CLEFT REGISTRY & AUDIT NETWORK

CRANE Database

2019 Annual Report

Results of the audit in England, Wales and Northern Ireland
for children born with a cleft
between January 2000 and December 2018

On behalf of the Cleft Development Group

www.crane-database.org.uk

Royal College
of Surgeons
ADVANCING SURGICAL CARE

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Hospital Episode Statistics (HES) data have been re-used with the permission of The Health and Social Care Information Centre. All rights reserved. Copyright © 2013.

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[1] Appendices 1 and 2 provide detail on the project team, and the project's Governance and funding arrangements. The Cleft Development Group CRANE web page provides detail on the CDG Membership and Terms of Reference.

Abbreviations

BCLP	Bilateral cleft lip and palate
CAPS-A	Cleft Audit Protocol for Speech—Augmented
CDG	Cleft Development Group
CEN	Clinical Excellence Network – previously referred to as Special Interest Group (SIG)
CSCs	Cleft Speech Characteristics
CFSGBI	Craniofacial Society of Great Britain and Ireland
CI	Confidence interval
CL	Cleft lip only
CLEFTSiS	The National Management Clinical Network for Cleft Service in Scotland
CP	Cleft palate only
CSAG	Clinical Standards Advisory Group
CRG	Clinical Reference Group
dmft	Decayed, missing and filled teeth at 5 years of age
DMFT	Decayed, missing and filled teeth at 10 years of age
DfE	Department for Education
DoH	Department of Health
EYFSP	Early Years Foundation Stage Profile – educational assessment at 5 years
ENT	Ear, nose and throat
ESQ	Experience of Service Questionnaire
GOSH	Great Ormond Street Hospital
FFT	Friends and Family Test
HES	Hospital Episode Statistics
ICD-10	International Classification of Disease 10th Revision
KS1	Key Stage 1 – educational assessment at 7 years
KS2	Key Stage 2 – educational assessment at 11 years
MCN	Managed Clinical Network
MDT	Multi-Disciplinary Team
NPD	National Pupil Database
OPCS-4	Classification of Surgical Operations and Procedures 4th Revision
PEDW	Patient Episode Database for Wales
PRS	Pierre Robin Sequence
RCPCH	Royal College of Paediatrics and Child Health
SCG	Specialised Commissioning Group
SD	Standard deviation

SDQ	Strengths and Difficulties Questionnaire
SLT	Speech and language therapy
TIM	Tiers of Involvement Measure
UCLP	Unilateral cleft lip and plate
WHO	World Health Organization

Glossary

Administrative Unit	A hospital that provides cleft surgery and submits data to the CRANE Database, sometimes as part of a wider cleft centre or network.
Alveolus / alveolar	The part of the jaw that supports the teeth and contains the tooth sockets.
Caries (dental)	Dental caries are also known as tooth decay / dental decay or a cavity.
Cleft	A failure of tissues to join during development.
Cleft care teams / Regions / Units	These terms are used interchangeably throughout this report and refer to the multidisciplinary group providing care for children with a cleft. See Appendix 3 for further information on Regional cleft units.
Cleft Development Group (CDG)	NHS National group representing all stakeholders in cleft care that is responsible for the CRANE Database as well as oversight and guidance on all aspects of the delivery of reorganised cleft care.
Cleft surgeon	A surgeon undertaking cleft repair surgery in a region / unit.
Clinical Standards Advisory Group (CSAG)	A group established in 1991 to act as an independent source of expert advice on standards of clinical care for, and access to and availability of services to, NHS patients.
Confidentiality Advisory Group (CAG)	An independent statutory body established to promote, improve and monitor information governance in health and adult social care. http://www.hra.nhs.uk/research-community/applying-for-approvals/confidentiality-advisory-group-cag/
Craniofacial anomalies	A diverse group of deformities in the growth of the head and facial bones.
Craniofacial Society of Great Britain and Ireland (CFSGBI)	An inter-specialty group set up to study cleft lip and palate and other craniofacial anomalies. www.cfsgb.org.uk
Denominator (see also Numerator)	In mathematical terms, the bottom number in a fraction. Considering that a fraction represents a part of a whole, the denominator represents the total number of parts created from the whole, for example 100 in 70/100. In the context of this report, we refer to the number of children in the cohort we are discussing that could meet a certain criteria. For example, children with a Cleft palate (CP) only.
Funnel Plot	A graph that identifies regions / units which are outliers, where the local situation might require closer inspection – either because an area is doing well or because there is some indication that it is performing poorly. In this report: • Each point on the funnel plot represents a region / unit. • Each funnel plot is for one outcome, with its values shown on the vertical/Y axis. • The size of the regions' /units' cohort is shown on the horizontal or X axis. • The benchmark value or overall national proportion is shown as a horizontal line through the centre of the graph. The graph shows two funnels that lie on either side of the benchmark and are called the control limits – similar to confidence intervals.

	• The inner lines show 2 standard deviations or 95% control limits. The outer lines represent 3 standard deviations or 99.8% control limits. • The funnel shape is formed because the control limits get narrower as the population size increases. The outer funnel is used to decide if an area is significantly different to the benchmark with 99.8% confidence. If a point lies within the funnel then we conclude that it is not significantly different to the benchmark. If it falls outside the funnel then we can say the value is significantly 'better' or significantly 'worse' than the benchmark, depending on the direction of the indicator/outcome. Funnel Plot Source: David Spiegelhalter, Medical Research Council Biostatistics Unit - http://www.erpho.org.uk/Download/Public/6990/1/INPHO%204%20Quantifying%20performance.pdf
General Population	In Epidemiological terms, all individuals without reference to any specific characteristic. In the context of this report, and to aid comparison, we sometimes refer to the latest national figures for children in the general population, which may also include children with a cleft or other health conditions. E.g. Gestational age and birth weight in the general population of England & Wales in 2016, according to the Office for National Statistics (ONS) (Section 3.4). In some instances, the latest national figures are based on a random sample of children in the general population, which, again, may include children with a cleft or other health conditions.
Hospital Episode Statistics (HES)	A national database containing records on all admissions to NHS hospitals in England.
Key Stage 1 (KS1)	An assessment of children's educational attainment across five subject areas at seven years of age.
Key Stage 2 (KS2)	An assessment of children's educational attainment across five subject areas at 11 years of age.
LAHSAL	A code used to classify clefts. Each letter (LAHSAL) relates to one of the six parts of the mouth that can be affected by a cleft.
Managed Clinical Network (MCN)	A formally organised network of clinicians.
National Pupil Database (NPD)	A database containing records on all pupils in England as they progress through primary and secondary education.
Numerator (see also Denominator)	In mathematical terms, the top number in a fraction. Considering that a fraction represents a part of a whole, the numerator represents how many parts of that whole are being considered, for example 70 in 70/100. In the context of this report, we refer to the number of children meeting a certain criteria. For example, receiving a certain type of care or meeting a standard.
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

CRANE
RCS
Cleft Registry & Audit NEtwork

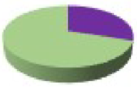
Annual Report 2019

CRANE aims to audit, evaluate and report on the delivery of cleft services to children in England, Wales and Northern Ireland with the congenital abnormality of cleft lip and/or palate.

Registrations, early care and diagnosis

20,013 children registered over the last nineteen years – from 2000 to 2018.

1,002 New registrations in 2018.



29.7% of children with a Cleft Palate (CP) received a delayed diagnosis, beyond 72 hrs after birth – from 2014 to 2018.

All parents / carers of children born in 2018 were approached for consent. Of the families that had made their decision, 99.2% of these provided consent. This is extremely positive, as consent is essential for the collection of a full dataset and the linkage to other datasets.

Outcomes for children aged 5 years and older

Dental health 41% of children with a cleft had at least one decayed, missing or filled tooth (>0 dmft) at the age of 5 years, which is higher than the rate in the general population (31%).

Dental health 23% of children with a cleft had at least one Decayed, Missing or Filled Tooth (>0 DMFT) at the age of 10 years.

Facial growth 26% of children had scores reflecting poor dental arch relationships at the age of 5 years.

Facial Growth & Speech No significant relationship was found between facial growth and speech among children with a complete unilateral cleft lip and palate at the age of 5 years.

Speech 61% of children with a cleft had speech scores that suggest their speech is not significantly different to their non-cleft peers at the age of 5 years. The proportion of children with 'normal' speech varies considerably according to cleft type.

Psychology 16% of children with a cleft had high or very high 'Strengths and Difficulties' scores¥ at the age of 5 years. This compares with 10% among their non-cleft peers.

Educational attainment Children born with isolated clefts had lower educational attainment than children in the general population in all subject areas throughout their primary education.

Data completeness

Cleft care teams should pay particular attention to recording the following information in the CRANE Database:

- Gestational age and birth weight (47% and 48% missing data, respectively),
- weight and height at the age of 5 years (61% missing data for both measures), and
- Decayed missing and filled teeth (DMFT) at the age of 10 years (86% missing data).

www.CRANE-Database.org.uk

@CRANE_News

Further information

¥Scores in the 'high' and 'very high' ranges indicate a greater level of difficulties than low scores; classified as being in the 'close to average' range (indicating no concern).

Recommendations

For cleft care teams within NHS organisations

CRANE registrations and early cleft care

1. Cleft care teams should specify on the CRANE Database:

- the cleft type for 100% of the babies receiving care at their unit, and
- the sex of 100% of the babies receiving care at their unit.

2. Cleft care teams should aim to establish contact with parents within 24 hours of referral.

3. Cleft care teams should aim to return to the families who have yet to make their decision about consent to confirm agreement/declining of consent (the consent status of 36.8% (N=374) of registrations in 2018 were unconfirmed).

Child growth

4. Cleft care teams should aim to assess children's weight and height around the age of 5 years and report this on the CRANE Database.

Dental health

5. Cleft care teams should aim to see all children with a cleft for:

- a decayed, missing or filled teeth (dmft) assessment at the age of 5 years, and
- a decayed, missing or filled teeth (DMFT) assessment at the age of 10 years.

Facial growth

6. Cleft care teams should aim to take dental impressions of all children with a complete unilateral cleft lip and palate (UCLP) around the age of 5 years, to allow for an assessment using the Five Year Old Index.

Speech

7. All children with an isolated cleft affecting the palate (CP, UCLP and BCLP) should have their speech assessed using all 16 CAPS-A parameters at the age of 5 years. All 16 scores should be reported to the CRANE Database.

- Differences in outcomes between units should be explored further to determine whether there are certain practices, such as the timing of the palate repair, that are associated with more favourable speech outcomes.

8. Cleft care teams should continue to assess and report speech outcomes among children with Pierre Robin Sequence (PRS) and other syndromes (e.g. sticklers syndrome and Van der Woude syndrome), where possible, so that we can examine differences in outcomes between clinical subgroups.

Psychology screening

9. Cleft care teams should aim to:

- see all children and families born with a cleft for a first face-to-face psychosocial screen before the age of 6 years,

- see all children and families born with a cleft for a ‘psychosocial screen at age five’ before the age of 6 years,
- screen all children born with a cleft, using the Tiers of Involvement Measure (TIM) and the Strengths and Difficulties Questionnaire (SDQ) ‘Total difficulties’ scale bands, before the age of 6 years.

Completeness of data items

10. Cleft care teams should:

- register all children with a cleft in the CRANE Database,
- record the time of referral and time of first contact for all children in the audit database, and
- record the following outcome information for consented children around the age of 5 years in the audit database:
    - dmft for all children with a cleft,
    - Five Year Old Index scores for all children with complete Unilateral Cleft Lip and Palate (UCLP),
    - all 16 CAPS-A parameters information for all children with a cleft affecting the palate, as well as for children with Pierre Robin Sequence (PRS) and other syndromes, where possible, and
    - psychology screening for all children with a cleft (specifically before the age of 6 years).

11. Cleft care teams should pay particular attention to recording the following information, with current rates of completeness below 60% in the audit database:

- Gestational age and birth weight (54% completeness for child growth at birth outcome data),
- weight and height at the age of 5 years (39% completeness for both measures),
- DMFT for all children with a cleft at the age of 10 years (14% complete),
- both Five Year Old Index and CAPS-A parameters for children with a non-syndromic complete UCLP at the age of 5 years (59% of cases had both outcomes reported) – this is particularly important to establish if facial growth and speech outcomes are associated, and
- psychology screening of children using the SDQ ‘Total difficulties’ scale bands, before the age of 6 years (54% complete) – this is particularly important to aid identification of all psychological concerns arising for children born with a cleft ant their families at age five years.

For professional organisations involved in the care of children with a cleft

Diagnosis

12. Some minor improvement has been observed in the timely detection of cleft palates since we drew attention to this issue seven years ago. Therefore, further collaboration with the medical workforce is required to identify barriers to the detection of cleft palate within 24 hours of birth. This will include further work with the Royal College of Paediatrics and Child Health to further develop the learning modules and training opportunities related to timely cleft palate detection.

Educational attainment

13. Further work is needed to identify risk factors associated with lower educational attainment in this clinical population. Those involved in commissioning and planning special educational services should be aware that children with a cleft may require additional academic support or specific support around the treatment pathway.

1. Introduction

The Cleft Registry & Audit Network (CRANE) Database is a national register that was established in 2000 to collect information on children born with a cleft lip and/or palate in England and Wales[2]. Northern Ireland officially joined the project in 2015. The geographical representation of the cleft regions / units is detailed in Appendix 3.

The Database collects birth, demographic and cleft diagnosis information. It also collects information about cleft-related treatment and outcomes. Hospital Episode Statistics (HES) and the National Pupil Database (NPD) are used to further examine treatment and outcomes for cleft lip and/or palate in England.

The aims of the CRANE Database are:

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

This Annual Report presents findings from data submitted to the CRANE Database[3] for children with a cleft lip and/or palate born in England, Wales and Northern Ireland between the 1 January 2000 and 31 December 2018. We describe:

- the timing of cleft diagnosis for children born in 2018 and the timing of referral and contact with cleft teams;
- cleft-related outcomes for children, registered in the CRANE Database, at five years of age (born 2004-2012);
- analyses of data from Hospital Episode Statistics (HES) data linked to the CRANE Database at the individual level for children born from 2004 to 2012. We describe the results exploring factors impacting on dental treatment and care among children with a cleft;
- analyses of the CRANE Database linked to the National Pupil Database (NPD), which contains records on all pupils in England as they progress through primary and secondary education. We describe differences in educational attainment (the 'attainment gap') between children with clefts and the general population at ages 5 (between 2006 and 2008), 7 and 11 years.

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

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[2] For further information on the background to the CRANE Database please visit https://www.crane-database.org.uk/

[3] Registered in the CRANE Database by the 10 July 2019.

2. Methods

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

2.1. CRANE

2.1.1. Data source

CRANE is an online custom-built secure database that holds information on children born with a cleft lip and/or palate in England, Wales and Northern Ireland. The CRANE Database 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. The CRANE Database also collects information about cleft-related treatment and outcomes. These data are reported to CRANE by the units that make up 10 Regional Cleft Centres / Managed Clinical Networks (as listed in Appendix 3). Each child born with a cleft in England, Wales and Northern Ireland should be referred to one of these units shortly after having their cleft diagnosed.

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

2.1.2. Patients

All data entered into the CRANE Database by 10 July 2019 pertaining to children born between 1 January 2000 and 31 December 2018 is included in the descriptions and analyses described in this Annual report. Patients whose parents have not consented to their data being used by CRANE have been excluded from the sections and tables in this report on: (1) 5-year outcomes, (2) HES analyses, and (3) NPD analyses (as the data presented in these sections and tables are not collected for non-consenting cases).

2.1.3. Data validation and cleaning

Logical and systematic data cleaning was undertaken to identify any potential data errors. Continuous data variables (birth weight, 5-year weight and 5-year height) were assessed in relation to valid ranges. Valid ranges for 5-year body weight and 5-year height have been defined according to growth charts published by the World Health Organisation (WHO)[4].

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[4] UK-WHO growth charts - 2-18 years (checked October 2019).
Available from: https://www.rcpch.ac.uk/resources/uk-who-growth-charts-2-18-years.

2.1.4. Analyses

Data have been analysed according to year of birth, unless otherwise stated. Five-year outcome data were restricted to children born between 2004 and 2012, depending on the outcome of interest. Children dying before five years of age were excluded from these analyses and from the denominator when calculating data completeness.

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 (9.5% of children born in 2018), cleft type is based on the type reported by the region/ unit registering the child. Children with a unilateral cleft lip and palate (UCLP) were categorised according to whether the UCLP was complete or incomplete. A complete UCLP was defined as LAHS or HSAL codes, indicating a complete cleft affecting all three components of the mouth on either the right or left side.

Decayed, missing and filled teeth (dmft at 5 years, DMFT at 10 years)

The dmft/DMFT score describes the dental caries experience of an individual and is a measure of oral health. A dmft/DMFT score reflects the total number of teeth that are decayed, missing or filled. Analyses on dmft data were restricted to consented children born between 2004 and 2012, and analyses on DMFT data were restricted to consented children born in 2007 and 2008 (excluding children with a submucous cleft palate).

Five Year Old Index

Dental models of 5-year old children with UCLP can be assessed using the Five Year Old Index to examine dental arch relationships. The index evaluates the effects of primary surgery on the facial growth of children with UCLP before any other interventions are performed, such as orthodontics or alveolar bone grafting, which may influence this growth further[5]. The CRANE Database collected both internal and external Five Year Old Index scores for consented children born between 2004 and 2012 with a complete UCLP (LAHSAL codes LAHS or HSAL). Some units score the models of children treated in their unit (internal scores) before they are sent off to be scored externally (external scores) by a blinded process undertaken by calibrated examiners. For this report we have analysed externally validated scores where available; where these were unavailable, internal scores are included in the analyses.

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

CAPS-A ratings collected at five years of age among children born with a cleft affecting the palate are reported to the CRANE Database for consented children only. The parameters of speech assessed include

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

resonance (hypernasality and hyponasality), nasal airflow (audible nasal emission and nasal turbulence) and twelve Cleft Speech Characteristics (CSCs) scores, including:

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

Speech outcomes are reported for non-syndromic children, born 2010-2012, who have had all 16 CAPS-A ratings reported to the CRANE Database.

For the first time, CAPS-A ratings have also been assessed for children born between 2007 and 2012 with a non-syndromic cleft palate and a diagnosis of Pierre Robin Sequence (PRS). Speech outcomes among those with PRS have been compared to those born across the same time period with a non-syndromic cleft palate without a PRS diagnosis.

Psychology

Children are screened by psychologists at 5 years of age (and sometimes prior to that) using the Tiers of Involvement Measure (TIM) and the Strengths and Difficulties Questionnaire (SDQ). The CRANE Database collected TIM and SDQ scores, as well as dates of psychological screening, for consented children born in 2011 and 2012 with all cleft types.

The TIM is used to record the tier (level) of involvement when a Psychologist sees a patient/family in a Cleft Multi-Disciplinary Team (MDT) Clinic. The tiers are as follows:

0. Patient not seen by Psychologist.
1. Patient seen and psychosocial screen completed.
    a. No psychological concerns requiring cleft psychological input.
    b. Psychological support and/or needs met by other services e.g. Child and Adult Mental Health services (CAMHs).
2. Psychological input provided in clinic.
    a. Preventative input only.
    b. Input in response to a problem/concern raised by family/child.
    c. Both preventative input and input in response to problem/concern raised by family/child.
3. Further action required by Psychologist but appointment not necessary (e.g. liaison with school, written information sent to family).
4. Psychologist appointment necessary (for the purposes of this report, TIM scores 4, 5 and 6 are included in this category[6]).

The SDQ is a brief behavioural screening questionnaire designed for use with 3-16 year olds. The SDQ asks about 25 attributes, some positive and others negative, which are divided between the following scales:

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[6] A score of 5 refers to a psychology appointment deemed as needed but resources do not allow for this to be offered in a timely way. A score of 6 refers to families who are already receiving psychology appointments when they are seen at age 5 years.

1. emotional symptoms (5 items)
2. conduct problems (5 items)
3. hyperactivity/inattention (5 items)
4. peer relationship problems (5 items)
5. prosocial behaviour (5 items)
6. scales 1 to 4 are added together to generate a 'Total difficulties' score (based on 20 items).

The CRANE Database collects the 'Total difficulties' score as well as the final scores for subscales 1 to 5, resulting from questionnaires completed by the parents of CRANE-registered children at 5 years of age[7].

Exploration of the data collected using the six SDQ scores has been conducted according to their categorisation into the following four bands:

1. close to average
2. slightly raised
3. high
4. very high.

Missing data

Missing data have been excluded from the denominators presented in all Tables/Figures and Appendices of this report, with the exception of Tables/Figures relating to data completeness. All units have some degree of missing data. The number of patients with missing data for 5-year outcomes can be high. A variety of reasons are reported by units. Reasons out of a unit's control include children not attending an appointment or moving away from the area.

2.2. Hospital Episode Statistics (HES)

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

The HES database holds diagnostic and procedure information on each patient, allowing us to identify those with a cleft lip and/or palate and those undergoing cleft-related treatment. In addition to being able to identify and confirm cleft type in the CRANE Dataset, HES is used by CRANE to identify any additional anomalies for the CRANE cohort (see Appendix 4 for a list of the HES diagnosis and procedure codes used by CRANE). This allows the categorisation of children in CRANE as 'non-syndromic' or 'syndromic'.

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[7] Using the parent version for 4-16 year olds. Goodman R (1997) The Strengths and Difficulties Questionnaire: A Research Note. Journal of Child Psychology and Psychiatry, 38, 581-586. For more information visit www.sdqinfo.com

2.3. National Pupil Database (NPD): Educational attainment

2.3.1. Data source and linkage

The National Pupil Database (NPD) is a national database containing records on educational outcomes for all pupils in England from the 1995/1996 school year onwards. The initial year for which Key Stage attainment data were first collected varies according to the examination of interest. The CRANE Database sought and was granted permission by the Department for Education (DfE), in accordance with their published application process [8], to link the information held in the CRANE Database with the NPD.

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

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

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

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

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

2.3.2. Patients

Consented, CRANE-registered children with a non-syndromic cleft, who were 5 years old between 1 September 2006 and 31 August 2008, and had CRANE-HES-NPD linked records were eligible for inclusion in our analyses of longitudinal school attainment.

2.3.3. Outcomes and analyses: Educational attainment

We analysed teacher-assessed academic attainment in four subject areas: 'Reading', 'Writing', 'Maths' and 'Science' (assessed as knowledge and understanding of the world at age 5). Teachers provided an assessment of each pupil's attainment against national expected levels of knowledge, skills and understanding in each subject at each assessment stage. Attainment was assessed using a scale ranging

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

from 0 to 9 points at age 5 years, and attainment levels at ages 7 and 11 years (Table 2.1). Attainment levels were converted to point scores to enable comparison across the three stages (Table 2.2).

