CRANE Database

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Annual Report on Cleft Lip and/or Palate 2014

On behalf of the Cleft Development Group

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Abbreviations

BCLP	Bilateral cleft lip and palate
BINOCAR	British Isles Network of Congenital Anomaly Registers
CAPS-A	Cleft Audit Protocol for Speech—Augmented
CARE	Craniofacial Anomalies Register
CDG	Cleft Development Group
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
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
MCN	Managed Clinical Network
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
SIG	Special Interest Group
UCLP	Unilateral cleft lip and plate
VTCT	Vocational Training Charitable Trust
WHO	World Health Organization

Glossary

Alveolus / alveolar	The part of the jaw that supports the teeth and contains the tooth sockets.
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.
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 an Administrative 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.
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
Hospital Episode Statistics (HES)	A national database containing records on all admissions to NHS hospitals in England.
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.
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/
Patient Episode Data Wales (PEDW)	A national database containing records on all admissions to hospitals in Wales.
Submucous Cleft Palate	The term submucous refers to the fact that the cleft is covered over by the lining (mucous membrane) of the roof of the mouth. This covering of mucosa makes the cleft difficult to see when looking in the mouth.

Executive summary

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

The 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 15 hospitals (otherwise known as Administrative 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 2013 in England, Wales and Northern Ireland. We examine trends in: Registrations and the timing of cleft diagnosis; patient referrals to Administrative Units; and first contact between Administrative Units and the parents of children born with a cleft.

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

- Information on cleft-related outcomes for children at five years of age (born 2004-2008). The completeness of these data, which is essential for CRANE to perform meaningful analyses, is presented according to Administrative Unit.
- Analyses of data from Hospital Episode Statistics (HES), a database containing records on all NHS hospital admissions in England. These data are used to derive information on children diagnosed with cleft lip and/or palate. This year we have examined instances of additional surgeries involving the palate, needed to improve speech or to close any residual fistulae, following primary cleft palate repairs. This was among children born between 1997 and 2004 in England; examined up until the age of 7 years. We present the results according to the presence and absence of additional anomalies or

[1] Stanier, P and Moore, G, Genetics of cleft lip and palate: syndromic genes contribute to the incidence of non-syndromic clefts. Human Molecular Genetics, 2004. 13: p. R73-R81.
[2] Mossey, PA, Little, J, Munger, RG, Dixon, MJ and Shaw, WC, Cleft lip and palate. The Lancet, 2009. 374(9703): p. 1773-1785.

syndromes, by cleft type classification, their age at the time of their surgeries, and by region where they underwent their procedure.

- 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 school. This data was linked to CRANE data for consented children. We describe the results of the linkage exercise, and make some preliminary comparisons between the educational outcomes at age 5 for the cleft cohort and the published national statistics for all children at the age of 5; based on assessments conducted at the end of their first year of school (at the end of reception) – known as the Early Years Foundation Stage Profile (EYFSP).

Key findings

Children born with a cleft lip and/or palate in 2013

Overall, 14,241 children born between 1 January 2000 and 31 December 2013 with a cleft lip and/or palate were registered on the CRANE Database by 19 September 2014; reflecting all children born with a cleft lip and/or palate, referred to one of the 15 Administrative Units in England, Wales and Northern Ireland, regardless of their consent status. Of these children, 1,121 were born in 2013; with CRANE receiving the highest number of registrations for births in 2012 and 2013 since 2008. This increase in registrations could be attributed to the registration of children before the verification of consent; and the improved function of the database as a national register of cleft births.

CRANE case ascertainment is very high, being around 95%, according to comparisons with HES and Patient Episode Data Wales (PEDW)[3]. Out of the 895 children born in 2013 whose consent status had been verified, the parental consent rate was 98.4% (ranging from 93.8% to 100% between Units). Out of all children registered, 20.2% (ranging from 0% to 69.2% between Units) had not had their consent status verified by the Administrative Unit at the time of writing this report.

Among children born in 2013, CRANE analyses revealed:

- 41.6% of all children with clefts and 63.3% 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 1.6% of children with cleft palate only (CP) were diagnosed during antenatal screening; 68% were diagnosed at birth, leaving 30.4% who were diagnosed late according to the national standard[4]. The proportion of children with CP diagnosed late has decreased by 1.7% compared with last year. Five and

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

a half percent of children with a cleft palate alone are diagnosed after one month of age. This proportion has reduced by 1.4% since last year.

- 81.9% of children were referred by a maternity unit to an Administrative Unit within 24 hours of birth; an almost 30% improvement on last year's rate of 52.6%.
- Referrals from maternity units within one day of birth varied from 66.7% to 90.6% according to the Administrative Unit receiving the referral. Some regions have seen substantial changes in this area over the last year, with improvements occurring in all regions.
- Administrative Units established contact with 92.8% of parents within 24 hours of their child's referral. This is an improvement of 2.4% since last year.

Cleft-related clinical outcomes at five years of age

CRANE collected clinical outcomes at five years of age among children born between 2004 and 2008. These outcomes include **height and weight** (2004-2008 births only); **the number of decayed, missing and filled teeth** (dmft), a measure of oral health; **Five Year Old Index scores**, which reflect dental arch relationships and the effects of primary cleft repair surgery on the facial growth of children with a complete unilateral cleft lip and palate (UCLP); and a **speech assessment recorded using the Cleft Audit Protocol for Speech – Augmented (CAPS-A) scoring system** (2006-2007 births only). Although there is still a high proportion of missing data, some Units have reported outcomes for more than 90% of their eligible patients, suggesting that the reporting of these outcome data is feasible. For those children with reported outcomes:

- 41.9% had at least one decayed, missing or filled tooth (>0 dmft), which, although still substantial, is only slightly higher than the rate in the general population. The proportion of children with >0 dmft varied significantly according to cleft type and Administrative Unit; although treatment indices of 67.2 to 100% across Units indicate that – in the majority of cases – Units have mechanisms in place to deal with any disease occurring. Bilateral cleft lip and palate (BCLP) was associated with the poorest oral health, with 49.5% of BCLP patients having >0 dmft at five years of age.
- Of the 239 children with a complete UCLP who had externally validated Five Year Old Index scores, 25% had scores of '4' or '5', reflecting poor dental arch relationships. This represents a substantial improvement compared to the CSAG findings that 36% of (223) cleft children had poor dental arch relationships at five years of age in 1996[5].
- Of the 415 of eligible children born in 2007 with a cleft affecting their palate, and for whom speech CAPS-A score had been reported across all 16 speech parameters measured, 58.8% had speech scores that would suggest their speech was not significantly different from their non-cleft peer group. 22.2%

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

of children received at least one score indicating a possible structural problem with the palate that may require further surgery.

Unfortunately, outcomes are still not collected consistently across Units:

- Height and weight measures are not collected routinely (in less than 20% of cases) by seven of the fifteen Administrative Units.
- Cambridge did not submit any dmft data as they had not appointed a paediatric dentist up until late in 2013 who could examine children to determine the dmft. However, the reporting of this outcome should improve in future years as this post has now been appointed by Cambridge. A couple of Units reported very few data to CRANE, despite collecting dmft. Inadequate administrative support has been cited previously as the reason for the lack of data submitted.
- The Five Year Old Index score was not reported by three Administrative Units. Of these, Belfast reported that their Orthodontist does not routinely see patients at five years of age.
- Speech measures were not collected consistently across all Administrative Units; with the proportion of eligible children with speech outcome scores ranging from 21.9% to 83.3%.
- CRANE will explore methods for improving communication and links with cleft teams to facilitate the submission of data to the database.

Secondary Speech Surgery

We analysed Hospital Episode Statistics (HES) data to examine instances of additional surgeries involving the palate, needed to improve speech or to close any residual fistulae (referred to as secondary speech surgeries), following primary cleft palate repairs. This was among children born between 1997 and 2004 with a cleft in England; examined up until the age of 7 years. A total of 982 children with a cleft palate, who had undergone a primary repair of the cleft palate, followed by at least one secondary speech surgery (after 6 months of their primary palate repair) were identified. We present the results according to the presence and absence of additional anomalies or syndromes, by cleft type classification, their age at the time of their surgeries, and by region where they underwent their procedure. The main findings are outlined below:

- 30.7% of children who undergo a secondary surgical procedure for the palate have additional anomalies or syndromes; with syndromic CP or BCLP types of cleft increasing the risk of more than one secondary speech repair.
- The majority of children had their secondary speech surgery before school age (of 5 years); with this pattern holding true for most regions – although the proportion of secondary speech surgeries conducted by school age varied substantially between some regions suggesting different patterns of delivery of care.

- These figures have to be interpreted with some caution as, despite data quality reports and checks, coding issues can still be identified in the data.

Educational achievement at five years

We analysed data from the National Pupil Database (NPD), a database containing records on all pupils in England as they progress through primary and secondary school, which was linked to the CRANE database at the individual pupil level – for consenting CRANE children born between 1 January 2000 and 31 December 2008 – in order to explore the impact of facial clefting on long term educational outcomes.

We describe the results of the linkage exercise – i.e. the success of the matching process and factors contributing to successful matching – and the utility of the dataset for making comparisons between the cleft cohort and published national statistics. We also compare the process of linking to a non-health data source such as the NPD with that of linking to a health data source such as HES.

Our initial comparisons between NPD and CRANE data focus initially on the data from assessments conducted at the end of their first year of school (at the end of reception) – known as the Early Years Foundation Stage Profile (EYFSP) – and make some preliminary comparisons between the educational outcomes at age 5 for the cleft cohort and the published national statistics for all children at the age of 5.

The main findings are outlined below:

- Details of 7,152 eligible consented CRANE registered patients born between 2000 and 2008 were available for linkage with NPD, and 56% of these could be linked to a NPD record.
- The NPD linkage rate did not vary across year of birth or by type of cleft. There was considerable variation in NPD linkage rates between the CRANE centres (39% - 87%). NPD linkage rates appeared to be correlated with the quality of postcode capture by CRANE.
- Over all academic years of 2007 to 2012 combined, the 'attainment' gap for all children in the non-syndromic cleft cohort is 5.0 points lower than the national mean total score. A similar 'attainment gap' between the cleft cohort and the national cohort is seen when girls and boys are considered separately.
- The attainment gap between the cleft cohort and the national cohort varies according to the type of cleft, with cleft lip (CL) being associated with the smallest attainment gap and clefts involving the palate (CP, UCLP, BCLP) with larger attainment gaps. This is the case for both girls and boys.
- This preliminary comparison of mean total scores suggest that children with a cleft have poorer educational outcomes on average than their peers nationally and that, as expected, children with a cleft involving the palate fare worse on average than those with a cleft involving the lip only.

Recommendations

Clinical care

- Late diagnosis of cleft palate (CP) remains an important issue that must be addressed. Among children born in 2013, 30.4% of those with CP were diagnosed late according to the national standard[6]. This represents a 1.7% reduction in late diagnosis compared with last year. 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) has set up a working group to develop a best practice guide and an e-learning module on the palate examination in the neonate. The RCPCH working group – including key partners such as the CRANE Database project team – are currently consulting on best practice guidance to aid healthcare professionals in the identification of cleft palate in neonates; and improve and standardise routine postnatal examination of the palate. The expected publication date is October 2014.

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

Outcome measures and reporting to CRANE

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

- CRANE's collection of CAPS-A data has been modified so that data can be analysed and reported more clearly. Analysis of this data will be further refined so as to report using the nationally agreed Speech Outcome Standards in future.

- 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.

    a) The Cleft Psychology Special Interest Group (SIG) were asked by the Craniofacial Society of Great Britain and Ireland (CFSGBI) Council to identify and pilot measurements to evaluate how patient and parent satisfaction could be measured nationally. The following were identified as potential measures: (1) the Friends and Family Test (FFT – developed by the Department of Health) and (2) the Experience of Service Questionnaire (CHI-ESQ – developed by the Commission for Health Improvement (CHI)) satisfaction assessment scales. The CRANE Database project team have drafted a proposal – to the Cleft Development Group (CDG) – to conduct a feasibility study to test

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

how best to collect, analyse and report on this Patient Reported Experience Measure (PREM) data nationally.

b) An international study is developing a patient reported outcome measure questionnaire for Cleft-Lip and/or Palate Patients aged 8 years and older; known as CLEFT-Q. The study team has been inviting comments on their current draft, which is being hosted on the CRANE Database website[7]. Once the CLEFT-Q has been finalised in 2015, the CRANE Database project team plans to develop a method for collecting this data as part of our core dataset.

- Following this first 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.

[7] http://www.crane-database.org.uk/news/

1. Introduction

Craniofacial abnormalities are among the most common of all birth defects[8]. 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[9].

The 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. The HES database contains records on all NHS hospital admissions in England. It 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.

This Annual Report presents data submitted to CRANE by 19 September 2014. It provides feedback to all stakeholders involved in cleft care with respect to children with a cleft lip and/or palate born in 2013 in England, Wales and Northern Ireland (i.e. Born by the 31 December 2013) and provides findings around data relating to cleft care and detailed outcomes – including:

- Trends in CRANE registrations over the last 10 years, comparing the 15 Administrative Units and the four different types of cleft.
- We also report the proportion of babies born in 2013 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 are presented; including height and weight, number of decayed, missing or filled teeth, Five Year Old Index scores, and speech-related outcomes at five years of age in the form of Cleft Audit Protocol for Speech—Augmented (CAPS-A) scores.
- This report also describes the analyses of data from Hospital Episode Statistics (HES), a database containing records on all NHS hospital admissions in England. These data are used to derive information on children diagnosed with cleft lip and/or palate. This year we have examined instances of additional surgeries involving the palate, needed to improve speech or to close any residual fistulae, following primary cleft palate repairs. This was among children born between 1997 and 2004 with an cleft in England; examined up until the age of 7 years. We present the results according to the

[8] 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.
[9] Mossey, PA, Little, J, Munger, RG, Dixon, MJ and Shaw, WC, Cleft lip and palate. The Lancet, 2009. 374(9703): p. 1773-1785.

presence and absence of additional anomalies or syndromes, by cleft type classification, their age at the time of their surgeries, and by region where they underwent their procedure.

- 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 school. This data was linked to CRANE data for consented children. We describe the results of the linkage exercise, and make some preliminary comparisons between the educational outcomes at age 5 for the cleft cohort and the published national statistics for all children at the age of 5; based on assessments conducted at the end of their first year of school (at the end of reception) – known as the Early Years Foundation Stage Profile (EYFSP).

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.

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[10]. 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'[11]. A high-quality national database could furthermore contribute to comparisons between countries.

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

The Cleft Development Group (CDG) is responsible for making arrangements for the running and commissioning of the CRANE Database (see Appendix 3 for CDG's membership). The funding for CRANE was provided by the Specialist Commissioners based on repeated two-year contracts. The CRANE team has responded to a number of requests for information from a commissioner led comprehensive review of all

[10] Clinical Standards Advisory Group, Clinical Standards Advisory Group. Report of a CSAG Committee on cleft lip and/or palate, 1998, The Stationery Office, London.
[11] Department of Health, HSC 1998/238: Cleft lip and palate services, 1998, Department of Health: Leeds.

databases relating to specialised services; the outcome of this has been an agreement to continue to fund CRANE in 2013/14.

