



The Cleft Collective
Closing the Gap in Cleft Research
A Scar Free Foundation Initiative



Public Involvement in The Cleft Collective Research Programme

A strategy document

The Cleft Collective Team

and

The Cleft Lip and Palate Association

Version 3

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**Bristol Speech and
Language Therapy
Research Unit**

Background to the Cleft Collective

The Cleft Collective is a national programme of research investigating the causes of, the best treatments for, and the psychological impact of cleft lip and/or palate (CL/P). Initiated by the Scar Free Foundation in 2012, the Cleft Collective is now the largest and most comprehensive CL/P research programme in the world.

A History of the Cleft Collective

In 1998, the **Clinical Standards Advisory Group (CSAG)** commissioned a research team to evaluate the care of children with CL/P in the United Kingdom (UK). The team found standards of care to be disappointing, and as a result, CL/P services were centralised in the UK. This process reduced the number of surgical sites from 57 to 17, and ensured that all health professionals working in CL/P services were dedicated specialists. The centralisation of CL/P care put the UK in a unique position to conduct large research studies across the different sites and disciplines.

Although centralisation allowed for more research activity, the **Craniofacial Society of Great Britain and Ireland (CFSGBI)** recognised that the amount of research taking place in the UK remained limited, and that there were no agreed research priorities for CL/P. Between 2005 and 2007, the CFSGBI funded a series of workshops for clinicians and researchers to discuss how to move CL/P research forward. It was suggested that a 'gene bank' be established in order to find out more about the causes of cleft, alongside a multidisciplinary cohort study that could follow families as their children grew up. The **Scar Free Foundation** (previously the Healing Foundation) consequently donated a significant sum to bring these plans to action. With the additional support of the **Vocational Training Charitable Trust Foundation (VTCTF)**, more than £2.4 million was pledged to establish the Cleft Collective research programme.

In 2012, the **James Lind Alliance (JLA)** brought patients, clinicians and researchers together to identify the most important unanswered questions in CL/P. A list of 12 research priorities was subsequently published, which subsequently informed the development of the aims of the Cleft Collective.

The Cleft Collective

The Cleft Collective research programme was set up as a collaboration between the **Bristol Dental School at the University of Bristol** and the **Centre for Appearance Research at the University of the West of England**. The Cleft Collective team also works in partnership with the **Bristol Speech and Language Therapy Research Unit**, which is part of the North Bristol NHS Trust. The Cleft Collective team are housed alongside the **MRC Integrative Epidemiology Unit**, and the **Avon Longitudinal**

Study of Parents and Children (ALSPAC) study team, another UK cohort study which has been running for more than 20 years.

The Cleft Collective is managed by the Scar Free Foundation Advisory Panel, which is made up of our funders, specialist CL/P clinicians from a range of disciplines, and a parent representative(s). The Cleft Collective is also supported by the **Cleft Lip and Palate Association** (CLAPA), who promote our research to potential participants, help us to carry out Public Involvement activities, and support the dissemination of the findings of our research.

Since the Cleft Collective recruited its first family in 2013, nearly 3,000 families have contributed biological samples (including saliva, blood, cord blood and discarded lip and palate tissue) and longitudinal questionnaire data. Access to other existing information, such as medical and educational records is also requested from enrolled families, to ensure we collect as complete a picture of family life as possible. This data collection is made possible by the support and dedication of every NHS cleft team in the UK.

We have secured funding for the core costs of the research programme until September 2020, and are currently applying for more. Our hope is that this study will continue to recruit and follow families for many years to come, so that as much can be learned about CL/P as possible, and so that the key unanswered questions which are important to families, clinicians, and researchers can be addressed.

Introduction to Public Involvement

Public Involvement (PI) concerns the active involvement of members of the public in the research process itself, as opposed to only being involved as a participant or 'subject' of the research.

The national research advisory group **INVOLVE** defines PI as *“research that is done **with** or **by** the public, rather than to, about, or for them”*.

In the case of the Cleft Collective, members of the public predominantly include individuals who were born with CL/P, their families, and the lay organisations that support them (e.g. CLAPA). When involved in research, these individuals are most often referred to as 'PI representatives'.

Key Principles of PI

PI is now an essential requirement of research funding. PI representatives should be involved in the development of the funding application itself, and there should be a clear plan in place for involving PI representatives in the research if the project does receive funding. People who are affected by research have a right to have a say in what and how research is undertaken, particularly when the research is funded by a public body.

Although these changes to policy are a positive step toward including members of the public in research, PI should never become a tick-box exercise that is hurried or ill thought through. Due time and consideration should be given to all PI activities, and the broader PI strategy of a research programme should be well planned and integral to the values and conduct of the project.

