether the parent would recommend the intervention to others. The scale has high internal consistency (α= .96), an item–total correlation of .66 and inter-item correlations of .30–.87 (Sanders, et al., 2000).

Table 1 shows the time-points at which the various outcome measures were administered.

Table 1. Time points for outcome measure data collection

	Baseline	Week 1	Week 2	Week 3	Week 4	Week 5	Week 6	Week 7	Week 8	Week 9	Week 10	One month Follow-up
TAQ-CF	X	x	x	x	x	X	X	x	x	X	x	X
PSA	X	x	x	x	x	X	X	x	x	X	x	X
PSOC	X	x				X					x	X
DASS-21	X	x				X					x	X
PIP	X	x				X					x	X
SQD	X	x				X						X
CSQ											x	

Treatment integrity

Participants also completed a Triple P module checklist (Appendix S) each week to record whether they had read the corresponding module, thereby assessing treatment integrity.

Acceptability and feasibility

Semi-structured interview data were collected from participating parents at the end of the Triple P intervention or at the point of withdrawal from the research study. Questions focussed on parents' experiences taking part in the programme, benefits and challenges, and ideas for improvements. Parents who did not take part in the intervention were also

invited to complete this interview in order to capture possible barriers to participation. A copy of the interview schedule can be found in Appendix T.

CF professionals who consented to take part in the study also participated in a semi-structured interview to further extend the acceptability and feasibility information collected. Interviews were recorded using an encrypted telephone enabled Dictaphone and were transcribed and stored on a secure university computer.

Procedure

Upon completion of the baseline period, participants received Triple P resources posted to their address. This included the self-directed Teen Triple P workbook (Ralph & Sanders, 2001) and Chronic Illness Tip Sheet to work through over the 10-week period (approximately 1 hour per week).

The Teen Triple P workbook is a self-directed behavioural parenting intervention that uses social learning theory principles to help parents build on their existing skills and information to practice positive parenting. A self-regulatory model is the basis of the 18 core skills, which fall into four main categories of skill building to: (1) increase positive parent–teenager relations, (2) increase desirable behaviour, (3) teach new behaviours and skills, and (4) manage problem behaviour. The workbook incorporates weekly exercises to help the parents to implement the strategies outlined in the workbook.

The first three weekly modules cover goal setting, increasing desirable behaviour and managing problem behaviour, which includes the use of reinforcement, such as praise and ignoring, behavioural contracts and tools for monitoring change. Modules 4–6

promote practice of these strategies, whilst providing guidance about how to monitor the effectiveness of these strategies and to alter where necessary. Module 7 provides strategies for dealing with risky behaviour, with modules 8–9 providing the chance to practice using these strategies. Module 10 reviews progress over the course of the program, identifying strategies to maintain progress and setting future goals.

The Chronic Illness Tip Sheet (Morawska & Sanders, 2010) demonstrates practical ways of tailoring advice in the workbook to solve common issues that may arise when supporting a child with a chronic illness. It summarises reasons for increased behavioural and emotional difficulties in chronic illness, prevention and coping advice for managing treatment routines, reducing family stress, helping siblings cope, and reducing anxiety.

Data analysis

The primary method of data analysis was visual inspection of data by graphing each individual's progress on the various outcome variables across the baseline, intervention, and follow-up periods. This method is commonly used in the case series literature and provides a stringent method of the treatment effect, because only unambiguous effects will be apparent (Parsonson & Baer, 1992). The small sample size in this study meant that the use of inferential statistics was inappropriate.

Qualitative content analysis (Elo & Kyngas, 2008; Hsieh & Shannon, 2005) was used to analyse the transcribed digital interview recordings. A process of open coding was used, whereby the researcher immersed themselves in the transcripts allowing manifest codes to emerge from the data (Kondracki, Wellman, & Amundson, 2002). The researcher made

comments and notes of potential codes throughout the transcripts following which lists of these codes were grouped together under higher order headings in order to organise these codes into meaningful categories. Definitions of each category were then developed from the data alongside identification of exemplars for each category.

Results

Participant characteristics

Demographic data for all consenting participants is provided in Table 2. Participant 1 dropped out following completing two weeks of the intervention, whilst participants 2, 3, and 5 all dropped out during the baseline phase of the intervention. Participants 4 and 6 completed the intervention. Baseline data for participants who dropped out of the intervention can be found in Appendix U. Insufficient data were available from these participants in order to evaluate the effects of the intervention. Observations of baseline periods for non-completers indicated that they generally had stable baselines and higher levels of treatment adherence than completers.

Table 2. Characteristics of participants

Parent Demographic	Participant 1	Participant 2	Participant 3	Participant 4	Participant 5	Participant 6
Relationship to child	Mother	Mother	Mother	Mother	Mother	Mother
Age	35	37	40	43	51	45
Child age	15	13	13	15	15	14
Ethnicity	White British	White British	White British	White British	White British	White British
Education level	High School	University degree	High School	University degree	University degree	University degree
Occupational status	Unemployed	Employed (part time)	Unemployed	Employed (full time)	Employed(full time)	Employed (part-time)
Marital status	Divorced	Married	Single	Married	Married	Married
Number of siblings	0	2	3	1	0	2

Treatment adherence

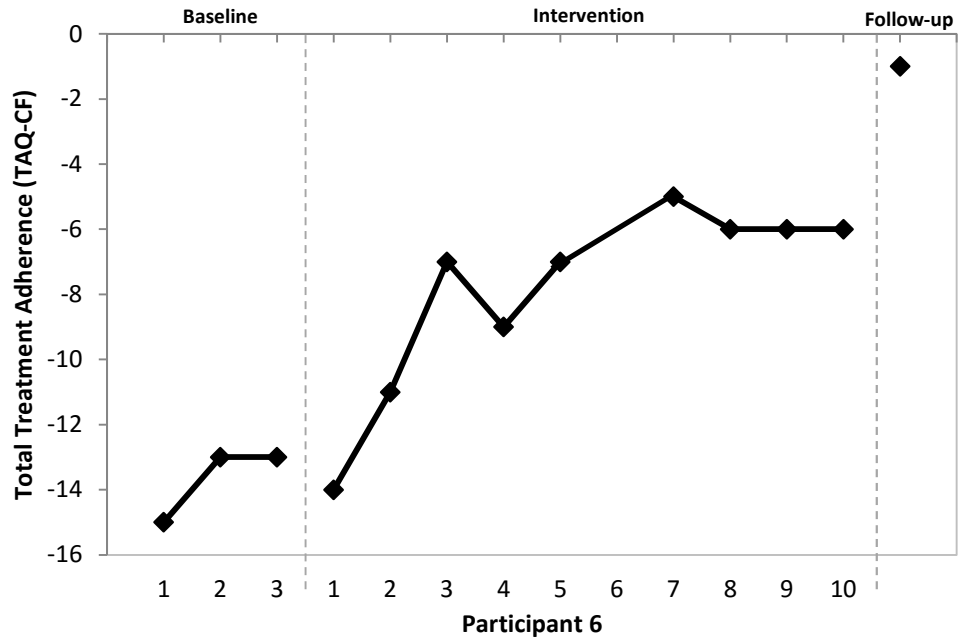
Participant 4 completed a two week baseline period and participant 6 completed a three week baseline period. Whilst Participant 6 demonstrated stability in parent-rated treatment adherence during baseline, as measured by the TAQ-CF, Participant 4 demonstrated an increasing trend during baseline (Figure 3). Upon commencement of the intervention clear improvements in treatment adherence can be observed for both participants. These increases continued throughout the intervention phase for both parents. For both participants gains made during the intervention phase were maintained at one-month follow-up. Participant 6 showed further increases in treatment adherence from post-intervention to one-month follow-up. In this case the parent reported that the child had become unwell and had been admitted to hospital, which may have provided increased motivation for the child to adhere to their treatments.

Parenting skills

Figure 4 highlights participants' scores on the Parenting Scale during baseline, intervention and follow-up phases of the study. Stability across the subscales of this measure can be observed during baseline assessments, with a decreasing trend emerging upon the instigation of the intervention. Here, both participants demonstrated an observable reduction in total Parenting Scale scores. Participant 4 showed comparable gradual declines in over-reactivity and laxness scores throughout the intervention. Although this participant showed a slight increase in total parenting scale score at one-month follow-up, this remained below the scores reported at baseline. Participant 6 demonstrated minimal change in parenting laxness throughout the intervention but a

reduction in over-reactivity throughout the intervention, which was maintained at one month follow-up.

Figure 3. Weekly TAQ-CF throughout baseline, intervention and follow-up



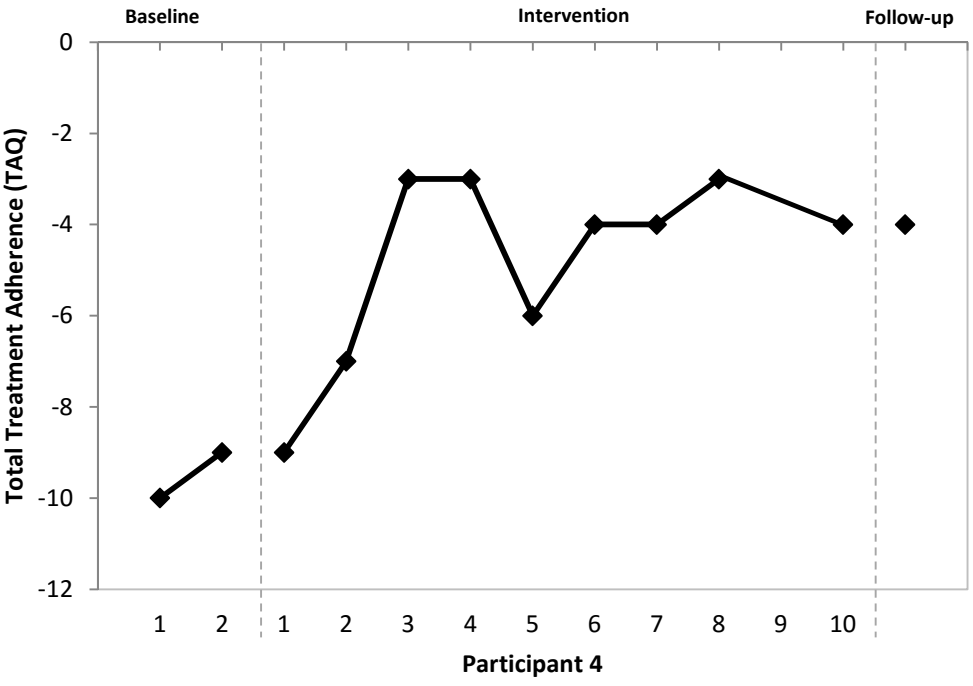
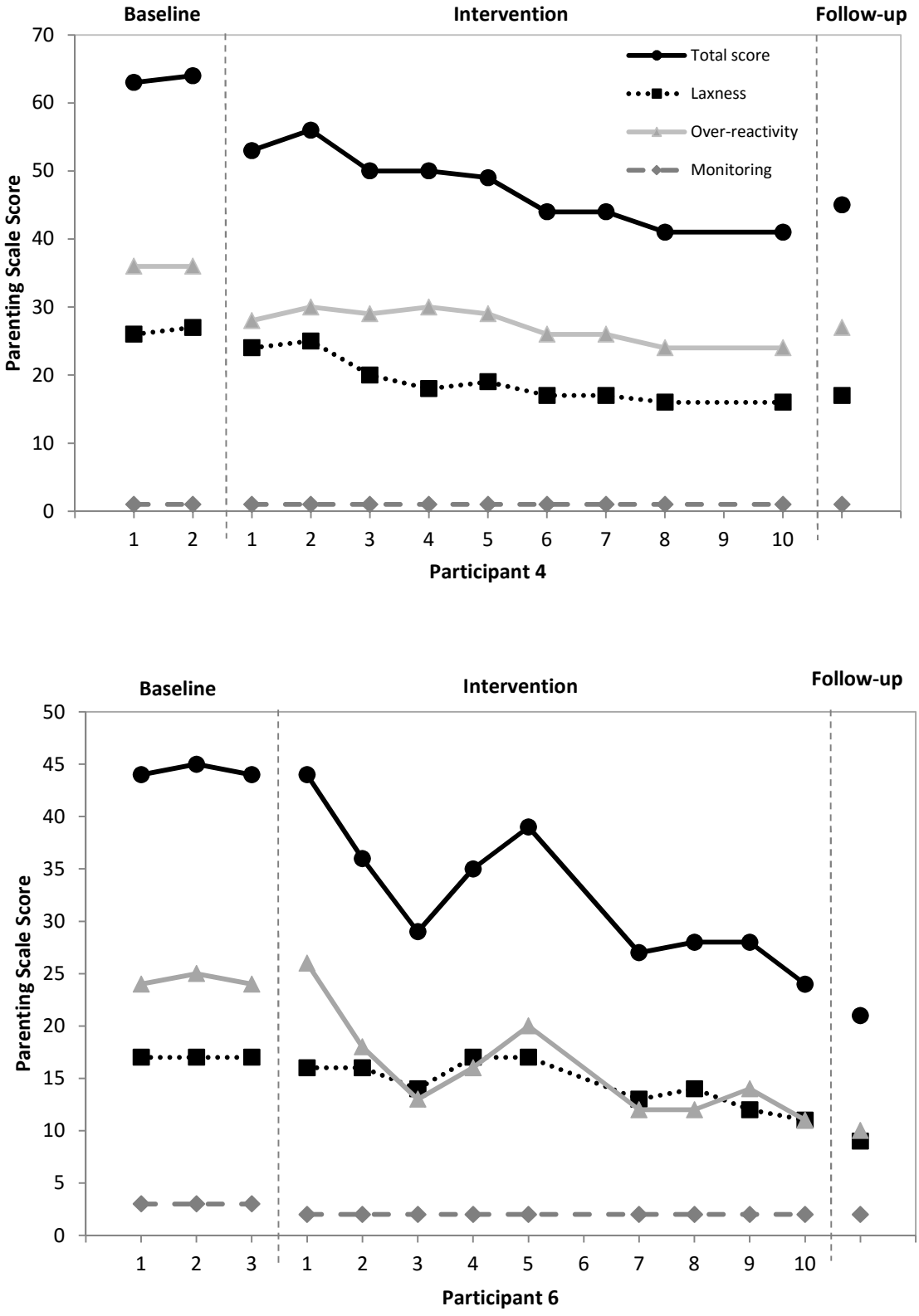


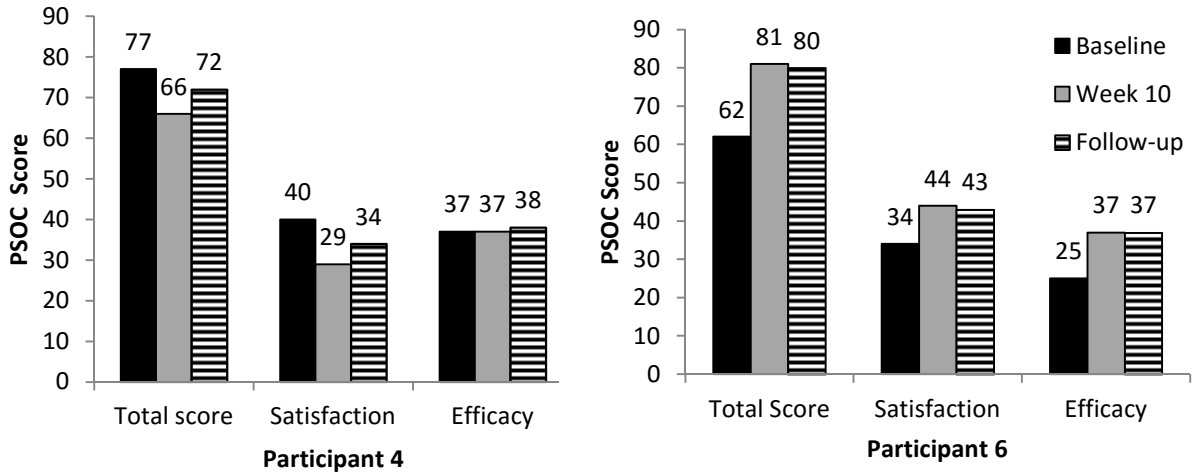
Figure 4. Weekly Parenting Scale scores throughout baseline, intervention, and follow-up



Parental Efficacy and Satisfaction

Visual comparison of participant 4's baseline and week 10 data PSOC scores (Figure 5) indicated a reduction in parental satisfaction scores from pre to post intervention. Although satisfaction increased at one-month follow-up, this remained below baseline levels. Participant 4 demonstrated minimal change in efficacy scores between baseline, week 10, and one-month follow-up. Conversely, participant 6 showed increases in overall parenting competence, parenting satisfaction and efficacy as the intervention progressed. Increases across all subscales were maintained at one-month follow-up.

