CLEFT REGISTRY & AUDIT NETWORK

CRANE Database

www.crane-database.org.uk

Annual Report on Cleft Lip and/or Palate 2015

On behalf of the Cleft Development Group

Royal College
of Surgeons
ADVANCING SURGICAL CARE

On behalf of the Cleft Development Group, this report was prepared by:

The Clinical Effectiveness Unit at the Royal College of Surgeons of England

Jibby Medina, Research Fellow
Kate Fitzsimons, Research Fellow
Lynn Copley, Data Manager
Scott Deacon, Clinical Project Lead
Jan van der Meulen, Clinical Epidemiologist

Clinical Effectiveness Unit
The Royal College of Surgeons of England
Email: crane@rcseng.ac.uk
Telephone: 020 7869 6610

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Abbreviations

BCLP	Bilateral cleft lip and palate
BINOCAR	British Isles Network of Congenital Anomaly Registers
CAPS-A	Cleft Audit Protocol for Speech—Augmented
CARE	Craniofacial Anomalies Register
CDG	Cleft Development Group
CEN	Clinical Excellence Network – previously referred to as Special Interest Group (SIG)
CFSGBI	Craniofacial Society of Great Britain and Ireland
CI	Confidence interval
CL	Cleft lip only
CLEFTSiS	The National Management Clinical Network for Cleft Service in Scotland
CP	Cleft palate only
CSAG	Clinical Standards Advisory Group
CRG	Clinical Reference Group
dmft	Decayed, missing and filled teeth
DfE	Department for Education
DoH	Department of Health
ENT	Ear, nose and throat
EYFSP	Early Years Foundation Stage Profile – educational assessment
GOSH	Great Ormond Street Hospital
HES	Hospital Episode Statistics
ICD-10	International Classification of Disease 10th Revision
KS1	Key Stage 1 – educational assessment
KS2	Key Stage 2 – educational assessment
MCN	Managed Clinical Network
NPD	National Pupil Database
OPCS-4	Classification of Surgical Operations and Procedures 4th Revision
PEDW	Patient Episode Data Wales
RCPCH	Royal College of Paediatrics and Child Health
SCG	Specialised Commissioning Group
SD	Standard deviation
SE	Standard error
SEN	Special Educational Needs
UCLP	Unilateral cleft lip and plate
VTCT	Vocational Training Charitable Trust
WHO	World Health Organization

Glossary

Administrative Unit	A hospital that provides cleft surgery and submits data to the CRANE Database, sometimes as part of a wider cleft centre or network.
Alveolus / alveolar	The part of the jaw that supports the teeth and contains the tooth sockets.
Carries (dental)	Dental caries are also known as tooth decay / dental decay or a cavity.
Cleft	A failure of tissues to join during development.
Cleft Development Group (CDG)	NHS National group representing all stakeholders in cleft care that is responsible for the CRANE Database as well as oversight and guidance on all aspects of the delivery of reorganised cleft care.
Cleft surgeon	A surgeon undertaking cleft repair surgery in a region / unit.
Clinical Standards Advisory Group (CSAG)	A group established in 1991 to act as an independent source of expert advice on standards of clinical care for, and access to and availability of services to, NHS patients.
Confidentiality Advisory Group (CAG)	An independent statutory body established to promote, improve and monitor information governance in health and adult social care. http://www.hra.nhs.uk/research-community/applying-for-approvals/confidentiality-advisory-group-cag/
Craniofacial anomalies	A diverse group of deformities in the growth of the head and facial bones.
Craniofacial Society of Great Britain and Ireland (CFSGBI)	An inter-specialty group set up to study cleft lip and palate and other craniofacial anomalies. www.cfsgb.org.uk
Early Years Foundation Stage Profile (EYFSP)	A teacher assessment of children's educational attainment across six areas of learning at 5 years of age.
Funnel Plot	A graph that identifies regions / units which are outliers, where the local situation might require closer inspection – either because an area is doing well or because there is some indication that it is performing poorly. In this report: • Each point on the funnel plot represents a region / unit. • Each funnel plot is for one outcome, with its values shown on the vertical/Y axis. • The size of the regions' /units' cohort is shown on the horizontal or X axis. • The benchmark value is shown as a horizontal line through the centre of the graph. The graph shows two funnels that lie on either side of the benchmark and are called the control limits – similar to confidence intervals. • The inner lines show 2 standard deviations or 95% control limits. The outer lines represent 3 standard deviations or 99.8% control limits. • The funnel shape is formed because the control limits get narrower as the population size increases. The outer funnel is used to decide if an area is significantly different to the benchmark with 99.8% confidence. If a point lies within the funnel then we conclude that it is not significantly different to the benchmark. If it falls outside the funnel then we can say the value is significantly 'better' or

	significantly ‘worse’ than the benchmark, depending on the direction of the indicator/outcome. Funnel Plot Source: David Spiegelhalter, Medical Research Council Biostatistics Unit - http://www.erpho.org.uk/Download/Public/6990/1/INPHO%204%20Quantifying%20performance.pdf
Hospital Episode Statistics (HES)	A national database containing records on all admissions to NHS hospitals in England.
Key Stage 1 (KS1)	An assessment of children’s educational attainment across five subject areas at 7 years of age.
LAHSAL	A code used to classify clefts. Each letter (LAHSAL) relates to one of the six parts of the mouth that can be affected by a cleft.
Managed Clinical Network (MCN)	A formally organised network of clinicians.
National Pupil Database (NPD)	A database containing records on all pupils in England as they progress through primary and secondary education.
Patient Episode Data Wales (PEDW)	A national database containing records on all admissions to hospitals in Wales.
Submucous Cleft Palate	The term submucous refers to the fact that the cleft is covered over by the lining (mucous membrane) of the roof of the mouth. This covering of mucosa makes the cleft difficult to see when looking in the mouth.

Executive summary

Craniofacial abnormalities are among the most common of all birth defects[1]. Cleft lip and/or palate can affect a variety of functions, including speech and hearing. Appearance and psychosocial health may also be compromised in those with a cleft. Typically, children with a cleft need multidisciplinary care from birth to adulthood, and they have higher morbidity and mortality throughout life compared with unaffected individuals[2].

The Cleft Registry & Audit Network (CRANE) Database is a national register that collects information on children born with a cleft lip and/or palate in England, Wales and Northern Ireland. The database was established in 2000 and transferred to the Clinical Effectiveness Unit of the Royal College of Surgeons in 2005. CRANE has two broad aims:

- To register birth and demographic data related to all children born in England, Wales and Northern Ireland with the congenital abnormality of cleft lip and/or palate;
- To record the treatment of children and adults with a cleft lip and/or palate and the outcome of such treatment.

Data are submitted to CRANE by the region / unit providing surgical treatment to cleft patients in England, Wales and Northern Ireland.

This Annual Report describes the results of on-going analyses of the CRANE Database with a specific focus on children born with a cleft lip and/or palate in 2014 in England, Wales and Northern Ireland. We examine trends in: Registrations and the timing of cleft diagnosis, patient referrals to units, and first contact between units and the parents of children born with a cleft.

This Annual Report also provides feedback to all stakeholders involved in cleft care, highlighting areas of success and areas requiring improvement in the future, and outlining the following:

- Information on cleft-related outcomes for children at five years of age (born 2004-2009). The completeness of these data, which is essential for CRANE to perform meaningful analyses, is presented according to region / unit.
- Analyses of data from the National Pupil Database, containing records on all pupils in England as they progress through primary and secondary education. These data were linked to the CRANE database at the individual level for consented children born between 2000 and 2008. We describe the results of the repeated linkage exercise and we present data on special educational needs and the educational achievement of non-syndromic children with a cleft at five years and seven years of age.

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[1] Stanier P, and Moore G. Genetics of cleft lip and palate: syndromic genes contribute to the incidence of non-syndromic clefts. Human Molecular Genetics, 2004. 13: p. R73-R81.

[2] Mossey PA, Little J, Munger RG, Dixon MJ and Shaw WC. Cleft lip and palate. The Lancet, 2009. 374(9703): p. 1773-1785.

Key findings

Children born with a cleft lip and/or palate in 2014

15,351 children were born over the last fifteen years, between 1 January 2000 and 31 December 2014, with a cleft lip and/or palate[3]. These children account for all children born with a cleft lip and/or palate, referred to one of the regions / units in England, Wales and Northern Ireland, regardless of their consent status. Of these children, 1,014 were born in 2014.

CRANE case ascertainment is very high, being around 95%, according to comparisons with HES and Patient Episode Data Wales (PEDW)[4]. Out of the 812 children born in 2014 whose consent status had been verified, the parental consent rate was 96.9% (ranging from 87.5% to 100% between units). Out of all children registered, 19.9% (ranging from 0% to 81% between units) had not had their consent status verified at the time of writing this report.

Among children born in 2014, CRANE analyses revealed:

- 70% of those with a cleft affecting the lip (CL) were diagnosed in the antenatal period through screening. The NHS Fetal Anomaly Screening Programme has a target of 75% for the antenatal diagnosis of clefts affecting the lip.

- Only 1% of children with cleft palate (CP) only were diagnosed during antenatal screening: 67.7% were diagnosed at birth, leaving 31.3% who were diagnosed late according to the national standard[5] – 8.2% of whom were diagnosed after one month of age. The proportion of children with CP diagnosed late has increased slightly by 0.9% compared with last year.

- 81.4% of children were referred by a maternity unit to a Cleft Administrative Unit within 24 hours of birth, maintaining approximately the same level of improvement as noted at this time last year (for 2013 births – versus 2012's rate of 52.6%).

- Referrals from maternity units within one day of birth varied from 70.4% to 88.5% according to the unit receiving the referral. Units went on to establish contact with 90.8% of parents within 24 hours of their child's referral.

Cleft-related clinical outcomes at five years of age

CRANE collected clinical outcomes at five years of age among children born between 2004 and 2009. These outcomes include **height and weight** (2004-2009 births), the number of **decayed, missing and filled teeth** (dmft), a measure of oral health (2004-2008 births), **Five Year Old Index scores**, reflecting dental arch

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[3] As registered on the CRANE Database by 6 August 2015.
[4] CRANE Project team on behalf of the Cleft Development Group, CRANE Database Annual Report 2011, 2011, Clinical Effectiveness Unit, The Royal College of Surgeons of England, London.
[5] Bannister P. Management of infants born with a cleft lip and palate. Part 1. Infant, 2008. 4(1): p. 5-8.

relationships and effects of primary cleft repair surgery on the facial growth of children with a complete unilateral cleft lip and palate (UCLP) (2004-2008 births), and a **speech assessed using the Cleft Audit Protocol for Speech – Augmented (CAPS-A) scoring system** (2007-2008 births). Although there is still a high proportion of missing data, some units have reported outcomes for more than 85% of their eligible patients, suggesting that the reporting of these outcome data is feasible. For those children with reported outcomes:

- 42.3% had at least one decayed, missing or filled tooth (>0 dmft), which is slightly higher than the rate in the general population (38.8%). The proportion of children with at least one dmft varied significantly according to cleft type and region / unit, although treatment indices (ranging from 54.7% to 96.6%) and care indices (ranging from 45.7% to 95.9%) across units indicate that, in the majority of cases, units have mechanisms in place to deal with any disease occurring. Bilateral cleft lip and palate (BCLP) was associated with the poorest oral health, with 51.6% of BCLP patients having at least one dmft at five years of age.
- Of the 522 children with a complete UCLP for whom Five Year Old Index scores had been reported, 25.1% had scores of ‘4’ or ‘5’, reflecting poor dental arch relationships. This represents a substantial improvement compared to the CSAG findings that 36% of (223) cleft children had poor dental arch relationships at five years of age in 1996[6].
- Of the 927 eligible children born in 2007-2008 with a cleft affecting their palate, and for whom speech CAPS-A scores had been reported across all 16 speech parameters measured, 58.3% had speech scores that would suggest their speech was not significantly different from their non-cleft peer group.

Unfortunately, outcomes are still not collected consistently across units:

- Height and weight measures are not collected routinely (in fewer than 20% of cases) by four of the fourteen units.
- Some regions such as the East and Trent submit low levels of dmft data for eligible cases (8.8% and 19.1% respectively). Nevertheless, this represents an improvement year on year (since reporting of 2013 and 2012 births in previous reports), which bodes well for continued improvement in reporting this outcome for future years.
- The Five Year Old Index scores were reported by all units (versus data not reported by three units at this time last year), with overall reporting of Five Year Old Index scores having increased by 11.5% in the last two years (from 62% for 2013 data and 57.5% for 2012 data), which is very encouraging.
- Speech measures were not collected consistently across all units but, given that this is only the second year that the expanded 16 CAPS-A speech outcome scores have been requested, we are encouraged by the fact that units have reported speech data on an average of 63.8% of cases.

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[6] Clinical Standards Advisory Group, Clinical Standards Advisory Group. Report of a CSAG Committee on cleft lip and/or palate, 1998, The Stationery Office, London.

- CRANE will continue to explore methods for improving communication and links with cleft teams to facilitate the submission of data to the database – the re-development of the CRANE Database and Website in 2015/16 is expected to play a key role in facilitating this (see Chapter 6 for further detail on this).

Educational achievement at five years

In 2014, individual-level data from the National Pupil Database (NPD) were linked to CRANE records of consented children born between 2000 and 2008. The initial linkage was successful for 56% of eligible CRANE-registered children. We found that linkage was largely determined by the quality of the postcode captured by CRANE. In 2015, we were able to update postcode records before repeating the linkage exercise. We describe the results of the second linkage, highlighting the linkage rate, and focusing on the special educational needs (SEN) and the educational attainment of children at the Early Years Foundation Stage Profile (EYFSP) assessment at age five years and at the end of Key Stage 1 (KS1) at seven years. Data are presented for non-syndromic children only.

The main findings are outlined below:

1. Data linkage: 6,194 (87%) eligible CRANE-registered children were linked to NPD records during the second linkage exercise between the two datasets. Linkage rates varied from 72% in Liverpool to 94% in Newcastle.

2. Early Years Foundation Stage Profile (EYFSP) at five years of age[7] (for academic years 2006/07 to 2011/12):

   - Special educational needs (SEN): 29.4% of non-syndromic children[8] with a cleft were identified in the NPD as having special educational needs at five years of age. This compares to 9.7% in the general population across the same time frame. The proportion of children with SEN varied according to cleft type, ranging from 13.2% of those with Cleft Lip (CL) to 47.6% of those with Bilateral Cleft Lip & Palate (BCLP). Speech, language and communication needs were the most common type of SEN, which was identified for almost three quarters of those with a SEN type specified.

   - Achieving a ‘good level of development’[9]: 42.2% of all non-syndromic children with a cleft were classified as having a good level of development. This compares to 54.3% of children in the

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[7] The EYFSP teacher assessment at five years of age consisted of 13 assessment scales across six areas of learning. These areas include 1. Personal, social and emotional development, 2. Communication, language and literacy, 3. Mathematical development, 4. Knowledge and understanding of the world, 5. Physical development, and 6. Creative development.

[8] 2,802 non-syndromic children with a cleft were included in the EYFSP analyses.

[9] Children achieving at least six points (out of a maximum of nine points) in each of the seven ‘personal, social and emotional development’ and ‘communication, language and literacy’ scales, and at least 78 points across all 13 scales, are classified as having a ‘good level of development’.

general population across the same time frame. 47.7% of those with a CL, 42% of those with a CP or UCLP and 27.7% of those with a BCLP were classified as having a good level of development.

- Year and sex-adjusted standardised scores (z-scores): Since the EYFSP assessment started in 2006/07, national mean results have improved each year. A significant gender gap also exists, with girls performing better than boys. To account for these differences when comparing outcomes among children with a cleft to those among all children in the general population, year and sex-adjusted standardised scores (z-scores) were calculated for each child for each of the 13 individual assessment scales and six areas of learning across the EYFSP.
    - Non-syndromic children with a cleft have z-scores that are significantly below the national average within each of the six areas of learning assessed.
    - The greatest differences appear to be within the ‘physical development area’, closely followed by the ‘communication, language and literacy’ area.
    - When assessing z-scores according to cleft type, children with a CL fare better than those with a cleft affecting the palate. Their z-scores were not significantly different to the national mean within the areas of ‘mathematical development’, ‘knowledge and understanding of the world’ and ‘creative development’.

3. Key Stage 1 (KS1) at seven years of age[10] (for academic years 2006/07 to 2014):

- Special educational needs: 40.4% of non-syndromic children[11] with a cleft were identified in the NPD as having special educational needs at seven years of age. This compares to 21% in the general population across the same time frame. The proportion of children with SEN varied significantly according to cleft type, ranging from 26.6% of those with CL to 57.3% of those with BCLP ($p<0.001$). Speech, language and communication needs were the most common type of SEN, which were identified for 70.2% of those with a SEN type specified. The second most common type of SEN was learning difficulties, which were documented for 24.1% of those with a type of SEN specified.
- Achieving Level 2 or above – the expected attainment level at the end of KS1:
    - Across all five assessed subject areas, a lower proportion of non-syndromic children with a cleft achieved Level 2 or above compared to the general population. The average attainment gap (the difference between the non-syndromic cleft cohort and the general population in terms of the proportion achieving Level 2 or above) between the two groups across the five subject areas was 7.5%.

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[10] The KS1 assessments at seven years of age cover five main subjects: reading, writing, speaking and listening, maths and science.

[11] 3,466 non-syndromic children with a cleft were included in the KS1 analyses.

  - The greatest difference was in the area of speaking and listening, where 78.5% of children with a cleft achieved Level 2 or above compared to 87.6% of those in the general population (9.1% attainment gap). The smallest difference was in the area of maths, where 83.8% of children with a cleft achieved Level 2 or above children compared to 90.3% in the general population (6.5% attainment gap).

  - Within each subject, a significant difference existed between cleft types in terms of the proportion of children achieving Level 2 or above ($p<0.001$). Children with CL were most likely to achieve Level 2 or above, while children with a BCLP appear to be most affected academically.

Recommendations

Clinical care

- Late diagnosis of cleft palate (CP) remains an important issue that must be addressed. Among children born in 2014, 31.3% of those with CP were diagnosed late according to the national standard[12]. Since the publication of our findings in our 2012 Annual Report, which highlighted the problem of late diagnosis, the Royal College of Paediatrics and Child Health (RCPCH) — in collaboration with key partners including the CRANE Database team — have drafted and published a best practice guide to help healthcare professionals identify cleft palate in neonates. This guide[13] provides recommendations to ensure early detection of a cleft palate, and to improve and standardise routine postnatal examination of the palate.

- Preventative dental support for children with a cleft seems essential to reduce dental decay, particularly among those with UCLP and BCLP who appear to be at the greatest risk of caries. However, further analyses on more complete data are recommended.

Outcome measures and reporting to CRANE

- Units must improve data submission to CRANE. This is related to the National Service Specification which now contractually obliges units to submit data. Although improvements in submission of data and reporting of outcomes have been noted this year, the submission of data *for all eligible patients* is still required so that CRANE can report data to the Quality Dashboard.

- CRANE’s collection of CAPS-A data has been modified so that data can be analysed and reported more clearly. Analysis of this data will continue to be refined to aid reports using the nationally agreed Speech Outcome Standards.

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[12] Bannister P. Management of infants born with a cleft lip and palate. Part 1. Infant, 2008. 4(1): p. 5-8.

[13] http://www.rcpch.ac.uk/improving-child-health/clinical-guidelines-and-standards/published-rcpch/inspection-neonatal-palate

- Further outcome measures need to be developed to reflect a wider age range of patients and a broader range of cleft-related outcomes, including hearing, psychology, and patient and/or parent satisfaction. Bearing this in mind, the Cleft Psychology Clinical Excellence Network (CEN), upon request by the Craniofacial Society of Great Britain and Ireland (CFSGBI) Council and the Cleft Development Group (CDG), identified and piloted measurements[14] to evaluate patient (and parent) reported experience. These measures were combined into one Patient (and Parent) Reported Experience Measure (PREM) questionnaire and, through piloting[15] by the Cleft Psychology CEN, adjusted to apply to cleft services. The CRANE project team and the Cleft Psychology CEN are collaborating to conduct a 12-month feasibility study (with a review point at 3-months). This feasibility study team aims to test PREM data collection, analysis and reporting, with a view to developing a method to implement data collection, analysis and reporting nationally.
- This is the third year of providing data for the Quality Dashboard. Further outcome development work should be considered with commissioner involvement. This should aim to capture data that can be used to inform the commissioning process for cleft-related services.

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[14] (1) The Friends and Family Test (FFT) – developed by the Department of Health, and (2) the Experience of Service Questionnaire (CHI-ESQ) satisfaction assessment scales – developed by the Commission for Health Improvement (CHI).

[15] For full copies of documentation around the Cleft Psychology CEN review and pilot please contact Vanessa Hammond, Chair of Cleft Psychology CEN on vanessa.hammond@wales.nhs.uk.

1. Introduction

Craniofacial abnormalities are among the most common of all birth defects[16]. Cleft lip and/or palate can affect a variety of functions, including speech and hearing. Appearance and psychosocial health may also be compromised in those with a cleft. Typically, children with a cleft need multidisciplinary care from birth to adulthood, and they have higher morbidity and mortality throughout life compared with unaffected individuals[17].

The Cleft Registry & Audit Network (CRANE) Database is a national register that was established in 2000 to collect information on children born with a cleft lip and/or palate in England, Wales and Northern Ireland. The Database collects birth, demographic and cleft diagnosis information. It also collects information about cleft-related treatment and outcomes. Hospital Episode Statistics (HES) is used to further examine treatment for cleft lip and/or palate in England, and data from the National Pupil Database (NPD) is used to further examine outcomes at five years of age.

This Annual Report presents findings from data submitted to CRANE by 6 August 2015 for children with a cleft lip and/or palate born in England, Wales and Northern Ireland between the 1 January 2000 and 31 December 2014. We describe:

- Trends in CRANE registrations over the last 10 years, comparing the ten regions and their units and the four different types of cleft;
- the proportion of babies born in 2014 who were diagnosed at birth, referred within 24 hours of birth, and contacted within 24 hours of referral;
- cleft-related outcomes at five years of age, including height and weight, number of decayed, missing or filled teeth (dmft), Five Year Old Index scores, and speech in the form of Cleft Audit Protocol for Speech—Augmented (CAPS-A) scores.

