Treatment Adherence in Adolescents with Chronic Illness: The Dyadic Experience of Adolescents and their Parents and the Efficacy of Psychological Interventions

Kelsey Odgers

Primary Supervisor: Dr Joanne Hodgekins

Secondary Supervisors: Dr Amy Carroll and Kiki Mastroyannopolou

Doctorate in Clinical Psychology

University of East Anglia

Faculty of Medicine and Health Sciences

Submission Date: 2nd March 2021

Thesis Word Count: 32,691

Candidate Registration Number: 100042365

This copy of the thesis has been supplied on condition that anyone who consults it is understood to recognise that its copyright rests with the author and that use of any information derived there-from must be in accordance with current UK Copyright Law. In addition, any quotation or extract must include full attribution

Thesis Portfolio Abstract

Background: Increasing numbers of adolescents are living with chronic illnesses which require adherence to multiple daily behaviours. Difficulties with adherence are common in every patient group. However, adolescents have higher rates of non-adherence than child and adult samples. Understanding the lived experience of managing adherence and the efficacy of interventions to promote adherence in adolescents is therefore imperative.

Aims: The first aim of this thesis was to explore the adolescent-parent dyadic experience when managing adherence to treatment in chronic illness. A second aim was to explore the efficacy of psychological interventions in promoting adherence to treatment, quality of life and family functioning in adolescents with chronic illness.

Methods: A systematic review of qualitative studies that explored adolescents and their parents' experiences of managing adherence was conducted. A second systematic review and meta-analysis was conducted which synthesised and pooled effect sizes from RCTs that examined the efficacy of psychological adherence-promoting interventions.

Results: Five 'analytic' themes were identified from nine studies in the qualitative systematic review. These highlighted the importance of relational factors including trust, negotiation and collaboration in enabling the dyad to manage adherence. Thirty-six studies were included in the second review. Several had missing data and were rated as high risk of bias. Nevertheless, significant small effects were found for adherence and quality of life outcomes at posttreatment. Follow-up, moderation and subgroup analyses were limited by the number of studies.

Conclusion: Future high-quality research, including qualitative research exploring the lived experience of adherence to treatment and adherence-promoting intervention research, is needed. Research should recruit adolescent samples with identified poor adherence across a range of chronic illnesses.

Access Condition and Agreement

Each deposit in UEA Digital Repository is protected by copyright and other intellectual property rights, and duplication or sale of all or part of any of the Data Collections is not permitted, except that material may be duplicated by you for your research use or for educational purposes in electronic or print form. You must obtain permission from the copyright holder, usually the author, for any other use. Exceptions only apply where a deposit may be explicitly provided under a stated licence, such as a Creative Commons licence or Open Government licence.

Electronic or print copies may not be offered, whether for sale or otherwise to anyone, unless explicitly stated under a Creative Commons or Open Government license. Unauthorised reproduction, editing or reformatting for resale purposes is explicitly prohibited (except where approved by the copyright holder themselves) and UEA reserves the right to take immediate ‘take down’ action on behalf of the copyright and/or rights holder if this Access condition of the UEA Digital Repository is breached. Any material in this database has been supplied on the understanding that it is copyright material and that no quotation from the material may be published without proper acknowledgement.

List of Contents

List of Tables

List of Figures

List of Appendices

Acknowledgements

I firstly want to thank my research supervisors, Jo Hodgekins, Amy Carroll and Kiki Mastroyannopoulou. Due to the COVID-19 pandemic, my original project unfortunately could not go ahead, so thank you all for supporting me through this difficult period. I couldn't have finished this thesis without your support, reassurance and belief in me! I would also like to thank Jo and Ari for all of your support in reading the papers and checking of quality/risk of bias assessment, I really appreciate your help. Thank you to Matthew Smith for all of your patience in helping me to develop the searches, and to Richard Meiser-Stedman for all of your teaching and support in running meta-analyses. I am thankful also to all of my trainee colleagues, but in particular Jo, Simone, Charlotte, Rachel T and Jinnie – I'm so lucky to have shared my training experience with you all. Thank you to my close friends and family – my mum, stepdad, grandparents and Imogen, I couldn't have done it without you! Finally, I want to thank my Fiancé, Jacob, who has been my rock throughout this process.

Chapter One: Thesis Portfolio Introduction

This chapter introduces and provides a definition of the key concepts of this thesis portfolio, namely chronic illness, adolescents and adherence. It outlines the key issues and theories in the context of adherence and self-management in adolescents with chronic illness. The rationale of the aims of the work in this thesis, with reference to the relevant literature, are also outlined in the context of the underlying epistemology and ontology of the first author.

Key Definitions

Chronic Illness

For the purposes of this thesis, chronic illness is defined as a disease with an expected duration of more than three months which requires on-going clinical intervention and self-management (Heath et al., 2017; Law et al., 2019; van der Lee et al., 2007). These illnesses include, but are not limited to, Epilepsy, Diabetes, Asthma, Sickle Cell Disease, Transplant, Spina Bifida, Cystic Fibrosis, Human Immunodeficiency Virus, Arthritis, Inflammatory Bowel Disease, Cancer, Chronic Pain, Gastrointestinal Disease, and Communicable Diseases.

This thesis is focused only on physical chronic illnesses, so studies focusing on samples of adolescents with psychological disorders (i.e., depression) or neurodevelopmental disorders (i.e., Autism) were not included. Although an important issue for examination, it is beyond the scope of the present thesis. Finally, studies on obesity were also not included, because there is debate as to whether obesity is conceptualised as a chronic illness, or a lifestyle variable and condition which can lead to the development of chronic illnesses (Graves et al., 2010).

Adolescents

For the purposes of this thesis, adolescents are defined as people aged between 10 and 19 years, which is in accordance the World Health Organisation's definition (World Health Organisation, n.d.).

Increasing numbers of adolescents are living with a chronic illness which significantly impacts their daily lives (Hanghøj & Boisen, 2013; Heath et al., 2017). Suris et al. (2004) estimated that 10% of adolescents suffer with a chronic illness and Hagell et al. (2015) reported that in the UK there are increasing numbers of adolescents diagnosed with a chronic illness.

Due to advances in treatment, more adolescents with a chronic illness will now reach adulthood (Heath et al., 2017). Living with and managing these illnesses often requires complex behaviours which adolescents and their families have to perform daily (Hanghøj & Boisen, 2013; Modi & Driscoll, 2020). These include monitoring symptoms (for example, daily blood glucose testing in Diabetes), following a specific diet or exercise regimens (for example, exercise regimens in Juvenile Idiopathic Arthritis) and taking medications or performing other treatments/therapies (for example, daily inhalers in Asthma).

Literature suggests that adolescents, regardless of the chronic illness, have more difficulties with adhering to treatment regimens than child and adult samples (DiMatteo, 2004; Hanghøj & Boisen, 2013; Rapoff, 2010). There are a range of theories and perspectives of adolescence and development which may explain why adolescence is a unique period for adherence compared to childhood or adulthood. A number of these will now be discussed to place the research in this thesis into the wider context.

First, theories of adolescent psychosocial development may explain why adolescents have difficulties with adhering to treatment. Adolescence is a unique developmental phase in which individuals experience physical, social and psychological

changes (Viner & Christie, 2005). Several models and theories define adolescence a unique period of human development. Piaget's (1936) theory suggests that from approximatively the age of 12 years, adolescents being to develop abstract reasoning abilities. More recent research suggests that the development of abstract thinking is gradual and not a linear process (Smith et al., 2011), which means that these skills and abilities are developing throughout adolescence. This cognitive ability is needed for adolescents to plan and prepare; to consider multiple outcomes of behaviour; and to think hypothetically about the future. Therefore, to manage their treatment independently, adolescents will need these skills (Viner & Christie, 2005). However, unlike children, adolescents are expected to be more autonomous in the management of their illness as parents begin to have less involvement in their child's treatment regimens (Lerch & Tharne, 2019), which may be problematic as they still developing these cognitive abilities.

Erikson's (1968) theory of psychosocial development suggests that during the period of adolescence there are more tensions around personal identity development, which he called the moratorium phase. This period, Erikson argues, is characterised by the adolescent experimenting with who they are, their values and roles and thus is it normal for adolescents to be uncertain about their decisions, rebel against parents and be uncooperative. This could explain why adolescents with chronic illness may be reluctant or refuse to adhere to treatment, as they are experimenting with who they are or rebelling against their parents (Suris et al., 2004). Whilst some will adopt roles and values that align with their parents, many will develop identities which align with their peers, as peers are central to identity development (Erikson, 1968). Therefore, the development of such relationships may conflict with treatment demands and thus the adolescent may prioritise 'fitting in' and time with peers over treatment tasks (Suris et al., 2004).

Finally, neurobiological theories may also explain why adolescents have difficulties with adherence. Blakemore (2019) argues that during adolescence the pre-frontal cortex which determines executive functioning, including planning, decision-making, and self-awareness, goes through a period of significant development. Due to this significant neurological development, adolescence is characterised by poor planning and increased risk-taking behaviour. Therefore, adolescents with chronic illnesses may not comprehend the long-term risks of non-adherence and thus may take more risks in relation to the management of their illness compared with adults (Suris et al., 2004).

Adherence and Self-Management

For the purposes of this thesis, adherence is defined as 'the extent to which a person's behaviour; taking medication, following a diet, and/or executing lifestyle changes, corresponds with agreed recommendations from a health care provider" (World Health Organisation, 2003, p.3). Adherence to multiple domains including self-care and/or self-management also form part of this definition (Kahana et al., 2008).

Rates of Adherence in Adolescence and Consequences of Poor Adherence

Adhering to treatment regimens can be problematic for every patient group but seems to be particularly problematic in adolescent samples (DiMatteo, 2004; Hanghøj & Boisen, 2013; Rapoff, 2010). Rapoff (2010) reported that rates of non-adherence average 50% in paediatric samples, with some studies reporting rates of non-adherence in adolescent samples as high as 75%. These rates have not changed over the past decade (Modi & Driscoll, 2020). This compares to lower rates of non-adherence in adult populations, which are between 30% and 50% (Peng et al., 2020).

Good adherence is vital in reducing healthcare utilisation and costs, decreasing the risk of morbidity and mortality and improving health outcomes (Kahana et al., 2008; Modi & Driscoll, 2020; Pai & McGrady, 2014). In addition to the physical health consequences

of poor adherence, studies have found that failure to adhere is associated with poor health-related quality of life (Fredericks et al., 2008) and family functioning (Psihogios et al., 2019).

Measuring Adherence and Self-Management

Many objective and subjective measures of adherence and self-management exist across various chronic illnesses. However, in both clinical practice and research, subjective measures are the most commonly used method (Plevinsky, Gutierrez-Colina et al., 2020).

Objective Measures

Objective measures include electronic monitors, such as insulin pumps and continuous glucose monitors in Type 1 Diabetes (Modi & Driscoll, 2020), technology which can record nebulizer use in Cystic Fibrosis (O'Toole, 2019), electronic pill boxes, and pharmacy refill data (Modi and Driscoll, 2020).

Other objective measures include biomarkers collected from urine and blood samples. Whilst these are objective, it is acknowledged that other factors aside from adherence can affect results from these samples (Modi & Driscoll, 2020).

Subjective Measures

Whilst objective adherence measures are considered the gold standard, they are often costly (Plevinsky, Gutierrez-Colina et al., 2020). Many self-report measures that have satisfactory psychometric properties exist across multiple chronic illnesses, as outlined in reviews by Plevinsky, Gutierrez-Colina et al. (2020) and Quittner et al. (2008). These measures vary across different chronic illness types and measure a range of constructs from specific adherence behaviours to general measures of daily adherence via diaries (Plevinsky, Gutierrez-Colina et al., 2020). These measures are often self-report (i.e., reported by youth themselves), but many parent-report measures exist.

The issues identified with these measures, due to their subjective nature, include social desirability bias and inaccurate recall. However, they are quickly administered and are low cost, so are often used in clinical settings (Plevinsky, Gutierrez-Colina et al., 2020).

Factors Influencing Adherence

Several factors that influence adherence have been explored in the literature. Given this, a brief outline of key findings related to the current thesis will be discussed in this introduction.

As aforementioned, evidence suggests that adolescents have more difficulties with adhering to treatment.

Illness beliefs and beliefs about treatment are important factors that influence adherence for adolescents with chronic illnesses. Several studies across different chronic illnesses, including Cystic Fibrosis (Bucks et al., 2009) and Asthma (Mammen et al., 2016), have found that adherence is influenced by beliefs held by adolescents about the benefits and side effects of treatment.

A systematic review conducted by Law et al. (2014) explored the extent to which illness perceptions relate to self-management in children and adolescents with chronic illness. They found that control beliefs (i.e., beliefs about controllability of the illness and its management) are strongly related to self-management. They also argue that illness beliefs amongst families and caregivers should be considered, given that they assume responsibility of adherence for young children and often maintain an active role in supporting their adolescent manage adherence to treatment.

Other family and caregiving factors have been found to be important in adherence in paediatric samples. A recent meta-analysis found that better adherence was significantly associated with better family functioning, including decreased family distress and conflict

and greater family cohesion in children and adolescents with a wide range of chronic illnesses (Psihogios et al., 2019).

The link between peer support and adherence in adolescents with chronic illness has been explored. It has been found that peers can offer a supportive role, but that this does not appear to impact adherence (Pendley et al., 2002). However, adolescents who anticipate negative reactions from peers are more likely to have difficulties with adherence (Hains et al., 2007).

Finally, healthcare professional factors that may influence adherence have been explored. Non-support from healthcare professionals was found to be linked to poorer adherence (Singh et al., 2013) and the significance of good adolescent and healthcare professional communication in adherence in Type 1 Diabetes has been discussed in a recent review by Patel et al. (2018).

Perspectives of Young People and Their Families

Whilst research that focuses on factors that influence adherence in paediatric samples is important, this body of research fails to understand adherence from the perspectives of young people and their families. This is an issue given that they are often expected to manage complex treatment regimens on a daily basis in their own homes (Lindsay et al., 2011). Studies that focus on both adolescent and parental perspectives on facilitators and barriers to adherence have been conducted.

Reviews by Lindsay et al. (2011) and Hanghøj and Boisen, (2013) synthesised both quantitative and qualitative research which examined barriers and facilitators to treatment adherence in adolescents with chronic illnesses. Key findings highlighted the importance of parental and peer relations (particularly around parental involvement in managing the illness), complexity of the regimen and beliefs about the illness and treatments. Hanghøj and Boisen (2013) argued that barriers were not unique to illnesses, but that adolescents

appear to have challenges with adherence unique to this developmental period. Santer et al. (2014) later conducted a systematic review of qualitative studies exploring the reasons for non-adherence among caregivers of children up to 12 years old living with a chronic illness. They highlighted that despite the challenges of complex treatment regimens which could threaten 'normal' family life, caregivers work hard to overcome their challenges to treatment adherence.

Whilst these findings are important, no previous review has captured both adolescent and parent perspectives on managing adherence to treatment. This is an important gap given that adolescent adherence is managed within a family context (Modi et al., 2012), with parents remaining involved in the treatment regimens into their child's young adult years (Pritlove et al., 2020).

Therefore, this thesis aims to fill this gap by conducting a systematic review and synthesis of qualitative studies that have explored the dyadic experience of adolescents and their parents when managing adherence to treatment.

Theories of Adherence and Self-Management

A wide range of theories and models that attempt to understand and explain adherence and self-management in paediatric samples exist. A detailed discussion of each theory and its application in pediatric adherence literature can be found in Modi and Driscoll (2020). Key models that have been applied to explain paediatric adherence include the Health Belief Model (HBM) and the Theory of Planned Behaviour (TPB) and Reasoned Action (TRA).

The HBM (Rosenstock, 1974) was applied to adherence behaviours by Becker and Maimam (1975). This model outlines key factors which influence behaviours related to health including perceived threat to developing an illness or becoming sick (perceived susceptibility), belief about the severity of the illness (perceived severity) and perceived

benefits and barriers of particular adherence or self-management behaviours (Modi & Driscoll, 2020).

Intention to perform a specific behaviour is thought to be key in Fishbein and Ajzen's (1975) TPB and later, TRA. This intention, it argues, is influenced by three factors: attitude towards and belief about the specific adherence behaviour; subjective norms and social pressures around whether the adherence behaviour would be viewed positively or negatively; and finally, self-efficacy around the individual's confidence to complete the task (Modi & Driscoll, 2020).

As discussed by Modi and Driscoll (2020), no theory applied to paediatric adherence has been proven to be best. Therefore, cotemporary theories have been developed which focus specifically on paediatric adherence. Both the Self and Family Management Model (Grey et al., 2015) and the Self and Family Self-Management Theory (Ryan & Sawkin, 2009) consider the systems around children and adolescents who are managing adherence to treatment (Modi & Driscoll, 2020). Both consider risk and protective factors (for both the individual and systems) which influence adherence and self-management and how these factors influence proximal (i.e., specific behaviours related to adherence) and distal outcomes (i.e., quality of life, healthcare cost).

Finally, the Pediatric Self-Management Model (Modi et al., 2012) has considered both the modifiable and non-modifiable individual, family and healthcare factors that influence adherence (Modi & Driscoll, 2020). It has therefore been applied in understanding populations that might be at high risk for poor adherence and in highlighting targets for adherence-promoting interventions (Modi & Driscoll, 2020).

This thesis draws on contemporary models of paediatric self-management by moving beyond the individual adolescent to consider adherence within the family system (Grey et al., 2015; Modi et al., 2012; Ryan & Sawkin, 2009). However, given that no

theory has been proven to be best (Modi & Driscoll, 2020), findings will be discussed in relation to other theories were appropriate.

Interventions to Promote Adherence

Due to the high rates of treatment non-adherence in young people with chronic illness and the health, wellbeing and overall costs of non-adherence, many psychological interventions have been developed and evaluated which aim to overcome the barriers to adherence and promote adherence in these populations (Drotar, 2000; Kahana et al., 2008; Modi & Driscoll, 2020). Psychologists have a key role in supporting adolescents and their families in improving adherence in chronic health illnesses given that they have unique expertise in addressing psychological challenges that affect adherence (Rapoff & Calkins-Smith, 2020). These interventions are defined as those which have a cognitive and/or behavioural, educational, organisational, problem solving or a family communication component including multisystemic therapy or those which use technology-based approaches (Kahana et al., 2008; Modi & Driscoll, 2020; Pai & McGrady, 2014).

Several meta-analyses examining the efficacy of psychological interventions to promote adherence and self-management in children and young people with chronic illness have been conducted (Graves et al., 2010; Kahana et al., 2008; Pai & McGrady, 2014). These studies have highlighted the significant amount of heterogeneity in the reporting in adherence-promoting intervention research and poor quality of studies. Taken together, they suggest that further transparency in reporting and standardisation is needed to improve quality of studies.

Given the methodological issues highlighted in previous systematic reviews and changes in more recent years in the way in which interventions are delivered and adherence is measured, a systematic review and meta-analysis to examine the efficacy of recent adherence-promoting interventions is needed. Further, to the author's knowledge, no

meta-analysis exists which focusses specifically on adolescents. This is despite adolescents appearing to have more difficulties with adhering to treatment regimens than children and adults (DiMatteo, 2004; Hanghøj & Boisen, 2013; Rapoff, 2010). Previous reviews have argued that examining the developmental age of the young person is needed (Pai & McGrady, 2014). In more recent years, adherence-promoting researchers recruit narrow age ranges in their samples and now many RCTs including adolescents only exist (e.g., Ellis et al., 2019; Kosse et al., 2019). This means that a systematic review and meta-analysis examining the efficacy of these interventions in adolescents specifically is possible.

Outline of the Thesis

With the above in mind, the current thesis aims to address these aforementioned gaps and make a clinically meaningful contribution to the field. First, the thesis aims to increase understanding of the lived experiences of both adolescents and their parents when managing adherence to treatment. Second, it aims to examine the efficacy of recent psychological interventions to promote adherence, quality of life and family functioning in adolescents with chronic illness.

To achieve these aims, a systematic review (chapter two) is reported which explored the adolescent-parent dyadic experience when managing adherence to treatment. Chapter three, a bridging chapter, outlines how the findings from this review can inform the development of interventions to promote adherence.

In chapter four, the methods and findings of the systematic review and meta-analysis which examined the efficacy of psychological interventions in promoting adherence, quality of life and family functioning in adolescents with chronic illness are reported. In chapter five, additional information regarding the methodology of both

reviews is presented, followed by chapter six which presents additional results from the meta-analysis.

In the final chapter, an overall discussion and critical evaluation of both the qualitative systematic review and systematic review and meta-analysis are presented. This brings the findings of both pieces of work together and positions them within the existing literature. Implications for clinical practice and future research are also discussed.

Epistemology and Ontology

Qualitative and quantitative research often contrast in their ontological and epistemological positions. However, combining these approaches is advocated, with researchers arguing that the most appropriate methods for answering research questions should be employed (McEvoy & Richard, 2006). Ontology examines the nature of reality and whether there is an objective reality, or truth, separate from human experience (Fletcher, 2017). Epistemology refers to knowledge about reality and how knowledge can be reached (Fletcher, 2017). Given that the current thesis uses both qualitative and quantitative methodology and therefore combines positions from both methods, the researcher's position needs to be considered and made explicit (McEvoy & Richard, 2006).

McEvoy & Richard (2006) argued that a critical realistic framework underpinning research using a combination qualitative and quantitative methods may overcome issues associated with "paradigm switching" (p. 66). This framework outlines that human knowledge or discourse can inform us about reality but does not consider reality as entirely constructed by discourse (Fletcher, 2017). By taking this approach, both constructionist and positivist approaches can be considered (McEvoy & Richards, 2006).

Chapter Two

A Systematic Review of Qualitative Studies Exploring the Dyadic Experience of Adolescents with Chronic Illness and Their Parents when Managing Adherence to Treatment

Kelsey Odgers[1], Dr Amy Carroll[1], Kiki Mastroyannopolou[1], Araminta Peters-Corbett[2] and Dr Joanne Hodgekins[1]

[1]Department of Clinical Psychology and Psychological Therapies, University of East Anglia, UK.

[2]Norfolk and Suffolk NHS Foundation Trust

Prepared for: Health Psychology Review (Author guidelines in Appendix A)

Word count excluding tables/figures: 9,420

Word count including tables/figures: 11,184

Author Note

This research has been conducted as part of the Clinical Psychology Doctorate course at the UEA. The first author would like to thank the Matthew Smith from the UEA for his support in the development of the search. They would also like to thank Simon Watts from the UEA their teaching on conducting qualitative analyses.

Correspondence concerning this article should be addressed to Kelsey Odgers, Norwich Medical School, University of East Anglia, Norwich, Norfolk, NR4 7TJ, UK. Email: K.Odgers@uea.ac.uk

Abstract

Background: Adolescence is a challenging period. For adolescents with chronic illness, managing adherence to treatment brings greater complexity. Chronic illnesses in adolescence are managed within a changing parent-adolescent relationship, so understanding adherence in this context is imperative.

Objective: The study aimed to understand the adolescent-parent dyadic experience when managing adherence to treatment in chronic illness by synthesising published qualitative studies including both adolescents and their parents.

Method: A thematic synthesis of qualitative studies was conducted. CINAHL, MEDLINE, and PsycINFO were systematically searched between 2000 and 2020. Included papers were quality assessed using the Critical Appraisal Skills Programme.

Results: Five 'analytic' themes were identified from nine studies: (1) Managing complexity whilst preserving 'normal' life (2) The relational consequences of forgetting (3) The social context of adherence (4) Family beliefs about adherence and (5) Responsibility, roles and relationships.

Conclusion: The review highlighted the importance of relational factors when managing adherence. Aspects of the parent-adolescent relationship including trust, negotiation and collaboration were identified as key in enabling the dyad to manage adherence. This suggests that adherence-promoting interventions for adolescents should include parents and future high-quality research should include multiple perspectives.

Keywords: adolescents; parents; chronic illness; adherence; relationship; qualitative

Introduction

Increasing numbers of adolescents are diagnosed with a chronic illness each year in the UK (Hagell et al., 2015). Chronic illnesses are those with a duration of more than three months which requires some form of on-going self-management (Heath et al., 2017; Law et al., 2019; van der Lee et al., 2007). Advances in treatment mean that more adolescents will reach adulthood but living with these illnesses require complex treatment regimens that adolescents and their families have to manage (Hanghøj & Boisen, 2013; Heath et al., 2017; Modi & Driscoll, 2020). Adherence to these treatments is problematic in every patient group, but rates of non-adherence appear to be higher in adolescent samples compared to child and adult samples (DiMatteo, 2004; Hanghøj & Boisen, 2013; Rapoff, 2010).

The cost of non-adherence to adolescents living with chronic illness, their families and society is significant (Modi & Driscoll, 2020). Several studies have explored barriers and facilitators to adherence from the perspectives of adolescents and their parents (Lindsay et al., 2011). However, given that these studies vary in their aims, samples and methodologies it is difficult for healthcare professionals to draw conclusions from this literature. Therefore, systematic reviews in the field exist.

Reviews by Lindsay et al. (2011) and Hanghøj and Boisen, (2013) synthesised quantitative and qualitative research which examined barriers from the perspectives of adolescents with chronic illnesses. Lindsay et al. (2011) also examined facilitators. The authors found that amongst 12-20-year-olds key barriers included forgetfulness, difficulty with the treatment regimen (including denial about its effectiveness and side effects), interference with activities, and embarrassment. Facilitators included social support, education about the illness, and learning self-management skills. Hanghøj and Boisen, (2013) examined self-reported barriers amongst 13-19-year-olds and similarly found that

relations with peers, worries about the treatment and forgetting were key barriers. They argued that barriers were not unique to illnesses, but those around peer and parent relations, were specific to adolescents. Both systematic reviews highlighted the importance of parental involvement in adherence, particularly around who carries the main responsibility, and how this can lead to conflict amongst adolescents and their parents.

A recent mixed method review by Lerch and Tharne (2019) explored the transition to self-management from the perspectives of adolescents. They found that perceptions of adolescent self-efficacy and their confidence support them in assuming responsibility. Important parental factors including being collaborative, resourceful and available also supported the transition.

These mixed method reviews, which mainly utilised data from quantitative studies using psychometric data and questionnaires, have provided insight into adolescents' experience of managing adherence to treatment. However, a systematic review including only qualitative studies may allow for a deeper and richer understanding of the qualities of adolescents' lived experience when managing adherence to treatment to be gained. Further, these reviews overlook parents and caregivers' experiences. This is an important gap given that parental involvement in managing adherence to treatment in adolescence is vital (Denison et al., 2015; Luo et al., 2020; Rapoff, 2010).

The author is aware of only one review which aimed to explore parents' experiences of adherence to treatment in chronic illnesses. Santer et al.'s (2014) thematic synthesis explored the reasons for treatment non-adherence among caregivers of children up to 12 years old. They highlighted that despite the challenges of complex treatments which could threaten 'normal' life, caregivers worked hard to overcome the challenges. Unlike previous reviews in this area, Santer et al.'s (2014) review included only qualitative studies which meant that a rich analysis could be conducted. This provided a deeper

understanding into the complexity of treatment adherence amongst parents of children that can inform interventions to promote adherence in young children. However, Santer et al.'s (2014) only synthesised studies which included parents of young children.

There remains a gap in the literature, with no previous review capturing both adolescent and parent perspectives. Contemporary theories of pediatric self-management move beyond the individual and consider adherence within the context of family systems (Modi et al., 2012). Adolescence is a period where individuals experience numerous changes and begin to move towards autonomy from their parents (Sanders et al., 2013; Viner & Christie, 2005). However, for adolescents with chronic illness this is more challenging due to the daily treatments these illnesses require (Lerch & Tharne, 2019). For many, parents remain involved in their treatment into their child's young adult years (Pritlove et al., 2020). Given that chronic illnesses during adolescence are managed within this changing parent-child relationship (Williams-Reade et al., 2019), understanding adherence in this context is imperative.

The current review aimed to fill the aforementioned gap by synthesising qualitative studies that explored the experience of adherence to treatment from both adolescents and their parents' perspectives. This is informed by both contemporary literature and theories of paediatric self-management (Grey et al., 2015; Modi et al., 2012; Ryan & Sawkin, 2009), which consider adherence within the context of the family systems. It is hoped that by bringing together adolescents and their parents' parallel experience, a deeper understanding of the nature of their dyadic relationship while managing treatment in chronic illnesses can be gained. This will inform clinical practice and future adherence-promoting intervention research.

Research Question

The research question was based on the SPIDER criteria (Sample, Phenomenon of Interest, Design, Evaluation, Research type, Cooke et al., 2012). This assisted in the developed of key search terms in order to identify relevant literature. These were:

Sample: Adolescents with chronic illness and their parent/caregivers.

Phenomenon of interest: The dyadic experience when managing adherence to treatment.

Design/research type: Qualitative methodology.

The research question is 'what are the lived experiences of the adolescent-parent dyad when managing adherence to treatment in chronic illness?'

Methods

Guiding Framework and Registration

This review adopted a thematic synthesis methodology, outlined by Thomas and Harden (2008). This was chosen as this approach has been deemed suitable for questions around the experience of treatment adherence (Santer et al., 2014) and it also aligns with a critical realist stance which is appropriate given the aim around gathering multiple perspectives of the same common or underlying 'reality'. The review did not hope to find the 'true reality' about the dyadic experience when managing adherence to treatment, which may in fact exist. However, it took the stance that acknowledging both perspectives can inform us about a common underlying 'reality' (McEvoy & Richards, 2006).

To ensure transparency in the methods, the Enhancing Transparency in Reporting the Synthesis of Qualitative Research (ENTREQ) Statement (Tong et al., 2012) and the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA, Moher et al., 2009) were adhered to. See supplementary material for details.

This review was registered on PROSPERO (registration number: CRD42020213097).

Search Strategy

The initial pre-planned systematic search was carried out on 20th July and searched the literature from January 2000, to ensure that studies had a contemporary focus, until 20th July 2020. This was across three electronic databases: CINAHL Complete (EBSCO), MEDLINE Complete (EBSCO) and PsycINFO (EBSCO). These were chosen as they contain relevant studies in this area. Before the search was conducted, the final search terms were agreed with the fifth author to ensure they were robust and rigorous. To capture additional studies, manual searches of the reference lists of relevant reviews, book chapters and included studies were conducted. Grey material was not included to ensure that included studies were peer-reviewed. The search was updated on 7th December 2020 before the analysis was conducted.

The final search terms are provided in Table 2.1 and a copy of the MEDLINE search can be found in Appendix B.

Table 2.1

Final Search Terms

Concept One Terms for parent/caregiver	Concept Two Terms for adolescent	Concept Three Terms for chronic illness	Concept Four Terms for adherence	Concept Five Terms for research type
Parent* or mother* or father* or guardian* or caregiver*	Child* OR adolescen* OR youth OR teen* OR "young people" OR infant OR "young adult*"	“Chronic illness*” OR "Chronic disease*" OR epilepsy OR diabetes OR asthma OR "sickle cell disease" OR transplant* OR "spina bifida" OR "cystic fibrosis" OR "human immunodeficiency virus" OR HIV OR arthritis OR "inflammatory bowel disease*" OR cancer OR “chronic pain” OR “gastrointestinal disease*” OR “communicable disease”	Adherence OR compliance OR concordance OR non-adherence OR self-management OR management OR "self care" OR "self-care" OR “poorly controlled”	Qualitative or experience* or perspective* or perception* or perceived or belief* or view* or narrative*

Note. MeSH terms were used where available. Free text was used to search titles and abstracts. “Apply related words” and “apply equivalent subjects” were used to increase the inclusiveness of the search. No language limits were put on the search, as the Librarian recommended. All concepts were searched with “AND”. The reason that terms for adolescents and parents were separated was to identify papers which included the rational aspect and explored both adolescents and their parent/caregivers’ perspectives. This was in order to answer the research question.

Inclusion and Exclusion Criteria

These criteria are in Table 2.2 and were based on the SPIDER criteria (Cooke et al., 2012).

Table 2.2

Inclusion and Exclusion Criteria

Inclusion Criteria	Exclusion Criteria
Sample Adolescents with a chronic illness (Heath et al., 2017; Law et al., 2019; van der Lee et al., 2007), aged between 10 and 19 years (World Health Organisation, n.d.) and their parents/caregivers of any age above 18 years (adults only). Caregivers must be the primary care provider to the adolescent, but do not have to be biological parents.	Sample Individuals who are either younger than 10 years of age or older than 19 years of age or there is a mixed sample of adults/children and adolescents and/or chronically ill and well adolescents. Parents and/or caregivers are not included, so the relational aspects when managing adherence to treatment are missed. The focus is not solely on adolescents and their parents/adult caregivers and data was obtained from others outside of this dyad, including healthcare professionals, teachers or other family members. Samples of adolescences with psychological disorders (i.e., depression) or neurodevelopmental disorders (i.e., Autism) only. Samples of adolescents with obesity only.
Phenomenon of Interest Qualitative studies which have explored adolescent and parent dyadic experience when managing adherence to treatment.	Phenomenon of Interest Those not explicitly focussed on the adolescent and parent dyadic experience when managing adherence to treatment. This includes those which focus on related issues, for example on coping with the illness generally. This is to ensure rich and in-depth data on the topic of interest in order to answer the research question.
Study Design Qualitative methodology only.	Study Design Quantitative and mixed method.
Research Type Manuscripts which have been written or translated into the English language.	Research Type Manuscripts that have been written in any language other than the

Inclusion Criteria	Exclusion Criteria
Manuscripts that have been peer-reviewed.	English language unless a translated version is available. Manuscripts that have not been peer-reviewed.

Note. Table 2.2 continued.

Study Selection

Studies identified by the searches were extracted into Microsoft Excel. After duplicates were removed, titles and abstracts of studies were screened for eligibility and removed if they clearly did not meet criteria. Full-text articles of the remaining studies were read to assess eligibility, and if they were excluded at this stage, each was coded to provide the reason for this. For inter-rater reliability, 30% of full-text articles were checked independently by the fourth author and any disagreements were taken to the research team.

Quality Appraisal

The Critical Appraisal Skills Programme (CASP, 2018) qualitative checklist, which includes 10 questions, was used to assess the quality of included papers. This was selected as it is the most widely used tool in qualitative systematic reviews and is consistent with Thomas and Harden's (2008) methodology.

The CASP tool itself does not produce results which provide an overall quality rating (Long et al., 2020). However, many reviewers use scoring guidelines to classify included papers in terms of their quality (e.g., Hendry et al., 2017; McCann et al., 2016; Rushbrooke et al., 2014). The rating criteria employed by previous reviews was used, which produced an overall score out of 20 for each included paper. For each question, a score was given. If studies gave a detailed response to the question, they scored two, if they provided a partial response, they scored one and if they gave little or no information, they scored zero points (Hendry et al., 2017; McCann et al., 2016; Rushbrooke et al., 2014).

In line with these guidelines, if studies scored ≥17, they were considered to be of high quality (Hendry et al., 2017; McCann et al., 2016); those which scored ≤10 were considered to be of low quality (Rushbrooke et al., 2014); and those scoring in between (i.e., 11 to 16) were considered to be of moderate quality. The first author independently conducted quality assessments. The fourth author independently conducted the ratings for a third (33.3%) of included studies. Any disagreements were discussed.

No study was excluded based on their quality, but a 'sensitivity analysis' was planned. This involves low quality studies being assessed for their impact on the findings (Thomas & Harden, 2008).

Data Extraction

A standardised form was used to extract data from the included articles. This was completed by the first author. The following categories were included: study setting; aim(s); key findings; participant information; method of data collection and type of analysis used. The fourth author independently completed the data extraction form for a third (33.3%) of included studies. Any disagreements were discussed. All data labelled 'findings' or 'results' (including participant quotes and author interpretations and themes) were extracted and subject to analysis (Thomas & Harden, 2008).

Data Analysis

Thomas and Harden's (2008) three stage thematic synthesis was used. The first stage involved inductive line-by-line coding of all extracted data following data familiarisation. This involved putting the review questions and preconceptions aside. Data were coded manually as the first author felt that this enabled them to remain inductive. As each study was coded, each line was put into pre-existing codes or additional codes were added. Both the first and fourth authors coded the data independently and, along with the second author, discussed initial codes.

The second stage involved ‘descriptive theme’ development through organising the codes into related areas. The first author did this independently before having discussions within the research team. The third stage involved the development of ‘analytic themes’ in order to ‘go beyond’ descriptions and the content of the included studies in order to produce a rich synthesis with new interpretations and understandings (Thomas & Harden, 2008). This was achieved through the descriptive themes being reviewed in relation to the research question and thus relational factors and both the adolescent and parent perspectives were considered within each theme. The first author independently considered these issues and had subsequent in-depth discussions within the research team. Through these discussions, analytic themes emerged. Quotes from papers were then purposely chosen by copying every quote which represented a theme into a table and selecting those which were representative of both parent and adolescent perspectives and each paper.

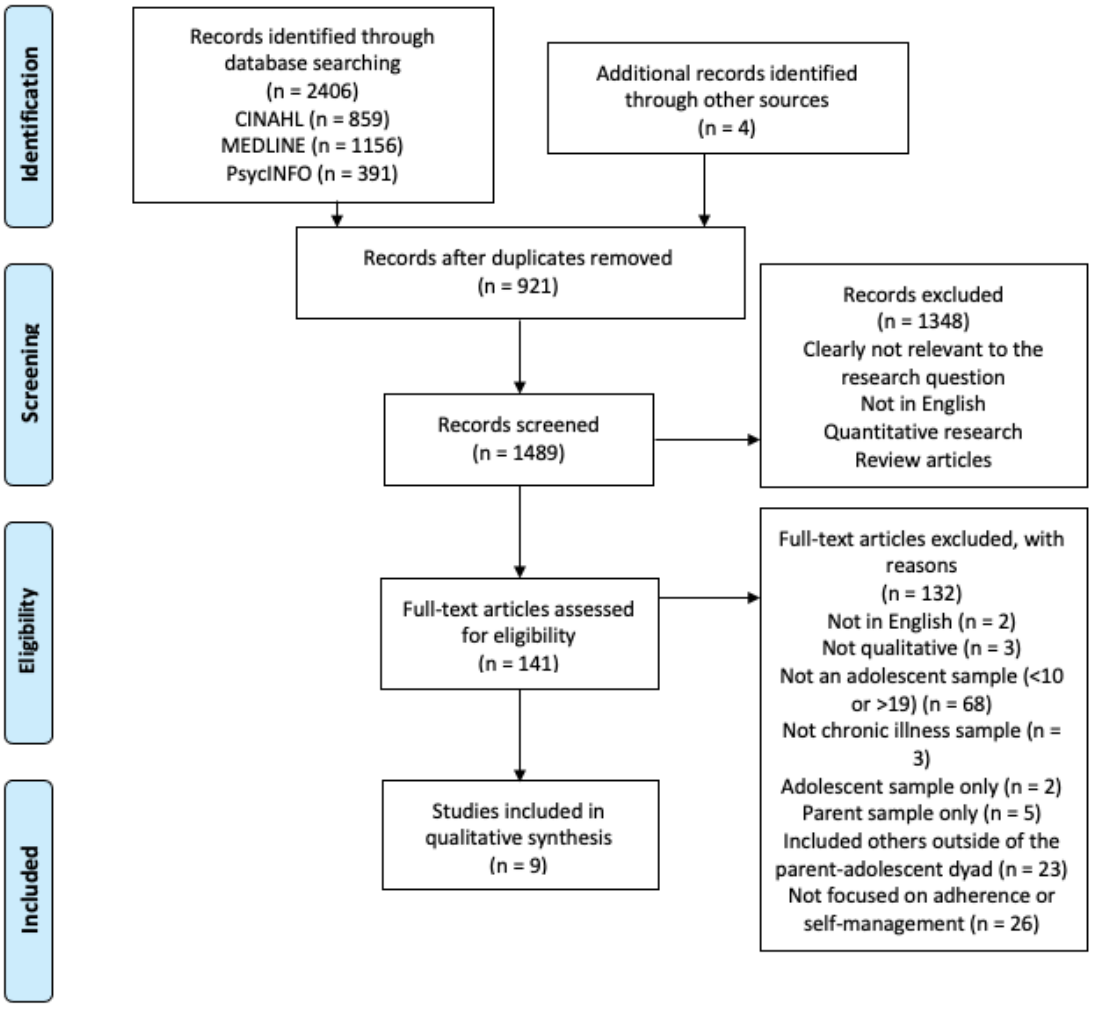
Results

Search Results

The PRISMA flow diagram is outlined in Figure 2.1. A total of 2410 papers were identified by the searches, leaving 1489 to be screened after duplicates were removed. Once titles and abstracts were screened, a total of 141 papers were read in full. This resulted in a total of nine papers for inclusion. Of the papers independently checked by fourth author, there was 97.5% agreement.

Figure 2.1

PRISMA Flow Diagram (Liberati et al., 2009)



Study Characteristics

Study characteristics of included studies are outlined in Table 2.3. Of the papers independently extracted, all extracted data was consistent with that completed by the first author. The studies were conducted across five countries: America (n = 5), UK (n = 1), Germany (n = 1), Brazil (n = 1) and Africa (n = 1). There was a total of 142 adolescents across the papers. For the parent sample there was a total of 64 mothers, 17 fathers and 29 were a mixture of mothers, fathers and other family members who were primary caregivers. It was not clear for 32 participants whether they were mothers or fathers. There

was a wide range of chronic illness diagnoses explored including Type 1 Diabetes (T1D, n = 3), Type 2 Diabetes (T2D, n = 1), Asthma (n = 1), Cystic Fibrosis (CF, n = 1), Human Immunodeficiency Virus (HIV, n = 2) and Inflammatory Bowel Disease (IBD, n = 1). Five studies purposely recruited participants with variable adherence rates in order to have a more representative sample and the remaining four did not report any data in relation to adherence rates of the sample.

Most studies used interviews to collect data (n = 6), others employed focus groups and data from discussions between adolescents and their parents. Of these, three collected the data from both adolescents and their parents together, whilst five collected it from both groups separately (one paper did not report this information). Most studies used content analysis (n = 7), with the remainder using grounded theory (n = 2).

Quality Appraisal

The first author completed the CASP checklist on all papers. For the studies independently rated, 80% were agreed upon. Differences were resolved via discussion. In all cases, differences in item ratings did not impact on the overall quality rating. Most studies were rated as being of moderate quality (n = 8), with one being of high quality (Hommel et al., 2010). As no studies were rated as low quality, a 'sensitivity analysis' (Thomas & Harden, 2008) was not conducted. Appendix C provides quality ratings.

All studies provided a clear statement of aims and qualitative methodology was deemed appropriate. Most papers selected an appropriate research design and recruitment strategy, however some lacked clarity on their design (Heyduck et al., 2015). All papers employed a data collection method which addressed their aims, with most clearly justifying the rationale for the methods used. However, this was less clear in some papers (Denison et al 2015; Ivey et al., 2007; Kourrouski & Lima, 2009).

None of the included studies adequately considered the relationship between the researcher and participants. Only one study (Sullivan-Bolyai et al., 2014) partially addressed this issue. All but two studies (Auslander et al., 2010; Ivey et al., 2007) described ethical issues adequately, but many were not explicit in how ethical issues were discussed with participants. No included study adequately conducted a rigorous analysis, although most provided details of the process and provided participant quotes. Main issues were around the role of the researcher not being considered and a lack of sufficient detail of the process. Finally, the majority of studies provided a clear statement of findings and discussed the value of them in context of the relevant literature.

