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A systematic review and meta-analysis of the effectiveness of interventions to improve psychological wellbeing in the parents of children with cerebral palsy

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ABSTRACT

Background: Caregivers of children with cerebral palsy (CP) face unique demands and challenges, with potential negative impact on psychological wellbeing and consequences for both caregiver and child. Timely support could improve parental wellbeing.
Aim: To evaluate the effectiveness of interventions aimed at improving the psychological well-being of caregivers of children with CP.
Methods and procedures: Systematic review and meta-analysis of randomised controlled trials of interventions to improve the psychological well-being of caregivers of children with cerebral palsy. Databases (including MEDLINE, EMBASE, PsycINFO, Cochrane) were searched for relevant English language publications between January 1990 and December 2017. Risk of bias was assessed including randomization, allocation concealment, incomplete outcome data and selective outcome reporting.
Outcome and results: We included 13 studies (1293 participants, 1/3 with CP). Six studies investigated a positive parenting intervention; the other seven studies covered a range of other interventions. Meta-analysis of 7 studies (662 participants) showed that interventions significantly improved parental wellbeing (standardised mean difference -0.61, 95% CI -0.92 to -0.30, $z = 3.84$, $p = 0.0001$).
Conclusions and implications: Limitations include small sample sizes and heterogeneity in study design; however, our results indicate that interventions can improve the wellbeing of parents of children with CP and should be made available.

What this paper adds?

Research has indicated the need for greater psychological support of parents of children with Cerebral Palsy (CP). This comprehensive systematic review and meta-analysis evaluates the effect of interventions on the psychological wellbeing of primary caregivers of children with CP. We found a significant benefit and highlighted that this can be an indirect effect, where the primary outcome focusses on the child (e.g. the child's behaviour). This review also highlights the paucity of studies evaluating early support

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for parents of infants with emerging CP and recommends areas for future research and improvement.

1. Introduction

In the UK, Cerebral Palsy (CP) occurs in around 1 in every 500 live births (Oskoui, Coutinho, Dykeman, Jette, & Pringsheim, 2013). CP is an umbrella term referring to a range of disorders affecting body movement, balance and posture which affects individuals differently. CP is caused by abnormal development or damage in one or more brain areas controlling muscle tone and motor activity, which can occur before, during or after birth (early acquired brain injury). The signs of cerebral palsy emerge over time following a lesion to the developing brain: historically this led to delays in the use of the term CP, though earlier use of the term in those identified as being at high risk of CP has been recommended (Novak et al., 2017).

In recent years, there has been a rise in the understanding of the impact of a diagnosis of CP in infants on caregivers (Ribeiro, Vandenberghe, Prudente, Vila, & Porto, 2016). Poor health outcomes in infants adversely affect health outcomes in their caregivers (Bella, Garcia, & Spadari-Bratfisch, 2011; Byrne, Hurley, Daly, & Cunningham, 2010; Levallois, 2015; Raina et al., 2005; Stetz & Brown, 2004; Vicentic et al., 2016). More specifically, caregivers report facing a heavy burden of care, significant time pressures and behavioural problems in their children, with adverse effects on caregiver psychological wellbeing (Sawyer et al., 2011; Whittingham, Sanders, McKinlay, & Boyd, 2013).

Caregiving has been proven to have adverse physical and psychological health effects such as stress (Parkes, Caravale, Marcelli, Franco, & Colver, 2011), depression (Sawyer et al., 2011), anxiety (Benutto, 2015) and physical health impacts (Gallagher, Phillips, Drayson, & Carroll, 2009). These are reported especially when there is a lack of support and with more demanding roles (Ribeiro et al., 2016). To combat such problems, there has been an increase in the development and implementation of interventions to increase caregiver wellbeing (Heller, Gibbons, & Fisher, 2015; Lindo, Kliemann, Combes, & Frank, 2016; Magana, Li, Miranda, & de Sayu, 2015; Rieger & McGrail, 2013; Riley & Rubarth, 2015). However, there is no standardised provision of these interventions and no agreed understanding of the most effective method of support. Consequently, there are gaps where little or even no support is provided.

The diagnosis time-period can be very confusing. Due to the variability in motor outcomes and likely comorbidity with other health complications, there is often a lack of understanding of future impacts and implications at diagnosis. During this time, caregivers begin to experience and struggle with the challenges CP poses, (Ribeiro et al., 2016), reporting greater stress and scarcity of resources than parents of older children with CP (Ribeiro et al., 2016). Furthermore, CP poses unique demands with high levels of perceived burden, stress and time pressure. Caregivers of those with CP report facing frequent challenges with sleeping, eating, communication, behaviour and health complications (Boucaut, Lang, Guppy, & Johnston, 2014; Bunning, Gona, Newton, & Hartley, 2014; Hadden & von Baeyer, 2002; Iversen, Graue, & Raheim, 2013; Whittingham, Wee, Sanders, & Boyd, 2011; Wijesinghe, Cunningham, Fonseka, Hewage, & Ostbye, 2015).

Additionally, the literature reports how a diagnosis of disability is often accompanied by the experience of grief (Eakes, Burke, & Hainsworth, 1998; Hobdell, 2004; Marwit & Kaye, 2006). The experiences of parents of children with CP can be explained through chronic sorrow theory, describing long-term sadness experienced due to the continual losses that come with a disability diagnosis (Lindgren, Burke, Hainsworth, & Eakes, 1992). It has been recommended that health practitioners be aware of these lasting effects which are experienced for several years after diagnosis (Whittingham, Wee, Sanders, & Boyd, 2013). A relationship between chronic sorrow and experiential avoidance, parental psychological symptoms and experienced parenting burden highlights the long-term impact of accepting a child's disability (Dahl, Tervo, & Symons, 2007).

The adverse effects of the pressures of caregiving on caregiver wellbeing have the potential to impact the outcome and wellbeing of the child (Lach et al., 2009). This has become more pertinent through the contemporary shift in the standardised care of CP towards home-based family interventions of rehabilitation. In recent years, there has been a move towards parent-delivered therapy aiming to increase recovery through early and intensive interventions (Basu, Pearse, Baggaley, Watson, & Rapley, 2017; Dirks & Hadders-Algra, 2011; Hinojosa & Anderson, 1991). For caregivers to deliver such interventions effectively, their own mental wellbeing must be supported (Lord, Rapley, Marcroft, Pearse, & Basu, 2018). This literature highlights the importance of the provision of effective support for caregivers for their own benefit and for the benefit of the infants.

In this systematic review, we aimed to identify and evaluate existing interventions for improving caregiver wellbeing, assessing their effectiveness. We also aimed to identify gaps in the literature through evidence mapping.

2. Methods

2.1. Protocol

The methods undertaken for this systematic review were outlined in our protocol which was prospectively registered with PROSPERO (ID: CRD42017082662).

2.2. Inclusion and exclusion criteria

2.2.1. Study dates

Publications identified in databases from January 1990 to December 2017 were explored. A scoping search of the literature found many outdated views on disability prior to 1990: this informed our choice of start date.

2.2.2. Participants

We initially aimed to restrict the studies in this systematic review to include the parents and caregivers of infants aged up to 2 years with CP, early acquired brain injury or hypoxic ischaemic encephalopathy, which are often antecedents of CP. As only one study met these criteria (Fig. 2) we increased the age range to include interventions for parents of older children and expanded the disability criteria to include other caregiver interventions nonexclusive to CP that include CP in the study population.

2.2.3. Types of interventions

Initially we included studies assessing interventions with the primary aim to improve the psychological wellbeing of the caregivers of infants and children with CP; however, this was expanded to include studies where this was the secondary aim.

2.2.4. Comparators

Eligible evidence included randomised controlled trials (RCT) where an intervention was compared with either no support (i.e. waiting list) or an alternative method of support (e.g. care as usual).

2.2.5. Outcomes

Outcomes eligible included those related to caregiver psychological wellbeing, e.g. anxiety, stress, depression.

2.2.6. Study design

Studies included were RCTs. If this search proved too limited, we planned to consider inclusion of other study designs.

2.2.7. Setting

We considered all study settings.

2.2.8. Language

We restricted the inclusion of studies to those published or translated into English due to limited resources for translation of studies published in other languages.

