CRANE Database

www.crane-database.org.uk

Annual Report on Cleft Lip and/or Palate 2016

On behalf of the Cleft Development Group

Royal College of Surgeons
ADVANCING SURGICAL CARE

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[1] Appendices 1 through 4 provide detail on the project team, the CDG Membership and Terms of Reference, and the project's Governance and funding arrangements.

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
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[2]. 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[3].

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 units 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 2015 in England, Wales and Northern Ireland.

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 2004-2010). 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 from 2000 onwards. We describe the results of exploring school absences at seven years of age.

[2] 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.
[3] 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 2015

16,730 children were born over the last sixteen years, between 1 January 2000 and 31 December 2015, with a cleft lip and/or palate[4]. 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, 1,098 were born in 2015.

For the 830 children born in 2015 whose consent status had been verified, the parental consent rate was high (98.2%); with parental consent at 95%+ for all units. Out of all children registered in CRANE in 2015, 24.4% had not had their consent status verified at the time of writing this report (ranging from 0% to 69.4% between units).

Among children born in 2015, CRANE analyses revealed:

- 68.3% 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 0.5% of children with cleft palate (CP) only were diagnosed during antenatal screening: 71.2% were diagnosed at birth, leaving 28.3% who were diagnosed late according to the national standard[5] – 4.3% of whom were diagnosed after one month of age.
- 81.3% of children were referred by a maternity unit to a Cleft Administrative Unit within 24 hours of birth, maintaining similar levels of improvement to those noted at this time last year. With the variation between units, ranging from 66.2% to 91.5%, being statistically significant.
- Units went on to establish contact with 97.4% of parents within 24 hours of their child's referral. There was no statistically significant difference between units receiving these referrals; with all units contacting at least 93.3% of patients within 24-hour of receiving the referral. This demonstrates the commitment of units to ensure timely response to new referrals of babies born with a cleft, to help support these babies and their families in the important initial stages.

Cleft-related clinical outcomes at five years of age

CRANE collected clinical outcomes at five years of age among children born between 2004 and 2010. These outcomes include **height and weight** (2004-2010 births), the number of **decayed, missing and filled teeth** (dmft), a measure of oral health (2004-2009 births), **Five Year Old Index scores**, reflecting dental arch relationships and effects of primary cleft repair surgery on the facial growth of children with a complete unilateral cleft lip and palate (UCLP) (2004-2009 births), and a **speech assessed using the Cleft Audit Protocol for Speech – Augmented (CAPS-A) scoring system** (2007-

[4] As registered on the CRANE Database by 29 December 2016.
[5] Bannister P. Management of infants born with a cleft lip and palate. Part 1. Infant, 2008. 4(1): p. 5-8.

2009 births). Although there is still a high proportion of missing data, some units have reported outcomes for 88.9% of their eligible patients, suggesting that the reporting of these outcome data is feasible. For those children with reported outcomes:

- 42.4% had at least one decayed, missing or filled tooth (>0 dmft), which is slightly higher than the rate in the general population (38.8%). Bilateral cleft lip and palate (BCLP) was associated with the poorest oral health, with 52.1% of BCLP patients having at least one dmft at five years of age. The proportion of children with at least one dmft varied significantly according to cleft type and region / unit, although treatment indices (ranging from 68.3% to 91.8%) and care indices (ranging from 59.2% to 89.4%) across units indicate that, in the majority of cases, units have mechanisms in place to deal with any dental disease occurring.

- Of the 652 children with a complete UCLP for whom Five Year Old Index scores had been reported, 24.6% had scores of '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 of age in 1996[6].

- Of the 1,318 eligible children born in 2007-2009 with a cleft affecting their palate, and for whom speech CAPS-A scores had been reported across all 16 speech parameters measured, 59.8% had speech scores that would suggest their speech was not significantly different from their non-cleft peer group. Therefore, the national speech outcome standard #1 target of 55%[7] of children being expected to have normal speech, has been exceeded (on average) by the CRANE cohort born in 2007-2009.

Unfortunately, outcomes are still not collected consistently across units:

- Height and weight measures are not collected routinely (in fewer than 15% 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 (18.6% and 21.6% respectively). Nevertheless, this represents an improvement year-on-year (ongoing since the 2012 Annual Report), and reflects the appointment of paediatric dentists in these regions[8]; which bodes well for continued improvement in future dmft reporting.

- CRANE will continue to explore methods for improving communication and links with units to facilitate the submission of data to the database – the recent re-development of the CRANE Database and Website in 2015-2016 is expected to play a key role in facilitating this (see Chapter 5 for further detail on this).

[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.

[7] Based on the national outcome mean 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.

[8] Recent appointment of a paediatric dentist to examine children (and determine dmft) has resulted in improved data completeness for 2009 births – specifically, 59% completeness for the East and 34% completeness for Trent.

School absence at seven years of age among children with a cleft in England

In order for children to fulfil their academic potential, they need to attend school regularly to benefit from their education. Missing lessons leaves pupils vulnerable to falling behind, and children with poor attendance tend to achieve less in both primary and secondary school[9].

We explored school absence among children with a cleft lip and/or palate in England using CRANE data linked to the National Pupil Database (NPD). We focused on children at Key Stage 1 (KS1), when they are in Year 2 and turning seven years of age. To our knowledge, this is the first time that school absence has been examined among children with a cleft.

Linkage

6,194 CRANE-registered children born from 2000 onwards were matched to NPD records. Of these, 4,928 had KS1 assessments and were in Year 2 between 2006/07 and 2013/14, and 4,920 had absence data reported.

School absence among children with a cleft

Children with a cleft in Year 2 of school, when they turn seven years of age, have a mean overall absence rate[10] of 6.1% and authorised absence rate of 5.3%. These rates are 1.3 and 1.7 times higher than the background rates in the general population. 5.1% of children with a cleft are classified as persistent absentees and this compares to 2.7% of Year 2 pupils in the general population.

Two thirds of children with a cleft who had absence broken down by reason missed at least one school session for a medical or dental appointment. Compared to the general population, absence for medical or dental appointments (0.9% of all possible sessions) was three times higher among pupils with a cleft, while absence for illness (3.7% of all possible sessions) was 1.3 times higher among those with a cleft.

School absence according to syndrome status

Overall absence and authorised absence were significantly higher among children with a syndromic cleft compared to those with a non-syndromic cleft (8.0% vs. 5.5% and 7.2% vs. 4.7%, respectively). 9.5% of children with syndromic clefts were classified as persistent absentees compared with 3.6% of children with non-syndromic clefts.

Of the children with their absence broken down by reason, 78.6% of those with syndromic clefts and 63.2% of those with non-syndromic clefts missed at least one school session for a medical or dental appointment, and the overall absence for these appointments was almost twice as high among children with syndromic clefts as those with non-syndromic clefts (1.5% vs. 0.8%). Children with

[9] Department for Education. *The link between absence and attainment at KS2 and KS4. 2012/13 academic year*. London: Department for Education.2015.
[10] Overall absence, calculated as a percentage of the total number of possible sessions available for the academic year.

syndromic clefts also took significantly more time off for illness than their non-syndromic counterparts (4.6% vs. 3.4%).

School absence according to cleft type group among children without additional anomalies or syndromes

Among non-syndromic children, overall absence and authorised absence increased with each increasing cleft type severity group (CL, CP, UCLP and BCLP) and the difference between groups was statistically significant. Differences between cleft type groups in the proportion of children considered persistent absentees were not statistically significant.

Of the children with their absence broken down by reason, the proportion of children who missed at least one school session for a medical or dental appointment increased significantly from 49.1% of those with CL to 78.0% of those with BCLP. Furthermore, the proportion of school sessions missed for these appointments increased significantly with each increasing severity of cleft type (from 0.4% among those with CL to 1.2% among those with BCLP). Absence for illness did not vary significantly between cleft type groups.

Recommendations

Clinical care

- Late diagnosis of cleft palate (CP) remains an important issue that must be addressed. Among children born in 2015, 28.3% of those with CP were diagnosed late according to the national standard[11]. 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 —published a best practice guide to help healthcare professionals identify cleft palate in neonates. This guide[12] provides recommendations to ensure early detection of a cleft palate, and to improve and standardise routine postnatal examination of the palate.

- Increased 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

[11] Bannister P. Management of infants born with a cleft lip and palate. Part 1. Infant, 2008. 4(1): p. 5-8.
[12] http://www.rcpch.ac.uk/improving-child-health/clinical-guidelines-and-standards/published-rcpch/inspection-neonatal-palate

data *for all eligible patients* is still required so that CRANE can report data to the Quality Dashboard.

- 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[13] to evaluate patient (and parent) reported experience. These measures were combined into one Patient (and Parent) Reported Experience Measure (PREM) questionnaire and, through piloting[14] by the Cleft Psychology CEN, adjusted to apply to cleft services. The CRANE project team and the Cleft Psychology CEN are collaborating with a 12-month feasibility study 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 fourth 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.

[13] (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).
[14] 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[15]. 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[16].

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 29 December 2016 for children with a cleft lip and/or palate born in England, Wales and Northern Ireland between the 1 January 2000 and 31 December 2015. 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 2015 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 from 2000 onwards. We present data on school absence rates among children with a cleft lip and/or palate in England during National Curriculum year 2, when children turn seven years of age and undergo their Key Stage (KS) 1 assessment.

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.

[15] 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.
[16] 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[17]. 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'[18]. 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 2 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, NHS Wales, and the Northern Ireland Health and Social Care Board. 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[19].

For the first time, in March 2016, we provided analysis of data from Scotland in comparison to the rest of the UK as part of benchmarking the current service. This was provided for a fixed fee and some discussions have been had with Cleft Care Executive Board as to how CRANE might support data collection and analysis with Scotland in the future.

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 treats an average of 108 new children born with a cleft lip and /or palate each year. Several of

[17] Clinical Standards Advisory Group, Clinical Standards Advisory Group. Report of a CSAG Committee on cleft lip and/or palate, 1998, The Stationery Office, London.
[18] Department of Health, HSC 1998/238: Cleft lip and palate services, 1998, Department of Health: Leeds.
[19] Current funding is confirmed up until the end of the 2015/16 financial year.

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).

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

Regional Cleft Centre / MCN	Administrative Unit
Northern & Yorkshire	Royal Victoria Infirmary, 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 29 December 2016 pertaining to children born between 1 January 2000 and 31 December 2015 is included in the descriptions and 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)[20].

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 2010, 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 (7.7% of children born in 2015), 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 dental caries experience of 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 2009 (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[21]. CRANE collected both internal and external Five Year Old Index scores for consented children born between 2004 and 2009 with a complete UCLP (LAHSAL codes LAHS or HSAL). Some units 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

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

[21] 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.

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 between 2007 and 2009 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[22] – 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 2 to 8 (and Appendices 6 to 11). 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 [23], to link the information held in the CRANE database with the NPD.

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:

[22] 2006 data is not included as only four Cleft Speech Categories (CSCs) summarising current 12 CSCs were collected for 2006 birth.
[23] https://www.gov.uk/national-pupil-database-apply-for-a-data-extract

- 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 and for school absences. 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.

2.2.3. Outcomes and analyses: School absence

This year, we have focussed on school absence rates among children with a cleft in England during National Curriculum Year 2, when children turn seven years of age and undergo their Key Stage (KS) 1 assessment. We have focused on this age group as absence data are not available for children in the reception year of school when they are in the Early Years Foundation Stage.

All state funded primary, secondary and special schools in England provide individual level attendance data for pupils of compulsory school age (ages 5 to 15 at the start of the school year), via the school census. A pupil's overall absence rate is the total number of overall absence sessions as a percentage of the total number of possible sessions during the academic year. Overall absence is the sum of authorised and unauthorised absence and one session is equal to half a day. Schools are required to report the number of authorised absence sessions accrued by each pupil. Since the academic year 2006/07, this can either be reported by a reason for absence breakdown, of which there are eight categories, or by an aggregated total. CRANE requested data on two authorised absence reason categories: i. illness and ii. medical or dental appointments. Some schools do not have the required software to provide absence data broken down by reason and are only able to provide overall totals. Unauthorised absence must also be reported and is absence without permission from a teacher or other authorised representative of the school.

Pupils are identified as persistent absentees if their overall number of absence sessions meets a standard threshold. From the academic year 2005/06 to 2009/10, this threshold was set at 20% (equivalent to missing more than six weeks of lessons a school year), and between 2010/11 and 2014/15 the threshold was reduced to 15% (equivalent to missing more than four weeks of lessons a school year).

Children attending Year 2 between 2006/07 and 2013/14 are included in the results. We report the following absence measures:

- The proportion of children missing at least one school session during the academic year;
- Overall absence, calculated as a percentage of the total number of possible sessions available for the academic year;
- Authorised absence, calculated as a percentage of the total number of possible sessions available for the academic year;
- The proportion of children classified as persistent absentees;
- The proportion of children with absence broken down by reason who missed at least one school session for medical or dental appointments;
- Absence for medical or dental appointments, and absence for illness, calculated as a percentage of the total number of possible sessions available for the academic year for those with absence broken down by reason.

The above measures are reported for all children with a cleft lip and/or palate, according to syndrome status, and according to cleft type among those without additional anomalies or syndromes. Published summary data from the general population are presented for comparison with the whole cleft cohort.

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 (see Appendix 5 for a list of the HES diagnosis and procedure codes used by CRANE).

