CRANE Database

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Annual Report on cleft lip and/or palate 2012

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
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
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.
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.
National Information Governance Board (NIGB)	An independent statutory body established to promote, improve and monitor information governance in health and adult social care. http://www.nigb.nhs.uk
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 these disorders 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 ongoing analyses of the CRANE Database, examining trends in registrations and the timing of cleft diagnosis, patient referral to Administrative Units and first contact between Administrative Units and the parents of children born with a cleft. It focuses primarily on children born in 2011.

For the second year running, we present information on cleft-related outcomes for children at five years of age (born 2004-2006). The completeness of these data has improved substantially since last year, which has allowed more meaningful analyses.

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, and receiving surgical treatment for, cleft lip and/or palate. We have developed our work on quantifying hospital care among children with cleft abnormalities. Preliminary analyses in this area, which focused on admissions for all causes in the first two years of life among patients without additional anomalies or syndromes, were presented in last year's report. This year, we have examined differences in

total time spent in hospital between those with and without additional anomalies, and we have examined cleft-related procedure admissions and emergency admissions according to the absence or presence of additional anomalies or associated syndromes, cleft type and Administrative Unit.

As an extension of this work, we are now looking at admissions for dental disease up to the age of seven years. Our preliminary analyses are presented in this report.

Key findings

Children born with a cleft lip and/or palate in 2011

Overall, 10,467 children born between 1 January 2000 and 31 December 2011 with a cleft lip and/or palate were registered on the CRANE Database by 3 September 2012. Of these, 832 were born in 2011. It is estimated that a further 250 patients born in 2011 will be registered in due course. CRANE case ascertainment is very high, being around 95%, according to comparisons with HES and Patient Episode Data Wales (PEDW) [3]. The parental consent rate for 2011 births is 98.8% (ranging from 82% to 100% between Units), according to reported figures from the Administrative Units.

Among children born in 2011, CRANE analyses revealed:

- 42% of children with clefts were diagnosed in the antenatal period through screening, which is the highest proportion since we started collecting the time of diagnosis.
- Only 1% of children with cleft palate only (CP) were diagnosed during antenatal screening; 71% were diagnosed at birth, leaving 28% who were diagnosed late according to the national standard [4]. This figure has improved by 4% since last year. Five per cent of children with a cleft palate alone are diagnosed after one month of age.
- 54% of children were referred by a maternity unit to an Administrative Unit within 24 hours of birth. This compares to 58% in 2010.
- Referrals from maternity units within one day of birth varied from 31% to 77% according to the Administrative Unit receiving the referral. Some regions have seen substantial improvements in this area over the last year.
- Administrative Units established contact with 90% of parents within 24 hours of their child's referral. This has not changed 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 2006. These outcomes include height and weight, the number of decayed, missing and filled teeth (a measure of oral health), and Five Year Old Index scores (2004-2005 births only), which can be used to assess dental arch relationships as a sign of how successful surgery has been. Although there is still a high proportion of missing data, there have been great improvements in the reporting of some of these outcomes since last year. For those children with reported outcomes:

- 42% had at least one decayed, missing or filled tooth (≥1 dmft), which, although still significant, is only slightly higher than the background rate in the general population. The proportion of children with ≥1 dmft varied significantly according to cleft type. BCLP was associated with the poorest oral health, with 50% of BCLP patients having ≥1 dmft at five years of age.
- Of the 147 children with a complete unilateral cleft lip and palate (UCLP) who had externally validated Five Year Old Index scores, 25% had scores of '4' or '5', reflecting poor dental arch relationships.

Unfortunately, outcomes are still not collected consistently across Units:

- Height and weight measures are not collected by five of the fifteen Administrative Units.
- The dmft score was not reported by two Administrative Units. Of these, Cambridge reported not having a paediatric dentist, who is required to determine the dmft score.
- The Five Year Old Index score was not collected by four Administrative Units. Of these, Belfast reported that their Orthodontist does not routinely see patients at five years of age.

Hospital admissions for children with a cleft lip and/or palate

We analysed HES data to examine hospital admissions and the total number of days spent in hospital by 10,892 cleft patients born between 1997 and 2008 who were treated in England. We focused on admissions in the first two years of life, as our preliminary analyses showed that the majority (~75%) of admissions in the first six years of life occurred before the age of two. The main findings are outlined below.

Admissions for all causes

- Non-syndromic children (children without a syndrome or associated medical condition) with a cleft had a median average of three admissions for all causes (including the birth episode) and spent a total of 10 days in hospital before the age of two.
- Syndromic children (children with additional complicating medical conditions or syndromes) with a cleft , who account for approximately 22% of all cleft patients, had a median of five all-cause admissions and spent 27 days in hospital, almost three times as long as non-syndromic children.

Admissions for cleft-related procedures

- Admissions for cleft-related procedures accounted for 37% of all-cause hospital admissions and 30% of the total time spent in hospital before the age of two years.
- The median number of admissions and days in hospital for cleft-related procedures did not differ between those with and those without additional anomalies or syndromes, although mean values were higher for the total number of days in hospital among syndromic children compared to non-syndromic children (8.5 vs. 6.3 days).
- Differences in admissions and total time in hospital for cleft-related procedures were observed between cleft types. Children with a cleft lip only (CL) or CP both had one admission and spent a total of four days in hospital for cleft-related procedures before the age of two years, whereas children with a UCLP and bilateral cleft lip and palate (BCLP) had two admissions and spent at least twice as long in hospital as those with either a CL or CP.

Emergency admissions for all causes

- All-cause emergency admissions represented 26% of all-cause admissions and 32% of all non-birth admissions within the first two years of life. Time spent in hospital for these admissions represented 23% of the total time spent in hospital before the age of two years.
- 42% of children with a cleft had at least one emergency admission in hospital for all-causes. There was a marked difference between non-syndromic and syndromic children. Of those without additional anomalies, 35% had at least one emergency admission; this

compares to 67% among syndromic children. Non-syndromic children spent a total of three days in hospital, while syndromic children spent nine unplanned days in hospital.

Cleft-treatment units (births 2006-2008)

- The average total number of days in hospital in the first two years of life varied significantly according to the hospital performing the primary repair. Non-syndromic children treated at Great Ormond Street Hospital (GOSH) and Guy's and St Thomas' spent the least amount of time in hospital (less than nine days), while those undergoing their repairs at Cambridge and Manchester spent the most time (14 days) in hospital for all causes.
- Differences between Administrative Units persisted when admissions in the first two years of life were restricted to cleft-related procedures, with total time in hospital ranging from three days for those treated at Guys and St Thomas' to seven days for those treated at Cambridge.
- Among children undergoing primary lip repairs, 2% were discharged the same day as their procedure. This was most commonly carried out at Oxford (10%) and GOSH (7%) and was occasionally carried out at five other centres. Day case admissions did not occur for other primary repairs.

Dental care before the age of seven

- For the first time, we have examined hospital admissions before the age of seven years for dental care, which we defined as an admission involving one of the following three procedures: surgical removal of teeth, simple extraction of teeth, or the restoration of teeth.
- 11% of cleft patients had at least one admission for dental care before the age of seven years. Syndromic children were more likely to have at least one dental care admission than non-syndromic children (18% vs. 10%), and they were also more like to have multiple admissions for dental care.
- Among non-syndromic children, the proportion having at least one admission for dental care increased with increasing severity of cleft type, with CL patients being the least likely and BCLP patients being the most likely to have at least one dental care admission in hospital.

- Dental care hospital admissions were also associated with deprivation; 8% of children in the least deprived quintile have at least one dental care admission compared to 18% of children in the most deprived quintile.

Recommendations

Clinical care

- 28% of the children born with a CP fail to be diagnosed at birth and 5% are not diagnosed until after one month of age. National and local guidelines for examining new born babies should be reviewed and recommendations should ensure proper visualisation of the palate to reduce the risk of a missed diagnosis and morbidity associated with late diagnosis.
- Just over half of all babies born with a cleft are referred to a cleft unit within 24 hours of birth. Administrative Units, together with maternity units, should ensure robust procedures are developed so that babies with a diagnosed cleft are referred promptly to the regional cleft team.
- The impact of additional anomalies and different cleft types should be considered when commissioning cleft services across England and Wales, as these factors directly affect both the total number of hospital admissions and the length of stay in hospital during the first two years of life.

Outcome measures and reporting to CRANE

- Gestational age and birth weight was reported to CRANE for approximately half of all registered cleft births in 2011. Further improvement in the reporting of these data is required.
- Height and weight should be measured at five years of age among all children with a cleft, and these data should be reported to CRANE.
- Children with a CP, UCLP and BCLP have a greater risk of dental decay than the general population, and they require greater support with decay prevention. All Administrative Units should have access to calibrated paediatric dentists who are able to examine children with clefts to determine their oral health status utilising decayed, missing and filled teeth (dmft) scoring. The collection of dmft scores at five years of age and the reporting of these to CRANE needs to improve even further in order to allow meaningful comparison between patient sub-groups and Units.
- The long-term collection of Five Year Old Index scores among children with a complete UCLP is required to allow for the comparison of dental arch relationships between Units.

- To enable the continuous monitoring of cleft care and outcomes on a national basis, adequate support to facilitate data entry on to the CRANE Database is required in all Administrative Units.

- There is an urgent need for the development of outcome measures in hearing, psychology and quality of life among children with a cleft.

1. Introduction

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 these disorders 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 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 includes trends in CRANE registrations over the last 10 years, comparing the 15 Administrative Units and the four different types of cleft. Using CRANE data, we also report the proportion of babies born in 2011 who were diagnosed at birth, referred within 24 hours of birth, and contacted within 24 hours of referral. For the second year running, cleft-related outcomes at five years of age are presented. These outcomes include height and weight, number of decayed, missing or filled teeth, and Five Year Old Index scores.

This year, we have developed our preliminary analyses of the burden of hospital care in early life using HES data. For the first time, we present differences in the total time spent in hospital between those with and without additional anomalies, and we have examined cleft-related procedure admissions according to the absence or presence of additional anomalies or associated syndromes, cleft type and Administrative Unit. We have also extended this work to examine admissions for dental disease up to the age of seven years.

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 [5]. 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, with which all patients should be registered [should] form the basis of national audit' [6]. 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 is provided by the Specialist Commissioners based on repeated two-year contracts. The CRANE team have responded to a number of requests for information from a commissioner led comprehensive review of all databases relating to specialised services; the outcome of this has been an agreement to continue to fund CRANE in 2012/13.

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 65 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
South Thames	Guy's Hospital, London
Northern Ireland	Royal Belfast Hospital for Sick Children, Belfast

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.