2.3. Study identification

We identified studies through electronic searches of online databases. Support was received from an information specialist to develop scoping searches used to develop the search terms (Table 1) for our search strategy. Electronic searches were conducted on MEDLINE, EMBASE, PsycINFO, PubMed, Scopus, Web of Science and CENTRAL Cochrane Library database and were last searched 5/3/18.

Once the initial electronic searches were complete we explored the grey literature using websites (such as Google) and grey literature databases (such as opengrey.eu; greylit.org). Following the identification and confirmation of the included studies, a citation search was carried out on the studies found, searching through the citations and references of these to identify further potentially eligible studies.

2.4. Study selection

Papers were screened independently by LI and CJ for eligibility. The inclusion and exclusion were assessed using the eligibility criteria on the title, abstract and further relevant sections, evaluating the full text when necessary. Any differences in opinion were resolved by AB to remove any bias in inclusion evaluation.

2.5. Data extraction

Data extraction was undertaken by LI and CJ using a standardised proforma based upon the Cochrane data extraction form,

Table 1
Study Identification Search Strategy.

Keywords	Alternatives
Parents	Parent OR carer OR caregiver OR mother OR father OR guardian
Support	Intervention OR therapy OR treatment OR psychological support OR psychological therapy OR caregiver support OR social support
Infant	Baby OR child OR paediatric OR early acquired OR paediatric OR toddler
Cerebral palsy	CP OR physical disability OR disability OR neurodevelopmental disorder OR brain injury OR prenatal stroke OR early acquired stroke OR early acquired brain injury OR HIE OR hypoxic ischaemic encephalopathy
Outcome	Effectiveness OR evaluation OR measurement OR outcome
Wellbeing	Coping OR stress OR time management OR burden OR acceptance OR health OR depression OR anxiety OR struggle
Additional terms	Diagnosis, parental support

Search terms were used in conjunction with 'OR' and 'AND' search functions.

prepared and informally piloted beforehand to ensure all relevant information was captured and documented through this process. The first two articles were extracted by both authors and then cross-checked for any major differences in style and type of data extracted. There was good agreement between data and the decision was made to split the papers between the authors for data extraction.

2.6. Evidence mapping

Evidence mapping can be used to identify gaps in the literature to guide future research. When formulating our evidence map we used our inclusion criteria as a template. We filtered and coded studies found through our search strategies in relation to the nature of the disability, age of children, type of intervention and level of evidence. From this we generated a pictorial evidence map to illustrate the focus of available literature.

2.7. Quality assessment

Quality assessment was undertaken using the Critical Appraisal Skills Programme (CASP) checklist for systematic reviews, available at https://casp-uk.net/casp-tools-checklists/. All studies were rated independently by LI and CJ for questions assessing the risk of bias, validity, results and applicability of the studies. Any discrepancies in marking were resolved through discussion and involvement of a third author AB.. Studies that scored below 7 on the checklist were excluded from the meta-analysis.

2.8. Risk of bias

We assessed any potential risk of bias using the Cochrane Risk of Bias tool (Higgins & Green, 2011) to examine studies for randomised sequence generation method, allocation concealment, methods of addressing incomplete outcome data, blinding of outcome assessments, potential selective reporting, and any other possible bias which could affect trial outcome. We acknowledged that blinding of participants was not possible for this type of intervention. Risk of bias across studies occurs when null or negative results are less likely to be published. An effort was made to identify all published and unpublished studies which met eligibility criteria.

2.9. Meta-analysis

Meta-analyses were conducted to determine the efficacy of improving parental well-being immediately post treatment. Where insufficient data were provided, the authors were contacted to request further information. As the studies did not consistently use a common scale to measure parental wellbeing and mental health, we used the standardised mean difference (SMD) to allow comparison of outcomes.

Where three-armed RCTs including two intervention arms and one common control were involved, we used the Cochrane recommended method of combining the intervention groups to find an overall effect size in comparison to the control group. This was appropriate, as our key aim was to distinguish if interventions improved parental wellbeing, Statistical analyses were conducted using the computer software program RevMan using the inverse variance random-effects model, as we expected some heterogeneity between the data and studies were often estimating different, yet related, intervention effects (Higgins & Green, 2011; Rosenthal, 1995). Statistical tests of heterogeneity were used including chi-squared tests and I^2 statistics. The impact of reporting bias was explored using funnel plots and interpretation of any asymmetry.

We re-expressed our results from the meta-analysis back into a common outcome measurement scale to highlight the clinical impact and relevance of the SMD scores obtained (Schünemann et al., 2011). We used the Depression, Anxiety and Stress Scale (DASS) as it was the most commonly used scale across the papers we selected. To do this we required the standard deviation (SD) of scores obtained on each component of the DASS outcome for the total population group we were investigating, parents of children with cerebral palsy. This was obtained by pooling the SDs of baseline scores in both control and intervention groups for total DASS, DASS-Depression, DASS-Stress and DASS-Anxiety using the papers that had DASS as an outcome measurement. The SDs obtained were similar to the normative data and SDs available for the DASS outcome measurement from a non-clinical sample of 1771 UK adults (Crawford & Henry, 2003). By multiplying our SMD results by the SDs calculated we yielded an estimate of the pooled effects shown by intervention and control groups expressed in the format of the DASS outcome measurement, including changes in DASS total score, DASS-Depression, DASS-Stress and DASS-Anxiety.

3. Results

3.1. Study selection

Fig. 1 shows the PRISMA flow diagram for the review. The search strategy for the selected online databases yielded 4557 studies. We found 104 study titles to be potentially relevant, 13 of which were eligible for inclusion after full inspection and 7 were eligible for meta-analysis regarding parental mental wellbeing. A full description of each of the final studies and their characteristics can be found in Table 2, alongside Table 3 which summarises the parental outcome measures used.

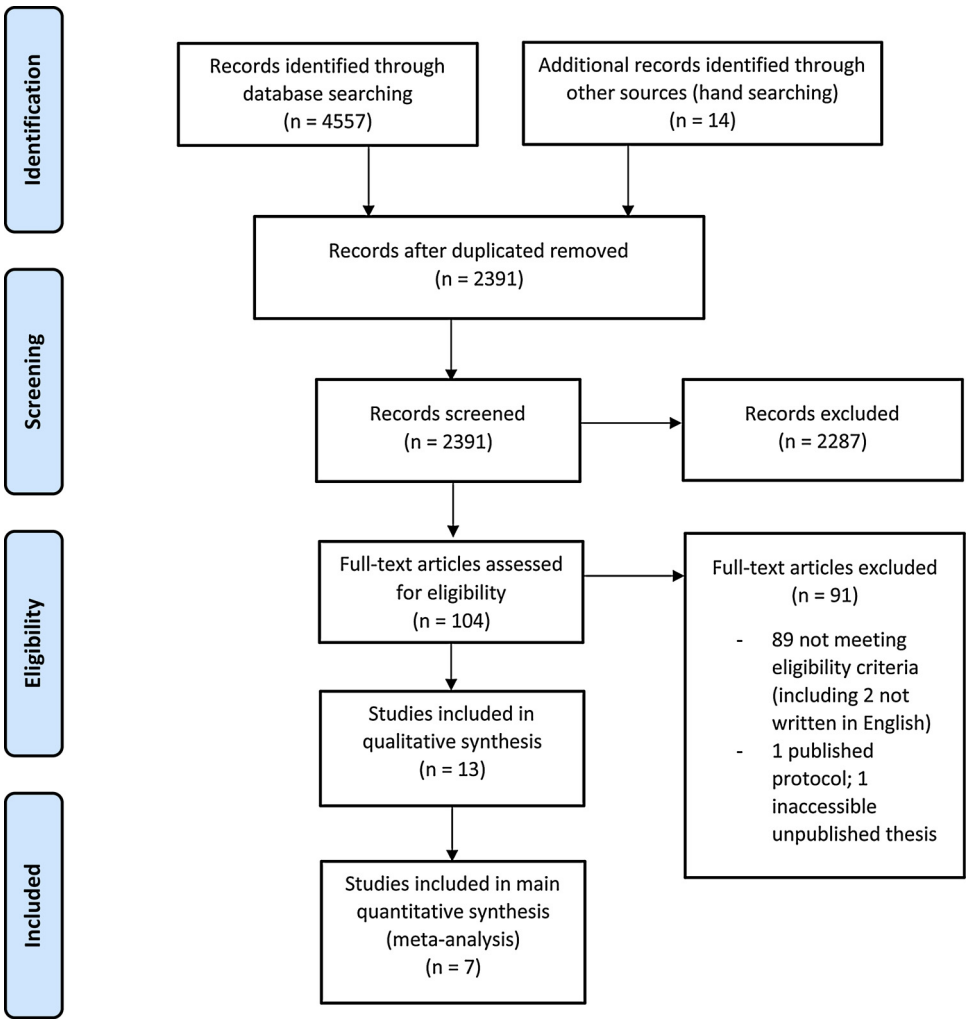

Fig. 1. PRISMA Flow Diagram of Study Selection.