Table 2.3

Study Characteristics Table

Study author (year)	Country and setting	Aim	Adolescent N; Adolescent age range (years); Parent N; Parent age range (years); Parent description	Chronic illness diagnosis	Method of Data Collection	Type of Analysis	Key Study Findings	Quality Rating
Table 2.3								
Auslander et al. (2010)	America; Pediatric clinics	"To identify psychosocial resources and barriers to self-management among African American adolescents with type 2 diabetes and their mothers" (p.613).	10; 14-19; 10; Not reported (mean 42.2); Mothers	Type 2 Diabetes	Separate adolescent and parent interviews	Grounded Theory	Resources included mother's role as the primary support person, emergence of greater self-efficacy and coping over time, family recognition of the seriousness of diabetes, and the presence of supportive peers. Barriers included comorbidity, dietary and other regimen challenges, negative peer influences, and financial problems. (p.613)	Moderate

Study author (year)	Country and setting	Aim	Adolescent N; Adolescent age range (years); Parent N; Parent age range (years); Parent description	Chronic illness diagnosis	Method of Data Collection	Type of Analysis	Key Study Findings	Quality Rating
Table 2.3								
Denison et al. (2015)	Africa; ART clinics	"To explore ART adherence from the perspectives and experiences of older ALHIV (aged 15_18) and their adult caregivers in Zambia" (p.2).	32; 15-18; 23; 23-70; Primary caregivers (19 female caregivers including aunts, mothers, sisters, grand-mothers and stepmothers and four male caregivers including fathers,	HIV	Semi-structured interviews (Not reported if together or separate)	Content analysis using a codebook	Barriers to ART adherence included fear of disclosure and anticipated stigma. Few youth were willing to take their drugs outside of the home, which led to missed doses of ART. Similarly, families tended to manage HIV within the home only. As a result, although caregivers and families were often the greatest source of emotional and instrumental support, they coped with HIV in isolation of other potential support from their communities, schools or churches. Factors that supported ART adherence	Moderate

Study author (year)	Country and setting	Aim	Adolescent N; Adolescent age range (years); Parent N; Parent age range (years); Parent description	Chronic illness diagnosis	Method of Data Collection	Type of Analysis	Key Study Findings	Quality Rating
Table 2.3								
			cousins, and uncles				included attending clinic sponsored youth groups, wanting to maintain one's health and using phone and clock alarms. Involvement of adult caregivers in HIV management varied greatly and was often based on the age and health status of the youth. Some caregivers struggled with letting the adolescents assume responsibility for their medication, and ALHIV had few self-management skills and tools to help them regularly take ART. (p.1)	

Study author (year)	Country and setting	Aim	Adolescent N; Adolescent age range (years); Parent N; Parent age range (years); Parent description	Chronic illness diagnosis	Method of Data Collection	Type of Analysis	Key Study Findings	Quality Rating
Table 2.3								
Hanna and Guthrie (2001)	USA; Diabetes specialist clinic	"To identify positive and negative dimensions of support related to adolescents' assumption of diabetes management responsibility from the perspectives of parents and adolescents" (p.212).	16; 11-18; 16; Not reported; Not reported (12 mothers and all primary caregivers)	Type 1 Diabetes	Separate adolescent and parent interviews	Manifest Content Analysis	"Both parents and adolescents describe directive guidance and tangible assistance as helpful and nonhelpful, depending on degree of directness and perceived need for help" (p.209).	Moderate
Heyduck et al. (2015)	Germany; Rehab centers	"To explore adolescents' and caregivers'	15; 11-17; 15; 37-55; Mothers	Asthma	Focus groups for teens and	"A multistep qualitative	The results demonstrated high complexity in the perceptions among	Moderate

Study author (year)	Country and setting	Aim	Adolescent N; Adolescent age range (years); Parent N; Parent age range (years); Parent description	Chronic illness diagnosis	Method of Data Collection	Type of Analysis	Key Study Findings	Quality Rating
Table 2.3								
		perceptions about asthma and asthma management and examine congruence and dissimilarity within the adolescent–caregiver dyads" (p.1227).			interviews for parents	content analysis procedure following the principles of Mayring (2000)" (p.1229).	adolescents and mothers and reflected 113 specific themes that could be assigned to four main topics: asthma beliefs, representations of asthma treatment, perceptions about individual asthma management and perceptions about family asthma management. Dyadic analyses revealed congruence in the adolescent–caregiver dyads in most of the themes. However, we also found issues where divergent perceptions became evident including, for example,	

Study author (year)	Country and setting	Aim	Adolescent N; Adolescent age range (years); Parent N; Parent age range (years); Parent description	Chronic illness diagnosis	Method of Data Collection	Type of Analysis	Key Study Findings	Quality Rating
Table 2.3								
							perceptions of asthma's general impact on adolescents' life or the question of who takes the main responsibility in asthma management. (p.1227)	
Hommel et al. (2010)	Gastro clinic; USA	"The objective of this study was to examine patient- and parent-perceived factors that impact adherence to inflammatory	16; 13-17; 16; Not reported (mean 46.44 mothers and 48.81 fathers); 12 mothers and 4 fathers	Inflammatory bowel disease	Interviews with teens and parents together	Directed content analysis	Parent–child dyads identified forgetting, interfering activities, parent–child conflict and oppositional behaviour and inadequate planning for treatment as challenges to adherence. Participants reported that family support and good parent–child relationships, routines,	High

Study author (year)	Country and setting	Aim	Adolescent N; Adolescent age range (years); Parent N; Parent age range (years); Parent description	Chronic illness diagnosis	Method of Data Collection	Type of Analysis	Key Study Findings	Quality Rating
Table 2.3								
		bowel disease treatment" (p.80).					monitoring and reminding and organisational tools such as pill boxes facilitated treatment adherence. Other issues that emerged included immediacy of treatment effects and parent-adolescent responsibility for treatment. Patients and parents experience a number of challenges related to adherence within behavioural, educational, organizational and health belief domains. (p.80)	
Ivey et al. (2007)	USA; Endo clinic	"The purpose of this study was to describe	28; 11-15; 28; Not reported;	Type 1 Diabetes	Transcript of 10-	Content Analysis	"Themes identified were frustration, fear, normalizing, trusting, and	Moderate

Study author (year)	Country and setting	Aim	Adolescent N; Adolescent age range (years); Parent N; Parent age range (years); Parent description	Chronic illness diagnosis	Method of Data Collection	Type of Analysis	Key Study Findings	Quality Rating
Table 2.3								
		the ways that parents and 11- to 15-year-old teens communicate and the recurrent themes and patterns of behaviour that were revealed during brief interactions about issues related to diabetes management" (p.10).	Not reported, but all parents		minute interaction		discounting. Trusting the adolescent to manage diabetes was difficult for the parents and was associated with frustration, fear, and discounting communication" (p.10).	

Study author (year)	Country and setting	Aim	Adolescent N; Adolescent age range (years); Parent N; Parent age range (years); Parent description	Chronic illness diagnosis	Method of Data Collection	Type of Analysis	Key Study Findings	Quality Rating
Table 2.3								
Kourrouski and Lima (2009)	Brazil; School hospital	"This study aimed to understand the experience of adolescents with HIV/AIDS concerning medication adherence" (p.947).	9; 12-18; 6; Not reported; Caregivers including mothers, grand-parents and aunts	HIV	Interviews with both the teen and parents together	Content Analysis	The results showed that adolescents have difficulties in medication adherence especially due to their side effects; they try to normalize their lives in such a way that stigma and discrimination do not compromise their quality of life and treatment adherence. (p.947)	Moderate
O'Toole et al. (2019)	UK; Regional Pediatric CF Centres	"To elicit the perspectives of adolescent patients with CF and their parents on the process of adhering to	6; 11-16; 6; Not reported (mean 42); 5 mothers and 1 father	Cystic Fibrosis	Separate adolescent and parent interviews	Grounded Theory	Parents and adolescents gave different but overlapping views of the aerosol regimen and the context in which adherence takes place. In particular, beliefs, emotional reactions, and behavioral strategies	Moderate

Study author (year)	Country and setting	Aim	Adolescent N; Adolescent age range (years); Parent N; Parent age range (years); Parent description	Chronic illness diagnosis	Method of Data Collection	Type of Analysis	Key Study Findings	Quality Rating
Table 2.3								
		inhalation therapy" (p.847).					connected to adherence were discernible in the accounts of both groups, and these interact with the parent and adolescent experience of one another, mediated largely by the strength of the relationship. (p.849)	
Sullivan-Bolyai et al. (2014)	US; Pediatric Diabetes Clinic	"To describe the perspectives of teens and their parents about self-management knowledge,	10; 13-17; 13; 33-58; 10 mothers and 3 fathers	Type 1 Diabetes	Separate teen and parent focus groups	Content Analysis	From the teens' perspective there was variation in interest in learning more about T1D and management. Those teens who had been diagnosed at a very young age reported not knowing anything else	Moderate

Study author (year)	Country and setting	Aim	Adolescent N; Adolescent age range (years); Parent N; Parent age range (years); Parent description	Chronic illness diagnosis	Method of Data Collection	Type of Analysis	Key Study Findings	Quality Rating
Table 2.3								
		behaviours (including division of labour associated with T1D management) and resources used to manage T1D" (p.178).					but diabetes, while those diagnosed later developmentally embraced the active learning process. Diabetes camp and peer group support were not seen as beneficial. All the teens were interested in "helping others" with diabetes. Parents shared the common struggle with transition of self-management, with variation in parenting styles. A small group of parents reported their "job" as a parent was to make sure their child was self-sufficient in self-management, but felt	

Study author (year)	Country and setting	Aim	Adolescent N; Adolescent age range (years); Parent N; Parent age range (years); Parent description	Chronic illness diagnosis	Method of Data Collection	Type of Analysis	Key Study Findings	Quality Rating
Table 2.3								
							pressure from the health care providers (HCPs) to physically do the care, defeating the purpose. Parents and teens reported wanting HCPs to be less focused on "numbers" (blood glucose levels) and more on the whole person. Scheduling appointment changes and long waiting times were reported as problematic by all participants. (p.178)	

Note. All included studies are referenced in a separate reference list in the supplementary material.

Thematic Synthesis

Five reoccurring 'analytic' themes were identified from the nine studies which provided a deeper understanding into the lived experiences of the parent-adolescent dyad when managing adherence to treatment. These were (1) Managing complexity whilst preserving 'normal' life (2) The relational consequences of forgetting (3) The social context of adherence (4) Family beliefs about adherence and (5) Responsibility, roles and relationships.

Managing Complexity Whilst Preserving 'Normal Life'

Both parents and adolescents discussed the complexity of the multiple treatment demands across all chronic illnesses and many perceived this as a barrier to adherence. "(16 y/o female): … at first it was difficult 'cause I had to take… over twenty pills a day, and I just didn't want to. So, I skipped it a lot." (Hommel et al., 2010, p. 84).
"You know it's alright for them to sit there and say he needs this nebulizer every day, this tablet every day, that tablet every day, they're not dishing them up, administering all the medicine. (Parent)" (O'Toole et al., 2019, p. 849).

This complexity was also discussed by parents in relation to how adherence requires them and their adolescent to manage their treatment on daily basis "without a break" (Sullivan-Bolyai, 2014, p. 186). They acknowledged is difficult for them whilst trying to live a 'normal' adolescent life.

> I think that's the difficult part. He's at an age now where he has an active social life. And… it's just a constant – there it is and you have to take it no matter what. It's part of his life and at the same time, he's trying to balance that with being sixteen. (Hommel et al., 2010, p. 84)

Interestingly, there was a difference between parents and adolescents in how they discussed the impact of trying to live a 'normal' life alongside the multiple treatment

demands of their chronic illness. For parents, they discussed ‘life’ getting in the way of adherence.

“[Name of health professional] once asked us ‘what gets in the way?’ And we said ‘life’” (O’Toole et al., 2019, p. 851).

However, for adolescents managing adherence to treatment was viewed as ‘getting in the way’ of them living a ‘normal’ adolescent life, such as carry out activities, particularly with friends.

“It kind of gets in the way of me playing on the computer endlessly or erm like if I’m going to my friends” (O’Toole et al., 2019, p. 851).

This suggests that there is a difference in priorities of parents and adolescents in relation to adherence, with adolescents prioritising the social and extra-curricular activities over adherence and parents prioritising adherence. Therefore, in an attempt to improve adherence, parents and adolescents tried to normalise the illness and the complex regimen to make it ‘part of life’.

> Another parent said, ‘‘It’s going to take work to make normal’’; a third said, ‘‘I just have to make it normal.’’ Negotiation was a part of reaching for normalcy, as one parent said to the adolescent, ‘‘What is your take?’’. (Ivey et al., 2007, p. 13)

However, this was not an easy task given the nature of the complexity of treatment regimens. Therefore, many parents used activities that adolescents enjoy as incentives to facilitate adherence and promote a ‘normal’ adolescent life.

> They kind of offered a little bit of incentive like “well, you know, we can’t let you go over to her house for the whole day if we can’t depend on you to check your blood sugar on your own”. (Hanna & Guthrie, 2001, p. 218)

This also encouraged negotiation within the parent-child relationship and enabled the adolescent to take responsibility for their own self-management.

The Relational Consequences of Forgetting

Forgetting was identified by both parents and adolescents a key barrier. This was identified across many contexts including school, home and during activities (particularly social activities), which suggests that factors involved in forgetting are complex.

"If I'm on Facebook and I see something interesting, I'll put it down to type something and I'll forget about it. (Child, aged 14 years)" (O'Toole et al., 2019, p. 853).

> (Mother of 16 y/o male): "There are times when…every other weekend he visits his father for the weekend. And normally I'll make sure he packs his pills. You know, I remind him. And I wasn't there to do that (one time), and he forgot them". (Hommel et al., 2010, p. 84)

The relational nature of the review highlighted the consequences of forgetting for the parent-child relationship. Both adolescents and parents discussed how this led to parents having to remind their adolescent constantly. Whilst this was acknowledged as a facilitator to adherence, ("All adolescents found these reminders helpful", Auslander et al., 2010, p. 617, author interpretation) adolescents often resented parental reminders and perceived their parents as 'nagging'. This 'nagging' often led to feelings of frustration and anger outbursts and also oppositional behaviour which led to more conflict within the parent-child relationship.

"Nagging was universally seen as 'not working.' The results were described as ending up with anger and outbursts. Teens shared descriptions of becoming frustrated with 'restrictions' put on them, and that makes them resist more and fight back" (Sullivan-Bolyai et al., 2014, p. 187, author interpretation).

Parents found this difficult to manage as, similarly to the adolescents, they acknowledged reminders as helpful but leading to feelings of frustration and anger in their child. Some parents were able to acknowledge that nagging wasn't helpful, so attempted to

get a balance between reminding versus nagging and used positive aspects of the parent-child relationship (e.g., humour) to encourage the adolescent to adhere.

"'All I can do is to remind, I can't restrict or nag. I am trying to pull back and not ride him. When I do that it doesn't work'" (Sullivan-Bolyai et al., 2014, p. 187).

"Approaches to subtle guidance included reminding rather than telling, having discussions, reasoning, and using jokes" (Hanna & Guthrie, 2001, p. 216, author interpretation).

Another way that parents and adolescents had found to manage forgetting in multiple contexts whilst ensuring the adolescent had autonomy in remembering their treatments was through organisational tools such as alarms and also setting a routine.

> Similarly, about a third of youth reported using a phone, watch alarm or clock as tools for remembering to take their medication. One 18-year-old female participant said, ''Especially if I am really busy with school or anything, I set an alarm so that I can remember coz I can easily forget to take my drugs". (Denison et al., 2015, p. 4)

Some parents discussed the importance of them still remaining involved even if their adolescents were using organisational tools to ensure that they had adequately performed tasks. They suggested having electronic tools that would send them data about their child's adherence in order to solve the issue of nagging and reduce parental concern.

The Social Context of Adherence

Both adolescents and parents discussed the "social context of adherence" (O'Toole et al., 2019, p. 852, author interpretation). Adolescents acknowledged that whilst some self-management tasks are easier and less embarrassing to perform in front of peers and family, others are more difficult.

"You could do that with a mate but you couldn't take your nebulizer with a friend obviously. (Child, aged 11 years)" (O'Toole et al., 2019, p. 852).

Due to the complexity of treatment regimens and difficulty in 'hiding' treatments from peers, adolescents are left with a sense of feeling different. Therefore, trying to avoid 'embarrassing' self-management tasks to 'fit in' could be a barrier to adherence.

> "When she's with her friends . . . she does buy candy, cookies, and stuff . . . . She knows that's not a part of her diet, and I don't think she wants her friends to know that she has to . . . do all this to control the diabetes. So she tries to fit in". (Auslander et al., 2010, p. 619, Mother)

Interestingly, in Ivey et al.'s (2007) study, which is the only study which used transcripts of an interactions between parents and adolescents on the subject of adherence, noted that "another couple threatened to embarrass a teen in front of his peers because he had not checked his blood sugar" (p. 13, author interpretation). This suggests that some parents' attempts to promote adherence may be reinforcing another barrier of adherence (i.e., reinforcing that adolescents should feel embarrassed about self-management tasks).

There was a similar theme in relation to stigma experienced by adolescents with HIV and their families. Both HIV studies noted the stigma and discrimination faced by individuals living with HIV (Denison et al., 2015; Kourrouski & Lima, 2009). However, the social stigma was also experienced by caregivers of adolescents and often meant that they attempted to keep their child's HIV status and treatment 'hidden'.

> Fear of unintentionally disclosing the youth's HIV status emerged as the most salient barrier to ART adherence. The majority of adolescents and adults felt that knowledge of an adolescent's HIV status should be kept within the home and within the family. (Denison et al., 2015, p. 3, author interpretation)

Some adolescents were able to share their diagnosis with friends. Those who did this received reminders from friends, which acted as a facilitator to adherence. Parents also described this as a being a positive support for them.

> "I felt relief because I was hiding a secret from them . . . and so when I finally told them . . . they weren't mad . . . so now they are reminding me to go take your sugar, go take your insulin and stuff like that." (Child) "And this year, she had a lot of friends who do know and do help . . . . That's a big support in the family that some of her friends worry about what she's eating . . . ." (Mother). (Auslander et al., 2010, p. 617)

Despite friends being a facilitator of adherence, adolescents discussed how they didn't want their illness to become a regular topic of conversation. Adolescents with Diabetes also discussed how meeting others with the same diagnosis was not helpful ("teens in general did not like meeting with others who had T1D and that diabetes camp was not well received" Sullivan-Bolyai et al., 2014, p. 187), whilst those with HIV discussed similar peer support environments as helpful.

Similarly, parents discussed the process of the focus groups conducted for the purposes of the research and how they would benefit from parent support groups. This was also evidenced in the difficulty researchers had in concluding the focus groups with parents (Sullivan-Bolyai et al., 2014).

Family Beliefs About Treatment

Parental and adolescent beliefs about adherence, including beliefs about treatment effectiveness, were important in either facilitating or hindering adherence. Adolescents expressed doubts about treatment effectiveness. Interestingly, some parents also doubted the effectiveness of treatments.

> I don't feel like it does much but I know like when I don't take it, I do notice that my chest feels a bit worse, but like for a day if I didn't take it then I wouldn't notice. I went through a phase of not taking it because I didn't think it was doing anything. (Child, aged 16 years). (O'Toole et al., 2019, p. 850)

"I can understand why some parents would think "stuff it, it's making no difference" because in some ways I feel like that. (Parent)" (O'Toole et al., 2019, p. 852).

This appeared to be mediated by the limited amount of knowledge and understanding both parents and adolescents had about the function of specific treatments, such as around how they work, and side effects of the treatments.

> (Mother of 16 y/o male): "Um, I just know the 6- MP lowers your immune system, so it also makes it easier for him to catch colds…. But, I know that without it, it seems that he has more flare-ups. I don't know if his intestines actually work against themselves, or how it works exactly". (Hommel et al., 2010, p. 84)

The perceived long-term benefits of adhering including wellness and reduced symptoms, increased participation in enjoyable and important activities (including school) and in some cases staying alive, were facilitators of adherence for both parents and adolescents.

> (14 y/o male): "It's important to stay healthy…for me, cause I play a lot of sports. You don't wanna miss practices… especially 'cause I'm going into high school, I don't want to fall behind in schoolwork. So it's important to make sure that you're… not missing school cause of stomach problems or something like that". (Hommel et al., 2010, p. 85)

Similarly, the fear of consequences of poor adherence, such as increased symptoms of the illness and for some chronic illnesses, including HIV and CF, death, were facilitators of adherence for both parents and children.

"At the end of the day if I don't take them I could die because I would become ill and that but it's keeping me alive . . . but I don't like doing them. (Child, aged 12 years)" (O'Toole et al., 2019, p. 850).

One parent who, like her daughter, had a diagnosis of T2D, articulated how she had learnt from her own experience of suffering with the consequences of poor adherence and how this motivated her to support her daughter to adhere:

> "I look at her, and I see me being a diabetic, wanting her at fourteen, she can live with diabetes, and she can live a healthy and full life, but at her age, she can control it if we work at it because I don't want the same thing to happen to her suffering strokes and be like I am, being a diabetic because had I known stuff about diabetes when I was diagnosed and . . . kept to a strict diet, I don't believe I would have had strokes and stuff". (Auslander et al., 2010, p. 617, Mother)

However, for some adolescents with HIV the consequences of the illness and poor adherence lead to a sense of hopelessness and 'giving up'.

"I'm too lazy to take the medication. If I'm gonna die, let me die. My mom bothers me all the time so I tell her to let it be, leave me alone (Adriana, 15 years old)" (Kourrouski & Lima, 2009, p. 950).

This highlighted important differences between the chronic illnesses in relation to the consequences of poor adherence (i.e., a stomach 'flare up' in IBD versus death in HIV) and how these are experienced by both adolescents living with these illnesses and their parents.

Responsibility, Roles and Relationships

Adolescents and parents discussed at length the adolescent's assumption of responsibility and taking a lead role in their own self-management with parents 'letting go' of sole responsibility. Many parents experienced difficulty in relinquishing control, comparing having an adolescent child to a small child, expressing that since their child had grown managing their illness is now more difficult.

> I like to know she's done it because if she hasn't done it I worry and again it's, she'll say I'm a control freak and I probably am a bit but having had all the years where I had complete control over her treatments to almost having no control at all now, erm yeah I like to remind her, just check in my own mind that she's done it. (Parent). (O'Toole et al., 2019, p. 852)

Parents often expressed anxiety and concern as their adolescent children assumed more responsibility and would become more aware signs of poor adherence. This often led to guilt and subsequently more reminders/oversight. This was centred around parents finding it difficult to trust their adolescent to manage their illness well. For the adolescents, this perceived lack of trust and increased reminders/oversight was frustrating.

> A young woman was able to tell her mother, "I don't think you trust me or something whenever I'm not with you, about eating right, checking my sugar or taking my shot or anything. I don't think there's a lot of trust, it's usually just about diabetes, that you don't think I'll check my sugars". (Ivey et al., 2007, p. 13)

Reciprocally, parents often perceived this frustration and anger from their adolescent as difficult and this led to more frustration. It appeared that often each half of the dyad was not aware of and didn't understand the other's position. In Ivey et al. (2007) they noted that only one family had an open discussion about trust and the difficulty in shared management:

"Another mother eloquently said, 'It makes me very sad that I can't do it for you, and it makes me angry when you get angry because I ask you!'" (p. 12).

There were differences between how parents approached their role within the management of the illness, with a broad spectrum of styles. At one end of the spectrum, parents provided all of the care e.g., "a control freak" (O'Toole et al., 2019, p. 852, Parent) or "'a psycho' about diabetes management" (Sullivan- Bolyai et al., 2014, pp. 186-187,

Parent). In the middle of the spectrum was shared-management "in this situation parents encouraged the teen to do most of the diabetes management, but the parents were still monitoring the care but more from a distance" (Sullivan- Bolyai et al., 2014, p. 187, author interpretation). At the other end, parents allowed the adolescent to have complete responsibility e.g., "'they used to remind me when I was young but now they have told me that since you have grown we don't need to be reminding you, you have to be taking medicine on your own'" (Denison et al., 2015, p. 4, Adolescent).

Many agreed, despite the challenges of relinquishing control, that "co-management" and shared responsibility was the best approach. This involved the adolescent assuming responsibility with their parent offering practical and emotional support when needed. This more subtle support often facilitated adherence, promoted adolescent self-efficacy and reduced conflict within the relationship.

"'They just keep me . . . encouraged each and everyday, mean, they're there when I need their help'" (Hanna & Guthrie, 2001, p. 218, Adolescent).

Interestingly, in the Sullivan et al. (2014) paper, parents described health-care professionals as having high expectations of them and experienced them as blaming of them if their adolescent had not adhered fully to treatments. Despite there being agreement between parents and adolescent that shared management was the best approach, some adolescents shared that health-care professionals had different expectations around what their parent's role should be and encouraged parents to be actively involved in performing the treatments.

"With a few sharing they felt their parents were treated poorly by the HCPs for not physically doing the teen's hands-on diabetes care such as blood glucose monitoring" (Sullivan- Bolyai et al., 2014, p. 185, author interpretation).

O'Toole et al.'s (2019) findings highlighted the importance of health-care professionals using "appropriate language…empathy, and tactfulness" (p. 850, author interpretation) in order to facilitator good adherence.

Another key theme in relation to roles and responsibility around adherence, was around the mother's role being primary in adolescent self-management. The father's role and their relationship with their adolescent was often not discussed.

> Among all except for one of the families, even among those with fathers living in the home, the role of the fathers in diabetes management was not discussed by either the adolescents or the mothers. However, mothers were not alone in providing support. There were a few examples of extended family members who provided support to the adolescent through reminders related to diet, blood glucose testing, and taking insulin and medication. (Auslander et al., 2010, pp. 617-618, author interpretation)

Interestingly, the role of extended family members was discussed by some families and it was agreed that they were all 'invested' in supporting the adolescent with the management of their chronic illness in order to facilitate good adherence.

Analytical Overview

The individual studies highlighted key issues in understanding of the experiences of adolescents with chronic illness and their parents when managing adherence to treatment. Whilst there was overlap between the themes identified in the review and those from the individual papers, synthesising data across studies led to the development of new themes which highlighted key relational issues. This provided a deeper understanding of the complex nature of managing adherence within this dyadic relationship. The review demonstrates that whilst trying to manage adherence and preserve 'normal' life within a social context, adolescents and their parents may have different priorities and beliefs about

treatment and adherence. Forgetting was identified as a key barrier to adherence, which has consequences for the parent-child relationship alongside the dyad experiencing transitions around their roles and responsibility in relation to adherence. Aspects of the relationship itself including trust, negotiation and collaboration are identified as key in enabling parents and adolescents navigate the complexity.

Discussion

This systematic review thematically synthesised qualitative studies exploring the adolescent-parent dyadic experience when managing adherence to treatment in chronic illness. The thematic synthesis of nine qualitative studies across six chronic illnesses led to the identification of five reoccurring 'analytic' themes. This provided a deeper understanding into the lived experiences of the parent-adolescent dyad. These themes were: (1) Managing complexity whilst preserving 'normal' life (2) The relational consequences of forgetting (3) The social context of adherence (4) Family beliefs about adherence and (5) Responsibility, roles and relationships. Informed by contemporary theories of pediatric self-management (Grey et al., 2015; Modi et al., 2012; Ryan & Sawkin, 2009), this was the first review which moved beyond the individual to explore adolescents and their parents' experience of treatment adherence. Whilst the review highlighted differences in the experiences of adolescents and their parents with different chronic illness diagnoses, it also suggests that the broad challenges they face appear transdiagnostic. This could have only been achieved through a synthesis of individual studies that tend to focus only on specific diagnoses.

In keeping with previous findings (e.g., Santer et al., 2014), the review highlighted that alongside managing multiple and complex treatment regimens, families tried to preserve 'normal' life. However, this review extended previous findings by suggesting that

adolescents and their parents have differing priorities when managing adherence to treatment which they attempt to manage through normalisation and negotiation.

Another key finding was in relation to parents and adolescent beliefs around treatment effectiveness, which has been found in previous reviews (Lindsay et al., 2011; Hanghøj & Boisen, 2013; Santer et al., 2014). It was interesting that parents, despite all adolescents being over 11 years of age, still doubted effectiveness of treatments and were unsure about how they worked. This theme fits with the Health Belief Model (HBM; Becker & Maimam, 1975), as both adolescent and parents' beliefs about the severity of the illness or becoming unwell (perceived susceptibility and severity in HBM) were identified as factors which both facilitated and hindered adherence.

Supporting adolescents and their parents to preserve 'normal' life would help them feel more similar to their peers whilst still managing their treatments. Given that feeling different and embarrassed in front of peers was highlighted in both the present review and previous reviews into barriers identified by adolescents (Lindsay et al., 2011; Hanghøj & Boisen, 2013), this is important. This is supported by Fishbein and Ajzen's (1975) Theory of Planned Behaviour, which suggests that adherence is influenced by social pressures around whether the adherence behaviour would be viewed positively or negatively. However, moving beyond the individual, the current review found that parents can have a role in contributing to their child's sense of embarrassment.

Although forgetting was a key barrier reported in previous reviews (Lindsay et al., 2011; Hanghøj & Boisen, 2013), this review extended the current knowledge and highlighted key relational consequences of forgetting for the parent-adolescent dyad. It was found that as a consequence, parents constantly remind adolescents which for the adolescent is frustrating and perceived as 'nagging'. This often led to conflict. To

overcome this, some parents were able to use positive aspects of the parent-child relationship (e.g., humour, skills in negotiation) to encourage rather than 'nag'.

The final theme around responsibility, roles and relationships supported what is previously known about adolescent's transition to self-management (Lerch & Thrane, 2019). However, this review extended this understanding by highlighting that often parents and adolescents did not understand the other's position. The child-parent relationship was important in supporting or hindering this transition and thus successful self-management. Parents' fear often lead to them not to trust the adolescent and subsequently take more control. This in turn frustrated the adolescent which led to conflict within the relationship and appeared to maintain parental fears that the adolescent was not adequately managing their illness. Parents and adolescents agreed that co-management reduced conflict and improved adherence.

Strengths and Limitations

Overall, the strength of the current qualitative review was that it provided a deeper understanding around the complex nature of managing adherence to treatment within the parent-child relationship across six chronic illness conditions. Given that chronic illnesses during adolescence are managed within a changing parent-child relationship, understanding adherence in this context was important (Pritlove et al., 2020; Williams-Reade et al., 2019). This allowed themes that had previously been overlooked in reviews which focussed solely on the adolescent experience to be identified.

The decision to synthesise studies across different contexts and chronic illness diagnoses highlighted that the broad challenges faced by adolescents and parents appear to be transdiagnostic. This decision meant that diagnoses and populations that are under-researched, including IBD (Hommel et al., 2010) and African Americans (Auslander et al., 2010), could be included. However, some would argue that themes identified in one

context may not be transferrable to others (e.g., Britten et al., 2002). Therefore, the validity of each transfer was questioned, and it was highlighted when differences appeared (Thomas & Harden, 2008).

It is important to note that whilst the review synthesised experiences of parents and adolescents managing six different chronic illnesses, most studies were conducted in English-speaking contexts, focussed on adolescents with Diabetes and mothers over fathers. Whilst this is a limitation of this body of research, a limitation of the review is that the findings may not be transferable to other contexts. Further, the focus of the current review was on the parent-child relationship when managing adherence to treatment, so the experiences of health-care professionals and other family members were not explored.

Finally, the experience of the first author when completing the synthesis needs acknowledging. They were conducting a related meta-analysis into the efficacy of psychological interventions to promote adherence in adolescents with chronic illnesses. This could mean that they had pre-conceived ideas about the experience of adolescents and parents when managing adherence to treatment and thus focussed heavily on some whilst overlooking other data. However, the inclusion of independent coders, with different experiences in this area, contributed to the overall rigour of findings. The first author acknowledging and reflecting on their experience also allowed them to remain inductive.

Clinical Implications

The review highlights clear implications for clinical practice and interventions to promote adherence in this population. First, health-care professionals should be aware of the findings outlined and the broad challenges identified by the parent-child dyad across multiple chronic illnesses. It suggests that adherence-promoting interventions for adolescents should always include parents.

As highlighted previously (Santer et al., 2014), it is important that health-care professionals support families to preserve 'normal life' and adapt treatment regimens to minimise the impact on relationships and other activities. Second, health-care professionals should be mindful of both the parent and adolescent perspective and relational issues when encouraging the use of organisational tools, such as alarms, to support the adolescent in remembering self-management tasks. It would be important to include both parents and adolescents in the use of organisational tools in order to maintain an open dialogue, reduce parental anxiety and adolescent frustration.

As highlighted previously (Hanghøj & Boisen, 2013), education about the effectiveness of treatments and how they work should be provided to adolescents. However, this review found that many parents often do not understand how treatments work and can sometimes doubt their effectiveness. This is important for health-care professionals to be aware of, because if parents do not believe treatments are effective then this may be a barrier to adolescent adherence.

Further, the findings highlighted that peer support interventions are not always well received by adolescents. Therefore, before offering peer support interventions, health-care professionals need to be aware of individual differences. However, it was noted that parents appeared to find peer support useful, and so interventions offered to parents, where possible, should be considered.

The current review found that despite parents and adolescents finding shared management the best approach, some shared that health-care professionals have different expectations about parents' role and involvement which can lead to feelings of blame. Therefore, health-care professionals should have an open, non-blaming and empathetic discussion with parents and adolescents about roles and responsibility in adherence. This is supported by a recent review (Patel et al., 2018).

Finally, the review highlighted different experiences of the consequences of poor adherence across the different illnesses included. Whilst for some this motivated adherence, for others this led to a sense of hopelessness and subsequent poor adherence. It is important that professionals supporting these adolescents are aware of these differences.

Research Implications

First, this review highlighted the importance of including both parental and adolescent perspectives when exploring adherence to treatment in chronic illnesses. Therefore, studies should continue to consider multiple perspectives when exploring this issue. The current review did not consider health-care professionals' perspectives (who are part of the therapeutic 'triad' of adherence, De Civita & Dobkin, 2004; Santer et al., 2014) nor extended family member's experience (such as siblings), which would be a useful avenue of exploration for future research.

Further, research should aim to explore the experiences of adolescents and parents when managing adherence to other chronic illnesses. Although the review covered a broad range of chronic illnesses, other commonly diagnosed illnesses were not represented. The experience of parents, particularly fathers, from non-English speaking contexts is also an important area for further exploration.

Finally, the use of the CASP in the current review highlighted key issues with the current literature particularly in relation to researcher reflexivity and the rigor of analyses, so future qualitative studies should aim to address the issues identified.

Conclusion

The aim of this review was to thematically synthesise qualitative studies exploring the adolescent-parent dyadic experience when managing adherence to treatment in chronic illness. The synthesis of nine qualitative studies across six different chronic illnesses led to the identification of five reoccurring 'analytic' themes. Overall, the review demonstrated

that whilst trying to manage adherence and preserve ‘normal’ life within a social context, adolescents and parents have different priorities and beliefs about treatment and adherence. Forgetting is a key barrier to adherence, which has consequences for the parent-child relationship. This is alongside the dyad experiencing transitions around their roles and responsibility in relation to adherence. Aspects of the relationship itself including trust, negotiation and collaboration are identified as key in enabling parents and adolescents navigate the complexity. This suggests that adolescent care should include parents and future research should continue to include multiple perspectives when exploring these issues, whilst ensuring quality in their methods.

Declaration of Interest Statement

No conflict of interest is reported.

References

Auslander, W. F., Sterzing, P. R., Zayas, L. E., & White, N. H. (2010). Psychosocial resources and barriers to self-management in African American adolescents with type 2 diabetes. *The Diabetes Educator, 36(4)*, 613-622. https://doi.org/10.1177/0145721710369706

Becker, M. H., & Maimam, L. A. (1975). Sociobehavioral determinants of compliance with health and medical care recommendations. *Medical Care, 13(1)*, 10-24. https://www.jstor.org/stable/3763271

Britten, N., Campbell, R., Pope, C., Donovan, J., Morgan, M., & Pill, R. (2002). Using meta ethnography to synthesise qualitative research: A worked example. *Journal of Health Services Research and Policy*, *7*(4), 209–215. https://doi.org/10.1258/135581902320432732

Cooke, A., Smith, D., & Booth, A. (2012). Beyond PICO: The SPIDER tool for qualitative evidence synthesis. *Qualitative Health Research 22(10)*, 1435-1443. https://doi.org/10.1177/1049732312452938

Critical Appraisal Skills Programme (2018). CASP Qualitative Checklist. [online]. https://casp-uk.net/wp-content/uploads/2018/03/CASP-Qualitative-Checklist-2018_fillable_form.pdf Accessed: 19th October 2020.

De Civita, M., & Dobkin, P. L. (2004). Pediatric adherence as a multidimensional and dynamic construct, involving a triadic partnership. *Journal of Pediatric Psychology, 29(3)*, 157-169. https://doi.org/10.1093/jpepsy/jsh018

Denison, J. A., Banda, H., Dennis, A. C., Packer, C., Nyambe, N., Stalter, R. M., Mwansa, J. K., Katayamoyo, P., & McCarraher, D. R. (2015). "The sky is the limit":

Adhering to antiretroviral therapy and HIV self-management from the perspectives of adolescents living with HIV and their adult caregivers. *Journal of the International AIDS Society, 18(19358)*, 1-6. http://dx.doi.org/10.7448/IAS.18.1.19358

DiMatteo, M. R. (2004). Variations in patients' adherence to medical recommendations: a quantitative review of 50 years of research. *Medical care*, 200-209. https://www.jstor.org/stable/4640729

Fishbein, M., & Ajzen, I. (1975). *Belief, attitude, intention, and behavior: An introduction to theory and research.* Addison-Wesley.

Grey, M., Schulman-Green, D., Knafl, K., & Reynolds, N. R. (2015). A revised self-and family management framework. *Nursing Outlook, 63(2)*, 162-170. https://doi.org/10.1016/j.outlook.2014.10.003

Hagell, A., Coleman, J., & Brooks, F. (2015). *Key Data on Adolescence*. Association for Young People's Health. http://www.youngpeopleshealth.org.uk/wp-content/uploads/2015/09/KeyData2015.pdf

Hanghøj, S., & Boisen, K. A. (2013). Self-reported barriers to medication adherence among chronically ill adolescents: A systematic review. *Journal of Adolescent Health, 54(2)*, 121-138. https://doi.org/10.1016/j.jadohealth.2013.08.009

Hanna, K. M., & Guthrie, D. (2001). Parents' and adolescents' perceptions of helpful and nonhelpful support for adolescents' assumption of diabetes management responsibility. *Issues in Comprehensive Pediatric Nursing, 24(4)*, 209-223. https://doi.org/10.1080/014608601753260317

Heath, G., Farre, A., & Shaw, K. (2017). Parenting a child with chronic illness as they transition into adulthood: a systematic review and thematic synthesis of parents'

experiences. *Patient education and counselling, 100(1)*, 76-92. https://doi.org/10.1016/j.pec.2016.08.011

Hendry, A., Snowden, A., & Brown, M. (2017). When holistic care is not holistic enough: The role of sexual health in mental health settings. *Journal of Clinical Nursing, 27(5-6)*, 1015-1027. https://doi.org/10.1111/jocn.14085

Heyduck, K., Bengel, J., Farin-Glattacker, E., & Glattacker, M. (2015). Adolescent and parental perceptions about asthma and asthma management: A dyadic qualitative analysis. *Child: Care, Health and Development, 41(6)*, 1227-1237. https://doi.org/10.1111/cch.12277

Hommel, K. A., Odell, S., Sander, E., Baldassano, R. N., & Barg, F. K. (2010). Treatment adherence in paediatric inflammatory bowel disease: perceptions from adolescent patients and their families. *Health & Social Care in the Community, 19(1)*, 80-88. https://doi.org/10.1111/j.1365-2524.2010.00951.x

Ivey, J. B., Wright, A., & Dashiff, C. J. (2007). Finding the balance: Adolescents with type 1 diabetes and their parents. *Journal of Pediatric Health Care, 23(1)*, 10-18. https://doi.org/10.1016/j.pedhc.2007.12.008

Kourrouski, M. F., & Lima, R. A. (2009). Treatment adherence: The experience of adolescents with HIV/AIDS. *Revista latino-americana de enfermagem, 17(6)*, 947-952. http://dx.doi.org/10.1590/S0104-11692009000600004

Law, E., Fisher, E., Eccleston, C., & Palermo, T. M. (2019). Psychological interventions for parents of children and adolescents with chronic illness. *Cochrane Database of Systematic Reviews, 3*. https://doi.org/10.1002/14651858.CD009660

Lerch, M. F., & Thrane, S. E. (2019). Adolescents with chronic illness and the transition to self-management: A systematic review. *Journal of Adolescence, 72*, 152-161. https://doi.org/10.1016/j.adolescence.2019.02.010

Liberati, A., Altman, D. G., Tetzlaff, j., Mulrow, C., Gøtzsche, P. C., Ioannidis, J. P., Clarke, J. P., Devereaux, P. J., Kleijnen, P., & Moher, D. (2009). The PRISMA Statement for Reporting Systematic Reviews and Meta-Analyses of Studies That Evaluate Health Care Interventions: Explanation and Elaboration. *Annuals of Internal Medicine, 151(4)*, 65-94. https://doi.org/10.1016/j.jclinepi.2009.06.006

Lindsay, S., Kingsnorth, S., & Hamdani, Y. (2011). Barriers and facilitators of chronic illness self-management among adolescents: a review and future directions. *Journal of Nursing and Healthcare of Chronic Illness, 3(3)*, 186-208. https://doi.org/10.1111/j.1752-9824.2011.01090.x

Long, H. A., French, D. P., & Brooks, J. M. (2020). Optimising the value of the critical appraisal skills programme (CASP) tool for quality appraisal in qualitative evidence synthesis. *Research Methods in Medicine & Health Sciences, 1(1)*, 31-42. https://doi.org/10.1177/2632084320947559

Luo, J., Guo, J., Yang, J., Ou, X., & Grey, M. (2020). Parent–Child Relationship Quality as a Mediator of the Association between Perceived Stress and Diabetes Self-Management in Adolescents With Type 1 Diabetes. *Journal of Family Nursing*, 1-10. https://doi.org/10.1177/1074840720971584

McCann, E., Lee, R., & Brown, M. (2016). The experiences and support needs of people with intellectual disabilities who identify as LGBT: A review of the literature. *Research in Developmental Disabilities, 57*, 39-53. https://doi.org/10.1016/j.ridd.2016.06.013

McEvoy, P., & Richards, D. (2006). A critical realist rationale for using a combination of quantitative and qualitative methods. *Journal of Research in Nursing, 11(1)*, 66-78. https://doi.org/10.1177/1744987106060192

Modi, A. C., & Driscoll, K. A. (2020). *Adherence and Self-Management in Pediatric Populations.* Elsevier Science & Technology.

Modi, A. C., Pai, A. L., Hommel, K. A., Hood, K. K., Cortina, S., Hilliard, M. E., Guilfoyle, S. M., Gray, W. N., & Drotar, D. (2012). Pediatric self-management: a framework for research, practice, and policy. *Pediatrics*, *129*(2), e473-e485. https://doi.org/10.1542/peds.2011-1635

Moher, D., Liberati, A., Tetzlaff, J., Altman, D. G., & The PRISMA Group. (2009). Preferred reporting items for systematic reviews and meta-analyses: The PRISMA statement. *PLoS Medicine, 6(7)*, e1000097. https://doi.org/10.1136/bmj.b2535

O'Toole, D. P., Latchford, G. J., Duff, A. J., Ball, R., McCormack, P., McNamara, P. S., Browniee, K. G., & Southern, K. W. (2019). Adherence to aerosol therapy in young people with cystic fibrosis: Patient and parent perspectives following electronic data capture. *Qualitative Health Research, 29(6)*, 846-856. https://doi.org/10.1177/1049732318805754

Patel, N. J., Datye, K. A., & Jaser, S. S. (2018). Importance of patient–provider communication to adherence in adolescents with type 1 diabetes. *Healthcare* 6(30), 1-11. https://doi.org/10.3390/healthcare6020030

Pritlove, C., Markowitz, B., Mukerji, G., Advani, A., & Parsons, J. A. (2020). Experiences and perspectives of the parents of emerging adults living with type 1 diabetes. *BMJ Open Diabetes Research and Care, 8(1)*, e001125. http://dx.doi.org/10.1136/bmjdrc-2019-001125

Rapoff, M. (2010). *Adherence to pediatric medical regimens.* Springer.

Rushbrooke, E., Murray, C. D., & Townsend, S. (2014). What difficulties are experienced by caregivers in relation to the sexuality of people with intellectual disabilities? A

qualitative meta-synthesis. *Research in Developmental Disabilities, 35(4)*, 871-886. https://doi.org/10.1016/j.ridd.2014.01.012

Ryan, P., & Sawin, K. J. (2009). The individual and family self-management theory: Background and perspectives on context, process, and outcomes. *Nursing Outlook, 22(10)*, 217-225. https://doi.org/10.1016/j.outlook.2008.10.004

Sanders R. A. (2013). Adolescent psychosocial, social, and cognitive development. *Pediatrics in review*, *34*(8), 354–359. https://doi.org/10.1542/pir.34-8-354

Santer, M., Ring, N., Yardley, L., Geraghty, A. W., & Wyke, S. (2014). Treatment non-adherence in pediatric long-term medical conditions: systematic review and synthesis of qualitative studies of caregivers' views. *BMC Pediatrics, 14(63)*, 1-10. https://doi.org/10.1186/1471-2431-14-63

Sullivan-Bolyai, S., Boya, C., Johnson, K., Cullen, K., Jaffarian, C., Quinn, D., Aroke, E. N., Crawford, S., Lee, M. M., & Gupta, O. (2014). Engaging teens and parents in collaborative practice: perspectives on diabetes self-management. *The Diabetes Educator, 40(2)*, 178-190. https://doi.org/10.1177/0145721713520568

Thomas, J., & Harden, A. (2008). Methods for the thematic synthesis of qualitative research in systematic reviews. *BMC Medical Research Methodology, 8(45)*, 1-10. https://doi.org/10.1186/1471-2288-8-45

Tong, A., Flemming, K., McInnes, E., Oliver, S., & Craig, J. (2012). Enhancing transparency in reporting the synthesis of qualitative research: ENTREQ . *BMC Medical Research Methodology, 12(181)*, 1-8. https://doi.org/10.1186/1471-2288-12-181

van der Lee, J. H., Mokkink, L. B., Grootenhuis, M. A., Heymans, H. S., & Offringa, M. (2007). Definitions and Measurement of Chronic Health Conditions in Childhood

A Systematic Review. *JAMA, 297(24)*, 2741-2751. https://doi.org/10.1001/jama.297.24.2741

Viner, R., & Christie, D. (2005). Adolescent development. *BMJ, 330(7486)*, 301-304. https://doi.org/10.1136/bmj.330.7486.301

Williams-Reade, J. M., Tapanes, D., Distelberg, B. J., & Montgomery, S. (2019). pediatric chronic illness management: A qualitative dyadic analysis of adolescent patient and parent illness narratives. *Journal of marital and family therapy*, *46*(1), 135-148. https://doi.org/10.1111/jmft.12377

World Health Organisation. (n.d.). *Adolescent Health*. https://www.who.int/health-topics/adolescent-health/#tab=tab_1

Supplementary Material

References of Included Studies

Auslander, W. F., Sterzing, P. R., Zayas, L. E., & White, N. H. (2010). Psychosocial resources and barriers to self-management in African American adolescents with type 2 diabetes. *The Diabetes Educator, 36(4)*, 613-622. https://doi.org/10.1177/0145721710369706

Denison, J. A., Banda, H., Dennis, A. C., Packer, C., Nyambe, N., Stalter, R. M., Mwansa, J. K., Katayamoyo, P., & McCarraher, D. R. (2015). "The sky is the limit": Adhering to antiretroviral therapy and HIV self-management from the perspectives of adolescents living with HIV and their adult caregivers. *Journal of the International AIDS Society, 18(19358)*, 1-6. http://dx.doi.org/10.7448/IAS.18.1.19358

Hanna, K. M., & Guthrie, D. (2001). Parents' and adolescents' perceptions of helpful and nonhelpful support for adolescents' assumption of diabetes management responsibility. *Issues in Comprehensive Pediatric Nursing, 24(4)*, 209-223. https://doi.org/10.1080/014608601753260317

Heyduck, K., Bengel, J., Farin-Glattacker, E., & Glattacker, M. (2015). Adolescent and parental perceptions about asthma and asthma management: A dyadic qualitative analysis. *Child: Care, Health and Development, 41(6)*, 1227-1237. https://doi.org/10.1111/cch.12277

Hommel, K. A., Odell, S., Sander, E., Baldassano, R. N., & Barg, F. K. (2010). Treatment adherence in paediatric inflammatory bowel disease: perceptions from adolescent patients and their families. *Health & Social Care in the Community, 19(1)*, 80-88. https://doi.org/10.1111/j.1365-2524.2010.00951.x

Ivey, J. B., Wright, A., & Dashiff, C. J. (2007). Finding the balance: Adolescents with type 1 diabetes and their parents. *Journal of Pediatric Health Care, 23(1)*, 10-18. https://doi.org/10.1016/j.pedhc.2007.12.008

Kourrouski, M. F., & Lima, R. A. (2009). Treatment adherence: The experience of adolescents with HIV/AIDS. *Revista latino-americana de enfermagem, 17(6)*, 947-952. http://dx.doi.org/10.1590/S0104-11692009000600004

O'Toole, D. P., Latchford, G. J., Duff, A. J., Ball, R., McCormack, P., McNamara, P. S., Browniee, K. G., & Southern, K. W. (2019). Adherence to aerosol therapy in young people with cystic fibrosis: Patient and parent perspectives following electronic data capture. *Qualitative Health Research, 29(6)*, 846-856. https://doi.org/10.1177/1049732318805754

Sullivan-Bolyai, S., Boya, C., Johnson, K., Cullen, K., Jaffarian, C., Quinn, D., Aroke, E. N., Crawford, S., Lee, M. M., & Gupta, O. (2014). Engaging teens and parents in collaborative practice: perspectives on diabetes self-management. *The Diabetes Educator, 40(2)*, 178-190.