Table 2.1. Expected attainment levels for children at the end of each Key Stage

Key Stage	Age at assessment (years)	Expected attainment at end of Key Stage within each scale/subject
Early Years Foundation Stage	5	Achieving at least 6 points
Key Stage 1	7	Level 2 (15 points)
Key Stage 2	11	Level 4 (27 points)

Table 2.2. Conversion of teacher-assessed attainment level to point scores [11]

Teacher Assessment level	Point score equivalent
Working towards level 1	3
1	9
2	15
3	21
4	27
5	33
6	39

At each age, the DfE summarise educational outcomes as the proportion achieving expected levels of attainment in each subject. At ages 7 and 11, national data on the proportions of children achieving each attainment level were converted to mean points scores and standard deviations (SD) using the conversions described in Table 2.2. For individual children with a cleft, attainment levels were also converted into mean point scores. Differences in attainment between children with a cleft and the general population at each age were summarised using Z-scores, calculated for each subject within each assessment year using published national data on educational attainment[9]. The calculated Z-scores represent the number of SDs by which observed individual scores differ from the national average in the general population, with a negative Z-score indicating a score below the national average and a positive score indicating a score above. Mean Z-scores and associated 95% confidence intervals (CI) are presented for each subject area and Key Stage. To determine whether there was any change in the size of the attainment gap with age, mean Z-scores for each subject were compared across the three Key Stages using repeated measures analysis of variance. All statistical analyses were conducted using Stata V.15 (StataCorp, College Station, Texas, USA).

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[9] Department for Education. *DCSF: Early Years Foundation Stage Profile Results in England, 2008/09*. 2009 [cited 19 February 2019]; Available from: https://webarchive.nationalarchives.gov.uk/20130323143251/https://www.education.gov.uk/researchandstatistics/statistics/allstatistics/a00196194/early-years-foundation-stage-profile-results-in-en; Department for Education. *National curriculum assessments at key stage 1: 2011* 2011 [cited 19 February 2019]; Available from: https://www.gov.uk/government/statistics/national-curriculum-assessments-at-key-stage-1-in-england-2011; Department for Education. *National curriculum assessments: key stage 2, 2014 (revised)* 2014 [cited 19 February 2019]; Available from: https://www.gov.uk/government/statistics/national-curriculum-assessments-at-key-stage-2-2014-revised.

3. Registrations and early care

In this chapter we present findings on children registered in the CRANE Database, with a cleft lip and/or palate, with regards to registrations and contact with cleft teams around the time of birth.

3.1. Characteristics of children registered in CRANE

Cohort summary	Data source	CRANE database
	Birth year	2018
	Denominator	1,002 children registered in the specified birth year.
	Numerator	Number of children meeting each patient characteristic.
	Data completeness	• 90.5% of registrations had a specified cleft type. • 98% of children had their sex reported to the audit database.
	Countries	England, Wales and Northern Ireland.
Benchmark	100% of children with a cleft should be registered in CRANE at birth, and 100% of registered children should have their cleft type and sex reported to the database.	
What did we find?	• Cleft Palate was the most common form of cleft, representing 39% of all cases with a known cleft type. • 9.5% of children had an unspecified cleft type. • 55% of children were boys. They were significantly more likely to have a CL, UCLP or BCLP than girls with a cleft.	
Recommendations	Units should aim to: • register all children with a cleft in the audit database, • specify the cleft type for 100% of the babies receiving care at their unit, and • specify the sex for 100% of the babies receiving care at their unit.	

Of the total 20,013 children born and registered in the CRANE Database over the last nineteen years[10], 1,002 were born in 2018[11]. Among these 1,002 children:

- Cleft palate (CP) continues to be the most common of the four cleft types[12], representing 39.3% of registrations.
- 21.0% of registrations were classified as unilateral cleft lip and palate (UCLP), of which 62.9% had complete UCLP (defined by either 'LAHS..' or '..HSAL' by the LAHSAL code).
- Cleft Lip (CL) represented 19.6% of registrations.
- Bilateral cleft lip and palate (BCLP) is the least common type, representing 10.6% of 2018 registrations.
- 9.5% of registrations did not have their type of cleft specified (i.e. it had yet to be reported by the region/unit registering the child).

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[10] 1 January 2000 and 31 December 2018.

[11] Among all these children, five deaths were reported to CRANE (0.5% of 2018 registrations). Most of these occurred between one month and one year of age. It is not known from CRANE whether these children had additional anomalies or syndromes.

[12] Cleft type is defined according to reported LAHSAL codes or, where LAHSAL has not been reported (for 9.5% of children registered in 2018), it is based on the cleft type reported by the region / unit registering the child.

- 55% percent of children born in 2018 were boys[13]. Boys were significantly more likely to have a CL, UCLP or BCLP than girls with a cleft ($p<0.001$)[14].
- CP was significantly more prevalent among girls (58% of CP cases were girls and 42% were boys, $p<0.001$).

Visit the CRANE Database website https://www.crane-database.org.uk/ to review the Tables on registrations over the last 10 years, by cleft type and year of birth, according to region / unit.

3.2. Contact with cleft teams

Cohort summary	Data source	CRANE database
	Birth year	2018
	Denominators	• 608 children with recorded time of referral to cleft teams. • 867 children with recorded time of first contact.
	Numerators	• 527 children referred to cleft unit within 24 hours. • 832 children contacted by cleft team within 24 hours of referral.
	Data completeness	Of the children born in 2018 and registered in CRANE (n=1,002): 60.7% had a recorded referral time, and 86.5% had a recorded time of first contact.
	Countries	England, Wales and Northern Ireland.
Benchmark	• Children with a cleft should be referred to cleft teams within 24 hours of birth. • Cleft teams should establish first contact with families of children with a cleft within 24 hours of receiving a referral.	
What did we find?	• 87% of families were referred to a cleft team within 24 hours of birth. This proportion varied significantly across cleft types. • 96% of families were contacted within 24 hours by their regional cleft unit. This proportion varied significantly across cleft teams.	
Recommendations	Units should aim to ensure that: • contact is established with parents within 24 hours of referral, and • the time of referral and time of first contact is recorded in the audit database.	

Out of 1,002 children born in 2018 and registered in CRANE, 608 (60.7%) children had a recorded referral time. Of these:

- 86.7% were referred to a Cleft Unit within 24 hours of birth.
- The proportion of referrals within 24 hours of birth varied significantly according to cleft type ($p<0.001$), with CP patients having the lowest proportion of early referrals, which is consistent with later diagnosis times for these children.
- The proportion of referrals within 24 hours of birth also varied according to cleft /administrative unit, although not significantly.

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[13] Twenty children did not have their sex reported to CRANE (2% of the total children registered)

[14] Males comprised 62.4% of CL cases, 42.1% of CP cases, 69.1%% of UCLP cases, and 65.7%% of BCLP cases.

- The patterns of referral according to time of diagnosis were consistent with patterns described in past years[15].

Of the 867 (86.5%) children with recorded time of first contact with cleft teams:

- Units established contact with 96% of parents within 24 hours of referral.
- The proportion of units establishing contact with parents within 24 hours of referral did not vary significantly according to cleft type. This is consistent with patterns described in past years[16]
- The proportion of units establishing contact with parents within 24 hours of referral varied significantly according to the cleft /administrative unit ($p<0.001$). Despite this statistically significant variation between units, overall rates of contact within 24 hours remain high (as for previous reporting years).
- Overall, units contacted at least four in five parents of their patients (78.4%) within 24 hours of receiving the referral. This demonstrates the commitment of units to ensure a timely response to new referrals of babies born with a cleft, to help support these babies and their families in the important initial stages.

3.3. CRANE consent

Cohort summary	Data source	CRANE database
	Birth year	2018
	Denominator	1,002 children born in the specified birth year and registered in CRANE.
	Numerators	1,002 families approached for consent. 633 families who had made decision about consent
	Data completeness	100% of families of children with a cleft were approached for consent for CRANE data collection.
	Countries	England, Wales and Northern Ireland.
Benchmark	• 100% of families of children with a cleft should be approached for consent.	
What did we find?	• 63.2% of families had reached a decision about providing (or declining) consent. • Of the families who had reached a decision, 99.2% provided their consent.	
Recommendations	Units should aim to return to the families who have yet to make their decision about consent to confirm agreement/declining of consent (36.8%, N=374 in 2018).	

With regards to families being approached for consent to collect data into childhood (beyond registration and diagnosis):

- The parents/carers of all 1,002 children born in 2018 had a record of being approached for consent.
- Of the families approached for consent, the decision to provide or decline consent had been made by 63.2% of families[17].
- Of those families that had reached a decision to provide or decline consent, 99.2% agreed to their children's data being collected by CRANE (by providing consent). This is extremely positive.
- The proportion of families that had reached a decision to provide or decline consent varied across the regions / units submitting data to CRANE (from 22.3% to 100%). This remained consistent with

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[15] For past CRANE Database Annual Reports please visit https://www.crane-database.org.uk

[16] For past CRANE Database Annual Reports please visit https://www.crane-database.org.uk

[17] Cleft care teams were awaiting decisions from 35.9% of families, and it had not been possible to consent 0.9% of families.

proportions reported in past years (for detail on this please consult previous CRANE Database Annual Reports).

- A marked improvement in the process for approaching parents for consent appears to have taken place in recent years, including in 2018. Although 36.8% of families approached in 2018[12] had yet to make their decision, regions' / units' approach to seeking consent in 2018 was extremely positive, as consent is essential for the collection of a full dataset and the linkage to other datasets.

3.4. Gestational age and birth weight

Cohort summary	Data source	CRANE database (consented cases only)
	Birth year	2018
	Size (N)	• 334 children with recorded gestational age, and • 326 children with recorded birth weight in the specified birth year.
	Data completeness	54% of eligible children (n=628) had recorded 'child growth at birth' (gestational age, birth weight, both) or a valid reason it was not collected [18].
	Countries	England, Wales and Northern Ireland.
Benchmark	• 7% of babies in the general population of England & Wales are born prematurely. • Healthy birth weights for babies born in England and Wales range from 2.5Kg to 4Kg[19]. • 100% of eligible children should have a recorded gestational age and birth weight (or a valid reason it was not collected).	
What did we find?	• The mean gestation period was 38.5 weeks. • 15% of babies were born prematurely. • The average birth weight was 3.1Kg.	• 46% of eligible consented babies (n=628) were missing recorded 'child growth at birth' (or a valid reason it was not collected).
Recommendations	Units should aim to improve completeness of recording of gestational age and birth weight in the audit database.	

Of the 628 children whose families had consented to their children's data being collected by CRANE in 2018, 334 (53.2%) had gestational age and 326 (51.9%) had birth weight reported to CRANE. Analyses revealed that:

- The mean gestation for those born in 2018 was 38.5 weeks (95% CI 38.2 to 38.8 weeks) and ranged from 23 to 45 weeks[20].
- Fifty (15.0%) babies were premature (born before 37 weeks' gestation), which is higher than the seven percent national average in England and Wales[21]. It should be noted that the gestation recorded in

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[18] Note: Only 323 babies had both a recorded gestational age & birth weight (51.4%), and 1 baby had a recorded 'reason not collected' (0.001%).

[19] Birthweights under 2.5Kg considered low and over 4Kg considered high - Office for National Statistics. Birth characteristics in England and Wales: 2016. Published 16 October 2017 (this is the latest release – checked October 2019). Available from: https://www.ons.gov.uk/peoplepopulationandcommunity/birthsdeathsandmarriages/livebirths/bulletins/birthcharacteristicsinenglandandwales/2016

[20] Gestational age: Reported for 334 (53%) of consented babies born in 2018. Highlighting need for improved data completeness.

[21] Office for National Statistics. Gestation-specific infant mortality. Part of Gestation-specific infant mortality in England and Wales, 2013. Published 14 October 2015 (this is the latest release – checked October 2019). Available from: http://www.ons.gov.uk/ons/rel/child-health/gestation-specific-infant-mortality-in-england-and-wales/2013/stb-gestation-specific-infant-mortality.html.

CRANE may not be representative of all babies born with a cleft lip and/or palate as 46.8% of consented children were missing this information.

- The mean birth weight was 3.1kg (95% CI 3.1 to 3.2kg), which is consistent with the national average in England & Wales.

4. Diagnosis

In this chapter we present findings on the timing of diagnosis for children registered in the CRANE Database, with a particular focus on cleft palate diagnoses.

4.1. Timing of diagnosis for all cleft types in 2018

The majority of all babies born with a cleft in 2018 were diagnosed antenatally (46.9%) or at birth (40.5%). The proportion of children diagnosed antenatally varied significantly according to cleft type ($p<0.001$), with only 2.6% of children with CP diagnosed antenatally compared with rates of 69%, 92.4% and 91.4% for CL, BCLP and UCLP, respectively.

Please view 'Table 3. Diagnosis time' on the CRANE Database website for detail on timing of diagnoses by cleft type for all 2000-2017 births and all 2018 births.

4.2. Diagnosis times for children with a cleft palate

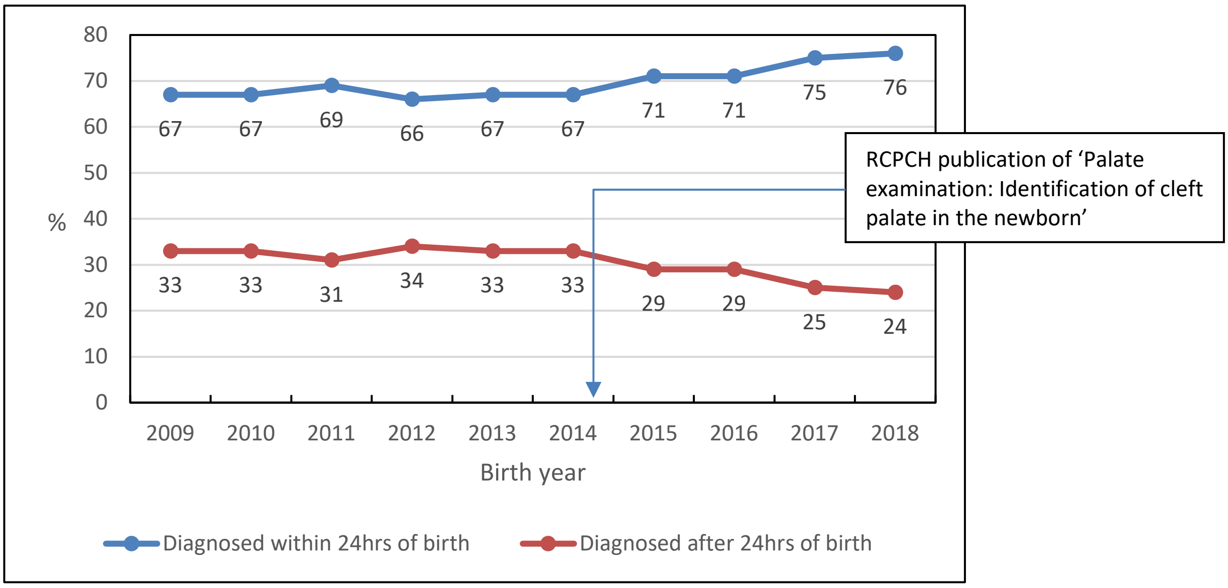
Cohort summary	Data source	CRANE database (consented cases only)
	Birth years	• 10 years: 2009 to 2018 to explore trends over time. • 5 years: 2014 to 2018 to explore regional differences.
	Denominators	• For 2009-18 births: 4,210 children with cleft palate alone, with a recorded timing of diagnosis. • For 2014-18 births: 2,040 children with cleft palate alone, with a recorded timing of diagnosis.
	Numerator	Number of children diagnosed at each time point with cleft palate alone.
	Data completeness	• 92% of children born 2009-18 with a CP (n=4,576) had a recorded timing of diagnosis. • 95% of children born 2014-18 with a CP (n=2,147) had a recorded timing of diagnosis.
	Countries	England, Wales and Northern Ireland.
Standard	100% of cleft palates should be diagnosed antenatally or within 24 hours of birth.	
What did we find?	• Some minor improvement has been observed in the timely detection of cleft palates in the last 10 years. CRANE will continue to monitor this to see if this an ongoing trend. • 72% of CPs of children born 2014-18 were diagnosed antenatally or at birth. • Diagnoses at birth by region ranged from 61.3% the South Thames to 81.8% in the West Midlands (for children born 2014-18). • CPs with complete soft/hard palate are more likely to be diagnosed before/at birth than cleft palate with incomplete soft/hard palate (for children born 2014-18).	
Recommendations	Some minor improvement has been observed in the timely detection of cleft palates since we drew attention to this issue seven years ago. Therefore, further collaboration with the medical workforce is required to identify barriers to the detection of cleft palate within 24 hours of birth. This will include further work with the Royal College of Paediatrics and Child Health to further develop the learning modules and training opportunities related to timely cleft palate detection.	

4.2.1. Diagnosis times for children with a cleft palate over the last 10 years

Our 2012 Annual Report highlighted the issue of late diagnosis among children with Cleft Palate (CP), reporting that over one quarter of children were diagnosed late according to the National Standard[22]. Because of this, we continue to investigate factors associated with a late CP diagnosis.

For the purpose of examining trends in diagnosis timing of CP, Figure 4.1 shows the proportion of CP cases who were diagnosed within 24 hours of birth (including antenatal diagnoses) and the proportion diagnosed beyond 24 hours of birth, according to years of birth since 2009. The line graph shows there have been small improvements in the diagnosis timing of CP over the last 10 years. The greatest improvements appear to have taken place after the publication of the Royal College of Paediatrics and Child Health guidance '*Palate examination: identification of cleft palate in the newborn'*[23] in October 2014. There was a 9% improvement in 24 hour diagnosis time between 2014 (67%) and the most recent year of CRANE data collection (76% in 2018).

Figure 4.1. % of CRANE-registered children with a cleft palate, born between 2009 and 2018, who were diagnosed within 24 hours of birth or diagnosed after 24 hours of birth, according to the year of birth.



RCPCH publication of 'Palate examination: Identification of cleft palate in the newborn'

4.2.2. Diagnosis times for children with a cleft palate by region, over the last 5 years

In order to have sufficient numbers to explore regional differences in CP 24-hour diagnosis rates, we have examined diagnosis time among CP patients born over the last five years, for all 2014-2018 births. Analyses of these most recent 5 years of data found a statistically significant difference between the five birth years ($p<0.05$), confirming that diagnosis times have improved in recent years.

Table 4.1 shows that, overall, 1.7% of CPs were diagnosed during antenatal screening and 70.3% were diagnosed at birth, leaving 28% who were diagnosed late according to the National Standard. Table 4.1 also shows that the proportion of CPs diagnosed before/at birth (72%) varied by region / unit, ranging from 63.5% (South Thames, 2.2% before and 61.3% at birth) to 82.6% (West Midlands, 0.8% before and 81.8% at

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

[23] Published October 2014 https://www.rcpch.ac.uk/resources/palate-examination-identification-cleft-palate-newborn-best-practice-guide

birth). This wide and significant variation (p<0.001) suggests that practice varies considerably between maternity units, with some better that others at identifying a cleft of the palate during the new-born examination.

Table 4.1. Number (%) of CRANE-registered children born between 2014 and 2018 with a cleft palate, according to the timing of diagnosis and region / unit.

Regional Cleft Centre / MCN	Administrative Unit	Time of diagnosis in relation to birth n (%)						
		Antenatal	At birth	≤1 week[a]	≤1 month	≤6 months	>6 mths	All
Northern &	Newcastle	1 (0.9)	73 (66.4)	11 (4.5)	8 (7.3)	15 (13.6)	2 (1.8)	110
Yorkshire	Leeds	1 (0.8)	99 (74.4)	14 (5.8)	8 (6)	6 (4.5)	5 (3.8)	133
North West &	Liverpool	1 (1)	72 (69.9)	19 (7.8)	5 (4.9)	5 (4.9)	1 (1)	103
North Wales	Manchester	1 (0.7)	105 (73.9)	21 (8.6)	7 (4.9)	4 (2.8)	4 (2.8)	142
Trent	Nottingham	0 (0)	130 (65.7)	37 (18.7)	9 (4.5)	17 (8.6)	5 (2.5)	198
West Midls.	Birmingham	2 (0.8)	198 (81.8)	26 (10.7)	9 (3.7)	6 (2.5)	1 (0.4)	242
East	Cambridge	0 (0)	105 (74.5)	21 (14.9)	9 (6.4)	6 (4.3)	0 (0)	141
North Thames	GOSH/Chelms	10 (4)	161 (64.1)	48 (19.1)	13 (5.2)	15 (6)	4 (1.6)	251
The Spires	Oxford/Salisbury	3 (1.7)	129 (75)	23 (13.4)	8 (4.7)	9 (5.2)	0 (0)	172
South Wales &	Swansea	1 (1.4)	56 (76.7)	9 (4.7)	4 (5.5)	2 (2.7)	1 (1.4)	73
South West	Bristol	8 (6.7)	83 (69.7)	15 (7.8)	7 (5.9)	6 (5)	0 (0)	119
South Thames	GSTT	6 (2.2)	171 (61.3)	70 (25.1)	14 (5)	12 (4.3)	6 (2.2)	279
N. Ireland	Belfast	0 (0)	53 (68.8)	8 (10.4)	4 (5.2)	3 (3.9)	9 (11.7)	77
All	**All**	**34 (1.7)**	**1,435 (70.3)**	**322 (15.8)**	**105 (5.1)**	**106 (5.2)**	**38 (1.9)**	**2,040**

Note: MCN - Managed Clinical Network.

[a] Recording of 'timing of diagnosis' within 72 hours commenced in May 2014 to align CRANE data collection with NIPE standards[24]. With only small numbers having been recorded using this timing, we report '≤72 hours' cases within the '≤1 week' timing (until recording of this timing is well established).

Overall, the most recent CRANE data show an encouraging trend with regards to timely detection of cleft palates. Nevertheless, 12.2% of children with a CP were not diagnosed until they were more than one week old, which is concerning given that the National Standard states that clefts should be diagnosed within 24 hours of birth to enable immediate referral to a specialist hospital. This helps to ensure the baby, and their family, receive appropriate care and support as soon as possible.

4.2.3. Relationship between type of cleft palate and diagnosis times

For third consecutive year, we conducted an exploration of the impact of different types of cleft palate based on the presentation (as recorded when reporting LAHSAL codes, as described in Chapter 2), on diagnosis times among children with cleft palate alone, born in the last five years between 1 January 2014 and 31 December 2018.

Table 4.2 shows that the completeness of the hard and soft palate impact on the timing of the CP diagnosis. Specifically:

- Children with CP who had complete hard palate involvement were significantly more likely to be identified at birth than those with incomplete hard palate involvement (p<0.001). This delay is addressed within the next week, by which point 92.9% of CP cases have had this identified.

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

- CP cases with any type of hard palate involvement were significantly more likely to be identified at birth (by approximately 19%) than CP cases where there was no hard palate involvement (p<0.001). Where there was no hard palate involvement, 23.2% of CP cases remain undiagnosed until after a week had elapsed since birth.
- CP cases with complete soft palates were significantly more likely to be identified at birth (by approximately 25%) than incomplete soft palates (p<0.001). With an incomplete soft palate, 29.8% of these CP cases remain undiagnosed until after a week had elapsed since birth.