In addition, we describe analyses of data from the National Pupil Database (NPD), a database containing records on all pupils in England as they progress through primary and secondary education. These data were linked to the CRANE database at the individual level for consented children born between 2000 and 2008. We describe the results of the repeated linkage exercise and we present data on special educational needs and the educational achievement of non-syndromic children with a cleft at five years and seven years of age.

This Annual Report aims to provide feedback to all stakeholders involved in cleft care, highlighting areas of success and areas requiring improvement in future reporting and in clinical practice.

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[16] Stanier P and Moore G. Genetics of cleft lip and palate: syndromic genes contribute to the incidence of non-syndromic clefts. Human Molecular Genetics, 2004. 13: p. R73-R81.

[17] Mossey PA, Little J, Munger RG, Dixon MJ and Shaw WC. Cleft lip and palate. The Lancet, 2009. 374(9703): p. 1773-1785.

1.1. Background to the CRANE Database

The CRANE Database was established in 2000 in response to the report of the Clinical Standards Advisory Group (CSAG) on cleft care in the UK in 1998[18]. The report suggested that the outcome of cleft care in the UK was inferior to other countries in Western Europe. The CRANE Database can be considered a continuation of the Craniofacial Anomalies Register (CARE) that since 1990 was maintained by the Craniofacial Society of Great Britain and Ireland (CFSGBI).

The CSAG report recommended that cleft care should be centralised into regional cleft teams that would treat larger numbers of patients. The rationale for this recommendation was that it would increase the experience of the cleft teams and facilitate genuine multi-disciplinary care. At the same time, it would also enable meaningful and statistically significant audit. The Health Services Circular 1998/238, which set out arrangements for commissioning cleft services according to the CSAG report, stated that 'a craniofacial anomalies register, within which all patients should be registered, should form the basis of national audit'[19]. A high-quality national database could furthermore contribute to comparisons between countries.

Currently, the CRANE Database collects information about children born with a cleft lip and/or palate in England, Wales and Northern Ireland. Scotland maintains a separate database which is part of CLEFTSiS, the National Management Clinical Network for Cleft Service in Scotland.

The Cleft Development Group (CDG) is responsible for making arrangements for the running and commissioning of the CRANE Database (see Appendix 3 for CDG's membership). The funding for CRANE was provided by the Specialist Commissioners based on an original contract. Currently this is extended annually by NHS England and NHS Wales. This year Northern Ireland has formally agreed to contribute to the project for the first time. The CRANE team has responded to a number of requests for information from a commissioner led comprehensive review of all databases relating to specialised services. The outcome of this has been an agreement to continue to fund CRANE on an annual basis[20].

1.2. Geographical representation of the cleft regions / units

The CRANE Database covers England, Wales and Northern Ireland. Cleft care is currently delivered by eight Regional Cleft Centres and two Managed Clinical Networks. Each of these 10 geographical hubs, with the exception of Northern Ireland, treats an average of 109 new children born with a cleft lip and /or palate each year. Several of the Regional Cleft Centres are split between two hospitals, where the primary surgery is usually undertaken, and therefore Hospitals/ Administrative Units in a region may submit data separately to the CRANE Database (see Table 1).

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[18] Clinical Standards Advisory Group, Clinical Standards Advisory Group. Report of a CSAG Committee on cleft lip and/or palate, 1998, The Stationery Office, London.

[19] Department of Health, HSC 1998/238: Cleft lip and palate services, 1998, Department of Health: Leeds.

[20] Current funding is confirmed up until the end of the 2015/16 financial year.

Table 1. Regional Cleft Centres and Managed Clinical Network and their associated regions / units.

Regional Cleft Centre / MCN	Administrative Unit
Northern & Yorkshire	Royal Victoria Hospital, Newcastle Leeds General Infirmary, Leeds
North West & North Wales & Isle of Man	Alder Hey Children's Hospital, Liverpool Royal Manchester Children's Hospital, Manchester
Trent	Nottingham City Hospital, Nottingham
West Midlands	Birmingham Children's Hospital, Birmingham
East	Addenbrooke's Hospital, Cambridge
North Thames	Great Ormond Street Hospital, London Broomfield Hospital, Chelmsford
The Spires	John Radcliffe Hospital, Oxford & Salisbury District Hospital, Salisbury
South Wales & South West	Morriston Hospital, Swansea University Hospitals Bristol*
South Thames	Guy's and St Thomas' Trust (GSTT), London
Northern Ireland	Royal Belfast Hospital for Sick Children, Belfast

Note: MCN – Managed Clinical Network. Data for Oxford and Salisbury units combined upon request by the Spires' Clinical Director (June 2015). *Frenchay Hospital, Bristol service moved to University Hospitals Bristol during 2014.

1.3. Aims and objectives of the CRANE Database

The aims of the CRANE Database are:

1. To register birth, demographic and epidemiological data related to all children born in England, Wales and Northern Ireland with the congenital abnormality of cleft lip and/or palate; and
2. to record the treatment of children and adults with a cleft lip and/or palate and the outcome of such treatment.

These data will provide the basis for national audit of cleft care. In line with these broad aims, the CRANE Database has the following specific objectives:

1. To ensure there is an up-to-date register of all children with cleft lip and/or palate;
2. to monitor the frequency and incidence of clefting in the population;
3. to audit and report on the quality of care for patients with clefts, thus promoting high standards in clinical management;
4. to seek and use linkage to other national databases for validation purposes, to augment the data recorded in CRANE, to reduce data collection burden for regional cleft centres, and be able to more thoroughly report on the impact of cleft care on patients' outcomes;
5. to work with and receive advice from the CFSGBI to improve the delivery of cleft care in the UK;
6. to work in partnership with Specialised Commissioning Groups (SCGs) to inform commissioning of cleft services; and
7. to support research and focused studies.

2. Methods

This report contains information on patterns of care and outcomes derived from three sources of data. These sources are (1) the CRANE Database, (2) the National Pupil Database (NPD) linked to both the CRANE Database and Hospital Episode Statistics (HES) data, and (3) HES linked to the Office for National Statistics (ONS) mortality dataset.

2.1. CRANE

2.1.1. Data source

CRANE is an online custom-built secure database that holds information on children born with a cleft lip and/or palate in England, Wales and Northern Ireland. CRANE collects data pertaining to a patient's birth, demographics, type of cleft, time of diagnosis, time of referral to a cleft team, and time of first contact between a patient and cleft team. CRANE also collects information about cleft-related treatment and outcomes. These data are reported to CRANE by the units that make up 10 Regional Cleft Centres / Managed Clinical Networks (as listed in Table 1). Each child born with a cleft in England, Wales and Northern Ireland should be referred to one of these units shortly after having their cleft diagnosed.

Since January 2012, CRANE has been able to act as a national register of cleft-affected births by collecting some basic information on all children born with a cleft and being treated by the specialist cleft units. Additional information, including cleft-related outcomes, is collected for children whose parents have consented to their child's data being submitted to the national database. Parental consent is usually obtained by units at some point between referral and the first primary repair. A coordinator within each unit submits data to CRANE on the children referred to them. Once a record has been created on CRANE for a particular child, it can later be updated with further information.

2.1.2. Patients

All data entered into the CRANE Database by 6 August 2015 pertaining to children born between 1 January 2014 and 31 December 2014 is included in the analyses described in this Annual report. Patients whose parents have not consented to their data being used by CRANE have been excluded from Tables 9 to 15 (as the data presented in these tables are not collected for non-consenting cases).

2.1.3. Data validation and cleaning

Logical and systematic data cleaning was undertaken to identify any potential data errors. Continuous data variables (birth weight, five-year weight and five-year height) were assessed in relation to valid ranges.

Valid ranges for five-year body weight and five-year height have been defined according to growth charts published by the World Health Organisation (WHO)[21].

2.1.4. Analyses

Data have been analysed according to year of birth, unless otherwise stated. Five-year outcome data were restricted to children born between 2004 and 2009, depending on the outcome of interest. Children dying before five years of age were excluded from these analyses.

Cleft type

Cleft type was defined according to reported LAHSAL codes. The LAHSAL code is used to classify clefts, with each letter relating to one of the six parts of the mouth that can be affected by a cleft:

L	**A**	**H**	**S**	**A**	**L**
Right Lip	Right Alveolus	Hard palate	Soft palate	Left Alveolus	Left Lip

The code also indicates whether there is a complete cleft (upper case letter, e.g. H), an incomplete cleft (lower case letter, e.g. h), or no cleft (left blank). Where LAHSAL has not been reported (5.8% of children born in 2014), cleft type is based on the type reported by the region/ unit registering the child. Children with a unilateral cleft lip and palate (UCLP) were categorised according to whether the UCLP was complete or incomplete. A complete UCLP was defined as LAHS or HSAL codes, indicating a complete cleft affecting all three components of the mouth on either the right or left side.

Decayed, missing and filled teeth (dmft)

The dmft score describes the amount of dental caries in an individual and is a measure of oral health. A dmft score reflects the total number of teeth that are decayed, missing or filled. Analyses on dmft data were restricted to consented children born between 2004 and 2008 (excluding children with a submucous cleft palate).

Five Year Old Index

Dental models of five-year old children with UCLP can be assessed using the Five Year Old Index to examine dental arch relationships. The index evaluates the effects of primary surgery on the facial growth of children with UCLP before any other interventions are performed, such as orthodontics or alveolar bone grafting, which may influence this growth further[22]. CRANE collected both internal and external Five Year Old Index scores for consented children born between 2004 and 2008 with a complete UCLP (LAHSAL codes

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[21] World Health Organization. The WHO Child Growth Standards 2011. Available from: http://www.who.int/childgrowth/standards/en/.

[22] Johnson N, Williams AC, Singer S, Southall P, Atack N and Sandy JR. Dentoalveolar relations in children born with a unilateral cleft lip and palate (UCLP) in Western Australia. The Cleft Palate-Craniofacial Journal, 2000. 37 (1): p. 12-16.

LAHS or HSAL). Some cleft teams score the models of children treated in their unit (internal scores) before they are sent off to be scored externally (external scores) by a blinded process undertaken by calibrated examiners. For the purpose of this report we have analysed externally validated scores where available, where these were unavailable internal scores are included in the analysis.

Cleft Audit Protocol for Speech – Augmented (CAPS-A)

CAPS-A scores collected at five years of age among children born in 2007 and 2008 were reported to CRANE for consented children only. The parameters of speech assessed include resonance (hypernasality and hyponasality), nasal airflow (audible nasal emission and nasal turbulence) and twelve Cleft Speech Characteristics (CSCs) scores[23] – including:

- Anterior oral CSCs – for dentalisation/interdentalisation, lateralisation/lateral, and palatalisation / Palatal characteristics;
- posterior oral CSCs – for double articulation and backed to velar/uvular characteristics;
- non-oral CSCs – for pharyngeal articulation, glottal articulation, active nasal fricatives, and double articulation characteristics; and
- passive CSCs – for weak and or nasalised consonants, nasal realisation of plosives, and gliding of fricatives.

Missing data

Missing data have been excluded from the denominators presented in Tables 5 to 20. All units have some degree of missing data. The number of patients with missing data for five-year outcomes is high. A variety of reasons were reported by units. Reasons out of a unit's control include children not attending an appointment or moving away from the area.

2.2. National Pupil Database (NPD)

2.2.1. Data source and linkage

The National Pupil Database (NPD) is a national database containing records on educational outcomes for all pupils in England from the 1995/1996 school year onwards. The initial year for which Key Stage attainment data were first collected varies according to the examination of interest. CRANE sought and was granted permission by the Department for Education (DfE), in accordance with their published application process [24], to link the information held in the CRANE database with the NPD.

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[23] 2006 data is not included as only four Cleft Speech Categories (CSCs) summarising current 12 CSCs were collected for 2006 birth.

[24] https://www.gov.uk/national-pupil-database-apply-for-a-data-extract

For eligible CRANE-registered children, personal identifiers (name, postcode and date of birth) were securely passed to the DfE, who performed the linkage between records. We requested the following datasets for each matched child:

- PLASC/School Census
- Early Years Foundation Stage Profile (age 5)
- Key Stage 1 (age 7)
- Key Stage 2 (age 11)
- Absence

NPD information on pupils who were matched to CRANE records was merged by the CRANE Data Manager with the existing CRANE-HES linked dataset. The CRANE-HES linked dataset provides information about the children's cleft type and the presence of additional anomalies, as well as treatment outcomes recorded in CRANE.

The first linkage exercise took place in 2014. In early 2015, postcode records in CRANE were updated before repeating the linkage for the second time.

National summary data are published for each National Curriculum assessment. We have used these data to draw comparisons with the CRANE cohort.

2.2.2. Patients

School-aged, consented, CRANE-registered children, born between 1 January 2000 to 31 December 2008, who had English residential postcodes and who were alive at five years of age were eligible for NPD linkage. Children were excluded from analyses if they had additional anomalies or syndromes identified in HES (see Appendix 5) or if it was not possible to determine their syndrome status. Children were also excluded if their gender or cleft type was not documented.

2.2.3. Outcomes and analyses

Special Educational Needs (SEN)

Pupils with SEN have learning difficulties or disabilities that make it harder for them to learn than most pupils of the same age. In the NPD, there are three categories of SEN:

1. School Action, where extra or different help is given from that provided as part of the school's usual curriculum;
2. school Action Plus, where the class teacher and the SEN coordinator receive advice or support from outside specialists (e.g. a specialist teacher or a health professional);

3. statements of SEN, where a pupil has had a formal assessment and a document setting out the child's needs and the extra help they should receive is in place [25].

In the NPD, there are 12 different types of SEN documented. These are recorded under 'primary SEN type' and, for those whose needs fall into more than one category, 'secondary SEN type'. For the purpose of this report, we have grouped these types into the following categories:

- Speech, language and communication needs
- Learning difficulties
- Behaviour, emotional and social difficulties
- Hearing impairment
- Other (including: Autistic spectrum disorder, multi-sensory impairment, other difficulty/disability, physical disability and visual impairment)

The proportion of children with SEN and the primary and secondary types of SEN documented in the NPD are reported for non-syndromic CRANE-registered children at five and at seven years of age

Early Years Foundation Stage Profile (EYFSP)

Between the six academic years 2006/2007 to 2011/2012, the EYFSP consisted of 13 assessment scales (see Section 4 from page 46). Children achieving at least six points in each of the seven Personal, social and emotional development (PSE) and Communication, language and literacy (CLL) scales and at least 78 points in total across all 13 scales are classified as having a good level of development. We calculated the proportion of children with a cleft achieving a good level of development at the EYFSP across the six academic years included.

Since the EYFSP assessment started in 2006/2007, national mean results have improved each year. A significant gender gap also exists, with girls achieving a higher level than boys. To account for these differences when comparing EYFSP outcomes among children with a cleft to national data on all children in the general population, year and sex-adjusted standardised scores (z-scores) were calculated for each child for each of the 13 individual assessment scales. Mean z-scores were also calculated for each of the six areas of learning. Z-scores were calculated using year and sex-specific national population means and standard deviations. The z-score presents the child's score as the number of standard deviations away from the general population mean.

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[25] Department for Education 2011 Statistical First Release – National Curriculum Assessments at Key Stage 1 in England, 2011. https://www.gov.uk/government/uploads/system/uploads/attachment_data/file/219034/main_20text_20sfr222011.pdf

Key Stage 1 (KS1)

KS1 attainment data for CRANE-registered children are available from 2007 to 2014. Five subject areas are assessed. These include reading, writing, speaking and listening, maths and science. We present the proportion of children achieving Level 2, which is the expected attainment level at the end of KS1.

We present the analyses as one cleft cohort and also by cleft type. Published summary data from the general population are presented for comparison. Differences between the cleft cohort and general population are sometimes referred to as *attainment gaps*.

2.3. Hospital Episode Statistics (HES)

HES is a national database containing records on all admissions to NHS hospitals in England. It includes data on private patients treated in NHS hospitals, patients who were resident outside of England and care delivered by treatment centres (including those in the independent sector) funded by the NHS. Data on admissions are available for every financial year from 1989/90 onwards. Since the 1997/98 financial year, a unique patient identifier has been available that enables records belonging to the same patient to be identified across years.

The HES database holds diagnostic and procedure information on each patient, allowing us to identify those with a cleft lip and/or palate and those undergoing cleft-related treatment. In addition to being able to identify and confirm cleft type in the CRANE Dataset, HES is used by CRANE to identify any additional anomalies for the CRANE cohort.

For this report, data were extracted from the HES database linked to the Office for national Statistics (ONS) mortality dataset. Diagnostic information is coded using the International Classification of Disease 10th revision (ICD-10), and procedure information is classified according to codes from the Classification of Surgical Operations and Procedures 4th Revision (OPCS-4). This HES-ONS dataset was then linked to CRANE data and NPD data for validation purposes (as described previously in section 2.2).

3. CRANE

In this chapter, we present data on children with a cleft lip and/or palate, born between 1 January 2004 and 31 December 2014 in England, Wales and Northern Ireland. Data entered into the CRANE Database by 6 August 2015 have been analysed to assess registration patterns, the timing of diagnosis, referral and contact with units around the time of birth, and cleft-related outcomes at five years of age.

3.1. Consent status

The consent status for the 1,014 children born in 2014 who have been referred to a Cleft Unit for treatment and registered on CRANE is presented in Table 2 and described below:

- The parents of 812 (80.1%) had been approached for consent. This figure varied across the regions and units submitting data to CRANE, ranging from 19% in the East region to 100% at the Leeds and Liverpool units.
- Of the 812 children whose parents had been through the consenting process, 96.9% provided consent for their child's data to be submitted to CRANE, which is extremely positive. This proportion ranged from 87.5% for the East to 100% for three units/regions (Liverpool, Chelmsford and Northern Ireland).
- Units registered a total of 202 (19.9%) children born in 2014 whose parents had not yet been approached for consent.
- Of these 202, it was not possible to obtain consent (verification) for eight (4%) children (0.8% of all children born in 2014).
- The proportion of children whose parents still need to be approached for consent is wide ranging across units (from 0% to 81%).
- Overall, the proportions described above are very similar to those described for the 2013 cohort at the time of last year's Annual Report[26].

Generally, the consent data are encouraging, with consent rates very high for those children whose parents have been approached. However, as highlighted in previous Annual Reports, there is still a relatively high proportion of children whose parents have not yet been through the consent process.

The variation in the number of parents approached to seek consent between units suggests different processes are being used between them. Units that have a large proportion of patients yet to be consented are encouraged to review their process for approaching parents for consent, as consent is essential for the collection of a full dataset and the linkage to other datasets.

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[26] CRANE Project team on behalf of the Cleft Development Group. CRANE Database Annual Report 2014. London: Clinical Effectiveness Unit, The Royal College of Surgeons of England, 2014.

Table 2. Number (%) of children born in 2014 with a cleft lip and/or palate in England, Wales and Northern Ireland registered on the CRANE Database[a], according to region / unit and consent status.

		Consent status n (%)				
		Consent status verified		**Consent status not verified**		
Regional centre / MCN	**Administrative Unit**	Consented	Refused	Awaiting verification	Not possible to verify	**All**
Northern	Newcastle	53 (91.4)	2 (3.4)	0 (0)	3 (5.2)	58
& Yorkshire	Leeds	66 (95.7)	3 (4.3)	0 (0)	0 (0)	69
North West	Liverpool	79 (100)	0 (0)	0 (0)	0 (0)	79
& North Wales	Manchester	64 (88.9)	5 (6.9)	3 (4.2)	0 (0)	72
Trent	Nottingham	80 (90.9)	2 (2.3)	6 (6.8)	0 (0)	88
West Midlands	Birmingham	93 (76.2)	2 (1.6)	24 (19.7)	3 (2.5)	122
East	Cambridge	7 (16.7)	1 (2.4)	34 (81)	0 (0)	42
North Thames	Great Ormond St	84 (86.6)	2 (2.1)	11 (11.3)	0 (0)	97
	Chelmsford	48 (94.1)	0 (0)	3 (5.9)	0 (0)	51
The Spires	Oxford & Salisbury	60 (77.9)	5 (6.5)	12 (15.6)	0 (0)	77
South Wales	Swansea	33 (89.2)	1 (2.7)	3 (8.1)	0 (0)	37
& South West	Bristol	47 (79.7)	1 (1.7)	11 (18.6)	0 (0)	59
South Thames	GSTT	39 (30.7)	1 (0.8)	85 (66.9)	2 (1.6)	127
Northern Ireland	Belfast	34 (94.4)	0 (0)	2 (5.6)	0 (0)	36
All	**All**	787 (77.6)	25 (2.5)	194 (19.1)	8 (0.8)	1,014

[a] Registered in CRANE by 6 August 2015. Note: MCN – Managed Clinical Network.

3.2. Number of registrations

A total of 15,351 children born over the last fifteen years, between 1 January 2000 and 31 December 2014, have been registered on the CRANE Database. 1,014 of these children were born and registered in 2014.

Table 3 shows the number of children born with a cleft lip and/or palate for each region / unit over the last 10 years. The North West & North Wales has registered the most births in this time frame (1,543 births).

The number of children born according to cleft type is shown in Table 4. Cleft type was defined according to reported LAHSAL codes or, where LAHSAL has not been reported (for 5.8% of children registered in 2014), it was based on the cleft type reported by the region / unit registering the child. This data is described below:

- The distribution of cleft type is consistent over time and with registrations in previous years. Cleft palate is the most common type of cleft, affecting on average 45% of the cleft population. Bilateral cleft lip and palate is the least common type, affecting on average 9% of people with clefts.
- A total of 141 children registered in 2014 had complete UCLP (defined by either ‘LAHS..’ or ‘..HSAL’ LAHSAL codes), representing 71.6% of the 197 children with UCLP.
- The proportion of children without a specified cleft type ranged between 1.1% in the Trent region to 18.6% at the Bristol unit.

Table 3. Number of children born between 2005 and 2014 with a cleft lip and/or palate in England, Wales and Northern Ireland registered on the CRANE Database[a], according to region / unit grouped within Regional Cleft Centre / Managed Clinical Network (MCN) and year of birth.

