

**Review title**

Improving the mental health of children and young people with long term conditions: Linked evidence syntheses

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**Background**

Rates of psychiatric disorder are up to four times greater in children with chronic, or long term, physical illness than in children who are physically well. Not only do psychiatric symptoms have considerable consequences for a child's quality of life, their behavioural, emotional, educational and social functioning, but mental health symptoms can have a larger impact on quality of life than the severity of the physical illness itself and mental ill health has been shown to impact upon management and medical consequences of the physical illness. Having a mental health problem may be associated with poor treatment adherence, which may exacerbate the physical condition, impair self-management and worsens long term outcomes.

Children with mental and physical illness need effective integrated healthcare, yet it has not been established whether, or which, interventions for mental health disorders are effective in children with chronic physical illness.

**Research Objectives**

The objectives are to evaluate the effectiveness of interventions aimed at improving the mental health of children and young people with long term physical health conditions, to explore the factors that may enhance, or limit, the beneficial delivery of such interventions and to assess whether treatments are good value for money.

The project will consist of two systematic reviews. We will undertake a systematic review of quantitative research evidence about the effectiveness and cost-effectiveness of relevant interventions and a review of qualitative research that will explore the attitudes to, and experiences of, receiving interventions, from the perspectives of the children and young people, their parents and families and those involved in delivering interventions. We will then bring together the findings from the two reviews to map out possible links between the treatments and their effects on mental health, gaps in the evidence and factors that seem to enhance or limit the success of interventions. A conceptual framework developed during the preparation of this protocol, will be used to facilitate the synthesis of the strands of evidence and will be further refined and developed as a result of this process.

### **Review question(s)**

- 1) What is the effectiveness and cost-effectiveness of interventions targeting mental health for children and young people with long term conditions and symptoms of mental distress?
- 2) What are the effects of such interventions on other key aspects of social and family functioning?
- 3) What are the factors that may enhance, or hinder, the effectiveness of interventions and / or the successful implementation of interventions intended to improve mental health for children and young people with long term conditions and symptoms of mental distress?

### **Searches**

The search methods will include extensive database searching and supplementary searching including forwards and backwards citation chasing, hand-searching of any key journals identified during the search process and additional searching on topic specific websites (if applicable).

A search strategy will be developed and tested in our suggested portfolio of resources. The strategy will use both controlled headings (e.g. MeSH) and free-text searching. Terms will be grouped according to three concepts:

- mental health terms
- chronic conditions terms
- study design terms (to describe a) randomised controlled trials and b) qualitative research)

Long term condition terms will be informed by a Cochrane review on interventions for parents of children and adolescents with chronic illness. Terms for mental health and chronic illness will be combined and searched together with a) terms to describe randomised controlled trials and b) terms for qualitative research (e.g. 'interviews', 'experiences' etc.). The search will therefore produce two separate bodies of literature one covering relevant quantitative research (to inform Review One) and the other covering relevant qualitative research (to inform Review Two).

Based on our scoping searches, we propose to search the following databases:

- MEDLINE including MEDLINE in-process (via OvidSp)
- EMBASE (via OvidSp)
- PsycINFO (via OvidSp)
- Cochrane Database of Systematic Reviews (via the Cochrane Library)
- CENTRAL (via the Cochrane Library)

- DARE (via the Cochrane Library)
- HTA database (via the Cochrane Library)
- NHS EED (via the Cochrane Library)
- CINAHL (via EBSCOhost)
- British Nursing Index (via ProQuest)
- HMIC (via OvidSp)
- Conference Proceedings Citation Index (via Web of Science)
- Science Citation Index (via Web of Science)

Supplementary searches: The citation lists of included references will be checked and forwards citation chasing (identifying where included references have been cited) will be carried out using Web of Science and Google Scholar. Any journals that are identified as being particularly pertinent in the field will be hand-searched. Targeted searches to identify “sibling” studies (process evaluations, economic studies and qualitative research) associated with included trials and based on trial names and first and last authors will also be conducted.