1.2. Geographical representation of the cleft Administrative Units

The CRANE Database covers England, Wales and Northern Ireland. Cleft care is currently delivered by eight Regional Cleft Centres and two Managed Clinical Networks. Each of these 10 geographical hubs, with the exception of Northern Ireland, treats at least 80 new children born with a cleft lip and /or palate each year. Several of the Regional Cleft Centres are split between two hospitals, where the primary surgery is usually undertaken. There are 15 Administrative Units (hospitals) who submit data to the CRANE Database (Table 1).

Table 1. Regional Cleft Centres and Managed Clinical Network and their associated Administrative Units

Regional centre / MCN	Administrative Unit
Northern & Yorkshire	Royal Victoria Hospital, Newcastle Leeds General Infirmary, Leeds
North West & North Wales & Isle of Man	Alder Hey Children's Hospital, Liverpool Royal Manchester Children's Hospital, Manchester
Trent	Nottingham City Hospital, Nottingham
West Midlands	Birmingham Children's Hospital, Birmingham
East	Addenbrooke's Hospital, Cambridge
North Thames	Great Ormond Street Hospital, London Broomfield Hospital, Chelmsford
The Spires	John Radcliffe Hospital, Oxford Salisbury District Hospital, Salisbury
South Wales & South West	Morriston Hospital, Swansea Frenchay Hospital, Bristol – Moved to University Hospitals Bristol during 2014
South Thames	Guy's and St Thomas' Hospital, London
Northern Ireland	Royal Belfast Hospital for Sick Children, Belfast

Note: MCN – Managed Clinical Network.

1.3. Aims and objectives of the CRANE Database

The aims of the CRANE Database are:

- 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;
- 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:

- to ensure there is an up-to-date register of all children with cleft lip and/or palate;
- to monitor the frequency and incidence of clefting in the population;
- to audit and report on the quality of care for patients with clefts, thus promoting high standards in clinical management;
- to work with and receive advice from the CFSGBI to improve the delivery of cleft care in the UK;
- to work in partnership with Specialised Commissioning Groups (SCGs) to inform commissioning of cleft services;
- 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) Hospital Episode Statistics (HES) linked to the Office for National Statistics (ONS) mortality dataset, and (3) the National Pupil Database (NPD) linked to both CRANE and HES data.

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 15 Administrative Units, 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 Administrative 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 obtained by the Administrative Unit, usually 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 19 September 2014 pertaining to children born between 1 January 2013 and 31 December 2013 have been included in the analyses described in this Annual report. Patients whose parents did not consent to their data being used by CRANE have been excluded from Tables 7 to 15 (as the data presented in these tables is 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 were defined according to growth charts published by the World Health Organisation (WHO)[12].

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 2008, 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 (3.3% of children born in 2013), cleft type is based on the type reported by the Administrative Unit registering the child. Children with a unilateral cleft lip and palate (UCLP) were categorised according to whether the UCLP was complete or incomplete. A complete UCLP was defined as LAHS or HSAL codes, indicating a complete cleft affecting all three components of the mouth on either the right or left side.

Decayed, missing and filled teeth (dmft)

The dmft score describes the amount of dental caries in an individual and is a measure of oral health. A dmft score reflects the total number of teeth that are decayed, missing or filled. Analyses on dmft data were restricted to consented children born between 2004 and 2007 (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

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

grafting, which may influence this growth further[13]. CRANE collected both internal and external Five Year Old Index scores for consented children born between 2004 and 2007 with a complete UCLP (LAHSAL codes LAHS or HSAL). Some cleft teams score the models of children treated in their Unit (internal scores) before they are sent off to be scored externally (external scores) by a blinded process undertaken by calibrated examiners. For the purpose of this report (for the first time), we have analysed externally validated scores where available; where these were unavailable internal scores are included in the analysis.

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

CAPS-A scores collected at five years of age among children born in 2006 and 2007 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 Cleft Speech Characteristics (CSCs) [14]. The twelve CSCs scores collected for the 2007 cohort – and analysed in detail in Section 3.6.5 of this report – include:

- 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.
- Passive CSCs – for weak and or nasalised consonants; nasal realisation of plosives; and gliding of fricatives.

Missing data

Missing data have been excluded from the denominators presented in Tables 5 to 15. 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.

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[13] 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.

[14] Only 4 Cleft Speech Categories (CSCs) summarising the full 12 CSCs (now being collected from 2007) were collected last year for 2006 births (these were known as anterior oral Cleft Speech Characteristics (CSCs), posterior oral CSCs, non-oral CSCs and passive CSCs). Because of the change in data collection 2006 data is not included in Section 3.6.5.

2.2. Hospital Episode Statistics (HES)

2.2.1. Data source

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.

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).

Using the linked dataset, we examined instances of additional surgeries involving the palate, needed to improve speech or to close any residual fistulae, following primary cleft palate repairs.

2.2.2. Patients

Patients born between 1 January 1997 and 31 December 2004 were included and defined as cleft patients if they had at least one HES record with a diagnosis code for cleft lip and/or palate (ICD-10 codes Q35, Q36, Q37 – as listed in Appendix 5). Patients who were identified in HES as 'private' with an 'unavailable/not applicable' postcode were excluded from analyses as they are unlikely to represent the 'typical' cleft patient in England, and follow up is unlikely to be possible.

2.2.3. Additional anomalies

Children were defined as having a syndrome or additional anomalies if any of their hospital episode or mortality records contained at least one of 33 ICD-10 diagnostic codes (listed in Appendix 5) representing congenital malformations and chromosomal abnormalities, in any one of the diagnosis or cause of death fields.

2.2.4. Cleft type

Clefts were grouped as cleft lip only (CL), cleft palate only (CP) and cleft lip and palate (CLP) according to the diagnosis codes (ICD-10) in the available HES records (as listed in Appendix 5). If a child's records provided more than one diagnosis code, the child was categorised according to the more severe diagnosis code. For example, if a child had both CL and CLP diagnoses in their records, they were categorised as having CLP. If they had separate records of CL and CP, they were also categorised as having CLP.

2.2.5. Analyses

Data were analysed according to the presence and absence of additional anomalies or syndromes, cleft type classification, children's age at the time of their surgeries, and by region where they underwent their procedure. All analyses were performed in Stata 11 (Statacorp, College Station, TX, USA).

2.3. National Pupil Database (NPD)

2.3.1. Data source

CRANE sought and was granted permission by the Department for Education (DfE) – in accordance with their published application process[15] – to link the information held in the CRANE database with the National Pupil Database (NPD).

NPD is a national database containing records on all 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. For example Key Stage 2 data was first collected in 1996 and Key Stage 5 data was first collected in 2002[16].

For this report, NPD data attained through linkage undertaken by the DfE at the individual pupil level with a dataset provided by the CRANE Database. Specifically, personal identifiers (name, postcode and date of birth) were securely passed to the DfE, who performed the linkage between records and provided information about educational attainment at the different stages for the matched cases. This educational information was then merged by the CRANE Data Manager with the existing CRANE-HES linked dataset for validation purposes, and to provide information about factors such as the patient's cleft type and the presence of additional anomalies, as well as the treatment outcomes recorded in CRANE.

Using the linked dataset, we describe the results of the linkage exercise, and make some preliminary comparisons between the educational outcomes at age 5 for the cleft cohort and the published national statistics for all children at the age of 5; based on assessments conducted at the end of their first year of school (at the end of reception) – known as the Early Years Foundation Stage Profile (EYFSP).

2.3.2. Patients

Consenting school aged children born with a cleft lip and/or palate in England between 1 January 2000 and 31 December 2008, as registered in the CRANE Database, with matched NPD data were included.

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[15] https://www.gov.uk/national-pupil-database-apply-for-a-data-extract
[16] http://www.adls.ac.uk/department-for-education/dcsf-npd/?detail

2.3.3. Exclusions

Non-consenting children born with a cleft lip and/or palate in England between 1 January 2000 and 31 December 2008, those children known to have died before age 5 and those with a foreign postcode as registered in the CRANE Database, were not included in the dataset sent for linkage.

2.3.4. Analyses

Data were analysed from assessments conducted at the end of their first year of school (at the end of reception) – known as the Early Years Foundation Stage Profile (EYFSP). Data were analysed according to the presence and absence of additional anomalies or syndromes (2.2.3) and cleft type classification (2.2.4). All analyses were performed in Stata 11 (Statacorp, College Station, TX, USA).

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 2013 in England, Wales and Northern Ireland. Data entered into the CRANE Database by 19 September 2014 have been analysed to assess registration patterns, the timing of diagnosis, referral and contact with Administrative Units around the time of birth, and cleft-related outcomes at five years of age.

The consent status for all children born in 2013 who have been referred to a Cleft Administrative Unit for treatment and registered on CRANE is presented below.

3.1. Consent status

Out of the 1,121 children born in 2013 with a cleft lip and/or palate, we can say the following about the completeness of their consent status data (see Table 2):

- The parents of 895 (79.8%) had been approached for consent. This figure varied across the 15 Administrative Units submitting data to CRANE, ranging from 30.8% at Cambridge to 100% at Belfast.
- Of the 895 children whose parents had been through the consenting process, 98.4% provided consent for their child's data to be submitted to CRANE, which is extremely positive. This proportion ranged from 93.8% for Manchester to 100% at eight Administrative Units (Newcastle, Leeds, Liverpool, Cambridge, Chelmsford, Oxford, Guys and Belfast).
- Administrative Units registered a total of 226 (20.2%) children born in 2013 whose parents had not yet been approached for consent.
- Of these 226, it was not possible to obtain consent (verification) for 23 (10.2%) children (2.1% of all children born in 2013).
- The proportion of children whose parents still need to be approached for consent is wide ranging across Administrative Units (from 0% to 62.6%).
- Overall, the proportions described above are very similar to those described for the 2012 cohort at the time of last year's Annual Report[17].

Generally, the consent data are encouraging; with consent rates very high for those children whose parents have been approached. However, as highlighted in previous Annual Reports, there are 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 Administrative Units suggests different processes are being used between them. Administrative 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.

[17] CRANE Project team on behalf of the Cleft Development Group. CRANE Database Annual Report 2013. London: Clinical Effectiveness Unit, The Royal College of Surgeons of England, 2013.

Table 2. Number of children born in 2013 with a cleft lip and/or palate in England, Wales and Northern Ireland registered on the CRANE Database[a], according to Administrative 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 & Yorkshire	Newcastle	66 (95.7)	0 (0)	0 (0)	3 (4.3)	69
	Leeds	68 (98.6)	0 (0)	0 (0)	1 (1.4)	69
North West & North Wales	Liverpool	61 (92.4)	0 (0)	5 (7.6)	0 (0)	66
	Manchester	60 (83.3)	4 (5.6)	8 (11.1)	0 (0)	72
Trent	Nottingham	89 (90.8)	1 (1)	8 (8.2)	0 (0)	98
West Midlands	Birmingham	96 (80.7)	4 (3.4)	17 (14.3)	2 (1.7)	119
East	Cambridge	28 (30.8)	0 (0)	57 (62.6)	6 (6.6)	91
North Thames	Gt Ormond St	81 (76.4)	2 (1.9)	22 (20.8)	1 (0.9)	106
	Chelmsford	44 (97.8)	0 (0)	1 (2.2)	0 (0)	45
The Spires	Oxford	42 (84)	0 (0)	3 (6)	5 (10)	50
	Salisbury	43 (84.3)	1 (2)	7 (13.7)	0 (0)	51
South Wales & South West	Swansea	34 (87.2)	1 (2.6)	2 (5.1)	2 (5.1)	39
	Bristol	73 (83)	1 (1.1)	13 (14.8)	1 (1.1)	88
South Thames	Guy's	51 (45.1)	0 (0)	60 (53.1)	2 (1.8)	113
Northern Ireland	Belfast	45 (100)	0 (0)	0 (0)	0 (0)	45
All	All	881 (78.6)	14 (1.2)	203 (18.1)	23 (2.1)	1,121

[a] Registered in CRANE by 19 September 2014. Note: MCN – Managed Clinical Network.

3.2. Number of registrations

A total of 14,241 children born over the last fourteen years – between 1 January 2000 and 31 December 2013 – have been registered on the CRANE Database, of whom 1,121 have been added for 2013 births.

Table 3 shows the number of children born with a cleft lip and/or palate for each cleft unit over the last 10 years (since 2004); registered on the CRANE Database by 19 September 2014.

Birmingham registered the most births over the last 10 years (1,134 births in the last 10 years). The North West and North Wales region, consisting of two Administrative Units, is the region that has the most registrations overall (1,536 in the last 10 years).

CRANE received the highest number of registrations for births in 2012 and 2013 since 2008. This increase in registrations could be attributed to the registration of children before the verification of consent.

The total number of registrations each year between 2004 and 2012 (reported in Table 3) has increased by about 10 registrations per year since these were reported on last year[18], which in itself is not unusual as

[18] CRANE Project team on behalf of the Cleft Development Group. CRANE Database Annual Report 2013. London: Clinical Effectiveness Unit, The Royal College of Surgeons of England, 2013.

records are constantly updated by administrative units. Nevertheless, there was a noticeable increase in registrations for the years of 2007 and 2008 (a difference of 58 and 74 cases respectively); potentially due to renewed attention given to reporting of outcomes at five years of age between 2004 and 2008.

Table 3. Number of children born between 2004 and 2013 with a cleft lip and/or palate in England, Wales and Northern Ireland registered on the CRANE Database[a], according to Administrative Unit grouped within Regional Cleft Centre / Managed Clinical Network (MCN) and year of birth.

Regional Cleft Centre / MCN	Administrative Unit	Year of birth (N)										
		2004	2005	2006	2007	2008	2009	2010	2011	2012	2013	All
Northern & Yorkshire	Newcastle	59	77	55	85	65	64	64	65	65	69	668
	Leeds	70	71	75	67	73	67	71	70	64	69	697
North West & North Wales	Liverpool	65	86	54	62	87	79	86	62	64	66	711
	Manchester	83	85	105	89	84	67	88	82	70	72	825
Trent	Nottingham	92	106	94	84	99	81	94	90	92	98	930
West Midlands	Birmingham	117	102	121	98	123	118	102	110	124	119	1134
East	Cambridge	82	87	81	82	84	84	81	66	87	91	825
North Thames	Gt Ormond St	45	79	94	130	155	107	86	111	136	106	1049
	Chelmsford	30	36	29	45	38	48	39	54	43	45	407
The Spires	Oxford	40	43	48	55	33	53	45	59	45	50	471
	Salisbury	58	46	67	71	56	39	54	52	51	51	545
South Wales & South West	Swansea	37	45	45	48	42	47	44	51	52	39	450
	Bristol	53	51	58	62	70	52	74	50	67	88	625
South Thames	Guy's	86	95	101	110	104	82	62	78	156	113	987
Northern Ireland	Belfast	29	37	42	41	31	34	37	36	32	45	364
All	All	946	1,046	1,069	1,129	1,144	1,022	1,027	1,036	1,148	1,121	10,688

[a] Registered in CRANE by 19 September 2014. Note: MCN – Managed Clinical Network.