Figure 5. Self-reported parent competence, satisfaction and efficacy as measured by the PSOC scale.

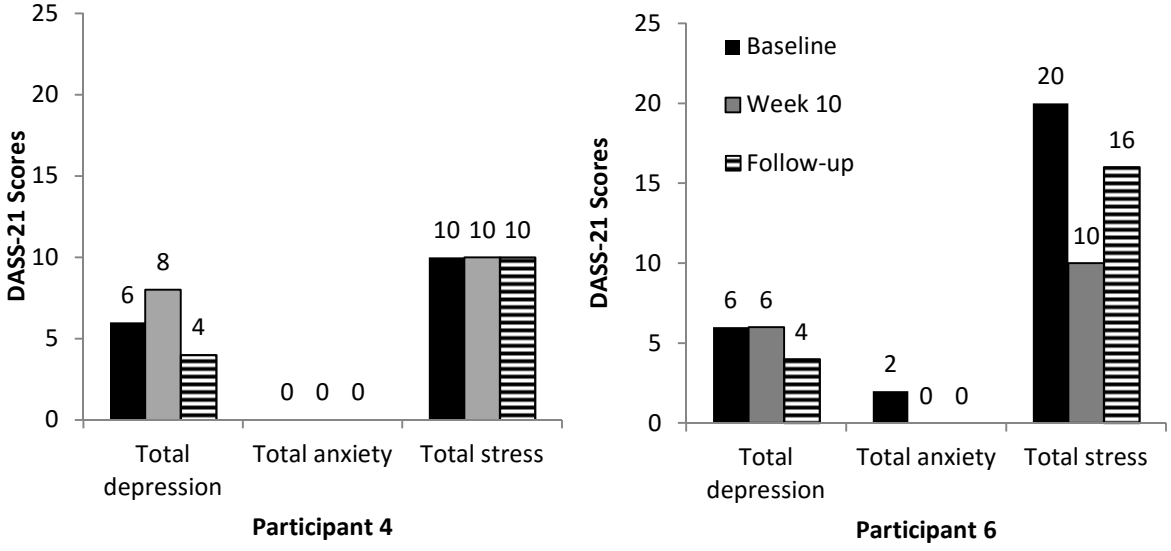


Parent wellbeing

Baseline, intervention and one-month follow-up scores on the DASS-21 are shown in Figure 6. Participant 4's scores on the depression subscale increased between baseline and post-intervention but reduced to below baseline levels at follow-up. However, these scores did not reach clinical caseness of depression at any point during the study.

Similarly, throughout baseline, intervention and follow-up, Participant 4's stress and anxiety subscale scores did not reach clinical caseness, with scores remaining very low all time points. Participant 6 did not demonstrate clinical caseness on the depression subscale at baseline, post-intervention and follow-up; although scores did show a slight reduction at one-month follow up. Participant 6 did not demonstrate clinical caseness of anxiety throughout the study, but a slight reduction in this score was demonstrated at one-month follow-up. Participant 6 reported 'moderate' levels of stress at baseline; however, by week 10 this score had reduced below the threshold of clinical caseness. Although follow-up stress scores for this participant remained below baseline levels following the implementation of the intervention, an increase was reported between post-intervention and one month follow-up. This may reflect the additional stress associated with the child's hospital admission during the follow-up period.

Figure 6. Parental emotional wellbeing as measured by the DASS-21



Participants' scores on the PIP throughout the course of the study can be found in Table 3. Participant 4's demonstrated minor increases in chronic illness related parenting stress between baseline and follow-up, which returned to baseline levels at one-month follow-up. Participant 6 demonstrated decreases in parenting stress on this measure between baseline and post-intervention. However, in line with DASS-21 scores, an increase in parenting stress was shown at follow-up.

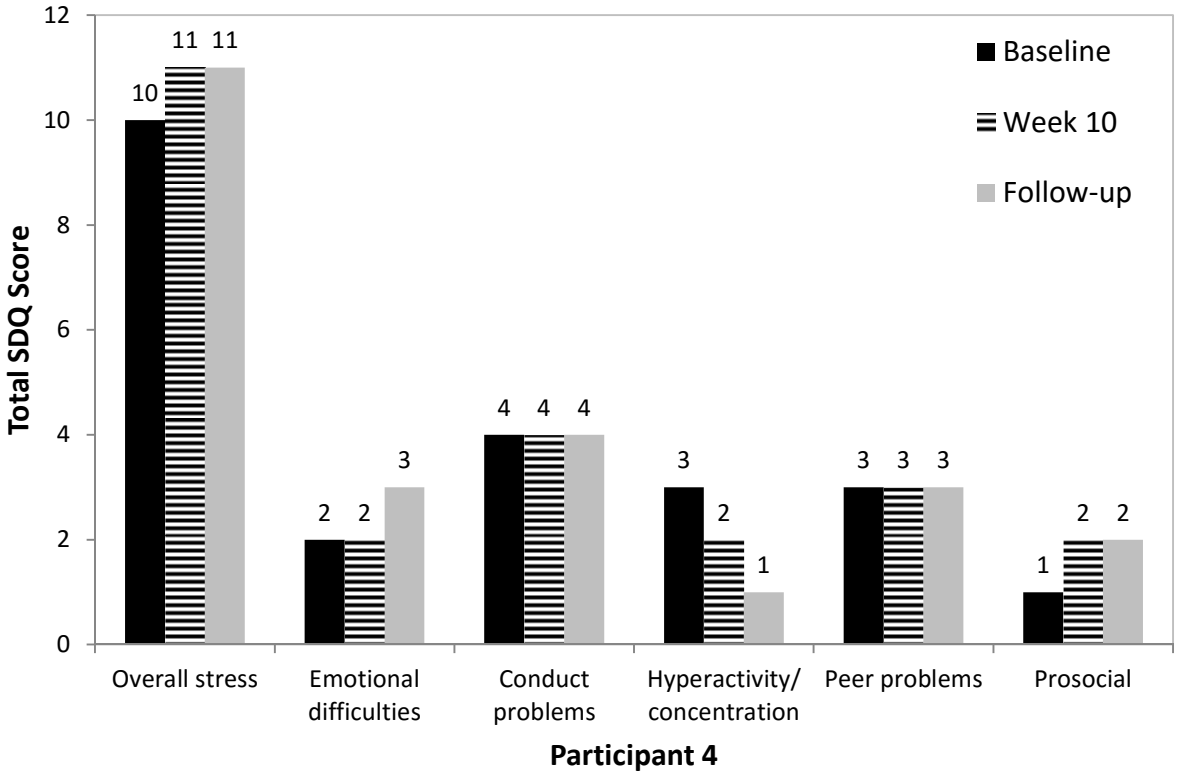
Table 3. Parental stress ratings as reported on the PIP

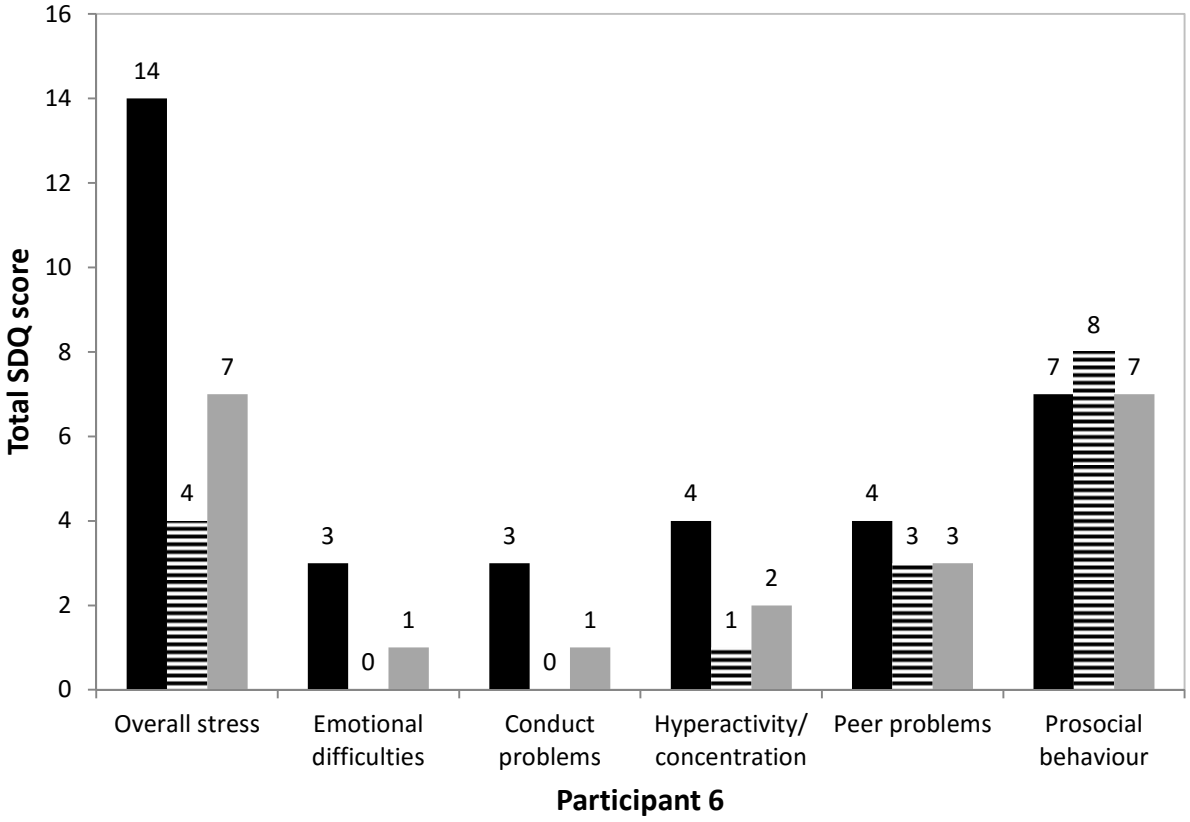
	Baseline	Week 10	Follow up
Participant 4	120	132	120
Participant 6	132	103	135

Child wellbeing

Figure 7 demonstrates parent-rated scores on the Strengths and Difficulties Questionnaire across baseline, intervention and follow-up phases of the study. At baseline, participant 4 reported 'average' scores on subscales assessing overall stress, emotional difficulties and hyperactivity, which remained within the 'average' range at the end of the intervention and at one-month follow-up. This participant reported 'high' levels of conduct difficulties, 'slightly raised' peer problems, and 'low' levels of prosocial behaviour at baseline. Scores on these subscales remained stable across the intervention and remained in their original clinical categorisations post-intervention and at follow-up.

Figure 7. Parent-rated SDQ subscale scores





At baseline, participant 6 reported that their child had 'slightly raised' overall stress and conduct difficulties, 'high' levels of peer related difficulties, and 'slightly low' levels of prosocial behaviour. Emotional and attentional functioning were reported within the 'average' range at all three time points. At the end of the intervention, substantial reductions in conduct and stress levels can be observed, with both scores falling in the 'average' range. These scores remained in the average range at one-month follow-up. There was minimal change in prosocial behaviour scores throughout the intervention.

Client satisfaction questionnaire

Participant 4 demonstrated an overall satisfaction score of 57 out of 91 and participant 6 rated her satisfaction at 78 out of 91. Participant 6 rated all domains of the experience at a score of 5 or above indicating high levels of satisfaction with the intervention and research process. Participant 4 rated most domains of the experience at 5 or above, again indicating high satisfaction. The remainder of domains were scored 4, indicating moderate satisfaction.

Parent and nurse feasibility interviews

A number of themes emerged from the interview data provided by parents and CF nurses, including the challenges of transitioning disease management during adolescence, barriers to participation, such as competing illness related time-demands and accessibility, the need for earlier intervention to support with transition. Additionally, alternative formats for parent interventions were discussed alongside benefits of the intervention.

Challenges of transitioning care

A key reason for participating in the study for three parents who started the intervention was wanting support with transferring treatment responsibility to the child. However, interviews highlighted juxtaposition between parents feeling a need to let go of responsibility whilst also maintaining high levels of control over their child's treatments. Two parents and both nurses felt that anxiety about the child becoming unwell was a driving force behind parents then taking charge of treatment responsibility. Both nurses reported that they felt parents struggled more with this transition that the young person, and that parents placing high expectations on their children and parents taking control were an instigator of "battles" in the parent-child-relationship. Unhelpful parenting strategies such as nagging, bribing, coercion and becoming angry and frustrated were reported by all parents when children resisted treatments and that these placed strain on their relationships with their child. Three parents stated that they hoped the Triple P intervention would support more positive relationships with their child due to the difficulties that treatment adherence created within these relationships. Significant changes in parental role as a result of reducing their responsibility over CF disease management were also reported by nurses to be an associated area of difficulty with parents at this time.

"You try to do everything for them. I think that has been a mistake that I have made actually [...] I have waited on her basically hand and foot. I've tried to make her life easier by me doing everything for her, because I've felt guilty that she has to do all these treatments […] So now I think I've made a rod for my own back really 'cause now when I ask her to do anything its "do I have to? "" (Parent interview 1)

Barriers to participation

Time: All parents talked about the demands of having a child with CF and how supporting their child with CF treatments took a considerable amount of time each day. This alongside the addition of needing to attend appointments, picking up regular prescriptions, and preparing treatments (i.e. sterilisation), as well as managing other normative task was the main reason why most parents declined participation. Two parents and both nurses felt that the unpredictability of CF made it hard to commit regular time to the intervention and that strict methodological issues such as the need for parents to consistently complete weekly modules in time for weekly data collection was pressurising and potentially off putting. It was felt that greater flexibility in the time provided to complete each module would improve retention.

"I'm on my own, so […] I work half of the week and do very long shifts so on them days there's absolutely no way I would do something like that. But you know, I might do on the other days if it is just an hour, but I am sure that people with other kids and that are working, if I am honest I don't think they would do it." (Parent interview 3)

Similarly, two parents suggested that the use of bite-sized chunks of information summarising headline Triple P strategies would be a more feasible adaptation of the intervention, as the manual was perceived to be "too wordy" and "repetitive" (Parent 5). This view was echoed by the CF nurses who reported that the size of the manual may have been off-putting for parents. Parents reported that the intervention involved too much "paperwork", and that the monitoring tasks in particular were too laborious and difficult to apply during challenging situations.

Parents not identifying the problem: CF nurses felt that parents would not open themselves up to the possibility of parenting support before they had themselves

acknowledged a difficulty with adherence. They felt that parents could feel confused or criticised if offered parenting support prior to this. Nurses felt that they would often become aware of the need for support before parents. There was a sense that parents would respond more positively if parenting support was initially introduced or offered via the CF team rather than an unknown researcher due to the closer relationship.

"I think what we have found is we can see, or we feel that we can see problems before that parent actually verbalises and acknowledges that there is an issue and it's not until they have realised themselves it's an issue that they are open to help. Whereas actually if you, if we had almost a tool box we could then go through and say "try this, try that", you know." (Nurse 1)

Accessibility: CF nurses discussed how the language used in the Triple P manual was likely to be above the reading level of many families within their catchment area. They suggested that presenting information in an array of formats such as video clips might facilitate accessibility, and reduce demands in reading abilities.

Earlier intervention: CF nurses felt that the Triple P intervention would be difficult to implement during adolescence, especially if families had functioned very differently prior to the commencement of the intervention. It was felt that starting such interventions earlier would allow parents to get more into a "habit" of parenting in this way and that there would be less difficulties in adjusting to a new approach. Despite treatment adherence difficulties becoming prevalent during adolescence, several parents discussed how they would still have benefitted from parenting advice and strategies earlier in childhood.

"Earlier so they are already in that mode of...just open to trying new things or saying... I think if you wait until they have got to the teenage years - if they have never done anything like that before I think it might be quite difficult." (Nurse 2)

Other formats of parenting support

Peer support from other parents: Four parents felt that support networks involving other parents would be beneficial in supporting them with their child's treatment adherence. Three parents talked about how such support would provide a sense of shared experience and reassurance. Four parents felt this would allow the sharing of strategies that other parents have found to be effective from their own experience and that these ideas would be more acceptable and meaningful as a result of coming from lived experience rather than a manualised approach.

"It's always better to come from someone who has been there themselves, who has experienced it. We can all read out of a text book. I know everything there is to know. But unless you have experienced it personally I don't think you can put it across the same." (Parent interview 2)

Online support

Four parents felt that making advice available via online forums would increase accessibility and allow parents to access resources more flexibly around other commitments. CF nurses also stated that struggling parents would often refer to CF related blogs and forums as a first line of enquiry when finding things challenging. Therefore, it was felt that posting parenting strategies via these forums would increase accessibility.