Grey literature: The databases CINAHL and HMIC will be searched, both of which index grey literature. We will also search Conference Proceedings Citation Index for abstracts that have only been presented at conferences. In addition we will search the website OpenGrey via <http://www.opengrey.eu/>

No language or date restrictions will be applied.

All references identified by the searches will be exported into EndNote X7 prior to de-duplication and screening.

The searches will be recorded using PRISMA guidelines. This will include the list of databases searched, recording of the date searched and the strategies used for each database.

### **Participants/population**

For the effectiveness review:

Children and young people (aged 0-25 years) with diagnosed *long term physical health conditions* and symptoms of poor mental health, emotional distress or behavioural problems.

*Long term physical health conditions* are defined as ‘physical health conditions that cannot be cured but are managed by medication and other therapies’ and will include chronic illness such as type I diabetes and asthma and also physical disability such as cerebral palsy and muscular dystrophy.

Studies of children with obesity will be excluded due to recently published evidence syntheses in this area.

Studies in children with moderate or severe learning/intellectual disabilities (moderate or severe mental retardation in ICD10 i.e. IQ < 50) will be excluded since the types of psychological difficulties and therefore interventions used in this population are likely to be sufficiently different to make extrapolation between studies difficult.

Children and young people may have a diagnosis of a mental health disorder (assessed by a validated measure and standardised measure) or may be at risk of such a diagnosis and experiencing symptoms of mental distress (defined as scoring above an established cut point on a validated

questionnaire). To be included in this review, a study will need to report findings for individuals with long term conditions experiencing symptoms of mental distress at baseline assessed using validated diagnostic or dimensional measures.

For the qualitative review:

- i) Children and young people (aged 0-25 years) receiving interventions to support mental health,
- ii) Their parents and families,
- iii) Those involved in the delivery of interventions to improve mental health in this patient group.

### **Intervention(s)**

Any intervention, delivered to children and young people, targeting improvement in mental health. Although our scoping of the literature and clinical experience suggests that many of the interventions will be psychological, we will include all other interventions that aim to improve mental health and anticipate that these will include pharmacological interventions and those that utilise a combination of psychological and pharmacological approaches.

Interventions delivered to parents and children or to families will be included if they report outcomes relating to the mental health of the children and young people. Studies of parent/child or family interventions in which only parent-focussed outcomes are reported will be excluded.

### **Comparator(s)/control**

Any control or comparator.

### **Types of study to be included**

For the effectiveness review only randomised controlled trials will be included.

For the assessment of cost effectiveness, we will include economic analyses and comparative cost studies of interventions meeting the inclusion criteria.

For the qualitative review any recognised method of qualitative data collection and qualitative data analysis will be included. Methods include interviews, focus groups and observational techniques and analysis includes thematic analysis, framework analysis and constant comparative method. This may be stand-alone qualitative research, or reported as part of a mixed methods intervention evaluation. We will include process and outcome evaluations.

### **Context**

For the qualitative review, only studies from OECD countries will be included. Consideration will be given to the degree of transferability of findings from non-UK settings to the NHS context.

### **Primary outcome(s)**

For the effectiveness review:

Any outcome describing the mental health of children and young people e.g. symptom of depression, anxiety, emotional distress or behavioural disorders, psychological health, psychological function, suicidal behaviour, health related quality of life, sleep quality and incidence of self-harm.

'Economic outcomes' will be collected from any study (whether ostensibly an effectiveness randomised controlled trial (RCT) or an economic evaluation) that reports the costs or resource implications for the included interventions and comparators. For example, frequency of service use or numbers of referrals will be included as economic outcomes, and better support an integrated assessment of effectiveness and cost-effectiveness.

For research question 3, we will include data from process evaluations conducted alongside included RCTs.

For the qualitative review:

- i) Attitudes, experiences, perceptions and understanding of children and young people with long term conditions and mental ill health, who have experienced interventions for their mental health.
- ii) Attitudes, experiences, perceptions and understanding of parents whose children have long term conditions and have experienced interventions for those with mental ill health.
- iii) Attitudes, experiences, perceptions and understandings of practitioners who have delivered interventions for children and young people with long term conditions and mental ill health.

