

STUDY PROTOCOL

Stage 1 Registered Report: The experiences and perceptions of parent-child interaction therapy for parents of young children with communication difficulties: A qualitative evidence synthesis protocol [version 2; peer review: 2 approved]

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Abstract

Background: Parent-child interaction therapy refers to a group of interventions mediated by trained parents to address areas of developmental difficulties in children. In the field of speech and language therapy it is used in early intervention for children with speech, language and communication difficulties. The intervention involves training parents and caregivers on the importance of responsivity and language input in daily interactions and coaches them on strategies to implement these with the children. As the success of the intervention is heavily influenced by caregiver engagement, understanding and acceptance, it is important to consider their views. However, to date there has been limited work on synthesising parental views of this intervention.

Methods: This is a protocol for a qualitative evidence synthesis of peer-reviewed qualitative papers addressing the experiences and perceptions of parent-child interaction therapy for parents of children with communication difficulties. We will complete a systematic search of 11 databases, review the reference lists and complete a cited reference search of all included studies. Two authors will independently screen tests for inclusion, initially by title and abstract, with full-text screening as necessary. Thematic synthesis will be used for all included studies. We will appraise the quality of included studies using CASP and confidence in the review findings using GRADE CERQual.

Discussion: As the views of parents are pivotal in the success of this intervention, the findings from this synthesis should help to guide best practice and policy for the future implementation of parent child interaction therapy for children with communication difficulties..

Open Peer Review Reviewer Status 🗸 🗸 **Invited Reviewers** 1 2 version 2 report report (revision) 20 May 2020 version 1 16 Dec 2019 report report 1 Lindsay Pennington (D), Newcastle University, Newcastle upon Tyne, UK 2 Rae Thomas (D), Bond University, Gold Coast, Australia Any reports and responses or comments on the article can be found at the end of the article.

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Keywords

Parent-child interaction therapy, Experiences and perceptions, Speech, language and communication difficulties, Systematic review, Qualitative Evidence Synthesis



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REVISED Amendments from Version 1

We would like to thank our reviewers for their detailed and thorough feedback. We have made a number of changes to the manuscript following this feedback, and hope that it has improved the clarity and quality of our protocol. The most substantive changes were adding more information on the theoretical underpinnings to the intervention, clarifying terminology and issues raised regarding the methodology. Changes were also made to the abstract to reflect those made in the body text.

Any further responses from the reviewers can be found at the end of the article

Background

Speech, language and communication difficulties in young children

Children are said to have a difficulty with speech, language and/ or communication when they are unable to listen, understand or speak in a developmentally appropriate way. Children born with conditions that are known to cause speech, language and communication difficulties, such as Down syndrome, are often identified early in development and so intervention begins early. Other children may not start to receive intervention until they are identified as having a delay or disorder in their language skills, often in the presence of age-appropriate milestones in other areas of development, such as motor skills. The benchmark of fewer than 50 words in their expressive vocabulary and failure to put two words together by two-years is often used to define late talking children (Dollaghan, 2013; Fenson et al., 1994). However, parents and caregivers may notice subtle differences earlier than this. For example, a child who fails to make or respond to eye contact or attend jointly with a caregiver to an event, toy or activity in the first year of life could cause parents to become concerned. Likewise, a child who does not smile, point or use gesture to act out what they want or express a feeling early into the 2nd year, may be identified early from screening of communication difficulties by a health visitor. There are also known risks associated with socialdemographic factors (e.g. gender or socioeconomic status), family history, parenting and child behaviour that are linked to speech, language and communication difficulties (Hammer et al., 2017). Either way, once the child has been identified as having language and communication difficulties, early intervention is a critical and often involves training parents on how to promote early language development effectively in everyday interactions (Barton & Fetting, 2013). This is because the long term negative educational, social and emotional consequences of having early speech and language difficulties early in development have been found to be improved through early intervention that focuses on parent-child interaction (Armstrong et al., 2017; Hammer et al., 2017).

Description of the intervention: Parent-child interaction therapy (PCIT)

Children develop within the context of their family and so parents and caregivers are best placed to support this development. Parent-child interaction therapy refers to a group of interventions mediated by trained parents to address areas of developmental difficulties in children. Although the intervention was originally developed to target challenging behaviour in children, it has since evolved to treat a range of difficulties, including speech language and communication (Lieneman et al., 2017). Language is acquired in everyday interactions between children and their parents, and as parents spend the most time interacting and communicating with their children, parent-child interaction therapy is considered to be ecologically valid and family-centred (Barton & Fetting, 2013). The intervention is mediated through parents and caregivers by training them about the importance of responsivity and the quality and quantity of their language input in daily interactions and coaching them on strategies to implement this. There is now evidence that interventions mediated through parents may be as effective as those mediated through clinicians when delivered with sufficient quality (Burgoyne et al., 2018; Law et al., 2003), are associated with improved outcomes in child language development (Roberts et al., 2019) and are recognised internationally as a valuable approach to remediating communication difficulties in young children (Law et al., 2019). We will use the term 'parent' in this paper to mean all caregivers who interact with children on a daily basis. Caregivers can include grandparents or other family caregiver who take on the 'parent' role for the purposes of the intervention. We will use the term parent-child interaction therapy to refer to those interventions targeting speech, language and communication difficulties in young children.