1.4. Current priorities of the CRANE Database

The agenda of the CRANE Database is constantly being updated. Currently, in addition to the ongoing CRANE remit, the main priorities are to:

- develop a new consent form and patient information sheet to reflect the recent changes brought about by the NIGB approval for CRANE to:
    - process a limited amount of patient-identifiable information without consent for the purpose of maintaining a register of cleft births, and
    - share CRANE data with other BINOCAR congenital anomaly registers
    - collect outcome data (with consent).
- introduce 5 and 10-year speech outcomes (CAPS-A) into CRANE;
- consult with cleft teams about the collection and reporting of 10-year cleft-related outcomes;
- re-run the linkage exercise between CRANE and HES with the latest HES dataset, which will include data up to 2012;
- run the linkage exercise between CRANE and PEDW
- examine the burden of care associated with dental disease among children with a cleft;
- establish collaborative working with the Cleft Collective, a new research programme set up to investigate cleft lip and palate;

2. Methods

This report contains information on patterns of care and outcomes derived from two sources of data: data from the CRANE Database and Hospital Episode Statistics (HES).

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.

Currently, CRANE only collects information on 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 consenting children referred to them. Units are also requested to notify CRANE of the number of non-consenting children referred to them each year (See Section 3.1). Once a record has been created on CRANE for a particular child, it can later be updated with further information.

Earlier in 2012, CRANE received approval from NIGB to collect a limited amount of patient-identifiable information without consent for the purpose of maintaining a register of cleft births. This means that soon, Administrative Units will be able to register all children with a cleft on CRANE.

2.1.2. Patients

All data entered into the CRANE Database by 3 September 2011 pertaining to children born between 1 January 2002 and 31 December 2011 have been included in the analyses described in this Annual report. Patients whose parents did not consent to their data being used by CRANE (1.2%) have been excluded from *Tables 3-11*.

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) [7].

2.1.4. Analyses

Data have been analysed according to year of birth, with Chapter 3 focusing on children born in 2011, unless otherwise stated. Five-year outcome data were restricted to children born in 2004 to 2006. 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 (4.2% of children registered in 2011), cleft type is based on the type reported by the Administrative Unit registering the child. Children with a 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 children born in 2004 to 2006 (excluding children with a submucous CP).

Currently, dmft data are not submitted by Nottingham and Cambridge. Cambridge does not have a paediatric dentist who would examine children to determine the dmft, and Nottingham has not had adequate administrative support to provide CRANE with dmft data.

Five Year Old Index

Dental models of five-year old children can be assessed using the Five Year Old Index to examine dental arch relationships. CRANE collected both internal and external Five Year Old Index scores for children born in 2004 and 2005 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, we have analysed external scores only.

Data on Five Year Old Index were not collected by Nottingham or Belfast. In Belfast, this is because children are not routinely seen by Orthodontists at five years of age. GOSH and Chelmsford had difficulty collating the data and coordinating submission to CRANE.

Missing data

Missing data have been excluded from the denominators presented in *Tables 5* to *11*. All Units have some degree of missing data. The number of patients with missing data for five-year outcomes is high. A variety of reasons were reported by units. Reasons out of a Unit's control include children not attending an appointment or moving away from the area. There was one Unit (Nottingham) who was unable to provide any five-year data. This centre collects the required outcomes but had difficulty collating the data and coordinating submission to CRANE. Nottingham is aware of this issue and is formulating an action plan to address this.

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, CRANE received an extract from HES on admissions for the 13 complete financial years between 1st April 1997 and 31st March 2010 and provisional data for the part-year between 1st April 2010 and 31st January 2011.

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).

We used HES data to identify hospital admissions and treatment for children with a cleft lip and/or palate. We focused on all-cause admissions, cleft-related procedure admissions, emergency admissions and admission for 'dental care'. HES also allowed us to estimate the treated incidence of clefting among births in England.

2.2.2. Patients

Patients were extracted for our analyses if they had at least one HES record of a hospital admission with a diagnosis code for cleft lip and/or palate (ICD-10 codes Q35, Q36 or Q37) and a procedure code for a primary cleft repair (OPCS-4 codes F031 or F291). ICD-10 codes were used to identify cleft patients with additional associated congenital anomalies or syndromes (see Appendix 5 for a list of these codes). Non-UK patients were excluded from analyses as they do not reflect a 'typical cleft patient' in the UK. Non-UK patients were identified in HES as a 'Private patient' with an 'unavailable/not applicable postcode'. All included patients were followed up until 31 January 2011, unless otherwise stated.

2.2.3. Analyses

Hospital admissions before the age of two years

Patients born between 1 January 1997 and 31 December 2008 were included in the analyses of hospital admissions. The total number of all-cause hospital admissions (including the birth episode) and the total number of days in hospital in the first two years of life were identified and included, regardless of the age at discharge. All included cleft patients were followed up for the first two years of life. An admission was defined as one complete spell within the same hospital, which may contain multiple episodes, each under a different consultant.

Admissions for cleft-related procedures were assessed to determine what proportion of all-cause admissions these represented and to explore differences between types of cleft and treatment centres. An admission was defined as a 'cleft-related procedure' if at least one of 24 specific procedure codes relating to the ear, respiratory tract or mouth (Appendix 5) was performed during a particular admission.

The proportion of all-cause admissions that were reported in HES as 'emergency' was determined by the reported method of admission, of which there are five options: *elective, emergency, birth, non-emergency transfer* and *unknown*.

Dental care hospital admissions before the age of seven years

Cleft patients born between 1st January 1997 and 31 December 2003 were included in the analyses of dental care admissions. All included patients were followed up for the first seven years of life, which is the period of primary dentition but is also prior to the commencement of dental care relating to alveolar bone grafting, which typically occurs between the ages of eight and 11 years of age. For the purpose of these analyses, a 'dental care' admission was defined as an inpatient episode with at least one of the following OPCS procedure codes:

F09	Surgical removal of tooth
F10	Simple extraction of tooth
F13	Restoration of tooth

Syndrome status

Patients were defined as 'syndromic' if any of their episode records had at least one of 33 ICD-10 diagnostic codes (listed in Appendix 5) representing congenital malformations and chromosomal abnormalities, in any one of the (fourteen) diagnosis code fields. Admissions and time spent in hospital was analysed separately for patients with additional congenital anomalies or syndromes, as the presence of these has a major impact on the frequency and duration of hospital admissions.

Cleft type

Clefts were grouped as cleft lip only (CL), cleft palate only (CP), unilateral cleft lip and palate (UCLP) or bilateral cleft lip and palate (BCLP) according to the presence of selected procedure codes (OPCS 4.5) and/or diagnosis codes (ICD-10) at any point in their HES record. A stepwise, hierarchical approach was employed. First, the cleft repair procedure codes (F03, F29, F30, F32) were used to identify three cleft type groups: CL, CP, Cleft lip and palate (CLP). Second, the diagnosis code information was used to distinguish between UCLP and BCLP cases in the CLP group.

Treatment centre

Admissions and time spent in hospital before the age of two years were also assessed according to the hospital performing the first primary cleft repair. Between 1998 and 2006, cleft services in England were centralised to 13 Administrative Units. To reflect care provided

by centralised services, only children born between 2006 and 2008 were included in analyses comparing the number of admissions and days spent in hospital between cleft centres.

Patients with additional congenital anomalies and syndromes and those whose total number of days in hospital exceeded the 95th percentile (>32 days) for non-syndromic patients were also excluded, as their admissions mostly reflected non-cleft-related care in the neonatal period (slow fetal growth and fetal malnutrition; disorders related to short gestation and low birth weight; birth asphyxia; respiratory distress of newborn). Analyses are presented according to cleft type and the Unit that performed the first primary repair. Analyses according to Unit include patients born between 1 January 2006 and 31 December 2008, as our analyses of HES data revealed that all cleft services had been centralised by the end of 2006.

Deprivation

Deprivation was explored by the application of an Index of Multiple Deprivation score based on postcode of residence and the corresponding Super Output area, as defined by the Office of National Statistics and is based on the entire population of England [8].

2.3. Statistical analyses

Data were analysed according to syndrome status, cleft type and Administrative Unit. The average number of admissions and total days in hospital are presented as medians, unless otherwise stated. Means and standard deviations are presented in the tables. Differences between treatment centres in the mean total time spent in hospital and the proportion of children with at least one emergency admission within the first two years of life were explored using funnel plots. The 95% and 99% confidence limits were calculated using the Binomial distribution [9]. Centres outside of these confidence limits are significantly different from the overall mean or proportion, depending on the outcome of interest. To avoid the means being heavily influenced by outliers (children staying in hospital for extreme lengths of time), the total time in hospital beyond the 95th percentile was set to a threshold of 32 days for all-cause admissions and 11 days for cleft-related admissions, equivalent to the 95th percentile for those born between 2006 and 2008 [10]. Each plot shows the mean total time spent in hospital within the first two years of life. All analyses were performed in Stata 10 (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 2002 and 31 December 2011 in England, Wales and Northern Ireland. The consent status for all children born in 2011 who have been referred to a Cleft Administrative Unit for treatment is presented below. Data entered into the CRANE Database by 3 September 2012 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.

3.1. Consent status

In June 2012, each Administrative Unit was asked to notify CRANE of the total number of children born with a cleft lip and/or palate in 2011 for whom:

- consent had been provided,
- consent had been refused,
- consent status had yet to be determined, and
- consent had not possible to determine, e.g. the child had died or had moved away.

Out of 1,092 children born in 2011 and being treated by the 14 Administrative Units reporting these figures to CRANE (Northern Ireland did not provide data) by 15 October 2012[*], the parents of 814 (74.5%) had been approached for consent. This figure ranged from 20.7% at Oxford to 100% at Newcastle (*Table 2*). Of the children whose parents had been through the consenting process, 98.8% provided consent for their child's data to be submitted to CRANE, which is extremely positive. This proportion ranged from 81.8% at Salisbury to 100% at eight Units (Newcastle, Nottingham, Cambridge, GOSH, Chelmsford, Oxford, Swansea and Bristol). Administrative Units reported a total of 278 children born in 2011 whose parents had not yet been approached for consent. Of these, it was not possible to obtain consent for 26 (9.4%) children (2.4% of all children born in 2011). The overall proportion of children whose parents still need to be approached for consent ranged from 0% at Newcastle to 79.3% at Oxford.

[*] The number of consented children reported by Administrative Units by 15 October 2012 may be fewer or greater than the number of children born in 2011 registered in CRANE by 3 September 2012 (the cut off for inclusion in analyses) due to differences in the time at which data were submitted to CRANE.