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 2015 in England, Wales and Northern Ireland. Data entered into the CRANE Database by 29 December 2016 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 1,098 children born in 2015 who have been referred to a Cleft Unit for treatment and registered on CRANE is described below (and detailed in Appendix 6):

- The parents of 830 (75.6%) had been approached for consent; this figure varied across the regions and units submitting data to CRANE, ranging from 30.6% (South Thames) to 100% (Leeds). In addition, 98.2% of children whose parents had been through the consenting process, provided consent for their child's data to be submitted to CRANE, which is extremely positive (with all units reporting consent for 95%+ of families approached).
- The parents of 268 (24.4%) children born in 2015 had not yet been approached for consent; this proportion ranged from 0% (Leeds) to 69.4% (South Thames) across units . Of these families, it had not been possible to obtain consent (verification) for 5.2% (1.3% of all children born in 2015).

Overall, the proportions described above – including high consent rates for those children whose parents have been approached – are consistent with proportions in previous Annual Reports. 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.

3.2. Number of registrations

A total of 16,730 children born with a cleft lip and/or palate over the last sixteen years, between 1 January 2000 and 31 December 2015, have been registered on the CRANE Database. Of these, 11,024 children were registered on CRANE over the last 10 years and 1,098 registered in 2015 (Appendix 7 provides detail on registrations over the last 10 years according to region / unit, by year of birth). The largest number of births are consistently registered by the North West & North Wales and North Thames regions year-on-year.

Cleft palate (CP) is the most common of the four types of cleft[24], affecting on average 43% of the cleft population, between 1 January 2000 and 31 December 2015. Bilateral cleft lip and palate (BCLP) is the least common type, affecting on average 9% of people with clefts. Of the 1,098 children registered on CRANE in 2015, 224 had unilateral cleft lip and palate (UCLP) of which 75% had complete UCLP (defined by either 'LAHS..' or '..HSAL' LAHSAL codes). In addition, 8.3% of 2015 registrations did not have their type of cleft specified (either by LAHSAL codes or by the units) (Appendix 7 provides detail on registrations over the last 10 years according to cleft type, by year of birth).

3.3. Characteristics of children born with a cleft lip and/or palate, 2015

Of the children born with a cleft in 2015, whose sex was reported to CRANE, most were boys (57%). Twenty-three children did not have their sex reported to CRANE (2% of the total children registered for 2015). There were significant gender differences in the distribution of cleft type ($p<0.001$), with CP more prevalent among females (53% vs. 47% in males), while CL, UCLP and BCLP is more prevalent among males (68% vs. 32%, 65% vs. 35%, and 63% vs. 37% respectively).

Gestational age was reported for 477 (58.5%) of the consented babies born in 2015. Therefore, further improvements in data completeness are required.

- The mean gestation for those born in 2015 was 38.8 weeks (95% CI 38.6 to 39 weeks) and ranged from 27 to 43 weeks.
- Forty-seven (9.9%) babies were premature (born before 37 weeks' gestation), which is higher than the seven per cent national average in England and Wales[25], 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 41.5% of registered (and consented) children were missing this information.
- A valid birth weight was reported for 480 (58.9%) consented babies born in 2015. The mean birth weight was 3.2kg (95% CI 3.1 to 3.3kg), which is consistent with the national average in England.

Among all the children born in 2015, four (0.4%) deaths were reported to CRANE. The majority of which (75%) occurred before one month of age. It is not known from CRANE whether these children had additional anomalies or syndromes.

[24] Cleft type is defined according to reported LAHSAL codes or, where LAHSAL has not been reported (for 7.7% of children registered in 2015), it is based on the cleft type reported by the region / unit registering the child.
[25] 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.

3.4. Timing of diagnosis

3.4.1. Diagnosis times among children born in 2015

Of the 1,098 children born in 2015 with a cleft diagnosis, 9% did not have the timing of their diagnosis reported to CRANE. Most units (10/14) had rates of missing diagnosis time data below 10%. Areas with high levels of missing diagnosis time data included Cambridge (26.4%) and Liverpool (24%).

Of the 999 children born in 2015 with a reported diagnosis time, 44.4% had their cleft diagnosed antenatally. The proportion of children diagnosed antenatally varying significantly according to cleft type ($p<0.001$), with only 0.5% of CP patients diagnosed antenatally (compared to rates of 68.3%, 85.5% and 87% for CL, BCLP and UCLP respectively), which demonstrates the difficulty of identifying this type of cleft with current antenatal screening techniques (see Appendix 8 for further detail on timing of diagnosis by cleft type).

Of the 555 children not diagnosed during the antenatal period, 74.2% were diagnosed at birth. The majority (81.2%) of the 125 children diagnosed postnatally with a CL, UCLP and BCLP had their cleft identified at the time of birth. The corresponding figure for the 401 children with a CP is 71.6%; and in 7% of cases the diagnosis occurred after one month of age. It should be noted that some children born in 2015 with a CP may not yet have had their cleft identified.

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[26]. 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 2015. No statistically significant differences were found between birth years ($p=0.22$), indicating diagnosis times have not improved in recent years.

Table 2 shows the CP diagnosis times according to the region / unit. The proportion of CPs diagnosed at birth ranged from 54.8% (South Thames) to over 78.1 (Northern Ireland). 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.

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

Table 2. Number (%) of CRANE-registered children born between 2011 and 2015 with a cleft palate, according to the timing of diagnosis and region / unit.

Regional Cleft Centre / MCN	Administrative Unit	Time of diagnosis in relation to birth[a] n (%)						
		Antenatal	At birth	≤1 week[b]	≤1 month	≤6 months	>6 months	**All**
Northern &	Newcastle	4 (2.6)	100 (64.1)	21 (6.9)	12 (7.7)	11 (7.1)	8 (5.1)	156
Yorkshire	Leeds	0 (0)	100 (68)	25 (8.3)	9 (6.1)	7 (4.8)	6 (4.1)	147
North West &	Liverpool	4 (3)	104 (77.6)	15 (5.4)	3 (2.2)	6 (4.5)	2 (1.5)	134
North Wales	Manchester	0 (0)	103 (72.5)	24 (8.7)	6 (4.2)	7 (4.9)	2 (1.4)	142
Trent	Nottingham	1 (0.6)	127 (70.2)	38 (21)	6 (3.3)	7 (3.9)	2 (1.1)	181
West Midls.	Birmingham	0 (0)	177 (74.7)	34 (14.3)	7 (3)	12 (5.1)	7 (3)	237
East	Cambridge	1 (0.7)	93 (68.4)	24 (17.6)	8 (5.9)	9 (6.6)	1 (0.7)	136
North Thames	Grt Ormond St	4 (2.1)	105 (55.3)	60 (20.6)	6 (3.2)	8 (4.2)	7 (3.7)	190
	Chelmsford	4 (4)	66 (65.3)	19 (6.5)	5 (5)	3 (3)	4 (4)	101
The Spires	Oxford/Salisbury	2 (1)	146 (74.5)	20 (10.2)	7 (3.6)	9 (4.6)	12 (6.1)	196
South Wales &	Swansea	0 (0)	69 (75)	12 (5.4)	8 (8.7)	2 (2.2)	1 (1.1)	92
South West	Bristol	4 (3)	86 (65.2)	14 (6.3)	15 (11.4)	10 (7.6)	3 (2.3)	132
South Thames	GSTT	2 (0.8)	138 (54.8)	72 (28.6)	18 (7.1)	16 (6.3)	6 (2.4)	252
N. Ireland	Belfast	0 (0)	57 (78.1)	7 (9.6)	0 (0)	4 (5.5)	5 (6.8)	73
All	All	26 (1.2)	1,471 (67.8)	385 (17.8)	110 (5.1)	111 (5.1)	66 (3)	2,169

[a] 148/2,317 (6.5%) missing diagnosis time and excluded from 'All' values. **Note:** MCN - Managed Clinical Network.
[b] Recording of 'timing of diagnosis' within 72 hours commenced in May 2014 to align CRANE data collection with NIPE standards[27]. With only small numbers having been recorded using this timing (n=47, 4.7%), we report '≤72 hours' cases within the '≤1 week' timing (until recording of this timing is well established).

3.5. Referral to and first contact with a cleft team

In August 2014 we received Confidentiality Advisory Group (CAG) approval to expand the notification dataset for non-consented patients to include all nine 'first contact information' timing fields. This expansion was built on the small changes introduced in January 2014 to allow us to collect the same information as reported in Quality Dashboard. After 2 years of this revised data collection, we report fully on 'first contact information' timing fields for both non-consented and consented cases in the two sections below.

3.5.1. Referral among children born in 2015

Out of the 1,098 children born in 2015, 28.2% were missing referral time; with the proportion of children missing referral time ranging from 2.9% (Leeds) to 70.8% (South Thames).

Of the 788 children with a reported referral time, 81.3% were referred to a Cleft Unit within 24 hours of birth.

95.2% of the 353 children whose clefts were diagnosed antenatally were referred to a Cleft Unit within 24 hours of birth, while 73.3% of the 435 children whose clefts were diagnosed after birth

[27] 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).

were referred to a Cleft Unit within 24 hours of birth. These high rates are consistent with rates reported last year.

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 (69.6% of those with a known cleft type; compared to rates of 89.5-96.7%+ for other cleft types), which corresponds with later diagnosis times for these children.

The proportion of referrals within 24 hours of birth also varied significantly according to **cleft /administrative unit** ($p<0.01$); ranging from 66.2% (GOSH) to 91.5% (Cambridge). Despite this statistically significant variation between units, overall rates of referral within 24 hours remain high (as for previous reporting years).

3.5.2. First contact between units and parents of children born in 2015

Out of the 1,098 children born in 2015, 10.9% were missing the first contact time between units; with the proportion of children missing first contact time ranging from 0% (Newcastle, Bristol & South Thames) to 44.4% (GOSH)[28].

Of the 978 children with a reported contact time, units established contact with 97.4% of parents within 24 hours of referral; representing another year-on-year improvement in registrations (95.7% for 2014 registrations and 92.5% for 2013 registrations).

The proportion of patients contacted within 24 hours of being referred to a unit did not vary significantly according to **cleft type** ($p=0.64$).

The proportion of contacts between units and parents of patients within 24 hours of referral also did not vary significantly according to the **cleft /administrative unit** receiving the referral ($p=0.32$); with contact rates varying between 93.3% and 100%. All units contacted at least 93.3% of their patients within 24 hours of receiving the referral. This demonstrates the commitment of units to ensure a timely response to new referrals of babies born with a cleft, to help support these babies and their families in the important initial stages.

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

Five year outcomes include height, weight, decayed missing and filled teeth (dmft), the Five Year Old Index, and the Cleft Audit Protocol for Speech – Augmented (CAPS-A) scores. These are reported for only consented children born between 2004 and 2010 (excluding children with submucous cleft palates)[29]. Information and analyses of these data are presented in the next six subsections.

[28] An improvement on 53.6% at the time of last year's report.
[29] Submucous cleft palate patients excluded from all five year outcomes as all/most teams do not audit these patients.

3.6.1. Reporting of outcomes

We describe the data completeness for outcomes at five years of age, according to region/unit, below:

- There is a high proportion of missing data for five-year old weight and height (for eligible children 64% and 65.5% missing respectively), this is despite continued small improvements year-on-year; with wide variation in reporting weight and height data across regions. Variation in reporting ranged from 72%+ for both weight & height (Northern & Yorkshire region), to less than 1% for both weight & height (Northern Ireland). This suggests that this data is not routinely collected in some regions.
- The proportion of eligible children with reported decayed, missing, filled teeth (dmft) index scores varied across regions from 18.6% (East) to 87.2% (West Midlands). Despite the East's low data completeness rate, this represents an almost 10% improvement on last year's reporting rate and reflects the appointment of a paediatric dentist in this region[30].
- The proportion of children with reported Five Year Old Index scores continues to increase year-on-year, which is encouraging. Nevertheless, there was wide variation in reporting of Five Year Old Index data across the regions/units from 10.8% (Northern Ireland) to 88.9% (The Spires). Although Northern Ireland submitted data for only 10.8% of their eligible patients, they have collected Five Year Old Index data for only 2 years – and therefore are likely to show improved data completion rates year-on-year.
- CRANE is encouraged by the fact that regions/units have shown increased rates in reported speech data year-on-year since the expanded 16 CAPS-A speech outcome scores were requested 3 years ago. The proportion of eligible children with Speech outcome scores ranged from 56% (GOSH) to 91% (Swansea).

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. Nevertheless, it is positive to note that reporting has increased for all outcomes at five years of age[31] since this time last year; and it is hoped that this trend will continue over the next few years (see Appendix 9 for detail on children born 2004-2010 with reported outcomes at five years of age).

3.6.2. Weight and height (2004-2009 births)

Five-year weight and height were reported for 36% and 34.5%, respectively, for the 6,533 children born in 2004-2010[32], who were alive at five years of age. The mean (SD) weight was 19.6kg (3.2kg) while the mean (SD) height was 111.7cm (5.9cm). A small difference in mean weight existed between the sexes (boys 19.8kg vs. girls 19.4kg), and boys were marginally taller than girls (112.2cm vs. 111cm).

[30] Recent appointment of a paediatric dentist to examine children (and determine dmft) has resulted in improved data completeness of 59% for 2009 births (for the East).
[31] 3.2% for weight, 2.9% for height, 0.3% for dmft, 1.6% for 5 year index and 2.1% for speech.
[32] Submucous cleft palate patients excluded from all five year outcomes as all/most teams do not audit these patients.

3.6.3. Decayed missing and filled teeth (dmft) (2004-2009 births)

The dmft describes the dental caries an individual has experienced 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[33]. We collect dmft data on CRANE-registered consented children at five years of age.