Table 4.2. Number (%) of CRANE-registered children born between 2014 and 2018 with a cleft palate, according to complete / incomplete hard and soft palates.

Palate type	Status	Time of diagnosis in relation to birth n (%)						
		Antenatal	At birth	≤1 week	≤1 month	≤6 months	>6 mths	All*
Hard palate	Incomplete (h)	14 (2)	495 (71.7)	119 (17.2)	30 (4.3)	26 (3.8)	6 (0.9)	690
	Complete (H)	15 (2.3)	533 (82.1)	67 (10.3)	21 (3.2)	12 (1.8)	1 (0.2)	649
All	All	29 (2.2)	1,028 (76.8)	186 (13.9)	51 (3.8)	38 (2.8)	7 (0.5)	1,339
Hard palate	No ‘h’ or ‘H’	5 (0.7)	407 (58.1)	136 (19.4)	54 (7.7)	68 (9.7)	31 (4.4)	701
	Either ‘h’ or ‘H’ present	29 (2.2)	1,028 (76.8)	186 (13.9)	51 (3.8)	38 (2.8)	7 (0.5)	1,339
All	All	34 (1.7)	1,435 (70.3)	322 (15.8)	105 (5.1)	106 (5.2)	38 (1.9)	2,040
Soft Palate	Incomplete (s)	7 (2.4)	143 (49)	55 (18.8)	23 (7.9)	43 (14.7)	21 (7.2)	292
	Complete (S)	24 (1.4)	1,277 (74.1)	264 (15.3)	80 (4.6)	62 (3.6)	17 (1)	1,724
All	All	31 (1.5)	1,420 (70.4)	319 (15.8)	103 (5.1)	105 (5.2)	38 (1.9)	2,016

*Totals for the sections of this table were based on where the hard and soft palate information had been recorded as part of the LAHSAL code(s). Missing data have resulted in the variation in denominator.

5. Weight and height at 5 years of age

Cohort summary	Data source	CRANE database (consented cases only) [25]
	Birth years	Ten years: 2004 to 2013.
	Size (N)	• 3,666 children with recorded weight at 5 years old. • 3,619 children with recorded height at 5 years old.
	Data completeness	45.8% of eligible children (n=9,357) had recorded 'child growth at 5 years' (weight, height, both) or a valid reason it was not collected[26].
	Countries	England, Wales and Northern Ireland.
Benchmarks	• For 5 year olds in the UK, healthy weights range from 14 to 22kg, and healthy heights range from 101cm to 115cm (3rd to 90th centiles for all ranges)[27]. • 100% of eligible children should have a recorded weight and height at the age of 5 years (or a valid reason it was not collected).	
What did we find?	• The average weight of these children was 19.6kg, and their average height was 111.8cm. • These figures should be interpreted with caution due to issues of data completeness: 54.2% of eligible children were missing a recorded weight and height at the age of 5 years (or a valid reason it was not collected). Some improvement has been observed in data completeness in the last 10 years. CRANE will continue to monitor this to see if this an ongoing trend.	
Recommendations	Units should aim to assess children's weight and height around the age of 5 years and improve the reporting of these measures in the audit database.	

Of the 9,357 children whose families had consented to their children's data being collected by CRANE in 2004 to 2013, 3,666 (39.2%) had a weight and 3,619 (38.7%) had a height reported to CRANE.

Analyses revealed that the mean weight for these children was 19.6kg (95% CI 19.5 to 19.7kg) and their mean height was 111.8cm (95% CI 111.6 to 112cm). These figures are consistent with UK national averages for 5 year olds in the general population. This is a positive finding that matches the typical birth weights we observed for children with a cleft (Section 3.4). This is encouraging considering the potential problems with feeding that some children with a cleft might experience in early life.

Nevertheless, it should be noted that the weight and height recorded in CRANE may not be representative of all children with a cleft lip and/or palate as approximately 54% of eligible children were missing growth information. When we examined the completeness of weight and height records, over the specified ten years, we found: **(a)** A statistically significant difference between birth years ($p<0.001$), indicating completeness of these data have improved in recent years, and that **(b)** 2013 was the best birth year for completeness of these data, with 48.6% of expected weight data and 47.7% of expected height data recorded vs. 16.3% and 15.9%, respectively, for births in 2004.

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[25] **Exclusions** (not mutually exclusive): No consent, children who died before the age of 5 years, and with submucous cleft palates.

[26] Note: Only 3,451 children had both a recorded gestational age & birth weight (36.9%), and 635 children had a recorded 'reason not collected' (6.8%).

[27] According to the UK-WHO growth charts - 2-18 years (checked October 2019). Available from: https://www.rcpch.ac.uk/resources/uk-who-growth-charts-2-18-years // See also English National Child Measurement Programme's healthy weight calculator. Published 31 August 2017 (checked October 2019). Available from: https://www.nhs.uk/live-well/healthy-weight/national-child-measurement-programme/

6. Dental health at 5 and 10 years of age

6.1. Decayed, missing or filled teeth (dmft) at 5 years of age

Cohort summary	Data source	CRANE database and Hospital Episode Statistics (HES) (consented cases only)
	Birth years	Nine years: 2004 to 2012
	Denominator	• 4,884 children with recorded dmft scores. • 4,709 children with scores for the calculation of treatment index[28]. • 4,720 children with scores for the calculation of care index[29].
	Numerator	• 2,000 children with at least one (>0) dmft. • Number of children diagnosed with each cleft type. • Number of children within each deprivation quintile.
	Data completeness	66% of eligible children[30] (n=8,440) had recorded dmft or a valid reason it was not collected.
	Countries	England*, Wales and Northern Ireland (*deprivation data were only available for children in England at the time of producing this report).
Benchmark	• 31% of children in the general population of England, Wales and Northern Ireland had at least one (>0) dmft[31] • The average treatment index reported for children in the general population in England is 25%[32]. • The average care index reported for children in the general population in England is 11.8%[33]. • 100% of eligible children should have recorded dmft outcome data at the age of 5 years (or a valid reason it was not collected).	
What did we find?	• 41% of children with a cleft had at least one (>0) dmft. • The average treatment index was 75% for children with a cleft assessed at 5 years of age. • The average care index was 68% for children with a cleft assessed at 5 years of age. • 34% of eligible consented children (n=8,440) were missing recorded dmft outcome data.	
Recommendations	Units should aim to see all children with a cleft for a dmft assessment at the age of 5 years, and this information should be recorded in the audit database.	

The dmft describes the dental caries an individual has experienced and is a measure of oral health. A dmft score reflects the total number of teeth that are decayed, missing or filled. The risk of dental caries is

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[28] The calculation of treatment index requires data on missing teeth (m), filled teeth (f), and dmft scores; or a dmft score of 0 (equating to a treatment index = 1).

[29] The calculation of care index requires data on filled teeth (f) and dmft scores; or a dmft score of 0 (equating to a care index = 1).

[30] Registered in the CRANE Database by the 10 July 2019. **Exclusions** (not mutually exclusive): No consent, children who died before the age of 5 years, and with submucous cleft palates.

[31] Child Dental Health Survey 2013, England, Wales and Northern Ireland - https://digital.nhs.uk/data-and-information/publications/statistical/children-s-dental-health-survey/child-dental-health-survey-2013-england-wales-and-northern-ireland

[32] National Dental Epidemiology Programme for England: oral health survey of five-year-old children 2017 (results) - https://assets.publishing.service.gov.uk/government/uploads/system/uploads/attachment_data/file/708159/NDEP_for_England_OH_Survey_5yr_2017_Results_FINAL_for_website.xlsx . Data on children in the general population in Wales and Northern Ireland were not available at the time of producing this report.

[33] National Dental Epidemiology Programme for England: oral health survey of five-year-old children 2017 (report) - https://assets.publishing.service.gov.uk/government/uploads/system/uploads/attachment_data/file/768368/NDEP_for_England_OH_Survey_5yr_2017_Report.pdf . Data on children in the general population in Wales and Northern Ireland were not available at the time of producing this report.

thought to be higher among children with a cleft lip and/or palate compared with children without an oral cleft[34]. We collect dmft data on CRANE-registered consented children at 5 years of age. Analyses revealed that:

- Of the 8,440 children whose families had consented to their children's data being collected by CRANE in 2004 to 2012, 5,605 (66%) had dmft outcome data recorded in CRANE[35], either in the form of dmft scores (n=4,884, 57.9%) or a valid reason why the outcome was not collected (n=721, 8.5%).
- Among children with reported dmft scores[36], 2,000 (41%) of children with a cleft had at least one (>0) dmft. The mean number of dmft at 5 years among children registered in CRANE was 2.1, with scores ranging from 0 to 20.
- There were 691 children (14.2%) who had a dmft score greater than 5; classified as having either extensive or severe caries or both conditions. This figure is higher than that reported in the general population (13%)[37].

6.1.1. Dental Treatment and Care Indices at 5 years

In this section, we describe our exploration of the impact of cleft type and deprivation on dental treatment and care (reflected by the treatment and care indices, respectively).

Of the 8,440 children whose families had consented to their children's data being collected by CRANE in 2004 to 2012:

- 4,709 (55.8%) had the information required to calculate treatment indices recorded in CRANE (on missing teeth (m), filled teeth (f), and dmft).
- 4,720 (55.9%) had the information required to calculate care indices recorded in CRANE (on filled teeth (f) and dmft).

HES data linked to the CRANE Database at the individual level for consented children born between 1 January 2004 and 31 December 2012 were used to explore the impact of deprivation on dental treatment and care indices; for children in England only. Deprivation data for children in Wales and Northern Ireland were not available at the time of producing this report.

Table 6.1 shows the average treatment index and care index for children according to cleft type and deprivation quintile[38].

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

[35] As per **exclusions footnote above**.

[36] Submucous cleft palate patients excluded from all 5 year outcomes as all/most teams do not audit these patients.

[37] Child Dental Health Survey 2013, England, Wales and Northern Ireland - https://digital.nhs.uk/data-and-information/publications/statistical/children-s-dental-health-survey/child-dental-health-survey-2013-england-wales-and-northern-ireland

[38] Each quintile represents 20% of the population being described. The first quintile represents the lowest fifth of the data (1% to 20% - and in this case the most deprived); the second quintile represents the second fifth (21% to 40%) and so forth. The fifth quintile represents the highest fifth of the data (81% to 100% - the least deprived).

Table 6.1. CRANE-registered consented children born between 2004 and 2012 with a cleft lip and/or palate, according to cleft type and deprivation, and their average treatment index and care index at age 5 years.

		Treatment Index[a]		**Care Index**[b]	
		Average (%)	**All (N)**	Average (%)	**All**[b] **(N)**
Cleft Type	CL	(78.3)	997	(74.8)	1,000
	CP	(75.7)	1,906	(68.3)	1,908
	UCLP	(73.5)	1,252	(66)	1,257
	BCLP	(74.4)	518	(60.5)	519
	Not specified	(72.9)	36	(67.4)	36
	All	**(75.5)**	**4,709**	**(68.2)**	**4,720**
Deprivation[c]	Q1 – Most deprived	(62.6)	874	(51.5)	876
	Q2	(72.1)	800	(64.6)	803
	Q3	(77.3)	731	(69.4)	733
	Q4	(80.4)	720	(75.7)	721
	Q5 – Least deprived	(85.6)	746	(82.7)	746
	All	**(75.1)**	**3,871**	**(68.1)**	**3,879**

[a and b] Exclusions from Treatment and Care Index (not mutually exclusive): No consent, children who died before the age of five, children with submucous clefts, and cases without a dmft score of 0[39] or all relevant dmft data items (to allow calculation of treatment and care index scores). [c] For children in England only, as deprivation data for children in Wales and Northern Ireland were not available at the time of producing this report.

Both treatment and care indices are calculated from the dmft[40], as raw dmft scores give a figure for dental disease experienced but do not distinguish between active and inactive disease at the time of exam (treatment index) or the proportion of children who have received care in the form of fillings (care index).

Furthermore, Table 6.1 shows the average treatment index for children with a cleft is 75.5% (75.1% for children in England only) and the average care index for children with a cleft is 68.2% (68.1% for children in England only); as assessed between 2009-2017 (considering these are 2004-2012 births). These figures are higher than the equivalent ones reported for children in the general population (of 25% and 11.8% for treatment and care indices, respectively, in England)[41].

Treatment index at 5 years

The treatment index reflects whether the mouth is dentally fit at that moment in time. i.e. if dental disease has occurred, the treatment index indicates the extent to which it has been dealt with and the degree to which the child has been rendered free from active decay. When calculated, treatment indices range from 0 to 1 and are usually expressed as a percentage[42]. Treatment indices with a value of 1 (100%) indicate that there is no untreated disease, which is the desired outcome. Furthermore, average treatment indices of 100% can be indicators of having mechanisms in place to deal with any disease occurring, and thereby provides the child with a dentition where the disease is controlled and the child has a pain free mouth.

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

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

[41] Data on children in the general population in peers in Wales and Northern Ireland were not available at the time of producing this report. For England report visit https://digital.nhs.uk/data-and-information/publications/statistical/children-s-dental-health-survey/child-dental-health-survey-2013-england-wales-and-northern-ireland

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

Treatment index scores by cleft type: As shown in Table 6.1, for the 4,709 children with dmft scores of 0 or scores for all three 'm', 'f' and 'dmft' data items – to allow calculation of the treatment index – there was significant variation in treatment index scores by cleft type ($p<0.01$). Children with UCLP and unspecified cleft types had the lowest proportion of treated dental disease (73.5% and 72.9 respectively; at least 2% less than the average for all cleft types), while children with CL had highest proportion of treated dental disease (78.3%, almost 3% more than the overall proportion for all cleft types).

Treatment index scores by deprivation: As shown in Table 6.1, for the 3,871 children with calculated treatment index and deprivation scores (in England only), there was significant variation in treatment index scores by deprivation ($p<0.001$). Children in the most deprived quintile had the lowest average proportion of treated dental disease (62.6%; at least 12% less than the average for all deprivation quintiles), while those from the least deprived quintile had highest average proportion of treated dental disease (85.6%, 10% more than the average for all deprivation quintiles). For the general population (including children with a cleft), children from the most deprived quintile have a higher caries rate and are least likely to be regularly accessing dental care.

Care index at 5 years

The care index reflects cases where children have experienced dental decay, which has been identified at the earliest possible stage (which is preferable), and have been provided with care in the least invasive form possible, i.e. fillings. When calculated, care indices also range from 0 to 1 and are usually expressed as a percentage[43]. Care indices with a value close to 1 (100%) indicate that there are high levels of care provided by fillings (not extraction or no treatment), which is the desired outcome. Conversely, in situations where levels of care are low (and decay could be addressed by filling but has not) the care index is close to 0%. Furthermore, average care indices of 100% can be indicators of having mechanisms for early identification of dental decay, and treatment with fillings rather than tooth removal or untreated dental decay.

Care index scores by cleft type: As shown in Table 6.1, for the 4,720 children with dmft scores of 0 or scores for both 'f' and 'dmft' data items, to allow calculation of the care index, there was significant variation in care index scores by cleft type ($p<0.001$). Children with BCLP had the lowest average care index (60.5%, approximately 8% less than the average for all cleft types), which means decay that might be treated by fillings has remained untreated or decay was so severe that extraction was the treatment of choice. Children with BCLP and UCLP may have a greater proportion of teeth with deficient enamel and dentine formation, and therefore the treatment option of choice is often extraction not restoration, due to the anatomy of the teeth. Meanwhile children with CL had the highest average proportion of children receiving care by fillings (74.8%, approximately 6% more than the average for all cleft types).

Care index scores by deprivation: As shown in Table 6.1, for the 3,879 children with calculated care index and deprivation scores (in England only), there was significant variation in care index scores by deprivation ($p<0.001$). Children in the most deprived quintile had the lowest average care index (51.5%, almost 17% less than the average for all deprivation quintiles), which means decay that might be treated by fillings has remained untreated or decay was so severe extraction was the treatment of choice. Meanwhile, children in

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

the least deprived quintile had the highest average proportion of children receiving care by fillings (82.7%, approximately 14% more than the average for all deprivation quintiles). As for the treatment index, all children (the whole population including cleft children) from the most deprived quintile have a higher caries rate and are least likely to be regularly accessing dental care.

It is worth considering that cleft type and deprivation 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 ethnicity, cultural differences in attitudes to dental health, water fluoridation levels, and mineralisation anomalies of the teeth that are more prevalent in children with clefts.

6.2. Decayed, missing or filled teeth (DMFT) at 10 years of age

Cohort summary	Data source	CRANE database (consented cases only)
	Birth years	Two years: 2007 and 2008
	Denominator	226 children with recorded DMFT scores.
	Numerator	52 children with at least one (>0) DMFT.
	Data completeness	14% of eligible[44] children (n=1,973) had recorded DMFT or a valid reason it was not collected.
	Countries	England, Wales and Northern Ireland.
Benchmark	• 100% of eligible children should have a recorded DMFT score at the age of 10 years (or a valid reason it was not collected).	
What did we find?	• 23% of 10 year old children with a cleft and DMFT score had at least one (>0) DMFT. • 86% of eligible consented children (n=1,979) were missing DMFT outcome data.	
Recommendations	Units should aim to see all children with a cleft for a DMFT assessment at the age of 10 years, and this information should be recorded in the audit database.	

Like the dmft (used to assess 5 year olds), the DMFT describes the dental caries an individual has experienced and is a measure of oral health. A DMFT score reflects the total number of adult 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 with children without an oral cleft[45]. We collect DMFT data on CRANE-registered consented children at 10 years of age.

This is the first year that DMFT, as an outcome for 10 year olds, has been reported in the CRANE Database Annual Report.

Of the 1,973 children whose families had consented to their children's data being collected by CRANE in 2007 and 2008, 207 (14%) had DMFT outcome data recorded in CRANE[46], either in the form of DMFT scores

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[44] Registered in the CRANE Database by the 10 July 2019. **Exclusions** (not mutually exclusive): No consent, children who died before the age of 10 years (n=3), and with submucous cleft palates.

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

[46] As per **exclusions footnote above**.

or a valid reason why the outcome was not collected. Data completeness varied by region / unit and ranged from 0% to 48%.

Among children with a reported DMFT score (n=226), 23% of 10 year old children with a cleft had at least one (>0) decayed, missing or filled tooth (DMFT). The mean number of DMFT at 10 years among children registered in CRANE was 0.7, with scores ranging from 0 to 15. Five children (2%) had a DMFT score greater than 5.

It is anticipated that we will be able to report on treatment and care index, at 10 years of age, for 2007 to 2009 births in the CRANE 2020 Annual Report and/or when recording of these data in CRANE improves.

The data reported for dmft /DMFT, Treatment and care indices shows that although dmft remains higher than the general population, the levels of Treatment index at more than three times that of the general population show that dental decay is being treated, with most children having no teeth with active decay in their mouth at the time of assessment d=0. Data collection for 10 year olds is still at a low level, policies for seeing children at this age varies across units and further discussion is taking place on how best to improve this.

7. Facial growth at 5 years of age

7.1. Five Year Old Index for children with a unilateral cleft lip and palate (UCLP)

Cohort summary	Data source	CRANE database (consented cases only)
	Birth years	Nine years: 2004 to 2012
	Denominator	954 children with complete UCLP and recorded Five Year Old Index scores.
	Numerator	Number of children with each of the five possible Five Year Old Index scores.
	Data completeness	74% of eligible[47] consented children (n=1,438) had recorded facial growth or a valid reason it was not collected.
	Countries	England, Wales and Northern Ireland.
Benchmark	• CSAG finding that 36% of children with a complete UCLP had poor dental arch relationships at 5 years old in 1998[48] • 100% of eligible children with a complete UCLP should have a recorded Five Year Old Index score (or a valid reason it was not collected).	
What did we find?	• 25.6% of children had scores reflecting poor dental arch relationships at 5 years old. • 26% of eligible consented children (n=1,438) were missing recorded Five Year Old Index outcome data.	
Recommendations	Cleft care teams should aim to take dental impressions of all children with a complete UCLP around the age of 5 years, to allow for an assessment using the Five Year Old Index.	

Dental models of 5-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 the use of any other interventions, such as orthodontics or alveolar bone grafting, which may influence this growth further[49]. Dental arch relationships at 5 years may predict treatment outcome in terms of facial growth on a population basis rather than at the individual child level[50]. The Five Year Old Index may, therefore, also be used to compare treatment outcomes between centres and surgeons. Patients scoring ‘1’ and ‘2’ on the index are considered to have the best possible outcomes, while those scoring ‘4’ and ‘5’ are thought to have poor outcomes in terms of facial growth, and they may benefit from further surgery to correct their facial disproportion once facial growth is complete.

Of the 1,438 children whose families had consented to their children’s data being collected by CRANE in 2004 to 2012, 1,068 (74%) had facial growth outcome data recorded in CRANE[51], either in the form of Five Year Old Index scores (n=954, 66.3%) or a valid reason why the outcome was not collected (n=114, 7.9%).

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[47] Registered in the CRANE Database by the 10 July 2019. **Exclusions** (not mutually exclusive): No consent, incomplete UCLP, children who died before the age of 5 years, and with submucous cleft palates.

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

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

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

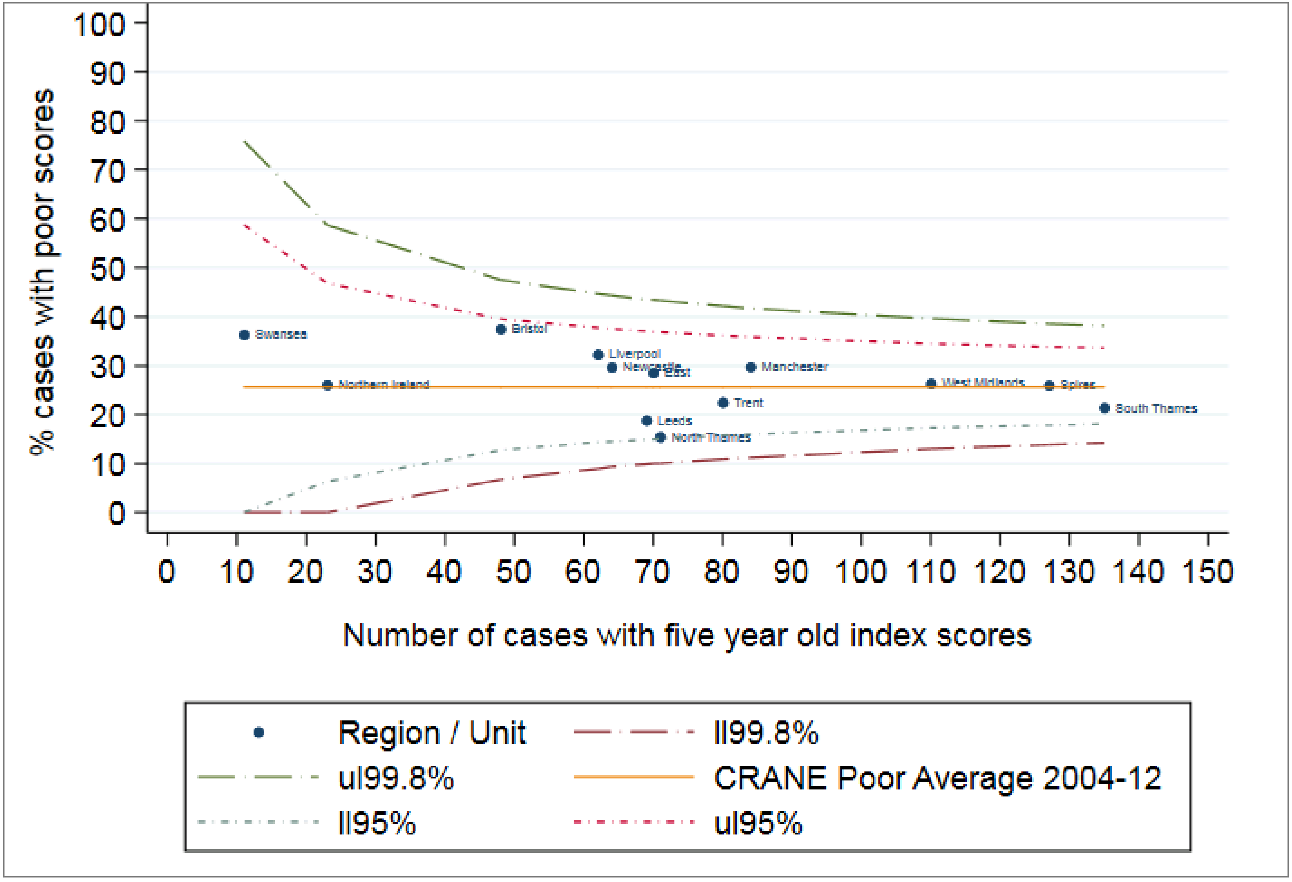
[51] As per **exclusions footnote above**.