Table 2. Number of children born in 2011 with a cleft lip and/or palate in England, Wales and Northern Ireland, 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	63 (100.0)	0 (0.0)	0 (0.0)	0 (0.0)	63
	Leeds	69 (94.5)	1 (1.4)	1 (1.4)	2 (2.7)	73
North West & North Wales	Liverpool	58 (86.6)	1 (1.5)	8 (11.9)	0 (0.0)	67
	Manchester	72 (88.9)	1 (1.2)	5 (6.2)	3 (3.7)	81
Trent	Nottingham	89 (81.7)	0 (0.0)	16 (14.7)	4 (3.7)	109
West Midlands	Birmingham	94 (77.0)	1 (0.8)	22 (18.0)	5 (4.1)	122
East	Cambridge	46 (70.8)	0 (0.0)	15 (23.1)	4 (6.2)	65
North Thames	Gt Ormond St	66 (65.3)	0 (0.0)	34 (33.7)	1 (1.0)	101
	Chelmsford	49 (92.5)	0 (0.0)	4 (7.5)	0 (0.0)	53
The Spires	Oxford	12 (20.7)	0 (0.0)	46 (79.3)	0 (0.0)	58
	Salisbury	18 (36.7)	4 (8.2)	26 (53.1)	1 (2.0)	49
South Wales & South West	Swansea	48 (94.1)	0 (0.0)	3 (5.9)	0 (0.0)	51
	Bristol	49 (80.3)	0 (0.0)	11 (18.0)	1 (1.6)	61
South Thames	Guy's	71 (51.1)	2 (1.4)	61 (43.9)	5 (3.6)	139
Northern Ireland	Belfast	-	-	-	-	-
All	All	804 (73.6)	10 (0.9)	252 (23.1)	26 (2.4)	1092

The consent figures are based on what has been reported by Administrative Units, according to their local records. There are some minor discrepancies between these figures and the number of consented registrations added to CRANE in time for inclusion in this report. These discrepancies are most likely due to differences in the time at which data were submitted to CRANE.

Overall, the consenting data are encouraging. The consent rate is very high for those children whose parents have been approached. However, there is still a relatively high proportion of children whose parents have not yet been through the consent process, and the varied rate between Units suggests different processes are being used across centres. We are aware that some Units do not obtain consent until the time of the primary repair, which may account for some of this lag, although we would expect the majority of children born with a cleft in 2011 to have had their first surgical procedure by the time of finalising this Report.

Fortunately, the issue of consent affecting the figures we publish in our Annual Reports, primarily because of the lag between birth or diagnosis and registration, will be of minimal concern next year, as we are now able to collect a minimum dataset on all children born with a cleft, regardless of their consent status [11]. This will allow CRANE to function fully as a national register of all cleft births in England, Wales and Northern Ireland.

3.2. CRANE registrations

A total of 10,467 children born between 1 January 2000 and 31 December 2011 have been registered on the CRANE Database, of whom 837 have been added since last year's Annual Report. A further 528 children born over the past twelve years whose parents withheld consent have also been registered with limited information. Non-consented children have been excluded from all analyses presented within the report.

Table 3 shows the number of registrations for each Cleft Unit over the last 10 years (since 2002). Birmingham registered the most births over the last 10 years. The Northern and Yorkshire region, consisting of two Administrative Units is the region that has the most registrations overall.

For births in 2011, 832 consenting children were registered with CRANE. This figure is similar to that published last year for 2010 registrations, and it is expected to increase, as there can be a time lag between birth or diagnosis and registration. There are several reasons for this lag. First, until now, parental consent had to be obtained before children were registered on

Table 3. Number of CRANE-registered babies born with a cleft lip and/or palate in England, Wales and Northern Ireland, according to Administrative Unit and year of birth, 2002-2011

		Year of birth										
Regional centre / MCN	**Administrative Unit**	2002	2003	2004	2005	2006	2007	2008	2009	2010	2011	All
Northern & Yorkshire	Newcastle	46	68	60	78	56	85	64	63	58	63	641
	Leeds	74	78	72	73	77	70	76	65	69	70	724
North West & North Wales	Liverpool	58	50	57	66	47	55	81	74	77	56	621
	Manchester	62	28	22	47	93	85	74	62	83	72	628
Trent	Nottingham	86	73	93	106	94	83	82	83	90	86	876
West Midlands	Birmingham	102	113	111	100	105	70	78	88	85	99	951
East	Cambridge	48	43	45	57	57	70	70	72	57	42	561
North Thames	Gt Ormond St	15	17	37	67	69	62	106	100	76	68	617
	Chelmsford	21	21	30	35	22	31	28	37	40	50	315
The Spires	Oxford	31	42	37	37	43	31	26	44	31	13	335
	Salisbury	39	36	42	39	60	59	45	29	40	18	407
South Wales & South West	Swansea	30	40	37	42	47	45	43	47	44	48	423
	Bristol	64	60	52	49	57	59	71	50	71	50	583
South Thames	Guy's	56	44	72	83	99	102	105	83	60	71	775
Northern Ireland	Belfast	24	26	29	36	38	37	28	33	37	26	314
All	All	756	739	796	915	964	944	977	930	918	832	8,771

Table 4. Number (%) of CRANE-registered babies born with a cleft lip and/or palate in England, Wales and Northern Ireland, according to cleft type and year of birth, 2002-2011

	Year of birth n (%)										
Cleft type	2002	2003	2004	2005	2006	2007	2008	2009	2010	2011	All
CL	175 (24.3)	145 (21.5)	171 (22.5)	163 (18.7)	211 (23.2)	222 (24.1)	234 (25.2)	192 (21.8)	225 (25.2)	204 (25.3)	1,942 (23.2)
CP	325 (45.1)	316 (46.8)	335 (44.0)	419 (48.1)	408 (44.8)	388 (42.2)	406 (43.7)	400 ((45.4)	380 (42.6)	345 (42.8)	3,722 (44.5)
UCLP	156 (21.6)	162 (24.0)	175 (23.0)	218 (25.0)	202 (22.2)	212 (23.0)	216 (23.2)	192 (21.8)	189 (21.2)	182 (22.6)	1,904 (22.8)
BCLP	65 (9.0)	52 (7.7)	80 (10.5)	71 (8.2)	90 (9.9)	98 (10.7)	74 (8.0)	97 (11.0)	99 (11.1)	75 (9.3)	801 (9.6)
Not specified	35	64 -	35 -	44 -	53 -	24 -	47 -	49 -	25 -	26 -	402 -
All	756 (100.0)	739 (100.0)	796 (100.0)	915 (100.0)	964 (100.0)	944 (100.0)	977 (100.0)	930 (100.0)	918 (100.0)	832 (100.0)	8,771 (100.0)

CL, cleft lip; CP, cleft palate; UCLP, unilateral cleft lip and palate; BCLP, bilateral cleft lip and palate

CRANE. Second, consent is sometimes not sought until the time of primary repair, which may take place up to one year after birth. Third, some clefts are diagnosed late. It is estimated that approximately 250 children born in 2011 will be added to CRANE in due course.

Registrations by Cambridge, GOSH, Oxford and Salisbury are substantially lower for 2011 births than preceding years, indicating that these centres may have the longest lag time between birth or diagnosis and CRANE registration.

The distribution of the four main cleft types is shown in *Table 4*. Cleft type was defined according to reported *LAHSAL* codes. Where *LAHSAL* has not been reported (4.1% of children registered in 2011), cleft type is based on the type reported by the Administrative Unit registering the child. Overall, 3.1% of the registered children born in 2011 did not have their type of cleft specified. These children were restricted to eight Cleft Units. Chelmsford had the highest proportion (12.0%) of patients whose cleft type was not specified. The other seven units had rates below 6%. Last year, Belfast and GOSH had the highest proportion of patients whose cleft type was not specified (19.1% and 13.6%, respectively). Their rates have improved substantially for 2011 (3.9% and 5.9%, respectively).

The distribution of cleft type is consistent over time. CP is the most common type of cleft, affecting just over 40% of the cleft population. This proportion is likely to increase to around 45% once late CP diagnoses are reported to CRANE. BCLP is the least common type, affecting around 10% of people with clefts. A total of 139 children registered in 2011 had complete UCLP (defined by either '*LAHS*..' or '..*HSAL*' *LAHSAL* codes), representing 76% of the 182 children with UCLP. This is similar to the proportion in recent previous years.

3.3. Characteristics of children born with a cleft lip and/or palate, 2011

Of the children born with a cleft in 2011, 44.0% were girls and 55.4% were boys. Sixteen children did not have their sex reported to CRANE. There are significant gender differences in the distribution of cleft type ($P<0.001$), as shown in the Annual Report published in 2010 [12]. Isolated CP is more prevalent among females (56.5% vs. 43.5% in males), while CL and UCLP is more prevalent among males (57.6% vs. 42.4% and 70.6% vs. 29.4%, respectively).

Gestational age was reported for 401 (48.2%) babies born in 2011. This represents a great improvement from last year, when only 10.2% of registered babies had their gestational age reported. The mean gestation for those born in 2011 was 38.8 weeks (95% CI 38.6 to 39.0 weeks) and ranged from 26 to 42 weeks. Forty babies (10.0%) were premature (born before

37 weeks' gestation), which is higher than the six per cent background rate in England [13], although it should be noted that the gestation recoded in CRANE may not be representative of all babies born with a cleft lip and/or palate as 51.8% of registered children were missing this information.
A valid birth weight was reported for 468 (56%) babies born in 2011. The median birth weight was 3.19kg (95% CI 3.14 to 3.25kg), which is consistent with the national average [13].

Among the children born in 2011, there were three (0.4%) deaths reported to CRANE. Of these, one child died within the first month of life and two died 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, subsequent referral to and first contact with a cleft team, 2011

Of the 832 consenting children born in 2011 with a cleft diagnosis, 34 (4.1%) did not have the timing of their diagnosis reported to CRANE. This is consistent with last year's figures, which have improved since 2009, when 13.4% of children born did not have the time of their diagnosis reported.

In total, 337 children born in 2011 had their cleft diagnosed during the antenatal period, representing 42.2% of all registered children – the highest proportion since we started collecting this information. The proportion of children diagnosed antenatally varied between cleft types, as shown in *Table 5*. Sixty percent of children with CL and approximately 80% of children with UCLP and BCLP were diagnosed in the antenatal period. Conversely, only 1.2% of children with a CP were diagnosed antenatally, which demonstrates the difficulty of identifying this type of cleft with current antenatal screening techniques.

Of the 407 children not diagnosed during the antenatal period, 76.8% were diagnosed at birth. This is 3% higher than the rate in 2010. Of those children who did not have their cleft identified antenatally, the majority (≥88%) with a CL, UCLP and BCLP were diagnosed at the time of birth; however, 28.7% of children with a CP were not identified until later, with 3.9% of all children with a CP being diagnosed between one and six months after birth. It should be noted that some children born in 2011 with a CP may not yet have had their cleft identified. Between 2006 and 2010, 40 children with a CP were diagnosed after six months of age.

