Institution: University of Bristol

Unit of Assessment: 3

Title of case study: Improving care for children born with cleft lip and palate in the UK and beyond

1. Summary of the impact (indicative maximum 100 words)

Around 1,000 children are born each year in the UK with cleft lip and/or palate. They need treatment from a range of clinical specialties. Bristol co-led a research survey commissioned by the Clinical Standards Advisory Group (CSAG) which was published in 1998. This informed a process of centralisation which by 2008 had reduced the number of centres from 57 to 11. The process of centralisation and service configuration has continued through 2008-13 leading to closer multi-disciplinary working, increased cases operated on per surgeon, less variation in surgical techniques and shorter stays in hospital. The impact for the individual is improved facial appearance, speech and psychological adjustment. This centralisation of care has resulted in care quality similar to the better cleft centres in Europe. Bone repairs to the palate were 85% successful in 2010 compared to only 58% in 1998 and there was poor jaw growth (linked to quality of surgery) in only 18% of cases in 2013 compared to 37% in 1998. The reach of this study has extended to several European countries and America.

2. Underpinning research (indicative maximum 500 words)

Cleft lip and palate is a common congenital anomaly in humans, with issues that relate to appearance, speech, hearing, general health and social integration requiring a range of healthcare professional skills. In the late 1980's and early 1990's there were indications that care for children born with a cleft lip and palate in the UK was varied and often suboptimal. Based on this, the Royal College of Surgeons Audit Unit and the Standing Dental Advisory Committee made appeals to the Department of Health that a Clinical Standards Advisory Group (CSAG) enquiry be established to determine national levels of care for those born with a cleft. The CSAG committee commissioned a research team to carry out a survey.

Logistics of the research survey

The research team was co-led by Jonathan Sandy (Professor in Orthodontics at U. Bristol, 1991 onwards) and Bill Shaw (Professor in Orthodontics at U. Manchester), both with track records in cleft research. The research described the care and outcome in non-syndromic cases of unilateral cleft lip and palate (UCLP) aged five and twelve years throughout the UK treated over a two year period. Information was collected on the process of care and key outcomes were measured including jaw growth, bone grafting, facial appearance, speech, hearing, dental health and child/parent satisfaction. Thus high quality research was used to inform decisions on the service. At that time there were 57 cleft centres operating on these children which required the researchers to drive a total of 40,000 miles to collect all of the data. Two part time research registrars (Alison Williams [RA, PhD student and Clinician Scientist Award at U. Bristol, 1996-2003] and David Bearn [Manchester] and two part time speech and language therapists (Sue Mildinhall [RA at Bristol 1995 -1997] and Terrie Murphy [Manchester] were appointed to collect and analyse the data. Bristol wrote the grant, developed the protocol, collected half of the data and led on the analysis and publication of the data.

Outcomes and cleft care in the UK

The research survey published in 1998 showed for the first time that the outcomes nationally were sub-optimal [1]. For example, poor growth of the top jaw was present in 37% of five year olds and 39% of twelve year olds. The implication of this finding was that around 40% of children were likely to need an operation to break, and move, the top jaw forward to correct an underlying discrepancy between the top and bottom jaws [2]. This compared with around 4% in children treated in leading European centres where this growth was better. Another example of poor outcome was seen in bone grafting in the upper jaw. This is a procedure undertaken around the age of 11 to unite the divided upper jaw and allow eruption of adjacent permanent teeth. There are very few, if any, children with a UCLP who would not be offered this operation. However, 16% of the UK twelve year olds had not received this bone graft, and of those who had a graft, only 58% were successful compared to a 92% success rate in some of the better European centres [2].