Regional Cleft Centre / MCN	Administrative Unit	Year of birth (n)										
		2005	2006	2007	2008	2009	2010	2011	2012	2013	2014	All
Northern & Yorkshire	Newcastle	77	56	86	66	63	64	64	65	69	58	668
	Leeds	71	75	67	73	66	71	71	66	69	69	698
North West & North Wales	Liverpool	86	54	63	87	79	86	62	64	66	79	726
	Manchester	84	105	89	86	68	89	82	70	72	72	817
Trent	Nottingham	106	95	84	105	94	94	91	92	98	88	947
West Midlands	Birmingham	102	121	98	122	116	103	110	127	119	122	1,140
East	Cambridge	87	80	83	83	83	81	66	86	91	42	782
North Thames	Great Ormond Street	81	93	130	153	108	86	111	136	108	97	1,103
	Chelmsford	37	29	45	39	48	39	54	42	45	51	429
The Spires	Oxford & Salisbury	93	116	125	99	93	98	114	98	100	77	1,013
South Wales & South West	Swansea	45	45	48	44	47	44	53	52	39	37	454
	Bristol	52	59	61	72	59	76	54	67	88	59	647
South Thames	GSTT	95	101	110	107	85	62	78	155	118	127	1,038
Northern Ireland	Belfast	38	42	42	37	35	37	37	32	46	36	382
All	**All**	1,054	1,071	1,131	1,173	1,044	1,030	1,047	1,152	1,128	1,014	10,844

[a] Registered in CRANE by 6 August 2015. Note: MCN – Managed Clinical Network.

Table 4. Number (%) of children born between 2005 and 2014 with a cleft lip and/or palate in England, Wales and Northern Ireland registered on the CRANE Database [a], according to cleft type and year of birth.

	Year of birth n (%)																					
Cleft type	2005		2006		2007		2008		2009		2010		2011		2012		2013		2014		All	
Cleft lip	195	(19.2)	236	(23)	265	(23.7)	273	(24.1)	218	(21.7)	246	(24.5)	248	(24.8)	249	(22.9)	294	(26.8)	244	(25.5)	2468	(23.7)
Cleft palate	498	(49.1)	472	(46.1)	491	(44)	529	(46.8)	469	(46.7)	443	(44)	449	(44.9)	483	(44.5)	478	(43.6)	413	(43.2)	4725	(45.3)
UCLP	242	(23.8)	219	(21.4)	247	(22.1)	244	(21.6)	210	(20.9)	209	(20.8)	217	(21.7)	254	(23.4)	221	(20.2)	197	(20.6)	2260	(21.7)
BCLP	80	(7.9)	97	(9.5)	114	(10.2)	85	(7.5)	108	(10.7)	108	(10.7)	86	(8.6)	99	(9.1)	103	(9.4)	101	(10.6)	981	(9.4)
Not specified	39	-	47	-	14	-	42	-	39	-	24	-	47	-	67	-	32	-	59	-	410	-
All	105	(100)	1,071	(100)	1,131	(100)	1,173	(100)	1,044	(100)	1,030	(100)	1,047	(100)	1,152	(100)	1,128	(100)	1,014	(100)	10,844	(100)

[a] Registered in CRANE by 6 August 2015. Note: CL - Cleft Lip, CP - Cleft Palate, UCLP - Unilateral cleft lip and palate, and BCLP - Bilateral cleft lip and palate.

Overall, rates of unspecified cleft types (detailed in Table 4) have remained consistent for 2014 registrations, with 4.8% of the 1,014 total registered children born in 2014 not having their type of cleft specified (either by LAHSAL codes or by the units). This represents a higher proportion of children than the 2.9% rate for 2013 at this time last year.

3.3. Characteristics of children born with a cleft lip and/or palate, 2014

Of the children born with a cleft in 2014, whose sex was reported to CRANE, 41% were girls and 59% were boys. Twenty-eight children did not have their sex reported to CRANE (3% of the total children registered for 2014). There are significant gender differences in the distribution of cleft type ($p<0.001$), with CP more prevalent among females (56% vs. 44% in males), while CL, UCLP and BCLP is more prevalent among males (65% vs. 35%, 71% vs. 29%, and 71% vs. 29% respectively).

Gestational age was reported for 531 (67.5%) of the consented babies born in 2014. This reporting has increased – by 5.9% since last year – however, further improvements are required.

- The mean gestation for those born in 2014 was 38.8 weeks (95% CI 38.6 to 38.9 weeks) and ranged from 26 to 42 weeks.
- Sixty-two (11.7%) babies were premature (born before 37 weeks' gestation), which is higher than the seven per cent national average in England and Wales[27], although it should be noted that the gestation recorded in CRANE may not be representative of all babies born with a cleft lip and/or palate as 32.5% of registered (and consented) children were missing this information.
- A valid birth weight was reported for 540 (68.6%) consented babies born in 2014. The mean birth weight was 3.22kg (95% CI 3.16 to 3.27kg), which is consistent with the national average in England.

Among all the children born in 2014, there were seven (0.7%) deaths reported to CRANE. The majority of deaths (57%) occurred between one month and one year of age. It is not known from CRANE whether these children had additional anomalies or syndromes.

3.4. Timing of diagnosis

3.4.1. Diagnosis times among children born in 2014

Of the 1,014 children born in 2014 with a cleft diagnosis, 57 (5.6%) did not have the timing of their diagnosis reported to CRANE. This is lower (1.2%) than last year's figures. Areas with high levels of missing diagnosis time data include the GOSH unit (15.5%) and the North West region (14.6%). All other regions had missing data rates below 10%.

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[27] Office for National Statistics. Gestation-specific infant mortality, 2013. Part of Gestation-specific infant mortality in England and Wales, 2013. Published 14 October 2015. Available from: http://www.ons.gov.uk/ons/rel/child-health/gestation-specific-infant-mortality-in-england-and-wales/2013/stb-gestation-specific-infant-mortality.html.

Of the 957 children born in 2014 with a reported diagnosis time, 438 (45.8%) had their cleft diagnosed during the antenatal period. This is slightly higher than last year's figure (41.6%). The proportion of children diagnosed antenatally varied between cleft types (as shown in Table 5), 70% per cent of children with CL and over 88% of children with UCLP and BCLP were diagnosed in the antenatal period. Conversely, only 1% of children with a CP were diagnosed antenatally, a statistically significant difference ($p<0.001$), which demonstrates the difficulty of identifying this type of cleft with current antenatal screening techniques.

Table 5. Number (%) of CRANE-registered children born in 2014 with a cleft lip and/or palate according to the timing of diagnosis and cleft type.

	Time of diagnosis in relation to birth[a] n (%)						
Cleft type	Antenatal	At birth	≤1 week[b]	≤1 month	≤6 months	>6 months	**All**
CL	161 (69.7)	57 (24.7)	9 (3.7)	0 (0)	4 (1.7)	0 (0)	231
CP	4 (1)	273 (67.7)	72 (17.4)	21 (5.2)	27 (6.7)	6 (1.5)	403
UCLP	173 (91.5)	14 (7.4)	1 (0.5)	1 (0.5)	0 (0)	0 (0)	189
BCLP	86 (88.7)	11 (11.3)	0 (0)	0 (0)	0 (0)	0 (0)	97
Not specified	14 (37.8)	13 (35.1)	1 (1.7)	3 (8.1)	5 (13.5)	1 (2.7)	37
All	438 (45.8)	368 (38.5)	83 (8.2)	25 (2.6)	36 (3.8)	7 (0.7)	957

[a] 57/1,014 (5.6%) missing diagnosis time and excluded from 'All' values. Note: CL - Cleft Lip, CP - Cleft Palate, UCLP - Unilateral cleft lip and palate, and BCLP - Bilateral cleft lip and palate.

[b] Recording of 'timing of diagnosis' within 72 hours commenced in May 2014 to align CRANE data collection with NIPE standards[28], with only small numbers having been recorded using this timing (n=35, 3.7%). Therefore we report '≤72 hours' cases within the '≤1 week' timing for this report and will report on '≤72 hours' referrals once this data item been functioning for a full 12 month period.

The distribution of timing of diagnosis shown in Table 5, for the children born in 2014, is very similar to that of 2013 births at this time last year.

Of the 519 children not diagnosed during the antenatal period, 368 (70.9%) were diagnosed at birth. The majority (79.2%) of the 120 children diagnosed postnatally with a CL, UCLP and BCLP had their cleft identified at the time of birth. The corresponding figure for the 399 children with a CP is 68.4%. A delayed diagnosis occurred for almost one third of those with a CP, and in 8.3% of cases the diagnosis occurred after one month of age. It should be noted that some children born in 2014 with a CP may not yet have had their cleft identified.

Up until 2012, around ten children with CP were diagnosed after six months of age. In the case of 2014 (and 2013 at the time of the last report) only six children with CP were diagnosed after six months of age – showing a small improvement when compared to previous years.

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[28] UK National Screening Committee Newborn and Infant Physical Examination (NIPE) Standards and Competencies 1 document (2008) – setting out the standard for 95% newborn to be screened by 72 hours after birth (page 13 of the document found at http://newbornphysical.screening.nhs.uk/getdata.php?id=10639).

3.4.2. Diagnosis times among children with a cleft palate alone

The 2012 Annual Report highlighted the issue of late diagnosis among children with CP, reporting that 1.1% were diagnosed during antenatal screening and 66.8% were diagnosed at birth, leaving 32.1% who were diagnosed late according to the National Standard[29]. This year (as for previous years), we have examined diagnosis time among CP patients born over the last five years, between 1 January 2010 and 31 December 2014. No statistically significant differences were found between birth years (p=0.11), indicating diagnosis times have not improved in recent years.

Table 6. Number (%) of CRANE-registered children born between 2010 and 2014 with a cleft palate, according to the timing of diagnosis and region / unit.

		Time of diagnosis in relation to birth[a] n (%)						
Regional Cleft Centre / MCN	**Administrative Unit**	Antenatal	At birth	≤1 week[b]	≤1 month	≤6 months	>6 months	**All**
Northern &	Newcastle	3 (2)	100 (66.2)	20 (6.8)	12 (7.9)	8 (5.3)	8 (5.3)	151
Yorkshire	Leeds	0 (0)	98 (67.6)	29 (9.8)	7 (4.8)	7 (4.8)	4 (2.8)	145
North West &	Liverpool	4 (2.8)	111 (76.6)	15 (5.1)	6 (4.1)	7 (4.8)	2 (1.4)	145
North Wales	Manchester	0 (0)	104 (70.3)	27 (9.2)	10 (6.8)	7 (4.7)	0 (0)	148
Trent	Nottingham	2 (1.1)	122 (69.3)	39 (22.2)	7 (4)	6 (3.4)	0 (0)	176
West Midls.	Birmingham	1 (0.4)	181 (76.4)	29 (12.2)	7 (3)	13 (5.5)	6 (2.5)	237
East	Cambridge	1 (0.8)	88 (66.2)	23 (17.3)	12 (9)	9 (6.8)	0 (0)	133
North Thames	Great Ormond St	3 (1.6)	98 (52.1)	69 (23.8)	6 (3.2)	8 (4.3)	4 (2.1)	188
	Chelmsford	2 (2)	67 (65.7)	22 (7.6)	3 (2.9)	4 (3.9)	4 (3.9)	102
The Spires	Oxford/Salisbury	3 (1.6)	141 (73.1)	17 (8.8)	7 (3.6)	12 (6.2)	13 (6.7)	193
South Wales &	Swansea	1 (1.1)	75 (78.9)	10 (4.4)	8 (8.4)	1 (1.1)	0 (0)	95
South West	Bristol	5 (3.8)	82 (62.1)	18 (7.9)	13 (9.8)	11 (8.3)	3 (2.3)	132
South Thames	GSTT	2 (0.9)	107 (50.5)	63 (29.7)	18 (8.5)	17 (8)	5 (2.4)	212
N. Ireland	Belfast	0 (0)	60 (83.3)	6 (8.3)	0 (0)	2 (2.8)	4 (5.6)	72
All	All	27 (1.3)	1,434 (67.4)	387 (18.2)	116 (5.4)	112 (5.3)	53 (2.5)	2,12

[a] 137/2,266 (6.1%) missing diagnosis time and excluded from 'All' values. Note: MCN - Managed Clinical Network.
[b] Recording of 'timing of diagnosis' within 72 hours only constitutes small numbers (n=31, 1.5%) – detail as per Table 5 footnote.

Table 6 shows the CP diagnosis times according to the region / unit. The proportion of CPs diagnosed at birth ranged from 50.5% among children registered by the South Thames region to over 75% among those registered by the Liverpool and Swansea units, and the West Midlands and Northern Ireland regions. This wide and significant variation (p<0.001) suggests that practice varies considerably between maternity units, with some better than others at identifying clefting of the palate during the newborn examination. Overall, 13.2% of children with a CP were not diagnosed until they were more than one week old, which is concerning given that the National Standard states that clefts should be diagnosed within 24 hours of birth to enable immediate referral to a specialist hospital. This helps to ensure the baby, and their family, receive appropriate care and support as soon as possible. Cleft services are advised to encourage their referring maternity units to identify all clefts as promptly as possible.

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[29] Bannister P. Management of infants born with a cleft lip and palate. Part 1. Infant, 2008. 4(1): p. 5-8.

3.5. Referral to and first contact with a cleft team

3.5.1. Referral among children born in 2014

Out of the 1,014 children born in 2014, 8.3% were missing referral time, reflecting an improvement on registrations for 2013 births (19.7%). The proportion of children missing referral time ranged from 0% at the Leeds unit to 19.6% at GOSH.

Of the 930 children with a reported referral time, 81.4% were referred to a Cleft Unit within 24 hours of birth[30]. 95.5% of the 418 children whose clefts were diagnosed antenatally were referred to a Cleft Unit within 24 hours of birth, while 69.9% of the 512 children whose clefts were diagnosed after birth were referred to a Cleft Unit within 24 hours of birth.

Table 7 shows that the proportion of referrals within 24 hours of birth varied significantly according to cleft type ($p<0.001$), with CP patients having the lowest proportion (67%) out of those with a known cleft type, which corresponds with later diagnosis times for these children.

Referrals within 24 hours of birth varied according to the region / unit receiving the referral (Table 8) although not significantly ($p=0.078$). Rates ranged from 70.4% of children registered by Bristol to 88.5% registered by Manchester referred from maternity units within 24 hours of birth. The increased rates of referral within 24 hours of birth noted at the time of last year's report has remained constant for 2014 births, with rates of referral within 24 hours remaining very similar in 2014 to the 2013 rates.

Table 7. Number (%) of CRANE-registered consented children born in 2014 with a cleft lip and/or palate who were referred within 24 hours of birth and contacted within 24 hours of referral, according to cleft type.

	Referral to Unit		**Contact between Unit and parents of patient**	
	Within 24h of birth	All[a]	Within 24h of referral to Unit	All[b]
Cleft type	n (%)	N	n (%)	N
CL	207 (90.4)	229	208 (97.2)	214
CP	264 (67)	394	355 (94.4)	376
UCLP	181 (95.8)	189	172 (96.6)	178
BCLP	94 (95.9)	98	92 (96.8)	95
Not specified	11 (55)	20	17 (89.5)	19
All	757 (81.4)	930	844 (95.7)	882

[a]84/1,014 (8.3%) missing referral time and [b]132/1,014 (13%) missing contact time and excluded in 'All' values. Note: CL - Cleft Lip, CP - Cleft Palate, UCLP - Unilateral cleft lip and palate, and BCLP - Bilateral cleft lip and palate.

3.5.2. First contact between the unit and parents of children born in 2014

Out of the 1,014 children born in 2014, 132 (13%) were missing the first contact time between units and parents – an improvement on registrations for 2013 births (25.7%) – with the proportion of children missing first contact time data ranging from 0% at the Leeds unit to 53.6% at GOSH.

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[30] A minor improvement on 2013 registrations (18.7%).

Of the 882 children with a reported contact time, units established contact with 95.7% of parents within 24 hours of referral (Table 7), a small improvement on 2013 registrations (92.5%). The proportion of patients contacted within 24 hours of being referred to a unit did not vary significantly between cleft types (p=0.27).

Contact between units and parents of patients within 24 hours of referral did vary significantly according to the unit receiving the referral (p<0.001) (Table 8), with rates varying between units (50% to 100.0%). The majority (10 units) contacted more than 90% of their patients within 24 hours of being referred.

Table 8. Number (%) of CRANE-registered consented children born in 2014 with a cleft lip and/or palate who were referred within 24 hours of birth and contacted within 24 hours of referral, according to region / unit.

		Referral to Unit		**Contact between Unit and parents of patient**	
Regional centre / MCN	**Administrative Unit**	Within 24h of birth n (%)	All[a] N	Within 24h of referral to Unit n (%)	All[b] N
Northern & Yorkshire	Newcastle	40 (74.1)	54	54 (100)	54
	Leeds	54 (78.3)	69	69 (100)	69
North West & North Wales	Liverpool	51 (79.7)	64	60 (93.8)	61
	Manchester	54 (88.5)	61	61 (100)	61
Trent	Nottingham	75 (88.2)	85	82 (96.5)	84
West Midlands	Birmingham	105 (87.5)	120	109 (90.8)	119
East	Cambridge	33 (84.6)	39	33 (84.6)	33
North Thames	Great Ormond St	60 (76.9)	78	39 (50)	45
	Chelmsford	42 (85.7)	49	46 (93.9)	48
The Spires	Oxford & Salisbury	63 (84)	75	65 (89)	73
South Wales & South West	Swansea	30 (83.3)	36	36 (100)	36
	Bristol	38 (70.4)	54	53 (98.1)	54
South Thames	GSTT	84 (74.3)	113	108 (95.6)	112
Northern Ireland	Belfast	28 (84.8)	33	29 (87.9)	33
All	All	757 (81.4)	930	844 (90.8)	882

[a] 84/1,014 (8.3%) missing referral time and [b] 132/1,014 (13%) missing contact time and excluded in 'All' values. Note: MCN - Managed Clinical Network.

3.6. Five-year outcomes among children born with a cleft lip and/or palate

Five-year outcomes include height and weight, decayed, missing and filled teeth (dmft), the Five Year Old Index, and the Cleft Audit Protocol for Speech – Augmented (CAPS-A) scores.

3.6.1. Reporting of outcomes

Table 9 shows the number of consented children born between 2004 and 2009 with reported outcomes at five years of age (excluding children with submucous cleft palates)[31], according to region / unit.

- There is still a very high proportion of missing data for five-year old weight and height (for eligible children 67.2% and 68.4% missing respectively), this is despite a small improvement in reporting of these data since last year's Annual Report (1.8% for weight and 1.3% for height). There was wide variation in reporting weight and height data across regions. This ranged from more than 75% for both weight & height in the Northern & Yorkshire region, to less than 5% for both weight & height in Northern Ireland and the Spires. This suggests that this data is not routinely collected in these regions.
- It is important to note improvements (of more than 8%) in reporting of weight and height by the regions of the East and the North Thames, which is encouraging.
- The proportion of eligible children with reported decayed, missing, filled teeth (dmft) index scores varied across regions from 8.8% (East) to 88.8% (West Midlands).
- It is acknowledged that sometimes there are reasons outside the units' control as to why outcome(s) data cannot be collected, and we encourage centres to report these. For example, the regions of Trent and the East have not submitted dmft data in past years as they have not had a paediatric dentist who can examine children to determine the dmft[32]. The East regional centre has recently appointed a paediatric dentist so it is anticipated that their data completeness will show improvement in future reports.
- There was wide variation in reporting of Five Year Old Index data across the regions/units. The proportion of children with a Five Year Old Index score reported has increased by 11.5% in the last two years (from 62% for 2013 data and 57.5% for 2012 data), which is very encouraging. It is hoped that this proportion will continue to increase over the next few years.
- Reporting of Five Year Old Index data across regions ranged from 12.1% (Northern Ireland) to 92.7% (the Spires). Although Northern Ireland submitted data for only 12.1% of their eligible patients, they have collected Five Year Old Index data for the first time – this indicates that study models are now being taken of 5 year olds in this region. This is a positive development for reporting this outcome.
- The proportion of eligible children with Speech outcome scores ranged from 48.1% at GOSH to 87.5% at Swansea. Given that this is only the second year that the expanded 16 CAPS-A speech outcome scores have been requested, CRANE is encouraged by the fact that regions/units have reported speech data on an average of 63.8% of cases.
- It is positive to note that reporting has increased for all outcomes at five years of age[33] since this time last year.

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[31] Submucous cleft palate patients excluded from all five year outcomes as all/most teams do not audit these patients.

[32] Trent has previously informed CRANE that they do not have a paediatric dentist to collect these records around the region, and have not had adequate administrative support to provide CRANE with dmft data.

[33] 1.8% for weight, 1.3% for height, 1.9% for dmft, 7% for 5 year index and 2.7% for speech.

Table 9. Number (%) of CRANE-registered[a] consented children born between 2004 and 2009[b] with reported outcomes at five years of age (excluding children with submucous cleft palates)[34], according to region / unit.

Regional centre / MCN	Administrative Unit		Weight		Height		dmft			5 year index			Speech		
			2004-2009 births[c]				2004-2008 births[d]			2004-2008 births[e]			2007-2008 births[f]		
			Reported		Reported			Reported			Reported[35]			Reported	
		N	n	(%)	n	(%)	N	n	(%)	N	n	(%)	N	n	(%)
Northern & Yorkshire	Newcastle	342	265	(77.5)	262	(76.6)	284	248	(87.3)	37	32	(86.5)	89	60	(67.4)
	Leeds	377	288	(76.4)	294	(78)	318	222	(69.8)	48	43	(89.6)	91	52	(57.1)
North West	Liverpool	367	64	(17.4)	64	(17.4)	295	223	(75.6)	63	48	(76.2)	107	56	(52.3)
	Manchester	415	145	(34.9)	142	(34.2)	352	267	(75.9)	47	39	(83)	112	75	(67)
Trent	Nottingham	523	75	(14.3)	76	(14.5)	440	84	(19.1)	70	35	(50)	123	83	(67.5)
West Midlands	Birmingham	593	271	(45.7)	264	(44.5)	493	438	(88.8)	89	74	(83.1)	146	102	(69.9)
East	Cambridge	378	89	(23.5)	83	(22)	306	27	(8.8)	67	41	(61.2)	111	66	(59.5)
North Thames	Great Ormond Street	457	111	(24.3)	111	(24.3)	355	156	(43.9)	44	20	(45.5)	131	63	(48.1)
	Chelmsford	199	69	(34.7)	68	(34.2)	155	63	(40.6)	18	12	(66.7)	44	26	(59.1)
The Spires	Oxford & Salisbury	530	24	(4.5)	24	(4.5)	449	316	(70.4)	82	76	(92.7)	138	91	(65.9)
South Wales & South West	Swansea	260	132	(50.8)	95	(36.5)	214	164	(76.6)	27	8	(29.6)	64	56	(87.5)
	Bristol	330	114	(34.5)	115	(34.8)	277	167	(60.3)	37	26	(70.3)	100	66	(66)
South Thames	Guy's and St Thomas'	530	154	(29.1)	139	(26.2)	448	250	(55.8)	94	64	(68.1)	140	85	(60.7)
Northern Ireland	Belfast	199	2	(1.0)	2	(1.0)	169	94	(55.6)	33	4	(12.1)	57	46	(80.7)
All	**All**	5,500	1,803	(32.8)	1,739	(31.6)	4,555	2,719	(59.7)	756	522	(69)	1,453	927	(63.8)

[a] Registered in CRANE by 6 August 2015. Note: MCN – Managed Clinical Network.