Table 5. Number (%) of CRANE-registered children born in 2011 with a cleft lip and/or palate according to the timing of diagnosis and cleft type

	Time of diagnosis in relation to birth n (%)						
Cleft type	Antenatal	At birth	≤1 week	≤1 month	≤6 months	>6 months	**All***
CL	118 (60.2)	69 (35.2)	6 (3.1)	0 (0.0)	3 (1.5)	0 (0.0)	196
CP	4 (1.2)	234 (70.5)	61 (18.4)	18 (5.4)	13 (3.9)	2 (0.6)	332
UCLP	146 (82.5)	30 (16.9)	0 (0.0)	0 (0.0)	1 (0.6)	0 (0.0)	177
BCLP	60 (80.0)	14 (18.7)	0 (0.0)	0 (0.0)	1 (1.3)	0 (0.0)	75
Not specified	9 (50.0)	7 (38.9)	0 (0.0)	1 (5.6)	0 (0.0)	1 (5.6)	18
All	337 (42.2)	354 (44.4)	67 (8.4)	19 (2.4)	18 (2.3)	3 (0.4)	798

CRANE, 2011

* 34/832 (4.1%) missing diagnosis time and excluded from table; CL, cleft lip; CP, cleft palate; UCLP, unilateral cleft lip and palate; BCLP, bilateral cleft lip and palate

Fifty-four per cent of all CRANE-registered children born in 2011 were referred to an Administrative Unit within 24 hours of birth. Seventy-two per cent of children whose clefts were diagnosed antenatally were referred to an Administrative Unit within 24 hours of birth. This compares to 41.7% of the 441 children without an antenatal diagnosis. This proportion is slightly lower than those born in 2010 (45.3%) and 2009 (46.1%), however these differences are not statistically significant.

Table 6. Number (%) of CRANE-registered children born in 2011 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*	Within 24h of referral to Unit	All[§]
Cleft type	n (%)	N	n (%)	N
CL	112 (62.6)	179	149 (89.2)	167
CP	115 (35.9)	320	266 (88.7)	300
UCLP	116 (73.4)	158	144 (93.5)	154
BCLP	51 (75.0)	68	59 (89.4)	66
Not specified	6 (37.5)	16	11 (78.6)	14
All	400 (54.0)	741	629 (89.7)	701

CRANE 2011

*91/832 (10.9%) missing referral time, [§]131/832 (15.8%) missing contact time. Missing excluded in 'All' values.

Table 6 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 (35.9%) out of those with a known cleft type, which corresponds with later diagnosis times for these children. A high proportion of patients were contacted within 24 hours of being referred to an Administrative Unit. This did not vary significantly between cleft types.

Referrals within one day of birth varied significantly according to the Administrative Unit receiving the referral ($p<0.001$). Seventy-seven per cent of children registered by Nottingham were referred from maternity units within 24 hours of birth, which is in contrast to only 30.8% of those referred to Oxford from maternity units. Progress in this area has been made in several regions. For example, Bristol had the lowest 24 hour referral rate at 22% for 2010 births; this increased to 52% for 2011 births.

Table 7. Number (%) of CRANE-registered children born in 2011 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* N	Within 24h of referral to Unit n (%)	All[§] N
Northern & Yorkshire	Newcastle	34 (54.8)	62	60 (96.8)	62
	Leeds	44 (63.8)	69	67 (97.1)	69
North West & North Wales	Liverpool	29 (63.0)	46	44 (95.7)	46
	Manchester	35 (50.0)	70	67 (98.5)	68
Trent	Nottingham	34 (77.3)	44	37 (100.0)	37
West Midlands	Birmingham	45 (47.4)	95	88 (93.6)	94
East	Cambridge	18 (72.0)	25	24 (57.1)	42
North Thames	Gt Ormond St	22 (35.5)	62	28 (56.0)	50
	Chelmsford	16 (33.3)	48	28 (80.0)	35
The Spires	Oxford	4 (30.8)	13	1 (100.0)	1
	Salisbury	10 (55.6)	18	12 (66.7)	18
South Wales & South West	Swansea	35 (72.9)	48	45 (97.8)	46
	Bristol	23 (47.9)	48	41 (97.6)	42
South Thames	Guy's	38 (54.3)	70	67 (95.7)	70
Northern Ireland	Belfast	13 (56.5)	23	20 (95.2)	21
All	All	400 (54.0)	741	629 (89.7)	701

CRANE, 2011

* 91/832 (10.9%) children missing referral time, [§] 131/832 (15.8%) children missing contact time. Missing excluded in 'All' values; MCN, managed clinical network.

Overall, Administrative Units established contact with 89.7% of all cleft patients within 24 hours of referral (*Table 7*). This has increased by almost 10% since 2007 (P=0.02). Rates varied between Units (56.0% to 100.0%), but the majority contacted greater than 90% of their patients within 24 hours of being referred.

As highlighted further above, very few children with a CP are diagnosed in the antenatal period, and a significant proportion remains undiagnosed at birth. For this reason, we examined in greater detail the diagnosis and referral of children born with a CP in 2010 and 2011, who were not diagnosed antenatally (*Table 8*). The proportion of children who had their CP diagnosed at birth varied from 41.7% to 94.4% according to the cleft Administrative

Table 8. Number (%) of CRANE-registered children with a cleft palate born in 2010-2011, without a prenatal diagnosis, who were diagnosed and referred within 24 hours of birth, according to Administrative Unit

		Diagnosis		**Referral**	
	Administrative	At birth	All*	Within 24h of birth	All[§]
Regional centre / MCN	**Unit**	n (%)	N	n (%)	N
Northern & Yorkshire	Newcastle	33 (67.3)	49	17 (34.7)	49
	Leeds	34 (73.9)	46	16 (35.6)	45
North West & North Wales	Liverpool	46 (79.3)	58	25 (43.9)	57
	Manchester	38 (55.9)	68	16 (23.5)	68
Trent	Nottingham	47 (81.0)	58	33 (71.7)	46
West Midlands	Birmingham	64 (90.1)	71	16 (22.5)	71
East	Cambridge	20 (60.6)	33	18 (52.9)	34
North Thames	Gt Ormond St	25 (41.7)	60	16 (26.2)	61
	Chelmsford	15 (42.9)	35	10 (28.6)	35
The Spires	Oxford	17 (94.4)	18	5 (26.3)	19
	Salisbury	19 (76.0)	25	12 (48.0)	25
South Wales & South West	Swansea	32 (80.0)	40	25 (62.5)	40
	Bristol	34 (69.4)	49	12 (23.1)	52
South Thames	Guy's	29 (56.9)	51	16 (31.4)	51
Northern Ireland	Belfast	17 (81.0)	21	6 (42.9)	14
All	All	470 (68.9)	682	243 (36.4)	667

CRANE, 2010-2011

8/725 (1.1%) children with a CP antenatal diagnosis excluded; * 35/725 (4.8%) children missing diagnosis time, [§] 50/725 (6.9%) children missing referral time. Missing excluded in 'All' values; MCN, managed clinical network.

Unit registering the child (P<0.001). The wide variation in the proportion of cleft palates diagnosed at birth suggests that some maternity units are better than others at identifying clefts during newborn examinations.

Referrals to a cleft team within 24 hours of birth ranged from 22.5% to 71.7% (P<0.001). Referral within 24 hours of birth was not necessarily dependent on a quick diagnosis. For example, although Oxford had the highest rate of CP diagnoses at birth, it had one of the lowest 24-hour referral rates, suggesting a delay between diagnosis and referral to the Administrative Unit.

3.5. Five-year outcomes among children born with a cleft lip and/or palate, 2004 to 2006

Five-year outcomes include height and weight, decayed, missing and filled teeth (dmft), and the Five Year Old Index. These outcomes were reported for the first time in last year's report [3].

3.5.1. Height and weight

Five-year height and weight were reported for 23% of the 2,623 children born in 2004-2006 who were alive at five years of age. The mean (SD) height was 111.3cm (5.9) while the mean weight was 19.75kg (3.01). There is a very high proportion of missing data for five-year height and weight. Nottingham, GOSH, Oxford, Salisbury and Belfast Cleft Units reported not collecting height and weight at five years of age.

3.5.2. Decayed missing and filled teeth (dmft)

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 [14, 15]. We collect dmft data on CRANE-registered children at five years of age. Out of 2,519 children born between 2004 and 2006 (excluding 52 children dying before five years of age and 104 with submucous CPs), dmft scores were provided for 1,379 (54.7%). Although almost half of the children are missing dmft scores, this situation has improved substantially since last year, when dmft scores were provided for only 27% of eligible children born in 2004 and 2005.

Among children with a reported dmft outcome, 41.9% of children with a cleft had at least one decayed, missing or filled tooth. The mean number of dmft at five years among children registered in CRANE was 2.0 (3.4), with scores ranging from 0 to 20. One hundred and eighty-two children (13.4%) had a dmft score greater than 5. The dmft data, obtained in 2005, are available for five-year old children in the general population in England and Wales. These data showed that 38.8% of five-year olds had ≥1 dmft, with a mean number of 1.5 [16]. The comparable figures for England and Wales among CRANE-registered children was 41.9% and a mean number of 2.0 dmft, which is significantly higher than the background rate.

Table 9. Number (%) of CRANE-registered children born in 2004-2006 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	n (%)	n (%)	95% CI	All*
CL	1.3	176 (65.4)	93 (34.6)	28.9 – 40.3	269
CP	2.3	318 (57.8)	232 (42.2)	38.0 – 46.3	550
UCLP	1.8	218 (54.9)	179 (45.1)	40.2 – 50.0	397
BCLP	2.4	63 (49.6)	64 (50.4)	41.6 – 59.2	127
Not specified	1.7	26 (72.2)	10 (27.8)	12.4 – 43.2	36
All	2.0	801 (58.1)	578 (41.9)	39.3 – 44.5	1379

CRANE, 2004-2006

106 children with submucous clefts excluded; 52 children who died before the age of five (of whom two had a submucous cleft) excluded; *1,140/2,519 (45.3%) children with missing dmft data excluded; UCLP, unilateral cleft lip and plate; BCLP, bilateral cleft lip and palate.

Table 9 shows the prevalence of dental caries according to cleft type. The proportion of children with ≥1 dmft varied according to cleft type (P=0.006). As we noted in last year's report, the proportion of children with a CL who had ≥1 dmft was smaller than the proportion in the general population. This could be due to raised parental awareness of dental caries and increased access to dental services through cleft teams, thereby increasing the level of caries prevention in a group not particularly at increased risk as compared to more severe cleft types. Children with a CP, UCLP and BCLP all had higher mean dmft scores than the general population. Children with a BCLP had the highest mean dmft score, at 2.4 (95% CI 1.8 to 3.1).