Table 4. Number (%) of children born between 2004 and 2013 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	2004		2005	2006	2007	2008	2009	2010	2011	2012		2013		All	
Cleft lip	209	(22.8)	196 (19.5)	236 (23.1)	264 (23.7)	272 (24.6)	214 (21.7)	245 (24.4)	247 (24.9)	249	(23.1)	293	(26.9)	2425	(23.5)
Cleft palate	414	(45.2)	495 (49.2)	472 (46.2)	490 (43.9)	513 (46.4)	461 (46.8)	442 (44.1)	441 (44.5)	479	(44.4)	473	(43.5)	4680	(45.4)
UCLP	204	(22.3)	236 (23.5)	217 (21.3)	248 (22.2)	238 (21.5)	205 (20.8)	208 (20.7)	216 (21.8)	253	(23.4)	220	(20.2)	2245	(21.8)
BCLP	89	(9.7)	79 (7.9)	96 (9.4)	114 (10.2)	82 (7.4)	106 (10.8)	108 (10.8)	86 (8.7)	99	(9.2)	102	(9.4)	961	(9.3)
Not specified	30	-	40 -	48 -	13 -	39 -	36 -	24 -	46 -	68	-	33	-	377	-
All	946	(100)	1046 (100)	1069 (100)	1129 (100)	1144 (100)	1022 (100)	1027 (100)	1036 (100)	1148	(100)	1121	(100)	10688	(100)

[a] Registered in CRANE by 19 September 2014. Note: CL - Cleft Lip; CP - Cleft Palate; UCLP - Unilateral cleft lip and palate; and BCLP - Bilateral cleft lip and palate.

With regards to the number and proportion of children born with a cleft lip and/or palate according to cleft type, the distribution of the four main cleft types is shown in Table 4. Cleft type was defined according to:

1. Reported LAHSAL codes, or
2. where LAHSAL has not been reported (3.3% of children registered in 2013) the cleft type is based on the type reported by the Administrative Unit registering the child, where provided.

Based on the reported LAHSAL codes, or cleft types reported by the Administrative Units where LAHSAL codes were not reported:

- The distribution of cleft type is consistent over time and with registrations in recent previous years. CP is the most common type of cleft, affecting over 45% of the cleft population. Bilateral cleft lip and palate (BCLP) is the least common type, affecting under 10% of people with clefts.
- A total of 157 children registered in 2013 had complete UCLP (defined by either 'LAHS..' or '..HSAL' LAHSAL codes), representing 71.4% of the 220 children with UCLP.
- The proportion of children without a specified cleft type ranged between Units from 0% (Birmingham, Chelmsford, Salisbury, Swansea and Belfast) to 14.2% at GOSH.
- Nevertheless, overall, rates have improved substantially for 2013 registrations – with only 3% of the 1,121 total registered children born in 2013 not having their type of cleft specified (either by LAHSAL codes or by the Administrative Units). This represents a much lower proportion of children than the 7.8% rate for 2012 at this time last year[19], which is a positive trend suggesting that Administrative Units are improving the recording of cleft type in CRANE at the point of registration.

3.3. Characteristics of children born with a cleft lip and/or palate, 2013

Of the children born with a cleft in 2013, whose sex was reported to CRANE, 43% were girls and 57% were boys. Eleven children did not have their sex reported to CRANE (1% of the total children registered for 2013). There are 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 (59% vs. 41%; 63% vs. 37% and 79% vs. 21%, respectively).

Gestational age was reported for 543 (61.6%) of the consented babies born in 2013. This reporting has increased substantially – by 10.1% since last year – however, further improvements are required.

- The mean gestation for those born in 2013 was 38.9 weeks (95% CI 38.7 to 39.1 weeks) and ranged from 25 to 43 weeks.

[19] CRANE Project team on behalf of the Cleft Development Group. CRANE Database Annual Report 2013. London: Clinical Effectiveness Unit, The Royal College of Surgeons of England, 2013.

- Forty-seven (8.7%) babies were premature (born before 37 weeks' gestation), which is higher than the six per cent national average in England[20], 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 38.4% of registered (and consented) children were missing this information.
- A valid birth weight was reported for 553 (62.8%) consented babies born in 2013. The mean birth weight was 3.22kg (95% CI 3.16 to 3.27kg), which is consistent with the national average in England.

Among all the children born in 2013, there were 20 (1.8%) deaths reported to CRANE. The majority of deaths (75%) occurred between one month and one year of age. It is not known from CRANE whether these children had additional anomalies or syndromes.

3.4. Timing of diagnosis

3.4.1. Diagnosis times among children born in 2013

Of the 1,121 children born in 2013 with a cleft diagnosis, 76 (6.8%) did not have the timing of their diagnosis reported to CRANE. This is only slightly higher (1.1%) than last year's figures. Units with high levels of missing diagnosis time data include Manchester (23.6%), Cambridge (22%), Liverpool (15.2%), Salisbury (11.8%) and GOSH (10.3%). All other Units had missing data rates below 10%.

Of the 1,045 children born in 2013 with a reported diagnosis time, 435 (41.6%) had their cleft diagnosed during the antenatal period. This is slightly higher than the 39.1% of children born in 2012 with an antenatal diagnosis. The proportion of children diagnosed antenatally varied between cleft types (as shown in Table 5); sixty-three per cent of children with CL and over 75% of children with UCLP and BCLP were diagnosed in the antenatal period. Conversely, only 1.6% of children with a CP were diagnosed antenatally, a statistically significant difference ($p<0.001$), which demonstrates the difficulty of identifying this type of cleft with current antenatal screening techniques.

[20] Hospital Episode Statistics. Maternity Data, 2009-2010. 2010 2011/11/18/; Available from: http://www.hesonline.nhs.uk/Ease/servlet/ContentServer?siteID=1937&categoryID=1475.

Table 5. Number (%) of CRANE-registered children born in 2013 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	≤72 hours[b]	≤1 week	≤1 month	≤6 months	>6 months	**All**
CL	178	(63.3)	90 (32)	0 (0)	5 (1.8)	1 (0.4)	5 (1.8)	2 (0.7)	281
CP	7	(1.6)	300 (68)	2 (0.5)	85 (19.3)	23 (5.2)	18 (4.1)	6 (1.4)	441
UCLP	174	(82.1)	37 (17.5)	0 (0)	1 (0.5)	0 (0)	0 (0)	0 (0)	212
BCLP	69	(75.8)	22 (24.2)	0 (0)	0 (0)	0 (0)	0 (0)	0 (0)	91
Not specified	7	(35)	5 (25)	0 (0)	5 (25)	2 (10)	0 (0)	1 (5.0)	20
All	435	(41.6)	454 (43.4)	2 (0.2)	96 (9.2)	26 (2.5)	23 (2.2)	9 (0.9)	1045

[a] 76/1121 (6.8%) 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] Small numbers reported within ≤72 hours to be expected as dataset expanded in May 2014 on request to allow recording of 'timing of diagnosis' within 72 hours; to align CRANE data collection with RCPCH national screening guidance[21].

The distribution of timing of diagnosis shown in Table 5, for the children born in 2013, is very similar to that of 2012 births at this time last year.

Of the 610 children not diagnosed during the antenatal period, 454 (74.4%) were diagnosed at birth. This is 2.1% higher than the rate in 2012. Of those children who did not have their cleft identified antenatally, the majority (87.5%) with a CL, UCLP and BCLP were diagnosed at the time of birth; with 30.9% of all children with CP not identified until after the time of birth (and 5.5% of all children with CP diagnosed beyond one month after birth). It should be noted that some children born in 2013 with a CP may not yet have had their cleft identified. Each year, around ten children with CP are diagnosed after six months of age – in the case of 2013 there were 6 children – showing a small improvement when compared to previous years.

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[22]. This year (as for last year), we have examined diagnosis time among CP patients born over the last five years – between 1 January 2009 and 31 December 2013. No statistically significant differences were found between birth years (p=0.31), indicating diagnosis times have not improved in recent years.

[21] UK National Screening Committee Newborn and Infant Physical Examination 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).
[22] Bannister, P, Management of infants born with a cleft lip and palate. Part 1. Infant, 2008. 4(1): p. 5-8.

Table 6. Number (%) of CRANE-registered children born between 2009 and 2013 with a cleft palate, according to the timing of diagnosis and Administrative Unit.

Regional Cleft Centre / MCN	Administrative Unit	Time of diagnosis in relation to birth[a] n (%)							All
		Antenatal	At birth	≤72 hrs		≤1 week	≤1 month	≤6 months	>6 mths	
Northern &	Newcastle	3 (2)	99 (66.4)	0	(0)	17 (11.4)	12 (8.1)	6 (4)	12 (8.1)	149
Yorkshire	Leeds	0 (0)	98 (68.1)	0	(0)	29 (20.1)	12 (8.3)	5 (3.5)	0 (0)	144
North West &	Liverpool	5 (3.2)	115 (73.7)	0	(0)	22 (14.1)	7 (4.5)	6 (3.8)	1 (0.6)	156
North Wales	Manchester	0 (0)	98 (67.1)	0	(0)	30 (20.5)	10 (6.8)	7 (4.8)	1 (0.7)	146
Trent	Nottingham	2 (1.3)	107 (71.8)	0	(0)	32 (21.5)	5 (3.4)	3 (2)	0 (0)	149
West Midlands	Birmingham	1 (0.4)	178 (77.4)	2	(0.9)	23 (10)	5 (2.2)	16 (7)	5 (2.2)	230
East	Cambridge	1 (0.7)	103 (68.2)	1	(0.7)	21 (13.9)	13 (8.6)	12 (7.9)	0 (0)	151
North Thames	Gt Ormond St	3 (1.6)	92 (48.7)	1	(0.5)	67 (35.4)	8 (4.2)	13 (6.9)	5 (2.6)	189
	Chelmsford	1 (1.1)	56 (58.9)	0	(0)	19 (20)	4 (4.2)	7 (7.4)	8 (8.4)	95
The Spires	Oxford	2 (1.8)	80 (71.4)	0	(0)	9 (8)	4 (3.6)	7 (6.3)	10 (8.9)	112
	Salisbury	2 (2.6)	60 (78.9)	1	(1.3)	8 (10.5)	2 (2.6)	2 (2.6)	1 (1.3)	76
South Wales &	Swansea	1 (1)	78 (78.8)	0	(0)	10 (10.1)	8 (8.1)	1 (1)	1 (1)	99
South West	Bristol	2 (1.7)	81 (66.9)	0	(0)	20 (16.5)	10 (8.3)	7 (5.8)	1 (0.8)	121
South Thames	Guy's	2 (1)	104 (53.9)	0	(0)	54 (28)	17 (8.8)	13 (6.7)	3 (1.6)	193
Northern Ireland	Belfast	1 (1.4)	56 (75.7)	0	(0)	9 (12.2)	0 (0)	3 (4.1)	5 (6.8)	74
All	All	26 (1.2)	1405 (67.4)	5	(0.2)	370 (17.8)	117 (5.6)	108 (5.2)	53 (2.5)	2084

[a] 212/2,296 (9.2%) missing diagnosis time and excluded from 'All' values. Note: MCN - Managed Clinical Network.
[b] Small numbers reported within ≤72 hours (as per Tables 5).

Table 6 shows the CP diagnosis times according to Administrative Unit. The proportion of CPs diagnosed at birth ranged from 48.7% among children registered by GOSH to over 75% among those registered by Birmingham, Salisbury, Swansea and Belfast. 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, 13.3% of children with a CP were not diagnosed until they were more than one week old, which is concerning given that the National Standard[23] 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. Administrative Units are advised to encourage their referring maternity units to identify all clefts as promptly as possible.

3.5. Referral to and first contact with a cleft team

3.5.1. Referral among children born in 2013

Out of the 881 consented children born in 2013, 23 (2.6%) were missing referral time; an improvement on registrations for 2012 at the time of the last Annual Report – when 11.7% were missing referral times. In

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

addition all units were missing less than 11% of their referral time data which represents a vast improvement on reporting of this data when compared to previous years.

Of the 858 children with a reported referral time, 81.9% were referred to an Administrative Unit within 24 hours of birth – a substantial improvement of 29.3% since this time last year. 94% of children whose clefts were diagnosed antenatally were referred to an Administrative Unit within 24 hours of birth; while 73% of the 500 children whose clefts were diagnosed after birth were referred to an Administrative Unit within 24 hours of birth.

Table 7 shows that the proportion of referrals within 24 hours of birth varied according to cleft type ($p<0.001$), with CP patients having the lowest proportion (69.9%) out of those with a known cleft type, which corresponds with later diagnosis times for these children.

Referrals within 24 hours of birth varied according to the Administrative Unit receiving the referral (Table 8) although not significantly ($p=0.055$); ranging from 66.7% of children registered by Chelmsford to 90.6% registered by Birmingham referred from maternity units within 24 hours of birth. Progress in this area has been made in all regions, with the greatest improvements observed at GOSH, Oxford, Swansea and Bristol (with increases of more than 35% in referrals within 24 hours since we reported on 2012 data).

Table 7. Number (%) of CRANE-registered consented children born in 2013 with a cleft lip and/or palate who were referred within 24 hours of birth to the Administrative Unit and contacted by the Administrative Unit within 24 hours of referral, according to cleft type.

	Referral to Unit		**Contact between Unit and parents of patient**	
	Within 24h of birth	All[a]	Within 24h of referral to Unit	All[b]
Cleft type	n (%)	N	n (%)	N
CL	202 (86.7)	233	201 (89.7)	224
CP	249 (69.9)	356	308 (92.5)	333
UCLP	174 (93.5)	186	172 (94.5)	182
BCLP	76 (97.4)	78	73 (98.6)	74
Not specified	2 (40)	5	5 (100)	5
All	703 (81.9)	858	759 (92.8)	818

[a]23/881 (2.6%) missing referral time and [b]63/881 (7.2%) missing contact time and excluded in 'All' values. Note: CL - Cleft Lip; CP - Cleft Palate; UCLP - Unilateral cleft lip and palate; and BCLP - Bilateral cleft lip and palate.

3.5.2. First contact between the Unit and parents of children born in 2012

Out of the 881 consented children born in 2013, 63 (7.2%) were missing the first contact time between Units and parents; an improvement on registrations for 2012 at the time of the last Annual Report – when 21.5% were missing first contact times. In addition 13 out of the 15 Administrative Units were missing less than 10% of their first contact time data, which represents a big improvement on reporting of this data when compared to years past. Cambridge and GOSH were the only units that showed high levels of missing first contact data (57.1% and 34.6% respectively).

Of the 818 consented children with a reported contact time, Units established contact with 92.8% within 24 hours of referral (Table 7). This is consistent with last year's rate of 90.4% for 2012 births. The proportion of

patients contacted within 24 hours of being referred to an Administrative Unit did not vary significantly between cleft types (p=0.09).