Indicators of Authentic PI

There are a number of key issues to consider when engaging in PI activities:

- **Diversity** – researchers should seek to recruit PI representatives with a range of backgrounds, perspectives, and skills.
- **Training** – PI representatives should be able to understand the aims of the project and their role within the project. PI representatives should be given opportunities to develop all the skills they need to contribute to the research effectively.
- **Inclusion** – PI activities should be structured around the needs of the PI representatives, rather than the needs of the researchers. This may include considerations such as the timing, location, and accessibility of events.
- **Equality** – PI representatives should have equal say and be seen as experts in their own experiences. Use of technical language should be avoided or clearly explained.
- **Reimbursement** – participation in PI should come at no cost to PI representatives, who should be covered for any travel expenses as needed. In some circumstances, PI representatives may also be offered salary support or child care costs.
- **Engagement** – the researchers should keep all PI representatives informed of how the project is progressing, and how their contributions are continuing to shape the research process.

Benefits of PI

PI can improve the research quality, enhance the credibility and cost-efficiency of the project, and ensure that the outputs of the research are relevant to the end users.

PI also gives members of the public an opportunity to have their voice heard, to influence the direction of research and clinical practice, to ‘give back’ to their community, and to support others in similar situations. PI representatives often develop new skills and experience opportunities that may not have been previously available to them.

The Research Pathway

There are several different stages of the research process in which PI may be helpful. These are shown in Figure 1 below. PI representatives may choose to become involved in one or all of these different stages.

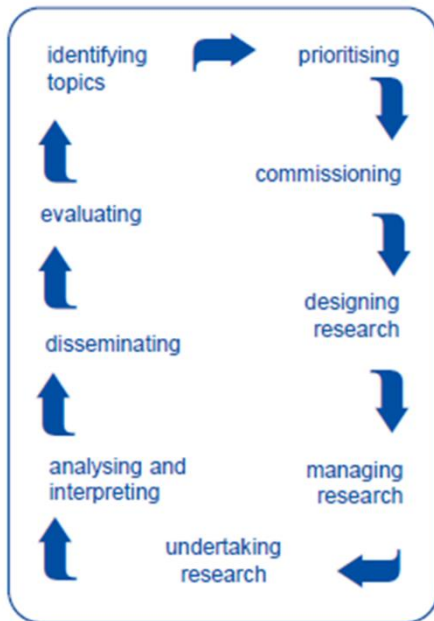


Figure 1: The Research Pathway (INVOLVE)

Levels of Involvement

There are three main levels of PI:

- **Consultation** – PI representatives will typically be asked to provide their feedback on something which is important to the research process, such as a funding application or a participant information leaflet.
- **Collaboration** – this represents an ongoing partnership between the researchers and the PI representative, such as a role on a project advisory panel.
- **User-controlled** – the research is actively directed and managed by PI representatives and/or lay-led organisations.

No one of these approaches are considered better than another; rather it depends on the nature of the task. A research project may use one, two, or all three of these approaches over time.

PI within the Cleft Collective Research Programme

The Cleft Collective research programme has engaged in a number of PI activities since it was first initiated, and the intention is to ensure that PI is an ongoing and integral feature of all future work.

PI Activities to Date

At the very beginning of the programme, the James Lind Alliance and other patient-focused organisations were involved in setting and prioritising the research questions which now drive the Cleft Collective agenda. CLAPA and its community were involved in the design of the research protocols, questionnaires, and other participant materials, to ensure they were relevant and accessible. A patient/parent representative has been crucial to the Advisory Panel, to provide guidance on the way the project is conducted from the patient perspective. Finally, patients, clinicians and families have helped with the interpretation and dissemination of research findings at conferences and events. These activities are illustrated below in Figure 2.

