Sainsbury, D. C. G., Davies, A., Wren, Y., Southby, L., Chadha, A., Slator, R., Stock, N. M. (2019). The Cleft Multidisciplinary Collaborative: Establishing a Network to Support Cleft Lip and Palate Research in the United Kingdom. *Cleft Palate-Craniofacial Journal*, 56(4), 502-507. <https://doi.org/10.1177/1055665618790174>

Peer reviewed version

Link to published version (if available):
[10.1177/1055665618790174](https://doi.org/10.1177/1055665618790174)

[Link to publication record in Explore Bristol Research](#)
PDF-document

This is the author accepted manuscript (AAM). The final published version (version of record) is available online via Sage at <http://journals.sagepub.com/doi/10.1177/1055665618790174>. Please refer to any applicable terms of use of the publisher.

University of Bristol - Explore Bristol Research

General rights

This document is made available in accordance with publisher policies. Please cite only the published version using the reference above. Full terms of use are available:
<http://www.bristol.ac.uk/red/research-policy/pure/user-guides/ebr-terms/>

The Cleft Multidisciplinary Collaborative: Establishing a network to support cleft lip and palate research in the United Kingdom

Abstract

Background: As a growing paradigm of health research, trainee collaboratives can influence clinical practice through the generation of cost-effective multicentre audit and research projects. The aims of the present article are to outline and discuss the establishment of a multidisciplinary collaborative in the context of cleft lip and/or palate (CL/P).

Methods: The Cleft Multidisciplinary Collaborative (CMC) was formed in April 2016 under the overarching supervision of the National Institute for Health Research. Membership of the CMC is open to all members of the CL/P multidisciplinary team, who are encouraged to submit ideas for new research projects which will benefit clinical practice.

Results: To date, 48 clinical participants are involved in the CMC. These participants represent all 17 cleft teams from the UK and encompass a wide range of disciplines. The CMC has undertaken two major projects thus far. The first involved collection of phenotype data to support a national cohort study. The second, still in progress, is a systematic review investigating factors associated with outcomes for velopharyngeal competence following cleft palate repair.

Conclusions: The concept of a multidisciplinary collaborative in CL/P has been demonstrated, through the generation of a UK-wide network of committed clinicians and researchers, and the effective delivery of two large research projects. As the CMC gathers momentum, it hopes to attract funding to support its activities, to promote more involvement from the allied health and nursing professions, to encourage a more ingrained research culture within the CL/P community, and to promote the wider ambition of a global collaborative.

Key words: trainee, collaboration, multicentre, research, audit, multidisciplinary

Introduction

The first trainee collaborative, the West Midlands Research Collaborative (<http://wmresearch.org.uk>), was established in 2007 by a group of general surgical trainees, with the aim of changing clinical practice through the generation of multicentre audit and research projects. Their success has been recognised, and other collaboratives have been established in the last decade, both across a range of surgical disciplines and within other medical specialities (Kolias et al., 2013; Skerrit and Hall, 2015). Through these supportive networks, clinical trainees are able to design, organise and disseminate both small and large scale collaborative projects across a wide geographical area (Nepogodiev et al., 2017).

As a growing paradigm of healthcare research, several studies have sought to evaluate the range of activity of trainee collaboratives (Bhangu et al., 2014; Dowswell et al., 2014; Skerrit and Hall, 2015; Jamjoom et al., 2016; Mehta et al., 2017; Nepogodiev et al., 2017). One large observational study of 24 surgical trainee research collaboratives in the United Kingdom (UK; Jamjoom et al., 2016) identified more than 80 projects, 41 percent of which had been completed and 59 percent of which were currently running or under development. Of these projects, the most common type of project was clinical audit (46%). However, a wide range of other projects were also identified, including randomised trials, surveys, cohort studies, and systematic reviews. A total of 35 publications attributed to the collaboratives were retrieved. These ranged from peer-reviewed journal articles to case reports, and had achieved 181 citations in all, with a median impact factor of 2.1 and a median h-index of 5. The median number of authors on each publication was seven. These findings demonstrate the wide-ranging outputs and potential impact of trainee collaboratives.

