Centralization of cleft care in the UK. Part 6: a tale of two studies

A. R. Ness
A. K. Wills
A. Waylen
R. Al-Ghatam
T. E. M. Jones
R. Preston
A. J. Ireland
M. Persson
J. Smallridge
A. J. Hall
D. Sell
J. R. Sandy

Authors’ affiliations:
A. R. Ness, D. Sell, National Institute for Health Research (NIHR) Biomedical Research Unit in Nutrition, Diet and Lifestyle at the University Hospitals Bristol NHS Foundation Trust and the University of Bristol, Bristol, UK
A. R. Ness, A. K. Wills, A. Waylen, R. Al-Ghatam, T. E. M. Jones, A. J. Ireland, J. R. Sandy, School of Oral and Dental Sciences, University of Bristol, Bristol, UK
R. Preston, Cleft Lip and Palate Association, London, UK
M. Persson, Centre for Appearance Research, University of the West of England, Bristol, UK
J. Smallridge, South Thames’ Cleft Unit, Guy’s and St Thomas Hospital, London, UK
J. Smallridge, Cleft Net East Cleft Network, Addenbrooke’s Hospital, Cambridge, UK
A. J. Hall, Children’s Hearing Centre, University Hospitals Bristol NHS Foundation Trust, Bristol, UK

Structured Abstract

Objectives – We summarize and critique the methodology and outcomes from a substantial study which has investigated the impact of reconfigured cleft care in the United Kingdom (UK) 15 years after the UK government started to implement the centralization of cleft care in response to an earlier survey in 1998, the Clinical Standards Advisory Group (CSAG).

Setting and Sample Population – A UK multicentre cross-sectional study of 5-year-olds born with non-syndromic unilateral cleft lip and palate. Data were collected from children born in the UK with a unilateral cleft lip and palate between 1 April 2005 and 31 March 2007.

Materials and Methods – We discuss and contextualize the outcomes from speech recordings, hearing, photographs, models, oral health and psychosocial factors in the current study. We refer to the earlier survey and other relevant studies.

Results – We present arguments for centralization of cleft care in healthcare systems, and we evidence this with improvements seen over a period of 15 years in the UK. We also make recommendations on how future audit and research may configure.

Conclusions – Outcomes for children with a unilateral cleft lip and palate have improved after the introduction of a centralized multidisciplinary
service, and other countries may benefit from this model. Predictors of early outcomes are still needed, and repeated cross-sectional studies, larger longitudinal studies and adequately powered trials are required to create a research-led evidence-based (centralized) service.

Key words: Cleft Lip; Cleft Palate; Cross-Sectional Studies

Introduction

In this supplement, we report on the results of a UK-wide cross-sectional survey of 5-year-old children with unilateral cleft lip and palate conducted between January 2011 and December 2012 – Cleft Care UK (CCUK) (1–5). We attempted as far as possible to follow the design of a previous survey conducted fifteen years ago that recruited from a similar target population of children with unilateral cleft lip and palate – the Clinical Standards Advisory Group (CSAG) study (6–11). Much has changed since this first survey was conducted. The 57 centres providing care for children with cleft lip and palate have reduced to 11 centres or managed clinical networks. Care in these centres is provided by multidisciplinary teams, and the surgeons in these teams operate on at least 35 cases each year (12–14). Preliminary studies suggest that outcomes have improved but these reports either were regional (rather than national), were specific to a single outcome or had incomplete data (15–17). We report here the results of a comprehensive nationwide survey and directly compare these results with those prior to centralization.

A tale of two studies

We were not able to replicate the previous survey exactly. We used the same inclusion criteria and measured the same attributes in the study children. But the children we studied fifteen years later were younger (despite using the same target age range), and some of the approaches to measurement have changed. Orthodontists now sometimes use photographs in place of study models, speech and language therapists assess speech using a modified protocol, and psychologists have changed the questions they ask. The similarities and differences between the two studies are summarized in Table 1 and described in more detail in the first paper in this supplement (1). Despite these differences, we believe that these studies are similar enough to allow us to describe changes in care and outcome over this time frame and thus to evaluate the impact of the move to a centralized multidisciplinary service.

