Promoting excellence in cleft care

CLEFT REGISTRY & AUDIT NETWORK

Cleft Registry and Audit NEtwork Database

Part of the Clinical Effectiveness Unit, of the Royal College of Surgeons of England

2020 Annual Report

Results of the audit in England, Wales and Northern Ireland for children
born with a cleft between January 2000 and December 2019

On behalf of the Cleft Development Group

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

The CRANE project team at the Clinical Effectiveness Unit, of the Royal College of Surgeons of England

Jibby Medina, Research Fellow
Kate Fitzsimons, Research Fellow
Craig Russell, Clinical Project Lead
Hussein Wahedally, Data Manager
Min Hae Park,   Assistant Professor
Jan van der Meulen, Clinical Epidemiologist

Clinical Effectiveness Unit
The Royal College of Surgeons of England
Website: www.CRANE-Database.org.uk
Email: crane@rcseng.ac.uk
Telephone: 020 7869 6639

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Craniofacial Society of Great Britain on Ireland (CFSGBI) enabling the best Cleft and Craniofacial Care.

For further information, please refer to the website
http://craniofacialsociety.co.uk/

Registered charity no: 1092782

The Cleft Lip and Palate Association (CLAPA) is a small charity working to improve the lives of people born with a cleft and their families in the UK. We tackle isolation by connecting the cleft community, and provide personal support and accessible information at all stages of the cleft journey. We empower parents and patients to have an impact on cleft care through participation in research, and act as the leading voice of people affected by cleft in the UK. Our vision is a society where everyone affected by cleft feels informed, connected, reassured and empowered. Visit www.clapa.com to find out more.

Registered charity no: 1108160 (England & Wales) & SC041034 (Scotland)

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Acknowledgements

We would like to thank the children, their families and the cleft services in England, Wales and Northern Ireland for the collection and submission of data into the Cleft Registry & Audit NEtwork (CRANE) Database.

We would also like to extend our thanks to the following people:

- members of the UK NHS Cleft Development Group for their advice and contributions in writing this report;
- Jackie Smallridge for her contribution towards the interpretation of the dental outcome data at 5 years of age – on behalf of the Dental Clinical Excellence Network (CEN);
- Lorraine Britton for her contribution towards the interpretation of the speech outcome data at 5 years of age – on behalf of the Speech and Language Therapy CEN; and
- Vanessa Hammond for her contribution towards the interpretation of the psychology outcome data at 5 years of age – on behalf of the Psychology CEN.

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.

Foreword

It has been a challenging time to join the CRANE team. In fact, it has been a challenging year for all.

When I formally took over Clinical Project Lead responsibility for the CRANE Database at the start of the year, most were just beginning to hear about the new COVID-19 virus that was affecting Wuhan Province, Central China. Little did I (and I assume most of you) realise that our lives would be forever changed. 2020 will be remembered as the year when we began to understand what the word 'pandemic' really means. We have all experienced restrictions in our daily activity, restrictions in our civil liberties, and many of us will know someone who has experienced, or have experienced ourselves, the loss of friends, family or loved ones to the virus.

For CRANE, 2020 was supposed to have been our coming of age. This was to be our 21st year of existence and we were predicting breaking the 21,000 patient registrations mark (20,946 children had been born in the UK with a cleft and registered on CRANE between 1st of January 2000 and 31st of December 2019). Instead of being able to share and celebrate these milestones with clinical colleagues in the spring at the Craniofacial Conference in Cardiff, we were all 'confined to barracks' as a UK wide lockdown was put in place.

With Cleft Development Group (CDG) approval, CRANE decided to alter its approach to conduct a review of the most recent 10 years of registrations, outcomes and findings; rather than comparing cleft care teams. In line with this approach, this year we decided to separate reporting CRANE activity into three parts: Registry, Outcomes and Research. Much time this year has been spent investigating modulators of risk relating to 5-year-old speech and dental outcomes. The results of these investigations form the basis of the research section of the report. However, this section does not detail all research and development activity at CRANE. The team has made many changes to the database including a new and improved website with increased search functionality. The team also continues to investigate and publish in the area of educational attainment. With increased understanding of the impact of having a cleft in this area and the potential for improved data linkage activity, CRANE hopes to be in the position to incorporate educational attainment (as a more holistic measure of cleft care outcome) into future annual reporting activity.

While this year has not exactly been what I was expecting for my start with the CRANE team, it has been hugely interesting, challenging and most of all professionally very satisfying. I have joined a fantastic team (with equally energetic partners) who have risen to the challenges presented by the virus, overcome many, circumnavigated others, and made a real success of the opportunity that restrictions to normal living and working have necessitated. I am proud of their response to these turbulent times and extremely appreciative of all the sacrifices they have made to bring this report to you. I hope you all find its contents as stimulating and thought provoking as I have.

Craig J. H. Russell
Clinical Project Lead,
The CRANE Database

Executive summary

Annual Report 2020

CLEFT REGISTRY & AUDIT NETwork

CRANE is a national registry and clinical audit. It aims to 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. This year's report focuses on data collected over the last ten years.

Registry Information

20,946 The total number of children registered in CRANE, over the last 20 years, since 2000.

1,077 Average number of registrations per year, over the last 10 years, since 2010.

12% of babies with a cleft were born prematurely (<37 weeks). This compares to 7% in the general population.

Cleft palate diagnosis times are improving, but almost one quarter are still missed in the first 24 hours after birth.

80% of families were referred to a cleft team within 24 hours of birth and 94% of families received contact within 24 hours of referral.

98% of families with verified consent status agreed to the collection of outcome data.

Audit Outcomes at 5 years of age
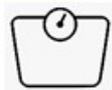
Body mass index

86% of children with a cleft had a healthy BMI. Rates of overweight and obesity were highest among children with CL.

Dental health

41% of children with a cleft had at least one decayed, missing or filled tooth (dmft>0), and 14% had at least six dmft (dmft>5).

Facial growth

41% of children with a complete unilateral cleft lip and palate (UCLP) had scores reflecting good dental arch relationships.

Speech

60% of children with a cleft affecting the palate had speech within the normal range. The proportion of children with 'normal' speech varied considerably according to cleft type.

Psychology

18% of children with a cleft had high or very high 'Strengths and Difficulties' scores*. This compares with 10% among their non-cleft peers.

*'High' and 'very high' scores indicate a greater level of difficulties than low scores, which are classified as being in the 'close to average' range (indicating no concern).

Research

What is the incidence of cleft lip and/or palate in England, Wales and Northern Ireland?

15 per 10,000 live births

This is the estimated mean incidence of clefting across England, Wales and Northern Ireland.

Is speech affected by the timing and number of primary palate repairs?

The most favourable speech outcomes were observed among children undergoing their last primary palate repair before 13 months of age. An increasing number of repairs was associated with poorer speech outcomes.

Is dental health associated with deprivation among children with a cleft?

Rates of dmft>0 and dmft>5 increased with increasing deprivation.
Treated dental disease and care index scores decreased with increasing deprivation.

