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**Experiences of Interventions Aiming to Improve the Mental Health and Wellbeing of  
Children and Young People with a Long-Term Physical Condition: A Systematic  
Review and Meta-ethnography**

**Short Title:**

**Qualitative synthesis of mental health and wellbeing interventions for children with  
long-term physical conditions**

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## **Abstract**

### **Background**

Children and young people with long term physical health conditions are at increased risk of experiencing mental health and wellbeing difficulties. However, there is a lack of research that explores the experiences of, and attitudes towards interventions aiming to improve their mental health and wellbeing. This systematic review seeks to address this gap in the literature by exploring what children and young people with long term conditions, their caregivers and health practitioners perceive to be important aspects of interventions aiming to improve their mental health and wellbeing.

### **Methods**

An information specialist searched five academic databases using pre-defined criteria for qualitative evaluations of interventions aiming to improve the mental health or wellbeing of children with long term physical conditions. Reviewers also performed supplementary citation and grey literature searches. Two reviewers independently screened titles, abstracts and full texts which met the inclusion criteria and conducted data extraction and quality assessment. Meta-ethnography was used to synthesise the findings.

### **Results**

Screening identified 60 relevant articles. We identified five overarching constructs through the synthesis: i) Getting In and Staying In, ii) Therapeutic Foundation, iii) Social Support, iv) A Hopeful Alternative and v) Empowerment. The line of argument which links these constructs together indicates that when interventions can provide an environment that allows young people to share their experiences and build empathetic relationships; it can enable participants to access social support and increase feelings of hope and empowerment.

### **Conclusion**

These findings may provide a framework to inform the development of mental health interventions for this population, and evaluate existing interventions which already include some of the components or processes identified by this research. Further research is needed to establish which of the constructs identified by the line of argument are most effective in improving the mental wellbeing of young people living with long term conditions.

## Introduction

Children and young people (CYP) living with long term physical health conditions (LTCs) are up to four times more likely to experience mental health or wellbeing difficulties than their physically healthy peers (Hysing et al., 2007; Piquart & Shen, 2011). The negative impact on quality of life of mental health problems alone can be as great as, or even more significant, than the impact of the LTC (Baca, 2011). Mental ill-health can also exacerbate physical health symptoms, which may then increase the risk of poorer long term outcomes, such as symptom control and medication adherence (Lustman & Clouse, 2005).

Between £8 and £13 billion of NHS spending in England is linked to co-morbid mental health problems among adults and children with LTCs (Naylor, 2012). Psychological interventions can reduce health-care costs associated with inpatient hospital admissions by up to 20 per cent (Chiles et al. 1999; Naylor et al. 2012). This highlights the importance of access to timely, evidence-based mental health and wellbeing treatments for people living with LTCs. Unfortunately, there is a paucity of research examining the effectiveness of psychological interventions for alleviating symptoms of mental ill-health, such as anxiety or depression, amongst CYP with LTCs (Bennett et al. 2015). Existing trials are underpowered and of low quality, with heterogeneity in intervention design and outcome measures limiting the confidence which can be placed in research findings (Bennett et al. 2015; Moore et al. in press).

In addition to research evaluating the effectiveness of interventions aiming to improve the mental health of CYP with LTCs, understanding the experiences and perceptions of CYP and their families and the people delivering interventions, is a crucial step in understanding the impact of existing interventions on the child themselves, as well as the family and social system around them. Patient and practitioner views provide a valuable addition to quantitative measures of effectiveness, by identifying factors that may act as barriers or facilitators to implementing evidence based medicine (Green & Britton, 1998). Patient

attitudes and experiences also offer valuable information regarding the wider context in which health services are delivered (Lorenz et al. 2012) and should inform the development and delivery of new health services (Department of Health, 2015; NHS England, 2014).

Whilst qualitative evidence syntheses have been conducted that explore CYP's experience of their LTCs and the impact on their lives (Lum et al. 2017; Venning et al. 2008), none have examined the views of key stakeholders on interventions aiming to improve the wellbeing of CYP with LTCs, despite the existence of primary qualitative research within this area. Here we aimed to explore the views and experiences of CYP with LTCs, their caregivers and practitioners of interventions intended to improve the mental health and wellbeing of CYP with LTCs. We utilise a systematic review methodology to address this aim in answer to a specific call from the National Institute of Health Research. This article reports the relationship between the key constructs arising from the evidence identified by the qualitative systematic review conducted by Moore et al (in press) and aims to provide an accessible summary of the factors CYP, their parents and professionals identify as being important within mental health and wellbeing interventions for CYP with LTCs.

### **Key definitions**

CYP were defined as individuals aged from birth to 25 years. We considered several of the most commonly used definitions in the literature (Perrin et al. 1993; Pless & Douglas, 1971; Stein et al. 1993) to define long-term physical health conditions as:

*Diagnosed physical health conditions, with an expected duration of at least three months, where cure is considered unlikely, causing limitations in ordinary activities and necessitating medical care, or related services, beyond what is usual for age in question.<sup>1</sup>*

This review considers the term “mental health” to encompass more than the absence of

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<sup>1</sup> This was the working definition used for this review. We recognise that for some LTCs, such as cancer, 5 year survival rates are increasing. CYP with learning/intellectual disabilities were excluded from this review as this population group require additional adaptation to both intervention content and delivery, making the review's scope too broad to incorporate findings into a meaningful synthesis.

psychiatric disorders, to include the concepts of adjustment, wellbeing and coping, as is consistent with other well-known definitions of mental health (Keyes, 2005; Nastasi & Borja, 2016). The current review was, therefore, not restricted to samples with emotional or behavioural symptoms.

### **Methods**

The review protocol was registered on PROSPERO (doi:10.15124/CRD42015027353), conducted according to best practice guidelines (Centre for Reviews and Dissemination, 2009) and reported according to the ENTREQ statement (Tong, 2012).

#### **Search strategy and article selection**

The pre-planned strategy used controlled headings (e.g. MeSH) and free-text searching. Search terms were grouped according to four concepts: children and young people, mental health, long term physical conditions and type of qualitative study design or analysis (See Appendix A for an example search strategy used within PsycINFO). An Information Specialist (MR) entered the search terms into five electronic databases; MEDLINE (including MEDLINE in-process), PsycINFO, CINAHL, HMIC and Conference Proceedings Citation Index, on the 19<sup>th</sup> April 2016 and searched the website OpenGrey via <http://www.opengrey.eu/> on 23<sup>rd</sup> June 2016. Two reviewers (LS, MN) and an Information Specialist (JTB) supplemented the electronic searches with forward and backward citation chasing based upon included studies and other key reviews identified through the search process. A CYP advisory group and clinical topic-experts (see Moore et al. (in press)) identified forty-six websites (see Appendix B) which were searched for primary qualitative research .

A group of eight reviewers (LS, DM, MN, IR, JTB, JTC, MR, VB) shared independent screening of the titles and abstracts of the identified records, with two reviewers screening each article against the pre-determined inclusion criteria outlined in Table 1. Full texts of

potentially eligible articles were screened in the same way. Disagreements were resolved through discussion between two members of the core review team (DM, MN, LS).

Reviewers recorded screening decisions within Endnote X7.

