**The effectiveness of psychosocial intervention for individuals with cleft lip and/or palate and their parents: A systematic review**

**Abstract**

***Objective:***The aim of this review was to assess the effectiveness of different psychological interventions for children and adults with cleft lip and/or palate and their parents.

***Design:***We searched six databases including MEDLINE and EMBASE to June 2013, and checked bibliographies. We included studies that evaluated any psychological intervention in studies that included at least 90% participants with cleft lip and/or palate or parents of those with cleft lip and/or palate. Studies containing less than 90% were excluded unless they reported results separately for those with CL±P, or raw data was available upon request from the authors. Inclusion assessment, data extraction, and risk of bias assessment were carried out independently by two reviewers.

***Results:***Seven studies were identified as inclusions, with only two studies being included in the full data analysis (one of which failed to meet the full inclusion criteria). The five remaining studies were included only in a narrative synthesis as data was available for people or parents of those with cleft lip and/or palate only. This highlights a distinct dearth of research into psychological intervention within the field of cleft lip and/or palate.

**Conclusions:** The review found no evidence to support any specific intervention. Key uncertainties need to be identified and addressed. Adequately powered, methodologically rigorous randomised controlled trials are needed to provide a secure evidence base for psychological intervention techniques in participants with cleft lip and/or palate and their parents.

**Key words:** psychological intervention, cleft palate, systematic review

# BACKGROUND

It is estimated that cleft lip and/or palate (CL±P) occurs in approximately 1 in 700 live births (Vieira, 2008). From infancy until early adulthood, care is provided by a large multidisciplinary team. The core members of the team as defined by the Clinical Standards Advisory group (CSAG, 1998) are cleft surgeons, speech and language therapists, orthodontists, paediatric dentists and nurses. In recognition of the challenges of CL±P, and following the centralisation of cleft care in the UK (CSAG, 1998), it was recommended that psychologists be included in every multidisciplinary cleft team.

Psychologists play an important role in the care of both children and adults with a CL±P. Looking and/or sounding different to their peers, social reaction to difference and the ongoing burden of treatment, can pose a number of challenges throughout an individual’s life (Rumsey and Stock, 2013). Other family members may also require psychological support, to cope with the emotions surrounding a diagnosis of CL±P and its treatment implications (Nelson et al., 2012). Literature exploring the psychosocial adjustment of those affected by CL±P has produced conflicting findings. While some studies suggested that individuals with a CL±P are more at risk of developing difficulties than their peers (Noar, 1991; 1992; Turner et al., 1997), others have found no differences between this group and the general population (Hunt et al., 2005; Collett and Speltz, 2006; Berger and Dalton, 2009). While these contrasting findings may be partly attributable to the multifactorial nature of psychosocial adjustment, a lack of consensus and comparability in relation to the methodology employed is also likely to play a role (Rumsey and Stock, 2013).

Nonetheless, some particular areas of difficulty have been identified. Those with CL±P may present with a number of cognitive, emotional and behavioural concerns (Endriga and Kapp-Simon, 1999), including difficulties with learning and development (linked to associated syndromes, time off school and impoverished social environments)(Lockhart, 2003), a higher risk of rejection from peers (Hearst and Middleton, 1997), a tendency to withdraw and internalise problems (Pope and Snyder, 2005), dissatisfaction with appearance (Billaud-Feragen and Borge, 2010), unrealistic expectations of cleft-related surgery (Cadogan and Bennum, 2011) and anxiety, depression and low self-esteem associated with facial difference more generally in relation to rejection by others (Thompson and Kent, 2001). Individuals with CL±P may therefore benefit from general support to improve quality of life and self-concept, increase social support, reduce social isolation and manage social anxiety. In addition, targeted interventions may be required to address concerns specifically linked to CL±P. A recent review of the psychosocial difficulties experienced by those born with a CL±P identified the need to address specific issues such as the management of appearance concerns, including expectations of surgery and orthodontic work, problems associated with speech, communication and education, and difficulties such as teasing and bullying (Rumsey and Stock, 2013). This review highlights the need for readily available intervention within an area with a paucity of service provision (CSAG, 1998).

Despite this important role in the provision of care for children and families affected by CL±P, evaluation of psychology services in reducing psychosocial difficulties has thus far been limited to internal audit data produced by individual clinics within the UK. This may be due in part to the tendency of clinicians to take a personalised approach to the content of interventions, in order to ensure that each individual receives support tailored to their own needs. While this level of flexibility is important, greater coherence in the development and evaluation of appropriate interventions would increase effectiveness and allow for direct comparisons. In addition, the James Lind Alliance (JLA) (a National institute for Health Research (NIHR) organisation which engages patients and clinicians in setting priorities for future research) recently identified the effectiveness and timing of psychosocial interventions to be the top ranking priority in CL±P research (JLA, 2012).