For further Information visit www.CRANE-Database.org.uk | @CRANE_News

Key findings and recommendations

Findings	Recommendations
REGISTRY	
Registrations in CRANE (Chapter 3, Section 3.1)	
• Over the last ten years, there have been an average of 1,077 new registrations per birth year. • Cleft Palate (CP) is the most common type of cleft, representing 45% of all cases with a known cleft type, followed by cleft lip (CL) (24%), unilateral cleft lip and palate (UCLP) (22%) and bilateral cleft lip and palate (BCLP) (10%).	1. CRANE will engage with the nursing Clinical Excellence Network (CEN) to ensure better assessment and recording of registrations. 2. CRANE will continue to record cleft births and validate case ascertainment using external datasets.
Timely diagnosis (Chapter 3, Section 3.2 & 3.3)	
• Over the last ten years, rates of antenatal diagnosis for CL, UCLP and BCLP have shown significant improvement. • Over the last ten years, significant improvement has been observed in the timely detection of CPs, particularly in the last 5 years. However almost 1 in 4 children with CP still have a diagnosis beyond 24 hours from birth.	3. CRANE will seek to collaborate further with clinical and non-clinical partners to identify ways of improving the timely diagnosis of CP within 24 hours of birth.
Gestational age and birth weight (Chapter 3, Section 3.4)	
• Over the last ten years, the mean gestational age of babies born with a cleft was 38.7 weeks. • 12% of babies were born prematurely (<37 weeks' gestation). This compares to 7% in the general population. • Children with cleft lip had a higher mean birth weight (3,296 g) than those with a cleft affecting the palate (3,154 g).	4. The research community should validate and further investigate the apparent association between cleft-affected pregnancies and prematurity at birth. 5. CRANE will engage with the RCM and Royal College of Obstetricians and Gynaecologists (RCOG) to communicate our findings.
Contact with cleft care teams (Chapter 3, Section 3.5)	
• Over the last ten years, 80% of families were referred to a cleft care team within 24 hours of birth. This rate varied significantly across cleft types. • 94% of families were contacted by a cleft care team within 24 hours of referral.	6. Cleft care teams should continue to work with referring obstetric, midwifery and neonatal units to improve timeliness of diagnosis and early referrals. 7. Cleft care teams with high levels of referrals within 24 hours should share their best practise recommendations.
CRANE Consent (Chapter 3, Section 3.6)	
• Over the last ten years, consent status was verified for 79% of families approached. This meant they had agreed or declined to their child's outcome data being recorded in CRANE. • The verified consent rate has fallen in recent years. • Of the families who had reached a decision, rate of agreed consent was 98%.	8. Cleft care teams should review their procedures to identify reasons for low rates of verified consent, and ensure that CRANE consent status is recorded for every child with a cleft. 9. CRANE will continue to work with cleft care teams and the Nursing CEN to improve consent status verification.
OUTCOMES	
Child growth at 5 years (Chapter 4, Section 4.1)	
• 86% had a healthy Body Mass Index (BMI). • The prevalence of overweight and obesity was found to be higher among children with CL than among those with a cleft affecting the palate. • The prevalence of underweight was higher among children with CP (2.5%) than among those with other cleft types.	10. Cleft care teams should aim to assess children's weight and height around the age of five years and improve the reporting of these measures in the CRANE database. This will facilitate more meaningful comparisons between subgroups in the future.
Dental health at 5 years (Chapter 4, Section 4.2)	
• Dental decay was experienced by 41% of children (having at least one decayed, missing or filled tooth (dmft >0)) and 14% of children were classified as having extensive caries (dmft >5). These rates increased significantly as cleft type became more involved. • The average Treatment Index (rate of treated disease) was 76%, and the average Care Index (having received the	11. All children with a cleft should have a recommended care plan established by collaborative work between the families' General Dental Practitioner (GDP) and cleft care teams to: (a) treat the child as per the high-risk category of the dental toolkit (delivering better oral health), (b) provide local dental care (GDP led), and (c) provide

appropriate care at the earliest possible stage) was 69%. These rates decreased significantly as cleft type became more involved.	age-specific dental development assessments and advice (cleft care team led).
Facial growth at 5 years (Chapter 4, Section 4.3)	
• 41% of children with complete UCLP had Five Year Old Index scores reflecting 'good' dental arch relationships. • Dental arch relationships remained stable over time.	12. The research community should undertake to compare UK facial growth outcomes with those in other countries.
Speech at 5 years (Chapter 4, Section 4.4)	
For children born with a cleft affecting the palate: • 60% achieved speech within the normal range. • 71% had speech without 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% had speech without cleft-related articulation difficulties. • These rates vary significantly according to cleft type. Children with a cleft affecting only the palate tend to fare better, while those with a bilateral cleft lip and palate tend to have the least favourable outcomes. • The proportion of children meeting the standards has not changed significantly over time.	13. Information given to parents by cleft care teams about expected speech outcomes should take into account the child's cleft type. 14. The research community should undertake to develop risk stratification models for analysing speech outcomes among children with a cleft.
Psychology screening at 5 years (Chapter 4, Section 4.5)	
• 96% of families were screened at least once before the target age of 6 years. • 86% were seen by a psychologist and a psychosocial screen was completed (TIM tiers 1 to 4). • 18% of children born with a cleft had 'high' or 'very high' SDQ scores. These rates are higher than the population norms.	15. Cleft care teams should aim to see all children and families at age 5, undertake a psychological screen and ensure psychological support is provided if appropriate (to be recorded as a TIM score).
RESEARCH	
Incidence of cleft (Chapter 5, Section 5.1)	
• The overall estimated incidence of cleft between 2010 and 2019 was 15.0 per 10,000 live births. • CP was the most common cleft type (estimated incidence for the period 6.3 per 10,000 live births). • Estimated Incidence of cleft was higher among girls than boys over the investigated period (17.0 per 10,000 live female births vs. 13.0 per 10,000 live male births)	16. The research community should undertake to validate these findings using the HES dataset and case ascertainment techniques.
Timing of repair, number of surgeries and speech at 5 years (Chapter 5, Section 5.2)	
The most favourable cleft-related speech outcomes were observed among children undergoing: • their last primary palate repair before 13 months of age, and • just one primary palate repair procedure on the palate. An increasing number of palate repair surgeries is associated with poorer outcomes.	For the most favourable speech outcomes: 17. Children should have their palate repaired by cleft surgeons before they are 13 months old. 18. Where possible, surgeons should aim to repair the cleft palate in one procedure. 19. The research community should validate these findings whilst controlling for potential confounders.
Deprivation and dental health at 5 years (Chapter 5, Section 5.3)	
• Rates of poor dental health (expressed by rates of dmft >0 and dmft >5) increased significantly as deprivation increased. • Treatment Index and Care Index scores increased significantly as deprivation decreased.	20. Cleft care teams should have mechanisms in place to identify and target children from the most deprived groups to ensure they receive help, such as Sure Start, to facilitate their access to appropriate dental care and advice.
DATA QUALITY – throughout report	
• There was significant variation in data completeness for registrations and also for specialty-specific outcomes across the period analysed for this report. • The COVID-19 pandemic has significantly affected some specialties' ability to submit 5-year-old outcome data for children born in 2013.	21. Cleft care teams, Clinical Excellence Networks (CENs) and CRANE should work together to identify barriers to collecting and submitting data. 22. Cleft care teams should pay particular attention to assessing outcomes and reporting these to CRANE.

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. Negotiations are ongoing with NHS Scotland and it is hoped that they will join the CRANE family in 2021. The geographical representation of the cleft regions / cleft care teams 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) data 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 for 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 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 1 January 2004 and 31 December 2019, and it has the following three main sections:

- **Registry Information:** This section details epidemiological findings, diagnosis times, early contact care information including timing of referral to and contact by cleft care teams, and consent status.
- **Audit Outcomes at 5 years of age:** This section details cleft-related outcomes for children, registered in the CRANE Database, at five years of age (born 2004-2013).
- **Research:** This section details the research and development activity undertaken by the CRANE team over the last 12 months.

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

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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 13 July 2020.

2. Methods

This report contains information on patterns of care and outcomes derived from two sources of data: (1) the CRANE Database, and (2) Hospital Episode Statistics (HES) data linked to CRANE 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 child's birth, demographics, type of cleft, time of diagnosis, time of referral to a cleft care team, and time of first contact between a patient and cleft care team. The CRANE Database also collects information about cleft-related treatment and outcomes. These data are reported to CRANE by the cleft care teams 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 cleft care teams 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 being treated by the specialist cleft care teams. 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 cleft care teams at some point between referral and the first primary repair. A coordinator within each cleft care team 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. CRANE cohort

All data entered into the CRANE Database by 13 July 2020 is included in the descriptions and analyses described in this Annual Report. Children 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) gestation and weight at birth, (2) 5-year outcomes, and (3) HES analyses (as the data presented in these sections and tables are not collected for non-consenting cases).

The children and timeframes covered in each chapter, and sub-section, are indicated in the summary tables at the beginning of each section (where relevant). Broadly, timeframes are the most recent ten years of available data:

- Registry Information is reported for children born between 1 January 2010 and 31 December 2019 (Chapter 3).
- Audit Outcomes at 5 years of age are reported for children born between 1 January 2004 and 31 December 2013 (Chapter 4) – with the exception of speech (2007-2013 births) and psychology (2011-2013 births) outcomes.