### **Data extraction and critical appraisal**

Four reviewers' (LS, MN, IR, DM) extracted data summarising each article's aims, participants, methods, intervention, findings and study quality into Microsoft Office Excel 2010. DM or LS checked each extraction, with disagreements resolved through discussion. The 13 quality appraisal items used were based upon the Wallace checklist (Wallace et al. 2004). We added an additional question: 'Are the interventions of interest clearly described?' based upon a previous review on intervention experience (Moore et al. 2016). Article quality was not used as a basis for inclusion in the review, but informed the confidence reviewers could place in the synthesis findings. Greater weight was given to papers containing a higher quantity of more conceptual or interpretative data within the synthesis, consistent with other systematic reviews using meta-ethnography (Cochrane Methods: Qualitative and Implementation, 2018).

### **Data synthesis**

Reviewers synthesised the data using meta-ethnography (Noblit & Hare, 1988). This approach has been applied in health research for interpreting findings across qualitative studies (e.g. Frost et al. 2014) and permits the comparison of different data sets; with the interpretation incorporated throughout the analytical process resulting in a final synthesis that constitutes more than "a sum of its parts" (Barnett-Page & Thomas, 2009 p8). Meta-ethnography consists of 4 stages of analysis:

1. *Reading and re-reading the included studies to ensure that reviewers were familiar with each article.*

Began during study selection and continued throughout data synthesis.

2. *Determining how the studies were related*

Structured summaries enabled comparisons of information across different articles (Frost et al. 2014). Summaries included the following information: participant's perspective, type of LTC, mental health intervention target (e.g. procedural anxiety, depression, coping) and any additional non-mental health intervention targets (e.g. LTC management, social-skills).

### *3. Reciprocal translation of studies*

LS extracted quotes and author interpretation of the material provided by study participants, known as first and second-order construct data (Atkins et al. 2008), from the results and discussion of each article. LS compared this data across included studies and developed concept maps to show the relationships between analytical ideas and themes. These were checked by DM. LS used NVivo v.11 software to conduct line-by-line coding, against a framework developed from these concept maps. This process identified new ideas which were either not included, or conflicted with, the initial coding framework, meaning each included article was re-examined in an iterative cycle of analysis.

Reviewers began by synthesising articles containing two or more pages of conceptually rich data. This prioritised articles which had the potential to have the greatest influence on developing constructs. The reviewers then used a maximum-variation purposive sampling strategy (Suri, 2011) to introduce articles containing less interpretive data into the reciprocal translation process. For detail regarding the purposive sampling approach see Table S1 and Appendix C.

### *4. Synthesising translations/creating a line of argument*

Reviewers initially synthesised the views of CYP, parents and practitioners separately to ensure any differences in the views and/or experiences between these groups were identified. LS and DM also considered the experiences of interventions targeting

procedural anxiety separately (see Appendix C). Due to the similarity between themes emerging from these populations, themes for each individual group were not reported. Reviewers considered the influence of CYP's age, LTC and type of intervention received during theme development.

LS grouped related themes together to form five overarching constructs, which were checked by two reviewers (DM, RG) and reported in full within the project report (Moore et al., in press). Each overarching construct was informed by at least 10 articles that achieved 10 or more positive ratings on the Wallace checklist (Wallace, 2004). A line of argument model was developed which describes the relationships between each overarching construct and the themes contributing to them.

## **Results**

Database searches identified 12,285 records, with 1,118 additional records found through a combination of grey literature searches, citation chasing and reference list searches of relevant reviews. Following the screening process outlined in the PRISMA diagram (See Figure 1), 60 articles (from 57 studies) were eligible for inclusion (see Appendix D). Articles from the same study typically described intervention experiences in different levels of detail; influencing the stage they entered the reciprocal translation process (Table S1).

### **Study characteristics**

The 57 included studies were published between 1991 and 2016 and conducted in seven different countries; the most common being the USA (n=25), Canada (n=13), and the United Kingdom (n=9).

The most common data collection strategies utilized within the primary studies were: interviews (n=48), focus groups (n=5) or analysis of posts made within online discussion forums (n=4). Median sample size was 23 participants (range 3-100). Twenty studies included only CYP, one study collected data from only the practitioners delivering the intervention and four studies collected data from parents alone. The remaining studies

included views from participants from more than one participant group. The most common types of intervention were categorised by reviewers (DM, LS, MN) as: Online support interventions (n=12), Camps (n=9) and Music therapy (n=6). Table S1 provides detail regarding the focus of each study, method of analysis and intervention type for each study.

The most common type of LTC were; cancer (n=14), HIV (n=8) and a mix of LTCs (n=10). Interventions focused on improving aspects of emotional wellbeing, such as coping (n=26), self-esteem (n=13), or providing emotional support (n=11), although symptoms of mental ill-health were not routinely measured. Other intervention aims included improving physical symptoms relating to the LTC and social skills. Table S2 (See Supplementary Materials) outlines the structure and setting of each intervention.

Only 13 articles scored positively on at least 12 quality appraisal questions (See Table S3). Articles typically described their data collection method, had clear research questions, used an appropriate study design and had findings supported by data. However, the theoretical or ideological perspective of the author was not reported in 45 articles, thus its influence upon the study design, methods or research findings could not be determined.

### **Line of argument**

Five overarching constructs arose from the translation of first and second order construct data across all 60 articles: 'Getting In and Staying In', 'A Therapeutic Foundation', 'Social Support', 'A Hopeful Alternative' and 'Empowerment'. Table 2 provides examples of participant quotes supporting each construct.

Figure 2 conceptualises the line of argument. A 'Therapeutic Foundation' is essential to enable CYP and their families to achieve benefit from an intervention. 'A Therapeutic Foundation' may support CYP and their families to access and provide Social Support as conceptualised within construct 3, which can help CYP living with physical health conditions to feel less isolated and contribute towards feelings of safety, being valued and unconstrained as described within construct 2.

Being able to access 'A Therapeutic Foundation' and/or 'Social Support' can help CYP develop positive self-esteem and learn new or practise existing skills, which over time can contribute towards feelings of empowerment (construct 5). The interaction between experiencing 'A Therapeutic Foundation', receiving 'Social Support' and experiencing 'Empowerment' can also help create a sense of hope for CYP, generated through meeting peers who are living fulling lives alongside their LTC, taking part in everyday activities and contributing positively within their social relationships, as illustrated by 'A Hopeful Alternative'.

Whilst some of the concepts discussed within each construct may be more pertinent at certain stages of an intervention, the relationships between constructs, 2, 3 and 4 are bidirectional and self-reinforcing and promote continued engagement with the intervention.

The longer CYP and families engage with an intervention, the more they are likely to develop therapeutic relationships with intervention deliverers and/or other participants and experience the therapeutic benefits associated with the intervention. Thus, the therapeutic effect of these bidirectional relationships could be regarded as accumulative over time. 'Getting In and Staying In' describes factors that may influence the availability and accessibility of an intervention as well as the ongoing engagement of people taking part and discusses how the social relationships, increased sense of empowerment and hope developed through taking part in an intervention are key to prolonging its benefits beyond its end-point.

### **Construct 1: Getting in and staying in**

This construct conceptualises CYP's ongoing interaction with an intervention aimed at improving their mental health; from the initial recognition of their mental health need(s) and support from professionals, maintaining engagement over time and retaining the effects of the intervention after it has ended.

To make interventions available to those who need them, service providers should ensure they have adequate funding for equipment (e.g. Nicholas et al. 2007), staff expertise in intervention design and delivery (Whittemore et al. 2010) and space to stage an intervention (Muskat et al. 2016). Health and social care staff play a key role in recognising the mental health needs of CYP and supporting them and their families to access an interventions that is appropriate for their needs (e.g. Desai et al. 2014; Fair et al. 2012; Lewis et al. 2016). As discussed within ‘A Therapeutic Foundation’ below, CYP find it validating when their emotions and experiences are recognised by others. This suggests that contact with health and social care practitioners who can recognise the impact of a LTC on CYP’s mental health and offer some emotional support, may have a beneficial effect on CYP’s mental wellbeing before they even access a more formalised mental health intervention.