The predominance of low volume operators (nearly 60% of surgeons were dealing with only one

UCLP case per year) and overall poor quality of results limited the detailed exploration of associations between volume and outcome but the survey suggested the more operations a surgeon did the better the results. There was thus evidence that the service needed to be centralised. The results were published as a report [1] (can be supplied upon request) and a series of papers [2, 4-6] in the only specialist journal for cleft clinicians and one in a leading general medical journal [3]. A grant was subsequently awarded (2009) by the NIHR to Andrew Ness (Professor in Epidemiology at U. Bristol, 2007 onwards) to examine the impact of centralisation of cleft services. Data collection is now complete and early analysis of poor growth of the top jaw (which was present in 37% of five year olds in the original CSAG) suggests this has fallen by 50%.

3. References to the research (indicative maximum of six references)

Outputs

[1] Clinical Standards Advisory Group Cleft Lip and/or Palate, 1998; HMSO, ISBN 0-11-322103-7. *Research grant funded by the Department of Health. Can be supplied upon request

[2] Cleft lip and palate care in the United Kingdom (UK) - The Clinical Standards Advisory Group (CSAG) Study: Part 2 - Dentofacial outcomes, psychosocial status and patient satisfaction. Williams AC, Bearn D, Mildinhall S, Murphy T, Sandy JR, Sell D, and Shaw WC. Cleft Palate Craniofac J. 2001;38:24-29. DOI: 10.1597/1545-1569(2001)038<0024:CLAPCI>2.0.CO;2.

[3] Influence of surgeon's experience on speech outcome in cleft lip and palate. Williams AC, Sandy JR, Thomas S, Sell D, Sterne JA. Lancet. 1999; 354:1697-8. DOI: 10.1016/S0140-6736(99)03748-4. *Demonstration that the more surgery you do the better the results

[4] Cleft lip and palate care in the United Kingdom (UK) - The Clinical Standards Advisory Group (CSAG) Study: Part 1 - Background and methodology. Sandy JR, Williams AC, Bearn D, Mildinhall S, Murphy T, Sell D, and Shaw WC. Cleft Palate Craniofac J. 2001; 38:20-23. DOI: 10.1597/1545-1569(2001)038<0020:CLAPCI>2.0.CO;2.

[5] Cleft lip and palate care in the United Kingdom (UK) - The Clinical Standards Advisory Group (CSAG) Study: Part 3 – Speech outcomes Sell D, Grunwell P, Mildinhall S, Murphy T, Cornish TA, Bearn D, Sandy JR, Shaw WC, Williams AC. Cleft Palate Craniofac J. 2001; 38:30-37. DOI: 10.1597/1545-1569(2001)038<0030:CLAPCI>2.0.CO;2.

[6] Cleft lip and palate care in the United Kingdom--the Clinical Standards Advisory Group (CSAG) Study. Part 4: outcome comparisons, training, and conclusions. Bearn D, Mildinhall S, Murphy T, Murray JJ, Sell D, Shaw WC, Williams AC, Sandy JR Cleft Palate Craniofac J. 2001;38:38-43. DOI: 10.1597/1545-1569(2001)038<0038:CLAPCI>2.0.CO;2.

Grants

[7] JR Sandy, WC Shaw, HB Devlin, Cleft lip and palate care in Britain – current status and future direction. Department of Health – Clinical Standards Advisory Group, October 2005 - September 2007, £160,000.

[8] AC Williams, JR Sandy, B Reeves. Outcome factors in the care of children born with a cleft lip and palate. Medical Research Council, October, 1997 – September 2001, £80,000

[9] A Ness, M Birchall, M Burton, S Fisher, C Nutting, T Peters, S Rogers, C Thompson, N Rumsey, JR Sandy, S Thomas, H Worthington. Head and neck cancer, cleft lip and palate. NIHR Programme Grant January 2009 – December 2014, £1.8 million