- CRANE Research is reported for children born between 1 January 2004 and 31 December 2019, as appropriate to the analysis in each of the three sub-sections (Chapter 5).

For full details of the methods used within this report, please see the CRANE Annual Report Methodology 2020 document, available online alongside this Annual Report (https://www.crane-database.org.uk/reports/crane-database-2020-annual-report/).

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 and Figures relating to data completeness (see Appendix 5 for a breakdown of those reported for each outcome).

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 may be used to identify any additional congenital anomalies and syndromes diagnosed for the CRANE cohort (see Appendix 4 for a list of the HES diagnosis and procedure codes used by CRANE). We use HES to identify whether a child should be classed as 'non-syndromic' or 'syndromic' for CRANE-HES linked research only.

3. Registry Information

This chapter details epidemiological findings and early contact care information, for children registered in the CRANE Database with a cleft lip and/or palate. This is key information for cleft care planning.

The sections in this chapter report on registrations, timing of diagnosis, gestation and weight at birth, and timing of referral to and contact with cleft care teams around the time of birth.

Timeframe: The most recent ten years of data – children born between 1 January 2010 and 31 December 2019.

Legal basis for data collection and analysis: The data used for this section is collected for all registered cases under a 'Section 251' exemption (of the NHS Act 2006 and its current regulations, the Health Service (Control of Patient Information (CPI)) Regulations 2002), with approval from the Confidentiality Advisory Group (CAG) for the disclosure of CPI held by the CRANE Database.

Gestation and birth weight is an exception to this exemption and is collected for all children whose families have given informed consent to outcomes data collection by the CRANE Database.

3.1. Registrations in CRANE

Cohort summary	Data source	CRANE Database
	Birth years	Ten years: 2010 to 2019
	Denominators	• 10,765 children registered in the CRANE database • 10,093 children with a record of cleft type • 10,595 children with a record of sex
	Numerator	Number of children meeting each patient characteristic
	Data completeness	• 94% of 10,765 registrations had a record of specified cleft type. • 98% of 10,765 registrations had a specified sex.
	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 (CP) was the most common form of cleft, representing 45% of all cases with a known cleft type, followed by cleft lip (CL) (24%), unilateral cleft lip and palate (UCLP) (22%) and bilateral cleft lip and palate (BCLP) (10%), respectively. 6.2% of children had an unspecified cleft type. • 55% of children were boys. They were more likely to have a CL, UCLP or BCLP than girls with a cleft. 1.6% of children did not have their sex specified.	
Recommendations	• Cleft care teams should aim to register all children with a cleft in the CRANE database. Cleft type and sex should be reported for every child registered. • CRANE will engage with the nursing Clinical Excellence Network (CEN) to ensure better assessment and recording of registrations. • CRANE will continue to record cleft births and validate case ascertainment using external datasets.	

3.1.1. Data completeness for sex and cleft type

A total of 10,765 children were born with a cleft and registered in the CRANE Database over the last ten years[4]. During this period, the mean number of children registered in CRANE Database was 1,077 per birth year (range: 897 to 1,168 children).

Figure 3.1. Percentage of CRANE-registered children with recorded cleft type and sex, by birth year.

	2010	2011	2012	2013	2014	2015	2016	2017	2018	2019
Cleft type	98.5%	95.8%	95.6%	97.4%	94.7%	91.8%	89.0%	90.6%	91.4%	92.1%
Sex	99.7%	97.0%	98.2%	99.1%	97.4%	98.1%	98.4%	99.0%	98.1%	99.2%
Total (N)	1,062	1,107	1,168	1,167	1,073	1,119	1,073	1,081	1,018	897

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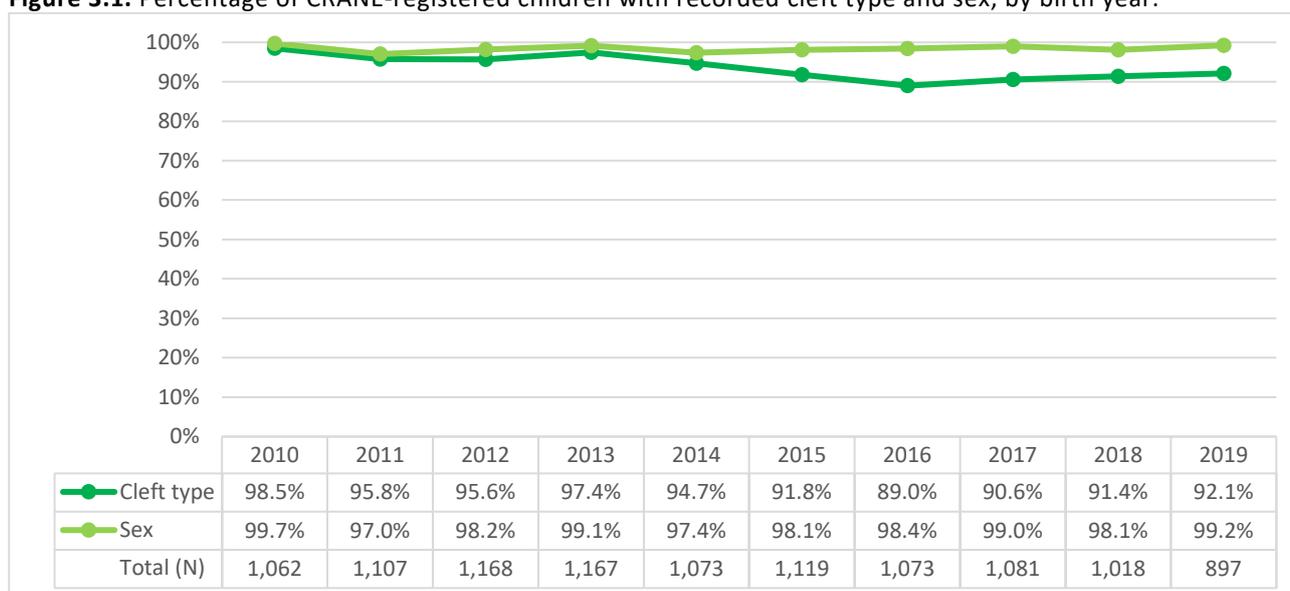
[4] Cohort includes all patients registered in CRANE Database between 1st January 2010 and 31st December 2019, inclusive.

Overall, 10,093 (93.8%) children had their type of cleft reported and 10,595 (98.4%) had their sex reported. Data completeness for cleft type varied by year of birth (p<0.001). Figure 3.1. shows that the latter five years had higher levels of missing data compared to the earlier five years. The reporting of sex was consistent over time.

3.1.2. Registrations by cleft type and sex

Over the last ten years, the percentage of children across each cleft type remained stable over time (Figure 3.2). CP was the most common of the four cleft types[5], representing 44.6% of all registrations with a known cleft type (n=10,093), followed by CL (23.9%), UCLP (21.7%) and BCLP (9.8%). There were 672 children (6.2%) with a non-specified cleft type. Of the registered UCLPs (n=2,888), 72% were complete, while 57% of all BCLPs (n=993) were complete.

Figure 3.2. The distribution of cleft types among CRANE-registered children, by birth year.