Among the 954 children with reported Five Year Old Index scores [52], 41.9% had Five Year Old Index scores in the two groups considered to have the best possible dental arch relationships (scores '1' or '2') while 25.6% of children had scores '4' or '5', reflecting poor dental arch relationships. This represents an improvement on the CSAG finding that 36% (of 223 children with a complete UCLP) had poor dental arch relationships at 5 years old in 1998[53] (see Appendix 5 for information on children born between 2004 and 2012 with a complete unilateral cleft lip and palate, according to Five Year Old Index scores and region / unit).

The majority of Five Year Old Index scores provided by all regions / units were externally validated (in 848/954 (88.9%) of eligible cases), and where externally validated scores were unavailable, internal scores were included in the analysis.

The funnel plot[54] in Figure 7.1 shows the proportion of CRANE 5-year olds with the poorest (4 and 5) Five Year Old Index outcome scores according to the number of children at each region / unit with index scores. It is centred on 25.7%[55], which is the national proportion of poor Five Year Old Index scores for CRANE 5-year olds across all units.

Figure 7.1. Funnel plot of 5-year olds (born between 2004 and 2012) with poor Five Year Old Index scores according to the number of children at each region / unit with index scores.



Note: Funnel plot centred on national proportion (for 2004-2012 births reported in CRANE) of poor Five Year Old Index scores across all units of 25.7%.

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[52] Submucous cleft palate patients excluded from all 5 year outcomes as all/most teams do not audit these patients.

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

[54] This funnel plot is calculated using valid data as denominators (not considering missing data), subject to the same inclusions and exclusions as data in Appendix 5. In addition, it is not adjusted (or risk adjusted) in any way.

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

The funnel plot in Figure 7.1 also shows that all regions' / units' rates of poor index scores fall within the expected range given the number of children (born between 2004-2012) with valid index scores at their unit. i.e. No unit has a 'poor index score rate' below the lower 99.8% control limit or above the upper 99.8% control limit (more information on funnel plots can be found in the Glossary at the front of this report).

The fact that recorded facial growth (or a valid reason it was not collected) were submitted for only 74% of eligible children, and the wide variation in the number of children within each region / unit (ranging from 11 to 135), means that the data presented in this section should be interpreted with caution, as it is possible that the overall findings from the limited data made available to CRANE may not be representative of the complete UCLP population. Analyses of data from a greater number of children are necessary to examine true differences that may exist between cleft care teams.

7.2. Relationship between facial growth and speech at 5 years

Question	To establish if facial growth and speech outcomes are associated.	
Cohort summary	Data source	CRANE database (consented cases only)
	Birth years	Six years: 2007 to 2012
	Denominator	521 5-year-old children with a complete UCLP and recorded Five Year Old Index scores as well as all 16 Cleft Audit Protocol for Speech – Augmented (CAPS-A) parameters reported.
	Numerator	Number of children meeting one of four categories based on: Good/poor facial growth scores and whether or not they had achieved normal speech.
	Data completeness	55% of eligible[56] consented children (n=946) had both facial growth and speech outcome data.
	Countries	England, Wales and Northern Ireland.
Benchmark	100% of consented children with a complete UCLP should have recorded facial growth and speech outcome data.	
What did we find?	• 29.5% of children had achieved scores indicating good facial growth and normal speech. • 20.5% of children had scores indicating poor facial growth and not achieving normal speech. • 45% of eligible children (n=946) were missing recorded facial growth and speech outcome data.	
Recommendations	Units should aim to take dental impressions of all children with a complete UCLP around the age of 5 years, to allow for an assessment using the Five Year Old Index. In addition, all children with a cleft affecting the palate should have their speech assessed at the age of 5 years. These outcome measures should be reported to the audit database.	

We sought to explore whether a relationship exists between facial growth and speech outcomes among children with a complete UCLP. Good outcomes for facial growth and speech have been defined as follows:

- Patients scoring '1' and '2' on the Five Year Old Index are considered to have good facial growth, while those scoring '4' and '5' are thought to have poor facial growth (detailed in the previous section on Five Year Old Index scores).

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[56] Registered in the CRANE Database by the 10 July 2019. **Exclusions** (not mutually exclusive): No consent, incomplete UCLP, children who died before the age of 5 years, with submucous cleft palates, and syndromic children.

- Normal speech is represented by ‘normal’ (green) scores across all 16 Cleft Audit Protocol for Speech – Augmented (CAPS-A) parameters (detailed further in the next section on CAPS-A ratings).

Table 7.1 shows that the proportion of children who have achieved normal speech is marginally higher among children classified as having good facial growth than those with poor facial growth; however, these differences were not statistically significant (p=0.64).

Overall, 29.5% (105/356) of consented children born between 2007 and 2012 had achieved scores indicating good facial growth and normal speech, while 20.5% (73/356) had scores indicating poor facial growth and not achieving normal speech.

Table 7.1. Number (%) of CRANE-registered consented children born between 2007 and 2011[a], with good or poor Five Year Old Index scores at 5 years of age, by those achieving/not achieving normal speech.

	Normal Speech				
Five Year Old Index scores	**Achieved**		**Not Achieved**		**Total**
	N	**(%)**	**N**	**(%)**	**N**
Good scores	105	(47.9)	114	(52.1)	219
Poor scores	64	(46.7)	73	(53.3)	137
Total	**169**	**(47.5)**	**187**	**(52.5)**	**356**

[a] Registered in CRANE by 10 July 2019. Excluding children who died before the age of 5 years, with an incomplete UCLP, children with submucous cleft palates, syndromic children, children missing Five Year Old Index scores data, and children missing one or more of all 16 CAPS-A data items.

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

1. Resonance: Hypernasality
2. Nasal Airflow: Audible Nasal Emission
3. Nasal Airflow: Nasal Turbulence
4. Passive Cleft Speech Characteristics (CSCs): Weak and or nasalised consonants
5. Passive CSCs: Nasal realisation of plosives
6. Passive CSCs: Gliding of fricatives.

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

Our results, based on data provided for eligible children with complete UCLP, suggest that facial growth is not associated with speech scores. The small sample size means these findings should be interpreted with caution. Data should continue to be revisited as the sample increases in size. A larger sample will become available as the collection of the full 16 CAPS-A outcome scores (which started six years ago) expands for births after 2012. In addition, as the data completeness of Five Year Old Index and CAPS-A ratings continues to improve, so will the sample size available for analyses.

8. Speech at 5 years of age

Cohort summary	Data source	CRANE database (consented cases only)
	Birth years	Three years: 2010 to 2012
	Denominator	1346 5-year-old children born with a non-syndromic cleft affecting the palate (CP, UCLP, BCLP) who had all 16 CAPS-A speech parameters reported to CRANE.
	Numerator	The number of children with a particular speech outcome or the number meeting a standard.
	Data completeness	• 69.7% of 1,931 eligible children had all 16 CAPS-A speech parameters reported • 2.1% had some but not all 16 CAPS-A speech parameters reported • 21.9% had a reason the speech outcomes were not collected • 6.4% were missing data or a reason for not collecting data
	Countries	England, Wales and Northern Ireland.
Standard	• The achievement of speech within the normal range (see text below for a full description of these standards[57]). • The absence of speech difficulties resulting from existing or previous structural anomalies. • The absence of significant cleft-related articulation difficulties.	
What did we find?	• There is wide variability between regions / units in the reporting of speech outcomes. • 60.8% achieved speech within the normal range. • 71.5% had no speech difficulties resulting from existing or previous structural anomalies. 17.8% of children had secondary surgery for speech purposes before the age of 5 years. • 68.0% had no cleft-related articulation difficulties. • The proportion of children meeting the standards in the 2010-2012 birth cohort represent improvements compared with the National Standards developed in 2009[58] and updated in 2016 based on the national proportions observed in the 2007-09 birth cohort[59].	
Recommendations	• All children with an isolated cleft affecting the palate should have their speech assessed and reported to CRANE. • Differences in outcomes between Units should be explored further to determine whether there are certain practices, such as timing of repair, that are associated with more favourable speech outcomes.	

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[57] Britton L, Albery L, Bowden M, Harding-Bell A, Phippen G, and Sell D (2016) National (UK) standards for speech for children born with cleft palate (+/-cleft lip /alveolus).

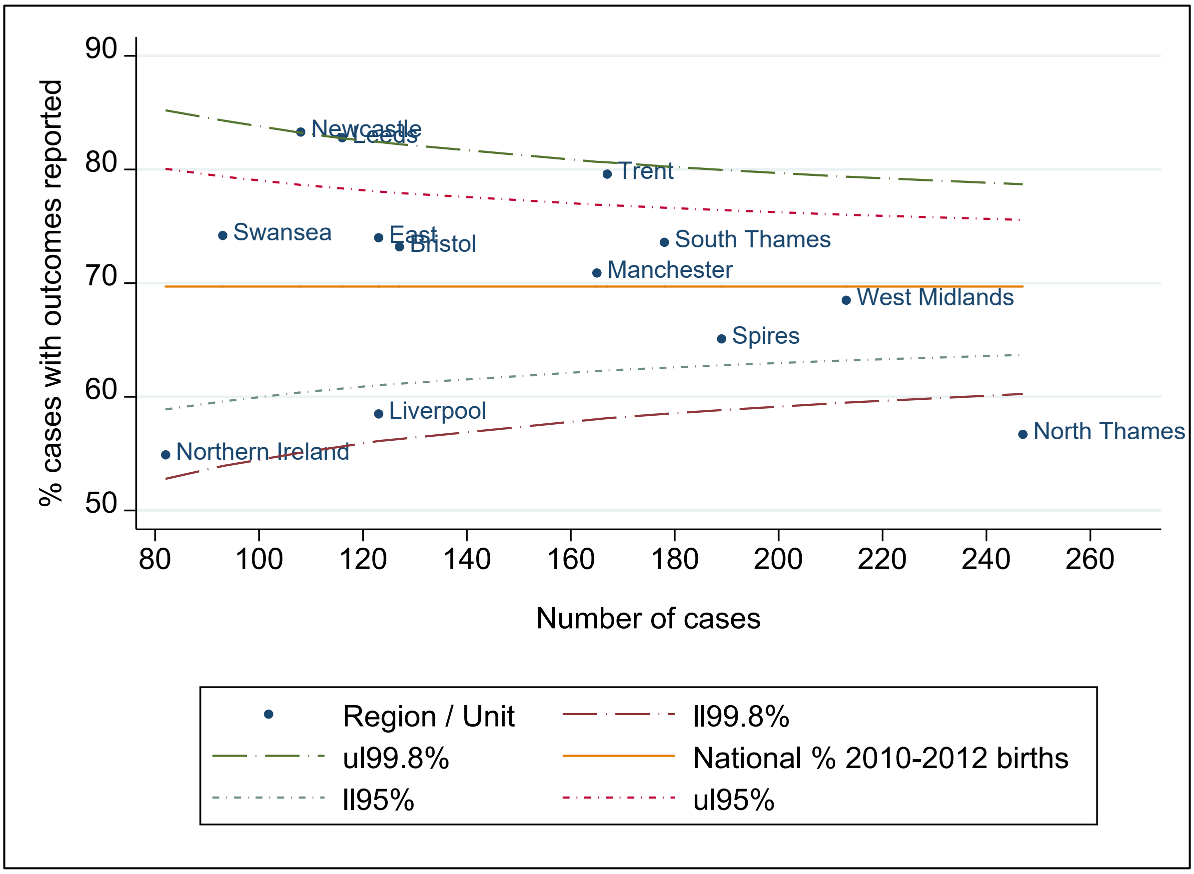
[58] Britton L, Albery L, Bowden M, Harding-Bell A, Phippen G, and Sell D (2014) A Cross-Sectional Cohort Study of Speech in Five-Year-Olds With Cleft Palate ± Lip to Support Development of National Audit Standards: Benchmarking Speech Standards in the United Kingdom. The Cleft Palate-Craniofacial Journal: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

[59] Britton L. (2017) National improvements in speech outcomes 2001-2009, presented at Craniofacial Society of Great Britain and Ireland Conference (Newcastle).

8.1. Data completeness

All regions / units are requested to report to CRANE all 5-year old 16 CAPS-A parameters for each consented child with an isolated cleft affecting the palate. If speech outcomes are not available, regions / units are asked to report a reason for this[2] (e.g. Patient transferred out of area or patient did not attend appointment). Appendix 6 shows the breakdown of speech reporting for each region / unit. Overall, the proportion of eligible cases accounted for ranged from 76.9% in North Thames to 99.5% in The Spires, with 9 out of 13 regions / units accounting for more than 95% of eligible cases. Figure 8.1 shows the variability in the reporting of all 16 CAPS-A parameters according to the number of eligible cases within each region / unit. The funnel plot shows that North Thames is an outlier for the proportion of eligible cases with all 16 CAPS-A parameters reported. Results from this region should therefore be interpreted with caution. Overall, 69.7% (54.9%-83.3% range between Units) of the 1,931 eligible cases had all 16 CAPS-A parameters reported. A further 2.1% of cases had some but not all 16 CAPS-A parameters reported, while 21.9% (13.8%-31.7% range) of cases had a reason why the outcome was not reported (e.g. Patient transferred in or out of area, etc[60]), and 8.4% (14.6%-2.4% range) were missing data or a reason.

Figure 8.1. Funnel plot showing the % of cases (born 2010-2012) with speech outcomes reported, according to the number of eligible children at each region / unit.



Note: Registered in CRANE by 10 July 2019. Exclusions (not mutually exclusive): children who died before the age of 5 years, with submucous cleft palates, born with either a CL or a non-specified cleft type, and syndromic children. Funnel plot centred on the overall national proportion (69.7%) of children (born 2010-2012) with speech outcomes reported. See Appendix 6 for the raw data used to create this funnel plot.

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[60] Patient deceased or emigrated; patient transferred in/out of area; clinically contraindicated; lack of staff/facilities/equipment; patient did not attend/cancelled/did not consent or cooperate; other reason

8.2. Speech outcomes

The Cleft Audit Protocol for Speech – Augmented (CAPS-A) tool has been used to assess speech among non-syndromic children with a cleft affecting the palate (CP, UCLP and BCLP). The 16 CAPS-A speech parameters assessed include:

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

The distribution of scores across the individual 16 CAPS-A speech parameters for those born 2010-2012 are presented in Appendix 7.

Resonance and Nasal Airflow

In terms of resonance, 6.1% of children had moderate or severe hypernasality i.e. nasal sounding speech[61]. This is indicative of velopharyngeal dysfunction (VPD), which is when the palate is unable to close off the nasal airway during speech. In addition, results of the Cleft Speech Characteristics show that 3.6% of children had 'weak and or nasalised consonants' and 1.8% of children had 'nasal realisation of plosives' (passive articulation errors) affecting three or more consonants, which are likely to be the consequence of VPD and is consistent with the hypernasality scorings.

It should be noted that, in order to achieve good speech, 17.8% of the children with reported surgical data[62] have had secondary surgery for speech purposes before the age of 5 years.

Eighty-four percent of children with reported ratings for all four resonance and nasal airflow parameters had ratings indicating that no structural problems existed in relation to these parameters[63].

Cleft Speech Characteristics (CSCs)

Out of the 1,346 children (born 2010-12) with reported ratings for all 12 cleft speech characteristics, 66.3% had ratings indicating they did not exhibit any CSCs[64]. 'Palatalisation / Palatal' anterior oral CSCs were the most commonly occurring CSC, affecting 23% of children (10.8% with ratings of one or two consonants affected (light green ratings) and 12.2% with three or more consonants affected (amber ratings)). These CSCs can vary in severity and may affect speech acceptability more than speech intelligibility. The cleft speech characteristics which are more likely to affect speech intelligibility are the posterior, non-oral and passive CSCs (see Table B in Appendix 6 for rates of these). Therapy would often be indicated for these children, and/or further investigation of structure and possible surgery.

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[61] With a hypernasality score of '3' or '4' (red ratings).
[62] VP surgery/fistula repair data was reported for 98.1% of eligible children.
[63] All green ratings of '0' or '1'.
[64] All green ratings of 'A' and in selected cases of 'B' – as per Appendix 7.

Nationally agreed Speech Outcome Standards

Further to reporting on the 16 CAPS-A speech parameters separately, we report on the proportion of 5-year olds meeting each of the following three nationally agreed Speech Outcome Standards[65]:

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

Cleft type

Table 8.1 shows that the proportion of children achieving each speech standard varies considerably according to the type of cleft. These differences are statistically significantly different (p<0.001). Children with a cleft affecting only the palate had the most favourable results, while those with a BCLP had the least favourable results. The greatest differences between cleft types were observed for Speech standards 1 and 3, whereby the proportion of children with a BCLP meeting the standards was approximately half that observed among children with a CP. Given that outcomes vary according to cleft type, it is important to consider that cleft type distribution may influence the results seen at particular regions/units. For more information on the cleft type distribution for 2010-2012 births at each region/unit, please see Table B in Appendix 8. Funnel plots displaying the proportion of CP, UCLP and BCLP cases are also shown in Appendix 8, showing that all units have rates that are within 3 standard deviations of the national proportion.

Table 8.1. Number (%) of CRANE-registered consented children born 2010 - 2012 with reported speech outcomes at 5 years of age, meeting each speech outcome standard, according to cleft type.

		1. Normal speech		2. No structurally-related speech difficulties		3. No cleft-related articulation difficulties	
Cleft type	N	N	(%)	N	(%)	n	(%)
CP	694	511	(73.6)	525	(75.7)	562	(81.0)
UCLP	456	236	(51.8)	317	(69.5)	275	(60.3)
BCLP	196	71	(36.2)	120	(61.2)	78	(39.8)
P value		*<0.001*		*<0.001*		*<0.001*	

Region and Unit variation

Normal speech

Out of the 1,346 children (born 2010-2012) with reported ratings for all 16 CAPS-A speech parameters, 60.8% of children across all units achieved the National Speech Outcome Standard 1. *They had normal*

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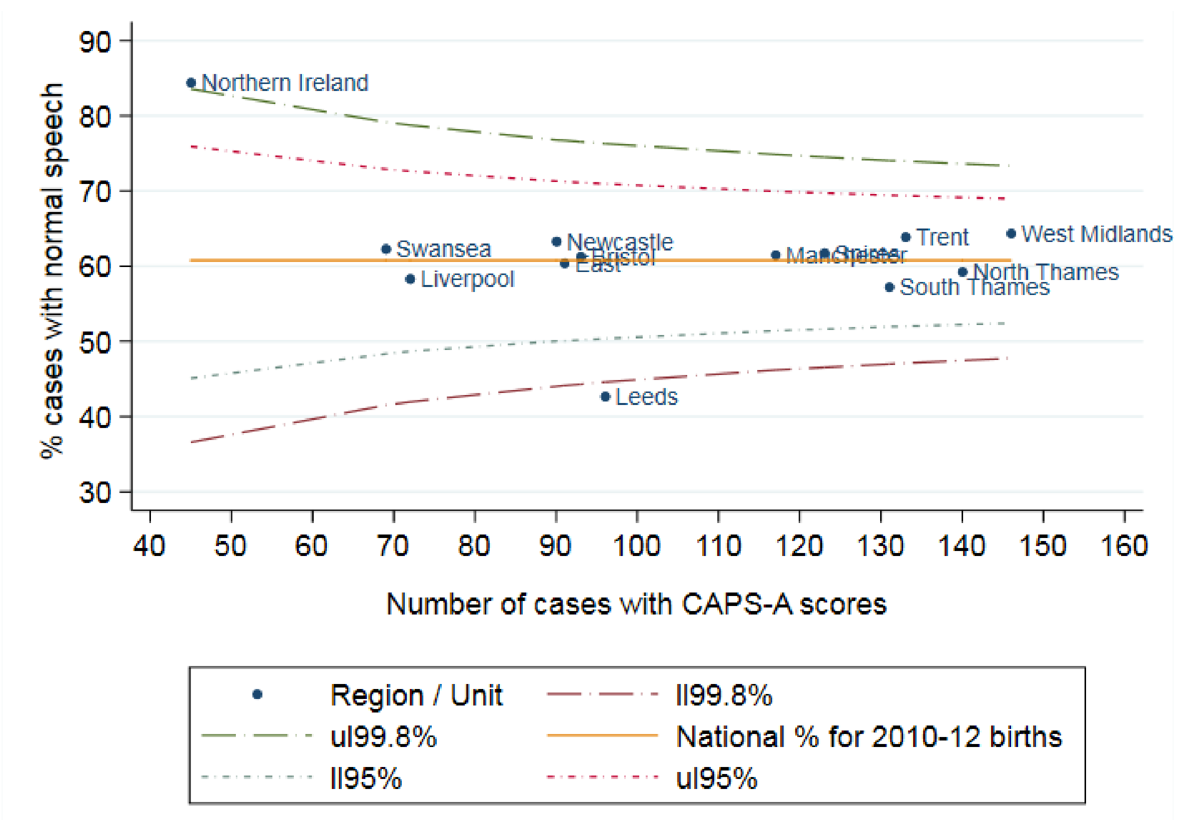
[65] Britton L, Albery L, Bowden M, Harding-Bell A, Phippen G, and Sell D(2014) A Cross-Sectional Cohort Study of Speech in Five-Year-Olds With Cleft Palate ± Lip to Support Development of National Audit Standards: Benchmarking Speech Standards in the United Kingdom. The Cleft Palate-Craniofacial Journal: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

(green) ratings across all 16 CAPS-A speech parameters. This means that the National Speech Outcome Standard 1 benchmark of 60% which was set in 2016[66], was achieved and exceeded by the CRANE cohort born in 2010-2012.

