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# Cleft Lip and Palate Research in the United Kingdom: Advances in Clinical Psychological Knowledge and Future Directions

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**Abstract**

In 2012, the James Lind Alliance (JLA) worked with individuals with cleft lip and/or palate (CL/P), their families and clinicians to identify priority areas for future research. This article reviews progress conducted in the United Kingdom in the 3 JLA priorities most closely related to Clinical Psychology. It then builds upon the original priorities to identify 4 future directions, based on contemporary literature and in-depth discussions between clinical and research experts. Finally, recommendations for next steps toward meeting these future directions are outlined, including consistent outcome measurement, engaging diverse groups of people with CL/P and embedding the principles of codesign.

**Keywords**

cleft lip and palate, psychosocial adjustment, evidence-based practice, mental health support

## Introduction

### *Background*

The impact of cleft lip and/or palate (CL/P), a congenital craniofacial condition, can include visible facial difference, early feeding difficulties, dental anomalies, and difficulties with hearing and speech. Individuals with CL/P typically receive multidisciplinary care throughout their lifespan.[1] The James Lind Alliance (JLA) is a United Kingdom–based initiative bringing together patients, carers, and clinicians in Priority Setting Partnerships. In 2012, the JLA collaborated with individuals with CL/P, their families and cleft clinicians in the UK, to identify the top research priorities within the field.[2] A survey of over 300 people, led to a shortlist of 26 research questions, which were subsequently prioritized to 12 questions. A number were related to psychosocial outcomes of individuals with CL/P (and/or their families; Table 1).

This paper starts with a review of recent research findings for research carried out in United Kingdom in relation to the 3 JLA priorities most relevant to Clinical Psychology. This paper then proposes future research directions, based on current literature and in-depth discussions between clinical and research experts.

### *Review of the 3 JLA Priorities Most Relevant to Clinical Psychology*

*James Lind Alliance Priority 1—What Types of Psychological Intervention (Individual Therapy, Community, or School Based) and at What Time (From Diagnosis to Adulthood) are Most Helpful for Patients With a CL/P and Their Families?.* Systematic reviews published between 2007 and 2024 examined psychological interventions for individuals with a range of congenital and acquired appearance-altering conditions and their parents, including CL/P.[3–8] Some evidence was found to support approaches based on cognitive behavioral therapy (CBT) and social skills training. However, many studies were of poor quality and no firm conclusions could be drawn. One online CBT- and social skills–based intervention originally designed for adults with visible differences (Face IT) and later adapted for young people (YP Face IT) has recently been trialed in 3 European countries.[9–12] Findings highlight likely utility with subgroups of people with appearance concerns. Evidence to support other forms

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**Table 1.** James Lind Alliance top 12 Priorities for Research in the Field of Cleft Lip and Palate.[a]

| | Priority research question |
|---|---|
| **Priority 1** | **What types of psychological intervention (individual therapy, community, or school based) and at what time (from diagnosis to adulthood) are most helpful for patients with a cleft of the lip and/or palate and their families?** |
| **Priority 2** | **What are the educational, employment and personal (eg, relationships) outcomes for individuals with a cleft of the lip and/or palate during childhood, adolescence and in the long term?** |
| Priority 3 | What is the best protocol for primary repair of both the lip and palate, including technique/timing and sequence? |
| Priority 4 | In individuals with a cleft of the lip and/or palate when is the most effective age to begin speech therapy? |
| Priority 5 | What is the best treatment for otitis media with effusion (glue ear) in individuals with a cleft of the lip and/or palate? |
| Priority 6 | Can stem cells be used to improve palate repair (both primary and secondary)? |
| Priority 7 | What interventions would enhance the educational outcomes for children with a cleft of the lip and/or palate? |
| **Priority 8** | **What is the impact of having a baby born with a cleft of the lip and/or palate on maternal/child attachment?** |
| Priority 9 | What is the best way to manage infants with a cleft of the lip and/or palate undergoing primary surgery before, during and after hospital (eg, Fluids, pain control, antibiotics, probiotics, arm splints, feeding practices)? |
| Priority 10 | What are the genetic and environmental causes of clefts of the lip and/or palate? |
| Priority 11 | How can we improve the diagnosis of cleft palate (without cleft lip)? |
| Priority 12 | What is the best way to prevent tooth decay in children with a cleft of the lip and/or palate? |

[a] Priorities relevant to the field of Clinical Psychology indicated in bold.

of intervention for other challenges related to CL/P is lacking and many methodological limitations exist. There remains a paucity of research into the effectiveness of novel interventions developed for individual with CL/P to address the range of psychosocial difficulties that can be experienced by this population and interventions developed with other clinical populations that have been adapted to meet the needs of those with CL/P.