Summary of findings

The treatment offered to children has changed over the last 15 years. The range of surgical procedures used is less varied, hearing aids are used more often, and grommets placed less frequently. Overall outcomes have improved. There have been marked improvements in dento-alveolar arch relationships and in speech, whereas the prevalence of dental caries and hearing loss is unchanged. These differences are summarized in Table 2 and described in more detail in the results papers in this supple-
ment (2–5). Though much improved, comparative data from other centres on dento-alveolar arch relationships and speech outcomes suggest these are still not as good as those achieved in the best centres in Europe (18–20). Further, there are still a proportion of children who do badly with up to 20% having poor results for important outcomes such as dento-alveolar arch relationships and intelligibility/distinctiveness of speech. The numbers with poor outcomes are summarized in Table 3 and described in more detail in the results papers in this supplement (2–5).

Other benefits of this programme of cleft research

This second cross-sectional survey was part of a larger programme of work funded by the UK National Institute for Health Research (NIHR). We ran a series of research workshops to design the study that also triggered a James Lind Alliance initiative (21) and a health talk project (22) in children with cleft lip and palate. We completed several systematic reviews on treatment for children with cleft lip and palate (23–25). We conducted a survey of cleft centres that has described the service provision (12, 13) and the process of centralization (26). We have been able to offer training opportunities. Three students have already completed taught doctorates using data collected as part of this cross-sectional survey and the survey of the centres. We have strengthened patient and public involvement among people with cleft by disseminating our work through the Cleft Lip and Palate Association and by running a workshop focussing on patient involvement. Our study (and programme) has thus paved the way for future

| Table 1. Comparison of methods and demographics between the Cleft Care UK (CCUK) study and the Clinical Standards Advisory Group (CSAG) study |
|---------------------------------|-----------------|-----------------|
|                                 | CCUK 2012       | CSAG 1998       |
| **Methods**                     |                 |                 |
| Type of activity                | Research        | Audit           |
| People collecting key outcomes  | Mainly local teams | Study team     |
| **Measures**                    |                 |                 |
| Appearance                      | Better quality digital images | Photographs |
| Dento-alveolar arch relationships | Study models and photographs | Study models |
| Oral health                     | British Association for the Study of Community Dentistry (BASCD) calibrated dental examination by consultant paediatric dentist | BASCD calibrated dental examination by an orthodontist |
| Hearing                         | Pure tone audiometry, tympanometry and otology assessment | Pure tone audiometry and otology assessment |
| Speech assessment               | Cleft Audit Protocol for Speech – Augmented (CAPS-A) | Cleft Audit Protocol for Speech (CAPS) |
| Psychology assessment           | Modified psychosocial questionnaire, 8 items; self-confidence response was 0–10 (0 = very negative effect; 5 = no difference; 10 = very positive effect) | Psychosocial questionnaire, 18 items; self-confidence response was yes/no |
| **Demographics**                |                 |                 |
| Year of birth                   | 2005–2007       | 1989–1991       |
| Eligible                        | 359             | 326             |
| Number recruited and response rate | 268 (75%)     | 239 (73%)       |
| Age (median and interquartile range) | 5.5 (5.4–5.7)  | 6.4 (5.9–6.9)  |
| Number of boys (percentage)     | 181 (67.5%)     | 159 (66.5%)     |
randomized trials and observation studies by reviewing the evidence and building research capacity in cleft centres in the UK.

**Implications for practice**

Our data show that a centralized multidisciplinary service improves outcomes albeit that some areas of cleft care still require improvement. This study will provide evidence for cleft teams to argue with commissioners for increased resource. In our view, centralized multidisciplinary services should be introduced in all countries. Local comprehensive surveys are not required to justify or guide this change. Earlier UK outcomes described in the original CSAG study were poor, and it was argued that this was because there was no centralized service. This does not prove at all that small centres with a low case load but very well-organized care are likely to have poor outcomes. The issue is that these small centres will have great difficulty in proving the quality of their outcomes because of a lack of statistical power. These arguments have been well rehearsed elsewhere with strong evidence to counter the continuation of low volume operating (10). However, it is not clear what the key component (or components) of centralization is. Is it the improvements in surgical training? Is it the increase in number of operations? Is it the implementation of multidisciplinary team working? Is it the creation of an audit culture that encourages reflective practice? Further analyses of the data in these two cross-sectional studies and future studies may refine our understanding, but this should not delay plans to rationalize services. It is also unclear how we should monitor outcome post-centralization. It would be useful to have process measures that predict outcome at age five, or earlier outcome measures that can be used before the age of