An up-to-date and systematic review of the literature, with a specific focus on the effectiveness of psychosocial interventions in CL±P is clearly required. This current review aims to address this need by specifically identifying and assessing the most useful approaches to psychosocial interventions within CL±P.

**AIMS &** **OBJECTIVES**

This systematic review aimed to 1) assess the evidence to support the effectiveness or efficacy of psychosocial therapy interventions compared to no intervention, 2) assess whether the focus and theoretical approach affect the relative effectiveness or efficacy of interventions (examples include, but are not limited to, cognitive behavioural therapy, solution-focussed therapy, mindfulness, person-centred therapy, and 3) assess whether the timing of psychosocial interventions and the frequency of sessions affect psychosocial outcomes in the short and longer term.

**METHODS**

### Search Strategy

The search aimed to identify all studies relating to psychosocial interventions for children or adults with CL±P or parents of children with CL±P. An extensive search strategy was used to search 10 databases, including MEDLINE, EMBASE, PsychINFO, and Cochrane central register of Controlled trials (CENTRAL) (See Appendix A for full search strategy and list of databases). The search was conducted on 11th June 2013. No language restrictions were applied. In addition websites including National Institute of Clinical Excellence (NICE) and the metaRegister of Controlled Trials (mRCT) were searched and reference lists of included papers were also searched. Search criteria were adapted to suit the search terms of each individual database.

### Inclusion Criteria

**Study design**

Eligible studies included both randomised and non-randomised controlled trials. Observational comparative studies, either within or between groups, were also eligible for inclusion.

**Population**

Individuals with non syndromic CL±P with/without cleft alveolus or individuals with syndromes with no known developmental delay (e.g. Sticklers, hemifacial microsomia) were eligible for inclusion. Intervention studies for parents of children with CL±P were also eligible for inclusion. Studies containing less than 90% children or adults with CL±P, or their parents, were excluded unless they reported results separately for those with CL±P, or raw data was available upon request from the authors.

**Intervention**

For the purposes of this review, a psychosocial intervention was defined as any attempt to develop solutions to the challenges individuals may experience to their psychological wellbeing when interacting with any element of the social environment. Within cleft care there is great variety in terms of the content and measurement of appropriate interventions. Therefore all interventions addressing psychosocial adjustment were included within the review.

**Comparators**

Studies that compared any psychosocial intervention with no intervention, or that compared different psychosocial interventions, or psychosocial interventions at different points in time were eligible for inclusion.

**Outcomes**

Any outcome measures related to psychosocial functioning were eligible for inclusion within the review.

**Analysis**

The authors used a qualitative approach to synthesise data across studies (Dixon-Woods et al., 2005). This narrative approach entails searching publications for information relating to key themes of interest to the reviewers and compiling them for comparison with other studies. In the instance of this review the reviewers searched for information pertaining to theoretical or therapeutic perspective of the intervention method of delivery (setting, person delivering the intervention) and timing of the intervention (age at intervention, intensity and frequency of the intervention). Where possible meta-analyses were performed on primary outcome measures (e.g.  preoccupation with appearance, anxiety, depression, confidence, quality of life, social integration, well-being). Mean differences (MDs) or Standard Mean differences (SMDs) together with 95% confidence intervals (CIs) were extracted. Effect sizes and confidence intervals were plotted using forest plots where appropriate. Study authors were contacted for additional information where necessary and possible.

**Assessment of Risk of Bias**

Two research assistants independently assessed bias in both trials and observational studies. Trials were assessed using the Cochrane Risk of Bias tool (Higgins and Green, 2011) to rate each of the following five components as high, low or unclear risk of bias: 1) method of sequence generation (how the randomisation sequence was generated, e.g. random number table), 2) method of allocation concealment (how the randomisation was concealed from researchers/clinicians), 3) method of blinding of the outcome assessor (how the study ensured the outcome assessor did not know to which condition the participant had been allocated), 4) selective reporting of outcome data (not reporting all outcomes in the results that are mentioned in the methods or failing to use standard outcome measures within a particular field of research) and 5) completeness of outcome data (were all attritions accounted for and was an intention to treat analysis performed?).

In the case of psychosocial interventions, it is not possible for the treating clinicians or participants to be blind to their treatment allocation, therefore, adequate blinding was considered to have taken place if both data analysis and outcome assessment were blinded. Selective reporting of outcome data was considered to have taken place when papers only reported outcomes which had a favourable result or if the outcome measures assessed did not include standard measures that experts in the area would expect to have been reported.