Supplementary Material Continued: PRISMA Statement (Moher et al., 2009)

From: Moher D, Liberati A, Tetzlaff J, Altman DG, The PRISMA Group (2009). Preferred Reporting Items for Systematic Reviews and Meta-Analyses: The PRISMA Statement. PLoS Med 6(7): e1000097. doi:10.1371/journal.pmed1000097
For more information, visit: **www.prisma-statement.org**.

This is under the Creative Commons Attribution License and has been appropriately cited and adapted for use in the present review.

Section/topic	#	Checklist item	Reported on page #
TITLE			
Title	1	Identify the report as a systematic review, meta-analysis, or both.	21
ABSTRACT			
Structured summary	2	Provide a structured summary including, as applicable: background; objectives; data sources; study eligibility criteria, participants, and interventions; study appraisal and synthesis methods; results; limitations; conclusions and implications of key findings; systematic review registration number.	22
INTRODUCTION			
Rationale	3	Describe the rationale for the review in the context of what is already known.	23-25
Objectives	4	Provide an explicit statement of questions being addressed with reference to participants, interventions, comparisons, outcomes, and study design (PICOS).	25-26 SPIDER used as an alternative to PICO
METHODS			

Protocol and registration	5	Indicate if a review protocol exists, if and where it can be accessed (e.g., Web address), and, if available, provide registration information including registration number.	26
Eligibility criteria	6	Specify study characteristics (e.g., PICOS, length of follow-up) and report characteristics (e.g., years considered, language, publication status) used as criteria for eligibility, giving rationale.	28-30
Information sources	7	Describe all information sources (e.g., databases with dates of coverage, contact with study authors to identify additional studies) in the search and date last searched.	26-27
Search	8	Present full electronic search strategy for at least one database, including any limits used, such that it could be repeated.	206-207
Study selection	9	State the process for selecting studies (i.e., screening, eligibility, included in systematic review, and, if applicable, included in the meta-analysis).	30
Data collection process	10	Describe method of data extraction from reports (e.g., piloted forms, independently, in duplicate) and any processes for obtaining and confirming data from investigators.	31
Data items	11	List and define all variables for which data were sought (e.g., PICOS, funding sources) and any assumptions and simplifications made.	31
Risk of bias in individual studies	12	Describe methods used for assessing risk of bias of individual studies (including specification of whether this was done at the study or outcome level), and how this information is to be used in any data synthesis.	30-31
Summary measures	13	State the principal summary measures (e.g., risk ratio, difference in means).	N/A
Synthesis of results	14	Describe the methods of handling data and combining results of studies, if done, including measures of consistency (e.g., I^2) for each meta-analysis.	31-32

Section/topic	#	Checklist item	Reported on page #
Risk of bias across studies	15	Specify any assessment of risk of bias that may affect the cumulative evidence (e.g., publication bias, selective reporting within studies).	30-31
Additional analyses	16	Describe methods of additional analyses (e.g., sensitivity or subgroup analyses, meta-regression), if done, indicating which were pre-specified.	N/A
RESULTS			
Study selection	17	Give numbers of studies screened, assessed for eligibility, and included in the review, with reasons for exclusions at each stage, ideally with a flow diagram.	32-33
Study characteristics	18	For each study, present characteristics for which data were extracted (e.g., study size, PICOS, follow-up period) and provide the citations.	36-47
Risk of bias within studies	19	Present data on risk of bias of each study and, if available, any outcome level assessment (see item 12).	36-47 and 208-209
Results of individual studies	20	For all outcomes considered (benefits or harms), present, for each study: (a) simple summary data for each intervention group (b) effect estimates and confidence intervals, ideally with a forest plot.	N/A
Synthesis of results	21	Present results of each meta-analysis done, including confidence intervals and measures of consistency.	N/A
Risk of bias across studies	22	Present results of any assessment of risk of bias across studies (see Item 15).	36-47 and 208-209
Additional analysis	23	Give results of additional analyses, if done (e.g., sensitivity or subgroup analyses, meta-regression [see Item 16]).	N/A
DISCUSSION			

Summary of evidence	24	Summarize the main findings including the strength of evidence for each main outcome; consider their relevance to key groups (e.g., healthcare providers, users, and policy makers).	59-64
Limitations	25	Discuss limitations at study and outcome level (e.g., risk of bias), and at review-level (e.g., incomplete retrieval of identified research, reporting bias).	61-62
Conclusions	26	Provide a general interpretation of the results in the context of other evidence, and implications for future research.	59-61
FUNDING			
Funding	27	Describe sources of funding for the systematic review and other support (e.g., supply of data); role of funders for the systematic review.	21

Supplementary Material Continued: ENTREQ (Tong et al., 2012)

From: Tong, A., Flemming, K., McInnes, E., Oliver, S., & Craig, J. (2012). Enhancing transparency in reporting the synthesis of qualitative research: ENTREQ . *BMC Medical Research Methodology, 12(181)*, 1-8. https://doi.org/10.1186/1471-2288-12-181

This is under the Creative Commons Attribution License and has been appropriately cited and adapted for use in the present review.

Tong *et al. BMC Medical Research Methodology* 2012, **12**:181
http://www.biomedcentral.com/1471-2288/12/181

Table 1 Enhancing transparency in reporting the synthesis of qualitative research: the ENTREQ statement

No	Item	Guide and description	Page no. In manuscript
1	Aim	State the research question the synthesis addresses.	26
2	Synthesis methodology	Identify the synthesis methodology or theoretical framework which underpins the synthesis, and describe the rationale for choice of methodology *(e.g. meta-ethnography, thematic synthesis, critical interpretive synthesis, grounded theory synthesis, realist synthesis, meta-aggregation, meta-study, framework synthesis).*	26
3	Approach to searching	Indicate whether the search was pre-planned (*comprehensive search strategies to seek all available studies)* or iterative (*to seek all available concepts until they theoretical saturation is achieved).*	27
4	Inclusion criteria	Specify the inclusion/exclusion criteria *(e.g. in terms of population, language, year limits, type of publication, study type).*	28-30
5	Data sources	Describe the information sources used (e.g. *electronic databases (MEDLINE, EMBASE, CINAHL, psycINFO, Econlit), grey literature databases (digital thesis, policy reports), relevant organisational websites, experts, information specialists, generic web searches (Google Scholar) hand searching, reference lists)* and when the searches conducted; provide the rationale for using the data sources.	26-27
6	Electronic Search strategy	Describe the literature search *(e.g. provide electronic search strategies with population terms, clinical or health topic terms, experiential or social phenomena related terms, filters for qualitative research, and search limits).*	27-28
7	Study screening methods	Describe the process of study screening and sifting *(e.g. title, abstract and full text review, number of independent reviewers who screened studies).*	30
8	Study characteristics	Present the characteristics of the included studies *(e.g. year of publication, country, population, number of participants, data collection, methodology, analysis, research questions).*	36-47
9	Study selection results	Identify the number of studies screened and provide reasons for study exclusion *(e,g, for comprehensive searching, provide numbers of studies screened and reasons for exclusion indicated in a figure/flowchart; for iterative searching describe reasons for study exclusion and inclusion based on modifications t the research question and/or contribution to theory development).*	32-33
10	Rationale for appraisal	Describe the rationale and approach used to appraise the included studies or selected findings *(e.g. assessment of conduct (validity and robustness), assessment of reporting (transparency), assessment of content and utility of the findings).*	30-31
11	Appraisal items	State the tools, frameworks and criteria used to appraise the studies or selected findings *(e.g. Existing tools: CASP, QARI, COREQ, Mays and Pope* [25]*; reviewer developed tools; describe the domains assessed: research team, study design, data analysis and interpretations, reporting).*	30-31
12	Appraisal process	Indicate whether the appraisal was conducted independently by more than one reviewer and if consensus was required.	31
13	Appraisal results	Present results of the quality assessment and indicate which articles, if any, were weighted/excluded based on the assessment and give the rationale.	31
14	Data extraction	Indicate which sections of the primary studies were analysed and how were the data extracted from the primary studies? *(e.g. all text under the headings "results /conclusions" were extracted electronically and entered into a computer software).*	31-32
15	Software	State the computer software used, if any.	31
16	Number of reviewers	Identify who was involved in coding and analysis.	31-32
17	Coding	Describe the process for coding of data *(e.g. line by line coding to search for concepts).*	31-32
18	Study comparison	Describe how were comparisons made within and across studies *(e.g. subsequent studies were coded into pre-existing concepts, and new concepts were created when deemed necessary).*	31
19	Derivation of themes	Explain whether the process of deriving the themes or constructs was inductive or deductive.	31
20	Quotations	Provide quotations from the primary studies to illustrate themes/constructs, and identify whether the quotations were participant quotations of the author's interpretation.	48-59
21	Synthesis output	Present rich, compelling and useful results that go beyond a summary of the primary studies (e.g. *new interpretation, models of evidence, conceptual models, analytical framework, development of a new theory or construct).*	48-59

Chapter Three: Bridging Chapter

The qualitative systematic review reported in chapter two outlined key issues that should be considered in the development of psychological interventions to promote adherence in adolescents with chronic illness. More specifically, it highlighted that both the parent and adolescent experiences of managing adherence to treatment are important. This suggests when designing interventions, researchers and clinicians should consider the perspectives of both the adolescent and their parents and relational issues between them. These findings are consistent with contemporary theories of pediatric self-management which suggest that interventions need to account for the relational interactions (e.g., Modi et al., 2012).

The meta-analysis presented in the next chapter examines the efficacy of psychological interventions in promoting adherence and improving quality of life and family functioning outcomes in adolescents with chronic illness. Some of the interventions included in the meta-analysis involve parents, whilst others are aimed at only the adolescent. The involvement of parents in these interventions was included as a moderator of intervention efficacy in the meta-analysis.

The results of the primary analysis which examined the efficacy of psychological interventions to promote adherence at posttreatment is presented in the main paper in the next chapter. The results of the secondary analyses, which examined the effects of adherence at follow-up and quality of life and family functioning outcomes at posttreatment and follow-up are also presented in the main paper. Exploratory analyses which examined the effects of different adherence measures at both posttreatment and follow-up are presented in an additional results chapter in chapter six. Sensitivity, moderator and subgroup analyses were performed where possible. The results of which are also reported.

Chapter Four

Psychological Interventions to Promote Adherence to Treatment in Adolescents Living with Chronic Illness: A Systematic Review and Meta-Analysis

Kelsey Odgers[1], Dr Amy Carroll[1], Kiki Mastroyannopolou[1], Araminta Peters-Corbett[2] and Dr Joanne Hodgekins[1]

[1]Department of Clinical Psychology and Psychological Therapies, University of East Anglia, UK.

[2]Norfolk and Suffolk NHS Foundation Trust

Prepared for: Health Psychology Review (Author guidelines in Appendix A)

Word count excluding tables/figures: 6,974

Word count including tables/figures: 10,460

Author Note

This research has been conducted as part of the Clinical Psychology Doctorate course at the UEA. The first author would like to thank Matthew Smith from the UEA for his support in the development of the search. They would also like to thank Jo Spauls for checking the inclusion of papers and quality assessment. Finally, they would like to thank Richard Meiser-Stedman for his teaching and support in conducting meta-analyses.

Correspondence concerning this article should be addressed to Kelsey Odgers, Norwich Medical School, University of East Anglia, Norwich, Norfolk, NR4 7TJ, UK. Email: K.Odgers@uea.ac.uk

Abstract

Background: Adolescents with chronic illnesses have higher rates of non-adherence compared to children and adults. This is due to the complexity of managing chronic illnesses during an already challenging period. No meta-analysis examining the efficacy of adherence-promoting psychological interventions in adolescents specifically exists.

Objective: To examine the efficacy of psychological interventions in promoting adherence and improving quality of life and family functioning in adolescents with chronic illness.

Method: A systematic review and meta-analysis of randomised controlled trials examining the efficacy of psychological interventions for adolescents with chronic illness was conducted. CINAHL, MEDLINE, and PsycINFO were systematically searched from 2007 until 2020. Risk of bias assessments were undertaken.

Results: Thirty-six studies met inclusion criteria. The exclusion of high risk of bias studies resulted in significant small effects on adherence outcomes ($g = 0.30$) and quality of life outcomes ($g = 0.14$) at posttreatment, with low heterogeneity. Follow-up, moderation and subgroup analyses were limited by the number of studies.

Conclusion: The findings suggest that psychological interventions for improving adherence in adolescents with chronic illness have limited efficacy. Future high-quality research recruiting adolescent samples with poor adherence is needed.

Keywords: adherence; chronic illness; adolescents; interventions; meta-analysis

Introduction

Increasing numbers of adolescents are living with a chronic illness which significantly impacts their daily lives (Hanghøj & Boisen, 2013; Heath et al., 2017). Chronic illnesses are those with a duration of more than three months which requires some form of on-going self-management (Heath et al., 2017; Law et al., 2019; van der Lee et al., 2007). Suris et al. (2004) estimated that 10% of adolescents worldwide suffer with a chronic illness and Hagell et al. (2015) reported that in the UK there are increasing numbers of adolescents diagnosed with a chronic illness. Due to advances in treatment, increasing numbers of these adolescents will reach adulthood (Heath et al., 2017), but living with these illnesses and managing them often requires complex daily behaviours (Hanghøj & Boisen, 2013; Modi & Driscoll, 2020).

Adherence is defined as "the extent to which a person's behaviour – taking medication, following a diet, and/or executing lifestyle changes, corresponds with agreed recommendations from a healthcare provider" (World Health Organisation, 2003, p.3). Adherence is vital in reducing healthcare utilisation and costs, decreasing the risk of morbidity and mortality and improving health outcomes (Kahana et al., 2008; Modi & Driscoll, 2020; Pai & McGrady, 2014). Despite this, adhering to treatment regimens is problematic for every patient group. However, it seems to be particularly problematic in adolescent samples (DiMatteo, 2004; Hanghøj & Boisen, 2013; Rapoff, 2010). Rapoff (2010) reported that rates of non-adherence average 50% in paediatric samples, with rates in adolescent samples specifically being as high as 75%. This compares to lower rates of non-adherence in adult populations, which are between 30% and 50% (Peng et al., 2020).

Due to the high rates of treatment non-adherence in young people with chronic illness, many psychological interventions have been developed and evaluated which aim to promote adherence in these populations (Drotar, 2000; Kahana et al., 2008; Modi & Driscoll, 2020).

Several meta-analyses examining the efficacy of these interventions to promote adherence in children and young people with chronic illness have been conducted. The first systematic reviews and meta-analyses in the field were conducted by Graves et al. (2010) and Kahana et al. (2008). Overall, they found that psychological interventions can effectively promote adherence in paediatric samples and result in positive health outcomes. These reviews highlighted the significant amount of heterogeneity in the reporting of the first adherence-promoting intervention research and argued that further transparency in reporting and standardisation was needed. An overall limitation of these early reviews is that only a small proportion of included studies were randomised controlled trials (RCTs).

The most recent systematic review and meta-analysis was conducted by Pai and McGrady (2014). They were more rigorous in their methods, as they included only RCTs. They found that studies published between 2007 and 2013 showed heterogeneous and relatively small effect sizes at posttreatment ($d = 0.20$) and follow up ($d = 0.29$). Quality assessments also suggested that there was great need for improvement. Pai and McGrady (2014) were unable to examine whether developmentally specific interventions, such as those delivered to adolescents only, produced larger treatment effects, because the majority of included studies included mixed child and adolescent samples.

Adolescents seem to have more difficulties with adhering to treatment regimens than children and adults (Hanghøj & Boisen, 2013; Rapoff, 2010). It has been suggested that poor adherence in this population results from the challenging nature and complexity of managing chronic illnesses coupled with an already challenging developmental phase where individuals experience considerable physical, social and psychological changes (Yeo & Sawyer, 2005; Viner & Christie, 2005). Adolescents with chronic illnesses often begin and are expected to have to be more autonomous in the management of their illness, as parents begin to have less involvement. Given the additional burden that managing

complex illnesses brings, adolescents may struggle with the transition from parental to self-management, particularly when such illnesses or their treatments may make them feel different from their peers (Lerch & Tharne, 2019; Yeo & Sawyer, 2005). Therefore, it is important to examine adherence-promoting interventions aimed at adolescents with chronic illness specifically, which has not been thoroughly examined to date.

Since the publication Pai and McGrady's (2014) meta-analysis, the adherence-promoting research field has seen significant growth and an increased rigour in methods (Modi & Driscoll, 2020). Due to advances in technology the research methods employed by adherence-promoting researchers has changed in recent years. Both the way that interventions are delivered, and the way adherence is measured has changed, as increasingly interventions are delivered via technology (e.g., web-based delivery compared with face-to-face methods) and studies use more rigorous and objective measures of adherence, such as electronic monitoring (Modi & Driscoll, 2020). Further, studies are recruiting narrow age ranges in their samples and now many RCTs recruiting adolescents only exist (e.g., Ellis et al., 2019; Kosse et al., 2019). Therefore, an updated systematic review and meta-analysis in this area, focussed specifically on adolescents, is needed.

The first aim of the current systematic review and meta-analysis is to address these aforementioned gaps by examining the efficacy of interventions, published within the last 14 years, aimed at promoting treatment adherence in adolescents with chronic illness.

Further, previous meta-analyses of adherence-promoting interventions in children and young people with chronic illness have focused solely on adherence and health outcomes (Graves et al., 2010; Pai & McGrady, 2014). However, adherence has been linked to other outcomes including health related quality of life and family functioning outcomes (Fredericks et al., 2008; Psihogios et al., 2019). Therefore, the secondary aim of

the current meta-analysis and systematic review is to examine whether interventions aimed at promoting treatment adherence in adolescents also improve adolescent quality of life and family functioning outcomes.

Research Questions

The research question was developed using the PICOS method (Richardson et al., 1995) and this method assisted in identifying the key search terms needed to answer the question. These were:

Patients and their problems: Adolescents (10–19 years) with chronic illness.

Intervention: Psychological intervention aimed at improving treatment adherence.

Comparison: Control groups.

Outcomes: Treatment adherence.

Study design: Randomised controlled trials.

The primary research question is ‘what is the efficacy of psychological interventions to promote adherence to treatment in adolescents with chronic illness?’

The secondary questions are 1) ‘Do psychological interventions aimed at promoting treatment adherence in adolescents with chronic illness improve adolescent quality of life and family functioning outcomes?’ 2) ‘Do moderator variables including parental involvement, intervention delivery, chronic illness type or sample (i.e., whether adolescents with poor adherence were targeted or not) affect the efficacy of adherence-promoting psychological interventions?’

Methods

Guiding Framework and Registration

This meta-analysis and systematic review were guided by the methodology set out by Khan et al. (2003) and Cuijpers’ (2016). The Preferred Reporting Items for Systematic

Reviews and Meta-Analyses (PRISMA, Moher et al., 2009) were adhered to using the 27-item checklist. See supplementary material for full details.

The review protocol was registered on PROSPERO (Registration number: CRD42020165792).

Search Strategy

The initial pre-planned systematic search was completed on 26th February and searched the literature from January 2007 until 26th February 2020 in three electronic databases: MEDLINE (EBSCO), CINAHL (EBSCO) and PsycINFO (EBSCO). Before implementation the search terms were agreed with the fifth author to ensure to it was robust and rigorous. The search was updated on 2nd November 2020 before the analysis was conducted.

In order to find RCTs, the search used the evidenced-based strategy recommended by Cochrane (Cochrane, 2020) in MEDLINE and PsychINFO and the strategy outlined in Glanville et al. (2019) in CINAHL. The final search terms used can be found in Table 4.1. A copy of the MEDLINE search string has been provided in Appendix D.

Additionally, the reference lists of any relevant reviews, books and included studies were hand searched. Grey material was not included to ensure that studies were subject to peer-review.

Table 4.1

Final Search Terms

Concept One Terms for population	Concept Two Terms for chronic illness	Concept Three Terms for adherence	Concept Four Terms for psychological intervention
Child* OR adolescen* OR youth OR teen* OR "young people" OR infant OR "young adult*"	"Chronic illness*" OR "Chronic disease*" OR epilepsy OR diabetes OR asthma OR "sickle cell disease" OR transplant* OR "spina bifida" OR "cystic fibrosis" OR "human immunodeficiency virus" OR HIV OR arthritis OR "inflammatory bowel disease*" OR obesity OR cancer OR "chronic pain" OR "gastrointestinal disease*" OR "communicable disease"	Adherence OR compliance OR concordance OR non-adherence OR self-management OR "poorly controlled"	Intervention* OR program* OR cognitive OR behaviour* OR behavior* or multisystemic or educat*

Note. MeSH terms were used where available. Free text was used to search titles and abstracts. "Apply related words" and "apply equivalent subjects" were used to increase the inclusiveness of the search. No language limits were put on the search, as the Librarian recommended.

Inclusion and Exclusion Criteria

These criteria are in Table 4.2 and were based on the PICOS acronym.

Table 4.2

Inclusion and Exclusion Criteria

Inclusion Criteria	Exclusion Criteria
Population Adolescents with a chronic illness (Heath et al., 2017; Law et al., 2019; van der Lee et al., 2007), aged between 10 and 19 years (World Health Organisation, n.d.) and/or their parents/caregivers.	Population Individuals who are either younger than 10 years of age or older than 19 years of age or there is a mixed sample of adults/children and adolescents and/or chronically ill and well adolescents. Samples of adolescences with psychological disorders (i.e., depression) or neurodevelopmental disorders (i.e., Autism) only. Samples of adolescents with obesity only.
Intervention A psychological intervention aiming to address adherence and/or self-management. This is defined in the literature as interventions which have a cognitive and/or behavioural, educational, organisational, social or a family component including multisystemic therapy or those which use technology-based approaches (Graves et al., 2010; Kahana et al., 2008; Modi & Driscoll, 2020; Pai & McGrady, 2014).	Intervention There is no intervention used. The intervention is a surgical, drug or dietary intervention only.
Comparison A control group is used to assess the effectiveness of the intervention, including waiting list groups, treatment as usual (TAU), standard care, and attention control groups.	Comparison No control group was used. The control group was another psychological intervention (e.g., comparing face-to-face delivered intervention to the same intervention delivered remotely).
Outcome A measure of adherence or adherence to multiple domains including self-care and/or self-management was included. This includes adherence to medication,	Outcome Adherence or self-management was not measured.

Inclusion Criteria	Exclusion Criteria
dietary, and exercise/behaviour regimens using either patient self-report, parent or other carer self-report, electronic monitors, pill refills or diaries.	
Study Design A randomised controlled trial (RCT) design only where data is available to compute effect sizes. RCTs must include a minimum of 10 participants randomised in each arm.	Study Design Non-RCT designs. RCTs that do not report the data needed to calculate effect sizes will be excluded from the meta-analyses. Less than 10 participants randomised in each arm.
Manuscript Manuscripts which have been written or translated into the English language. Manuscripts that have been peer-reviewed.	Manuscript Manuscripts that have been written in any language other than the English language unless a translated version is available. Manuscripts that have not been peer-reviewed.

Note. Table 4.2 continued.

Study Selection

Studies identified by the searches were extracted into Microsoft Excel. After duplicates were removed, titles and abstracts were screened for eligibility and removed if it was clear that they did not meet criteria. Full-text articles were then read to assess eligibility and, if excluded, each was coded in order to provide the reason for this. Approximately 20% were read and checked with the second author. For inter-rater reliability, a further 25% of full-text articles were checked independently and any disagreements were discussed.

Quality Assessment

The Cochrane Collaboration's risk of bias tool was used to assess study quality (Higgins & Green, 2008). This included the consideration of sequence generation; allocation concealment; blinding of outcome assessors; incomplete outcome data and

selective outcome reporting. The Cochrane risk of bias tool for cluster-randomised designs (Eldridge et al., 2016) was used for cluster RCTs. An overall risk of bias rating was given using the criteria by Higgins et al. (2020). The first author independently conducted risk of bias assessment. For inter-rater reliability, 25% of included articles were checked independently and any disagreements were discussed.

Data Extraction

A standardised form was used to extract data from the included articles. The Cochrane Effective Practice and Organisation of Care (EPOC, 2020) extraction form was consulted. The following categories were included: general information; study setting; sample size and completion rates; diagnoses and demographic information; relevant primary and secondary outcome measures; intervention characteristics; type of control group; immediate posttreatment means, standard deviations and numbers of participants for each relevant outcome; follow-up means, standard deviations and numbers for each relevant outcome (if applicable); and information for assessment of risk of bias. The first author independently conducted the data extraction for each study. For inter-rater reliability, 25% of included articles were extracted independently.

Data Analysis

A narrative synthesis was completed for the systematic review. Following this, meta-analyses were conducted to estimate effect sizes. The MAVIS computer package (version 1.1.3; Hamilton et al., 2017) was used to analyse all study data. The primary analysis explored effects of treatment on adherence outcomes at posttreatment (i.e., the first assessment time point following the end of the intervention). Where data were available, Cohen's d effect sizes were calculated for each study and transformed to Hedge's *g*. Random effects models were used to account for study heterogeneity (Cooper et al., 2009). In line with guidance from Fritz et al. (2012), an effect size of 0.2 was considered to

be small, with 0.5 being a medium effect and 0.8 being large. Sensitivity analyses were used to assess the impact of study bias and cluster-randomised controlled trials.

Heterogeneity was investigated using the I^2 statistic (Higgins et al., 2003). A I^2 statistic of 25% or less was considered low heterogeneity, 50% was considered moderate and above 75% was considered high heterogeneity (Higgins et al., 2003). The *Q*-statistic was also used to assess study heterogeneity; if this was significant ($p < .05$), then it indicated that heterogeneity was higher than that expected by chance.

Moderator and Subgroup Analyses

Several moderator and subgroup analyses were conducted. Using MAVIS, the *Qb* statistic and its significance were used, which indicates the impact of the moderator on the variance across groups. The moderators were 1) sample (identified adolescents with poor adherence versus other); 2) type of chronic illness diagnosis (i.e., Diabetes versus other, Asthma versus other, etc.); 3) parental involvement (adolescent only versus parental involved); 4) intervention delivery (face-to-face delivery versus remote delivery). To improve the reliability of findings, all high risk of bias studies were excluded and only those with at least four studies in each subgroup were conducted. To correct for multiple analyses, the Holm-Bonferroni method (Holm, 1979) was used using the Gaetano (2018) Microsoft Excel calculator.

Secondary Analyses

Secondary analyses explored 1) the effects of adherence outcomes at follow-up; 2) the effects of the quality of life and family outcomes (secondary outcomes) at posttreatment; 3) the effects of secondary outcomes at follow-up. Sensitivity analyses as well as moderation and subgroup analyses were conducted if there were sufficient studies (i.e., at least four in each subgroup). Forrest plots were created for each analysis.

Publication Bias

Publication bias was assessed in two ways using MAVIS. First, a rank correlation tests for the funnel plot asymmetry were conducted, where a significant correlation ($p < 0.5$) indicated funnel plot asymmetry and publication bias. Second, the trim and fill procedure were used to indicate if there were any missing null studies that could account for significant funnel plot asymmetry (Hamilton et al., 2017).

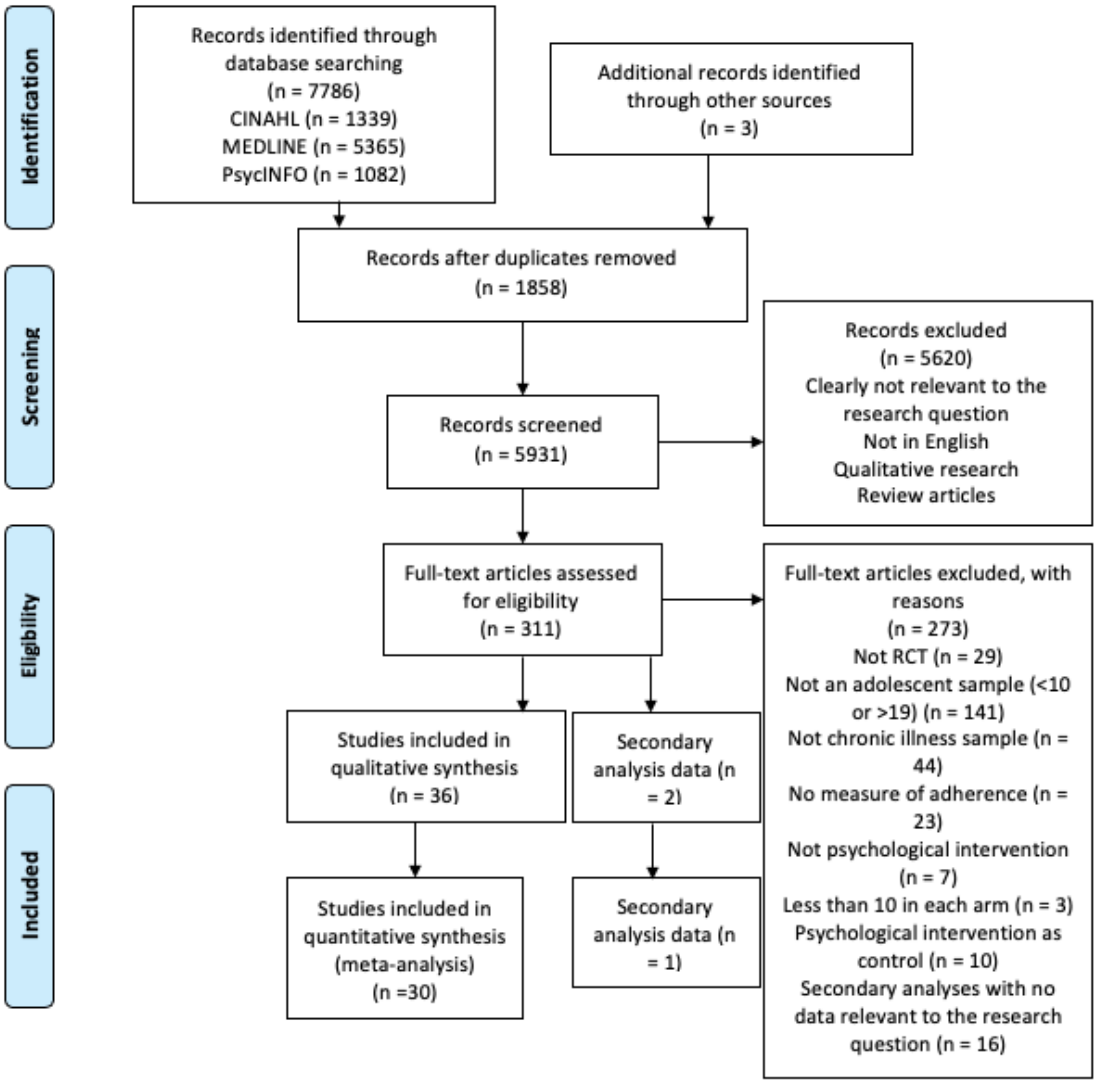
Results

Search Results

The PRISMA flow diagram is outlined in Figure 4.1. A total of 7789 papers were identified by the searches, leaving 5931 after duplicates were removed. Following the screening of titles and subtracts, a total of 311 full-text papers were read. This resulted in a total of 38 papers, describing 36 studies (as two papers provided data for the secondary outcomes of two included studies). Thirty of these studies (with a total of 31 papers) were included in the quantitative synthesis. There was 96.77% agreement on inclusion of studies.

Figure 4.1

PRISMA Flow Diagram (Liberati et al., 2009)



Missing Data

Twenty-three studies did not provide all data needed for the analyses. Therefore, the corresponding authors of all of these studies were contacted. Full details of the data provided by authors and missing data for each study is in the study characteristics table in Table 4.3. For the posttreatment adherence data, nine studies had missing data and were therefore excluded from the primary analysis.

Study Characteristics

Of the papers independently extracted, all extracted data was consistent with that completed by the first author.

Study Design

The 36 studies were conducted across seven countries, including US (n = 24), Canada (n = 5), Denmark (n = 1), The Netherlands (n = 2), Northern Taiwan (n = 1), Zimbabwe (n = 1) and South Africa (n = 2). They were conducted across a range of settings, the majority of which were paediatric clinics (n = 24) but also included schools (n = 3), pharmacies (n = 1), hospitals (n = 6), and medical centres (n = 2). Control groups were mostly usual/standard care (n = 17) but also included attention control (n = 9), waitlist control (n = 6), no treatment control/inactive control group (n = 1), publicly available website (n = 2) or in one study a non-contingent group where participants earned incentives at random. For the 12 studies which included a follow-up time point, the average was 9.17 months (range 2-18).

Participants

In total, 3971 (mean = 110.31, range = 24-345) participants were recruited. On average, 56.66% of the participants were female (one study did not report this). There were a wide range of chronic illness diagnoses in the studies including Type 1 Diabetes (T1D, n = 15), Type 2 Diabetes (T2D, n = 1), Asthma (n = 10), Human Immunodeficiency Virus (HIV, n = 3) and Inflammatory Bowel Disease (IBD, n = 3), Juvenile Idiopathic Arthritis (JIA, n = 3), Chronic Pain (n = 1) and Sickle Cell Disease (n = 1). Only eleven of the studies specifically targeted adolescents with poor or suboptimal adherence. Three studies did not report the retention at the longest follow-up time point, but for studies who did there was an average of 86.64% retention (range 62.26-100%).

Outcomes

Fifteen studies had more than one measure of adherence. For those with adherence data at posttreatment (n = 27), the measures of adherence selected included adolescent self-report of specific chronic illness adherence/self-management (n = 16), objective measures (including electronic monitoring, n = 7) and joint adolescent and parent measures (including report and interviews, n = 4). For those with adherence data at follow-up (n = 11), the measures of adherence selected included adolescent report (n = 5), objective measures (n = 5) and adolescent and parent measures (n = 1). Eighteen studies included a quality of life measure. Of these, 14 studies had data at posttreatment and seven at follow-up. A total of eight studies had family functioning data including family conflict (n = 4), family impact (n = 1), communication (n = 1), family relationships (n = 2). Of these, six studies had data at posttreatment and four at follow-up.

Intervention Characteristics

Interventions in 21 studies included adolescents and their caregivers, whilst 15 included adolescents only. Seventeen of the studies were delivered remotely (either via telehealth, video or telephone calls), 12 were face-to-face and seven were both face-to-face with a remote element (including text messages, telephone and video calls). Twenty-nine of the studies included individual sessions, six included group sessions and one had both group and individual sessions. The average number of planned sessions for those studies who reported this (n = 6 did not report this information), was 10.11 (range 1-40). Three studies reported that they did not have a planned number of sessions and were based on the needs of individual participants, for these three studies the average number of sessions delivered was 40.26 (range 27.09-48).

Table 4.3

Study Characteristics Table

Study author (year)	Country and setting	N randomised; % retention (at longest follow-up)	Chronic illness diagnosis; participant age range; % female	Intervention name; number of sessions (mean); parental/family involvement; delivery; format	Control	Adherence behaviour and measure; Quality of Life (QoL) measure; Family Functioning (FF) measure	Longest follow-up in months	Overall quality rating
Table 4.3								
Bhana et al. (2014)	South Africa, Hospital	65; Not reported	HIV; 10-14; 51%	The VUKA family program; 6 sessions (Not reported); Family intervention; Face-to-face; Groups (multi-family groups)	Wait-list control	Adherence to HIV medication, measured using a single item (adolescent report) [a]; No QoL measure; Caregiver Communication Frequency and Caregiver Communication Comfort using the Family Environment Scale/Family Assessment Measure (FES/FAM) [ab]	N/A	High

Study author (year)	Country and setting	N randomised; % retention (at longest follow-up)	Chronic illness diagnosis; participant age range; % female	Intervention name; number of sessions (mean); parental/family involvement; delivery; format	Control	Adherence behaviour and measure; Quality of Life (QoL) measure; Family Functioning (FF) measure	Longest follow-up in months	Overall quality rating
Table 4.3								
Bruzzee et al. (2008)	US, school	24; 95.8% (data based on students)	Asthma; 11-14; 46%	It's a family affair! A school-based intervention; 6 child sessions (6) and 5 parent sessions (3); Parental involvement; Face-to-face; groups	No treatment control	Asthma management; two indices measuring management behaviours: attack management and symptom Prevention [b], (adolescent only); No QoL measure; Parent-Adolescent Relationship Questionnaire (communication, problem-solving and hostility/warmth subscales) [b]	2 [c]	Moderate (some concerns)
Bruzzee et al. (2010)	US, schools	345; 81.45%	Asthma; 14-16; 70.44%	Asthma Self-management for adolescents (ASMA); 3	Wait-list control	Asthma management; two indices measuring management	12	Moderate (some concerns)

Study author (year)	Country and setting	N randomised; % retention (at longest follow-up)	Chronic illness diagnosis; participant age range; % female	Intervention name; number of sessions (mean); parental/family involvement; delivery; format	Control	Adherence behaviour and measure; Quality of Life (QoL) measure; Family Functioning (FF) measure	Longest follow-up in months	Overall quality rating
Table 4.3								
				workshops (2.8) and 5 individual coaching sessions (4.9); No parental involvement; Face-to-face; groups and individual sessions		behaviours: attack management and symptom prevention (adolescent report) [ab] Pediatric Asthma Quality of life Questionnaire [a]; No FF measure		
Carlsen et al. (2017)	Denmark, pediatric clinic	53; 62.26%	IBD; 10-17; 58.5%	Interactive web-based disease monitoring tool; Not reported; Parental involvement; Online; Individual use with peer discussions	Standard care	IBD medication adherence; The Medication Adherence Report Scale (MARS) and visual analogue scale (VAS) [d]; IMPACT III self-reported Health-Related QoL [d]; No FF measure	N/A	Moderate (some concerns)

Study author (year)	Country and setting	N randomised; % retention (at longest follow-up)	Chronic illness diagnosis; participant age range; % female	Intervention name; number of sessions (mean); parental/family involvement; delivery; format	Control	Adherence behaviour and measure; Quality of Life (QoL) measure; Family Functioning (FF) measure	Longest follow-up in months	Overall quality rating
Table 4.3								
Chawana et al. (2017)	South Africa, public health clinic	50; 100%	HIV (poor control); 10-18; 54%	Home-based enhanced adherence intervention with text messages; 17 (Not reported); No parental involvement; Face-to-face and text messages; Individual sessions	Standard care	ART adherence; AIDS Clinical Trials Group (ACTG) adherence follow-up questionnaire (QLO702) and visual analogue scale (VAS; adolescent report) [a]; No QoL or FF measures	N/A	Moderate (some concerns)
Davis et al. (2019)	US, pediatric clinics	319; Not reported	Persistent asthma; 1-17; 65.5%	Question prompt list and educational video intervention; 1 (Not reported); Parental involvement; Face-to-face; Individual sessions	Usual care	Asthma medication adherence; Visual analog scale (VAS) [ef]; No QoL or FF measures	N/A	High

Study author (year)	Country and setting	N randomised; % retention (at longest follow-up)	Chronic illness diagnosis; participant age range; % female	Intervention name; number of sessions (mean); parental/family involvement; delivery; format	Control	Adherence behaviour and measure; Quality of Life (QoL) measure; Family Functioning (FF) measure	Longest follow-up in months	Overall quality rating
Table 4.3								
Ellis et al. (2007a)	Endocrinology clinic, US	127; 79.53%	Poorly controlled Type 1 Diabetes; 10-17; 51%	Multisystemic therapy; Not a set number of sessions (48 for treatment completers and 9 for dropouts); Family intervention; Face-to-face; Individual family sessions	Standard care	Blood glucose monitoring; electronic monitor; No QoL measure; Diabetes Family Behavior Checklist (DFBC) and Family Relationship Index (data extracted from Ellis et al., 2007b) [d]	6	Moderate (some concerns)
Ellis et al. (2012)	University-affiliated pediatric endocrinology clinic, US	146; 98%	Poorly controlled Type 1 Diabetes; 10-18; 82%	Multisystemic therapy; Not a set number of sessions (45.7); Family intervention; Face-to-face; Individual family sessions	Attention control	Diabetes regimen adherence; Diabetes Management Scale [d]; No QoL or FF data	12	Moderate (some concerns)
Ellis et al. (2019)	Pediatric endocrinology clinic, US	50; 91%	Poorly controlled Type 1 Diabetes;	REACH for Control (adaption of MST-healthcare); The	Standard care	Blood glucose monitoring; electronic monitor [g] (The Diabetes	N/A	Moderate (some concerns)

Study author (year)	Country and setting	N randomised; % retention (at longest follow-up)	Chronic illness diagnosis; participant age range; % female	Intervention name; number of sessions (mean); parental/family involvement; delivery; format	Control	Adherence behaviour and measure; Quality of Life (QoL) measure; Family Functioning (FF) measure	Longest follow-up in months	Overall quality rating
Table 4.3								
			10-18; 62%	expected number of primary sessions was 20 (16.6 for completers and 4.7 for dropouts), while the expected number of follow-up sessions was 16 (1.6 completers and 0.3 dropouts); Family intervention; Face-to-face; Individual family sessions		Management Scale); Diabetes quality of life-youth scale (DQOL-Y); No FF measure		
Goyal et al. (2017)	Two pediatric endocrinology centers, Canada	92; 95.65%	Type 1 Diabetes (struggling with glycemic control); 11-16; 55.43%	A mobile app for the self-management of Type 1 Diabetes; NA; No parental involvement; Online; Individual	Treatment as usual	Blood glucose monitoring; electronic monitor [g] (The Self-Care Inventory); The Diabetes Quality of Life for Youth	12 [c]	Moderate (some concerns)

Study author (year)	Country and setting	N randomised; % retention (at longest follow-up)	Chronic illness diagnosis; participant age range; % female	Intervention name; number of sessions (mean); parental/family involvement; delivery; format	Control	Adherence behaviour and measure; Quality of Life (QoL) measure; Family Functioning (FF) measure	Longest follow-up in months	Overall quality rating
Table 4.3								
				use with peer discussions		(DQOLY) [b]; No FF measure		
Green et al. (2017)	Two pediatric sickle cell clinical sites; US	28; 89.29%	Sickle cell disease, (poor adherence) ; 10-18; 42.9%	"HABIT" Community healthcare workers performed adherence support through home visits, augmented by tailored text messages; Not reported (4.9); Parental involvement; Face-to-face and text messages; Individual sessions	Attention control	Hydroxyurea adherence; Percentage decrease from HbF [gh] (Prescription refill [PDC] and Morisky self-report scale for both parent and child); PedsQL Generic Core Scale (self-report) [eh] (data extracted from Smaldone et al., 2018); No FF measure	N/A	Moderate (some concerns)
Greenley et al. (2015)	Outpatient clinic, US	76; 88.15%	IBD; 11-18; 45%	Problem solving skills training; 2 (Not reported); Parental	Wait list	Oral medication adherence; MEMS track caps electronic monitor [d]; PedsQL	N/A	Moderate (some concerns)

Study author (year)	Country and setting	N randomised; % retention (at longest follow-up)	Chronic illness diagnosis; participant age range; % female	Intervention name; number of sessions (mean); parental/family involvement; delivery; format	Control	Adherence behaviour and measure; Quality of Life (QoL) measure; Family Functioning (FF) measure	Longest follow-up in months	Overall quality rating
Table 4.3								
				involvement; Telephone sessions; Individual sessions		generic core scales [d]; No FF measure		
Hommel et al. (2012)	Outpatient clinic, US	41; 97.56%	IBD; 11-18; 50%	Family based group behavioural intervention; 4 session (Not reported); Parental involvement; Face-to-face; Group sessions	Usual care	IBD medication adherence; Pill count, Treatment Regiment Adherence Questionnaire; MEMS track caps electronic monitor [d]; No QoL or FF measures	N/A	Moderate (some concerns)
Jaser et al. (2014)	Outpatient pediatric diabetes clinic, US	39; 92.31%	Type 1 Diabetes; 13-17; 51.28%	Positive psychology intervention; 4 (Not reported); Parental involvement; Telephone sessions and text messages; Individual sessions	Attention control	Blood glucose monitoring; Electronic monitor [ag] (The Self-Care Inventory and SCI Blood Glucose Regulation Scale both with parent and	6	Moderate (some concerns)

Study author (year)	Country and setting	N randomised; % retention (at longest follow-up)	Chronic illness diagnosis; participant age range; % female	Intervention name; number of sessions (mean); parental/family involvement; delivery; format	Control	Adherence behaviour and measure; Quality of Life (QoL) measure; Family Functioning (FF) measure	Longest follow-up in months	Overall quality rating
Table 4.3								
						adolescent report [a]; PedsQL Diabetes [a]; Diabetes Family Conflict Scale [ai]		
Jaser et al. (2019)	Academic medical center, US	120; 83.33% (based on child data)	Type 1 Diabetes; 13-17; 52.5%	Positive affect intervention; 8 (Not reported); Parental involvement; Telephone sessions and text messages; Individual sessions	Attention control	Blood glucose monitoring; Electronic monitor [g] (The Self-Care Inventory both adolescent and parent); PedsQL Diabetes; No FF measure	6	Moderate (some concerns)
Jaser et al. (2020)	Diabetes clinic, US	39; 89.74%	Type 1 Diabetes; 13-17; 53%	Sleep coach intervention; 3 (Not reported); No parental involvement; Telephone	Standard care	Diabetes management; The Self-Care Inventory [e]; No QoL or FF measures	N/A	Moderate (some concerns)