Among children with a reported dmft outcome[34] , 42.4% 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. Four hundred and seventy-five children (14.3%) had a dmft score greater than 5.

Dental caries according to cleft type

Table 3 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[35]. The comparable figure of 42.4% among CRANE-registered children (shown in Tables 3 and 4) 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 (33.5% versus 38.8%). Despite this, the mean dmft was higher among CP, UCLP and BCLP patients compared to the general population's mean (2.3, 2 and 2.7, respectively, compared to a mean of 1.5).

Table 3. Number (%) of CRANE-registered consented children born 2004-2009 with a cleft lip and/or palate according to the number of decayed, missing or filled teeth (dmft) at 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	1.3 (1.1 to 1.5)	454 (66.5)	229 (33.5)	(29.9 to 37.1)	683
CP	2.3 (2.1 to 2.6)	793 (58.9)	553 (41.1)	(38.5 to 43.7)	1346
UCLP	2.0 (1.8 to 2.2)	482 (52.7)	433 (47.3)	(44.1 to 50.6)	915
BCLP	2.7 (2.3 to 3.1)	172 (47.9)	187 (52.1)	(46.9 to 57.3)	359
Not specified	2.5 (0.7 to 4.4)	17 (58.6)	12 (41.4)	(22.3 to 60.4)	29
All	2.1 (1.9 to 2.2)	1,918 (57.6)	1,414 (42.4)	(40.8 to 44.1)	3,332

[33] (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.

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

[35] 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.

[a] Exclusions from 'All' values (not mutually exclusive): 2,221/5,553 (40%) children with missing dmft data, 209/5,867 (3.6%) children with submucous clefts[36], and 105/5,658 (1.9%) children who died before the age of five.
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 60% 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

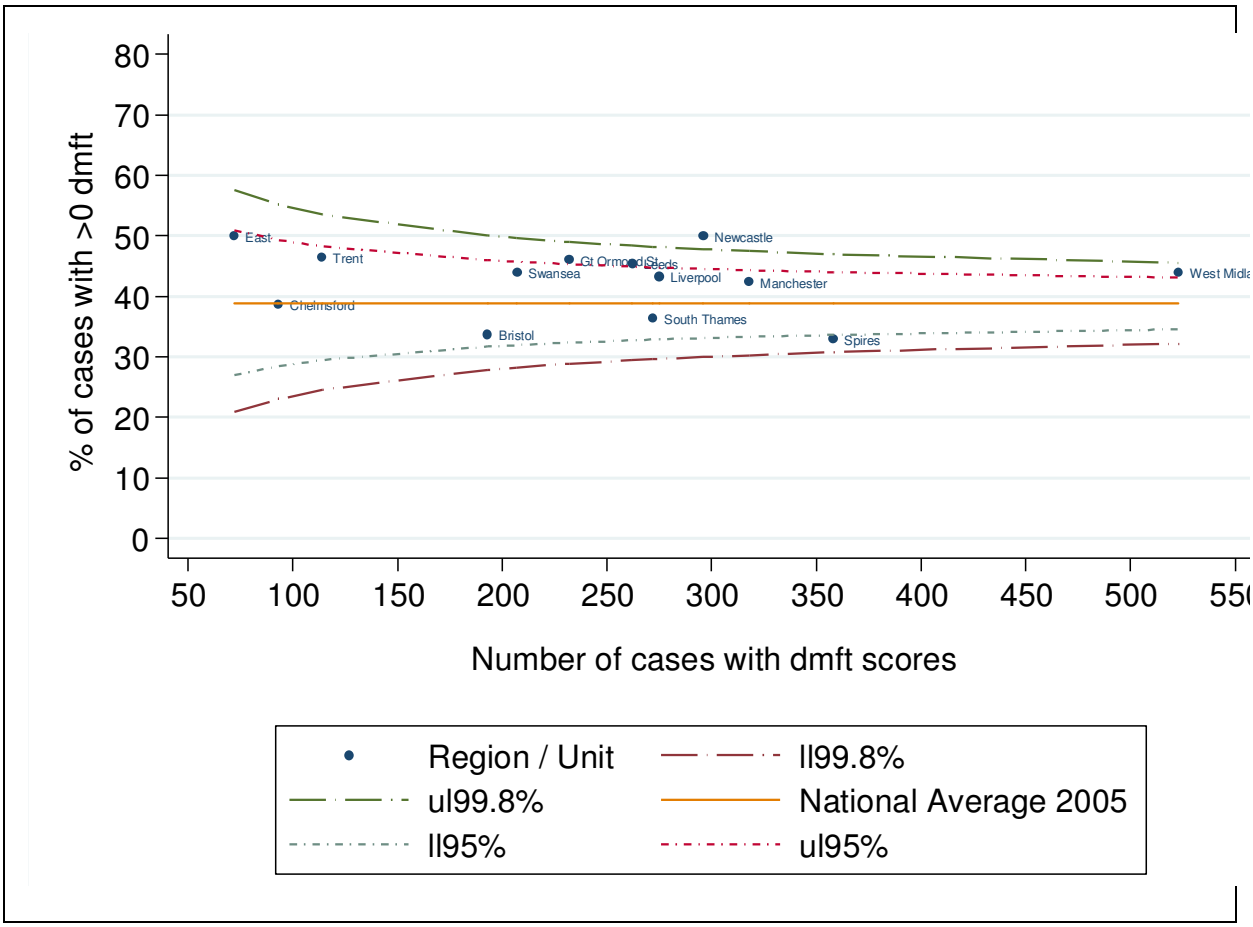
Table 4 shows the prevalence of dental caries according to region / unit[37]. There was a significant variation in dmft scores across Units ($p<0.001$). Children registered in the Trent region, the East region, and in Newcastle, had the highest numbers of mean dmft (2.9, 2.8 and 2.8 respectively), which were significantly different to the overall mean (2.1). It should be noted that Trent and the East submitted data for few patients[38], 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. Bristol, Chelmsford, and the Spires region had mean dmft values that were significantly lower than the overall mean (1.3, 1.5 and 1.5 respectively).

In terms of the proportion of cleft children with at least one dmft (>0 dmft), the Spires region had the lowest proportion (33%), 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[39].

The funnel plot[40] 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[41].

[36] Submucous cleft palate patients excluded from all five year outcomes as all/most teams do not audit these patients.
[37] Currently there is some difference in the cleft calibration of dmft assessors.
[38] Note that dmft data were reported for only 21.6% of eligible children registered by the Trent region and 18.6% of eligible children registered by the East region (as reported in Appendix 9).
[39] 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.
[40] 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 4. In addition, it is not adjusted (or risk adjusted) in any way.
[41] 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.

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: Funnel plot 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 (only). Therefore only 13 units shown as Northern Ireland data excluded.

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[42] and is worth investigating (more information on funnel plots can be found in the Glossary at the front of this report).

[42] Regional differences in the levels of dental disease are not only be affected by the dental care received by children. Oral health is also affected by deprivation, ethnicity, cultural differences in attitudes to dental health and water fluoridation levels.

Table 4. Number (%) of CRANE-registered consented children born between 2004 and 2009 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]		All[b]		All[b]
		Mean (95% CI)	n (%)	n (%)	95% CI	(N)	Average (%)	(N)	Average (%)	(N)
Northern & Yorkshire	Newcastle[§¥]	2.8 (2.3 to 3.3)	148 (50)	148 (50)	(44.3 to 55.7)	296	(69)	266	(59.2)	268
	Leeds[§¥]	2.6 (2.1 to 3.1)	143 (54.6)	119 (45.4)	(39.4 to 51.5)	262	(70.4)	262	(63.1)	262
North West & North Wales	Liverpool	1.9 (1.5 to 2.3)	156 (56.7)	119 (43.3)	(37.4 to 49.2)	275	(68.3)	271	(61.4)	271
	Manchester	2.0 (1.6 to 2.5)	183 (57.5)	135 (42.5)	(37 to 48)	318	(71)	317	(65.1)	317
Trent	Nottingham[¥*]	2.9 (2 to 3.8)	61 (53.5)	53 (46.5)	(37.2 to 55.8)	114	(79)	111	(62.6)	112
West Midlands	Birmingham[§¥]	2.1 (1.8 to 2.4)	293 (56)	230 (44)	(39.7 to 48.2)	523	(68.4)	522	(61.7)	522
East	Cambridge[*]	2.8 (1.8 to 3.8)	36.0 (50)	36.0 (50)	(38.2 to 61.8)	72	(70)	70	(61.2)	70
North Thames	Gr Ormond St[¥]	2.1 (1.7 to 2.6)	125 (53.9)	107 (46.1)	(39.7 to 52.6)	232	(73.1)	230	(60.4)	230
	Chelmsford	1.5 (0.9 to 2.2)	57 (61.3)	36 (38.7)	(28.6 to 48.9)	93	(83.6)	75	(80.3)	75
The Spires	Oxford/Salis.	1.5 (1.2 to 1.8)	240 (67)	118 (33)	(28.1 to 37.9)	358	(91.8)	274	(89.4)	274
South Wales & South West	Swansea[§¥]	1.9 (1.5 to 2.3)	116 (56)	91 (44)	(37.1 to 50.8)	207	(76.5)	205	(64)	207
	Bristol[§¥]	1.3 (0.9 to 1.6)	128 (66.3)	65 (33.7)	(26.9 to 40.4)	193	(80.9)	189	(75.4)	189
South Thames	GSTT[§¥]	1.6 (1.3 to 1.9)	173 (63.6)	99 (36.4)	(30.6 to 42.2)	272	(84.2)	268	(78.4)	269
Nrthn. Ireland	Belfast	2.6 (1.9 to 3.4)	59 (50.4)	58 (49.6)	(40.4 to 58.8)	117	(75.2)	116	(62.1)	116
All	All	2.1 (1.9 to 2.2)	1,918 (57.6)	1,414 (42.4)	(40.8 to 44.1)	3,332	(74.8)	3,176	(67.1)	3,182

[a] Exclusions from dmft values (not mutually exclusive): 2,221/5,553 (40%) children with missing dmft data, 209/5,867 (3.6%) children with submucous clefts[43], and 105/5,658 (1.9%) children who died before the age of five.

Note: MCN – Managed Clinical Network.

[b] Exclusions from Treatment and Care Index (not mutually exclusive): Children who died before the age of five, children with submucous clefts, and cases without a dmft score of 0[44] or all relevant dmft data items (to allow calculation of treatment and care index scores).

Individual unit considerations: [§]Cleft 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.

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

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

Dental Treatment and Care Indices

Table 4 also shows the average treatment index and care index (reported for the second time) for children according to region / unit. Both indices are calculated from the dmft[45], 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[46]. 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 3,176 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 Liverpool and in the West Midlands had the lowest average proportion of treated dental disease (68.3 and 68.4 respectively; equal to 6.5% and 6.4% less than the national average), while the Spires region had highest average proportion of treated dental disease (91.8% equal to 17% 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[47]. 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 3,182 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 Newcastle had the lowest average care index (59.2% equal to 7.9% less than the national average), which means decay that might be treated by fillings has remained untreated or decay was so severe extraction was the treatment of choice. Meanwhile the Spires region had the highest average proportion of children receiving care by fillings (89.4% equal to 22.3% more than the national average).

[45] 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).

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

[47] If a dmft score for an individual is 0 then the care index is 1 (100%) as there is no dental 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, ethnicity, 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[48]. 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%)[49]. 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 (2004-2009 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[50]. 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[51]. The Five Year Old Index may, therefore, also be used to compare treatment outcomes between centres and surgeons. Patients scoring '1' and '2' on the index are considered to have the best possible outcomes, while those scoring '4' and '5' are thought to have poor outcomes in terms of facial growth, and they may benefit from further surgery to correct their facial disproportion once facial growth is complete.

The majority of Five Year Old Index scores provided by all regions/units were externally validated (in 591/652 (90.6%) of eligible cases), and where externally validated scores were unavailable, internal scores were included in the analysis (in 61/652 (9.4%) of cases). Overall, 44.6% of complete UCLP patients born between 2004 and 2009 had Five Year Old Index scores in the two groups considered to have the best possible dental arch relationships (scores '1' or '2') while 24.6% 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[52]. (see Appendix 10 for detail on children born between 2004 and 2009 with a complete unilateral cleft lip and palate, according to Five Year Old Index scores and region / unit).

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[48] 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.

[49] 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.