Recognising the social and family context of CYP was key to ensure they had the appropriate social support available to enable them to maintain engagement with an intervention over time. Parents highlighted the importance of flexibility in the delivery of interventions, such as considering the time of day (e.g. Brodeur, 2005) and setting (e.g. Muskat et al. 2016). Providing opportunities for CYP to maintain the peer relationships formed during an intervention via social media (e.g. Kirk & Milnes, 2016) or organised events (e.g. Desai et al. 2014; Lewis et al. 2016) may help maintain the beneficial effects of the social support received through an intervention (as discussed within the construct ‘Social Support’) beyond its scheduled end-point.

Consideration of the factors that may influence the availability and accessibility of interventions increases the likelihood that participants engage with an intervention long enough to access its therapeutic potential; a concept described within the construct ‘A Therapeutic Foundation’.

## **Construct 2: A therapeutic foundation**

This construct describes the emotional experience that results from the interaction between CYP feeling safe, unconstrained by their LTC and having the opportunity to develop ‘therapeutic’ relationships with intervention deliverers and/or fellow intervention recipients.

The presence of familiar people (e.g. Ayres et al.; Dennison et al. 2010) and ensuring that there are rules encouraging appropriate behaviour (e.g. Brodeur 2005; Desai et al. 2014) and protect the privacy and/or anonymity of CYPs (e.g. Nicholas et al. 2007; Stewart et al. 2013a) can support participants to feel secure within an intervention setting. This perception of safety can help CYP ‘escape’ the stigma and worry associated with their LTC experienced during their daily lives (e.g. Muskat et al. 2016). This feeling of being unconstrained can be facilitated by activities which distract CYP from the pain or boredom associated with long hospital stays (O’Callaghan et al 2013;Docherty et al 2013) or help them overcome physical limitations associated with their LTC (Gillard et al 2013). CYP with stigmatised diagnoses such as HIV (e.g. Gillard & Allsop. 2016), or altered appearances (e.g. Bluebond-Langer et al 1991; Tiemens et al. 2007) appear to particularly value this release from everyday worries or routine.

Feeling unconstrained within a safe space supports CYP to freely share their emotions and experiences (e.g. Fair et al. 2012). Given enough time, the unconstrained sharing of emotions and experiences can lead to the development of supportive ‘Therapeutic Relationships’ between CYP taking part in the intervention (e.g. Tiemens et al. 2007). These relationships seem to be characterised by CYP feeling understood, cared for and valued (e.g. Brodeur, 2005; Fair et al. 2012; Gillard et al. 2011). Together the concepts described above can create a therapeutic atmosphere, which we suggest could enhance the wellbeing of CYP with LTCs by itself. This ‘Therapeutic Foundation’ also creates the potential for those

receiving the intervention to engage with the social support offered by peers and experience feelings of hope and empowerment.

### **Construct 3: Social support**

This construct conceptualises ‘Social Support’ as the sense of belonging experienced by CYP as they meet other peers with a LTC, and the opportunity to learn new skills and improve relationships with healthy peers and family outside of the intervention setting.

Sharing experiences with intervention deliverers, peers and/or family within a safe environment allows CYP to receive reassurance that they are not alone living with the difficulties they are experiencing (e.g. Campbell et al. 2010; Gan et al. 2010). CYP described experiencing powerful feelings of belonging within interventions that allowed them to meet peers similar to themselves (e.g. Gillard & Allsop, 2016). This experience of belonging can further contribute to the therapeutic atmosphere described within ‘A Therapeutic Foundation’.

Meeting similar peers also helps CYP learn how to replicate the empathetic, understanding and validating relationships they valued as part of an intervention, and improve their relationships with others outside of the intervention setting (e.g. Bignall et al. 2015; Desai et al. 2014). This can be achieved directly by involving family with the intervention (e.g. Gan et al. 2010), or indirectly through CYP applying the skills they learned outside of the intervention setting (e.g. Hosek et al. 2012; Stewart et al 2011a/b). The type of social support most appreciated by CYP with LTCs may vary according to their individual preference, age, LTC and developmental level. The need for contact with similar peers may increase as CYP become more aware of their social surroundings and encounter the typical challenges associated with this developmental stage.

Over the course of the intervention, there seems to be a transition from building therapeutic relationships with peers and staff within an intervention setting, to replicating

these types of relationships within the CYPs everyday family and social systems. This process may be facilitated by the feelings of hope and empowerment generated through participation in an intervention, as detailed within ‘A Hopeful Alternative’ and ‘Empowerment’ below.

#### **Construct 4: A hopeful alternative**

● This construct describes how meeting peers and taking part in everyday activities can help CYP develop a sense of hope for the future and an identity not solely focused on their LTC.

CYP described how having the opportunity to meet other CYP who are currently managing the challenges of living with their LTC (e.g. Barnetz & Feigin, 2012; White, 2014) can result in a realisation that it is possible to live an enjoyable life despite the presence of a LTC. This may in turn motivate them to engage with others participating in the intervention and both teach and learn new skills to manage their physical and mental health.

CYP appear to value recognition that they are able to support others (e.g. Desai et al. 2014; Nicholas et al 2007; Tiemens et al. 2007) and not merely the passive recipients of help. This realisation may enable participant to offer support to others as depicted under ‘A Therapeutic Foundation’ and ‘Social Support’. Thus, taking part in an intervention can be experienced as a collaborative process, where everyone brings expertise.

Consideration of the family and social systems that CYP belong to within the intervention (as discussed within ‘Getting In and Staying In’) may support the recognition that the child is more than their physical illness. Integrating aspects of daily life into interventions, such as completion of everyday chores, hobbies and non-LTC orientated conversation with family members (e.g. Dennison et al. 2010; Nicholas et al. 2007), appeared to aid CYP’s realisation that they could be ‘normal’ despite their LTC. However, the needs of CYP who require end-of-life care may be different and should be met accordingly.

It appears having the opportunity to observe peers successfully managing life with a LTC; take part in everyday activities; and contribute meaningfully to their relationships with others, can help CYP visualise a hopeful future for themselves, which can have a positive influence their mental health.

### **Construct 5: Empowerment**

● The interlinked and self-reinforcing relationship between a young person's increased sense of their own ability to manage themselves and increased self-esteem are conceptualised here as "Empowerment".

Social support can help CYP acquire knowledge and skills regarding how to manage their physical, emotional and social difficulties (Stewart et al. 2013a; Marsac et al. 2012). As they master these skills, CYP develop a sense of confidence in their own abilities to manage their future physical (e.g. Barnett & Feigin, 2012) and mental (e.g. Jaser et al. 2014) health needs. There appears to be an interaction between feelings of increased hope and empowerment. Feeling empowered to manage their own wellbeing may provide CYP with increased hope for their future. In turn, the experience of successfully completing fun, challenging and/or normative activities not associated with their LTC (e.g. Moola et al. 2015) can lead to feelings of empowerment and increased self-esteem. We suggest there needs to be a balance between participating in activities that challenge them, with the need for these activities to be tailored to their developmental and health needs. This balance will ensure that CYP with LTCs can experience success, and feel empowered.

Feelings of empowerment, hope and being valued within an intervention's therapeutic relationships can have a positive impact on CYP's long-term mental health and their ability to access ongoing social support from family and peers both during an intervention and after it has ended.