**Table 2. Comparison of good outcomes between the Cleft Care UK (CCUK) study and the Clinical Standards Advisory Group (CSAG) study**

|                      | CCUK | CSAG | P-value |
|----------------------|------|------|---------|
| Structural outcomes  |      |      |         |
| Facial appearance (% good or excellent) | 36   | 32   | 0.107*  |
| Dento-alveolar relationships (% good or excellent) | 53†  | 30   | <0.001† |
| Oral health          |      |      |         |
| Caries-free (dmft = 0) (%) | 48   | 45   | 0.6†    |
| Hearing              |      |      |         |
| None or mild hearing loss in better ear (%) | 78   | 79   | 0.7†    |
| Speech               |      |      |         |
| No hypernasality (%) | 90   | 82   | 0.018†  |
| Intelligibility/distinctiveness (% normal) | 56   | 20   | <0.001† |
| Psychosocial outcome |      |      |         |
| Child’s self-confidence not affected (%) | 92   | 81   | <0.001† |

*Mixed effects logistic regression to account for the non-independence of observations from multiple observers.
†This is based on the 198 children with models as the 49 children with photographs were excluded.
‡Chi-squared test.

**Table 3. Comparison of poor outcomes between the Cleft Care UK (CCUK) study and the Clinical Standards Advisory Group (CSAG) study**

|                      | CCUK | CSAG | P-value |
|----------------------|------|------|---------|
| Structural outcomes  |      |      |         |
| Facial appearance (% poor or very poor) | 22   | 28   | 0.013*  |
| Dento-alveolar relationships (% poor or very poor) | 19†  | 36   | <0.001‡ |
| Oral health          |      |      |         |
| Caries present (dmft ≥1) (%) | 52   | 55   | 0.6‡    |
| Hearing              |      |      |         |
| Moderate or worse hearing loss in better ear (%) | 22   | 21   | 0.7‡    |
| Speech               |      |      |         |
| Hypernasality (%)    | 10   | 18   | 0.018‡  |
| Intelligibility/distinctiveness (% just intelligible or less) | 17   | 19   | 0.6‡    |
| Psychosocial outcome |      |      |         |
| Child’s self-confidence affected (%) | 8    | 19   | <0.001‡ |

*Mixed effects logistic regression to account for the non-independence of observations from multiple observers.
†This is based on the 198 children with models as the 49 children with photographs were excluded.
‡Chi-squared test.
5 years or both. In the UK, we rely on data collected at audit clinics when the child is 5 years old. These data therefore reflect outcome sometime after the primary surgery, and the numbers treated in any one centre are small. Obtaining standardized measures from all centres routinely is challenging and expensive (17). In our view, the best approach is to encourage audit clinics locally (where these do not already happen) that provide training and encourage review of personal practice – this will certainly be able to detect extreme variations in outcome – and to carry out intermittent comprehensive surveys nationally that describe care and outcomes. It is essential to continue to participate in international comparative studies and to be mindful of the better European centres where care is still being delivered at higher standards with better outcomes.

Implications for research

We plan to conduct further analyses of these data to look at the impact of centre characteristics such as size, time to centralization, surgical throughput and multidisciplinary working on outcome. We also intend to look at predictors of specific outcomes in an attempt to quantify the role of individual and treatment factors that predict both good and poor outcomes. We will also describe the costs incurred by families and people’s choice preferences. This cross-sectional study is a resource, and we are trying to encourage future collaborations to ensure it is fully exploited. We are currently creating a detailed data dictionary and formalizing access arrangements. A further national cross-sectional survey should be considered in 5–10 years to confirm that there have been further improvements in service provision and outcome such that cleft care in the UK is the best in Europe. Longitudinal studies to describe trajectories of children with cleft and to identify early outcome measures or predictors of outcome would be valuable. Well-designed adequately powered trials informed by the priorities identified through the recent James Lind Alliance initiative should be conducted (21). There is also now an opportunity for the cleft teams in the UK to participate in larger international studies of outcomes and to participate in clinical trials. These collaborations are key with a low incidence anomaly as all aspects of aetiology and care will benefit from increasing sample sizes. The diversity of genetic and environmental factors in the causation of clefting will only be explored through multicentre collaborations and international epidemiological approaches.