In the case of observational studies two reviewers (AN and AM) used the RAMbo assessment tool (Chen and Wang, 2009) to assess the quality of randomisation (R), whether missing data was accounted for (A) and whether the type of measurement was appropriate (M).

**RESULTS**

**Search Results**

The search results identified 2320 potential references for inclusion.After duplicates were removed, this was reduced to 1367 unique papers. Four independent reviewers (AN, MP, NS and HW)checked the remaining list for inclusion and identified 23 possible references for inclusion based on titles and abstracts.On retrieval of the full papers, two reviewers (AN and MP) independently identified seven potentially eligible studies that met the inclusion criteria. This process can be seen in Figure 1.

**Excluded studies**

Of the seven studies identified as potentially eligible for inclusion five studies could only be included in the narrative synthesis of this review (Robinson et al., 1996; Newell and Clarke 2000; Pelchat et al., 2004; Maddern et al., 2006; Kleve et al., 2002). These studies met all inclusion criteria, except that they did not contain data from at least 90% participants with CL±P, or 90% parents of those with CL±P. The authors sought to retrieve the CL±P-only participant data from each of these studies. However, due to the age of the papers this data was no longer readily available. Therefore, these papers were excluded from the analysis below, but are included in the narrative synthesis.

**Included Studies**

Two studies were included within the review (Kapp-Simon et al., 2005; Bessell et al., 2012). As with the excluded studies, each contained less than 90% participants with CL±P. The Kapp-Simon (2005) paper focused on children with craniofacial conditions; the judgement was therefore made that the findings from this study were likely to be relevant to children with CL±P more specifically. The Bessell et al. (2012) paper was included, as it was possible to access the raw CL±P participant data provided by the first author, and to focus the analysis on participants with a CL±P. As this paper was written by the first author of this review, data extraction and risk of bias assessments were conducted by three different reviewers (MP, AM and HW) to ensure objectivity was maintained.

**Risk of Bias Assessment**

Two studies were assessed using the Cochrane risk of bias assessment tool (Higgins and Green, 2011): Bessell et al. (2012) and Newell and Clarke (2000).

**Randomisation Procedure & Allocation Concealment:** Both papers were found to be at low risk of bias with regards to randomisation sequence and sequence allocation (See table 1).

**Blinding of outcome assessment:** Both studies were found to be at low risk of bias.

**Attrition:** All rates of attrition were adequately documented in the papers.

**Selective reporting of outcome measures:** All outcomes reported in the studies were reported in the results.

**RAMbo Assessment of observational studies**

The remaining five studies were assessed for bias using the RAMbo assessment technique (Chen and Wang, 2009).

**Randomisation:** None of the six studies employed a randomisation procedure to allocate participants within the study. All studies were therefore rated at high risk of bias (see table 2).

**Accountability of participants:** All studies provided information about missing participants where possible, although detailed information about the reasons for drop out was not included in the studies. No drop outs were present in the Kapp-Simon et al. (2005) study. Unfortunately in the Robinson et al. (1996) study, the rate of attrition at follow-up was so high that an insufficient number of participants were included in the study. Therefore, this study was rated at high risk of bias.

**Measurement:** It is unclear from the Pelchat et al. (2004) studies who was responsible for data analysis and a self-designed questionnaire was used rather than a validated questionnaire. The other papers were found to have adequate and appropriate measurement (Maddern et al., 2006; Kapp-Simon et al., 2005; Kleve et al., 2002, Robinson et al., 1996).

**Analysis**

Only one of the studies with usable data was a randomised controlled trial (RCT) (Bessell et al., 2012). As eight participants had a CL±P in this study and as there were no comparison studies, it was neither possible nor appropriate to perform a meta-analysis. Instead, a narrative synthesis was conducted on the two included studies and the six studies where data for CL±P alone were not available.

**Description of Studies**

**Therapeutic approach**

**Cognitive-Behavioural Therapy:** Five studies assessed the effectiveness of a CBT-based approach(Bessell et al., 2012; Maddern et al., 2006; Pelchat, 2004; Kleve et al., 2002; Newell and Clarke, 2000). These studies assessed CBT-based interventions in a range of adults with visible differences (Bessell et al., 2012; Maddern et al., 2006; Kleve et al., 2002; Newell and Clarke, 2000) or parents of children with CL±P or Downs Syndrome (Pelchat et al., 2004). See Table 3 for details.