Several advantages of trainee collaboratives have been identified. First, and from a methodological standpoint, a multicentre approach limits bias and increases the external validity of results (Bhangu et al., 2014). Second, trainee collaboratives offer the opportunity for high quality research to be carried out with few setup or running costs, and with minimal infrastructure (Mehta et al., 2017; Nepogodiev et al., 2017). While the level of input required from consultants is low, they may also benefit from adopting trainee projects into their portfolio (Skerrit and Hall, 2015). Third, high relevance to clinical practice is assured, increasing the likelihood that findings will be translated into practice and have an impact on patient outcomes (Dowswell et al., 2014; Skerrit and Hall, 2015). Finally, there are many benefits for the trainees themselves. Collaborative models make research opportunities more accessible to trainees at an earlier stage of their career, and offer trainees experience in protocol writing, gaining ethical approvals, contributing to funding applications, interaction with professionals

from other disciplines, and involvement in peer-reviewed publications (Bhangu et al., 2014; Dowswell et al., 2014; Skerrit and Hall, 2015). Equally, trainees can gain a sense of greater project ownership and widespread recognition for their efforts (Mehta et al., 2017). In combination, these strategies may serve to engrain a research culture into routine clinical practice (Bhangu et al., 2014).

Existing evidence suggests that trainee collaboratives can provide a valuable contribution to knowledge through the collection of multicentre research data, and supports the case for collaborative-led audit and research to be trialled within the field of cleft lip and/or palate (CL/P). Research within the field of CL/P has long been criticised for its lack of multicentre studies (Stock et al., 2018), and input from trainee collaboratives may therefore help to overcome this ongoing challenge. Trainee collaboratives have historically been medical in nature, while CL/P requires a wider multidisciplinary approach. Further, centralised organisation of CL/P services in the UK and the specialist knowledge of the clinicians delivering this care lends itself well to a trainee collaborative model. Although the majority of clinicians working in the field would not necessarily describe themselves as ‘trainees’, many are still developing their specialist expertise, are keen to participate in multicentre research, and wish to gain further research experience. Most collaboratives are unfunded but productive, and therefore cost effective by contributing to the evidence base/advancing knowledge without the need for grant money. Their success relies on individuals committing their time and effort to a project unpaid but in exchange for the advantages described earlier. Dividing the work and time required between the members of the collaborative reduces the time required from any individual, making a large project manageable. In particular, a collaborative approach reduces the costs and complexities of multicentre working as individuals local to each centre can contribute rather than a single researcher or research team having to travel.

The aims of the present article are to outline the establishment of a multidisciplinary collaborative in the context of CL/P, to demonstrate the Cleft Multidisciplinary Collaborative’s (CMC) successes to date; and to discuss the future potential of the CMC.

Methods

The National Institute of Health Research (NIHR) funds health and care research in the UK. As part of the NIHR structure, the NIHR Clinical Research Network (CRN) Coordinating Centre manages the provision of facilities

and people in 30 clinical specialties. One of these specialties (CRN: Children) manages fourteen Clinical Studies Groups (CSGs), the aims of which are to promote high quality research in conditions specifically affecting children. The Cleft and Craniofacial Conditions CSG was formed in May 2012, and later founded the Early Career Researcher Group (ECRG) to support and develop clinicians interested in building their research skills. The CMC was formed in April 2016 as a sub-group of the Cleft and Craniofacial Conditions ECRG (Figure 1). Building upon existing UK surgical trainee collaboratives, and expanding the remit to allow all clinical disciplines to contribute, the goal of the CMC was to develop a national network and infrastructure for delivering multicentre audit and research in the field of CL/P.

From its inception, the CMC has therefore been open to all members of the CL/P multidisciplinary team (MDT) interested in cleft-related research, including and not limited to oral and maxillofacial, Ear, Nose and Throat and plastic surgery; paediatric and restorative dentistry; orthodontics; speech and language therapy; psychology; and nursing. Additionally, clinicians with a desire to incorporate clinical research into their roles can be at any stage of their clinical career. Interested parties are advised to read and agree to the terms of reference and authorship policy. It is also recommended that if a clinician expresses an interest in becoming involved with a CMC project, they first gain the approval of their Training Programme Director and Assigned Educational Supervisor within their designated training scheme. For those invited professions who do not have this management structure in place, approval from their line manager is required. As standard, clinical participants should complete the Good Clinical Practice training provided by the NIHR prior to undertaking research.