The fact that dmft were submitted for only 54.7% of children means that these data should be interpreted with caution. Two Administrative Units 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.

Table 10. Number (%) of CRANE-registered children born in 2004-2006 with a cleft lip and/or palate according to the number of decayed, missing or filled teeth (dmft) at age five years and Administrative Unit

		Number of decayed, missing or filled teeth (dmft)			
		0	>0		
Regional centre / MCN	**Administrative Unit**	n (%)	n (%)	95% CI	All*
Northern & Yorkshire	Newcastle	67 (48.6)	71 (51.4)	43.0 – 59.9	138
	Leeds	73 (53.7)	63 (46.3)	37.8 – 54.8	136
North West & North Wales	Liverpool	70 (57.9)	51 (42.1)	33.2 – 51.1	121
	Manchester	58 (59.8)	39 (40.2)	23.1 – 50.4	97
Trent	Nottingham	_	_	_	_
West Midlands	Birmingham	165 (59.6)	112 (40.4)	34.6 – 46.3	277
East	Cambridge	_	_	_	_
North Thames	Gt Ormond St	29 (61.7)	18 (38.3)	23.9 – 52.7	47
	Chelmsford	26 (54.2)	22 (45.8)	31.2 – 60.5	48
The Spires	Oxford[§]	47 (58.8)	33 (41.3)	30.2 – 52.3	80
	Salisbury[§]	67 (72.8)	25 (27.2)	17.9 – 36.4	92
South Wales & South West	Swansea	32 (49.2)	33 (50.8)	38.3 – 63.3	65
	Bristol	56 (65.9)	29 (34.1)	23.8 – 44.4	85
South Thames	Guy's	91 (61.9)	56 (38.1)	30.2 – 46.0	147
Northern Ireland	Belfast	20 (43.5)	26 (56.5)	41.6 – 71.4	46
All	All	801 (58.1)	578 (41.9)	39.3 – 44.5	1,379

CRANE, 2004-2006

106 children with submucous clefts excluded; 52 children who died before the age of five (of whom two had a submucous cleft) excluded; *1,140/2,519 (45.3%) children with missing dmft data excluded; [§]Non-BASCD calibration; UCLP, unilateral cleft lip and plate; BCLP, bilateral cleft lip and palate.

Table 10 shows the number and proportion of five-year old children with ≥1 dmft according to Administrative Unit. The proportion of cleft children with ≥1 dmft was the lowest in Salisbury at 27.2%, which was significantly lower than the overall proportion among cleft

children. The corresponding figure for Newcastle (51.4%) was significantly higher than the overall rate. Whilst the proportion of cleft children with ≥1 dmft varies substantially between regions, for the majority of regions their rate does not seem to differ substantially from their region's background rate [17]. The only substantial difference appears to be for Birmingham, whose cleft rate is a third higher than their background rate of 30.7%.

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 (see Section 4.2.1) 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 [18]. Fluoridation levels vary between regions throughout the UK. For example, parts of the West Midlands and 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 ≥1 dmft in the general population; however the North East had the highest proportion (50%) [17].

3.5.3. Five Year Old Index (2004-2005)

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 [19]. Dental arch relationships at five years are thought to predict treatment outcome in terms of facial growth on a population basis rather at the individual child level [20]. The Five Year Old Index may, therefore, also be used to compare treatment outcomes between centres and surgeons. Patients categorised as '1' and '2' on the index are considered to have the best possible outcome, while those categorised as '4' and '5' are thought to have very poor outcomes in terms of facial growth, and they may benefit from further surgery to correct their facial disproportion once facial growth is complete.

CRANE collected Five Year Old Index scores for children born in 2004 and 2005 with a complete UCLP. Five children who died before their fifth birthday were excluded. Externally validated scores were provided for 147 (53.8%) eligible children by 11 of the 15 Administrative Units (*Table 11*).

Only Leeds, Birmingham, Oxford, Salisbury and Guy's 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, thus, scores

for other Units should be interpreted with caution. The small number of patients within each Unit (1-29) means that statistical comparison between Units is not currently appropriate. CRANE will continue to collect these outcomes over the next few years, and as numbers increase, meaningful comparison between Units will become possible.

Overall, 41.5% of complete UCLP patients born in 2004 and 2005 had Five Year Old Index scores in the two groups considered to have the best possible dental arch relationships (scores '1' or '2') while 25.2% of children had scores '4' or '5', reflecting poor dental arch relationships. This compares to 36% (of 239 children) with poor dental arch relationships at five years old in 1996 [5].

Table 11. Number (%) of CRANE-registered children born in 2004-2005 with a complete unilateral cleft lip and palate, according to Five Year Old Index scores and Administrative Unit

		Five Year Old Index n (%)					
Regional centre / MCN	**Administrative Unit**	1	2	3	4	5	All*
Northern & Yorkshire	Newcastle	1 (14.3)	0 (0.0)	5 (71.4)	0 (0.0)	1 (14.3)	7
	Leeds	0 (0.0)	8 (36.4)	8 (36.4)	5 (22.7)	1 (4.5)	22
North West & North Wales	Liverpool	1 (10.0)	3 (30.0)	4 (40.0)	2 (20.0)	0 (0.0)	10
	Manchester	0 (0.0)	1 (100.0)	0 (0.0)	0 (0.0)	0 (0.0)	1
Trent	Nottingham	_	_	_	_	_	_
West Midlands	Birmingham	1 (3.7)	13 (48.1)	7 (25.9)	3 (11.1)	3 (11.1)	27
East	Cambridge	0 (0.0)	4 (66.7)	2 (33.3)	0 (0.0)	0 (0.0)	6
North Thames	Gt Ormond St	_	_	_	_	_	_
	Chelmsford	_	_	_	_	_	_
The Spires	Oxford	0 (0.0)	7 (50.0)	4 (28.6)	2 (14.3)	1 (7.1)	14
	Salisbury	1 (6.3)	7 (43.8)	2 (12.5)	6 (37.5)	0 (0.0)	16
South Wales & South West	Swansea	0 (0.0)	1 (14.3)	3 (42.9)	2 (28.6)	1 (14.3)	7
	Bristol	0 (0.0)	1 (12.5)	3 (37.5)	3 (37.5)	1 (12.5)	8
South Thames	Guy's	1 (3.4)	11 (37.9)	11 (37.9)	3 (10.3)	3 (10.3)	29
Northern Ireland	Belfast	_	_	_	_	_	_
All	All	5 (3.4)	56 (38.1)	49 (33.3)	26 (17.7)	11 (7.5)	147

CRANE, 2004-2005

* 126/273 (46.2%) children with missing Five Year Old Index scores excluded; MCN, managed clinical network.

4. Hospital Episode Statistics

In this section, we present data on children who have at least one HES record of an English NHS hospital admission with a diagnosis code for cleft lip and/or palate as well as a procedure code for a primary cleft repair (that is, a first repair of the lip and/or palatal cleft).

4.1. Admissions and time in hospital in the first two years of life among children treated with a cleft lip and/or palate in England

This section presents data reflecting the number of all-cause hospital admissions and days spent in hospital up to two years of age. We have focused on admissions within the first two years, as the majority (~75%) of admissions in the first six years occur by the age of two years [3].

4.1.1. All-cause admissions and time in hospital

Non-syndromic children born between 1997 and 2008 with a cleft had an average of three all-cause hospital admissions and spent a total of 10 days in hospital by the age of two years (*Table 12*). Excluding the birth episode, children had an average of two admissions and seven days in hospital. The average number of admissions and total time spent in hospital before the age of two years has not changed significantly for births between 1997 and 2008.

Children with a cleft and additional anomalies or associated syndromes represented 22% of all children with a cleft who had undergone a primary surgical repair. These children had five admissions and spent a total of 27 days in hospital before their second birthday, almost three times as long as their non-syndromic counterparts (*Table 12*). Excluding the birth episode, syndromic children had an average of four admissions and spent a total of 15 days in hospital. At birth, syndromic children spent twice as long in hospital as those without additional anomalies or syndromes (eight vs. four days).

4.1.2. Cleft type

Admissions and days in hospital varied according to the type of cleft and appeared to be correlated with the severity of the abnormality. Out of the four main cleft types among non-syndromic children, CL was associated with the fewest admissions and total days in hospital, while children with a BCLP had the highest number of admissions and days in hospital (*Table*

12). Despite a greater number of overall admissions and days in hospital, a similar pattern was observed for syndromic children.

Table 12. Total number of all-cause admissions and days in hospital for a) non-syndromic and b) syndromic cleft patients in the first two years of life, according to cleft type; year of birth 1997-2008

a) Non-syndromic patients

		Total number of admissions			Total number of days in hospital		
Cleft type	N*	Median	Mean	(SD)	Median	Mean	(SD)
CL	2,342	2	2.6	(1.7)	7	9.2	(14.5)
CP	3,378	2	2.8	(2.2)	9	12.6	(20.3)
UCLP	1,881	4	4.0	(1.9)	13	16.1	(16.8)
BCLP	881	4	4.7	(2.1)	16	19.7	(15.5)
All	8,482	3	3.2	(2.1)	10	13.2	(17.9)

b) Syndromic patients

		Total number of admissions			Total number of days in hospital		
Cleft type	N*	Median	Mean	(SD)	Median	Mean	(SD)
CL	187	4	6.6	(6.7)	18	55.3	(121.8)
CP	1,827	5	6.4	(6.2)	28	48.5	(64.6)
UCLP	215	5	7.5	(5.4)	26	58.5	(88.2)
BCLP	181	7	8.8	(6.3)	35	67.5	(94.3)
All	2,410	5	6.7	(6.2)	27	51.4	(75.5)

Hospital Episode Statistics, England 1997-2011

* Private, non-UK patients excluded; SD, standard deviation; CL, cleft lip; CP, cleft palate; UCLP, unilateral cleft lip and plate; BCLP, bilateral cleft lip and palate.

4.1.3. Cleft-related procedure admissions

Overall, cleft-related procedure admissions accounted for 37.2% of all hospital admissions and 29.6% of the total number of days spent in hospital among children with a cleft in the first two years of life. The median number of admissions and days in hospital for cleft-related procedures did not differ between those with and those without additional anomalies or syndromes, although mean values were higher for the total number of days in hospital among syndromic children compared to non-syndromic children (8.5 vs. 6.3 days).

Differences in admissions and total time in hospital were observed between cleft types (*Table 13*). Children with a CL or CP both had one admission and spent a total of four days in hospital for cleft-related procedures before the age of two years, whereas children with UCLP and BCLP had two admissions and spent at least twice as long in hospital as those with either a CL or CP.