Contact between units and parents of patients within 24 hours of referral did vary significantly according to the Administrative Unit receiving the referral (p<0.001) (Table 8); with rates varying between Units (56.1% to 100.0%), but the majority (12/15) contacted more than 90% of their patients within 24 hours of being referred.

Table 8. Number (%) of CRANE-registered consented children born in 2013 with a cleft lip and/or palate who were referred within 24 hours of birth and contacted within 24 hours of referral, according to Administrative Unit.

		Referral to Unit		Contact between Unit and parents of patient	
Regional centre / MCN	**Administrative Unit**	Within 24h of birth n (%)	All[a] N	Within 24h of referral to Unit n (%)	All[b] N
Northern & Yorkshire	Newcastle	53 (81.5)	65	65 (100)	65
	Leeds	52 (76.5)	68	67 (100)	67
North West & North Wales	Liverpool	47 (81)	58	57 (98.3)	58
	Manchester	47 (85.5)	55	54 (100)	54
Trent	Nottingham	78 (87.6)	89	89 (100)	89
West Midlands	Birmingham	87 (90.6)	96	88 (92.6)	95
East	Cambridge	21 (84)	25	12 (100)	12
North Thames	Gt Ormond St	58 (76.3)	76	37 (69.8)	53
	Chelmsford	28 (66.7)	42	23 (56.1)	41
The Spires	Oxford	35 (83.3)	42	39 (92.9)	42
	Salisbury	35 (85.4)	41	37 (90.2)	41
South Wales & South West	Swansea	28 (82.4)	34	34 (100)	34
	Bristol	57 (78.1)	73	70 (95.9)	73
South Thames	Guy's	46 (90.2)	51	49 (96.1)	51
Northern Ireland	Belfast	31 (72.1)	43	38 (88.4)	43
All	All	703 (81.9)	858	759 (92.8)	818

[a]23/881 (2.6%) missing referral time and [b]63/881 (7.2%) missing contact time and excluded in 'All' values. Note: MCN - Managed Clinical Network.

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

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

3.6.1. Reporting of outcomes

Table 9 shows the number of consented children born between 2004 and 2008 with reported outcomes at five years of age (excluding children with submucous cleft palates)[24], according to Administrative Unit.

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

- There is still a very high proportion of missing data for five-year old height and weight (for eligible children; 69.7% and 69% missing respectively); this is despite a small improvement in reporting of these data since last year's Annual Report (4.5% increase in reporting of height and 4.8% for weight). Belfast was the only Administrative Unit to not submit any height or weight data. There was wide variation in completeness of data across the Administrative Units; ranging from 0.5% (Oxford) and 0.8% (Salisbury) reporting for both height and weight, suggesting that these measures are not routinely collected, to 80.1% and 81.2% for height and weight respectively (both report rates for Newcastle).
- It is important to note improvements in reporting of height and weight by seven of the Administrative Units – of approximately 10% – in Newcastle, Manchester, Nottingham, GOSH, Chelmsford, Swansea and Bristol, which is very encouraging.
- The proportion of eligible children with reported decayed, missing, filled teeth (dmft) index scores has increased since this last time last year by 6%; and ranged from 0% (Cambridge) to 93% (Birmingham). Nottingham submitted data for only 12.2% of their eligible patients[25] – nevertheless this represents a positive improvement since last year (when data submission was at 6.5%), with their outcome data submission continuing to improve year on year.
- It is acknowledged that sometimes there are reasons outside the units' control as to why outcome(s) data cannot be collected, and we encourage centres to report these. For example, Cambridge did not submit any dmft data as they had not appointed a paediatric dentist up until late in 2013 who could examine children to determine the dmft.
- There was wide variation in reporting of Five Year Old Index data across the Administrative Units. The proportion of children with a Five Year Old Index reported has increased by 4.5% since last year's report (from 57.5% for 2012 data), which is encouraging. It is hoped that this proportion will continue to increase over the next few years.
- Five Year Old Index data are not collected by Belfast because children are not routinely seen by Orthodontists at five years of age. Apart from this, reporting of Five Year Old Index data ranged from 0% (GOSH and Chelmsford) to 90.9% (Oxford).
- The proportion of eligible children with Speech outcome scores ranged from 21.9% at GOSH to 83.3% at Swansea. Given that this is only the second year that speech outcome scores have been requested, and changes to the data collected were made recently in 2014, CRANE is encouraged by the fact that (1) all Administrative Units have reported speech data and (2) overall reporting has increased by more than 12% (from 48.4% to 61.1% this year).
- It is positive to note that reporting has increased for all outcomes at five years of age[26] since this time last year.

[25] Nottingham have previously informed CRANE that they have not had adequate administrative support to provide CRANE with dmft data.

[26] 4.8% for weight; 4.5% for height; 6% for dmft; 4.5% for 5 year index and 12.7% for speech.

Table 9. Number (%) of CRANE-registered [a] consented children born between 2004 and 2008 with reported outcomes at five years of age (excluding children with submucous cleft palates)[27], according to Administrative Unit.

Regional centre / MCN	Administrative Unit	Children alive at 5 years[b]	Weight[c]		Height[c]		dmft[d]			5 year index[e]			Speech[f]		
			Reported		Reported			Reported			Reported[28]			Reported	
		N	n	(%)	n	(%)	N	n	(%)	N	N	(%)	N	n	(%)
Northern & Yorkshire	Newcastle	282	229	(81.2)	226	(80.1)	226	196	(86.7)	29	25	(86.2)	87	66	(75.9)
	Leeds	317	247	(77.9)	254	(80.1)	257	180	(70)	39	34	(87.2)	97	64	(66)
North West & North Wales	Liverpool	292	39	(13.4)	38	(13)	213	151	(70.9)	42	24	(57.1)	79	47	(59.5)
	Manchester	345	97	(28.1)	96	(27.8)	270	196	(72.6)	36	30	(83.3)	123	74	(60.2)
Trent	Nottingham	438	68	(15.5)	70	(16)	352	43	(12.2)	59	27	(45.8)	133	77	(57.9)
West Midlands	Birmingham	490	270	(55.1)	263	(53.7)	385	358	(93)	69	57	(82.6)	138	95	(68.8)
East	Cambridge	307	22	(7.2)	18	(5.9)	236	0	(0)	49	18	(36.7)	99	45	(45.5)
North Thames	Gt Ormond St	350	53	(15.1)	54	(15.4)	248	95	(38.3)	31	0	(0)	105	23	(21.9)
	Chelmsford	152	39	(25.7)	39	(25.7)	122	51	(41.8)	15	0	(0)	37	13	(35.1)
The Spires	Oxford	193	1	(0.5)	1	(0.5)	165	116	(70.3)	33	30	(90.9)	69	44	(63.8)
	Salisbury	250	2	(0.8)	2	(0.8)	197	145	(73.6)	32	29	(90.6)	85	57	(67.1)
South Wales & South West	Swansea	214	102	(47.7)	87	(40.7)	172	136	(79.1)	24	8	(33.3)	72	60	(83.3)
	Bristol	276	102	(37)	104	(37.7)	207	119	(57.5)	30	20	(66.7)	82	60	(73.2)
South Thames	Guy's and St Thomas'	450	130	(28.9)	116	(25.8)	348	208	(59.8)	72	62	(86.1)	139	84	(60.4)
Northern Ireland	Belfast	166	0	(0)	0	(0)	138	51	(37)	27	0	(0)	62	50	(80.6)
All	All	4,522	1,401	(31)	1,368	(30.3)	3,536	2,045	(57.8)	587	364	(62)	1,407	859	(61.1)

[a] Registered in CRANE by 19 September 2014. MCN - Managed Clinical Network.
[b] 82/4,604 (1.8%) children died before 5 years and are excluded from table.
[c] Children born in 2004-2008: 167/4,689 (3.6%) children with submucous cleft palates are excluded from weight and height data.
[d] Children born in 2004-2007 only: 142/3,678 (3.9%) children with submucous cleft palates are excluded from dmft data.
[e] Children born in 2004-2007 only: 237/826 (28.7%) children with submucous cleft palates & incomplete UCLPs are excluded from 5-year old index data.
[f] Children born in 2006-2007 only: 487/1,894 (25.7%) children with submucous cleft palates & born with either a CL (23.8%) or a non-specified cleft type (1.9%) are excluded from speech data.

[27] Submucous cleft palate patients excluded from all the five year outcomes as all/most teams do not audit these patients.
[28] Only 7/15 of the Administrative 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 Administrative Unit (0-62) means that statistical comparison between Administrative Units is not currently appropriate. CRANE will continue to collect these outcomes over the next few years, and as numbers increase, meaningful comparison between Units will become possible.

3.6.2. Height and weight (2004-2008 births)

Five-year height and weight were reported for 30.3% and 31% (respectively) of the 4,522 children born in 2004-2008[29], who were alive at five years of age. The mean (SD) height was 111.7cm (5.9cm) while the mean (SD) weight was 19.7kg (3.2kg). Boys were marginally taller than girls (112.3 cm vs. 110.9cm) and a small difference in mean weight existed between the sexes (boys 19.8Kg vs. girls 19.5Kg).

3.6.3. Decayed missing and filled teeth (dmft) (2004-2007 births)

The dmft describes the amount of dental caries in an individual and is a measure of oral health. A dmft score reflects the total number of teeth that are decayed, missing or filled. The risk of dental caries is thought to be higher among children with a cleft lip and/or palate compared to children without an oral cleft[30]. We collect dmft data on CRANE-registered consented children at five years of age.

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

Among children with a reported dmft outcome[31] , 41.9% of children with a cleft had at least one (>0) decayed, missing or filled tooth. The mean number of dmft at five years among children registered in CRANE was 2, with scores ranging from 0 to 20. Two hundred and eighty-four children (13.9%) had a dmft score greater than 5.

The dmft data, obtained in 2005, available for five-year old children in the general population in England and Wales show that 38.8% of five-year olds had >0 dmft, with a mean number of 1.5[32]. The comparable figure of 41.9% among CRANE-registered children (shown in Table 10) is quite close, although slightly higher, to that of the general population; in part probably due to the fact that the number of dmft among children with a CL was lower than the general population (33.2% versus 38.8%). Despite this, the mean dmft was substantially higher among CP, UCLP and BCLP patients compared to the general population (2.4, 2.0 and 2.6 versus a mean of 1.5).

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

[30] 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; and 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.

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

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

Table 10. Number (%) of CRANE-registered consented children born between 2004 and 2007 with a cleft lip and/or palate according to the number of decayed, missing or filled teeth (dmft) at age five years and cleft type.

	Number of decayed, missing or filled teeth (dmft)				
		0	>0		
Cleft type	Mean (95% CI)	n (%)	n (%)	(95% CI)	**All**[a]
CL	1.2 (1.0 to 1.5)	286 (66.8)	142 (33.2)	(28.7 to 37.7)	428
CP	2.4 (2.1 to 2.6)	478 (58.5)	339 (41.5)	(38.1 to 44.9)	817
UCLP	2.0 (1.7 to 2.2)	304 (53.1)	268 (46.9)	(42.8 to 51)	572
BCLP	2.6 (2.0 to 3.1)	105 (50.5)	103 (49.5)	(42.7 to 56.4)	208
Not specified	0.9 (0 to 1.8)	15 (75)	5 (25)	(4.2 to 45.8)	20
All	2.0 (1.9 to 2.2)	1188 (58.1)	857 (41.9)	(39.8 to 44)	2045

[a] 1,491/3,536 (42.2%) children with missing dmft data, 145/3,749 (3.9%) children with submucous clefts, and 68/3,605 (1.9%) children who died before the age of five were excluded in 'All' values. Note: CL - Cleft Lip; CP - Cleft Palate; UCLP - Unilateral cleft lip and palate; and BCLP - Bilateral cleft lip and palate.

The fact that dmft were submitted for only 57.8% of children means that these data should be interpreted with caution. One Administrative Unit (Cambridge) did not provide dmft data for any of their patients. Thus, 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.

Table 11 shows the prevalence of dmft according to Administrative Unit[33]. There was a significant variation in dmft scores across Units ($p<0.05$). Children registered by Nottingham[34] had the highest number of mean dmft, which was significantly different to the overall mean. It should be noted that Nottingham submitted data for very few patients, 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 Nottingham are required. Chelmsford, Salisbury, Bristol and Guy's had mean dmft values that were significantly lower than the overall mean. In terms of the proportion of cleft children with >0 dmft, Salisbury had the lowest proportion (28.3%), which was significantly different to the overall proportion among cleft children. Whilst the proportion of cleft children with >0 dmft varies between regions, for the majority of regions their rate does not seem to differ substantially from their region's total population rate[35]. The only substantial difference appears to be for Birmingham, whose cleft rate is a third higher than their total population rate of 31.4%.

[33] Submucous cleft palate patients excluded from all five year outcomes as all/most teams do not audit these patients.
[34] Note that dmft data were reported for only 12.2% of eligible children registered by Nottingham.
[35] 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.

Regional differences in the levels of dental disease will not only be affected by the dental care received by children. Oral health will also be affected by deprivation, cultural differences in attitudes to dental health and water fluoridation levels. A systematic review found that water fluoridation is associated with an increased proportion of children without caries and a reduction in the number of teeth affected by caries[36]. 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; however the North East had the highest proportion (50%)[37]. Accurate water fluoridation data will be useful for interpreting dmft regional differences and allowing for risk adjustment in the long term.

Table 11 also shows the average treatment index for children according to Administrative Unit for the first time in the annual report. The treatment index is a figure calculated from the dmft[38], as raw dmft scores give a figure for dental disease experience but do not distinguish if there is active disease present at the time or not.

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[39]. 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 1881 children with dmft scores of 0 or scores for all three ‘d’, ‘m’ and ‘dmft’ data items (to allow calculation of treatment index scores), there was no significant variation in treatment indices scores across Units (p=0.44). Children registered by Leeds had the lowest average proportion of treated disease (67.2%), while Chelmsford and Oxford had highest average proportion of treated disease possible (100%). Further analysis of treatment index with cleft type and deprivation is planned for next year’s annual report.

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[36] 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.

[37] 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.

[38] Calculated as = (Total number of decayed teeth in primary dentition (d) + Total number of missing teeth in primary dentition (m)) / ‘Total number of decayed, missing or filled teeth in primary dentition (dmft).

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

Table 11. Number (%) of CRANE-registered consented children born between 2004 and 2007 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 Administrative Unit.