Study author (year)	Country and setting	N randomised; % retention (at longest follow-up)	Chronic illness diagnosis; participant age range; % female	Intervention name; number of sessions (mean); parental/family involvement; delivery; format	Control	Adherence behaviour and measure; Quality of Life (QoL) measure; Family Functioning (FF) measure	Longest follow-up in months	Overall quality rating
Table 4.3								
				sessions; Individual sessions				
Johnson et al. (2016)	Pediatric outpatient setting, US	98; 90.82%	Asthma; 12-17; 49.4%	MyMediHealth (MMH) website and short messaging service; Over three weeks (2.5 times log in; 12 text reminders); No parental involvement; Online and text messages; Individual sessions; Individual use	Standard care	Compliance with asthma controller; Self-report about compliance during the past seven days [a]; Mini PAQLQ [a]; No FF measure	N/A	Moderate (some concerns)
Joseph et al. (2007)	School, US	314; 87%	Asthma; 14-16; 63.4%	Puff city, web-based asthma management program; 4 (Not reported); No	Generic, publicly available asthma websites	Controller adherence; Self-report [d]; Juniper self-report measure; No FF measure	N/A	High

Study author (year)	Country and setting	N randomised; % retention (at longest follow-up)	Chronic illness diagnosis; participant age range; % female	Intervention name; number of sessions (mean); parental/family involvement; delivery; format	Control	Adherence behaviour and measure; Quality of Life (QoL) measure; Family Functioning (FF) measure	Longest follow-up in months	Overall quality rating
Table 4.3								
				parental involvement; Online; Individual use				
Kichler et al. (2013)	Diabetes clinic, US	30; 83.33%	Type 1 Diabetes; 13-17; 53%	Diabetes adjustment and coping group therapy program; 6 (Not reported); Parental involvement; Face-to-face; Groups	Wait-list control	Diabetes management; The Self-Care Inventory [ae]; PedsQL General [aeh]; PedsQL Family Impact [a]	N/A	High
Kohut et al. (2016)	Large pediatric tertiary hospital; Canada	30; 93.33%	Chronic pain; 12-18; 93%	iPeer2peer program, a peer mentorship program that provides modelling and reinforcement by peers online; 10 (Not reported); No parental	Wait-list control	Self-management of chronic pain; Self-management skills assessment guide (SMSAG); No QoL or FF measures	N/A	Moderate (some concerns)

Study author (year) Table 4.3	Country and setting	N randomised; % retention (at longest follow-up)	Chronic illness diagnosis; participant age range; % female	Intervention name; number of sessions (mean); parental/family involvement; delivery; format	Control	Adherence behaviour and measure; Quality of Life (QoL) measure; Family Functioning (FF) measure	Longest follow-up in months	Overall quality rating
				involvement; Skype calls; Individual sessions				
Kosse et al. (2019)[j]	Community pharmacies, The Netherlands	66 pharmacies (253 patients signed up); 92.49%	Asthma; 12-18; 52.6%	The Adolescent adherence patient tool (ADAPT), an interactive mobile health (mHealth) intervention; 6-month access (Not reported); No parental involvement; Online; Personal use but with a peer chat function	Usual care	Asthma medication adherence; Medication Adherence Report Scale (MARS); Pediatric asthma quality of life questionnaire (PAQLQ); No FF measure	N/A	High
Mayer-Davis et al. (2018)	Pediatric endocrinology	258; 93.41%	Type 1 Diabetes;	Flexible Lifestyles Empowering Change (FLEX),	Usual care	Diabetes self-management behaviours; The	N/A	Moderate (some concerns)

Study author (year) Table 4.3	Country and setting	N randomised; % retention (at longest follow-up)	Chronic illness diagnosis; participant age range; % female	Intervention name; number of sessions (mean); parental/family involvement; delivery; format	Control	Adherence behaviour and measure; Quality of Life (QoL) measure; Family Functioning (FF) measure	Longest follow-up in months	Overall quality rating
	diabetes clinics, US		13-16; 49.65%	adaptive behavioural intervention; 4 to 8 sessions (as well as brief contacts as needed, Not reported); Parental involvement; Face-to-face and use of technology (telephone or video calls); Individual sessions		Diabetes Self-management assessment Profile (DSMP-SR) [e]; PedsQL–generic [e]; The diabetes family conflict scale (DFCS) [i]		
Mosnaim et al. (2013)	Outpatient setting, US	68; 85.29%	Asthma (poor adherence); 11-16; 52.9%	Peer group intervention and mP3 peer-recorded asthma messages; 8 (Not reported); No parental involvement; Face-to-face and some	Attention control	ICS adherence; Electronic monitor [dg] (self-reported adherence); No QoL or FF measures	N/A	Moderate (some concerns)

Study author (year)	Country and setting	N randomised; % retention (at longest follow-up)	Chronic illness diagnosis; participant age range; % female	Intervention name; number of sessions (mean); parental/family involvement; delivery; format	Control	Adherence behaviour and measure; Quality of Life (QoL) measure; Family Functioning (FF) measure	Longest follow-up in months	Overall quality rating
Table 4.3								
				MP3 messages; Groups				
Mulvaney et al. (2010)	Pediatric Diabetes Clinic, US	52; Not reported	Type 1 Diabetes; 13-17; Not reported	Internet-based self-management support intervention; 6 (5.2); No parental involvement; Online; Individual use but also social networking via peer platform	Usual care	Diabetes self-management; Diabetes Behaviour Rating Scale; No QoL or FF measures	N/A	High
Naar-King et al. (2014)	University-affiliated pediatric asthma clinic or during inpatient hospitalization, US	170; 93.53%	Asthma; 12-16; 38.92%	Multisystemic therapy-healthcare; Not a set number of sessions (27.09); family intervention; face-to-face; Individual family sessions	Attention control	Asthma medication adherence; The family asthma management system scale (FAMSS) [fh] (Daily Phone Diary, adolescent self-report); No QoL or FF measures	N/A	Moderate (some concerns)

Study author (year)	Country and setting	N randomised; % retention (at longest follow-up)	Chronic illness diagnosis; participant age range; % female	Intervention name; number of sessions (mean); parental/family involvement; delivery; format	Control	Adherence behaviour and measure; Quality of Life (QoL) measure; Family Functioning (FF) measure	Longest follow-up in months	Overall quality rating
Table 4.3								
Nansel et al. (2007)	Pediatric endocrinology clinics, US	81; 93%	Type 1 Diabetes; 11-16; 55.55%	Diabetes personal trainer intervention; 6 with subsequent telephone calls (Not reported); Parental involvement; Face-to-face with additional telephone calls; Individual sessions	Attention control	Diabetes self-management; Diabetes Self-Management Profile [ae]; The Diabetes Quality of Life Scale [ac]; No FF measure	6	Moderate (some concerns)
Raiff et al. (2016)	Urban outpatient's diabetes center, US	52; 78.85%	Type 1 Diabetes (non-adherent); 13-18; 41.75%	Internet-based incentives to meet web camera-verified SMBG goals to earn incentives plus brief motivational interviewing; 1 MI session and 20 days	Non-contingent (NS) groups where they earned incentives at random	Blood glucose monitoring; electronic monitor [a]; No QoL or FF measures	N/A	High

Study author (year)	Country and setting	N randomised; % retention (at longest follow-up)	Chronic illness diagnosis; participant age range; % female	Intervention name; number of sessions (mean); parental/family involvement; delivery; format	Control	Adherence behaviour and measure; Quality of Life (QoL) measure; Family Functioning (FF) measure	Longest follow-up in months	Overall quality rating
Table 4.3								
				treatment (Not reported); No parental involvement; Online; Individual sessions				
Rikkers-Mutsaerts et al. (2012)	University medical centre, The Netherlands	90; 83.33%	Asthma (poorly controlled) ; 12-18; 50%	Internet-based self-management (IBSM) intervention; Not a set number of sessions (Not reported); No parental involvement; Face-to-face and web-based including telephone calls and text messages; Group sessions	Usual care	Medication adherence; Self-report (adolescent only) [a]; Pediatric asthma quality of life questionnaire PAQLQ [c]; No FF measure	N/A	Moderate (some concerns)

Study author (year)	Country and setting	N randomised; % retention (at longest follow-up)	Chronic illness diagnosis; participant age range; % female	Intervention name; number of sessions (mean); parental/family involvement; delivery; format	Control	Adherence behaviour and measure; Quality of Life (QoL) measure; Family Functioning (FF) measure	Longest follow-up in months	Overall quality rating
Table 4.3								
Stanger et al. (2018)	Endocrinology clinics in a children's hospital, US	61; 98.36%	Type 1 Diabetes (poorly controlled) ; 13-17; 42.6%	Web-delivered multicomponent intervention (WebRx); 40 (Not reported); Parental involvement; Face-to-face; Individual	Usual care	Blood glucose monitoring; electronic monitor [a]; No QoL; Revised Diabetes Family Conflict Scale [a]	12	Moderate (some concerns) for adherence data High for FF data
Stinson et al. (2010)	Tertiary care centres, Canada	46; 86.96%	JIA; 12-18; 69.65%	An internet-based multicomponent intervention; 12 (Not reported); Parental involvement; Online and telephone sessions; Individual use and telephone calls, but with social support	Attention control	JIA treatment adherence; JIA-specific Child Adherence Report Questionnaire (CARQ) and the Parent Adherence Report Questionnaire (PARQ) [b]; Juvenile Arthritis Quality of Life Questionnaire	N/A	High

Study author (year)	Country and setting	N randomised; % retention (at longest follow-up)	Chronic illness diagnosis; participant age range; % female	Intervention name; number of sessions (mean); parental/family involvement; delivery; format	Control	Adherence behaviour and measure; Quality of Life (QoL) measure; Family Functioning (FF) measure	Longest follow-up in months	Overall quality rating
Table 4.3								
						(JAQQ) adolescents only; No FF measure		
Stinson et al. (2016)	Pediatric tertiary hospital, Canada	39; 76.92%	JIA; 12-18; 96.7%	iPeer2peer program, a peer mentorship program that provides modelling and reinforcement by peers online; 10 (Not reported); No parental involvement; Skype calls; Individual sessions	Wait-list control	JIA treatment adherence; Medical issues, Exercise, Pain and Social support questionnaire (MEPS) (self-report, adolescent only); PedsQL Arthritis module [b]; No FF measure	N/A	High
Stinson et al. (2020)	Pediatric rheumatology centers, Canada	333; 65.77%	JIA; 12-18; 70.3%	Teens taking charge web-based self-management intervention; 12 module website plus monthly telephone calls	Attention control	JIA treatment adherence; Child Adherence Report Questionnaire [be]; PedsQL Rheumatology	12	High

Study author (year)	Country and setting	N randomised; % retention (at longest follow-up)	Chronic illness diagnosis; participant age range; % female	Intervention name; number of sessions (mean); parental/family involvement; delivery; format	Control	Adherence behaviour and measure; Quality of Life (QoL) measure; Family Functioning (FF) measure	Longest follow-up in months	Overall quality rating
Table 4.3								
				(Not reported); Parental involvement; Online and telephone sessions; Individual use and sessions		Modules [be]; No FF measure		
Tseng et al. (2020)	Tertiary hospital, Northern Taiwan	90; 92.22%	Asthma; 12-18; 43.4%	Theoretical asthma self-management program; 3 (Not reported); Parental involvement; Face-to-face with text messages and telephone calls; Individual sessions	Usual care	Asthma self-management; Asthma Prevent and Management Index [b]; No QoL or FF measures	N/A	Moderate (some concerns)
Whittemore et al. (2016)	Diabetes clinics, US	124; 69.35%	Type 1 Diabetes; 11-14; 63%	Teens.connect program, an interactive internet program; 10 lessons (14 logons	Attention control (publicly available website	Diabetes management; The Self-Care	6	High

Study author (year)	Country and setting	N randomised; % retention (at longest follow-up)	Chronic illness diagnosis; participant age range; % female	Intervention name; number of sessions (mean); parental/family involvement; delivery; format	Control	Adherence behaviour and measure; Quality of Life (QoL) measure; Family Functioning (FF) measure	Longest follow-up in months	Overall quality rating
Table 4.3								
				per teen); No parental involvement; Online; Individual use but included discussion board	with no lessons)	Inventory [a]; PedsQL Diabetes [a]; No FF measure		
Willis et al. (2019)	HIV clinics, Zimbabwe	100; 68%	HIV; 10-15; 61%	Community Adolescent Treatment Supporters (CATS) who deliver adherence and psychosocial support; Weekly visits over 12 months (Not reported); No parental involvement; Face-to-face, individual sessions	Standard care	ART adherence; Self-report [d]; No QoL or FF measures	N/A	High

Study author (year)	Country and setting	N randomised; % retention (at longest follow-up)	Chronic illness diagnosis; participant age range; % female	Intervention name; number of sessions (mean); parental/family involvement; delivery; format	Control	Adherence behaviour and measure; Quality of Life (QoL) measure; Family Functioning (FF) measure	Longest follow-up in months	Overall quality rating
Table 4.3								
Wysocki et al. (2007)	Pediatric centers, US	68 (from two arms analysed); 83.33%	Type 1 Diabetes or insulin-treated Type 2 Diabetes (poorly controlled) ; 11-16; 45.59%	Behavioural family systems therapy; 12 (Not reported); Family intervention; Face-to-face; Individual family sessions	Standard care	Diabetes self-management; Diabetes Self-Management Profile; No QoL measure; Interaction Behaviour Code (data extracted from Wysocki et al., 2008)	18	High

Note. All included studies are referenced in a separate reference list in the supplementary material.
[a] Authors provided data upon request. [b] Subscales averaged. [c] No data for posttreatment was available. [d] No data available for any relevant analysis. [e] Adolescent report chosen over parent report for primary analysis. [f] Data calculated using data available in the published paper. [g] Objective measure chosen over adolescent and/or parent report for primary analysis. [h] Chosen over a less reliable measure. [I] Parent report chosen over adolescent report. [j] Cluster RCT.

Quality Assessment

Overall, 14 studies were rated as high risk of bias. An additional paper (Stanger et al., 2018) was rated as high risk for the family functioning outcome only. All other papers were rated as moderate risk which is due to many studies being rated as unclear in at least one of the domains. For studies independently rated, 94.4% were agreed upon and any disagreements were resolved via discussions. See Appendix E for quality ratings.

Meta-Analysis Findings

Primary Analyses

Effects for the primary outcome of adherence are presented in Table 4.4. A total of 27 studies, compromising 2524 participants were included. Overall, a significant small effect ($g = 0.21$) was found where those participants who received the psychological intervention had better posttreatment adherence than participants in the control group. However, significant heterogeneity ($p<.001$) was found.

Impact of Study Bias. A sensitivity analysis which excluded the high risk of bias studies are also presented in Table 4.4. The outcomes indicate that study bias had a substantial impact on the findings, as the effect size became larger ($g = 0.30$), and heterogeneity ceased to be significant. The forest plot for this analysis is presented in Figure 4.2.

Impact of Cluster RCTs. A sensitivity analysis which excluded one cluster RCT study from the primary analysis did not lead to any considerable changes (see Table 4.4).

Table 4.4

Outcome of Adherence at Posttreatment Analysis Including Moderation and Subgroup Analyses

		k	*g*	95% CI	p-value	I^2 (*Q, p-value*)
Main analysis (N = 2524)		27	0.21	0.06 to 0.36	**.006**	68% (68.46, **<.001**)
Excluding high risk of bias studies (*N* = 1496)		16	0.30	0.20 to 0.41	**<.001**	0% (22.57, .094)
Excluding cluster study (N = 2290)		26	0.22	0.06 to 0.38	**.009**	70% (68.22, **<.001**)
Moderators	Subgroups					
Sample (*Qb* = 5.85, *p* = 0.016*)	Poor adherence	6	0.54	0.31 to 0.76	**<.001**	0% (2.80, .731)
	Other	10	0.22	0.09 to 0.35	**<.001**	34% (22.57, .146)
Diabetes (*Qb* = 2.62, *p* = 0.106)	Diabetes	8	0.21	0.03 to 0.40	.057	53% (14.88, **.038**)
	Other	8	0.43	0.24 to 0.61	**<.001**	0% (4.75, .690)
Asthma (*Qb* = 0.24, *p* = 0.623)	Asthma	5	0.37	0.15 to 0.59	**.001**	0% (1.53, .821)
	Other	11	0.29	0.11 to 0.48	**.002**	51% (20.58, **.024**)
Parental involvement (*Qb* = 0.29, *p* = 0.592)	Adolescent-only	6	0.34	0.17 to 0.51	**<.001**	0% (4.94, .424)
	Parents involved	10	0.28	0.15 to 0.41	**<.001**	48% (17.34, **.044**)
Delivery (*Qb* = 0.24, *p* = 0.627)	Face to face	10	0.35	0.18 to 0.51	**<.001**	15% (10.60, .304)
	Remote	6	0.27	0.01 to 0.53	.045*	58% (11.84, **.037**)

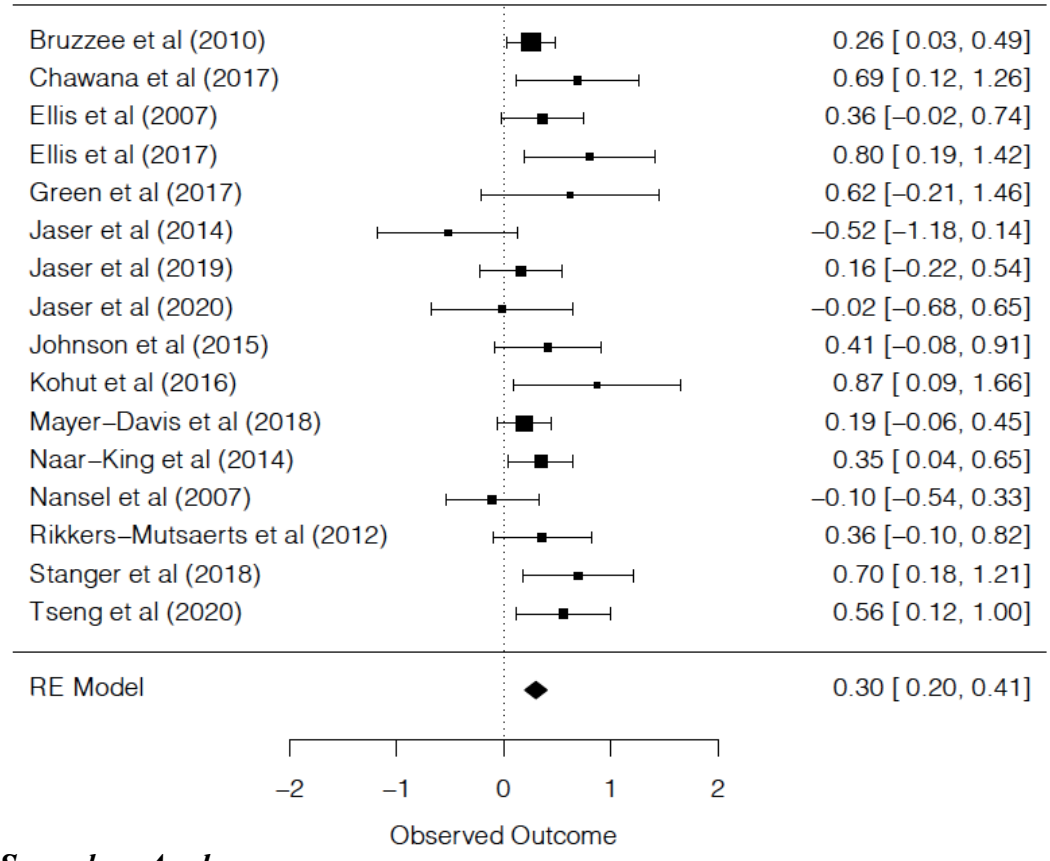
Note. N = number of participants. k = number of studies. *g* = Hedges' *g*. CI = confidence interval. p-value = significance. Significant effects are indicated in bold. Positive *g* indicates that the participants in the intervention group had better adherence. Moderator and subgroup analyses excluded high risk of bias studies.

*Non-significant following the Holm-Bonferroni correction method.

Moderation and Subgroup Analyses. The results of planned moderation and subgroup analyses are also presented in Table 4.4. Following the Holm-Bonferroni correction method, no moderators were significant, but descriptive differences can be observed between effect sizes for each subgroup. Of note, the pooled effect size of studies specifically targeting adolescents with poor adherence is considerably larger. Studies which targeted adolescents with Diabetes had the smallest effect and those studies which used remote delivery also produce smaller effects.

Figure 4.2

Forest Plot for Primary Analysis Excluding High Risk of Bias Studies



Secondary Analyses

Primary Outcomes at Follow-Up. Effects of adherence outcomes at follow-up are presented in Table 4.5. This analysis yielded a non-significant, negligible effect size. A cluster sensitivity analysis was not needed, and no moderation or subgroup analyses could be performed due to an insufficient number of studies.

Impact of Study Bias. The removal of high risk of bias studies did not considerably impact findings. The forest plot for this analysis is presented in Figure 4.3.

Table 4.5.

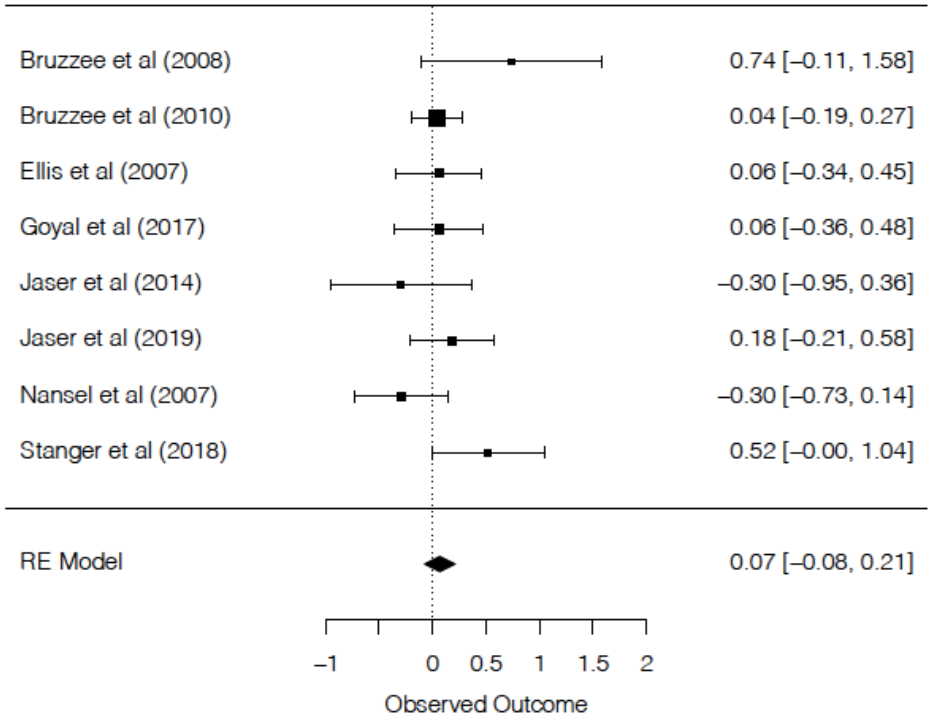
Outcome of the Adherence Measures at Follow-up Analysis

	k	*g*	95% CI	p-value	I^2 (*Q*, *p-value*)
Adherence					
Main analysis (N = 983)	11	0.05	-0.08 to 0.17	.466	.01% (13.20, .212)
Excluding high risk of bias studies (*N* = 763)	8	0.07	-0.08 to 0.21	.355	0% (9.46, .221)

Note. N = number of participants. k = number of studies. *g* = Hedges' *g*. CI = confidence interval. p-value = significance. Significant effects are indicated in bold. Positive g indicates that the participants in the intervention group had better adherence. No moderator or subgroup analyses were possible due to less than four studies per subgroup.

Figure 4.3

Forest Plot for Adherence Data at Follow-up Excluding High Risk of Bias Studies



Secondary Outcomes. Effects of secondary outcomes are presented in Table 4.6. Forest plots are presented in Figures 4.4 and 4.5. Quality of life outcomes at posttreatment yielded a small non-significant effect. Sensitivity analyses resulted in the small effects becoming significant. At follow-up, quality of life outcomes yielded a negligible, non-significant effect. The removal of high risk of bias studies did not lead to any considerable changes. Family functioning outcomes at posttreatment yielded a small, non-significant effect. At follow-up, family functioning outcomes yielded a negligible, non-significant effect. No sensitivity analysis excluding high risk of bias studies could be performed on the family functioning data due to an insufficient number of studies. No moderation or subgroup analyses could be performed due to an insufficient number of studies.

Table 4.6

Outcome of Secondary Outcome Analyses at Posttreatment and Follow-up

	k	*g*	95% CI	p-value	I^2 (*Q, p-value*)
Quality of Life Posttreatment					
Main analysis (N = 1677)	14	0.10	-.008 to 0.20	.071	10% (15.50, .277)
Excluding high risk of bias studies (N = 812)	7	0.15	0.012 to 0.29	**.033**	0% (8.32, .216)
Excluding cluster study (N = 1443)	13	0.14	0.031 to 0.24	**.011**	0% (12.49, .407)
Family Functioning Posttreatment					
Main analysis (N = 467)	6	0.10	-0.35 to 0.55	.660	78% (20.15, **.001**)
Quality of Life Follow-up					
Main analysis (N = 901)	7	0.04	-0.14 to 022	.665	40% (10.16, .118)
Excluding high risk of bias studies (N = 646)	5	0.02	-0.23 to 0.27	.873	52% (8.22, .084)
Family Functioning Follow-up					
Main analysis (N = 170)	4	0.06	-.295 to 0.41	.754	24% (3.74, .291)

Note. N = number of participants. k = number of studies. *g* = Hedges' *g*. CI = confidence interval. p-value = significance. Significant effects are indicated in bold. Positive *g* indicates that the participants in the intervention group had better quality of life or family functioning. No moderator or subgroup analyses were possible due to less than four studies per subgroup.

Figure 4.4

Forest Plot for Quality of Life Outcomes at Posttreatment (Excluding High Risk of Bias Studies)

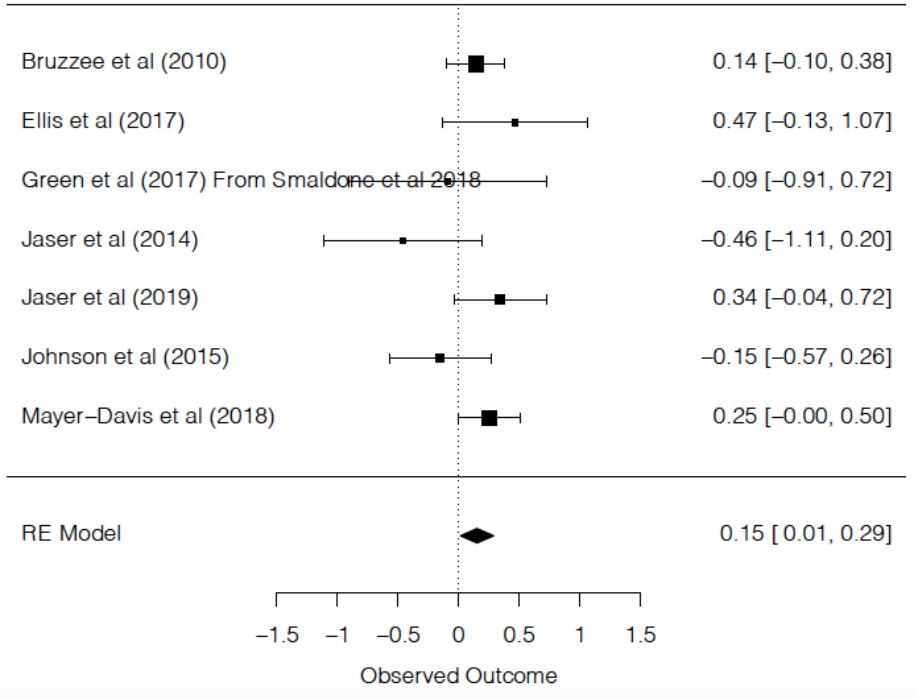
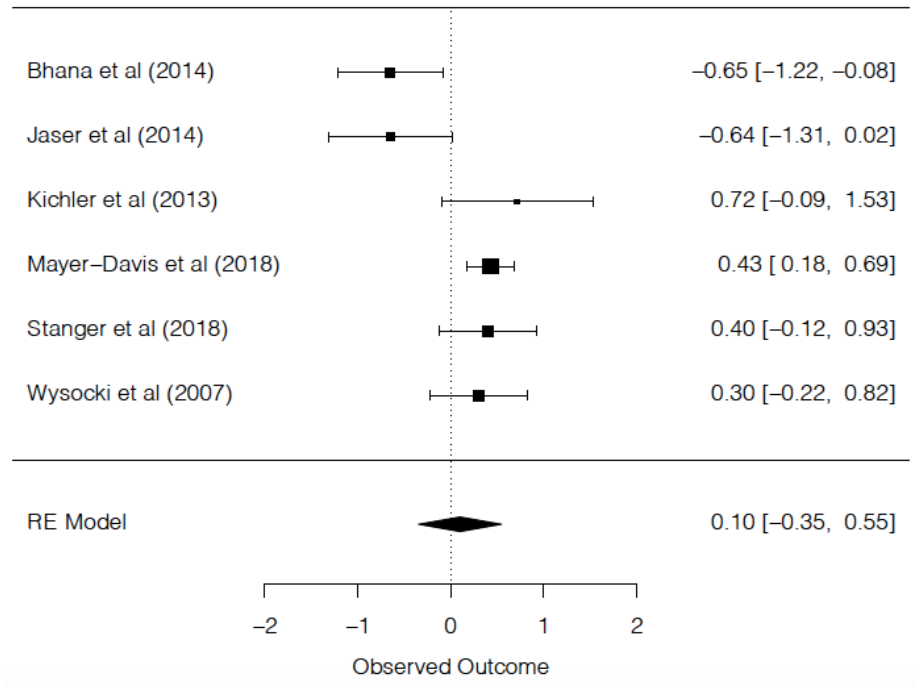


Figure 4.5

Forest Plot for Family Functioning Outcomes at Posttreatment



Publication Bias

The results of the rank correlation tests were non-significant for all measures, except for quality of life outcomes at follow-up. The funnel plot for this outcome estimated two missing studies, which is suggestive of bias. Missing studies were also estimated for quality of life outcomes at posttreatment and adherence outcomes at follow-up, but asymmetry was non-significant. Overall, this suggests that publication bias was limited. See Appendix F for full details.

Discussion

This systematic review and meta-analysis examined literature published since 2007 which examined the efficacy of psychological interventions to promote adherence in adolescents with chronic illness. The findings suggest a small but significant effect of such interventions on improving adherence posttreatment, but not follow-up, when compared to control groups. The effect size increased when only including low risk of bias studies. This indicates that psychological interventions to promote adherence for adolescents with chronic illness currently have limited efficacy. Findings also suggest that study bias had a significant impact on effect sizes found. Therefore, future high-quality research is needed.

The systematic review highlighted that most RCTs examining the efficacy of interventions to promote adherence in adolescents with chronic illnesses have been conducted in the US and in paediatric clinic settings. They mainly employed usual care control groups and used an adolescent self-report measure of adherence. A considerable number of included studies recruited adolescents with Diabetes and very few specifically targeted adolescents with poor/suboptimal adherence or had a follow-up time point. Most interventions were delivered individually, rather than in groups, and included adolescents and their parents/family members.

The findings for posttreatment adherence for adolescents specifically are consistent with Pai and McGrady's (2014) findings, which also found significant small effects in paediatric samples. After correcting for multiple comparisons, no moderation analyses were significant. However, the findings from subgroup analyses indicated that outcomes were most efficacious when studies specifically recruited adolescents who had difficulties with adherence and also indicated that outcomes for adolescents with Diabetes are less efficacious.

Findings from subgroup analyses may provide explanations for the small effects observed. First, few studies included in the primary analysis specifically targeted adolescents with poor/suboptimal adherence, so small effects found might be explained by ceiling effects. Further, the majority of studies targeted adolescents with Diabetes, which produced the smallest effects.

The subgroup analyses indicated that interventions delivered face-to-face produced only slightly larger effects than those that were remotely delivered. Similarly, those that were aimed at adolescents only produced slightly larger effects than those which also included parents/family members. These small differences were not statistically significant.

Unlike Pai and McGrady (2014), adherence outcomes at follow-up produced negligible, non-significant effects. This suggests that there is little evidence for intervention efficacy at follow-up. However, very few studies had a follow-up time point and those that did varied in their longest follow-up time point.

In terms of secondary outcomes, a significant small effect was found for quality of life at posttreatment and effect sizes were negligible and non-significant at follow-up. In terms of family functioning outcomes, small non-significant effects were found at both posttreatment and follow-up. Overall, these findings suggest that psychological

interventions to promote adherence are not efficacious for quality of life or family functioning outcomes. However, a small number of studies measured these outcomes and there was considerable variability in measures used.

Strengths and Limitations

Overall, the strength of the current review and meta-analysis is that it thoroughly examined recent intervention literature targeting adherence in adolescents with chronic illness (Hanghøj & Boisen, 2013; Rapoff, 2010). Compared to the most recent meta-analysis in the field, who included a total of 23 studies (Pai & McGrady, 2014), a larger number of studies was included. Due to the differences in the aims and criteria of the present review compared to Pai and McGrady's (2014) review, only eight studies were included in both reviews. The present review also included additional secondary outcomes and explored several potential moderators of treatment efficacy.

There was a high number of missing data. This had implications for the analyses that were possible, and therefore, effect sizes reported may not be an accurate representation of true treatment effects and thus need to be interpreted with caution. Study bias had an impact on effects found and heterogeneity and this is an issue given the quality issues found with the studies.

The decision to synthesise studies across different contexts and chronic illness diagnoses was taken due to adolescents having more difficulties and unique challenges with adherence. This decision meant that moderator and subgroup variables were possible. It also resulted in a broad range of chronic illness diagnoses and countries being represented, which meant that diagnoses and populations which are under-researched in this area, such as Sickle Cell Disease (Green et al., 2017), could be included.

Moderator and subgroup analyses were conducted to explore specific factors which may influence intervention effectiveness. Although there were multiple moderator and

subgroup analyses, these were planned a-priori and corrections for multiple comparisons was made, which is a strength. A limitation of these additional analyses is that important moderators, such as intervention components and theoretical frameworks may have been missed. However, given the differences between studies at present, these analyses would likely have been difficult.

Research Implications

The findings highlight specific implications for future research examining interventions to promote adherence in adolescents with chronic illnesses.

First, the review highlighted key issues with study quality. Most studies were rated as moderate risk of bias due to them not being specific in their reporting. Despite previous meta-analyses in the field highlighting similar issues with study quality (e.g., Pai & McGrady, 2014), the current review highlights that this body of literature has not been conducted to a high standard. Therefore, future research in the field should address these issues.

Further, very few studies included a follow-up time point and those which did varied greatly in when these were conducted. This has two implications for future research. First, studies, where possible, should include a follow-up time point in order for the maintenance of effects to be examined. This is important given that adolescents will likely be managing adherence into adulthood. Second, as recommended by Pai and McGrady (2014), studies should carry out follow-up time point consistently so that comparisons can be made. They should be grounded in a clear clinical rationale so that they can have greater applicability to clinical settings.

Third, in line with the Open Science Agenda (European Commission, 2020), future studies should report descriptive data to ensure that all available data is included in future

meta-analysis. This will ensure that effect sizes reported in future meta-analysis accurately represent treatment effects.

Moreover, due to missing data and a lack of studies for several chronic illnesses it was not possible to explore effect sizes in other chronic illness, except for Diabetes and Asthma. Several common chronic illnesses were not represented at all. Further, most studies were conducted in the US and in paediatric clinics and did not specifically target adolescents with poor adherence. Generalisability of the findings of the current review are limited by several common chronic illnesses (such as Epilepsy), countries (particularly European countries) and settings (e.g., schools) not being well represented. Therefore, future studies should conduct interventions across settings and in underrepresented illnesses where adherence is a challenge for adolescents, including Epilepsy (Carbone et al., 2013) and Cancer (Rohan et al., 2017).

Further, the review indicated that psychological interventions to promote adherence in adolescents are more efficacious for those with identified adherence difficulties. Therefore, given that adolescents with identified adherence difficulties are those who receive these interventions in healthcare settings, future research should attempt to recruit those with suboptimal adherence.

Several moderation variables, which may have been important, such as intervention components and theoretical frameworks, were not explored. Therefore, more exploratory research at an earlier stage of intervention development (i.e., pre-RCT) is needed to explore effective treatment components and guiding frameworks. Future studies should explicitly ground their interventions on established theoretical frameworks and provide a clear rationale for intervention components. In a topical review, McGrady, Ryan, Brown et al. (2015) applied the theoretical domains framework (TDF), which is an adult behaviour change theory, to paediatric adherence-promoting interventions. This was an

effective way of reducing variability between studies and if future research used this framework, it would allow for future meta-analysis to examine additional moderators of efficacy of treatment effect.

More specifically, to progress the field and fill the current gaps in the evidence base, a future trial should have a targeted recruitment strategy in order to recruit participants with adherence difficulties. It is acknowledged that it may be difficult to recruit these adolescents, particularly for a powered trial, so adolescents could be recruited across a range of chronic illnesses. As aforementioned, the intervention in such a trial should be linked to theoretical models of adherence and as proposed by McGrady, Ryan, Brown et al. (2015), the trial could apply the TDF and include specific intervention components included in this framework, including psychoeducation, skill development and a social component. This could be compared to an attention control group, such as telephone support (Ellis et al., 2012), in order to control for non-specific intervention aspects, such as regular contact with a clinician. It would be imperative that such a trial examines the mechanism of change through examining which components of the intervention account for an increase in adherence to treatment in that trial. A process evaluation (Oakley et al., 2006), examining the implementation of the intervention through exploring the lived experiences of those involved, would also be beneficial. Future research can then take small steps in testing future intervention components, based on findings from the previous trial. An objective measure of adherence should be included. Finally, the trial should include short-, medium- and long-term follow up so that maintenance of treatment effects can be examined.

Clinical Implications

Due to the issues identified within this current body of literature, specific recommendations based on these findings are difficult to make until research in the field begins to address these.

Overall, findings suggest that there is limited evidence of efficacy of psychological interventions in promoting adherence to treatment in this group. However, the findings of the subgroup analyses suggested that they were efficacious for adolescents with poor adherence. Therefore, healthcare professionals should be delivering psychological interventions to this subgroup. However, other interventions may be necessary to target quality of life and family functioning outcomes.

There are resource-related barriers to implementing psychological interventions to promote adherence in paediatric populations (e.g., clinic space, McGrady, Ryan, Gutiérrez-Colina et al., 2015). Whilst findings suggest that face-to-face interventions are slightly more efficacious than remotely delivered interventions, the difference was small and not statistically significant. Therefore, healthcare professionals should deliver interventions face-to-face in the first instance. However, where this is not possible or safe, such as in current COVID-19 pandemic, healthcare professionals could employ web-based delivery similar to that of several studies in this review to overcome these barriers.

Conclusion

This is the first meta-analysis to examine the efficacy of psychological interventions to promote adherence, quality of life and family functioning outcomes in adolescents with chronic illness. Thirty-six studies met inclusion criteria, but several could not be included in quantitative synthesis due to missing data. The findings suggest a small but significant effect of such interventions on improving adherence at posttreatment, but not follow-up, when compared to control groups. Overall, these findings suggest there is limited evidence of efficacy of psychological interventions in promoting adherence in

adolescents specifically. There was also high heterogeneity between study design and study bias had a significant impact on effect sizes found. Moderation and subgroup analyses indicated that outcomes were best when studies specifically recruited adolescents who had suboptimal adherence and that outcomes for adolescents with Diabetes (which made up most of the included studies) are less efficacious. There was little evidence of efficacy on quality of life or family functioning outcomes. Future high-quality research recruiting adolescent samples with suboptimal adherence across a range of chronic illnesses is needed.

Declaration of Interest Statement

No conflict of interest is reported.

References

Carbone, L., Zebrack, B., Plegue, M., Joshi, S., & Shellhaas, R. (2013). Treatment adherence among adolescents with epilepsy: What really matters?. *Epilepsy & Behavior*, *27*(1), 59-63. https://doi.org/10.1016/j.yebeh.2012.11.047

Cochrane. (2020, February 3). *RCT filters for different databases*. Cochrane Work: https://work.cochrane.org/rct-filters-different-databases

Cooper, H. M., Hedges, L. V., & Valentine, J. C. (2009). *The handbook of research synthesis and meta-analysis.* Russel Sage Foundation.

Cuijpers, P. (2016). *Meta-analyses in mental health research: A practical guide.* Pim Cuijpers Uitgeverij.

DiMatteo, M. R. (2004). Variations in patients' adherence to medical recommendations: a quantitative review of 50 years of research. *Medical care*, 200-209. https://www.jstor.org/stable/4640729

Drotar, D. (2000). *Promoting adherence to medical treatment in childhood chronic illness: Concepts, methods, and interventions.* Lawrence Erlbaum Associates.

Eldridge, S., Campbell, M., Campbell, M., Dahota, A., Giraudeau, B., Higgins, J., Reeves, B., & Seigfried, N. (2016). *Revised Cochrane Risk of Bias Tool for Randomised Trials (RoB 2.0): Additional Considerations for Cluster-Randomised Trials. https://sites.google.com/site/riskofbiastool/welcome/rob-2-0-tool/archive-rob-2-0-cluster-randomized-trials-2016*

Ellis, D. A., Carcone, A. I., Naar-King, S., Rajkumar, D., Palmisano, G., & Moltz, K. (2019). Adaptation of an evidence-based diabetes management intervention for delivery in community settings: Findings from a pilot randomized effectiveness trial. *Journal of Pediatric Psychology, 44(1)*, 110-125. https://doi.org/10.1093/jpepsy/jsx144

Ellis, D. A., Naar-King, S., Chen, X., Moltz, K., Cunningham, P. B., & Idalski-Carcone, A. (2012). Multisystemic therapy compared to telephone support for youth with poorly controlled diabetes: Findings from a randomized controlled trial. *Annals of Behavioral Medicine*, *44*(2), 207-215. https://doi-org.uea.idm.oclc.org/10.1007/s12160-012-9378-1

EPOC. (2020, May 16). *EPOC Resources for review authors.* Cochrane Effectice Practice and Organisation of Care (EPOC): https://epoc.cochrane.org/resources/epoc-resources-review-authors

European Commission. (2020, October 29). *Open Science*. https://ec.europa.eu/digital-single-market/en/open-science

Fredericks, e. m., Magee, J. C., Opipari-Arrigan, L., Shieck, V., Well, A., & Lopez, M. J. (2008). Adherence and health-related quality of life in adolescent liver transplant recipients. *Pediatric Transplantation, 12(3)*, 289-299. https://doi.org/10.1111/j.1399-3046.2008.00901.x

Fritz, C. O., Morris, P. E., & Richler, J. J. (2012). Effect Size Estimates: Current Use, Calculations, and Interpretations. *Journal of Experimental Psychology: General, 141*(1), 2-18. https://doi.org/10.1037/a0024338

Gaetano, J. (2018). *Holm-Bonferroni sequential correction: An Excel calculator* (1.3). ResearchGate. https://bit.ly/2VQOqcn

Glanville, J., Dooley, G., Wisniewski, S., Foxlee, R., & Noel-Storr, A. (2019). Development of a search filter to identify reports of controlled clinical trials within CINAHL Plus. *Health Information & Libraries Journal, 36(1)*, 73-90. https://doi.org/10.1111/hir.12251

Graves, M. M., Roberts, M. C., Rapoff, M., & Boyer, A. (2010). The Efficacy of Adherence Interventions for Chronically Ill Children: A Meta-Analytic Review.

Journal of Pediatric Psychology, 35(4), 368-382. https://doi.org/10.1093/jpepsy/jsp072

Green, N. S., Manwani, D., Matos, S., Hicks, A., Soto, L., Castillo, Y., Ireland, K., Stennett, Y., Findley, S., Jia, H., & Smaldone, A. (2017). Randomized feasibility trial to improve hydroxyurea adherence in youth ages 10–18 years through community health workers: The HABIT study. *Pediatric blood & cancer*, *64*(12), e26689. https://doi-org.uea.idm.oclc.org/10.1002/pbc.26689

Hagell, A., Coleman, J., & Brooks, F. (2015). *Key Data on Adolescence*. Association for Young People's Health: http://www.youngpeopleshealth.org.uk/wp-content/uploads/2015/09/KeyData2015.pdf

Hamilton, K. W., Aydin, B., Mizumoto, A., Coburn, K., Zelinksy, N. (2017). MAVIS: Meta-Analysis via Shiny. R Package Version 1.1.3

Hanghøj, S., & Boisen, K. A. (2013). Self-reported barriers to medication adherence among chronically ill adolescents: A systematic review. *Journal of Adolescent Health, 54(2)*, 121-138. https://doi.org/10.1016/j.jadohealth.2013.08.009

Heath, G., Farre, A., & Shaw, K. (2017). Parenting a child with chronic illness as they transition into adulthood: a systematic review and thematic synthesis of parents' experiences. *Patient education and counselling, 100(1)*, 76-92. https://doi.org/10.1016/j.pec.2016.08.011

Higgins, J. P., & Green, S. (2008). *Cochrane handbook for systematic reviews of interventions.* John Wiley & Sons.

Higgins, J. P., Savović, J., Page , M. J., Elbers, R. G., & Sterne, J. A. (2020). *Chapter 8: Assessing risk of bias in a randomized trial*. Cochrane Training: https://training.cochrane.org/handbook/current/chapter-08

Higgins, J. P., Thompson, S. G., Deeks, J. J., & Altman, D. G. (2003). Measuring inconsistency in meta-analyses. *British Medical Journal, 327(7414)*, 557-560. https://doi.org/10.1136/bmj.327.7414.557

Holm, S. (1979). A simple sequentially rejective method procedure. *Scandinavian Journal of Statistics, 6(2)*, 65-70. https://www.jstor.org/stable/4615733

Kahana, S., Drotar, D., & Frazier, T. (2008). Meta-analysis of psychological interventions to promote adherence to treatment in pediatric chronic health conditions. *Journal of Pediatric Psychology, 33(6)*, 590-611. https://doi.org/10.1093/jpepsy/jsm128

Khan, K. S., Kunz, R., Kleijnen, J., & Antes, G. (2003). Five steps to conducting a systematic review. *Journal of the Royal Society of Medicine*, 118-121.