Table 13. Total number of cleft-related procedure admissions and days in hospital for a) non-syndromic and b) syndromic cleft patients in the first two years of life, according to cleft type; year of birth 1997-2008

a) Non-syndromic patients

		Total number of admissions			Total number of days in hospital		
Cleft type	N*	Median	Mean	(SD)	Median	Mean	(SD)
CL	2,326	1	1.2	(0.4)	4	4.0	(2.0)
CP	3,004	1	1.2	(0.5)	4	5.0	(3.8)
UCLP	1,877	2	2.3	(0.7)	8	9.0	(4.4)
BCLP	880	2	2.6	(0.9)	10	11.2	(8.5)
All	8,087[§]	1	1.6	(0.8)	5	6.3	(5.0)

b) Syndromic patients

		Total number of admissions			Total number of days in hospital		
Cleft type	N*	Median	Mean	(SD)	Median	Mean	(SD)
CL	180	1	1.4	(0.7)	4	11.0	(40.0)
CP	1,623	1	1.2	(0.5)	5	6.4	(12.7)
UCLP	211	2	2.2	(0.8)	9	13.7	(23.7)
BCLP	180	2	2.6	(1.0)	11	19.1	(43.3)
All	2,194[§]	1	1.5	(0.8)	5	8.5	(21.7)

Hospital Episode Statistics, England 1997-2011

* Private, non-UK patients excluded; [§] Not all patients included in the analyses had a cleft related procedure admission within the first two years of life. SD, standard deviation; CL, cleft lip; CP, cleft palate; UCLP, unilateral cleft lip and plate; BCLP, bilateral cleft lip and palate.

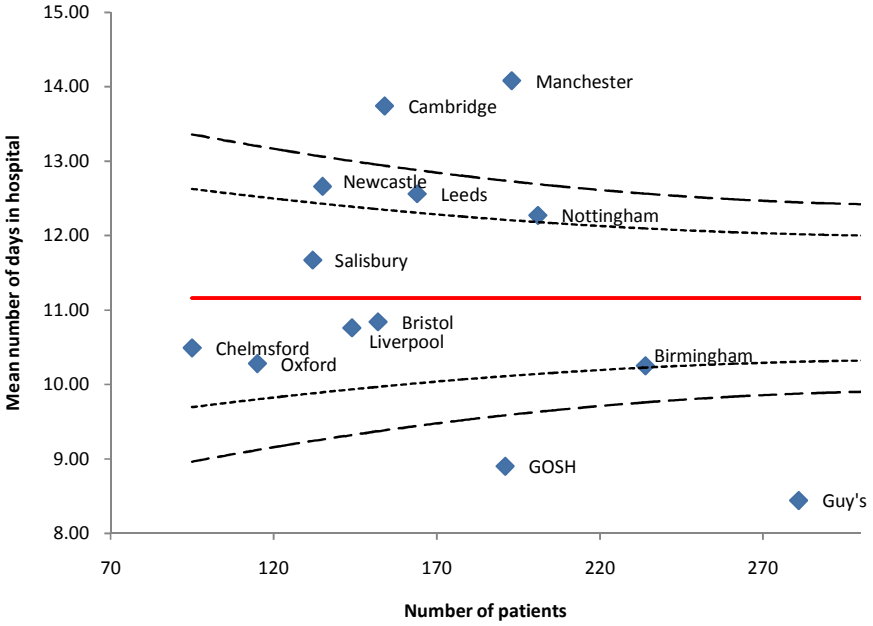
4.1.4. Cleft-treatment centres

Since 2006, there have been 13 hospitals (Administrative Units) performing cleft surgery in England. To reflect care provided by centralised services, only children born between 2006 and 2008 have been included in these analyses. The average number of all-cause admissions in the first two years of life was three for all Administrative Units, with the exception of Birmingham, which had an average of four admissions. A greater range was observed for the total number of days spent in hospital, where mean values ranged from 8.4 to 14.1 (Figure 1.1). Children receiving their primary repair at Guys and St Thomas' and GOSH Administrative Units spent, on average, the shortest total time in hospital, while those undergoing their repairs at Cambridge and Manchester spent almost twice as much time in hospital for all causes.

Differences between Administrative Units persisted when admissions were restricted to cleft-related procedures (Figure 1.2). Although the total time in hospital was reduced, patients at Manchester and Cambridge still spent twice as long in hospital as those at Guys and St Thomas' and GOSH.

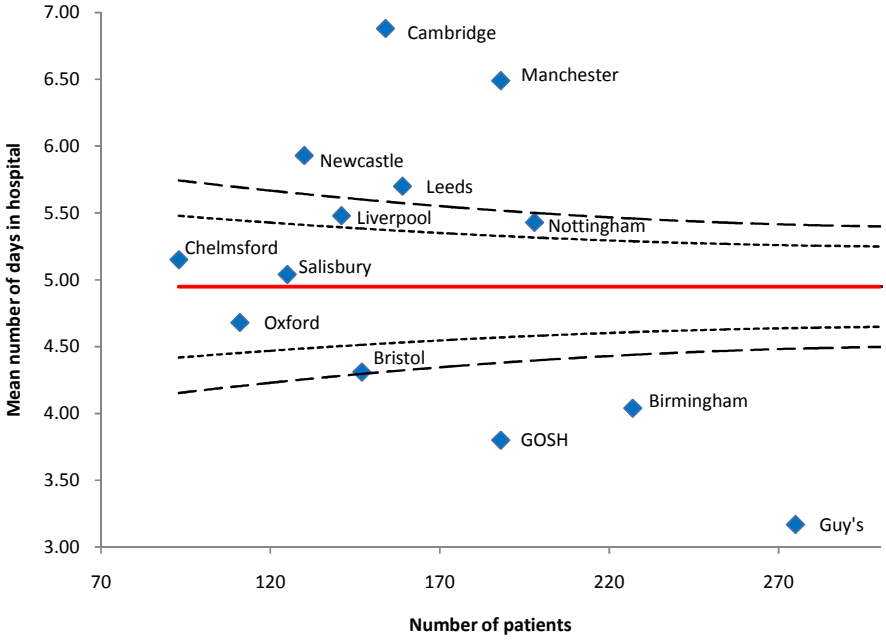
Figure 1. Number of days in hospital for all-causes in the first two years of life, according to Administrative Unit, births 2006-2008

1.1 *All-causes*



Solid line, mean number of days; triangle, Administrative Unit; wide dotted line, 99.8% confidence limit; narrow dotted line, 95% confidence limit. Extreme values limited to the 95th percentile (32 days)

1.2 Cleft-related procedures



Solid line, mean number of days; triangle, Administrative Unit; wide dotted line, 99.8% confidence limit; narrow dotted line, 95% confidence limit. Extreme values limited to the 95th percentile (11 days)

The funnel plots in Figure 1 show that the mean total time spent in hospital by patients receiving their primary repairs at these four centres was significantly different to the overall mean.

Emergency admissions also differed between Administrative Units for those born between 2006 and 2008. Overall, 37.1% of non-syndromic children had at least one unplanned admission before the age of two years; this proportion varied between treatment centres, however the cause of emergency admissions appears to be non-cleft-related in the majority of cases.

Although there are some differences in cleft type distribution between Administrative Units, these do not appear to explain the observed differences between Units in terms of the number of days in hospital. For example, although children undergoing primary repairs at GOSH spent the shortest time in hospital, GOSH had one of the highest proportions of children with BCLP, which is associated with the most time in hospital.

It is important to recognise that differences between hospitals do not necessarily reflect differences in the quality of care, but may instead reflect differences in treatment protocols, particularly with regards to repairing the cleft lip and palate during the same operation or on separate occasions. For instance, according to HES coding, Cambridge never performs combined lip and palate repairs, whereas 32% of all UCLP and BCLP primary repairs in Liverpool are combined lip and palate repairs. Geographical differences may also account for some variation. Children who live further away from Administrative Units may be discharged later than those who live closer as they would have to travel further to return to the Unit in the event of problems or complications.

4.1.5. Deprivation

Deprivation was not associated with admissions or total time spent in hospital before the age of two years.

4.2. Dental extractions with the first seven years of life among children treated with a cleft lip and/or palate in England

This section presents data reflecting admissions to hospital for dental care involving the surgical removal of teeth, simple extraction of teeth, or the restoration of teeth before the age of seven years. We have focused on the first seven years of life as this is the period of

primary dentition and is also prior to the commencement of dental care relating to alveolar bone grafting, which typically occurs between the ages of eight and 11 years.

4.2.1. Dental care admissions

A total of 6,551 children born between 1997 and 2003 who were treated for a cleft lip and/or palate in England were identified. Among these children, there were 858 dental care admissions before the age of seven years. A total of 746 (11.4%) patients had at least one hospital admission for dental care before the age of seven years. Children with syndromes or additional anomalies (syndromic), who represented 21.5% of the sample, were more likely to have at least one dental care admission before the age of seven than non-syndromic children (17.5% vs. 9.7%). Syndromic children were also more likely to have multiple admissions for dental care compared to their non-syndromic counterparts (*Table 14*).

Table 14. Number (%) of dental care hospital admissions before the age of seven years, according to syndrome status, year of birth 1997-2003

	Number of admissions for dental care N (%)					
Syndrome status	0	1	2	3	4	All
Non-syndromic	4,642 (90.3)	446 (8.7)	49 (1.0)	4 (0.1)	0 (0.0)	5,141
Syndromic	1,163 (82.5)	201 (14.3)	38 (2.7)	7 (0.5)	1 (0.1)	1,410
All	5,805 (88.6)	647 (9.9)	87 (1.3)	11 (0.2)	1 (0.0)	6,551

Hospital Episode Statistics, England 1997-2011

Of the non-syndromic children, the proportion having at least one episode of dental care in hospital varied according to the type of cleft, with CL patients being the least likely to have an admission and BCLP patients the most likely (Table 15), which reflects the degree of dental decay (dmft) between different cleft types, as stated earlier in this report. Furthermore, patients with a BCLP were more likely to have multiple admissions for dental care compared to patients with other types of cleft.

The proportion of patients having at least one admission for dental care varied according to index of multiple deprivation (IMD). Analyses using quintiles of IMD revealed that the proportion of children having at least one dental care admission increased with increasing deprivation: 8% of children in the least deprived quintile have at least one dental care admission compared to 18% of children in the most deprived quintile. The relatively small

number of children having multiple dental care admissions prevents us being able to establish a relationship with deprivation quintiles.