The challenges of clinical research in cleft

The evidence base to inform treatment of children with cleft lip and palate is limited. There are few well-designed adequately powered randomized trials or prospective observational studies. Clinical research should be easier in a centralized service particularly in a state-funded health system on a small heavily populated island like the UK. We were able to recruit children to this study within a narrower age range than previously, but our response rates were similar. Furthermore, we faced challenges trying to collect data that were directly comparable with the previous study. Families do have further to travel to reach a cleft centre, and this may have reduced their willingness to come to clinic and to complete questionnaires. Interestingly, families did not report that it is any more difficult to attend the cleft centre (5). We decided to run this project as research rather than audit. This allowed us to obtain consent to carry out follow-up through record linkage, to collect additional data, to standardize and control data quality and to be able to contact families about participation in other studies. We had to navigate research approval processes which led to delays (27), we incurred extra costs, and we had to obtain consent from the parents and assent from the children. Few families declined to participate in the research project, but response rates for self-completed questionnaires were disappointing. So, our experience suggests that a centralized service does make it easier to
Conclusions

Outcomes for children with a unilateral cleft lip and palate have improved after the introduction of a centralized multidisciplinary service. This process of centralization should be introduced in other countries with a less centralized service for children with clefts. Further analyses will be conducted to explore centre-level effects and individual and treatment factors that influence outcome. There is a need for earlier outcome measures or predictors of outcome that could be used to audit practice and monitor service quality in a more timely fashion. Repeated cross-sectional studies, larger longitudinal studies and adequately powered trials are required to create the research-led evidence-based (centralized) service that children born with cleft lip and palate deserve.

Acknowledgements: We would like to thank the families that took part in this study. We would also like to thank the clinicians and staff in the cleft centres who supported this project. This publication presents independent research commissioned by the National Institute for Health Research (NIHR) under its Programme Grants for Applied Research scheme (RP-PG-0707-10034). The views expressed in this publication are those of the author(s) and not necessarily those of the NHS, the NIHR or the Department of Health.

References
1. Persson M, Sandy JR, Waylen A, Wills AK, Al-Ghatam R, Hall A et al. A cross-sectional survey of five-year old children with non-syndromic unilateral cleft lip and palate: cleft Care UK. Orthod Craniofac Res 2015;18(S2):1–13.
2. Al-Ghatam R, Jones TEM, Ireland AJ, Atack NE, Chawla O, Deacon S et al. Structural outcomes in the cleft care UK study – dentofacial outcomes. Orthod Craniofac Res 2015;18(S2):14–24.
3. Smallridge J, Hall AJ, Chorbaki R, Parfect V, Persson M, Ireland A et al. Functional outcomes in the Cleft Care UK study: oral health and audiology. Orthod Craniofac Res 2015;18(S2):25–35.
4. Sell DA, Mildinhall S, Albery L, Wills AK, Sandy JR, Ness AR. Perceptual speech outcomes in the Cleft Care UK study. Orthod Craniofac Res 2015;18(S2):36–46.
5. Waylen A, Ness AR, Wills AK, Persson M, Rumsey N, Sandy JR. Child psychosocial outcomes and satisfaction with cleft services in the Cleft Care UK study. Orthod Craniofac Res 2015;18(S2):47–55.
6. Clinical Standards Advisory Group. Cleft Lip and/or Palate, HSMO, ISBN 0-11-322103-7 London, 1998.
7. Sandy JR, Williams AC, Bearn D, Mildinhall S, Murphy T, Sell D et al. Cleft lip and palate care in the United Kingdom—the Clinical Standards Advisory Group (CSAG) Study. Part 1: background and methodology. Cleft Palate Craniofac J 2001;38:20–3.
8. Williams AC, Bearn D, Mildinhall S, Murphy T, Sell D, Shaw WC et al. Cleft lip and palate care in the United Kingdom—the Clinical Standards Advisory Group (CSAG) Study. Part 2: dentofacial outcomes and patient satisfaction. Cleft Palate Craniofac J 2001;38:24–9.
9. Sell D, Grunwell P, Mildinhall S, Murphy T, Cornish TA, Bearn D et al. Cleft lip and palate care in the United Kingdom—the Clinical Standards Advisory Group (CSAG) Study. Part 3: speech outcomes. Cleft Palate Craniofac J 2001;38:30–7.
10. Bearn D, Mildinhall S, Murphy T, Murray JJ, Sell D, Shaw WC et al. Cleft lip and palate care in the United Kingdom—the Clinical Standards Advisory Group (CSAG) Study. Part 4: outcome comparisons, training, and conclusions. Cleft Palate Craniofac J 2001;38:38–43.
11. Williams AC, Sandy JR, Thomas S, Sell D, Sterne JA. Influence of surgeon’s experience on speech outcome in cleft lip and palate. Lancet 1999;354:1697–8.
12. Scott JK, Leary SD, Ness A, Sandy J, Persson M, Kilpatrick N et al. Centralisation of services for children born with orofacial clefts in the United Kingdom: a cross sectional survey. Cleft Palate Craniofac J 2014;51:e102–9.
13. Scott JK, Leary SD, Ness AR, Sandy JR, Persson M, Kilpatrick N et al. Perceptions of team members working in cleft services in the United Kingdom: a pilot study. Cleft Palate Craniofac J 2015;52:e1–7.
14. Fitzsimons KJ, Mukarram S, Copley LP, Deacon SA, van der Meulen JH. Centralisation of services for children with cleft lip or palate in England: a study of hospital episode statistics. BMC Health Serv Res 2012;12:148.
15. Hathorn IS, Atack NE, Butcher G, Dickson J, Durning P, Hammond M et al. Centralisation of services: standard setting and outcomes. Cleft Palate Craniofac J 2006;43:401–5.
16. Revington PJ, McNamara C, Mukarram S, Perera E, Shah HV, Deacon SA. Alveolar bone grafting: results of a national outcome study. *Ann R Coll Surg Engl* 2010;92:643–6.