Parent-child interaction therapy (PCIT) is known by various names, including: '(interactive) focused stimulation'; 'socialinteraction therapy'; 'responsive education/ teaching'; 'naturalistic teaching' or 'milieu teaching' (O'Toole et al., 2018). The aim of all programmes is to train parents to recognise and respond to verbal and nonverbal communication and interaction in their children in order to encourage an increase in these behaviours (Warren et al., 2008). One example of this intervention is the Hanen programme for parents 'It Takes Two to Talk®' (Girolametto & Weitzman, 2006), which educates parents about the importance of child-oriented behaviours to promote joint attention and reciprocal interaction, and helps them to apply language facilitation strategies in natural, everyday interactions. This programme is delivered through classroom-based training of parents in groups in addition to individual coaching of parents with their children, but does not involve direct clinician-child intervention. Enhanced Milieu Teaching (EMT) or prelinguistic milieu teaching is another version of the intervention, which combines elements of responsivity education with behavioural strategies and milieu teaching through modelling and environmental arrangements in order to promote verbal and/or nonverbal language and communication (Hancock & Kaiser, 2007). This intervention is mostly delivered intensively through one-to-one sessions and involves the clinician working directly with the child in addition to coaching the parent. Responsive Teaching is a relationship-based intervention that helps parents to engage in reciprocal interaction and respond contingently to their children's behaviour with high levels of positive affect matched to their children's development, interests, and behavioural style (Mahoney & Perales, 2019). It focuses on the development of childhood pivotal behaviours in the areas of cognition (e.g. exploration and problem-solving), communication (e.g. vocalisation and joint attention) and social emotional functioning (e.g. empathy and cooperation). It can be delivered in groups, but most of the efficacy research has involved individual weekly one-hour sessions over a 3–6-month period. Similar to EMT, parents are first provided with a model of the desired strategy before being coached on their implementation by the clinician as they interact with their child. Finally, many programmes use an intervention known as 'dialogic reading' whereby parents are trained on how to read 'with' and not 'to' their child by engaging in active discussion and strategic questions when sharing a book (Vally et al., 2015).

How the intervention might work

PCIT comes from social-interactionist/constructivist theories of language development, which state that children naturally learn to communicate based on how adults in their environment respond to and interact with them in daily activities (Klatte et al., 2019; Warren et al., 2008). This theory holds that helping children to intentionally use broader and more frequent verbal and/or nonverbal means of communicating and by helping parents to recognise and respond to this in appropriate ways enhances language learning. The effect of this intervention is therefore bi-directional, in that children and adults reciprocally change how they interact as the child's ability to communicate increases. The intervention is also in line with the transactional theory of development (Sameroff & Fiese, 2000) which acknowledges the mutual effect of the child, caregiver and environment on a child's developmental outcome. In the case of children with speech and language difficulties, it is assumed that communication may be more subtle than for typically developing children (e.g. through gestures, movements or vocalisations (Pennington et al., 2018) which can in turn affect how parents recognise and respond to their children. Therefore, this intervention works by training parents to be more aware of all means of communication and how to respond using naturalistic strategies with a greater frequency and intensity to help develop their children's communication. When parents become more responsive to their children's attempt to communicate and respond contingently to the child's message, they improve the language learning environment. This is because any competing attentional and cognitive processing are reduced, allowing the child to focus on the appropriately delivered language input by the parent (for more see Levickis et al., 2018). Pennington et al. (2018) and Oono et al. (2013) also note that additional benefits of the intervention may be to increase parents' confidence and skills in how they communicate with their children as well as reduce parental stress and child frustration as communication becomes more successful for all. The aims of PCIT or parent-mediated interventions are as follows:

- 1. To foster and increase adult-child interaction and joint attention through child-centred activities.
- 2. To promote the frequency and complexity of adult responsivity to non-verbal and verbal communication.

3. To facilitate appropriate language modelling and prompting from adults that help the child to understand and produce language O'Toole *et al.*, 2018.

Roberts & Kaiser (2011) describe PCIT as 'triadic' as it involves the engagement of a clinician, parent and child. They also describe it as having a 'cascading effect' as an experienced clinician trains parents to use the interaction- and language-promoting strategies to a high degree of fidelity and consistency with their child, leading to enhanced language development in the child. This means that there are many aspects that can influence the overall effectiveness of the intervention, including the clinician's experience, how the intervention is delivered, parental implementation of the strategies, and the child's baseline language and cognitive skills and overall (Roberts & Kaiser, 2011; Siller *et al.*, 2013). However, as the agent of change for the intervention is the parent, their engagement, reflection, understanding and acceptance of the intervention have a significant influence on the success of the programme.

Parental experiences and perceptions of PCIT

Two recent papers have investigated the observations of Speech and Language Therapists (SLTs) about parental experiences with this intervention. Klatte et al. (2019) interviewed ten SLTs about their views on the facilitators and barriers towards parental engagement in PCIT for children with Developmental Language Disorder (DLD). They identified that the SLTs expected that they would reach a mutual understanding with parents about each other's roles and expectations. The therapists also expected that they would create a constructive relationship though a supportive environment so that parents would feel empowered to understand their influence on the child's progress. They also identified barriers to the intervention. These included physical (e.g. time, travel, childcare) and biopsychosocial (e.g. depression, illness and learning potential) issues. Davies et al. (2019) interviewed SLTs about their views on parental roles in intervention more broadly. Their findings indicated that SLTs see parents as a 'helper' and expect them to carry out home activities planned by the SLT. This is in contrast Klatte et al. (2019) paper that discussed parental 'empowerment'. Although it could be argued that this was because Davies et al., 2019 did not focus on PCIT, the views do go against the motivation behind parent-led intervention where the parent is a learner or adaptor of the intervention according to their own situation and their child's changing development, and the SLT is in a coaching role.