Table 15. Number (%) of dental care hospital admissions among non-syndromic children before the age of seven years, according to cleft type, year of birth 1997-2003

	Number of admissions for dental care N (%)					
Cleft type	0	1	2	3	4	All
CL	1,291 (95.0)	65 (4.8)	2 (0.1)	1 (0.1)	0 (0.0)	1,359
CP	2,022 (91.4)	172 (7.8)	17 (0.8)	2 (0.1)	0 (0.0)	2,213
UCLP	920 (86.5)	128 (12.0)	14 (1.3)	1 (0.1)	0 (0.0)	1,063
BCLP	409 (80.8)	81 (16.0)	16 (3.2)	0 (0.0)	0 (0.0)	506
All	4,642 (90.3)	446 (8.7)	49 (1.0)	4 (0.1)	0 (0.0)	5,141

Hospital Episode Statistics, England 1997-2011

5. Linkage between CRANE and HES

5.1. Introduction

Linkage of CRANE to HES offers several advantages. First, information obtained through HES on co-morbidities and procedures can be used to verify cases in CRANE and validate the information recorded in CRANE e.g. cleft type. Second, linked HES data can provide additional information about the patient that is not currently collected by CRANE e.g. ethnicity, presence of additional anomalies, complications following a repair procedure, non-cleft related hospital admissions. These factors can then be considered when examining variation in the CRANE recorded patient outcomes. Third, provided that high case ascertainment and linkage rates are maintained, it is conceivable that HES data could be used in place of some aspects of direct data collection by CRANE (e.g. procedure information), thereby reducing the burden on those submitting data to CRANE.

5.2. Methods

We aimed to match each CRANE registered patient born between 2000 and 2009 to their corresponding HES identifier. We then explored:

- The success of the matching process
- The factors contributing to successful matching
- The level of agreement in cleft type between the two data sources

5.2.1. Linkage

Patient identifiers from CRANE were securely passed to the Trusted Data Linkage Service (TDLS) of the Health and Social Care Information Centre, who performed the linkage between records. Data were linked using a hierarchical deterministic approach, which involved matching patient records using the various patient identifiers: NHS number, sex, date of birth, and postcode

5.2.2. Data correspondence

For linked cases, the level of agreement in cleft type between the HES and CRANE data sources was examined. Where we found an inconsistency in the cleft type between the two data sources, we asked the centres to validate that the cleft type information recorded in CRANE was correct.

5.3. Results

Details of 8,966 CRANE-registered patients born between 1 January 2000 and 31 December 2009 were provided to TDLS and 84.8% of these could be linked to a HES record.

The linkage rate varied according to the presence of NHS number in the CRANE record; 92.6% of patients with NHS number present could be linked. This fell to 47.8% of patients where NHS number was not present. A total of 41 duplicate records were identified in CRANE as a result of the same HES identifier being linked to two different CRANE records.

For linked cases, there was a high level of agreement (>92%) in reported cleft type between the HES and CRANE data sources. This varied according to type of cleft; agreement was highest for the isolated cleft palate group (95%) and lowest for the bilateral cleft lip & palate group (81%).

The exercise to validate cleft type led to the information recorded in CRANE being changed in some instances. The CRANE cleft type error rate varied according to centre.

5.4. Conclusions

CRANE has been successful in linking 85% of its cases to HES. Linkage rates can be further improved through better capture of NHS number in CRANE. Cleft type as recorded in CRANE and the cleft type that can be derived from the patient's HES record are highly consistent. The small number of cases where an inconsistency does exist between HES and CRANE can be targeted as part of a validation process. Targeted validation using linked data allows the burden of the data checking process on the centres to be reduced.

6. Development of the CRANE Database and future directions

6.1. Development of a new consent form and patient information leaflet

In light of the recent NIGB approval CRANE received to collect and process a limited amount of patient-identifiable information without consent for the purpose of maintaining a register of cleft births, we will be revising the existing consent and patient information forms. Some Administrative Units will be consulted during this process. This is an immediate priority for CRANE as the new changes to the database will soon be live. These changes will allow Administrative Units to register new cases regardless of consent status.

6.2. Data linkage

The exercise to link the CRANE Database to HES was described previously in the 2011 Progress Report [21]. In September 2011 we were able to perform a corresponding linkage exercise to the Patient Episode Data for Wales (PEDW), the administrative database of admissions to the NHS hospitals in Wales, for patients registered by Swansea. It should be noted that cleft patients from North Wales have their surgery in Liverpool under the North West and North Wales Regional Centre.

The success of the linkage process relies on the completeness of patient identifiers and this has improved since our last report, as highlighted in the 2012 Progress Report [11].

CRANE has recently received an updated HES extract, containing hospital records up to 2012. The linkage exercises will be repeated annually.

The analysis of the linked data will:

- support data validation by assessing the correspondence of the recorded cleft type between the two data sources (CRANE Database and HES/PEDW);
- enable the examination of risk factors for delayed diagnosis of clefts in England and Wales;
- enable more extensive reporting of outcomes and treatment for children with clefting.

6.3. Future analyses

Surgical care for hearing

We intend to use HES and PEDW data to examine the trends and current delivery of surgical interventions for resolving otitis media with effusion in children with cleft palate with or without a cleft lip. We will assess if the NICE guidelines published in 2008 on this surgical procedure [22] had any impact on the delivery of care for this cohort of patients.

Evaluation of dental treatment

As highlighted in Section 4.2.1, CRANE has examined HES data to explore the burden of hospital care for dental disease. We hope to develop these analyses, examining CRANE-HES linked data to examine burden of care with CRANE outcome data, such as dmft scores.

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.

6.4. Exploring the potential of CRANE and HES

We intend to explore further the potential use of CRANE and HES data for the future reporting of cleft-related care. 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 quality dashboards, outcomes and performance reporting.

The inclusion of submitting data to CRANE as a requirement to the proposed National Service Specification, developed by the Clinical Reference Group (CRG), would be a welcome advance in improving the quality and completeness of data held in the CRANE database.

6.5. Collaboration

CRANE is collaborating with a number of individuals and organisations:

- 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 is currently collaborating with clinicians from the Paediatric Dentistry SIG of the CFSGBI on a paper focusing on dental disease among patients with a cleft in England.
- 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 from 2012. 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 determine whether CRANE could collect data on behalf of the 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

6.6. 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.
As part of the continuing development of recording outcomes on the Database, a number of groups of the CFSGBI have been approached to identify and recommend outcome measures which are appropriate for evaluating cleft care on a national basis.

Speech

The Cleft Audit Protocol for Speech—Augmented (CAPS-A) tool, a valid and reliable measure of speech outcome [23], has been piloted against agreed national speech standards derived by the Lead Speech and Language Therapy group in the last 24 months. Previous

recommendations about training and increasing the robustness of the measuring tool have been described [24]. This development within the UK is in contrast to the varied measures and reporting used in the US cleft teams [25]. We are in process of adding this outcome measure to the database. Administrative Units will be required to submit CAPS-A data at five years of age and at 10 years of age.

Psychology

The current lack of well defined valid and reliable measures among cleft patients is preventing the project from reporting on a range of cleft-related outcomes. Discussions have been ongoing with the appropriate SIG, who has advised CRANE that there are currently no agreed measures used consistently across all Administrative Units. Further, not all Units have access to psychologists. Work is currently underway with the Cleft Collective, who aims to have finalised psychosocial measures for their study by Spring 2013. CRANE will review these measures to consider whether any meaningful psychosocial outcome can be incorporated into the CRANE database.

6.7. CRANE Database meeting with users

CRANE organises meetings with representatives of the Administrative Units approximately once a year. The last meeting took place on 31 January 2012. Key points from the meeting are listed below.

Consent and NIGB approvals

- Changes to data entry screens to permit submission of 'notification data' without consent will be introduced later this year following NIGB review of CRANE security documentation (currently in progress). Teams will be kept informed of developments.
- Submission of outcomes data for existing registered patients where consent has not yet been validated will also be permitted.
- CRANE will provide teams with a form of wording to be used to explain to parents who decline consent for the treatment outcomes Database that notification data will still be submitted to CRANE and why this is permitted.

- CRANE will be introducing a system of more frequent password changes for users. Teams are reminded to advise CRANE immediately of any staff changes to the users at their centre.

Data collection

- The processes and barriers to collecting 5-year outcomes data (weight and height, dmft, 5-year index for complete UCLP cases) were discussed. Teams requested that CRANE advise them in the preceding April which cohort of patients will require outcomes data to be submitted by the data entry cut-off for the annual report. This reminder will be sent to all users, not just the clinical directors.

- The importance of verifying the patient's LAHSAL code in CRANE was emphasised. This is especially critical prior to submitting outcomes data. CRANE will provide further guidance on how and when this verification of LAHSAL code should be undertaken.

- Clarification of the definition of an 'At birth' diagnosis timing will be provided by CRANE.

- Submission of NHS number has improved. Teams are requested to continue monitoring the completeness of this data item in order to allow the highest possible proportion of CRANE patients to be linked to the HES database. The linkage exercises will be repeated annually.

The next CRANE Database Users' meeting is scheduled for Spring 2013.

6.8. Publications and presentations related to the CRANE Database

Publications

The following paper has been published:
Fitzsimons K, Mukarram S, Copley L, Deacon S and van der Meulen J. (2012) *Centralisation of services for children with cleft lip or palate in England: a study of hospital episode statistics.* BMC Health Services Research. 12(1): 148-155

The following paper is being prepared for peer review:
Fitzsimons K, Copley L, Deacon S and van der Meulen J. *Burden of hospital care among children born with a cleft lip and/or palate between 1997 and 2008: An analysis of Hospital Episode Statistics in England.*

Oral presentations

Fitzsimons K, Copley L, Deacon S, van der Meulen J. "Burden of hospital care among children born with a cleft lip and/or palate between 1997 and 2008: An analysis of Hospital Episode Statistics in England" **BINOCAR** (25 September 2012)

Fitzsimons K, Copley L, Deacon S, van der Meulen J. "Burden of hospital care among children born with a cleft lip and/or palate between 1997 and 2008: An analysis of Hospital Episode Statistics in England" **Craniofacial Society of Great Britain and Ireland (CFSGBI) Annual Conference** (26 April 2012)

Poster presentations

Copley L, Fitzsimons K, Deacon S, van der Meulen J. "Enhancing the potential of anomaly registers using linked Hospital Episode Statistics data" **BINOCAR** (25 September 2012)

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.