## Discussion

Synthesis of first and second order construct data across the 60 articles in this review yielded five overarching constructs; 'Getting in and Staying In', 'Therapeutic Foundation', 'Social Support', 'A Hopeful Alternative' and 'Empowerment'. The line of argument presented in Figure 2 explores the relationship between these five constructs. It highlights how the social support received from service professionals, family members and peers was highly valued by CYP with LTCs. Social support can facilitate CYP's initial engagement with an intervention, the development of therapeutic relationships and sense of belonging, as well as facilitating the learning of skills. This is consistent with the social support model proposed by Stewart et al (2013a; b), which differentiates between affirmational, emotional and informational support. Gaysynsky et al (2015), propose the concept of 'esteem support' which encompasses "validation" and "compliment", ideas included within the constructs 'Therapeutic Foundation' and 'Hope and Inspiration' emerging from this review. Health and social care staff play a key role in identifying and signposting of CYP and their families towards age appropriate social support. Recommendations to increase the mental health support for young people in schools and community health services (Joint Commissioning Panel for Mental Health, 2013) may provide an opportunity for services who support CYP with LTCs to utilise a more holistic approach and encourage CYP to develop their social connections with peers.

The value of hope was a particularly striking finding of this review, complementing a review by Venning et al (2008), which highlighted how young people feel their chronic illness restricts their participation in "normal" life and the importance of developing a sense of hope. Our findings suggest the development of hope and acceptance can be facilitated by CYP with LTCs having the opportunity to meet peers with a LTC, improve relationships with healthy peers and incorporating fun or challenging activities unrelated to their LTC into their daily lives. This may then contribute towards building an identity not centred on their LTC.

### **Strengths and limitations**

This review was conducted according to best practice guidelines (CRD, 2009). The purposive sampling procedure described within the methods section meant each of the 60 articles included in this review contributed towards the final synthesis. Whilst the large number of studies and variety of interventions included in the review may have made it harder to report nuances within the data, each of the five overarching constructs was informed by a large number of articles, with at least ten of these receiving a “high” quality rating. We relied upon the self-report and reflexivity of the authors of primary studies to identify how their ideological approach influenced their results, which may have influenced our findings.

The constructs identified within this synthesis may represent core therapeutic components common across different types of intervention, however we cannot assume these are applicable across all interventions and all CYP with any LTC. A more focused review would be required for clinicians wishing to understand experiences of/attitudes towards a specific intervention or views in relation to specific psychiatric diagnoses. The absence of data regarding participants’ experiences of mental health difficulties means we do not know how the findings of this review can be applied to CYP with different mental health needs; from those with a diagnosed mental health disorder to those who may need support to maintain their mental wellbeing.

### **Recommendations for future research and practise**

This reviews findings review suggest that interventions to assist CYP with a LTC to improve their social support networks, develop a sense of hope and feel empowered may improve their mental health and wellbeing. The five constructs identified by this review offer a potential framework for health and social care professionals to consider when developing or refining existing interventions to improve and/or maintain the mental health of CYP with LTCs. In their linked-evidence synthesis, Moore et al. (in press) highlight the paucity of

research evaluating the effectiveness of the type of interventions included within this review, as only four of the included studies conducted their qualitative evaluation of an intervention as part of a randomised controlled trial (RCT) ([Barry et al. 2010](#); [Bignall et al. 2015](#); [Dennison et al. 2010](#); [Jaser et al. 2014](#)). Further RCTs are needed to evaluate whether interventions including the components/processes identified by this review are more effective at improving the mental health of CYP with LTCs than those which do not. These should be carried out alongside process evaluations to ensure the experience and practicalities of implementing these types of an intervention within a UK setting are fully understood.

### **Conclusion**

This review explored the attitudes and experiences of CYP, caregivers and practitioners towards interventions aiming to improve the mental health of CYP with LTCs. The synthesis elicited five key constructs that suggest important areas to consider during the development and delivery of mental health interventions. Future research should implement mixed methods approaches to evaluate such interventions in light of components identified by this research with CYP, in order to better understand what influences the success of interventions in CYP with different physical and mental health needs.

## Key messages

- The line of argument presented within this paper explores the relationship between the five constructs developed through the qualitative evidence synthesis: ‘Getting In and Staying In’, ‘A Therapeutic Foundation’, ‘Social Support’, ‘Hope and Inspiration’ and ‘Empowerment’.
- Social support from peers and the people delivering the intervention appear to play a key role in supporting CYP to access an intervention, gain new skills and develop an alternative, more hopeful view for the future.
- Ensuring that interventions are sustainable, accessible and relevant to CYP’s mental health needs can create the opportunity for CYP to engage with the therapeutic potential of an intervention.
- A ‘Therapeutic Foundation’ where CYP feel safe, valued and able to express themselves is a core component of mental health interventions.
- The interaction between feelings of increased hope, empowerment and improved social support may help sustain improvements to CYP’s mental health beyond the end of the intervention.
- Process evaluations should be incorporated alongside randomised controlled trials to explore which interventions are effective, and for whom.