**Social Skills Training:** Two studies assessed the effectiveness of social skills training (Kapp-Simon et al., 2005; Robinson et al., 1996), whilst an additional study included a mixed CBT and SST approach (Bessell et al., 2012). Of these studies two assessed the effects of this model in adults with visible difference**,** whilst the final study assessed an SST approach in adolescents with craniofacial conditions. See Table 3 for details.

**Method of Delivery**

**Self-help:** Two studies assessed the effectiveness of self-help interventions. The Bessell et al. (2012) paper compared face-to-face delivery of a CBT intervention against an online delivery with minimal facilitation from an assistant psychologist or counsellor.The Newell and Clarke (2000) intervention consisted of a CBT-based self-help booklet consisting of 18 A5 pages. The self-help booklet was mailed to participants for them to use in their own homes.

**Group-based (facilitated):** Two studies adopted a group-based approach to intervention. The Kapp-Simon et al. (2005) study assessed the effectiveness of a group-based social skills intervention for adolescents. The setting of the Kapp-Simon et al. (2005) intervention was unclear regarding the method of delivery (i.e. who delivered it). Robinson et al. (1996) assessed the effectiveness of a group-based social skills workshop for individuals with visible differences. No information was provided about who delivered the intervention.

**Face-to-face individual:** Four studies assessed the effectiveness of individual CBT-based interventions (Bessell et al., 2012; Maddern et al., 2006; Pelchat et al., 2004; Kleve et al., 2002). The Bessell et al. (2012) paper also assessed the effectiveness of a face-to-face delivery of a CBT/SST intervention administered by a trained counsellor or an assistant psychologist. This intervention took place within clinics. Maddern et al. (2006) assessed the effectiveness of a face-to-face CBT delivery for children with visible differences delivered by a clinical psychologist or assistant psychologist within clinics. Pelchat et al. (2004) assessed the effectiveness of an individual parent support programme for parents of children with CL±P or Down’s syndrome delivered by nurses partially in clinics and partially at home. Kleve et al. (2002) evaluated the effectiveness of a CBT-based therapy to address the psychosocial issues experienced by adults with visible differences. The intervention was delivered by the clinical psychology team at Outlook, Bristol.

**Timing of Intervention**

This review attempted to identify the optimal duration and intensity of intervention. The studies included generally varied in duration from two sessions (Robinson et al., 1996), through to 12 sessions (Kapp-Simon et al., 2005). Full details of intervention duration can be found in Table 3.

Where stated, the intensity of the interventions also varied from daily (Robinson et al., 1996) to weekly (Bessell et al., 2012; Kleve et al., 2002). The length of sessions varied from one hour (Bessell et al., 2012) to whole days (Robinson et al., 1996) (see Table 3 for full details of intensity).

**DISCUSSION**

**Summary of results**

This systematic review identified only two papers that matched the majority of the inclusion criteria. One paper focused on addressing appearance-related concerns amongst adults with visible differences, including adults with CL±P, and the second paper reported a social skills intervention for adolescents with a variety of craniofacial conditions, including CL±P. Only one intervention was tested using a RCT design, whilst the other was an observational study. Overall 28 participants with CL±P or other craniofacial conditions were represented within the two papers. Therefore, the two studies did not provide sufficient data to draw any firm conclusions regarding the best forms of psychosocial interventions for either adults or children affected by CL±P, the optimal method of delivery or intensity and timing of intervention.

**Therapeutic approach**

The interventions included within the current systematic review focused on social skills training or CBT, or a combination of the two approaches. Evidence from the wider psychological literature suggests that CBT and SST may prove useful psychosocial intervention models to address many of the concerns experienced by those with CL±P including reduced social communication and support, teasing and bullying and poor self-esteem. Social skills training for the general population have been found to be effective at increasing social support, through increasing social communication, and also reducing social isolation (Roth and Fonagy 2005). SST has also been highly effective in addressing social anxiety and is a technique that has been used in the general population for many years, both with adults (Roth and Fonagy, 2005), children and adolescents (Roth et al., 2002). SST can also help individuals to manage teasing and bullying by boosting self-confidence and improving social communication (Roth et al., 2002). There is also an argument that helping to increase individuals’ social support network and increase their social contact with others will in turn improve their quality of life, at least in relation to social elements (Kapp-Simon et al., 2005). The use of the SST and CBT approaches have been evaluated in a systematic review assessing interventions for those with visible differences more generally (Bessell and Moss, 2007). The poor methodological quality of the papers included in this review and the small numbers of participants involved made it difficult to draw any firm conclusions, but the review highlighted SST as a potentially useful technique for managing appearance concerns. The review also concluded that the incorporation of elements of CBT into these interventions had the greatest potential to promote adjustment in terms of improving social skills, reducing anxiety and reducing appearance-related distress. This review also highlighted the general paucity of intervention for individuals with visible difference, a difficulty also present within CL±P services, at least within the UK (CSAG, 1998).