The funnel plot[67] in Figure 8.2 (see Appendix 8 for raw data) shows the proportion of children (born in 2010-2012) achieving *normal speech* according to the number of auditable children at each region / unit with ratings for all 16 CAPS-A speech parameters (more information on funnel plots can be found in the Glossary at the front of this report). It shows that most (11/13) regions / units had a rate of normal speech that was within 4% of the overall national proportion of 60.8%, and that these units fell well within the 95% control limits of the funnel plot. Northern Ireland had a normal speech rate of 84.4% while the corresponding figure in Leeds was 42.7%. The funnel plot shows that these units fall outside of the upper and lower 99.8% control limits, respectively. It should be noted that Northern Ireland had the fewest patients and that only 55% of their eligible cases had outcomes reported (see Figure 8.1). Leeds reported outcomes for 83% of eligible cases, which was one of the highest proportions out of all units. Appendix 8 contains a table and funnel plots showing the distribution of cleft types (CP, UCLP and BCLP) across each unit, which do not appear to explain the difference in rates of speech within the normal range between Leeds and Northern Ireland.

Figure 8.2. Funnel plot of 5-year olds (born 2010-2012) with ratings suggesting speech within the normal range, according to the number of children at each region / unit with CAPS-A ratings.



Note: Funnel plot centred on the overall national proportion (60.8%) of children (born 2010-2012) with speech rated within the normal range. See Appendix 8 for the raw data used to create this funnel plot.

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[66] Britton L. (2017) National improvements in speech outcomes 2001-2009, presented at Craniofacial Society of Great Britain and Ireland Conference (Newcastle)

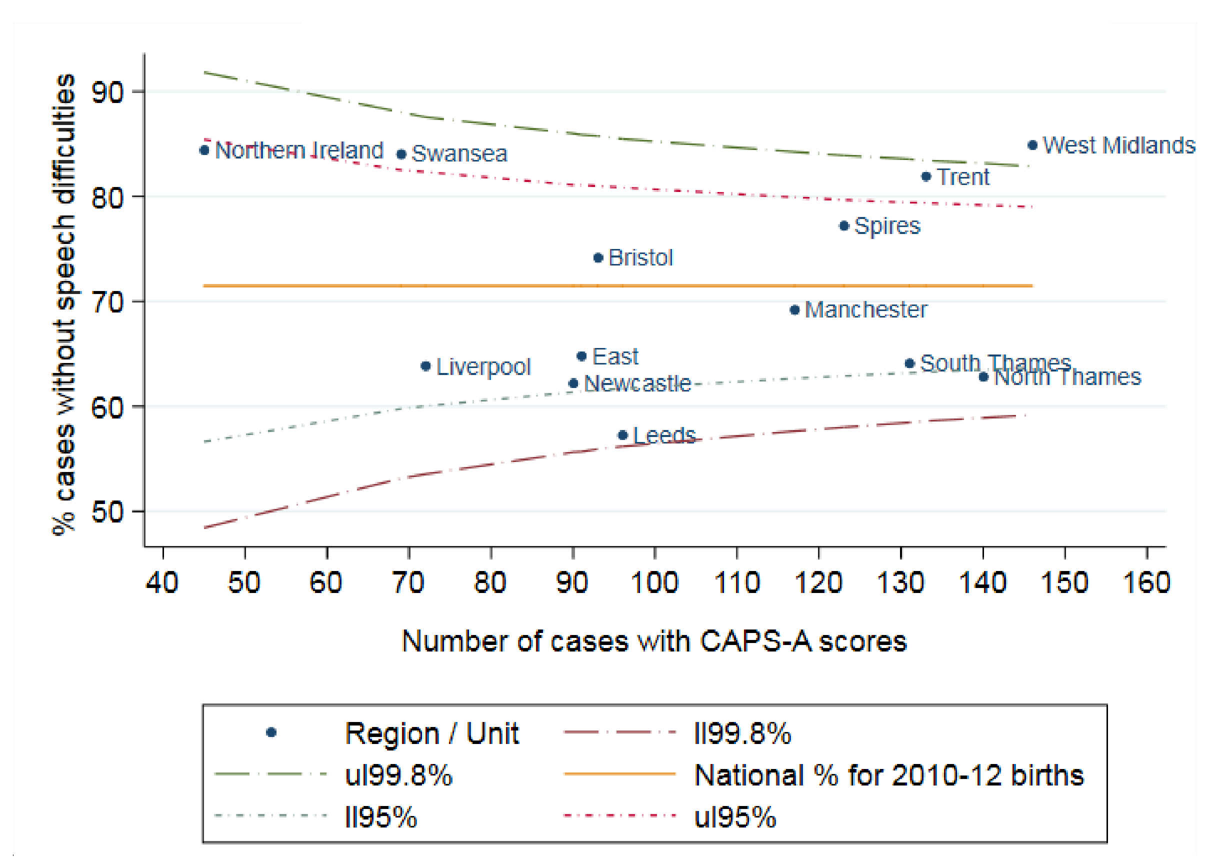
[67] This funnel plot is calculated using valid data as denominators (not considering missing data) and is not adjusted (or risk adjusted) in any way.

Absence of structurally-related speech difficulties

Out of the 1,346 children (born 2010-2012) with reported ratings for all 16 CAPS-A speech parameters, 71.5% of children across all units achieved the National Speech Outcome Standard 2a. *They had no reported history of surgery for speech purposes and normal (green) ratings across the following six CAPS-A speech parameters: Hypernasal resonance, both nasal airflow parameters (audible nasal emission and nasal turbulence), and all three Passive CSCs.* This means the National Speech Outcome Standard 2a benchmark of 70%, which was set in 2016[68], was achieved and exceeded by the CRANE cohort born 2010-2012.

Figure 8.3 (see Appendix 8 for raw data) shows the proportion of 5-year olds with speech ratings that suggest they do not have structurally-related speech difficulties [69], according to the number of children at each region / unit with CAPS-A ratings. It is centred on the overall national proportion of 71.5% for 2010-2012 births, and shows that there is a lot of variability between regions / units in the proportion of children without structurally-related speech difficulties. In particular, the 84.9% in the West Midlands was above the upper 99.8% control limit, and the care and service provision offered by this Unit may be worth investigating for best practice recommendations.

Figure 8.3. Funnel plot of 5-year olds (born 2010-2012) with ratings suggesting no structurally-related speech difficulties, according to the number of children at each Region/Unit with CAPS-A ratings.



Note: Funnel plot centred on the overall national proportion (71.5%) of children (born 2010-2012) without structurally-related speech difficulties. See Appendix 8 for the table of data used to create this funnel plot.

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[68] Britton L (2017) National improvements in speech outcomes 2001-2009, presented at Craniofacial Society of Great Britain and Ireland Conference (Newcastle)

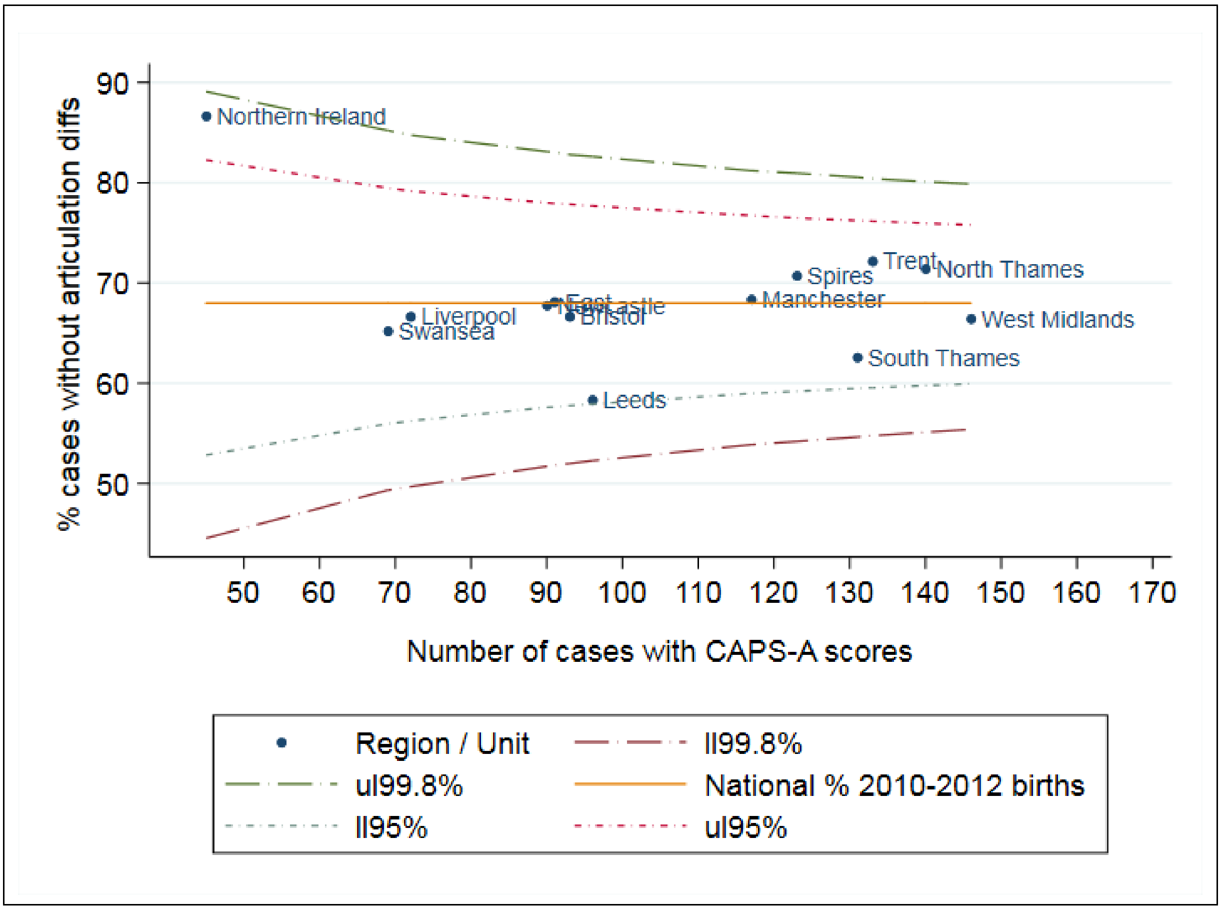
[69] As a result of existing or previous structural anomalies – specifically there is no evidence of a structurally-related problem and they have not had VP surgery or fistula repair for speech.

Absence of cleft-related articulation difficulties

Out of the 1,346 children (born 2010-2012) with reported ratings for all 16 CAPS-A speech parameters, 68.0% of children across all units achieved the National Speech Outcome Standard 3: *They had normal (green) ratings across the following 10 CSCs: All three Anterior Oral CSCs, both Posterior Oral CSCs, all four Non Oral CSCs, and gliding of fricatives (a Passive CSC).* This means the national Speech Outcome Standard 3 benchmark of 68%, which was set in 2016, was achieved by the CRANE cohort born 2010-2012. There has been minimal change, at a national level, over recent years.

Figure 8.4 (see Appendix 8 for raw data) shows the proportion of 5-year olds with speech ratings that suggest they do not have cleft-related articulation difficulties[70], according to the number of children at each region / unit with CAPS-A ratings. It is centred on the overall national proportion (68%) of children, born 2010-2012, who met this standard. All units fall within the 99.8% control limits.

Figure 8.4. Funnel plot of 5-year olds (born 2010-2012) with ratings suggesting no cleft-related articulation difficulties, according to the number of children at each Region/Unit with CAPS-A ratings.



Note: Funnel plot centred on the overall national proportion (68.0%) of children (born 2010-2012) without cleft-related articulation difficulties. See Appendix 8 for the table of data used to create this funnel plot.

Despite the considerations around missing data and the lack of formal adjustment for potential confounding factors, as described above, presenting the data in Figures 8.2, 8.3 and 8.4 as funnel plots centred on overall national proportions is the most straightforward method (at this time[71]) of checking whether or not any units deviate significantly from the expected standards. However, the results should be

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[70] No cleft type speech characteristics requiring SLT and/or surgery.

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

interpreted with caution in those regions / units where there is a high level of missing speech data. Nevertheless, it is recommended that differences in outcomes between Units without a high volume of missing data should be explored further to determine whether there are certain practices, such as timing of repair, that are associated with more favourable speech outcomes.

Box 1. Leeds Cleft Team Response to ‘Speech Outcomes at Five Years’.

We would like to extend to CRANE our gratitude for the opportunity to provide a response to the reported speech outcomes (births 2010-2012) within the CRANE Annual Report 2019.

In reviewing our unit’s speech outcomes for this period, we are systematically examining the data, and practices and processes at our unit, in order to identify factors that may have influenced the outcomes. Possible factors include a high proportion of the more challenging types of cleft, such as bilateral cleft lip and palate and Robin Sequence cleft palate, and associated healing difficulties. We welcome the additional analysis in this year’s report of speech outcomes in children with Robin-associated cleft palate and recommend that cleft type and Robin-association are included as important risk-adjustment factors in future reports.

The report shows there is wide variation in reporting rates by units nationally and low reporting rates at some units. This limits meaningful interpretation of nationally reported speech outcomes and therefore we recommend that reporting rate is included as a risk-adjustment factor in the future. The reporting rate from Leeds is one of the highest. In future reports, it would be useful if CRANE could document a more detailed breakdown of ‘reasons reported for not collecting outcome’ in order for us to better understand this in relation to the nature of exclusions across units.

We support all attempts by CRANE to explore differences in outcomes between units and in their continuing efforts to maximise the quality of data analysis within future reports. At our unit, we will continue with our own investigation in a systematic and transparent manner in order to identify any changes to practice or process that will optimise outcomes for our patients in the future.

Leeds Cleft Lip and Palate Unit, December 2019.

8.3. Pierre Robin Sequence

Cohort summary	Data source	CRANE database
	Birth years	Six years: 2007 to 2012
	Denominator	227 children born with a cleft affecting only the palate (CP) plus a diagnosis of Pierre Robin Sequence (and no additional syndromes) who had all 16 CAPS-A speech parameters reported to CRANE.
	Numerator	The number of children born with isolated CP + Pierre Robin Sequence (PRS) who met each speech standard.
	Data completeness	• 65.6% of 346 eligible children had all 16 CAPS-A speech parameters reported • 2.3% had some but not all 16 CAPS-A speech parameters reported • 26.0% had a reason the speech outcomes were not collected • 6.1% were missing data or a reason for not collecting data
	Countries	England, Wales and Northern Ireland.
Standard	• The achievement of speech within the normal range. • The absence of speech difficulties as a result of existing or previous structural anomalies. • The absence of significant cleft-related articulation difficulties.	
What did we find?	• 60.4% achieved speech within the normal range. • 69.6% had no speech difficulties resulting from existing or previous structural anomalies. • 68.3% had no cleft-related articulation difficulties. • These rates are significantly lower than those observed among children with an isolated cleft palate without Pierre Robin Sequence. The proportions of children with PRS meeting the standards are more comparable with rates observed among children without PRS who have an isolated UCLP.	
Recommendations	• Cleft care teams should continue to assess and report to CRANE the speech outcomes among children with PRS and other syndromes (e.g. Stickler syndrome, Van der Woude syndrome and Di George syndrome), where possible. This will allow us to examine differences in outcomes between clinical subgroups.	

For the first time, CRANE has examined the three national speech standards in children reported to have Pierre Robin Sequence (PRS). We restricted our sample to children with an isolated cleft palate (which excluded two children reported to have other non-syndromic types of cleft) and compared the proportion of children with PRS meeting the standards with the corresponding proportions among those without PRS born across the same time period. Between 2007 and 2012, there were 346 consented children born with a cleft affecting only the palate who were reported to CRANE as having PRS and no further syndromes. Of these, 227 (65.6%) had all 16 CAPS-A speech parameters reported. There was no significant difference in the proportion of children with speech outcomes reported between those with PRS and those without PRS (of whom 62.8% had speech outcomes reported). Eight children (2.3%) had some but not all 16 CAPS-A parameters reported, while 90 children (26.0%) had reasons for not collecting the speech parameters provided, leaving 21 (6.1%) with missing data.

Table 8.2 shows the proportion of children with PRS meeting each speech standard. For comparison, we have included the proportion of children without PRS meeting each speech standard, according to non-syndromic cleft type. These data indicate, for the first time, that children born with PRS do have statistically significant differences in their speech outcomes based on the three nationally agreed speech outcome standards compared with their non-PRS cleft palate peers. The proportion of children with PRS meeting the standards are, however, more comparable to those observed in children without PRS who have an isolated UCLP. Finally, the data show that children with PRS have higher rates of normal speech and the absence of cleft-related articulation difficulties than those with BCLP. We encourage regions / units to continue submitting speech data for children with PRS so that we can continue to track their outcomes and build risk adjusted models.

Table 8.2. Number (%) of CRANE-registered consented children born 2007 - 2012 with an isolated cleft palate and reported speech outcomes at 5 years of age, meeting each speech outcome standard, according to reported Pierre Robin Sequence status and cleft type.

Pierre Robin Sequence (PRS) status and cleft type	N	1. Normal speech N	(%)	2. No structurally-related speech difficulties N	(%)	3. No cleft-related articulation difficulties N	(%)
1. CP plus PRS	227	137	(60.4)	158	(69.6)	155	(68.3)
2. CP without PRS	1158	857	(74.0)	878	(75.8)	953	(82.3)
P value for difference between 1 & 2		***<0.001***		***0.049***		***<0.001***	
3. UCLP without PRS	912	486	(53.3)	621	(68.0)	564	(61.8)
P value for difference between 1 & 3		*0.058*		*0.697*		*0.078*	
4. BCLP without PRS	386	139	(36.0)	246	(63.7)	153	(39.6)
P value for difference between 1 & 4		***<0.001***		*0.149*		***<0.001***	

Although we have been able to explore speech outcomes in children with PRS, the numbers of children with other syndromes, including Van der Woude syndrome, Stickler syndrome and Di George syndrome (22Q11 deletion), are still too low for meaningful analyses of speech data. We recommend that regions / units continue to record speech outcomes (where appropriate) for these children and report them to CRANE so that we can, over time, explore how these children fare in terms of speech.

9. Psychology screening at 5 years of age

This is the second year that Psychology outcomes have been reported in the CRANE Database Annual Report. The assessed parameters of psychology include:

a. date of first face-to-face psychosocial screening,
b. date of psychosocial screening at age five,
c. the Tiers of Involvement Measure (TIM),
d. the Strengths and Difficulties Questionnaire (SDQ), which contributes to a total score, and
e. where the above were not collected, a reason as to why the outcome was not collected (as for all other CRANE outcomes).

The Tiers of Involvement Measure (TIM) is used to record the tier (level) of involvement when a psychologist sees a patient/family in a Cleft Multi-Disciplinary Meeting (MDT) Clinic. The tiers range from 0 to 4 and are described in full in the Methods section (2.1.4) of this report.

The Strengths and Difficulties Questionnaire (SDQ) is a brief behavioural screening questionnaire designed for use with 3-16 year olds. The SDQ asks about 25 attributes, some positive and others negative, which are divided between six scales described in full in the Methods section (2.1.4) of this report.

The CRANE Database collects the ‘Total difficulties’ score as well as the final scores for subscales 1 to 5, resulting from questionnaires completed by the parents of CRANE-registered children at 5 years of age[72].

Exploration of the data collected using the SDQ scores has been conducted according to their categorisation into the following four bands:

1. close to average
2. slightly raised
3. high
4. very high.

Low scores, indicating no concern, are classified as being in the ‘close to average’ range. Scores in the ‘high’ and ‘very high’ ranges indicate a greater level of difficulties.

Of the 1,754 children whose families had consented to their children’s data being collected by CRANE in 2011 and 2012, 1,517 (86.5%) had psychology screening outcome data recorded in CRANE[73], either in the form of screening scores or a valid reason why the outcome was not collected. Data completeness for these cases is shown in Appendix 9, by region/unit.

The figures presented throughout this section of the CRANE report are calculated using valid data as denominators (see Appendix 9 for detail of missing data).

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[72] Using the parent version for 4-16 year olds. Goodman R (1997) The Strengths and Difficulties Questionnaire: A Research Note. Journal of Child Psychology and Psychiatry, 38, 581-586. For more information visit www.sdqinfo.com

[73] Registered in the CRANE Database by the 10 July 2019, and eligible for Psychology outcomes data to be added. **Elxclusions** (not mutually exclusive): No consent, children who died before the age of 5 years, with submucous cleft palates, and syndromic children.

9.1. Date of first face-to-face psychosocial screening

Cohort summary	Data source	CRANE database (consented cases only)
	Birth years	Two years: 2011 and 2012
	Denominator	1,144 children with recorded 'date of first face-to-face screening'.
	Numerator	Number of children with a particular age at first face-to-face screening.
	Data completeness	65% of eligible[74] consented children (n=1,754) had a recorded date of first face-to-face screening.
	Countries	England, Wales and Northern Ireland.
Standard	• 100% of eligible children should be screened at least once before the age of 6 years[75]. • 100% of eligible children should have a recorded date of first face-to-face screening before the age of 6 years (or a recorded reason outcome not provided).	
What did we find?	• 66% of families were seen for a 1st psychosocial screen within the first year of the child's life. • 99% of families were seen before the target age of six years. • 35% of eligible consented children (n=1,754) were missing a recorded date of their first face-to-face screening.	
Recommendations	Units should aim to see all children and families born with a cleft for a first face-to-face psychosocial screen before the age of six years, and this information should be recorded in the audit database.	

The date of the first face-to-face screening was recorded in CRANE for 1,144 children (65% of the total 1,754 eligible children)[76].

- The average age of children at the time of their 'first face-to-face psychosocial screen' was 1 year and 6 months, with half of these children having their first screening before they were 6-months-old[77].
- 66% of families were seen before their child's first birthday.
- 82% of children had their first face-to-face psychosocial screen before the age of five, and 99% before the age of six[78].

Although clinical psychologists have a target to see all children and families born with a cleft for a face-to-face psychosocial screen before the age of six years, families would ideally be seen earlier than this, where team resources and structures allow. This is in order to:

- introduce and normalise psychology as part of the cleft team early on, thereby increasing accessibility of psychology services to families, and
- offer psychological support and intervention, alongside multi-disciplinary colleagues, around issues such as adjustment to diagnosis, parental anxiety around surgery, and managing comments, questions and staring from others.

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[74] As per **exclusions footnote above**.

[75] Clinical Psychological & Counselling Services Standards Core Standard (#38). NHS Standard Contract - Cleft lip and / or palate services including non-cleft velopharyngeal dysfunction (all ages). NHS Commissioning Board, 2013. https://www.england.nhs.uk/specialised-commissioning-document-library/service-specifications/

[76] 8 cases had a recorded date of 'first face-to-face psychosocial screen' prior to their date of birth. These are excluded from reporting.