4. Details of the impact (indicative maximum 750 words)

The CSAG committee prepared a report (Section 3, ref [1]) based on the evidence collected by the research team (co-led by Jonathan Sandy). The recommendations of this report were accepted by Ministers (Health Services Circular [HSC] 1998/002) and the Department of Health announced the establishment of the cleft implementation group (CIG) in HSC 1998/087. This was the first time that a CSAG report had been followed by a formal process of implementation and included the development and maintenance of a national database (CRANE) to record the births of children with a cleft as well as key audit points with established care indicators [a]. There were several reasons for this besides the strength and scientific robustness of the evidence collected. These included the fact that there was unanimous clinical support for service rationalisation and that the proposed changes were supported by an active and well informed user/clinician group (Cleft Lip and Palate Association, CLAPA). The speed of centralisation varied with the earliest of the nine regions reconfiguring services in 2001 and the last in 2008. The process of centralisation has continued through 2008 – 13 as centres have recruited recommended specialist staff, developed team working and set up multicentre audits. Most of the CSAG recommendations have now been implemented and care across the UK is now provided through multidisciplinary teams [b]. For the individual, this means better care resulting in improvements in facial appearance, speech and ultimately psychological adjustment. The number of surgeons responsible for repairs reduced from 98 before centralisation to 26 in 2008. Furthermore 92% of cleft units operate on at least 40 patients a year compared to an average of 6 per year before CSAG centralisation [c]. The extensive research also identified areas of care that lacked provision within the service. These deficiencies were highlighted in the CSAG report and as centralisation moved forward, strong arguments have been made to improve staff and capital resources in the newly designated centres. The nature and extent of the impact is becoming apparent with preliminary evidence that outcomes, collected through audits set up as part of the implementation of the CSAG report, have improved. Success rates for upper jaw bone grafting were 85% in 2010 compared to 58% pre-CSAG [d]. In addition, less than 20% of children have poor upper and lower jaw relations when treated within a centralised service compared to 40% pre-CSAG 40% [e]. Several geographically proximate regions have come together to run regular tri-centre audits to review outcomes. This demonstrates a commitment to review the outcomes and evaluate the service through continued audit. Early nursing input at diagnosis before or after birth has improved immeasurably post centralisation as has outreach nursing and feeding support [f].

User awareness of the impact of CSAG

In 2006 CLAPA surveyed parents of children born with a cleft between 2002 and 2005. This was similar to the survey carried out pre-CSAG in 1996. The 2007 report recognised that the changes in cleft care over the last five years mean that the needs of families and children born with clefts are better catered for than ever before. Ten years ago it was still largely other parents, under the umbrella of CLAPA, who guided and advised on some of the early care, especially around feeding. Specialist cleft nurses now provide that early care and tried and tested surgical protocols are in place meaning that babies receive surgery to a high standard, provided by a surgeon who has well documented expertise and experience [g]. A further survey of members by CLAPA in 2010 suggested that there had been further improvements in care. The survey reported that there was now a closer relationship with teams enabling patient representatives to be part of the team. Furthermore, CLAPA had helped draft the new National Service Specification as well as engaging media on issues such as late diagnosis of cleft palate [h].

Impact on service commissioning and specification in the UK

The NHS commissioning board in issuing the 2012/13 NHS standard contract (service specification D7) for Cleft Lip and Palate Services uses the 1998 CSAG report to summarise the key issues and challenges to be addressed by providers and commissioners [i]. The specification builds on the recommendations and does not replicate or supersede the original CSAG report. This formally recognises the importance of the CSAG research and its central role in policy formation. The USA national medical policy (2011) uses CSAG data to inform service configuration [j]. These latter policies are based upon a review of the available clinical information including clinical outcome studies in the peer-reviewed published medical literature and evidence-based guidelines of governmental bodies.

Global impact

The CSAG study was widely recognised as an important step in the evaluation of cleft care services including the WHO in their documentation of global strategies to reduce the health-care burden of craniofacial anomalies (2001). After CSAG, other countries became interested in conducting similar surveys. The Americleft study has produced results from one centre that are worse than any of the pre-CSAG UK centres. There is now an inevitable process of centralisation with care being moved towards University Hospitals [k]. The data from CSAG provided a basis and logic for centralisation which a number of countries used without gathering the same level of information. There is evidence in Europe that the influence of the original CSAG has led to the creation of centres of excellence in France (2013) which has been driven by the Ministry of Health (http://www.fente-labio-palatine.fr/). In Bulgaria and Romania similar service recommendations have been made with single high volume units, specialist speech therapists and national registers (2012). These decisions used the CSAG data to inform their decisions.