Kosse, R. C., Bouvy, M. L., de Vries, T. W., & Koster, E. S. (2019). Effect of a mHealth intervention on adherence in adolescents with asthma: A randomized controlled trial. *Respiratory Medicine, 149*, 45-51. https://doi.org/10.1016/j.rmed.2019.02.009

Law, E., Fisher, E., Eccleston, C., & Palermo, T. M. (2019). Psychological interventions for parents of children and adolescents with chronic illness. *Cochrane Database of Systematic Reviews, 3*. https://doi.org/10.1002/14651858.CD009660

Lerch, M. F., & Thrane, S. E. (2019). Adolescents with chronic illness and the transition to self-management: A systematic review. *Journal of Adolescence, 72*, 152-161.

Liberati, A., Altman, D. G., Tetzlaff, j., Mulrow, C., Gøtzsche, P. C., Ioannidis, J. P., Clarke, J. P., Devereaux, P. J., Kleijnen, P., & Moher, D. (2009). The PRISMA Statement for Reporting Systematic Reviews and Meta-Analyses of Studies That Evaluate Health Care Interventions: Explanation and Elaboration. *Annuals of Internal Medicine, 151(4)*, 65-94. https://doi.org/10.1016/j.jclinepi.2009.06.006

McGrady, M. E., Ryan, J. L., Brown, G. A., & Cushing, C. C. (2015). Topical review: theoretical frameworks in pediatric adherence-promotion interventions: research

findings and methodological implications. *Journal of pediatric psychology*, *40*(8), 721-726. https://doi.org/10.1093/jpepsy/jsv025

McGrady, M. E., Ryan, J. L., Gutiérrez-Colina, A. M., Fredericks, E. M., Towner, E. K., & Pai, A. L. (2015). The impact of effective paediatric adherence promotion interventions: systematic review and meta-analysis. *Child: care, health and development, 41(6)*, 789-802. https://doi.org/10.1111/cch.12271

Modi, A. C., & Driscoll, K. A. (2020). *Adherence and Self-Management in Pediatric Populations.* Elsevier Science & Technology.

Moher, D., Liberati, A., Tetzlaff, J., Altman, D. G., & The PRISMA Group. (2009). Preferred reporting items for systematic reviews and meta-analyses: The PRISMA statement. *PLoS Medicine, 6(7)*, e1000097. https://doi.org/10.1136/bmj.b2535

Oakley, A., Strange, V., Bonell, C., Allen, E., & Stephenson, J. (2006). Process evaluation in randomised controlled trials of complex interventions. *BMJ*, *332*(7538), 413-416. https://doi.org/10.1136/bmj.332.7538.413

Pai, A. L., & McGrady, M. (2014). Systematic review and meta-analysis of psychological interventions to promote treatment adherence in children, adolescents, and young adults with chronic illness. *Journal of Pediatric Psychology, 39(8)*, 918-931. https://doi.org/10.1093/jpepsy/jsu038

Peng, Y., Wang, H., Fang, Q., Xie, L., Shu, L., Sun, W., & Liu, Q. (2020). Effectiveness of mobile applications on medication adherence in adults with chronic diseases: a systematic review and meta-analysis. *Journal of managed care & specialty pharmacy*, *26*(4), 550-561. https://doi.org/10.18553/jmcp.2020.26.4.550

Psihogios, A. M., Fellmeth, H., Schwartz, L. A., & Barakat, L. P. (2019). Family Functioning and Medical Adherence Across Children and Adolescents With

Chronic Health Conditions: A Meta-Analysis. *Journal of Pediatric Psychology, 44(1)*, 84-97. https://doi.org/10.1093/jpepsy/jsy044

Rapoff, M. (2010). *Adherence to pediatric medical regimens.* Springer.

Richardson, W. S., Wilson, M. C., Nishikawa, J., & Hayward, R. S. (1995). The well-built clinical question: a key to evidence-based decisions. *Acp j club*, *123*(3), A12-A13. https://doi.org/10.7326/ACPJC-1995-123-3-A12

Rohan, J. M., Fukuda, T., Alderfer, M. A., Wetherington Donewar, C., Ewing, L., Katz, E. R., Muriel, A. C., Vinks, A. A., & Drotar, D. (2017). Measuring medication adherence in pediatric cancer: an approach to validation. *Journal of pediatric psychology*, *42*(2), 232-244. https://doi.org/10.1093/jpepsy/jsw039

Stanger, C., Lansing, A. H., Scherer, E., Budney, A., Christiano, A. S., & Casella, S. J. (2018). A web-delivered multicomponent intervention for adolescents with poorly controlled Type 1 diabetes: a pilot randomized controlled trial. *Annals of Behavioral Medicine*, *52*(12), 1010-1022. https://doi.org/10.1093/abm/kay005

Suris, J.-C., Michaud, P.-A., & Viner, R. (2004). The adolescent with a chronic condition. Part I: developmental issues. *Archives of Disease in Childhood, 89(10)*, 938-942. http://dx.doi.org/10.1136/adc.2003.045369

van der Lee, J. H., Mokkink, L. B., Grootenhuis, M. A., Heymans, H. S., & Offringa, M. (2007). Definitions and Measurement of Chronic Health Conditions in Childhood A Systematic Review. *JAMA, 297(24)*, 2741-2751. https://doi.org/10.1001/jama.297.24.2741

Viner, R., & Christie, D. (2005). Adolescent development. *BMJ, 330(7486)*, 301-304. https://doi.org/10.1136/bmj.330.7486.301

World Health Organisation. (2003). *Adherence to Long-Term Therapies: Evidence for Action.* https://www.who.int/chp/knowledge/publications/adherence_full_report.pdf?ua=1

World Health Organisation. (n.d.). *Adolescent Health*. https://www.who.int/health-topics/adolescent-health/#tab=tab_1

Yeo, M., & Sawyer, S. (2005). Chronic illness and disability. *British Medical Journal, 330(7493)*, 721-723. https://doi.org/10.1136/bmj.330.7493.721

Supplementary Material

References of Included Studies

Bhana, A., Mellins, C. A., Petersen, I., Alicea, S., Myeza, N., Holst, H., Abrams, E., John, S., Chhagan, M., Nestadt, D. F., Leu, C-S., & Leu, C. S. (2014). The VUKA family program: piloting a family-based psychosocial intervention to promote health and mental health among HIV infected early adolescents in South Africa. *AIDS care*, *26*(1), 1-11. https://doi-org.uea.idm.oclc.org/10.1080/09540121.2013.806770

Bruzzese, J. M., Sheares, B. J., Vincent, E. J., Du, Y., Sadeghi, H., Levison, M. J., Mellins, R. B., & Evans, D. (2011). Effects of a school-based intervention for urban adolescents with asthma: a controlled trial. *American journal of respiratory and critical care medicine*, *183*(8), 998-1006. https://doi-org.uea.idm.oclc.org/10.1164/rccm.201003-0429OC

Bruzzese, J. M., Unikel, L., Gallagher, R., Evans, D., & Colland, V. (2008). Feasibility and impact of a school-based intervention for families of urban adolescents with asthma: Results from a randomized pilot trial. *Family process*, *47*(1), 95-113. https://doi-org.uea.idm.oclc.org/10.1111/j.1545-5300.2008.00241.x

Carlsen, K., Jakobsen, C., Houen, G., Kallemose, T., Paerregaard, A., Riis, L. B., Munkholm, P., & Wewer, V. (2017). Self-managed eHealth disease monitoring in children and adolescents with inflammatory bowel disease: a randomized controlled trial. *Inflammatory bowel diseases*, *23*(3), 357-365. https://doi-org.uea.idm.oclc.org/10.1097/MIB.0000000000001026

Chawana, T. D., Katzenstein, D., Nathoo, K., Ngara, B., & Nhachi, C. F. B. (2017). Evaluating an enhanced adherence intervention among HIV positive adolescents failing atazanavir/ritonavir-based second line antiretroviral treatment at a public

health clinic. *Journal of AIDS and HIV research (Online)*, *9*(1), 17. https://doi-org/10.5897/JAHR2016.0406

Davis, S. A., Carpenter, D., Lee, C., Garcia, N., Reuland, D. S., Tudor, G., ... & Sleath, B. (2019). Effect of an Asthma Question Prompt List and Video Intervention on Adolescents' Medication Adherence 12 Months Later. *Annals of Pharmacotherapy*, *53*(7), 683-689. https://doi-org.uea.idm.oclc.org/10.1177/1060028019831259

Ellis, D. A., Carcone, A. I., Naar-King, S., Rajkumar, D., Palmisano, G., & Moltz, K. (2019). Adaptation of an evidence-based diabetes management intervention for delivery in community settings: Findings from a pilot randomized effectiveness trial. *Journal of Pediatric Psychology, 44(1)*, 110-125. https://doi.org/10.1093/jpepsy/jsx144

Ellis, D. A., Naar-King, S., Chen, X., Moltz, K., Cunningham, P. B., & Idalski-Carcone, A. (2012). Multisystemic therapy compared to telephone support for youth with poorly controlled diabetes: Findings from a randomized controlled trial. *Annals of Behavioral Medicine*, *44*(2), 207-215. https://doi-org.uea.idm.oclc.org/10.1007/s12160-012-9378-1

Ellis, D. A., Templin, T., Naar-King, S., Frey, M. A., Cunningham, P. B., Podolski, C. L., & Cakan, N. (2007a). Multisystemic therapy for adolescents with poorly controlled type I diabetes: Stability of treatment effects in a randomized controlled trial. *Journal of consulting and clinical psychology*, *75*(1), 168. https://doi.org/10.1037/0022-006X.75.1.168

Ellis, D. A., Yopp, J., Templin, T., Naar-King, S., Frey, M. A., Cunningham, P. B., Idalski, A., & Niec, L. N. (2007b). Family mediators and moderators of treatment outcomes among youths with poorly controlled type 1 diabetes: results from a randomized

controlled trial. *Journal of pediatric psychology*, *32*(2), 194-205. https://doi-org.uea.idm.oclc.org/10.1093/jpepsy/jsj116

Goyal, S., Nunn, C. A., Rotondi, M., Couperthwaite, A. B., Reiser, S., Simone, A., Katzman, D. K., Cafazzo, J. A., & Palmert, M. R. (2017). A mobile app for the self-management of type 1 diabetes among adolescents: a randomized controlled trial. *JMIR mHealth and uHealth*, *5*(6), e82. https://doi.org/10.2196/mhealth.7336

Green, N. S., Manwani, D., Matos, S., Hicks, A., Soto, L., Castillo, Y., Ireland, K., Stennett, Y., Findley, S., Jia, H., & Smaldone, A. (2017). Randomized feasibility trial to improve hydroxyurea adherence in youth ages 10–18 years through community health workers: The HABIT study. *Pediatric blood & cancer*, *64*(12), e26689. https://doi-org.uea.idm.oclc.org/10.1002/pbc.26689

Greenley, R. N., Gumidyala, A. P., Nguyen, E., Plevinsky, J. M., Poulopoulos, N., Thomason, M. M., Walter, J. G., Wojtowicz, A. A., Blank, E., Gokhale, R., & Kirschner, B. S. (2015). Can you teach a teen new tricks? Problem solving skills training improves oral medication adherence in pediatric patients with inflammatory bowel disease participating in a randomized trial. *Inflammatory bowel diseases*, *21*(11), 2649-2657. https://doi-org.uea.idm.oclc.org/10.1097/MIB.0000000000000530

Hommel, K. A., Hente, E. A., Odell, S., Herzer, M., Ingerski, L. M., Guilfoyle, S. M., & Denson, L. A. (2012). Evaluation of a group-based behavioral intervention to promote adherence in adolescents with inflammatory bowel disease. *European journal of gastroenterology & hepatology*, *24*(1), 64. https://doi-org/10.1097/MEG.0b013e32834d09f1

Jaser, S. S., Hamburger, E. R., Bergner, E. M., Williams, R., Slaughter, J. C., Simmons, J. H., & Malow, B. A. (2020). Sleep coach intervention for teens with type 1 diabetes:

Randomized pilot study. *Pediatric Diabetes*, *21*(3), 473-478. https://doi-org.uea.idm.oclc.org/10.1111/pedi.12991

Jaser, S. S., Patel, N., Rothman, R. L., Choi, L., & Whittemore, R. (2014). Check it! A randomized pilot of a positive psychology intervention to improve adherence in adolescents with type 1 diabetes. *The Diabetes Educator*, *40*(5), 659-667. https://doi.org/10.1177/0145721714535990

Jaser, S. S., Whittemore, R., Choi, L., Nwosu, S., & Russell, W. E. (2019). Randomized Trial of a Positive Psychology Intervention for Adolescents With Type 1 Diabetes. *Journal of pediatric psychology*, *44*(5), 620-629. https://doi-org.uea.idm.oclc.org/10.1093/jpepsy/jsz006

Johnson, K. B., Patterson, B. L., Ho, Y. X., Chen, Q., Nian, H., Davison, C. L., Stagle, J., & Mulvaney, S. A. (2016). The feasibility of text reminders to improve medication adherence in adolescents with asthma. *Journal of the American Medical Informatics Association*, *23*(3), 449-455. https://doi-org.uea.idm.oclc.org/10.1093/jamia/ocv158

Joseph, C. L., Peterson, E., Havstad, S., Johnson, C. C., Hoerauf, S., Stringer, S., Gibson-Scipio, W., Ownby, D. R., Elston-Lafata, J., Pallonen, U., & Strecher, V. (2007). A web-based, tailored asthma management program for urban African-American high school students. *American journal of respiratory and critical care medicine*, *175*(9), 888-895. https://doi.org/10.1164/rccm.200608-1244OC

Kichler, J. C., Kaugars, A. S., Marik, P., Nabors, L., & Alemzadeh, R. (2013). Effectiveness of groups for adolescents with type 1 diabetes mellitus and their parents. *Families, Systems, & Health*, *31*(3), 280. https://doi.org/10.1037/a0033039

Kohut, S. A., Stinson, J. N., Ruskin, D., Forgeron, P., Harris, L., van Wyk, M., Luca, S., & Campbell, F. (2016). iPeer2Peer program: a pilot feasibility study in adolescents

with chronic pain. *Pain*, *157*(5), 1146-1155. https://doi-org/10.1097/j.pain.0000000000000496

Kosse, R. C., Bouvy, M. L., de Vries, T. W., & Koster, E. S. (2019). Effect of a mHealth intervention on adherence in adolescents with asthma: A randomized controlled trial. *Respiratory Medicine, 149*, 45-51. https://doi.org/10.1016/j.rmed.2019.02.009

Mayer-Davis, E. J., Maahs, D. M., Seid, M., Crandell, J., Bishop, F. K., Driscoll, K. A., Hunter, C. M., Kichler, J. C., Standiford, D., & Thomas, J. M. (2018). The Flexible Lifestyles Empowering Change (FLEX) Intervention for Adolescents with Type 1 Diabetes: Randomized Clinical Trial Results for Effect on Metabolic Status, Diabetes Related Behaviors, and Quality of Life. *The Lancet. Child & adolescent health*, *2*(9), 635. https://doi-org/10.1016/S2352-4642(18)30208-6

Mosnaim, G., Li, H., Martin, M., Richardson, D., Belice, P. J., Avery, E., Ryan, N., Bender. B., & Powell, L. (2013). The impact of peer support and mp3 messaging on adherence to inhaled corticosteroids in minority adolescents with asthma: a randomized, controlled trial. *The Journal of Allergy and Clinical Immunology: In Practice*, *1*(5), 485-493. https://doi.org/10.1016/j.jaip.2013.06.010

Mulvaney, S. A., Rothman, R. L., Wallston, K. A., Lybarger, C., & Dietrich, M. S. (2010). An internet-based program to improve self-management in adolescents with type 1 diabetes. *Diabetes care*, *33*(3), 602-604. https://doi.org/10.2337/dc09-1881

Naar-King, S., Ellis, D., King, P. S., Lam, P., Cunningham, P., Secord, E., Bruzzese, J. M., & Templin, T. (2014). Multisystemic therapy for high-risk African American adolescents with asthma: a randomized clinical trial. *Journal of consulting and clinical psychology*, *82*(3), 536. https://doi-org/10.1037/a0036092

Nansel, T. R., Iannotti, R. J., Simons-Morton, B. G., Cox, C., Plotnick, L. P., Clark, L. M., & Zeitzoff, L. (2007). Diabetes personal trainer outcomes: short-term and 1-year

outcomes of a diabetes personal trainer intervention among youth with type 1 diabetes. *Diabetes care*, *30*(10), 2471-2477. https://doi-org.uea.idm.oclc.org/10.2337/dc06-2621

Raiff, B. R., Barrry, V. B., Ridenour, T. A., & Jitnarin, N. (2016). Internet-based incentives increase blood glucose testing with a non-adherent, diverse sample of teens with type 1 diabetes mellitus: a randomized controlled Trial. *Translational behavioral medicine*, *6*(2), 179-188. https://doi-org.uea.idm.oclc.org/10.1007/s13142-016-0397-5

Rikkers-Mutsaerts, E. R. V. M., Winters, A. E., Bakker, M. J., van Stel, H. F., van der Meer, V., de Jongste, J. C., Sont, J. K., & SMASHING Study Group. (2012). Internet-based self-management compared with usual care in adolescents with asthma: A randomized controlled trial. *Pediatric pulmonology*, *47*(12), 1170-1179. https://doi-org.uea.idm.oclc.org/10.1002/ppul.22575

Smaldone, A., Findley, S., Manwani, D., Jia, H., & Green, N. S. (2018). HABIT, a randomized feasibility trial to increase hydroxyurea adherence, suggests improved health-related quality of life in youths with sickle cell disease. *The Journal of pediatrics*, *197*, 177-185. https://doi.org/10.1016/j.jpeds.2018.01.054

Stanger, C., Lansing, A. H., Scherer, E., Budney, A., Christiano, A. S., & Casella, S. J. (2018). A web-delivered multicomponent intervention for adolescents with poorly controlled Type 1 diabetes: a pilot randomized controlled trial. *Annals of Behavioral Medicine*, *52*(12), 1010-1022. https://doi.org/10.1093/abm/kay005

Stinson, J., Kohut, S. A., Forgeron, P., Amaria, K., Bell, M., Kaufman, M., Luca, N., Luca, S., Harris, L., Victor, C., & Spiegel, L. (2016). The iPeer2Peer program: a pilot randomized controlled trial in adolescents with juvenile idiopathic

arthritis. *Pediatric Rheumatology*, *14*(1), 48. https://doi-org.uea.idm.oclc.org/10.1186/s12969-016-0108-2

Stinson, J. N., Lalloo, C., Hundert, A. S., Campillo, S., Cellucci, T., Dancey, P., Duffy, C., Ellsworth, J., Feldman, B. M., Hulber, A. M., & Johnson, N. (2020). Teens Taking Charge: A Randomized Controlled Trial of a Web-Based Self-Management Program With Telephone Support for Adolescents With Juvenile Idiopathic Arthritis. *Journal of medical Internet research*, *22*(7), e16234. https://doi-org/10.2196/16234

Stinson, J. N., McGrath, P. J., Hodnett, E. D., Feldman, B. M., Duffy, C. M., Huber, A. M., Tucker, L. B., Hetherington, C. R., Tse, S. M., Spiegel, L. R., & Campillo, S. (2010). An internet-based self-management program with telephone support for adolescents with arthritis: a pilot randomized controlled trial. *The Journal of Rheumatology*, *37*(9), 1944-1952. https://doi.org/10.3899/jrheum.091327

Tseng, T. J., & Chang, A. M. (2020). Theoretical asthma self-management program for Taiwanese adolescents with self-efficacy, outcome-expectancy, health behaviour, and asthma symptoms: A randomized controlled trial. *Contemporary Clinical Trials Communications*, *19*, 100624. https://doi.org/10.1016/j.conctc.2020.100624

Whittemore, R., Liberti, L. S., Jeon, S., Chao, A., Minges, K. E., Murphy, K., & Grey, M. (2016). Efficacy and implementation of an Internet psychoeducational program for teens with type 1 diabetes. *Pediatric diabetes*, *17*(8), 567-575. https://doi-org.uea.idm.oclc.org/10.1111/pedi.12338

Willis, N., Milanzi, A., Mawodzeke, M., Dziwa, C., Armstrong, A., Yekeye, I., Mtshali, P., & James, V. (2019). Effectiveness of community adolescent treatment supporters (CATS) interventions in improving linkage and retention in care, adherence to ART and psychosocial well-being: a randomised trial among adolescents living with HIV

in rural Zimbabwe. *BMC Public Health*, *19*(1), 117. https://doi-org.uea.idm.oclc.org/10.1186/s12889-019-6447-4

Wysocki, T., Harris, M. A., Buckloh, L. M., Mertlich, D., Lochrie, A. S., Mauras, N., & White, N. H. (2007). Randomized trial of behavioral family systems therapy for diabetes: maintenance of effects on diabetes outcomes in adolescents. *Diabetes Care*, *30*(3), 555-560. https://doi-org.uea.idm.oclc.org/10.2337/dc06-1613

Supplementary Material Continued: PRISMA Statement (Moher et al., 2009)

From: Moher D, Liberati A, Tetzlaff J, Altman DG, The PRISMA Group (2009). Preferred Reporting Items for Systematic Reviews and Meta-Analyses: The PRISMA Statement. PLoS Med 6(7): e1000097. doi:10.1371/journal.pmed1000097
For more information, visit: **www.prisma-statement.org**.

This is under the Creative Commons Attribution License and has been appropriately cited and adapted for use in the present review.

Section/topic	#	Checklist item	Reported on page #
TITLE			
Title	1	Identify the report as a systematic review, meta-analysis, or both.	81
ABSTRACT			
Structured summary	2	Provide a structured summary including, as applicable: background; objectives; data sources; study eligibility criteria, participants, and interventions; study appraisal and synthesis methods; results; limitations; conclusions and implications of key findings; systematic review registration number.	82
INTRODUCTION			
Rationale	3	Describe the rationale for the review in the context of what is already known.	83-86
Objectives	4	Provide an explicit statement of questions being addressed with reference to participants, interventions, comparisons, outcomes, and study design (PICOS).	86
METHODS			

Protocol and registration	5	Indicate if a review protocol exists, if and where it can be accessed (e.g., Web address), and, if available, provide registration information including registration number.	87
Eligibility criteria	6	Specify study characteristics (e.g., PICOS, length of follow-up) and report characteristics (e.g., years considered, language, publication status) used as criteria for eligibility, giving rationale.	89-90
Information sources	7	Describe all information sources (e.g., databases with dates of coverage, contact with study authors to identify additional studies) in the search and date last searched.	87
Search	8	Present full electronic search strategy for at least one database, including any limits used, such that it could be repeated.	210-215
Study selection	9	State the process for selecting studies (i.e., screening, eligibility, included in systematic review, and, if applicable, included in the meta-analysis).	90
Data collection process	10	Describe method of data extraction from reports (e.g., piloted forms, independently, in duplicate) and any processes for obtaining and confirming data from investigators.	91
Data items	11	List and define all variables for which data were sought (e.g., PICOS, funding sources) and any assumptions and simplifications made.	91
Risk of bias in individual studies	12	Describe methods used for assessing risk of bias of individual studies (including specification of whether this was done at the study or outcome level), and how this information is to be used in any data synthesis.	90-91
Summary measures	13	State the principal summary measures (e.g., risk ratio, difference in means).	91-92

Synthesis of results	14	Describe the methods of handling data and combining results of studies, if done, including measures of consistency (e.g., I^2) for each meta-analysis.	91-92
Section/topic	**#**	**Checklist item**	**Reported on page #**
Risk of bias across studies	15	Specify any assessment of risk of bias that may affect the cumulative evidence (e.g., publication bias, selective reporting within studies).	91-93
Additional analyses	16	Describe methods of additional analyses (e.g., sensitivity or subgroup analyses, meta-regression), if done, indicating which were pre-specified.	91-93
RESULTS			
Study selection	17	Give numbers of studies screened, assessed for eligibility, and included in the review, with reasons for exclusions at each stage, ideally with a flow diagram.	93-94
Study characteristics	18	For each study, present characteristics for which data were extracted (e.g., study size, PICOS, follow-up period) and provide the citations.	97-117
Risk of bias within studies	19	Present data on risk of bias of each study and, if available, any outcome level assessment (see item 12).	97-118 and 216-218
Results of individual studies	20	For all outcomes considered (benefits or harms), present, for each study: (a) simple summary data for each intervention group (b) effect estimates and confidence intervals, ideally with a forest plot.	118-126
Synthesis of results	21	Present results of each meta-analysis done, including confidence intervals and measures of consistency.	118-126

Risk of bias across studies	22	Present results of any assessment of risk of bias across studies (see Item 15).	97-118 and 216-218
Additional analysis	23	Give results of additional analyses, if done (e.g., sensitivity or subgroup analyses, meta-regression [see Item 16]).	118-126
DISCUSSION			
Summary of evidence	24	Summarize the main findings including the strength of evidence for each main outcome; consider their relevance to key groups (e.g., healthcare providers, users, and policy makers).	126-128
Limitations	25	Discuss limitations at study and outcome level (e.g., risk of bias), and at review-level (e.g., incomplete retrieval of identified research, reporting bias).	128-129
Conclusions	26	Provide a general interpretation of the results in the context of other evidence, and implications for future research.	129-133
FUNDING			
Funding	27	Describe sources of funding for the systematic review and other support (e.g., supply of data); role of funders for the systematic review.	81

Chapter Five: Additional Methodology

This chapter contains additional information about the methods employed in both reviews that are not included in the publications. Although there are not any length restrictions imposed by the chosen journal, this additional information was not included to ensure the papers were succinct and focussed.

Qualitative Systematic Review: Additional Methodology

Development of the Inclusion and Exclusion Criteria

Many discussions within the research team were had around the inclusion and exclusion criteria during the preliminary searching and protocol development stages. Initially, the research team considered including papers which had some data related to adolescents' and their parents' experience of managing treatment adherence, even if the focus of the paper was not on this topic specifically. However, after reading some papers it was decided that those which did not focus directly on the issue, such as those which explored the experience of coping with a chronic illness or the adolescent-parent relationship more generally, did not provide enough rich and in-depth data on the topic of interest and therefore did not answer the review question.

Further, including papers which only included one half of the dyad's experience (i.e., either adolescents or their parents) or those which included others' experiences outside of the dyad were also considered. However, when reading these papers in the development stage, it was felt that they did not directly answer the research question as they did not address relational issues between the dyad. Further, if these papers were included, it would have meant that rather the entire findings section being extracted, only direct quotes from adolescents and/or their parents would be extracted. This would have meant that contextual data would have been lost. The studies excluded based on these decisions may have offered additional data and contributed the overall synthesis, but they

are justified as it meant that included papers were exploring similar questions to that of the review and lead to an in-depth and rich analysis of the entire findings section of included papers.

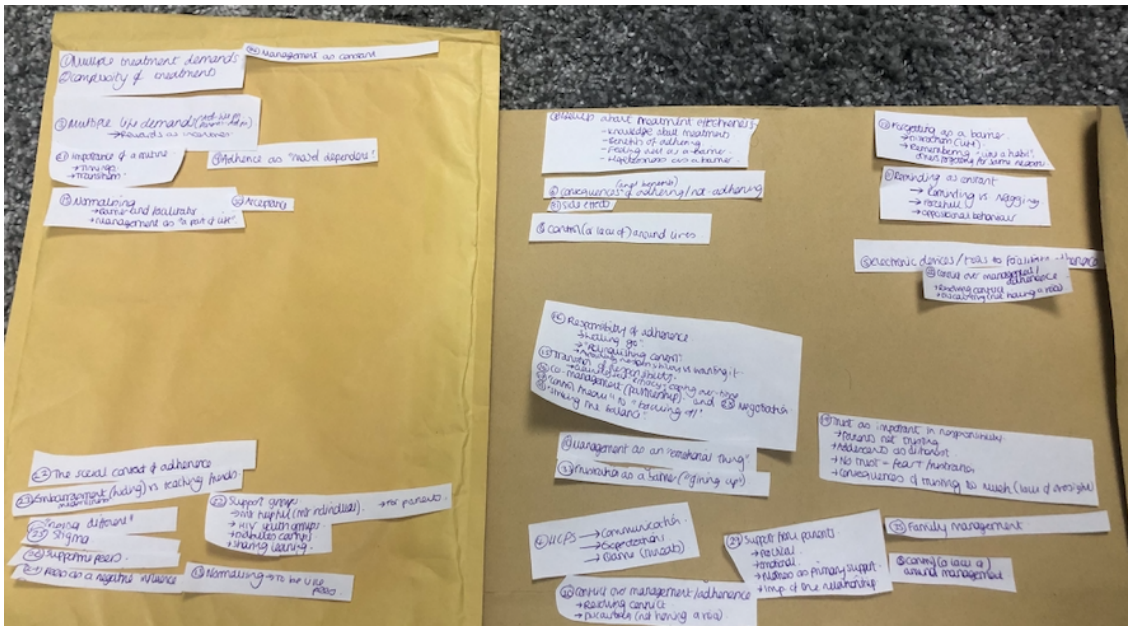
Further Details on the Analytic Process

Data Analysis Process and Data Transparency. Thomas and Harden's (2008) three stage thematic synthesis was followed, which included 1) line-by-line coding, 2) 'descriptive theme' development and finally, 3) development of analytic themes.

1) All data, including parents' and adolescents' direct quotes and authors interpretations of the data was coded. The codes were a word or very brief statement which summarised the meaning of or nature of the text. Some examples of codes include:
    a. Multiple treatments
    b. Responsibility
    c. Consequences
2) Once data from all included studies had been coded, the codes were organised into related areas and further refined. The first author did this independently before having in depth discussions with both the second and fourth authors and making changes to this (see Figure 5.1 for a photo of this process).
3) Finally, the third stage involved the development of 'analytic themes' in order to 'go beyond' the descriptions. This was achieved through the descriptive themes being reviewed in relation to the review questions and thus considering relational factors and both the adolescent and parent perspectives within each theme. Multiple discussions within the research team were had around the analytic themes and the first author made detailed notes of thoughts and ideas.

Figure 5.1

Photo of the Second Stage of the Analysis Process



Personal Reflective Account of the Analytic Process. When initially reading papers (during the data familiarisation stage), the first author was struck by the lack of researcher reflexivity and contextual information about participants. Whilst the lack of reflexivity was captured by the quality ratings, it was also felt that there was not enough information about the process of data collection (i.e., about the process of 'being with' participants in the room, particularly given the relational nature of the review). The first author reflected on this during the analysis process and compared it to their previous experiences of conducting qualitative analysis when they had collected the data themselves and had detailed contextual data. This had implications for the analysis, because it was more difficult for them to remain inductive and aligned to the critical realist positioning (where contextual data is important). This made them reflect on the power of researchers in relation to reporting and missing the 'voices' of adolescents and their families and thus the importance of future researchers considering reflexivity and contextual data.

Systematic Review and Meta-Analysis: Additional Methodology

Justification for Quality Assessment Tool

The Cochrane Collaboration's risk of bias tool (Higgins & Green, 2008) was used to assess study quality. This was decided within the research team during the development of the protocol, as it is considered the gold standard for assessing quality of RCTs. This is the quality assessment tool recommended by Cuijpers' (2016), which was the practical guide consulted throughout the meta-analyses process. The choice of using this tool was also made because the most recent meta-analysis in the area (Pai & McGrady, 2014) also used this. Therefore, comparisons in ratings between this review and that of the previous meta-analysis could be made.

Missing Data

When data needed for the meta-analyses were not fully reported in the papers, study authors were contacted by the first author. They were emailed up to four times over a two-and-a-half-month period. This was to ensure that multiple attempts to access the data were made. Where numbers and means were presented, standard deviations were calculated using other available data in the manuscripts (i.e., standard errors) if authors did not respond to requests made. This was calculated by the first author using a standardised Microsoft Excel spreadsheet and were checked by the fourth author for accuracy.

Additional Information about Data Extraction

Several decisions were made within the research team, whilst consulting relevant guidance (i.e., Cochrane), when extracting data to ensure a systematic approach was taken. First, when studies had multiple follow-ups of the same participants, data from the longest follow-up point were extracted. Taking the longest follow-up was proposed by Cochrane as a way to overcome the issue of multiple follow-up points, but it is important to note that this decision could have led to increased heterogeneity (Higgins, Li, et al., 2020). Second, if studies only had pre- and post-intervention time points, then the posttreatment data was

included in the posttreatment meta-analysis. Third, if studies had a posttreatment and follow-up time point, but reported only the follow-up data, this was included only in the follow-up meta-analysis if the posttreatment data could not be obtained. Fourth, if studies had more than two groups, then the Cochrane guidance on handling this issue was consulted (Higgins, Eldridge et al., 2020).

Finally, where studies included multiple measures of adherence (i.e., an objective and self-report measure of adherence), all data was extracted. However, for the main analysis, a selection procedure agreed by the research team was utilised (see Appendix G for adherence measure outcome selection procedure), as this is one of the established ways to deal with this issue (Cuijpers, 2016). Separate exploratory analyses for the additional outcomes not included in this main analysis were conducted, so no data was lost. For the secondary outcomes, namely quality of life and family functioning outcomes, a selection procedure was agreed in order to select the most reliable and valid measure (see Appendix H for selection procedures) and only data from the selected outcome was extracted. These selection procedures were developed through consultation with the relevant literature around different types of adherence measures to ensure the most objective and validated measure was chosen. This approach is consistent to that of previous meta-analyses in the area (e.g., Pai & McGrady, 2014). Pooling all of the papers together in one analysis was justified, as it meant that moderation and subgroup analyses could be conducted and that exploratory analyses examining the effects of different measurements of adherence could also be conducted.

Of note, where only means and standard deviations of each item of a relevant measure were reported, multiple attempts were made to study authors for the descriptive data for the total outcome mean and standard deviation. When this wasn't provided, the first author averaged effect sizes of each item to create one effect size for the outcome.

Exploratory Analyses

Given that many studies used multiple measures of adherence, exploratory analyses examined the impact these different types of adherence measurement. All studies with an adolescent self-report measure of adherence, all studies with a parent self-report measure, all studies with an objective measure of adherence and finally all studies with a joint adolescent and parent measure were pooled separately. This meant that studies with multiple measures could be included in multiple analyses, but to ensure that data from the same participants were not included in the same analysis, a paper was only included once in each analysis.

Chapter Six: Additional Results

This chapter contains additional information about results from the systematic review and meta-analysis presented in chapter four that are not included in the publication to ensure that it was succinct and focussed.

Systematic Review and Meta-Analysis: Additional Results

Missing Data

Twenty-three studies did not provide all data needed for the analyses. Therefore, the corresponding authors of all of these studies were contacted.

For the primary analysis, data for 11 studies were provided by study authors. Three were calculated using other data reported in the papers (i.e., standard errors). Data for nine studies were not provided and could not be calculated and were excluded from the analysis.

For studies that included a follow-up time period and included a measure of adherence, authors were contacted where there were missing data. Data for five studies were provided. One author could not provide data for one study, so this was therefore removed from this secondary analysis.

For the quality of life outcomes at posttreatment, data for four studies were provided by authors, but data for a further four were not provided and thus these studies were excluded from this analysis. At follow-up, data for four studies were provided by authors, but data for one study could not be provided.

Finally, for the family functioning outcomes, data for four studies were provided at posttreatment, but data for a further two studies could not be provided. At follow-up, data for four studies were provided, but data for another study (which was one of the secondary analysis papers) were not provided by the author and was therefore excluded from this secondary analysis.

Taken together, of the 36 studies (from 38 papers) included in the qualitative synthesis, a total of 30 studies (from 31 papers) were included in the quantitative analysis. This is due to seven studies not having any data for any of the analyses. However, a decision was taken within the research team that studies with any relevant data would be included in the relevant analyses. This meant that only 27 were included in the main analysis, as three papers did not provide data needed to be included for this analysis but had relevant data for other meta-analyses.

Additional Information About Characteristics of Included Studies

Twenty-three studies had a broad age range of adolescent samples (with five years or more in between the lowest and oldest participant), whilst 13 studies had a narrow age range (with four or less years between the oldest and youngest participant).

Additional information regarding the interventions of included studies, including theoretical framework/theory, professional delivering the intervention and treatment components can be found in Table 6.1.

Only 13 interventions were grounded explicitly in a theoretical model or framework and of these, many were grounded in multiple theories and/or models. These included social cognitive theory (n = 4), cognitive-behavioural theory (n = 2), family systems theory (n = 1), broad-based ecological framework (n = 1), social-ecological model (n =1), broaden and build theory (n = 2), transtheoretical model (n =2), health-belief model (n = 2), learning theory (n = 1), self-determination theory (n = 1), the common-sense model of self-regulation (n = 1), theory of reasoned action (n = 1), an integration of social and health psychology theory (n = 1) and Bandura's self-efficacy model (n = 1).

Of those studies who reported the healthcare professional delivering the intervention (n = 4 not reported), there was a broad range of professionals and non-professionals including psychologists (n = 6), social workers (n = 3), researchers (n = 3),

pharmacists (n = 1), other healthcare workers (n = 4), trained workers (including peer mentors, n = 6), masters-level clinicians (including psychology and social work n = 4) and non-healthcare professionals with an undergraduate degree in psychology or healthcare (n = 2). Finally, five studies were delivered by an automated web-based component.

Overall, this suggests that very few were grounded in a theoretical framework, a broad range of professionals delivered the interventions, and a range of treatment approaches and components were used. Given this variability across all of the included studies, no additional exploratory analyses were conducted on this data, but it highlights issues within this body of research.

Table 6.1

Additional Details about Study Interventions

Study Table 6.1	Theoretical framework	Professional delivering the intervention	Intervention components [a]
Bhana et al (2014)	N/A	Lay counsellors and one masters-level psychologist	F, S, E, C
Bruzzee et al (2008)	Social cognition theory, cognitive behavioural theory, and two forms of family systems theory, parenting styles, and behavioural family systems theory	Psychologists	C-B, B, F
Bruzzee et al (2010)	Social cognitive theory	Trained health educators	C, S, E
Carlsen et al (2017)	N/A	Not reported	E, F, T, S
Chawana et al (2017)	N/A	Trained field workers	T, O, B, S
Davis et al (2019)	Social cognitive theory	Not reported	E, F, B, C
Ellis et al (2007)	Cognitive and behaviour theory	Psychologist/social worker	B, E, C, S, F, O
Ellis et al (2012)	Broad-based ecological framework	Five masters-level therapists with varied backgrounds (three psychologists, two social workers)	F, C-B, B, E, O

Study	Theoretical framework	Professional delivering the intervention	Intervention components [a]
Table 6.1			
Ellis et al (2019)	N/A	Community health workers	B, C-B, F, S, E, O
Goyal et al (2017)	N/A	Automated Web-based content	S, B, T, E
Green et al (2017)	N/A	Community healthcare workers	S, F, T, E, B
Greenley et al (2015)	N/A	Doctorate clinical psychology students	B, F, T
Hommel et al (2012)	N/A	Doctoral level clinical psychologists or postdoctoral psychology fellows	B, F, O, E
Jaser et al (2014)	Broaden and build theory	Not reported	F, C, B, T
Jaser et al (2019)	Broaden and build theory	Research assistants	E, B, F, T, C
Jaser et al (2020)	N/A	Trained member of the research team	E, C, B, T
Johnson et al (2015)	N/A	Automated Web-based content	T, B, E
Joseph et al (2007)	Transtheoretical model and health belief model	Automated Web-based content	B, E, T
Kichler et al (2013)	N/A	Licensed psychologist	F, S, E, B, C, C-B
Kohut et al (2016)	N/A	Trained peer mentors	T, S, E, B

Study	Theoretical framework	Professional delivering the intervention	Intervention components [a]
Table 6.1			
Kosse et al (2019)	The common-sense model of self-regulation	Pharmacists	E, M, B, T, S
Mayer-Davis et al (2018)	Health belief model, transtheoretical model and theory of reasoned action and integration of theory in social and health psychology	'Coaches' who were already members of an existing T1D medical care team (e.g., dietitian, nurse, and certified diabetes educator)	C, B, E, S, T, F
Mosnaim et al (2013)	N/A	Social workers	B, S, T, E, C
Mulvaney et al (2010)	Learning, social-cognitive and self-determination theories	Automated Web-based content	T, S, C, B
Naar-King et al (2014)	Social-ecological model	Master's-level therapists with varied backgrounds (one psychologist, three social workers)	F, C-B, B, E, O
Nansel et al (2007)	N/A	Diabetes personal trainer (non-professionals) – bachelor's degree and/or graduate students in health-related fields	B, C-B, E
Raiff et al (2016)	N/A	Clinician or research assistant	B, T, C
Rikkers-Mutsaerts et al (2012)	N/A	Not reported	T, E, B, S, O
Stanger et al (2018)	N/A	Master level clinicians	C-B, F, T

Study	Theoretical framework	Professional delivering the intervention	Intervention components [a]
Table 6.1			
Stinson et al (2010)	N/A	A non-health-care professional with an undergraduate degree in psychology	E, S, B, C, T, F
Stinson et al (2016)	N/A	Trained peer mentors (trained by research staff)	T, S, E, B
Stinson et al (2020)	N/A	Trained health coach – non-healthcare professional	C-B, T, S, E, F
Tseng et al (2020)	Bandura's self-efficacy model	Researcher	T, F, E, B
Whittemore et al (2016)	N/A	Automated Web-based content	T, S, E, C-B
Willis et al (2019)	N/A	Community Adolescent Treatment Supporters (peer supporters)	S, O, E
Wysocki et al (2007)	N/A	Psychologist or licensed social worker	B, E, C, F

Note. This table outlines additional information regarding study interventions including the explicit theory outlined in the papers, the professionals who delivered the interventions and intervention components.

[a] Using Graves et al.'s (2010); Kahana et al.'s (2008); and Pai and McGrady's (2014) classifications: E = Educational element e.g., provided some teaching and/or information/psychoeducation about the chronic illness and its treatment; B = Behavioural element e.g., problem-solving, rewards, positive reinforcement; C = Cognitive element e.g., motivation enhancing, positive psychology techniques; C-B = Cognitive-behavioural element e.g., The intervention was based (even in part) on cognitive-behavioural principles; F = Family element e.g., family therapy, parent sessions and/or training; S = Social element e.g., peer support, peer chat functions, groups; T = Technology-based element e.g., text messages, websites; O = Organisational element e.g., reducing barriers to adherence, simplifying the regimen, changing ways of working in the systems surrounding the adolescent.

Additional Risk of Bias Information

No studies were deemed to be high risk of bias in relation to sequence generation, and thus none were excluded. Some provided limited information, so were rated as unclear risk on this domain. Similarly, studies were only rated as low or unclear risk in relation to allocation concealment and those where it was unclear did not provide enough information. Only two studies were rated as high risk on blinding on outcome assessment, as they specifically reported that outcome assessors were unblinded, the remaining studies were all rated as low or unclear on this domain. Many studies did not report if outcome assessors were blinded. Most studies were rated as low risk for incomplete outcome data as they provided adequate information on the number of dropouts and reasons for this and only two studies were rated as high risk on this domain. In terms of selective outcome reporting, no studies were rated as high risk, and many were unclear due to a published protocol not being available. No other biases were identified. The one cluster RCT (Kosse et al., 2019) was rated as high risk due to issues with the timing and identification of participants, measurement of outcomes and selection of the reported result.

Results of Exploratory Analyses

Effects of all exploratory analyses at posttreatment and follow-up are presented in Table 6.2.

Different Types of Adherence Measurement at Posttreatment.

Adolescent Self-Report. Overall, a significant small effect was found, with significant heterogeneity. A sensitivity analysis which excluded the high risk of bias studies are also presented and the forest plot for this analysis is presented in Figure 6.1. The outcomes indicate that study bias had an impact on the effect size found, as this increased, but heterogeneity remained significant. A sensitivity analysis which excluded the cluster RCT study did not lead to any considerable changes. The results of the

moderation and subgroup analyses are also presented in Table 6.2. Following the Holm-Bonferroni correction method, no moderators were significant.

Parent Self-Report. This analysis yielded a negligible, non-significant effect. The removal of high risk of bias studies did not lead to any considerable changes and the forest plot for this analysis is presented in Figure 6.2.