[77] The age of the oldest child children at the time of their 'first face-to-face psychosocial screen' was 6 years and 4 months.

[78] 17% of children had their first face-to-face psychosocial screen at the age of 5.

9.2. Date of psychosocial screening at age five

Cohort summary	Data source	CRANE database
	Birth years	Two years: 2011 and 2012
	Denominator	1,083 children with recorded date of 'psychosocial screening at age five'.
	Numerator	Number of children meeting one of three screening age categories –before the age of 5, at the age of 5, or after the age of 6 years.
	Data completeness	62% of eligible[79] consented children (n=1,754) had a recorded date of psychosocial screening at age five.
	Countries	England, Wales and Northern Ireland.
Standard	• 100% of children /families should have their 'psychosocial screening at age 5' for the minimum audit at age 5. • 100% of eligible children should have a recorded date of psychosocial screening at age 5, before the age of 6 years (or a recorded reason outcome not provided).	
What did we find?	• 97% had their 'psychosocial screen at age 5' before the age of 6 years. • 38% of eligible consented children (n=1,754) were missing a recorded date of screening at age 5.	
Recommendations	Units should aim to see all children and families born with a cleft for a 'psychosocial screen at age 5' before the age of 6 years, and this information should be recorded in the audit database.	

The date of the psychosocial screening at age 5 was recorded in CRANE for 1,083 children (62% of the total 1,754 eligible children)[80].

- Ninety-seven percent of these children and families had their 'psychosocial screening at age 5' before the age of 6, thereby meeting the target for the collection of outcome audit data at age 5.

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[79] As per **exclusions footnote above**.

9.3. The Tiers of Involvement Measure (TIM)

Cohort summary	Data source	CRANE database
	Birth years	Two years: 2011 and 2012
	Denominator	1,044 children with recorded TIM scores.
	Numerator	Number of children with a particular TIM level of psychological involvement.
	Data completeness	60% of eligible[81] consented children (n=1,754) had a recorded TIM score.
	Countries	England, Wales and Northern Ireland.
Standard	• 100% of children should be seen by a psychologist and have a TIM assessment at 5 years of age[82] • 100% of eligible children should have a recorded TIM score, at the age of 5 (or a recorded reason outcome not provided).	
What did we find?	• 93% were seen by a psychologist and a psychosocial screen was completed (TIM tiers 1 to 4). Of these children, 53% had no psychological concerns identified (TIM tier 1a). • Of those receiving psychological input in clinic (tier 2): ○ 82% received preventative input (tiers 2a and 2c). ○ 44% received input in response to a concern raised by the family (tiers 2b and 2c). ○ 27% received both preventative input and input in response to a concern (tier 2c). • Children who had a UCLP were more likely to be offered an additional psychology appointment (TIM tier 4). • There were no sex differences across TIM levels. • 40% of eligible consented children (n=1,754) were missing a recorded TIM score.	
Recommendations	Units should aim to screen all children born with a cleft, using the TIM, before the age of six years, and this information should be recorded in the audit database	

9.3.1. Proportion of children by TIM level

The TIM was recorded in CRANE for 1,044 children (60% of 1,754 eligible children). Among these children, our analyses revealed that:

- 93% were seen by a psychologist and a psychosocial screen was completed (tiers 1 to 4).
- 41% received psychological input, either in clinic or after clinic (tiers 2, 3 and 4).

These data are shown in Figure 9.1 below.

- Of those seen by a psychologist and who had a psychosocial screen completed (tiers 1 to 4, n=966), 53% had no psychological concerns identified (tier 1a, n=510).
- Of those seen by a psychologist and had a psychosocial screen completed but were not receiving psychological input (all tier 1, n=536), the majority (95%) had no psychological concerns identified (tier 1a, n=510). A small group were having their psychological concerns or needs met by another service (tier 1b, n=26).
- Of those receiving psychological input in clinic (tier 2, n=343):

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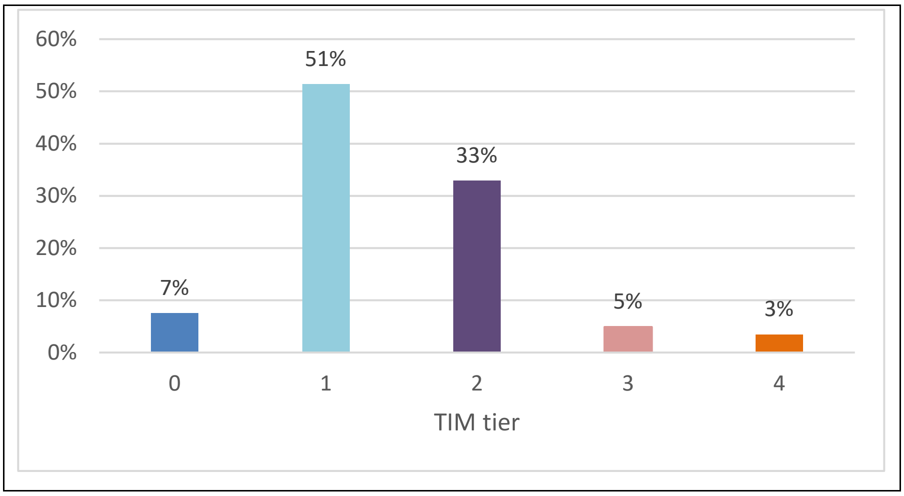
[81] As per **exclusions footnote above**.

[82] Clinical Psychological & Counselling Services Standards Core Standard (#38). NHS Standard Contract - Cleft lip and / or palate services including non-cleft velopharyngeal dysfunction (all ages). NHS Commissioning Board, 2013. https://www.england.nhs.uk/specialised-commissioning-document-library/service-specifications/

  - 82% received preventative input (tiers 2a and 2c, n=191 and n=91, respectively). This is most likely to be in relation to helping children prepare for dealing with comments and questions from others about cleft-related factors but will also include other psychosocial issues.

Figure 9.1. Proportion of CRANE-registered consented children[83] born with a cleft lip or palate (2011 and 2012), according to the Tiers of Involvement Measure (TIM) levels of psychological involvement / input received.



Key:
0 - Child and family not seen by psychologist; 1 - Child and family seen by a psychologist and a psychosocial screen carried out with no further psychological input required; 2 - Psychological input provided in clinic; 3 - Further psychological input required from psychologist after clinic but appointment not necessary; and 4 - Psychology appointment required.

  - 44% received input in response to a concern raised by the family (tiers 2b and 2c, n=61 and n=91, respectively). Examples of concerns include behaviour or developmental concerns.
  - 27% received both preventative input and input in response to a concern (tier 2c, n=91).
- 8% of all children with recorded TIM scores received further input after clinic (tiers 3 and 4, n=51 and n=36, respectively), with a minority being offered a separate psychological appointment (tier 4).

9.3.2. TIM levels by cleft type

As shown in Table 9.1, generally TIM scores were spread evenly across cleft types, in similar proportions as would be expected from general cleft type prevalence data, with a few exceptions and notable points.

- Of those patients not seen (tier 0), 30% had a cleft palate (a proportion approximately 10% smaller than the proportion expected from general cleft type prevalence (~40%)).
- Of those receiving psychological input in clinic (tier 2, n=343), patients with a cleft lip only were more likely to have received preventative psychological input only (2a only).
- Patients who were offered a psychological appointment (tier 4, n=36), were more likely to have a UCLP (39% of children scored in tier 4 had UCLP compared to 27% of children across all TIM tiers had UCLP). The numbers of families involved is fairly small (n=36) but this suggests that, for this cohort, families of children who have a UCLP may benefit from an additional psychology appointment.

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[83] Exclusions (not mutually exclusive): No consent, children who died before the age of 5 years, with submucous cleft palates, and syndromic children.

Table 9.1. Number (%) of CRANE-registered consented children[84] born with a cleft lip or palate (2011 and 2012), according to Tiers of Involvement Measure (TIM) levels and cleft type.

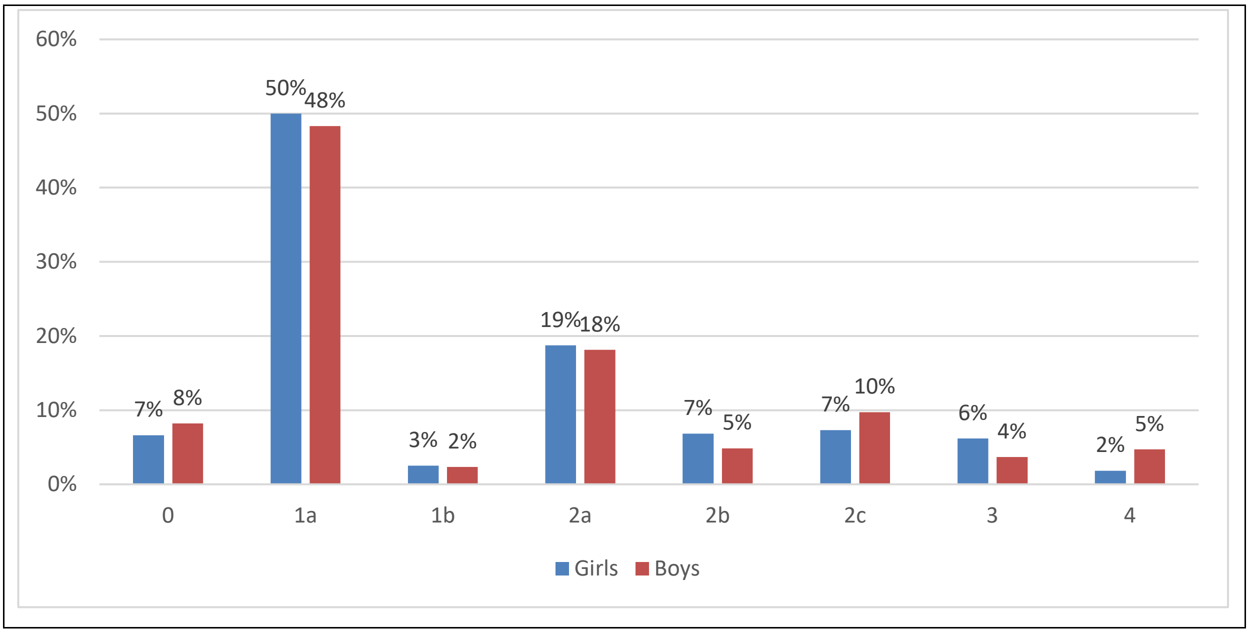
	n (%)				
TIM level	CL	CP	UCLP	BCLP	**All**
0	21 (26.9)	23 (29.5)	24 (30.8)	10 (12.8)	**78 (100)**
1a	103 (20.4)	229 (45.3)	127 (25.1)	46 (9.1)	**505 (100)**
1b	4 (15.4)	12 (46.2)	7 (26.9)	3 (11.5)	**26 (100)**
2a	75 (39.3)	47 (24.6)	51 (26.7)	18 (9.4)	**191 (100)**
2b	6 (9.8)	28 (45.9)	19 (31.1)	8 (13.1)	**61 (100)**
2c	19 (20.9)	29 (31.9)	30 (33)	13 (14.3)	**91 (100)**
3	5 (10.6)	25 (53.2)	9 (19.1)	8 (17)	**47 (100)**
4	6 (16.7)	14 (38.9)	14 (38.9)	2 (5.6)	**36 (100)**
All	239 (23.1)	407 (39.3)	281 (27.1)	108 (10.4)	**1035 (100)**

Key: 0 - Child & family not seen by psychologist; 1 - Child & family seen by a psychologist and a psychosocial screen carried out with no further psychological input required (1a. No input required; 1b. Needs met by another service); 2 - Psychological input provided in clinic (2a. preventative input only; 2b. In response to problem or concern; 2c. As per both 2a & 2b); 3 - Further psych input required from psychologist after clinic but appointment not necessary; and 4 - Psychology appointment required.

9.3.3. TIM levels by Sex

Figure 9.2 shows the proportions of girls and boys by TIM level. For most TIM scores there was an equal split of girls and boys.

Figure 9.2. Proportion of CRANE-registered consented girls and boys[85] born with a cleft lip or palate (2011 and 2012), according to the Tiers of Involvement Measure (TIM) levels of psychological involvement / input received.



Key: 0 - Child & family not seen by psychologist; 1 - Child & family seen by a psychologist and a psychosocial screen carried out with no further psychological input required (1a. No input required; 1b. Needs met by another service); 2 - Psychological input provided in clinic (2a. preventative input only; 2b. In response to problem or concern; 2c. As per both 2a & 2b); 3 - Further psychological input required from psychologist after clinic but appointment not necessary; and 4 - Psychology appointment required.

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[84] As per **exclusions footnote above**.

[85] As per **exclusions footnote above**.

9.4. Strengths and Difficulties Questionnaire (SDQ) 'Total difficulties' scale bands

Cohort summary	Data source	CRANE database
	Birth years	Two years: 2011 and 2012
	Denominator	951 children with recorded SDQ scores.
	Numerator	Number of children with a particular range of SDQ 'Total difficulties' scores.
	Data completeness	54% of eligible[86] consented children (n=1,754) had a recorded SDQ score.
	Countries	England, Wales and Northern Ireland.
Standard	• SDQ population norms[87]: 10% of children aged 5 to 10 years old have SDQ scores that are 'high' (5%) or 'very high' (5%), and 7.6% of girls and 12.2% of boys score 'high' or 'very high'. • 100% of eligible children should have a recorded SDQ score, before the age of 6 years (or a recorded reason outcome not provided).	
What did we find?	• 16% of children born with a cleft had 'high' (7%) or 'very high' (9%) SDQ scores. These proportions are higher than the population norms. • Boys and children with a cleft affecting the palate had a higher proportion of high/very high SDQ scores than the population norms. • 46% of children with a high or very high SDQ score had seen a psychologist and received psychological input in response to a concern, either in clinic or after clinic (including that provided by another service) (TIM tiers 1b, 2b, 2c, 3 and 4). • 18% of children with lower scores on the SDQ had psychosocial concerns identified, highlighting the fact that the SDQ does not identify all psychological concerns arising for children born with a cleft and their families at age 5 years. • 46% of eligible consented children (n=1,754) were missing a recorded SDQ score.	
Recommendations	• As the SDQ does not identify all psychological concerns arising for children born with a cleft and their families at age 5 years, all Units should aim to provide all children born with a cleft, and their families, a face-to-face psychosocial screen and discussion. • Units should aim to screen all children born with a cleft, using the SDQ 'Total difficulties' scale bands, before the age of six years, and this information should be recorded in the audit database.	

The SDQ has been recorded in CRANE for 951 children (54% of 1,754 eligible cases).

SDQ total scores were compared with population norms for the cohort[88] as a whole and were looked at by cleft type and sex. The patterns highlighted below need to be interpreted cautiously because the number of children in some of the groups is small, making it difficult to infer statistical or clinical significance. Furthermore, the population norms cover the age range of 5 to 10 years and our cohort is at the extreme end of this range, all being aged 5 years.

- 16% of children had SDQ total scores within the 'high' and 'very high' ranges combined (7% and 9% respectively). This compares with 10% (5% in each range) in the population norms.

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[86] As per **exclusions footnote above**.

[87] The sample are described in more detail in: Meltzer, H., Gatward, R., Goodman, R., and Ford, F. (2000) Mental health of children and adolescents in Great Britain. London: The Stationery Office.

[88] The normative data in question were collected in 2000. Therefore, it may be that scores for the population norms would have increased over this period.

- 9% of children born with a cleft lip only had SDQ total scores in the ‘high’ and ‘very high’ ranges, which is just below that expected from the population norms. Children born with all other cleft types had a higher proportion than population norms scoring in the ‘high’ and ‘very high’ ranges (CP 18%, UCLP 19% and BCLP 13%).
- 13% of girls had ‘high’ or ‘very high’ SDQ total scores, as compared with 18% of boys. These rates are higher than the corresponding 7.6% of girls and 12.2% of boys scoring in these ranges in the population norms.

9.4.1. TIM levels by SDQ ‘Total difficulties’ scale bands

We explored the relationship between the TIM levels and SDQ ‘Total difficulties’ scale bands.

Table 9.2 shows the proportion of children within each TIM level according to collapsed SDQ Total difficulties scale bands. Children scoring in the ‘high’ and ‘very high’ range on the SDQ are grouped together (n=144), and children scoring in the ‘close to average’ or ‘slightly raised’ ranges are grouped together (n=766). Please see Appendix 8 for the detailed breakdown by the four SDQ Total difficulties scale bands.

Table 9.2. Number (%) of CRANE-registered consented children[89] born with a cleft lip or palate (2011 and 2012), according to grouped Tiers of Involvement Measure (TIM) levels and SDQ Total difficulties scale bands.

	TIM n (%)								
SDQ	0	1a	1b	2a	2b	2c	3	4.	**All**
Cl to aver./ Sl raised	34 (4.4)	424 (55.4)	10 (1.3)	153 (20)	48 (6.3)	51 (6.7)	29 (3.8)	17 (2.2)	**766 (100)**
High / Very high	23 (16)	38 (26.4)	12 (8.3)	17 (11.8)	8 (5.6)	16 (11.1)	12 (8.3)	18 (12.5)	**144 (100)**
All	57 (6.3)	462 (50.8)	22 (2.4)	170 (18.7)	56 (6.2)	67 (7.4)	41 (4.5)	35 (3.8)	**910 (100)**

Key: 0 - Child & family not seen by psychologist; 1 - Child & family seen by a psychologist and a psychosocial screen carried out with no further psychological input required (1a. No input required; 1b. Needs met by another service); 2 - Psychological input provided in clinic (2a. preventative input only; 2b. In response to problem or concern; 2c. As per both 2a & 2b); 3 - Further psychological input required from psychologist after clinic but appointment not necessary; and 4 - Psychology appointment required.

- Of the small number of families who were recorded as not having been seen by a psychologist (tier 0) at age five, most (60%, 34 of 57) of the children had a ‘close to average’ SDQ Total score.
- Of those children scoring in the ‘high’ and ‘very high’ range on the SDQ (n=144):
    - 33% were seen by a psychologist and either had their psychological needs met by another service (tier 1b, 8%), received psychological input in response to a concern in the clinic (tier 2b and 2c, 17%), or after clinic (tier 3, 8%).
    - 13% were offered a separate psychology appointment (tier 4).
    - 38% were seen by a psychologist and either no psychological concerns were identified (tier 1a, 26%), or they were provided with preventative psychological input in the clinic (tier 2a, 12%)
    - Only 16% were not seen by a psychologist (tier 0).
    - *Therefore, for the majority of children with an elevated SDQ score (38%), the* cleft team *psychologist identified that they either had no psychological concerns or required input in the clinic that was preventative only.*
- Of those children with SDQ scores in the ‘close to average’ or ‘slightly raised’ ranges (n=766):

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[89] Exclusions (not mutually exclusive): No consent, children who died before the age of 5 years, with submucous cleft palates, and syndromic children.

    - 18% were seen by a psychologist and either had their psychological needs met by another service (tier 1b, 1%), received psychological input in response to a concern in the clinic (tier 2b and 2c, 13%), or after clinic (tier 3, 4%).
    - 2% were offered a separate psychology appointment (tier4).
    - *This demonstrates that the SDQ does not identify all psychological concerns arising for children born with a cleft and their families at age 5 years, highlighting the importance of a face-to-face psychosocial screen and discussion.*

9.5. Summary, considerations and limitations

There was a considerable variation between cleft teams' data completeness and a decision was taken, for this second year of reporting psychology data, to report on the national picture. The more detailed data will be used by the psychology Clinical Excellence Network (CEN) to gain a better understanding of the reasons for the variation.

With regards to the patient journey, where data have been supplied:

- Families were almost all seen for a face-to-face psychosocial screen before the target age of six years and the majority (66%) were seen for a psychosocial screen within the first year of the child's life.
- 97% of children and families had their 'psychosocial screen at age five' before the age of six, meeting the target for the collection of outcome audit data.

With regards to the level of psychological input (measured with the TIM) provided to families at age five when they were seen by a psychologist and psychosocial screen carried out:

- 53% had no psychological concerns identified.
- 33% received psychological input in the clinic (tier 2), of whom:
    - 82% received preventative input (tiers 2a and 2c),
    - 44% received input in response to a concern they raised (tiers 2b and 2c), and
    - 27% received both preventative input and input in response to a concern (tier 2c).
- 8% received psychological input after the clinic, with a minority receiving a psychology appointment.
- Cleft type did not predict those families who required or would have potentially benefited from an additional psychology appointment.

Looking at scores on the SDQ and comparing this with the level of psychological input provided to families at age five:

- For the majority children with an elevated SDQ score, the cleft team psychologist identified psychological concerns and provided psychological input to the family.
- The SDQ does not identify all psychological concerns arising for children born with a cleft and their families at age 5 years; however, 18% of children with lower scores on the SDQ had psychosocial concerns identified, highlighting the importance of a face-to-face psychosocial screen and discussion.

The SDQ Total scores identified a higher proportion of children with a cleft affecting the palate scoring in the 'high' and 'very high' ranges compared with population norms. A higher proportion of boys than girls scored in the 'high' and 'very high' ranges for the SDQ Total score and for all subscales. However, these findings should be considered with caution due to factors detailed above.

10. Educational attainment at 5, 7 and 11 years of age

Cohort summary	Data source	CRANE database (consented cases only) – Hospital Episode Statistics (HES) – National Pupil Database (NPD)
	Inclusion criteria	CRANE-consented children born with an isolated (non-syndromic) oral cleft with CRANE-HES-NPD linked data who had teacher-based assessments at 5, 7 and 11 years.
	Years	5 years old and in reception between 01/09/06 and 31/08/08 and followed up until age 11 years (2012 to 2014) when they undergo Key Stage 2 assessments.
	Size (N)	832
	Countries	England
Outcomes	• The proportion of children born with a cleft achieving the expected levels for Reading, Writing, Maths and Science at each time point: (1) Early Years Foundation Stage at age 5 years, (2) Key Stage 1 at 7 years, and (3) Key Stage 2 and 11 years. • The attainment gap between children born with a cleft and the general population, as summarised using z-scores.	
What did we find?	• Children born with isolated clefts have lower educational attainment than children in the general population in all subject areas throughout their primary education. • The size of the attainment gap does not appear to change with age or between subjects.	
Recommendations	• Further work is needed to identify risk factors associated with lower educational attainment in this clinical population. • Those involved in commissioning and planning special educational services should be aware that children with a cleft may require academic support or specific support around the treatment pathway.	

10.1. Background

Several cross-sectional studies have shown that children and adolescents with clefts are at increased risk of learning difficulties and lower levels of achievement at school than their unaffected peers[90]. The largest population-based study to date, conducted in a cohort of 2,802 children born with an isolated cleft in England between 2001 and 2007[91], identified that children with clefts had lower educational attainment than the general population across all areas of learning at age 5 years. The aim of the current study was to describe longitudinal educational attainment among children with a cleft, and to describe changes in the attainment gap between those with a cleft and the general population.