Recruitment to the CMC is performed using a variety of methods. Initially, these included a direct email to potentially interested clinical participants, an email to the leads for the various CL/P specialist clinical networks who subsequently cascaded this information to their membership, advertisements at relevant meetings and conferences, notices posted in specialist information bulletins and websites (such as the Craniofacial Society for Great Britain and Ireland, the British Association of Plastic and Reconstructive Aesthetic Surgeons and the Association of Oral and Maxillofacial Surgery), and via social media (@cleft_collab). All of the above activities were performed once at the outset of the formation of the CMC during the spring/summer of 2016. Subsequently, deliberate recruitment activity has also been delivered via oral presentation at the annual congress of the Craniofacial Society for Great Britain and Ireland. Here, specific emphasis was placed on recruitment from non-medical/surgical and non-dental/orthodontic disciplines. Another important factor is likely to be word of mouth.

Since the CMC is unfunded, all involvement remains entirely voluntary, and with flexible participation as a fundamental principle.

Initial support and guidance is provided by senior academics and NHS consultants, with the day-to-day leadership and running of the CMC managed by a consultant 'guide' (DS). The CMC is also supported by the ECRG, which holds biannual face-to-face meetings allowing like-minded clinicians and researchers to discuss ongoing and potential projects, and to be given advice from experts on topics such as Patient and Public Involvement, gaining ethical approval and performing systematic reviews. The CMC generally operates as a project based, virtual group/network. For each project the project leaders use email, phone and web conferencing to develop the strategy and to keep in touch. Potential collaborators who have expressed interest are contacted using email to confirm their involvement and / or telephone if further discussion is required. This demonstrates that it is possible to perform multi-centre collaborative studies without direct meetings; such methodology potentially enables global initiatives. Other external support to date has been received from the Reconstructive Surgery Trials Network (www.reconstructivesurgerytrials.net); the UK-based charity, the Cleft Lip and Palate Association (www.clapa.com); and the Cleft Collective research programme (www.bristol.ac.uk/cleft-collective). Ideas for new projects can be submitted by any member of the CMC to stimulate discussion, and to collect useful feedback from the ECRG and the CSG. Ideas driven by the findings of the UK James Lind Alliance Research Priority Setting Exercise (Petit-Zeman and Cowan, 2013) for CL/P are particularly encouraged. The CMC also sits within the broader registry of the National Research Collaborative (www.nationalresearch.org.uk).

The core aims of the CMC are as follows: 1) To inspire clinicians to become involved in cleft related research; 2) To establish a clinical specialist-led national network of those disciplines involved in cleft care to conduct high quality, multi-centre research, audit and quality improvement projects; 3) To provide a channel of communication and continuity allowing clinical participants to develop their involvement in research and audit projects as they undertake their training or develop their clinical careers; 4) To provide opportunities to develop research skills for the educational benefit of its members; 5) To attract national funding for research and audit work; 6) To foster a collaborative spirit for national and international cleft-related projects; and 7) To keep clinical participants up to date with the latest research developments from the international community.

Results

To date, 48 clinicians have expressed an interest in being involved in the CMC. These clinical participants represent all of the 17 specialist cleft teams in the UK, and encompass the following disciplines: plastic surgery ($n = 16$), oral and maxillofacial surgery ($n = 15$), restorative dentistry ($n = 7$), paediatric dentistry ($n = 4$), orthodontics ($n = 3$), research nursing ($n = 1$), speech and language therapy ($n = 1$) and general medicine ($n = 1$). The CMC has been involved in two major projects thus far. The first of these was the collection of CL/P phenotype data to support the Cleft Collective Cohort Studies. The Cleft Collective is the world's largest CL/P research programme, an initiative of the Scar Free Foundation and supported entirely by voluntary donations. At the core of the Cleft Collective programme are two national cohort studies, with the overarching aim of investigating the biological and environmental causes of CL/P, the best treatments for CL/P, and the psychological impact of CL/P on those affected and their families (Stock et al., 2016). With data being collected from a variety of sources, and across a wide geographical area, some gaps in data collection are unavoidable. The CMC therefore took the opportunity to assist the Cleft Collective to collect phenotype data for those participants who had explicitly consented to their data being collected from medical records. This project will enable the Cleft Collective to backfill missing baseline phenotypes, and will help to validate phenotypic data already collected from different sources. The Cleft Collective developed a secure online method of entering and transferring this missing data, which were retrieved directly from medical records by members of the CMC. Following a pilot conducted in two UK cleft teams, this project was rolled out nationally across ten CL/P surgical sites, and achieved a data collection rate of between 68 and 100 percent. Due to the phased set-up of the Cleft Collective cohort studies, the amount of data requested at each surgical site varied. Data collected from this project will be cross-referenced with those data already held by the Cleft Collective to ensure accuracy. It is hoped that data collected by other relevant organisations can also be cross-referenced.