5. Sources to corroborate the impact (indicative maximum of 10 references)

[a] Crane Database Annual Reports, Clinical Effectiveness Unit, The Royal College of Surgeons of England London. https://www.crane-database.org.uk

Website shows that a national database for recording children born with cleft lip and palate has been established.

[b] Caring for children with clefts: UK clinical team approaches and perceptions. JK Scott.

Doctorate of Dental Science in Orthodontics 2011; University of Bristol.

*Grant funded by the NIHR.

Survey of centres showing that cleft care is provided by multidisciplinary teams and most CSAG recommendations have been implemented.

[c] Centralisation of services for children with cleft lip or palate in England: a study of hospital episode statistics. Fitzsimmons K, Mukarram S, Copley L, Deacon S, van der Meulen J. BMC Health Services Research. 2012; 12: 148-156. DOI: 10.1186/1472-6963-12-148

Report on national inpatient data showing reduced numbers of surgeons operating on children born with a cleft and an increased volume of operations carried out per surgeon.

[d] Alveolar bone grafting: results of a national outcome study. Revington PJ, McNamara C, Mukarram S, Perera E, Shah HV, Deacon SA. Ann R Coll Surg Engl. 2010; 92:643-6. DOI: 10.1308/003588410X12699663904790

Survey showing that success rates, after centralisation, have improved nationally for the operation to join the divided palate (bone grafting).

[e] Centralisation of services: standard setting and outcomes. Hathorn IS, Atack NE, Butcher G, Dickson J, Durning P, Hammond M, Knight H, Mitchell N, Nixon F, Shinn D, Sandy JR. Cleft Palate Craniofac J. 2006; 43:401-5. DOI: 10.1597/04-198.1

Report showing improvements in jaw growth in a centralised service such that operations to move the jaws to a normal relationship have fallen by 50%.

[f] Letter from Chair of the Clinical Reference Group for Cleft Lip and Palate and the Chair of the National UK NHS Cleft Development Group 2013.

Letter stating that cleft care is being examined through continued audit and that early nursing input at diagnosis before or after birth has improved immeasurably post centralisation.

[g] Regionalisation of cleft lip and palate services: has it worked? A report on users' perspectives. Cleft Lip and Palate Association. www.clapa.com

User survey reporting that specialist cleft nurses and surgeons are now in post in UK cleft centres and that they carry out specialist cleft surgery skills using similar surgical protocols.

[h] Letter from Chief Executive of CLAPA 2013.

Letter confirming patient and public involvement in service specification development and in highlighting issues in treatment and care.

[i] NHS Commissioning Board 2012/13 - Standard Contract for Acute Ambulance, Community and Mental Health and Learning Disabilities (multilateral). Section B part 1- service specifications D7, Cleft Lip and/or Palate services including Non- cleft Velopharyngeal Dysfunction.

UK Service specification that uses CSAG data to identify key issues to be addressed by providers and commissioners in developing commissioning arrangements and contracts.

[j] Cleft Palate and other Craniofacial Disorders, National Medical Policy, Cleft Palate and other Craniofacial Disorders - Policy Number: NMP113. Effective Date: June 2011.

US National policy document that used CSAG data to inform service configuration.

[k] The Americleft study: an inter-center study of treatment outcomes for patients with unilateral cleft lip and palate part 1. Principles and study design. Long RE Jr, Hathaway R, Daskalogiannakis J, Mercado A, Russell K, Cohen M, Semb G, Shaw W. Cleft Palate Craniofac J. 2011;48:239-43. DOI: 10.1597/09-180.1

A US study using methods based on CSAG to inform centralisation of cleft care and thus illustrating the reach of the work.