Specific issues in CL±P may include the burden of multidisciplinary care and the impact of multiple surgeries. Also, as a CL±P can be associated with additional challenges such as speech and language difficulties, hearing loss and/or delays in cognitive development, the additive effects of these factors may result in distress. Further investigation is required to highlight the best models for intervention in this specific area.

**Method of Delivery**

Methods of delivery fit broadly within the following categories: group self-help, group- healthcare facilitated, individual self-help or individual facilitated intervention. Future research should assess which techniques of delivery are most appropriate for people and families.

The findings lend minimal support for a CBT or SST, or combined approach for this population, but the limited sample sizes make it impossible to draw any firm conclusions. Due to the differences in methodological design, it was also difficult to draw firm conclusions about the optimal delivery of psychosocial interventions. Each intervention was delivered using different facilitators (for example assistant psychologists, clinical psychologists, or nurse or researchers). Therefore, the review cannot recommend whether any particular individuals should be responsible for delivering these psychosocial interventions.

**Timing of intervention**

Due to the differing intensity and duration across the studies, it is difficult to draw firm conclusions regarding the optimal length and intensity of therapy. However, most studies opted for between 6 - 10 sessions administered weekly for 1-1.5 hours. Therefore, future trials should consider using this level of intensity and duration at a minimum whist ensuring that timing of interventions for this specific population are more rigorously evaluated. This also matches recommendations for the minimum intensity of therapies in the general population (Roth and Fonagy, 2005).

Evidence from the wider psychological literature based on CBT and SST interventions show that therapy varies considerably in length from 6 to 20+ sessions (Roth and Fonagy, 2005). The question of how interventions are delivered and the optimum duration and intensity depends at least in part on the degree of psychosocial distress the individual is experiencing (Roth and Fonagy, 2005) . At lower levels of distress, less intense, perhaps remote forms of intervention may be suitable, whereas moderate-to-severe levels of distress more intensive face-to-face support is needed (National Institute for Health Research, 2007). This tiered model of psychological care is based on the concept that following assessment/screening for psychological distress the individual is matched to an appropriate level of intervention based on level of distress and expressed need (i.e. the type of intervention they wish to receive). This model of stepped-care requires testing with the CL±P population. Clinical opinion suggests that most CBT/SST approaches should employ weekly or bi-weekly sessions (Roth and Fonagy, 2005). This model ensures that clients have enough time between each session to engage in at home assignments and to reflect adequately on the intervention sessions, but does not leave such a large period of time to run the risk of clients forgetting basic information or feeling unsupported (Roth and Fonagy, 2005). Despite this, there may be occasions, especially towards the end of the intervention when sessions become less regular and perhaps move to monthly sessions to provide individuals with “top up” support (Carr, 2009). The exact number of sessions required will always depend to some extent on individual need (Roth and Fonagy, 2005). In the absence of evaluations of interventions specifically relating to CL±P, guidance can only be drawn from the results of evaluations of interventions for people challenged by other conditions.

Due to the limited number of studies, no comment can be made of the optimal timing of intervention with regards to the age of participants. There were not enough studies to conclude whether intervention should take place in childhood or adulthood, or should be focused at parents in the early stages of an infant’s life. Further study is required and it is possible that the type of difficulty encountered may dictate the optimal age of intervention.

**Study Design**

All systematic reviews base the evidence for intervention models on a hierarchy of methodological designs (Centre for Reviews and Dissemination (CRD), 2001). The studies included in this review consisted mainly of observational studies. Only one study consisted of an RCT. The gold standard for evidence of effectiveness is taken from high quality RCTs with large sample sizes. In this field there is no RCT evidence on which to inform clinical practice. Although RCTs are the gold standard, they are not always possible to execute. In these instances observational studies can provide useful evidence, but they should be conducted to the highest methodological standard with sample sizes based on calculations that allow the detection of realistic and clinically important effects.

**Limitations of the Review**

The review conducted a comprehensive literature search across numerous relevant databases. All inclusion assessments were checked by at least two researchers. Additionally, reference sections of identified papers were also checked. In addition no language restrictions were applied. Therefore, it is unlikely that this review failed to include any studies conducted within this area. A further limitation is the small number of papers and small sample size represented within this review.