Regional centre / MCN	Administrative Unit	Number of decayed, missing or filled teeth (dmft)				Treatment Index		
			0	>0			Average	
		Mean (95% CI)	n (%)	n (%)	95% CI	All[a]	Percentage	All[b]
Northern & Yorkshire	Newcastle[§¥]	2.7 (2.1 to 3.3)	103 (52.6)	93 (47.4)	(40.4 to 54.5)	196	73.5	167
	Leeds[§¥]	2.7 (2.1 to 3.4)	96 (53.3)	84 (46.7)	(39.3 to 54)	180	67.2	180
North West & North Wales	Liverpool	2.2 (1.6 to 2.8)	86 (57)	65 (43)	(35.1 to 51)	151	70.9	147
	Manchester	2.2 (1.7 to 2.7)	109 (55.6)	87 (44.4)	(37.4 to 51.4)	196	68.6	195
Trent	Nottingham[¥]	4.0 (2.3 to 5.8)	18 (41.9)	25 (58.1)	(42.8 to 73.5)	43	72.8	40
West Midlands	Birmingham[§¥]	2.0 (1.6 to 2.4)	206 (57.5)	152 (42.5)	(37.3 to 47.6)	358	68.9	357
East	Cambridge	- -	- -	- -	-	-	-	-
North Thames	GOSH[§¥]	2.3 (1.5 to 3)	53 (55.8)	42 (44.2)	(34 to 54.4)	95	80.5	76
	Chelmsford	1.3 (0.7 to 1.9)	29 (56.9)	22 (43.1)	(29.1 to 57.2)	51	100	29
The Spires	Oxford[§]	2.0 (1.3 to 2.7)	69 (59.5)	47 (40.5)	(31.4 to 49.6)	116	100	70
	Salisbury[§]	1.0 (0.6 to 1.2)	104 (71.7)	41 (28.3)	(20.9 to 35.7)	145	69.2	110
South Wales & South West	Swansea[§¥]	2.0 (1.5 to 2.5)	77 (56.6)	59 (43.4)	(34.9 to 51.8)	136	78.6	135
	Bristol[§¥]	1.3 (0.9 to 1.8)	78 (65.5)	41 (34.5)	(25.8 to 43.1)	119	80.3	117
South Thames	GSTT[§¥]	1.4 (1.0 to 1.7)	138 (66.3)	70 (33.7)	(27.2 to 40.1)	208	84.2	208
Northern Ireland	Belfast	2.0 (1.2 to 2.8)	22 (43.1)	29 (56.9)	(42.8 to 70.9)	51	69.5	50
All	All	2.0 (1.9 to 2.2)	118 (58.1)	857 (41.9)	(39.8 to 44)	2045	76.2	1881

[a] 1,491/3,536 (42.2%) children with missing dmft data, 145/3,749 (3.9%) children with submucous clefts, and 68/3,605 (1.9%) children who died before the age of five were excluded in 'All' values. [§]British Association for the Study of Community Dentistry (BASCD) calibrated assessor. [¥]Specialist paediatric dentist. Note: MCN – Managed Clinical Network.
[b] Only including children who were alive after the age of five, without submucous clefts, and who had either a dmft score of 0[40] or scores for all three 'd', 'm' and 'dmft' data items (to allow calculation of treatment index scores).

3.6.4. Five Year Old Index (2004-2007 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[41]. 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[42]. The Five Year Old Index may, therefore, also be used to compare treatment outcomes between centres and surgeons. Patients categorised as '1' and '2' on the index are considered to have the best possible outcome, while those categorised as '4' and '5' are thought

[40] If a dmft score for an individual is 0 then the treatment index is 1 (100%) as there is no untreated disease.
[41] 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.
[42] 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.

to have very poor outcomes in terms of facial growth, and they may benefit from further surgery to correct their facial disproportion once facial growth is complete.

Table 12 shows the distribution of externally validated Five Year Old Index scores where available (in 233/364 (88.5%) of eligible cases) as provided by 12 of the 15 Administrative Units – where externally validated scores were unavailable, internal scores were included in the analysis (in 42/364 (11.5%) of cases)[43].

Overall, 44.5% of complete UCLP patients born between 2004 and 2007 had Five Year Old Index scores in the two groups considered to have the best possible dental arch relationships (scores '1' or '2') while 25% 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 1996[44]. Comparisons between Units in five year old index scores are not appropriate because of the small number of children within each group.

Table 12. Number (%) of CRANE-registered consented children born between 2004 and 2007 with a complete unilateral cleft lip and palate[45]; according to Five Year Old Index scores and Administrative Unit.

Regional centre / MCN	Administrative Unit	Five Year Old Index n (%)					All[a]
		1	2	3	4	5	
Northern & Yorkshire	Newcastle	4 (16)	2 (8)	13 (52)	3 (12)	3 (12)	25
	Leeds	1 (2.9)	13 (38.2)	10 (29.4)	9 (26.5)	1 (2.9)	34
North West & North Wales	Liverpool	2 (8.3)	7 (29.2)	9 (37.5)	5 (20.8)	1 (4.2)	24
	Manchester	1 (3.3)	12 (40)	6 (20)	8 (26.7)	3 (10)	30
Trent	Nottingham	2 (7.4)	14 (51.9)	6 (22.2)	2 (7.4)	3 (11.1)	27
West Midlands	Birmingham	6 (10.5)	20 (35.1)	17 (29.8)	9 (15.8)	5 (8.8)	57
East	Cambridge	0 (0)	7 (38.9)	6 (33.3)	4 (22.2)	1 (5.6)	18
North Thames	Gt Ormond St	- -	- -	- -	- -	- -	-
	Chelmsford	- -	- -	- -	- -	- -	-
The Spires	Oxford	1 (3.3)	13 (43.3)	8 (26.7)	5 (16.7)	3 (10)	30
	Salisbury	5 (17.2)	10 (34.5)	7 (24.1)	7 (24.1)	0 (0)	29
South Wales & South West	Swansea	0 (0)	1 (12.5)	4 (50)	2 (25)	1 (12.5)	8
	Bristol	1 (5)	8 (40)	4 (20)	6 (30)	1 (5)	20
South Thames	Guy's and St Thomas'	6 (9.7)	26 (41.9)	21 (33.9)	6 (9.7)	3 (4.8)	62
Northern Ireland	Belfast	- -	- -	- -	- -	- -	-
All	All	29 (8)	133 (36.5)	111 (30.5)	66 (18.1)	25 (6.9)	364

[a] 237/826(28.7%) children with an incomplete UCLP; 9/596 (1.5%) children who died before the age of five, and 223/587 (38%) children missing Five Year Old Index scores data excluded in 'All' values. Note: MCN – Managed Clinical Network.

[43] Note: Externally validated scores only were used in previous reports.
[44] Clinical Standards Advisory Group, Clinical Standards Advisory Group. Report of a CSAG Committee on cleft lip and/or palate, 1998, The Stationery Office, London.
[45] Submucous cleft palate patients excluded from all five year outcomes as all/most teams do not audit these patients.

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

For the second time, we are reporting speech outcomes assessed at five years of age. 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). 2013 was the first year that CAPS-A scores have been requested, and changes to the CAPS-A data collected were made recently in 2014. Several parameters of speech are assessed including:

- Resonance (hypernasality and hyponasality) and nasal airflow (audible nasal emission and nasal turbulence) (listed in Table 14). These are structurally-related speech difficulties 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 (listed in Table 15). These reflect articulation patterns which can affect the clarity and intelligibility of a child's speech. This set of 12 individual CSCs are being reported on in full, for the 2007 birth cohort, rather than the shortened version reported on last year.

Therefore, this section is reporting only on children born in 2007 – assessed across a total of 16 speech parameters.

Table 13 shows the number of consented children born in 2007 with reported speech outcomes and for whom exclusion reasons have been provided at five years of age, according to Administrative Unit (excluding children with submucous cleft palates[46]).

A total of 460 (63.8%) out of 721[47] consented children born with a CP, UCLP or BCLP in 2007 had at least one speech score reported across the 16 speech parameters. The scores for each assessed speech parameter can be seen in Tables 14 and 15.

There are reasons why speech scores are not reported for children, such as (1) when children's data is excluded – because of patients being deceased, having emigrated or transferred out of the area[48] – or (2) when children's data is not available; such as when the patient was not seen (e.g. due to a lack of staff or facilities) or it was not possible to take a record (e.g. for reasons unrelated to the patient). As can be seen in Table 13, these reasons accounted for 19.5% of reports on speech data.

Rates exclusion/non-availability (for children born in 2007), varied widely between Administrative Units; ranging from 51.1% of speech data being accounted for in Liverpool to 100% of speech data being accounted for in Belfast.

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

[47] 1,407 CRANE-registered consented children born between 2006 and 2007 with reported speech outcomes at five years of age (according to Administrative Unit) were reported in Table 9 – 686 were from 2006 and 721 from 2007.

[48] Plus: Clinically contraindicated (this record type for this patient) and other reasons.

Table 13. Number (%) of CRANE-registered[a] consented children born with a cleft palate in 2007, with reported speech outcomes and exclusion reasons at five years of age, according to Administrative Unit.

Regional centre / MCN	Administrative Unit	Speech[b]							
			Reported		Exclusion reason		Not Available Reason		Total
		N	n	(%)	n	(%)	n	(%)	(%)
Northern & Yorkshire	Newcastle	50	34	(68)	8	(16)	7	(14)	(98)
	Leeds	47	27	(57.4)	4	(8.5)	4	(8.5)	(74.5)
North West & North Wales	Liverpool	45	23	(51.1)	0	(0)	0	(0)	(51.1)
	Manchester	63	40	(63.5)	20	(31.7)	0	(0)	(95.2)
Trent	Nottingham	60	43	(71.7)	13	(21.7)	0	(0)	(93.3)
West Midlands	Birmingham	62	41	(66.1)	0	(0)	0	(0)	(66.1)
East	Cambridge	54	27	(50.0)	24	(44.4)	3	(5.6)	(100)
North Thames	Gt Ormond St	57	23	(40.4)	13	(22.8)	15	(26.3)	(89.5)
	Chelmsford	21	13	(61.9)	2	(9.5)	5	(23.8)	(95.2)
The Spires	Oxford	34	21	(61.8)	0	(0)	1	(2.9)	(64.7)
	Salisbury	39	26	(66.7)	0	(0)	4	(10.3)	(76.9)
South Wales & South West	Swansea	41	35	(85.4)	4	(9.8)	0	(0)	(95.1)
	Bristol	43	30	(69.8)	5	(11.6)	2	(4.7)	(86)
South Thames	Guy's and St Thomas'	72	50	(69.4)	0	(0)	1	(1.4)	(70.8)
Northern Ireland	Belfast	33	27	(81.8)	6	(18.2)	0	(0)	(100)
All	All	721	460	(63.8)	99	(13.7)	42	5.8%[49]	(83.4)

[a] Registered in CRANE by 19 September 2014. Note: MCN - Managed Clinical Network.
[b] Children born in 2007 only – excluding 237/958 (24.7%) children because: They were born a CL (24.2% of 958 cases) or a non-specified cleft type (0.5% of 958 cases); or they had submucous cleft palates (3%)[50]; or they had died before the age of 5 years (0.8%).

Resonance and Nasal Airflow

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

[49] There was an overlap of 14 cases with a code for 'Exclusion reason' and a code for 'Not available reason' to account for reasons why the speech outcomes were not collected – although completion of these sections should be mutually exclusive. Therefore those with an 'Exclusion reason' were excluded from the 'Not available reason' data.
[50] Submucous cleft palate patients excluded from all the outcomes as all/most teams do not audit these patients.

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

Description	Score	N[b]	(%)
RESONANCE – HYPERNASALITY			
Absent	0	349	(77.2)
Borderline – minimal	1	50	(11.1)
Mild – evident on close vowels	2	29	(6.4)
Moderate – evident on open and close vowels	3	13	(2.9)
Severe – evident on vowels and voiced consonants	4	11	(2.4)
Total		**452**	**(100)**
RESONANCE – HYPONASALITY			
Absent	0	384	(84)
Mild – partial dentalization of nasal consonants and adjacent vowels	1	65	(14.2)
Marked – dentalization of nasal consonants and adjacent vowels	2	8	(1.8)
Total		**457**	**(100)**
NASAL AIRFLOW – AUDIBLE NASAL EMISSION			
Absent on pressure consonants	0	425	(92.6)
Occasional: pressure consonants affected <10% of the sample	1	26	(5.7)
Frequent: pressure consonants affected >10% of the sample	2	8	(1.7)
Total		**459**	**(100)**
NASAL AIRFLOW – NASAL TURBULENCE			
Absent on pressure consonants	0	354	(77.6)
Occasional: pressure consonants affected <10% of the sample	1	81	(17.8)
Frequent: pressure consonants affected >10% of the sample	2	21	(4.6)
Total		**456**	**(100)**

[a] Registered in CRANE by 19 September 2014.
[b] Number of eligible children (as specified for Table 13) – excluding children with missing scores: 269/721 (37.3%) children missing hypernasality scores; 264/721 (36.6%) children missing hyponasality scores; 262/721 (36.3%) children missing audible nasal emission scores; 265/721 (36.7%) children missing nasal turbulence scores.

In terms of resonance, 5.3% of children with a hypernasality score had a score of '3' or '4', which means they had moderate or severe hypernasality i.e. nasal sounding speech (Table 14). This is indicative of velopharyngeal dysfunction (VPD), which is when the palate is unable to close off the nasal airway during speech. In addition, results of the Cleft Speech Characteristics (in Table 15) show that 2.6% of children had 'Weak and or nasalised consonants' passive articulation errors and 1.6% 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 – 91/ 418 (21.8%) of the children with reported surgical data[51] have had surgery for speech purposes (referred to as secondary speech surgery) before the age of 5 years.

In addition, out of the 449 children with a reported score for all 4 resonance and nasal airflow parameters listed in Table 14 (62.3% of eligible children and 97.6% of those with at least 1/16 speech scores reported[52]), 374 children (83.3%) had all green scores – indicating that no structural problems existed in relation to these parameters.

Cleft Speech Characteristics (CSCs)

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

[51] VP surgery/fistula repair data was only reported for 418/721 (58%) of eligible children (as specified for Table 13) – I.e. 42% of eligible cases did not have reported secondary speech surgery data.
[52] Of 721 eligible children born in 2007; and of the 460 children born in 2007 with at least 1/16 speech scores reported.

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

Cleft Speech Characteristics (CSCs)		Score	N[b]	(%)
ANTERIOR ORAL CSCs	1. Dentalisation / Interdentalisation	A	347	(81.3)
		B	80	(18.7)
		Total	**427**	**(100)**
	2. Lateralisation / Lateral	A	393	(91.8)
		B	19	(4.5)
		C	16	(3.7)
		Total	**428**	**(100)**
	3 Palatalisation / Palatal	A	334	(78.2)
		B	53	(12.4)
		C	40	(9.4)
		Total	**427**	**(100)**
POSTERIOR ORAL CSCs	4. Double Articulation	A	420	(98.4)
		B	7	(1.6)
		C	0	(0)
		Total	**427**	**(100)**
	5. Backed to Velar / Uvular	A	360	(84.3)
		C	21	(4.9)
		D	46	(10.8)
		Total	**427**	**(100)**
NON ORAL CSCs	6. Pharyngeal Articulation	A	423	(99.1)
		C	3	(0.7)
		D	1	(0.2)
		Total	**427**	**(100)**
	7. Glottal Articulation	A	391	(91.6)
		C	16	(3.7)
		D	20	(4.7)
		Total	**427**	**(100)**
	8. Active Nasal Fricatives	A	392	(91.8)
		C	21	(4.9)
		D	14	(3.3)
		Total	**427**	**(100)**
	9. Double Articulation	A	421	(98.6)
		C	4	(0.9)
		D	2	(0.5)
		Total	**427**	**(100)**
PASSIVE CSCs	10. Weak and or nasalised consonants	A	401	(93.9)
		C	15	(3.5)
		D	11	(2.6)
		Total	**427**	**(100)**
	11. Nasal realisation of plosives	A	412	(96.5)
		C	8	(1.9)
		D	7	(1.6)
		Total	**427**	**(100)**
	12. Gliding of fricatives	A	414	(97.2)
		C	11	(2.6)
		D	1	(0.2)
		Total	**426**	**(100)**

[a] Registered in CRANE by 19 September 2014.