[b] 96/5,493 (1.7%) children died before 5 years and are excluded from all measures presented in this table (accounting for minor discrepancies between totals above and footnotes below).

[c] Children born in 2004-2009: 199/5,699 (3.5%) children with submucous cleft palates are excluded from weight and height data.

[d] Children born in 2004-2008: 170/4,725 (3.6%) children with submucous cleft palates are excluded from dmft data.

[e] Children born in 2004-2008: Only complete unilateral cleft lip & palate (UCLP) reported as children with all other cleft types (301/1,060 = 28.4%) are excluded from 5-year old index data.

[f] Children born in 2007-2008: 588/2,013 (29.2%) children with submucous cleft palates (2.6%), missing one or more of all 16 CAPS-A data items (1.4%), or born with either a CL (24%) or a non-specified cleft type (1.2%) are excluded from speech data. Details of additional excluded cases can be found in Table 13.

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[34] Submucous cleft palate patients excluded from all the five year outcomes as all/most teams do not audit these patients.

[35] Only 6/14 units provided data for more than 75% of their eligible patients, as recommended by the Orthodontic Special Interest Group at the 2012 Craniofacial Society of Great Britain and Ireland Annual Conference, and therefore scores should be interpreted with caution. The small number of patients with reported scores within each region / unit (4-76) means that statistical comparison between units is not currently appropriate. CRANE will continue to collect these outcomes over the next few years, and as numbers increase, meaningful comparison between units will become possible.

3.6.2. Weight and height (2004-2009 births)

Five-year weight and height were reported for 32.8% and 31.6%, respectively, for the 5,500 children born in 2004-2009[36], who were alive at five years of age. The mean (SD) weight was 19.6kg (3.2kg) while the mean (SD) height was 111.8cm (5.9cm). A small difference in mean weight existed between the sexes (boys 19.8kg vs. girls 19.5kg), and boys were marginally taller than girls (112.3 cm vs. 111.1cm).

3.6.3. Decayed missing and filled teeth (dmft) (2004-2008 births)

The dmft describes the amount of dental caries in an individual and is a measure of oral health. A dmft score reflects the total number of teeth that are decayed, missing or filled. The risk of dental caries is thought to be higher among children with a cleft lip and/or palate compared to children without an oral cleft[37]. We collect dmft data on CRANE-registered consented children at five years of age.

Among children with a reported dmft outcome[38] , 42.3% of children with a cleft had at least one (>0) decayed, missing or filled tooth. The mean number of dmft at five years among children registered in CRANE was 2, with scores ranging from 0 to 20. Two hundred and eighty-four children (17.1%) had a dmft score greater than 5.

Dental caries according to cleft type

Table 10 shows the prevalence of dental caries according to cleft type, with the mean dmft and the proportion of children with at least one dmft (>0 dmft) varying significantly according to cleft type ($p<0.001$).

The dmft data, obtained in 2005, available for five-year old children in the general population in England and Wales shows that 38.8% of five-year olds had at least one dmft, with a mean number of 1.5[39]. The comparable figure of 42.3% among CRANE-registered children (shown in Tables 10 and 11) is close (although slightly higher) to that of the general population. This is likely to be due to the fact that the number of dmft among children with a CL was lower than the general population (33.9% versus 38.8%). Despite this, the mean dmft was higher among CP, UCLP and BCLP patients compared to the general population's mean (0.9, 0.5 and 1.1, respectively, compared to a mean of 1.5).

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[36] Submucous cleft palate patients excluded from all five year outcomes as all/most teams do not audit these patients.

[37] (1) Al-Dajani M. Comparison of dental caries prevalence in patients with cleft lip and/or palate and their sibling controls. The Cleft Palate-Craniofacial Journal, 2009. 46(5): p. 529-531. (2) Britton, KF and Welbury, RR, Dental caries prevalence in children with cleft lip/palate aged between 6 months and 6 years in the West of Scotland. European Archives of Paediatric Dentistry, 2010. 11 (5): p. 236-241.

[38] Submucous cleft palate patients excluded from all five year outcomes as all/most teams do not audit these patients.

[39] Dental Health Services Research Unit from National Health Service - British Society for the Study of Community Dentistry. Dental caries experience of 5-year-old children in Great Britain 2005/2006. 2011, Available from: http://www.dundee.ac.uk/tuith/search/bdsearch.html.

Table 10. Number (%) of CRANE-registered consented children born between 2004 and 2008 with a cleft lip and/or palate according to the number of decayed, missing or filled teeth (dmft) at age five years and cleft type.

	Number of decayed, missing or filled teeth (dmft)				
		0	>0		
Cleft type	Mean (95% CI)	n (%)	n (%)	(95% CI)	**All**[a]
CL	1.2 (1.0 to 1.4)	369 (66.1)	189 (33.9)	(29.9 to 37.8)	558
CP	2.4 (2.1 to 2.6)	644 (59)	447 (41)	(38 to 43.9)	1091
UCLP	2.0 (1.8 to 2.2)	405 (52.9)	360 (47.1)	(43.5 to 50.6)	765
BCLP	2.6 (2.2 to 3.1)	135 (48.4)	144 (51.6)	(45.7 to 57.5)	279
Not specified	1.6 (0.04 to 3.1)	17 (65.4)	9 (34.6)	(15 to 54.2)	26
All	2.0 (1.9 to 2.2)	1,570 (57.7)	1,149 (42.3)	(40.4 to 44.1)	2,719

[a] 1,836/4,555 (40.3%) children with missing dmft data, 173/4,812 (3.6%) children with submucous clefts, and 84/4,639 (1.8%) children who died before the age of five were excluded in 'All' values. Note: CL - Cleft Lip, CP - Cleft Palate, UCLP - Unilateral cleft lip and palate, and BCLP - Bilateral cleft lip and palate.

The fact that dmft were submitted for only 59.7% of children means that these data should be interpreted with caution, as it is possible that the overall findings from the limited data made available to CRANE may not be representative of the entire cleft population. Analyses of data from a greater number of children are necessary to examine true differences that may exist between the cleft population and general population, and between cleft types.

Dental caries according to region / unit

Table 11 shows the prevalence of dental caries according to region / unit. There was a significant variation in dmft scores across Units (p<0.001). Children registered in the Trent and East regions had the highest numbers of mean dmft (3.1 and 3.0 respectively), which were significantly different to the overall mean (2.0). It should be noted that Trent and the East submitted data for few patients[40], and it is possible that dmft data were collected for only those who were referred to the dentist because of problems. This could explain their high caries rate. Data from a larger and more representative sample from Trent and the East are required. The Bristol unit and Spires region had mean dmft values that were significantly lower than the overall mean (1.3 and 1.4 respectively[41]).

In terms of the proportion of cleft children with at least one dmft (>0 dmft), the Spires region had the lowest proportion (32.6%), which was significantly different to the overall proportion among cleft children. Whilst the proportion of cleft children with at least one dmft varied between regions, for the majority of regions their rate does not seem to differ substantially from their region's total population rate[42].

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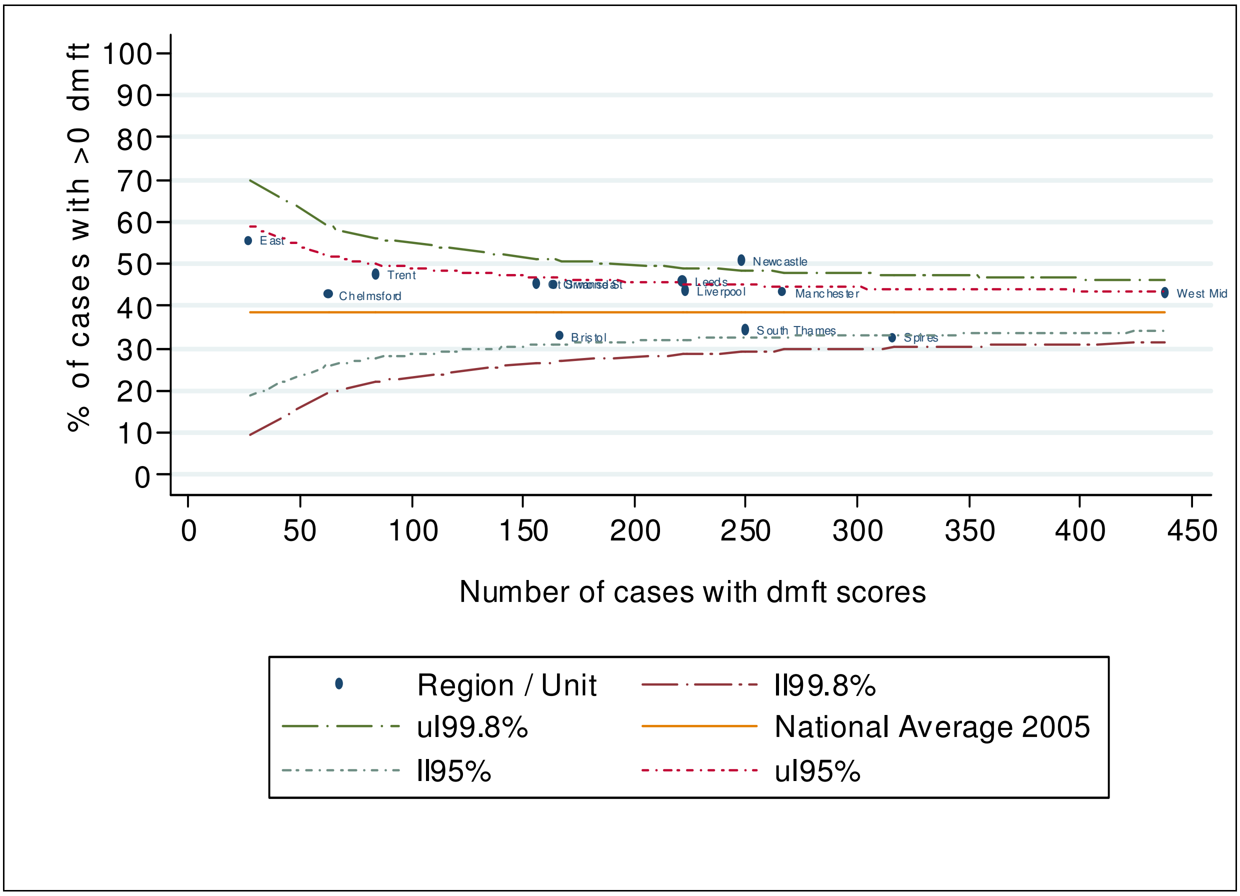
[40] Note that dmft data were reported for only 19.1% of eligible children registered by the Trent region and 8.8% of eligible children registered by the East region (as reported in Table 9).

[41] With dmft data reported for more than 60% of eligible children at these sites (as reported in Table 9).

[42] Dental Health Services Research Unit from National Health Service - British Society for the Study of Community Dentistry data. Dental Caries Experience of 5-year-old Children in Great Britain 2005 / 2006. Available from: http://www.app.dundee.ac.uk/tuith/search/tables/tab2005_6.htm.

The funnel plot[43] in Figure 1 further demonstrates the proportion of five-year olds with at least one decayed missing or filled tooth (>0 dmft) according to the number of children with valid dmft scores at each region/ unit. This funnel plot is centred on the national average of 38.8% (with at least one dmft), obtained in 2005, for five-year old children in the general population in England and Wales[44].

Figure 1. Funnel plot of five-year olds (born between 2004 and 2008) with at least one dmft (>0 dmft), according to the number of children at each region / unit with dmft scores.



Note: This funnel plot is centred on the national average of 38.8% (with at least one dmft), obtained in 2005, for five-year old children in the general population in England and Wales. Plus, only 13 units shown in the Figure above as Northern Ireland data excluded (as centring national average is for England and Wales only).

Figure 1 shows that most regions' / units' rates of at least one dmft (>0 dmft) fall within the expected range given the number of children with valid dmft scores at their region / unit. No site has a >0 dmft rate below the lower 99.8% control limit, and one unit (Newcastle) has a >0 dmft rate above the upper 99.8% control limit. This means they have significantly high rates of children with >0 dmft – which is unlikely to be as a result of chance and is worth investigating (more information on funnel plots can be found in the Glossary at the front of this report).

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[43] This funnel plot is calculated using valid data as denominators (not considering missing data), subject to the same inclusions and exclusions as data in Table 11. In addition, it is not adjusted (or risk adjusted) in any way.

[44] Dental Health Services Research Unit from National Health Service - British Society for the Study of Community Dentistry. Dental caries experience of 5-year-old children in Great Britain 2005/2006. 2011. Available from: http://www.dundee.ac.uk/tuith/search/bdsearch.html.

Table 11. Number (%) of CRANE-registered consented children born between 2004 and 2008 with a cleft lip and/or palate – according to the number of decayed, missing or filled teeth (dmft) and the average treatment index at age five years by region / unit.

Regional centre / MCN	Administrative Unit	Number of decayed, missing or filled teeth (dmft)					Treatment Index		Care Index	
			0	>0		All[a] (N)		All[b] (N)		All[b] (N)
		Mean (95% CI)	n (%)	n (%)	95% CI		Average (%)		Average (%)	
Northern & Yorkshire	Newcastle§¥	2.8 (2.2 to 3.3)	122 (49.2)	126 (50.8)	(44.5 to 57.1)	248	(68.1)	218	(59.3)	220
	Leeds§¥	2.6 (2.1 to 3.2)	120 (54.1)	102 (45.9)	(39.3 to 52.6)	222	(69.1)	221	(61.7)	221
North West & North Wales	Liverpool	2.1 (1.6 to 2.5)	125 (56.1)	98 (43.9)	(37.4 to 50.5)	223	(68.1)	219	(60.7)	219
	Manchester	2.1 (1.7 to 2.6)	151 (56.6)	116 (43.4)	(37.5 to 49.4)	267	(70)	266	(63.6)	266
Trent	Nottingham¥*	3.1 (2 to 4.2)	44 (52.4)	40 (47.6)	(36.7 to 58.5)	84	(76.3)	81	(61.3)	82
West Midlands	Birmingham§¥	2.0 (1.7 to 2.4)	249 (56.8)	189 (43.2)	(38.5 to 47.8)	438	(68.9)	437	(62)	437
East	Cambridge*	3.0 (1.4 to 4.7)	12.0 (44.4)	15.0 (55.6)	(35.5 to 75.6)	27	(54.7)	27	(45.7)	27
North Thames	Gr Ormond St¥	2.2 (1.6 to 2.8)	85 (54.5)	71 (45.5)	(37.6 to 53.4)	156	(72.3)	154	(58.5)	154
	Chelmsforc	1.5 (0.8 to 2.2)	36 (57.1)	27 (42.9)	(30.3 to 55.4)	63	(83.7)	45	(82.9)	45
The Spires	Oxford/Salis.	1.4 (1.1 to 1.7)	213 (67.4)	103 (32.6)	(27.4 to 37.8)	316	(96.6)	225	(95.9)	225
South Wales & South West	Swansea§¥	2.0 (1.5 to 2.4)	90 (54.9)	74 (45.1)	(37.4 to 52.8)	164	(75.9)	163	(63.1)	164
	Bristol§¥	1.3 (0.9 to 1.6)	112 (67.1)	55 (32.9)	(25.7 to 40.1)	167	(82.3)	164	(76.9)	164
South Thames	GSTT§¥	1.5 (1.1 to 1.8)	164 (65.6)	86 (34.4)	(28.5 to 40.3)	250	(84.2)	246	(79.2)	247
Nrthn. Ireland	Belfast	2.4 (1.6 to 3.2)	47 (50)	47 (50)	(39.7 to 60.3)	94	(75.6)	93	(63.3)	93
All	All	2.0 (1.9 to 2.2)	1,570 (57.7)	1,149 (42.3)	(40.4 to 44.1)	2,719	(74.9)	2559	(67.5)	2,564

[a] 1,836/4,555 (40.3%) children with missing dmft data, 173/4,812 (3.6%) children with submucous clefts[45], and 84/4,639 (1.8%) children who died before the age of five were excluded in 'All' values. Note: MCN – Managed Clinical Network.

[b] Only including children who were alive after the age of five, without submucous clefts, and who had either a dmft score of 0[46] or scores for all relevant dmft data items (to allow calculation of treatment and care index scores).

Individual unit considerations: §British Association for the Study of Community Dentistry (BASCD) calibrated assessor. *dmft data not been submitted in years past as no paediatric dentist in place to examine children (determining dmft) or no administrative support in place to submit data to CRANE – it is anticipated these units' data completeness will show improvement in future reports. ¥Specialist paediatric dentist.

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[45] Submucous cleft palate patients excluded from all five year outcomes as all/most teams do not audit these patients.

[46] If a dmft score for an individual is 0 then the treatment index and care index = 1 (100%) as there is no untreated disease.

Dental Treatment and Care Indices

Table 11 also shows the average treatment index and care index (reported for the first time) for children according to region / unit. Both indices are calculated from the dmft[47], as raw dmft scores give a figure for dental disease experienced but do not distinguish if there is active disease present at the time or not (treatment index) or the proportion of children who have received care in the form of fillings (care index).

The treatment index reflects whether the mouth is dentally fit at that moment in time. i.e. If dental disease has occurred, the treatment index indicates the extent to which it has been dealt with and the degree to which the child has been rendered free from active decay. When calculated, treatment indices range from 0 to 1 and are usually expressed as a percentage[48]. Treatment indices with a value of 1 (100%) indicate that there is no untreated disease, which is the desired outcome. Furthermore, average treatment indices of 100% can be indicators of having mechanisms in place to deal with any disease occurring, and thereby providing the child with a dentition where the disease is controlled and the child has a pain free mouth.

For the 2,559 children with dmft scores of 0 or scores for all three ‘m’, ‘f’ and ‘dmft’ data items – to allow calculation of the treatment index – there was significant variation in treatment index scores across units ($p<0.001$). Children registered by the East region[49] had the lowest average proportion of treated disease (54.7% equal to 20.2% less than the national average), while the Spires region had highest average proportion of treated disease (96.6% equal to 21.7% more than the national average).

The care index reflects cases where children have experience dental decay, which has been identified at the earliest possible stage (which is preferable), and have been provided with care in the least invasive form possible – in the form of fillings. When calculated, care indices also range from 0 to 1 and are usually expressed as a percentage[50]. Care indices with a values close to 1 (100%) indicate that there are high levels of care provided by filling (not extraction or no treatment), which is the desired outcome. Conversely in situations where levels of care low (and decay could be addressed by filling but has not) the care index is close to 0%. Furthermore, average care indices of 100% can be indicators of having mechanisms in place to increase levels of care in relation to fillings.

For the 2,564 children with dmft scores of 0 or scores for both ‘f’ and ‘dmft’ data items, to allow calculation of the care index, there was significant variation in care index scores across Units ($p<0.001$). Children registered by the East region had the lowest average care index (45.7% equal to 21.8% less than the national average), which means decay that could be treated by fillings has remained untreated. Meanwhile the Spires region had the highest average proportion of children receiving care by filling (95.9% equal to 28.4% more than the national average).

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[47] Treatment Index calculated as = (Total number of missing teeth in primary dentition (m) + Total number of filled teeth in primary dentition (f)) / ‘Total number of decayed, missing or filled teeth in primary dentition (dmft).
Care Index calculated as = Total number of filled teeth in primary dentition (f) / ‘Total number of decayed, missing or filled teeth in primary dentition (dmft).

[48] If a dmft score for an individual is 0 then the treatment index is 1 (100%) as there is no untreated disease.

[49] Recent appointment of a paediatric dentist to examine children (and determine dmft) is expected to result in improved data completeness published in future reports.

[50] If a dmft score for an individual is 0 then the care index is 1 (100%) as there is no disease.

It is also worth considering that regional differences in the levels of dental disease will not only be affected by the dental care received by children. Oral health will also be affected by deprivation, cultural differences in attitudes to dental health and water fluoridation levels. A systematic review found that water fluoridation is associated with an increased proportion of children without caries and a reduction in the number of teeth affected by caries[51]. Fluoridation levels vary within and between regions throughout the UK. For example, parts of the West Midlands and parts of the North East receive fluoridated water, whereas other areas do not. Interestingly, data from 2005 revealed the West Midlands had one of the lowest proportions of five year olds with >0 dmft in the general population, while the North East had the highest proportion (50%)[52]. Accurate water fluoridation data will be useful for interpreting dmft regional differences and allowing for risk adjustment in the long term.

3.6.4. Five Year Old Index (2004-2008 births)

Dental models of five-year old children with a complete UCLP were assessed using the Five Year Old Index to examine dental arch relationships. The index evaluates the effects of primary surgery on the facial growth of children with UCLP before any other interventions, such as orthodontics or alveolar bone grafting, which may influence this growth further[53]. Dental arch relationships at five years are thought to predict treatment outcome in terms of facial growth on a population basis rather than at the individual child level[54]. The Five Year Old Index may, therefore, also be used to compare treatment outcomes between centres and surgeons. Patients categorised as ‘1’ and ‘2’ on the index are considered to have the best possible outcome, while those categorised as ‘4’ and ‘5’ are thought to have very poor outcomes in terms of facial growth, and they may benefit from further surgery to correct their facial disproportion once facial growth is complete.

Table 12 shows the distribution of Five Year Old Index scores as provided by all regions/units. The majority of these scores were externally validated (in 472/522 (90.4%) of eligible cases), and where externally validated scores were unavailable, internal scores were included in the analysis (in 50/522 (9.6%) of cases). Overall, 43.9% of complete UCLP patients born between 2004 and 2008 had Five Year Old Index scores in the two groups considered to have the best possible dental arch relationships (scores ‘1’ or ‘2’) while 25.1% of children had scores ‘4’ or ‘5’, reflecting poor dental arch relationships. This represents an improvement, compared to the CSAG findings that 36% (of 223 cleft children) had poor dental arch relationships at five years old in 1998[55].