Benefits of the intervention

A number of benefits were reported by parents who completed the intervention. Participant 6 reported that it had helped her to manage her own expectations around transitioning CF disease management to her son and to respect his decisions as a young adult. She felt it helped her to realise that previous conflict had arisen out of her son's reaction to high parental expectations, rather than a problem lying solely within her child. She stated that changing her own expectations and parenting behaviours had led to a more positive and mutually respectful relationship with her son, and had facilitated with the transfer of treatment responsibility.

"Before I started doing the programme I felt that he had a problem taking responsibility and having done the programme I would fully say now that he was only reacting to my problem […] And part of what this has done for me is enabled me to just accept that he is on a learning curve and he is not going to do it perfectly." (Parent interview 6)

Participants reported the development of a core set of skills as a result of the intervention, such as active listening, stating viewpoints calmly and consistently and not being drawn into arguments. One parent reported that this approach meant that she became angry and upset less frequently leading to improved parent-adolescent relationships. Participant 6 also discussed how the Triple P had helped her to reconnect with her teenager and to appreciate the need for and benefits of providing praise and positive attention.

"In my mind [treatment behaviours] were obvious behaviours and I thought they weren't to be rewarded because they were to be expected […] little things like physical attention and touch, smiles, things that seem terribly obvious weren't happening so nothing was reciprocated. So the simplest little things like touch and smile can make significant difference and I hadn't noticed that before." (Parent interview 6)

Discussion

This study is the first to report the outcomes and feasibility of a developmentally appropriate self-directed parenting intervention in adolescent CF. Overall, results from two participating parents suggest this intervention could be associated with improving treatment adherence and positive parenting practices. Visual inspection of the data indicated that both children's treatment adherence increased following the implementation of the intervention and continued to increase throughout the intervention period. However, the increasing trend in participant 4's baseline treatment adherence scores makes it difficult to determine whether further increases shown during the intervention were due to the intervention itself or due to an extension of a pre-existing increasing trend.

Furthermore, both parents demonstrated a reduction in the use of negative and unhelpful parenting practices following the implementation of the intervention, with one parent showing clear increases in parenting competence throughout the intervention. The results are consistent with previous research with younger children with CF, which has consistently shown notable and significant improvements in treatment adherence as a result of behaviourally informed interventions (Hourigan et al., 2013; McClellan et al., 2009; Stark et al., 2003; Stark et al., 2009). The results are also consistent with previous research that highlights the positive role that parents can play in promoting adherence throughout adolescence (Laursen & Collins, 2009; Taylor et al., 2008). These encouraging findings suggest that further larger scale investigations of parenting support interventions within this age group would be beneficial.

Overall both parents who completed the intervention seemed to be functioning emotionally well prior to the intervention, as indicated by DASS-21 scores. However, one parent did show a reduction in clinically elevated stress levels, which returned within the normal range by the end of the intervention, providing preliminary evidence that the intervention may have had positive effects of parental wellbeing. The fact that both parents seemed to be functioning psychologically well prior to the intervention reflects previous research, which suggests that parents of children with CF are a resilient group who demonstrate no higher stress levels than the rest of the population (Ullrich, Bobis, & Bewig, 2016). However, it is also possible that parents who have the most significant psychological difficulties supporting their adolescent with disease management behaviours are the least likely to put themselves forward for support, for example, due to increased time demands as a result of more complex treatment regimes. Research within the wider parenting literature has reported the difficulties in engaging those families most in need of parenting support (Ingoldsby, 2010; Morawska & Sanders, 2006). Further research is needed to explore possible emotional factors that might differ between participants and non-participants in supportive interventions in order to be able to provide interventions for those families most in need of psychological support.

The small sample size and different trends demonstrated in the two children's emotional and behavioural functioning makes it difficult to draw any firm conclusions regarding the interventions effects on parent-reported child wellbeing. One child demonstrated normal levels of child emotional and behavioural functioning prior to the intervention indicating no difficulties within this area. However, participant 6 did show clinically elevated scores on these measures prior to intervention and demonstrated a reduction in measures of

child stress, conduct and peer difficulties post-intervention. Therefore it is possible that this intervention has the potential to have a positive influence on child wellbeing, but a larger sample is required to confirm this.

Whilst the results of this research tentatively suggest that parenting interventions may have a positive influence on treatment adherence and positive parenting practices, retention data alongside parent and staff interviews indicate low feasibility and acceptability of this intervention within its current form. The burden on families to commit to weekly hour-length sessions was reported by parents and staff to be a significant barrier to the intervention. This is consistent with the retention difficulties found in previous parenting interventions for younger children with CF, where time has been reported as a key barrier (e.g., Hourigan et al., 2013). Furthermore, the need for parents to complete weekly work in time for weekly telephone data collection was perceived to be pressurising and indicates that the research methodology itself may have had a negative influence on the acceptability and feasibility of the intervention. Therefore, future researchers will need to be mindful of the time consuming and unpredictable nature of parenting an adolescent with CF and develop appropriately flexible research methodologies in order to optimise uptake and retention.

Condensing the key elements of the intervention into a more manageable format given the time demands placed on carers of adolescents with CF was a key theme that emerged from the parent interviews. Parenting interventions such as Self-Directed Teen Triple P comprise a multi-faceted package of parenting knowledge and skills and there is a lack of research examining exactly which components are most essential (Gardner et al., 2010).

This is perhaps even more apparent within the field of chronic illness where fewer parenting intervention studies have been conducted. Westen, Novotny and Thompson-Brenner (2004) have suggested that there is a need to move beyond examining complete parenting packages to examining individual strategies and processes of change. It is suggested that such analyses will optimise intervention effectiveness by integrating components that are reliably associated with greater effectiveness, and eliminating or reducing emphasis on less effective components. Meta-analysis is one approach that has recently been used to examine the active ingredients of parenting interventions within the wider child conduct difficulty literature (Kaminski, Valle, Filene, & Boyle, 2008). A similar approach could be suitably applied to the chronic illness literature, where different strategies and mediators are likely to exist. Future research using such approaches alongside component and moderator analyses will be important in ensuring that parenting interventions are suitably adapted to reducing participant burden and increase cost-effectiveness and efficacy (Gardner et al., 2010).

Low uptake to the current intervention needs to be situated within similar intervention studies conducted within paediatric chronic illness. Similar recruitment difficulties have been observed within the paediatric asthma population, whereby only 10% of families approached completed a web based Triple P intervention (Clarke & Calam, 2012). Interestingly, another similar study within the adolescent diabetes population had no difficulties in recruiting to a randomised controlled trial evaluating the effectiveness of Self-directed Teen Triple P (Doherty et al., 2013). Taken together, these findings suggest that there may be important psychosocial differences between chronic illness groups that might need to be taken into account when developing and implementing parenting

interventions. It is possible that the severity and endurance of symptoms and the nature and demands of different treatment regimens might place different demands on parents of children with different chronic illnesses (Clarke & Calam, 2012). In this way more individualised and specially tailored interventions may be needed rather than broad-spectrum parenting approaches.

One area to consider when developing tailored parenting interventions within the adolescent CF population is the issue of letting go of treatment responsibility. This was a key theme raised by all parents interviewed, and was associated with conflict and strained parent-child relationships. This is consistent with previous research findings within the wider chronic illness literature (Schilling, Knafl, & Grey, 2006). The experience of letting go of treatment responsibility expressed by parents fits within the grounded theory reported by Williams, Mukhopadhyay, Dowell and Coyle (2007). This theory posits that changes in the child's health status influence the transition of treatment responsibility from parent to child. As symptom severity increases, parental anxiety about allowing their child to self-manage also increases, leading to a renegotiation in disease management roles and associated parent-child conflict.

Importantly, parents who completed the intervention reported that it had facilitated this transfer of responsibility. It would therefore be helpful to explore which elements of the intervention specifically facilitated this process in order to streamline future interventions. Additionally, future research should explore the psychological factors and processes that facilitate parental acceptance and transfer of treatment responsibility, for example, by interviewing families who have successfully managed this transition. Most

available research has focussed around transition from paediatric to adult CF services (Boyle, 2001; Brumfield & Lansbury, 2004), which is surprising given that most transition occurs within the domestic environment (Williams, 2007). A fuller understanding of these issues and processes as experienced by parents and adolescents is required if interventions are to be appropriately responsive. Indeed the process of treatment responsibility transition is complex, involving a multitude of disease related factors, parent and child illness beliefs and attributional factors, parental role shifts, and dynamic family factors (Leeman, Sandelowski, Havill, & Knafl, 2015) and it is therefore probable that additional support structures will be required alongside parenting interventions.

As well as considering which elements of parenting interventions are essential for effectiveness and necessary for meeting the specific needs of parents of children with CF, the current research has highlighted that issues regarding intervention facilitator and intervention format are also important considerations in order to maximise acceptability and feasibility. Nurses suggested that parents may not have been open to the intervention due to not acknowledging that there were adherence difficulties despite such problems being identified by CF nurses. It is possible in these circumstances that an unknown researcher presenting parents with a parenting intervention could be perceived as confusing and invasive. Previous research has also suggested that parenting interventions can be perceived by parents as stigmatising (Koerting et al., 2013). This may be even more prevalent in medical settings whereby a psychological approach is less dominant. Given clinical psychologists' increasing role in teaching, training and consultation they are in a prime position to provide training to CF nurses and other

medical colleagues in order for them to develop an increased understanding of the relational elements associated with treatment adherence and to provide emotional and practical parenting support to parents within the context of a pre-established close professional relationship. Such an approach may be more acceptable for parents. It has also been shown to improve psychological thinking and practice amongst paediatric teams, as well as increasing staff confidence in managing complex and difficult scenarios (Douglas & Benson, 2015).

Both CF nurses and parents felt that support and strategies were more likely to be taken on board if they came from other parents who had tried similar strategies The use of testimonies from parents who have completed the intervention might therefore improve uptake (Morawska et al., 2011) as might the inclusion of more specific examples of how Triple P strategies can be applied to the adolescent CF population. Furthermore, providing summarised Triple P information and advice sheets via CF web forums may also increase uptake because the current research suggests that parents would usually turn to such forums first at the point of wanting support. The addition of visual materials and video clips that supplement information provided in the Tip Sheet and may also reduce literacy barriers and offer other stimulating ways to engage parents.

Limitations of the research

The use of case series designs is based upon the findings of a small number of participants, in this case two parents. Whilst such designs can be informative when exploring the feasibility and acceptability of new and previously un-researched

interventions (Wells & Papageorgiou, 2001), the use of such methods limits the generalisability of any intervention effects. Data from two participants is insufficient to draw firm conclusions about the effectiveness of the Triple P programme within this population. Additionally, the lack of stability in Participant 4's baseline treatment adherence scores makes it difficult to be confident that subsequent increases in treatment adherence scores were a result of the intervention or due to other extraneous factors. Therefore, the findings discussed should be interpreted with significant caution. Future research utilising larger sample sizes and an attention control group would increase the reliability and rigor of the research findings presented. However, the current research has shown that further pilot work to streamline parenting interventions within this populations is warranted prior to investing in more costly designs. Involving parents and adolescents in the design phase of research studies and interventions is likely to lead to more acceptable and feasible support options for families.

The authors also acknowledge that there may be limitations with the analysis used for the case series data. A number of authors suggest that visual analysis should be the sole, or at least primary method for the analysis of case series data (Baer,1977; Parsonson & Baer, 1978; 1986), and that such an analysis will reveal any intervention effects large enough to be important for clinicians. However, Kazdin (1982) has stated that statistical analysis may be of additional value when there is a lack of baseline stability, as is the case in the current research, and when statistical control is needed for extraneous factors that are inherent within naturalistic studies. The rigor and reliability of the present case series results could therefore have been improved via the addition of appropriate statistical methods such as randomisation tests, time-series analysis, or trend analysis.

The use of parent reported treatment adherence measures introduces other limitations. The increasing autonomy of adolescents often means that they spend longer periods of time away from their parents. As a result parent reports may be inaccurate or incomplete, and may be subject to social desirability effects as a result of completing these measures over the phone with the researcher. The addition of child adherence reports would have been useful to increase the reliability of these reports.

The self-directed nature of the intervention made it difficult to determine treatment integrity. Although beyond the scope of the current research, future researchers may wish to include observational measures of parenting practices and parent-child interactions to increase the measurement of intervention integrity. Furthermore, the use of adolescent self-report measures (e.g., child self-report SDQ) and additional semi-structured interview data from adolescents would have increased the richness of the feasibility information. Longer term follow-up data collection would also be clinically useful in order to determine the longevity of intervention effects.

Conclusion

The results of this study provide very tentative evidence to suggest the potential role of self-directed Teen Triple P for improving treatment adherence and positive parenting practices within this population. However, adaptations are required in order to increase acceptability. Consideration of more flexible and creative approaches such as the use of psychological consultation and providing supportive information via well-established CF web forums may increase uptake to parenting interventions. Given the paucity of research within this area future research should continue to develop an evidence base for

developmentally appropriate parenting interventions within this age group. As retention to parenting interventions across the paediatric CF age range is problematic, more research is needed to explore potential barriers to engagement.

Compliance with ethical standards

The authors declare that they have no conflict of interest and there were no funding sources for this study. All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. Informed consent was obtained from all individual participants included in the study. This article does not contain any studies with animals performed by any of the authors.

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Paper 3

Critical Appraisal

Word Count: 4966 (excluding references)

Introduction to Paper 3

This paper offers a critical review of the many different elements of this research project. It offers critical reflections from the start of the research process, through to the development, completion and evaluation of the literature review (Paper 1) and empirical paper (Paper 2). Consideration is given to the strengths and weaknesses of each paper as well as the challenges that were faced. In addition, implications for future research and clinical practice are discussed.

Paper 1 – Literature review

Rationale for review topic

The development of interventions to support treatment adherence in chronic health conditions, such as CF, have been assigned high priority by the World Health Organisation (Sabaté, 2003). Whilst there is a lack of formalised standards of psychosocial care for people with CF, developing European guidelines state that psychologists should take the lead on the management of partial treatment adherence, and participate in the application of evidence based interventions to support with this (Kerem, Conway, Elborn, & Heijerman, 2005). During childhood, parents play an integral role in promoting treatment adherence and should therefore be a key target for psychological support and intervention. Whilst a number of literature reviews have highlighted the positive role that parenting interventions can play in promoting treatment adherence across paediatric chronic illnesses (Kahana, Drotar, & Frazier, 2008; Law, Fisher, Fales, Noel, & Eccleston, 2014), very little research has examined the use of such interventions within CF.

Although an initial scoping exercise revealed that a literature review had been undertaken in this area (Bernard & Cohen, 2004), this review was not conducted systematically, making it difficult to ascertain the quality, strengths and weaknesses and reliability of the findings discussed. Furthermore, this review was conducted over a decade ago. It is only within the last ten years or so that the median survival rate in CF has exceeded 30 years due to medical advances (Sawicki & Tiddens, 2012). Therefore in the years since the previous review, clinicians and researchers may have become increasingly aware of the importance that treatment adherence plays in preserving physical functioning and quality of life, and may have greeted research within this area with greater enthusiasm. An up to date systematic review of the literature regarding parenting interventions in CF was deemed beneficial in order to be able to paint a current and holistic picture of the evidence base for these interventions.

Literature search

Due to the number of different terms used to describe parenting interventions an over-inclusive approach was adopted during the literature search. This was recommended following consultation with a specialist University librarian and via examining search terms used in previous reviews within other chronic illnesses. Using a diverse array of search terms reduced the risk of missing relevant papers. However, a large number of irrelevant papers were retrieved, indicating that search terms may well have been too broad. Search terms need to be balanced between breadth and focus to ensure that relevant papers are not missed, whilst ensuring that a minimal number of irrelevant papers are retrieved (Smith, Devane, Begley, & Clarke, 2011). More thorough testing of

the search terms prior to undertaking the review might have been helpful to streamline this process.

A further challenge regarded the inclusion and exclusion criteria. It is generally accepted that criteria should be set to limit the kinds of evidence included in a systematic review as the risk of bias varies across studies (Reeves, Deeks, Higgins, & Wells, 2008). It is generally recommended that systematic reviews limit their searches to randomised controlled trials because they are regarded as the gold standard for research evidence. However, the lack of research within the area covered in Paper 1 meant that such a restriction could not be placed on the search. The researcher therefore consulted systematic review guidance (Reeves et al., 2008) and discovered that in these circumstances it is recommended that researchers include a synthesis of the best available evidence, whereby the inclusion of non-randomised and observational studies is permitted. As such all study designs were permitted for inclusion. In order to promote transparency the findings of randomised and observational research designs were reported separately in line with systematic review recommendations (Reeves et al., 2008). It was hoped that this would reduce the potential for additional bias being added to interpretation of the findings.