**Implications for practice**

This review was unable to provide evidence to support the use of any specific model of psychosocial intervention in relation to either the therapeutic approach, the duration of the intervention, the age at which interventions should be conducted or the setting, intensity and delivery of the interventions. Therefore, as a first step to understanding service provision an audit of current practice, along with a description of intervention models used would help to inform the direction of future practice. As the need to identify effective psychological interventions was the top priority in the recent James Lind Alliance (JLA) Priority Setting Exercise (JLA, 2012), a programme of research should be put in place to provide the evidence required to inform practice. Clinicians and researchers will need to collaborate in an effort to identify the factors which may contribute to the development and persistence of distress and resilience in those born with CL±P, in order to target these factors through interventions.

**Implications for research**

Researchers within the field of psychosocial intervention face many challenges in addressing the key uncertainties within the literature and in developing adequately designed studies that are well reported. In order for reviews to be useful in the future it is important that studies are conducted that formally assess therapeutic approaches and evaluate the effectiveness of psychosocial interventions. These papers need to include detailed information about the intervention and its underlying therapeutic approach. It is also necessary to describe the intervention setting, who provided it, and the frequency and intensity of the intervention in sufficient details to allow for replication (Abraham et al., 2008). A consensus is required in relation to the content and timing of interventions and in the choice of the measures used to assess the efficacy of these interventions.

Future studies also need to address the issue of small sample sizes. One way of ensuring larger sample sizes when working with a small population is to engage in multi-centre trials to increase access to participants. While multidisciplinary cleft services (in the UK) are well-placed to achieve this (with over 1000 new cases per year and the creation of larger clinical centres of excellence (Sandy et al., 2011)), the lifelong nature of CL±P, the complexity of psychosocial adjustment and the broad range of therapeutic approaches practised provide challenges for the clinical and research communities. An important step will be to achieve a consensus between clinicians and researchers in relation to the elements to be targeted by therapeutic intervention, and in the measures used to evaluate change, as well as substantial cooperation to achieve the necessary designs and sample sizes. Another option is the use of cohort multiple RCTs that may increase recruitment rates within trials and therefore would be especially useful for small populations (Relton et al., 2010). These studies take the form of large observational cohorts of patients whose outcomes are measured on a regular basis. The pool of patients is then used for any future RCTs, whereby the cohort is used to identify eligible patients, some of which receive the trial intervention (selection is done using a randomisation process). The outcomes of these patients are then compared against the outcomes of the eligible patients who were not randomised to receive intervention (i.e. those receiving usual care) (Relton et al., 2010). These trials use principles of RCTs but incorporate them into existing clinical practice models to ensure they represent real-world delivery. The recent establishment of a UK wide cleft cohort study provides a potential platform for this work, as it will generate longitudinal data about psychological adjustment to cleft and provide a platform from which representative samples can be studied using RCTs with ‘waiting list’ controls.

Where possible, future studies should take the form of RCTs and use core outcome measures as standard. These may include generic measures to assess adjustment in relation to other conditions and norm groups, and specific measures to tap into the issues that are particularly relevant to CL±P.

**Conclusion**

In identifying only seven studies suitable for a systematic review of psychosocial interventions in the field of CL±P, the findings of this review have limited clinical value. However, this paper does bring into sharp focus the current lack of research and consensus in the area and the paucity of current service provision. On the basis of the available research findings and evidence from the wider psychological literature, this review has highlighted that SST and CBT approaches are worthy of further investigation as techniques for addressing many of the difficulties associated with having a cleft. Pilot studies are required to assess the needs of children and adults with CL±P and their parents separately to ensure future interventions meet are targeted to the population. It has also highlighted that the need for a broader programme of work to inform the content and timing of future interventions. In light of the recent JLA priority setting exercise, it is clear that such a research programme is a priority in order to provide evidence-based interventions to address the challenges faced by families, children and adults affected by CL±P. RCTs are the way forward but can present challenges particularly when working with small clinical populations where large multi-centred trials are required. It is hoped that the establishment of a UK wide cohort study will serve as a platform for future collaborative RCT studies within the UK and will potentially aid with issues such as equipoise and blinding to interventions.

**Acknowledgments**

Thank you to the UK cleft psychology special interest group for their advice and support during the review process. Thank you to Amir Massoumian, Nastasha-Ann Deprosse & Jo-Lee Hankinson for their help with title and abstracts searches and risk of bias assessment.

**References**

Berger ZE, Dalton LJ. Coping with a cleft: psychosocial adjustment of adolescents with a cleft lip and palate and their parents. *Cleft Palate-Cran J.* 2009; 29: 352-356.

Bessell A, Brough V, Clarke A, Harcourt D, Moss TP, Rumsey N. Evaluation of the effectiveness of Face IT, a computer-based psychosocial intervention for disfigurement-related distress. *Psychol Health Med*. 2012; 5: 565-577.