	2010	2011	2012	2013	2014	2015	2016	2017	2018	2019
CL	23.9%	24.2%	22.8%	25.3%	24.6%	24.8%	24.3%	21.8%	21.8%	25.3%
CP	44.5%	45.9%	45.3%	44.9%	44.8%	44.4%	44.6%	44.3%	43.3%	43.5%
UCLP	20.9%	21.5%	22.8%	20.4%	20.5%	22.3%	21.2%	22.5%	23.2%	21.7%
BCLP	10.7%	8.4%	9.0%	9.4%	10.1%	8.5%	9.9%	11.4%	11.6%	9.6%
Total (N)	1,046	1,060	1,117	1,137	1,016	1,027	955	979	930	826
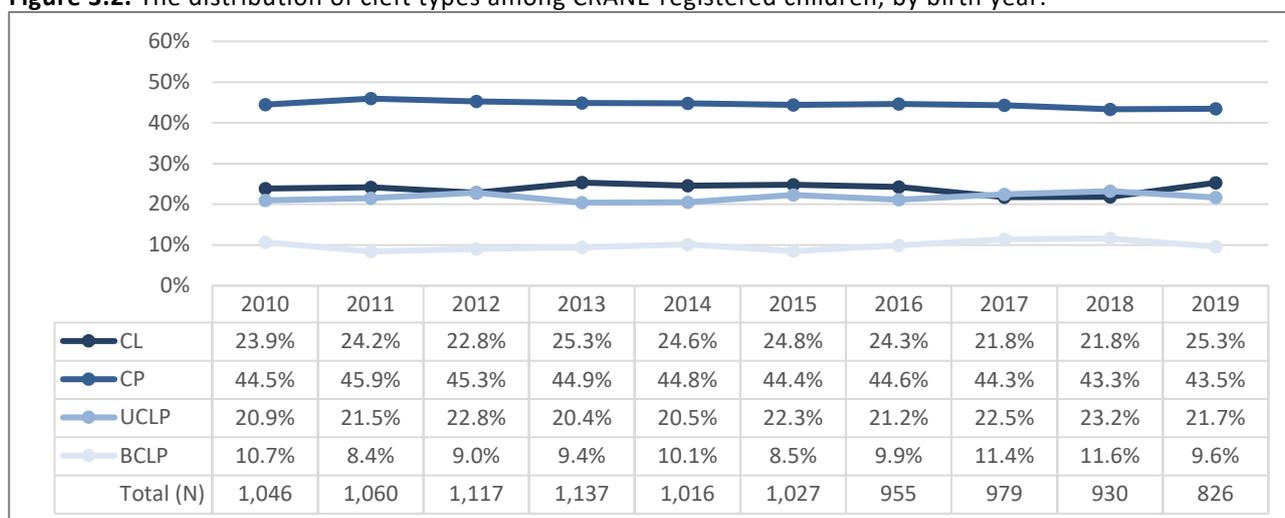
Note: Children without a known cleft type are excluded.

Overall, more boys (56.0%) than girls (44.0%) were registered in the CRANE Database between 2010 and 2019. The sex ratio has remained relatively stable over time, with the exception of births in 2014, when boys represented 58.8% of registrations and girls represented 41.2% of registrations (see Figure 3.3).

Figure 3.3. Sex ratio* among CRANE-registered children, by birth year.

	2010	2011	2012	2013	2014	2015	2016	2017	2018	2019
Boys	55.1%	54.5%	54.3%	56.6%	58.8%	57.0%	56.1%	56.3%	55.4%	56.1%
Girls	44.9%	45.5%	45.7%	43.4%	41.2%	43.0%	43.9%	43.7%	44.6%	43.9%
Total (N)	1,059	1,074	1,147	1,157	1,045	1,098	1,056	1,070	999	890
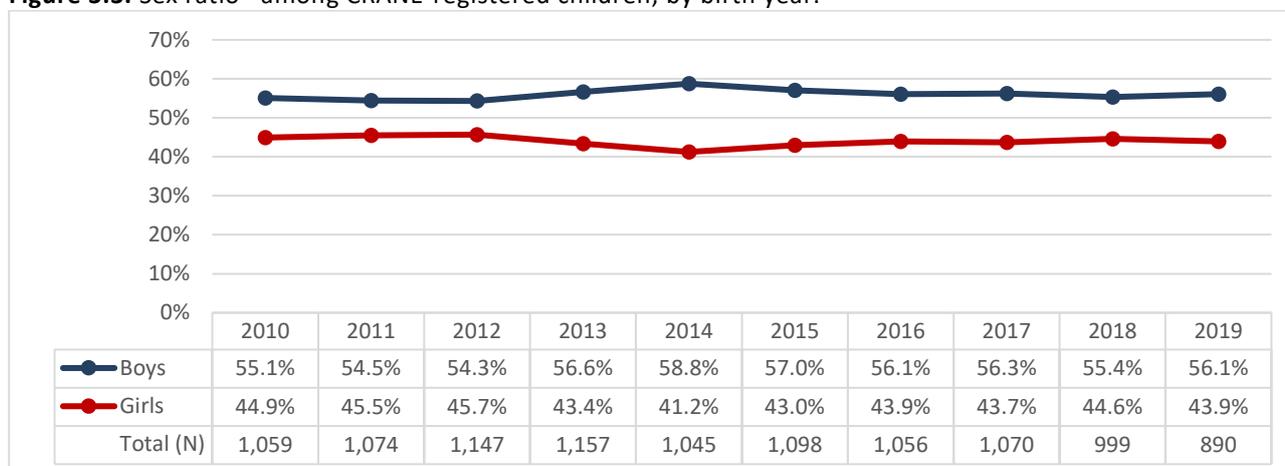
Note: Children missing sex information are excluded.

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[5] Cleft type is defined according to reported LAHSAL codes or, where LAHSAL has not been reported (for 6.2% of children registered between 2010 and 2019), it is based on the cleft type reported by the region / unit registering the child.

The sex ratio varied according to cleft type. On average, more boys had CL, UCLP and BCLP than girls, while more girls than boys had CP (Table 3.1).

Table 3.1. Sex ratio among CRANE-registered children born between 2010 and 2019, by cleft type.

Cleft Type	No. of Children	Boys		Girls	
		N	%	N	%
CL	2,393	1,487	(62.1)	906	(37.9)
CP	4,457	1,996	(44.8)	2,461	(55.2)
UCLP	2,175	1,459	(67.1)	716	(32.9)
BCLP	988	675	(68.3)	313	(31.7)
Total	10,013	5,617	(56.1)	4,396	(43.9)

Note: Children missing cleft type and sex information are excluded.

3.2. Timing of diagnosis for all cleft types

Cohort summary	Data source	CRANE Database
	Birth years	10 years: 2010 to 2019
	Denominator	9,968 children with a recorded diagnosis time
	Numerator	Number of children diagnosed at each time point
	Exclusion	Children without a recorded diagnosis time
	Data completeness	93% of 10,765 CRANE-registered children had a recorded diagnosis time.
	Countries	England, Wales and Northern Ireland
Standard	• 100% of children with a cleft should have a recorded diagnosis time. • 100% of clefts should be diagnosed in a timely manner, either antenatally (clefts involving the lip) or within 24 hours of birth.	
What did we find?	**Antenatal diagnoses** • Rates of antenatal diagnoses for CL, UCLP, BCLP have shown significant improvement over the last 10 years of registrations. There has been no significant change in the low rates of antenatal diagnoses of CP. **Timely diagnoses** • Overall, timely detection of babies' clefts has improved significantly over the last ten years. 84.8% of all babies born with a cleft in the last 10 years were diagnosed in a timely manner i.e. antenatally or within 24 hours of birth.	
Recommendations	• CRANE will seek to collaborate further with the medical workforce to identify barriers to recording timing of diagnosis for all registrations in CRANE. • CRANE will continue to monitor rates of antenatal and timely diagnoses to ensure issues are highlighted, as well as opportunities for learning and areas for improvement identified.	

3.2.1. Data completeness

Out of the 10,765 children born and registered in CRANE between 2010 and 2019, 92.6% of them had a recorded diagnosis time (range: 88.3% in 2011 - 97.2% in 2019). Figure 3.4 shows that data completeness of diagnosis time has improved steadily and significantly over time (p<0.001).

When individual cleft types are examined: 96.1% of CL, 93.5% of CP, 95.8% of UCLP and 95.9% of BCLP had a recorded diagnosis time. All four cleft types showed significant improvement in data completeness over time (p<0.001 for CL, CP and UCLP; p=0.003 for BCLP). This improvement has been greatest for children

born with CP (range: 88.7 in 2011 - 98.3% in 2018) and BCLP (89.3% in 2010 - 100% in 2019). Improvement has also been particularly noticeable in the most recent three years of CRANE data collection, for all cleft types (compared to 2016 rates). Cleft care teams should be commended for their commitment to this improvement.