Selecting an appropriate quality appraisal tool

The inclusion of an array of research designs within the current review made it difficult to determine the most appropriate method of assessing study quality. Initially, separate design-specific tools were considered for the evaluation of different types of study design. Whilst the rigor of quality assessment is generally increased via the separate application of such tools, it creates difficulties when trying to evaluate the overall quality

of the body of evidence discussed due to the high levels of diversity in different scoring criteria (Katrak, Bialocerkowski, Massy-Westropp, Kumar, & Grimmer, 2004). The Quality Assessment Tool for Diverse Designs (Sirriyeh et al., 2012) was therefore chosen to assess research quality, as this is an inclusive tool that facilitates the synthesis of evidence across allied health research designs. Although this tool was chosen the researcher acknowledges that there are a number of limitations also inherent in inclusive quality appraisal tools. Firstly, such tools lack specificity in asking the 'hard' questions about research quality related to specific research design features (Katrak et al., 2004). For instance, such tools do not consider design-specific methodological issues such as baseline length or stability and frequency of measurement intervals within case series designs. Secondly, questions have been raised regarding how clinically useful these tools are because of the generalist nature of their items, and variable interpretation of items across research designs (Andresen, 2000; Katrak et al., 2004). In light of these limitations, the author used this tool as a guide to aid in the critical appraisal of research quality but supplemented this with more detailed discussion of the methodological limitations throughout Paper 1.

Findings and limitations

The results of the systematic review suggested that treatment adherence and associated physical health outcomes can be improved following the implementation of behaviourally oriented parenting interventions, with both randomised controlled trials and observational studies demonstrating encouraging findings. These findings provide preliminary evidence for the potential role of these interventions within routine clinical practice; however, more research using rigorous research designs and sufficient sample

sizes is required to increase reliability and confidence. The majority of studies were observational in nature and the small sample sizes used in such studies necessitates caution when interpreting results because there is limited generalisability of such findings. Furthermore, when including observational study evidence, systematic reviewers have to consider the possibility of extraneous variables that may have an influence on the results. This makes it more difficult to draw reliable conclusions with regards the effectiveness of the interventions under investigation. For instance, the lack of attention control groups within observational studies makes it difficult to ascertain whether the intervention itself triggers improvements in adherence, or whether more general contact with a psychological professional could account for such improvements. Additionally, it is possible that increased monitoring of treatment adherence and parenting practices may have influenced parental and treatment behaviours. Additional limitations regarding the studies included in the literature review and of the literature review method itself are provided in Paper 1.

Paper 2 - Empirical paper

Rationale for the research topic

The supervisor's recent research within the domain of parenting interventions and asthma (e.g., Clarke & Calam, 2012) had highlighted the potential for developing parenting interventions within other respiratory conditions, such as CF. In order to further develop and refine the research question a meeting was convened with a local paediatric CF centre. The aim of this meeting was to explore whether parenting interventions were of potential usefulness to the CF team, and to ascertain within which age groups or sub-populations such interventions were deemed to be potentially beneficial. The CF team

were receptive to the idea of parenting support and highlighted that adolescence was an age at which treatment adherence and associated family and parenting issues became more challenging. Therefore the design and rationale for the empirical study had their roots firmly within the needs and experiences of CF teams. The researcher then explored the literature and discovered that there was a clear lack of parenting interventions specifically designed to support treatment adherence during adolescence, alongside consistent reports of increased adherence difficulties within this age group. Taking the findings of the literature and the views of CF professionals together, the researcher felt that there was a clear need to explore the use of parenting interventions to support adolescent treatment adherence.

The research supervisor has an extensive background in examining Triple P parenting interventions and had recently been involved in a doctorate student project that had reported positive findings regarding the use of the Self-Directed Teen Triple P intervention within adolescent diabetes (Doherty, Calam & Sanders, 2013). This intervention therefore offered a promising, flexible, and developmentally tailored intervention that was worthy of investigation within adolescent CF.

Why a case series design?

Within the hierarchy of research evidence, case series designs do not rank highly. The lack of control subjects makes them prone to biases, such as selection bias, and this limits the generalisability of findings obtained. In spite of these limitations, case series designs offer a resource-effective and feasible approach to inform researchers about the preliminary effectiveness of new interventions (Wells, Fisher, Myers, Wheatley, Patel, & Brewin,

2009). They are also helpful in refining new interventions prior to the initiation of more advanced, costly and resource intensive trials. Given that Paper 2 was, to the author's knowledge, the first study of its kind to investigate the use of Self-Directed Teen Triple P for parents of adolescents with CF, the case series design therefore provided a useful methodology to explore the initial effectiveness and feasibility of this intervention within this population.

Case series designs – Methodological considerations

In an attempt to increase the reliability and generalisability of case series data, a number of recommendations (Barlow & Hersen, 1984) were considered during the design phase of the study. The first of these recommendations was the use of an A-B-A-B reversal design whereby, following a baseline period (A), the intervention (B) is introduced and then removed following change being observed in the data. Such a strategy allows a researcher to confirm a treatment effect by showing that behaviour changes systematically with conditions of No Intervention and Intervention (Barlow & Hersen, 1984). However, the learning that occurs during the Triple P intervention is irreversible meaning that carry over effects would contaminate this withdrawal phase. A second option that was therefore considered was the use of a multiple baseline across subjects' designs. This type of design does not require the removal or reversal of a treatment condition. Instead interval validity is ensured by the multiple replications of the intervention being delivered to different subjects after different length baselines. Each transition from baseline to intervention is an opportunity to observe the effects of the treatment. Because each participant makes this transition at different times, it allows the

researcher to rule out alternative explanations for any behaviour changes that occur during treatment (Morgan & Morgan, 2009).

In line with the multiple baseline design initial participants were randomly allocated to baselines of differing lengths (ranging from 3-8 weeks); however, due to significant recruitment difficulties reducing the project time, this had to be adapted. The two participants who were able to complete the intervention were recruited late in the research process and therefore baseline length had to be capped at 2 weeks for participant 6 and 3 weeks for participant 4. The researcher acknowledges that this limits the conclusions that can be drawn from the research, but such changes were unavoidable within the timescale of the project.

Recruitment

A preliminary meeting with a local NHS CF team secured recruitment support for the study prior to applying for NHS ethical approval. This team examined their patient lists and felt that there would be sufficient interest in the study to allow the use of a case series approach. This CF team was able to contact three other North West CF teams directly in order to gain preliminary permission for their involvement in the research. The researcher was able to liaise with CF colleagues from these teams via face-to-face meetings and telephone calls in order to engage teams within the research process. Following NHS Research Ethics Committee approval, individual Research and Development (R&D) approvals from four NHS Trusts were coordinated in a stepwise fashion in order to reduce any possible ethical issues associated with over-recruitment and the need to potentially turn parents away from the intervention (see Appendix T for

individual R&D approval letters). Therefore only two NHS Trusts were initially involved. The first two CF teams who provided local ethical approval were very proactive and 68% of all potential participants were identified from these sites. Whilst this was initially promising, it proved very difficult to contact parents and a number of parents dropped out during the first weeks and months of recruitment. This experience was unnerving for the researcher who was constantly oscillating between a place of satisfaction with recruitment (and therefore stopping recruiting other potential families) and pressure as a result of attrition.

After several weeks of slow recruitment it was decided that approval should be applied for from the two additional NHS Trusts. Unfortunately, these approvals also took considerable time and further reduced the timescale of the research project. Furthermore, due to pressures within these CF teams recruitment was understandably not a team priority, therefore meaning that additional recruitment was slow. The researcher was mindful of balancing recruitment concerns with being amenable to the competing time demands faced by CF teams.

In December 2015 the researcher contacted the CF Trust in a final attempt to increase recruitment. They were able to post an advertisement about the research project on their Facebook page. Whilst this received a substantial number of 'likes', only a handful of families contacted the researcher for more information and only two parents were fully engaged in the programme by early 2016. Whilst the researcher was hoping to have more families involved in the intervention, low uptake and attrition provided informative data and highlighted the researcher to the possibility that the Triple P intervention in its

original form may not be acceptable and feasible for parents. It was therefore felt that gaining additional perspectives from parents who were unable to participate in the intervention or who withdrew would be a more clinically useful trajectory to pursue during the remaining research timescale and would inform the development of more agreeable and tailored interventions. Therefore, in early 2016 an amendment was put through NHS ethics in order to allow for the capture of this information from parents and CF staff via semi-structured interviews.

The importance of using a consumer perspective approach

Despite the researcher's initial disappointment regarding the low uptake to the intervention, the process of gaining additional consumer experiences and views was very informative and pointed the researcher to the importance of including such perspectives within health service developments. Very few CF related parenting intervention studies have included formalised investigations of feasibility and acceptability, despite consistent reports of poor uptake and high attrition, as highlighted in Paper 1. Therefore, the additional aim of investigating feasibility and acceptability of the Self-Directed Teen Triple P intervention is considered a key strength of Paper 2. A qualitative approach was chosen in order to be able to capture rich and individualised information regarding parents' experiences using the intervention and barriers to use. Although parents were the active recipients of the intervention, healthcare providers are also considered to be important consumers (Sanders & Kirby, 2012) due to their key role in supporting families with the demands of CF treatments. Additional insight from a small number of CF nurses regarding their professional experience and views about the intervention were deemed important in increasing the richness of feasibility information, as well as providing additional

information regarding the applicability of this intervention within routine CF care. It is noted that feasibility and acceptability information could have been extended to include the views of adolescents.

The benefits of adopting a consumer perspective approach within the development of interventions in new populations have been documented in several studies (Metzler et al., 2012; Sanders & Kirby, 2012). Within the current research, consumer perspectives were collected from parents who had dropped out of or completed the Self-Directed Teen Triple P parenting intervention. This is in line with recent Medical Research Council Guidelines (Craig et al., 2008) which suggests that existing interventions should be trialled prior to the development of new ones in order to increase resource and cost-effectiveness. In retrospect, it may have been more informative to include consumer perspectives from an earlier stage in the development of the intervention, particularly considering the low uptake and retention to the intervention. A number of theories within the consumer perspectives approach, such as the Participatory Action Research paradigm (PAR) (Whyte, Greenwood, & Lazes, 1989) and Diffusion of Innovations Theory (Rogers, 2003) are being increasingly used in social sciences and health research. These theories advocate for the direct involvement of consumers from the outset of intervention and research design in order to facilitate in the development of more valid and meaningful products and interventions (Greenhalgh, Robert, Macfarlane, Bate, & Kyriakidou, 2004) and to promote consumer interest in the success of the intervention, cooperation and fidelity. In line with these suggestions, consultation with parents and staff during the design phase of the research may have highlighted that the Triple P resources required considerable adaptation in order to be feasible and acceptable. Earlier

consultation may have resulted in the development of a different but related research project that may have avoided the recruitment difficulties experienced in the study, and may have focussed more around the development of a more tailored and CF specific parenting intervention which may ultimately have greater feasibility and acceptability.

Sample characteristics

All participating parents were mothers. This is consistent with other research which demonstrates that mothers are highly represented in research due to assumptions that they undertake the main caregiver role (Phares, Lopez, Fields, Kamboukos, & Duhig, 2005). However, given that some research indicates that paternal reports of psychological distress and child behavioural difficulties are greater than those reported by mothers (Sanders, Haslam, Calam, Southwell, & Stallman, 2011), research is required that identifies factors that increase paternal involvement in order to ensure they are adequately represented and supported.

In addition to this, both parents who completed the intervention came from highly educated backgrounds, with both having at least undergraduate level degrees. This is consistent with other research which shows that self-selecting samples tend to result in greater proportions of white females with higher levels of formal education accessing interventions (Buis, Janney, Hess, Culver, & Richardson, 2009; Stopponi et al., 2009). These individuals are more likely to have been involved in research previously, and may therefore have been more likely to take part. Both parents were also married and in employment. This might suggest that these parents were able to share the parenting burden of supporting a child with CF and may potentially have lower extraneous stressors

such as financial difficulties, which may impact upon the ability to cope with the challenges of parenting a child with CF, as well as to commit time to research projects. Whilst research has indicated that socioeconomic status has an impact on parental wellbeing and child health status (Adler et al., 1994), there was insufficient scope and participant numbers within the current research to systematically explore socioeconomic factors.

Difficulties in measuring treatment adherence

Research has highlighted that a major stumbling block in the development of methodologically sound adherence studies is the problem of objectively measuring treatment adherence (Quittner et al., 2000). The use of self-report measures, such as the Treatment Adherence Questionnaire (TAQ-CF), is associated with an increased risk of social desirability biases (McEwan, Davis, MacKenzie, & Mullen, 2009). Research has demonstrated that self-report measures overinflate treatment adherence estimates when compared to objective electronic monitored measurements (Modi et al., 2006). Such biases may have been even more prevalent due to questionnaires being completed over the telephone with the researcher. In hindsight this measure could have been supplemented with a measure of social desirability effects, for example, the Marlow-Crowne Social Desirability Scale (Loo & Thorpe, 2000; Reynolds, 1982; Strahan & Gerbasi, 1972).

Furthermore, it is recommended that a range of treatment adherence measures are utilised and triangulated (Quittner et al., 2000). The use of daily phone diaries reduces reliance on protracted retrospective accounts of adherence which may be inaccurate

(Quittner & Espelage, 1999). Additionally, the development of new electronic monitoring systems has the potential to provide objective data for the performance of certain elements of the CF treatment regime. Whilst triangulation of such methods would likely increase the reliability of treatment adherence measurement, such an approach was impractical within the scope of the current research. Firstly the researcher was mindful of the burden of placing increasing demands on each participant's time. Furthermore, the use of electronic monitoring is limited to particular aspects of CF treatment regime, therefore precluding the objective measurement of other CF treatments. Additionally, as some parents were recruited from charities appropriate ethical approvals were not in place to capture this information.

Selection of measures

It is acknowledged that parents were required to complete a number of different outcome measures throughout the intervention and the researcher was mindful of ensuring a balance between comprehensive data collection and reducing burden on participant time. Given the interactions that have been demonstrated to exist between treatment adherence, parenting practices, parental emotional wellbeing, and parental self-efficacy (see Paper 2 for more details), inclusion of measures assessing these variables was deemed to be informative, allowing for the investigation of possible wide spread psychosocial influences of the parenting intervention.

Having explored the parenting literature a number of measures were located that would allow investigation of these factors. The Triple P programme routinely uses a core set of parenting questionnaires including the Parenting Scale- Adolescent version (Irvine, Biglan,

Smolkowski & Ary, 1999), Parent Sense of Competency Scale (Johnson & Mash, 1989), Depression, Anxiety and Stress Scale (Lovibond & Lovibond, 1995a) and Strengths and Difficulties Questionnaire (Goodman, 1997). It was felt that the inclusion of these measures would allow for direct comparison to other Triple P studies, therefore situating any changes within the wider parenting literature. However, a limitation of these measures is that they are normed on outcomes from the general population and therefore lack the specific challenges posed by CF. Research has shown that parents of children with CF are generally a highly resilient group who cope remarkably well in the face of considerable psychosocial stressors (Ullrich et al., 2016). It is also well known within the CF literature that parents demonstrate positive biases in their views about their own wellbeing which reflect a 'need to be normal' and the presence of a different personal scale upon which stressors are evaluated (Ullrich et al., 2016). These findings suggest that parents of children with CF may show differences in their experience and reporting of stress and emotional wellbeing compared to normative samples. In hindsight, it may therefore have been beneficial to include more disease specific measures of parental and child wellbeing.

Whilst the Pediatric Inventory for Parents (PIP) (Streisand et al., 2001) was utilised, this has been normed across a heterogeneous group of chronic illnesses. The combination of the progressive nature of CF alongside the chronic enduring symptoms and disease management associated with the disease means that the nature, frequency and extent of various stressors are likely to be quite different to other conditions, for example, such as asthma where symptoms and disease management is more intermittent (Clarke & Calam, 2012). These factors may mean that the PIP was not the most appropriate measure to

assess change associated with the intervention. A wider measure of parental quality of life may have been more informative and would be more likely to capture how well parents are able to manage in spite of continual unforeseen stressors. The Cystic Fibrosis Questionnaire (Henry, Aussage, Grosskopf, & Launois, 1996) is a quality of life measure which may have offered clinically meaningful findings.