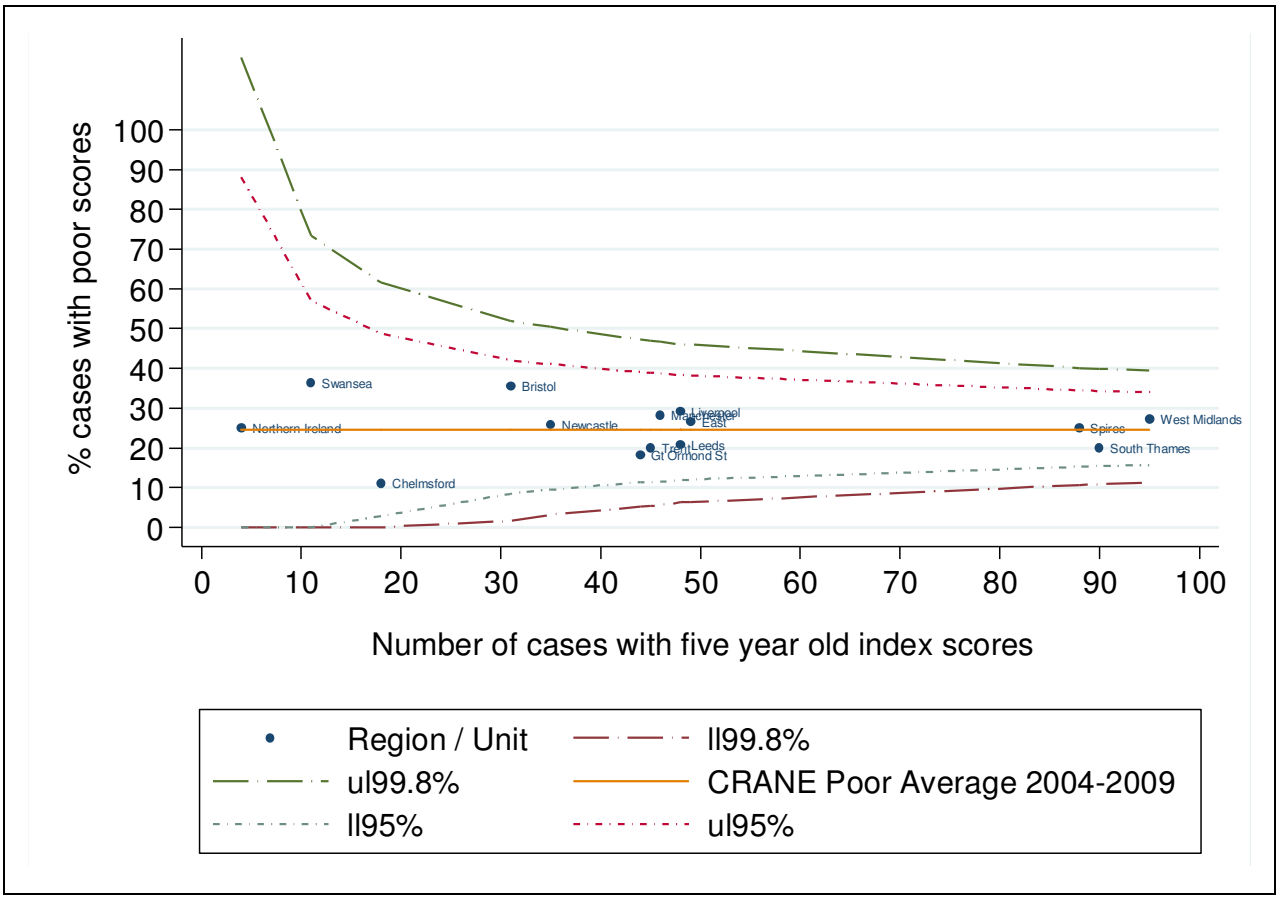
[50] Johnson N, Williams AC, Singer S, Southall P, Atack N and Sandy JR. Dentoalveolar relations in children born with a unilateral cleft lip and palate (UCLP) in Western Australia. The Cleft Palate-Craniofacial Journal, 2000. 37 (1): p. 12-16.

[51] 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.

[52] 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 funnel plot[53] in Figure 2 further demonstrates the proportion of CRANE 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 for CRANE five-year olds across all units of 24.6%[54]. It also shows that all regions' / units' rates of poor index scores fall within the expected range given the number of children (born between 2004-2009) 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).

Figure 2. Funnel plot of five-year olds (born between 2004 and 2009) with poor Five Year Old Index scores according to the number of children at each region / unit with index scores.



Note: Funnel plot centred on national average (for 2004-2009 births reported in CRANE) of poor Five Year Old Index scores across all units of 24.6%.

The fact that Five Year Old Index scores were submitted for only 70.6% of children, and the wide variation in the number of children within each region / unit (ranging from 4 to 88), 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.

[53] This funnel plot is calculated using valid data as denominators (not considering missing data), subject to the same inclusions and exclusions as data in Appendix 10. In addition, it is not adjusted (or risk adjusted) in any way.
[54] 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.

3.6.5. Cleft Audit Protocol for Speech – Augmented scores (2007-2009 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 the third year that 16 CAPS-A speech outcome scores have been requested[55]. The parameters of speech assessed include:

- Resonance (hypernasality and hyponasality) and nasal airflow (audible nasal emission and nasal turbulence). These are structurally related speech characteristics 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 65.9% of consented children born between 2007 and 2009 had reported speech outcomes for all 16 CAPS-A parameters, and 22.8% had reported reasons why speech data was not collected (e.g. Patient deceased or emigrated, transferred in or out of area, etc[56]). Table 13 also shows the distribution of proportions of reported speech outcomes (or reasons why not collected) by region / unit.

Table 5. Number (%) of CRANE-registered[a] consented children born with a cleft palate (2007-2009), with speech outcome data or reasons this outcome was not collected at five years of age, according to region / unit.

Regional centre / MCN	Administrative Unit	Speech[b]					
			Reported		Reason outcome not collected		Total cases acc. for
		N	n	(%)	N	(%)	(%)
Northern	Newcastle	100	74	(74)	12	(12)	(86)
& Yorkshire	Leeds	114	72	(63.2)	39	(34.2)	(97.4)
North West	Liverpool	158	85	(53.8)	64	(40.5)	(94.3)
& North Wales	Manchester	146	102	(69.9)	37	(25.3)	(95.2)
Trent	Nottingham	192	122	(63.5)	50	(26)	(89.6)
West Midlands	Birmingham	192	142	(74)	19	(9.9)	(83.9)
East	Cambridge	163	101	(62)	50	(30.7)	(92.6)
North Thames	Great Ormond Street	191	107	(56)	41	(21.5)	(77.5)
	Chelmsford	70	47	(67.1)	12	(17.1)	(84.3)
The Spires	Oxford & Salisbury	184	121	(65.8)	32	(17.4)	(83.2)
South Wales	Swansea	89	81	(91)	6	(6.7)	(97.8)
& South West	Bristol	121	80	(66.1)	19	(15.7)	(81.8)
South Thames	Guy's and St Thomas'	199	119	(59.8)	66	(33.2)	(93)
Northern Ireland	Belfast	81	65	(80.2)	8	(9.9)	(90.1)
All	**All**	2,000	1,318	(65.9)	455	(22.8)	(88.7)

[a] Registered in CRANE by 29 December 2016. Note: MCN - Managed Clinical Network.
[b] Exclusions (not mutually exclusive): children who died before the age of 5 years, with submucous cleft palates, missing one or more of all 16 CAPS-A data items, born with either a CL or a non-specified cleft type, and syndromic children. For more detail on exclusions see Appendix 9.

[55] Four Cleft Speech Categories (CSCs) summarising the all 12 CSCs were collected for births prior to 2007. Because of this, speech data collected before 2007 is not included in this report. Changes to expand data collection to 12 CSCs were made in 2014.
[56] Plus: Syndromic Diagnosis; Clinically contraindicated (other than syndromic) - this record type for this patient; Lack of staff / facilities / equipment; Patient DNA / cancelled / did not consent / cooperate; or Other reason.

Overall, rates of data completeness ranged from 77.5% of children being accounted for at GOSH to 97.8% of children being accounted for in Swansea (see Appendix 11 for detail of missing data).

The scores for each of the individual 16 CAPS-A assessed are presented in Tables 6 and 7, found in the next two sections.

Resonance and Nasal Airflow

In Table 6, 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 6. Number (%) of CRANE-registered [a] consented children born with a cleft palate in 2007-2009, according to the four parameters for resonance and nasal airflow

Description	Score	N[b]	(%)
RESONANCE – HYPERNASALITY			
Absent	0	1,015	(77)
Borderline – minimal	1	155	(11.8)
Mild – evident on close vowels	2	92	(7)
Moderate – evident on open and close vowels	3	36	(2.7)
Severe – evident on vowels and voiced consonants	4	20	(1.5)
RESONANCE – HYPONASALITY			
Absent	0	1,099	(83.4)
Mild – partial dentalization of nasal consonants and adjacent vowels	1	201	(15.3)
Marked – dentalization of nasal consonants and adjacent vowels	2	18	(1.4)
NASAL AIRFLOW – AUDIBLE NASAL EMISSION			
Absent on pressure consonants	0	1,207	(91.6)
Occasional: pressure consonants affected <10% of the sample	1	82	(6.2)
Frequent: pressure consonants affected >10% of the sample	2	29	(2.2)
NASAL AIRFLOW – NASAL TURBULENCE			
Absent on pressure consonants	0	1,035	(78.5)
Occasional: pressure consonants affected <10% of the sample	1	234	(17.8)
Frequent: pressure consonants affected >10% of the sample	2	49	(3.7)
TOTAL		**1,318**	**(100)**

[a] Registered in CRANE by 29 December 2016. [b] Number of eligible children (as specified for Table 13).

In terms of resonance, 4.2% 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 6). 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 7) show that 2.9% of children had 'weak and or nasalised consonants' 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, 249/1,318 (18.9%) of the children with

reported surgical data[57] have had surgery for speech purposes (referred to as secondary speech surgery) before the age of five years.

In addition, 1,111 out of the 1,318 (84.3%) children with reported scores for all four resonance and nasal airflow parameters listed in Table 6 had all green scores, indicating that no structural problems existed in relation to these parameters.

Cleft Speech Characteristics (CSCs)

Table 7 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.

Table 7. Number (%) of CRANE-registered[a] consented children born with a cleft palate in 2007-2009, according to the twelve Cleft Speech Characteristics (CSCs) parameters.

Cleft Speech Characteristics (CSCs)		Score	N[b]	(%)
ANTERIOR ORAL CSCs	1. Dentalisation / Interdentalisation	A	1072	(81.3)
		B	246	(18.7)
	2. Lateralisation / Lateral	A	1,228	(93.2)
		B	52	(3.9)
		C	38	(2.9)
	3 Palatalisation / Palatal	A	1,010	(76.6)
		B	161	(12.2)
		C	147	(11.2)
POSTERIOR ORAL CSCs	4. Double Articulation	A	1,280	(97.1)
		B	34	(2.6)
		C	4	(0.3)
	5. Backed to Velar / Uvular	A	1,122	(85.1)
		C	81	(6.1)
		D	115	(8.7)
NON ORAL CSCs	6. Pharyngeal Articulation	A	1,292	(98)
		C	17	(1.3)
		D	9	(0.7)
	7. Glottal Articulation	A	1,241	(94.2)
		C	38	(2.9)
		D	39	(3)
	8. Active Nasal Fricatives	A	1,202	(91.2)
		C	74	(5.6)
		D	42	(3.2)
	9. Double Articulation	A	1,295	(98.3)
		C	15	(1.1)
		D	8	(0.6)
PASSIVE CSCs	10. Weak and or nasalised consonants	A	1,246	(94.5)
		C	34	(2.6)
		D	38	(2.9)
	11. Nasal realisation of plosives	A	1,279	(97)
		C	21	(1.6)
		D	18	(1.4)
	12. Gliding of fricatives	A	1,291	(98)
		C	21	(1.6)
		D	6	(0.5)
		TOTAL	**1,318**	**(100)**

[a] Registered in CRANE by 29 December 2016. [b] Number of eligible children (as specified for Table 5).

[57] VP surgery/fistula repair data was only reported for 1,313/1,318 (99.6%) of eligible children (as specified for Table 5 to 7).

'Palatalisation / Palatal' anterior oral CSCs were the most commonly occurring CSC, affecting 23.4% of children (12.2% with scores of B and 11.2% 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 875/1,318 (66.4%) children with reported scores for all 12 CSC parameters listed in Table 7 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[58]:

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 7.
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 7.

Out of the 1,318 children with reported scores for all 16 CAPS-A speech parameters listed in Tables 6 & 7, 788 (59.8%) 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%[59] was met and exceeded on average by the CRANE cohort born in 2007-2009. 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* (70% of children with CP – with reported speech score – achieved *normal speech*) and significantly fewer children with BCLP achieved *normal speech* (only 36% of children with BCLP had achieved *normal speech* by age 5).