Accepted

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Figure 1: PRISMA diagram

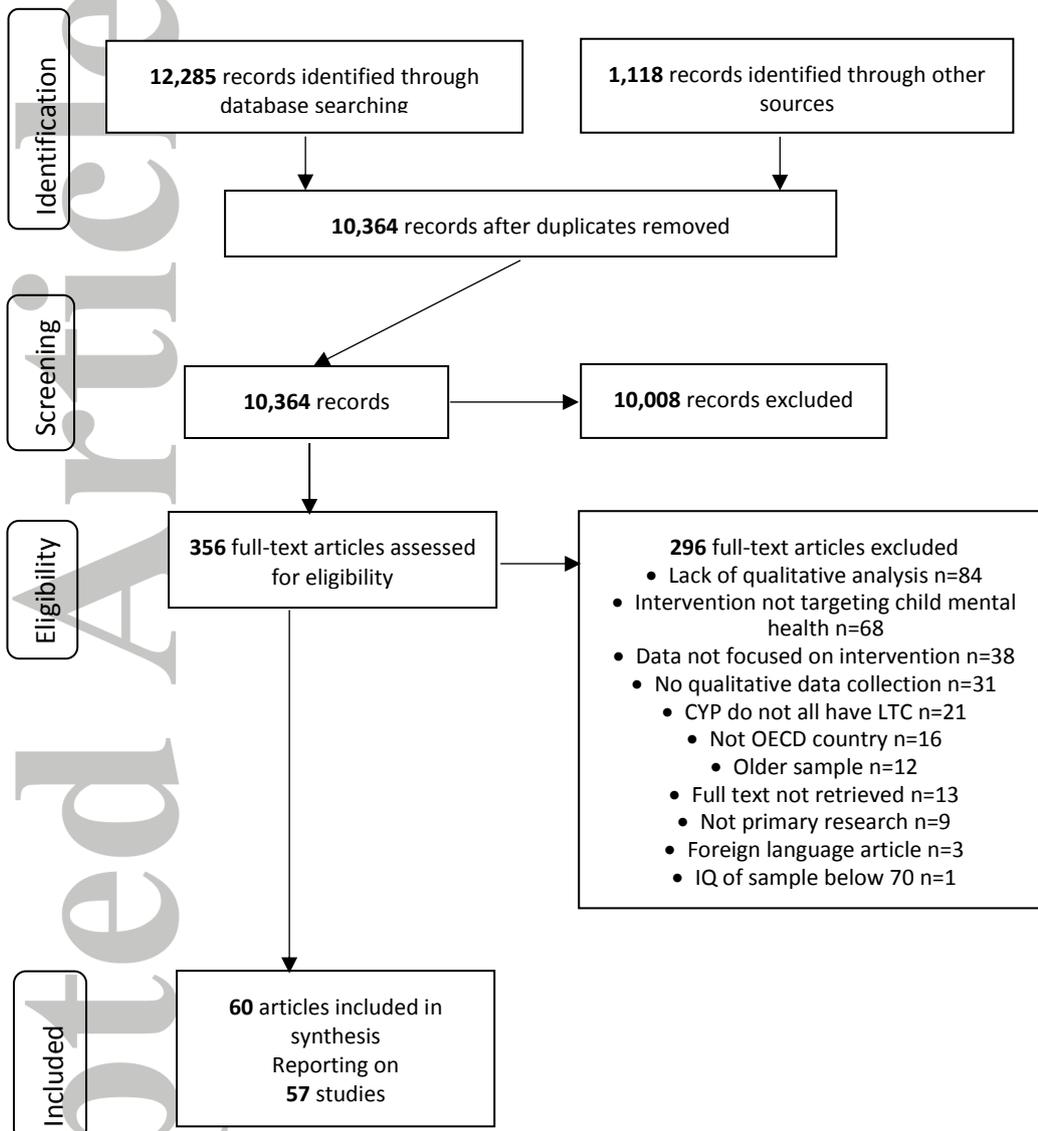
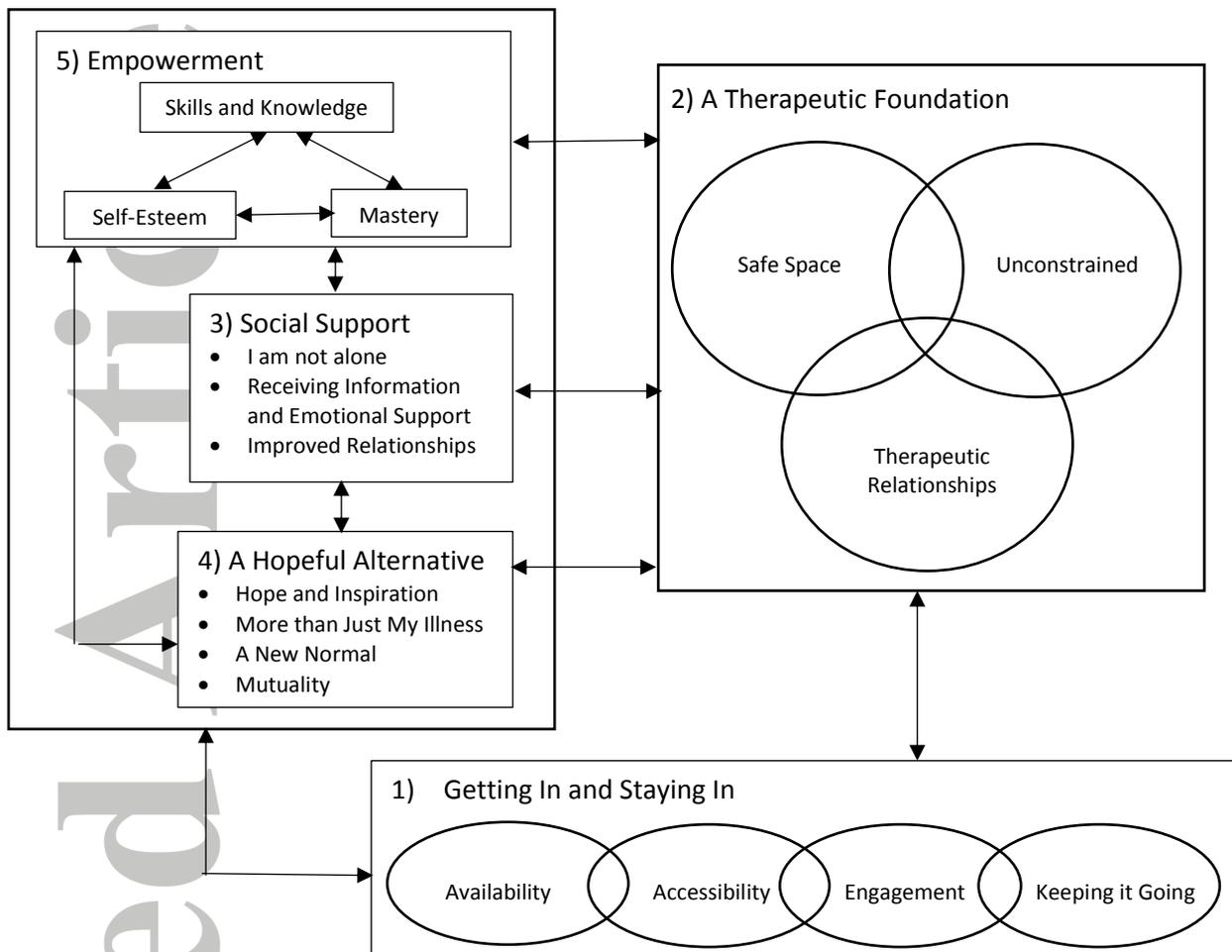


Figure 2: Line of argument



Accepted

Table 1: Inclusion and exclusion criteria

Criteria	Specification
<b>Population</b>	<p>Included if:</p> <ul style="list-style-type: none"> <li>○ CYP aged <math>\leq 25</math> years old with any LTC.</li> <li>○ The parents and families of CYP aged <math>\leq 25</math> years old with any LTC.</li> <li>○ Those involved in the delivery of interventions to improve mental health and wellbeing in CYP with LTCs.</li> </ul> <p>Excluded if:</p> <ul style="list-style-type: none"> <li>○ LTC was obesity.</li> <li>○ All participants had learning/intellectual disabilities (i.e. IQ &lt; 70).</li> </ul>
<b>Intervention</b>	<p>Included if:</p> <ul style="list-style-type: none"> <li>○ Intervention aimed to improve CYP's mental health and wellbeing.</li> <li>○ Intervention targeted CYP's mental health directly (i.e. CYP were recipients) or indirectly (e.g. parenting interventions).</li> <li>○ Attitudes towards an intervention in development or interventions they chose not to receive.</li> </ul> <p>Exclude if:</p> <ul style="list-style-type: none"> <li>○ Focus on mental health <i>service provision</i>, rather than specific interventions.</li> </ul>
<b>Outcomes</b>	<p>Include:</p> <ul style="list-style-type: none"> <li>○ Attitudes, experiences, perceptions and understanding of CYP with LTCs, their parents or the practitioners who have delivered such interventions regarding interventions focused on the mental health/wellbeing of CYP with LTCs.</li> </ul>
<b>Study design</b>	<p>Include if:</p> <ul style="list-style-type: none"> <li>○ Qualitative data collection e.g. interviews, focus groups.</li> </ul>

- 
- Qualitative data analysis e.g. thematic analysis, framework analysis, constant comparative method.
  - May be stand-alone qualitative research, or reported as part of a mixed methods intervention evaluation or process evaluation.

Exclude if:

- Qualitative data only provided through open-ended questionnaire items.