Format of data collection

It was felt that paper-based methods of data collection would increase the likelihood of attrition. The additional demands for parents to post weekly questionnaires back to the researcher may have been off-putting and may have resulted in unnecessary delays in data collection due to forgetting, for example. Whilst web-based approaches for questionnaire completion were also considered favourably, liaison with a web technician led to the conclusion that the amount of questionnaires used and the diversity within their formats would make designing a web-based survey too challenging. In light of this, it was decided that telephone questionnaire completion would be the most advantageous and practical way to collect data. A major advantage of this approach was that the researcher was able to prompt parents and ensure that they completed relevant modules of the intervention in line with timely data collection. It also facilitated the process of engagement, which due to the demands of looking after a child with CF and the demands of the intervention, has been considered to be an essential part of designing and conducting interventions within this client group (Quittner et al., 2000).

Data analysis

Statistical advice was sought throughout the study. Initially it was hoped that 10 parents could be included in the case series in order to increase reliability and to allow the use of simple inferential statistics on pre- and post-intervention data. However, due to the unforeseen low uptake and continuation difficulties this was not possible. Therefore visual inspection of results via graphical representations was used to analyse the case series data. This is in line with recommendations from the case series literature (Barlow & Hersen, 1984). It has been acknowledged earlier in this paper that the generalisability and reliability of the research findings is therefore limited and results should be interpreted with caution. In order to optimise reliability, weekly measures were collected rather than limiting data collection to pre and post intervention time intervals.

The method of analysis of semi-structured interview data was debated. Due to this aspect of the methodology being added relatively late on in the research process, it was felt that there was insufficient time to complete an in depth qualitative analysis (i.e. interpretive phenomenological analysis, grounded theory). Furthermore, a lower level of interpretation was all that was required from the data because the focus was primarily around describing participants' experiences of the acceptability and usefulness of the intervention. Qualitative content analysis (Elo & Kyngas, 2008) was therefore chosen as an appropriate analytical framework because this method focusses primarily on the manifest content of interviews. It also allows for the quantification of the themes discussed, thereby providing a proxy measure of clinical significance by highlighting the proportion of participants for whom the theme was relevant (Vaismoradi, Turunen, & Bondas, 2013). However, it is important to note that qualitative analysis does not exist

within a value free framework, and although the use of inter-coder reliability checks is recommended to improve the rigor and reliability of interpretation (Cavanagh, 1997), time constraints did not allow for this within the current research.

Overall findings and clinical implications

Parenting interventions tailored to the needs of chronically ill adolescents are not routinely available in the NHS. Within CF this is a relatively new line of research enquiry, and feasibility studies examining readily adaptable interventions offer valuable insight for clinicians planning future interventions. The study described in Paper 2 provides preliminary support for the effectiveness of such interventions to promote treatment adherence and positive parenting practices within CF. However, the current research also raises important questions about parent engagement and the inclusion of service users and other consumers within the design of such interventions. Despite the adaptation of readily available broad-spectrum interventions being the most time and cost-effective approach, the current research suggests that we may need to go back to the drawing board and include the views of service users and consumers from the outset of intervention design. This is needed to fully appreciate parents' experiences and to successfully develop far reaching, acceptable and feasible interventions for parents who have specific demands and needs that may not be addressed by broad-spectrum approaches. It is hoped that the current research will encourage future researchers to utilise a consumer perspective approach within the design of intervention studies and to conduct more research examining the active ingredients of parenting interventions in order to streamline interventions and reduce the participant burden. It is hoped that the creative and indirect approaches discussed in Paper 2 (i.e. staff consultation and training,

the use of social media forums, etc.) will provide researchers with new ways to disseminate and evaluate parenting support and advice within CF. Finally, the views expressed by parents within the current research should prompt future researchers to develop suitably flexible research methodologies that carefully consider the daily demands of parents with CF in order to prevent the research design itself from becoming a barrier to retention.

Final reflections

Undertaking this large scale piece of research has provided many learning and development opportunities. The skills learnt and experiences gained will enhance my work both as a researcher and as a clinician. The insight gained from working with parents and CF professionals has increased my understanding and empathy for the pressures faced by these individuals. Recruitment challenges have provided me with a new appreciation of the demands of conducting applied health research and I have learnt the vital role that service user involvement can play in ensuring acceptable and feasible interventions. I hope that these skills will facilitate any future involvement that I have in service development projects as a qualified clinician.

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Appendices

Appendix A: Author Guidelines for 'Journal of Clinical Psychology in Medical Settings'

Instructions for Authors

Journal of Clinical Psychology in Medical Settings

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In general, the journal follows the recommendations of the 2010 Publication Manual of the American Psychological Association (Sixth Edition), and it is suggested that contributors refer to this publication.

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- and suggested running head

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- and state (or nation)

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References

List references alphabetically at the end of the paper and refer to them in the text by name and year in parentheses. References should include (in this order):

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- title of article
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- volume number
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- Journal Article

  Burns, J. W., & Katkin, E. S. (1993). Psychological, situational, and gender predictors of cardiovascular reactivity to stress: A multivariate approach. *Journal of Behavioral Medicine, 16,* 445−465.
- Book

  Ray, R. (2006): *Chronic Pain and Family: A Clinical Perspective.* New York: Springer.
- Contribution to a Book

  Bleiberg, J., Ciulla, R., & Katz, B. L. (1991). Psychological components of rehabilitation programs for brain−injured and spinal−cord−injured patients. In J. J. Sweet, R. H. Rozensky, & S. M. Tovian (Eds.), *Handbook of clinical psychology in medical settings*(pp. 375−400). New York: Plenum Press.

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Appendix B. Literature Review Data Extraction Proforma

Author	Article Title	Country of origin	Aims/ objectives	Study design	Inclusion/ Exclusion criteria	Recruitment processes (e.g. randomisation , blinding etc)	Unit of allocation (e.g. parents or parents and children)	Number of particpants	Parent and child age

Parent and child gender	Parent and child ethnicity	SES	No of Ps in experimental group	No of Ps in control group	Mean/ median characteristic values	Intervention setting	Dose/ length of intervention	Interventionist details	Route of administration (i.e. group, individual)

Theoretical Basis of intervention and key components covered	Details of control group interventionist and intervention	Unit of assessment / analysis	frequency of data collection	Statistical tests used	Dealing with missing data?	Length of follow-up and follow-up details	Ps enrolled/ included in analysis	Withdrawals, exclusions, lost to follow-uo?	Summary outcome data (e.g. p values, mean differences, confidence intervals)

Additional outcomes	Conclusions

Table 1 Quality assessment tool and scoring

guidance notes	Criteria	0 = Not at all	2 = Moderately	3 = Complete
1 = Very slightly				
Reference to broad theoretical basis.	Explicit theoretical framework	No mention at all.	Reference to a specific theoretical basis.	Explicit statement of theoretical framework and/or constructs applied to the research.
General reference to aim/objective at some point in the report including abstract.	Statement of aims/objectives in main body of report	No mention at all.	Reference to broad aims/objectives in main body of report.	Explicit statement of aims/objectives in main body of report.
General description of research area and background, e.g. 'in primary care'.	Clear description of research setting	No mention at all.	General description of research problem in the target population, e.g. 'among GPs in primary care'.	Specific description of the research problem and target population in the context of the study, e.g. nurses and doctors from GP practices in the east midlands.
Basic explanation for choice of sample size. Evidence that size of the sample has been considered in study design.	Evidence of sample size considered in terms of analysis	No mention at all.	Evidence of consideration of sample size in terms of saturation/information redundancy or to fit generic analytical requirements.	Explicit statement of data being gathered until information redundancy/saturation was reached or to fit exact calculations for analytical requirements.
Sample is limited but represents some of the target group or representative but very small.	Representative sample of target group of a reasonable size	No statement of target group.	Sample is somewhat diverse but not entirely representative, e.g. inclusive of all age groups, experience but only one workplace. Requires discussion of target population to determine what sample is required to be representative.	Sample includes individuals to represent a cross section of the target population, considering factors such as experience, age and workplace.
Very basic and brief outline of data collection procedure, e.g. 'using a questionnaire distributed to staff'.	Description of procedure for data collection	No mention at all.	States each stage of data collection procedure but with limited detail, or states some stages in details but omits others.	Detailed description of each stage of the data collection procedure, including when, where and how data were gathered.
Very limited explanation for choice of data collection tool(s).	Rationale for choice of data collection tool(s)	No mention at all.	Basic explanation of rationale for choice of data collection tool(s), e.g. based on use in a prior similar study.	Detailed explanation of rationale for choice of data collection tool(s), e.g. relevance to the study aims and assessments of tool quality either statistically, e.g. for reliability & validity, or relevant qualitative assessment.
Minimal recruitment data, e.g. no. of questionnaire sent and no. returned.	Detailed recruitment data	No mention at all.	Some recruitment information but not complete account of the recruitment process, e.g. recruitment figures but no information on strategy used.	Complete data regarding no. approached, no. recruited, attrition data where relevant, method of recruitment.
Reliability and validity of measurement tool(s) discussed, but not statistically assessed.	Statistical assessment of reliability and validity of measurement tool(s) (Quantitative only)	No mention at all.	Some attempt to assess reliability and validity of measurement tool(s) but insufficient, e.g. attempt to establish test–retest reliability is unsuccessful but no action is taken.	Suitable and thorough statistical assessment of reliability and validity of measurement tool(s) with reference to the quality of evidence as a result of the measures used.
Method of data collection can only address some aspects of the research question.	Fit between stated research question and method of data collection (Quantitative)	No research question stated.	Method of data collection can address the research question but there is a more suitable alternative that could have been used or used in addition.	Method of data collection selected is the most suitable approach to attempt answer the research question
Structure and/or content only suitable to address the research question in some aspects or superficially.	Fit between stated research question and format and content of data collection tool e.g. interview schedule (Qualitative)	No research question stated.	Structure & content allows for data to be gathered broadly addressing the stated research question(s) but could benefit from greater detail.	Structure & content allows for detailed data to be gathered around all relevant issues required to address the stated research question(s).
Method of analysis can only address the research question basically or broadly.	Fit between research question and method of analysis	No mention at all.	Method of analysis can address the research question but there is a more suitable alternative that could have been used or used in addition to offer greater detail.	Method of analysis selected is the most suitable approach to attempt answer the research question in detail, e.g. for qualitative IPA preferable for experiences vs. content analysis to elicit frequency of occurrence of events, etc.
Basic explanation for choice of analytical method	Good justification for analytical method selected	No mention at all.	Fairly detailed explanation of choice of analytical method.	Detailed explanation for choice of analytical method based on nature of research question(s).
More than one researcher involved in the analytical process but no further reliability assessment.	Assessment of reliability of analytical process (Qualitative only)	No mention at all.	Limited attempt to assess reliability, e.g. reliance on one method.	Use of a range of methods to assess reliability, e.g. triangulation, multiple researchers, varying research backgrounds.
Use of pilot study but no involvement in planning stages of study design.	Evidence of user involvement in design	No mention at all.	Pilot study with feedback from users informing changes to the design.	Explicit consultation with steering group or statement or formal consultation with users in planning of study design.
Very limited mention of strengths and limitations with omissions of many key issues.	Strengths and limitations critically discussed	No mention at all.	Discussion of some of the key strengths and weaknesses of the study but not complete.	Discussion of strengths and limitations of all aspects of study including design, measures, procedure, sample & analysis.

Study	Q1	Q2	Q3	Q4	Q5	Q6	Q7	Q8	Q9	Q10	Q11	Q12	Q13	Q14	Q15	Q16	Total score	Percentage score	Rating
Powers (2015)	3	3	2	0	1	3	2	3	1	3	n/a	3	3	n/a	1	3	32	76.19047619	high
Bernard (2009)	2	3	3	2	1	3	2	3	3	3	n/a	3	1	n/a	0	2	32	76.19047619	high
Hourigan (2013)	3	3	3	1	1	3	2	3	3	3	n/a	3	1	n/a	0	2	32	76.19047619	high
McClellan (2009)	2	3	3	3	1	3	2	1	2	3	n/a	3	2	n/a	0	2	31	73.80952381	good
Powers (2006)	2	3	3	2	2	3	3	3	1	3	n/a	3	0	n/a	0	2	32	76.19047619	high
Piazza-Waggoner (2005)	1	0	1	0	1	2	0	2	0	2	n/a	1	0	n/a	0	1	12	28.57142857	mod
Stark (2009)	2	2	3	3	3	2	3	3	2	3	n/a	3	3	n/a	0	2	37	88.0952381	high
Powers (2003)	3	3	3	1	1	2	2	3	2	3	n/a	2	1	n/a	0	2	29	69.04761905	good
Stark (2003)	3	3	3	0	1	3	3	3	3	3	n/a	2	1	n/a	0	1	30	71.42857143	good
Stark (1996)	2	2	3	0	1	3	1	1	2	3	n/a	3	2	n/a	0	1	25	59.52380952	good
Stark (1994)	2	3	3	0	1	3	2	0	2	3	n/a	2	0	n/a	0	2	24	57.14285714	good
Stark (1993)	2	2	3	0	1	3	2	1	2	3	n/a	1	1	n/a	0	2	24	57.14285714	good
Stark (1990)	2	3	3	0	1	3	2	1	2	3	n/a	2	1	n/a	0	1	25	59.52380952	good
Singer (1991)	1	0	3	0	1	2	0	1	0	2	n/a	1	0	n/a	0	0	12	28.57142857	mod
Bartholomew (1997)	1	3	2	2	3	3	1	1	3	3	n/a	2	2	n/a	0	2	31	73.80952381	good

Appendix E: NHS Ethics Approval Letter

Part of the research infrastructure for Wales funded by the National Institute for Social Care and Health Research, Welsh Government.
Yn rhan o seilwaith ymchwil Cymru a ariannir gan y Sefydliad Cenedlaethol ar gyfer Ymchwil Gofal Cymdeithasol ac Iechyd, Llywodraeth Cymru

NISCHR
Gwasanaeth Moeseg Ymchwil | RES | Research Ethics Service

WALES REC 7
PO Box 108
Building 1
Jobswell Road
St David's Park
Carmarthen SA31 3WY
(for sat nav/courier purposes SA31 3HB)

Telephone : 01267 225045
E-mail : sue.byng@wales.nhs.uk
Website : www.hra.nhs.uk

Professor Rachel Calam
School of Psychological Sciences
Zochonis Building, Brunswick Street
University of Manchester
Manchester
M13 9PL

9 April 2015

Dear Professor Calam

Study title:	**A case series examining the impact of a positive parenting program (Self Directed Teen Triple P) on treatment adherence in families with an adolescent with Cystic Fibrosis**
REC reference:	**15/WA/0096**
IRAS project ID:	**170263**

I acknowledge receipt of Emma Wells' email of 7 April 2015, responding to the Proportionate Review Sub-Committee's request for changes to the documentation for the above study.

The revised documentation has been reviewed and approved by the Vice-Chair of the PR sub-committee.

We plan to publish your research summary wording for the above study on the HRA website, together with your contact details. Publication will be no earlier than three months from the date of this favourable opinion letter. The expectation is that this information will be published for all studies that receive an ethical opinion but should you wish to provide a substitute contact point, wish to make a request to defer, or require further information, please contact the REC Manager Ms Sue Byng. Under very limited circumstances (e.g. for student research which has received an unfavourable opinion), it may be possible to grant an exemption to the publication of the study.

Confirmation of ethical opinion

On behalf of the Committee, I am pleased to confirm a favourable ethical opinion for the above research on the basis described in the application form, protocol and supporting documentation as revised.

Conditions of the favourable opinion

The favourable opinion is subject to the following conditions being met prior to the start of the study.

Management permission or approval must be obtained from each host organisation prior to the start of the study at the site concerned.

Management permission ("R&D approval") should be sought from all NHS organisations involved in the study in accordance with NHS research governance arrangements.

Guidance on applying for NHS permission for research is available in the Integrated Research Application System or at http://www.rdforum.nhs.uk.

Where a NHS organisation's role in the study is limited to identifying and referring potential participants to research sites ("participant identification centre"), guidance should be sought from the R&D office on the information it requires to give permission for this activity.

For non-NHS sites, site management permission should be obtained in accordance with the procedures of the relevant host organisation.

Sponsors are not required to notify the Committee of approvals from host organisations.

Registration of Clinical Trials

All clinical trials (defined as the first four categories on the IRAS filter page) must be registered on a publically accessible database. This should be before the first participant is recruited but no later than 6 weeks after recruitment of the first participant.

There is no requirement to separately notify the REC but you should do so at the earliest opportunity e.g. when submitting an amendment. We will audit the registration details as part of the annual progress reporting process.

To ensure transparency in research, we strongly recommend that all research is registered but for non-clinical trials this is not currently mandatory.