The CMC's second major project is a systematic review of factors influencing outcomes for velopharyngeal competence following cleft palate (CP) repair (PROSPERO project number: 51624). Velopharyngeal competence that supports normal speech production and the absence of fistula formation are early principal outcome measures following CP repair (Smith and Losee, 2014). The objective of this systematic review is therefore to identify which pre-operative factors may support or hinder velopharyngeal competence following the primary CP surgery. This review is currently in process and will be documented soon.

Discussion

The aims of the present article were to outline the establishment of a multidisciplinary collaborative in the context of CL/P, to demonstrate the CMC's successes to date; and to discuss the future potential of the CMC. While the CMC is still in its infancy, the authors believe that the appetite for, and the concept of a multidisciplinary collaborative in the field of CL/P has been proven, through the generation of a UK-wide network of committed clinicians and researchers, and the successful delivery of two major research projects.

The CMC model offers an appealing antidote to numerous research-based challenges in CL/P, including the bias inherent in single-centre research, and high running costs (Bos et al., 2007; Bhangu et al., 2014; Mehta et al., 2017; Nepogodiev et al., 2017), as well as several benefits to consultants, large research projects, clinical practice and the clinical participants themselves (Bhangu et al., 2014; Dowswell et al., 2014; Skerrit and Hall, 2015; Mehta et al., 2017). The CMC offers opportunities for clinicians to learn and develop key research skills and to complete research projects that will result in tangible outputs. Some are now undertaking formal research training via a higher degree, in order to enhance their qualifications and improve their job prospects in a competitive market. This is especially important given that research activities are often subsidiary to clinical commitments, particularly in light of recent reforms such as the European Working Time Directive (Chapman et al., 2014). Not only do the CMC's efforts have the potential to impact directly upon patient care, but they also benefit the clinician for the remainder of their career through improved critical appraisal skills, continued desire for academic enquiry and a passion for research; success which can then propagate. Additional support from initiatives such as the CSG and ECRG help to demonstrate what a clinical academic career can offer and provide several stages of peer review and quality control to projects undertaken by the CMC.

Establishing a wide geographical network of collaborators is not without its challenges (Skerrit and Hall, 2015), and engaged, enthusiastic participants are prerequisites for a successful collaborative (Dowswell et al. 2014). In the case of the CMC's inaugural study, which collected phenotype data for the Cleft Collective research programme, the infrastructure was already established, and a support network already in place. Dowswell and colleagues (2014) describe the requirement of a "critical mass", and similar parallels can be seen with our experience of this study. While this was a relatively simple data collection task, it was vital to the future capability of the Cleft Collective programme to deliver meaningful results. Further, this inaugural study, in addition to the

second project carried out by the CMC, demonstrated a clear potential for professional development and tangible outputs.

An ongoing priority for the CMC will be to increase the representation of disciplines other than medicine/surgery and dentistry/orthodontics in the CMC. Speech and language therapists, psychologists, nurses, and geneticists, as well as a number of other allied professions are all vital members of the clinical teams, and would bring a more holistic approach to the investigations carried out by the CMC; a unique feature of this collaborative when compared to previous trainee collaboratives. Discussions with members of these professions who are currently working in a junior specialist capacity suggest there is no lack of enthusiasm, but that a different professional development structure exists, in which expectations of, and capacity for research activity is more limited. A move toward a more ingrained research culture and the ongoing support of managers will be needed, in order to release clinical members of staff to participate in research activity. Some representation of speech and language therapy and nursing is currently evident within the CMC, and therefore identifying the ways in which these individuals have been successful in their involvement will inform future efforts to encourage more diversity. While the CMC works to encourage greater involvement from the other professions in the MDT, consultation/feedback on specific projects from many of these professions can be sought from the CSG and ECRG where they are currently better represented.