The views of parents about their role in speech and language therapy intervention have also been reviewed in many studies. For example, Glogowska & Campbell (2000) found that parents do expect to have some role in intervention, but that this needs to made clear to them early on. This can be achieved through discussion about their perceptions, needs and concerns and then negotiated into what can be achieved with the therapist so as to avoid any misunderstanding. Parents often have different expectations about the therapy process, particularly that it will involve direct contact between the therapist and the child, and

may not expect to be so heavily involved in the intervention themselves. For example, Baxendale et al. (2001) completed questionnaires followed by in-depth interviews involving eight parents who participated in traditional clinicbased therapy, and ten who took part in the Hanen Parent Programme focusing on PCIT. All of the children involved were aged between 30 and 42 months and had a diagnosis of language impairment. Parents in the PCIT group initially had difficulty accepting the philosophy behind this indirect approach, but later appreciated that it was more appropriate for children of this age. They also were able to attribute their child's progress to changes they had made in their own interaction styles more than those in the clinic-based group. Although there were aspects of the intervention such as role play that were not viewed positively, the use of video feedback and support from other parents was welcomed. Davies et al. (2017) also interviewed parents about their role in SLT interventions and although parents were initially uncertain, as they gained greater experience they understood the importance of their role as an intervener. Similarly, Carroll (2010) reviewed parents' expectations of speech and language therapy for their children with intellectual disability. It was noteworthy that parents in this study saw the therapist as the expert on their child in line with more of a medical model of treatment and expected the therapist to make decisions and carry out interventions to 'fix' their child's speech difficulty. They were more ambivalent about their own role, indicating a possibly mismatch between their own expectations and that of the therapist.

Lyons et al. (2010) completed focus groups with parents about their expectations and experiences before and after engaging in an early intervention programme. Similar to previous studies, they noted that parents had expected to be guided about how to facilitate their child's speech and language development but that their role would be more 'observational'. They were therefore uncertain about why the activities were focused on their own behaviour instead of the child's. Lyons et al. (2010) concluded that therapists need to move away from an 'expert' role, and engage in discussion with parents about their expectations before therapy starts so that they can work out what is to be involved together. This will ultimately affect parental perceptions and engagement with therapy. Finally, a recent qualitative systematised review looked at parental engagement in early speech-language pathology interventions (the term for speech and language therapy used in the US and Australia) (Melvin et al., 2019). 'Engagement' in this paper related to parental investment and involvement in therapy, which was found to be a complex process. The review found that parents need time and ongoing support in order to become empowered and engaged in early interventions. Furthermore, each parent engaged with the interventions in a unique way, and so time, open communication and trust building were identified as important aspects.

Why is it important to do this synthesis?

Parental perceptions of this intervention are central to understanding the complex factors that make this intervention work. As discussed above, research has noted that parents do not always expect to have to be so heavily involved in their child's treatment, but rather may assume that the SLT will 'fix' their

child (Carroll, 2010; Goodhue et al., 2010). In addition, as the SLT will often not treat the child directly, the intervention is considered 'indirect', which can be confusing and unsatisfactory for parents (Klatte & Roulstone, 2016). Although many quantitative systematic reviews exist on the effectiveness of this intervention (Oono et al., 2013; O'Toole et al., 2018; Pennington et al., 2018; Roberts & Kaiser, 2011; Roberts et al., 2019; Zwi et al., 2011), few have considered the qualitative evidence around the understanding, acceptability and implementation challenges that exist for parents. A recently published review by Melvin et al. (2019) did examine parental engagement in early interventions, but focused more broadly at the area of early intervention and not PCIT specifically. As PCIT requires parents to take on the role of the clinician and be coached in delivery of the intervention, parents are involved more heavily than in other interventions. Therefore, understanding the experiences of parents in this intervention in particular is worth investigating.

As outlined in the previous section, a number of qualitative studies have been published in this field. We need to gather and synthesize this evidence to describe parental experiences of this intervention using a systematic and rigorous approach. Integrating these findings with the previously published systematic reviews will provide a substantial evidence base for this intervention (Schlosser, 2004). This will ultimately ensure that parents can be informed decision makers where they are provided with all of the relevant information so that they can actively collaboration with professionals and advocate for their children (Crais *et al.*, 2006). The findings from the synthesis will be useful in guiding practice and policy for the implementation of PCIT, by capturing the parent voice in how they view their role in improving their child's language and communication outcomes.

Objectives

The aim of this review is to examine the experiences and perceptions of PCIT for parents of children with speech, language and communication difficulties using qualitative evidence synthesis. The objectives of the review are to:

- Describe the experiences and perceptions of PCIT for parents of children with speech, language and communication difficulties. The intervention targets the language and interaction patterns of parents and their children in everyday interactions and can involve group and/or individual training. It can take place in community, clinical, preschool/ school or home-based settings. Interventions that involve telehealth or connected health technologies into the family home will also be included.
- Examine the potential implications of the themes from the parental views that emerge from this synthesis for policy, regulation and practice in providing PCIT for children with communication difficulties.

Protocol

This protocol has been submitted to the International Prospective Register of Systematic Reviews (PROSPERO) and this article will be updated with the identification number once the protocol has been accepted by PROSPERO.

Criteria for considering studies for this synthesis

Types of studies. We will consider primary research studies that use qualitative design methods such as ethnography, phenomenology, case studies and grounded theory for inclusion. The study design and analysis method (e.g. thematic analysis) must be clearly reported and must be qualitative to be included in the review. Mixed-methods studies will be included if it is possible to extract the qualitative data. All studies must be peerreviewed articles. Studies which collect data qualitatively but analysed it quantitatively, and studies where the full text is not available will be excluded.

Types of participants. Study participants will be parents and caregivers of children with any type of speech, language or communication difficulty. The children will have a range of conditions such as autism spectrum disorder, Down syndrome, Cerebral Palsy, Intellectual Disability, DLD or can be late-talking children with a communication difficulty of unknown origin. Studies will be included once the author(s) have stated that the children have a speech, language and communication difficulty or delay/disorder as diagnosed by a speech and language therapist. Children in the studies must be between the ages of birth to six year of age. There is the possibility of subgroup analysis within this population, for example analysis of perspectives of parents of children with autism spectrum disorder compared to children with other developmental/communication difficulties; group vs. individually-delivered interventions or clinic vs. home-based interventions (see Subgroup Analysis below).