[b] Number of eligible children (as specified for Table 13) – excluding children with missing scores: 293/721 (40.6%) children missing 'Lateralisation / Lateral' anterior oral CSC scores (#2 in the table above); 295/721 (40.9%) children missing 'gliding of fricatives' passive CSC scores (#12) ; and 294/721 (40.8%) children missing scores for all remaining ten CSC parameters.

'Palatalisation / Palatal' anterior oral CSCs were the most commonly occurring CSC, affecting 21.8% of children (12.4% with scores of B and 9.4% 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 425 children with a reported score for all 12 CSC parameters (58.9% of eligible children and 92.4% of those with at least 1/16 speech scores reported[53]), 279 children (65.6%) had all green scores – indicating they did not exhibit cleft speech characteristics.

Further to reporting on the 16 CAPS-A speech parameters separately, we anticipate reporting using the nationally agreed Speech Outcome Standards in the next CRANE Annual Report for both children born in 2007 and 2008. Specifically, we plan to report on

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)
3. The presence of cleft-related articulation difficulties (speech outcome standard #3)

However the data reported on in this chapter allows us to ascertain that, out of the 415 children with reported scores for all 16 assessed speech parameters listed in Table 15 (57.6% of eligible children and 90.2% of those with at least 1/16 speech scores reported[54]):

- 244 (58.8%) children had speech scores that would suggest their speech is within the normal range (all green scores) by the age of 5 years – and not substantially different to their non-cleft peer group
- While 92 (22.2%) children received at least one (red) score indicating a possible structural problem with the palate that may require further surgery.

[53] Of 721 eligible children born in 2007; and of the 460 children born in 2007 with at least 1/16 speech scores reported.
[54] Of 721 eligible children born in 2007; and of the 460 children born in 2007 with at least 1/16 speech scores reported.

4. Hospital Episode Statistics

In this section, we present some results of analyses of the data for children who have at least one HES record of an English NHS hospital admission with a diagnosis code for cleft palate (CP), unilateral cleft lip and palate (UCLP) or bilateral cleft lip and palate (BCLP); born between 1 January 1997 and 31 December 2004.

4.1. Secondary speech surgery

Cleft palates (CP) are usually repaired within the first year of life – as identified from HES analysis presented in the 2010 CRANE Annual Report[55] – and with a minimum of one surgical episode usually being required (separate from any related cleft lip (CL) repair). CP repair aims to restore the muscle anatomy of the palate to reduce the chance of future speech problems.

Sometimes additional surgeries involving the palate are needed to improve speech or to close any residual fistulae (a hole between the mouth and nose which can appear if the original repair breaks down), following primary CP repair. We refer to these as secondary speech surgery.

Our aim was to examine the instances of secondary speech surgery after first cleft palate repairs (primary palate repair) among children with a cleft palate (CP) up to the age of seven[56]. These included children with a unilateral cleft lip and palate (UCLP) and bilateral cleft lip and palate (BCLP).

We have presented instances of secondary speech surgery according to the presence and absence of additional anomalies or syndromes, by cleft type classification, by the child's age at the time of their surgeries, and by the region where they underwent their procedure.

4.1.1. All children with a cleft palate

We identified 982 children[57], born between 1 January 1997 and 31 December 2004 who:

- Had a 'primary cleft diagnosis' of CP, UCLP or BCLP; i.e. had at least one HES record with a diagnosis code for Orofacial Cleft (according to the International Classification of Diseases 10th Revision (ICD-10) codes Q35, Q36, Q37) [58].

[55] CRANE Project team on behalf of the Cleft Development Group. CRANE Database Annual Report 2010. London: Clinical Effectiveness Unit, The Royal College of Surgeons of England, 2010.
[56] By December 2011 – the date of the HES extract used – all children born in 2004 would be 7 years old.
[57] Live births, and cases still alive 1month after birth included. Deaths under 1month of age were excluded.
[58] Submucous cleft palate patients were not excluded as there is not a good enough linkage rate between HES and CRANE (from which submucous classification is calculated) to have accurate classification.

- Had undergone a primary repair of the cleft palate (according to the Office of Population, Censuses and Surveys Classification of Surgical Operations and Procedures 4th Revision (OPCS-4) code F291).
- Had undergone secondary speech surgery (according to OPCS-4 codes F292 for 'a revision of repair of cleft palate' and E21 for 'a repair of pharynx').
- Had received their second speech surgery more than 6 months after their primary palate repair[59].

4.1.2. Children with additional anomalies or syndromes by cleft type classification

Of the 982 children with a cleft palate we identified that 301 (30.7%) children had additional anomalies or syndromes – according to ICD-10 diagnosis and OPCS-4 procedure codes listed in Appendix 5. In addition, the majority had been diagnosed with a CP (56.6%) when compared to 24.9% of cases diagnosed with UCLP and 18.5% diagnosed with BCLP (see Table 16).

4.1.3. Number of secondary speech surgeries

Of the children who had undergone primary CP repair, 982 (18.3%)[60] were identified as having received secondary speech surgery. For some of these children, additional surgeries were needed to improve speech or to close any residual fistulae following their 1st secondary speech surgery.

Our cohort of 982 children underwent anywhere between 1 and 5 secondary speech surgeries; with only 20.3% of them needing to undergo more than one secondary speech surgery (see Table 16).

Table 16 shows the number and percentage of children who have undergone one or more secondary speech surgeries according to both their syndrome status and cleft type.

[59] Of a total of 1059 cases identified, 77 cases coded as undergoing secondary speech surgery within 0 to 6months were excluded to avoid reporting procedures related to postoperative complications resulting from the primary surgery.

[60] 5373 children born between 1 January 1997 and 31 December 2004 with a primary cleft diagnosis of CP, UCLP or BCLP had undergone primary CP repair.

Table 16. Number (%) of children born in England between 1997 and 2004, who have undergone 1 or more secondary speech surgeries according to syndrome status and cleft type.

Cleft type	N	Number of secondary speech surgeries N (%)			
		1		>1	
Children with additional anomalies or syndromes					
CP	235	188	(80)	47	(20)
UCLP	30	21	(70)	9	(30)
BCLP	36	26	(72.22)	10	(27.78)
All	301	235	(78.07)	66	(21.93)
Children without additional anomalies or syndromes					
CP	321	275	(85.67)	46	(14.33)
UCLP	214	168	(78.5)	46	(21.5)
BCLP	146	105	(71.92)	41	(28.08)
All	681	548	(80.47)	133	(19.53)
All children					
CP	556	463	(83.27)	93	(16.73)
UCLP	244	189	(77.46)	55	(22.54)
BCLP	182	131	(71.98)	51	(28.02)
All	982	783	(79.74)	199	(20.26)

CP – Cleft palate; UCLP – Unilateral cleft lip and palate; and BCLP – Bilateral cleft lip and palate.

As shown in Table 16, children with a syndromic cleft palate are more likely to undergo more than one secondary speech repair when compared to children with a non-syndromic cleft palate; and children with a BCLP are more likely to undergo more than one secondary speech repair.

4.1.4. Age at the time of palate surgeries

The average age for this cohort's primary CP repair was 11 months of age, and the average age for their 1st secondary speech surgery was 4 years and 1month of age – with the average number of years between children's primary CP repair and a secondary speech surgery being 3 years and 2 months.

Table 17 shows the number and percentage of children who have undergone secondary speech surgery, according to their age at the time of both their primary CP repair and a 1st secondary speech surgery, and shows that:

- 67.1% of all children have their secondary speech surgery before school age (> 5 years).
- The majority of those who have their primary repair under the age of 1 year, have their secondary speech surgery between the ages of 3 and 5 years (43.4%)

Table 17. Number (%) of children with a cleft palate diagnosis born in England between 1997 and 2004, who have undergone secondary speech surgery, according to their age at the time of their secondary and primary surgery.

Secondary speech surgery	0 to 1 years N (%)	>1 to ≤3 years N (%)	>3 to ≤5 years N (%)	>5 to <7 years N (%)	**Total N**
Primary CP repair					
0 to 1 years	12 (1.52)	203 (25.66)	343 (43.36)	233 (29.46)	791
>1 to ≤3 years	0 (0)	26 (19.7)	56 (42.42)	50 (37.88)	132
>3 to ≤5 years	0 (0)	0 (0)	19 (37.25)	32 (62.75)	51
>5 to <7 years	0 (0)	0 (0)	0 (0)	8 (100)	8
Total N of children	12 (1.22)	229 (23.32)	418 (42.57)	323 (32.89)	982

4.1.5. Region

Considering children approaching school age and the audit time point for speech nationally, the proportion of secondary speech surgeries conducted by the age of 5 years, ranged by region from 54.4% in the West Midlands to 77.8% in the North Thames region (as shown in Table 18).

In the case of the West Midlands most children undergo their secondary speech surgery between the ages of 5 and 7 years (45.6%); while most children in the North Thames and South & West undergo secondary speech surgery between the ages of 1 and 3 years (59.3% and 37.9% respectively). Most regions undertake these procedures between 3 and 5 years (such as the South Thames and London regions).

Table 18. Number (%) of children with a cleft palate diagnosis born in England between 1997 and 2004, who have undergone a secondary speech surgery, according to their age and region.

Region	**Secondary speech surgery (Age)**				
	0 to 1 years N (%)	>1 to ≤3 years N (%)	>3 to ≤5 years N (%)	>5 to <7 years N (%)	**Total N**
Northern and Yorkshire	0 (0)	17 (15.74)	**50 (46.3)**	41 (37.96)	108
Trent	0 (0)	18 (25.71)	**32 (45.71)**	20 (28.57)	70
Anglia and Oxford	1 (2.63)	12 (31.58)	**13 (34.21)**	12 (31.58)	38
North Thames	0 (0)	**16 (59.26)**	5 (18.52)	6 (22.22)	27
South Thames	0 (0)	5 (16.67)	**18 (60)**	7 (23.33)	30
South and West	2 (6.9)	**11 (37.93)**	6 (20.69)	10 (34.48)	29
West Midlands	0 (0)	28 (22.4)	40 (32)	**57 (45.6)**	125
North West	7 (3.14)	44 (19.73)	**108 (48.43)**	64 (28.7)	223
Eastern	1 (1.2)	22 (26.51)	**40 (48.19)**	20 (24.1)	83
London	0 (0)	16 (23.19)	**34 (49.28)**	19 (27.54)	69
South East	0 (0)	26 (20.63)	**52 (41.27)**	48 (38.1)	126
South West	1 (1.89)	14 (26.42)	**20 (37.74)**	18 (33.96)	53
All Regions	**12 (1.22)**	229 (23.34)	418 (42.61)	322 (32.82)	981 (100)

4.1.6. Summary and considerations

This analysis found that 30.7% of children who undergo a secondary surgical procedure for the palate have additional anomalies or syndromes; with syndromic CP or BCLP types of cleft increasing the risk of more than one secondary speech repair. Also, the majority of children had their secondary speech surgery before school age (of 5 years); with this pattern holding true for most regions – although the proportion of secondary speech surgeries conducted by school age varied substantially between some regions suggesting different patterns of delivery of care.

These figures have to be interpreted with some caution as, despite data quality reports and checks, coding issues can still be identified in the data. For example a total of 94 cases had to be excluded from this analysis as children were coded as undergoing their secondary speech surgery before their primary CP repair (in 17 cases) or coded as undergoing secondary speech surgery within 0 to 6 months of their primary CP repair (in 77 cases), which suggested either a second primary repair following adhesion surgery or a procedure related to postoperative complications rather than secondary procedures.

It should also be noted that this chapter reports on data between 1 January 1997 and 31 December 2004; including data from both before and after the reconfiguration of cleft services / centralisation of cleft services recommended in 1998[61] – therefore these results may change when more current cohorts are analysed. This analysis has demonstrated that these surgical procedures can be identified and warrants further work once an updated HES extract is available.

[61] Clinical Standards Advisory Group (1998). Cleft lip and / or palate. Report. HMSO, London.

5. National Pupil Database

In this section, we describe the results of a pilot project to link the CRANE records of children with a cleft lip and/or palate born between 1 January 2000 and 31 December 2008 to the National Pupil Database.

5.1. Introduction

Up until now, little has been understood about the impact of facial clefting on non-health outcomes such as educational achievement. This has been identified as a priority for cleft research from patient, carer and clinical perspectives[62].

The National Pupil Database (NPD), held by the Department for Education (DfE), holds a wide range of information about pupils who attend schools and colleges in England[63].

As described in the Progress Report 2014[64], CRANE received permission from the Department for Education to link the CRANE Database to the National Pupil Database at the individual pupil level.

In this section, we describe the results of the linkage exercise and make some preliminary comparisons between the educational outcomes at age 5 for the cleft cohort and the published national statistics for all children.

5.2. Methods

We aimed to match each consented CRANE registered patient from England born between 2000 and 2008 to their corresponding NPD identifier. We requested information about three educational assessment types, where available, for each child:

- Early Years Foundation Stage Profile (age 5)
- Key Stage 1 (age 7)
- Key Stage 2 (age 11)

[62] http://www.lindalliance.org/CleftPSP.asp // http://www.craniofacialsociety.org.uk/Launch_CleftPSP.PDF
[63] https://www.gov.uk/national-pupil-database-apply-for-a-data-extract
[64] CRANE Project team on behalf of the Cleft Development Group. CRANE Database Progress Report 2014. London: Clinical Effectiveness Unit, The Royal College of Surgeons of England, 2014.

We then explored:

- the success of the matching process
- the factors contributing to successful matching
- the utility of the dataset for making comparisons between the cleft cohort and published national statistics, focussing initially on the Early Years Foundation Stage Profile

5.2.1. Linkage

Personal identifiers (name, postcode and date of birth) were securely passed to the Department for Education, who performed the linkage between records and provided information about educational attainment at the different stages for the matched cases. This educational information was then merged by the CRANE team with the existing CRANE-HES linked dataset to provide information about factors such as the patient's cleft type and the presence of additional anomalies, as well as the treatment outcomes recorded in CRANE.

5.2.2. Early Years Foundation Stage Profile

The Early Years Foundation Stage Profile (EYFSP) is a National Curriculum teacher assessment of children's development at the end of the Early Years Foundation Stage, usually the academic year in which the child turns five. Further information is available from the Administrative Data Liaison Service[65].

The analysis was restricted to the six academic years 2006/2007 to 2011/2012. During this time, the EYFSP consisted of 13 assessment scales grouped into 6 areas of learning, scored from 0 to 9; with the maximum possible total score being 117.