3.2. Evidence mapping

Fig. 2 shows the evidence map of the 104 potentially relevant titles for which full text versions were screened for eligibility. We found only one RCT evaluating the effectiveness of an intervention addressing the wellbeing of parents of infants with CP.

3.3. Quality assessment and risk of bias analyses

Two studies (Gammon & Rose, 1991; Sofronoff, Jahnel, & Sanders, 2011) were excluded at the stage of the meta-analysis due to high risk of bias as examined by the CASP and Cochrane tool for assessing bias. As there were no baseline characteristic tables, measures of treatment effect or estimates of precision of the treatment effect, both papers scored low (< 7) on the CASP. Additionally they both included a very low or unspecified number of children with CP. The quality assessment of all 13 papers using the Cochrane tool is summarised in Fig. 3.

3.4. Study findings

Table 4 provides a qualitative summary of the individual study findings. Of the 13 studies, 6 (Brown, Whittingham, Boyd, McKinlay, & Sofronoff, 2015; Plant & Sanders, 2007; Roberts, Mazzucchelli, Studman, & Sanders, 2006; Roux, Sofronoff, & Sanders, 2013; Sofronoff et al., 2011; Whittingham, Sanders, McKinlay, & Boyd, 2016) investigated the effectiveness of Stepping Stones Triple P therapy (SSTP) (+/- an adjunct). SSTP is a behavioural family intervention. In some studies (Brown et al., 2015; Whittingham et al., 2016), SSTP was combined with Acceptance and Commitment Therapy (ACT), which is a form of cognitive-behavioural therapy aiming to foster psychological flexibility and reduce experiential avoidance. The combination of SSTP and ACT led to

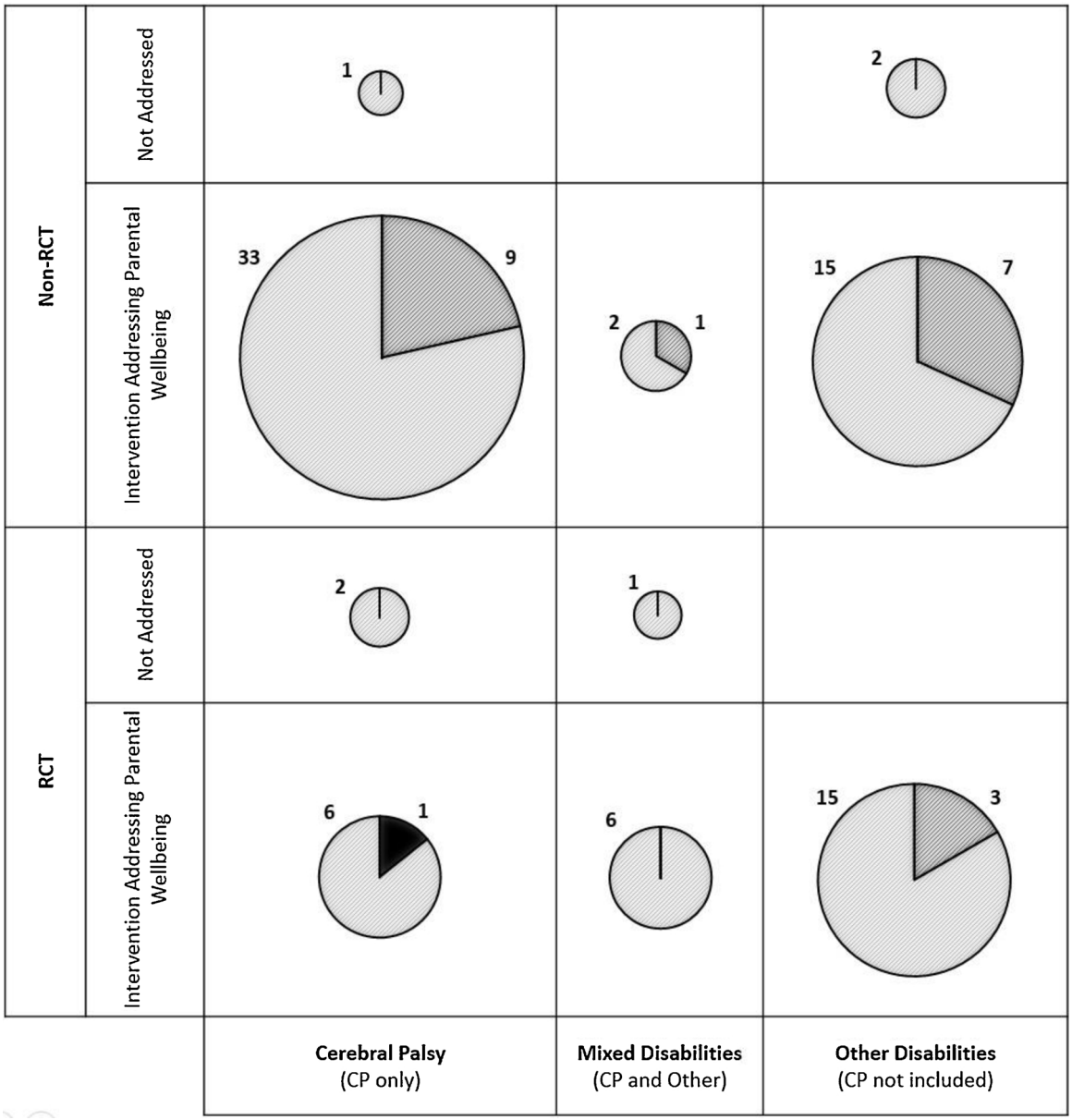

Fig. 2. Evidence Map Presenting Properties of the 104 full text articles reviewed.
Size-proportional charts in relation to the number of studies in each area, with darker shaded areas representing studies of infants (aged < 2y) and lighter shaded areas for older children. The area highlighted in black represents our initial targeted area of literature prior to scoping searches (RCTs of infants addressing wellbeing of parents of infants aged < 2y with CP).

improvements in parental psychological outcomes, however SSTP alone did not improve depressive symptoms except when used in a group setting (Roux et al., 2013). SSTP alone led to some improvement in relationship quality, conflict and parental competence (Plant & Sanders, 2007; Roberts et al., 2006; Roux et al., 2013; Sofronoff et al., 2011). In all studies recording child outcome measures, SSTP (+/- an adjunct) improved child behaviour or wellbeing (Plant & Sanders, 2007; Roberts et al., 2006; Roux et al., 2013; Sofronoff et al., 2011; Whittingham et al., 2016).

The other 7 studies investigated a wide range of interventions including an empowerment programme (Zare et al., 2017), a health-education intervention (Magana et al., 2015), life skills (Kirkham & Schilling, 1990), parent-to-parent support programmes (Singer et al., 1999), a cognitive-behavioural training programme (Gammon & Rose, 1991), involvement of a family support worker and physiotherapy assistant (Weindling, Cunningham, Glenn, Edwards, & Reeves, 2007) and a training and support programme including child massage skills (Barlow, Powell, Gilchrist, & Fotiadou, 2008).

Table 5 summarises the statistical findings for each outcome by intervention type; the interventions are shaded according to whether the primary outcome focused on the parent or child. No consistent pattern was evident in relation to intervention type and

Table 2
Study Characteristics.