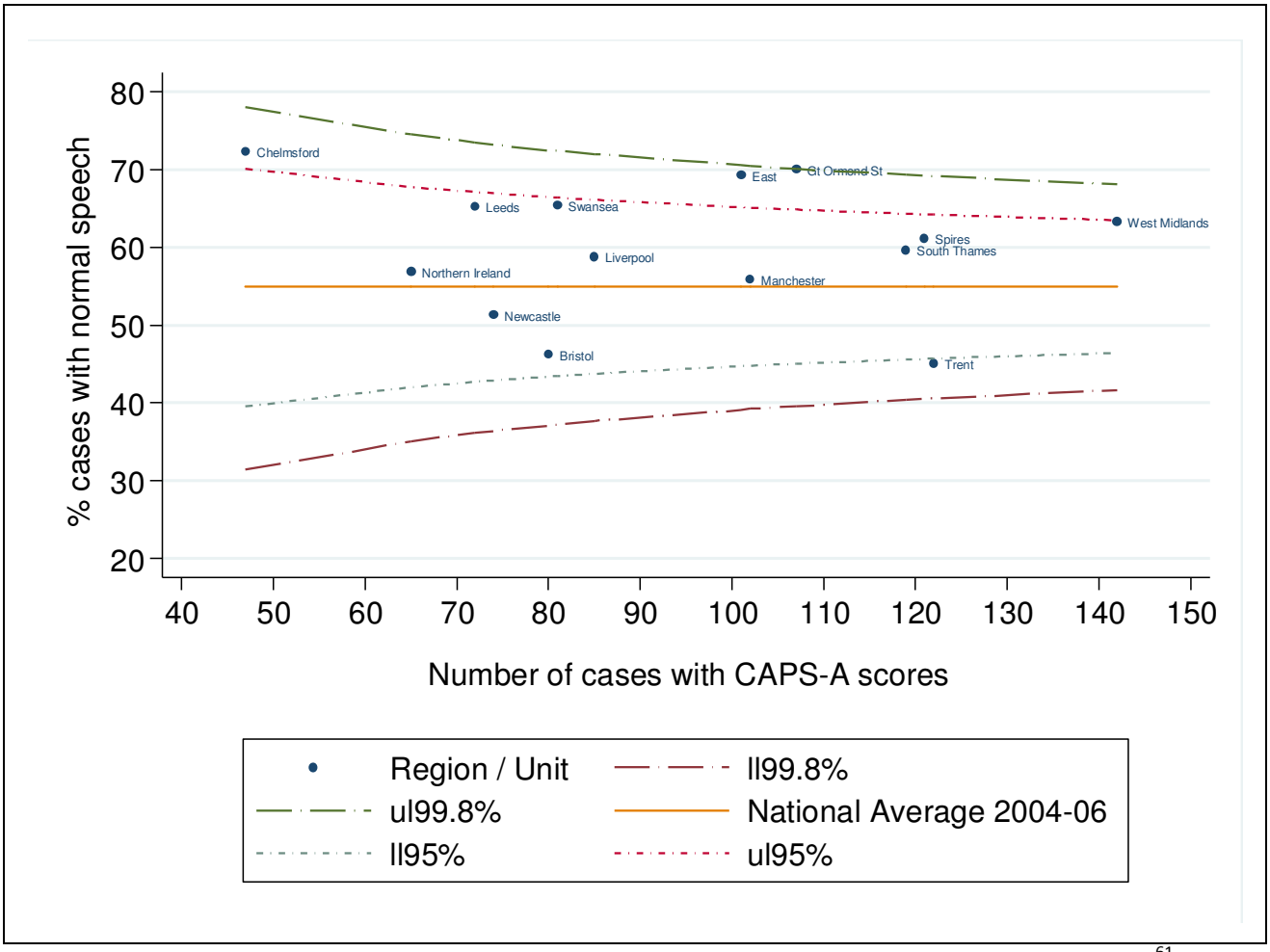
[58] Britton L, Albery L, Bowden M, Harding-Bell A, Phippen G, and Sell D(2014) A Cross-Sectional Cohort Study of Speech in Five-Year-Olds With Cleft Palate ± Lip to Support Development of National Audit Standards: Benchmarking Speech Standards in the United Kingdom. The Cleft Palate-Craniofacial Journal: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

[59] Based on the national outcome mean 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.

The funnel plot[60] in Figure 3 goes on to show the proportion of children (born in 2007-2009) achieving *normal speech* according to the number of auditable children at each region / 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). Figure 3 shows that rates of *normal speech*, for all regions / units, fall within the expected range given it is centred on the agreed national average of 55% 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 graphical representation of the data showing high levels of *normal speech* achieved at GOSH (70%) and lower levels of *normal speech* achieved in the Trent region (45%) – should be interpreted with caution. This is because there is wide variation in rates of missing data between regions/units (range of 2.2% to 22.5% – as detailed in Table 5 and Appendix 11)), and speech outcomes at 5 years of age are indicative of historical rather than current service provision.

Figure 3. Funnel plot of five-year olds (born between 2007 and 2009) with scores suggesting normal speech, according to the number of children at each region / unit with CAPS-A scores.



Note: Funnel plot centred on national average identified as the national outcome mean of 2004-06 speech outcome data[61] of 55%.

[60] 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 5 to 7. In addition, it is not adjusted (or risk adjusted) in any way.

[61] 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 above, presenting the data in Figure 3 as a funnel plot centred on the national average of 55%[62] is the most conservative method (at this time[63]) of checking whether or not any units deviate significantly from the expected standard (of 55%) and they do not.

3.6.6. Relationship between speech and facial growth (2007-2009 births)

In this section we describe analyses exploring the if any relationship between children's speech and facial growth outcomes exists for UCLP children. This is to establish whether or not there is a relationship between these children achieving good / normal speech outcomes and good or poor outcomes in terms of facial growth. Good outcomes for speech and facial growth have been defined as follows:

- Normal speech is achieved in cases where patients score 'normal' (green) scores across all 16 Cleft Audit Protocol for Speech – Augmented (CAPS-A) speech parameters (detailed further in section 3.6.5 on CAPS-A scores).
- Patients scoring '1' and '2' on the Five Year Old Index are considered to have the best possible outcome (good outcomes), while those scoring '4' and '5' are thought to have poor outcomes in terms of facial growth (detailed in section 3.5.4 on Five Year Old Index scores).

Table 8 shows that the proportion of children with good Five Year Index scores who have/have not achieved normal speech do not differ significantly from the proportion of children with poor scores (p=0.8). Overall, 30% (59/194) of consented children born between 2007 and 2009 had achieved scores indicating good facial growth and normal speech; while 18% (35/194) had scores indicating poor facial growth and not achieving normal speech.

Table 8. Number (%) of CRANE-registered[a] consented children born between 2007 and 2009[b], with good or poor Five Year Old Index scores at five years of age, by those achieving/not achieving normal speech.

Five Year Old Index scores	Normal Speech					
	Achieved		Not Achieved		Total	
	N	(%)	N	(%)	N	(%)
Good scores	59	(30)	67	(34.6)	126	(64.6)
Poor scores	33	(17.4)	35	(18)	68	(35.4)
Total	**92**	**(47.4)**	**102**	**(52.6)**	**194**	**(100)**

[a] Registered in CRANE by 29 December 2016.
[b] Excluding children who died before the age of 5 years, children with an incomplete UCLP, children with submucous cleft palates, syndromic children, children missing Five Year Old Index scores data, and children missing one or more of all 16 CAPS-A data items.

We also explored the relationship between facial growth and children's scores for the following 6 individual structurally related CAPS-A speech parameters – whose poor scores are indicative of structural issues of the palate / poor surgery:

1. Resonance: Hypernasality.
2. Nasal Airflow: Audible Nasal Emission.
3. Nasal Airflow: Nasal Turbulence.

[62] 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.
[63] No consensus has been reached on the factors that should be incorporated into an adjustment (or risk adjustment) of this data.

4. Passive CSCs: Weak and or nasalised consonants.
5. Passive CSCs: Nasal realisation of plosives.
6. Passive CSCs: Gliding of fricatives.

The proportion of children with good Five Year Index scores who had achieved good (green) scores for the above 6 CAPS-A speech parameters did not differ significantly from the proportion of children with poor (amber or red) CAPS-A scores.

Therefore there currently is no evidence of positive or negative correlation between facial growth and normal speech scores.

These initial findings should be interpreted with caution due to the small sample size. The analyses should be revisited in future with a larger sample. A larger sample will become available as the collection of the full 16 CAPS-A outcome scores (which started 3 years ago) expands for births after 2009; along with the yearly expansion of the collection of Five Year Index scores. In addition, as the data completeness of Five Year Old Index and CAPS-A scores continues to improve year-on-year (currently at 70.6% and 65.9% respectively – as detailed in Appendix 9); so will the sample size available for analyses.

4. National Pupil Database

The linkage of CRANE records to the National Pupil Database, at the individual level, has allowed us to explore educational achievement and special educational needs among children with a cleft lip and/or palate in England (see the 2014 and 2015 CRANE Annual Reports). In this section, we describe school absence rates among children with a cleft lip and/or palate in England during National Curriculum year 2, when children turn seven years of age and undergo their Key Stage (KS) 1 assessment. This is the first time that school absence has been examined in a cleft population.

4.1. Introduction

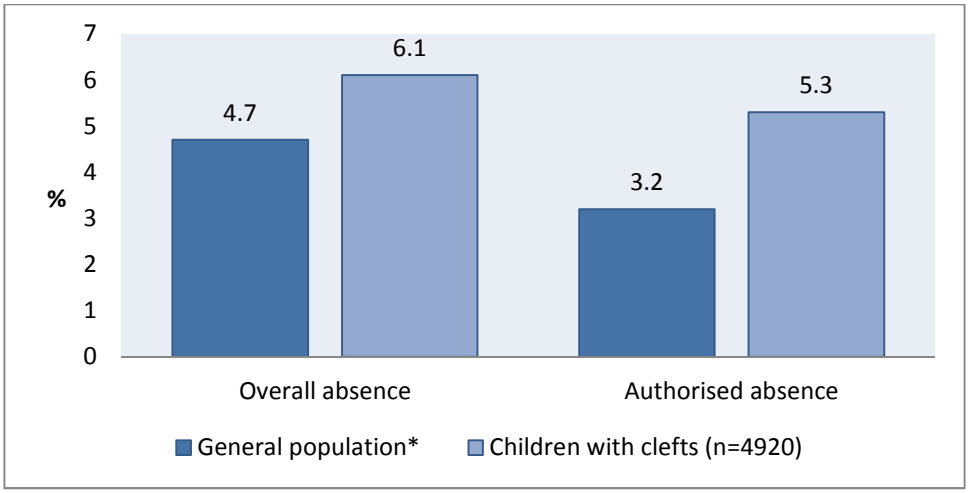
In order for children to fulfil their academic potential, they need to attend school regularly to benefit from their education. Missing lessons leaves pupils vulnerable to falling behind, and children with poor attendance tend to achieve less in both primary and secondary school[64,65,66,67]. As levels of pupil absences increase, the proportion of pupils reaching the expected levels of attainment at Key Stage 2 (age 11 years) and Key Stage 4 (age 16 years) decrease[68]. Our aim was to explore whether children with a cleft miss more school than their peers in the general population, and we sought to compare absence rates according to syndrome status and cleft type. For information on how the absence measures were calculated, please refer to the NPD section within the Methods chapter.

4.2. Absence among children with a cleft lip and/or palate in Year 2 (KS1) in England

There were 6,194 CRANE-registered children born from 2000 onwards who were matched to NPD records (87% linkage rate). Of these, 4,928 had KS1 assessments and were in Year 2 between 2006/07 and 2013/14. Among the 4,920 children who had absence data reported, 4,743 (96.4%) missed at least one school session in the academic year. This is higher than the 91% of the general population who miss at least one session per year[69].

[64] Department for Education. *The link between absence and attainment at KS2 and KS4. 2012/13 academic year*. London: Department for Education.2015.
[65] Arthurs N, Patterson J, Bentley A. Achievement for Students Who are Persistently Absent: Missing School, Missing Out? *The Urban Review,* 2014; 46(5):860-76. http://dx.doi.org/10.1007/s11256-014-0307-4
[66] Gottfried MA. The Detrimental Effects of Missing School: Evidence from Urban Siblings. *American Journal of Education,* 2011; 117(2):147-82. http://www.jstor.org/stable/10.1086/657886
[67] Hancock, K. J., Shepherd, C. C. J., Lawrence, D., & Zubrick, S. R. (2013). *Student attendance and educational outcomes: Every day counts*. Report for the Department of Education, Employment and Workplace Relations, Canberra.
[68] Department for Education. *A profile of pupil absence in England*. London: Department for Education.2011.
[6969] Department for Education. *Statistical First Release: pupil absence in schools in England, including pupil characteristics: 2011/12*. London: Department for Education. 2013.

Figure 4. Overall and authorised absence among children with a cleft in Year 2 compared with the general population in England.

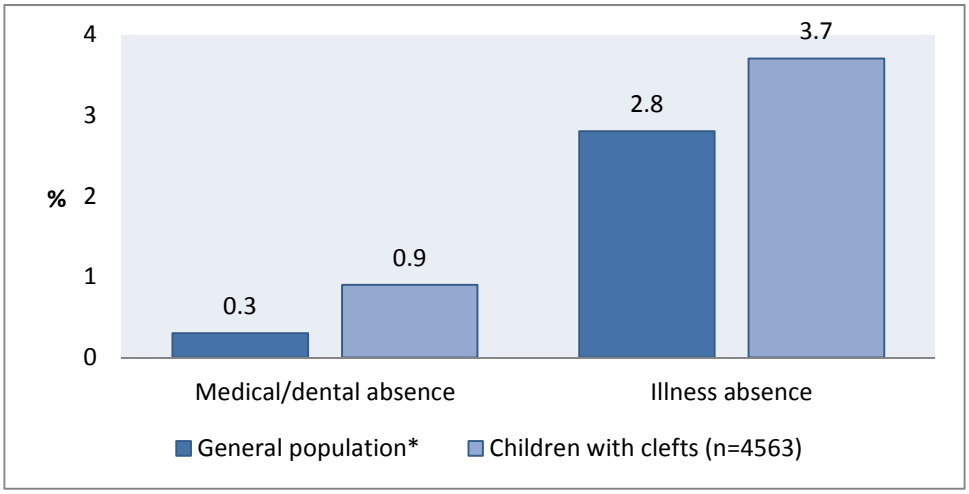


*General population data from 2010-2014.

The mean overall proportion of possible sessions missed during the academic year was 6.1% and the mean proportion of possible sessions taken as authorised absence was 5.3%. These rates are 1.3 and 1.7 times higher than the respective rates in the general population[70] (See Figure 4). 5.1% of Year 2 pupils with a cleft were classified as persistent absentees[71], which compares to 2.7% of Year 2 pupils in the general population.

Of the 4,563 (92.7%) children with absence broken down by reason[72], 3,049 (66.8%) missed at least one school session for medical or dental appointments. The mean percentage of all possible school sessions not attended because of medical or dental appointments was 0.9% and for illness it was 3.7%. These are higher than the corresponding figures for the general population (see Figure 5).

Figure 5. Reasons for absence among children with a cleft in Year 2 compared with the general population in England.