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[90] Gallagher ER, Collett BR. (2019) Neurodevelopmental and academic outcomes in children with orofacial clefts: A Systematic Review. *Pediatrics* :e20184027.

[91] Fitzsimons et al. (2018) Early academic attainment in children with isolated clefts: a population-based study in England. *Archives of Disease in Childhood.* 103(4):356-362

10.2. Data source

The National Pupil Database (NPD) contains individual-level records of educational outcomes for all children attending state schools in England. CRANE-registered children whose parents provided consent for linkage with other datasets had their CRANE records matched with NPD records using key identifiers. This allowed us to analyse teacher-assessed educational attainment across Reading, Writing Maths and Science at Early Years Foundation Stage (assessed at age 5 years, the first year of primary education), Key Stage 1 (7 years) and Key Stage 2 (11 years, the last year of primary education). CRANE-NPD linked records were also matched with records from Hospital Episode Statistics (HES) in order to identify children with an isolated (non-syndromic) cleft and exclude those with additional anomalies and/or syndromes.

10.3. Study sample

There were 2,307 consented children in the CRANE-National Pupil Database (NPD) linked dataset who were 5 years old between 1 September 2006 and 31 August 2008. Of these, 2,122 (92.0%) had a linked record in the Hospital Episode Statistics (HES) database, which at the time of analyses contained records on all admissions to National Health Service (NHS) hospitals in England up to 31 March 2015.

Linkage to HES was used to exclude 500 children (23.6%) who were identified as having additional anomalies or syndromes (ie, a non-isolated cleft) based on the presence of 33 specific International Classification of Diseases, 10th edition (ICD-10) diagnostic codes (see Appendix 4) representing congenital malformations and chromosomal abnormalities in any of the diagnosis code fields of their HES records.

A further 44 children were excluded as their cleft type was unknown. Data from 1,578 children were reviewed to exclude cases with missing attainment records. A cohort of 832 children with isolated clefts and who had Reading, Writing, Maths and Science attainment data at ages 5, 7 and 11 years formed the study sample, representing two academic year cohorts (age 5 in 2006/07 and 2007/08).

10.4. Analyses

At each age, educational outcomes for children with a cleft were summarised as the proportion achieving expected levels of attainment in each subject. To compare attainment levels with the general population, mean attainment scores were converted into z scores using national population means and standard deviations[92]. A z-score represents the signed number of SD by which the child’s actual score is above the national average. A negative z-score indicates that the score is below the national average. A repeated measures analysis of variance was used to determine whether there was any change in the size of the attainment gap with age.

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[92] Department for Education. *DCSF: Early Years Foundation Stage Profile Results in England, 2008/09*. 2009 [cited 19 February 2019]; Available from: https://webarchive.nationalarchives.gov.uk/20130323143251/https://www.education.gov.uk/researchandstatistics/statistics/allstatistics/a00196194/early-years-foundation-stage-profile-results-in-en; Department for Education. *National curriculum assessments at key stage 1: 2011* 2011 [cited 19 February 2019]; Available from: https://www.gov.uk/government/statistics/national-curriculum-assessments-at-key-stage-1-in-england-2011; Department for Education. *National curriculum assessments: key stage 2, 2014 (revised)* 2014 [cited 19 February 2019]; Available from: https://www.gov.uk/government/statistics/national-curriculum-assessments-at-key-stage-2-2014-revised.

10.5. Results

10.5.1. School attainment among children with isolated clefts

Among children with an isolated cleft, the proportion achieving expected levels of attainment across different subjects is shown in Table 10.1. Writing had the lowest proportion of children with a cleft achieving the expected level of attainment at all three time points. This is consistent with data in the general population (see figures in brackets in Table 10.1). Across all subjects and time points, the cleft population underperformed in comparison to the general population. Differences in the rates of achieving expected levels between the cleft and general population ranged from 6.4% (Maths and Science at KS1) to 11.4% (Reading at EYFS). The average difference in the proportion achieving expected levels between the cleft and general population across all subjects combined was 10.1%, 7.0% and 8.4% at EYFS, KS1 and KS2, respectively.

Table 10.1 Percentage of children born with isolated oral clefts achieving expected levels of attainment in Reading, Writing, Maths and Science at each assessment stage, based on teacher assessment. Corresponding rates from the general population across the same time period are shown in brackets.[93]

	EYFS (5 years)		KS1 (7 years)		KS2 (11 years)	
Subject	Cleft pop.	General pop.	Cleft pop.	General pop.	Cleft pop.	General pop.
Reading	59.0	(69.5)	77.8	(84.5)	81.4	(88.0)
Writing	48.1	(59.5)	72.4	(81.0)	75.7	(84.0)
Maths	57.6	(67.0)	82.6	(89.0)	76.7	(87.5)
Science	68.9	(78.0)	82.6	(89.0)	80.3	(88.0)

EYFS – Early Years Foundation Stage, age 5 years; KS1 – Key Stage 1, age 7 years; KS2 – Key Stage 2, age 11 years.

10.5.2. Attainment gap between children with clefts and the general population

Children with isolated oral clefts had attainment Z-scores that were statistically significantly lower than the national average in all four subject areas, ranging from -0.30 (95% CI -0.38 to -0.23) to -0.22 (-0.29 to -0.15) (Figure 10.1). Based on national distributions of attainment scores, a Z-score of 1 was equivalent to approximately 1.5 points at age 5, 4 points at age 7 and 5.5 points at age 11 (see Table 2.1 and 2.2 in Methods section 2.3.1). The size of the attainment gap (measured as Z-scores) did not vary across assessment ages for any of the subject areas (Figure 10.1), indicating the attainment gap observed at age 5, when children begin their school journey, persists until at least age 11 years.

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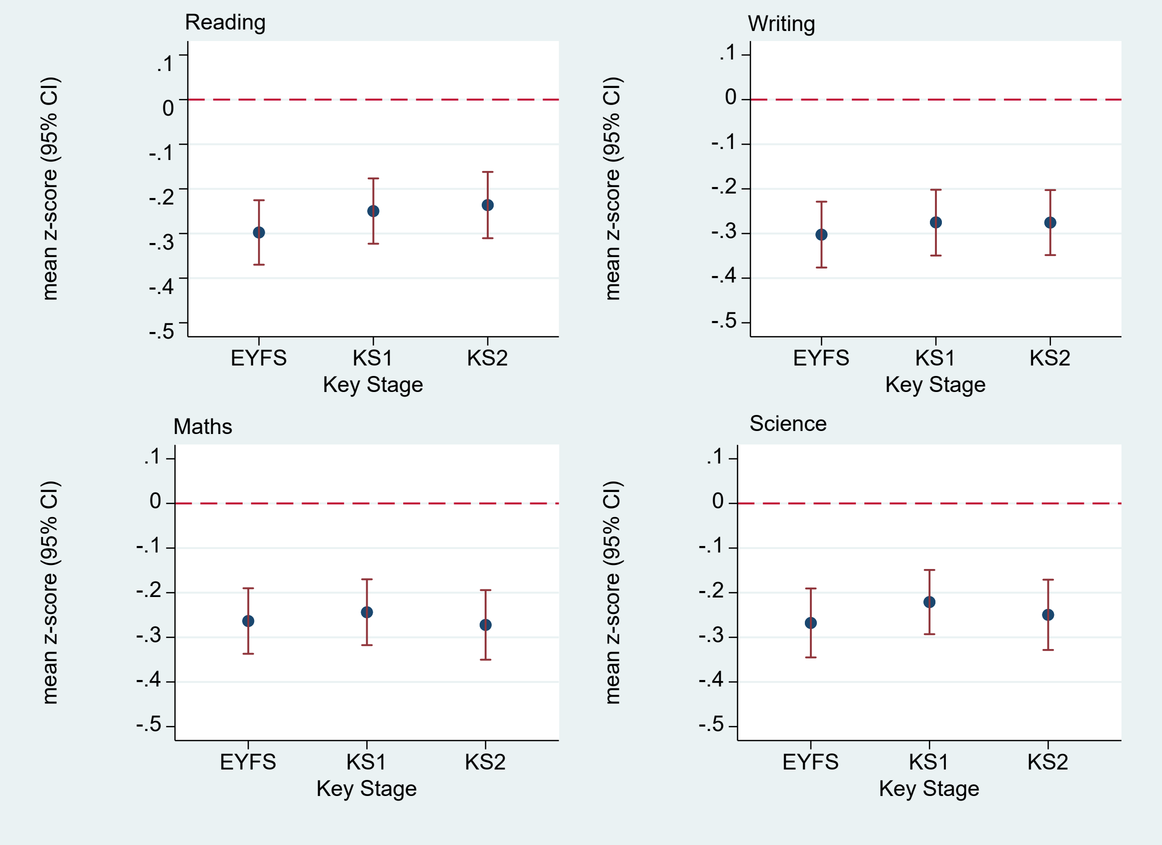
[93] https://www.gov.uk/government/collections/statistics-early-years-foundation-stage-profile
https://www.gov.uk/government/collections/statistics-key-stage-1
https://www.gov.uk/government/collections/statistics-key-stage-2

Figure 10.1 Mean Z-scores and 95% confidence intervals at each Key Stage comparing children with an isolated cleft to general population, by subject



10.6. Conclusions

Children born with an isolated oral cleft in England have lower educational attainment than children in the general population at ages 5, 7 and 11 years. The effect is similar across the four assessed subject areas (Reading, Writing, Maths and Science). The size of the attainment gap is consistent at each assessment stage, and does not appear to narrow or broaden over the course of children's primary education. The persistent nature of the attainment gap throughout primary education suggests that children with isolated oral clefts may be a group that would benefit from early, targeted interventions. Further research is needed to identify effective interventions for children at risk of lower school attainment and we recommend further evaluation of educational data to determine whether attainment gaps persist into secondary education.

11. Development of CRANE Database and future directions

11.1. Future development of the CRANE Database and website

CRANE is negotiating with NHS England a new contract to sustainably support the project going forward. This will fund the registry and audit function of the database. Funding of significant research activity beyond these functions will continue to be sought through collaborative applications.

A new contract has been agreed with our IT provider that will involve a 12-month period of transitioning of the platform to a UK based product with enhanced flexibility. Upgrades/agreed developments of the database will also happen during this period. Transitioning to a UK-based platform will ensure sustainability and compliance irrespective of the BREXIT outcome.

Over the period of transitioning, CRANE will continue work on specifying data collection in the following sections for the database, as proposed by our stakeholders:

- LAHSAL data collection items changed to collect LAHSHAL data to increase the phenotypic data available for analysis and linkage to other projects.
- Dental Defects of Enamel (DDE) section/items (at 5 and 10yrs) as proposed by the Paediatric Dental CEN of CFSGBI.
- Surgical data collection. This will be taken forward in collaboration with the Surgical CEN of CFSGBI

11.2. Scotland

NHS Scotland management have reaffirmed their intention to submit data to the CRANE Database. We are currently engaged in the process of setting up the necessary permissions for sharing data with Scotland for a start date of 1st April 2020. NHS Scotland Management have also indicated a wish to not only prospectively enter data relating to new births but also investigate the possibility of retrospective entry for children consented at 5 year old audit. This will allow the project to achieve full UK coverage and is a significant step forward.

11.3. ICHOM

CRANE is currently awaiting memorandum of understanding from the ICHOM group on what type of relationship they seek with the cleft community in the UK. Upon receipt, there will be a need to share and discuss this document with the UK cleft community on the practicalities/extent of potential involvement. If involvement is supported by CDG/Cleft teams, discussion will be required on what additional resource this will require over and above the current CRANE activity.

11.4. Outcome measures

11.4.1 Risk Stratification

Despite the significant improvement in both volume and quality of data collection across the spectrum of cleft care, the lack of ability to risk stratify data continues to undermine the valid comparison of the outcomes achieved across the UK. A volume of data is now available within CRANE and is starting to be utilised for investigating risk stratification of cleft outcomes in the UK. Moving forward, the database intends to look more closely at risk stratification with the aim of incorporating agreed risk models into future reports. Thereby, allowing valid risk adjusted comparison of cleft outcomes in the UK.

11.4.2 Young People and Adult Outcomes

The clinical directors group of the CFSGB&I have previously asked CRANE to lead on a project to agree an outcome set for young people and adults. A multiphase Delphi consultation using different methods of engagement to develop consensus and identify valid and robust measures is currently in progress. It is hoped that this process will be complete for inclusion in the 2020 report.

11.5. Data sources and future analyses

Over the last 12 months CRANE has collaborated with the Cleft Collective on a funding bid that would allow the Cleft collective to be linked to various data sources including the CRANE Database. Unfortunately, this first application has been unsuccessful in its first funding bid. However, the desire to develop such collaboration remains, and alternate funding streams will be sought to allow the strengths of this type of collaboration to benefit the cleft community.

11.5.1. National Pupil Database (NPD)

CRANE continues its work with NPD and CRANE-HES-linked data and is seeking new linkage following changes to the process by the Department for Education (DfE) after GDPR introduction.

We are currently exploring school absence and its relationship with academic attainment among children with a cleft. We also plan to develop our initial analyses of longitudinal educational attainment among children with a cleft. Tracking educational attainment over time has potential to be utilised as part of CRANE outcome analyses demonstrating efficacy of cleft care delivery in the UK.

11.5.2. Hospital Episode Statistics (HES)

CRANE is in the process of applying to NHS Digital to allow for linkage of the full CRANE dataset to HES data from the year 2000 (the year CRANE started registering patients).

One of the first analyses planned once such linkage is available will be the investigation of the timing of cleft repairs and its association with 5 year old speech and growth audit outcomes.

CRANE through the CEU already has access to a rolling retrospective 10 year HES dataset. This allows for analysis and comparison of recent historic cleft-related activity in NHS hospital in England with similar activity for non-cleft patients. CRANE intends to use such access and its experience with HES data to investigate geographic variations in adult cleft hospital activity over the last 10 years. This will, for the first time, comprehensively inform patients/clinicians and commissioners as to what care has taken place across NHS England over the recent past. This data has the potential to provide a resource for future planning of young people and adult services.

11.5.3. Newborn Hearing Screening Programme (NHSP)

We have approval for linkage between our CRANE Database and the Newborn Hearing Screening Programme (NHSP)[94] data – via Public Health England (PHE) – with the purpose of looking at the relationship between clefts and Permanent Childhood Hearing Impairment (PCHI) and the effect of PCHI on children's outcomes. We are just awaiting confirmation of resource allocation at PHE to undertake the linkage process.

11.6. Quality Dashboard

The CRANE project team have submitted data for the 2016/17, 2017/18 and 2018/19 quality dashboards. This was done for the following five out of the six items requested:

- Measure Number CLP00: The number of CRANE-registered children born within a specified quarter of the calendar year (refreshed every quarter).
- Measure Number CLP01: The number of Parents contacted by a Cleft Team Clinical Nurse Specialist (CNS) within 24 hours of referral with an antenatal diagnosis of Cleft Lip and/or Palate – born within a specified quarter of the calendar year (refreshed every quarter).
- Measure Number CLP02: The number of Parents receiving visit from a Cleft Team CNS within 24 hours of first referral (provided the child has not reached the age of one year) – born within a specified quarter of the calendar year (refreshed every quarter).
- Measure Number CLP06: The number of 5 year old children with a decayed, missing and filled teeth (dmft) index score, as a percentage of all 5 year old children (refreshed annually).
- Measure Number CLP09: The number of 5 year old children with 5 year old index scores 1 or 2 (as indicator of maxillary growth in patients with complete UCLP[95]) – as a percentage of the number of 5 year old children with a 5 year old index score (refreshed annually) [previously numbered CLP08].

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

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

Future productions of Quality Dashboard CRANE tables have been confirmed – potentially including speech data. Discussions on timelines for data submission have been had and are due to be circulated by Methods in the near future.

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

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

11.7. CRANE Communications

11.7.1 Dissemination of 2019 findings

- Publication of the Annual Report will be announced via the regular December eNewsletter. The report will be available on the CRANE website from this time.
- A Summary of Findings for Patients and Parents/Carers from this 2019 Annual Report will be produced in collaboration with CLAPA. CRANE aims to publish this contemporaneously with the main report. The summary will also be made available on the CRANE website
- A Twitter feed for the project, to help highlight and share activity, developments and outputs throughout the year, has been active since August 2019 (@CRANE_News)

11.7.2 Publications and Presentations related to the CRANE Database

Peer reviewed Publications

Early academic achievement in children with isolated clefts: a population-based study in England
Fitzsimons KJ, Copley LP, Setakis E, Charman SC, **Deacon** SA, Dearden L, van der Meulen JH.
Arch Dis Child. 2018 Apr;103(4):356-362. doi: 10.1136/archdischild-2017-313777. Epub 2017 Nov 2.

Oral Presentations

Consultation on older children outcomes: Update on a multiphase Delphi method approach
Medina J, Fitzsimons KJ, Park MH, Van Der Meulen J and Deacon SA
CFSGBI Annual Scientific Conference April 2019, Birmingham

Longitudinal Educational Attainment among children with isolated orofacial cleft
Park MH, Fitzsimons KJ, Medina J, Deacon SA and Van Der Meulen J
CFSGBI Annual Scientific Conference April 2019, Birmingham

What and When to measure in cleft care: my experiences in the UK
Deacon SA
European Cleft Palate and Craniofacial Association Scientific Congress, June 2019, Utrecht

11.6.3 Public Interaction

As a new initiative, CRANE has decided to actively participate in scientific conferences through the manning of a stand. This will allow direct dissemination of findings with patients, clinicians and scientists attending the conferences, offer direct training opportunities to cleft clinicians / administrators from around the UK and allow for active conversations about the opportunities of CRANE collaboration. Furthermore, in the era of GDPR, such activity also allows for contact consent to be obtained directly from individuals attending the conferences. Direct (e-mail/phone/twitter) contact from the Project will further strengthen attempts to widen distribution of the Databases findings/ publications.

Scientific conferences to be attended:

1. CFSGBI Annual Scientific Conference April 2020, Cardiff.
2. RCPCH Conference April 2020, Liverpool (TBC).

12. Conclusion

As it moves into its 20th year of existence, CRANE looks forward to building on past successes for the betterment of patients, and the clinicians who work for them. We aim to continue to provide good and robust data that delivers quality information that clinicians can use to make treatment decisions with their patients. It allows commissioners and patients access to independent information on the activity and outcomes achieved by each of the cleft regions/units in England, Wales and Northern Ireland and in due course Scotland.

CRANE now holds a data repository relating to over 20,000 patients. Some data quality issues do exist with missing data which reduces the 'real number of' patient outcomes available for analysis to less than the 20,000 patients. Also there is a 5-7 year lag from initial patient registration until outcome data is available on CRANE to report. We will continue to work with cleft centres to improve data completeness but we are now in a position, with a sufficient volume of data, to more accurately inform about features pertaining to better cleft outcomes. For example, whilst previously understood by clinicians on an anecdotal basis, this year we have been able to inform (in a statistically robust manner) that patients with different cleft types have different likelihood of good speech outcomes (in line with non-cleft peers) by age 5. This is the first step in building good risk stratification models that will allow for valid comparison of the clinical care provided by different cleft centres. We hope to extend this work over the next 12 months to explore other features that are linked with good speech outcomes and investigate features influencing outcomes across all other aspects of cleft care.

We now have an exciting future at CRANE with the ever-increasing size of our own data set, experience gained with data linkage with other national data sets (health, education and others), and the continued enthusiasm of Cleft units for data collection and submission. (**Alongside the requirement in the National Service Specification for cleft lip and/or palate services to continue to improve the quality and completeness of data held in the CRANE Database.)** We are in key position to collaborate with the Cleft Collective, the young researchers collaborative of the Craniofacial Society of Great Britain and Ireland, members of cleft teams, patient groups and cleft clinicians. This means an exciting future exists for CRANE.

We look forward to communicating all progress in both future annual reports and in real-time via our Twitter feed.

Best wishes,

The CRANE Project Team.

Appendices

Appendix 1: CRANE Project Team

Members of CRANE Project Team

Scott Deacon	Clinical Project Lead / Interim Clinical Director Bristol Dental Hospital (Until December 2019)	Clinical Effectiveness Unit / South West Cleft Unit, University Hospital Bristol NHS Foundation Trust University of Bristol
Jibby Medina	Research Fellow	Clinical Effectiveness Unit
Kate Fitzsimons	Research Fellow	Clinical Effectiveness Unit
Craig Russell	Clinical Project Lead / Consultant Surgeon at (From September 2019)	Clinical Effectiveness Unit / NHS Greater Glasgow and Clyde
Hussein Wahedally	Data Manager	Clinical Effectiveness Unit
Jan van der Meulen	Clinical Epidemiologist	Clinical Effectiveness Unit / London School of Hygiene and Tropical Medicine
Min Hae Park	Assistant Professor (From June 2018)	Clinical Effectiveness Unit
Catherine Foster	CEU Research Coordinator (From December 2017)	Clinical Effectiveness Unit

Appendix 2: Governance and funding

Ownership

The database is funded by the National Health Service through the Specialist Service Commissioners, who have responsibility for the delivery of care to children born with cleft lip and palate in England and Wales. An independent body, the Cleft Development Group, which represents patient representative groups, clinicians and commissioners, has the overall responsibility for running the database..

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.

The Cleft Development Group CRANE web page provides detail on the CDG Membership and Terms of Reference.

Funding

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

Appendix 3: Regional Cleft Centres and Managed Clinical Network and their associated regions / units

The CRANE Database covers England, Wales and Northern Ireland. Cleft care is currently delivered by eight Regional Cleft Centres and two Managed Clinical Networks. Several of the Regional Cleft Centres are split between two hospitals, where the primary surgery is usually undertaken, and therefore Hospitals/ Administrative Units in a region may submit data separately to the CRANE Database, as shown in the Table below.

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

Notes:
MCN – Managed Clinical Network.
*Data for GOSH and Broomfield units combined upon request by the Spires' Clinical Director (January 2017).
**Data for Oxford and Salisbury units combined upon request by the Spires' Clinical Director (June 2016).
***Frenchay Hospital, Bristol service moved to University Hospitals Bristol during 2014.

Appendix 4: Diagnosis and Procedure Codes, Hospital Episode Statistics (HES)

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

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

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

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

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

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

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

Appendix 5: Five Year Old Index scores detail

Number (%) of CRANE-registered consented children born between 2004 and 2012 with a complete unilateral cleft lip and palate, according to Five Year Old Index scores and region / unit.