*James Lind Alliance Priority 2—What Are the Educational, Employment, and Personal Outcomes for Individuals With a CL/P During Childhood, Adolescence and in the Long Term?.* In some children, a poorer trajectory has been found in relation to behavior,[13–16] cognitive functioning,[17] reading and language development,[17–20] and lower overall educational achievement in comparison to peers and siblings without a CL/P.[21–24] Higher rates of developmental delay and neurological differences also exist.[22,25] The negative impact of CL/P on quality of life appears low, especially after completion of primary surgeries. However, some children experience emotional, social, and appearance-related concerns,[16,26] especially for those with cleft palate or additional conditions.[26–28] Among adolescents with a CL/P, evidence suggests that the average prevalence of cognitive, behavioral, emotional (eg, mood and anxiety), social, and appearance-related concerns is consistent with the general population.[14,29] However, small subgroups exhibit higher risk.[29] In some adults, a higher prevalence of psychological difficulties, elevated emotional and interpersonal challenges,[30–32] a desire for further treatment due to ongoing concerns about appearance,[33,34] worries around starting a family and a range of functional impairments are reported.[35,36]

*James Lind Alliance Priority 8—What is the Impact of Having a Baby With a CL/P on Maternal/Child Attachment?.* Literature is mixed as to whether parent–infant interactions and attachment are affected by CL/P. Parent–infant interactions may be negatively affected in the short-term, but long-term attachment is unaffected,[37–40] albeit can be influenced by maternal well-being and feeding difficulties.[37,41] The presence of a cleft lip has been identified as a risk factor in some studies,[37] but not in others,[40] perhaps highlighting the complexity of parental adjustment to having a baby with a visible facial difference.

## Proposed Future Directions in Light of Developments in Research and Clinical Practice

Clinical Psychologists are integrated within the UK multidisciplinary cleft services to provide clinical input aimed at enhancing psychosocial outcomes for individuals with CL/P.[42,43] A research subgroup for the UK Cleft Clinical Psychology Clinical Excellence Network (CEN) was established in 2011, led by academic partners at the Centre for Appearance Research (CAR) at the University of the West of England, with the goal of establishing recommendations for the core set of psychological well-being measures for use in the Cleft Collective Cohort Studies.[44,45] The research subgroup has since evolved to continue the joint clinical research interests of CAR and members from the CEN.

As part of this reform, a review of research addressing the 3 JLA priorities most relevant to Clinical Psychology was carried out. Members of the research subgroup worked with Clinical Psychologists within the CEN, to identify gaps and barriers in cleft psychology research and to identify the areas of research which could lead to improvements in clinical practice. Four updated psychosocial research future directions were identified and these are described below along with recent work by the research subgroup to address these future directions.

*Future Direction 1—What is the Impact of CL/P on Parent and Family Well-Being and Functioning? (Building upon JLA Priorities 2 and 8).* Rather than focusing only on attachment, an understanding how CL/P impacts parent and family functioning more broadly was identified as an important future direction. Firstly, data collected from 1163 mothers and fathers of infants with CL/P enrolled in the Cleft Collective Cohort Studies between birth and the infants' primary cleft surgery were analyzed.[46] Mothers reported higher anxiety and depression compared to norms and both mothers and fathers reported higher perceived stress. Nonetheless, parents appeared to adjust well to their child's diagnosis. Investigations are currently underway by members of the research subgroup to assess the longer-term impact of CL/P on parental well-being now that longitudinal Cleft Collective data are available.

Members of the research subgroup have recently updated the review published in 2012 by Nelson et al, which aimed to understand parental experiences of caring for a child with CL/P.[47] This updated review included 126 new papers and summarized the known emotional impact of caring for a child with CL/P as well as describing parents' experiences of social situations and care delivery.[48]

Further research could focus on the impact on siblings (and sibling relationships), the relationships between parental dyads and the generational impact of being born with a CL/P (eg, relationships between different generations within a family affected by a CL/P or experiences of parents born with a CL/P themselves who go on to have their own child with a CL/P). Longitudinal or prospective cohort studies are needed, in addition to qualitative research to provide a nuanced insight.