CRANE data showed that 42% of all babies born with a cleft were diagnosed in the antenatal period, which is the highest proportion since we started collecting this information. Seventy-three per cent of CLs, with or without a CP, were diagnosed antenatally, which is just below the recommended antenatal target detection rate of 75% set by the NHS Fetal Anomaly Screening Programme [26]. CPs are notoriously difficult to identify during antenatal screening (1% are diagnosed antenatally). National Standards state that clefts should be diagnosed within 24 hours of birth to enable immediate referral to a specialist cleft Unit [4]. In 2011, 28% of CPs remained undiagnosed at birth, with 5% remaining undiagnosed one month after birth. In 2010, one third of CPs were not diagnosed within 24 hours of birth, indicating that detection rates have improved, although further progress is required. Delays in diagnosis are concerning because they may lead to problems with feeding, breathing, nasal regurgitation and growth [27]. Furthermore, a CLAPA survey found that parents whose children had received a late diagnosis expressed feelings of distress, bitterness and resentment [28]. The finding that over one quarter of cleft palates are missed at birth reinforces our recommendation in both the 2010 and 2011 Annual Reports [3, 12], which called for maternity guidelines to include guidance on proper visualisation of the palate during examinations of the newborn child at delivery.

Referral times from maternity units varied according to Administrative Unit. Overall, 54% of all CRANE-registered children born in 2011 were referred to an Administrative Unit within 24 hours of birth. This represents a 4% reduction since 2010. Once children were referred, 90% were contacted by the Administrative Unit within 24 hours, with 10 out of the 15 Units contacting >95% of their patients within 24 hours of referral.

For the second year running, CRANE has reported outcomes at five years of age among children with a cleft. Although there is still a high proportion of missing data for these outcomes (45-77%, depending on outcome), this has improved substantially since last year (73-86%), when we first started collecting these outcomes. Height and weight were provided for 23% (14% last year) of children, data on the number of decayed missing and filled teeth (dmft) were provided for 55% (27% last year) of eligible children, and Five Year Old Index scores were submitted for 54% (23% last year) of eligible children. Administrative Units

reported a variety of reasons for the missing data. Five Units reported not collecting height and weight at five years of age, one Unit did not have a calibrated paediatric dentist who is required to determine dmft, and two Units reported not collecting Five Year Old Index. There are three Units who collected required outcomes but did not submit these to CRANE. One of these Units reported insufficient administrative support to allow submission to CRANE and is actively addressing this issue to allow submission in the future. The inconsistent collection and reporting of cleft-related outcomes between Administrative Units limits our ability to comment on the quality of care delivered and this continues to be a concern.

Of those children with reported outcomes, 41.9% of five year olds (born 2004-2006) had ≥1 dmft. This proportion is significantly higher than the 38.8% of five year olds in the general population [16]. Further, a clear and significant relationship between cleft type and dmft was observed; 35% of CL patients, 42% of CP patients, 45% of UCLP patients and 50% of BCLP patients had ≥1 dmft. Although the proportion of CL and CP patients with ≥1 dmft is not significantly different to the general population, children with a cleft receive additional dental care, with particular focus on the prevention of caries. Thus, it may be argued that children with clefts should have better oral health than the general population. The analysis of data from a greater number of children is necessary to further explore true differences that may exist between different cleft types and between the cleft population and general population.

Twenty-five per cent of children with a complete UCLP had externally validated Five Year Old Index Scores of '4' or '5', reflecting poor dental arch relationships. The number of children within each Administrative Unit with reported Five Year Old Index Scores ranged from 0 (four Units) to 29. CRANE will continue to collect these outcomes and meaningful comparison between Units will become possible once the number of children for whom we have outcome data increases.

Since last year's report, we have extended our burden of hospital care analyses using Hospital Episode Statistics (HES) to examine cleft-related procedure admissions and emergency admissions. We have also examined burden of care among syndromic cleft patients and have compared their admissions and total time spent in hospital with non-syndromic cleft patients. We found that non-syndromic children had a median of three all-cause admissions and spent 10 days in hospital in the first two years of life. In contrast, syndromic children had a median of five admissions and spent almost three times as long in hospital (27 days) for all causes. However, for cleft-related procedure admissions and time in hospital, there was no difference between the two groups. Admissions and days in hospital varied significantly according to the type of cleft, with BCLP patients having the most all-cause and cleft-related procedure admissions and total time in hospital. All-cause, cleft-

related procedure and emergency admissions and time in hospital also varied substantially between Administrative Units. These differences could not be explained by differences in cleft type distribution or deprivation.

It is important to recognise that differences between hospitals do not necessarily reflect differences in the quality of care, but may instead reflect differences in treatment protocols or geographic areas covered by each Unit. The factors we have highlighted that influence the number of admissions and time spent in hospital in the first two years of life should be considered when commissioning services and discussed with parents of children recently diagnosed with a cleft when providing counselling on future cleft care.

For the first time, we have extended our burden of care analyses to examine admissions to hospital for dental care, involving the surgical removal of teeth, simple extractions of teeth or the restoration of teeth before the age of seven years. We found that 11% of cleft patients had at least one hospital admission for dental care before their seventh birthday. There were differences between non-syndromic and syndromic children. Among non-syndromic children, 10% had at least one dental care hospital admission. The corresponding proportion among syndromic children was 18%. These children were also more likely to have multiple admissions for dental care. Differences in the proportion of children admitted at least once also varied according to cleft type and deprivation, with almost one fifth of children in the most deprived quintile having at least one dental care admission.

We recommend that health professionals involved in the care of patients with a cleft lip and/or palate review the key findings within this report and identify areas in which local improvements, particularly in terms of data completeness, may be required.

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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 North Bristol NHS Trust; University of Bristol
Kate Fitzsimons	Research Fellow	Clinical Effectiveness Unit
Lynn Copley	Data Manager	Clinical Effectiveness Unit
Jan van der Meulen	Clinical Epidemiologist	Clinical Effectiveness Unit; London School of Hygiene and Tropical Medicine
Jackie Horrocks	CRANE Administrator	Clinical Effectiveness Unit

Appendix 2: Governance and funding

Ownership

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

Cleft Development Group

The Cleft Development Group is a body with two distinct roles. Firstly, it is responsible for making arrangements for the running and commissioning of the CRANE Database.
Secondly, it is responsible for providing guidance on all aspects of the delivery of cleft care in England and Wales. It includes representatives from all the stakeholders in cleft care in England and Wales, including commissioners, public health consultants/regional cleft leads, specialists in the provision of cleft care, and parents and patients. It also has representatives from the health services in Wales, Scotland and Northern Ireland, as well as a representative from the Republic of Ireland cleft service.

Funding

Funding of the CRANE Database is currently coordinated and agreed by representatives of the national Specialised Commissioning Group for England and the Wales Specialised Health Services Committee. 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 Cleft Development Group

Adrian Sugar	Chair / Wales Clinicians
Jon Currington	Vice Chair / East Midlands Specialised Commissioning Group
Liz Albery	Leads Group of the Speech and Language Therapy SIG CFSGBI
Geoffrey Carroll	Medical Director, Wales Health Specialised Services Committee
Sue Carroll	Cleft Lip and Palate Association (CLAPA) Acting Chief Executive
Michelle Collard	Paediatric Dentistry (Special Interest Group (SIG) CFSGBI)
Scott Deacon	CRANE Clinical Project Lead
Mark Devlin	Scotland Clinicians
Adrian Drake-Lee	Cleft Surgery Interface Committee
Mandy Elder	East of England Specialised Commissioning Group
Sue Gregory	Department of Health (Dept. CDO England)
Per Hall	Cleft Surgeon (British Association of Plastic, Reconstructive and Aesthetic Surgeons (BAPRAS))
Chris Hill	Northern Ireland Clinicians
Nichola Hudson	Specialist Cleft Nurses (SIG CFSGBI)
David Landes	North of England Dental Public Health Consultant
Fiona Mackison	South East Coast SCG
Fiona Marley	National Specialised Commissioning Group
Kate Le Marechal	Clinical Psychologists (SIG CFSGBI)
Jan van der Meulen	Clinical Epidemiologist
David Orr	Ireland Clinicians
Stephen Robinson	Orthodontics (SIG CFSGBI)
Alison Sims	Cleft Co-ordinators and Managers (SIG)
Rona Slator	President of the CFSGBI
Peter Hodgkinson	Chair, Cleft Centres Clinical Directors/Managers Group;
Alistair Smyth	Cleft Surgeon (British Association of Oral and Maxillofacial Surgeons)
Mike Winter	Medical Director, National Services Division, Scotland
Ken Wragg	East Midlands Dental Public Health Consultant
Christopher Allen	Deputy for Ken Wragg and David Landes
Jackie Horrocks	Minutes Secretary, CRANE/Clinical Effectiveness Unit

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 organisations, 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.
3. 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".
4. The Database was UK wide when run by the Craniofacial Society of Great Britain and Ireland and before it became the responsibility of the CRANE Levy Board. Devolution of government in the UK resulted in four distinct health services and as a result CDG came to be responsible for a national Database for the recording of all children with clefts of the lip and/or palate born and treated in England and Wales, as the health service in Wales indicated its support for this development at an early stage. It has since then successfully sought to include in its work strong relationships also with the cleft services in Scotland, Northern Ireland and the Irish Republic.
5. 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.
6. 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.

7. 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.
8. 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 three years. The **Group** will elect its own Chair, who will remain in office for three 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 specialised commissioners from Specialised Commissioning Groups in England (nominated nationally), 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) (one to be 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 Interface Group on cleft surgery training
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 Dentists Special Interest Group)
12. The Co-ordinator/Chair of the UK Cleft Centres Clinical Directors' Group (1)

13. A Cleft Co-ordinator/Manager (1) (to be nominated by the Cleft Coordinators and Managers 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 England (1) / Northern Ireland (1) / Scotland (1) / Wales (1) (as appropriate, if not already represented) (to be nominated by those countries). There may also be a representative of the cleft service in Ireland.
16. There may be representation as determined by CDG as considered appropriate of any national bodies representative of Audit (1) and Research (1)
17. 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.
18. 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.
19. A representative of the DoH 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.
20. 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 specialist 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.

CDG amended and approved 21 Oct 2011

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

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

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

Classification of Surgical Operations and Procedures 4th Revision (OPCS-4) codes used to define a 'cleft-related procedure' admission for patients with a cleft diagnosis and history of a primary surgical cleft repair

OPCS 3-char code	Description
Chapter D:	**Ear**
D14	Repair of eardrum
D15	Drainage of middle ear
D20	Other operations on middle ear
Chapter E:	**Respiratory tract**
E02	Plastic operations on nose
E03	Plastic operations on nose
E07	Other plastic operations on nose
E08	Other operations on internal nose
E09	Operations on external nose
E10	Other operations on nose
E21	Repair of pharynx
Chapter F:	**Mouth**
F01	Partial excision of lip
F02	Extirpation of lesion of lip
F03	Correction of deformity of lip
F04	Other reconstruction of lip
F05	Other repair of lip
F06	Other operations on lip
F09	Surgical removal of tooth
F10	Simple extraction of tooth
F11	Preprosthetic oral surgery
F14	Orthodontic operations
F29	Correction of deformity of palate
F30	Other repair of palate
F32	Other operations on palate
F42	Other operations on mouth

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
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