As the CMC continues to gain momentum, it is hoped that funding will become available to support future activities. Elsewhere, trainee-led trials are beginning to attract funding from organisations such as the NIHR, and the CMC is determined to continue this tradition. Furthermore, the CMC hopes to utilise the current impetus among the CL/P community to establish a more ingrained research culture, and to inspire full MDT involvement in future studies. In addition, the authors hope to capitalise on the momentum generated thus far, and aspire to create a global collaborative of clinical participants interested in CL/P and other craniofacial research. The authors would therefore welcome interest from professionals around the globe who are currently developing a specialism within cleft and craniofacial care.

Conclusions

The concept of, and the enthusiasm for a multidisciplinary collaborative in the field of CL/P has been demonstrated, through the generation of a UK-wide network of committed clinicians and researchers, and the

successful delivery of two major research projects to date. With ongoing support from other organisations and networks, the CMC offers a platform from which clinicians, consultants, existing research projects and patients can benefit. The development of a global collaborative is a key future ambition.

References

- Bhangu A, Kolias AG, Pinkney T, Hall NJ, Fitzgerald JE. Surgical research collaboratives in the UK. *Lancet*. 2013;382:1091–1092.
- Bos N, Zimmerman A, Olson J, Yew J, Yerkie J, Dahl E, Olson G. From shared databases to communities of practice: A taxonomy of collaboratories. *J Comp-Mediated Comm*. 2007;12:652-672.
- Chapman SK, Bhangu A, Fitzgerald JE. European Working Time Directive: More harm than good for surgical trainees. *BMJ*. 2014;349:g7323
- Dowswell G, Bartlett DC, Futaba K, Whisker L, Pinkney TD, West Midlands Research Collaborative. How to set up and manage a trainee-led research collaborative. *BMC Med Educ*. 2014;14:94.
- Jamjoom AA, Phan PN, Hutchinson PJ, Kolias AG. Surgical trainee research collaboratives in the UK: an observational study of research activity and publication productivity. *BMJ Open*. 2016;6:p.e010374.
- Kolias AG, Cowie CJ, Tarnaris A, Hutchinson PJ, Brennan PM, British Neurosurgical Trainee Research Collaborative. Ensuring a bright future for clinical research in surgery with trainee led research networks. *BMJ*. 2013;21:f5225.
- Mehta N, Williams RJ, Smith ME, Hall A, Hardman JC, Cheung L, Ellis MP, Fussey JM, Lakhani R, McLaren O, et al. Can trainees design and deliver a national audit of epistaxis management? A pilot of a secure web-based audit tool and research trainee collaboratives. *J Laryngol Otol*. 2017;131:518-522.
- Nepogodiev D, Chapman SJ, Kolias AG, Fitzgerald JE, Lee M, Blencowe NS, Chari A, Jamjoom A, Kasivisvanathan V, D'Auria M, et al. The effect of trainee research collaboratives in the UK. *Lancet Gastroenterol Hepatol*. 2017;2:247-248.
- Petit-Zeman S, Cowan K. Patients/carers and clinicians can set joint priorities for research in cleft lip and palate. *Int J Pediatr Otorhinolaryngol*. 2013;77:309-310.
- Skerritt C, Hall NJ. The Value of Trainee Networks in Pediatric Surgical Research. *Eur J Pediatr Surg*. 2015;25:504-508.

Smith DM, Losee JE. Cleft palate repair. *Clin Plast Surg*. 2014;41:189–210.

Stock NM, Feragen KB, Moss TP, Rumsey N. Toward a conceptual and methodological shift in craniofacial research. *Cleft Palate Craniofac J*. 2018;55:105-111.

Stock NM, Humphries K, St Pourcain B, Bailey M, Persson M, Ho KM, Ring S, Marsh C, Albery L, Rumsey N, et al. Opportunities and challenges in establishing a cohort study: An example from cleft lip/palate research in the United Kingdom. *Cleft Palate Craniofac J*. 2016;53:317-325.