Authors (year) Location	Objective	Method/ design (n = total)	Participants	Number of parents Number of CP children (% total children)	Intervention Delivery	Time points (Total intervention duration)	Outcomes assessed (measures used)
Brown et al. (2015) Australia	Evaluate efficacy of a behavioural family intervention, Stepping Stones Triple P (SSTP), combined with an Acceptance and Commitment Therapy (ACT) workshop in improving parent, family and couple outcomes following paediatric acquired brain injury (ABI).	RCT/ parallel-group, 2 arms: SSTP + ACT (n = 30) WLC (n = 29)	Parents of children with ABI aged 2-12y, at least three months post-injury/ and demonstrating at least one parent-reported emotional or behavioural difficulty.	n = 59 CP: n = 4 (2.36%)	SSTP programme: 6*2 -h group sessions + 3*30-minute individual telephone sessions. ACT workshop: 2*2 -h group sessions.	**T1:** 0 weeks/ pre-intervention **T2:** 10 weeks/ post-intervention **T3:** 6-month follow-up (10 weeks)	**Parent:** Family and parent adjustment (DASS, FAD), Parenting confidence (PTC), Inter-parental relationship (PPC, RQI), Treatment process (AAABIQ, PTFQ).
Whittingham et al. (2016) Australia	Examine effects of SSTP and ACT on child functioning, quality of life, and parental adjustment (secondary outcomes).	RCT/ parallel-group, 3-arms: SSTP (n = 22) SSTP + ACT (n = 23) WLC (n = 22)	Parents/carers of children aged 2–12 years, with CP who believed they would benefit from a parenting intervention.	n = 67 CP: n = 67 (100%)	SSTP programme: 6*2 -h group sessions + 3*30-minute individual telephone sessions. ACT: 2*2 -h group sessions. Eleven families received the intervention as an outreach weekend workshop.	**T1:** 0 weeks/ pre-intervention **T2:** 10 weeks/ post-intervention **T3:** 6-month follow-up (10 weeks)	**Parent:** Parental adjustment (DASS), Parental confidence (CPDPTC). **Child:** Child functional performance (PEDI), Child quality of life (CP QOL-child).
Roberts et al. (2006) Australia	Evaluate effects of SSTP on child behaviour problems and family outcomes for mothers and fathers such as parenting style, parental stress, and treatment satisfaction.	RCT/ parallel-group, 2 arms: SSTP (n = 24 families), 29 parents WLC (n = 23 families), 26 parents	Parents of preschool children with development and behaviour problems registered with the Western Australian Disability Services Commission.	n = 55 (47 families) CP: n = 5 (9.80%)	Standard SSTP delivered in 10 sessions; clinic sessions lasted 2 hours, home visits (3-4/family) lasted 1 hour. Families with additional needs participated in 1-2 enhanced triple P modules 'partner support' and 'coping skills'.	**T1:** 0 weeks/ pre-intervention **T2:** 16 weeks/ post-intervention **T3:** 6-month follow-up (16 weeks)	**Parent:** Parental stress (DASS), Parental behaviour (PS, FOS-RIII), Intervention acceptability (CSQ). **Child:** Child behaviour (DBC-parent, FOS-RIII).
Plant et al. (2005) Australia	Evaluate effectiveness of Stepping Stones Triple P-Enhanced (SSTP-E) versus Stepping Stones Triple P-Standard (SSTP-S) on child behaviour outcomes and parenting outcomes such as parenting skills and ability, and parental adjustment.	RCT/ Parallel-group, 3 arms: SSTP-S (n = 52) SSTP-E (n = 48) WLC (n = 48)	Parents of preschool children (< 6 years) with developmental disability, who rated their child's behaviour as being in the elevated range on the Eyberg Child Behaviour Inventory (ECBI).	n = 142 CP: n = 5 (6.8%)	SSTP-S: 10 individual sessions (intensive behavioural parent training) with a practitioner (60–90 min/week). SSTP-E: as per SSTP-S plus six SSTP-E sessions on caregiver coping strategies (60-90 min each).	**T1:** 0 weeks/ pre-intervention **T2:** 10-16 weeks/ post-intervention **T3:** 1-year follow-up (10/16 weeks)	**Parent:** Parental adjustment (DASS), Parent-child interaction (FOS-RIII), Parenting skills and ability (PS), Parenting competence (PSOC), Relationship satisfaction (ADAS), Intervention acceptability (CSQ). **Child:** Parent-child interaction (FOS-RIII), Child behaviour (ECBI, DBC-Parent, CPC).
Roux et al. (2013) Australia	Evaluate parent-perceived efficacy of the Group SSTP Program for families of children in a mixed-disability group on child behaviour and parenting style.	RCT/ Parallel-group, 2 arms: SSTP (28 = families) WLC (27 = families)	Parents/carers of children aged 2-9 with ASD, Down Syndrome, CP or intellectual disability.	n = 52 families CP: n = 3 (11.11%)	Nine sessions: six group sessions each 2-2½ hours and three individual telephone sessions for 15– 30 min.	**T1:** 0 weeks/pre-intervention **T2:** 9 weeks/ post-intervention **T3:** 6-month follow-up (9 weeks)	**Parent:** Parental wellbeing (DASS), Parent behaviour (PS), Inter-parental relationship (PPC, RQI), Goal attainment (GAQ), Intervention acceptability (CSQ). **Child:** Child behaviour (ECBI, DBC-Parent).
	Evaluate the efficacy of SSTP seminars for parents of a child	RCT/Parallel-group, 2 arms.	Parents of a child with a disability.	n = 70 CP: (3.8%)	SSTP programme: 2*2 -h group seminars attended	**T1:** 0 weeks/pre-intervention	**Parent:** Parental adjustment and family functioning (FAD, DASS),

(*continued on next page*)

Table 2 (*continued*)