Bessell A, Moss TP. Evaluating the effectiveness of psychosocial interventions for individuals with visible differences: A systematic review of the empirical literature. *Body Image.* 2007; 4: 227-238.

Billaud Feragen K, Borge AIH. Peer harassment and satisfaction with appearance in children with and without a facial difference. *Body Image*. 2010; 7:97-105

Cadogan J, Bennun I. Face value: An exploration of the psychological impact of orthognathic surgery. *Br J Oral Maxillofac Surg*. 2011; 49: 376-380.

Carr A. *What works with children, adolescents and adults: A review of research on the effectiveness of psychotherapy.* Hove, East Sussex: Routledge; 2009.

Centre for Reviews and Dissemination (CRD). Undertaking systematic reviews of research on effectiveness, CRD Report Number 4 (2nd Edition). Available at: http://www.york.ac.uk/inst/crd/report4.htm. Accessed January 19 2006.

Chen JF, Wang CF. Introduction of critical appraisal of medical literature. *Medical Dispute Mediation.* 2009; 42: 19-24.

Clinical Standard Advisory Group*. Cleft lip and or palate. Report of a CSAG Committee*. London: Department of Health; 1998.

Collett BR, Speltz ML. Social-emotional development of infants and young children with orofacial clefts. *Infant young child*. 2006; 19(4): 262-291.

Dixon-Woods MS, Agarwal S, Jones D, Young B, Sutton A. Synthesising qualitative and quantitative evidence: a review of possible methods. *J Health Serv Res Policy*. 2005; 10(1):45-53.

Endriga MC, Kapp-Simon KA. Psychological issues in craniofacial care: state of the art. *Cleft Palate-Cran J.* 1999; 36: 3-11

Hearst D, Middleton J. Psychological intervention and models of current working practice. In R Lansdown, N Rumsey, E Bradbury, T Carr & J Partridge (Eds.), *Visibly different: Coping with disfigurement.* Oxford: Butterworth-Heinemann; 1997

Higgins JPT, Green S. *Cochrane handbook for systematic reviews of interventions*. Oxford: The Cochrane Collaboration; 2011.

Hunt O, Burden D, Hepper P, Johnston C. The psychosocial effects of cleft lip and palate: A systematic review. *Euro J Orthodont*. 2005; 27: 274-285.

James Lind Alliance (JLA). Cleft lip and palate: A JLA priority setting partnership. Available at: <http://www.lindalliance.org/CleftPSP.asp>. Accessed June 28 2013.

Kapp-Simon KA, McGuire DE, Long BC, Simon DJ. Addressing quality of life issues in adolescents: Social skills interventions. *Cleft Palate-Cran J.* 2005; 42(1): 45-50.

Kent G. Understanding experiences of people with disfigurement: An integration of four models of social and psychological functioning. *Psychol Health Med*. 2000; 5: 117-129.

Leary MR. A brief version of the Fear of Negative Evaluation Scale. *Pers Soc Psychol B.* 1983; 9: 371-376.

Kleve L, Rumsey N, Wyn-Williams M, White P. The effectiveness of cognitive-behavioural interventions provided at Outlook: A disfigurement support unit. *Journal of the Evaluation of Clinical Practice.* 2002; 8(4): 387-395.

Lockhart E. The mental health needs of children and adolescents with cleft lip and/or palate. *Clinical Child Psychology and Psychiatry.* 2003; 8: 7-16.

Maddern LH, Cadogan JC, Emerson MP. Outlook: A psychological service for children with a different appearance. *Clin Child Psychol Psyc.* 2006; 11: 431-443.

National Institute for Health and Clinical Excellence (NICE). *Behaviour change at population, community and individual levels. NICE Public Health guidance 6.* London: NICE; 2007.

Nelson P, Glenny A-M, Kirk S, Caress A-L. Parents’ experiences of caring for a child with a cleft lip and/or palate: A review of the literature. Child Care Health Dev. 2012; 38: 6-20.

Newell R, Clarke M. Evaluation of a self-help leaflet in treatment of social difficulties following facial disfigurement. *Int J Nurs Stud.* 2000;3: 381-388.

Noar JH. A questionnaire survey of attitudes and concerns of three professional groups involved in the cleft palate team. *Cleft Palate-Cran J.* 1992; 29: 92-95.

Noar JH Questionnaire survey of attitudes and concerns of patients with cleft lip and palate and their parents. *Cleft Palate-Cran J.* 1991; 28: 279-284.