If a sponsor wishes to request a deferral for study registration within the required timeframe, they should contact hra.studyregistration@nhs.net. The expectation is that all clinical trials will be registered, however, in exceptional circumstances non registration may be permissible with prior agreement from NRES. Guidance on where to register is provided on the HRA website.

It is the responsibility of the sponsor to ensure that all the conditions are complied with before the start of the study or its initiation at a particular site (as applicable).

Ethical review of research sites

The favourable opinion applies to all NHS sites taking part in the study, subject to management permission being obtained from the NHS/HSC R&D office prior to the start of the study (see "Conditions of the favourable opinion" above).

Approved documents

The documents reviewed and approved by the Committee are:

Document	Version	Date
REC application form		26 February 2015
Research Protocol	3	18 March 2015
CV for CI Professor Rachel Calam		13 January 2015
CV for Student Ms Emma Wells		17 February 2015
CV for Academic Supervisor Dr Clare Murray		02 January 2015
Participant Information Sheet – Adult	2	16 February 2015
Participant Consent Form – Adult	2	16 February 2015
Evidence of insurance – University confirmation of insurance cover		16 February 2015
Letter from sponsor		16 February 2015
Referee's report		19 November 2014
Validated questionnaires – Weekly Measures Pack – amended Family Background Questionnaire, My CF Treatment, Parenting Scale, Parenting Sense of Competence Scale, Paediatric Inventory for Parents, DASS 21, Strengths and Difficulties Questionnaire, Client Satisfaction Questionnaire	3	07 March 2015
Interview Schedule	1	16 February 2015
Participant Information Sheet – Child	1	17 March 2015
Assent Form – Child	2	17 March 2015

Statement of compliance

The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

After ethical review

Reporting requirements

The attached document "After ethical review – guidance for researchers" gives detailed guidance on reporting requirements for studies with a favourable opinion, including:

- Notifying substantial amendments
- Adding new sites and investigators
- Notification of serious breaches of the protocol
- Progress and safety reports
- Notifying the end of the study

The HRA website also provides guidance on these topics, which is updated in the light of changes in reporting requirements or procedures.

Feedback

You are invited to give your view of the service that you have received from the National Research Ethics Service and the application procedure. If you wish to make your views known please use the feedback form available on the HRA website:
http://www.hra.nhs.uk/about-the-hra/governance/quality-assurance

We are pleased to welcome researchers and R & D staff at our NRES committee members' training days – see details at http://www.hra.nhs.uk/hra-training/

15/WA/0096	Please quote this number on all correspondence

With the Committee's best wishes for the success of this project.

Yours sincerely

pp

Mr Derek Lassetter
Vice-Chair

Email: sue.byng@wales.nhs.uk

Enclosures: *"After ethical review – guidance for researchers"*

Copy to: *Lynne Macrae*
Lorraine Broadfoot, Central Manchester University Hospitals NHS Trust

Participant Information Sheet

The impact of the Teen Triple P programme on medication adherence in families with a teenager with cystic fibrosis

We would like to invite you to take part in our research study. Before you decide whether you would like to be involved, we would like you to understand why the research is being done and what it would involve for you. The researcher will go through the information sheet with you and answer any questions you have.

Part 1 tells you the purpose of this study and what will happen to you if you take part.

Part 2 gives you more detailed information about the conduct of the study.

Part 1

What is the purpose of the study?

As children get older and become teenagers, all families go through a number of big changes. Children go thorough rapid physical and emotional changes and parents start to take a step back to allow their child/ teenager to become more independent. It is not surprising then that the teenage years are often challenging for families, as both parents and children learn to adjust to these changes.

Chronic illnesses like cystic fibrosis (CF) can impact on families in many ways during this time. CF requires adherence to an illness management plan including medications and physiotherapy, and sometimes older children and teenagers may struggle with the demands of this. This might be because they want to fit in with their friends or because they find their treatments unpleasant. It is important therefore, to provide appropriate support that the teenager, siblings, and parents can benefit from.

Our research team is running a project based on the Triple P - Positive Parenting Programme. It has been adapted for use with families of older children and teenagers with a chronic illness with the aim of promoting a healthy and happy family life. Research in other chronic illnesses like diabetes has found this programme to help families to manage their child's illness and to improve family relationships (for instance, by reducing arguments and disputes between parents and children). We would like to see if similar benefits can be found in families with an older child or teenager with CF. In particular, we would like to see if the Triple P programme can help teenagers to stick to their medical, dietary, and physiotherapy treatments by helping to provide parents with support and skills.

Why have I been invited?

We are inviting parents and primary care givers of children and teenagers aged 11-16 years with CF to take part in this research developing Triple P for use with CF. In particular, we are interested in inviting parents whose child may be finding it hard to stick to all their treatments to take part in this study. This is relatively common for older children and teenagers with CF, and we would like to see if the Teen Triple P programme can help to provide support to parents and teenagers to help them to take their treatments better and improve parent and child health and wellbeing.

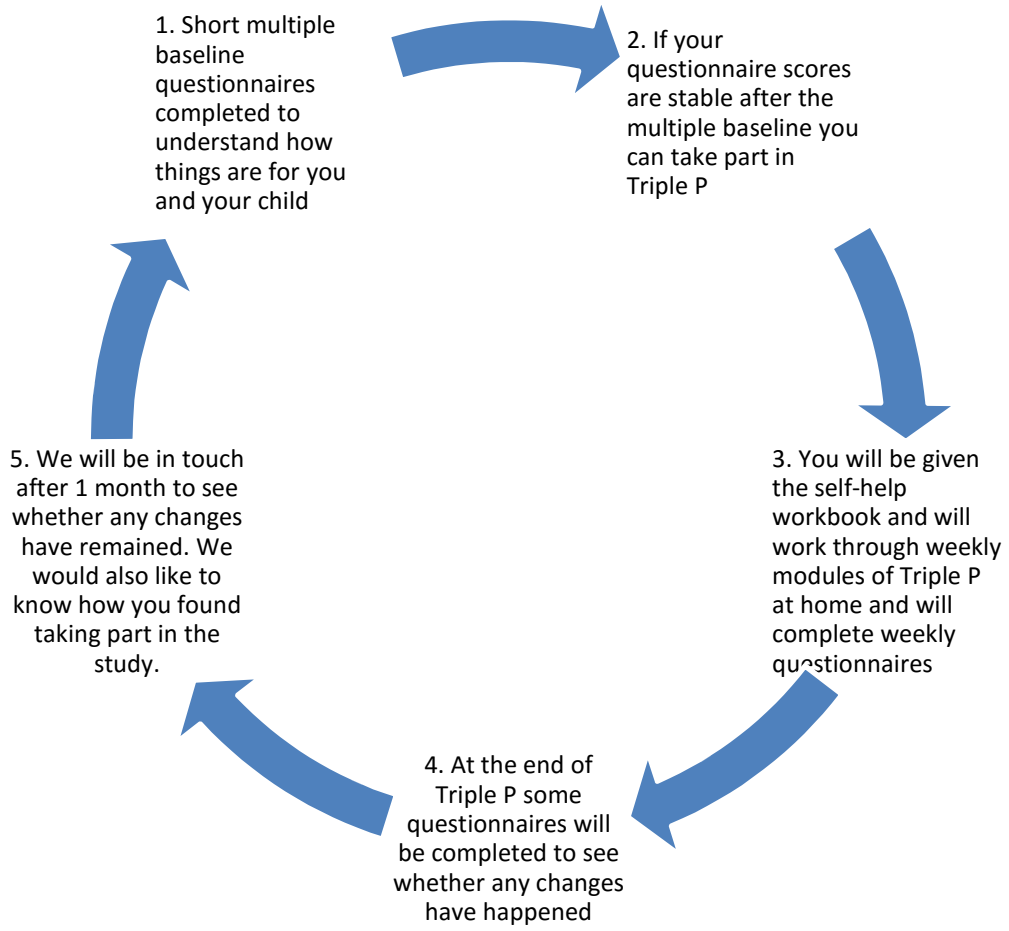
Because this is the first research study to look at using Teen Triple P in CF, we are expecting that around 10-15 families will take part.

Do I have to take part?

It is up to you whether you decide to join the study. If you are interested in taking part we will describe the study in person and go through this information sheet. You are invited to ask questions at any time. If you agree to take part, we will then ask you to sign a consent form. You are free to withdraw at any time, without giving a reason and this will not affect any current or future treatments you or your child receive.

What will happen to me if I take part?

If you decide to take part then the study will involve the following stages:



You will be able to complete all parts of the Triple P programme at home and will not need to attend any extra appointments at the hospital as part of the research. The study will last between 16 and 20 weeks.

1. The 'baseline' period: 2-6 weeks (30-40 minutes each week)

Before we provide you with the Triple P programme materials, we would like to know more about what things are like for your family. In order to do this, we will ask for some basic information about your family (i.e. who lives in your house, names, ages, relationships to each other) and about your child/ teenager's CF. With your permission, we may also speak to your child's care team to get some basic information about their CF (e.g. weight, lung function, age when diagnosed, INeb/ nebulised medication measurements).

Then we would like you to complete a set of questionnaires each week for between 2 and 6 weeks. They will ask you about how well your child/ teenager sticks to their treatment, how well things are going for you and your family, yours and your child/ teenagers wellbeing, and how confident you feel in your parenting skills and abilities. You will be able to choose whether you would like to be sent paper copies of the questionnaires, or whether you would prefer to complete them on-line or over the phone with the researcher.

If after the baseline period the scores on your questionnaires are stable, you will be able to start the Triple P programme. If they are not, we may ask you to complete the same questionnaires on a few more occasions so that your scores are stable before starting the programme.

2. Teen Triple P: 10 weeks (one hour per week)

Following on from the baseline period, you will be posted the Teen Triple P manual. At this point, we would like you to work through weekly modules outlined in the manual and complete a number of tasks and exercises along the way. These modules will help you to build upon skills that you as parents already have and will also help to provide new skills to help manage challenging teenage behaviours, build positive family relationships and help you to support your child to become independent in their own care. Each module should take you one hour each week to complete.

You will also be given the chronic illness tip sheet which addresses common themes that can arise with a chronic illness. This includes prevention and coping advice for: reducing family stress; helping siblings cope; and reducing anxiety.

As well as completing weekly one hour modules of Teen Triple P, you will be asked to complete a set of weekly questionnaires (10 minutes each week, except for weeks 5 and 10 where questionnaires will take around 30 minutes to complete). These questionnaires will ask you about your child/ teenagers treatment adherence and how confident and skilled you feel as a parent. These questionnaires can be completed on paper forms that we can post to you, or online or over the phone with the researcher.

3. End of study questionnaires

Once the Triple P programme has finished we will wait for four weeks before contacting you again. After four weeks the researcher will contact you one more time to complete some more questionnaires. This will help us to see if the Triple P programme has helped your family and if any benefits are carrying on after the programme has finished. This will take around 30 to 40 minutes to complete.

Because we are interested in how families feel about taking part in the study, the researcher will also ask a few questions about how you found the Triple P intervention. The researcher will ask these questions over the phone at a time that is convenient for you and will note down your comments. These comments may be used anonymously in reports once the research has finished.

What are the possible disadvantages and risks of taking part?

We appreciate that you will be putting some valuable time aside each week to take part in the study; however, we hope that you find the Triple P intervention useful. If at any point you feel that you can no longer commit to the time required to take part in the study, you will be free to leave the study at any time.

We do not expect you to experience any risks as a result of taking part in the study. However, if you experience any distress linked to your child's condition we can signpost you to relevant agencies for support.

What are the possible benefits of taking part?

We are hoping that the Triple P programme will help children and teenagers to stick to their treatment better, improve family relationships, improve parents confidence and skills, as well as improving parent and child wellbeing. However, whilst research in other chronic health conditions has found Teen Triple P to be helpful, no research has been carried out for us to know for certain that this intervention will help families of teenagers with CF.

By taking part in this study you will be helping researchers to understand what types of support may or may not be helpful in improving the quality of life in families of a teenager with cystic fibrosis.

What happens when the research study stops?

Once the study has finished you will be able to keep the Teen Triple P manual and chronic illness tip sheet for you to continue to use in the future if you wish. The research team will not need to contact you again once the study has stopped and you will not be asked to complete any more questionnaires. The researcher will answer any questions you may have about the study or Triple P once it has finished.

If you would like to be notified of the results of the study once they have been compiled, you can leave your contact details with the researcher.

What if there is a problem?

Any complaint about the way you have been dealt with during the study or any possible harm you might suffer will be addressed. The detailed information on this

is given in Part 2.

Will my taking part in the study be kept confidential?

Yes. We will follow ethical and legal practice and all information about you will be handled in confidence. The details are included in Part 2.

If the information in Part 1 has interested you and you are considering participation, please read the additional information in Part 2 before making any decision.

Part 2

What if relevant new information becomes available?

If at any point in the research study relevant new information becomes available, you will be contacted by the researcher who will pass this information on to you.

What will happen if I don't want to carry on with the study?

You are free to withdraw from the study at any time, without having to give a reason. This will not affect the medical care or treatment received by you or your child. We will include any results that we get from you up to the point that you leave the study in the final report as this will still be useful to us. This may include anonymised quotes from any interview questions that you answer. However, if you would not like this to happen we will destroy all your data if you ask us to. We would also like to invite you to answer some questions about why you chose to withdraw from the study. This will help us to understand what types of support and interventions are most suitable and acceptable for families and to see what families think about Teen Triple P. However, you do not have to answer these questions if you would prefer not to.

What if there is a problem?

You should contact Emma Wells (contact details at the end of this information sheet) if you have any queries or concerns in relation to the research project.

The following services may also provide further assistance if required:

Medical issues: - You can contact your GP

- Your child's Cystic Fibrosis Care Team, or

- NHS Direct. Tel: 0845 4647 (24 hour health advice)

Support network: - Cystic Fibrosis Trust Help Line. Tel: 0300 373 1000 (Monday–Friday, 9am–5pm)

Further psychological help/parenting support: If you feel you need further help, contact your GP or Cystic Fibrosis Care Team.

If the researchers are unable to answer your concerns, or you wish to make a

formal complaint about the conduct of the research you should contact: Head of the Research Office, Christie Building, University of Manchester, Oxford Road, Manchester, M13 9PL.

Complaints

If you have a concern about any aspect of this study, you should ask to speak to the researchers who will do their best to answer your questions. If they are unable to resolve your concern or you wish to make a complaint regarding the study, please contact a University Research Practice and Governance Co-ordinator on 0161 2757583 or 0161 2758093 or by email to research.complaints@manchester.ac.uk

Will my taking part in this study be kept confidential?

Yes. During the study, your data will be recorded by the experimenter and via the computer. It will only ever be associated with a participant number and never your name. The data will be stored on University of Manchester secure computers and will be accessible only to the research team. The data will be analysed and the results presented in research papers, and no individual will ever be identified in these. The data will be retained for seven years after the publication of the data, after which time it will be securely disposed of. For monitoring and auditing purposes, study data and material may be looked at by individuals from the University of Manchester, from regulatory authorities or from the NHS Trust, and this may include access to personal information.

What will happen to the results of the research study?

The results will be published in academic journals, and no individual will ever be identified in these articles. There will also be a short summary of the results that can be sent to you after you have completed the research, and you can tell us if you want to receive this at the end of the study.

Involvement of the General Practitioner/Family doctor

If you agree, we would like to inform your GP that you are taking part in this research project.

Who is organising and funding the research?

This research is being organised by the University of Manchester. This research is being conducted as part of a thesis as part of a doctorate programme in clinical psychology being undertaken by the researcher.

Who has reviewed the study?

All research in the NHS is looked at by independent group of people, called a Research Ethics Committee, to protect your interests. This study has been reviewed and given favourable opinion by Wales 7 Ethics Committee.

You will be given a copy of this information sheet and a signed consent form to

keep.