Authors (year) Location	Objective	Method/ design (n = total)	Participants	Number of parents Number of CP children (% total children)	Intervention Delivery	Time points (Total intervention duration)	Outcomes assessed (measures used)
Sofronoff et al. (2011) Australia	with a disability on child behavior and parenting styles, satisfaction and efficacy.	SSTP (n = 35) WLC (n = 35)			(90 minute presentation +30 minute questions).	**T2:** 6 weeks/ post-intervention **T3:** 3-month follow-up (6 weeks)	Parent behaviour (PS), Parenting confidence (PSOC), Inter-parental relationship (PPC, RQI). **Child:** Child behaviour (ECBI).
Magana et al. (2015) USA	Test efficacy of an 8-week manualised health education intervention at increasing health related self-efficacy, healthy behaviours and reducing depressive symptoms and care-giver burden.	RCT/ Parallel-group, 2 arms: Intervention (n = 50) WLC (n = 50)	Mothers of a child diagnosed with intellectual and developmental disability and of Latin American descent, in their midlife or older.	n = 100 CP: n = 25 (36.76%)	Eight sessions each 1-2 hours, delivered at participant's home.	**T1:** 0 weeks/ pre-intervention **T2:** 8 weeks/ post-intervention (8 weeks)	**Parent:** Depressive symptoms (CED-D), Health-related self-efficacy (CDSES-adapted), Positive health behaviours (FHS-adaptation), Caregiver burden (CAM-adapted).
Zare et al. (2017) Iran	Compare effect of intervention program based on self-management empowerment model (vs. no intervention) on anger and social isolation of mothers with children with CP.	RCT/ Parallel-group, 2 arms: Intervention (n = 36) Control (n = 36)	Mothers, aged 20-45 y, with a child aged 2-10 y with moderate or severe CP.	n = 72 CP: n = 64 (89%)	Two 2 -h sessions covering 5 key steps. Goal-setting and planning were delivered individually, other steps were group-based.	**T1:** 0 weeks/ pre-intervention **T2:** 6 week follow-up (6 weeks)	**Parent:** Loneliness (UCLA Loneliness Scale), Aggression (BP-AQ).
Barlow et al. (2008) U.K.	Determine effectiveness of 'Training and Support Programme' (TSP) for parents of children with disability.	RCT/ Parallel-group, 2 arms: TSP with basic massage skills (n = 98) WLC (n = 46)	Main carers of children (< 16y) with disability.	n = 188 CP: n = 88 (47%)	Eight weekly sessions, each 1 hour. Individual sessions with therapist.	**T1:** 0 weeks/ pre-intervention **T2:** 4-month follow-up (8 weeks)	**Parent:** Parental wellbeing (HADS, PSS10), Self-efficacy (GSES, PSES), Life satisfaction (SWLS). **Child:** Child Behaviour (VAS).
Gammon et al. (1991) USA	Determine the effectiveness of a 'Coping Skills Training Programme' (CSTP), a cognitive-behaviour training package, in helping parents deal with the problems they face raising children with development disabilities.	RCT/Parallel-group, 2 arms: CSTP (n = 24) WLC (n = 18)	Parents of children with known disability (aware of disability for more than 6-months) and that have verbally indicated experiencing difficulties related to being a parent of a child with a disability.	n = 42 Children with CP were included but number with CP not specified.	Group setting with 4-8 parents. Each session 2 hours and delivered over 10 weeks by social worker (senior author of the programme).	**T1:** 0 weeks/ pre-intervention **T2:** 10-weeks/ post-intervention (10 weeks)	**Parent:** Parental stress (POMS, QRS), Problem Solving (PSI*), Interpersonal communication skills (COMTS), Positive Self-Regard (PSRTS), Goal attainment (Individualised Goal Scaling).
Kirkham et al. (1990) USA	Compare effects of a skills-building intervention aimed at improving coping and communication vs. a traditional parent support group for mothers of disabled children.	RCT/ Parallel group, 2-arms: Skills building group (n = 143) Traditional support group	Mothers of children with a variety of developmental disabilities, aged 2-14y.	n = 230 CP: n = 23 (10%)	Skills building intervention: nine sessions in total, each 2 hours, delivered twice a week in a group format. Traditional support group: intensity/frequency not specified.	**T1:** 0 weeks/ pre-intervention **T2:** 5 weeks/ post-intervention (5 weeks)	**Parent:** Parental wellbeing (QRS, BDI), Self-reinforcement (SRA), life satisfaction (IPE), Communication skills (written answers to vignettes).
Singer et al. (1999) USA	Multi-site evaluation of parent-to-parent programmes to determine effect on problem-focused coping, and cognitive and emotional adaptation.	RCT/ Parallel group, 2 arms: Parent to parent programs WLC	Parents, foster parents, or grandparents of children with a disability or chronic health condition.	n = 128 Children with CP were included but number with CP not specified.	Support conducted via telephone. Supporting parents were asked to make four calls to the help-seeking parent over a 2-month period.	**T1:** 0 weeks/ pre-intervention **T2:** 8 weeks/ post-intervention (8 weeks)	**Parent:** Cognitive adaptation (KIPP – strength and family closeness subscale), Empowerment (FES), Coping efficacy (PCES), Progress on presenting problem (Likert scale

(*continued on next page*)

Table 2 (*continued*)

Authors (year) Location	Objective	Method/ design (n = total)	Participants	Number of parents Number of CP children (% total children)	Intervention Delivery	Time points (Total intervention duration)	Outcomes assessed (measures used)
							questionnaire), Perceived helpfulness (Likert scale questionnaire). Qualitative interview.
Weindling et al. (2007) U.K.	Investigate additional support by (a) a physiotherapy assistant (PA) improved physical function in young children with spastic CP and (b) a family support worker (FSW) improved family functioning.	RCT/ Parallel group, 3 arms: FSW group (n = 31) PA group (n = 28) Care as usual group (n = 29)	Primary carers of children, aged < 4y, with predominantly spastic CP	n = 88 families CP: n = 88	In addition to their regular physiotherapy, each week the PA group received 1 hour of extra physiotherapy, and the FSWG group received a 1 hour home visit from a family support worker.	**T1:** 0 weeks/ pre-intervention **T2:** 6-months/ post-intervention **T3:** 12-month post-intervention follow-up (24 weeks)	**Parent:** Stress (PSI), Family identified needs (FNS), Intervention acceptability (Interview). **Child:** Gross motor ability (GMFM), Developmental status (GMDS), Adaptive functioning (VABS – daily living and socialisation domains).

Legend: IG - Intervention Group, CG - Control Group, WLC - Wait List Control, RCT – Randomised Control Trial. AAABIQ - Acceptance and Action for Acquired Brain Injury Questionnaire, ADAS – Abbreviated Dyadic Adjustment Scale, BDI – Beck Depression Inventory, BP—AQ – Buss-Perry Aggression Questionnaire, CAM – Caregiver Appraisal Measure, CDSES - Chronic Disease Self-Efficacy Scale, CED-D – Centre for Epidemiologic Studies Depression Scale, CPC – Care-giving Problem Checklist, CPDPTC – Cerebral Palsy Daily Parenting Tasks Checklist, CP QOL-Child – Cerebral Palsy Quality of Life Child, COMTS – Communication Skills Total Score, CSQ – Client Satisfaction Questionnaire, DASS – Depression Anxiety Stress Scale, DBC – Developmental Behaviour Checklist (Parent Version), FAD – Family Assessment Device, FES - Family Empowerment Scale, FHS - Family Habits Scale, FNS – Family Needs Scale, FOS-RIII – Family Observation Schedule–Revised III, GAQ – Goal Attainment Questionnaire, GMDS – Griffiths Mental Development Scales, GMFM – Gross Motor Function Measure, GSES – Generalised Self-Efficacy Scale, HADS – Hospital Anxiety and Depression Scale, IPE – Inventory of Parent Experiences, KIPP – Kansas Inventory of Parental Perceptions, PCES – Parent Coping Efficacy Scale, PEDI – Paediatric Evaluation of Disability Inventory, PPC – Parent Problem Checklist, PS – Parenting Scale, PSES – Parent's Self-Efficacy Scale, PSI – Parenting Stress Index, PSI* - Problem Solving Index, PSOC – Parenting Sense of Competence Scale, PSRTS – Positive Self-Regard Total Score, PSS10 – Perceived Stress Scale, PTC – Parenting Tasks Checklist, PTFQ – Parent Thoughts and Feelings Questionnaire, POMS - Profile of Mood States, QRS – Questionnaire on Resources and Stress, RQI – Relationship Quality Index, SRA – Self-Reinforcement Attitudes Questionnaire, SWLS – Satisfaction with Life Scale, UCLA Loneliness Scale - University California, Los Angeles Loneliness Scale, VABS – Vineland Adaptive Behaviour Scales, VAS – Visual Analogue Scale.

Table 3
Parental Outcome Measures.

Outcome	Measures
Aggression	BP-AQ
Anxiety	DASS, HADS
Stress	DASS, POMS, PSI, PSS10, QRS
Depression	BDI, CED-D, DASS, HADS
Behaviour (Parent)	PS, FOS-RIII
Caregiver Burden	CAM-adapted
Conflict	COMTS, PPC
Coping	PCES, PCES, SRA
Family Functioning	FAD, FNS
Loneliness	UCLA-Loneliness Scale
Physical Health	CDSES-adapted, FHS-adapted
Psychological Flexibility	AAABIQ, KIPP, PFTQ, PSRTS
Relationship	ADAS, PPC, RQI
Satisfaction	IPE, SWLS
Self-efficacy	CPDPTC, FES, GAQ, Individualised Goal Scaling, GSES, PSES, PSI*, PSOC, PTC

For abbreviations see Table 2 legend.

parental outcome effects. However, interventions where the primary outcome focused on the child (SSTP +/- adjunct) showed consistent improvements in child related outcomes.

3.5. Meta-analysis

Four papers were excluded from the meta-analysis: in one of the studies it was not possible to obtain the required data regarding the SD of the baseline scores (Barlow et al., 2008); in another, there was no corresponding outcome measure that could be included in meta-analysis (Singer et al., 1999); and two of the papers (Gammon & Rose, 1991; Sofronoff et al., 2011) were excluded due to the high risk of bias. For the remaining papers, we analysed data in relation to four categories of outcome: parental wellbeing, depression, anxiety and stress.