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[51] McDonagh M, Whiting P, Bradley M, Cooper J, Sutton A, Chestnutt I, et al. A systematic review of public water fluoridation. BMJ, 2000. 321: p. 855-859.

[52] Dental Health Services Research Unit from National Health Service - British Society for the Study of Community Dentistry data. Dental Caries Experience of 5-year-old Children in Great Britain 2005 / 2006. Available from: http://www.app.dundee.ac.uk/tuith/search/tables/tab2005_6.htm.

[53] Johnson N, Williams AC, Singer S, Southall P, Atack N and Sandy JR. Dentoalveolar relations in children born with a unilateral cleft lip and palate (UCLP) in Western Australia. The Cleft Palate-Craniofacial Journal, 2000. 37 (1): p. 12-16.

[54] Atack N, Hathorn IS, Semb G, Dowell T and Sandy JR. A new index for assessing surgical outcome in unilateral cleft lip and palate subjects aged five: reproducibility and validity. The Cleft Palate-Craniofacial Journal, 1997. 34 (3): p. 242-246.

[55] Clinical Standards Advisory Group, Clinical Standards Advisory Group. Report of a CSAG Committee on cleft lip and/or palate, 1998, The Stationery Office, London.

Table 12. Number (%) of CRANE-registered consented children born between 2004 and 2008 with a complete unilateral cleft lip and palate[56], according to Five Year Old Index scores and region / unit.

Regional centre / MCN	Administrative Unit	Five Year Old Index n (%)					
		1	2	3	4	5	All[a]
Northern & Yorkshire	Newcastle	5 (15.6)	4 (12.5)	14 (43.8)	5 (15.6)	4 (12.5)	32
	Leeds	5 (11.6)	16 (37.2)	12 (27.9)	9 (20.9)	1 (2.3)	43
North West & North Wales	Liverpool	4 (8.3)	15 (31.3)	15 (31.3)	10 (20.8)	4 (8.3)	48
	Manchester	2 (5.1)	15 (38.5)	10 (25.6)	8 (20.5)	4 (10.3)	39
Trent	Nottingham	2 (5.7)	15 (42.9)	9 (25.7)	4 (11.4)	5 (14.3)	35
West Midlands	Birmingham	7 (9.5)	24 (32.4)	25 (33.8)	13 (17.6)	5 (6.8)	74
East	Cambridge	3 (7.3)	14 (34.1)	14 (34.1)	9 (22)	1 (2.4)	41
North Thames	Great Ormond Street	1.0 (5)	9.0 (45)	6.0 (30)	4.0 (20)	0.0 (0)	20
	Chelmsford	2.0 (16.7)	5.0 (41.7)	3.0 (25)	2.0 (16.7)	0.0 (0)	12
The Spires	Oxford & Salisbury	9 (11.8)	25 (32.9)	22 (28.9)	16 (21.1)	4 (5.3)	76
South Wales & South West	Swansea	0 (0)	1 (12.5)	4 (50)	2 (25)	1 (12.5)	8
	Bristol	1 (3.8)	10 (38.5)	5 (19.2)	8 (30.8)	2 (7.7)	26
South Thames	Guy's and St Thomas'	6 (9.4)	28 (43.8)	21 (32.8)	6 (9.4)	3 (4.7)	64
Northern Ireland	Belfast	0.0 (0)	1.0 (25)	2.0 (50)	1.0 (25)	0.0 (0)	4
All	All	47 (9)	182 (34.9)	162 (31)	97 (18.6)	34 (6.5)	522

[a] 301/1,060 (28.4%) children with an incomplete UCLP, 11/767 (1.4%) children who died before the age of five, and 234/756 (30.9%) children missing Five Year Old Index scores data excluded in 'All' values. Note: MCN – Managed Clinical Network.

The funnel plot[57] in Figure 2 further demonstrates the proportion of five-year olds with the poorest (4 and 5) Five Year Old Index outcome scores according to the number of children at each region / unit with index scores. It is centred on the national average of poor Five Year Old Index scores across all units of 25.1%[58].

Figure 2 shows that all regions' / units' rates of poor index scores fall within the expected range given the number of children (born between 2004-2008) with valid index scores at their unit. i.e. No unit has a 'poor index score rate' below the lower 99.8% control limit or above the upper 99.8% control limit (more information on funnel plots can be found in the Glossary at the front of this report).

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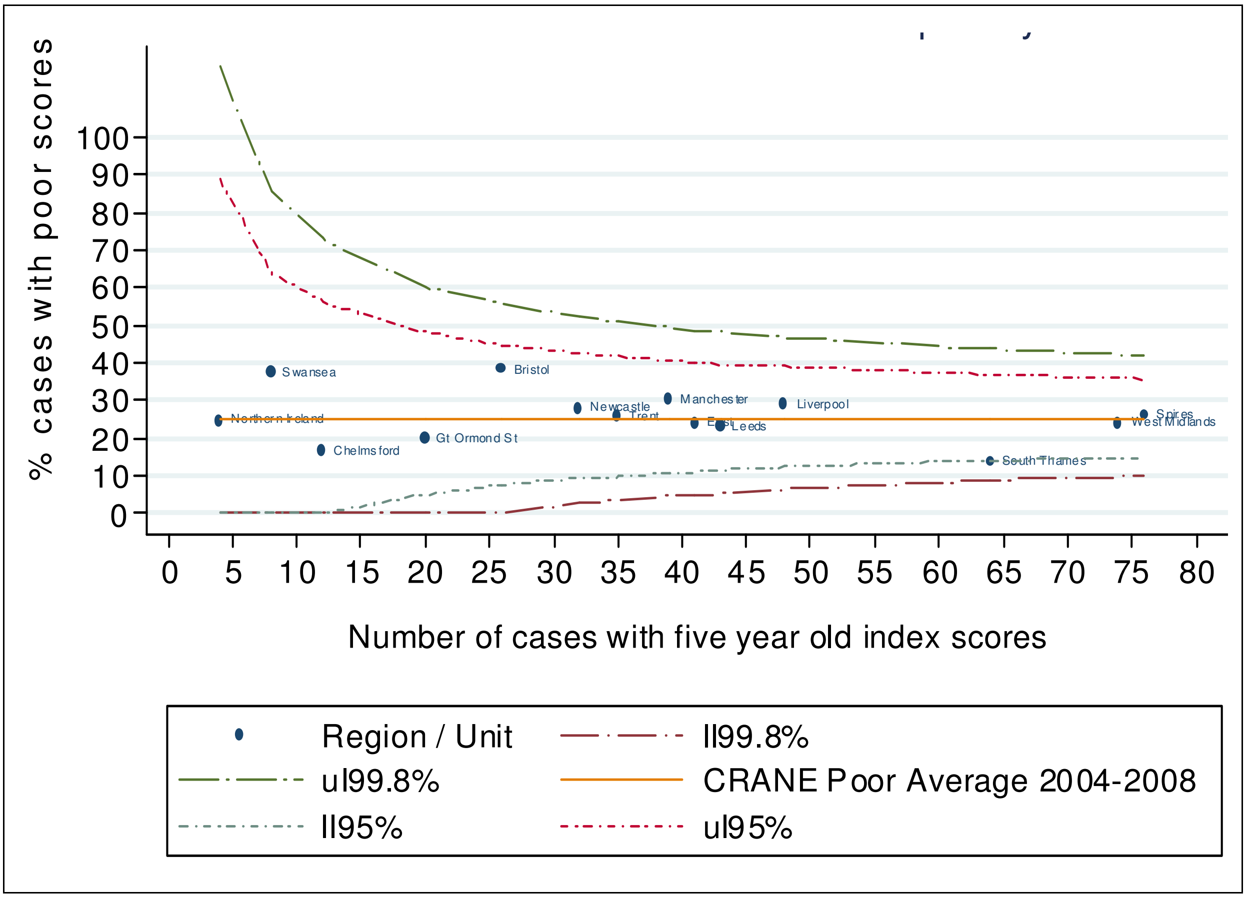
[56] Submucous cleft palate patients excluded from all five year outcomes as all/most teams do not audit these patients.

[57] This funnel plot is calculated using valid data as denominators (not considering missing data), subject to the same inclusions and exclusions as data in Table 12. In addition, it is not adjusted (or risk adjusted) in any way.

[58] Versus the 36% national average identified by Clinical Standards Advisory Group (CSAG). Report of a CSAG Committee on cleft lip and/or palate, 1998, The Stationery Office, London.

Figure 2. Funnel plot of five-year olds (born between 2004 and 2008) with poor Five Year Old Index scores according to the number of children at each region / unit with index scores.



Note: This funnel plot is centred on the national average (for 2004-2008 births reported in CRANE) of poor Five Year Old Index scores across all units of 25.1%.

The fact that Five Year Old Index scores were submitted for only 69.1% of children, and the small number of children within each region / unit (ranging from 4 to 76), means that the data presented in this section should be interpreted with caution, as it is possible that the overall findings from the limited data made available to CRANE may not be representative of the cleft population. Analyses of data from a greater number of children are necessary to examine true differences that may exist between the cleft population and general population, and between cleft types.

3.6.5. Cleft Audit Protocol for Speech – Augmented scores (2007-2008 births)

The Cleft Audit Protocol for Speech – Augmented (CAPS-A) score has been used to assess speech among children with a cleft affecting the palate (CP, UCLP and BCLP). This is only the second year that 16 CAPS-A speech outcome scores have been requested[59]. The parameters of speech assessed include:

- Resonance (hypernasality and hyponasality) and nasal airflow (audible nasal emission and nasal turbulence). These are structurally related speech difficulties reflecting aspects such as the ability of the palate to close off the nasal airway during speech.
- 12 individual cleft speech characteristics (CSCs) grouped into four categories of CSCs – anterior oral, posterior oral, non-oral and passive – are also assessed. These reflect articulation patterns which can affect the clarity and intelligibility of a child's speech.

Table 13 shows that 63.8% of consented children born in 2007 and 2008 had reported speech outcomes for all 16 CAPS-A parameters, and the distribution of proportions by region / unit. Reasons why speech scores were *excluded* or *unavailable* were provided by regions / units for 27.7% of children.

Table 13. Number (%) of CRANE-registered[a] consented children born with a cleft palate (2007-2008), with speech outcome data and exclusion reasons at five years of age, according to region / unit.

Regional centre / MCN	Administrative Unit	Speech[b]							
			Reported		Exclusion reason		Not Available Reason		Total Acc. For
		N	n	(%)	n	(%)	n	(%)	(%)
Northern	Newcastle	89	60	(67.4)	17	(19.1)	10	(11.2)	(97.8)
& Yorkshire	Leeds	91	52	(57.1)	18	(19.8)	19	(20.9)	(97.8)
North West	Liverpool	107	56	(52.3)	17	(15.9)	31	(29)	(97.2)
& North Wales	Manchester	112	75	(67)	32	(28.6)	2	(1.8)	(97.3)
Trent	Nottingham	123	83	(67.5)	27	(22)	8	(6.5)	(95.9)
West Midlands	Birmingham	146	102	(69.9)	7	(4.8)	9	(6.2)	(80.8)
East	Cambridge	111	66	(59.5)	25	(22.5)	15	(13.5)	(95.5)
North Thames	Great Ormond Street	131	63	(48.1)	15	(11.5)	15	(11.5)	(71)
	Chelmsford	44	26	(59.1)	8	(18.2)	8	(18.2)	(95.5)
The Spires	Oxford & Salisbury	138	91	(65.9)	19	(13.8)	22	(15.9)	(95.7)
South Wales	Swansea	64	56	(87.5)	7	(10.9)	1	(1.6)	(100)
& South West	Bristol	100	66	(66)	8	(8)	5	(5)	(79)
South Thames	Guy's and St Thomas'	140	85	(60.7)	29	(20.7)	21	(15)	(96.4)
Northern Ireland	Belfast	57	46	(80.7)	8	(14)	0	(0)	(94.7)
All	**All**	1,453	927	(63.8)	237	(16.3)	166	(11.4)	(91.5)

[a] Registered in CRANE by 6 August 2015. Note: MCN - Managed Clinical Network. [60]

[b] Excluding children who died before the age of 5 years and 588/2,013 (29.2%) children with submucous cleft palates (2.6%), missing one or more of all 16 CAPS-A data items (1.4%), or born with either a CL (24%) or a non-specified cleft type (1.2%) are excluded from speech data.

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[59] Only four Cleft Speech Categories (CSCs) summarising the all 12 CSCs were collected for 2006 births. Because of this, 2006 speech data is not included in this report. Changes to expand data collection to 12 CSCs were made in 2014.

[60] 80 cases had both 'Exclusion reason' and 'Not available reason' codes – as completion of these sections is not currently mutually exclusive. Therefore those with an 'Exclusion reason' were excluded from the 'Not available reason' data.

Consented children born in 2007 and 2008 were reported as *excluded* in 16.3% of cases where, for example, patients had emigrated or transferred out of the area[61]. In 11.4% of cases children's data was *unavailable*, such as when patients were not seen (e.g. due to a lack of staff or facilities) or when it was not possible to take a record (e.g. for reasons unrelated to the patient). Rates of *excluded* or *unavailable* data varied widely between regions / units. This ranged from 45% in the Liverpool to 11% in the West Midlands.

Overall, rates of data completeness ranged from 71% of children being accounted for at GOSH to 100% of children being accounted for in Swansea. This was either in the form of speech data or *excluded/ unavailable* data (see Appendix 6 for detail of missing data).

The scores for each of the individual 16 CAPS-A assessed are presented in Tables 14 and 15, found in the next two sections.

Resonance and Nasal Airflow

In Table 14, scores are colour-coded as green when the child's palate is functioning well in terms of the assessed parameter. No action, either speech therapy or surgery, would be required with green scores. Amber for hyponasality is indicative of nasal obstruction, while amber or red for hypernasality, nasal emission or nasal turbulence are indicative of structurally-related speech difficulties that may involve palate function and/or palatal fistulae. These difficulties may require surgical treatment.

Table 14. Number (%) of CRANE-registered [a] consented children born with a cleft palate in 2007-2008, according to the four parameters for resonance and nasal airflow

Description	**Score**	**N[b]**	**(%)**
RESONANCE – HYPERNASALITY			
Absent	**0**	709	(76.5)
Borderline – minimal	**1**	109	(11.8)
Mild – evident on close vowels	**2**	61	(6.6)
Moderate – evident on open and close vowels	**3**	31	(3.3)
Severe – evident on vowels and voiced consonants	**4**	17	(1.8)
RESONANCE – HYPONASALITY			
Absent	**0**	772	(83.3)
Mild – partial dentalization of nasal consonants and adjacent vowels	**1**	140	(15.1)
Marked – dentalization of nasal consonants and adjacent vowels	**2**	15	(1.6)
NASAL AIRFLOW – AUDIBLE NASAL EMISSION			
Absent on pressure consonants	**0**	852	(91.9)
Occasional: pressure consonants affected <10% of the sample	**1**	60	(6.5)
Frequent: pressure consonants affected >10% of the sample	**2**	15	(1.6)
NASAL AIRFLOW – NASAL TURBULENCE			
Absent on pressure consonants	**0**	720	(77.7)
Occasional: pressure consonants affected <10% of the sample	**1**	168	(18.1)
Frequent: pressure consonants affected >10% of the sample	**2**	39	(4.2)
TOTAL		**927**	**(100)**

[a] Registered in CRANE by 6 August 2015. [b] Number of eligible children (as specified for Table 13).

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[61] Plus: Patient deceased, clinically contraindicated (this record type for this patient) and other reasons.

In terms of resonance, 5.1% of children with a hypernasality score had a score of ‘3’ or ‘4’, which means they had moderate or severe hypernasality i.e. nasal sounding speech (Table 14). This is indicative of velopharyngeal dysfunction (VPD), which is when the palate is unable to close off the nasal airway during speech. In addition, results of the Cleft Speech Characteristics (in Table 15) show that 2.9% of children had ‘weak and or nasalised consonants’ passive articulation errors and 1.4% of children had ‘nasal realisation of plosives’ passive articulation errors affecting three or more consonants, which are likely to be the consequence of VPD and is consistent with the hypernasality scorings.

It should be noted that, in order to achieve these outcomes, 178/921 (19.3%) of the children with reported surgical data[62] have had surgery for speech purposes (referred to as secondary speech surgery) before the age of five years.

In addition, 773 out of the 927 (83.4%) children with reported scores for all four resonance and nasal airflow parameters listed in Table 14 had all green scores, indicating that no structural problems existed in relation to these parameters.

Cleft Speech Characteristics (CSCs)

Table 15 presents the cleft speech characteristics (CSCs). A colour coding of green indicates the CSC is absent or considered to be a minor speech characteristic unlikely to require intervention. A colour coding of amber or red indicates the CSC is affecting one or more consonants to the extent that therapy and / or surgery may be required.

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[62] VP surgery/fistula repair data was only reported for 921/927 (99.3%) of eligible children (as specified for Table 13).

Table 15. Number (%) of CRANE-registered[a] consented children born with a cleft palate in 2007-2008, according to the twelve Cleft Speech Characteristics (CSCs) parameters.

Cleft Speech Characteristics (CSCs)		Score	N[b]	(%)
ANTERIOR ORAL CSCs	1. Dentalisation / Interdentalisation	A	747	(80.6)
		B	180	(19.4)
	2. Lateralisation / Lateral	A	866	(93.4)
		B	38	(4.1)
		C	23	(2.5)
	3 Palatalisation / Palatal	A	712	(76.8)
		B	112	(12.1)
		C	103	(11.1)
POSTERIOR ORAL CSCs	4. Double Articulation	A	901	(97.2)
		B	24	(2.6)
		C	2	(0.2)
	5. Backed to Velar / Uvular	A	788	(85)
		C	54	(5.8)
		D	85	(9.2)
NON ORAL CSCs	6. Pharyngeal Articulation	A	910	(98.2)
		C	13	(1.4)
		D	4	(0.4)
	7. Glottal Articulation	A	863	(93.1)
		C	29	(3.1)
		D	35	(3.8)
	8. Active Nasal Fricatives	A	851	(91.8)
		C	48	(5.2)
		D	28	(3)
	9. Double Articulation	A	911	(98.3)
		C	9	(1)
		D	7	(0.8)
PASSIVE CSCs	10. Weak and or nasalised consonants	A	870	(93.9)
		C	30	(3.2)
		D	27	(2.9)
	11. Nasal realisation of plosives	A	893	(96.3)
		C	21	(2.3)
		D	13	(1.4)
	12. Gliding of fricatives	A	904	(97.5)
		C	19	(2)
		D	4	(0.4)
		TOTAL	**927**	**(100)**

[a] Registered in CRANE by 6 August 2015. [b] Number of eligible children (as specified for Table 13).

‘Palatalisation / Palatal’ anterior oral CSCs were the most commonly occurring CSC, affecting 23.2% of children (12.1% with scores of B and 11.1% with scores of C). However, these may only have a minor effect on speech intelligibility, and, if treatment is indicated, this would probably involve speech therapy only. The more significant characteristics are the posterior, non-oral and passive CSCs, which are more likely to affect a child’s intelligibility. Therapy would often be indicated for these children, and/or further investigation of structure and possible surgery.

In addition, out of the 613/927 (66.1%) children with reported scores for all 12 CSC parameters listed in Table 15 had all green scores, indicating they did not exhibit cleft speech characteristics.

Nationally agreed Speech Outcome Standards

Further to reporting on the 16 CAPS-A speech parameters separately, we anticipate expanding our reporting of speech outcomes assessed at five years of age year on year. Specifically, we aim to report on the proportion of five-year olds meeting each of the following three nationally agreed Speech Outcome Standards[63]:

1. The achievement of *normal speech* (speech outcome standard #1) – this standard is achieved in cases where patients have normal (green) scores across all 16 CAPS-A speech parameters.
2. The presence of speech difficulties likely to be the result of existing or previous structural anomalies (speech outcome standard #2) – this standard is achieved in cases where patients have no reported history of surgery for speech purposes and have normal (green) scores across the following six CAPS-A speech parameters: Hypernasal resonance, both nasal airflow parameters (audible nasal emission and nasal turbulence), and all three Passive CSCs – as listed in Table 15.
3. The presence of cleft-related articulation difficulties (speech outcome standard #3) – this standard is achieved in cases where patients have normal (green) scores across the following 10 CSCs: All three Anterior Oral CSCs, both Posterior Oral CSCs, all four Non Oral CSCs, and gliding of fricatives (a Passive CSC) – as listed in Table 15.

Out of the 927 children with reported scores for all 16 CAPS-A speech parameters listed in Tables 14 and 15, 540 (58.3%) of children across all units had normal (green) scores across all 16 parameters. This means that the national speech outcome standard #1 target of 55% was met and exceeded on average by the CRANE cohort born in 2007-2008 (with the target of 55% based on the national outcome mean of 2004-06 speech outcome data[64]). Closer examination of the proportion of children achieving *normal speech* across the cleft types audited for speech (CP, UCLP & BCLP) highlighted a statistically significant difference between rates of *normal speech* reported by cleft type ($p<0.01$). This meant that significantly more children with a CP achieved *normal speech* (68% of children with CP achieved *normal speech*) and significantly fewer children with BCLP achieved *normal speech* (only 34.7% of children with BCLP had achieved *normal speech* by age 5).

The funnel plot[65] in Figure 3 goes on to show the proportion of children (born in 2007-2008) achieving *normal speech* according to the number of auditable children at each administrative unit with scores for all 16 CAPS-A speech parameters (more information on funnel plots can be found in the Glossary at the front of this report).

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[63] Britton L, Albery L, Bowden M, Harding-Bell A, Phippen G, and Sell D(2014) A Cross-Sectional Cohort Study of Speech in Five-Year-Olds With Cleft Palate ± Lip to Support Development of National Audit Standards: Benchmarking Speech Standards in the United Kingdom. The Cleft Palate-Craniofacial Journal: July 2014, Vol. 51, No. 4, pp. 431-451.