Regional centre / MCN	**Administrative Unit**	**Five Year Old Index*** n (%)					**All**[a]
		1	2	3	4	5	
Northern & Yorkshire	Newcastle	8 (12.5)	9 (14.1)	28 (43.8)	13 (20.3)	6 (9.4)	64
	Leeds	8 (11.6)	27 (39.1)	21 (30.4)	12 (17.4)	1 (1.4)	69
North West & North Wales	Liverpool	4 (6.5)	18 (29)	20 (32.3)	14 (22.6)	6 (9.7)	62
	Manchester	4 (4.8)	33 (39.3)	22 (26.2)	14 (16.7)	11 (13.1)	84
Trent	Nottingham	6 (7.5)	25 (31.3)	31 (38.8)	10 (12.5)	8 (10)	80
West Midlands	Birmingham	9 (8.2)	36 (32.7)	36 (32.7)	22 (20)	7 (6.4)	110
East	Cambridge	4 (5.7)	24 (34.3)	22 (31.4)	17 (24.3)	3 (4.3)	70
North Thames	GOSH & Chelms.	6 (8.5)	30 (42.3)	24 (33.8)	11 (15.5)	0 (0)	71
The Spires	Oxford & Salisbury	13 (10.2)	42 (33.1)	39 (30.7)	26 (20.5)	7 (5.5)	127
South Wales & South West	Swansea	0 (0)	2 (18.2)	5 (45.5)	2 (18.2)	2 (18.2)	11
	Bristol	1 (2.1)	17 (35.4)	12 (25)	11 (22.9)	7 (14.6)	48
South Thames	Guy’s and St Thomas’	11 (8.1)	56 (41.5)	39 (28.9)	19 (14.1)	10 (7.4)	135
Northern Ireland	Belfast	1 (4.3)	5 (21.7)	11 (47.8)	4 (17.4)	2 (8.7)	23
All	All	75 (7.9)	324 (34)	310 (32.5)	175 (18.3)	70 (7.3)	954

Appendix 6: 5-year old speech outcome data completeness

Number (%) of CRANE-registered[a] consented children born with a cleft palate in 2010-2012, with speech outcome data or reasons this outcome was not collected at 5 years of age, according to region / unit.

		Consented eligible cases[b]	All 16 CAPS-A scores reported		<16 CAPS-A scores reported		Reason reported for not collecting outcome		Total cases acc. for	Missing data	
Region/MCN	**Administrative Unit**	**N**	**n**	**(%)**	**n**	**(%)**	**n**	**(%)**	**(%)**	**n**	**(%)**
Northern	Newcastle	108	90	(83.3)	2	(1.9)	15	(13.9)	(99.1)	1	(0.9)
& Yorkshire	Leeds	116	96	(82.8)	2	(1.7)	16	(13.8)	(98.3)	2	(1.7)
North West	Liverpool	123	72	(58.5)	8	(6.5)	28	(22.8)	(87.8)	15	(12.2)
& North Wales	Manchester	165	117	(70.9)	5	(3.0)	39	(23.6)	(97.6)	4	(2.4)
Trent	Nottingham	167	133	(79.6)	2	(1.2)	30	(18)	(98.8)	2	(1.2)
West Midlands	Birmingham	213	146	(68.5)	3	(1.4)	50	(23.5)	(93.4)	14	(6.6)
East	Cambridge	123	91	(74.0)	4	(3.3)	26	(21.1)	(98.4)	2	(1.6)
North Thames	GOSH & Chelmsford	247	140	(56.7)	4	(1.6)	46	(18.6)	(76.9)	57	(23.1)
The Spires	Oxford & Salisbury	189	123	(65.1)	5	(2.7)	60	(31.7)	(99.5)	1	(0.5)
South Wales	Swansea	93	69	(74.2)	1	(1.1)	21	(22.6)	(97.8)	2	(2.2)
& South West	Bristol	127	93	(73.2)	3	(2.4)	27	(21.3)	(96.9)	4	(3.1)
South Thames	Guy's and St Thomas'	178	131	(73.6)	0	(0)	39	(21.9)	(95.5)	8	(4.5)
Northern Ireland	Belfast	82	45	(54.9)	1	(1.2)	25	(30.5)	(86.6)	11	(13.4)
All	**All**	**1,931**	**1,346**	**(69.7)**	**40**	**(2.1)**	**422**	**(21.9)**	**(93.6)**	123	(6.4)

[a] Registered in CRANE by 10 July 2019. Note: MCN - Managed Clinical Network.

[b] Exclusions (not mutually exclusive): children who died before the age of 5 years, with submucous cleft palates, missing one or more of all 16 CAPS-A data items, born with either a CL or a non-specified cleft type, and syndromic children.

Appendix 7: Cleft Audit Protocol for Speech: Ratings for individual 16 CAPS-A parameters

Resonance and Nasal Airflow

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

Table A. Number (%) of CRANE-registered [a] consented children born with a cleft palate in 2010-2012, according to the four parameters for resonance and nasal airflow

Description	Score	N	(%)
RESONANCE – HYPERNASALITY			
Absent	0	1,035	(76.9)
Borderline – minimal	1	153	(11.4)
Mild – evident on close vowels	2	76	(5.6)
Moderate – evident on open and close vowels	3	38	(2.8)
Severe – evident on vowels and voiced consonants	4	44	(3.3)
RESONANCE – HYPONASALITY			
Absent	0	1,123	(83.4)
Mild – partial dentalization of nasal consonants and adjacent vowels	1	202	(15)
Marked – dentalization of nasal consonants and adjacent vowels	2	21	(1.6)
NASAL AIRFLOW – AUDIBLE NASAL EMISSION			
Absent on pressure consonants	0	1,229	(91.3)
Occasional: pressure consonants affected <10% of the sample	1	88	(6.5)
Frequent: pressure consonants affected >10% of the sample	2	29	(2.2)
NASAL AIRFLOW – NASAL TURBULENCE			
Absent on pressure consonants	0	1,067	(79.3)
Occasional: pressure consonants affected <10% of the sample	1	246	(18.3)
Frequent: pressure consonants affected >10% of the sample	2	33	(2.5)
TOTAL		**1,346**	**(100)**

[a] Registered in CRANE by 10 July 2019.

Cleft Speech Characteristics (CSCs)

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

Table B. Number (%) of CRANE-registered[a] consented children born with a cleft palate in 2010-2012, according to the twelve Cleft Speech Characteristics (CSCs) parameters.

Cleft Speech Characteristics (CSCs)		Score	N	(%)
ANTERIOR ORAL CSCs	1. Dentalisation / Interdentalisation	A	1,068	(79.3)
		B	278	(20.7)
	2. Lateralisation / Lateral	A	1,237	(91.9)
		B	65	(4.8)
		C	44	(3.3)
	3 Palatalisation / Palatal	A	1,036	(77)
		B	146	(10.8)
		C	164	(12.2)
POSTERIOR ORAL CSCs	4. Double Articulation	A	1,297	(96.4)
		B	42	(3.1)
		C	7	(0.5)
	5. Backed to Velar / Uvular	A	1,183	(87.9)
		C	65	(4.8)
		D	98	(7.3)
NON ORAL CSCs	6. Pharyngeal Articulation	A	1,320	(98.1)
		C	9	(0.7)
		D	17	(1.3)
	7. Glottal Articulation	A	1,253	(93.1)
		C	44	(3.3)
		D	49	(3.6)
	8. Active Nasal Fricatives	A	1,208	(89.7)
		C	76	(5.6)
		D	62	(4.6)
	9. Double Articulation	A	1,310	(97.3)
		C	22	(1.6)
		D	14	(1)
PASSIVE CSCs	10. Weak and or nasalised consonants	A	1,264	(93.9)
		C	33	(2.5)
		D	49	(3.6)
	11. Nasal realisation of plosives	A	1,298	(96.4)
		C	24	(1.8)
		D	24	(1.8)
	12. Gliding of fricatives	A	1,325	(98.4)
		C	13	(1)
		D	8	(0.6)
		TOTAL	**1,346**	**(100)**

[a] Registered in CRANE by 10 July 2019.

Appendix 8: Cleft Audit Protocol for Speech: Speech Outcome Standards

Table A. Raw data for funnel plots. Number (%) of CRANE-registered consented children born in 2010 - 2012 with reported speech outcomes at 5 years of age, meeting each speech outcome standard, according to Regional Centre / Administrative Unit.

Regional Centre	Admin Unit	Reported[**]	Standard 1. Normal speech n	(%)	Standard 2: No structurally-related speech difficulties n	(%)	Standard 3: No cleft-related articulation difficulties n	(%)
Northern & Yorkshire	Newcastle	90	57	63.3	56	62.2	61	67.8
	Leeds	96	41	42.7	55	57.3	56	58.3
North West & North Wales	Liverpool	72	42	58.3	46	63.9	48	66.7
	Manchester	117	72	61.5	81	69.2	80	68.4
Trent	Nottingham	133	85	63.9	109	82.0	96	72.2
West Midlands	Birmingham	146	94	64.4	124	84.9	97	66.4
East	Cambridge	91	55	60.4	59	64.8	62	68.1
North Thames	GOSH & Chelmsford	140	83	59.3	88	62.9	100	71.4
The Spires	Oxford & Salisbury	123	76	61.8	95	77.2	87	70.7
South Wales & South West	Swansea	69	43	62.3	58	84.1	45	65.2
	Bristol	93	57	61.3	69	74.2	62	66.7
South Thames	Guy's and St Thomas'	131	75	57.3	84	64.1	82	62.6
Northern Ireland	Belfast	45	38	84.4	38	84.4	39	86.7
All 2010-2012 births		**1,346**	**818**	**60.8**	**962**	**71.5**	**915**	**68.0**
All 2007-2009 births		**1,317**	**789**	**59.9**	**925**	**70.2**	**898**	**68.2**
2004-2006 births[$]				**55**		**67**		**65**
2001-2003 births[$$]				**48**		**66**		**60**

[**] Exclusions (not mutually exclusive): children who died before the age of 5 years, with submucous cleft palates, missing one or more of all 16 CAPS-A data items, born with either a CL or a non-specified cleft type, and syndromic children. [$] Based on the national outcome resulting from statistical analysis on 2004-06 Speech Outcome data – completed by the Lead Speech and Language Therapy Group, with statistical support from the Cleft Collective in Manchester, and presented in April 2014 to the Leads group. [$$] Britton L, Albery L, Bowden M, Harding-Bell A, Phippen G, and Sell D(2014) A Cross-Sectional Cohort Study of Speech in 5-year-Olds With Cleft Palate ± Lip to Support Development of National Audit Standards: Benchmarking Speech Standards in the United Kingdom. The Cleft Palate-Craniofacial Journal: July 2014, Vol. 51, No. 4, pp. 431-451.

Table B. Number (%) of CRANE-registered consented children born in 2010 - 2012 with reported speech outcomes at 5 years of age, according to Regional Centre / Administrative Unit and cleft type

Regional Centre	Admin Unit	Reported[**]	Cleft palate n	(%)	Unilateral cleft lip and palate n	(%)	Bilateral cleft lip and palate n	(%)
Northern & Yorkshire	Newcastle	90	56	62.2	22	24.4	12	13.3
	Leeds	96	44	45.8	35	36.5	17	17.7
North West & North Wales	Liverpool	72	42	58.3	20	27.8	10	13.9
	Manchester	117	53	45.3	40	34.2	24	20.5
Trent	Nottingham	133	71	53.4	45	33.8	17	12.8
West Midlands	Birmingham	146	67	45.9	53	36.3	26	17.8
East	Cambridge	91	43	47.3	33	36.3	15	16.5
North Thames	GOSH & Chelmsford	140	85	60.7	41	29.3	14	10.0
The Spires	Oxford & Salisbury	123	66	53.7	45	36.6	12	9.8
South Wales & South West	Swansea	69	37	53.6	25	36.2	7	10.1
	Bristol	93	54	58.1	29	31.2	10	10.8
South Thames	Guy's and St Thomas'	131	54	41.2	53	40.5	24	18.3
Northern Ireland	Belfast	45	22	48.9	15	33.3	8	17.8
All 2010-2012 births		**1,346**	**694**	**51.6**	**456**	**33.9**	**196**	**14.6**

[**] Exclusions (not mutually exclusive): children who died before the age of 5 years, with submucous cleft palates, missing one or more of all 16 CAPS-A data items, born with either a CL or a non-specified cleft type, and syndromic children. [$] Based on the national outcome resulting from statistical analysis on 2004-06 Speech Outcome data – completed by the Lead Speech and Language Therapy Group, with statistical support from the Cleft Collective in Manchester, and presented in April 2014 to the Leads group.

Figure A. Funnel plot showing the proportion of children with a cleft affecting the palate who have speech outcomes reported who are classified as Cleft Palate cases

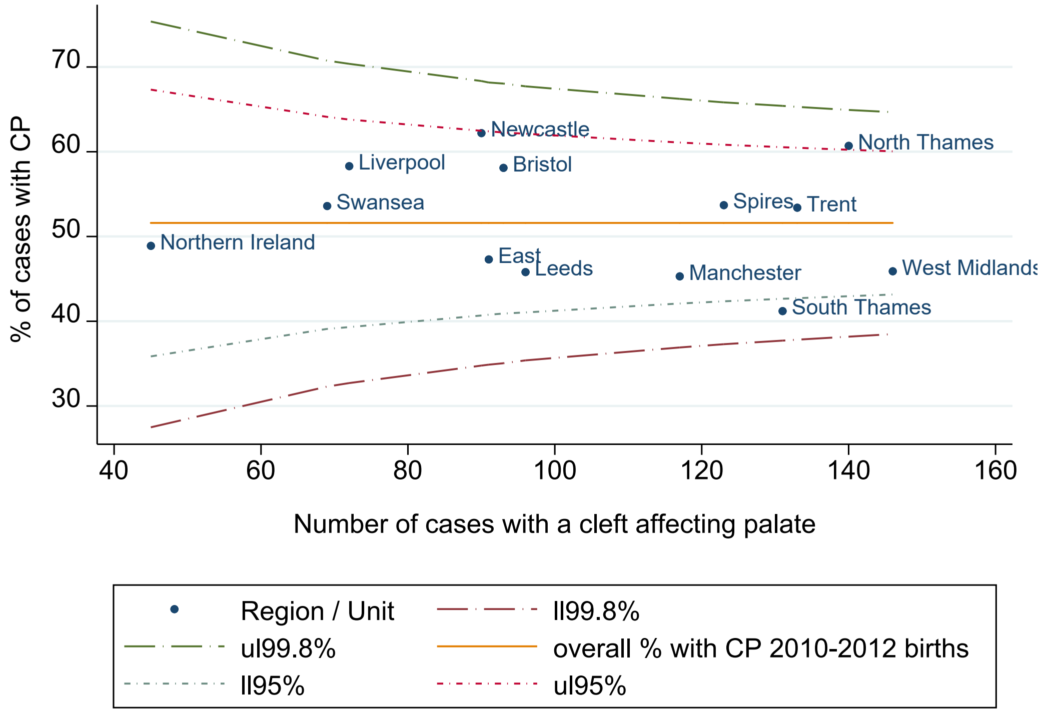


Figure B. Funnel plot showing the proportion of children with a cleft affecting the palate who have speech outcomes reported who are classified as Unilateral cleft lip and palate cases

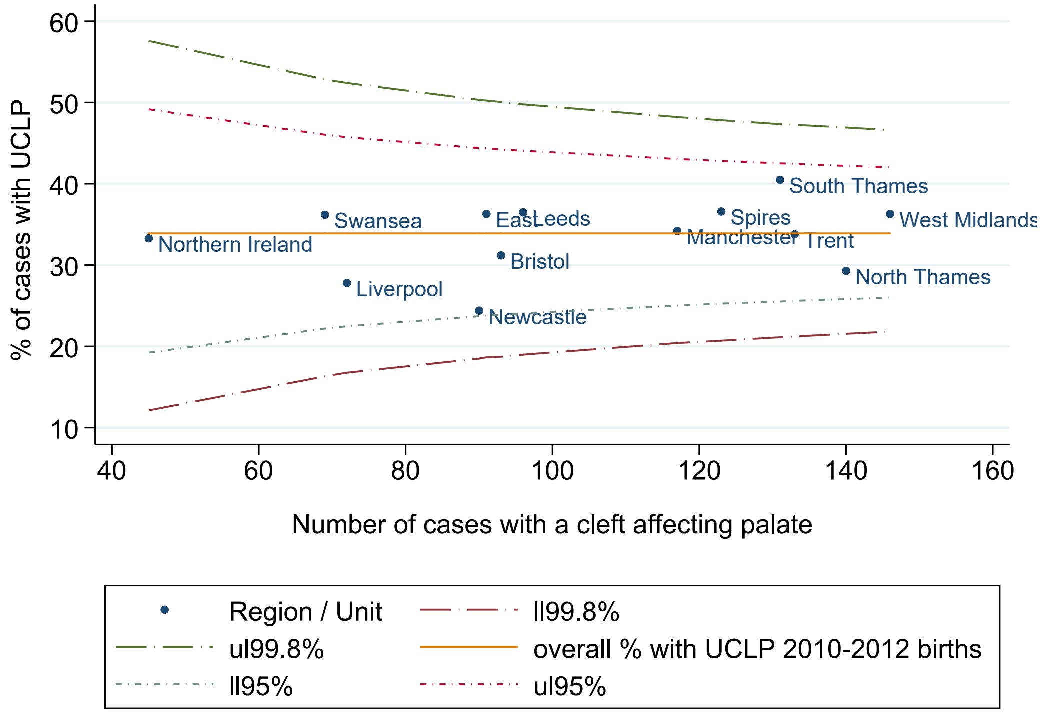
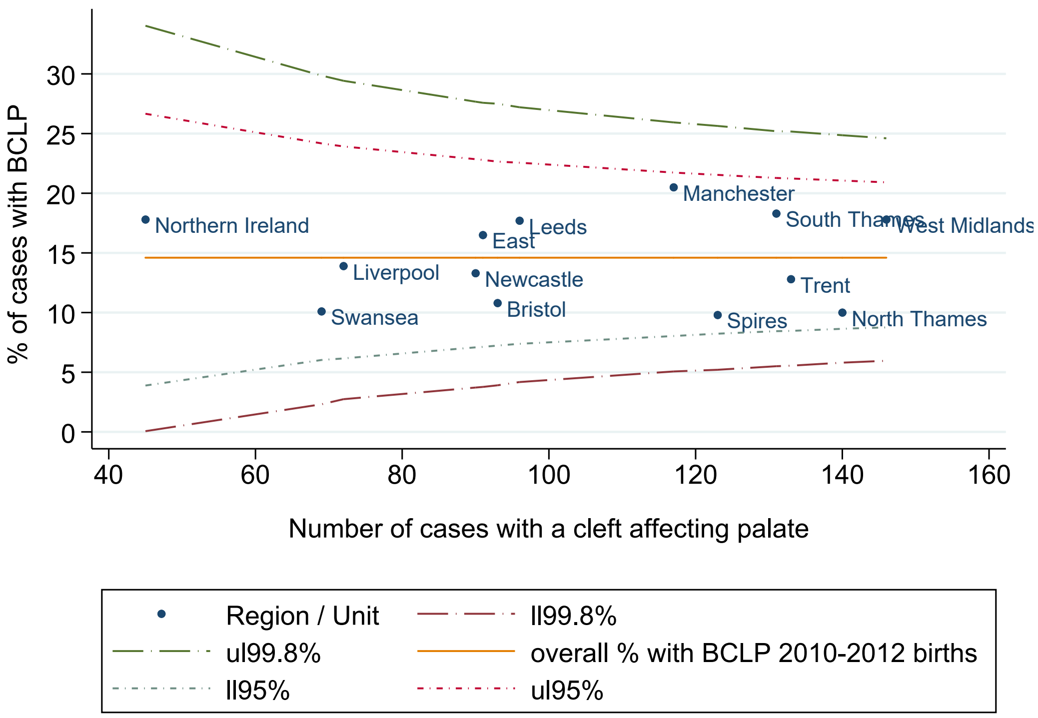


Figure C. Funnel plot showing the proportion of children with a cleft affecting the palate who have speech outcomes reported who are classified as Bilateral cleft lip and palate cases



Appendix 9: Psychology screening scores detail

Data completeness

Number (%) of CRANE-registered consented children born with a cleft lip or palate (2011 and 2012), with psychology outcome data or reasons this outcome was not collected at 5 years of age, according to region / unit.

		Psychology***							
Regional centre / MCN	**Administrative Unit**	**Eligible cases**	**Outcome collected: At least 1 of 9 items**		**Outcome not collected: reason provided**		**Total cases acc. for**	**Missing Data**	
		N	**n**	**(%)**	**n**	**(%)**	**(%)**	**N**	**(%)**
Northern	Newcastle	101	83	82.2%	18	17.8%	100%	0	(0)
& Yorkshire	Leeds	110	83	75.5%	27	24.5%	100%	0	(0)
North West	Liverpool	103	78	75.7%	11	10.7%	86.4%	14	(13.6)
& North Wales	Manchester	129	74	57.4%	14	10.9%	68.2%	41	(31.8)
Trent	Nottingham	159	0	0%	68	42.8%	42.8%	91	(57.2)
West Midlands	Birmingham	191	164	85.9%	25	13.1%	99%	2	(1)
East	Cambridge	108	84	77.8%	23	21.3%	99.1%	1	(0.9)
North Thames	GOSH & Chelms.	261	236	90.4%	18	6.9%	97.3%	7	(2.7)
The Spires	Oxford & Salisbury	170	105	61.8%	47	27.6%	89.4%	18	(10.6)
South Wales	Swansea	81	74	91.4%	2	2.5%	93.8%	5	(6.2)
& South West	Bristol	111	76	68.5%	21	18.9%	87.4%	14	(12.6)
South Thames	Guy's and St Thomas'	160	110	68.8%	48	30%	98.8%	2	(1.3)
Northern Ireland	Belfast	70	17	24.3%	11	15.7%	40%	42	(60)
All	**All**	**1,754**	**1,184**	**67.5%**	**333**	**19%**	**86.5%**	**237**	**(13.5)**

*** Exclusions (not mutually exclusive): No consent, children who died before the age of 5 years, with submucous cleft palates, and syndromic children.

Relationship between TIM and SDQ scores

Number (%) of CRANE-registered consented children born with a cleft lip or palate (2011 and 2012), within each of the Tiers of Involvement Measure (TIM) levels according to SDQ Total difficulties bands.

	TIM n (%)												
SDQ	0		1a	1b	2a	2b	2c	3		4.		**All**	
Close to aver.	29	(4.2)	396 (56.8)	7 (1)	144 (20.7)	39 (5.6)	46 (6.6)	25	(3.6)	11	(1.6)	**697**	**(100)**
Slightly raised	5	(7.2)	28 (40.6)	3 (4.3)	9 (13)	9 (13)	5 (7.2)	4	(5.8)	6	(8.7)	**69**	**(100)**
High	9	(15)	20 (33.3)	2 (3.3)	6 (10)	4 (6.7)	7 (11.7)	6	(10)	6	(10)	**60**	**(100)**
Very high	14	(16.7)	18 (21.4)	10 (11.9)	11 (13.1)	4 (4.8)	9 (10.7)	6	(7.1)	12	(14.3)	**84**	**(100)**
All	57	(6.3)	462 (50.8)	22 (2.4)	170 (18.7)	56 (6.2)	67 (7.4)	41	(4.5)	35	(3.8)	**910**	**(100)**

Exclusions (not mutually exclusive): No consent, children who died before the age of 5 years, with submucous cleft palates, and syndromic children.