3.5.1. Parental wellbeing

Outcomes related to parental wellbeing included any that addressed either the subjective or objective total measures of improvements in depression, aggressive behaviour, stress or anxiety, self-actualisation and contentment/happiness. Seven studies could be analysed (n = 622; 369 in the intervention group (IG) and 253 in the control group (CG)). These used the DASS-T (Brown et al., 2015; Plant & Sanders, 2007; Roux et al., 2013; Whittingham et al., 2016), Beck Depression Inventory (BDI)(Kirkham & Schilling, 1990), Centre for Epidemiologic Studies Depression Scale (CED-D)(Magana et al., 2015), Buss-Perry Aggression Questionnaire (BP-AQ) (Zare et al., 2017). Fig. 4 highlights the effect of intervention on wellbeing.

3.5.2. Depression

Four studies could be included (n = 424; IG = 255 and CG = 169) (Brown et al., 2015; Kirkham & Schilling, 1990; Magana et al., 2015; Whittingham et al., 2016). Fig. 5 highlights the effect of intervention specifically on depression.

3.5.3. Anxiety

For anxiety, only 2 studies could be included (n = 119; IG = 70 and CG = 49) (Brown et al., 2015; Whittingham et al., 2016) as these provided details of DASS anxiety scores. Fig. 6 highlights the effect of intervention specifically on anxiety.

3.5.4. Stress

For stress, 4 studies were analysed (n = 216; IG = 116 and CG = 100), using scores from the DASS-S (Brown et al., 2015; Roberts et al., 2006; Whittingham et al., 2016) and Parenting Stress Index (Weindling et al., 2007). Fig. 7 highlights the effect of intervention specifically on stress.

3.5.5. Re-expression of SMDs using the DASS

The DASS is a 42-item questionnaire; each item scores a value between 0-3. DASS scores are most commonly interpreted using the separate components of the scale rather than the total score (Lovibond & Lovibond, 1995). The 42 items are divided equally between the three subscales (Depression, Anxiety and Stress): for each subscale the maximum score (most severe clinical presentation) is 42. Whilst each condition represents a continuum, cutoff levels have been suggested representing normal, mild, moderate, severe and extremely severe presentations for each subscale.

Multiplication of the calculated SMDs for Depression, Anxiety and Stress with the among-person SD for the DASS scale for the included datasets (Higgins & Green, 2011) revealed the estimates for the difference in mean outcome scores on the DASS scales between intervention and control groups. For the DASS total score the mean difference was -10.53 (95%CI -15.9 to -5.18). For the

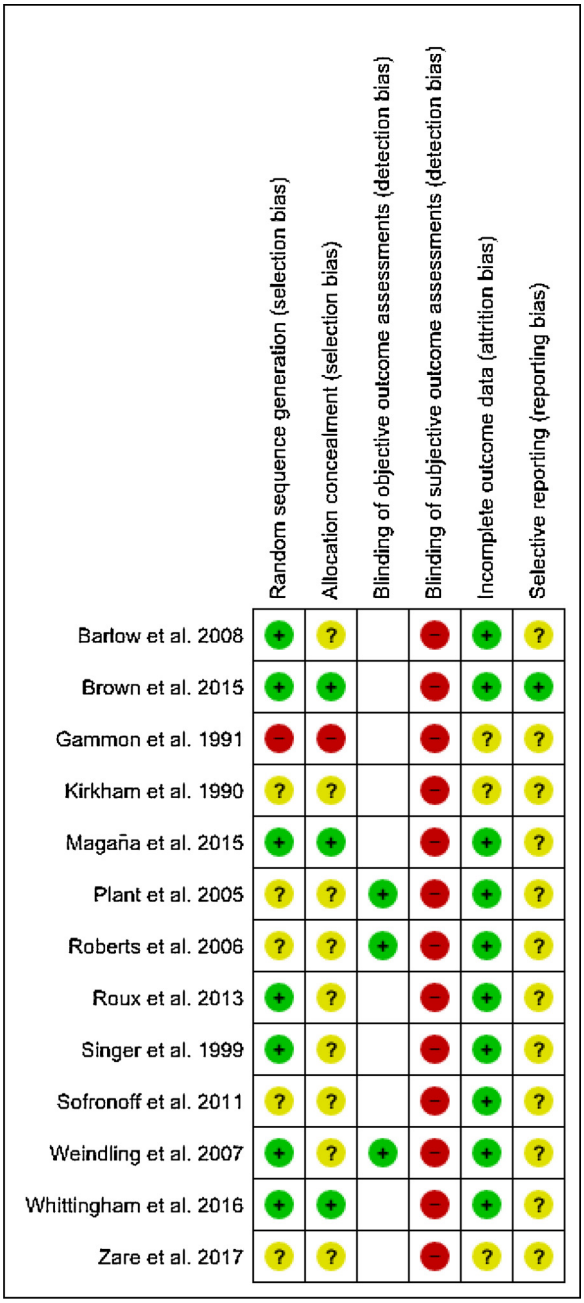

Fig. 3. Risk of Bias Summary.
Green "plus" sign: low risk of bias; red "minus" sign: high risk of bias; yellow "?" sign: risk could not be clearly ascertained.

Depression, Anxiety and Stress subscales these values were -1.96 (-3.74 to -0.18), -2.17 (-4.14 to -0.26), and -2.53 (-5.06 to 0.084) respectively. These represent relatively small changes in clinical status, given the information about the DASS summarized above.

3.6. Publication bias

We were unable to obtain access to two unpublished studies and as a result could not complete moderator analysis. A funnel plot was obtained for each of the meta-analyses conducted; visual inspection of these suggested the possibility of publication bias. We were unable to conduct tests to analyse possible funnel plot asymmetry due to the use of SMD in our meta-analysis (Schünemann et al., 2011) and the relatively small number of studies. Possible explanations of the visualised funnel plot asymmetry include selection bias, poor methodological quality and chance.

Table 4
Summary of Intervention Outcomes for Parents.

Study	Initial results	Follow-up results	Overall findings / conclusions
Brown et al. (2015)	Significant between-group changes in parenting confidence (PTC), family and parent adjustment (FAD and DASS anxiety, stress but not depression subscale), inter-parental relationship (PPC) and parenting psychological flexibility (AAABIQ and PTFQ).	Effects on psychological flexibility retained at 6 months.	ACT + SSTP produced short-term improvements in parenting confidence, psychological distress, family and parent adjustment and couple disagreements; and sustained improvement in parental psychological flexibility.
Whittingham et al. (2016)	SSTP alone showed no significant effect on parental measures in comparison to CG. SSTP + ACT group showed significant reductions in parental adjustment compared with CG (DASS depression and stress but no change in DASS anxiety) but not in parental confidence (CPDPTC).	Effects not retained at 6 months.	Parents receiving SSTP + ACT showed reductions in depressive symptoms and stress.
Roberts et al. (2006)	No significant effects for parental stress (DASS). There were significant effects in parental behaviour; IG fathers reduced verbose and lax disciplinary styles and mothers reduced over-reactivity.	No deteriorations in reported parental stress levels at 6 months	SSTP was associated with reductions in child behaviour problems. Parent behaviour changed but not as consistently as child behaviour, with differences between mothers and fathers.
Plant et al. (2005)	SSTP-S and SSTP-E (enhanced) showed comparable effects. Neither led to reduction in depressive symptoms. Both improved dysfunctional parenting styles and parenting sense of competence.	Effects retained at 1 year	SSTP-E was not superior to SSTP-S. Neither reduced depressive symptoms.
Roux et al. (2013)	Parenting Scale and Parent Problem Checklist: significant changes. No improvement in relationship quality. Significant improvement in DASS scores, however pre-intervention scores fell within the normal range.	Effects retained at 6 months	Group-SSTP is an effective intervention for parenting difficulties in mixed-disability groups. Significant effect on DASS despite normal baseline DASS scores.
Sofronoff et al. (2011)	IG showed significant reduction in child behaviour problems, but not in intensity of problem behaviours (ECBI). Significantly lower level of conflict (PPC). No significant improvement parental satisfaction, efficacy (PSOC) or parental and family adjustment (DASS, FAD).	At 3 months most significant effects maintained and new significant change in ECBI *Intensity* scale for intervention. Effect for PPC not maintained.	Participation in SSTP workshop seminars were effective in improving child behaviour, conflicts over child-rearing and reducing dysfunctional parenting styles, however had little effect on parental wellbeing scores.
Magana et al. (2015)	Both groups reported lower depressive symptoms and reduced caregiver burden. IG showed greater increases in health-related self-efficacy, self-care, nutrition and overall health-related behaviours.	No follow-up	Health education intervention program delivered by community health workers increased maternal health-related self-efficacy and positive health behaviours compared with providing program content manual alone.
Zare et al. (2017)	Significant difference in change in mean aggression scores; no improvement in isolation and loneliness.	No follow-up	Implementation of the empowerment program significantly reduced aggression.
Barlow et al. (2008)	Significant improvement in parental self-efficacy in IG vs CG, as well as reduction in depressed mood, both at 4 months	No follow-up	Training and Support Programme improved parental self-efficacy and depressed mood.
Gammon et al. (1991)	IG showed significant improvements in problem-solving skills, interpersonal skills (total score) and reported perceptions of goal achievement. No significant improvement for overall results related to positive self-regard and stress levels.	No follow-up	Coping Skills Training Programme (CSTP) delivered by social work group services useful for improving problem-solving abilities, achievement of Individualised goals and inter-personal skills. CSTP has little meaningful impact on stress experienced by parents and positive self-regard.
Kirkham et al. (1990)	IG reported significantly larger improvements in coping, communication, satisfaction with family support and near-significant reduction in depression and stress levels.	No follow-up	Life skills-building group sessions led to improved coping and communication skills and greater satisfaction with social support networks compared with traditional parent support group.
Singer et al. (1999)	IG showed improvements in Source of Strength and Family Closeness subscale of the KIPP, suggesting parental cognitive adaptation to disability. Improvements in Perceptions of Coping Efficacy in the IG were greater than in CG for participants with low baseline coping efficacy. IG showed greater perceived progress with goals.	No follow-up	Parent to Parent support helped parents feel better able to cope and to view family/ circumstances more positively, as well as making progress on goals.