For each academic year, we compared the mean total EYFSP score (across all 13 assessment scales) in the CRANE linked cohort with the corresponding national statistics for all children and for girls and boys separately. The analysis was restricted to children having no additional anomalies identified in their HES record[66].

We then described the mean total EYFSP score in the CRANE linked cohort according to cleft type by combining data across all six academic years.

[65] http://www.adls.ac.uk/department-for-education/dcsf-npd/?detail
[66] http://www.hscic.gov.uk/hes

5.3. Results

5.3.1. Linkage

Details of 7,152 eligible consented CRANE registered patients born between 1 January 2000 and 31 December 2008 were provided to the DfE and 56% of these could be linked to a NPD record. The NPD linkage rate did not vary across year of birth or by type of cleft. There was considerable variation in NPD linkage rates between the CRANE centres (39% - 87%). NPD linkage rates appeared to be correlated with the quality of postcode capture by CRANE.

5.3.2. Early Years Foundation Stage Profile

Table 19 shows the national mean total Early Years Foundation Stage Profile (EYFSP) score for each academic year from 2007 to 2012 and compares it to the mean for the corresponding cohort of non-syndromic cleft patients.

We see increasing mean scores across time for both the national and the cleft cohorts. On average, girls perform better than boys nationally and this is also reflected in the cleft cohort. The mean score for the cleft cohort is less than the national mean in each year for all children combined and for girls and boys separately. We refer to this difference as the 'attainment gap' between the cleft cohort and the national cohort.

Table 20 shows the national mean total EYFSP score for all academic years (2007-2012) combined and compares it to the mean for the corresponding cohort of non-syndromic cleft patients overall and by cleft type.

Over all years combined, the attainment gap for all children in the cleft cohort is 5.0 points. A similar attainment gap between the cleft cohort and the national cohort can be seen when girls and boys are considered separately. The attainment gap between the cleft cohort and the national cohort varies according to the type of cleft, CL being associated with the smallest attainment gap and clefts involving the palate (CP, UCLP, BCLP) with larger attainment gaps. This is the case for both girls and boys.

These results suggest that children with a cleft have poorer educational outcomes on average than their peers nationally and that, as expected, children with a cleft involving the palate fare worse on average than those with a cleft involving the lip only.

We would caution that these results are very preliminary and should be considered as an exploratory step towards demonstrating the value of using educational results as an outcome measure for cleft patients.

There are many considerations to bear in mind, such as which of the many educational assessment types that are collected would represent the most appropriate outcome measure for the cleft

population and whether our linked cleft cohort is representative of the cleft population as a whole. We must also account for trends over time and school effects.

Future analysis is planned to look in detail at the impact of clefting on specific subscales of the EYFSP – for instance we expect the type of cleft to impact especially on the communication, language and literacy component. We will also examine KS1 and KS2 results.

We will apply a methodology that, as well as possible, addresses a number of deficiencies in the current results (e.g. using regression techniques to combine data while allowing for differences between boys and girls after having transformed the results into z-scores to adjust for systematic differences in outcomes over academic years).

Table 19. Children's Early Years Foundation Stage Profile (EYFSP) mean total scores in England from 2007 to 2012: National means versus CRANE cohort means, by academic year.

	Mean score for ALL children Across all 13 assessment scales			Mean score for GIRLS Across all 13 assessment scales			Mean score for BOYS Across all 13 assessment scales		
	National Mean	CRANE Mean*	Att Gap vs Nat Mean	National Mean	CRANE Mean*	Att Gap vs Nat Mean	National Mean	CRANE Mean*	Att Gap vs Nat Mean
2007	85.4	80.5	4.9	88.3	83.0	5.3	82.5	78.5	4.0
2008	85.7	79.6	6.1	88.4	80.5	7.9	83.0	78.9	4.1
2009	86.1	80.5	5.6	89.0	84.5	4.5	83.6	77.6	6.0
2010	86.9	81.8	5.1	89.6	83.3	6.3	84.5	80.9	3.6
2011	87.5	84.0	3.5	90.1	87.6	2.5	85.1	81.6	3.5
2012	88.6	83.7	4.9	91.3	88.7	2.6	86.0	80.2	5.8
All years	86.7	81.7	5.0	89.5	84.7	4.8	84.1	79.7	4.4

* Non-syndromic cleft patients

Table 20. Children's Early Years Foundation Stage Profile (EYFSP) mean total scores in England from 2007 to 2012: National mean versus CRANE cohort mean for all academic years combined, by cleft type.

	ALL CHILDREN				ALL GIRLS				ALL BOYS			
	N	Mean	Att Gap vs Nat Mean	95% CI	N	Mean	Att Gap vs Nat Mean	95% CI	N	Mean	Att Gap vs Nat Mean	95% CI
National		**86.7**				**89.5**				**84.1**		
CRANE*	1779	81.7	5.0	(80.7 to 82.7)	729	84.7	4.8	(83.3 to 86)	1050	79.7	4.4	(78.4 to 80.9)
Cleft Type												
CL	464	85.1	1.6	(83.5 to 86.7)	167	87.9	1.6	(85.5 to 90.2)	297	83.5	0.6	(81.4 to 85.6)
CP	616	80.1	6.6	(78.4 to 81.9)	339	83.8	5.7	(81.7 to 85.8)	277	75.7	8.4	(72.9 to 78.5)
UCLP	492	81.7	5.0	(79.8 to 83.5)	165	83.3	6.2	(80.1 to 86.5)	327	80.8	3.3	(78.6 to 83)
BCLP	181	78.9	7.8	(76.1 to 81.6)	50	85.7	3.8	(80.7 to 90.6)	131	76.3	7.8	(73.1 to 79.4)

* Non-syndromic cleft patients

6. Development of CRANE Database and future directions

6.1. Data sources and future analyses

6.1.1. National Pupil Database (NPD)

As described in Chapter 5, details of 7,152 eligible consented CRANE registered patients born between 1 January 2000 and 31 December 2008 were provided to the Department for Education and 56% of these could be linked to a NPD record; with NPD linkage rates appearing to be correlated with the quality of postcode capture by CRANE.

We will investigate and pursue methods to improve the accuracy of postcode data and seek to improve this 56% linkage rate for future analysis.

6.1.2. Hospital Episode Statistics (HES)

The Clinical Effectiveness Unit has recently received a new HES extract – containing hospital data updated up until April 2014 – which we will link to CRANE. This will allow us to refresh our analyses, which have involved HES data, such as those on examining mortality, secondary speech surgery, grommets and the NPD.

6.1.3. Newborn Hearing Screening Programme (NHSP)

We are exploring the option of requesting linkage between our CRANE Database and the Newborn Hearing Screening Programme (NHSP)[67] 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.

6.1.4. 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.

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

6.1.5. Mapping boundaries of Regional Cleft Networks

We plan on attempting to create a map of the boundaries of all Regional Cleft Networks using CRANE data; aiming to draw from the experience of colleagues within the Clinical Effectiveness Unit, such as Prostate Cancer Audit project team who are conducting similar work. This will be useful information for Commissioners about this group of patients whose treatment they fund.

6.2. 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.

6.2.1. Speech

Cleft Audit Protocol for Speech – Augmented (CAPS-A) scores have been used to assess speech among 5 year old children with a cleft affecting the palate (CP, UCLP and BCLP) and were collected by CRANE for the first time last year (for 2006 births).

This year, and for the second time, we collected speech outcomes; but changes to the CAPS-A data collected by CRANE were made recently in 2014. Therefore we only reported on children born in 2007 (see section 3.6.5). Specifically, CRANE now collects data to assess a total of 16 speech parameters including:

- Resonance (hypernasality and hyponasality) and nasal airflow (audible nasal emission and nasal turbulence) (listed in Table 14). These are structurally-related speech difficulties 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 (listed in Table 15). These reflect articulation patterns which can affect the clarity and intelligibility of a child's speech.

Further to reporting on the 16 CAPS-A speech parameters, we anticipate reporting using the nationally agreed Speech Outcome Standards in the next CRANE Annual Report for both children born in 2007 and 2008.

6.2.2. Notification data

From January 2014 we introduced small changes to data collection for notification data around birth to allow us to collect the same information as reported in Quality Dashboard. In August 2014 we received Confidentiality Advisory Group (CAG) approval to expand the notification dataset for non-consented cases to include all nine 'first contact information' timing fields.

We will monitor the progress of data collection with these changes, and plan on fully reporting on 'first contact information' timing fields for non-consented cases in next year's reports.

6.2.3. 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 RCPCH national screening guidance[68]. Preliminary analysis including data on this timing can be seen in Section 3.4. We anticipate being able to report on this data more meaningfully in next year's reports.

6.2.4. Patient and parent satisfaction

The Cleft Psychology Special Interest Group (SIG) were asked by the Craniofacial Society of Great Britain and Ireland (CFSGBI) Council to identify and pilot measurements to evaluate how patient and parent satisfaction could be measured nationally. The following were identified as potential measures: (1) the Friends and Family Test (FFT – developed by the Department of Health) and (2) the Experience of Service Questionnaire (CHI-ESQ – developed by the Commission for Health Improvement (CHI)) satisfaction assessment scales. The CRANE Database project team have drafted a proposal – to the Cleft Development Group (CDG) – to conduct a feasibility study to test how best to collect, analyse and report on this PREM data nationally.

6.2.5. Patient Reported Outcome Measure

An international study is developing a patient reported outcome measure questionnaire for Cleft-Lip and/or Palate Patients aged 8 years and older; known as CLEFT-Q. The study team has been inviting comments on their current draft (until the end of October 2014).

Because of the immense value of this work, CRANE offered to host the current version of the CLEFT-Q 'Patient Version feedback document' on our CRANE Database website; so that clinicians and patients could provide feedback on the draft CLEFT-Q.

Once the CLEFT-Q has been finalised in 2015, the CRANE Database project team plans develop a method for collecting this data as part of our core dataset.

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[68] In line with a statement of the UK National Screening Committee Newborn and Infant Physical Examination 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).

6.3. CRANE Database and website

6.3.1. CRANE Website

We have been making updates to the CRANE website; such as revising the information on the CRANE "Information for Parents" web page. This has been edited for clarity and the updated version is now called "Information for Parents and Carers" to provide information on:

- How CRANE data been used to improve children's care so far
- How CRANE plans to continue to influence children's care – with a positive quote endorsing this work, by Rosanna Preston, Chief Executive of the Cleft Lip & Palate Association (CLAPA).
- CRANE linkage to National Pupil Database (NPD) data – with clear guidance on participation.

This information has the aim of helping to allay and respond to any concerns around data confidentiality and the right to opt out of the linkage.

In addition, in order to communicate submission deadlines and updates to the database to cleft teams, we have added a page to the CRANE website containing "Information for Cleft Teams". This is a non- restricted page of the CRANE website, which was flagged to all teams via email communication[69].

6.3.2. CRANE Database

We are reaching the point where CRANE's database and website IT systems need to be updated. This is because the current system is becoming out-dated (it will be 10-years-old in 2015); and experiences limitations stemming from the approach taken when the system was built in 2005. Specifically:

- We currently have a website that cannot be amended easily and reactively by the project team in response to demand for information (direct amendment of websites is now commonplace for most current/new register sites).
- We also have a database that is limited in how it can be expanded to collect new data such as additional information around outcome measures.
- Furthermore, the current database cannot accommodate a Picture Archive feature, for instance, which we aspire to incorporate into CRANE. The Picture Archive feature would facilitate a national process for valid and reliable outcome assessments by panels of calibrated examiners, to allow them to score images of clefts in a secure web based platform, adding value for CRANE users. The picture archive would replicate some of the functionality of the Birmingham Institute of Paediatric Plastic Surgery (BIPPS) web-based version of the 'Oxford Aesthetic Audit' survey. We are currently seeking to explore the following considerations with the BIPPS team: (1) the burden of resource and cost required to create such a system, (2) the ease of uploading new images and related

[69] http://www.crane-database.org.uk/info_cleftteams/

functionality, and (3) the potential future proofing of this type of system to cope with year-on-year increases in storage/capacity.

6.4. Clinical Reference Group (CRG)

The inclusion of submitting data to CRANE as a requirement to the D07/S/a National Service Specification (Cleft Lip and or Palate Services including Non-cleft Velopharyngeal Dysfunction (VPD) (All Ages)), developed by the CRG, is a welcome advance in helping to improve the quality and completeness of data held in the CRANE database. CRANE is exploring ways to develop communication and links with cleft teams that should also 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.

6.4.1. Quality Dashboard

The project team have submitted data for the quality dashboard in November 2013, February 2014 and April 2014. The data required for the Cleft Lip and Palate Quality Dashboard includes:

- Measure Number CLP00: The number of CRANE-registered children born within a specified calendar year period (refreshed every quarter). E.g. Data submitted for the April 2014 was for children born 01 July 2012 to 30 June 2013.
- Measure Number CLP06: The number of 5 year old children with a decayed, missing, filled teeth (dmft) index score – as a percentage of all 5 year old children (annual).
- Measure Number CLP007: CRANE will provide data for this in the future - The number of 5 year old children with green Cleft Audit Protocol for Speech – Augmented CAPS-A scores[70] – as a percentage of the number of 5 year old children with a CAPS-A score (annual).
- Measure Number CLP08: The number of 5 year old children with five year old index scores 1 or 2[71] – as a percentage of the number of 5 year old children with a five year old index score (annual).

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

[70] Scores colour-coded as green indicate that 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. (CRANE Project team on behalf of the Cleft Development Group. CRANE Database Annual Report 2013. London: Clinical Effectiveness Unit, The Royal College of Surgeons of England, 2013.)

[71] Atack NE, Hathorn IS, Semb G, Dowell T, 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.

6.5. Public Health England

CRANE has been involved in a scoping exercise across existing congenital anomaly registers and disease specific registers by Public Health England in 2013 and 2014. This is to examine the feasibility of developing a national congenital anomaly register that will capture all congenital anomalies in England. We await the decision from Public Health England but have expressed our intention to comply with any data submission requirements to this project.

6.6. 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 set up a working group to develop a best practice guide and an e-learning module on the palate examination in the neonate. The overall aim is to increase the proportion of timely detections of CP by promoting a visual technique of examination of the mouth and palate, supplemented by palpation where appropriate, as well as to aid clinical awareness by alerting all health care professionals responsible for the newborn examination to symptoms associated with cleft palate. The RCPCH working group launched in November 2013 – including key partners such as the CRANE Database project team – are currently consulting on best practice guidance to aid healthcare professionals in the identification of cleft palate in neonates; and improve and standardise routine postnatal examination of the palate. The expected publication date is October 2014.

- CRANE has agreed to share data with other registers affiliated with the British Isles Network of Congenital Anomaly Registers (BINOCAR), with the aim of improving the completeness of anomaly reporting.

- CRANE will be involved with a multidisciplinary group from the CFSGBI evaluating previously collected national speech data to identify possible risk adjustment factors for the speech outcome data which could be utilised when reporting surgeon- or team-specific data in the future.