Figure 3.4. Percentage of CRANE-registered children with recorded diagnosis time, by cleft type and birth year.

	2010	2011	2012	2013	2014	2015	2016	2017	2018	2019
Recorded diagnosis time (all)	90.5%	88.3%	92.8%	93.1%	92.6%	90.7%	89.6%	95.0%	97.2%	97.2%
CL	94.0%	93.4%	96.5%	95.5%	95.2%	96.9%	92.2%	99.5%	100.0%	99.5%
CP	88.8%	88.7%	94.1%	93.5%	95.2%	90.6%	93.4%	96.5%	98.3%	97.8%
UCLP	94.1%	91.7%	97.3%	95.7%	94.7%	96.1%	92.1%	98.2%	99.1%	98.9%
BCLP	89.3%	97.8%	96.0%	92.5%	98.1%	94.3%	95.8%	97.3%	99.1%	100.0%
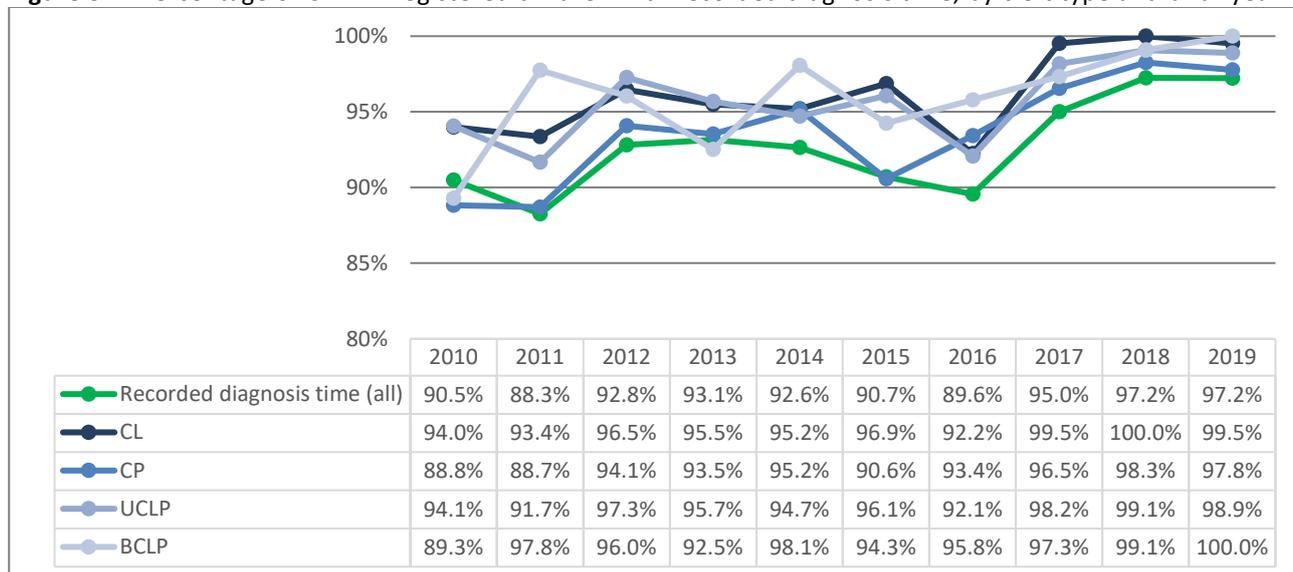
Note: Children missing diagnosis time and without a known cleft type are excluded.

3.2.2. Antenatal diagnosis of all cleft types

The variation in antenatal diagnoses by cleft type and birth year, over the last ten years, were explored and are presented in Figure 3.5. Rates of antenatal diagnoses of BCLP have shown particular and significant improvement, from 75% of cases in 2010 to 92.5% in 2018 (p=0.001). The increase in antenatal diagnoses for children with UCLP and CL was also statistically significant (p<0.001 and p=0.013, respectively).

Figure 3.5. Rates of antenatal diagnosis among CRANE-registered children, by cleft type and birth year.

	2010	2011	2012	2013	2014	2015	2016	2017	2018	2019
CL antenatal diagnosis	64.3%	59.8%	55.7%	63.3%	67.6%	67.6%	69.6%	69.3%	69.0%	69.2%
CP antenatal diagnosis	1.0%	0.9%	1.3%	1.7%	1.2%	0.5%	1.3%	2.9%	2.5%	2.0%
UCLP antenatal diagnosis	78.6%	84.2%	76.2%	82.0%	90.9%	87.3%	87.1%	88.9%	91.1%	87.6%
BCLP antenatal diagnosis	75.0%	77.0%	78.4%	72.7%	85.1%	85.4%	82.4%	86.2%	92.5%	92.4%
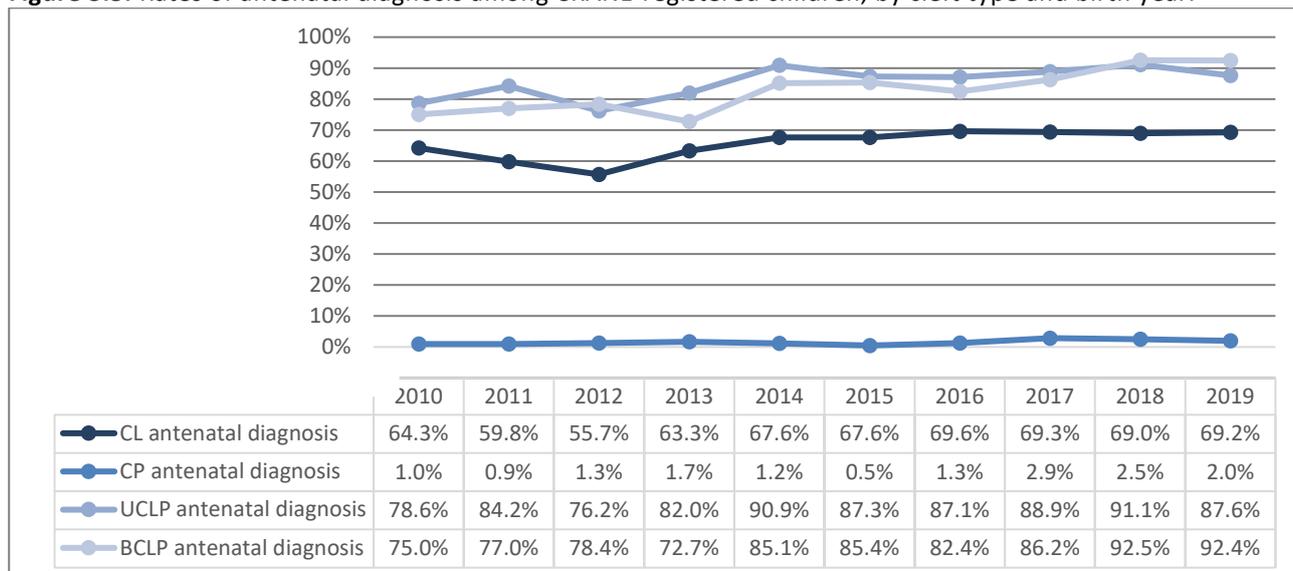
Note: Children missing diagnosis time and without a known cleft type are excluded.

There was no significant change in the low rates of antenatal diagnoses of CP. Clinically, the diagnosis of cleft palates antenatally is extremely difficult on standard 20-week anomaly ultrasound scans. As such we would not expect changes in antenatal diagnosis rates of CP over this reporting period. Over the last ten

years, antenatal diagnosis rates for CP were 1.5%, CL (65%), UCLP (85%) and BCLP (83%), indicating that the more involved the cleft type is (BCLP>UCLP>CL), the higher the rate of antenatal detection. Because of the importance of timely diagnosis of CPs, the next section in this chapter focuses on CP diagnoses within 24 hours of birth.