Types of interventions. All types of PCIT interventions designed to improve the communication, interaction and language input of parents for their children with communication and language difficulties. The intervention will involve coaching, supervision and support from a clinician and will take place either on an individual or group basis. The interventions will take place in naturalistic contexts and focus on everyday interactions between parents and their children. If the intervention involves parent-mediated intervention delivered in conjunction clinician-mediated intervention, we will include it as long as the perspectives of parents is presented. However, we will exclude interventions where the clinician is the main provider of the intervention and parents are encouraged to do 'home practice' or 'homework' only, but no coaching is provided. We will also exclude interventions that focus only on speech sound production or stuttering/stammering as they tend to be more behavioural in their orientation. Finally, interventions that are parent-mediated but focus on other developmental aspects such as disruptive behaviour, motor development, social-emotional, cognitive or self-help skills will be excluded.

Phenomenon of interest. The phenomenon of interest in this study is the experiences and perceptions of parents who take part in PCIT, which include studies of acceptability, engagement, understanding and importance of the intervention, facilitators and barriers to the intervention, parental role in changing their child's language and communication, and any quality of life or stress indicators. Studies that focus on broader aspects of

parenting children with communication difficulties will be excluded.

Search methods for identification of studies

The review will systematically search the literature using electronic databases and also purposively sample papers using citation searching, contacting key authors and following up of references lists as outlined in Booth, (2016).

Electronic searches

We will search the following electronic databases:

- Scopus
- · Web of Science
- EBSCO- CINAHL
- ERIC
- · PsychINFO
- Embase
- · Cochrane
- · PubMed
- · Academic Search Complete
- · ProQuest Dissertations and Theses
- SpeechBITE

Using guidelines developed by the Cochrane Qualitative Research Methods Group for searching qualitative evidence (Harris *et al.*, 2018), we will develop search strategies for each database in consultation with an expert librarian (DOD). We will not apply any limits on language, date or location. The search will be conducted by one author (COT) on all databases over one week.

A summary of the electronic search string is presented in Table 1. Certain terms will be truncated, for example parent* or child* to ensure all spellings are captured. We will adapt our searching of title and abstracts to the individual databases. We will report the results of searching, screening and included studies using the PRISMA flowchart (Moher *et al.*, 2009).

Searching other resources. We will review the reference lists of all the included studies and key references. We will conduct a cited reference search of all included studies identified by the initial search using Google Scholar's *Cited by* option (Booth, 2016). We will contact authors of included studies to clarify reported published information and contact researchers with expertise relevant to the synthesis topic to request studies that might be eligible.

Data collection, management and synthesis

Selection of studies. We will import all references into Endnote X9 and remove duplicates. Two authors (COT and RL) will screen titles and abstracts independently to evaluate

Table 1. Search strings.

	experienc* OR perception*
AND	"parent-child interact*" OR focus*near/2 stimulation OR natural* near/2 teaching OR milieu near/2 teaching OR responsiv* near/2 education OR responsiv* near/2 teaching OR Hanen OR parent near/2 mediat* OR dialogic near/2 reading
AND	coach* OR educat* OR intervention* OR learn* OR program* OR teach* OR train* OR therap*
AND	parent* OR maternal* OR mother* OR father* OR paternal* OR carer* OR caregiver* OR care-giver*
AND	child* OR speech* or languag* or communicat*
AND	delay* OR disorder* OR disability* OR difficult* OR impair* OR need OR problem*
AND	qualitative OR "mixed*method*" OR narrative OR phenomenol* OR ethno* OR questionnaire OR "grounded theory" OR "case*study*" OR "action research" OR "focus group" OR thematic OR construction* OR hermeneutic OR heurist*

eligibility against our inclusion/exclusion criteria using Covidence systematic review management system. Where it is not possible to determine whether to include an article or not, the full text of the article will be retrieved. One author (COT) will review all full-text articles and another author (RL) will share second screening of all full-text articles. Disagreements between authors will be solved via discussion, or if required, in consultation with a third team member (CH). As necessary and appropriate, we will contact authors of potential included studies for further information. We will include a table listing the studies excluded from our synthesis at the full text stage and the main reasons for exclusion. We will collate multiple reports of the same study. We will report the outcome of the search strategy in a PRISMA flowchart (Moher et al., 2009).

Sampling of studies. As qualitative evidence synthesis aims for variation in concepts rather than an exhaustive sample, and because large numbers of studies can affect the quality of the analysis, if the review retrieves more than 40 eligible papers, we will develop a sampling framework to ensure richness and representativeness, as used by Ames et al. (2019). This will involve extracting data from all studies found such as participant characteristics (e.g. mother/father, socioeconomic status and diagnosis of the child); study setting (e.g. home, clinic or tele-practice delivery); geographical location; type of PCIT (e.g. EMT or Hanen Parent Programme); data richness (quantity and quality of data provided) and study objectives in order to develop a sampling framework. Data richness will be assessed on a scale developed by EPOC, (2017). We will aim to ensure that the sample consists of a wide range of participants, rich data and a focus that resembles our synthesis objectives. We will report how we developed any sampling framework in the full review.

Data extraction and management. We will import all full text articles into NVivo Version 12. Data extraction and the thematic synthesis will be facilitated within NVivo (Houghton *et al.*, 2017). We will use categories such as author, year,

location, study setting, sample characteristics (parents and children), intervention type, intervention setting, design, ethics, data collection and analysis methods, results/themes/findings including supporting quotations and sources of support.

Data synthesis. Based on our consideration of the RETREAT criteria for selected qualitative evidence synthesis approaches, the review will synthesise the included qualitative studies using thematic synthesis (Booth et al., 2018). According to Thomas & Harden (2008), thematic synthesis is an inductive approach using three steps, which start with line by line coding of primary data from the included studies to develop descriptive themes and generate broader analytical themes. The primary data will include direct quotes from participants and the author interpretations of the data. This will be conducted within NVivo with guidance from a previous synthesis on how best to use the coding software (Houghton et al., 2017) and allows for transparency and clarity in the synthesis process. We chose thematic synthesis because it fits with the aim of this review, which is to provide information for policy and practice by presenting the voice of those who experience the intervention (Barnett-Page & Thomas, 2009). One author (COT) will carry out the thematic synthesis with continuous input from two other authors (RL and CH) authors at each stage. All of the authors will read and make contributions to the final paper.

