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Closing the loop on centralisation of cleft care in the UK

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Running title: **Centralisation of cleft care in the UK**

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Abstract

Objective: We highlight a major study which investigated the impact of reconfigured cleft care in the United Kingdom (UK) some 15 years after centralisation. We argue that centralisation as an intervention has a major impact on outcomes.

Setting: Audit clinics held in Cleft Centres in the UK.

Patients, Participants: Five year olds born between 1 April 2005 and 31 March 2007 with non-syndromic unilateral cleft lip and palate.

Interventions: Centralisation of cleft care.

Main Outcome Measure(s): We collected routine clinical measures (speech recordings, hearing, photographs, models, oral health, psychosocial factors) in a very similar way to a previous survey.

Results: We identified 359 eligible children and recruited 268 (74.7%) to the study. Overall their outcomes were better post-centralisation. There have been marked improvements in dento-alveolar arch relationships and in speech whereas the prevalence of dental caries and hearing loss are unchanged.

Conclusions: Centralised cleft care has changed UK outcomes considerably and there is no argument for returning to a dispersed model of treatment.

Key Words: cleft care; unilateral cleft lip and palate outcomes; United Kingdom

In this brief article we highlight the impact of changes in UK cleft services over a fifteen year period. In 1996, we collected data which informed the Clinical Standards Advisory Group (CSAG) report which was published in 1998 (CSAG, 1998). The organisation and outcomes of cleft care across the UK at that time were described in a series of publications (Sandy et al., 2001; Williams et al., 2001; Sell et al., 2001; Bearn et al., 2001). Subsequently the government acted to centralise cleft care by reducing the 57 cleft units down to 8-15 centres (Sandy et al., 2012). We now highlight a repeat of this study (called Cleft Care UK [CCUK]) which has had an impact on outcomes for these children.

Treatment for children with cleft lip and palate in the UK is now provided through interdisciplinary teams in 11 centres or managed clinical networks. Surgeons in these teams operate on at least 35 cases each year (Fitzsimons et al., 2012; Scott et al., 2014; Scott et al., 2015). The process of centralisation of care was variable in relation to speed and implementation with not all teams having essential team members or facilities immediately available (Scott et al., 2014). Some teams, following the recommendations of CSAG, were able to very rapidly change their personnel and structures, as well as addressing shortfalls in staffing. This was not true for all of these teams and since the initial centralisation, with increasing financial pressures some centres have found it difficult to maintain staffing. The other challenges were in reducing the number of centres and clinicians involved in cleft care (Sandy et al., 2012). Training of surgeons was also an issue (Bearn et al., 2001) and through the Royal Colleges of Surgeons, the Training Interface Group, comprising Oral and Maxillofacial Surgery, Otolaryngology and Plastic Surgery, recognised a number of national advanced training posts in cleft surgery. These posts are open to trainees of these three specialties for the last two years of their training. Despite these variations and perturbations there have been major changes in the way that care is provided in the UK and this centralised

inter-disciplinary model is effective with demonstrable improvements in outcome 15 years after centralisation took place.

To examine the impact of this change we conducted a UK-wide cross-sectional survey (CCUK) of five year old children with unilateral cleft lip and palate (UCLP), between January 2011 and December 2012 (Persson et al., 2015; Al-Ghatam et al., 2015; Sell et al., 2015; Smallridge et al., 2015; Waylen et al., 2015; Ness et al., 2015). We based the study design on the original survey. CCUK followed principles outlined in the Declaration of Helsinki with ethical approval from the Local Institutional Review Board (REC reference number: 10/H0107/33, South West 5 REC) and appropriate informed consent from participants. There were some differences in the studies despite trying to use the same methodologies, but we believe that these studies were similar enough to allow us to evaluate the impact of the centralised inter-disciplinary service (Persson et al., 2015).

We have now reported the results of this nationwide survey and compared these with the results of the previous survey. The table summarises the study demographics and outcomes. There were differences in practice between the two surveys with less variation in the types of surgical procedures used, more frequent treatment of dental caries, more hearing aids used and fewer grommets placed. Overall the treatment outcomes were better with the most obvious improvements in speech (Sell et al., 2015) and dento-alveolar relationships (Al-Ghatam et al., 2015). Fewer parents in CCUK perceived their children as having poor self-confidence than in the previous study. At least 81% of parents reported that they were happy with their child's facial features after surgery and parental satisfaction with service was high. Interestingly, families did not report that it was any more difficult to attend the cleft centre despite inevitable increased distances to travel when care is centralised (Waylen et al., 2015).