Further information and contact details

If you have any queries during the course of the study please contact the researcher

Emma Wells: Emma.Wells-2@postgrad.manchester.ac.uk

MANCHESTER 1824
The University of Manchester

PARTICIPANT CONSENT FORM

Title of Project: A case series examining the impact of a Positive Parenting Program (Self Directed Teen Triple P) on treatment adherence in families with an adolescent with Cystic Fibrosis

Name of Chief Investigator: Professor Rachel Calam

Name of student Researcher: Emma Wells

Please initial box

1. I confirm that I have read and understand the information sheet (Version 2.0; dated 16/02/2015) for the above study and have had the opportunity think about it and to ask questions. *The information sheet is for you to keep and refer to at any time. Please read it carefully.* ☐

2. I understand that my participation is voluntary and that I am free to withdraw at any time without giving any reason, without mine or my child's medical care or legal rights being affected. ☐

3. I understand that the information I give will only be seen by members of the research team and their supervisors, and that this information will be stored anonymously and securely on university computers. ☐

4. I understand that for monitoring and auditing purposes, study data and materials may be looked at by individuals from the University of Manchester, from regulatory authorities or from the NHS Trust. ☐

5. I agree for quotes (i.e. from interviews with the research team) to be used in any possible publications of the research findings and that the researcher will make attempts to anonymise quotes as much as possible, but that full anonymisation is not guaranteed. ☐

6. I agree for the researcher to send me weekly text or telephone reminders to complete weekly study tasks. ☐

7. I understand that the researcher may speak to my child's care team in order to gain some information that may be used for research purposes (i.e. height, weight, lung function, use of nebulized medication), and that this information will be stored anonymously by the research team on secure university computers. ☐

8. I agree to my General Practitioner being informed of my participation in the study. ☐

9. I would like to receive a written summary of the overall findings of the study ☐

10. I agree to take part in the above study ☐

Name of Participant	Name of child	Date	Signature

Name of Person taking consent	Date	Signature

Participant Information Sheet

The impact of the Teen Triple P programme on medication adherence in families with a teenager with cystic fibrosis

Your parent/ carer has agreed to take part in some research that we are doing.

This research is looking at ways that we can help your parent/ carer to support you with your cystic fibrosis (CF) and it's treatments.

We would like to collect some information about your CF from your parents.

We would also like to let your doctor know that you and your parents are taking part in our research.

Before you decide whether you are happy for us to collect this information, we would like to give you some information about the research. The researcher can go through this information with you and answer any questions you may have.

What is the research about?

Illnesses like cystic fibrosis (CF) can affect families in many ways, especially during older child and teenage years (also known as adolescence)

During adolescence there are lots of bodily changes and emotional changes. Parents also have to start to let their children take more charge of their CF treatments. This can be difficult for children, teenagers, and their parents.

CF involves many treatments including medication and physiotherapy, which can be difficult and challenging to stick to.

Because CF treatments are very important in keeping young people healthy and well, we are trying to find new ways that we can help you and your parents to manage them better.

Our team is running a research project based on something called the 'Triple P - Positive Parenting Programme'.

Triple P aims to help parents/carers to support children and teenagers to live a happy and healthy life.

Research in other illnesses has found that Triple P can help parents to support their children/teenagers with their illnesses and its treatments. It has also helped to reduce arguing and falling out between parents and their children.

We would like to see if the 'Teen Triple P –Positive Parenting Programme' could also be helpful for you and your family. We are keen to see if it can help older children and teenagers with CF.

Why have my parents and I been asked to take part?

We are inviting parents and carers of children and teenagers aged 11-16 years with CF to take part in this research. Your CF team are passing out leaflets about the research to parents of all children in this age group.

It is common for children and young people to find it hard to stick to their treatments at this age. Your parent/ carer has agreed that they would like to take part in the research to help them to support you to improve your CF care.

Do I have to take part?

Your parent/ carer has already agreed that they would like to take part in the study.

We would like to know whether you are happy for us to get some information about your CF from your parents and care team.

We would also like to know whether you are happy for us to let your family doctor know that you and your parents are taking part in our research study.

You can ask us any questions about this before you decide. If you agree for us to collect this information, we will ask you to fill in a form to show that you are OK with this. You can change your mind about this at any time even after the study has started. You do not have to give us a reason why you have changed your mind.

What will happen to me if I take part?

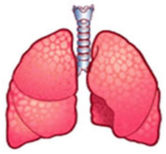
You do not need to do anything; your parent/carer is the one who will do all the hard work. They will be asked to take part in a number of tasks, questionnaires, and activities as part of the research.

Before they begin the research, we would like to ask them more about what things are like for you and your family. We will ask them some information about your family such as:

- who lives at your house
- names
- ages
- relationships etc.

With your permission we may also speak to your care team about your CF. For instance, we may ask about your:

- lung function
- weight
- age when you were diagnosed
- medications you take.



Your parent/carer will then be posted a Teen Triple P booklet. They will follow 10 weekly 1 hour learning sessions from the booklet.

This booklet gives parents information and tasks that can help them to support you with your CF treatments, and support you and your parents to get along well and manage difficult situations.

Each week your parent/carer will be asked to complete some questionnaires asking about how they think the Triple P booklet is helping them and how well it is or isn't helping them to support you with your CF.

What are the possible disadvantages and risks of taking part?

We do not expect there are any risks in taking part in the study.

What are the possible benefits of taking part?

We are hoping that the Triple P programme will help older children and teenagers to stick to their treatment better, improve family relationships, improve parents confidence and skills, as well as improving parent and child wellbeing.

No research has been carried out for us to know for certain that Triple P will help families of teenagers with CF. This is why we have asked you and your parent/ carer to take part.

By taking part you and your parent/ carer will be helping us to understand what types of support could be helpful in improving life for children and teenagers with CF, and their parents/carers.

What happens when the research study stops?

Once the study has finished your parent/ carer will be able to keep the Teen Triple P booklet if they wish. They will be able to carry on using it if they find it helpful. You may also want to have a look at it.

We will not need to contact you again once the study has stopped.

Further information and contact details

If you have any queries during the course of the study please contact the researcher Emma Wells: Emma.Wells-2@postgrad.manchester.ac.uk

Appendix I: Child Assent Form

Participant Identification Number for this trial: Study Number: **170263**

A case series examining the impact of a Positive Parenting Program (Self Directed Teen Triple P) on treatment adherence in families with an adolescent with Cystic Fibrosis

ASSENT FORM

To be completed by the young person. Please put you initials in the box if you agree with the statement.

Please initial box

1. I have read (or had read to me) the information about this study. A researcher has explained it to me. I understand what the study is about and I have been able to ask questions. ☐
2. I understand that it's OK to stop taking part at any time. ☐
3. I am happy for the researcher to get information about me and my CF care from my parents and my CF team. ☐
4. I agree to my family doctor (GP) being told about my parents and I taking part in this study. ☐
5. I would like hear about the results of the study when it is finished. ☐

Name	Date	Signature
Name of Person taking consent	Date	Signature

MANCHESTER 1824
The University of Manchester

CF PROFESSIONALS INTERVIEW CONSENT FORM

Title of Project: A case series examining the impact of a Positive Parenting Program (Self Directed Teen Triple P) on treatment adherence in families with an adolescent with Cystic Fibrosis

Name of Chief Investigator: Professor Rachel Calam

Name of student Researcher: Emma Wells

Please initial box

1. I confirm that I have read and understand the information sheet (**Version 2.0; dated 31/03/2016**) for the above study and have had the opportunity think about it and to ask questions. *The information sheet is for you to keep and refer to at any time. Please read it carefully.* ☐

2. I agree for the researcher to ask me some questions about my professional experiences of working with parents who might be having difficulties supporting their child with cystic fibrosis treatments ☐

3. I understand that my participation is voluntary and that I am free to withdraw at any time without giving any reason. ☐

4. I understand that the interview information that I give will only be seen by members of the research team, and that this information will be anonymised as much as possible and stored securely on university computers. ☐

5. I understand that my interview will be recorded and transcribed for use in research papers that may be published. I agree for quotes to be used in any possible publications, but that full anonymisation is not guaranteed. ☐

6. I understand that for monitoring and auditing purposes, study data and materials may be looked at by individuals from the University of Manchester, from regulatory authorities or from the NHS Trust. ☐

7. I would like to receive a written summary of the overall findings of the study ☐

Name of Participant	Role	Date	Signature

Name of Person taking consent	Date	Signature

Appendix K: Family Background Questionnaire

INTERNATIONAL FAMILY BACKGROUND QUESTIONNAIRE

Participant ID number: _____ *Date:* _____

This questionnaire collects information about your family. Please read and answer every question.

1. **Date**: _____/_____/_____

   (Day) (Month) (Year)

2. **Your age today:** _____ (years)

3. **Child's gender:** Male ☐ Female ☐

4. **Child's age today**: _____ (years)

5. **Does your child experience any of the following problems:**

A chronic illness e.g., asthma, eczema?	Yes ☐	No ☐
A physical disability?	Yes ☐	No ☐
An intellectual disability?	Yes ☐	No ☐
A developmental delay?	Yes ☐	No ☐

If Yes to any of the above, please provide details :

_____

6. **Your relationship to this child:**

Mother (biological or adoptive)	☐	Father (biological or adoptive)	☐
Step-mother	☐	Step-father	☐
Foster mother	☐	Foster father	☐

Other (please describe)_____

Your current marital status:

Married ☐ Divorced/separated ☐ Single ☐

Cohabiting ☐ Widow/er ☐

Other (please describe)_____

8. Which best describes the household in which your child is presently living?

Original family (both biological or adoptive parents present) ☐

Step family (two parents, one being a step parent) ☐

Sole parent family ☐

Other (please describe) _____ ☐

9. At present who lives at home with your child (e.g. parents, siblings, grandparents), including yourself?

Relationship to child	Age
1.	
2.	
3.	
4.	
5.	

10. How would you describe your child's ethnic background? Please choose one

☐ White ☐ Mixed ☐ Indian ☐ Pakistani ☐ Bangladeshi ☐ Other Asian

☐ Black Caribbean ☐ Black African ☐ Other Black ☐ Chinese ☐ Other

11. **Your highest level of education:**

☐ primary school or less ☐ some high school ☐ completed high school

☐ trade/technical college qualification ☐ university degree ☐ post-graduate degree

12. **Your partner's highest level of education** (if applicable):

☐ primary school or less ☐ some high school ☐ completed high school

☐ trade/technical college qualification ☐ university degree ☐ post-graduate degree

13. **Are you working outside the home right now?**

☐ yes, full time ☐ yes, part time☐ not working, but looking for a job

☐ home based paid work (child care, sewing, internet or phone-based work, etc)

☐ not working (includes stay at home parents, retired)

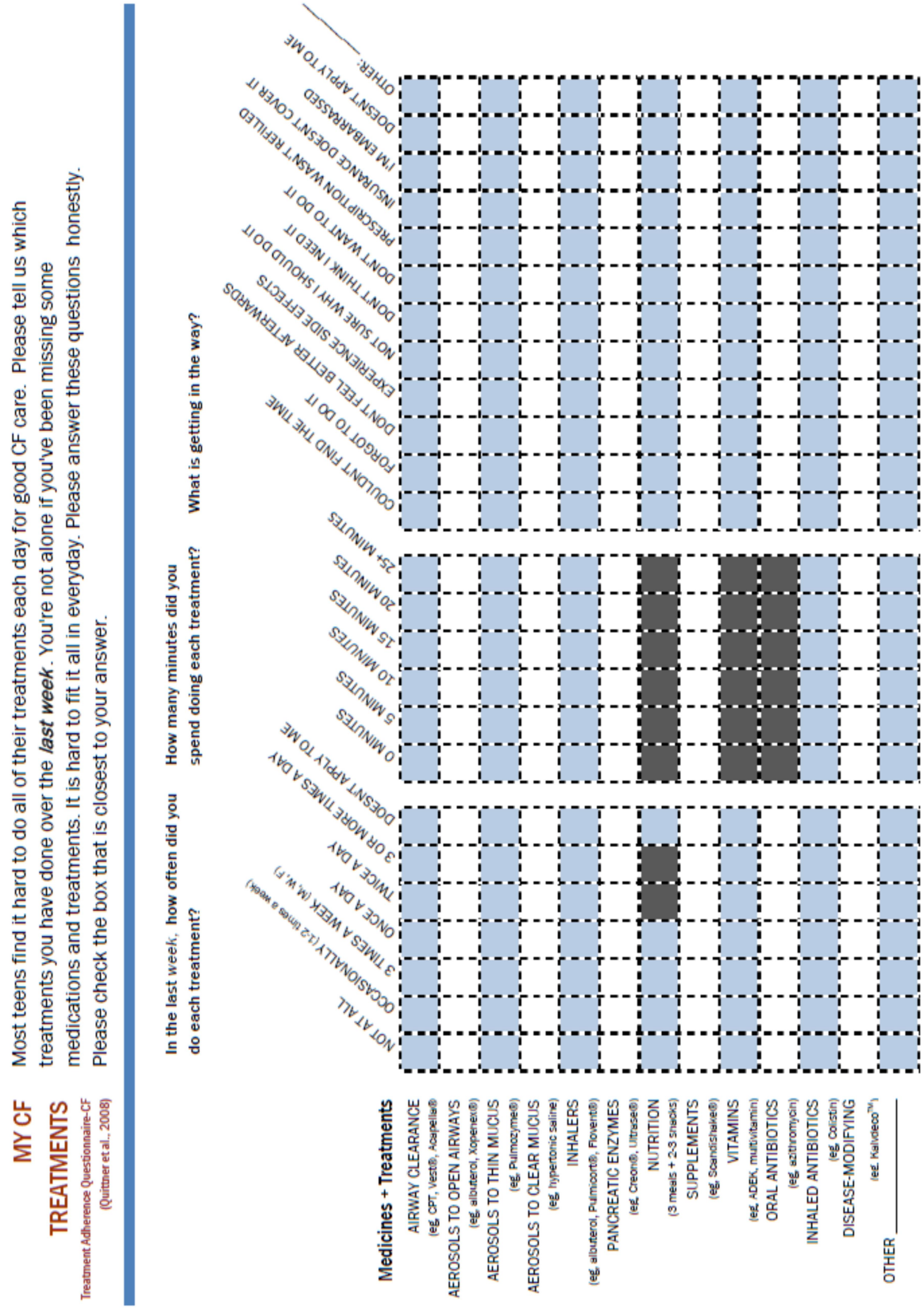
. **Is your partner working outside the home right now**? (if applicable)

☐ yes, full time ☐ yes, part time☐ not working, but looking for a job

☐ home based paid work (child care, sewing, internet or phone-based work, etc)

☐ not working (includes stay at home parents, retired)

Appendix L: Treatment Adherence Questionnaire – CF



PARENTING SCALE — ADOLESCENTS

At one time or another, all children misbehave or do things that could be harmful, that are "wrong", or that parents don't like. Examples include: hitting someone, whingeing or complaining, damaging things, forgetting homework, leaving things lying around, lying, being over-emotional, refusing to follow requests, breaking family rules, swearing, taking other people's things, staying out late.

Parents have many different ways or styles of dealing with these types of problems. Below are items that describe some styles of parenting. For each item, circle the number that best describes your style of parenting during the past 2 months with your teenager.

Sample Item

At meal time...

I let my teenager decide what to eat.	1	(2)	3	4	5	6	7	I decide what my teenager eats.

1. When I give fair threat or warning…

I often don't carry it out.	1	2	3	4	5	6	7	I always do what I said

2. If my teenager gets upset when I say 'no'…

I back down and give in to my teenager.	1	2	3	4	5	6	7	I stick to what I said

3. When my teenager doesn't do what I asked…

I often let it go or end up doing it myself.	1	2	3	4	5	6	7	I take some other action

4. When I say my teenager can't do something…

I let my teenager do it anyway.	1	2	3	4	5	6	7	I stick to what I said.

5. If saying 'no' doesn't work…

I take some other kind of action.	1	2	3	4	5	6	7	I offer my teenager something nice so he or she will behave

6. When my teenager does something I don't like…

I do something about it every time it happens.	1	2	3	4	5	6	7	I often let it go

7. When my teenager misbehaves…

I raise my voice or yell.	1	2	3	4	5	6	7	I speak to my teenager calmly.

8. When my teenager misbehaves…

I handle it without getting upset	1	2	3	4	5	6	7	I get so frustrated or angry my teenager can see I'm upset.

9. When there is a problem with my teenager…

Things build up and I do things I don't mean to.	1	2	3	4	5	6	7	Things don't get out of hand.

10. When my teenager does something I don't like, I insult my teenager, say mean things, or call my teenager names…

Never or rarely	1	2	3	4	5	6	7	Most of the time

11. When my teenager misbehaves…

I usually get into a long argument with my teenager.	1	2	3	4	5	6	7	I don't get into an argument.

12. When I am upset or under stress…

I am picky and on my teenager's back.	1	2	3	4	5	6	7	I am no more picky than usual.

13. When my teenager is out of sight…

I often don't know what my teenager is doing.	1	2	3	4	5	6	7	I always have a good idea of what my teenager is doing.

Appendix N: Parent Sense of Competency Scale

Parenting Sense of Competence Scale

(Gibaud-Wallston & Wandersman, 1978)

Please rate the extent to which you agree or disagree with each of the following statements.