Subgroup analysis. The potential for subgroup analysis will be determined inductively through the synthesis. Where there are sufficient numbers of studies found, this may include subgroup analysis based on the diagnosis of the children (e.g. children with autism vs. developmental language disorder); the nature of the intervention (group vs. individual) the setting in which it is delivered (clinic vs. home-based) or geographical location. It is generally middle-class, Western parents that participate in these studies, although more recently interventions have targeted at risk groups of families from lower socio-economic settings. Therefore, further subgroup analysis

might look at the differences between those from lower and middle/upper socioeconomic groups as measured by maternal education and/or income levels as described by the studies.

Assessment of methodological limitations in included studies. Two review authors (COT and RL) will independently assess the quality of the included studies using the Critical Appraisal Skills Programme (CASP) tool (CASP, 2018). This tool examines a number of aspects of quality including the methodology, research design, data collection, relationship between researcher and participant, data analysis, findings and the value of the research among others. Although we will not exclude studies on the basis of their methodological limitations, this assessment will inform our overall confidence in the review findings.

Assessment of confidence in the synthesis findings. Two authors (COT and CH) will use the GRADE CERQual (Confidence in the Evidence from Reviews of Qualitative research) approach to summarise our confidence in each finding (Lewin et al., 2018). This involves examining four main elements:

- 1. Methodological limitations of included studies.
- 2. Coherence of the review findings to the review question.
- Adequacy of the data is in supporting the review finding.
- Relevance of the included studies to the review question.

After assessing each of the four components, we will make a judgement about the overall confidence in the evidence supporting the review finding. We will judge confidence as high moderate, lower or very low. The final assessment will be based on consensus among the review authors. We will then include a sensitivity analysis to examine the contribution of the poorer quality studies to the overall findings (Houghton *et al.*, 2017; Thomas & Harden, 2008).

Reporting

This review will be reported in line with the ENTREQ guidelines (Tong *et al.*, 2012). A completed PRISMA-P checklist is available from the Open Science Framework at https://www.doi.org/10.17605/OSF.IO/FDP3W (O'Toole, 2019).

Dissemination of findings

The findings will be submitted to a peer-reviewed journal for publication. They will also be integrated into a wider study that is currently planned to look at the feasibility of an intensive parent-child interaction therapy for Irish children with Down syndrome. The findings will also be shared with stakeholders, including parent and therapist groups via newsletters, social media and professional bodies.

Study status

The study has not yet started.

Discussion

Understanding the perspectives of parents in parent-child interaction therapy (PCIT) is important in deciphering facilitators and barriers to the behaviour change techniques that needs to take place in order to improve children's speech, language and communication (Justice *et al.*, 2015). The purpose of this protocol is to systematically assesses peer-reviewed qualitative studies of the perceptions of parents in implementing PCIT.

Furthermore, PCIT requires parents to alter how they talk to and interact with their children. It is therefore important to consider parental experiences and perceptions of the intervention in light of theories regarding the factors that might influence their implementation and maintenance. These include Normalisation Process Theory (May et al., 2009) which addresses how practices are put into action and embedded into everyday social routines and contexts and maintained over time. Furthermore, the Theoretical Framework of Acceptability (Sekhon et al., 2017) considers factors such as attitude, burden, perceived effectiveness, and self-efficacy which influence how people experience and respond to an intervention. Finally, a recent paper identified factors related the context, mechanism and outcomes that should be reviewed when looking at theories of collaborative work practices between parents and SLTs (Klatte et al. (2020). It will be important to evaluate these theories in light of the evidence found in this synthesis.

The outcomes of this synthesis will add to the existing quantitative evidence for this intervention and give greater recognition to the parent's perspectives which will untimely help to facilitate their involvement and increase their satisfaction with their child's therapy (Glogowska & Campbell, 2000). In this way the results should be included for future guidance for practice, regulation and policy in providing PCIT for all children with communication difficulties.

Data availability

Underlying data

No data is associated with this article.

Reporting guidelines

Open Science Framework: PRISMA-P checklist for 'The experiences and perceptions of parent-child interaction therapy for parents of young children with communication difficulties: A qualitative evidence synthesis protocol', https://www.doi.org/10.17605/OSF.IO/FDP3W (O'Toole, 2019).

Data are available under the terms of the Creative Commons Zero "No rights reserved" data waiver (CC0 1.0 Public domain dedication).

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Open Peer Review

Current Peer Review Status:





Version 2

Reviewer Report 08 June 2020

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Lindsay Pennington (ii)



Institute of Health and Society, Newcastle University, Newcastle upon Tyne, UK

The revised article is very clearly presented and well argued. I look forward to reading the results of the study in due course.

Competing Interests: No competing interests were disclosed.

I confirm that I have read this submission and believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard.

Reviewer Report 29 May 2020

https://doi.org/10.21956/hrbopenres.14161.r27429

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Rae Thomas (1)



Institute for Evidence-Based Healthcare, Bond University, Gold Coast, Australia

The authors have done a good job at addressing my queries and articulating the definition of PCIT in the field of speech and language therapy.

Although I agree that views of intervention participants (in this case parents) should inform policy and practice I am less convinced they often do. Does the reference for Barnett-Page and Thomas, 2009 report policies that have been modified because of participant views or is it theoretical? If the former, it would be good, in the discussion to demonstrate how this might be achieved in concrete rather than abstract forms,

particularly if this is an objective.

Thank you for the opportunity again to review the manuscript. I believe it is fine for indexing.

Competing Interests: No competing interests were disclosed.

Reviewer Expertise: Psychology, implementing behaviour management interventions, conducting systematic and scoping reviews.

I confirm that I have read this submission and believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard.