(*continued on next page*)

Table 4 (*continued*)

Study	Initial results	Follow-up results	Overall findings / conclusions
Weindling et al. (2007)	No significant improvements in parental stress and level of family needs, though parental satisfaction with interventions was high.	Significant improvements in Family Needs Scale at 12 months in the group with family support worker (not seen at 6 months)	Input by a family support worker did not improve parental stress or level of family needs to a clinically significant level despite reaching statistical significance in the latter at 12 months.

For abbreviations see Table 2 legend.

4. Discussion

Our primary aim was to determine the efficacy of existing interventions for improving psychological wellbeing in the primary caregivers of children with cerebral palsy and identify gaps in the literature through the process of evidence mapping. This review summarised 13 studies indicating that psychological interventions may significantly reduce the levels of depression, stress and anxiety in parents of children with cerebral palsy. Evidence mapping highlighted that only a small proportion of RCTs address psychological interventions for parents of young children with cerebral palsy.

4.1. Population

4.1.1. Age

The lack of RCTs including young children with a diagnosis of CP likely reflects historical delays in making this diagnosis at an early age. The situation is changing, with earlier diagnosis now possible and greatly encouraged (Novak et al., 2017). Even with early diagnosis, prognosis (e.g. in terms of severity and effects on function) often remains uncertain for some time as clinical features of the condition gradually emerge. The time of diagnosis has been referred to as a 'crisis event' for parents in which their previous views of their child's future are disrupted (Dagenais et al., 2006; Sloper & Turner, 1993), and grief reactions similar to those of bereavement are experienced (Blacher, 1984; Cameron, Snowdon, & Orr, 1992). Whilst studies have identified these and other issues including the increased parental stress that can occur with poor communication around diagnosis (Dagenais et al., 2006; Schuengel et al., 2009), there does not appear to be a corresponding amount of literature on support strategies and interventions for parents at this time though there is recognition that support is needed (Hedderly, Baird, & McConachie, 2003; Rahi, Manaras, Tuomainen, & Hundt, 2004). It is important to direct future research in this area to ensure that clinicians are clearly and correctly advised on the best interventions to help parents of young children with cerebral palsy to cope during this difficult time.

4.1.2. Diagnosis

Evidence mapping highlighted literature exploring the efficacy of interventions for parents of children with neurodevelopmental disabilities, but only 4 out of the 13 studies identified for our review focused solely on parents of children with cerebral palsy. This is despite literature available showing that parents of children with cerebral palsy have high rates of stress, anxiety and depression (Basaran, Karadavut, Uneri, Balbaloglu, & Atasoy, 2013; Cheshire, Barlow, & Powell, 2010; Yilmaz, Erkin, & Nalbant, 2013). Given the paucity of literature specific to CP, we included studies covering a mixture of disabilities that also encompassed children with cerebral palsy.

4.1.3. Demographics

Parents in the included studies were predominantly female and white, with most studies occurring in Australia. Little is known about the psychological wellbeing of fathers of children with disabilities: effects on mothers have been researched to a greater extent. However, gender differences exist between emotions experienced by mothers and fathers of children with neurodevelopmental disorders. For example, among parents of children with autism, fathers experienced more anger and mothers more sadness (Gray, 2003). Other studies indicate that mothers of a child with a health problem feel stress more intensely and show a greater tendency to express their emotions than fathers, who tend to harbour and repress emotion (McGrath & Chesler, 2004; Pelchat, Lefebvre, & Levert, 2007). However, fathers are not immune to the challenges faced with raising a child with a disability; they report higher levels of parenting stress and have diminished psychological well-being (Simmerman, Blacher, & Baker, 2001). The relative lack of inclusion of fathers in the intervention studies may be because of their higher employment rates than mothers as demonstrated in the baseline demographic data of the individual trials where this is detailed (Plant & Sanders, 2007; Weindling et al., 2007), leaving less time for commitment to such studies, particularly if attendance at sessions would conflict with working hours on a regular basis. It would be helpful to obtain the views of fathers on the type of support they would value.

4.1.4. Baseline characteristics

Studies indicate high rates of depression and stress in parents of children with cerebral palsy (Basaran et al., 2013; Cheshire et al., 2010; Garip et al., 2017; Sajedi, Alizad, Malekkhosravi, Karimlou, & Vameghi, 2010). However, many of the parents in the current studies were not depressed, anxious or stressed prior to the intervention being conducted, as demonstrated by their baseline scores (Table S1).

Table 5
Overview of Statistical Findings for Outcomes by Intervention Type.

Approach	Educational							Educational and Psychological				Support from Parent Expert	
	Life Skills Training	Massage skills	Health education	SSTP				SSTP + ACT		Empower-ment	Cognitive-behavioural training	Support over the phone	Home visit
Outcomes	Kirkham et al. (1990)	Barlow et al. (2008)	Magana et al. (2015)	Roberts et al. (2006)	Plant et al. (2005)	Roux et al. (2013)	Sofronoff et al. (2011)	Brown et al. (2015)	Whittingham et al. (2016)	Zare et al. (2017)	Gammon et al. (1991)	Singer et al. (1999)	Weindling et al. (2007)
PARENT													
Aggression										+			
Anxiety		**0**			**0**	+	**0**	+	**0**				
Depression	**0**	+	**0**		**0**			**0**	+				
Stress	**0**	**0**		**0**	**0**			+	+		**0**		**0**
Behaviour (Parent)				+		+	+						
Caregiver Burden			**0**										
Conflict	+					+	+	+					
Coping	+							**0**				+	
Family Functioning							**0**	+					**0**
Loneliness										**0**			
Physical Health			+										
Psychological Flexibility								+			**0**	+	
Relationship	+				**0**	**0**	**0**	**0**					
Satisfaction	+	**0**											
Self-efficacy		+			+	+	**0**	+	**0**		+	+	
CHILD													
Behaviour		**0**		+	+	+	+						
Communication		**0**											
Development													**0**
Motor Function									+				**0**
Wellbeing									+				

[0] : No significant effect (neutral).
+ : Statistically significant ($p < 0.05$) positive effect (benefit) on measured outcome, i.e. improved coping or reduced depression, anxiety etc.
- : Statistically significant ($p < 0.05$) negative effect (detriment) on measured outcome, i.e. increased stress, anger etc.
Shaded column = primary outcomes focussed on parent.
Unshaded column = primary outcomes focussed on child.
Column with Both Shaded and Unshaded cells = primary outcomes focussed on both parent and child.