Table 6.2

Outcomes of the Exploratory Analyses at Posttreatment and Follow-up

		k	*g*	95% CI	p-value	I^2 (*Q, p-value*)
Adolescent Self-Report Posttreatment						
Main analysis (N = 2189)		21	0.18	0.05 to 0.30	**.005**	45% (39.19, **.006**)
Excluding high risk of bias studies (*N* = 1299)		13	0.24	0.07 to 0.40	**.005**	46% (24.31, **.018**)
Excluding cluster study (N = 1955)		20	0.18	0.04 to 0.32	**.011**	50% (39.08, **.004**)
Moderators [a]	Subgroups					
Sample (*Qb* = 0.55, *p* = 0.46)	Poor adherence	4	0.35	-0.01 to 0.72	.059	0% (2.54, .468)
	Other	9	0.20	-.003 to 0.39	.054	62% (21.10, **.007**)
Diabetes (*Qb* = 5.18, *p* = 0.0228*)	Diabetes	6	0.04	-0.19 to 0.27	.749	63% (13.45, **.020**)
	Other	7	0.40	0.19 to 0.60	**<.001**	0% (4.40, .623)
Asthma (*Qb* = 3.35, *p* = 0.067)	Asthma	5	0.40	0.16 to 0.63	**.001**	0% (2.00, .735)
	Other	8	0.24	-0.13 to 0.32	.412	60% (17.66, **.014**)
Parental involvement (*Qb* = 0.92, *p* = 0.3388)	Adolescent-only	5	0.34	0.05 to 0.63	.021*	0% (3.06, .548)
	Parents involved	8	0.16	-.067 to 0.39	.166	66% (20.53, **.005**)
Delivery (*Qb* = 1.73, *p* = 0.1887)	Face to face	9	0.30	0.10 to 0.50	**.003**	13% (9.23, .323)
	Remote	4	0.04	-0.30 to 0.38	.838	78% (13.37, **.004**)
Adolescent Self-Report Follow-up [b]						
Main analysis (N = 778)		8	-0.05	-0.27 to 0.18	.683	52% (15.42, **.031**)
Excluding high risk of bias studies (N = 614)		6	-0.01	-0.34 to 0.32	.949	70% (13.90, **.016**)
Parent Report Posttreatment [b]						
Main analysis (N = 986)		10	0.05	-0.07 to 0.18	.393	0% (5.52, .480)
Excluding high risk of bias studies (*N* = 570)		7	0.01	-0.15 to 0.18	.900	0% (8.10, .523)

	k	*g*	95% CI	p-value	I^2 (*Q, p-value*)
Parent Report Follow-up [b]					
Main analysis (N = 301)	4	-0.06	-.487 to 0.36	.772	69% (8.94, **.030**)
Objective Posttreatment [b]					
Main analysis (N = 423)	7	0.08	-0.54 to 0.71	.790	89% (39.02, **<.001**)
Excluding high risk of bias studies (*N* = 382)	6	0.35	.0002 to 0.70	**.049**	62% (11.91, **.360**)
Objective Follow-up [b]					
Main analysis (N = 301)	5	0.13	-.076 to 0.32	.221	0% (4.04, .401)
Joint Adolescent and Parent Measures [b]					
Main analysis (N = 292)	4	0.41	0.18 to 0.64	**<.001**	.01% (2.95, .400)

Note. Table 6.2 continued. *N* = number of participants. k = number of studies. *g* = Hedges' *g*. CI = confidence interval. p-value = significance. Significant effects are indicated in bold. Positive *g* indicates that the participants in the intervention group had better adherence.

[a] Moderator and subgroup analyses excluded high risk of bias studies. [b] No moderator or subgroup analyses were possible due to less than four studies per subgroup.

[*]Non-significant following the Holm-Bonferroni correction method.

Figure 6.1

Forest Plot for Adolescent Self-Report Measures at Posttreatment (Excluding High Risk of Bias Studies)

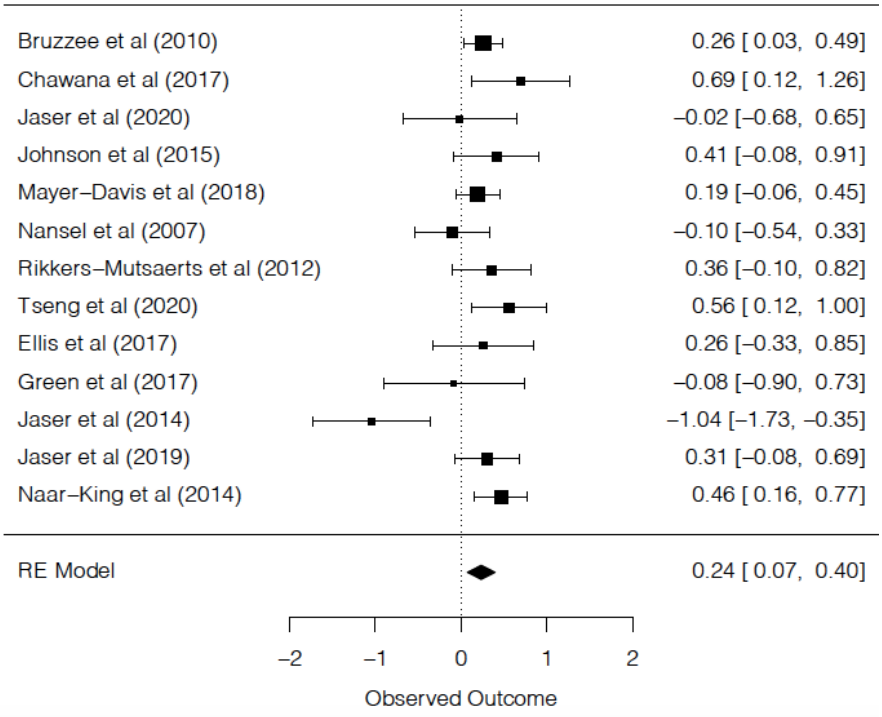
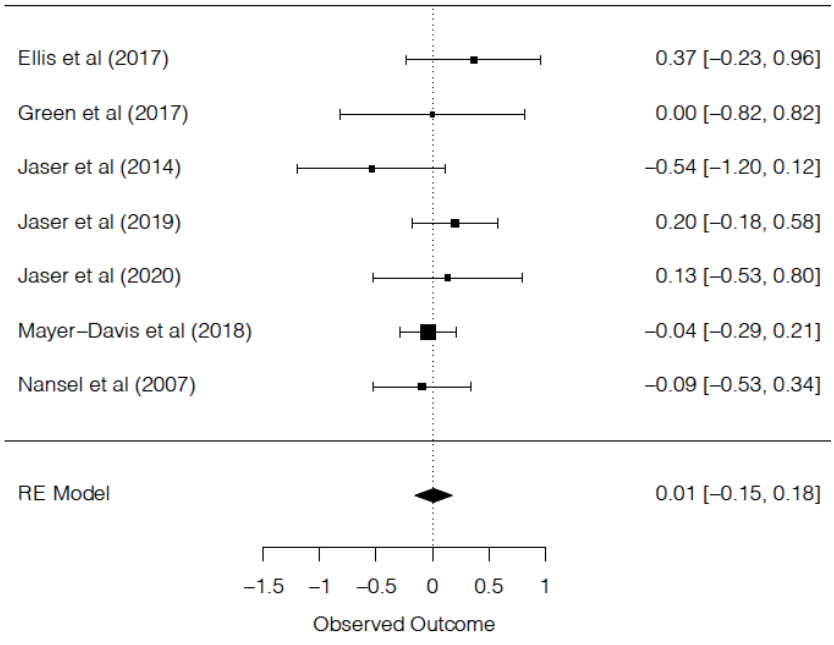


Figure 6.2

Forest Plot for Parent Report Measures at Posttreatment (Excluding High Risk of Bias Studies)

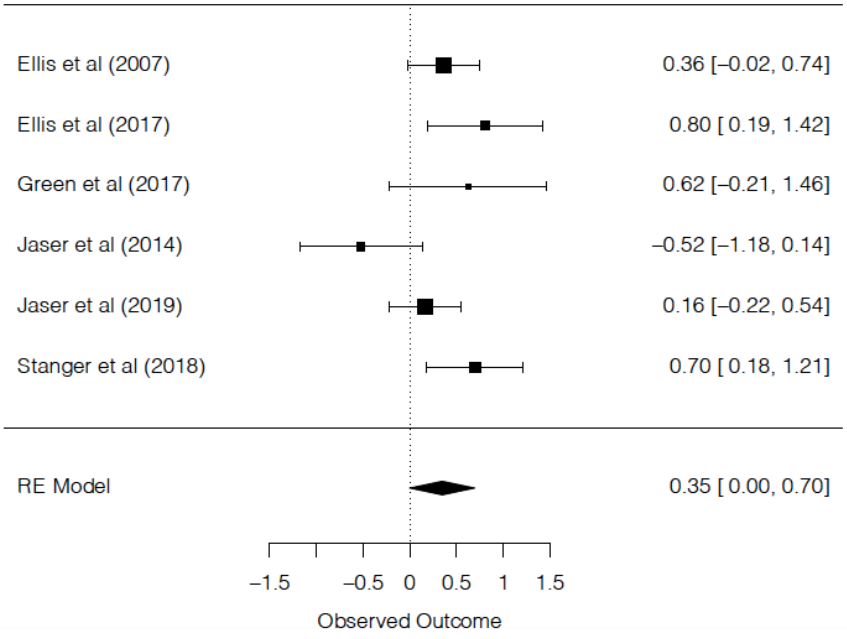


Objective Measures. This analysis yielded a negligible, non-significant effect. However, the removal of high risk of bias studies considerably affected findings and resulted in a significant small effect, but heterogeneity remained significant. The forest plot for this analysis is presented in Figure 6.3.

For both parent report and objective measures, a cluster sensitivity analysis was not needed, and no moderation or subgroup analyses could be performed due to an insufficient number of studies in each group.

Figure 6.3

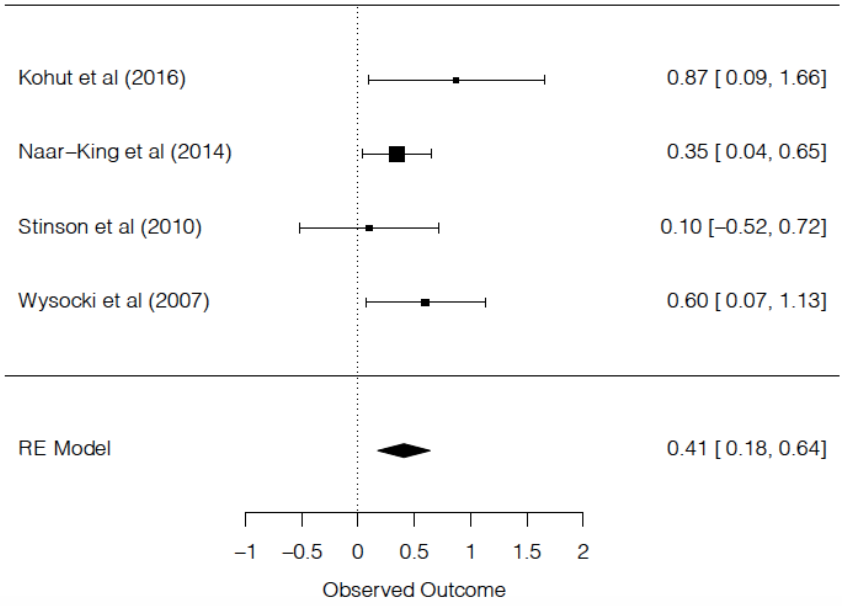
Forest Plot for Objective Measures at Posttreatment (Excluding High Risk of Bias Studies)



Joint Adolescent and Parent Measures. This yielded a significant small effect. The forest plot for this analysis is presented in Figure 6.4. It was not possible to perform any sensitivity analyses, moderator or subgroup analyses due to an insufficient number of studies.

Figure 6.4

Forest Plot for Joint Adolescent and Parent Measures at Posttreatment (Including High Risk of Bias Studies)



Different Types of Adherence Measurement at Follow-Up.

Adolescent Self-Report. This yielded a negative but negligible non-significant effect. The removal of high risk of bias studies did not lead to any considerable changes.

Parent Self-Report. This yielded a negative but negligible non-significant effect. It was not possible to perform any sensitivity analyses, moderator or subgroup analyses due to an insufficient number of studies.

Objective Measures. This yielded a small, non-significant effect. No sensitivity analyses were needed, and no moderation or subgroup analyses could be performed due to an insufficient number of studies in each group.

Joint Adolescent and Parent Measures. It was not possible to perform any analyses as there was only one study who used a joint adolescent and parent measure at follow-up.

Overall Findings. Exploratory analyses examined effect sizes of different adherence measures at both posttreatment and follow-up. Objective measures (with high

risk of bias studies removed), adolescent self-report and joint adolescent and parent reports showed significant small effects at posttreatment. Although not compared statistically, effect sizes were largest on joint adolescent and parent outcome measures and were negligible for parent report. This suggests that there are some differences between effects of different types of adherence measurement.

At follow-up, adolescent and parent report measures and objective measures produced small, non-significant effects. There were insufficient studies to calculate effect sizes for joint measures at follow-up. These findings indicate that there is little evidence for intervention efficacy at follow-up, which is consistent with the findings from the primary and secondary analyses. Interestingly, objective measures produced the largest effects at follow-up, but these were still small and non-significant.

Moderation and subgroup analyses were only possible on adolescent report studies. Similarly to the primary analysis, after correcting for multiple comparisons, no moderation analyses were significant. However, effect sizes were larger for the subgroups of studies which specifically targeted adolescents with poor adherence, those which delivered interventions face-to-face and were delivered only to adolescents. They were more efficacious for samples of adolescents that did not have a diagnosis of Diabetes and for those with Asthma.

Assessment of Publication Bias

The outcome of all rank correlation tests, and funnel plots can be found in Appendix F. Publication bias for the primary and secondary analyses are outlined in chapter four. The funnel plots indicated that missing studies were estimated for parent report measures and joint measures at posttreatment and objective adherence measures at follow-up, but asymmetry was non-significant.

Holm-Bonferroni Correction

Appendix I provides details about the Holm-Bonferroni correction (Holm, 1979) used for moderation and subgroup analyses. Due to an insufficient number of studies, these

were only possible for the primary analysis at posttreatment and for those studies which used an adolescent report of adherence at posttreatment in the exploratory analysis.

Chapter Seven: Discussion and Critical Evaluation

The purpose of this final chapter is to pull together the whole body of work outlined in this thesis. An overview of the rationale and findings of both reviews, will be discussed, followed by synthesis of the findings. Overall strengths and limitations of the work and how this could be improved in relation to clinical, theoretical and research implications will also be outlined.

Overall Discussion

This thesis aimed to fill the aforementioned gaps in the literature, namely to synthesise and examine existing literature which explored adolescent-parent dyadic experience of adhering to treatment in chronic illness. Second, to examine the efficacy of psychological interventions to promote adherence, quality of life and family functioning outcomes in this population. As aforementioned, adherence is particularly problematic for adolescents with chronic illness. This is thought to be related to the complex and challenging nature of adherence during a period of considerable developmental changes in which adolescents are expected to be more autonomous in the management of their illness (Yeo & Sawyer, 2005; Viner & Christie, 2005). Given the impact of poor adherence not only on adolescent's physical health and quality of life, but on healthcare utilisation and costs (Kahana et al., 2008; Modi & Driscoll, 2020; Pai & McGrady, 2014), it is vital that research in this area is synthesised and understood.

A qualitative systematic review was needed in the area in order to understand how adherence is managed within a changing parent-adolescent relationship during this period. This had been overlooked by previous reviews in the area (i.e., Hanghøj & Boisen, 2013; Lerch & Tharne, 2019; Lindsay et al., 2011), which had focussed predominately on adolescents' experience. These reviews had included both qualitative and quantitative studies, which prevented the possibility of a rich qualitative analysis. These gaps were problematic given that parents play a vital role in their adolescent's adherence (Denison et al., 2015; Luo et al., 2020; Rapoff, 2010; Williams-Reade et al., 2019). Therefore, the

review outlined in chapter two filled these aforementioned gaps. This review drew on ideas from contemporary theories of paediatric self-management that have highlighted the importance of going beyond the individual and understanding adherence within the context of the family system (Grey et al., 2015; Ryan & Sawkin, 2009). Through including only qualitative studies, a deeper understanding into the relational issues and complexity around managing adherence within this dyadic relationship was gained.

More specifically, the review found that trying to manage adherence and preserve 'normal' life within a social context, adolescents and parents have different priorities and beliefs about treatment and adherence. Forgetting was a key barrier to adherence, which had consequences for the parent-child relationship. This is alongside the dyad experiencing transitions around their roles and responsibility in relation to management of the illness. Aspects of the relationship itself including trust, negotiation and collaboration were identified as key in enabling parents and adolescents navigate the complexity.

The systematic review and meta-analysis presented in chapter four was justified given that despite a wealth of adherence intervention research, effect sizes observed in the most recent meta-analysis remain relatively small with significant heterogeneity (Pai & McGrady, 2014). McGrady, Ryan, Brown et al. (2015) argued that "while the iterative nature of intervention development should lead to increasing effect sizes, the observed magnitude of intervention effects has remained stagnant over the past 25 years" (pp. 721-722). Thus, exploring the developmental age of youth, which had not been adequately examined in previous meta-analyses, may have increased treatment effects. This is important given that adolescents appear to have more difficulties with adherence than other groups, which may have addressed ceiling effects reported in previous meta-analyses (Pai & McGrady, 2014). Due to there being an increasing number of RCTs recruiting only adolescent samples, a meta-analysis examining the efficacy of interventions to promote adherence in adolescents was possible and conducted as part of this thesis.

Overall, the data from thirty-six RCTs was extracted and analysed in random effects meta-analyses. Many included studies had missing data and there were issues with the quality of the studies identified. The exclusion of high risk of bias studies led to significant small effects for adherence outcomes ($g = 0.30$) at posttreatment, with non-significant heterogeneity. The small effects suggest that psychological interventions to promote adherence in adolescents specifically have limited efficacy. These effects increase when excluding studies with a high risk of bias and when examining those which targeted samples of adolescents with adherence difficulties. Therefore, future high-quality research is needed in this area.

Secondary outcomes of quality of life and family functioning were examined. The exclusion of high risk of bias studies lead to significant small effects for quality of life outcomes ($g = 0.14$) at posttreatment, with non-significant heterogeneity. Family functioning outcome produced non-significant effects. This suggests that there is little evidence of efficacy of these interventions in promoting quality of life and family functioning. However, few studies measured these secondary outcomes.

Findings indicated that there is little evidence of efficacy at follow-up. However, few studies included a follow-up time point. Moderators of intervention effectiveness were also included, which were informed by research and clinical practice in order to support dissemination. Whilst no moderators were significant, subgroup analyses indicated that effects are larger for studies that recruited adolescents with suboptimal/poor adherence and smaller for those recruiting adolescents with Diabetes (which made up the majority of the studies). These analyses were limited by the number of studies with available data. Overall, the findings indicated that future high-quality research recruiting adolescent samples with poor adherence across a range of chronic illnesses is needed.

Given that this thesis used both qualitative and quantitative research methods, the first author took a critical realistic stance. This methodological approach meant that both quantitative and qualitative studies in this body of literature could be synthesised. This

allowed for deeper understanding of the experiences of adolescents and parents when managing treatment to be gained and highlighted clear implications for adherence-promoting interventions in this population. It also informed a comprehensive examination of interventions aimed to improve adherence and adolescents' and their parents' lives, which was the overarching aim of this thesis. This means that findings of both reviews can be synthesised in order to discuss how many of the RCTs in this current body of research addressed the key suggestions outlined in the qualitative review around what adherence-promoting interventions should include. This will now be discussed in more detail.

First, the findings of the qualitative review suggested that parents should be involved in adherence-promoting interventions. Most of the 36 studies included in the meta-analysis included parents in their interventions (n = 21). However, in contrast to the findings in the qualitative review, the meta-analysis found that interventions delivered to adolescents only produced slightly larger effects than those that also involved parents. It is important to note, however, that interventions that included parents differed significantly in how much they were involved in the intervention. More specifically, most studies only included parents in some of the adolescent's sessions or offered them a separate session (n = 13), whist only eight delivered specific family-based interventions, such as multisystemic therapy (Ellis et al., 2012). It was only in these family-based interventions where the parent-adolescent relationship and communication in the context of adherence was considered, which were suggested in the qualitative review as important components for interventions to consider. The efficacy of these family-based intervention studies specifically was not quantitatively examined. However, the findings of the qualitative review suggest this could be an important avenue for future reviews as the field develops.

Second, findings from the review suggest that adolescents can perceive parental reminders as 'nagging'. Text message reminders were a central part of the interventions in seven studies included in the meta-analysis. It may be suggested that some adolescents could perceive these reminders as 'nagging' too. Therefore, researchers should consider

this when offering reminders as part of their interventions. Further, the qualitative review highlighted that in order to reduce parental anxiety and adolescent frustration in relation to 'nagging', organisational tools to support adherence, such as alarms, should involve parents as well as adolescents (i.e., parents should also receive reminders). However, only three of the eight studies which used text reminders also involved parents, where four were sent only to the adolescent. This also needs to be considered by researchers moving forward.

Third, most studies (n = 29) included in the meta-analysis included an education component in their interventions, 17 of which involved parents. This is consistent with the qualitative review which suggested that it was important for adherence-promoting interventions to include an educational component to parents as well as adolescents.

Finally, the findings from the qualitative review suggested that it is important that interventions and treatment regimens are adapted to support families to preserve 'normal life' and minimise the impact on relationships and other activities. However, only six studies explicitly used approaches in their interventions in order to adapt the intervention to suit the needs of adolescents and their families. More specifically, five of these studies gave participants the choice of where to deliver the intervention, such as in their home, school or local community, in order to adapt to the needs of individual families. However, only one study (Hommel et al., 2012) explicitly discussed simplifying the treatment regimen as part of their intervention in order to reduce the impact of it on families. This therefore needs further consideration and examination in future adherence-promoting intervention research in this population.

Strengths and Limitations

Strengths and limitations of both reviews have been highlighted in the individual publications, so the focus here will be on wider strengths and limitations of the work.

Both reviews were highly comprehensive syntheses of existing literature across a range of chronic illnesses in multiple settings. They were transparent in their methods,

following PRISMA guidelines and for the qualitative review, the ENTREG guidelines, with published protocols and comprehensive searches. There is always a risk in systematic review searching that studies were missed. However, both searches were developed with an experienced academic Librarian, were reviewed within the research team and manual searching of relevant literature was also undertaken to safeguard against this.

For both reviews, the inclusion and exclusion criteria were carefully deliberated and reviewed within the research team. Rationales for final decisions made to the criteria were provided, but it is acknowledged that these decisions may have had implications for the findings and thus conclusions drawn. For the qualitative review outlined in the chapter two, the decision not to include grey literature meant that unpublished theses and dissertations were not included. Further, some mixed-method studies, those which included only the adolescent or parental perspective and studies which included perspectives of others outside of the dyad were also excluded due to the strict criteria. These papers may have offered additional rich data and contributed the overall synthesis. However, the decisions made meant that the study sample was more homogenous, and that the entire findings/results section could be subject to analysis. For the review presented in chapter four, studies which randomised less than 10 participants to each arm or included mixed samples of adolescents and children were excluded. These studies may also have contained additional data.

Further, the definition of adolescents (i.e., 10 to 19 years) was decided a-priori and based on the World Health Organisation's definition (World Health Organisation, n.d.). However, this age range is broad. It is acknowledged that adolescents within this definition may have different challenges and experiences in relation to treatment adherence and therefore may need different types of adherence-promoting interventions. For example, it is much more likely that a 10-year-old will still have parental involvement or oversight in their treatment regimens compared to a 19-year-old. This highlights an important limitation of the thesis overall and of the body of research synthesised, as most included broad age

ranges in their samples. As future reviews and meta-analysis become more focussed in the field, it would be appropriate for specific developmental challenges and experiences to be explored and for developmentally specific interventions to be examined.

For the qualitative systematic presented in chapter two, multiple members of the research team, with differing experiences of the field, were involved in the thematic synthesis. This improved the overall rigour of the analysis and enabled researchers to remain inductive. Discussions within the team allowed the development of analytic themes which moved beyond the findings of individual studies to create new understandings (Thomas & Harden, 2008). This resulted in a deeper understanding of adolescents and their parents' experiences when managing adherence to treatment.

For the meta-analysis outlined in chapter four, the development of the protocol was an iterative process, as PRISMA recognises (Moher et al., 2009). During this phase, the research team carefully deliberated the chosen outcomes, interventions that came under the definition in the literature and moderator and subgroup analyses. These decisions meant that the effectiveness of a range of interventions to promote adherence in adolescents included RCTs across a range of chronic illnesses and settings. This breadth meant that several clinically and research informed moderator, subgroup and exploratory analyses could be conducted. These were methodologically rigorous given the existence of a registered protocol and that corrections were made (Holm, 1979). However, it is acknowledged that several other potential moderators of treatment efficacy may have been missed.

There were some limitations of the body of literature synthesised, which means that the findings and conclusions drawn from the work needs to be interpreted with a degree of caution. First, most studies recruited adolescents with Diabetes and were conducted in the US. Several common chronic illnesses where adherence is problematic, including Epilepsy (Carbone et al., 2013) and Cancer (Rohan et al., 2017), were not represented in either review. Therefore, the findings may not be transferable or generalisable to different

populations of adolescents with chronic illness. Further, there is also an issue of cross-cultural generalisability of the findings. This is particularly important as there are key differences in healthcare contexts between countries. This may therefore mean that adolescents and their parents may have different experiences of managing adherence to treatment and therefore may require different interventions. Cross-cultural generalisation may also be limited due to cultural differences in parenting and developmental contexts. More specifically, there are differences in how cultures approach parenting and view adolescent development. For example, in countries such as China and Kenya adolescent obedience is expected using authoritarian parenting styles whilst in others, such as Sweden and Jordan, parenting is viewed as encouraging adolescents to have more autonomy (Bornstein, 2012; Kapetanovic et al., 2020; Putnick et al., 2012). This therefore suggests that adolescents and parents in these different settings will have different experiences of adherence to treatment and therefore will require different interventions.

Second, there were quality issues in the body of research synthesised. No included studies in the meta-analysis were rated to be low risk of bias, which means that caution must be taken when interpreting the findings. Finally, many studies included in the meta-analysis had descriptive data missing, which meant that they could not be included in the analyses. This meant that follow-up, moderation and subgroup analyses were limited and may mean that the effect sizes found may not accurately represent true treatment effects.

Clinical, Research and Theoretical Implications

The findings of both the qualitative review and meta-analysis have clinical, research and theoretical implications. First, the studies included in both reviews rarely recruited adolescents with poor adherence. The meta-analysis indicated that interventions targeting adolescents with poor adherence specifically produce larger effects than those which do not, which suggests that ceiling effects may, in part, explain the small effects found. This is consistent with previous findings (Pai & McGrady, 2014). Therefore, given that adolescents with identified adherence difficulties are those who will receive these

interventions in healthcare settings (Rapoff & Calkins-Smith, 2020), future research, both exploring experiences and examining intervention efficacy, should aim recruit adolescents with known adherence difficulties. This will allow for dissemination to healthcare settings. It is acknowledged that researchers may face difficulty in recruiting these adolescents and their families, particularly for powered RCTs. Therefore, future research should be creative in recruiting adolescents with poor adherence. Some suggestions include having multiple recruitment sites; involving adolescents with chronic illness and their families in the development of studies; and sharing experiences of effective recruitment strategies within the field. Further, given that many included studies recruited adolescents with Diabetes and were conducted in the US, future research should aim to recruit adolescents with a range of chronic illness diagnoses across multiple countries and settings.

Moderators chosen in the meta-analysis presented in chapter four were driven by clinical applicability. However, other important moderators, such as intervention components and theoretical frameworks, may have been missed. However, due to variability between studies, it would have been difficult to examine these factors quantitively. This has been an issue highlighted in a topical review by McGrady, Ryan, Brown et al. (2015). They argued that stagnant intervention effects found in meta-analyses in the field could, in part, be explained by this research field not becoming more focussed. In their topical review, they found that over two-thirds of adherence-promoting research did not cite a guiding framework and when they did, they often cite multiple overlapping theories. This was supported by findings in the meta-analysis. Therefore, future research needs to be driven by theoretical and clinical rationales in order to progress the field. McGrady, Ryan, Brown et al. (2015) applied the theoretical domains framework (TDF), which is an adult behaviour change theory, to interventions in the included studies. This reduced the variability between domains and theories in the interventions.

Therefore, the findings of their review and from the present thesis indicate that more exploratory research at an earlier stage of intervention development (i.e., pre-RCT) is

needed to explore effective treatment components and aspects of theoretical frameworks. In order to increase consistency, future intervention research could apply the TDF so that future meta-analyses can examine specific intervention domains. However, it is recognised, holding in mind the relational factors involved in adolescent adherence highlighted in chapter two, that the TDF does not consider systemic issues. Therefore, the findings from the qualitative synthesis would support McGrady, Ryan, Brown et al.'s (2015) recommendation for this to be further refined to include the roles of caregivers and peers. This is particularly important to ensure that interventions are developmentally appropriate.

To date almost all intervention research to promote adherence in adolescents has been designed and targeted for specific chronic illnesses. This is a finding which is supported in paediatric adherence-promoting intervention research generally (Pai & McGrady, 2014). Findings from the review presented in chapter two suggest that challenges are transdiagnostic and avenues for future research may be for researchers recruiting samples trans-diagnostically. This may allow for specific intervention domains, grounded explicitly in theories, to be examined in adolescent samples with identified poor adherence. Then, as the field progresses in regard to understanding which treatment components and aspects of theoretical frameworks optimise effectiveness, interventions for specific adherence behaviours in specific chronic illnesses can be developed. This direction is important to improve consistency in the field so that specific intervention and theoretical domains can be examined in order to inform clinical practice.

Further, future research also needs to overcome issues with study quality and reporting. More specifically, future qualitative researchers should consider their relationship to the participants and report detailed demographic and contextual data. Future RCTs need to be more explicit in their methods, including the randomisation process and blinding, and also ensure they have a published protocol available. In order to overcome

the limitation of missing data in the meta-analysis presented in this thesis, it is vital that future RCTs report descriptive data.

It is important for healthcare professionals to be aware of the aforementioned limitations of the work and gaps in the field. As the field progresses more specific recommendations for clinical practice can be reported. It is important for healthcare professionals working with adolescents to be aware of the relational difficulties in adherence across multiple chronic illnesses. They should have an open, non-blaming and empathetic discussion with families about roles and responsibility in adherence.

Moreover, is acknowledged that there are resource-related barriers to implementing psychological interventions to promote adherence in paediatric populations (e.g., clinic space, McGrady, Ryan, Gutiérrez-Colina et al., 2015). Whilst the findings of the meta-analysis suggest that face-to-face interventions are slightly more efficacious than remotely delivered interventions, this difference is small and not statistically significant. Therefore, healthcare professionals should deliver interventions face-to-face in the first instance. However, where this is not possible, healthcare professionals could employ web-based delivery similar to that of several studies in the meta-analysis. This form of delivery would overcome resource barriers and would be particularly beneficial in the current COVID-19 pandemic, as supported by recent commentary by Plevinsky, Young et al. (2020).

Exploratory analyses examining effect sizes of different adherence measures at both posttreatment and follow-up were reported in chapter six. These indicated that there are some differences between effects of different types of adherence measurement. Joint adolescent and parent measures produced the largest effects whilst parent reports produced the smallest, negligible effects. However, very few studies included an objective measure or joint parent and adolescent measures, so it is difficult to draw firm conclusions. Interestingly, objective measures of adherence only showed a small, significant effect after high risk of bias studies were removed, which suggests that bias significantly affected the findings. Therefore, future research is needed to examine differences between objective,

adolescent self-report and parent measures of adherence in adolescents. This will inform future intervention research and clinical practice, which often relies on self-report measures, due to resource limitations (Plevinsky, Gutierrez-Colina et al., 2020). An important question for future interventions to consider is the construct that they are measuring and whether, based on the aims of their intervention, is it enough that self-reported adherence improves or whether objective measures should always be included.

Overall, the mixed method nature of this thesis and synthesis of the findings showed the merit in using both qualitative and quantitative methods to improve the lives of adolescents with chronic illness and their families when managing adherence. Therefore, future qualitative and theoretical research exploring adolescents and their parents' experience should be considered when developing feasibility and later, randomised trials examining intervention effectiveness. Hommel et al.'s (2010, 2012) studies included in both reviews show a good example of how qualitative exploratory and quantitative intervention research can be used to complement one another in this area. In this example, both approaches were utilised when developing an adherence-promoting intervention for adolescents with Inflammatory Bowel Disease. The synthesis of the findings of both reviews aforementioned highlighted key issues that researchers and clinicians should consider when developing interventions and examining their effectiveness in this population. Most importantly, adolescents and their parents' perspectives should be considered during the development and evaluation of such interventions both in research and clinical practice.

Overall Conclusion

Improving adherence in adolescents with chronic illnesses in adolescents is a priority. This thesis utilised a mixed method approach in order to explore adolescents and their parents' experience of adherence to treatment and examined the efficacy of psychological interventions. In the first systematic review, qualitative studies exploring the parent-adolescent dyadic experience when managing adherence to treatment were

synthesised. In the separate, but related systematic review and meta-analysis, the effect sizes from RCTs examining the effectiveness of psychological adherence-promoting interventions in adolescents with chronic illness were pooled. Findings from the qualitative review highlighted the importance of relational factors including trust, negotiation and collaboration in enabling the parent-child dyad to navigate the complexity of adherence. The meta-analysis indicated that psychological interventions for promoting adherence in adolescents specifically have limited efficacy. Overall, future high-quality research, including qualitative research exploring multiple perspectives and adherence-promoting intervention research, is needed. Key limitations of the current literature, as well as the methods applied in this thesis, were discussed. Recommendations for future research and clinical practice were also provided. Moving forward, researchers in this field should aim to recruit adolescent samples with suboptimal adherence across a range of chronic illnesses.

References

Becker, M. H., & Maimam, L. A. (1975). Sociobehavioral determinants of compliance with health and medical care recommendations. *Medical Care, 13(1)*, 10-24. https://www.jstor.org/stable/3763271

Blakemore, S-J. (2019). *Inventing Ourselves: The Secret Life of the Teenage Brain.* Penguin Random House UK.

Bornstein, M. H. (2012). Cultural approaches to parenting. *Parenting: Science and Practice, 12(3-4),* 212-221. https://doi.org/10.1080/15295192.2012.683359

Bucks, R. S., Hawkins, K., Skinner, T. C., Horn, S., Seddon, P., & Horne, R. (2009). Adherence to treatment in adolescents with cystic fibrosis: the role of illness perceptions and treatment beliefs. *Journal of pediatric psychology*, *34*(8), 893-902. https://doi.org/10.1093/jpepsy/jsn135

Carbone, L., Zebrack, B., Plegue, M., Joshi, S., & Shellhaas, R. (2013). Treatment adherence among adolescents with epilepsy: What really matters?. *Epilepsy & Behavior*, *27*(1), 59-63. https://doi.org/10.1016/j.yebeh.2012.11.047

Cuijpers, P. (2016). *Meta-analyses in mental health research: A practical guide.* Pim Cuijpers Uitgeverij.

Denison, J. A., Banda, H., Dennis, A. C., Packer, C., Nyambe, N., Stalter, R. M., Mwansa, J. K., Katayamoyo, P., & McCarraher, D. R. (2015). "The sky is the limit": Adhering to antiretroviral therapy and HIV self-management from the perspectives of adolescents living with HIV and their adult caregivers. *Journal of the International AIDS Society, 18(19358)*, 1-6. http://dx.doi.org/10.7448/IAS.18.1.19358

DiMatteo, M. R. (2004). Variations in patients' adherence to medical recommendations: a quantitative review of 50 years of research. *Medical care*, 200-209. https://www.jstor.org/stable/4640729

Drotar, D. (2000). *Promoting adherence to medical treatment in childhood chronic illness: Concepts, methods, and interventions.* Lawrence Erlbaum Associates.

Ellis, D. A., Carcone, A. I., Naar-King, S., Rajkumar, D., Palmisano, G., & Moltz, K. (2019). Adaptation of an evidence-based diabetes management intervention for delivery in community settings: Findings from a pilot randomized effectiveness trial. *Journal of Pediatric Psychology, 44(1)*, 110-125. https://doi.org/10.1093/jpepsy/jsx144

Ellis, D. A., Naar-King, S., Chen, X., Moltz, K., Cunningham, P. B., & Idalski-Carcone, A. (2012). Multisystemic therapy compared to telephone support for youth with poorly controlled diabetes: Findings from a randomized controlled trial. *Annals of Behavioral Medicine*, *44*(2), 207-215. https://doi-org.uea.idm.oclc.org/10.1007/s12160-012-9378-1

Erikson, E. H. (1968). *Identity: Youth and crisis.* Norton.

Fishbein, M., & Ajzen, I. (1975). *Belief, attitude, intention, and behavior: An introduction to theory and research.* Addison-Wesley.

Fletcher, A. J. (2017). Applying critical realism in qualitative research: methodology meets method. *International Journal of Social Research Methodology*, *20*(2), 181–194. https://doi.org/10.1080/13645579.2016.1144401

Fredericks, e. m., Magee, J. C., Opipari-Arrigan, L., Shieck, V., Well, A., & Lopez, M. J. (2008). Adherence and health-related quality of life in adolescent liver transplant recipients. *Pediatric Transplantation, 12(3)*, 289-299. https://doi.org/10.1111/j.1399-3046.2008.00901.x

Graves, M. M., Roberts, M. C., Rapoff, M., & Boyer, A. (2010). The Efficacy of Adherence Interventions for Chronically Ill Children: A Meta-Analytic Review. *Journal of Pediatric Psychology, 35(4)*, 368-382. https://doi.org/10.1093/jpepsy/jsp072

Grey, M., Schulman-Green, D., Knafl, K., & Reynolds, N. R. (2015). A revised self-and family management framework. *Nursing Outlook, 63(2)*, 162-170. https://doi.org/10.1016/j.outlook.2014.10.003

Hagell, A., Coleman, J., & Brooks, F. (2015). *Key Data on Adolescence*. Association for Young People's Health: http://www.youngpeopleshealth.org.uk/wp-content/uploads/2015/09/KeyData2015.pdf

Hains, A. A., Berlin, K. S., Hobart Davies, W., Smothers, M. K., Sato, A. F., & Alemzadeh, R. (2007). Attributions of adolescents with type 1 diabetes related to performing diabetes care around friends and peers: the moderating role of friend support. *Journal of pediatric psychology*, *32*(5), 561-570. https://doi.org/10.1093/jpepsy/jsl040

Hanghøj, S., & Boisen, K. A. (2013). Self-reported barriers to medication adherence among chronically ill adolescents: A systematic review. *Journal of Adolescent Health, 54(2)*, 121-138. https://doi.org/10.1016/j.jadohealth.2013.08.009

Heath, G., Farre, A., & Shaw, K. (2017). Parenting a child with chronic illness as they transition into adulthood: a systematic review and thematic synthesis of parents' experiences. *Patient education and counselling, 100(1)*, 76-92. https://doi.org/10.1016/j.pec.2016.08.011

Higgins, J. P., Eldridge, S., & Li, T. (2020). *Chapter 23: Including variants on randomized trials*. Cochrane Training: https://training.cochrane.org/handbook/archive/v6/chapter-23

Higgins, J. P., & Green, S. (2008). *Cochrane handbook for systematic reviews of interventions.* John Wiley & Sons.

Higgins, J. P., Li, T., & Deeks, J. J. (2020). *Chapter 6: Choosing effect measures and computing estimates of effect*. Cochrane Training: https://training.cochrane.org/handbook/current/chapter-06

Holm, S. (1979). A simple sequentially rejective method procedure. *Scandinavian Journal of Statistics, 6(2)*, 65-70. https://www.jstor.org/stable/4615733

Hommel, K. A., Hente, E. A., Odell, S., Herzer, M., Ingerski, L. M., Guilfoyle, S. M., & Denson, L. A. (2012). Evaluation of a group-based behavioral intervention to promote adherence in adolescents with inflammatory bowel disease. *European journal of gastroenterology & hepatology*, *24*(1), 64. https://doi-org/10.1097/MEG.0b013e32834d09f1

Hommel, K. A., Odell, S., Sander, E., Baldassano, R. N., & Barg, F. K. (2010). Treatment adherence in paediatric inflammatory bowel disease: perceptions from adolescent patients and their families. *Health & Social Care in the Community, 19(1)*, 80-88. https://doi.org/10.1111/j.1365-2524.2010.00951.x

Kahana, S., Drotar, D., & Frazier, T. (2008). Meta-analysis of psychological interventions to promote adherence to treatment in pediatric chronic health conditions. *Journal of Pediatric Psychology, 33(6)*, 590-611. https://doi.org/10.1093/jpepsy/jsm128

Kapetanovic, S., Rothenberg, W. A., Lansford, J. E., Bornstein, M. H., Chang, L., Deater-Deckard, K., Di Giunta, L., Dodge, K. A., Gurdal, S., Malone, P. S., Oburu, P., Pastorelli, C., Skinner, A. T., Sorbring, E., Steinberg, L., Tapanya, S., Uribe Tirado, L. M., Yotanyamaneewong, S., Alampay, L. P., … & Bacchini, D. (2020). Cross-cultural examination of links between parent–adolescent communication and adolescent psychological problems in 12 cultural groups. *Journal of youth and adolescence, 49,* 1225-1244. https://doi.org/10.1007/s10964-020-01212-2

Kosse, R. C., Bouvy, M. L., de Vries, T. W., & Koster, E. S. (2019). Effect of a mHealth intervention on adherence in adolescents with asthma: A randomized controlled trial. *Respiratory Medicine, 149*, 45-51. https://doi.org/10.1016/j.rmed.2019.02.009

Law, E., Fisher, E., Eccleston, C., & Palermo, T. M. (2019). Psychological interventions for parents of children and adolescents with chronic illness. *Cochrane Database of Systematic Reviews, 3*. https://doi.org/10.1002/14651858.CD009660

Law, G. U., Tolgyesi, C. S., & Howard, R. A. (2014). Illness beliefs and self-management in children and young people with chronic illness: a systematic review. *Health psychology review*, *8*(3), 362-380. https://doi.org/10.1080/17437199.2012.747123

Lerch, M. F., & Thrane, S. E. (2019). Adolescents with chronic illness and the transition to self-management: A systematic review. *Journal of Adolescence, 72*, 152-161. https://doi.org/10.1016/j.adolescence.2019.02.010

Lindsay, S., Kingsnorth, S., & Hamdani, Y. (2011). Barriers and facilitators of chronic illness self-management among adolescents: a review and future directions. *Journal of Nursing and Healthcare of Chronic Illness, 3(3)*, 186-208. https://doi.org/10.1111/j.1752-9824.2011.01090.x

Luo, J., Guo, J., Yang, J., Ou, X., & Grey, M. (2020). Parent–Child Relationship Quality as a Mediator of the Association between Perceived Stress and Diabetes Self-Management in Adolescents With Type 1 Diabetes. *Journal of Family Nursing*, 1-10. https://doi.org/10.1177/1074840720971584

Mammen, J. R., Rhee, H., Norton, S. A., & Butz, A. M. (2016). Perceptions and experiences underlying self-management and reporting of symptoms in teens with asthma. *Journal of Asthma*, *54*(2), 143-152. https://doi.org/10.1080/02770903.2016.1201835

McEvoy, P., & Richards, D. (2006). A critical realist rationale for using a combination of quantitative and qualitative methods. *Journal of Research in Nursing, 11(1)*, 66-78. https://doi.org/10.1177/1744987106060192

McGrady, M. E., Ryan, J. L., Brown, G. A., & Cushing, C. C. (2015). Topical review: theoretical frameworks in pediatric adherence-promotion interventions: research findings and methodological implications. *Journal of pediatric psychology*, *40*(8), 721-726. https://doi.org/10.1093/jpepsy/jsv025

McGrady, M. E., Ryan, J. L., Gutiérrez-Colina, A. M., Fredericks, E. M., Towner, E. K., & Pai, A. L. (2015). The impact of effective paediatric adherence promotion

interventions: systematic review and meta-analysis. *Child: care, health and development, 41(6)*, 789-802. https://doi.org/10.1111/cch.12271

Modi, A. C., & Driscoll, K. A. (2020). *Adherence and Self-Management in Pediatric Populations.* Elsevier Science & Technology.

Modi, A. C., Pai, A. L., Hommel, K. A., Hood, K. K., Cortina, S., Hilliard, M. E., Guilfoyle, S. M., Gray, W. N., & Drotar, D. (2012). Pediatric self-management: a framework for research, practice, and policy. *Pediatrics*, *129*(2), e473-e485. https://doi.org/10.1542/peds.2011-1635

Moher, D., Liberati, A., Tetzlaff, J., Altman, D. G., & The PRISMA Group. (2009). Preferred reporting items for systematic reviews and meta-analyses: The PRISMA statement. *PLoS Medicine, 6(7)*, e1000097. https://doi.org/10.1136/bmj.b2535

O'Toole, D. P., Latchford, G. J., Duff, A. J., Ball, R., McCormack, P., McNamara, P. S., Browniee, K. G., & Southern, K. W. (2019). Adherence to aerosol therapy in young people with cystic fibrosis: Patient and parent perspectives following electronic data capture. *Qualitative Health Research, 29(6)*, 846-856. https://doi.org/10.1177/1049732318805754

Pai, A. L., & McGrady, M. (2014). Systematic review and meta-analysis of psychological interventions to promote treatment adherence in children, adolescents, and young adults with chronic illness. *Journal of Pediatric Psychology, 39(8)*, 918-931. https://doi.org/10.1093/jpepsy/jsu038

Patel, N. J., Datye, K. A., & Jaser, S. S. (2018). Importance of patient–provider communication to adherence in adolescents with type 1 diabetes. *Healthcare* 6(30), 1-11. https://doi.org/10.3390/healthcare6020030

Pendley, J. S., Kasmen, L. J., Miller, D. L., Donze, J., Swenson, C., & Reeves, G. (2002). Peer and family support in children and adolescents with type 1 diabetes. *Journal of pediatric psychology*, *27*(5), 429-438. https://doi.org/10.1093/jpepsy/27.5.429

Peng, Y., Wang, H., Fang, Q., Xie, L., Shu, L., Sun, W., & Liu, Q. (2020). Effectiveness of mobile applications on medication adherence in adults with chronic diseases: a systematic review and meta-analysis. *Journal of managed care & specialty pharmacy*, *26*(4), 550-561. https://doi.org/10.18553/jmcp.2020.26.4.550

Piaget, J. (1936). *The origin of intelligence in the child.* Penguin.