Pelchat D, Lefebvre H, Proulx M, Reidy M. Parental satisfaction with an early family intervention program. *J perinat neonat nur.* 2004; 18(2): 128-144.

Pope AW, Snyder HT Psychosocial adjustment in children and adolescents with a craniofacial anomaly: Age and sex patterns. *Cleft Palate-Cran J.* 2005; 42: 349-354

Relton C, Torgerson D, O’Cathain A, Nicholl J. Rethinking pragmatic randomised controlled trials: introducing the “cohort multiple randomised controlled trial” design. *Brit Med J.* 2010; 340: 963-967

Robinson E, Rumsey N, Partridge J. An evaluation of the impact of social integration skills training for facially disfigured people. *Brit J Plast Surg. 1996; 49:* 281-289.

Roth A, Fonagy P. *What works for whom? (2nd ed):* A critical review of psychotherapy research. New York: The Guilford Press; 2005.

Roth A, Target M, Cottrell D, Phillips J, Kurtz Z. *What works for whom: a critical review of treatments for children and adolescents.* New York: The Guilford Press; 2002.

Rumsey N, Stock NM. Living with a cleft: Psychological challenges, support and intervention. In: S Berkowitz, ed. Cleft Lip and Palate: Diagnosis and Management (3rd edition). Springer-Verlag Berlin Heidelberg; 2013/in press.

Sandy J, Kilpatrick N, Persson M, Bessell A, Waylen A, Ness A, Ireland A. Why are multi-centre clinical observational studies still so difficult to run? *Brit Dent J.* 2011; 211(2): 59-61.

Thompson A, Kent G. Adjusting to disfigurement: processes involved in dealing with being visibly different. *Clin Psychol Rev.* 2001; 21: 663-682.

Turner SR, Thomas PWN, Dowell T, Rumsey N, Sandy J R. Psychological outcomes amongst cleft patients and their families. *Brit J Plast Surg*. 1997; 50: 1-9.

Vieira AR. Unravelling human cleft lip and palate research. J Dent R. 2008; 87: 119-125.

**Figure 1: Diagram of review process**

### Appendix A: Sample search strategy (for Medline) and databases searched

The following search terms will be used to identify articles. Only English language papers will be considered. The Databases will be searched using the MEDLINE database strategy outlined below. This approach will be adapted to run on the other selected databases.

Medline strategy:

Cleft Palate/ (15858)

2 Cleft Lip/ (11262)

3 hare lip$.tw. (92)

4 harelip$.tw. (341)

5 Palatoschisis.tw. (83)

6 cleft lip$.tw. (8300)

7 cleft palate$.tw. (8313)

8 orofacial$ cleft$.tw. (544)

9 facial cleft$.tw. (824)

10 oral cleft$.tw. (441)

11 craniofacial cleft$.tw. (107)

12 Velopharyngeal Insufficiency/ (1336)

13 Velopharyngeal Insufficiency.tw. (534)

14 or/1-13 (22163)

15 exp Adaptation, Psychological/ (88681)

16 exp Psychotherapy/ (133902)

17 exp Counseling/ (28798)

18 "Self-Help Groups"/ (7073)

19 "Social Support"/ (42051)

20 ((psychosocial$ or psycho-social$) adj5 (intervention$ or treatment$ or therap$ or program$)).tw. (6601)

21 counsel$.tw. (54194)

22 (behavi$ adj5 (therap$ or treatment$ or program$ or intervention$)).tw. (36684)

23 (cognitiv$ adj5 (therap$ or treatment$ or program$ or intervention$)).tw. (15149)

24 (psychologic$ adj5 (therap$ or treatment$ or program$ or intervention$)).tw. (9679)

25 (mindfulness adj5 (therap$ or treatment$ or program$ or intervention$)).tw. (347)

26 "Early Intervention (Education)"/ (1312)

27 Patient Education as Topic/ (62111)

28 support group$.tw. (3710)

29 self-help.tw. (3764)

30 psychotherap$.tw. (26937)

31 group therap$.tw. (3177)

32 Social Adjustment/ (19616)

33 person-cent$ therap$.tw. (5)

34 solution-based therap$.tw. (0)

35 or/15-34 (412834)

36 14 and 35 (668)

The databases will be searched from 1937 to present. In cases where databases do not go back as far as 1937, searches will be conducted from inception. The following databases will be searched: Medline, Embase, Cochrane Database of Systematic Reviews (CDSR), Cochrane Central Register of Controlled Trials (CCTR), Web of Science, Database of Abstracts of Reviews of Effects (DARE), National Institute of Clinical Excellence (NICE), Psychinfo, and CINAHL.