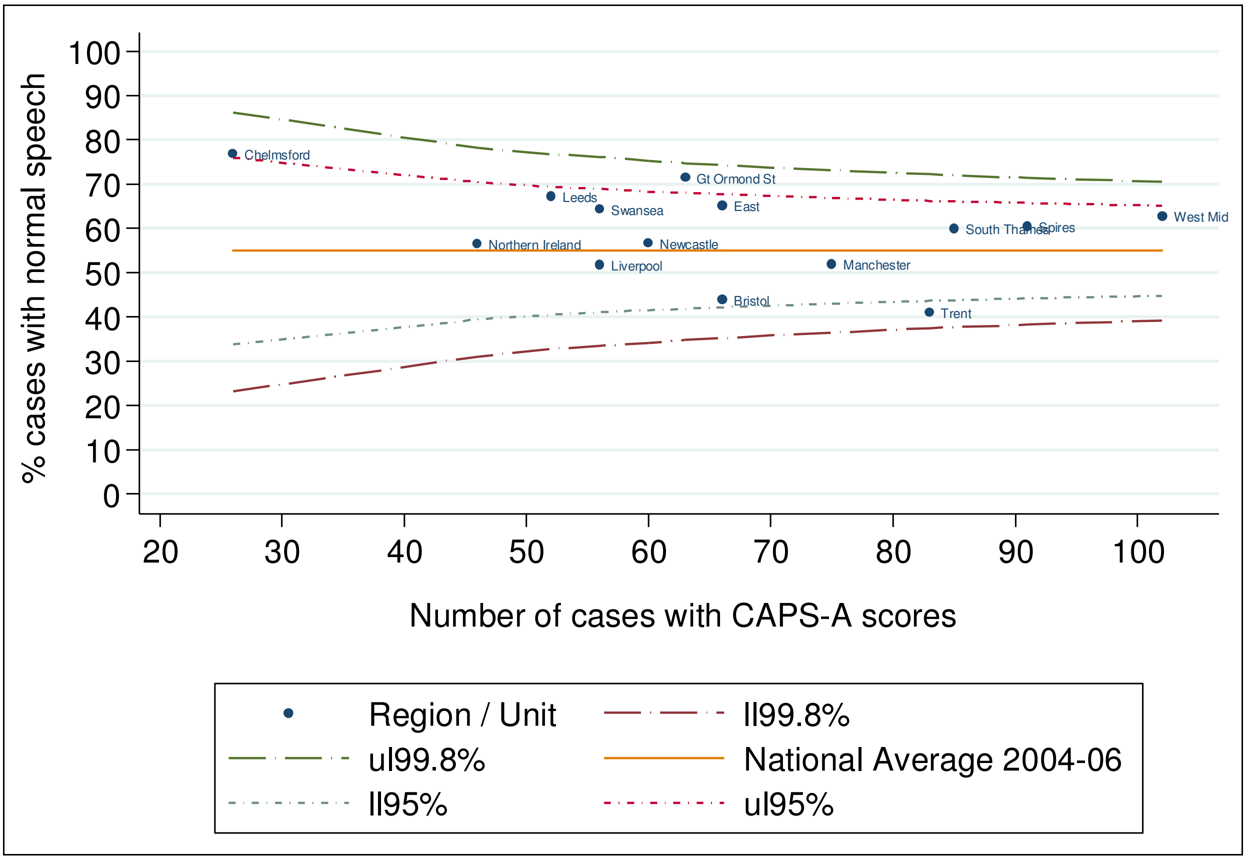
[64] Resulting from statistical analysis on 2004-06 Speech Outcome data completed by the Lead Speech and Language Therapy Group, with statistical support from the Cleft Collective in Manchester, and presented in April 2014 to the Leads group.

[65] This funnel plot is calculated using valid data as denominators (not considering missing data), subject to the same inclusions and exclusions as data in Tables 13, 14 & 15. In addition, it is not adjusted (or risk adjusted) in any way.

The funnel plot shows that rates of *normal speech*, for all administrative units, fall within the expected range given it is centred on the agreed national average of 55%[66] and the number of children with valid speech scores at each unit. I.e. no unit has *normal speech* rates below the lower 99.8% control limit or above the upper 99.8% control limit, which is positive.

Nevertheless, this graphics representation of the data showing high levels of *normal speech* achieved at GOSH (71.4%) and lower levels of *normal speech* achieved in the Trent region (41%) – should be interpreted with caution. This is because some regions/units have significantly more missing data[67] (p<0.01) than the overall average of 8.5% across all units (as detailed in Table 13 and Appendix 6). Areas with significantly more missing data included the GOSH and Bristol units, and the West Midlands region (missing 29%, 21% and 19.2% respectively). Meanwhile 10 units including the Trent region had little (less that 5%) or no missing data (Swansea accounted for 100% of their auditable cases – see Table 13 & Appendix 6 for detail).

Figure 3. Funnel plot of five-year olds (born between 2007 and 2008) with scores suggesting normal speech, according to the number of children at each region / unit with CAPS-A scores.



Note: This funnel plot is centred on the national average identified as the national outcome mean of 2004-2006 speech outcome data[68] of 55%.

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[66] Resulting from statistical analysis on 2004-06 Speech Outcome data completed by the Lead Speech and Language Therapy Group, with statistical support from the Cleft Collective in Manchester, and presented in April 2014 to the Leads group.

[67] Children with no record of CAPS-A scores data or legitimate exclusions data – as detailed in Table 13.

[68] Resulting from statistical analysis on 2004-06 Speech Outcome data completed by the Lead Speech and Language Therapy Group, with statistical support from the Cleft Collective in Manchester, and presented in April 2014 to the Leads group.

Despite the considerations around missing data described earlier, presenting the data in Figure 3 as a funnel plot centred on the national average of 55%[69] is the most conservative method of checking (at this time[70]) whether or not any units deviated significantly from the expected standard (of 55%) and they are not ($p<0.01$). Additional checks of the speech data submitted by cleft type (CP, UCLP & BCLP) and by regions allowed us to positively confirm the following:

a) The distribution of missing cases by cleft type for all units is on average similar. I.e. No individual units have reported on, for example, more children with CP who are missing data (on average) than other units. Also, no statistically significant differences were found between proportions of missing or valid speech data[71] reported by units for each cleft type audited for speech ($p=0.3$).

b) The distribution of children achieving *normal speech* by cleft type for all units is on average similar. I.e. No individual units have reported on, for example, more children with CP who have normal speech scores (on average) than other units. Also, no statistically significant differences were found between proportions of children with *normal speech* reported by units for each cleft type audited for speech ($p=0.8$).

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[69] Resulting from statistical analysis on 2004-06 Speech Outcome data completed by the Lead Speech and Language Therapy Group, with statistical support from the Cleft Collective in Manchester, and presented in April 2014 to the Leads group.

[70] No consensus has been reached on the factors that should be incorporated into an adjustment (or risk adjustment) of this data.

[71] Specifically either CAPS-A scores or legitimate exclusions –as detailed in Table 13.

4. National Pupil Database

In this section, we describe the results of linking CRANE records to the National Pupil Database (NPD) at the individual level. The linkage was initially performed in 2014 and has been repeated recently. We describe improved linkage rates and we present data on special educational needs and the educational achievement of children with a cleft at five years and seven years of age.

4.1. Introduction

Up until now, little has been understood about the impact of facial clefting on non-health outcomes such as educational achievement. This has been identified as a priority for cleft research from patient, carer and clinical perspectives[72].

The NPD, held by the Department for Education (DfE), contains a wide range of information about pupils who attend schools and colleges in England[73]. It is considered to be one of the richest educational datasets in the world. The database includes detailed information about pupils' test and exam results, and prior attainment and progression at each key stage (see below). It also includes information about the characteristics of pupils, such as gender, ethnicity, first language, eligibility for free school meals, special educational needs and absences and exclusions.

As described in the Progress Report 2014[74], CRANE received permission from the DfE to link the CRANE Database to the NPD at the individual pupil level. The first linkage exercise was performed in 2014. The linkage rates appeared to be correlated with the quality of the postcode captured by CRANE. In 2015 we were able to update postcode records in CRANE before repeating the linkage exercise.

Last year, we described the educational attainment of CRANE-registered children at five years of age. This was based on preliminary analyses of the Early Years Foundation Stage Profile (EYFSP) assessment. This year, we have extended our analyses of the EYFSP and we have also performed some preliminary analyses on the results of the Key Stage 1 (KS1) assessment at seven years of age. Both of these assessments are described below.

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[72] http://www.lindalliance.org/CleftPSP.asp // http://www.craniofacialsociety.org.uk/Launch_CleftPSP.PDF

[73] https://www.gov.uk/national-pupil-database-apply-for-a-data-extract

[74] CRANE Project team on behalf of the Cleft Development Group. CRANE Database Progress Report 2014. London: Clinical Effectiveness Unit, The Royal College of Surgeons of England, 2014.

4.1.1. Early Years Foundation Stage Profile

The EYFSP is a National Curriculum teacher assessment of children's development at the end of the Early Years Foundation Stage, usually the academic year in which the child turns five. Between the academic years 2006/2007 to 2011/2012, the EYFSP consisted of 13 assessment scales, with pupils being able to achieve a maximum of nine points within each scale. The 13 scales are grouped into six areas of learning (see Table 16).

Table 16. The Early Years Foundational Stage profile (EYFSP) Area of learning and assessment scales.

Area of Learning	Assessment scale
Personal, social and emotional development	Dispositions and attitudes Social development Emotional development
Communication, language and literacy	Language for communication and thinking Linking sounds and letters Reading Writing
Mathematical development	Numbers as labels for counting Calculating Shape, space and measures
Knowledge and understanding of the world	Knowledge and understanding of the world
Physical development	Physical development
Creative development	Creative development

4.1.2. Key Stage 1

The KS1 teacher assessment measures pupils' attainment against the levels set by the National Curriculum. The assessments are undertaken by schools at the end of Year 2, when most pupils are seven years old. Five main subjects are assessed:

- Reading
- Writing
- Speaking and listening
- Maths
- Science

The majority of pupils are expected to reach level 2 at the end of KS1.

Further information about the NPD and the national assessments is available from the Administrative Data Liaison Service[75].

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[75] http://www.adls.ac.uk/department-for-education/dcsf-npd/?detail

4.2. Results

4.2.1. Linkage

Details of 7,152 eligible consented CRANE-registered patients born between 1 January 2000 and 31 December 2008 were provided to the DfE. Fifty-six percent were linked to an NPD record during the first linkage exercise in 2014. The second linkage, performed in 2015 with updated postcode information, resulted in the linkage of 6,194 (87%) CRANE records. The proportion of CRANE records successfully linked to the NPD varied across cleft centres, ranging from 72% in Liverpool to 94% in Newcastle.

4.2.2. Early Years Foundation Stage Profile at five years of age

Exclusions

Of the 6,194 CRANE-registered children matched to NPD records, 2,254 we excluded because they did not have an EYFSP assessment between 2006/07 and 2011/12. A further 1,098 children were excluded because they had additional anomalies or syndromes identified in HES (n=817) (see Appendix 5) or it was not possible to determine their syndrome status (n=211). Of the remaining 2,842 children, 40 were also excluded because either their gender (n=2) or their cleft type (n=38) was not known. A total of 2,802 non-syndromic children with a cleft were included in the analyses of EYFSP data.

Special educational needs

Out of the 2,802 non-syndromic children with a cleft and an EYFSP assessment, the SEN status was documented for 2,769 (98.8%). Of these, 815 (29.4%) were identified in the NPD as having special educational needs (SEN) at five years of age. This compares to 9.7% of children in the general population classified as having SEN at the EYFSP assessment across the same time frame.

Table 17 shows that the proportion of children with SEN varied according to cleft type (p<0.001). Children with a CL were least likely to have SEN while almost half of those with a BCLP were identified as having SEN.

Of the children with SEN documented in the NPD, 565 (66.6%) had the type of SEN specified and 105 (18.6%) of these had more than one type of SEN recorded. Speech, language and communication needs were the most common type of SEN, which was identified for almost three quarters of those with a SEN type specified (see Table 18).

Table 17. The number and percentage of non-syndromic children according to cleft type and special educational needs status at age five years (assessment years 2007-2012).

Cleft type	No SEN		SEN		Total
	N	(%)	N	(%)	N
Cleft lip	658	(86.8)	100	(13.2)	758
Cleft palate	642	(66.9)	318	(33.1)	960
Unilateral cleft lip and plate	504	(65.9)	261	(34.1)	765
Bilateral cleft lip and palate	150	(52.5)	136	(47.6)	286
Total	1954	(70.6)	815	(29.4)	2769
General population		**(90.3)**		**(9.7)**	

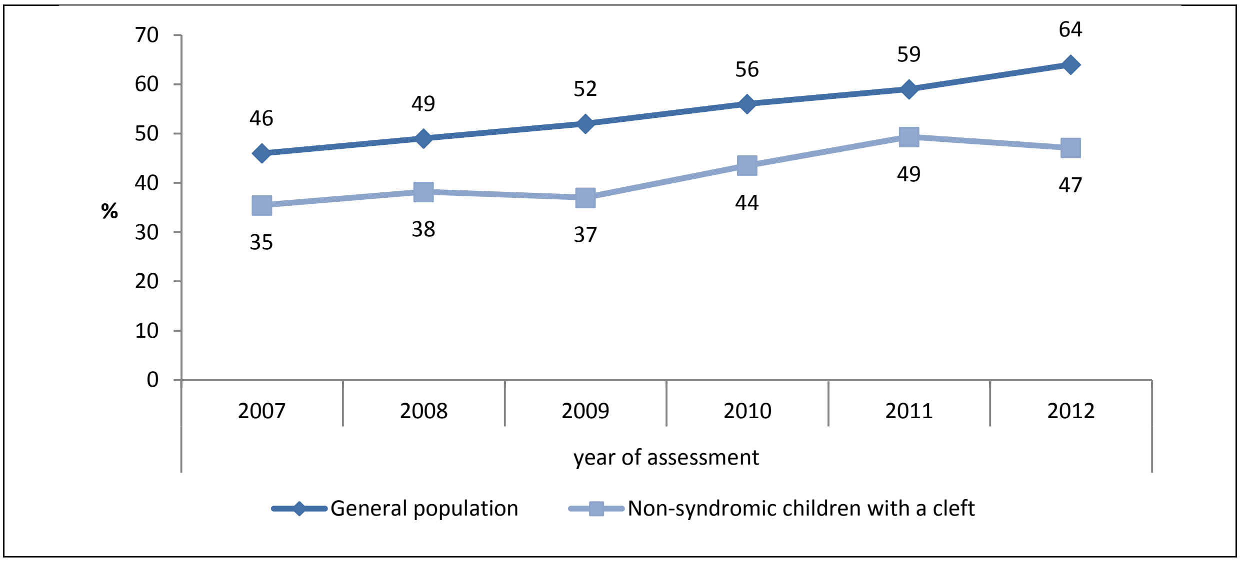
Table 18. The number and percentage[76] of non-syndromic children according to the type of special educational needs documented in the NPD at five years of age (assessment years 2007-2012).

SEN type	N	(%)
Behaviour, Emotional & Social Difficulties	52	(9.2)
Hearing Impairment	43	(7.6)
Learning Difficulties	77	(13.6)
Speech, Language and Communication Needs	416	(73.6)
Other	81	(14.3)

National marker of achievement

Figure 4 shows a clear difference between non-syndromic children with a cleft and the general population in terms of the proportion being classified as having a *good level of development* in each assessment year. In both cohorts, this proportion increased over time. Overall, 42.2% of non-syndromic children with a cleft were classified as having a *good level of development* at five years of age. This compares to 54.3% of children in the general population. The mean difference or attainment gap between the two groups across all six years was 12.1%.

Figure 4. The percentage of non-syndromic children achieving a *good level of development* at five years of age, by year of assessment and compared to the general population.



When combining all six assessment years (2006/07 to 2011/12) together, a significant difference (p<0.001) was observed between cleft types in terms of the proportion of children achieving a *good level of development*. Almost a half (47.7%) of those with a CL, 42% of those with a CP or UCLP and 27.7% of those with a BCLP achieved this level of development.

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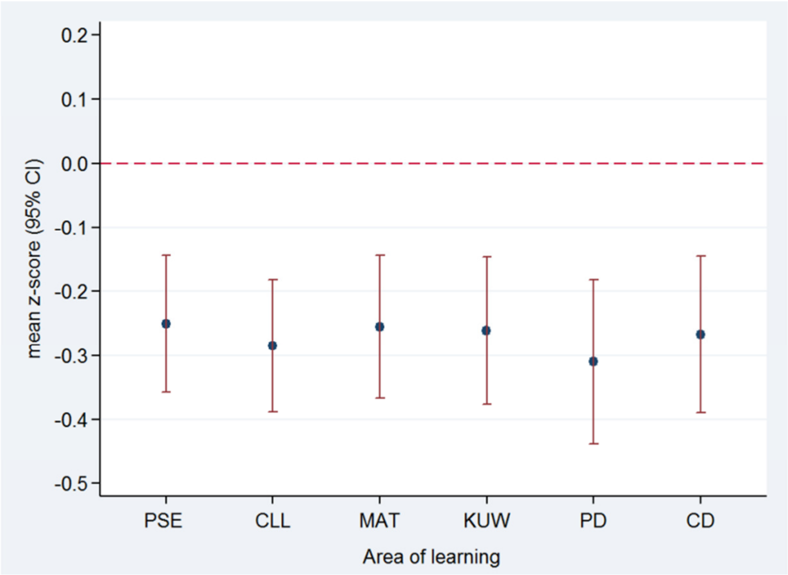
[76] Percentage reflects the number of children out of the 565 with a type of SEN specified in the NPD at the time of the EYFSP assessment. Some children had more than one type of SEN specified, so the percentages total more than 100.

Z-scores

Figure 5 shows the mean z-score and 95% confidence interval for each of the six areas of learning for all non-syndromic children with a cleft combined. The red hyphenated line on the graph represents the national population mean (a z-score of 0). The graph shows that children with a cleft have scores that are significantly below the national average across all six areas of learning. The greatest differences appear to be within the physical development (PD) area, closely followed by the communication, language and literacy area.

Figure 6 shows the mean z-score and 95% confidence interval by cleft type for each area of learning. Z-scores for children with a CL were not significantly different to the national mean within the areas of mathematical development (MAT), knowledge and understanding of the world (KUW) and creative development (CD). On average, children with a cleft affecting the palate (CP, UCLP and BCLP) have scores that are significantly lower than the national population mean within each area of learning assessed at five years of age.

Figure 5. Mean z-scores and 95% confidence intervals for each area of learning[77] in the EYFSP at five years of age among non-syndromic children with a cleft compared to the national population mean (assessment years 2007-2012).



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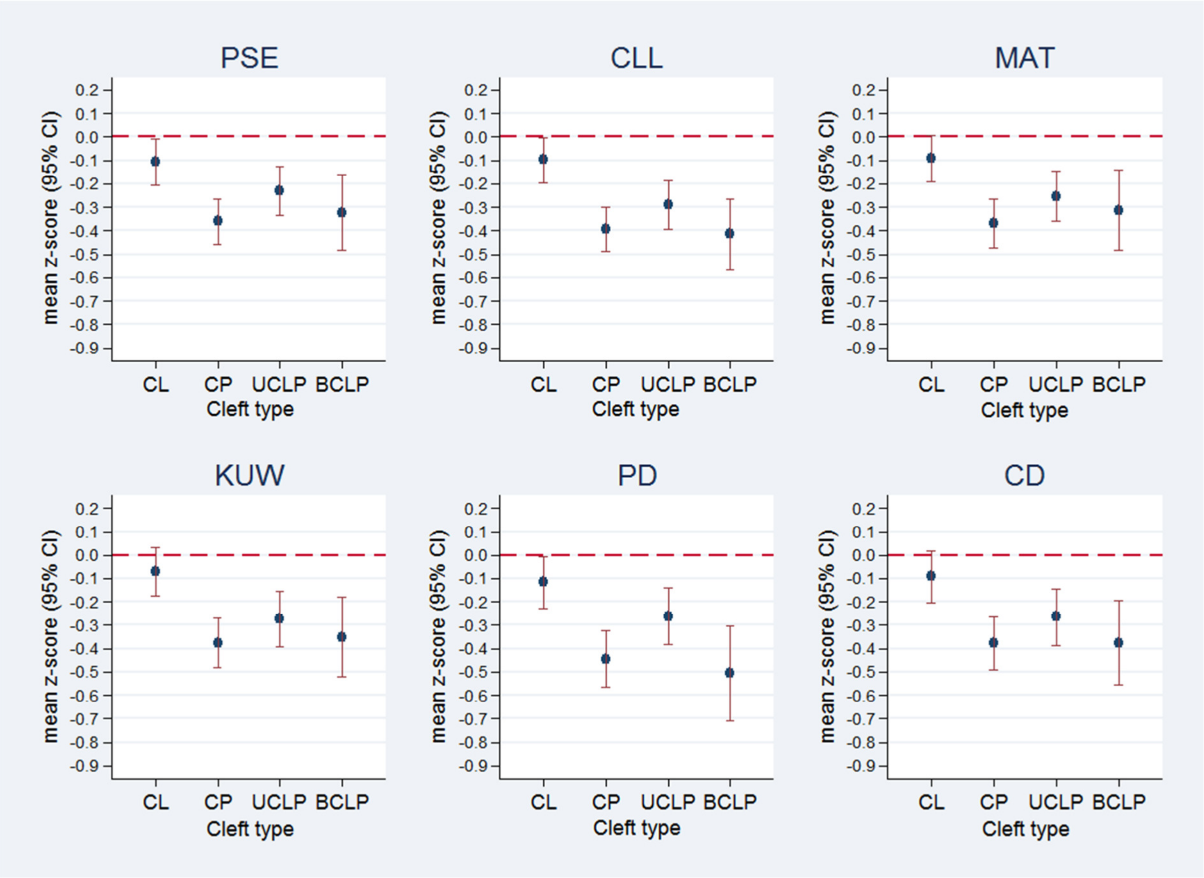
[77] PSE, Personal, social and emotional development; CLL, Communication, language and literacy; MAT, Mathematical development; KUW, Knowledge and understanding of the world; PD, Physical development; CD, creative development

Figure 6. Mean z-scores and 95% confidence intervals according to cleft type for each area of learning in the EYFSP at five years of age among non-syndromic children with a cleft compared to the national population mean (assessment years 2007-2012).



4.2.3. Key Stage 1 at seven years of age

Exclusions

Of the 6,194 CRANE-registered children matched to NPD records, 1,264 were excluded because they did not have a KS1 assessment between 2006/07 and 2013/14. A further 1,401 children were excluded because they had additional anomalies or syndromes identified in HES (n=1,059) (see Appendix 5) or it was not possible to determine their syndrome status (n=342). Of the remaining 3,529 children, 63 were also excluded because either their gender (n=2) or their cleft type (n=61) was not known. A total of 3,466 non-syndromic children with a cleft were included in the analyses of KS1 data.