17. Fitzsimons K, Deacon S, Copley L, van der Meulen J. *CRANE Database: Annual Report on cleft lip and/or palate* 2013. www.crane-database.org.uk.

18. Sinko K, Caacbay E, Jagsch R, Turhani D, Baumann A, Mars M. The GOSLON yardstick in patients with unilateral cleft lip and palate: review of a Vienna sample. *Cleft Palate Craniofac J* 2008;45:87–92.

19. Lilja J, Mars M, Elander A, Enocson L, Hagberg C, Worrell E et al. Analysis of dental arch relationships in Swedish unilateral cleft lip and palate subjects: 20-year longitudinal consecutive series treated with delayed hard palate closure. *Cleft Palate Craniofac J* 2006;43:606–11.

20. Lohmander A. Surgical intervention and speech outcomes in cleft lip and palate. In: Howard S, Lohmander A, editors. *Cleft Palate Speech*. Chichester: John Wiley & sons; 2011. pp. 55–85.

21. Petit-Zeman S, Cowan K. Patients/carer and clinicians can set joint priorities for research in cleft lip and palate. *Int J Paediatr Otorhinolaryngol* 2013;77:309–10.

22. Healthtalk.org. Cleft lip and palate. [Online] Available from: http://www.healthtalk.org/peoples-experiences/pregnancy-children/cleft-lip-and-palate/topics [Accessed February 2015].

23. Bessell A, Sell D, Whiting P, Roulstone S, Albery L, Persson M et al. Speech and language therapy interventions for children with cleft palate: a systematic review. *Cleft Palate Craniofac J* 2013;50: e1–17.

24. Norman A, Persson M, Stock N, Rumsey N, Sandy J, Waylen A et al. The effectiveness of psychosocial intervention for individuals with cleft lip and/or palate and their parents: a systematic review. *Cleft Palate Craniofac J* 2015;52:301–10.

25. Pepper H, Perry R, Ness A, Sandy J, Thomas S. A Systematic review of skin closure techniques in primary cleft lip surgery. *Oral Surgery* 2015;8:10–7.

26. Searle A, Scott J, Sandy J, Ness AR, Waylen A. Clinical directors’ views of centralisation and commissioning of cleft services in the UK. *BMC Oral Health* 2015;22:12.

27. Sandy J, Kilpatrick N, Persson M, Bessel A, Waylen A, Ness A et al. Why are multi-centre clinical observational studies still so difficult to run? *Br Dent J* 2011;211:59–61.

28. Bonevski B, Randell M, Paul C, Chapman K, Twyman L, Bryant J et al. Reaching the hard-to-reach: a systematic review of strategies for improving health and medical research with socially disadvantaged groups. *BMC Med Res Methodol* 2014;14:42.

29. Sandy J, Rumsey N, Persson M, Waylen A, Kilpatrick N, Ireland T et al. Using service rationalisation to build a research network: lessons from the centralisation of UK services for children with cleft lip and palate. *Br Dent J* 2012;212:553–5.