*Future Direction 2—Which Factors Predict Psychosocial Adjustment in Individuals With a CL/P and Their Families? (Building upon JLA Priorities 2 and 8).* It is important to understand the factors which *predict* outcomes, to identify those most at risk and to drive the development of psychological interventions. Initial progress by the research subgroup includes the analysis of Cleft Collective data, which identified optimism, satisfaction with healthcare, social support, and relationship satisfaction as predictive of psychological outcomes in new parents of infants with CL/P. Other risk factors for mothers included the presence of a prior mental health condition and stressful life events during pregnancy. Risk factors for fathers included being older at the time of conception and having unexpected absences from work.[46]

Parent-reported Ages and Stages Questionnaire data extracted for 322 children enrolled in the Cleft Collective Cohort Study highlighted the following factors as factors associated with poorer overall development at 18 months: problems with physical development, feeding method (nasogastric tube and formula feeding combined), mother's (higher) levels of anxiety and depression, and marital status (single).[49]

A third study by members of the research subgroup in collaboration with the Cleft Lip and Palate Association (CLAPA) examined factors related to psychosocial adjustment in adults with CL/P, drawing on data collected from a national UK sample.[50] Predictive factors included reports of a happy childhood, talking about CL/P with family, close friendships, comfort in public spaces, satisfaction with appearance, and a positive life orientation. Psychological distress was associated with a desire for further surgery to improve appearance and/or function. Interviews with clinicians working in UK cleft services identified similar risk and protective factors for psychological distress in affected families and individuals, based on clinical experience.[51]

The research subgroup is currently investigating promising psychological factors that may inform future intervention development, including parental psychological flexibility,[52] and the longitudinal role of parental adjustment on child development and well-being.

*Future Direction 3—What Aspects of Cleft Care Need be Improved for People With CL/P and Their Families and How Might This be Achieved? (Building upon JLA Priority 1).* Research that leads to improvements in clinical care in the National Health Service providing cleft treatments was not included in the original JLA priorities. Members of the research subgroup explored healthcare satisfaction in parents using data collected from 517 parent dyads enrolled in the Cleft Collective Cohort Studies.[53] Parents reported being satisfied with their overall care. However, less favorable ratings of satisfaction were made in relation to the information parents had received and how included fathers felt in relation to their child's treatment pathway.

In rapid response to the Covid-19 pandemic, members of the research subgroup conducted interviews with new parents of babies born with CL/P to explore their experiences.[54] The findings identified parental concerns about delays in primary surgery and leant further support to the need for multidisciplinary care for families from diagnosis onward, the importance of timeliness for primary surgery, the need for routine screening for parental psychological distress, and the potential for remote support to reduce the burden of care and increase accessibility.

Two further studies by the research subgroup interviewed young people aged 17 to 25 years with CL/P about their experiences of transition from child to adult care in UK cleft services.[55,56] These studies indicated that some experienced transition as overwhelming and that improvements in transitioning care were needed to ensure young people are prepared to access and manage their care into adulthood and to meet the standards of care outlined by the National Institute for Health and Care Excellence.

More research is needed to understand the factors that influence surgical/treatment decision-making in those with CL/P (and their families). There are limited insights into the psychological processes related to how surgical/treatment decisions are made and how these processes might be associated with post-surgical satisfaction or psychosocial outcomes. Interventions aimed at supporting people with surgical/

treatment decision-making and enhancing post-surgery satisfaction and psychosocial outcomes are lacking.

*Future Direction 4—How Acceptable and Effective are Psychological Interventions Aimed at Improving the Psychosocial Well-Being of Individuals With CL/P and Their Families? (Building upon JLA Priority 1).* To understand the approaches that were being used to support families by clinicians working in UK cleft services, members of the research subgroup conducted in-depth interviews with Clinical Nurse Specialists and Clinical Psychologists. Clinical Psychologists drew upon a range of therapeutic approaches, most commonly CBT and systemic approaches, as well as Acceptance and Commitment Therapy (ACT), Compassion-Focused Therapy (CFT), and Narrative Therapy.

There is an evidence base for a range of psychological approaches, particularly for CBT, ACT, and CFT, which are likely to be of benefit to individuals with C/P and their families. However, it is currently unknown at what time points interventions would be most acceptable or effective, for what type(s) of difficulties (eg, appearance concerns, self-esteem/social confidence, procedural distress) or whether standard approaches to these broad therapeutic modalities need to be tailored to suit the cleft population and their families (and if so, how).