Plevinsky, J. M., Gutierrez-Colina, A. M., Carmody, J. K., Hommel, K. A., Crosby, L. E., McGrady, M. E., Pai, A. L., & Modi, A. C. (2020). Patient-reported outcomes for pediatric adherence and self-management: a systematic review. *Journal of Pediatric Psychology*, *45*(3), 340-357. https://doi.org/10.1093/jpepsy/jsz096

Plevinsky, J. M., Young, M. A., Carmody, J. K., Durkin, L. K., Gamwell, K. L., Klages, K. L., Ghosh, S., & Hommel, K. A. (2020). The Impact of COVID-19 on Pediatric Adherence and Self-Management. *Journal of pediatric psychology*, *45*(9), 977-982. https://doi-org.uea.idm.oclc.org/10.1093/jpepsy/jsaa079

Pritlove, C., Markowitz, B., Mukerji, G., Advani, A., & Parsons, J. A. (2020). Experiences and perspectives of the parents of emerging adults living with type 1 diabetes. *BMJ Open Diabetes Research and Care, 8(1)*, e001125. http://dx.doi.org/10.1136/bmjdrc-2019-001125

Psihogios, A. M., Fellmeth, H., Schwartz, L. A., & Barakat, L. P. (2019). Family Functioning and Medical Adherence Across Children and Adolescents With Chronic Health Conditions: A Meta-Analysis. *Journal of Pediatric Psychology, 44(1)*, 84-97. https://doi.org/10.1093/jpepsy/jsy044

Putnick, D. L., Bornstein, M. H., Lansford, J. E., Chang, L., Deater-Deckard, K., Di Giunta, L., Gurdal, S., Dodge, K. A., Malone, P. S., Oburu, P. O., Pastorelli, C., Skinner, A. T., Sorbring, E., Tapanya, S., Uribe Tirado, L. M., Zelli, A., Alampay, L. P., Al-Hassan, S. M., Bacchini, D., & Bombi, A. S. (2012). Agreement in mother and father acceptance-rejection, warmth, and hostility/rejection/neglect of children

across nine countries. *Cross-Cultural Research*, *46*(3), 191-223. https://doi.org/10.1177/1069397112440931

Quittner, A. L., Modi, A. C., Lemanek, K. L., Ievers-Landis, C. E., & Rapoff, M. A. (2008). Evidence-based assessment of adherence to medical treatments in pediatric psychology. *Journal of pediatric psychology*, *33*(9), 916-936. https://doi.org/10.1093/jpepsy/jsm064

Rapoff, M. (2010). *Adherence to pediatric medical regimens.* Springer.

Rapoff, M. A., & Calkins-Smith, A. (2020). Enhancing Treatment Adherence in Young People with Chronic Diseases. In A. Hadler, S. Sutton, & L. Osterberg, *The Wiley Handbook of Healthcare Treatment Engagement: Theory, Research, and Clinical Practice* (pp. 354-364). Wiley-Blackwell.

Rohan, J. M., Fukuda, T., Alderfer, M. A., Wetherington Donewar, C., Ewing, L., Katz, E. R., Muriel, A. C., Vinks, A. A., & Drotar, D. (2017). Measuring medication adherence in pediatric cancer: an approach to validation. *Journal of pediatric psychology*, *42*(2), 232-244. https://doi.org/10.1093/jpepsy/jsw039

Rosenstock, I. M. (1974). Historical origins of the health belief model. *Health education monographs*, *2*(4), 328-335. https://doi.org/10.1177/109019817400200403

Ryan, P., & Sawin, K. J. (2009). The individual and family self-management theory: Background and perspectives on context, process, and outcomes. *Nursing Outlook, 22(10)*, 217-225. https://doi.org/10.1016/j.outlook.2008.10.004

Santer, M., Ring, N., Yardley, L., Geraghty, A. W., & Wyke, S. (2014). Treatment non-adherence in pediatric long-term medical conditions: systematic review and synthesis of qualitative studies of caregivers' views. *BMC Pediatrics, 14(63)*, 1-10. https://doi.org/10.1186/1471-2431-14-63

Singh, E., Farruggia, S. P., & Peterson, E. R. (2013). Adolescents with diabetes: support from healthcare teams and families. *International journal of adolescent medicine and health*, *25*(1), 91-96. https://doi.org/10.1515/ijamh-2013-0013

Smith, P. K., Cowie, H., & Blades, M. (2011). *Understanding children's development*. John Wiley & Sons.

Suris, J.-C., Michaud, P.-A., & Viner, R. (2004). The adolescent with a chronic condition. Part I: developmental issues. *Archives of Disease in Childhood, 89(10)*, 938-942. http://dx.doi.org/10.1136/adc.2003.045369

Thomas, J., & Harden, A. (2008). Methods for the thematic synthesis of qualitative research in systematic reviews. *BMC Medical Research Methodology, 8(45)*, 1-10. https://doi.org/10.1186/1471-2288-8-45

van der Lee, J. H., Mokkink, L. B., Grootenhuis, M. A., Heymans, H. S., & Offringa, M. (2007). Definitions and Measurement of Chronic Health Conditions in Childhood A Systematic Review. *JAMA, 297(24)*, 2741-2751. https://doi.org/10.1001/jama.297.24.2741

Viner, R., & Christie, D. (2005). Adolescent development. *BMJ, 330(7486)*, 301-304. https://doi.org/10.1136/bmj.330.7486.301

Williams-Reade, J. M., Tapanes, D., Distelberg, B. J., & Montgomery, S. (2019). pediatric chronic illness management: A qualitative dyadic analysis of adolescent patient and parent illness narratives. *Journal of marital and family therapy*, *46*(1), 135-148. https://doi.org/10.1111/jmft.12377

World Health Organisation. (2003). *Adherence to Long-Term Therapies: Evidence for Action.* https://www.who.int/chp/knowledge/publications/adherence_full_report.pdf?ua=1

World Health Organisation. (n.d.). *Adolescent Health*. https://www.who.int/health-topics/adolescent-health/#tab=tab_1

Yeo, M., & Sawyer, S. (2005). Chronic illness and disability. *British Medical Journal, 330(7493)*, 721-723. https://doi.org/10.1136/bmj.330.7493.721

Appendices

Appendix A: Health Review Psychology Instructions to Authors

Information copied from:

https://www.tandfonline.com/action/authorSubmission?show=instructions&journalCode=rhpr20

Instructions for authors

COVID-19 impact on peer review
As a result of the significant disruption that is being caused by the COVID-19 pandemic we understand that many authors and peer reviewers will be making adjustments to their professional and personal lives. As a result they may have difficulty in meeting the timelines associated with our peer review process. Please let the journal editorial office know if you need additional time. Our systems will continue to remind you of the original timelines but we intend to be flexible.

Thank you for choosing to submit your paper to us. These instructions will ensure we have everything required so your paper can move through peer review, production and publication smoothly. Please take the time to read and follow them as closely as possible, as doing so will ensure your paper matches the journal's requirements.

AUTHOR SERVICES
Supporting Taylor & Francis authors

For general guidance on every stage of the publication process, please visit our Author Services website.

EDITING SERVICES
Supporting Taylor & Francis authors

For editing support, including translation and language polishing, explore our Editing Services website

SCHOLARONE MANUSCRIPTS™

This journal uses ScholarOne Manuscripts (previously Manuscript Central) to peer review manuscript submissions. Please read the guide for ScholarOne authors before making a submission. Complete guidelines for preparing and submitting your manuscript to this journal are provided below.

This title utilises format-free submission. Authors may submit their paper in any scholarly format or layout. References can be in any style or format, so long as a consistent scholarly citation format is applied. For more detail see the format-free submission section below.

Contents

About the Journal

Health Psychology Review is an international, peer-reviewed journal publishing high-quality, original research. Please see the journal's Aims & Scope for information about its focus and peer-review policy.

Open Access

You have the option to publish open access in this journal via our Open Select publishing program. Publishing open access means that your article will be free to access online immediately on publication, increasing the visibility, readership and impact of your research. Articles published Open Select with Taylor & Francis typically receive 32% more citations* and over 6 times as many downloads** compared to those that are not published Open Select.

Your research funder or your institution may require you to publish your article open access. Visit our Author Serviceswebsite to find out more about open access policies and how you can comply with these.

You will be asked to pay an article publishing charge (APC) to make your article open access and this cost can often be covered by your institution or funder. Use our APC finder to view the APC for this journal.

Please visit our Author Services websiteor contact openaccess@tandf.co.uk if you would like more information about our Open Select Program.

*Citations received up to Jan 31st 2020 for articles published in 2015-2019 in journals listed in Web of Science®.
**Usage in 2017-2019 for articles published in 2015-2019.

Peer Review

Taylor & Francis is committed to peer-review integrity and upholding the highest standards of review. Once your paper has been assessed for suitability by the editor, it will then be single blind peer reviewed by independent, anonymous expert referees. Find out more about what to expect during peer review and read our guidance on publishing ethics.

Preparing Your Paper

Manuscripts must be written in English. American or British spelling and punctuation are acceptable, provided authors apply the style consistently throughout the manuscript. Manuscripts with incorrect grammar or errors will be returned to authors. Authors are encouraged to proofread their manuscript prior to submission. Authors requiring English language editing services are directed to the Taylor and Francis Author Services website.

Manuscript Length

The editorial team acknowledge that review articles are usually longer than articles reporting findings of primary research. Health psychology review does not impose any length restrictions on submitted articles. However, it is also recognised that articles should be appropriately concise and pithy so that the main focus is not lost and the argument is not encumbered by unnecessary detail. Authors can include supplemental materials such as figures and tables not directly germane to the main argument of the manuscript as online supplemental materials. For meta-analyses and systematic reviews, references for studies included in the review should be only appear in a separate supplemental list that the journal will make available as an online supplement. These materials will not count toward the page length of the manuscript, but will be included as a permanent record of supplemental materials alongside the online version of the manuscript (see later). Manuscripts should be compiled in the following order: title page including acknowledgements and funding details as an author note; abstract; keywords; main text; references; table(s) with caption(s) (on individual pages); figures and figure caption(s).

Meta-analyses and systematic reviews

In order to comply with international standards and for academic transparency, authors of meta-analyses and systematic reviews submitted to Health Psychology Review are required to include a statement in accordance with the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) statement (http://www.prisma-statement.org/) as a supplemental file for review (the final document will be included as online supplemental material). In addition, authors of meta-analyses should include the information recommended by the APA's Meta-Analysis Reporting Methods (MARS) which can be found here (http://www.apastyle.org/manual/related/JARS-MARS.pdf)

Open Science Policies

Open Data

Health psychology review is committed to the principles of open science. Authors are therefore required to publish any raw data and any code or syntax used in data

analysis in the interests of full disclosure and transparency. Please see a recent article outlining the recommendations: Peters, G.-J. Y., Abraham, C., & Crutzen, R. (2012). Full disclosure: Doing behavioural science necessitates sharing. European Health Psychologist, 14, 77-84. Authors of articles that make use of data (e.g., meta-analysis, systematic reviews) are required to make all raw data files and code or syntax used in data analysis available when submitting the manuscript. This can be done using the HPR online submission portal. Authors should upload files as supplemental materials (for review). Authors should choose formats that are able to be read using commonly available software (e.g., text or rtf files). Authors of articles accepted for publication can opt to have the data and analysis files published as supplemental materials permanently linked with the online version of the article, or with another archival service provider such as the Open Science Framework website or Dryad, or both.

Pre-Registration

From January 1, 2021 all reviews with empirical content (e.g., systematic reviews, meta-analyses) will be required to be pre-registered on an appropriate independent, institutional registry such as Prospero https://www.crd.york.ac.uk/prospero/, the Open Science Framework https://osf.io/ or other registry (e.g., http://clinicaltrials.gov/, http://socialscienceregistry.org/, http://egap.org/designregistration/, http://ridie.3ieimpact.org/). Pre-registration of studies involves registering the study design, variables, and treatment conditions prior to conducting the research. Authors must confirm that the study was registered prior to conducting the research, and that the preregistration adheres to the disclosure requirements of the institutional registry or those required for the preregistered 'badge'maintained by the Center for Open Science: see https://osf.io/tvyxz/wiki/home/. Authors must report the web link to the timestamped pre-registration at the institutional registry or the pre-registration trial number prominently (e.g., in the Methods section) in their manuscript. For registries that do not provide a standard template for pre-registering a review, authors are advised to use a template that includes the required information. An example is provided here. There will be a one year grace period between January 1, 2019 and December 31, 2020 in which pre-registration is not a requirement for submission, but strongly encouraged. From January 1, 2021 pre-registration will be a mandatory requirement for submission for all empirical reviews. It is acceptable to pre-register a review after the initial search has been completed, but not before any screening according to eligibility criteria has been conducted. Post-registration is not acceptable. Conceptual and narrative reviews do not need to be pre-registered.

Structure

Your paper should be compiled in the following order: title page; abstract; keywords; main text introduction, materials and methods, results, discussion; acknowledgments; declaration of interest statement; references; appendices (as appropriate); table(s) with caption(s) (on individual pages); figures; figure captions (as a list).

Format-Free Submission

Authors may submit their paper in any scholarly format or layout. Manuscripts may be supplied as single or multiple files. These can be Word, rich text format (rtf), open document format (odt), or PDF files. Figures and tables can be placed within the text or submitted as separate documents. Figures should be of sufficient resolution to enable refereeing.

- There are no strict formatting requirements, but all manuscripts must contain the essential elements needed to evaluate a manuscript: abstract, author affiliation, figures, tables, funder information, and references. Further details may be requested upon acceptance.
- References can be in any style or format, so long as a consistent scholarly citation format is applied. Author name(s), journal or book title, article or chapter title, year of publication, volume and issue (where appropriate) and page numbers are essential. All bibliographic entries must contain a corresponding in-text citation. The addition of DOI (Digital Object Identifier) numbers is recommended but not essential.
- The journal reference style will be applied to the paper post-acceptance by Taylor & Francis.
- Spelling can be US or UK English so long as usage is consistent.

Note that, regardless of the file format of the original submission, an editable version of the article must be supplied at the revision stage.

Taylor & Francis Editing Services

To help you improve your manuscript and prepare it for submission, Taylor & Francis provides a range of editing services. Choose from options such as English Language Editing, which will ensure that your article is free of spelling and grammar errors, Translation, and Artwork Preparation. For more information, including pricing, visit this website.

Checklist: What to Include

1. **The Title Page** (p. 1) should contain the article title, authors' names and affiliations and the address for manuscript correspondence (including e-mail, address and telephone and fax numbers). No information that would indicate authors' identity or affiliation should be contained in the manuscript itself; all such information should be included on the title page only. Read more on authorship.
2. **The Abstract** (p. 2) must be a single paragraph that summarizes the main findings of the paper in fewer than 200 words. After the abstract a list of six **Keywords** that will be useful for indexing or searching should be included.

3. **Figures.** Figures may be embedded in the manuscript file (preferred), provided at the end, or saved as separate files. All figures must be numbered in the order in which they appear in the manuscript (e.g. Figure 1, Figure 2) and have appropriate captions. Figures should be of sufficient resolution to enable refereeing.

4. **Tables.** Tables may be embedded in the manuscript file (preferred), provided at the end, or saved as separate files. All tables must be numbered in the order in which they appear in the manuscript (e.g. Table 1, Table 2) and have appropriate captions.
5. **Authors' contributions.** All authors are expected to have made substantive intellectual contributions to, and to have been involved in drafting or revising the manuscript. Each author should have participated sufficiently in the work to take public responsibility for appropriate portions of the content. Acquisition of funding, collection of data, or general supervision of the research group, alone, does not justify authorship. With the submission of a manuscript, it is assumed that all authors have read and approved the final manuscript.
6. **Acknowledgements.** All contributors who do not meet the above criteria for authorship, should be listed in an acknowledgements section in accordance with the APA guidelines. The acknowledgements should be contained on the title page of the manuscript as making acknowledgements available to reviewers will compromise the masked peer-review process. Examples of those who might be acknowledged include those who provided general, technical, or writing assistance. Acknowledgement of funding/grants are also included in this section.
7. **Funding details.** Please supply all details required by your funding and grant-awarding bodies as follows:
   For single agency grants
   This work was supported by the [Funding Agency] under Grant [number xxxx].
   For multiple agency grants
   This work was supported by the [Funding Agency #1] under Grant [number xxxx]; [Funding Agency #2] under Grant [number xxxx]; and [Funding Agency #3] under Grant [number xxxx].
8. **Supplemental online material.** Supplemental material can be a video, dataset, fileset, sound file or anything which supports (and is pertinent to) your paper. We publish supplemental material online via Figshare. Find out more about supplemental material and how to submit it with your article.
9. **Disclosure statement.** This is to acknowledge any financial interest or benefit that has arisen from the direct applications of your research. Further guidance on what is a conflict of interest and how to disclose it.
10. **Data availability statement.** If there is a data set associated with the paper, please provide information about where the data supporting the results or analyses presented in the paper can be found. Where applicable, this should include the hyperlink, DOI or other persistent identifier associated with the data set(s). Templates are also available to support authors.
11. **Data deposition.** If you choose to share or make the data underlying the study open, please deposit your data in a recognized data repository prior to or at the time of submission. You will be asked to provide the DOI, pre-reserved DOI, or other persistent identifier for the data set.
12. **Equations.** If you are submitting your manuscript as a Word document, please ensure that equations are editable. More information about mathematical symbols and equations.
13. **Units.** Please use SI units (non-italicized).

Using Third-Party Material in your Paper

You must obtain the necessary permission to reuse third-party material in your article. The use of short extracts of text and some other types of material is usually permitted, on a limited basis, for the purposes of criticism and review without securing formal permission. If you wish to include any material in your paper for which you do not hold copyright, and which is not covered by this informal agreement, you will need to obtain written permission from the copyright owner prior to submission. More information on requesting permission to reproduce work(s) under copyright.

Submitting Your Paper

This journal uses ScholarOne Manuscripts to manage the peer-review process. If you haven't submitted a paper to this journal before, you will need to create an account in ScholarOne. Please read the guidelines above and then submit your paper in the relevant Author Centre, where you will find user guides and a helpdesk.

If you are submitting in LaTeX, please convert the files to PDF beforehand (you will also need to upload your LaTeX source files with the PDF).

Please note that *Health Psychology Review* uses Crossref™ to screen papers for unoriginal material. By submitting your paper to *Health Psychology Review* you are agreeing to originality checks during the peer-review and production processes.

On acceptance, we recommend that you keep a copy of your Accepted Manuscript. Find out more about sharing your work.

Data Sharing Policy

This journal applies the Taylor & Francis Basic Data Sharing Policy. Authors are encouraged to share or make open the data supporting the results or analyses presented in their paper where this does not violate the protection of human subjects or other valid privacy or security concerns.

Authors are encouraged to deposit the dataset(s) in a recognized data repository that can mint a persistent digital identifier, preferably a digital object identifier (DOI) and recognizes a long-term preservation plan. If you are uncertain about where to deposit your data, please see this informationregarding repositories.

Authors are further encouraged to cite any data sets referenced in the article and provide a Data Availability Statement.

At the point of submission, you will be asked if there is a data set associated with the paper. If you reply yes, you will be asked to provide the DOI, pre-registered DOI, hyperlink, or other persistent identifier associated with the data set(s). If you have selected to provide a pre-registered DOI, please be prepared to share the reviewer URL associated with your data deposit, upon request by reviewers.

Where one or multiple data sets are associated with a manuscript, these are not formally peer reviewed as a part of the journal submission process. It is the

author's responsibility to ensure the soundness of data. Any errors in the data rest solely with the producers of the data set(s).

Publication Charges

There are no submission fees, publication fees or page charges for this journal.

Colour figures will be reproduced in colour in your online article free of charge. If it is necessary for the figures to be reproduced in colour in the print version, a charge will apply.

Charges for colour figures in print are £300 per figure ($400 US Dollars; $500 Australian Dollars; €350). For more than 4 colour figures, figures 5 and above will be charged at £50 per figure ($75 US Dollars; $100 Australian Dollars; €65). Depending on your location, these charges may be subject to local taxes.

Copyright Options

Copyright allows you to protect your original material, and stop others from using your work without your permission. Taylor & Francis offers a number of different license and reuse options, including Creative Commons licenses when publishing open access. Read more on publishing agreements.

Complying with Funding Agencies

We will deposit all National Institutes of Health or Wellcome Trust-funded papers into PubMedCentral on behalf of authors, meeting the requirements of their respective open access policies. If this applies to you, please tell our production team when you receive your article proofs, so we can do this for you. Check funders' open access policy mandates here. Find out more about sharing your work.

Accepted Manuscripts Online

This journal posts manuscripts online as rapidly as possible, as a PDF of the final, accepted (but unedited and uncorrected) paper. This is clearly identified as an unedited manuscript and is referred to as the Accepted Manuscript Online (AMO). No changes will be made to the content of the original paper for the AMO version but, after copy-editing, typesetting, and review of the resulting proof, the final corrected version (the Version of Record [VoR]), will be published, replacing the AMO version.

The VoR is the article in its final, definitive and citable form (this may not be immediately paginated, but is the version that will appear in an issue of the journal). Both the AMO version and VoR can be cited using the same DOI (digital object identifier). To ensure rapid publication, we ask you to return your signed publishing agreement as quickly as possible, and return corrections within 48 hours of receiving your proofs.

My Authored Works

On publication, you will be able to view, download and check your article's metrics (downloads, citations and Altmetric data) via My Authored Works on Taylor & Francis Online. This is where you can access every article you have published with us, as well as your free eprints link, so you can quickly and easily share your work with friends and colleagues.

We are committed to promoting and increasing the visibility of your article. Here are some tips and ideas on how you can work with us to promote your research.

Article Reprints

You will be sent a link to order article reprints via your account in our production system. For enquiries about reprints, please contact the Taylor & Francis Author Services team at reprints@tandf.co.uk. You can also order print copies of the journal issue in which your article appears.

Queries

Should you have any queries, please visit our Author Services website or contact us here.

Updated 31-07-2020

Appendix B: MEDLINE Search String for the Qualitative Review

EBSCOhost

Monday, July 20, 2020 7:59:35 AM

#	Query	Limiters/Expanders	Last Run Via	Results
S34	S4 AND S8 AND S12 AND S29 AND S32	Limiters - Date of Publication: 20000101-20201231 Expanders - Apply related words; Apply equivalent subjects Search modes - Find all my search terms	Interface - EBSCOhost Research Databases Search Screen - Advanced Search Database - MEDLINE Complete	1,113
S33	S4 AND S8 AND S12 AND S29 AND S32	Expanders - Apply related words; Apply equivalent subjects Search modes - Find all my search terms	Interface - EBSCOhost Research Databases Search Screen - Advanced Search Database - MEDLINE Complete	Display
S32	S30 OR S31	Expanders - Apply related words; Apply equivalent subjects Search modes - Find all my search terms	Interface - EBSCOhost Research Databases Search Screen - Advanced Search Database - MEDLINE Complete	Display
S31	TI Qualitative or experience* or perspective* or perception* or perceived or belief* or view* or narrative*	Expanders - Apply related words; Apply equivalent subjects Search modes - Find all my search terms	Interface - EBSCOhost Research Databases Search Screen - Advanced Search Database - MEDLINE Complete	Display
S30	(MH "Qualitative Research")	Expanders - Apply related words; Apply equivalent subjects Search modes - Find all my search terms	Interface - EBSCOhost Research Databases Search Screen - Advanced Search Database - MEDLINE Complete	Display
S29	S13 OR S14 OR S15 OR S16 OR S17 OR S18 OR S19 OR S20 OR S21 OR S22 OR S23 OR S24 OR S25 OR S26 OR S27 OR S28	Expanders - Apply related words; Apply equivalent subjects Search modes - Find all my search terms	Interface - EBSCOhost Research Databases Search Screen - Advanced Search Database - MEDLINE Complete	Display
S28	AB "Chronic illness*" OR "Chronic disease*" OR epilepsy OR diabetes OR asthma OR "sickle cell disease" OR transplant* OR "spina bifida" OR "cystic fibrosis" OR "human immunodeficiency virus" OR HIV OR arthritis OR "inflammatory bowel disease*" OR cancer OR "chronic pain" OR "gastrointestinal disease*" OR "communicable disease"	Expanders - Apply related words; Apply equivalent subjects Search modes - Find all my search terms	Interface - EBSCOhost Research Databases Search Screen - Advanced Search Database - MEDLINE Complete	Display
S27	TI "Chronic illness*" OR "Chronic disease*" OR epilepsy OR diabetes OR asthma OR "sickle cell disease" OR transplant* OR "spina bifida" OR "cystic fibrosis" OR "human immunodeficiency virus" OR HIV OR arthritis OR "inflammatory bowel disease*" OR cancer OR "chronic pain" OR "gastrointestinal disease*" OR "communicable disease"	Expanders - Apply related words; Apply equivalent subjects Search modes - Find all my search terms	Interface - EBSCOhost Research Databases Search Screen - Advanced Search Database - MEDLINE Complete	Display
S26	(MH "Gastrointestinal Diseases")	Expanders - Apply related words; Apply equivalent subjects Search modes - Find all my search terms	Interface - EBSCOhost Research Databases Search Screen - Advanced Search Database - MEDLINE Complete	Display
S25	(MH "Chronic Pain")	Expanders - Apply related words; Apply equivalent subjects Search modes - Find all my search terms	Interface - EBSCOhost Research Databases Search Screen - Advanced Search Database - MEDLINE Complete	Display
S24	(MH "Neoplasms")	Expanders - Apply related words; Apply equivalent subjects Search modes - Find all my search terms	Interface - EBSCOhost Research Databases Search Screen - Advanced Search Database - MEDLINE Complete	Display
S23	(MH "Inflammatory Bowel Diseases")	Expanders - Apply related words; Apply equivalent subjects Search modes - Find all my search terms	Interface - EBSCOhost Research Databases Search Screen - Advanced Search Database - MEDLINE Complete	Display
S22	(MH "Arthritis, Juvenile")	Expanders - Apply related words; Apply equivalent subjects Search modes - Find all my search terms	Interface - EBSCOhost Research Databases Search Screen - Advanced Search Database - MEDLINE Complete	Display
S21	(MH "HIV")	Expanders - Apply related words; Apply equivalent subjects Search modes - Find all my search terms	Interface - EBSCOhost Research Databases Search Screen - Advanced Search Database - MEDLINE Complete	Display
S20	(MH "Cystic Fibrosis")	Expanders - Apply related words; Apply equivalent subjects Search modes - Find all my search terms	Interface - EBSCOhost Research Databases Search Screen - Advanced Search Database - MEDLINE Complete	Display
S19	(MH "Spinal Dysraphism")	Expanders - Apply related words; Apply equivalent subjects Search modes - Find all my search terms	Interface - EBSCOhost Research Databases Search Screen - Advanced Search Database - MEDLINE Complete	Display
S18	(MH "Transplants")	Expanders - Apply related words; Apply equivalent subjects Search modes - Find all my search terms	Interface - EBSCOhost Research Databases Search Screen - Advanced Search Database - MEDLINE Complete	Display
S17	(MH "Asthma")	Expanders - Apply related words; Apply equivalent subjects Search modes - Find all my search terms	Interface - EBSCOhost Research Databases Search Screen - Advanced Search Database - MEDLINE Complete	Display
S16	(MH "Anemia, Sickle Cell")	Expanders - Apply related words; Apply equivalent subjects Search modes - Find all my search terms	Interface - EBSCOhost Research Databases Search Screen - Advanced Search Database - MEDLINE Complete	Display
S15	(MH "Diabetes Mellitus")	Expanders - Apply related words; Apply equivalent subjects Search modes - Find all my search terms	Interface - EBSCOhost Research Databases Search Screen - Advanced Search Database - MEDLINE Complete	Display

S14	(MH "Epilepsy")	Expanders - Apply related words; Apply equivalent subjects Search modes - Find all my search terms	Interface - EBSCOhost Research Databases Search Screen - Advanced Search Database - MEDLINE Complete	Display
S13	(MH "Chronic Disease")	Expanders - Apply related words; Apply equivalent subjects Search modes - Find all my search terms	Interface - EBSCOhost Research Databases Search Screen - Advanced Search Database - MEDLINE Complete	Display
S12	S9 OR S10 OR S11	Expanders - Apply related words; Apply equivalent subjects Search modes - Find all my search terms	Interface - EBSCOhost Research Databases Search Screen - Advanced Search Database - MEDLINE Complete	Display
S11	AB Adherence OR compliance OR concordance OR non-adherence OR self-management OR management OR "self care" OR "self-care" OR "poorly controlled"	Expanders - Apply related words; Apply equivalent subjects Search modes - Find all my search terms	Interface - EBSCOhost Research Databases Search Screen - Advanced Search Database - MEDLINE Complete	Display
S10	TI Adherence OR compliance OR concordance OR non-adherence OR self-management OR management OR "self care" OR "self-care" OR "poorly controlled"	Expanders - Apply related words; Apply equivalent subjects Search modes - Find all my search terms	Interface - EBSCOhost Research Databases Search Screen - Advanced Search Database - MEDLINE Complete	Display
S9	(MH "Medication Adherence") OR (MH "Patient Compliance") OR (MH "Treatment Adherence and Compliance")	Expanders - Apply related words; Apply equivalent subjects Search modes - Find all my search terms	Interface - EBSCOhost Research Databases Search Screen - Advanced Search Database - MEDLINE Complete	Display
S8	S5 OR S6 OR S7	Expanders - Apply related words; Apply equivalent subjects Search modes - Find all my search terms	Interface - EBSCOhost Research Databases Search Screen - Advanced Search Database - MEDLINE Complete	Display
S7	AB Parent* or mother* or father* or guardian* or caregiver*	Expanders - Apply related words; Apply equivalent subjects Search modes - Find all my search terms	Interface - EBSCOhost Research Databases Search Screen - Advanced Search Database - MEDLINE Complete	Display
S6	TI Parent* or mother* or father* or guardian* or caregiver*	Expanders - Apply related words; Apply equivalent subjects Search modes - Find all my search terms	Interface - EBSCOhost Research Databases Search Screen - Advanced Search Database - MEDLINE Complete	Display
S5	(MH "Parents") OR (MH "Mothers") OR (MH "Fathers")	Expanders - Apply related words; Apply equivalent subjects Search modes - Find all my search terms	Interface - EBSCOhost Research Databases Search Screen - Advanced Search Database - MEDLINE Complete	Display
S4	S1 OR S2 OR S3	Expanders - Apply related words; Apply equivalent subjects Search modes - Find all my search terms	Interface - EBSCOhost Research Databases Search Screen - Advanced Search Database - MEDLINE Complete	Display
S3	AB Child* OR adolescen* OR youth OR teen* OR "young people" OR infant OR "young adult*"	Expanders - Apply related words; Apply equivalent subjects Search modes - Find all my search terms	Interface - EBSCOhost Research Databases Search Screen - Advanced Search Database - MEDLINE Complete	Display
S2	TI Child* OR adolescen* OR youth OR teen* OR "young people" OR infant OR "young adult*"	Expanders - Apply related words; Apply equivalent subjects Search modes - Find all my search terms	Interface - EBSCOhost Research Databases Search Screen - Advanced Search Database - MEDLINE Complete	Display
S1	(MH "Infant") OR (MH "Child") OR (MH "Adolescent")	Expanders - Apply related words; Apply equivalent subjects Search modes - Find all my search terms	Interface - EBSCOhost Research Databases Search Screen - Advanced Search Database - MEDLINE Complete	Display

Appendix C: Individual Quality Ratings for the Qualitative Review

Study Appendix C	Clear aims?	Appropriate qualitative methodology?	Appropriate research design?	Appropriate recruitment strategy?	Appropriate data collection to address question?	Has the relationship between the researcher and participants been considered?	Have ethical issues been considered?	Was the data analysis rigorous?	Is there a clear statement of findings?	How valuable is the research?	Overall quality rating
Auslander et al. (2010)	2	2	2	2	2	0	0	1	2	2	15 (Moderate)
Denison et al. (2015)	1	2	1	2	1	0	1	1	2	2	13 (Moderate)
Hanna and Guthrie (2001)	2	2	1	1	2	0	1	1	2	2	14 (Moderate)
Heyduck et al. (2015)	2	2	0	1	2	0	2	1	2	2	14 (Moderate)
Hommel et al. (2010)	2	2	2	2	2	0	2	1	2	2	17 (High)
Ivey et al. (2007)	2	2	1	1	1	0	0	1	2	2	12 (Moderate)
Kourrouski and Lima (2009)	2	2	1	1	1	0	2	1	1	1	12 (Moderate)
O'Toole et al. (2019)	2	2	1	1	2	0	2	1	2	2	15 (Moderate)
	2	2	2	2	2	1	1	1	1	2	15 (Moderate)

Study Appendix C	Clear aims?	Appropriate qualitative methodology?	Appropriate research design?	Appropriate recruitment strategy?	Appropriate data collection to address question?	Has the relationship between the researcher and participants been considered?	Have ethical issues been considered?	Was the data analysis rigorous?	Is there a clear statement of findings?	How valuable is the research?	Overall quality rating
Sullivan-Boyai et al. (2014)											

Appendix D: MEDLINE Search String for the Meta-Analysis

EBSCOhost

Thursday, February 27, 2020 5:17:37 AM

#	Query	Limiters/Expanders	Last Run Via	Results
S37	S1 AND S18 AND S21 AND S34 AND S35	Limiters - Date of Publication: 20070101-20201231 Expanders - Apply related words; Apply equivalent subjects Search modes - Find all my search terms	Interface - EBSCOhost Research Databases Search Screen - Advanced Search Database - MEDLINE Complete	Display
S36	S1 AND S18 AND S21 AND S34 AND S35	Expanders - Apply related words; Apply equivalent subjects Search modes - Find all my search terms	Interface - EBSCOhost Research Databases Search Screen - Advanced Search Database - MEDLINE Complete	Display
S35	TI (Intervention* OR program* OR cognitive OR behaviour* OR behavior* or multisystemic or educat*) OR AB (Intervention* OR program* OR cognitive OR behaviour* OR behavior* or multisystemic or educat*)	Expanders - Apply related words; Apply equivalent subjects Search modes - Find all my search terms	Interface - EBSCOhost Research Databases Search Screen - Advanced Search Database - MEDLINE Complete	Display
S34	S30 NOT S33	Expanders - Apply related words; Apply equivalent subjects Search modes - Find all my search terms	Interface - EBSCOhost Research Databases Search Screen - Advanced Search Database - MEDLINE Complete	Display
S33	S31 NOT S32	Expanders - Apply related words; Apply equivalent subjects Search modes - Find all my search terms	Interface - EBSCOhost Research Databases Search Screen - Advanced Search Database - MEDLINE Complete	Display
S32	(MH "Humans")	Expanders - Apply related words; Apply equivalent subjects Search modes - Find all my search terms	Interface - EBSCOhost Research Databases Search Screen - Advanced Search Database - MEDLINE Complete	Display
S31	(MH "Animals+")	Expanders - Apply related words; Apply equivalent subjects Search modes - Find all my search terms	Interface - EBSCOhost Research Databases Search Screen - Advanced Search Database - MEDLINE Complete	Display

S30	S22 OR S23 OR S24 OR S25 OR S26 OR S27 OR S28 OR S29	Expanders - Apply related words; Apply equivalent subjects Search modes - Find all my search terms	Interface - EBSCOhost Research Databases Search Screen - Advanced Search Database - MEDLINE Complete	Display
S29	TI groups OR AB groups	Expanders - Apply related words; Apply equivalent subjects Search modes - Find all my search terms	Interface - EBSCOhost Research Databases Search Screen - Advanced Search Database - MEDLINE Complete	Display
S28	TI trial OR AB trial	Expanders - Apply related words; Apply equivalent subjects Search modes - Find all my search terms	Interface - EBSCOhost Research Databases Search Screen - Advanced Search Database - MEDLINE Complete	Display
S27	TI randomly OR AB randomly	Expanders - Apply related words; Apply equivalent subjects Search modes - Find all my search terms	Interface - EBSCOhost Research Databases Search Screen - Advanced Search Database - MEDLINE Complete	Display
S26	SU drug therapy	Expanders - Apply related words; Apply equivalent subjects Search modes - Find all my search terms	Interface - EBSCOhost Research Databases Search Screen - Advanced Search Database - MEDLINE Complete	Display
S25	TI placebo OR AB placebo	Expanders - Apply related words; Apply equivalent subjects Search modes - Find all my search terms	Interface - EBSCOhost Research Databases Search Screen - Advanced Search Database - MEDLINE Complete	Display

S24	TI randomized OR AB randomized	Expanders - Apply related words; Apply equivalent subjects Search modes - Find all my search terms	Interface - EBSCOhost Research Databases Search Screen - Advanced Search Database - MEDLINE Complete	Display
S23	PT controlled clinical trial	Expanders - Apply related words; Apply equivalent subjects Search modes - Find all my search terms	Interface - EBSCOhost Research Databases Search Screen - Advanced Search Database - MEDLINE Complete	Display
S22	PT randomized controlled trial	Expanders - Apply related words; Apply equivalent subjects Search modes - Find all my search terms	Interface - EBSCOhost Research Databases Search Screen - Advanced Search Database - MEDLINE Complete	Display
S21	S19 OR S20	Expanders - Apply related words; Apply equivalent subjects Search modes - Find all my search terms	Interface - EBSCOhost Research Databases Search Screen - Advanced Search Database - MEDLINE Complete	Display
S20	TI (Adherence OR compliance OR concordance OR non- adherence OR self-management OR "poorly controlled") OR AB (Adherence OR compliance OR concordance OR non- adherence OR self-management OR "poorly controlled")	Expanders - Apply related words; Apply equivalent subjects Search modes - Find all my search terms	Interface - EBSCOhost Research Databases Search Screen - Advanced Search Database - MEDLINE Complete	Display

S19	(MH "Medication Adherence") OR (MH "Patient Compliance") OR (MH "Treatment Adherence and Compliance")	Expanders - Apply related words; Apply equivalent subjects Search modes - Find all my search terms	Interface - EBSCOhost Research Databases Search Screen - Advanced Search Database - MEDLINE Complete	Display
S18	S2 OR S3 OR S4 OR S5 OR S6 OR S7 OR S8 OR S9 OR S10 OR S11 OR S12 OR S13 OR S14 OR S15 OR S16 OR S17	Expanders - Apply related words; Apply equivalent subjects Search modes - Find all my search terms	Interface - EBSCOhost Research Databases Search Screen - Advanced Search Database - MEDLINE Complete	Display
S17	TI ("Chronic illness*" OR "Chronic disease*" OR epilepsy OR diabetes OR asthma OR "sickle cell disease" OR transplant* OR "spina bifida" OR "cystic fibrosis" OR "human immunodeficiency virus" OR HIV OR arthritis OR "inflammatory bowel disease*" OR obesity OR cancer OR "chronic pain" OR "gastrointestinal disease*" OR "communicable disease") OR AB ("Chronic illness*" OR "Chronic disease*" OR epilepsy OR diabetes OR asthma OR "sickle cell disease" OR transplant* OR "spina bifida" OR "cystic fibrosis" OR "human immunodeficiency virus" OR HIV OR arthritis OR	Expanders - Apply related words; Apply equivalent subjects Search modes - Find all my search terms	Interface - EBSCOhost Research Databases Search Screen - Advanced Search Database - MEDLINE Complete	Display

	arthritis OR "inflammatory			
S11	(MH "Arthritis") OR (MH "Arthritis, Juvenile") OR (MH "Arthritis, Rheumatoid")	Expanders - Apply related words; Apply equivalent subjects Search modes - Find all my search terms	Interface - EBSCOhost Research Databases Search Screen - Advanced Search Database - MEDLINE Complete	Display
S10	(MH "HIV")	Expanders - Apply related words; Apply equivalent subjects Search modes - Find all my search terms	Interface - EBSCOhost Research Databases Search Screen - Advanced Search Database - MEDLINE Complete	Display
S9	(MH "Cystic Fibrosis")	Expanders - Apply related words; Apply equivalent subjects Search modes - Find all my search terms	Interface - EBSCOhost Research Databases Search Screen - Advanced Search Database - MEDLINE Complete	Display
S8	(MH "Spinal Dysraphism")	Expanders - Apply related words; Apply equivalent subjects Search modes - Find all my search terms	Interface - EBSCOhost Research Databases Search Screen - Advanced Search Database - MEDLINE Complete	Display
S7	(MH "Transplants")	Expanders - Apply related words; Apply equivalent subjects Search modes - Find all my search terms	Interface - EBSCOhost Research Databases Search Screen - Advanced Search Database - MEDLINE Complete	Display
S6	(MH "Anemia, Sickle Cell")	Expanders - Apply related words; Apply equivalent subjects Search modes - Find all my search terms	Interface - EBSCOhost Research Databases Search Screen - Advanced Search Database - MEDLINE Complete	Display

S5	(MH "Asthma")	Expanders - Apply related words; Apply equivalent subjects Search modes - Find all my search terms	Interface - EBSCOhost Research Databases Search Screen - Advanced Search Database - MEDLINE Complete	Display
S4	(MH "Diabetes Mellitus")	Expanders - Apply related words; Apply equivalent subjects Search modes - Find all my search terms	Interface - EBSCOhost Research Databases Search Screen - Advanced Search Database - MEDLINE Complete	Display
S3	(MH "Epilepsy")	Expanders - Apply related words; Apply equivalent subjects Search modes - Find all my search terms	Interface - EBSCOhost Research Databases Search Screen - Advanced Search Database - MEDLINE Complete	Display
S2	(MH "Chronic Disease") OR (MH "Noncommunicable Diseases")	Expanders - Apply related words; Apply equivalent subjects Search modes - Find all my search terms	Interface - EBSCOhost Research Databases Search Screen - Advanced Search Database - MEDLINE Complete	Display
S1	((MH "Infant") OR (MH "Child") OR (MH "Child, Preschool") OR (MH "Adolescent")) OR TI (Child* OR adolescen* OR youth OR teen* OR "young people" OR infant OR "young adult*") OR AB (Child* OR adolescen* OR youth OR teen* OR "young people" OR infant OR "young adult*")	Expanders - Apply related words; Apply equivalent subjects Search modes - Find all my search terms	Interface - EBSCOhost Research Databases Search Screen - Advanced Search Database - MEDLINE Complete	Display

Appendix E: Individual Risk of Bias Ratings for the Meta-Analysis

Study	Random sequence generation	Allocation concealment	Blinding of outcome assessment	Incomplete outcome data	Selective outcome reporting	Other bias?	Overall rating
Appendix E							
Bhana et al (2014)	U	U	U	U	U	L	H
Bruzzese et al (2008)	U	L	U	L	U	L	M
Bruzzese et al (2010)	L	L	U	L	U	L	M
Carlsen et al (2017)	L	L	U	L	L	L	M
Chawana et al (2017)	L	L	U	L	L	L	M
Davis et al (2019)	U	U	U	U	U	L	H
Ellis et al (2007a)	U	L	L	L	U	L	M
Ellis et al (2012)	L	L	U	L	L	L	M
Ellis et al (2019)	L	L	Objective adherence data – L Self-report adherence and QoL data - U	L	U	L	M
Goyal et al (2017)	U	U	Objective adherence data – L Self-report adherence and QoL data - U	L	L	L	M
Green et al (2017)	L	U	Objective adherence data – L Self-report adherence and QoL data – U	L	L	L	M
Greenley et al (2015)	L	L	Objective adherence data – L QoL – U	L	U	L	M
Hommel et al (2012)	U	U	Objective adherence data – L Self-report adherence data – U	L	L	L	M
Jaser et al (2014)	L	U	U	L	U	L	M
	L	U		L	L	L	M

Study	Random sequence generation	Allocation concealment	Blinding of outcome assessment	Incomplete outcome data	Selective outcome reporting	Other bias?	Overall rating
Appendix E							
Jaser et al (2019)			Objective adherence data – L Self-report adherence and QoL data – U				
Jaser et al (2020)	L	U	U	L	L	L	M
Johnson et al (2015)	L	U	U	L	L	L	M
Joseph et al (2007)	L	U	U	U	U	L	H
Kichler et al (2013)	U	U	U	H	U	L	H
Kohut et al (2016)	L	L	U	L	U	L	M
Mayer-Davis et al (2018)	L	U	U	L	L	L	M
Mosnaim et al (2013)	L	U	Objective adherence data – L Self-report adherence data – U	L	L	L	M
Mulvaney et al (2010)	U	L	U	U	U	L	H
Naar-King et al (2014)	U	U	U	L	L	L	M
Nansel et al (2007)	L	U	U	L	L	L	M
Raiff et al (2016)	U	U	L	H	U	L	H
Rikkers-Mutsaerts et al (2012)	L	L	U	L	U	L	M
Stanger et al (2018)	L	U	Objective adherence data – L Family functioning self-report data – H	L	L	L	M for A H for FF
Stinson et al (2010)	L	L	H	L	L	L	H
Stinson et al (2016)	L	L	U	H	U	L	H
Stinson et al (2020)	L	L	U	H	L	L	H

Study	Random sequence generation	Allocation concealment	Blinding of outcome assessment	Incomplete outcome data	Selective outcome reporting	Other bias?	Overall rating
Appendix E							
Tseng et al (2020)	L	L	U	L	L	L	M
Whittemore et al (2016)	L	L	U	H	L	L	H
Willis et al (2019)	L	U	U	U	U	L	H
Wysocki et al (2007)	U	U	U	L	U	L	H

Cluster RCT	Random process	Timing and identification of participants	Intended interventions	Missing outcomes data	Measurements of outcomes	Selection of the reported result	Overall rating
Kosse et al (2019)	Some concerns	High risk	Some concerns	Some concerns	High risk	High risk	H

Appendix F: Publication Bias Inspection

Rank correlation test outcomes for primary and secondary outcomes and exploratory outcomes at posttreatment and follow-up

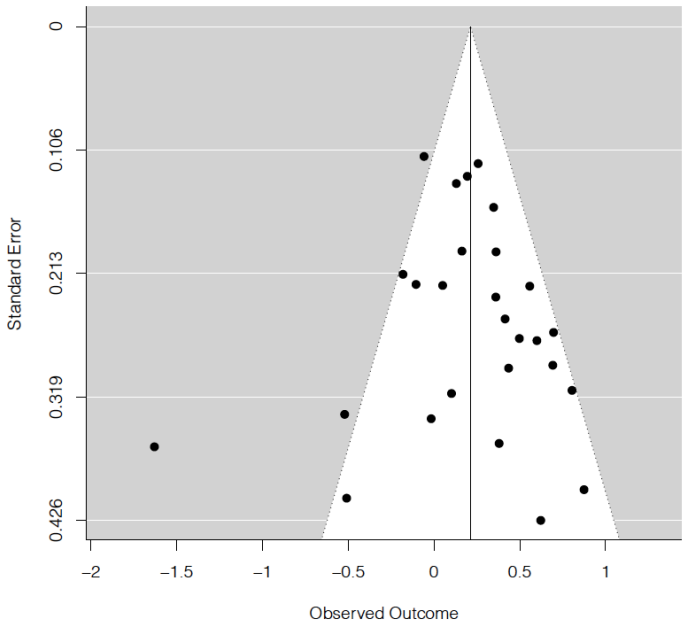
Outcome	Kendall's tau (τ)	Significant
Primary		
Adherence Posttreatment	0.0940	0.5084
Adherence Follow-up	0.0909	0.7612
Secondary		
Quality of Life Posttreatment	-0.2967	0.1572
Family Functioning Posttreatment	-0.2000	0.7194
Quality of Life Follow-up	-0.7143.	**0.0302**
Family Functioning Follow-up	-0.3333	0.7500
Exploratory		
Objective Measures Posttreatment	-0.2381	0.5619
Objective Measures Follow-up	-0.2000	0.8167
Adolescent Self-Report Posttreatment	-0.0667	0.6982

Outcome	Kendall's tau (τ)	Significant
Adolescent Self-Report Follow-up	-0.4286	0.1789
Parent Self-Report Posttreatment	0.0222	1.0000
Parent Self-Report Follow-up	-1.0000	0.0833
Adolescent and Parent Measures Posttreatment	0.3333	0.7500
Adolescent and Parent Measures Follow-up	N/A	N/A

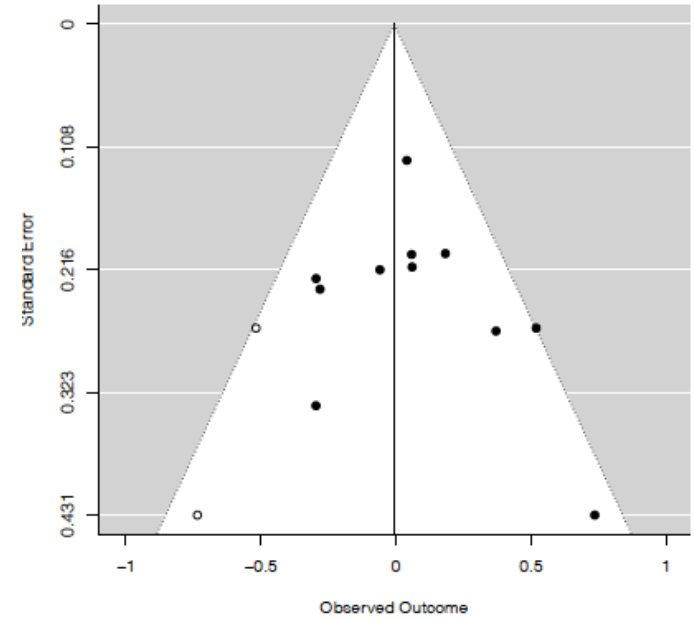
Note. Appendix F continued.