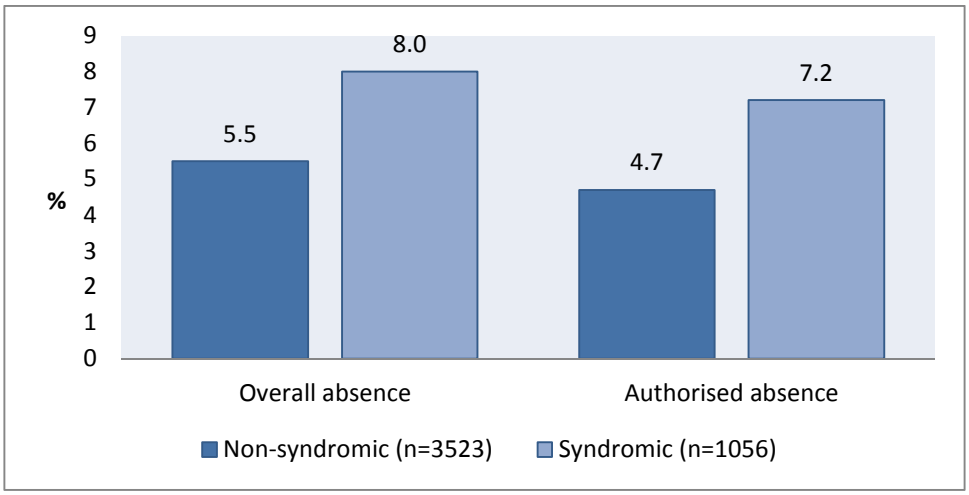
*General population data from 2010-2014.

[70] General population data available from 2010-2014.
[71] From 05/06 to 09/10 the persistent absence threshold was 20% of all possible sessions and from 10/11-14/15 is was 15%.
[72] Children with absence broken down by reason are those with >0 sessions missed for medical/dental appointments, or >0 sessions missed for illness. CRANE does not have access to other reasons for authorised absence.

4.3. Absence among children with a cleft lip and/or palate according to syndrome status

A total of 4,586 children (93.2%) linked to NPD at KS1/Year 2 were also successfully linked to HES records to verify their syndrome status. There were 3,527 (76.9%) children with a cleft alone and 1,059 (23.1%) with additional anomalies or syndromes. Absence data were available for 3,523 and 1,056 children, respectively.

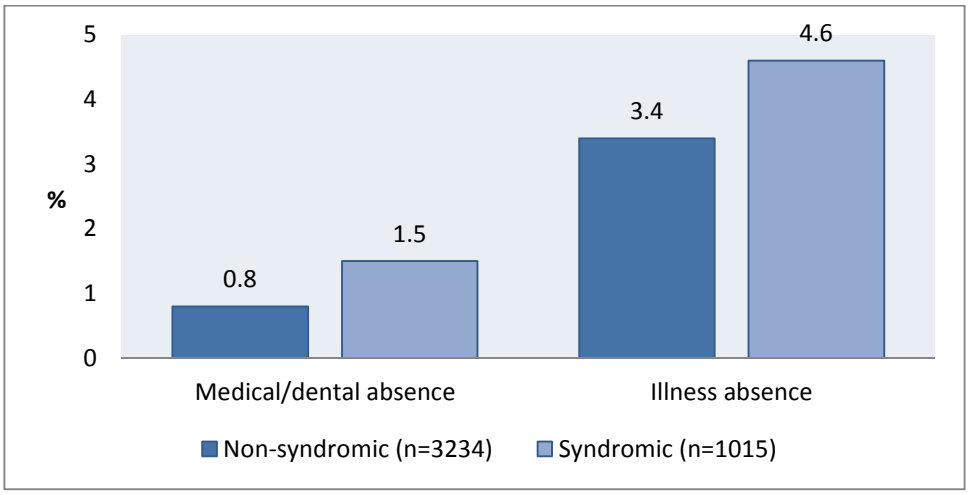
Figure 6. Overall and authorised absence among children with a cleft in Year 2, according to syndrome status.



Ninety-six percent of all non-syndromic children and 98.0% of those with a syndromic cleft missed at least one session during Year 2 (p=0.002 between groups). Compared with non-syndromic children, those with a syndromic cleft had significantly higher overall absence and authorised absence rates (p<0.0001) (see Figure 6). Furthermore, compare with non-syndromic children, a significantly greater proportion of children with syndromic clefts were identified as persistent absentees (9.5% vs. 3.6%, p<0.0001).

Of the 3,234 (91.8%) children with non-syndromic clefts and 1,015 (96.2%) children with syndromic clefts, for whom absence was broken down by reason, 63.2% and 78.6%, respectively, missed at least one school session for medical or dental appointments (p<0.0001). The difference between non-syndromic and syndromic groups in the proportion of possible sessions missed for medical and/or dental appointments and those missed for illness were statistically significant (p<0.0001) (see Figure 7).

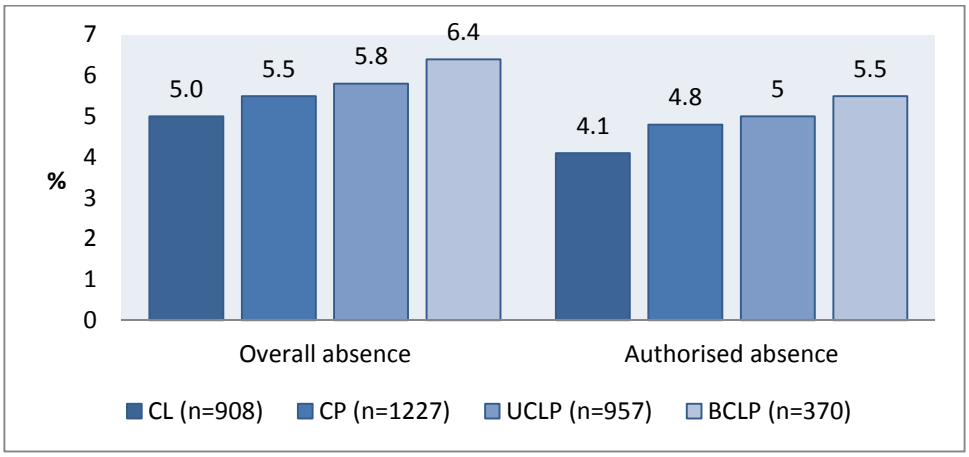
Figure 7. Reasons for absence among children with a cleft in Year 2, according to syndrome status.



4.4. Absence among children with a non-syndromic cleft lip and/or palate according to cleft type

We explored absence data according to cleft type among 3,462 children at KS1/Year 2 with a non-syndromic cleft and known cleft type. The proportion of children who were absent for at least one session in Year 2 varied from 94.7% in the cleft lip group to 98.1% in the BCLP group (p=0.017). There were also significant differences in overall absence (p=0.0001) and authorised absence (p<0.0001) rates between the four cleft type groups, with increasing rates with each increasing cleft type severity group (see Figure 8).

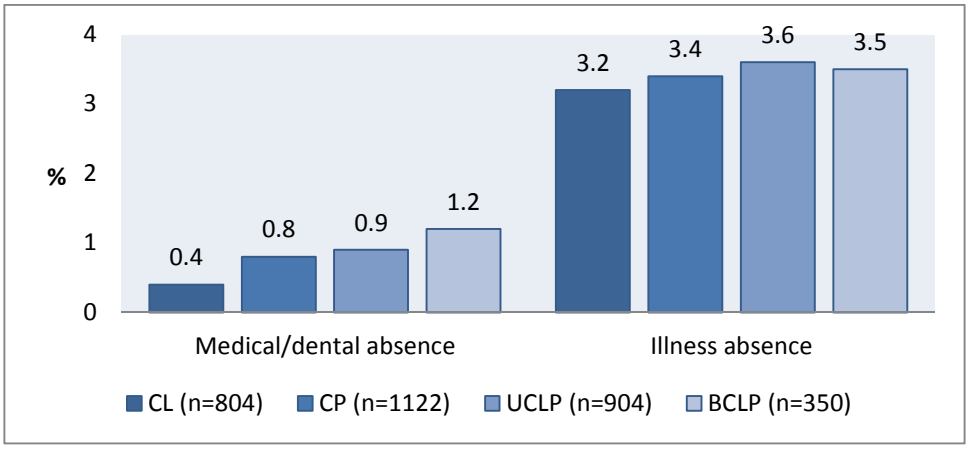
Figure 8. Overall and authorised absence among children with a cleft in Year 2, according to cleft type.



The proportion of children considered persistent absentees ranged from 3.2% among children with a cleft lip to 5.1% among those with a bilateral cleft lip and palate; however, the differences between groups were not statistically significant (p=0.370).

Absence broken down by reason was available for 3,180 children (91.6%) with non-syndromic clefts (see figure 9 for numbers by cleft type). The proportion of children missing at least one session for medical or dental appointments was 49.1% among pupils with a cleft lip, 64.0% in pupils with cleft palate, 69.0% in those with UCLP and 78.0% among those with a BCLP (p<0.0001). Figure 9 shows that the mean percentage of all possible school sessions not attended because of medical or dental appointments increased with each increasing severity of cleft type (p<0.0001). The differences between cleft groups in the percentage of sessions not attended because of illness were not statistically significant (p=0.1538).

Figure 9. Reasons for absence among children with a cleft in Year 2, according to cleft type.



4.5. Summary

For the first time, school absence has been explored among children with a cleft lip and/or palate in England. We found that children with a cleft have higher overall rates of absence, authorised absence, absence for medical or dental appointments, absence for illness, and persistent absenteeism than the general population. There were also significant differences in these rates between non-syndromic and syndromic cleft groups, with the latter taking more time off school. Finally, we found that there were significant trends for increasing overall absence, authorised absence and absence for medical or dental appointments with each increasing cleft type severity group among those without additional anomalies or syndromes.

These exploratory analyses have several strengths. First, we were able to achieve a high linkage rate (87%) between CRANE and the NPD. This means that the reported school absence data among children in Year 2 are likely to be representative of most children with a cleft attending maintained schools in England. Second, we were able to use general population data as a comparison to children with a cleft. Third, we were able to link to HES to explore absence rates according to syndrome status. This also allowed us to examine absence according to cleft type among children with non-syndromic clefts only, thus removing the potential influence of other health issues causing the difference in absence rates between cleft type subgroups. Finally, this is the first time that school absence has been reported among a cleft population, and these data indicate the potential impact that a cleft may have on school attendance for children around the age of seven years.

A limitation to this work is that CRANE requested data on two authorised absence reason categories out of a possible eight. Not all schools are able to provide absence data broken down by reason, so to identify whether a pupil had reason for absence reported, we classified those with >0 sessions missed for medical or dental appointments OR >0 sessions missed for illness as having reasons broken down. This method identified that 92.7% of the linked cleft cohort had absence broken down. Thus, the results of absence broken down by reason may slightly overestimate the proportion of children missing school for medical/dental appointments and illness as some children without any authorised absence or only absence for other reasons will have been excluded from our analyses. In the future we will request all eight categories to overcome this limitation.

Future analyses will examine school absence during other school years and also the impact of school absence on academic attainment within the cleft population.

5. Development of CRANE Database and future directions

5.1. Re-development of the CRANE Database and website

5.1.1. Timetable

As described in last year's report, in early 2015 the CRANE project team identified the need to re-develop the CRANE Database and Website IT systems. Crown Informatics Limited were appointed as system developer and the contract with Crown was finalised in August 2015. Development work by Crown took place from September 2015 and a User Acceptance Testing (UAT) site was made available by Crown to the Crane Project Team staff in order to test functionality and review progress with reference to the specified requirements documentation. Three rounds of user acceptance testing took place:

1. First UAT site made available to CRANE Project Team staff on 15 December 2015.
2. Second round of UAT by project team started 26 January 2016.
3. Third round, involving staff from cleft / administrative units, between 9 March 2016 and 22 March 2016. Feedback from unit staff was collated and passed on to Crown.

The old CRANE website was shut down on 6 May 2016, for an 18-day period during which time data was migrated from the old to the new site. Existing users were invited to pre-register for an account on the new site. The new website launched on 24 May 2016. The timing of the closedown was scheduled to maximise the time till the next data entry deadline. We identified that browser compatibility was likely to be an issue and took steps to highlight this to new users.

5.1.2. Operation since launch

The site has proved reliable being operational >99.9% of the time. Unplanned downtime has been minimal.

71 users have completed the registration process to end December 2016. . The majority of users are using MS IE9 and above, Firefox or Chrome. Some users initially used MS IE8 but have upgraded or changed browsers, which is encouraging. Some users continue to use MS IE8 which is not officially supported and does not provide the best user experience, so are advised to upgrade or use a different supported browser.

We are pleased to note that users appear to be using the new site without difficulty. User requests for help have been lower than expected and the majority of these could be resolved by directing users to the User Guides. The number of new patient registrations each month has been in line with expectations with a total of 764 new registrations in the period from launch to end December 2016

User feedback regarding the new site has generally been extremely positive. We encourage users to report any issues or difficulties with the site to the project team so that these can be investigated and addressed promptly.

5.1.3. Dataset since launch

The CRANE Database dataset has been refined where possible; and expanded where appropriate. Examples of key changes include:

- Since May 2016, in addition to standard CRANE consent, the **Patient Registration** section now captures data on parental consent to link CRANE data to health and education data.
- Additional outcomes are now collected at 5 and 10 years of age – specifically **Psychology at 5 years of age** and **Paediatric Dentistry at 10 years of age**.
- Prior to May 2016, the CRANE Database had the ability to capture **Speech outcomes at 10 years** of age – for future reporting (to be agreed with key stakeholder). After instructing the CRANE system developer to retain this feature (in late 2015), it came to CRANE's attention that there is some concern regarding the burden of collection and analysis of speech data at 10 years (Craniofacial Society of Great Britain and Ireland (CFSGBI) Meeting on 4 February 2016). Therefore the recording and reporting of this data will need to be re-considered with guidance from the CDG going forwards.