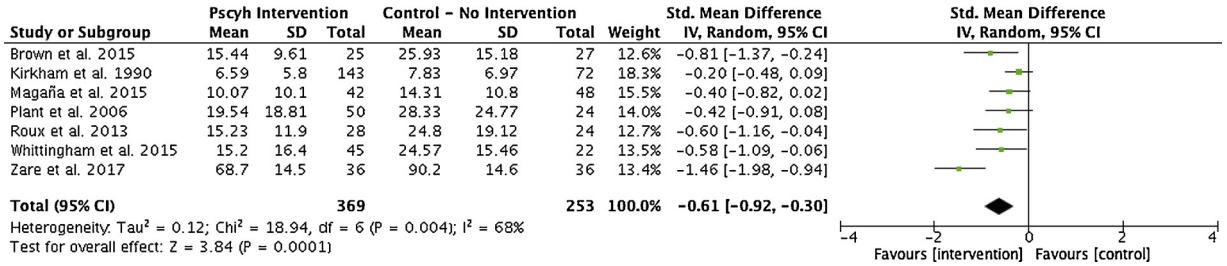

Fig. 4. 'Wellbeing' Meta-Analysis - Intervention vs. Control.

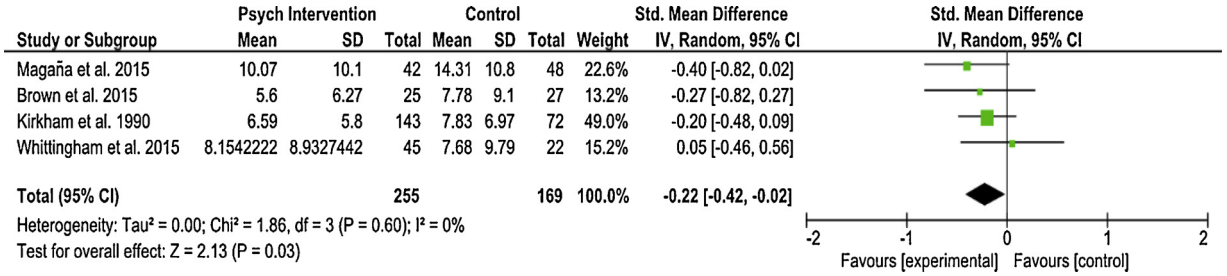

Fig. 5. Depression Subgroup Meta-Analysis - Intervention vs. Control.

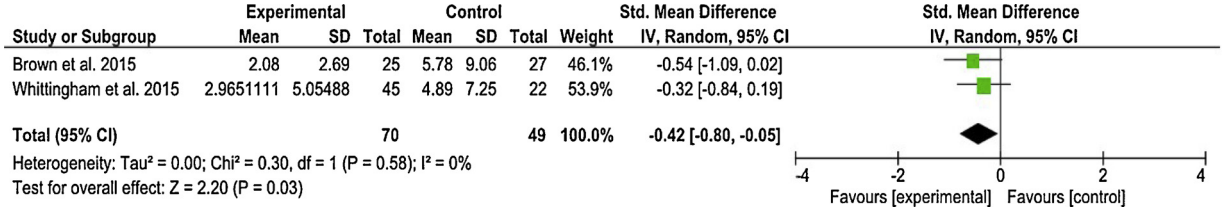

Fig. 6. Anxiety Subgroup Meta-Analysis - Intervention vs. Control.

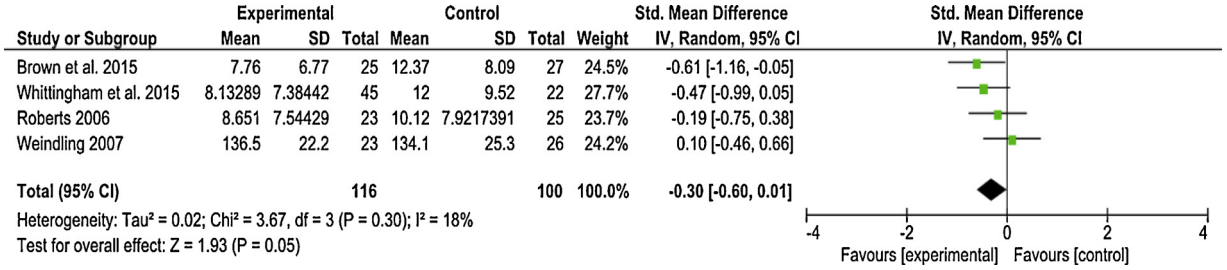

Fig. 7. Stress Subgroup Meta-Analysis - Intervention vs. Control.

4.2. Interventions

Whilst all the interventions purported to influence parental wellbeing, for some of these this was a secondary aim, achieved through indirect means such as improvements in the child's behavior (Brown et al., 2015; Plant & Sanders, 2007; Roberts et al., 2006; Roux et al., 2013; Sofronoff et al., 2011; Whittingham et al., 2016). Thus, the inclusion criteria were focused around the child, not the parental mental health needs. The effects on parental mental wellbeing remain important and can be considered as preventative; however, they may not translate into relevant benefits for parents with greater baseline mental health needs. It would be beneficial to have more data on the effects of interventions with parental mental wellbeing as a primary outcome.

Collectively, psychological interventions improved parental mental wellbeing; however there was no consistent pattern in relation to the type of intervention and its approach, and the observed parental outcomes. The only evident pattern was that SSTP (which had the primary aim of improving child behaviour), consistently benefitted child behaviour outcomes specifically. Interventions that focused primarily on the parent did not appear to consistently show a pattern of improved parental outcomes, such as mental wellbeing.

4.3. Comparator groups

Comparator groups were in most cases care as usual or wait-list controls: this precluded blinding of the participants. Inability to blind participants to allocation is a common problem in trials of complex interventions. Furthermore, control interventions requiring a large time commitment from the parents and family would be unethical. An alternative approach would be to use large-scale observational studies rather than RCTs to trial interventions aimed at psychological wellbeing (Palermo, 2014; Morley, Williams & Eccleston 2014). At present it is unclear whether this would be a more beneficial approach than continuing to use unblinded RCTs.

4.4. Outcomes

There was a marked lack of standardization of the assessments used to measure parental mental wellbeing outcomes. We attempted to correct for this using standardized mean differences, mapping these on to the DASS score and subscales to understand the clinical relevance of any changes. In future, standardization of outcome measures used in such trials will likely be achieved through the Core Outcome Measures in Effectiveness Trials (COMET) initiative http://www.comet-initiative.org/ (Williamson & Clarke, 2012).

4.5. Study size and number of studies

Key limitations include the size of individual trials, in conjunction with the small number of studies in this field. We were also unable to include studies not written in the English language. Furthermore, we were unable to access the level of detail in the data required for inclusion in meta-analysis for all studies, despite contacting the authors.

4.6. Authors' conclusions

The findings of the included studies indicate that these interventions do significantly increase parental mental wellbeing; however due to the paucity, quality and quantity of research application of this evidence is limited, especially for parents of young infants with emerging CP. The effect size of individual interventions appears relatively small and few studies have a primary aim of improving parental mental wellbeing.

4.7. Implications for practice

It was hoped that the completion of this study would allow us to guide clinicians to support the mental wellbeing of parents of children with cerebral palsy around the time of diagnosis, using approaches with proven efficacy. Unfortunately, this is an area that requires further research. The review included research showing that parental mental wellbeing can be indirectly improved through certain interventions primarily aimed at the child.

4.8. Implications for research

This review highlights several areas in need of exploration. More work is required to understand effective approaches to support fathers of children with CP, as their emotional responses and needs may differ from those of mothers. Another area to be addressed concerns the provision of effective early support around the time of diagnosis of CP. Furthermore, it is crucial to develop interventions that focus primarily on the psychological wellbeing of the parents rather than addressing this as a secondary aim. Such interventions ideally require flexibility and accessibility; and should be proportionate in terms of time commitment. Future studies should also be adequately powered, use standard outcome measures and address longer term outcomes for the parents and children.

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Appendix A. Supplementary data

Supplementary material related to this article can be found, in the online version, at doi:https://doi.org/10.1016/j.ridd.2019.103511.

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