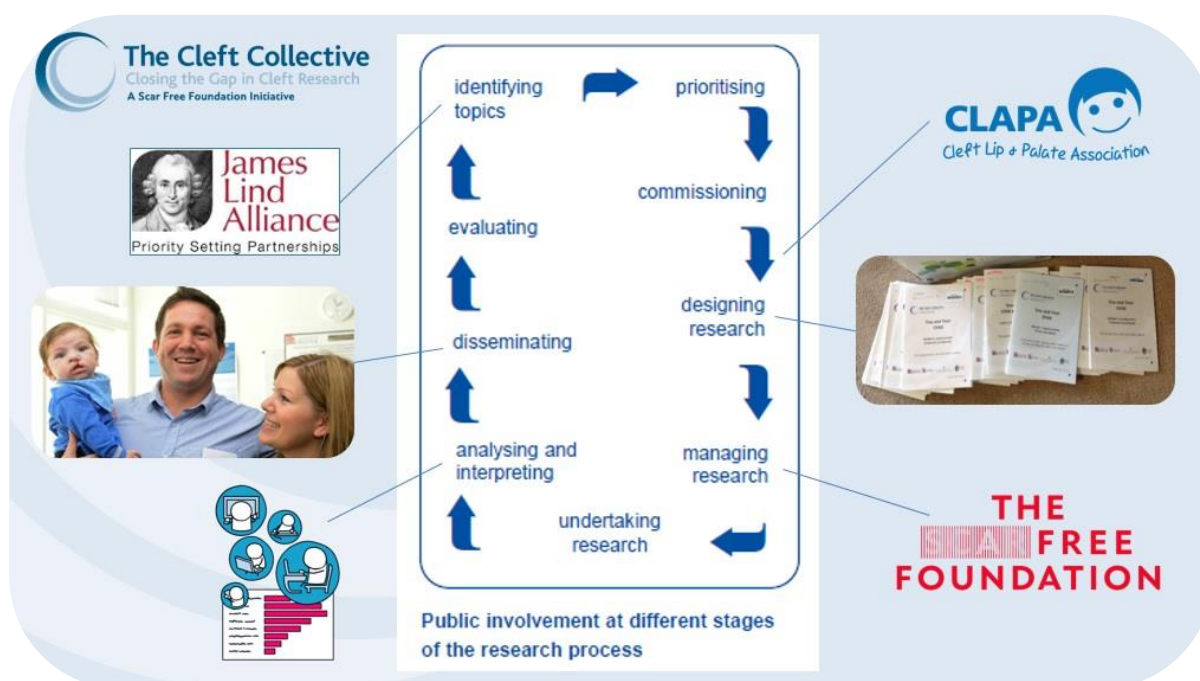


Figure 2: PI in the Cleft Collective to date

The Cleft Collective team have also conducted a series of qualitative studies to inform the development of the research agenda and the content of participant questionnaires. These references are available upon request.

In September 2017 a research workshop was held in London in collaboration with CLAPA. Feedback from PI representatives was gathered in relation to participation in the cohort studies, continuation of recruitment and data collection, and the proposed research agendas of the three main work

streams (molecular epidemiology, speech and language, and psychology). A summary of the key points taken from this workshop are available in a separate document upon request.

Proposed Future PI Activities

PI has played an integral role in the establishment of the Cleft Collective research programme. Now that data are being collected successfully, the programme is moving into a more research-focused phase. As such, further consideration of the role of PI in the Cleft Collective going forward is necessary.

Operations

PI will be crucial in the management of day-to-day activity within the Cleft Collective programme. This includes, but is not limited to: participant recruitment, data collection, and public engagement. The following core components are proposed:

- PI input into any significant protocol changes
- PI input into the design of all participant materials
- PI input into the communication of significant newsworthy items
- The inclusion of a parent/patient column in the Cleft Collective newsletter
- Opportunities to contribute to the Cleft Collective Image Bank and other resources

Research

PI will be essential to the ongoing development of the broader research agenda, as well as to the running of individual research projects. The following core components are proposed:

- PI input into the development of research questions and research priority setting
- PI input into the content of participant questionnaires
- PI review of all grant applications and co-construction of lay summaries
- PI review of all research proposals received
- PI input into the communication of research findings, including authorship on reports and publications where appropriate
- PI representation in mass media engagement

These activities will be achieved through:

- Ensuring a minimum of one patient/parent representative in the membership of the Scar Free Foundation Advisory Panel. Ideally, this/these representative(s) should also be a participant(s) in the cohort study.

- Ongoing recruitment to and maintenance of the Cleft Collective **Participant Pool** (*also see the Cleft Collective Communication Strategy*)
- Collaboration with the **CLAPA Cleft Collective Patient Consultation Group**

The CLAPA Cleft Collective Patient Consultation Group

The CLAPA Cleft Collective Patient Consultation Group was established by CLAPA in 2018 and is comprised of several patient and parent representatives located across the UK. The Cleft Collective will work in collaboration with CLAPA to access this group for discussion and feedback on research-related issues. Where possible, meetings of the CLAPA Cleft Collective Patient Consultation Group will be attended by a member of the Scar Free Foundation Advisory Panel, to ensure the continuity and accuracy of feedback between groups.