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**Country and language**

Include if:

- Full text is in English
  - OECD setting
-

Table 2: Constructs contributing to line-of-argument

Overarching Construct	Second Order Construct Data (Author interpretation extracted from articles included in review)	Supporting Quotes
Getting In and Staying In	<p>Clearly, greater availability of a range of educational materials and group or individual interventions combined with ease of access will be of paramount importance in the future (Barlow, 1999)</p> <p>The habitat of fun consisted of abundance and opportunities for transgressions, which were grounded in an unceasing focus on campers' enjoyment and engagement. (Gillard et al. 2011)</p> <p>Again, some participants would have changed the meeting time to make it more convenient for families. (Brodeur, 2005)</p>	<p>“They said they only had one social worker for the outpatient department and it was all very difficult for them to see him.” [Parent, Mixed LTCs] (Lewis et al. 2016, p 2546)</p> <p>“I think [this camp is] unique because you're in a setting with so many kids, and you're in a setting where you're scheduled to have fun constantly and you don't have as much down time as at home, so it creates a habitat of fun and constantly going that you don't get anywhere else.” [Camp counsellor, LTC] (Gillard et al. 2011 p)</p> <p>“The only thing I would change, and that’s from having older kids and mine were older than most of the others, would have been a weekend or a Friday night because of the homework and getting downtown and coming back. That was pretty much a 2-1/2</p>

hour, 3-hour venture by the time we left and came back. Now that I'm working, it would be almost impossible." [Parent, Mixed LTCs] (Brodeur, 2005 p242)

**Therapeutic  
Foundation**

Children and parents said how important it was that parents support their child during needle-procedures. They provided children with familiarity, reassurance, security, and practical support. {Ayers, 2011 p}

If I actually knew the nurse it would be okay, but if I didn't I would be like 'what's going on? I don't even know you!' [CYP, Cystic Fibrosis] (Ayers, 2011 p. 337)

"And I personally wouldn't have coped very well if they had just invited me to go to somewhere. I needed my mum there." [CYP, CFS]. (Dennison et al. 2010 p175)

Feeling like there are others who share experiences, who talk about topics that cannot be discussed elsewhere and where one feels that they are just like everyone, are important for all children and teens. Feeling "normal" is even more important when one lives

"My family, not really, and outside like friends no, cause you don't really know who to trust. And even in my family it's awkward to talk about but here I know I can talk about it with these people and that's really good. I like feeling that I can talk to someone,

with a condition that is often kept secret due to associated shame and stigma. (Muskat et al. 2016)

it's really good, yeah ... Here you feel like it's not taboo, you know" [CYP, HIV] (Muskat et al. 2012 p8)

Campers expressed feeling a sense of love, respect, happiness, and caring throughout their time at camp. (Gillard et al. 2016)

"I would take from camp the vibe that I get – the vibe of caring and respect and love that is just emanating through everything, through every activity in the cabins and everywhere." [CYP, Mixed LTCs] Gillard et al. 2016 p116)

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**Social Support**

Several participants talked about how they felt the program had brought their family closer together. (Brodeur, 2005)

"We realized the necessity just to set aside time. How well we do it is a different story. But realized the necessity to sort of just set everything aside and just be together and play Monopoly or whatever." [Parent, Mixed LTCs] (Brodeur, 2005 p. 170)

Loneliness and social dissatisfaction decreased. (Stewart et al. 2013)

"It was nice for her to see that other people were in her shoes, that what I think was the best part, to see that other people are dealing with the same things she was dealing with. You don't feel like you're alone." [Parent, Allergies/Asthma] (Stewart et al. 2013a p181)

The mentors told them about their troubles and fears (at group meetings too) and the mentees were amazed to see diabetics, who had the same fears as their own, managing to function well and share their feelings, difficulties and successes. This experience provided a sense of relief, optimism and hope (Bartnetz et al. 2012)

“You can share deep experiences and fears that other people won’t understand or don’t know how to calm.” [CYP, T1D] (Bartnetz et al. 2012 p475)

“Helped us to discuss concerns and feelings that otherwise might not surface.” [Parent, ABI] (Gan et al. 2010 p659)

**A Hopeful  
Alternative**

The mentees admired their mentors. Numerous mentees reported adopting and emulating parts of their mentors’ behaviour regarding diabetes. {Bartnetz, 2012 p}

It was fun like it’s never been before, I saw someone who is in control of the situation and isn’t afraid of diabetes. If he can do it, I’ve got to succeed...I’m less afraid because of the project; I met people there with amazing abilities. (CYP quote: {Bartnetz, 2012 p. 474})

When asked what young HIV-positive women need to make healthier life choices and decrease risky behaviors, participants emphasized the need for comprehensive programs that extended beyond HIV specific topics. They requested programs that address a

“My perfect program wouldn’t just not only be focused on the infection. It would just really be building self-worth, building self-esteem like all the way around. So many youth have a hard time just making that transition perhaps to the college, and still be like

wide range of issues impacting their lives such as self-esteem, self-confidence, self-worth, living with HIV, sexuality, coping mechanisms, handling adversity, and developing and maintaining healthy relationships. (Hosek et al. 2012)

During the program, all participants noted that they had increased their overall daily physical activity, incorporated planned physical activities into their daily schedules (Table 7), and experienced increased motivation to go out or be with their friends even if they had pain. (Kashikar-Zuck et al. 2016)

They also saw themselves as able to give information to help other children. (Bluebond Langer et al. 1992)

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**Empowerment**

Several participants also discussed the benefits of skills-based education and learning strategies to manage diabetes in public. (Serlachius et al. 2012)

do they have to take care of themselves, either by nutrition..."[CYP, HIV] (Hosek et al. 2012 p293)

"I used activity pacing didn't push myself too far and I was still able to stay with my friends and do what they were doing." [CYP, LTC] (Kashikar-Zuck et al. 2016 p74)

"Well, Kim is on the kind of therapy now that just finished, so now she can come to me and ask me what it's like, and I can tell her." [CYP, Cancer] (Bluebond-Langer et al. 1992 p75)

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"Like strategies, or ways to deal with the public thing. Like developing a skill where you stop worrying about what strangers would think, for

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example at the footy.” [CYP, T1D]

(Serlachius et al. 2012 p319)

As part of the rigorous program within the camp, participants realized success in mastering difficult tasks. (Tiemans et al. 2007)

“I was like, ‘wow’ you know, hard things that you overcome, you are, kind of feel that you are such a good person.” [CYP, CFD] (Tiemans et al. 2007 p65)

Initial uncertainty or reservation was replaced by a sense of pride, competence, and mastery. (Burns et al. 2010)

“So it was that short little brief of ‘I am in control...this is my project’ was really, really good for him.” [Parent, Cancer] (Burns et al. 2010 e24)

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ABI = Acquired Brain Injury, CF = Cystic Fibrosis, CFD = Craniofacial Differences, CFS = Chronic Fatigue Syndrome, CHD = Congenital Heart Disease, CP = Cerebral Palsy, T1D = Type 1 Diabetes,

## Appendix A: Example search strategy within the PsycINFO (via OvidSp) database

#	Searches	Results
1	Chronic Disease/	0
2	Neoplasms/	28712
3	Diabetes Mellitus/	4470
4	Asthma/	4034
5	exp Respiratory Tract Diseases/	0
6	Cystic Fibrosis/	732
7	Cerebral Palsy/	4202
8	exp Epilepsy/	22725
9	exp Muscular Diseases/	0
10	exp Endocrine System Diseases/	0
11	exp Immune System Diseases/	0
12	exp HIV Infections/	0
13	exp Cardiovascular Diseases/	0
14	exp Nervous System Diseases/	0
15	exp Skin Diseases/	0
16	exp Digestive System Diseases/	0
17	exp Hematologic Diseases/	0
18	exp Otorhinolaryngologic Diseases/	0
19	exp Stomatognathic Diseases/	0
20	exp Eye Diseases/	0
21	exp Pain/	47653
22	Disabled Children/	0
23	((chronic* or longterm or long-term or "long standing" or physical) adj2 (condition* or ill* or disease* or disorder* or syndrome*)).ti,ab.	45152
24	(cancer* or neoplas* or tumor* or tumour* or malignan* or carcinoma* or "bone marrow" or leukaemia or leukemia).ti,ab.	63603
25	diabet*.ti,ab.	23493
26	asthma*.ti,ab.	6465
27	(respiratory adj2 (illness* or disease* or condition*)).ti,ab.	1201