Strongly Disagree	Somewhat Disagree	Disagree	Agree	Somewhat Agree	Strongly Agree
1	2	3	4	5	6

1. The problems of taking care of a child are easy to solve once you know how your actions affect your child, an understanding I have acquired. 1 2 3 4 5 6

2. Even though being a parent could be rewarding, I am frustrated now while my child is at his / her present age. 1 2 3 4 5 6

3. I go to bed the same way I wake up in the morning, feeling I have not accomplished a whole lot. 1 2 3 4 5 6

4. I do not know why it is, but sometimes when I'm supposed to be in control, I feel more like the one being manipulated. 1 2 3 4 5 6

5. My mother was better prepared to be a good mother than I am. 1 2 3 4 5 6

6. I would make a fine model for a new mother to follow in order to learn what she would need to know in order to be a good parent. 1 2 3 4 5 6

7. Being a parent is manageable, and any problems are easily solved. 1 2 3 4 5 6

8. A difficult problem in being a parent is not knowing whether you're doing a good job or a bad one. 1 2 3 4 5 6

9. Sometimes I feel like I'm not getting anything done. 1 2 3 4 5

10. I meet by own personal expectations for expertise in caring for my child. 1 2 3 4 5 6

11. If anyone can find the answer to what is troubling my child, I am the one. 1 2 3 4 5 6

12. My talents and interests are in other areas, not being a parent. 1 2 3 4 5 6

13. Considering how long I've been a mother, I feel thoroughly familiar with this role. 1 2 3 4 5 6

14. If being a mother of a child were only more interesting, I would be motivated to do a better job as a parent. 1 2 3 4 5 6

15. I honestly believe I have all the skills necessary to be a good mother to my child. 1 2 3 4 5 6

16. Being a parent makes me tense and anxious. 1 2 3 4 5 6

17. Being a good mother is a reward in itself. 1 2 3 4 5 6

Appendix 0: Strengths and Difficulties Questionnaire

Strengths and Difficulties Questionnaire

For each item, please mark the box for Not True, Somewhat True or Certainly True. It would help us if you answered all items as best you can even if you are not absolutely certain or the item seems daft! Please give your answers on the basis of the child's behaviour over the last six months or this school year.

Child's Name ..... Male/Female

Date of Birth.....

	Not True	Somewhat True	Certainly True
Considerate of other people's feelings	☐	☐	☐
Restless, overactive, cannot stay still for long	☐	☐	☐
Often complains of headaches, stomach-aches or sickness	☐	☐	☐
Shares readily with other children (treats, toys, pencils etc.)	☐	☐	☐
Often has temper tantrums or hot tempers	☐	☐	☐
Rather solitary, tends to play alone	☐	☐	☐
Generally obedient, usually does what adults request	☐	☐	☐
Many worries, often seems worried	☐	☐	☐
Helpful if someone is hurt, upset or feeling ill	☐	☐	☐
Constantly fidgeting or squirming	☐	☐	☐
Has at least one good friend	☐	☐	☐
Often fights with other children or bullies them	☐	☐	☐
Often unhappy, down-hearted or tearful	☐	☐	☐
Generally liked by other children	☐	☐	☐
Easily distracted, concentration wanders	☐	☐	☐
Nervous or clingy in new situations, easily loses confidence	☐	☐	☐
Kind to younger children	☐	☐	☐
Often lies or cheats	☐	☐	☐
Picked on or bullied by other children	☐	☐	☐
Often volunteers to help others (parents, teachers, other children)	☐	☐	☐
Thinks things out before acting	☐	☐	☐
Steals from home, school or elsewhere	☐	☐	☐
Gets on better with adults than with other children	☐	☐	☐
Many fears, easily scared	☐	☐	☐
Sees tasks through to the end, good attention span	☐	☐	☐

Signature ..... Date .....

Appendix P: Depression Anxiety Stress Scale (21-item version)

DASS 21

NAME _____ DATE _____

BLACK DOG INSTITUTE

Please read each statement and circle a number 0, 1, 2 or 3 which indicates how much the statement applied to you over the past week. There are no right or wrong answers. Do not spend too much time on any statement.

The rating scale is as follows:

0 Did not apply to me at all - NEVER

1 Applied to me to some degree, or some of the time - SOMETIMES

2 Applied to me to a considerable degree, or a good part of time - OFTEN

3 Applied to me very much, or most of the time - ALMOST ALWAYS

		N	S	O	AA	FOR OFFICE USE D	A	S
1	I found it hard to wind down	0	1	2	3			
2	I was aware of dryness of my mouth	0	1	2	3			
3	I couldn't seem to experience any positive feeling at all	0	1	2	3			
4	I experienced breathing difficulty (eg, excessively rapid breathing, breathlessness in the absence of physical exertion)	0	1	2	3			
5	I found it difficult to work up the initiative to do things	0	1	2	3			
6	I tended to over-react to situations	0	1	2	3			
7	I experienced trembling (eg, in the hands)	0	1	2	3			
8	I felt that I was using a lot of nervous energy	0	1	2	3			
9	I was worried about situations in which I might panic and make a fool of myself	0	1	2	3			
10	I felt that I had nothing to look forward to	0	1	2	3			
11	I found myself getting agitated	0	1	2	3			
12	I found it difficult to relax	0	1	2	3			
13	I felt down-hearted and blue	0	1	2	3			
14	I was intolerant of anything that kept me from getting on with what I was doing	0	1	2	3			
15	I felt I was close to panic	0	1	2	3			
16	I was unable to become enthusiastic about anything	0	1	2	3			
17	I felt I wasn't worth much as a person	0	1	2	3			
18	I felt that I was rather touchy	0	1	2	3			
19	I was aware of the action of my heart in the absence of physicalexertion (eg, sense of heart rate increase, heart missing a beat)	0	1	2	3			
20	I felt scared without any good reason	0	1	2	3			
21	I felt that life was meaningless	0	1	2	3			
	TOTALS							

PEDIATRIC INVENTORY FOR PARENTS

Below is a list of difficult events which parents of children who have (or have had) a serious illness sometimes face. Please read each event carefully, and circle HOW OFTEN the event has occurred for you in the past 7 days, using the 5 point scale below. Afterwards, please rate how DIFFICULT it was/or generally is for you, also using the 5 point scale. Please complete both columns for each item.

EVENT	HOW OFTEN? 1=Never, 2=Rarely, 3=Sometimes, 4=Often, 5=Very often					HOW DIFFICULT? 1=Not at all, 2=A little, 3=Somewhat, 4=Very much, 5=Extremely				
1. Difficulty sleeping	1	2	3	4	5	1	2	3	4	5
2. Arguing with family member(s)	1	2	3	4	5	1	2	3	4	5
3. Bringing my child to the clinic or hospital	1	2	3	4	5	1	2	3	4	5
4. Learning upsetting news	1	2	3	4	5	1	2	3	4	5
5. Being unable to go to work/job	1	2	3	4	5	1	2	3	4	5
6. Seeing my child's mood change quickly	1	2	3	4	5	1	2	3	4	5
7. Speaking with doctor	1	2	3	4	5	1	2	3	4	5
8. Watching my child have trouble eating	1	2	3	4	5	1	2	3	4	5
9. Waiting for my child's test results	1	2	3	4	5	1	2	3	4	5
10. Having money/financial troubles	1	2	3	4	5	1	2	3	4	5
11. Trying not to think about my family's difficulties	1	2	3	4	5	1	2	3	4	5
12. Feeling confused about medical information	1	2	3	4	5	1	2	3	4	5
13. Being with my child during medical procedures	1	2	3	4	5	1	2	3	4	5
14. Knowing my child is hurting or in pain	1	2	3	4	5	1	2	3	4	5
15. Trying to attend to the needs of other family members	1	2	3	4	5	1	2	3	4	5
16. Seeing my child sad or scared	1	2	3	4	5	1	2	3	4	5

EVENT	HOW OFTEN? 1=Never, 2=Rarely, 3=Sometimes, 4=Often, 5=Very often					HOW DIFFICULT? 1=Not at all, 2=A little, 3=Somewhat, 4=Very much, 5=Extremely				
18. Making decisions about medical care or medicines	1	2	3	4	5	1	2	3	4	5
19. Thinking about my child being isolated from others	1	2	3	4	5	1	2	3	4	5
20. Being far away from family and/or friends	1	2	3	4	5	1	2	3	4	5
21. Feeling numb inside	1	2	3	4	5	1	2	3	4	5
22. Disagreeing with a member of the health care team	1	2	3	4	5	1	2	3	4	5
23. Helping my child with his/her hygiene needs	1	2	3	4	5	1	2	3	4	5
24. Worrying about the long term impact of the illness	1	2	3	4	5	1	2	3	4	5
25. Having little time to take care of my own needs	1	2	3	4	5	1	2	3	4	5
26. Feeling helpless over my child's condition	1	2	3	4	5	1	2	3	4	5
27. Feeling misunderstood by family/friends as to the severity of my child's illness	1	2	3	4	5	1	2	3	4	5
28. Handling changes in my child's daily medical routines	1	2	3	4	5	1	2	3	4	5
29. Feeling uncertain about the future	1	2	3	4	5	1	2	3	4	5
30. Being in the hospital over weekends/holidays	1	2	3	4	5	1	2	3	4	5
31. Thinking about other children who have been seriously ill	1	2	3	4	5	1	2	3	4	5
32. Speaking with my child about his/her illness	1	2	3	4	5	1	2	3	4	5
33. Helping my child with medical	1	2	3	4	5	1	2	3	4	5

Appendix R: Client Satisfaction Questionnaire

CLIENT SATISFACTION QUESTIONNAIRE

Your child's age: _____ Relationship to Child:(Circle one) Parent Caregiver Grandparent Other _____

Your Gender: M F Your Age: 15-24 25-34 35-44 45-54 55-64 65+

City / Town: _____ Postal Code: _____

This questionnaire will help us to evaluate and continually improve the services we offer. We are interested in your **honest opinions** *about the services you have received, whether they are positive or negative. Please answer all the questions.*

Please circle the response that best describes how you honestly feel.

1. How would you rate the quality of the service you and your child received?

7	6	5	4	3	2	1
Excellent		Good		Fair		Poor

2. Did you receive the type of help you wanted from the program?

1	2	3	4	5	6	7
No definitely not		No not really		Yes generally		Yes definitely

3. To what extent has the program met you *child's needs*?

7	6	5	4	3	2	1
Almost all needs have been met		Most needs have been met		Only a few needs have been met		No needs have been met

4. To what extent has the program met *your* needs?

7	6	5	4	3	2	1
Almost all needs have been met		Most needs have been met		Only a few needs have been met		No needs have been met

5. How satisfied were you with the ***amount of help*** you and your child received?

1	2	3	4	5	6	7
Quite Dissatisfied		Dissatisfied		Satisfied		Very Satisfied

6. Has the program helped you deal more effectively with your child's behaviour?

7	6	5	4	3	2	1
Yes, it has helped a great deal		Yes, it has helped somewhat		No, it hasn't helped much		No, it made things worse

7. Has the program helped you to deal more effectively with problems that arise in your family?

7	6	5	4	3	2	1
Yes, it has helped a great deal		Yes, it has helped somewhat		No, it hasn't helped much		No, it made things worse

8. Do you think your relationship with your partner has been improved by the program?

1	2	3	4	5	6	7
No definitely not		No not really		Yes generally		Yes definitely

9. In an overall sense, how satisfied are you with the program you and your child received?

7	6	5	4	3	2	1
Very Satisfied		Satisfied		Dissatisfied		Very Dissatisfied

10. If you were to seek help again, would you come back to Triple P?

1	2	3	4	5	6	7
No definitely not		No, I Don't think so		Yes, I think so		Yes, Definitely

11. Has the program helped you to develop skills that can be applied to other family members?

1	2	3	4	5	6	7
No definitely not		No, I Don't think so		Yes, I think so		Yes, Definitely

12. In your opinion, how is your child's behaviour at this point?

1	2	3	4	5	6	7
Considerably Worse	Worse	Slightly Worse	The same	Slightly Improved	Improved	Greatly Improved

13. How would you describe your feelings at this point about your child's progress?

7	6	5	4	3	2	1
Very Satisfied	Satisfied	Slightly satisfied	Neutral	Slightly dissatisfied	Dissatisfied	Very dissatisfied

14. Since beginning this program, have you sought further assistance for your child's behaviour or for you family from any other source. If so, please describe.

_____

_____

_____

_____

15. Have you had any other problems with your child which you feel may be related to the original difficulty?

_____

_____

_____

_____

16. Do you have any other comments about this program?

_____

_____

_____

_____

Appendix S: Treatment Integrity Measure

Module completion:

It is helpful for us to know how many of the modules in the workbook you have been able to complete and how you found them. Equally if you have not been able to complete certain modules, it is helpful for us to know why this has been the case.

	I completed the module this week (Yes/No)	I understood the module (Yes/No/ Not applicable)	I felt this module was useful (Yes/No/ Not applicable)	I felt this module was relevant (Yes/No/ Not applicable)	I had time to complete the module this week (Yes/No)	I did not read the modules for another reason (please state)	I intend to complete module at a later date (Yes/No/ Not applicable)
Module 1							
Module 2							
Module 3							
Module 4							
Module 5							
Module 6							
Module 7							
Module 8							
Module 9							
Module 10							

Parent Interview

A. Explanation of the aims of the interview and the topics to be covered.
B. Explanation of ground rules during interview (e.g. anonymity, value of opinions regardless of how unusual, no right or wrong answers, taking notes, format of the interview).
C. Ask if they have any question before beginning the interview

The following questions are indicative of the areas to be covered in the interview. (* indicates questions to be completed by parents who do not consent to participate in the intervention but who are happy to have a discussion about reasons why not taking part)

Reasons for showing interest in the research study

1. What initially interested you in putting forward your contact details for this study?*

2. What are/ were you hoping for or looking for from the parenting intervention?*

Overall parenting experience

3. How was your relationship with your son/daughter before taking part in the program? How did you find supporting your child with their CF treatments?* *(Main difficulties, concerns, worries, problems affecting the family in general)*

4. Would you say that the experience of taking part so far has helped you better manage your own problems?

Effectiveness of the program

5. How was your experience of the program? And how does this compare to the information you normally receive about parenting? *(Can you tell me positive / negative experiences about it?)*

6. Overall, how do you think that the program helped you? (*In relation to parenting & cystic fibrosis*).

7. What kind of change have you seen in your child's behaviour or in the relationship with your child?

8. What strategies are you putting into practice with your family? How are they working for you?*

Relevance to Population of Parents

9. What do you think of the materials and the way they were presented, do you

think they have helped with your personal difficulties?
(How attractive were the materials? Were they relevant to you and your family? Were they understandable?)

10. Do you think there are any modifications to be made for this program which would make it more relevant to you (or more able/ willing to take part)? *Or help others in your position (What would you modify to make it more relevant to you?)**

Experience after the program

11. Can you bring to mind some time when you became angry or frustrated with your child recently? If yes: What happened? And after it was over? Is this different from before you took part in the program? If so, how?

For parents who opted out of the study from the outset ONLY

12. Can you tell me a bit about why you chose not to take part in the programme?

13. How much time would you want to/ be able to commit to parent support interventions and what mediums of support would be most useful (i.e. groups, social media, face to face contact, booklets etc.)?*

CF Professionals Feasibility Interview

At the start of the interview/ focus group, service providers will be asked basic demographic information (e.g., age, gender, ethnicity, role, and number of years working in their role).

Prior to the interview, professionals will also be provided with a summary/ copy of the content and structure of the Triple P intervention in order to inform their knowledge of this and to aid with the below questions.

1. What are your experiences of working with parents who are experiencing difficulties supporting their child with their CF treatments?
2. What are your experiences of the needs of parents experiencing such difficulties?(prompt: explore things such as specific issues that might be prevalent within the adolescent population, family relationships, parent and child wellbeing, areas that seem to be more common difficulties)
3. Having heard briefly about the self-directed Teen Triple P intervention, can you think of any strengths or limitations of integrating such an approach into the care plans of parents who are struggling to cope and support their child with their CF treatments?

4. The uptake of the Teen Triple P intervention has not been as successful as we thought it might be, do you have any ideas why this might be the case? (prompt: potential barriers to involvement, issues with intervention format, parental motivation, etc.)
5. Parents could potentially receive support for an array of things and in an array of formats. What other types and formats of support do you think could help to support parents who might be having difficulties supporting their child with their CF treatments?
   (prompt: online, face-to-face, individual, group, telephone, facilitated by CF team or external bodies)
6. Based on your experiences of working with children across the age range, do you think there are any specific issues that occur for parents when specifically supporting teenagers with their CF treatments that support packages need to be mindful of?
7. Are there any other things that we have not covered that you would like to discuss?

Appendix U: Baseline TAQ-CF and Total Parenting Scale Scores for Competing (Participants 4 & 6) and Withdrawing (Participants 1,2,3 & 5) Participants