Special educational needs

Out of the 3,466 non-syndromic children with a cleft and a KS1 assessment, the SEN status was documented for 3,458 (99.8%). Of these, 1,397 (40.4%) were identified as having SEN at seven years of age. This compares to approximately 21% in the general population who were classified as having SEN at the KS1 assessment across the same time frame. The proportion of children with SEN varied significantly from 26.6% of those with a CL to 57.3% of those with a BCLP (p<0.001).

Of the children with SEN identified in the NPD, 885 (63.4%) had the type of SEN documented and 235 (26.6%) of these had more than one type of SEN specified. Speech, language and communication needs were documented for 621 children (70.2% of those with a type of SEN specified) and were the most common type of SEN among non-syndromic seven year olds with a cleft. The second most common type were learning difficulties, which were documented for almost one quarter of those with a type of SEN specified (see Table 19).

Table 19. The number and percentage[78] of non-syndromic children according to the type of special educational needs documented in the NPD at seven years of age (assessment years 2007-2014).

SEN type	N	(%)
Behaviour, Emotional & Social Difficulties	106	(12.0)
Hearing Impairment	88	(9.9)
Learning Difficulties	213	(24.1)
Speech, Language and Communication Needs	621	(70.2)
Other	135	(15.3)

Achieving Level 2 or above

Across all five subject areas that are assessed at Key Stage 1, a lower proportion of non-syndromic children with a cleft achieved Level 2 or above compared to the general population. The greatest difference was in the area of speaking and listening, with an attainment gap of 9.1%, while the smallest difference was in the area of maths, with an attainment gap of 6.5%. The average attainment gap between the cleft cohort and the general population across the five subject areas was 7.5%.

Table 20. Percentage of non-syndromic children achieving Level 2 or above in each of the five subject areas of the Key Stage 1 assessment at seven years of age, by cleft type and compared to the general population (assessment years 2007-2014).

Cleft type	N	Reading	Writing	Speaking & listening	Maths	Science
CL	909	83.7	79.2	83.8	88.7	86.8
CP	1,230	77.3	70.7	76.5	80.7	80.0
UCLP	957	78.6	74.6	77.6	83.8	83.2
BCLP	370	74.6	69.7	74.1	82.4	79.2
All children with a cleft	3,466	79.1	73.9	78.5	83.8	82.6
General population		**86.0**	**82.1**	**87.6**	**90.3**	**89.4**

Within each subject, a significant difference existed between cleft types in terms of the proportion of children achieving Level 2 or above ($p<0.001$). Children with a cleft lip were most likely to achieve the expected level of 2 or above, while children with a BCLP appear to be most affected academically (see Table 20).

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[78] Percentage reflects the number of children out of the 885 with a type of SEN specified in the NPD at the time of the KS1 assessment. Some children had more than one type of SEN specified, so the percentages total more than 100.

4.3. Summary

We have been able to achieve a high linkage rate between CRANE and the NPD. This means that the reported educational attainment of CRANE-registered children is likely to be representative of most non-syndromic children with a cleft attending schools in England.

A large proportion of children with a cleft who are not identified in HES as having additional anomalies or syndromes are classified as having special educational needs in the NPD. This proportion increased from 29% at age five years to 40% at age seven years. These rates are much higher compared to those in the general population (10% and 21%, respectively) across the same time frames. The majority of children with the type of special educational needs documented, have speech, language and communication needs.

Our analyses revealed significant differences in educational achievement between cleft types. In general, children with a cleft affecting only the lip do better than those with a cleft affecting the palate. These differences were seen at both five and seven years of age.

We found an attainment gap of 12% between the cleft cohort and the general population when comparing the proportion of children considered to have a good level of development at age five years. An attainment gap of 7.5% was found across the five subjects when comparing the proportion of children achieving the expected Level 2 or above at the end of KS1 between those with a cleft and the general population. These preliminary results may suggest that some catch up occurs between the age of five and seven years among children with a cleft. However, differences in the scoring systems at the different assessments mean it is not appropriate to draw comparisons based on the magnitude of the attainment gaps.

We have demonstrated the feasibility of linking CRANE with the NPD, and we will continue our work with NPD and CRANE-HES-linked data.

5. Development of CRANE Database and future directions

5.1. Re-development of the CRANE Database and website

In early 2015 the CRANE project team identified the need to re-develop the CRANE Database and Website IT systems. This was because the current systems are out-dated (they are 10-years-old) and because of reduced access to IT support from the RCS team currently maintaining CRANE (due to a number of unavoidable circumstances). These issues were identified as putting CRANE at risk of (1) compromised access to the CRANE Database, (2) disrupted maintenance, and (3) not being able to amend or expand data collection (such as additional outcome measures).

On the other hand, this challenge presented an opportunity to ensure the new database and website are more user friendly for cleft teams and patients (and parents/carers), as well as the CRANE Project team as administrators of the systems.

To address this, the CRANE project team compiled a specification to inform re-development and expansion of the capabilities of the CRANE database and website. Following this, System Developers were invited to review this specification and provide cost estimates for this work. This resulted in Crown Informatics Limited being appointed to work in collaboration with CRANE to complete this work.

Following the appointment of a system developer, the CRANE project team held a 'Consultation on Database & Website changes' meeting on the 9 June 2015. Representatives of all cleft services and CLAPA were invited to this consultation meeting to provide their input to identify useful changes / features for the re-development of the CRANE Database and the CRANE Website. Their feedback included the following suggestions:

1. CRANE should look into expanding data collection beyond its current emphasis on five year old outcomes – to collect data at pre-school age, and for older children – into adulthood.
2. The CRANE Database system should help make it easier for cleft teams to enter data required by allowing them to identify the gaps in their data.
3. In the long term, CRANE should seek to reduce duplication of effort for teams entering data into multiple systems – including hospital systems, CRANE and other local and national audits.
4. CRANE should ensure reports are more useful for patients and parents/carers – while considering the wider picture, and the fact that CRANE may possibly not be able to be 'all things to all people'.

The detailed feedback provided at the consultation meeting went on to inform a 'functional requirements document' and 'detailed requirements specification' – key to finalizing the contract with Crown Informatics Limited in August 2015.

The new CRANE website and database will be under development through the autumn of 2015, with user acceptance testing followed by the launch of the new system expected to take place in the winter of 2015/16.

5.2. Consent form and patient information leaflet

In 2014 we published translated versions of the CRANE Information Leaflet and Parental Consent Form for speakers of Welsh, Polish and Urdu, as well as producing an audio version of the English language information leaflet (found at http://www.cranedatabase.org.uk/info_cleftteams/). This was to acknowledge the diversity of languages spoken by patients and family accessing cleft services, and expand opportunities for access to information about CRANE.

Anonymised feedback on these new versions, provided by cleft teams in 2015, was summarised for the Cleft Development Group (CDG). It suggested that translations are used infrequently, but where they are used they are found to be useful. Cleft teams also recommended languages for future translations, suggesting there may be value in producing additional translations in Portuguese, Czech, Farsi and Russian in future. Our aim is to undertake up to three translations each year to spread the overall costs until we have covered the more common languages encountered by teams.

We will consider the feedback and input provided by cleft teams in 2016 when producing updated versions of the CRANE Information Leaflet and Parental Consent Form, as well as a new ‘Information Leaflet on Data Linkage’, to coincide with the launch of the re-developed CRANE Website and Database in 2016. We are also considering methods to inform older children and young adults about their data stored on the CRANE database.

5.3. Outcome measures

Currently the outcome section of the Database is hampered by the lack of agreed measures which have been shown to be valid and reliable in assessing the outcome of cleft care. Therefore, the future plans outlined below are being pursued.

5.3.1. Newborn screening

The CRANE Dataset was expanded in May 2014 on request to allow recording of ‘timing of diagnosis’ within 72 hours to align CRANE data collection with Newborn and Infant Physical Examination (NIPE) standards [79]. Analysis including data on this timing was not conducted for 2014 births (for this Annual Report) as only

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[79] In line with a statement of the UK National Screening Committee Newborn and Infant Physical Examination (NIPE) Standards and Competencies 1 document (2008) – setting out the standard for 95% newborn to be screened by 72 hours after birth (page 13 of the document found at http://newbornphysical.screening.nhs.uk/getdata.php?id=10639).

small numbers were recorded using the '≤72 hours' timing (n=35, 3.7%)[80]. We anticipate being able to report this next year when we have a full calendar year of data.

5.3.2. Speech

Cleft Audit Protocol for Speech – Augmented (CAPS-A) scores have been used to assess speech among five year old children with a cleft affecting the palate (CP, UCLP and BCLP). This is only the second year that 16 CAPS-A speech outcome scores have been collected (see Section 3.6.5 for a description of the 16 CAPS-A speech parameters).

Further to reporting on the 16 CAPS-A speech parameters separately, we anticipate expanding our reporting of speech outcomes assessed at five years of age year on year. Specifically we aim to report on the proportion of five-year olds meeting the following three nationally agreed Speech Outcome Standards[81]:

1. The achievement of *normal speech* (speech outcome standard #1);
2. the presence of speech difficulties likely to be the result of existing or previous structural anomalies (speech outcome standard #2); and
3. the presence of cleft-related articulation difficulties (speech outcome standard #3).

5.3.3. Patient and Parent Reported Experience

The Cleft Psychology Clinical Excellence Network (CEN), upon request by the Craniofacial Society of Great Britain and Ireland (CFSGBI) Council and the Cleft Development Group (CDG), identified and piloted measurements to evaluate patient (and parent) reported experience[82]. These measures were combined into one Patient (and Parent) Reported Experience Measure (PREM) questionnaire and, through piloting by the Cleft Psychology CEN, adjusted to apply to cleft services.

The CRANE project team and the Cleft Psychology CEN are collaborating to conduct a 12-month feasibility study (with a review point at 3-months). This feasibility study team aims to test PREM data collection, analysis and reporting, with a view to developing a method to implement these nationally. This feasibility study will also inform decisions around potentially collecting PREM data via the CRANE Database.

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[80] Therefore we included '≤72 hours' cases within the '≤1 week' timing for this report and will report on '≤72 hours' referrals once this data item has come fully into use.

[81] Britton L, Albery L, Bowden M, Harding-Bell A, Phippen G, and Sell D(2014) A Cross-Sectional Cohort Study of Speech in Five-Year-Olds With Cleft Palate ± Lip to Support Development of National Audit Standards: Benchmarking Speech Standards in the United Kingdom. The Cleft Palate-Craniofacial Journal: July 2014, Vol. 51, No. 4, pp. 431-451.

[82] Including: (1) the Friends and Family Test (FFT) – developed by the Department of Health) and (2) the Experience of Service Questionnaire (CHI-ESQ) satisfaction assessment scales – developed by the Commission for Health Improvement (CHI).

5.4. Data sources and future analyses

5.4.1. National Pupil Database (NPD)

The improved linkage between NPD and CRANE resulted from improving the quality of the postcode data held by CRANE. We are continuing our work with NPD and CRANE-HES-linked data. Future analyses will involve exploring in more detail children's educational attainment at seven years of age and we will also begin to explore educational attainment at Key Stage 2, when children are 11 years of age. Tracking children's educational attainment across different assessments as they age will allow us to study whether attainment gaps persist and to what extent, or whether children with a cleft do catch up with their peers in the general population.

Future analyses will also aim to examine the correlation between educational outcomes and treatment outcomes recorded in CRANE, such as speech quality, and we are interested in exploring whether there are aspects of the cleft treatment pathway, such as timing of repair, that may explain observed differences in educational outcomes within the cleft cohort.

We are collaborating with the Institute of Education research group, who have substantial previous experience in analysing NPD data and adjusting for the multiple factors that affect educational achievement.

5.4.2. Hospital Episode Statistics (HES)

The Clinical Effectiveness Unit received a HES extract – containing hospital data updated up until 2013 – which we have linked to CRANE. This will allow us to refresh our analyses involving HES data, such as those involving the NPD, mortality, secondary speech surgery, and grommets.

5.4.3. Newborn Hearing Screening Programme (NHSP)

We are continuing to explore the option of requesting linkage between our CRANE Database and the Newborn Hearing Screening Programme (NHSP)[83] data – via Public Health England (PHE) – with the purpose of looking at the relationship between clefts and Permanent Childhood Hearing Impairment (PCHI) and the effect of PCHI on children's outcomes.

5.4.4. National Child Measurement Programme (NCMP)

We are continuing to explore the option of requesting linkage between our CRANE Database and the National Child Measurement Programme (NCMP) data[84], with the purpose of improving height and weight outcome measures at five years of age (which are still not collected and recorded routinely in CRANE – for fewer than 30% of cases by half of units at the time of this report). This improvement of the data held by

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[83] http://hearing.screening.nhs.uk/

[84] Via either the Health and Social Care Information Centre (HSCIC) or our links at Public Health England (PHE), with whom we are seeking linkage to the NHSP (see item 5.1.3).

CRANE could be substantial as the NCMP has collected measures of weight and height of over 90% of all children in reception class (aged four to five years) in England (since 2005-06).

5.4.5. Equity and treatment and outcomes

We plan to explore possible associations between socio-demographic factors (index of multiple deprivation and ethnicity) and burden of care and outcomes using linked CRANE-HES data.

5.4.6. Mapping boundaries of Regional Cleft Networks

We are undertaking an analysis to map the boundaries of all Regional Cleft Networks using CRANE -HES linked data. This will allow us to report on patient flow between cleft teams. The mapping will also be useful information for Commissioners about this group of patients whose treatment they fund. We are conducting this work in collaboration with the London School of Hygiene and Tropical Medicine (LSHTM), drawing on the experience of colleagues within the Clinical Effectiveness Unit (CEU), such as the Prostate Cancer Audit project team who are conducting similar work.

5.5. Clinical Reference Group (CRG)

The inclusion of submitting data to CRANE as a requirement to the D07/S/a National Service Specification (Cleft Lip and or Palate Services including Non-cleft Velopharyngeal Dysfunction (VPD) (All Ages)), developed by the CRG, is a welcome advance in helping to improve the quality and completeness of data held in the CRANE database. It is anticipated that the new CRANE Website and Database (and engagement with our stakeholders during this re-development) will help develop communication and links with cleft teams and help to improve data submission.

The CRANE team is committed to working with commissioners to make sure that its outputs are consistent with current and future commissioning requirements, which may in the future include areas such as performance reporting.

Quality Dashboard

The CRANE project team submitted data for the quality dashboard in February 2015 for Quarter 1 and Quarter 2 of the 2014/15 year (Q1 – April to June 2014 & Q2 – July to September 2014), and in May 2015 for Quarter 3 and Quarter 4 of the 2014/15 year (Q3 – October to December 2014 & Q4 – January to March 2015). This was done for the following five out of the six items requested – two of which were new requests (CLP01 and CLP02):

- Measure Number CLP00: The number of CRANE-registered children born within a specified quarter of the calendar year (refreshed every quarter).
- Measure Number CLP01: The number of Parents contacted by a Cleft team Clinical Nurse Specialist (CNS) within 24 hours of referral with an antenatal diagnosis of Cleft Lip and/or Palate – born within a specified quarter of the calendar year (refreshed every quarter).

- Measure Number CLP02: The number of Parents receiving visit from a Cleft team CNS within 24 hours of first referral (provided the child has not reached the age of one year) – born within a specified quarter of the calendar year (refreshed every quarter).
- Measure Number CLP06: The number of 5 year old children with a decayed, missing and filled teeth (dmft) index score, as a percentage of all 5 year old children (refreshed annually).
- Measure Number CLP09: The number of five year old children with 5 year old index scores 1 or 2 (as indicator of maxillary growth in patients with complete UCLP[85]) – as a percentage of the number of 5 year old children with a 5 year old index score (refreshed annually) [previously numbered CLP08].

The sixth item requested by Methods – the speech data – was once again provided directly by the centres. Specifically:

- Measure Number CLP07: The number of 5 year old children with green Cleft Audit Protocol for Speech – Augmented CAPS-A scores – (who have speech within normal range) as a percentage of the number of 5 year old children with a CAPS-A score (refreshed annually).

Future productions of Quality Dashboard CRANE tables have been confirmed – potentially including speech data – but future dates have yet to be agreed.

5.6. Public Health England

CRANE has been involved in work conducted by Public Health England in 2015 to develop a National Congenital Anomaly Register (CAR) to capture all congenital anomalies in England. We have expressed our intention to comply with any data submission requirements to this project.

5.7. Collaboration

CRANE is collaborating with a number of individuals and organisations:

- Since the publication of our annual report in 2012, which highlighted the problem of late diagnosis of CP, the Royal College of Paediatrics and Child Health (RCPCH) has, in collaboration with key partners including the CRANE Database team, drafted and published a best practice guide to help healthcare professionals identify cleft palate in neonates. This guide provides recommendations to ensure early detection of a cleft palate, and to improve and standardise routine postnatal examination of the palate. A parent/carer guide is also available. (http://www.rcpch.ac.uk/improving-child-health/clinical-guidelines-and-standards/published-rcpch/inspection-neonatal-palate)
- The Healing Foundation Cleft Gene Bank and Cohort Study supported by the Vocational Training Charitable Trust (VTCT) called the Cleft Collective (www.cleftcollective.org.uk) will be the world's

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[85] Atack NE, Hathorn IS, Semb G, Dowell T and Sandy JR. A new index for assessing surgical outcome in unilateral cleft lip and palate subjects aged five: reproducibility and validity. Cleft Palate Craniofac J. 1997 May;34(3):242-6.

largest cleft lip and palate research programme, which is taking place in the UK. Up to 5,000 children and their families are being recruited to the Birth Cohort Study hosted by the University of Bristol. The Centre for Appearance Research at the University of the West of England will be working on the psychological issues associated with cleft lip and palate and the support needed by families and children. We are currently working with the Cleft Collective team to establish whether CRANE could share data with this research project.

- The development of an Outcome Measure for Aesthetics at five years of age following lip repair is being considered by the Birmingham Institute of Paediatric Plastic Surgery (BIPPS). This is based on the original pilot conducted by surgeon from Oxford which has been significantly developed by surgeons from Birmingham. This audit tool allows the review and scoring of images of children's lip repairs. CRANE looks forward to becoming involved in this work at an appropriate time in the development of this measure, as it could become a valid and reliable tool which could be used nationally to audit results of surgery.

5.8. CRANE Communications

5.8.1. Dissemination of 2015 findings

- Publication of the Annual Report will be announced via our regular eNewsletter, which will be circulated in November, and it will be available on our website.

- We will also work with our close collaborators – such as the CFSGBI and CLAPA – to expand the reach of our eNewsletter (and the report).

- Once the *Summary of Findings for Patients and Parents/Carers* from last year's 2014 Annual Report has been signed off by the CDG along with this report, we will seek to publish it in collaboration with CLAPA. In 2016 we will produce a summary of this 2015 Annual Report, and aim to publish it in the summer of 2016.

5.8.2. Publications and presentations related to the CRANE Database

Publications

The following paper has been peer-reviewed and accepted for publication (date to be confirmed):

- Fitzsimons KJ, Copley LP, van der Meulen JH, Panagamuwa C, Deacon S. "Grommet Surgery in Children with Orofacial Clefts in England". The Cleft Palate-Craniofacial Journal.

Oral presentations

- Deacon S, Copley LP, Charman S, Fitzsimons KJ, Medina J, van der Meulen J. "CRANE Database: Educational outcomes of patients with a cleft in England" Craniofacial Society of Great Britain and Ireland (CFSGBI) Annual Conference (April 2015).

- Medina J, Copley LP, Deacon S, van der Meulen J. “Exploration of rates of secondary speech surgeries in patients with cleft palate involvement in England” Craniofacial Society of Great Britain and Ireland (CFSGBI) Annual Conference (April 2015).
- Deacon S, Charman S, Copley LP, Fitzsimons KJ, Medina J, van der Meulen J. “CRANE Database: Educational outcomes of patients with a cleft in England” 10th European Craniofacial Congress: ECC 2015 (June 2015).
- Deacon S, Medina J, Copley LP, Fitzsimons KJ, van der Meulen J. “CRANE Database: Our experience of data linkage in England” 10th European Craniofacial Congress: ECC 2015 (June 2015).
- Deacon S, Medina J, Copley LP, Fitzsimons S, van der Meulen J. “A Summary Report of Five Year Outcome Data from the CRANE Database”. Prepared for Norcleft Quadcentre Audit Day (June 2015).
- Deacon S, Medina J, Copley LP, Fitzsimons KJ, van der Meulen J. “A Summary Report of Five Year Outcome Data from the CRANE Database”. Prepared for the West Midlands, South West and South Wales Centres-Tri Centre Audit Day (July 2015).
- Deacon S, Medina J, Copley LP, Fitzsimons KJ, van der Meulen J. “A Summary Report of Five Year Outcome Data from the CRANE Database”. Prepared for the North Thames, South Thames and Cleft Net East Audit Day (November 2015)

Poster Presentations

- Fitzsimons KJ, Copley LP, Charman S, Medina J, Deacon S, van der Meulen J. “CRANE Database: Examination of the educational achievement of a national cohort of children with a cleft lip and/or cleft palate” Closer (Cohort & Longitudinal Studies Enhancement Resources) Conference – The importance of early years, childhood and adolescence: Evidence from longitudinal studies. (November 2014).

6. Conclusions

This Annual Report presents national-level data on children born with a cleft lip and/or palate in England, Wales and Northern Ireland.

A total of 1,014 children born with a cleft in 2014 had been registered on CRANE at the time of preparing this report. This equates to an incidence of approximately one in every 713 live births in England, Wales and Northern Ireland[86].

Although children can now be registered with CRANE prior to obtaining parental consent, consent must still be obtained so that complete data, including outcomes, can be collected and reported by CRANE. The consent rate is very high among patients who have been through the consent process, which is encouraging. However, almost one fifth of the children born in 2014 had not been consented at the time of preparing this report. Further, this proportion ranged from 0% to 81% between units. Units with a high proportion of unconsented patients are encouraged to review their consent-taking process, with the aim of obtaining consent in a timely fashion to enable the reporting of complete data.

The majority of units collect all the data items requested by CRANE. However, the reporting of some data, in particular outcomes at five years of age, is variable between units. Some units have provided outcome data for more than 85% of their eligible patients, suggesting that the reporting of outcomes is feasible. CRANE will continue to explore ways to improve communication and links with units to improve the submission of data in the future. The re-development of the CRANE Database and Website in 2015/16 is expected to play a key role in facilitating this (see Chapter 6 for further detail on this).