3.2.3. Timely diagnosis of all cleft types

Overall, the majority of all babies born with a cleft in the last ten years were diagnosed in a timely manner (84.8%), either antenatally (42.7%) or within 24 hours of birth (42.1%). As shown in Figure 3.6, overall rates of timely diagnoses have shown significant improvement over the last ten years, from 81.8% in 2012 to 87.5% in 2019 (p=0.002).

Figure 3.6 also shows the variation in timely diagnoses of individual specified cleft types. Rates of timely diagnoses of UCLP have fluctuated significantly over the last ten years (p=0.037). Improvement appears to have taken place from 97.2% timely diagnoses in 2012 to 100% in 2015. Rates of timely diagnoses have not fallen below 97.7% since 2015. The variation in timely diagnoses for children with CL and BCLP were not statistically significant (p=0.055 and p=0.533, respectively). Timely diagnosis of CP is explored in the next section of this report.

Figure 3.6. Rates of timely diagnosis (antenatal and within 24 hours of birth) among CRANE-registered children, by cleft type and birth year.

	2010	2011	2012	2013	2014	2015	2016	2017	2018	2019
Timely diagnosis (all)	83.7%	84.6%	81.8%	83.3%	83.3%	84.8%	84.7%	87.3%	87.1%	87.5%
CL	96.2%	95.8%	91.1%	94.2%	94.5%	93.1%	97.2%	95.8%	95.1%	97.6%
CP	66.3%	68.8%	66.0%	66.7%	67.0%	70.7%	70.4%	74.7%	75.8%	75.8%
UCLP	98.5%	99.5%	97.2%	99.5%	99.0%	100.0%	98.9%	100.0%	99.5%	97.7%
BCLP	98.0%	98.9%	99.0%	100.0%	100.0%	98.8%	100.0%	100.0%	99.1%	97.5%
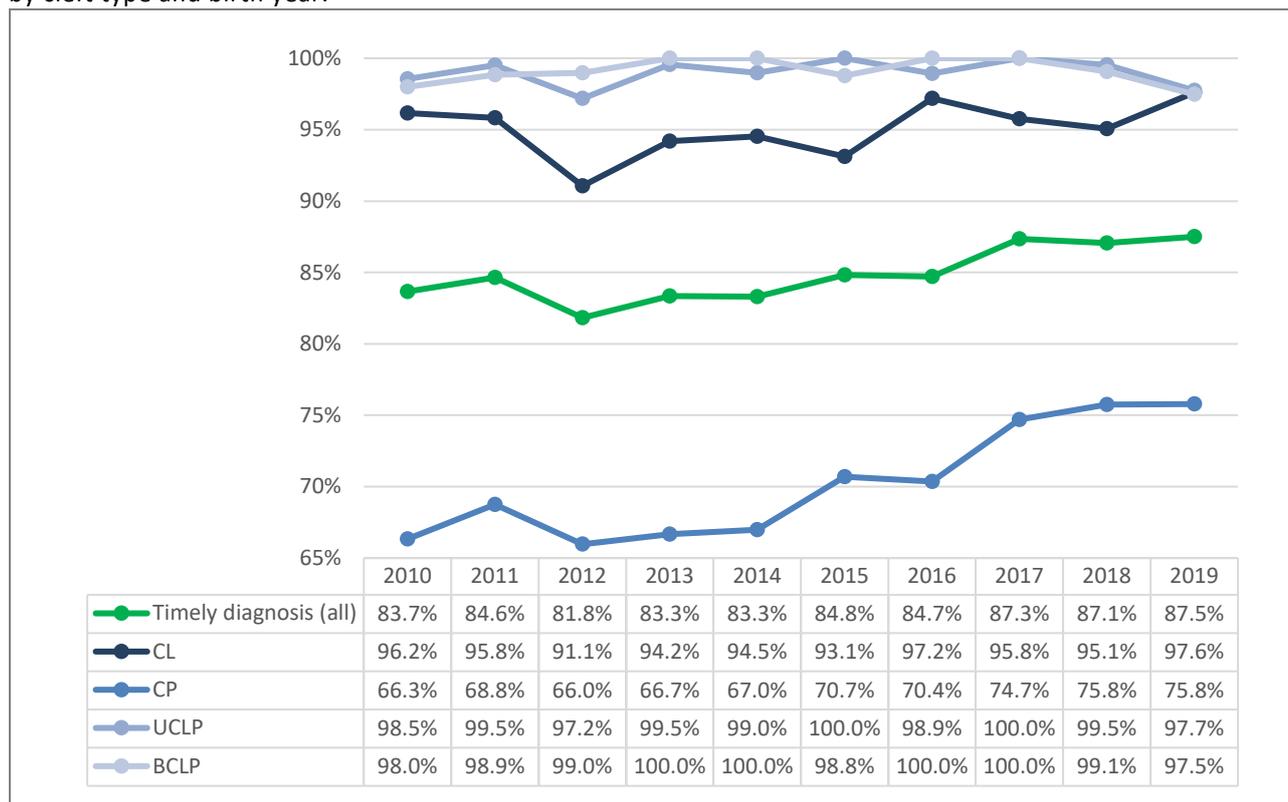
Note: Children missing diagnosis time are excluded.

3.3. Timely diagnosis of cleft palate

Cohort summary	Data source	CRANE Database
	Birth years	10 years: 2010 to 2019
	Denominator	4,208 children with cleft palate alone and a recorded diagnosis time
	Numerator	Number of children diagnosed at each time point with cleft palate alone
	Exclusions (not mutually exclusive)	• Children with CL, UCLP, BCLP or an unspecified cleft type • Children without a recorded diagnosis time
	Data completeness	94% of children born with CP (n=4,501) had a recorded diagnosis time.
	Countries	England, Wales and Northern Ireland
Standard	100% of cleft palates should be diagnosed in a timely manner, either antenatally or within 24 hours of birth.	
What did we find?	Over the last ten years, significant improvement has been observed in the timely detection of CPs, particularly in the last 5 years. However almost 1 in 4 children with CP still have a diagnosis beyond 24 hours from birth.	
Recommendations	CRANE will seek to collaborate further with clinical and non-clinical partners to identify ways of improving the timely diagnosis of CP within 24 hours of birth.	

Our 2012 Annual Report was the first to highlight the issue of diagnosis beyond 24 hours from birth among children with cleft palate (CP), reporting that nearly one third of children were diagnosed late according to the National Standard (for 2011 births)[6]. Because of this, we continue to investigate factors associated with a late CP diagnosis.

Figure 3.7 shows that there have been significant improvements in the rates of timely diagnosis of CP, either antenatally or within 24 hours of birth, over the last 10 years (p<0.001) with rates increasing from 66% for children born in 2012 to 76% for those born in 2019. This improvement commenced the year after CRANE first highlighted the issue of diagnosis beyond 24 hours after birth and was further helped by the publication of the Royal College of Paediatrics and Child Health (RCPCH) guidance '*Palate examination: identification of cleft palate in the newborn*'[7] in October 2014 (and its formal launch in 2015). This guidance was drafted in response to the CRANE findings and in collaboration with key partners including CRANE.

Exploration of the CRANE data not only demonstrated improvement over the full 10 years examined. It showed that mean diagnosis rates for the 5 years post introduction of RCPCH guidance were significantly better that the 5 years before (73.4% for 2015-19 births vs. 66.9% for 2010-14 births, p=0.001), indicating the value of audit findings informing best practice guidance with subsequent clinical benefit.

Despite the encouraging improvements in timely diagnosis rates, almost 1 in 4 children with CP continue to have a diagnosis beyond 24 hours from birth.

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[6] Bannister P. Management of infants born with a cleft lip and palate. Part 1. Infant, 2008. 4(1): p. 5-8.
[7] Published October 2014 https://www.rcpch.ac.uk/resources/palate-examination-identification-cleft-palate-newborn-best-practice-guide

Figure 3.7. Rates of timely diagnosis (antenatal and within 24 hours of birth) among CRANE-registered children with CP, by birth year.