Author Response 04 Jun 2020

Ciara O'Toole, University College Cork, Cork, Ireland

Thank you for your comments on our revised paper and for approving the same. In relation to the query about Barton-Page and Thomas (2009) review of methods for synthesising qualitative research, this was commissioned by the Department of Health in the UK to inform policy in practice. In this case, it is more theoretical- presenting the evidence of using thematic synthesis in a way "that could inform" policy although as you point out, there are as yet no concrete examples of this.

Competing Interests: No competing interests were disclosed.

Version 1

Reviewer Report 07 May 2020

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Institute for Evidence-Based Healthcare, Bond University, Gold Coast, Australia

Overall comment: Thank you for the opportunity to review this manuscript. There is thoroughness in the proposed review method, however the definition of the intervention is confusing.

The authors appear to take a broad definition of Parent-Child Interaction Therapy (PCIT) and appear to encompass a range of different therapies. However, I also note the authors use the name PCIT as an actual intervention ("PCIT is <u>an</u> early intervention"; As the success of <u>the</u> intervention" etc). PCIT is a behavioural intervention developed in the 1980s for children with behavioural challenges, not specifically for children with communication difficulties and their parents (http://www.pcit.org). It would conceivably work for children with these challenges and their parents but it was not developed for that purpose.

Confusion regarding the specific intervention known as PCIT versus interventions that proritise and intervene in parent-child interactions is underscored by not including any references from the intervention known as PCIT. This needs to be clarified and is my major concern with the protocol and is reiterated in several sections of my review. I note the authors have published a Cochrane SR on "Parent-mediated interventions for promoting communication and language development in young children with Down syndrome" and some references are the same. Is this what is meant by PCIT?

Given the broad focus of the research study (the long list of phenomenon of interest, page 6), it might be better to consider this a scoping review.

Title:

1. It is convention for the study design to be part of the title. Do the authors intend for this to be a systematic review? It is set up like one and has it as a key word but it is not mentioned in either the title or abstract.

Abstract:

- 1. The authors will need to clarify what they mean by PCIT throughout the manuscript. This is the major concern I have with the manuscript in its present format.
- 2. The first sentence of the Methods part of the abstract is not necessary there because it is an aim and not a method.
- 3. In the Discussion section of the abstract, what do the authors mean be "help guide best practice and policy"? How do the authors think experiences and perceptions of people involved in PCIT will guide these two areas?

Introduction:

- 1. Please define PCIT. It is very different to the therapy conventionally known as PCIT.
- 2. References are missing in some areas For example, "parent-child interaction therapy is considered to be ecologically valid and family-centred" (page 3) and "Parent-child interaction therapy (PCIT) is known by various names, including xxx, xxx etc". I note the authors have published a Cochrane SR on parent-mediated interventions for promoting communication and language development. Is this what they mean by PCIT? If so, please use this term consistently.
- 3. There is no documentation as to how the authors intend to meet Objective 2 "Examine the potential implications of this synthesis for policy, regulation and practice in providing PCIT for children with communication difficulties". Please describe how this is to be done in the Data collection, management and synthesis section.

Protocol:

- 1. The authors state: "Finally, interventions that are parent-mediated but focus on other developmental aspects such as disruptive behaviour, motor development or self-help skills will be excluded." What do the authors mean when they say "other" developmental aspects? What developmental focus do they intend to include?
- Normally I would suggest the large number of databases is unnecessary but as the authors have already conducted a Cochrane review with these databases, I assume they have a fair idea of the number of articles they need to screen. Having said that this review is more broad than Down syndrome.
- 3. In the data synthesis section, what will you extract? The quotes from the papers or the interpretation of those quotes to a framework? Should I assume "primary data" are the actual

quotes?

4. I think the heading "Subgroup analysis and heterogeneity" should be modified as this typically relates to quantitative analyses. I would caution the authors in doing too many "subgroups" as the included number of articles is likely small.

Discussion:

1. Please describe how this will provide guidance for regulation and policy.

Thank you.

Have the authors pre-specified sufficient outcome-neutral tests for ensuring that the results obtained can test the stated hypotheses, including positive controls and quality checks? Yes

Is the rationale for, and objectives of, the study clearly described? Partly

Is the study design appropriate for the research question? Yes

Are sufficient details of the methods provided to allow replication by others? Yes

Are the datasets clearly presented in a useable and accessible format? Not applicable

Competing Interests: No competing interests were disclosed.

Reviewer Expertise: Psychology, implementing behaviour management interventions, conducting systematic and scoping reviews.

I confirm that I have read this submission and believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard, however I have significant reservations, as outlined above.

Author Response 14 May 2020

Ciara O'Toole, University College Cork, Cork, Ireland

General Response:

We would like to thank our reviewers for their detailed and thorough feedback. We have made a number of changes to the manuscript following this feedback, and hope that it has improved the clarity and quality of our protocol. The most substantive changes were adding more information on the theoretical underpinnings to the intervention, clarifying terminology and issues raised regarding the methodology.

We have responded to each reviewer's comment in turn using the following codes: R1 = reviewer 1; R2= Reviewer 2; C1 = comment 1 etc.

R2CI: The authors appear to take a broad definition of Parent-Child Interaction Therapy (PCIT) and

appear to encompass a range of different therapies. However, I also note the authors use the name PCIT as an actual intervention ("PCIT is an early intervention"; As the success of the intervention" etc). PCIT is a behavioural intervention developed in the 1980s for children with behavioural challenges, not specifically for children with communication difficulties and their parents (http://www.pcit.org). It would conceivably work for children with these challenges and their parents but it was not developed for that purpose. Confusion regarding the specific intervention known as PCIT versus interventions that proritise and intervene in parent-child interactions is underscored by not including any references from the intervention known as PCIT. This needs to be clarified and is my major concern with the protocol and is reiterated in several sections of my review. I note the authors have published a Cochrane SR on "Parent-mediated interventions for promoting communication and language development in young children with Down syndrome" and some references are the same. Is this what is meant by PCIT?

Given the broad focus of the research study (the long list of phenomenon of interest, page 6), it might be better to consider this a scoping review.