The reason for this is that many of the repeat visits for care (orthodontics, dentistry, speech and language therapy) are delivered locally and the central clinics are very focussed and decisive on future care pathways. However, not all outcomes improved, the prevalence of dental caries and hearing loss were unchanged (Smallridge et al., 2015). Furthermore, a proportion of children (approximately 20%) still have difficulties with speech and poor dento-alveolar relations. So further service improvements are required if the UK is to achieve outcomes similar to those reported from the best European centres (Lilja et al., 2006; Sinko et al., 2008; Lohmander, 2011).

In the UK, outcomes for children born with a cleft have improved as a result of the introduction of a centralised inter-disciplinary service. These improvements probably reflect a combination of better surgical training, increased number of surgical procedures per surgeon with the elimination of low volume operators (Fitzsimons et al., 2012; Scott et al., 2014; Scott et al., 2015), an investment in support staff including clinical nurse specialists, clinical psychologists and specialist speech and language therapists, and the implementation of inter-disciplinary team working. The relative importance of these factors needs clarification but this is not a reason to delay rationalising services. We believe that the argument for a dispersed model of care is no longer tenable and that centralisation should now be introduced in other countries. We would further argue that local comprehensive surveys are not required to justify or guide this change. Establishing base levels of outcomes before centralisation will delay, unnecessarily, the improvements that these children and their families can and should expect. Thereafter comprehensive national surveys that describe care and outcomes are needed to refine and monitor service improvements. These surveys can also help build research expertise and understanding in cleft teams. A research-led evidence-based (centralised) service for children born with cleft lip and palate will not only offer better

care but the infrastructure to conduct much needed adequately-powered trials and robust serial cross-sectional and longitudinal studies.

References

Al-Ghatam R, Jones TE, Ireland AJ, Attack NE, Chawla O, Deacon S, Albery L, Cobb AR, Cadogan J, Leary S et al., Structural Outcomes in the Cleft Care UK study – Dentofacial Outcomes. *Orthod Craniofac Res.* 2015; 18 Suppl 2:56-62

Attack N, Hathorn I, Mars M and Sandy J. Study models of 5 year old children as predictors of surgical outcome in unilateral cleft lip and palate. *Eur J Orthod* 1997; 19:165-70.

Bearn D, Mildinhall S, Murphy T, Murray JJ, Sell D, Shaw WC, Williams AC, Sandy JR. 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.

Clinical Standards Advisory Group Cleft Lip and/or Palate, HSMO, ISBN 0-11-322103-7 London, 1998.

Fitzsimons KJ, Mukarram S, Copley LP, Deacon SA and van der Meulen JH. Centralisation of services for children with cleft lip or palate in England: a study of hospital episode statistics. *BMC Health Services Research.* 2012; 12:148.

John A, Sell D, Sweeney T and Williams A. The Cleft Audit Protocol for Speech – Augmented: a validated and reliable measure for auditing cleft speech. *Cleft Palate Craniofac J.* 2006; 43:272-288.

Kim JB, Strike P and Cadier MC. A simple assessment method for auditing multi-centre unilateral cleft lip repairs. *J Plast Reconstr Aesthet Surg* 2011; 64:195-200.

Lilja J, Mars M, Elander A, Enocson L, Hagberg C, Worrell E, Batra P, Friede H. 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: 606-611.

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

Ness AR, Wills AK, Waylen A, Al-Ghatam R, Jones TE, Preston R, Ireland AJ, Persson M, Smallridge J, Hall AJ et al., Centralization of cleft care in the UK. Part 6: a tale of two studies *Orthod Craniofac Res*. 2015; 18 Suppl 2:56-62.

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 Suppl 2:1-13.

Sandy JR, Williams AC, Bearn D, Mildinhall S, Murphy T, Sell D, Murray JJ, Shaw WC. 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; 38:20-23.

Sandy J, Rumsey N, Persson M, Waylen A, Kilpatrick N, Ireland T, Ness A. 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: 553-555.

Scott JK, Leary SD, Ness A, Sandy J, Persson M, Kilpatrick N, Waylen A. Centralisation of services for children born with orofacial clefts in the United Kingdom: a cross sectional survey. *Cleft Palate Craniofac J.* 2014; 51:e102-109.

Scott JK, Leary SD, Ness AR, Sandy JR, Persson M, Kilpatrick N, Waylen AE. Perceptions of team members working in cleft services in the United Kingdom: a pilot study. *Cleft Palate Craniofac J.* 2015; 52: e1-7.

Sell D, Grunwell P, Mildinhall S, Murphy T, Cornish TA, Bearn D, Shaw WC, Murray JJ, Williams AC, Sandy JR. 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: 30-37.

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 Suppl 2:36-46.

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.

Smallridge J, Hall AJ, Chorbachi R, Parfect V, Persson M, Ireland AJ, Wills AK, Ness AR, Sandy JR. Functional outcomes in the Cleft Care UK study: Oral Health and Audiology. *Orthod Craniofac Res.* 2015;18 Suppl 2:25-35.

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 Suppl 2:47-55.