28	cystic fibrosis.ti,ab.	993
29	cerebral palsy.ti,ab.	5460
30	quadriplegi*.ti,ab.	491
31	tetraplegi*.ti,ab.	291
32	diplegi*.ti,ab.	378
33	spinal cord injur*.ti,ab.	4595
34	muscular dystrophy.ti,ab.	1123
35	epilep*.ti,ab.	33297
36	seizure*.ti,ab.	25791
37	spina bifida.ti,ab.	868
38	(heart adj2 (condition* or disease* or disorder* or defect*)).ti,ab.	9188
39	(cardiac adj2 (condition* or disease* or disorder* or defect*)).ti,ab.	1390
40	(cardiovascular adj2 (condition* or disease* or disorder* or defect*)).ti,ab.	8891
41	(skin adj2 (condition* or disease* or disorder*)).ti,ab.	966
42	eczema.ti,ab.	330
43	(allergies or allergy).ti,ab.	1529
44	dermatitis.ti,ab.	532
45	(gastrointestinal adj (disorder* or disease*)).ti,ab.	669
46	((stomach or abdominal or gastrointestinal) adj pain).ti,ab.	1342
47	(bowel* adj2 inflammatory adj2 (condition* or disease* or illness*)).ti,ab.	602
48	(liver adj (disease* or transplant*)).ti,ab.	1621
49	hepatitis.ti,ab.	3515
50	(disabilit* adj5 child*).ti,ab.	13184
51	(human immunodeficiency virus or HIV).ti,ab.	42006
52	AIDS.ti,ab.	30925
53	(hyperthyroidism or hypothyroidism).ti,ab.	1635
54	an?emia.ti,ab.	1617
55	h?emophilia.ti,ab.	368
56	sickle.ti,ab.	1162
57	((renal or kidney) adj (disease* or disorder*)).ti,ab.	1900
58	nephrotic syndrome.ti,ab.	55

59	encephalomyelitis.ti,ab.	1604
60	chronic fatigue syndrome.ti,ab.	1946
61	((cleft or palate) adj lip).ti,ab.	295
62	craniofacial.ti,ab.	527
63	(deaf or deafness).ti,ab.	13257
64	(hearing adj (defect* or disorder*)).ti,ab.	347
65	blindness.ti,ab.	5420
66	((vision or visually or visual) adj (impaired or impairment*)).ti,ab.	4261
67	((persistent or chronic or recurring or frequent) adj (headache* or migraine*)).ti,ab.	1720
68	chronic pain.ti,ab.	11547
69	fibromyalgia.ti,ab.	2541
70	medically unexplained symptoms.ti,ab.	452
71	(spinal adj injur*).ti,ab.	301
72	or/1-71	335060
73	(child or children*).ti,ab.	529036
74	(adolescent or adolescents).ti,ab.	169875
75	teen*.ti,ab.	18103
76	(young adj (adult* or people)).ti,ab.	53922
77	youth*.ti,ab.	73029
78	73 or 74 or 75 or 76 or 77	701901
79	Mental Health/	47847
80	(psychological adj (illness* or disorder* or difficulties or problems or distress)).ti,ab.	23105
81	(mental adj (health or illness* or disorder* or distress or problem*)).ti,ab.	186832
82	(depression or depressed or depressive).ti,ab.	235411
83	((disruptive or challenging or antisocial) adj behavior?).ti,ab.	11412
84	(behavior?r adj problem*).ti,ab.	12771
85	(anxiety or anxious).ti,ab.	157623
86	feelings.ti,ab.	57562
87	Internaliz*.ti,ab.	3
88	(wellbeing or well being).ti,ab.	61891
89	happiness.ti,ab.	11767

90	worry.ti,ab.	7030
91	distress.ti,ab.	48777
92	satisfaction.ti,ab.	82607
93	emotional.ti,ab.	174569
94	coping.ti,ab.	61721
95	or/79-94	828319
96	qualitative research/	6724
97	Ethnology/	1943
98	exp Questionnaires/	15996
99	phenomenology/	11126
100	Attitudes/	23102
101	interviewing/	3181
102	interview*.ti,ab.	250183
103	qualitative.ti,ab.	110757
104	(talked or asked).ti,ab.	81112
105	focus group*.ti,ab.	24246
106	ethnograph*.ti,ab.	20873
107	grounded theory.ti,ab.	11222
108	thematic.ti,ab.	14905
109	(barriers and (facilitators or enablers)).ti,ab.	2521
110	process evaluation.ti,ab.	1094
111	group discussion*.ti,ab.	7342
112	perception*.ti,ab.	232038
113	attitude*.ti,ab.	171590
114	views.ti,ab.	56281
115	experience*.ti,ab.	486970
116	or/96-115	1107429
117	intervention.ti,ab.	171181
118	psychotherapy.ti,ab.	77346
119	(support adj3 (group* or network)).ti,ab.	13125
120	therapy.ti,ab.	194016

121 counselling.ti,ab.	9299
122 peer support.ti,ab.	2673
123 social support.ti,ab.	35639
124 program*.ti,ab.	318905
125 (mental adj3 service*).ti,ab.	22848
126 training.ti,ab.	210131
127 technique*.ti,ab.	158051
128 treatment*.ti,ab.	508051
129 or/117-128	1226579
130 72 and 78 and 95 and 116 and 129	4011