Response: In the field of speech and language therapy, the term PCIT is the overarching term used to refer to a number of interventions that are mediated by trained parents to improve the speech, language and communication of children. However we appreciate that the term is also used in other fields, particularly those used to target challenging behaviour. We have therefore acknowledged the various uses of the term in our abstract and introduction and clarified what we mean by PCIT in the context of this review. The focus of the review is therefore only on PCIT that targets language and communication and so we will continue to explore it as a qualitative evidence synthesis.

R2 C2: Title: It is convention for the study design to be part of the title. Do the authors intend for this to be a systematic review? It is set up like one and has it as a key word but it is not mentioned in either the title or abstract.

Response: This is a qualitative evidence synthesis, which is an umbrella term for the methodologies associated with the systematic review of qualitative research evidence (Carroll, 2017) and so it is important that this is retained in title. It is also in the methods section of the abstract. This is now clarified in the body of the text.

R2 C3: Abstract: The authors will need to clarify what they mean by PCIT throughout the manuscript. This is the major concern I have with the manuscript in its present format. **Response:** This term is now explained in the abstract and introduction.

R2 C4: The first sentence of the Methods part of the abstract is not necessary there because it is an aim and not a method.

Response: As qualitative evidence synthesis is the method, it has been retained in this sentence, and the term 'qualitative' added.

R2 C5: In the Discussion section of the abstract, what do the authors mean by "help guide best practice and policy"? How do the authors think experiences and perceptions of people involved in PCIT will guide these two areas?

Response: Parental views should form part of best practice and policy. This sentence has been rewritten to clarify this. We have also expanded this in the section *Data synthesis* as thematic synthesis is regarded as a useful intervention for informing policy.

R2 C6: Introduction: Please define PCIT. It is very different to the therapy conventionally known as PCIT.

Response: We have now defined what we mean by PCIT in the context of speech and language therapy interventions, acknowledging that it can be used for other areas of behaviour.

R2 C7: References are missing in some areas For example, "parent-child interaction therapy is considered to be ecologically valid and family-centred" (page 3) and "Parent-child interaction therapy (PCIT) is known by various names, including xxx, xxx etc". I note the authors have published a Cochrane SR on parent-mediated interventions for promoting communication and language development. Is this what they mean by PCIT? If so, please use this term consistently. Response: References now added. We have clarified what is meant by PCIT in the introduction and is the same meaning that was taken in the Cochrane review. More recent descriptions of the intervention have started to use the term PCIT (e.g. Klatte et al., 2019 as referenced in this paper).

R2 C8: There is no documentation as to how the authors intend to meet Objective 2 "Examine the potential implications of this synthesis for policy, regulation and practice in providing PCIT for children with communication difficulties". Please describe how this is to be done in the Data collection, management and synthesis section.

Response: Thematic synthesis is known to help inform policy and practice. We have highlighted this in the Data synthesis section with the sentence: We chose thematic synthesis because it fits with the aim of this review, which is to provide information for policy and practice by presenting the voice of those who experience the intervention (Barnett-Page & Thomas, 2009).

R2 C9: Protocol: The authors state: "Finally, interventions that are parent-mediated but focus on other developmental aspects such as disruptive behaviour, motor development or self-help skills will be excluded." What do the authors mean when they say "other" developmental aspects? What developmental focus do they intend to include?

Response: The other areas of development are those listed and social-emotional and cognitive development which have been added. The only focus will be on speech, language and communication. Anything outside of this will be excluded.

R2 C10: Normally I would suggest the large number of databases is unnecessary but as the authors have already conducted a Cochrane review with these databases, I assume they have a fair idea of the number of articles they need to screen. Having said that this review is more broad than Down syndrome.

Response: We have retained the planned search strategy to ensure that a wide range of papers is captured.

R2 C11: In the data synthesis section, what will you extract? The quotes from the papers or the interpretation of those quotes to a framework? Should I assume "primary data" are the actual quotes?

Response: This has been clarified with the sentence: *The primary data will include direct quotes from participants and the author interpretations of the data*

R2 C12: I think the heading "Subgroup analysis and heterogeneity" should be modified as this typically relates to quantitative analyses. I would caution the authors in doing too many "subgroups" as the included number of articles is likely small.

Response: This heading has been changed to 'subgroup analysis'. A caveat of not including too many subgroups is provided.

R2 C13: Discussion: Please describe how this will provide guidance for regulation and policy.

Response: The link between including parental perspectives for future iterations of policy has been added in the final sentence.

Competing Interests: No competing interests were disclosed.

Reviewer Report 11 February 2020

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Lindsay Pennington

Institute of Health and Society, Newcastle University, Newcastle upon Tyne, UK

This study will examine parents' perceptions and experiences of training to facilitate their child's language and communication development provided by speech and language therapists. This type of intervention is often used in clinical practice, but varies in the methods of delivery, content and dosage. The effectiveness of the interventions is yet to be fully determined. This review will provide timely and important information to guide clinical practice and further research.

The protocol is clearly written and the design of the review to be conducted is well thought through. The search strategy is appropriate and comprehensive. The analysis plan is appropriate. The authors should state if analysis will be inductive or both inductive and deductive. Theories of factors influencing implementation of interventions could be helpful in assessing factors that may influence parents' experiences and perceptions of the intervention, highlighting factors that have been raised in research to date and those that may be influential but have not yet been assessed. (e.g. May *et al.* (2009)¹; Sekhon *et al.* (2017)²).

Please state the characteristics on which studies will be sampled if over 40 fit the review criteria.

Subgroup analysis could include setting – at least one of the named interventions has been delivered outside North America, Europe and Australia (where the bulk of the research takes place). How will socio-economic group be defined?

I have some minor suggestions for changes to the text:

- In the first section of the Background, what is the evidence for parent-child interaction training for children 'at risk' versus children with diagnosed speech, language or communication difficulty?
- N.B. children with biomedical conditions such as Down syndrome are not always diagnosed early.
- Description of the intervention: Explain the meaning of 'pivotal behaviours' and add a supporting reference.
- How the intervention may work: The authors could provide more detail on how increased parental responsivity may facilitate children's communication. Theories of communication and language

development could be added here.