It is anticipated that the CRANE dataset will continue to evolve in collaboration with stakeholders. For example, there is a need to better define a way to capture information on Surgical Procedures and encourage recording of this data. Therefore, new data collection fields for Surgical Procedures must be agreed, specified and added as part of a future development package.

The CRANE Project Team will continue to engage with opportunities for positive listening to the professional groups served by CRANE, such as at key stakeholder meetings and consultations (E.g. CFSGBI meetings), to inform future developments of the CRANE Database and website.

5.1.4. Key features of the new site

The new site allows users to perform a number of functions for themselves that were previously undertaken by project team staff. These include:

- Transferring patients into or out of their unit.
- Deleting a record.
- Ability to import files containing outcome information for existing patients.

Other features include:

- Advanced searching/export.
- Improved validation checks.
- Live reports.
- Dedicated User Acceptance Testing site.
- Easier management of site content by project team staff.
- Improved management of user accounts

We would particularly encourage users to try the import facility as an alternative to editing individual records one at a time. Users may prepare files containing outcomes information offline and upload these

to the site. This enables a number of records to be updated at once. Template files specifying the format required for each outcome are provided on the site.

In addition, short guides for individual features have been created for users and provided via the new website on the following: Exports, Imports and Duplicates (in the Help section of - https://www.crane-database.org.uk/).

5.1.5. Future developments

The development and introduction of data downloads and real time reports accessible to teams through the web based platform for the database has led us to consider what we report annually from the CRANE project team. Originally we were contracted to deliver a written annual report and a progress report 6 months apart. This concept was based on delivery before real time reporting was available and we have also developed other methods of communication with teams and stakeholders in recent years. These include patient and parent friendly reports of key data and eNewsletters to clinical teams and managers.

In light of these developments we are proposing to reduce the quantity and length of reporting undertaken. The project aims to produce the following as alternatives to these written reports:

- Real time reporting of delayed CP diagnosis available on the website to the public.
- Access to teams to funnel plots of key outcome data.
- Access to teams to demographic data regarding births in England, Wales and Northern Ireland.

This will allow us to devote more of the project team's time into analysis of data for commissioners and teams and subsequent submission of peer reviewed publications. We aim to review the impact of these changes after 12 months with feedback from both clinical teams and commissioners.

5.2. Consent form and patient information leaflet

Informed by feedback and input provided by cleft teams in 2016, we produced separate information English language leaflets (1) for the Database and (2) for Data Linkage, and a revised consent form. These were reviewed by the Confidentiality Advisory Group (CAG); the Cleft Lip And Palate Association (CLAPA); the plain English Campaign; and signed off by the CDG in May 2016 – before being hosted on the new CRANE Website.

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 [73]. Analysis including data on this timing was not conducted for 2015 births (for this Annual Report) as only small numbers were recorded using the '≤72 hours' timing (n=47, 4.7%)[74]. We anticipate being able to report this in future.

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 the third 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[75]:

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[76] to evaluate patient (and parent) reported experience. These measures were combined into one Patient (and Parent) Reported Experience Measure (PREM) questionnaire and, through piloting[77] by the Cleft Psychology CEN, adjusted to apply to cleft services.

[73] 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).

[74] 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.

[75] 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.

[76] (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).

[77] 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.

The CRANE project team and the Cleft Psychology CEN are collaborating with a 12-month feasibility study – running until the end of January 2017 – to test PREM data collection, analysis and reporting, with a view to developing a method to implement data collection, analysis and reporting nationally.

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 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 has very recently received an updated HES extract – containing hospital data for 2014 & 2015. This will allow us to refresh our analyses involving HES data, such as those involving the NPD, mortality, secondary speech surgery, and grommets. We will apply to NHS Digital to link these recent HES Data to CRANE in order to include in our analyses those births that have been registered in CRANE since the last linkage exercise to HES which was conducted in March 2013.

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)[78] 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.

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

5.4.4. 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.

5.4.5. 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 units. 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, was 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 units 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 have submitted data for the 2015/16 quality dashboards, as well as started to do so for the 2016/17 quality dashboards. 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[79]) – 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 (future dates have yet to be agreed).

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 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 linkage of CRANE data with data collected by the Cleft Collective.

5.8. CRANE Communications

5.8.1. Dissemination of 2016 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.

[79] 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.

- We will also work with our close collaborators – such as the CFSGBI and CLAPA – to expand the reach of our eNewsletter (and the report).
- A *Summary of Findings for Patients and Parents/Carers* from this 2016 Annual Report will be produced, with the aim of publishing it in the summer of 2017.

5.8.2. Publications and presentations related to the CRANE Database

Publication(s)

Kate J. Fitzsimons, Lynn P. Copley, Jan H. van der Meulen, Channa Panagamuwa, Scott A. Deacon, Grommet Surgery in Children With Orofacial Clefts in England, The Cleft Palate-Craniofacial Journal. 2017;54(1):80-89. doi: http://dx.doi.org/10.1597/15-047

Oral presentation(s)

Setakis, E., Fitzsimons, K., Copley, L.,, Deacon S., Gilbert, R., and van der Meulen, J. "Using postcode histories when linking cohorts to the National Pupil Database (NPD)" Administrative Data Research Network, London. (3 June 2016).

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 1,098 children born with a cleft in 2015 had been registered on CRANE at the time of preparing this report. This equates to an incidence of approximately one in every 639 live births in England, Wales and Northern Ireland[80].

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 quarter of the children born in 2015 had not been approached for consent at the time of preparing this report[81]. 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 up to 88.9%[82] 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 recent re-development of the CRANE Database and Website in 2015/16 is expected to play a key role in facilitating this (see Chapter 5 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 continue to 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

[80] 24,215 Births in NI in 2015 – Available from Northern Ireland Statistics & Research Agency (NISRA): http://www.nisra.gov.uk/demography/default.asp8.htm. // 697,852 Births in England & Wales in 2015 – Available from the Office for National Statistics. Characteristics of Birth 1, England and Wales: https://www.ons.gov.uk/peoplepopulationandcommunity/birthsdeathsandmarriages/livebirths/datasets/birthsummarytables

[81] See Appendix 6 for further detail.

[82] See Appendix 9 for further detail.

Anomaly Screening Programme target detection rate of 75%[83].

2. Less than a third of children with a cleft palate alone (28.3%) 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[84]. This proportion has dropped since last year (was 31.3%).

3. 81.3% of the children born in 2015 with a cleft were referred by a maternity unit to an Cleft Unit within 24 hours of birth. This proportion varied significantly according to the unit receiving the referrals (ranging from 66.2% to 91.5%). These high rates are consistent with those reported this time last year; and although rates continue to improve year-on-year, 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 97.4% of parents within 24 hours of their child's referral; with no statistically significant difference between units receiving the referrals. In fact, all units contacted at least 93.3% of their patients within 24-hour of receiving the referral. This is positive and demonstrates the commitment of units to ensure timely response to new referrals of babies born with a cleft, to help support these babies and their families in the important initial stages.

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 74.8% and 67.1% respectively) across units indicate that, in the majority of cases, units have mechanisms in place to deal with any dental disease occurring.

6. Approximately one quarter of children with a complete UCLP have poor dental arch relationships (24.6%) 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[85].

7. Almost three quarters of children (59.8%) with a complete speech assessment had speech scores that would suggest their speech is not significantly different from their non-cleft peer group, with scores suggesting they have achieved *normal speech*. This means that the national speech outcome standard #1 target of 55%[86] was not only met, but it was exceeded (on average) by the CRANE cohort born in 2007-2009.

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[83] 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.

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

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

[86] Based on the national outcome mean 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.

Educational achievement at five and seven years

6,194 CRANE-registered children born from 2000 onwards were matched to NPD records. Of these, 4,928 had KS1 assessments and were in Year 2 between 2006/07 and 2013/14, and 4,920 had absence data reported. Using this NPD and CRANE-HES-linked dataset, we examined school absence at seven years of age among children with a cleft in England. This is the first time that school absence has been reported among a cleft population, and these data indicate the potential impact that a cleft may have on school attendance for children around the age of seven years.

The key findings are outlined below:

8. Children with a cleft have higher overall rates of absence, authorised absence, absence for medical or dental appointments, absence for illness, and persistent absenteeism than the general population.

9. There were also significant differences in these rates between non-syndromic and syndromic cleft groups, with the latter taking more time off school.

10. Finally, we found that there were significant trends for increasing overall absence, authorised absence and absence for medical or dental appointments with each increasing cleft type severity group among those without additional anomalies or syndromes.

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: Members of the Cleft Development Group

Members of the Cleft Development Group (CDG)

Simon van Eeden	Chair, CDG & Clinical Lead, North West, IoM & North Wales Cleft Network
Elizabeth Adamson	Joint Representative for CDs and Managers Group
Victoria Beale	Consultant Cleft & Maxillofacial Surgeon
Lorraine Britton	Lead Speech and Language Therapist, Trent Regional Cleft Lip & Palate Service
Alec Cash	Clinical Lead, South Thames Cleft Service
Chris Coslett	Specialised Planner, Women and Children's Services, Welsh Health Specialised Services
Sinead Davis	Chair, CEN for Cleft ENT and Hearing and Consultant ENT Surgeon
Scott Deacon	CRANE Clinical Project Leader
Mark Devlin	Lead Clinician, Scottish Cleft Surgical Service
David Drake	Cleft Surgery Training Interface Group
Yvette Edwards	Joint Representative of CDs and Managers Group
Toby Gillgrass	Lead Clinician of Cleft Care Scotland
Per Hall	Cleft Surgeon (BAPRAS) and Consultant Cleft Surgeon, CleftNetEast
Norman Hay	Clinical Lead, North Thames Cleft Service
Chris Hill	Northern Ireland Clinicians
Peter Hodgkinson	President, Craniofacial Society, Clinical Lead, Newcastle Site, Northern and Yorkshire Cleft Service & Chair Cleft Centres
Nichola Hudson	Lead Clinical Nurse Specialist
David Landes	Public Health Consultant
Karine Latter	Nursing deputy for Nichola Hudson
Kate le Marechal	Clinical Psychologists CEN
Sian Lewis	Acting Medical Director - Welsh Health Specialised Services Committee
Jason Neil-Dwyer	Clinical Director, Trent Cleft Service
David Orr	Cleft Services in the Republic of Ireland
Susan Parekh	Paediatric Dentistry CEN
Marie Pinkstone	Lead Speech & Language Therapists
Sandip Popat	Restorative Dentistry CEN
Stephen Robinson	Outgoing Chair, CDG & Clinical Director Spires Cleft Service
Jonathan Sandy	Lead, Cleft Collective Birth Cohort and Gene Bank Study
Ian Sharp	Clinical Director, West Midlands Cleft Centre
Bill Shaw	Lead at Manchester Clinical Trials Centre
Jackie Smallridge	Consultant Paediatric Dentist, CleftNetEast
Alistair Smyth	Cleft Surgeon (BAOMS)
David Steel	Chair Programme Director, National Services Division, NHS Scotland
David Stokes	CLAPA Chief Executive
Adrian Sugar	Chair, CDG & Clinical Lead, North West, IoM & North Wales Cleft Network
Jan van der Meulen	Senior Epidemiologist, Clinical Effectiveness Unit
Jennifer Williams	Deputy for Per Hall and Lead Clinical Nurse Specialist, CleftNetEast
Mike Winter	Medical Director, National Services Division, Scotland

Appendix 3: NHS UK Cleft Development Group – Terms of Reference

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 (the Craniofacial Society Anomalies Register) and then CRANE (Cleft Registry and Audit Network). 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 Implementation 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 Implementation Monitoring Group which were 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 the funding of the CRANE Register and is responsible for ensuring that there is a contact in place with NHS England and that monies are paid annually to the hosting Clinical Effectiveness Unit at the Royal College of Surgeons. 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 4 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 and demographic 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, and 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 member's ex-officio who will be members during their terms of that office whether it is 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. The Group may decide to re-elect the holders or extend the period of office.

The composition will be:

1. Commissioners of Cleft Care. These should include a commissioner from NHS England, one from Wales, one from Scotland and one from Northern Ireland (each nominated by their equivalent national specialist commissioning body).
2. Public Health Consultants. These should include a representative of commissioning areas who are actively involved in cleft commissioning, and will normally be Consultants in Dental Public Health.
3. A Lay representative from a Parent Support Group (1) (to be nominated by CLAPA).
4. Cleft surgeons (2) (nominated by the surgical CEN).
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 Clinical Nurse Specialist (1) (to be nominated by the lead 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 Service Manager/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 one or more 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. paediatricians, anaesthetists and 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 October 2016

Appendix 4: 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, the Wales Specialised Health Services Committee, and the Northern Ireland Specialist Services Commissioning Team. 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 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: Consent status detail

Number (%) of children born in 2015 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 (93)	1 (1.8)	0 (0)	3 (5.3)	57
& Yorkshire	Leeds	70 (100)	0 (0)	0 (0)	0 (0)	70
North West	Liverpool	60 (80)	1 (1.3)	14 (18.7)	0 (0)	75
& North Wales	Manchester	59 (84.3)	0 (0)	10 (14.3)	1 (1.4)	70
Trent	Nottingham	80 (88.9)	1 (1.1)	9 (10)	0 (0)	90
West Midlands	Birmingham	78 (64.5)	4 (3.3)	36 (29.8)	3 (2.5)	121
East	Cambridge	54 (62.1)	2 (2.3)	29 (33.3)	2 (2.3)	87
North Thames	Great Ormond St	71 (78.9)	1 (1.1)	18 (20)	0 (0)	90
	Chelmsford	40 (93)	0 (0)	2 (4.7)	1 (2.3)	43
The Spires	Oxford & Salisbury	83 (85.6)	3 (3.1)	11 (11.3)	0 (0)	97
South Wales	Swansea	31 (73.8)	0 (0)	10 (23.8)	1 (2.4)	42
& South West	Bristol	61 (80.3)	0 (0)	14 (18.4)	1 (1.3)	76
South Thames	GSTT	43 (29.3)	2 (1.4)	100 (68)	2 (1.4)	147
Northern Ireland	Belfast	32 (97)	0 (0)	1 (3)	0 (0)	33
All	**All**	815 (74.2)	15 (1.4)	254 (23.1)	14 (1.3)	1,098

[a] Registered in CRANE by 29 December 2016. Note: MCN – Managed Clinical Network.