	2010	2011	2012	2013	2014	2015	2016	2017	2018	2019
Diagnosed within 24hrs of birth	66.3%	68.8%	66.0%	66.7%	67.0%	70.7%	70.4%	74.7%	75.8%	75.8%
Diagnosed after 24hrs of birth	33.7%	31.0%	34.0%	33.0%	33.0%	29.0%	30.0%	25.0%	24.0%	24.0%
Total (N)	413	432	476	477	433	413	398	419	396	351
Diagnosed within 72hrs of birth						80.6%	80.9%	83.1%	86.1%	88.3%
Diagnosed after 72hrs of birth						19.4%	19.1%	16.9%	13.9%	11.7%

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

Note: Children missing diagnosis time are excluded.

Current RCPCH guidance is that full newborn examinations should be undertaken within 72 hours of birth[8]. This now includes a complete palate examination as per the 2014 palate examination document.

Figure 3.7 (above) demonstrates a significant improvement in the rate of CP diagnosis within 72 hours (88.3% in 2019 vs. 80.6% in 2015, p=0.015). This means that approximately 8 of every 9 live births with an isolated CP are now diagnosed within 3 days of birth. In comparing the two datasets – diagnoses within 24 hours and diagnoses within 72 hours – we see that the magnitude of increase over the last 5 years has been greater in the 72 hours data. While it is encouraging to see this improvement within the parameters of the formal full newborn examination, the cleft clinical community still supports an emphasis to improve earlier diagnosis within the first 24 hours from birth.

CRANE will continue to encourage work with partners involved in development of the original palate examination guidance, to see if it is practical to include formal palate examination within current guidance on infant feeding within the first 24 hours from birth.

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[8] National Institute for Health and Clinical Excellence. Guideline on Postnatal care up to 8 weeks after birth. NICE Guideline (CG37). London: NICE, 2006. [Last updated in 2015] https://www.nice.org.uk/guidance/cg37/ifp/chapter/During-the-first-week

3.4. Gestational age and birth weight

Cohort summary	Data source	CRANE Database (consented cases only)
	Birth years	10 years: 2010 to 2019
	Denominators	• 4,248 children with a recorded gestational age • 4,738 children with a recorded birth weight • 4,241 children born at term (>=37 weeks) with a recorded birth weight
	Numerators	• 514 children born prematurely (<37 weeks' gestation) • 315 children born at term with a low birth weight (<2500 g) • 422 children born at term with a high birth weight (>=4000 g)
	Exclusions (not mutually exclusive)	• Children without consent to data collection • Children without a recorded gestational age • Children without a recorded birth weight
	Data completeness	Gestational age: • 51% of 8,394 eligible children had a recorded gestational age. • 1% had a reason gestational age was not collected. • 48% were missing gestational age data or a reason for not collecting data. Birth weight: • 57% of 8,394 eligible children had a recorded birth weight. • 1% had a valid reason birth weight was not collected. • 42% were missing birth weight data or a reason for not collecting data.
	Countries	England, Wales and Northern Ireland
Benchmark		• 100% of eligible children should have a recorded gestational age and birth weight (or a valid reason it was not collected). • 7% of babies in the general population of England & Wales are born prematurely. • 7% of babies in the general population of England & Wales are born with a low birth weight (LBW). 10.5% are born with a high birth weight (HBW)[9].
What did we find?		• The mean gestational age was 38.7 weeks. • The percentage of premature births among children born with a cleft was higher than in the general population (12% compared to 7%). • The average birth weight was 3188 g. Children with CL had a higher mean birth weight (3296 g) than those with a cleft affecting the palate (3154 g). • 7.4% of children born at term (>=37 weeks) weighed <2500 g (LBW) and 10.0% weighed >=4000 g (HBW).
Recommendations		• Cleft care teams should aim to improve the reporting of gestational age and birth weight in the CRANE Database. This is essential in monitoring the rate of premature births and the rate of babies with LBW and HBW among the cleft population. • The research community should validate and further investigate the apparent association between cleft-affected pregnancies and prematurity at birth. • CRANE will engage with the RCM and RCOG to communicate our findings.

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[9]https://www.ons.gov.uk/peoplepopulationandcommunity/birthsdeathsandmarriages/livebirths/bulletins/birthcharacteristicsinenglandandwales/2016/pdf

3.4.1. Data completeness for gestational age

Of the 8,394 children born between 2010 and 2019 whose families had consented to their children's data being collected by CRANE, 4,248 (50.6%) children had gestational age reported to CRANE. This number varied significantly by birth year (p<0.001), ranging between 24.3% in 2010 and 67.4% in 2014. Since 2014 the rate has fallen again, as shown in Figure 3.8 below.

Figure 3.8. Percentage of CRANE-consented children with recorded gestational age, by birth year.

	2010	2011	2012	2013	2014	2015	2016	2017	2018	2019
Gestational age data	24.3%	45.8%	52.6%	60.7%	67.4%	57.7%	52.2%	50.6%	51.2%	46.5%
Reason for missing data	2.5%	1.9%	2.6%	1.8%	0.5%	0.2%	0.0%	0.3%	0.1%	0.2%
Missing data	73.2%	52.3%	44.9%	37.6%	32.2%	42.1%	47.8%	49.2%	48.7%	53.3%
Toal (N)	1,062	1,107	1,168	1,167	1,073	1,119	1,073	1,081	1,018	897
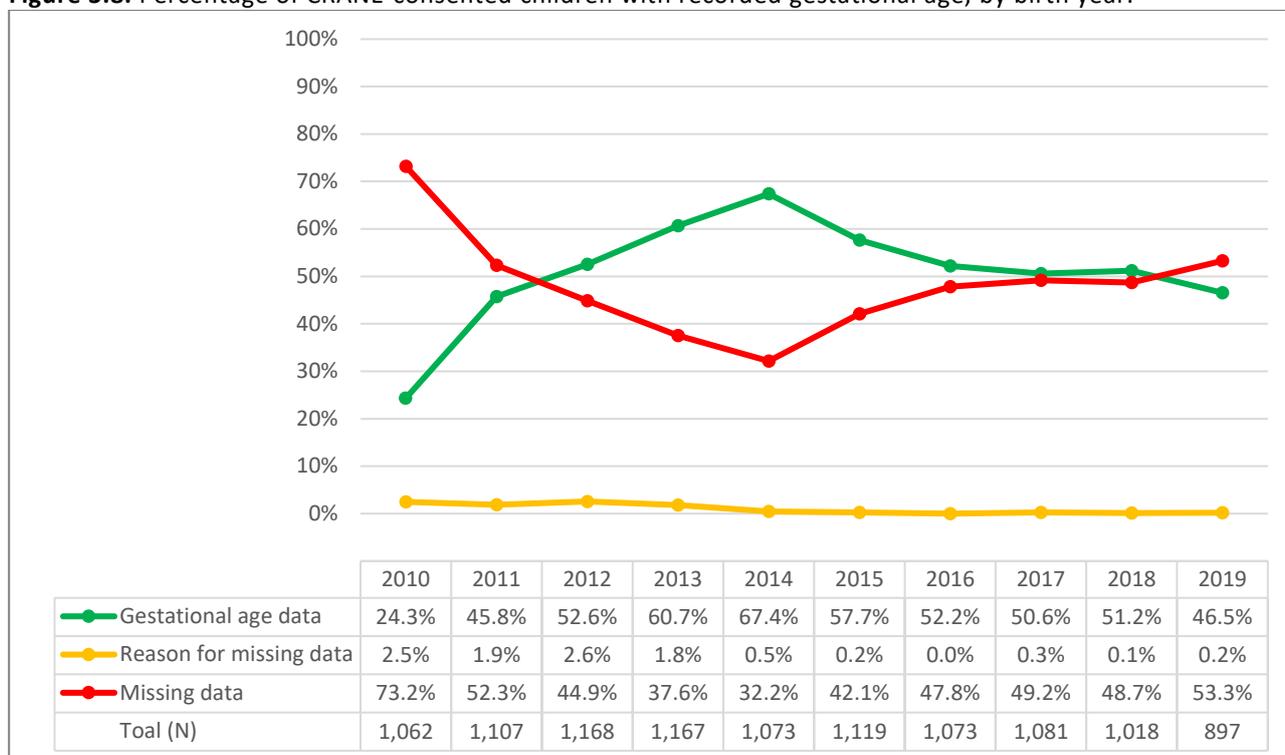
3.4.2. Gestational age

The mean gestational age was 38.7 weeks (95% confidence interval 38.6 to 38.8) and was significantly higher among children with cleft lip (39.0 weeks, p<0.001) when compared to cleft palate (38.6 weeks), UCLP (38.6 weeks) and BCLP (38.4 weeks). Of babies with known gestational age at birth (n=4,248), 12.1% were born prematurely (born before 37 weeks' gestation). This percentage varied significantly by birth year (p=0.019), ranging from 9% to 15%. This is higher than the 7.0% national average in England and Wales[10]. It should be noted that the gestational age recorded in CRANE may not be representative of all babies born with a cleft lip and/or palate as almost half of all consented children were missing information on their gestational age at birth.