The responsibilities of **CLAPA** in relation to this collaboration will be as follows:

- To recruit volunteers to the CLAPA Cleft Collective Patient Consultation Group as needed, to ensure diverse and sufficiently sizeable representation
- To provide standardised volunteer training to all members of the CLAPA Cleft Collective Patient Consultation Group
- To support the Cleft Collective to organise one accessible face-to-face meeting per year (September), supported by one/two teleconferences per year (January and May), or as needed
- To send out Cleft Collective documents to CLAPA Cleft Collective Patient Consultation Group members as needed
- To offer the Cleft Collective access to the Zoom conferencing service
- To be the first point of contact for any general issues relating to the CLAPA Cleft Collective Patient Consultation Group
- To alert the Cleft Collective to any changes to the group structure or membership

The responsibilities of the **Cleft Collective** in relation to this collaboration will be as follows:

- To provide supplementary research training to CLAPA Cleft Collective Patient Consultation Group members
- To provide CLAPA Cleft Collective Patient Consultation Group members with appropriate supporting documents a minimum of one week prior to each meeting
- To provide a summary of key points and agreed actions after each meeting
- To be the first point of contact for any research-related issues

The costs of one member of CLAPA staff to organise and attend each meeting will be covered by the Cleft Collective on a full cost-recovery basis (see CLAPA Research Consultancy Fees document).

The responsibilities of **PI representatives** in relation to the Cleft Collective will be as follows:

- To attend standardised research training with the Cleft Collective. This training may be delivered in person or over the telephone, depending on needs and resources.
- To attend a minimum of one meeting per year
- To read through any supporting documents in advance of each meeting
- To provide reasonable notification if they cannot attend a meeting
- To give permission for their comments at meetings to be audio recorded for internal anonymised use

PI Research Training

Research training of potential PI representatives should include:

- An overview of research, including different methods and approaches
- An overview of PI in research, including the INVOLVE definition, the different stages of the research process, the benefits of PI, the different levels of involvement, and indicators of authentic PI
- An overview of the Cleft Collective research programme, including its history, overarching aims, protocols, work streams, team members, progress to date, and future ambitions
- A description of the role PI has played in the Cleft Collective to date, and an overview of the current PI strategy
- An overview of the expectations and responsibilities of Cleft Collective and CLAPA staff, and PI representatives
- Opportunities for any questions and discussion

Cleft Collective staff will also be expected to ensure that their PI knowledge and skills are current, via the undertaking of suitable PI training and research, and/or attendance at relevant events.

Roles of Individual Staff

The PI lead(s) for the Cleft Collective will:

- Be the first point of contact for any PI queries from staff
- Liaise with CLAPA on behalf of the Cleft Collective team
- Prepare any supporting documents prior to each meeting, and prepare summary documents following each meeting

- Lead on the delivery of research training where possible
- Chair PI meetings where possible
- Ensure all research documents are up to date
- Ensure appropriate records are kept and ongoing PI activities are appropriately reported and evaluated

Every member of the Cleft Collective team is responsible for engaging in appropriate PI. Administrative backing may also be required to support the organisation of meetings and the dissemination of activities.

Monitoring the Impact of PI

Regular monitoring of the impact of PI activities within the Cleft Collective research programme is essential for maintaining standards. PI activity will be monitored by the Cleft Collective in the following ways:

- PI will be a standing item agenda in Cleft Collective research meetings
- Standardised feedback will be collected from PI representatives following each CLAPA Cleft Collective Patient Consultation Group meeting
- Records of all PI activity will be kept in the form of summary documents, which will be shared with all PI representatives, and Cleft Collective and CLAPA staff members
- PI activity will be posted on the Cleft Collective website, in newsletters, and on social media
- PI activity will be reported in all external reports and publications, and the contribution of PI representatives will be acknowledged
- This PI strategy will be reviewed every 12 months and updated as necessary
- A member of the Cleft Collective will attend one CLAPA team meeting per year (~July) to reflect on joint progress

Also see the NIHR National Standards for Involvement document (March 2018): https://www.nihr.ac.uk/news-and-events/documents/Public_Involvement_Standards_March%202018_WEB.pdf.

Useful Links

INVOLVE: www.invo.org.uk.

The Scar Free Foundation: www.scarfree.org.uk.

Vocational Training Charitable Trust Foundation: www.vtctfoundation.org.uk.

Craniofacial Society of Great Britain and Ireland: www.craniofacialsociety.co.uk.

Centre for Appearance Research: www.uwe.ac.uk/car.

The James Lind Alliance: www.jla.nihr.ac.uk.

Bristol Dental School: www.bristol.ac.uk/dental.

Bristol Speech and Language Therapy Research Unit: www.nbt.nhs.uk/bristol-speech-language-therapy-research-unit/about-bsltru.

Avon Longitudinal Study of Parents and Children: www.bristol.ac.uk/alspac.

MRC Integrative Epidemiology Unit: www.bristol.ac.uk/integrative-epidemiology.

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The Cleft Collective: www.bristol.ac.uk/cleft-collective

The Cleft Lip and Palate Association: www.clapa.com