### **Secondary outcomes**

For the effectiveness review, outcomes describing attendance at school, adherence with treatment for the primary health condition, costs and resource use will also be included where reported.

### **Study selection**

Inclusion and exclusion criteria will be applied to the title and abstract of each identified citation independently by two reviewers with disagreements being settled by discussion with a third. The full text will be obtained for papers that appear to meet the criteria and those for which a decision is not possible based on the information contained within the title and abstract alone. The full text of each paper will be assessed independently for inclusion by two reviewers. Extent of agreement will be measured using the kappa statistic and disagreements settled by discussion with a third reviewer. A PRISMA-style flowchart will be produced to detail the study selection process and reasons for exclusion of each full-text paper will be reported.

### **Data extraction**

For the effectiveness review:

A standardised, piloted data extraction form will be used to collect data from each included paper. Data extraction will be performed by one reviewer and checked by a second, with disagreements being settled through discussion with a third.

For the qualitative review:

Details of the studies' methods and findings will be extracted into a pre-designed and piloted data extraction form. The extraction of data will be conducted by one reviewer and checked by a second with discussion where necessary. Depending on the number of papers identified, we may upload included papers into NVIVO for coding to facilitate analysis and synthesis.

**Risk of bias (quality) assessment**

For the effectiveness review:

We will use the Cochrane Risk of Bias tool to critically appraise all included papers that assess the effectiveness of interventions. Cost effectiveness papers will be critically assessed using accepted frameworks, such as the consensus-developed list of criteria developed by Evers and colleagues and those produced by Drummond and colleagues. Quality assessment will be performed independently by two reviewers, with recourse to a third in case of disagreement. Where insufficient detail is provided in the published paper to adequately assess the risk of bias, authors will be contacted and asked to provide additional information.

For the qualitative review:

We will use the Wallace checklist for quality assessment. The checklist will be supplemented by critical reading of each study. The quality of studies will be independently quality assessed by two reviewers. Any disagreement will be resolved by consensus and if necessary a third reviewer will be consulted. We also anticipate, however, that the value of each study will be judged through its contribution to the synthesis.

**Strategy for data synthesis**

For the effectiveness review:

Data will be tabulated and discussed narratively in the first instance. Data tables for the effectiveness studies will include details of the intervention type and content, the setting and the provider, sample characteristics of the included population and the type of outcomes measured. Studies will be grouped by comparator, by intervention and/or by co-morbidity if appropriate.

The methods and findings from included economic evaluations will be summarised in a tabular format, noting the type of evaluation carried out, the setting and perspective. Findings will be synthesised in a narrative review (i.e. we will not quantitatively synthesise ICERs or other summary measures of economic evaluation) which will pay particular regard to issues relating to generalisability of findings to the UK.

For the effectiveness studies, if the data allow, meta-analysis will be used to estimate summary measures of effect on relevant outcomes, based on data from intention to treat analyses in contributing studies. If data allow, we will explore the impact of study quality factors (e.g. control for potential confounding factors) using meta-regression. Heterogeneity will be explored through consideration of the study populations, methods and interventions by visualisation of results and, in statistical terms, by the chi-squared ( $\chi^2$ ) test for heterogeneity and I-squared (I<sup>2</sup>) statistic and, where possible, using meta-regression.

For the qualitative review:

Precise methods of synthesis will be determined in response to the nature of the findings in the identified studies. Preliminary analysis will involve reading and re-reading the findings of included papers, in order to consolidate understandings of the themes and concepts and their relations within and between studies. A structured summary for each paper will also be produced which will aid discussion of the emerging synthesis amongst the review team.

Assuming sufficient conceptual data is available, we will undertake a meta-ethnography. The aim of meta-ethnography is to identify where similar themes and concepts from different papers refer to the same concepts (congruent synthesis) or identify opposing findings (refutational synthesis), this process is referred to as 'translation'. Study concepts may also be linked to create a 'line of argument', developing ideas across more than one study.