Collecting and reporting outcomes among children with a cleft is important for evaluating treatment, drawing comparisons between different groups of patients, providing information to patients and parents, and for planning future services. The inclusion of submitting data to CRANE as a requirement in the National Service Specification for cleft lip and/or palate services should improve the quality and completeness of data held in the CRANE database.

Based on the data reported to CRANE, we have highlighted some areas that should be addressed by maternity, paediatric, cleft and dental services to improve care and outcomes.

Diagnosis, Referral and Contact

1. Antenatal diagnosis rates of cleft lip, with or without cleft palate, are still falling below the NHS Fetal Anomaly Screening Programme target detection rate of 75%[87].

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[86] 24,394 Births in NI in 2014 – Available from Northern Ireland Statistics & Research Agency (NISRA): http://www.nisra.gov.uk/demography/default.asp8.htm. // 698,512 Births in England & Wales in 2013 (2014 release has been cancelled so figure for 2013 used as unlikely to be drastically different) – Available from the Office for National Statistics. Characteristics of Birth 1, England and Wales: http://www.ons.gov.uk/ons/rel/vsob1/characteristics-of-birth-1--england-and-wales/index.html

2. Just under a third of children with a cleft palate alone (31.3%) are being diagnosed late according to the national standard, which states that clefts should be diagnosed within 24 hours of birth to enable immediate referral to a specialist hospital[88]. This proportion has increased since last year (30.4%).

3. 81.4% of the children born in 2014 with a cleft were referred by a maternity unit to an Cleft Unit within 24 hours of birth. This proportion varied according to the unit receiving the referrals (ranging from 70.4% to 88.5%). These figures show approximately the same level of improvement as noted at this time last year (for 2013 births – versus 2012's rate of 52.6%). Although rates have improved over the last two years, prompt referral is still recommended to ensure that the baby and their family receive appropriate care and support as soon as possible.

4. Units established contact with 90.8% of families within 24 hours of their child's referral.

Cleft-related outcomes at five years

5. Children with a cleft are at increased risk of poor oral health. Children with a cleft affecting both the lip and palate are at the greatest risk of caries and may benefit from targeted preventive intervention. Nevertheless, average treatment and care indices (of 74.9% and 67.5% respectively) across units indicate that, in the majority of cases, units have mechanisms in place to deal with any disease occurring.

6. Approximately one quarter of children with a complete UCLP have poor dental arch relationships (25.1%) that may benefit from further surgery to correct facial disproportion. While there is room for improvement, this proportion is substantially lower than the 36% of five year old children with a cleft who were reported by CSAG to have poor dental arch relationships in 1996[89].

7. More than two fifths of children (41.7%) with a complete speech assessment had speech scores that would suggest their speech is significantly different from their non-cleft peer group, with scores suggesting they have not achieved *normal speech*.

Educational achievement at five and seven years

We were able to successfully link 87% of eligible CRANE-registered children born between 2000 and 2008 to NPD records. Using the NPD and CRANE-HES-linked dataset, we examined special educational needs and educational achievement among non-syndromic children with a cleft at five and seven years of age.

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[87] Donna Kirwan and NHS Fetal Anomaly Screening Programme in collaboration with the Royal College of Obstetricians and Gynaecologists (RCOG), British Maternal and Fetal Medicine Society (BMFMS) and the Society and College of Radiographers (SCoR), NHS Fetal Anomaly Screening Programme. 18+0 to 20+6 Weeks Fetal Anomaly Scan National Standards and Guidance for England, 2010, NHS Fetal Anomaly Screening Programme: Exeter.

[88] Bannister P. Management of infants born with a cleft lip and palate. Part 1. Infant, 2008. 4(1): p. 5-8.

[89] Clinical Standards Advisory Group, Clinical Standards Advisory Group. Report of a CSAG Committee on cleft lip and/or palate, 1998, The Stationery Office, London.

The key findings are outlined below:

8. A large proportion of children with a cleft who were not identified in HES as having additional anomalies or syndromes were classified as having special educational needs in the NPD. This proportion increased from 29% at age five years to 40% at age seven years. These rates are much higher compared to those in the general population (10% and 21%, respectively) across the same time frames. The majority of children with the type of special educational needs documented, have speech, language and communication needs.
9. Non-syndromic children with a cleft have standardised EYFSP scores that are significantly below the general population average within each of the six areas of learning assessed at five years of age. The greatest differences appear to be within the ‘physical development area’, closely followed by the ‘communication, language and literacy’ area.
10. Across all five KS1 subject areas assessed at seven years of age, a lower proportion of non-syndromic children with a cleft achieved the expected Level 2 or above compared to the general population. The greatest difference was in ‘speaking and listening’, where there was an attainment gap of 9.1%.
11. Our analyses revealed significant differences in the educational achievement between cleft types. In general, children with a cleft affecting only the lip do better than those with a cleft affecting the palate. These differences were seen at both five and seven years of age.

Appendices

Appendix 1: CRANE Project Team

Members of CRANE project team

Scott Deacon	Clinical Project Lead / Lead Consultant Orthodontist	Clinical Effectiveness Unit / South West Cleft Unit, University Hospital Bristol NHS Foundation Trust University of Bristol
Jibby Medina	Research Fellow	Clinical Effectiveness Unit
Kate Fitzsimons	Research Fellow	Clinical Effectiveness Unit
Lynn Copley	Data Manager	Clinical Effectiveness Unit
Jan van der Meulen	Clinical Epidemiologist	Clinical Effectiveness Unit / London School of Hygiene and Tropical Medicine
Jackie Horrocks	CRANE Administrator	Clinical Effectiveness Unit

Appendix 2: Governance and funding

Ownership

It has been agreed that the “ownership” of the CRANE Database lies with the Craniofacial Society of Great Britain and Ireland (CFSGBI) as it represents the multidisciplinary group of professionals involved in the care of patients with a cleft lip and/or palate.

Cleft Development Group

The Cleft Development Group is a body with two distinct roles. Firstly, it is responsible for making arrangements for the running and commissioning of the CRANE Database.

Secondly, it is responsible for providing guidance on all aspects of the delivery of cleft care in England and Wales. It includes representatives from all the stakeholders in cleft care in England and Wales, including commissioners, public health consultants/regional cleft leads, specialists in the provision of cleft care, and parents and patients. It also has representatives from the health services in Wales, Scotland and Northern Ireland, as well as a representative from the Republic of Ireland cleft service.

Funding

Funding of the CRANE Database is currently coordinated and agreed by representatives of the national Specialised Commissioning Group for England and the Wales Specialised Health Services Committee. This year Northern Ireland agreed to formally fund a contribution to the project for the first time. Funds are raised through a levy calculated on a weighted per capita basis from the commissioning bodies in England, Wales and Northern Ireland. The levy is currently collected by Specialised Commissioning (East Midlands).

Appendix 3: Members of the Cleft Development Group

Members of the Cleft Development Group (CDG)

Stephen Robinson	Chair / Clinical Director Spires Cleft Service
Victoria Beale	Deputy for Simon van Eeden
Geoffrey Carroll	Medical Director, Welsh Health Specialised Services
Alex Cash	Clinical Lead, South Thames Cleft Service
Michele Davis	Regional Programme of Care Manager London / University Hospitals Birmingham NHS Foundation Trust
Scott Deacon	CRANE Clinical Project Leader
Mark Devlin	Clinical Lead for the Cleft Surgery Programme Board, Scotland
David Drake	Cleft Surgery Training Interface Group
Toby Gillgrass	Lead Clinician of Cleft Care Scotland
Per Hall	Cleft Surgeon (BAPRAS)
Norman Hay	Clinical Lead, North Thames Cleft Service
Chris Hill	Northern Ireland Clinicians
Peter Hodgkinson	Clinical Lead, Newcastle Site, Northern and Yorkshire Cleft Service & Chair Cleft Centres
Jackie Horrocks	Minute Secretary, Clinical Effectiveness Unit
Nichola Hudson	Lead Clinical Nurse Specialist
Thayalan Kandiah	Paediatric Dentistry SIG
Natalie Kirk	Network Business Manager for Cleft Net East
David Landes	Public Health Consultant
Kate le Marechal	Clinical Psychologists / Clinical Excellence Network (CEN) CFSGBI
Jason Neil-Dwyer	Clinical Director, Trent Cleft Service
David Orr	Cleft Services in the Republic of Ireland
Norma Patterson	CDs and Managers Group
Marie Pinkstone	Lead Speech & Language Therapists
Rosanna Preston	CLAPA Chief Executive
Jonathan Sandy	Lead, Cleft Collective Birth Cohort and Gene Bank Study
William Shaw	Manchester Lead, Cleft Collective Birth Cohort and Gene Bank Study
Rona Slator	Clinical Lead, West Midlands Cleft Centre
Jackie Smallridge	Consultant Paediatric Dentist
Alistair Smyth	Cleft Surgeon (BAOMS)
David Steel	Chair Programme Director, National Services Division, NHS Scotland,
Adrian Sugar	CRG Chair/ Wales Clinicians
Jan van der Meulen	Senior Epidemiologist, Clinical Effectiveness Unit

Simon van Eeden	Clinical Lead, North West, IoM & North Wales Cleft Network
Mike Winter	Medical Director, National Services Division,, Scotland
Ken Wragg	Dental Public Health Consultant

Appendix 4: Terms of Reference for the Cleft Development Group

The Origins of the Cleft Development Group (CDG)

The NHS Cleft Development Group was formed in November 2004 out of the previous CRANE/Cleft Levy Board, the CRANE Management Group and their Advisory bodies. These groups and bodies had been responsible for the national cleft database, CARE and then CRANE. The implementation of the DoH's guidance regarding the re-organisation of cleft services in the UK which stemmed from the DoH Clinical Standards Advisory Group report into the care of patients with Clefts of the Lip and/or Palate (1998) was the responsibility of the Cleft Implementation Group (CIG). When this group was terminated by the DoH, a new body took over its role, the Cleft Monitoring Group. When that body was terminated, the Cleft Development Group (CDG) was asked to take over its role too.

The Roles of the CDG

The CDG has two distinct roles which arise from its origins.

1. The CDG is responsible for guidance on all aspects of the delivery of re-organised cleft care in England and Wales and, when asked, by Scotland and Northern Ireland. It gives advice to the cleft centres, to health authorities, trusts, boards, commissioning groups and consortia and to the Departments of Health in England and the devolved administrations. It represents all stakeholders in cleft care and works with all to ensure the highest quality of cleft care in the UK to all patients who need it. It inherits the responsibilities of the Cleft Implementation Group and the Cleft Monitoring Group which were largely advisory.

2. The CDG is responsible for the commissioning of, the strategic governance of and is ultimately responsible for the national cleft database which used to be called CARE and is now called CRANE. It must negotiate and agree a contract for the running of CRANE and have operational oversight of the implementation of that contract. It is responsible for funding of the CRANE Register and is responsible for ensuring that the agreed levy is collected annually through the NHS Specialist Commissioners. It will approve an annual budget and business plan for CRANE drawn up with the contract holders and will review income and expenditure and ensure that the terms of reference are implemented. It will determine the location of the register and will appoint the Clinical Director/Project Leader who will be accountable to the Group.

The CDG's responsibility stems from Health Services Circular 1998/238 which states that "A CARE Register, with which all patients should be registered, will be maintained by the Craniofacial Society of Great Britain – this will form the basis for national audit".

The database was UK wide when run by the Craniofacial Society of Great Britain and Ireland and before it became the responsibility of the CRANE Levy Board. Devolution of government in the UK resulted in four distinct health services and as a result CDG came to be responsible for a national database for the recording of all children with clefts of the lip and/or palate born and treated in England and Wales, as the health service in Wales indicated its support for this development at an early stage. It has since then successfully sought to include in its work strong relationships also with the cleft services in Scotland, Northern Ireland and the Irish Republic.

The CDG is responsible for providing data for cleft births and cleft treatment for England and Wales and it also endeavours, with the cooperation of the health services in Scotland and Northern Ireland, to do so for the whole of the UK.

The national CRANE database has two primary functions:-

a. the recording of all birth, demographic and epidemiological data related to children born in England and Wales with the congenital abnormality of clefting of the lip and/or palate, and where possible extending this to the whole of the UK and Ireland

b. the recording of all treatment of children and adults in England and Wales with clefts of the lip and/or palate and the outcome of such treatment, and where possible extending this to the whole of the UK and Ireland

The data from (a) will provide the same kind of information as other congenital anomaly registers and will be the basis for reports, audit and research in that area. The data from (b) will provide the basis for national cleft audit which is intended to be a major and integral role of CRANE.

The relationships between the bodies involved in the national cleft database, CRANE, are defined by a Tripartite Agreement (2007) between the Cleft Development Group, the NHS Specialist Commissioners and the Craniofacial Society of Great Britain and Ireland.

Composition of the Cleft Development Group

The composition of the Cleft Development Group should reflect all stakeholders involved in cleft care. Consequently its composition (and consequently these Terms of Reference) will need to be changed from time to time. The Members of the Cleft Development Group will normally and primarily be active clinical members of a designated Cleft Team, public health consultants, commissioners of cleft care and representatives of parent/patient organisations. Membership of the Group will be for a term of three years which can be extended at the behest of the nominating organisation, except for members ex-officio who will be members during their terms of that office whether it be less or more than 3 years. The Group will elect its own Chair, who will remain in office for 3 years. The Group will also elect a Vice Chair. Either the Chair or the Vice Chair should be a Specialist Commissioner. The Group may decide to re-elect the holders of these offices.

The composition will be:

1. Commissioners of Cleft Care. These should include at least two commissioners from Specialist Commissioning Groups in England (nominated by the National Specialist Commissioning Group for England), one from Wales, one from Scotland and one from Northern Ireland (each nominated by their equivalent national specialist commissioning body). It is intended that there should be no more than six specialist commissioners in total to be agreed and appointed by the bodies which

contribute data to the database (in the case of Scotland by sharing its data with CDG). Only those commissioning groups which pay the levy may vote on issues relating to CRANE.

2. Public Health Consultants. These should include representatives of commissioning areas who are actively involved in cleft commissioning, and will normally be Consultants in Dental Public Health. There should be at least two (to be nominated by the BASCD Consultants in Dental Public Health Group).
3. A Lay representative from a Parent Support Group (1) (to be nominated by CLAPA)
4. Cleft surgeons (2) (presently one nominated by BAOMS and one by BAPRAS)
5. The President of the Craniofacial Society of Great Britain and Ireland
6. The Chair of the Cleft Surgery Training Interface Group
7. A Speech & language therapist (1) (to be nominated by the Lead Cleft Speech and Language Therapy Group)
8. An Orthodontist (1) (to be nominated by the Cleft Orthodontists Clinical Excellence Network).
9. A Specialist Cleft nurse (1) (to be nominated by the Cleft Nurses Clinical Excellence Network)
10. A Psychologist (1) (to be nominated by the Cleft Psychologists Clinical Excellence Network)
11. A Paediatric Dentist (1) (to be nominated by the Cleft Paediatric Dentist Clinical Excellence Network)
12. The Co-ordinator/Chair of the UK Cleft Centres Clinical Directors' Group (1)
13. A Cleft Co-ordinator (1) (to be nominated by the Cleft Coordinators Clinical Excellence Network).
14. A Representative from the group of 'other' specialities involved in cleft care (1) (to be nominated by CFS Council).
15. A Clinical representative from Northern Ireland (1) / Scotland (1) / Wales (1) / England (as appropriate, if not already represented) (to be nominated by those countries)
16. There may be representation, as determined by CDG to be appropriate, of any national bodies representative of Audit (1) and Research (1)
17. Clinical Directors/Clinical Leads of UK Cleft Centres not otherwise represented on CDG shall be invited to attend and become voting members so that all centres will be represented.
18. The Clinical Director/Project Leader of the CRANE service will be in attendance at Group meetings to which he/she will report, except when required to be absent because their own position is being discussed/decided. This individual will not be a voting member of the Group unless in another capacity and will not be eligible to become Chair.

19. The Director of the body which holds the contract for CRANE will be in attendance at Group meetings to which he/she will report, except when required to be absent because their own position is being discussed/decided. The Director will not be a voting member of the Board and will not be eligible to become the Chair.

20. A representative of the DH will always be invited to meetings and will receive minutes but will not be a voting member of the Board and will not be eligible to become the Chair.

21. Such other people who from time to time would serve the interests of the Cleft Development Group may be co-opted for a period of one year at a time.

Deputies for members may be appointed from time to time provided they are done so formally in writing by the nominating body to the CDG Chair. Where an individual comes to represent two positions on CDG, that person will continue to fulfil those roles and no additional person will be elected.

Additional representation will be considered (e.g. cleft paediatricians, cleft anaesthetists, cleft ENT and Audiology, cleft genetics) as and when those disciplines have formally established national Clinical Excellence Networks which genuinely represent those disciplines.

Meetings

Meetings will normally be held three times per year but must be held at least twice yearly with administrative support provided by the body which holds the CRANE contract, or the DoH or NHS bodies.

Amended May 2012

Appendix 5: Diagnosis and Procedure Codes, Hospital Episode Statistics (HES)

International classification of Disease 10th Revision (ICD-10) diagnostic codes for cleft lip and/or palate.

Code	Description
Q35	Cleft palate
Q36	Cleft lip
Q37	Cleft palate with cleft lip

Classification of Surgical Operations and Procedures 4th Revision (OPCS-4) codes for cleft lip and cleft palate repairs.

Code	Description
F031	Correction of deformity to lip
F291	Correction of deformity to palate

International classification of Disease 10th Revision (ICD-10) diagnostic codes for syndromes and anomalies used to identify ‘syndromic’ cleft patients. Patients were defined as ‘syndromic’ if there was a record of any of the following codes in any of the fourteen diagnosis code fields for any of that patient’s HES episodes.

Code	Description
D821	Di George's syndrome
	Congenital malformations of the nervous system (Q00-Q07)
Q00	Anencephaly and similar malformations
Q01	Encephalocele
Q02	Microcephaly
Q03	Congenital hydrocephalus
Q04	Other congenital malformations of brain
Q05	Spina bifida
Q06	Other congenital malformations of spinal cord
Q07	Other congenital malformations of nervous system
Q16	Congenital malformations of ear causing impairment of hearing
Q18	Other congenital malformations of face and neck
	Congenital malformations of the circulatory system (Q20-Q28)
Q20	Congenital malformations of cardiac chambers and connections
Q21	Congenital malformations of cardiac septa
Q22	Congenital malformations of pulmonary and tricuspid valves
Q23	Congenital malformations of aortic and mitral valves
Q24	Other congenital malformations of heart
Q25	Congenital malformations of great arteries
Q26	Congenital malformations of great veins
Q27	Other congenital malformations of peripheral vascular system
Q28	Other congenital malformations of circulatory system
Q380	Congenital malformations of lips, not elsewhere classified
Q75	Other congenital malformations of skull and face bones
Q86	Congenital malformation syndromes due to known exogenous causes, not elsewhere classified
Q87	Other specified congenital malformation syndromes affecting multiple systems

Continued on next page...

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Code	Description
	Chromosomal abnormalities, not elsewhere classified (Q90-99)
Q90	Down's syndrome
Q91	Edwards' syndrome and Patau's syndrome
Q92	Other trisomies and partial trisomies of the autosomes, not elsewhere classified
Q93	Monosomies and deletions from the autosomes, not elsewhere classified
Q95	Balanced rearrangements and structural markers, not elsewhere classified
Q96	Turner's syndrome
Q97	Other sex chromosome abnormalities, female phenotype, not elsewhere classified
Q98	Other sex chromosome abnormalities, male phenotype, not elsewhere classified
Q99	Other chromosome abnormalities, not elsewhere classified

Appendix 6: Cleft Audit Protocol for Speech – Augmented scores detail

Number (%) of CRANE-registered[a] consented children born with a cleft palate in 2007-2008, with reported speech outcomes, exclusion reasons and missing data at five years of age, according to region / unit.

Regional centre / MCN	Administrative Unit	Speech[b]									
			Reported		Exclusion reason		Not Available Reason		Total Acc. For	Missing Data	
		N	n	(%)	n	(%)	n	(%)	(%)	n	(%)
Northern & Yorkshire	Newcastle	89	60	(67.4)	17	(19.1)	10	(11.2)	(97.8)	2	(2.2)
	Leeds	91	52	(57.1)	18	(19.8)	19	(20.9)	(97.8)	2	(2.2)
North West & North Wales	Liverpool	107	56	(52.3)	17	(15.9)	31	(29)	(97.2)	3	(2.8)
	Manchester	112	75	(67)	32	(28.6)	2	(1.8)	(97.3)	3	(2.7)
Trent	Nottingham	123	83	(67.5)	27	(22)	8	(6.5)	(95.9)	5	(4.1)
West Midlands	Birmingham	146	102	(69.9)	7	(4.8)	9	(6.2)	(80.8)	28	(19.2)
East	Cambridge	111	66	(59.5)	25	(22.5)	15	(13.5)	(95.5)	5	(4.5)
North Thames	Great Ormond Street	131	63	(48.1)	15	(11.5)	15	(11.5)	(71)	38	(29)
	Chelmsford	44	26	(59.1)	8	(18.2)	8	(18.2)	(95.5)	2	(4.5)
The Spires	Oxford & Salisbury	138	91	(65.9)	19	(13.8)	22	(15.9)	(95.7)	6	(4.3)
South Wales & South West	Swansea	64	56	(87.5)	7	(10.9)	1	(1.6)	(100)	0	(0)
	Bristol	100	66	(66)	8	(8)	5	(5)	(79)	21	(21)
South Thames	Guy's and St Thomas'	140	85	(60.7)	29	(20.7)	21	(15)	(96.4)	5	(3.6)
Northern Ireland	Belfast	57	46	(80.7)	8	(14)	0	(0)	(94.7)	3	(5.3)
All	**All**	1,453	927	(63.8)	237	(16.3)	166	(11.4)	(91.5)	123	(8.5)

[a] Registered in CRANE by 6 August 2015. Note: MCN - Managed Clinical Network. [90]

[b] Excluding 588/2,013 (29.2%) children with submucous cleft palates (2.6%), missing one or more of all 16 CAPS-A data items (1.4%), or born with either a CL (24%) or a non-specified cleft type (1.2%) are excluded from speech data.

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[90] 80 cases had both 'Exclusion reason' and 'Not available reason' codes – as completion of these sections is not currently mutually exclusive. Therefore those with an 'Exclusion reason' were excluded from the 'Not available reason' data.