Appendix 7: Number of registrations detail

Table A. Number of children born between 2006 and 2015 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)										
		2006	2007	2008	2009	2010	2011	2012	2013	2014	2015	All
Northern & Yorkshire	Newcastle	67	91	71	64	63	65	66	74	58	57	676
	Leeds	75	71	70	65	70	73	65	69	67	70	695
North West	Liverpool	55	63	86	79	86	63	63	73	88	75	731
& North Wales	Manchester	105	89	85	68	90	82	70	73	74	70	806
Trent	Nottingham	94	85	107	96	103	106	95	99	87	90	962
West Midlands	Birmingham	121	100	123	118	108	113	129	120	121	121	1174
East	Cambridge	79	83	82	84	81	66	88	92	57	87	799
North Thames	Great Ormond Street	95	129	153	106	89	112	135	109	102	90	1120
	Chelmsford	28	46	38	46	38	54	43	45	51	43	432
The Spires	Oxford & Salisbury	115	125	99	89	96	111	99	98	79	97	1008
South Wales	Swansea	45	48	44	47	45	53	51	37	38	42	450
& South West	Bristol	60	61	72	59	78	55	69	91	59	76	680
South Thames	GSTT	102	112	107	85	69	81	150	120	130	147	1103
Northern Ireland	Belfast	44	42	37	36	38	41	33	47	37	33	388
All	**All**	1,085	1,145	1,174	1,042	1,054	1,075	1,156	1,147	1,048	1,098	11,024

Table B. Number (%) of children born between 2006 and 2015 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	2006		2007		2008		2009		2010		2011		2012		2013		2014		2015		All	
Cleft lip	238	(23.2)	266	(23.7)	274	(24.2)	216	(21.3)	249	(24.2)	251	(24.5)	250	(23)	293	(26.4)	248	(25.1)	254	(25.2)	2539	(24.1)
Cleft palate	472	(45.9)	493	(43.9)	527	(46.6)	474	(46.7)	455	(44.2)	464	(45.3)	486	(44.6)	491	(44.3)	435	(43.9)	441	(43.8)	4738	(44.9)
UCLP	221	(21.5)	247	(22)	242	(21.4)	215	(21.2)	215	(20.9)	222	(21.7)	254	(23.3)	223	(20.1)	206	(20.8)	224	(22.2)	2269	(21.5)
BCLP	97	(9.4)	117	(10.4)	88	(7.8)	111	(10.9)	110	(10.7)	87	(8.5)	99	(9.1)	102	(9.2)	101	(10.2)	88	(8.7)	1000	(9.5)
Not specified	57	-	22	-	43	-	26	-	25	-	51	-	67	-	38	-	58	-	91	-	478	-
All	1,085	(100)	1,145	(100)	1,174	(100)	1,042	(100)	1,054	(100)	1,075	(100)	1,156	(100)	1,147	(100)	1,048	(100)	1,098	(100)	11,024	(100)

[a] Registered in CRANE by 29 December 2016. Note: MCN – Managed Clinical Network. CL - Cleft Lip, CP - Cleft Palate, UCLP - Unilateral cleft lip and palate, and BCLP - Bilateral cleft lip and palate.

Appendix 8: Timing of diagnosis detail

Number (%) of CRANE-registered children born in 2015 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	168	(68.3)	62 (25.2)	7 (2.8)	2 (0.8)	6 (2.4)	1 (0.4)	246
CP	2	(0.5)	287 (71.2)	67 (16.6)	19 (4.7)	23 (5.7)	5 (1.2)	403
UCLP	187	(87)	28 (13)	0 (0)	0 (0)	0 (0)	0 (0)	215
BCLP	71	(85.5)	11 (13.3)	1 (1.2)	0 (0)	0 (0)	0 (0)	83
Not specified	16	(30.8)	24 (46.2)	2 (3.8)	2 (3.8)	5 (9.6)	3 (5.8)	52
All	444	(44.4)	412 (41.2)	77 (7.7)	23 (2.3)	34 (3.4)	9 (0.9)	999

[a] 99/1,098 (9%) 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[87]. With only small numbers having been recorded using this timing (n=47, 4.7%), we report '≤72 hours' cases within the '≤1 week' timing (until recording of this timing is well established).

[87] 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).

Appendix 9: Reported five-year outcomes for children born with a cleft lip and/or palate detail

Number (%) of CRANE-registered [a] consented children born between 2004 and 2010[b] with reported outcomes at five years of age (excluding children with submucous cleft palates)[88], according to region / unit.

Regional centre / MCN	Administrative Unit		Weight		Height		dmft			5 Year Old Index			Speech		
			2004-2010 births[c]				2004-2009 births[d]			2004-2009 births[e]			2007-2009 births[f]		
			Reported		Reported		Reported			Reported[89]			Reported		
		N	n	(%)	n	(%)	N	n	(%)	N	n	(%)	N	n	(%)
Northern & Yorkshire	Newcastle	437	318	(72.8)	315	(72.1)	380	296	(77.9)	48	35	(72.9)	100	74	(74)
	Leeds	443	349	(78.8)	358	(80.8)	377	262	(69.5)	57	48	(84.2)	114	72	(63.2)
North West	Liverpool	448	66	(14.7)	66	(14.7)	369	275	(74.5)	72	48	(66.7)	158	85	(53.8)
	Manchester	505	222	(44)	221	(43.8)	418	318	(76.1)	57	46	(80.7)	146	102	(69.9)
Trent	Nottingham	621	76	(12.2)	77	(12.4)	527	114	(21.6)	84	45	(53.6)	192	122	(63.5)
West Midlands	Birmingham	699	357	(51.1)	344	(49.2)	600	523	(87.2)	112	95	(84.8)	192	142	(74)
East	Cambridge	450	132	(29.3)	118	(26.2)	387	72	(18.6)	84	49	(58.3)	163	101	(62)
North Thames	Great Ormond Street	535	170	(31.8)	165	(30.8)	450	232	(51.6)	66	44	(66.7)	191	107	(56)
	Chelmsford	235	85	(36.2)	84	(35.7)	197	93	(47.2)	26	18	(69.2)	70	47	(67.1)
The Spires	Oxford & Salisbury	612	64	(10.5)	64	(10.5)	525	358	(68.2)	99	88	(88.9)	184	121	(65.8)
South Wales & South West	Swansea	304	180	(59.2)	118	(38.8)	260	207	(79.6)	31	11	(35.5)	89	81	(91)
	Bristol	412	153	(37.1)	155	(37.6)	335	193	(57.6)	46	31	(67.4)	121	80	(66.1)
South Thames	Guy's and St Thomas'	596	179	(30)	164	(27.5)	528	272	(51.5)	105	90	(85.7)	199	119	(59.8)
Northern Ireland	Belfast	236	2	(0.8)	2	(0.8)	200	117	(58.5)	37	4	(10.8)	81	65	(80.2)
All	**All**	6,533	2,353	(36)	2,251	(34.5)	5,553	3,332	(60)	924	652	(70.6)	2,000	1,318	(65.9)

[a] Registered in CRANE by 29 December 2016. **Note:** MCN – Managed Clinical Network.

[b] Exclusions from all five year outcomes / all measures presented in this table: Submucous cleft palate patients as all/most teams do not audit these patients. Patients that died before 5yrs (113/6,646 (1.7%)). Plus, cases fitting the exclusion criteria detailed below; which are **not mutually exclusive**:

[c] Weight and height data for children born in 2004-2010: 221/6,754 (3.3%) cases with submucous cleft palates are excluded.

[d] dmft data for children born in 2004-2009: 206/5,759 (3.6%) cases with submucous cleft palates are excluded.

[e] 5 Year Old Index scores for children born in 2004-2009: 342/1,266 (27%) cases with incomplete unilateral cleft lip & palate (UCLP) are excluded.

[f] Speech data for children born in 2007-2009: 1,000/3,000 (33.3%) children with submucous cleft palates (2.7%), missing one or more of all 16 CAPS-A data items (1.3%), born with either a CL (23.1%) or a non-specified cleft type (1.6%), and syndromic children (6.8%%) are excluded from speech data. Details of additional excluded cases can be found in Table 5.

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

[89] Only 5/14 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 (4-95) 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.

Appendix 10: Five Year Old Index scores detail

Number (%) of CRANE-registered consented children born between 2004 and 2009 with a complete unilateral cleft lip and palate[90], 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	6 (17.1)	5 (14.3)	15 (42.9)	6 (17.1)	3 (8.6)	35
	Leeds	6 (12.5)	19 (39.6)	13 (27.1)	9 (18.8)	1 (2.1)	48
North West & North Wales	Liverpool	4 (8.3)	15 (31.3)	15 (31.3)	10 (20.8)	4 (8.3)	48
	Manchester	2 (4.3)	16 (34.8)	15 (32.6)	8 (17.4)	5 (10.9)	46
Trent	Nottingham	3 (6.7)	19 (42.2)	14 (31.1)	4 (8.9)	5 (11.1)	45
West Midlands	Birmingham	8 (8.4)	33 (34.7)	28 (29.5)	21 (22.1)	5 (5.3)	9
East	Cambridge	3 (6.1)	17 (34.7)	16 (32.7)	11 (22.4)	2 (4.1)	4
North Thames	Great Ormond Street	3 (6.8)	21 (47.7)	12 (27.3)	8 (18.2)	0 (0)	44
	Chelmsford	2 (11.1)	7 (38.9)	7 (38.9)	2 (11.1)	0 (0)	18
The Spires	Oxford & Salisbury	9 (10.2)	32 (36.4)	25 (28.4)	18 (20.5)	4 (4.5)	88
South Wales & South West	Swansea	0 (0)	2 (18.2)	5 (45.5)	2 (18.2)	2 (18.2)	11
	Bristol	1 (3.2)	14 (45.2)	5 (16.1)	8 (25.8)	3 (9.7)	31
South Thames	Guy's and St Thomas'	9 (10)	34 (37.8)	29 (32.2)	11 (12.2)	7 (7.8)	90
Northern Ireland	Belfast	0 (0)	1 (25)	2 (50)	1 (25)	0 (0)	4
All	All	56 (8.6)	235 (36)	201 (30.8)	119 (18.3)	41 (6.3)	652

[a] Exclusions from 'All' values (not mutually exclusive): 342/1,266 (27%) children with an incomplete UCLP, 11/935 (1.2%) children who died before the age of five, and 272/924 (29.4%) children missing Five Year Old Index scores data.

Note: MCN – Managed Clinical Network.

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

Appendix 11: Cleft Audit Protocol for Speech – Augmented scores detail

Number (%) of CRANE-registered[a] consented children born with a cleft palate in 2007-2009, 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		Reason outcome not collected		Total cases acc. for	Missing Data	
		N	n	(%)	n	(%)	(%)	n	(%)
Northern	Newcastle	100	74	(74)	12	(12)	(86)	14	(14)
& Yorkshire	Leeds	114	72	(63.2)	39	(34.2)	(97.4)	3	(2.6)
North West	Liverpool	158	85	(53.8)	64	(40.5)	(94.3)	9	(5.7)
& North Wales	Manchester	146	102	(69.9)	37	(25.3)	(95.2)	7	(4.8)
Trent	Nottingham	192	122	(63.5)	50	(26)	(89.6)	20	(10.4)
West Midlands	Birmingham	192	142	(74)	19	(9.9)	(83.9)	31	(16.1)
East	Cambridge	163	101	(62)	50	(30.7)	(92.6)	12	(7.4)
North Thames	Great Ormond Street	191	107	(56)	41	(21.5)	(77.5)	43	(22.5)
	Chelmsford	70	47	(67.1)	12	(17.1)	(84.3)	11	(15.7)
The Spires	Oxford & Salisbury	184	121	(65.8)	32	(17.4)	(83.2)	31	(16.8)
South Wales	Swansea	89	81	(91)	6	(6.7)	(97.8)	2	(2.2)
& South West	Bristol	121	80	(66.1)	19	(15.7)	(81.8)	22	(18.2)
South Thames	Guy's and St Thomas'	199	119	(59.8)	66	(33.2)	(93)	14	(7)
Northern Ireland	Belfast	81	65	(80.2)	8	(9.9)	(90.1)	8	(9.9)
All	**All**	2,000	1,318	(65.9)	455	(22.8)	(88.7)	227	(11.4)

[a] Registered in CRANE by 29 December 2016. Note: MCN - Managed Clinical Network.

[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.