If findings are more descriptive, we will conduct a thematic synthesis. Where the evidence base consists of a mixture of more and less conceptual analyses, it may be necessary to thematically analyse the more descriptive papers first, before incorporating these into a meta-ethnography.

For the overarching synthesis:

In order to synthesise the findings across the two reviews, we will seek to understand how the interrelation between mental health and chronic physical ill-health is experienced for children and young people and how this may impact on the access, acceptability and effectiveness of mental health treatments for this group. This will involve synthesis of the findings from both reviews through the systematic development of the conceptual framework. We also plan to explore whether heterogeneity in the findings may be explained through these understandings and as well as illustrating gaps in the evidence and factors that seem to enhance or limit intervention success.

#### **Analysis of subgroups or subsets**

If data allow, we will explore sub-group analyses by type of LTC, age of child/adolescent, intervention and common intervention and delivery components.

#### **Consultation**

Following development and refinement of the framework we will consult with clinicians and practitioners involved in the provision of mental health and physical health services for this population, children and young people and their families. We will use face to face discussion with individuals and groups and also email to achieve a widespread distribution of different views and experiences. The aim of this process will be to seek feedback from stakeholders (on the credibility of the findings, the clarity of the model and the extent to which it illustrates their experience).

#### **Dissemination**

It will be important to share our findings with all those working in services for children who have long term conditions and either have, or are at risk of, developing mental ill health. We have begun to develop a plan of dissemination that aims to reach those involved in the clinical care of children, charities, academics, policy makers, as well as children and young people and their families. The dissemination plan consists of three main components; plain language summaries, academic journal articles and presentations at key national and regional meetings. As the findings of the reviews emerge the dissemination plan will be further developed with the identification of the key messages for each audience and the most appropriate mechanism for disseminating those messages.

**End-user involvement**

End-user involvement will be an integral part of our project. Children and young people with long term conditions and mental ill health and their families will be invited to join a Children & Young People Research Advisory Group, which will be a sub-group of the Project Advisory Group. Experience suggests three key stages of an evidence synthesis project at which involvement is most valuable – i) at the beginning of the project, once funding is in place but while the protocol is still being finalised to ensure that the focus of the questions and definitions remain focussed on that which is important to service users, ii) making sense of preliminary results for the same reason and iii) during the preparation of the final report and dissemination materials. Meetings will be arranged by the group members in consultation with the core research team to suit project progress, but are likely to occur in months 1, 7, 12 and 14. Members of the core research team will travel to London to contribute to the discussions at these meetings. In addition, there will be a period of intensive engagement with the group during the consultation phase of the project in which the conceptual framework will be discussed.

Role descriptions will be created at the inception of the group and group members will generate their own terms of reference. Basic training in systematic review methods and in being part of an advisory group will be provided. Representatives from the group will be invited to take part in whole team meetings with appropriate preparation and support.

We believe the benefits of the proposed active involvement are that the research outputs will be relevant to the concerns of children and young people with long term condition and mental health need, and their families and the findings disseminated across mediums accessed by children and young people with long term conditions, and their families.



Table mapping research questions onto reviews.

<b>Research Question</b>	<b>Type of evidence to be considered</b>	<b>Addressed by</b>
1) What is the effectiveness and cost-effectiveness of interventions targeting mental health for children and young people with long term conditions and symptoms of mental distress?	Effectiveness outcomes collected from RCTs  Economic outcomes including economic evaluations and cost comparison studies collected from any study that reports the costs or resource implications for the included interventions and comparators.	Review One
2) What are the effects of such interventions on other aspects of social and family functioning?	Effectiveness outcomes collected from RCTs  Information about the experience of those receiving relevant interventions collected from qualitative studies and process evaluations	Review One  Review Two
3) What are the factors that may enhance, or hinder, the successful implementation and / or effectiveness of interventions intended to improve mental health for children and young people with long term conditions and symptoms of mental distress?	Intervention and delivery components collected from RCTs  Information collected from qualitative studies of the experience of interventions  Information from process evaluations of included RCTs	Review One  Review Two