Random effects model funnel plots for outcomes at posttreatment and follow-up for all analyses. Using the trim-and-fill methods, open circles show any estimated missing null studies

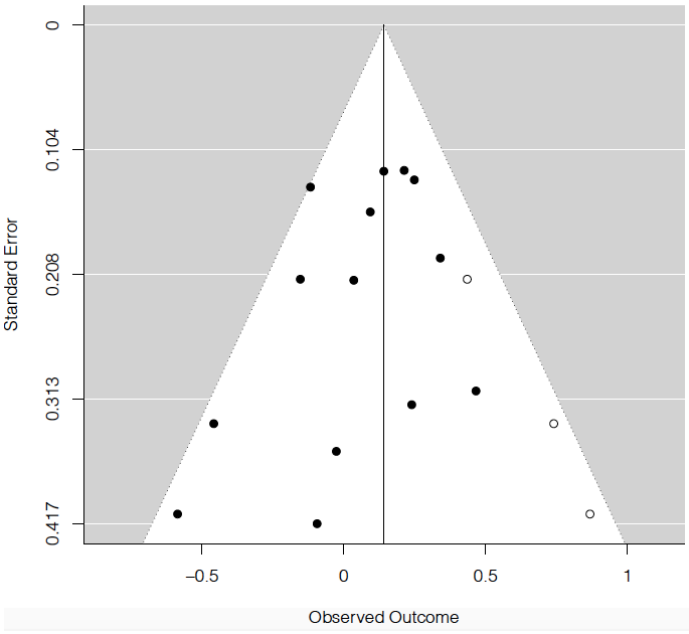
Adherence Posttreatment



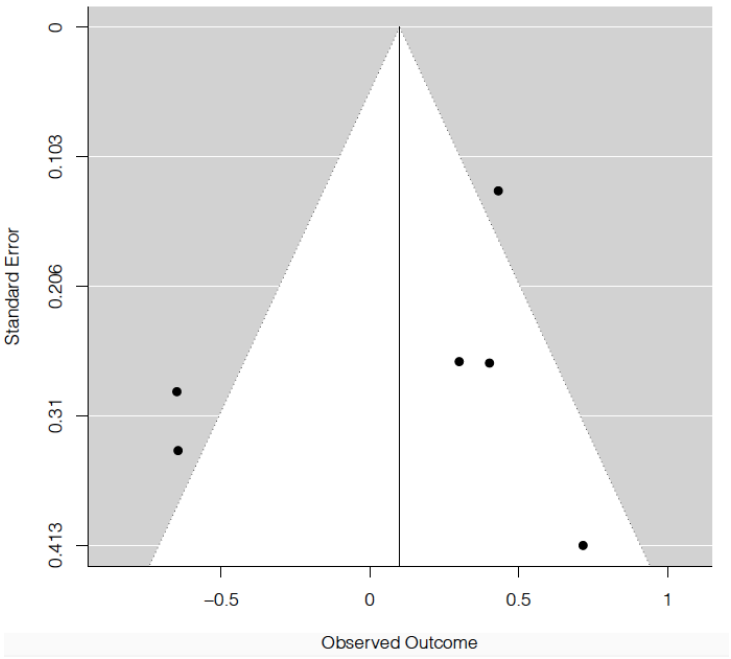
Adherence Follow-up



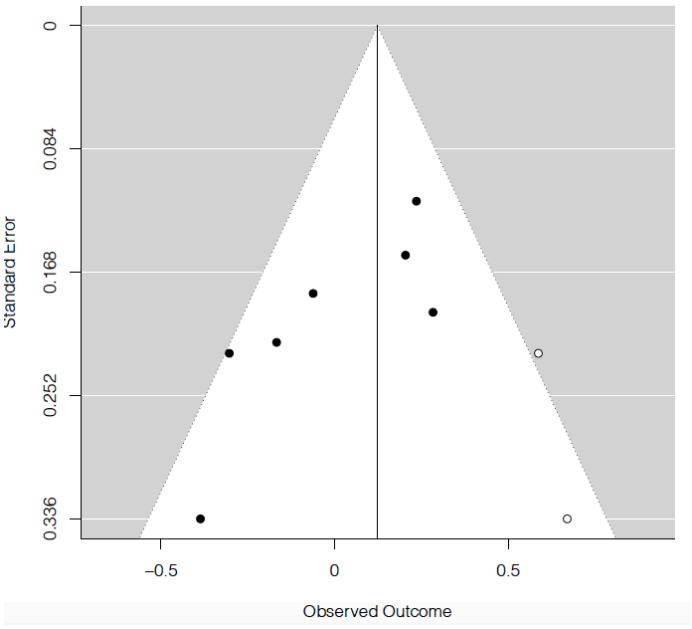
Quality of life outcomes at posttreatment



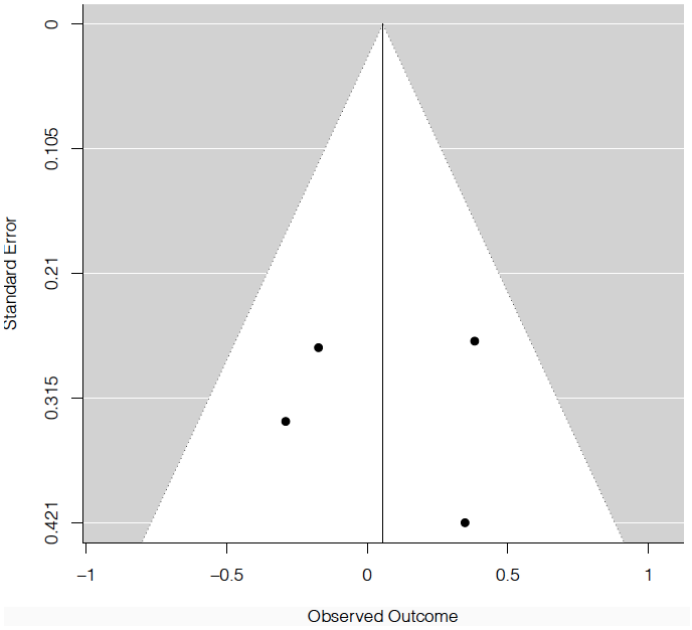
Family functioning outcomes at posttreatment



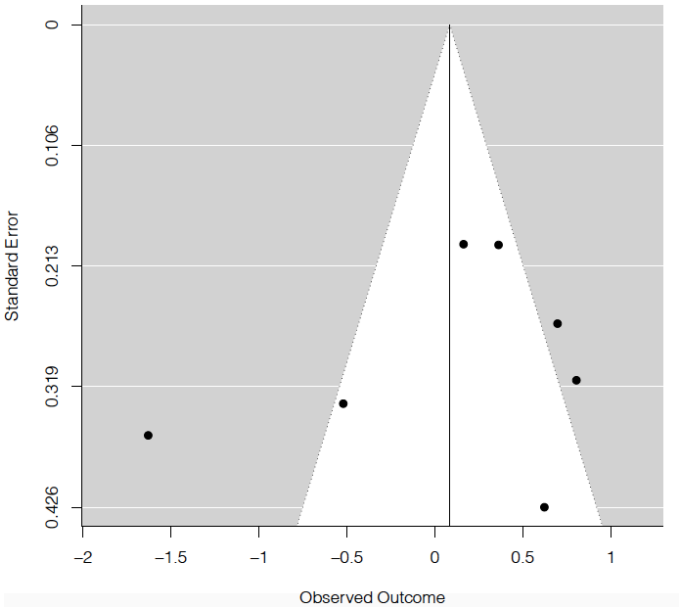
Quality of life outcomes at follow-up



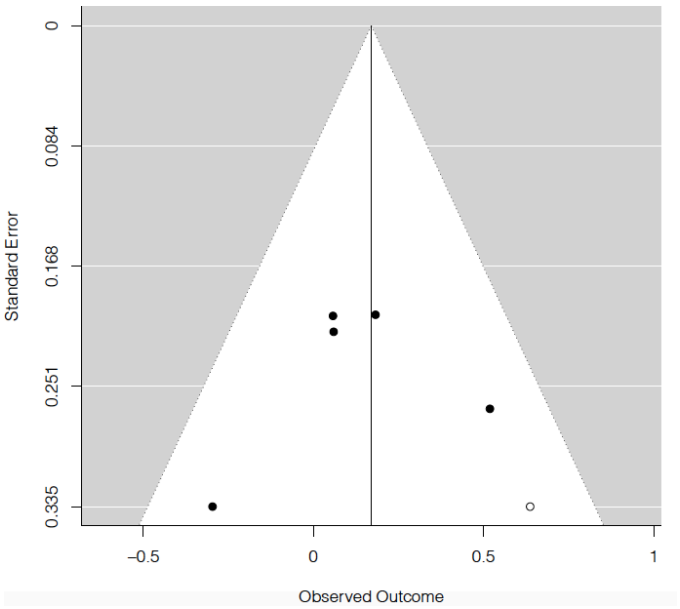
Family functioning outcomes at follow-up



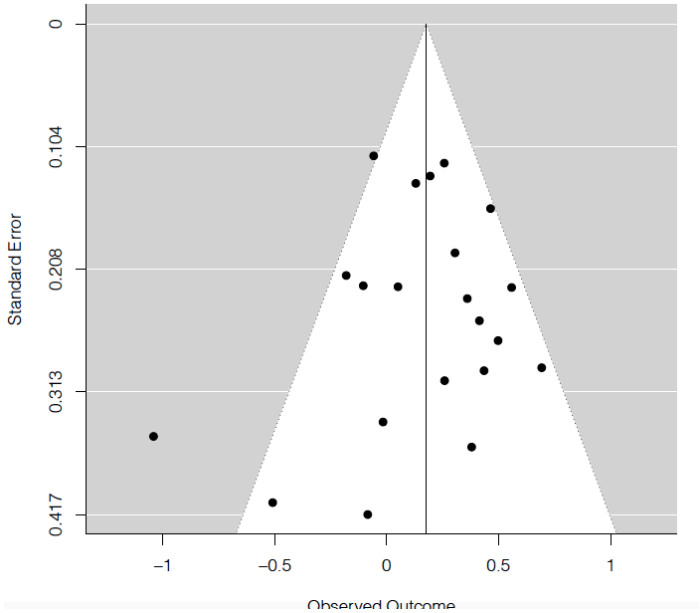
Objective Measures (Adherence) Posttreatment



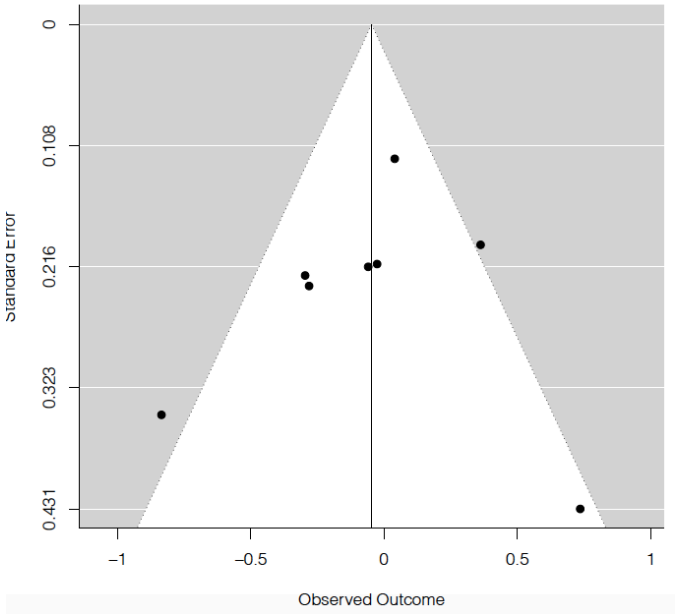
Objective Measures (Adherence) Follow-up



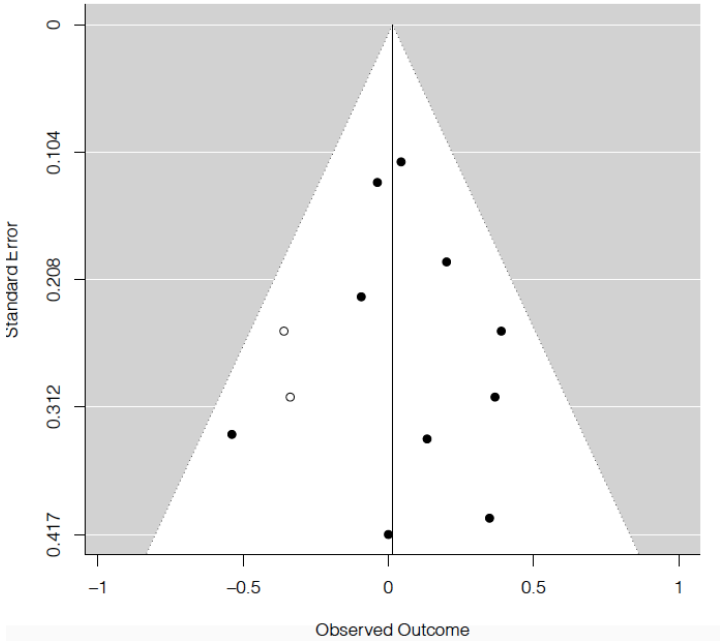
Adolescent Self-Report Measures (Adherence) Posttreatment



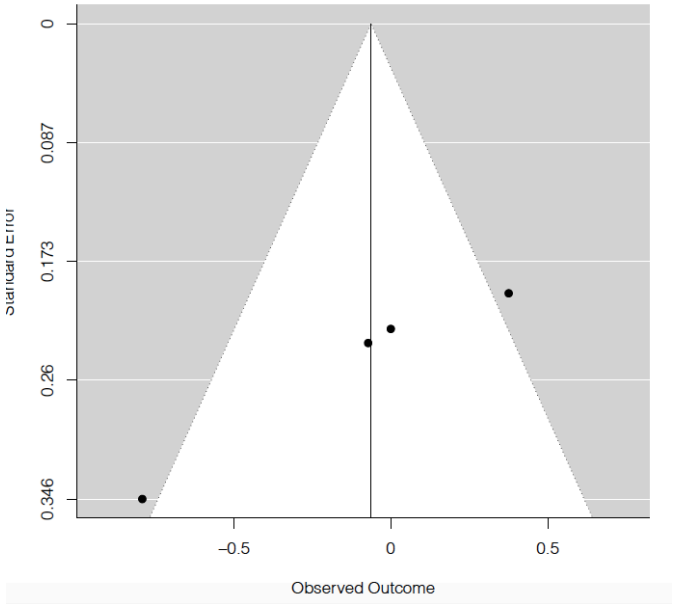
Adolescent Self-Report Measures (Adherence) Follow-up



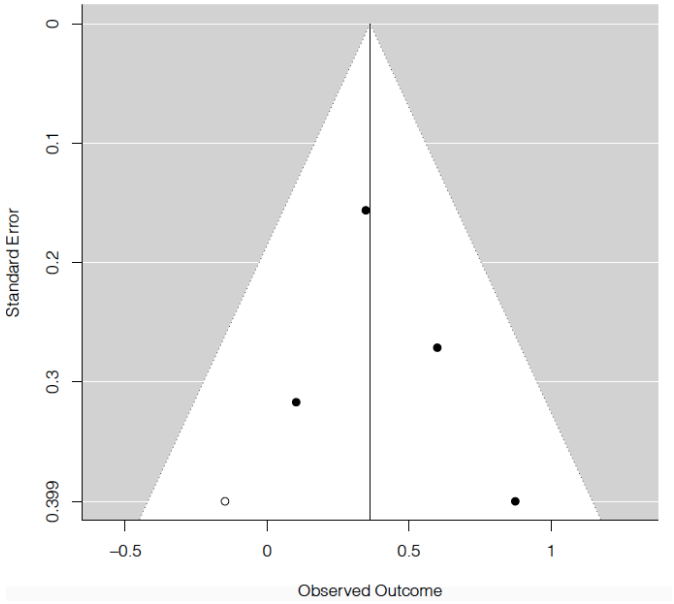
Parent Self-Report Measures (Adherence) Posttreatment



Parent Self-Report Measures (Adherence) Follow-up



Joint Adolescent and Parent Measures (Adherence) Posttreatment



Joint Adolescent and Parent Measures (Adherence) Follow-up

N/A

Appendix G: Outcome Selection Procedure for Adherence Measures

1. Similar to Pai and McGrady (2014), when the study included multiple adherence measures, the most well-established outcome should be selected. More specifically, objective electronic monitoring measures are to be selected over self-report measures (Quittner et al., 2008).

2. The most well-established, validated and reliable outcome for the chronic illness being investigated in the paper should be selected, based on reviews by Plevinsky, Gutierrez-Colina et al. (2020) and Quittner et al. (2008).

3. Select the measure that the authors cite as their primary outcome measure.

4. Use a random number generator to select the measure. This is unlikely given the decisions made before this final resort.

Note. If there are multiple respondents for the same outcome, including teen, parent or physician report then teen-reported data is preferably extracted first, followed by parent and then finally followed by physician report. This a similar procedure to a recent meta-analysis including adolescents with chronic illness and their parents (Law et al., 2019). If there are only subscales instead of total scores reported and authors do not respond and provide total score data, then subscales will be averaged to calculate one effect size for each study.

Appendix H: Outcome Selection Procedure for Quality of Life and Family Functioning Outcomes, Respectively

1. If there are multiple quality of life outcomes, the most well-established, validated and reliable outcome should be selected. This will be based on the psychometric properties of the measured used.

2. Select the measure that the authors cite as their primary outcome measure.

3 Use a random number generator to select the measure. This is unlikely given the decisions made before this final resort.

Note. If there are multiple respondents for the same outcome, including teen, parent or physician report then teen-reported data is preferably extracted first, followed by parent and then finally followed by physician report. This a similar procedure to a recent meta-analysis (Law et al., 2019). If there are only subscales instead of total scores reported and authors do not respond and provide total score data, then subscales will be averaged to calculate one effect size for each study.

1. If there are multiple family functioning outcomes, the most well-established, validated and reliable outcome should be selected. This will be based on the psychometric properties of the measured used.

2. Select the measure that the authors cite as their primary outcome measure.

3. Use a random number generator to select the measure. This is unlikely given the decisions made before this final resort.

Note. If there are multiple respondents for the same outcome, including teen, parent or other family member report then parent/caregiver-reported data is preferably extracted first, followed by teen and then finally followed by other family member report. This a similar procedure to a recent meta-analysis (Law et al., 2019) and was decided given that parents will likely have a broader view of the functioning of the family.

Appendix I: Holm-Bonferroni Correction for All Analyses

Holm-Bonferroni correction used for all possible moderation analyses

Outcome	Number of comparisons	Adjusted p-value[a]	First p-value that was non-significant
Adherence posttreatment	5	0.01	First
Adolescent self-report measures posttreatment	5	0.01	First

[a]Using the Bonferroni method, any p-value were non-significant if equal to or greater than this.

Holm-Bonferroni correction used for all possible subgroup analyses

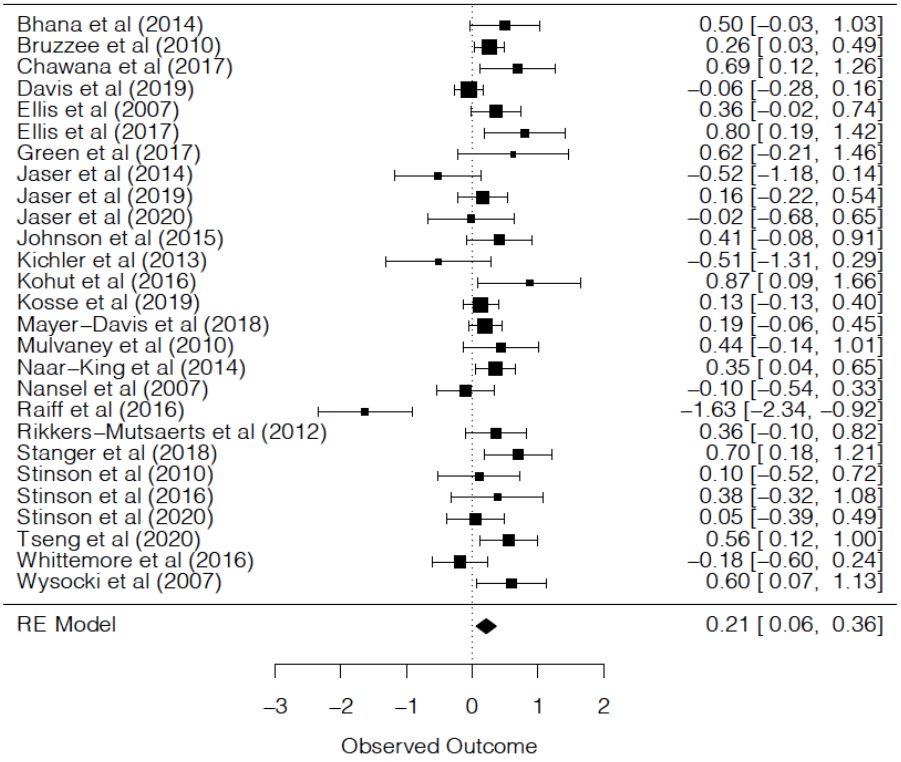
Outcome	Number of comparisons	Adjusted p-value[a]	First p-value that was non-significant
Adherence posttreatment	10	0.025	Ninth
Adolescent self-report measures posttreatment	10	0.007	Fourth

[a]Using the Bonferroni method, any p-value were non-significant if equal to or greater than this.

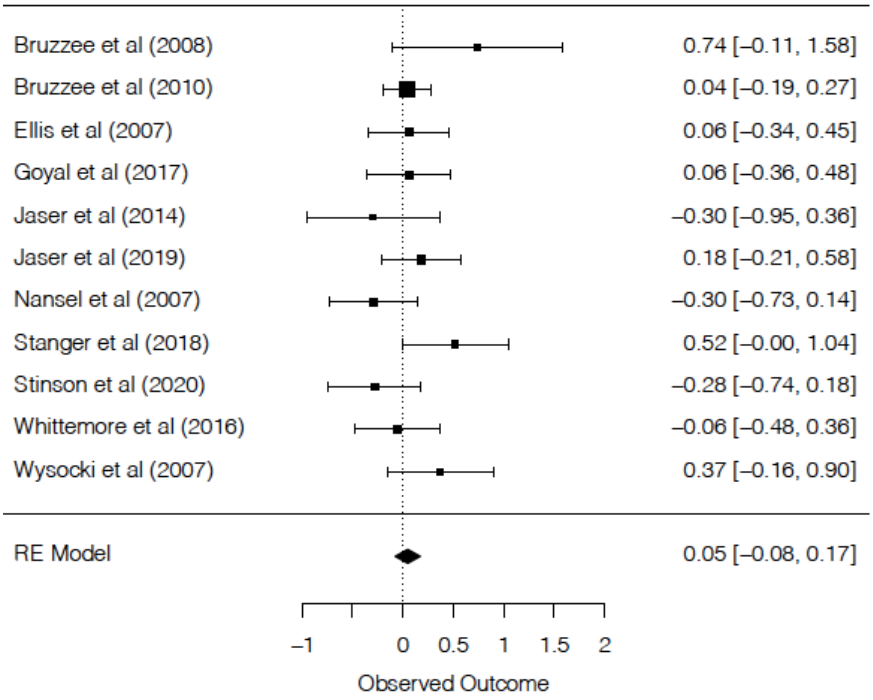
Appendix J: Additional Forest Plots for Primary and Secondary Analyses

Forest plots not included in the main paper due to formatting.

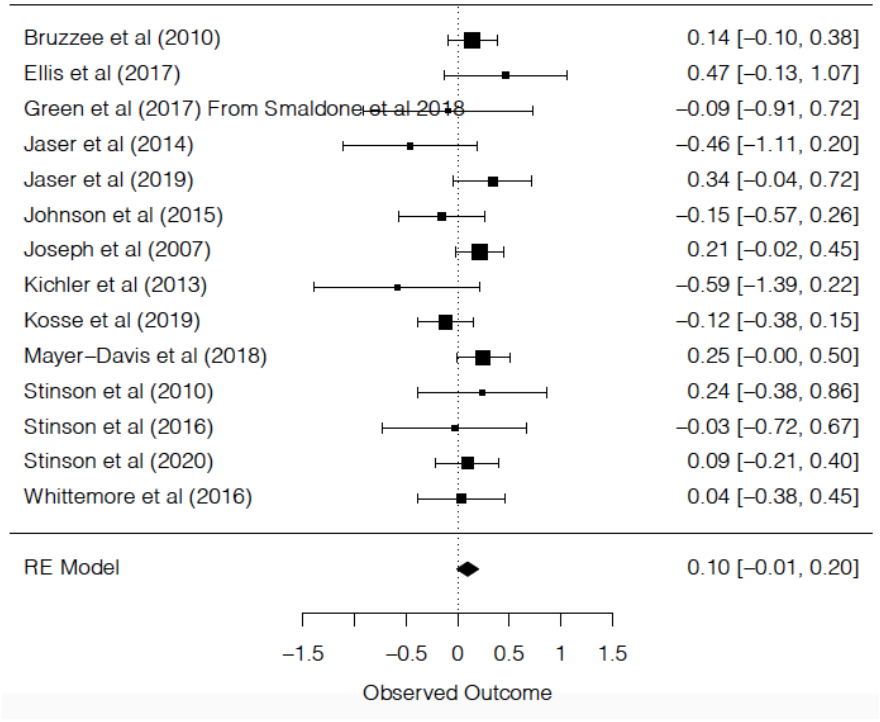
Adherence posttreatment (including high risk of bias studies)



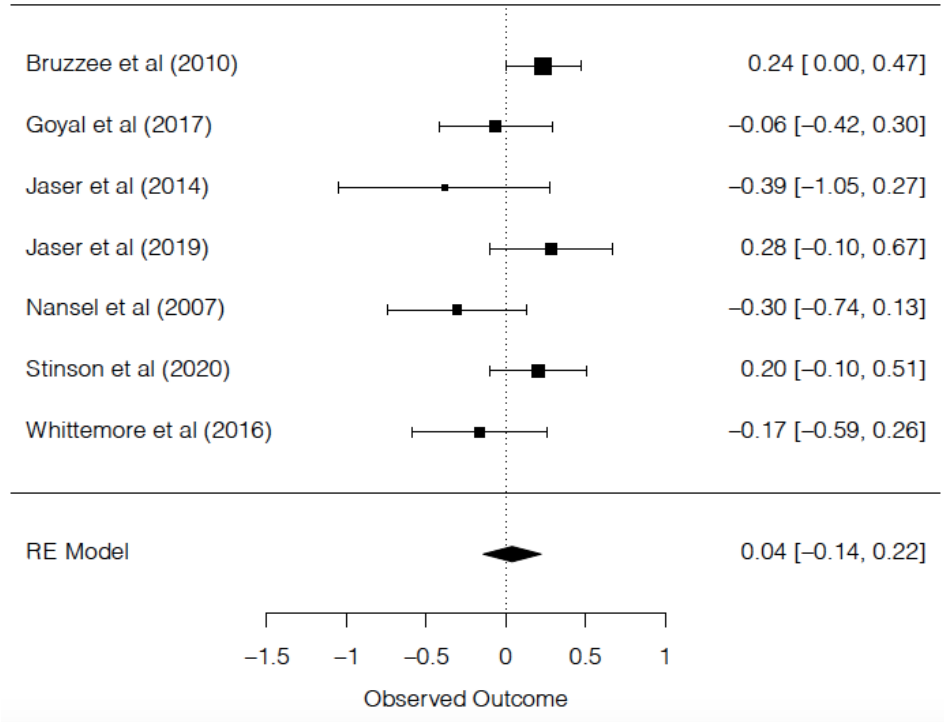
Adherence follow-up (including high risk of bias studies)



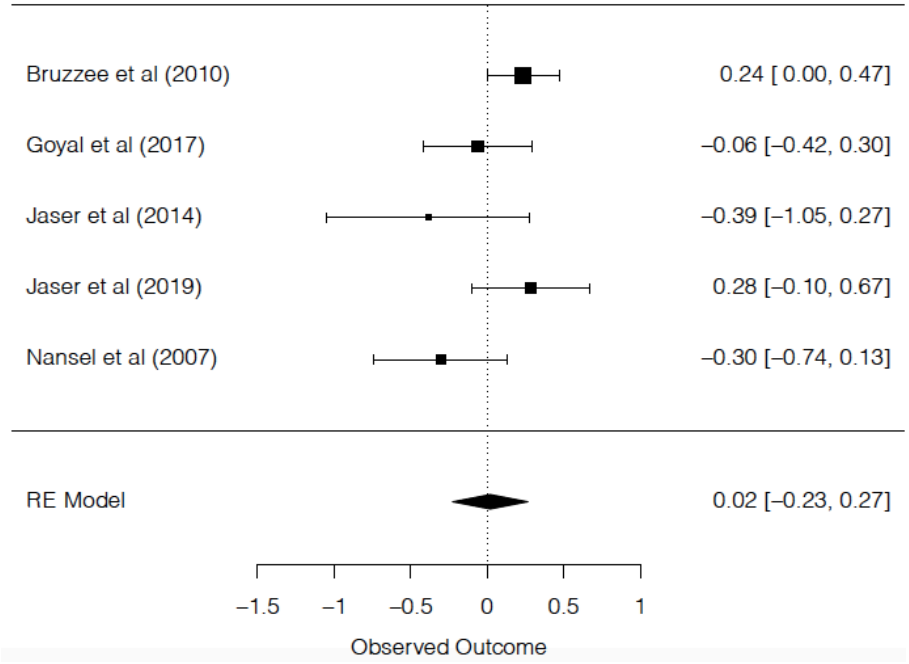
Quality of life outcomes posttreatment (including high risk of bias studies)



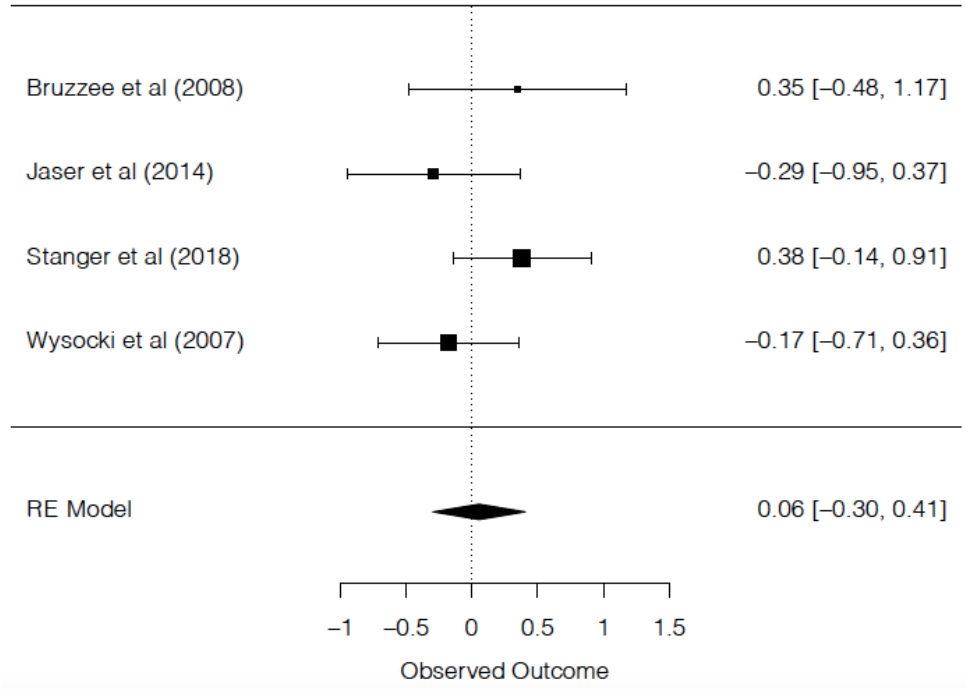
Quality of life outcomes follow-up (including high risk of bias studies)



Quality of life outcomes follow-up (excluding high risk of bias studies)



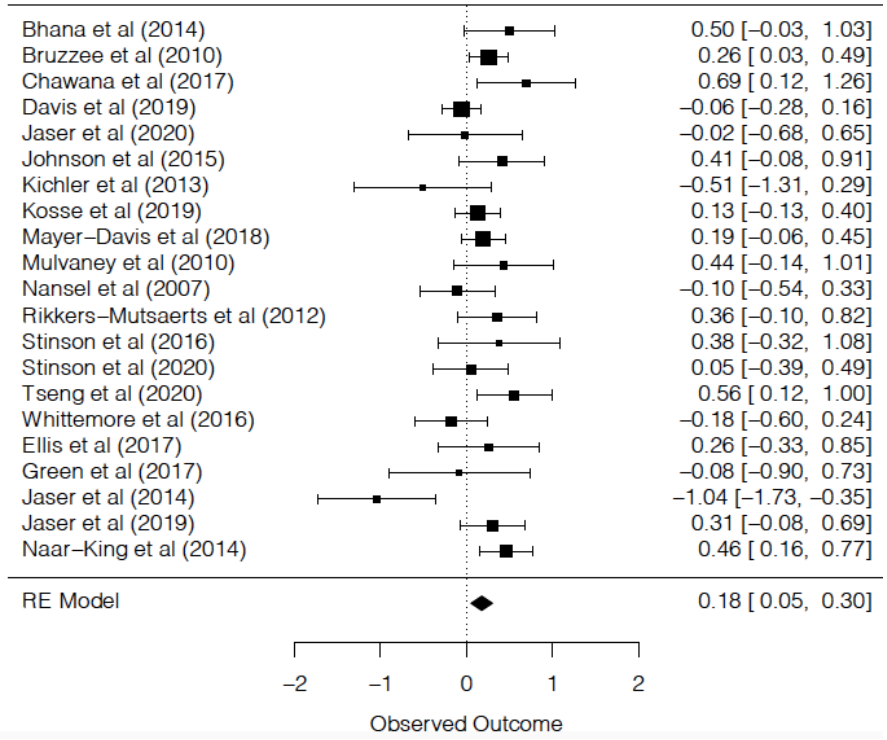
Family functioning outcomes follow-up (including high risk of bias studies)



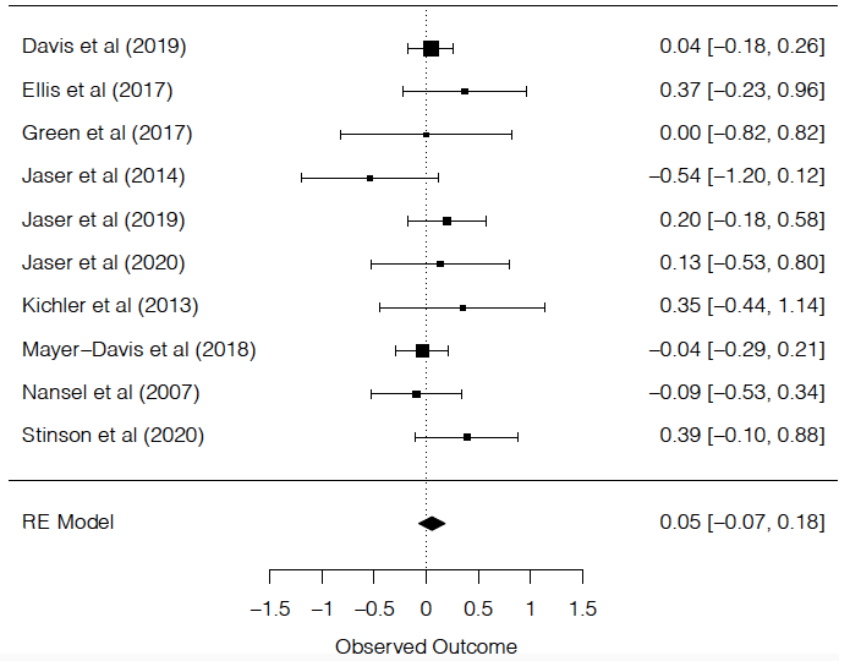
Appendix K: Additional Forest Plots for Exploratory Analyses

Forest plots not included in the main paper due to formatting.

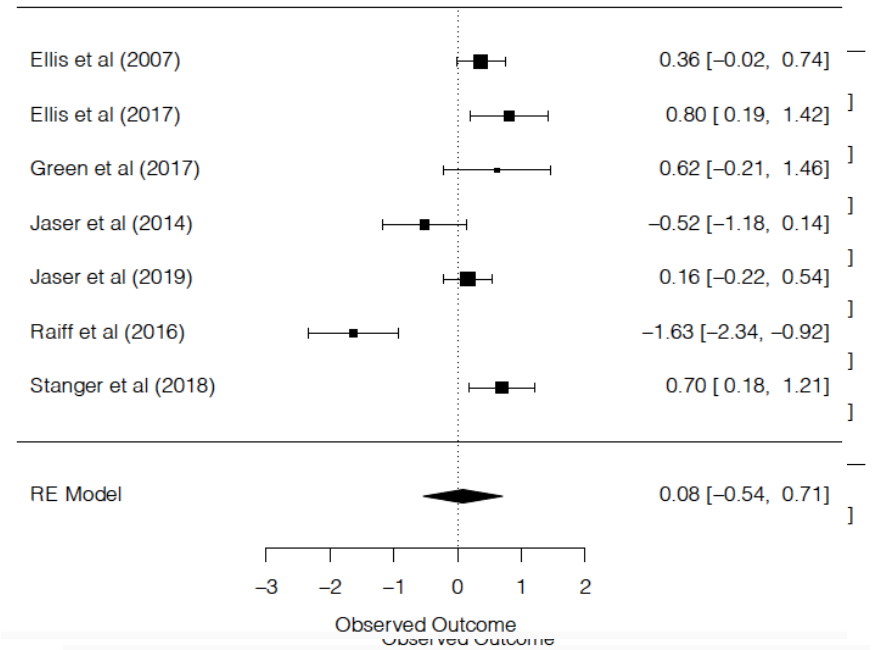
Adolescent self-report posttreatment (including high risk of bias studies)



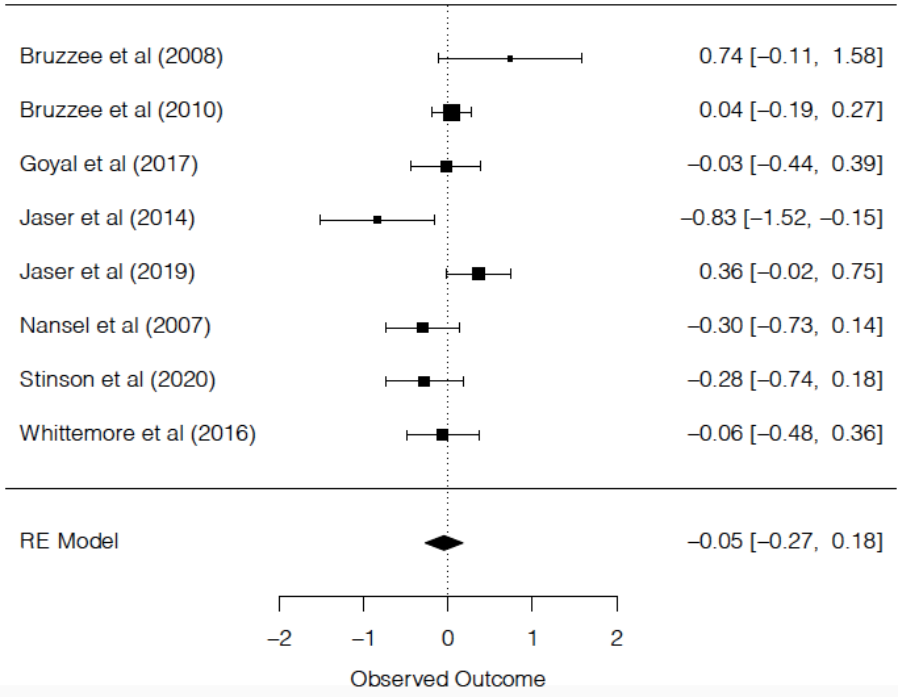
Parent report posttreatment (including high risk of bias studies)



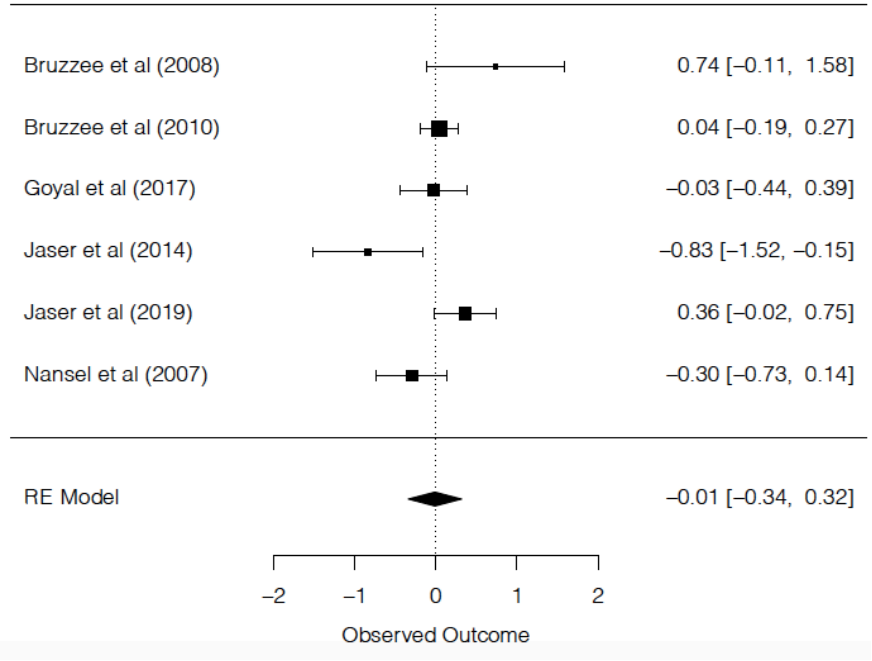
Objective measures posttreatment (including high risk of bias studies)



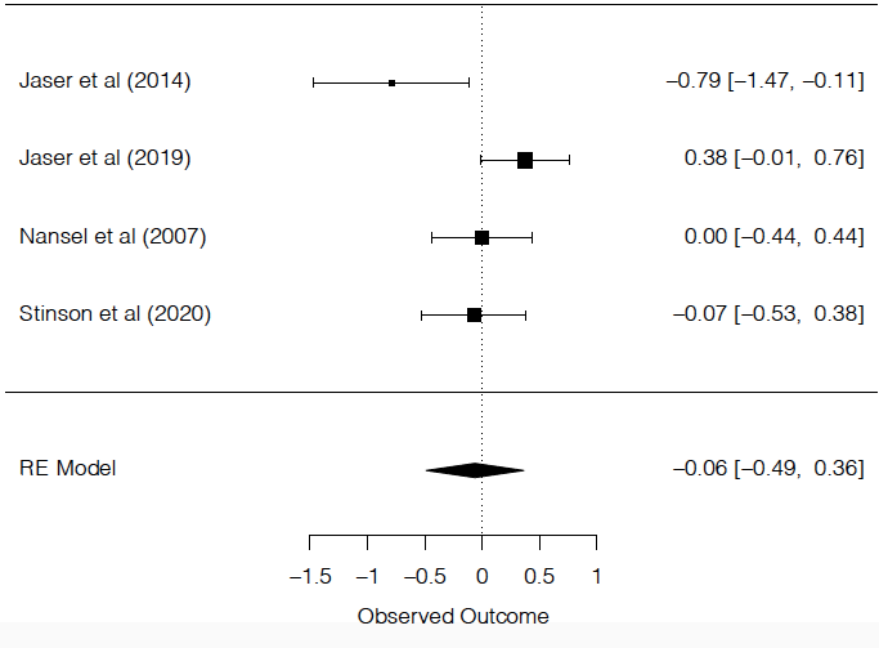
Adolescent self-report measures follow-up (including high risk of bias studies)



Adolescent self-report measures follow-up (excluding high risk of bias studies)



Parent report measures follow-up (including high risk of bias studies)



Objective measures follow-up (including high risk of bias studies)