Interventions can range from support for appearance concerns, through to procedural distress or low self-esteem. This presents considerable challenges for evaluation, as interventions are rarely homogeneous, but formulation-driven and person-centered, meaning that no 2 interventions are the same. One solution to this challenge is the implementation of Goal-Based Outcomes, which have high face validity, with many users advocating the flexibility of goals.[57,58]

The research subgroup has begun to address the gap in evidence for intervention by exploring the concepts of acceptability and feasibility. Members of the research subgroup were involved in the evaluation of 2 new community-based programs of service delivery by CLAPA. The first program focused on using a new regional service to increase local support. The mixed-methods evaluation demonstrated that volunteers felt more confident in their role and more able to support others, while caregivers, children, young people, and adults with CL/P felt more connected to a local support network and better able to cope with the challenges of CL/P.[59] The second program aimed to increase information and support specifically for adults with CL/P by developing a podcast, a conference, and a leaver's pack of resources to support the transition to adult care. These interventions were viewed as valuable by adults with CL/P and health professionals.[60]

A recent pilot study by members of research subgroup investigated the potential acceptability of the Promoting Resilience in Stress Management-Parent, originally developed for young people with chronic conditions. Given the recurring presence of optimism as a leading protective factor for psychological distress, targeting resiliency in those with CL/P and their families could prove beneficial. In this pilot study, acceptability was rated highly, and qualitative data highlighted areas for adaptation for this population to enhance feasibility that can be implemented in future research designs.[61]

Clinical Psychologists can also indirectly influence the psychosocial outcomes of those with CL/P through consultation, education, and supervision within the wider cleft team.[43] While it can be challenging to quantify the impact of these ways of working, future research could focus on understanding patients' and professionals' experience of the impact of psychologically informed working within teams.

## Addressing These Future Directions

There is a need to move beyond the simple question of 'What is the psychosocial impact of CL/P?' and instead focus on understanding what predicts, prevents, reduces, or prevents this impact and what fosters positive psychological adjustment, resilience, and satisfaction with cleft care. Furthermore, we need to understand what we can learn about the wider systemic impact and how services can be improved.

Historically, research within cleft care has been deficit-focused, with insufficient attention paid to strengths and resilience.[62] Studies which help identify the ways in people individuals with CL/P have similar, or better, outcomes than control groups can also be highly informative for patient care. There are also key considerations regarding inclusion and exclusion criteria in research.[63] While narrow inclusion criteria optimize the homogeneity of samples and subsequently help identify the specific impact of certain factors, such as cleft type, these often have unintended consequences.[27] For example, individuals with conditions additional to CL/P are often excluded from research in the pursuit of sample homogeneity. However, these are the individuals who may be at higher risk for experiencing psychological difficulties.[27]

While there have been advances in achieving consensus in the measurement of psychological adjustment to CL/P, consistency in measurement remains a key issue.[44,45] Researchers are encouraged to continue to aim for consensus in measurement, as this facilitates comparison and continuity across studies with the aim of meta-analyses being possible in the future.[62]

Clinical Psychologists face barriers conducting research within their clinical roles in cleft services,[51] including clinicians lacking confidence and time. Ongoing collaboration with academic partners, as demonstrated by the research subgroup described in this paper, is one solution to this challenge.

It may be helpful to examine broader literature and advancements in the related fields of visible difference, body image, chronic illness, rare conditions, mental health, and disability. This may further inform our understanding of psychological adjustment to CL/P and allow us to draw on effective outcome measures, tools, and interventions.

Finally, future research should be representative of the CL/P population as a whole. This means strides need to be taken to ensure CL/P research is as inclusive as possible, across a range of demographics, including but not limited to: ethnicity, gender, intellectual ability, neurodiversity, religion, and sexuality. As such, intersectionality can be appropriately considered, leading to better and more relevant psychological support.[64]

Although the future directions presented in this paper are influenced by the accounts of individuals with CL/P and their families as part of the JLA priority-setting process and through qualitative research, a limitation of this paper was a lack of direct consultation with individuals with CL/P and their families. A future step must be to consult with individuals with CL/P and their families, to determine their perceptions of the relevance and impact of research resulting from this updated direction-setting process. Indeed, future research should strive to involve codesign with those with CL/P. Another limitation of this paper was the focus on research conducted within the UK and as such it is unclear as to whether findings are generalizable across different cultural contexts. We encourage psychologists in other countries to undertake similar direction-setting exercises, which will also allow for cross-cultural comparison.

## Conclusions

In summary, despite considerable advances in the field of cleft Clinical Psychology since the publication of the JLA priorities, key research questions remain unanswered. This paper has outlined future directions for cleft psychology research in the UK. It is important that future research capitalizes on the opportunities presented by collaborative and multidisciplinary research and aims to represent the outcomes and experiences of all people with CL/P and their families. The next step should be to ensure that these future directions reflect the priorities of this population.

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### Authors' Contribution

Matthew Hotton: conceptualization (equal); investigation (equal); methodology (equal); project administration (equal); writing—original draft (equal); and review and editing (equal). Laura Shepherd: writing—original draft (supporting) and review and editing (equal). Nicola Stock: conceptualization (equal); investigation (equal); methodology (equal); project administration (equal); and writing—original draft (equal), review and editing (equal).

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