Accepted Article

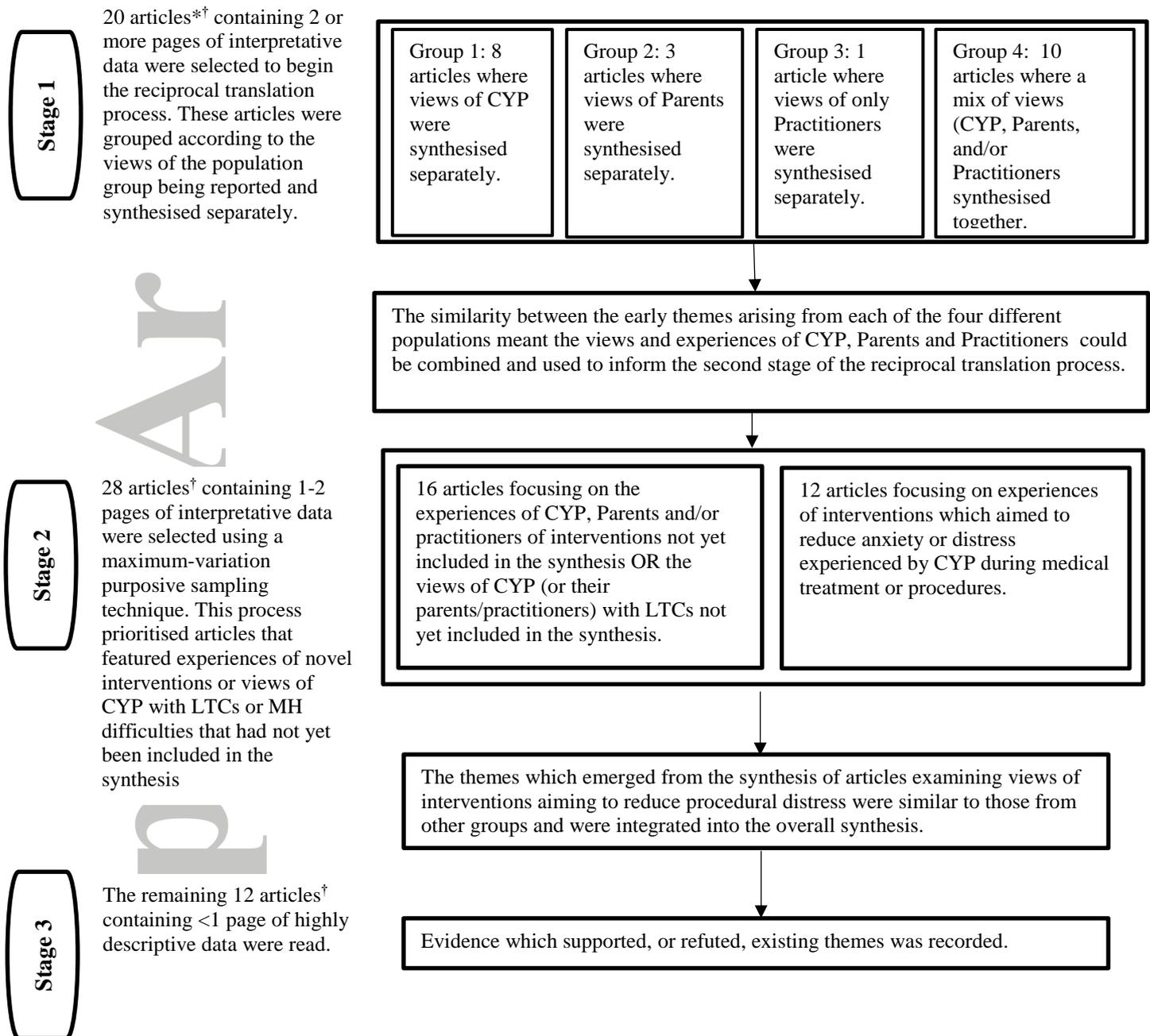
## Appendix B: List of websites searched

Mental Health Foundation <https://www.mentalhealth.org.uk/>  
Young Minds <http://www.youngminds.org.uk/>  
Department of Child and Adolescent Mental Health Service (CAMHS) at Great Ormond Street Hospital <http://www.gosh.nhs.uk/medical-information/clinical-specialties/child-and-adolescent-mental-health-services-camhs-information-parents-and-visitors/research-and-publications>  
Child & Adolescent Mental Health Services (CAMHS) Research Unit <http://www.cpcs.org.uk/index.php?page=high-needs-project>  
NIHR CRN: Children and young people's mental health research <https://www.crn.nihr.ac.uk/mentalhealth/about-mental-health-research/children-and-young-peoples-mental-health-research/>  
NIMH Child and Adolescent Mental Health [http://www.nimh.nih.gov/health/topics/child-and-adolescent-mental-health/index.shtml#part\\_152583](http://www.nimh.nih.gov/health/topics/child-and-adolescent-mental-health/index.shtml#part_152583)  
RCP CAMHS Resource Library <http://www.rcpsych.ac.uk/quality/qualityandaccreditation/childandadolescent/communitycamhsqnce/amhsresource/library.aspx>  
National Child and Maternal Health Intelligence Network <http://www.chimat.org.uk/camhs>  
National Children's Bureau <http://www.ncb.org.uk/healthy-care/useful-links/mental-health-and-emotional-well-being>  
Association for Children's Mental Health <http://www.acmh-mi.org/get-information/>  
Substance Abuse and Mental Health Services Administration <http://www.samhsa.gov/data/>  
American Academy of Pediatrics <https://www.aap.org/en-us/professional-resources/Research/Pages/Research.aspx>  
Child and Youth Health <http://www.cyh.com/HealthTopics/HealthTopicDetails.aspx>  
The Kings Fund: Mental Health <http://www.kingsfund.org.uk/topics/mental-health>  
Young Epilepsy: <http://www.youngepilepsy.org.uk/>  
Epilepsy Society: [https://www.epilepsysociety.org.uk/young-people-and-epilepsy#.Vz8FjuTAN\\_A](https://www.epilepsysociety.org.uk/young-people-and-epilepsy#.Vz8FjuTAN_A)  
Epilepsy Action: <https://www.epilepsy.org.uk/info/young-people>  
Cancer Research UK: <http://www.cancerresearchuk.org/health-professional/cancer-statistics/teenagers-and-young-adults-cancers>  
Macmillan: <http://www.macmillan.org.uk/cancerinformation/teensandyoungadults/infoforteensandyoungadults.aspx>  
Teenage Cancer Trust: <https://www.teenagecancertrust.org/get-help/ive-got-cancer/stories-others>  
Teenagers and Young Adults with Cancer: <http://www.tyac.org.uk/>  
Sickle Cell and young stroke survivors: <http://www.scyss.org/>  
Sickle Cell Society: <http://sicklecellsociety.org/resources/did-you-know-age-11-16-an-information-booklet-for-young-people/>  
AVERT (HIV): <http://www.avert.org/living-with-hiv/health-wellbeing/being-young-positive>  
Young people with Aids: <http://www.amfar.org/about-hiv-and-aids/young-people-and-hiv/young-people-and-hiv-aids/>  
Pozitude: <http://www.pozitude.co.uk/>  
Children and Young People HIV network: <http://www.ncb.org.uk/hiv>  
Child's Brain Injury Trust: <http://childbraininjurytrust.org.uk/how-we-help/young-people/>  
Stroke Association: <https://www.stroke.org.uk/finding-support/information-about-stroke-childhood>  
National Institute of Neurological Disorders and Stroke: [http://www.ninds.nih.gov/disorders/tbi/detail\\_tbi.htm](http://www.ninds.nih.gov/disorders/tbi/detail_tbi.htm)  
Cerbera: <http://w3.cerebra.org.uk/>  
Association of young people with M.E.: <http://www.ayme.org.uk/>  
Away with Pain: <http://www.awaywithpain.co.uk/young-people>  
Fibromyalgia: <http://www.fmauk.org/information-packs-mainmenu-58/booklet-mainmenu-135/232-children-and-young-people>  
<http://www.fmscommunity.org/pediatric.htm>

Healing Well: <http://www.healingwell.com/community/default.aspx?f=24&m=1813258>  
Arthritis research UK: <http://www.arthritisresearchuk.org/arthritis-information/young-people.aspx>  
Youth Health: <http://www.healthtalk.org/young-peoples-experiences/arthritis/what-arthritis-and-what-are-early-symptoms>  
Arthritis Care: <https://www.arthritiscare.org.uk/managing-arthritis/children-with-arthritis>  
CLIC Sargent <http://www.clicsargent.org.uk/content/help-and-support>  
Cerebral Palsy <http://www.cerebralpalsy.org/information/child-care>  
Asthma UK <https://www.asthma.org.uk/research/>  
British Lung Foundation <https://www.blf.org.uk/what-we-do/research>  
Diabetes UK <https://www.diabetes.org.uk/Research/>  
British Deaf Association <https://www.bda.org.uk/publications>  
Mental Elf <http://www.nationalelfservice.net/>

Accepted Article

## Appendix C: Description of how articles meeting the inclusion criteria for the review were entered into the reciprocal translation process



CYP=Children and young people, MH=Mental health,

\*Articles which reported views of multiple population groups, but synthesized the views of CYP, Parent and/or practitioner views separately, were included in multiple groups within this synthesis. Thus, total number of articles included across different groups exceeds 20. †See Table S1 for details regarding at which stage each included article entered the reciprocal translation process.