- Type of Participants: cerebral palsy, Down syndrome etc. are not 'types of speech, language or communication difficulty'. The first part of this section should be rewritten.
- 'Goldbard' should be Goldbart.
- Speech and language therapy is used in the main to describe the workforce, but on P5 'speech
 pathology' is referred to. It would be helpful to explain this additional term.
- There are a few typos in the protocol and it requires some more proof reading.

References

- 1. May CR, Mair F, Finch T, MacFarlane A, et al.: Development of a theory of implementation and integration: Normalization Process Theory. *Implement Sci.* 2009; **4**: 29 PubMed Abstract | Publisher Full Text
- 2. Sekhon M, Cartwright M, Francis J: Acceptability of healthcare interventions: an overview of reviews and development of a theoretical framework. *BMC Health Services Research*. 2017; **17** (1). Publisher Full Text

Have the authors pre-specified sufficient outcome-neutral tests for ensuring that the results obtained can test the stated hypotheses, including positive controls and quality checks? Yes

Is the rationale for, and objectives of, the study clearly described? Yes

Is the study design appropriate for the research question? Yes

Are sufficient details of the methods provided to allow replication by others? Partly

Are the datasets clearly presented in a useable and accessible format? $\begin{tabular}{l} Yes \end{tabular}$

Competing Interests: No competing interests were disclosed.

I confirm that I have read this submission and believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard, however I have significant reservations, as outlined above.

Author Response 14 May 2020

Ciara O'Toole, University College Cork, Cork, Ireland

General Response:

We would like to thank our reviewers for their detailed and thorough feedback. We have made a number of changes to the manuscript following this feedback, and hope that it has improved the clarity and quality of our protocol. The most substantive changes were adding more information on the theoretical underpinnings to the intervention, clarifying terminology and issues raised regarding the methodology.

We have responded to each reviewer's comment in turn using the following codes: R1 = reviewer 1; R2= Reviewer 2; C1 = comment 1 etc.

R1C1: The authors should state if analysis will be inductive or both inductive and deductive. **Response:** We have clarified that the analysis is inductive, and amended the working on page 7 to be "Thematic synthesis is an *inductive* approach using three steps".

R1C2: Theories of factors influencing implementation of interventions could be helpful in assessing factors that may influence parents' experiences and perceptions of the intervention, highlighting factors that have been raised in research to date and those that may be influential but have not yet been assessed. (e.g. May et al. (2009)1; Sekhon et al. (2017)2).

Response: Thank you for directing us to this literature. It will be very relevant when reviewing our findings from the synthesis and taking them forward. We have included reference to these studies and the theories on factors that influence implementation of these interventions on page 7 under 'Discussion. However as we are not using a framework synthesis which involves a more deductive approach we will not be analysing our results to an existing theoretical model.

R1C3: Please state the characteristics on which studies will be sampled if over 40 fit the review criteria.

Response: We have added greater detail on this based on Ames criteria under 'sampling of studies' and referring to the to the data richness scale as developed by the Cochrane Effective Practice and Organisation of Care (EPOC) group. We have included the following text: This will involve extracting data from all studies found such as participant characteristics (e.g. mother/father, socioeconomic status and diagnosis of the child); study setting (e.g. home, clinic or tele-practice delivery); geographical location; type of PCIT (e.g. EMT or Hanen Parent Programme); data richness (quantity and quality of data provided) and study objectives in order to develop a sampling framework. Data richness will be assessed on a scale developed by EPOC (2017). We will aim to ensure that the sample consists of a wide range of participants, rich data and a focus that resembles our synthesis objectives. We will report how we developed any sampling framework in the full review.

R1C4: Subgroup analysis could include setting – at least one of the named interventions has been delivered outside North America, Europe and Australia (where the bulk of the research takes place). How will socio-economic group be defined?

Response: We have added geographical location as a potential subgroup analysis, and clarified that SES will be defined by maternal education and/or income level as described in the studies.

R1C5: In the first section of the Background, what is the evidence for parent-child interaction training for children 'at risk' versus children with diagnosed speech, language or communication difficulty?

Response: The terms 'at risk' has now been removed and clarification added that early intervention is important for all children with language and communication difficulties.

R1C6: N.B. children with biomedical conditions such as Down syndrome are not always diagnosed early.

Response: We have clarified to say that they are 'often' diagnosed early in development to acknowledge that this does not always happen.

R1C7: Description of the intervention: Explain the meaning of 'pivotal behaviours' and add a supporting reference.

Response: Examples of pivotal behaviours are now added in the sentence "It focuses on the development of childhood pivotal behaviours in the areas of cognitive (e.g. exploration and problem-solving), communication (e.g. vocalisation and joint attention) and social emotional functioning (e.g. empathy and cooperation)." Supporting reference of related manual for the intervention (Mahoney & Perales, 2019) is now provided.

R1C8: How the intervention may work: The authors could provide more detail on how increased parental responsivity may facilitate children's communication. Theories of communication and language development could be added here.

Response: Further information on how parental responsibility facilitates communication is provided under 'How the intervention may work'. Theories such as social interactionist/constructive and views of language development and the transactional theory of development are explored.

R1C9: Type of Participants: cerebral palsy, Down syndrome etc. are not 'types of speech, language or communication difficulty'. The first part of this section should be rewritten.

Response: This section has now been re-written

R1C10: 'Goldbard' should be Goldbart.

Response: Amended

R1C11: Speech and language therapy is used in the main to describe the workforce, but on P5 'speech pathology' is referred to. It would be helpful to explain this additional term.

Response: This is the terms for speech and language therapy used in the US and Australia. This explanation has been added

R1C12: There are a few typos in the protocol and it requires some more proof reading.

Response: The protocol has been full proofread.

Competing Interests: No competing interests were disclosed.