- 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 and many are being invited to take part in clinical trials and other studies coordinated by the Clinical Trials Unit, at the University of Manchester and the Royal Manchester Children's Hospital. The Centre for Appearance Research at the University of the West of England will be working on the psychological issues associated with cleft lip and palate and the support needed by families and children. We are currently working with the Cleft Collective team to establish whether CRANE could share data with this research project.

- We are also exploring ways to support feasibility studies conducted by The Healing Foundation Cleft and Craniofacial Clinical Research Centre supported by VTCT based at the University of Manchester
- Developing an Outcome Measure for Aesthetics at 5 years of age following lip repair. This Outcome Measure would be based on pilot work carried out by the Birmingham Institute of Paediatric Plastic Surgery (BIPPS) of an online version of the 'Oxford Aesthetic Audit Scoring System'. This audit tool, developed by a surgeon in Oxford, allows the review and scoring of images of children's lip repairs.

6.7. CRANE Communications

6.7.1. Dissemination of 2014 findings

- Publication of the Annual Report will announced via our regular eNewsletter (launched in April 20014) which will be circulated in early November, and our website.
- We will also work with our close collaborators – such as the CFSGBI and CLAPA – to expand the reach of our eNewsletter (and the report).
- Further to this, we will be drafting a 'Parent and Young People / Easy Access version' of the findings after the 2014 Annual Report has been completed, signed off and published – this will be carried out in collaboration with CLAPA and potentially published in late 2014 / early 2015.

6.7.2. Addressing equality and diversity

During 2014, forms and patient information leaflets for gaining consent were translated into the most common languages identified with cleft teams in 2013. This was to acknowledge the diversity of languages spoken by patients and family accessing cleft services, and expand opportunities for access to information about CRANE.

We will undertake a similar exercise later in 2014 to identify next year's program of translations required. Our aim is undertake 3 of these each year to spread the overall costs each year until we have covered the more common languages encountered by teams.

6.7.3. Publications and presentations related to the CRANE Database

Poster presentations

- Fitzsimons K, Copley L, van der Meulen J, Panagamuwa C, Deacon S. "Surgical management of otitis media with effusion in children with cleft lip and/or palate born in England between 1997 and 2005" Craniofacial Society of Great Britain and Ireland (CFSGBI) Annual Conference (April 2014)

Oral Presentations

- Copley L, Medina J, Deacon S, van der Meulen J. “Crane database – feasibility of linkage to the National Pupil Database to explore long term educational outcomes in patients with a cleft lip and / or palate” British Isles Network of Congenital Anomaly Registers (BINOCAR) Biennial Scientific Meeting (October 2014).

Publications

The following paper is awaiting publication:

- Fitzsimons K, Copley L, van der Meulen J, Panagamuwa C, Deacon S. “Grommet Surgery in Children with Orofacial Clefts in England”.

7. 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 1,121 children born with a cleft in 2013 had been registered on CRANE at the time of preparing this report; with CRANE receiving the highest number of registrations for births in 2012 and 2013 since 2008. High numbers should not be interpreted as an increase in clefting incidence, but, instead, it represents the improved function of the database as a national register of cleft births. The number of registered cleft births in 2013 equates to an incidence of approximately one in every 645 live births in England, Wales and Northern Ireland[72].

Although children can now be registered with CRANE prior to obtaining parental consent, consent must still be obtained so that complete data, including outcomes, can be collected and reported by CRANE. The consent rate is very high among patients who have been through the consent process, which is encouraging. However, almost one fifth of the children born in 2013 had not been consented at the time of preparing this report. Further, this proportion ranged from 0% to 69.2% 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. A few Units have provided outcome data for more than 90% of their eligible patients, suggesting that the reporting of outcomes is feasible. CRANE is exploring ways to improve communication and links with Units to improve the submission of data in the future.

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 will 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:

[72] 24,279 Births in NI in 2013 – Available from Northern Ireland Statistics & Research Agency (NISRA): http://www.nisra.gov.uk/demography/default.asp8.htm. // 698,512 Births in England & Wales in 2013 – Available from the Office for National Statistics. Characteristics of Birth 1, England and Wales - 2013: http://www.ons.gov.uk/ons/rel/vsob1/characteristics-of-birth-1--england-and-wales/2013/index.html

Diagnosis, Referral and Contact

1. Antenatal diagnosis rates of cleft lip, with or without cleft palate, are still falling below the NHS Fetal Anomaly Screening Programme target detection rate of 75%[73].

2. Just under a third of children (30.4%) with a cleft palate alone 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[74]. This proportion has increased since the previous year.

3. 81.9% of the children born in 2013 with a cleft were referred to a Cleft Unit within 24 hours of birth. This proportion varied substantially according to the Unit receiving the referrals (ranging from 66.7% to 90.6%). Although rates have improved substantially since 2012, prompt referral is still recommended to ensure that the baby and their family receive appropriate care and support as soon as possible. Once referred, Cleft Units established contact with 92.8% of families within 24 hours, which is encouraging.

Cleft-related outcomes at five years

4. 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, treatment indices of 67.2 to 100% across Administrative Units indicate that – in the majority of cases – Units have mechanisms in place to deal with any disease occurring.

5. One quarter of children with a complete UCLP have poor dental arch relationships 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[75].

6. Almost one quarter of children (22.2%) with a complete speech assessment received at least one score indicating a possible structural problem with the palate that may require further surgery.

Secondary Speech Surgery

As a result of our analyses of HES data linked with CRANE data, we have been able to report on secondary speech surgeries for 982 children according to the presence and absence of additional anomalies or

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[73] 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.
[74] Bannister, P, Management of infants born with a cleft lip and palate. Part 1. Infant, 2008. 4(1): p. 5-8.
[75] Clinical Standards Advisory Group, Clinical Standards Advisory Group. Report of a CSAG Committee on cleft lip and/or palate, 1998, The Stationery Office, London.

syndromes, by cleft type classification, their age at the time of their surgeries, and by region where they underwent their procedure. Although these data have limited implications for clinical practice, they should be of interest to those involved in the care of children with a cleft lip and/or palate. The key findings were:

7. 18.2% of the total children required a secondary surgical procedure of whom 30.7% had additional anomalies or syndromes; with syndromic CP or BCLP types of cleft increasing the risk of more than one secondary speech repair.

8. The majority of children had their secondary speech surgery before school age (of 5 years); with this pattern holding true for most regions – although the proportion of secondary speech surgeries conducted by school age varied substantially between some regions suggesting different patterns of delivery of care.

9. These figures have to be interpreted with some caution as, despite data quality reports and checks, coding issues can still be identified in the data.

Education achievement at five years

As a result of our analyses of NPD data linked with CRANE and HES data, we have been able to describe the results of the linkage exercise, and make some preliminary comparisons between the educational outcomes at age 5 for the cleft cohort and the published national statistics for all children at the age of 5. The key findings were:

10. Details of 7,152 eligible consented CRANE registered patients born between 2000 and 2008 were available for linkage with NPD, and 56% of these could be linked to a NPD record.

11. The NPD linkage rate did not vary across year of birth or by type of cleft. There was considerable variation in NPD linkage rates between the CRANE centres (39% - 87%). NPD linkage rates appeared to be correlated with the quality of postcode capture by CRANE.

12. Over all academic years of 2007 to 2012 combined, the 'attainment' gap for all children in the non-syndromic cleft cohort is 5 points lower than the national mean total score. The attainment gap between the cleft cohort and the national cohort also varies according to the type of cleft, with cleft lip (CL) being associated with the smallest attainment gap and clefts involving the palate (CP, UCLP, BCLP) with larger attainment gaps. These difference in 'attainment gap' between the cleft cohort and the national cohort are also seen when girls and boys are considered separately.

13. This preliminary comparison of mean total scores suggest that children with a cleft have poorer educational outcomes on average than their peers nationally and that, as expected, children with a cleft involving the palate fare worse on average than those with a cleft involving the lip only.

Cleft Units should review the findings in this report and identify areas in which local improvements are required to help ensure the provision of high quality care for children with a cleft.

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
Lynn Copley	Data Manager	Clinical Effectiveness Unit
Jan van der Meulen	Clinical Epidemiologist	Clinical Effectiveness Unit; London School of Hygiene and Tropical Medicine
Jackie Horrocks	CRANE Administrator	Clinical Effectiveness Unit

Appendix 2: Governance and funding

Ownership

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

Cleft Development Group

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

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

Funding

Funding of the CRANE Database is currently coordinated and agreed by representatives of the national Specialised Commissioning Group for England and the Wales Specialised Health Services Committee. Funds are raised through a levy calculated on a weighted per capita basis from the commissioning bodies in England and Wales. The levy is currently collected by Derbyshire County PCT.

Appendix 3: Members of the Cleft Development Group

Members of the Cleft Development Group (CDG)

Stephen Robinson	Chair / Clinical Director Spires Cleft Service
Liz Albery	Clinical Director North Bristol NHS Trust
Geoffrey Carroll	Medical Director, Wales Health Specialised Services Committee
Mechelle Collard	Paediatric Dentistry (Special Interest Group (SIG) CFSGBI)
Michele Davis	Regional Programme of Care Manager London
Scott Deacon	CRANE Clinical Project Lead
Mark Devlin	Clinical lead for the Scottish Network
David Drake	Cleft Surgery Training Interface Group
Sue Gregory	Department of Health (Deputy CDO for England)
Piet Haers	Clinical Lead, South Thames Cleft Service
Per Hall	Cleft Surgeon, British Association of Plastic, Reconstructive and Aesthetic Surgeons
Chris Hill	Northern Ireland Clinicians
Peter Hodgkinson	Clinical Lead, Newcastle Site, Northern and Yorkshire Cleft Service & Chair Cleft Centres
Jackie Horrocks	Minutes Secretary, CRANE/Clinical Effectiveness Unit
Nichola Hudson	Specialist Cleft Nurses (SIG CFSGBI)
Loshan Kangesu	Clinical Lead, North Thames Cleft Service
David Landes	North of England Dental Public Health Consultant
Kate Le Maréchal	Clinical Psychologists (SIG CFSGBI) / President CFSGBI
Fiona Mackison	South East Coast SCG
Fiona Marley	National Specialised Commissioning Group
David Orr	Cleft Services in the Republic of Ireland
Norma Patterson	CDs and Managers Group
Marie Pinkstone	Lead Speech & Language Therapists Group
Rosanna Preston	CLAPA Chief Executive
John Rowson	Clinical Director, Trent Cleft Service
Jonathan Sandy	Lead, Cleft Collective Birth Cohort and Gene Bank Study
William Shaw	Manchester Lead, Cleft Collective Birth Cohort and Gene Bank Study
Rona Slator	Clinical Director, West Midlands Cleft Centre
Jackie Smallridge	Consultant Paediatric Dentist, South Thames Cleft Service / President CFSGBI
Alistair Smyth	Cleft Surgeon (British Association of Oral and Maxillofacial Surgeons) / Clinical Lead, Leeds Site, Northern and Yorkshire Cleft Service
David Steel	Programme Director, National Services Division, NHS Scotland
Adrian Sugar	Wales Clinicians

Jan van der Meulen	Clinical Epidemiologist, CRANE/Clinical Effectiveness Unit
Simon van Eeden	Clinical Lead, North West, IoM & North Wales Cleft Network
Mike Winter	Medical Director, National Services Division, Scotland
Ken Wragg	East Midlands Dental Public Health Consultant

Appendix 4: Terms of Reference for the Cleft Development Group

The Origins of the Cleft Development Group (CDG)

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

The Roles of the CDG

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

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

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

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

The database was UK wide when run by the Craniofacial Society of Great Britain and Ireland and before it became the responsibility of the CRANE Levy Board. Devolution of government in the UK resulted in 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, demographic and epidemiological data related to children born in England and Wales with the congenital abnormality of clefting of the lip and/or palate, and where possible extending this to the whole of the UK and Ireland

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

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

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

Composition of the Cleft Development Group

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

The composition will be:

1. Commissioners of Cleft Care. These should include at least two commissioners from Specialist Commissioning Groups in England (nominated by the National Specialist Commissioning Group for England), one from Wales, one from Scotland and one from Northern Ireland (each nominated by

their equivalent national specialist commissioning body). It is intended that there should be no more than six specialist commissioners in total to be agreed and appointed by the bodies which contribute data to the database (in the case of Scotland by sharing its data with CDG). Only those commissioning groups which pay the levy may vote on issues relating to CRANE.

2. Public Health Consultants. These should include representatives of commissioning areas who are actively involved in cleft commissioning, and will normally be Consultants in Dental Public Health. There should be at least two (to be nominated by the BASCD Consultants in Dental Public Health Group).

3. A Lay representative from a Parent Support Group (1) (to be nominated by CLAPA)

4. Cleft surgeons (2) (presently one nominated by BAOMS and one by BAPRAS)

5. The President of the Craniofacial Society of Great Britain and Ireland

6. The Chair of the Cleft Surgery Training Interface Group

7. A Speech & language therapist (1) (to be nominated by the Lead Cleft Speech and Language Therapy Group)

8. An Orthodontist (1) (to be nominated by the Cleft Orthodontists Special Interest Group).

9. A Specialist Cleft nurse (1) (to be nominated by the Cleft Nurses Special Interest Group)

10. A Psychologist (1) (to be nominated by the Cleft Psychologists Special Interest Group)

11. A Paediatric Dentist (1) (to be nominated by the Cleft Paediatric Dentist Special Interest Group)

12. The Co-ordinator/Chair of the UK Cleft Centres Clinical Directors' Group (1)

13. A Cleft Co-ordinator (1) (to be nominated by the Cleft Coordinators Special Interest Group).

14. A Representative from the group of 'other' specialities involved in cleft care (1) (to be nominated by CFS Council).

15. A Clinical representative from Northern Ireland (1) / Scotland (1) / Wales (1) / England (as appropriate, if not already represented) (to be nominated by those countries)

16. There may be representation, as determined by CDG to be appropriate, of any national bodies representative of Audit (1) and Research (1)

17. Clinical Directors/Clinical Leads of UK Cleft Centres not otherwise represented on CDG shall be invited to attend and become voting members so that all centres will be represented.

18. The Clinical Director/Project Leader of the CRANE service will be in attendance at Group meetings to which he/she will report, except when required to be absent because their own position is being discussed/decided. This individual will not be a voting member of the Group unless in another capacity and will not be eligible to become Chair.

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

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

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

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

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

Meetings

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

Amended May 2012

Appendix 5: Diagnosis and procedure codes, Hospital Episode Statistics

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

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
	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

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Code	Description
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

Classification of Surgical Operations and Procedures 4th Revision (OPCS-4) codes used to define primary and secondary cleft palate surgery.

Primary cleft palate surgery

Code	Description
F291	Primary repair of cleft palate

Secondary cleft palate surgery

Code	Description
F292	Revision of repair of cleft palate
E21	Repair of pharynx (Includes: Nasopharynx) – Including:
E21.1	Pharyngoplasty using posterior pharyngeal implant
E21.2	Pharyngoplasty using posterior pharyngeal flap
E21.3	Pharyngoplasty using lateral pharyngeal flap
E21.4	Plastic repair of pharynx NEC
E21.8	Other specified
E21.9	Unspecified