3.4.3. Data completeness for birth weight

Of the 8,394 children born between 2010 and 2019 whose families had consented to their children's data being collected by CRANE, 4,738 (56.4%) children had their birth weight recorded in the CRANE Database. This percentage varied significantly by birth year (p<0.001). Figure 3.9 shows that from 2011, reporting steadily improved until 2014, when 69.0% of consented children had their birth weight reported. Since

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[10] Office for National Statistics. Gestational-specific infant mortality. Part of Gestational-specific infant mortality in England and Wales, 2014. Published 14 October 2015 and updated on 20 April 2017 (this is the latest release – checked October 2020). Available

2014, however, reporting has continued to decline, with birth weight reported for only 47.4% of consented children born in 2019. Over the same period, there was no significant variation in data completeness for birth weight by cleft type (p=0.579).

Figure 3.9. Percentage of CRANE-consented children with recorded birth weight, by birth year.

	2010	2011	2012	2013	2014	2015	2016	2017	2018	2019
Birthweight data	61.6%	52.7%	54.4%	61.6%	69.0%	58.0%	52.3%	50.7%	50.0%	47.4%
Reason for missing data	2.5%	1.9%	2.6%	1.8%	0.5%	0.2%	0.0%	0.3%	0.1%	0.2%
Missing data	35.9%	45.4%	43.0%	36.6%	30.5%	41.8%	47.7%	49.0%	49.9%	52.4%
Total (N)	1,011	1,005	976	948	877	855	759	712	690	561
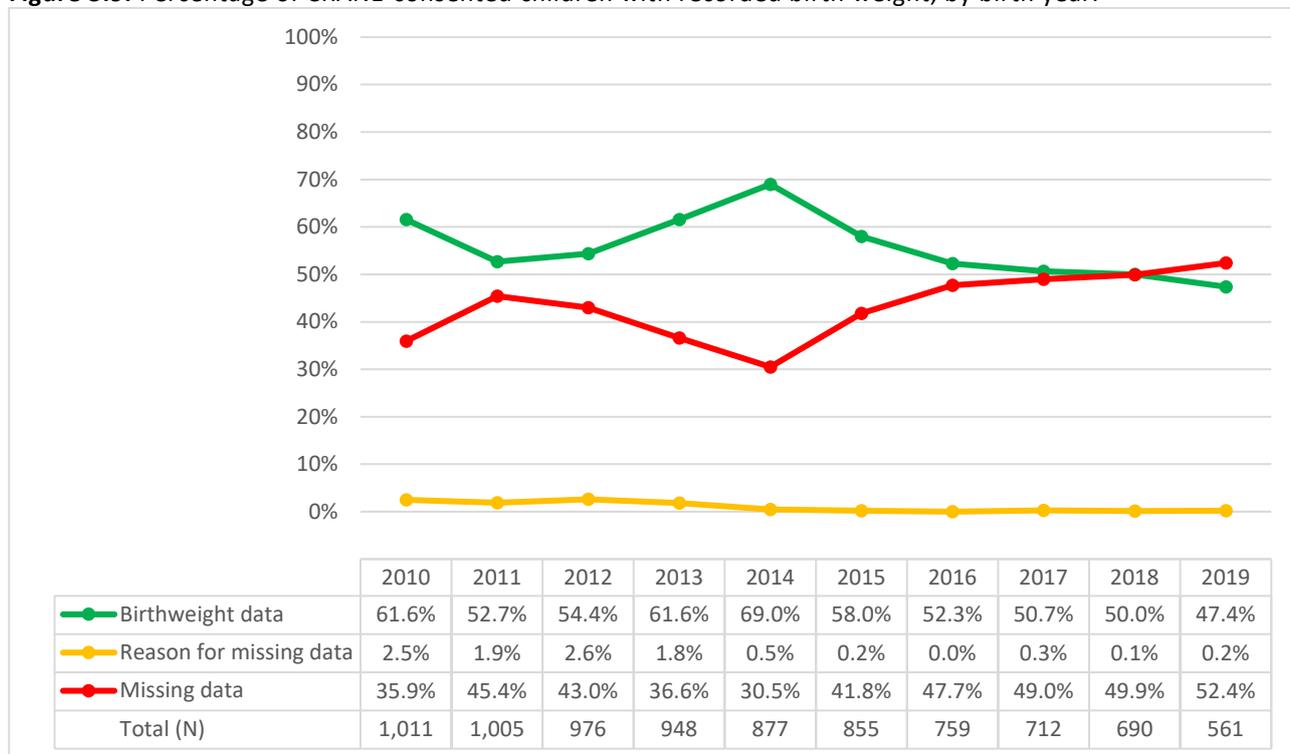
3.4.4. Birth weight

Over the last ten years, the overall mean birth weight was 3188 g (95% CI 3169 g to 3207 g). Mean birth weight varied significantly between those with CL (3296 g, 95% CI 3261 g to 3331 g) and those with a cleft affecting the palate (CP, UCLP and BCLP) (3154 g 95% CI 3132 g to 3177 g) (p<0.0001). This was also the case when comparing birthweights among babies born at term only. There was no statistically significant difference in birth weight between those with CP, UCLP or BCLP (p=0.128). For children born at term (>=37 weeks' gestation), 7.5% were born with a low birth weight (<2500 g) and 10.0% were born with a high birth weight (>=4000 g). The corresponding rates in the general population are 7.0% and 10.5%, respectively[11] It should be noted that the birth weight recorded in CRANE may not be representative of all babies born with a cleft lip and/or palate as almost 42% of all consented children were missing this information.

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[11]https://www.ons.gov.uk/peoplepopulationandcommunity/birthsdeathsandmarriages/livebirths/bulletins/birthcharacteristicsinenglandandwales/2016/pdf

3.5. Contact with cleft care teams

Cohort summary	Data source	CRANE Database
	Birth years	Ten years: 2010 to 2019
	Denominators	• 7,779 children with a recorded time of referral to cleft care teams • 8,729 children with a recorded time of first contact
	Numerators	• 6,249 children referred to cleft care team within 24 hours of birth • 8,203 children contacted by cleft care team within 24 hours of referral
	Data completeness	• 72% of 10,765 eligible children had a recorded referral time. • 81% had a recorded time of first contact.
	Countries	England, Wales and Northern Ireland
Benchmark	• Children with a cleft should be referred to cleft care teams within 24 hours of birth. • Cleft care teams should establish first contact with families of children within 24 hours of receiving a referral.	
What did we find?	• Over the last ten years, 80% of families were referred to a cleft care team within 24 hours of birth. This percentage varied significantly across cleft types. • 94% of families were contacted by a cleft care team within 24 hours of referral.	
Recommendations	• Cleft care teams should continue to work with referring obstetric, midwifery and neonatal units to improve timeliness of diagnosis and early referrals. • Cleft care teams with high levels of referrals within 24 hours should share their best practise recommendations.	

3.5.1. Data completeness

Out of 10,765 children born and registered in CRANE between 2010 and 2019, 72.3% (range: 61% - 85%) had a recorded referral time to a cleft care team. Figure 3.10 shows that data completeness for referral time has decreased over time, from 85.3% in 2010 to 60.8% in 2019 (p<0.001).

Figure 3.10. Percentage of CRANE-registered children with referral time and time of first contact with cleft care team, by birth year.

|	2010	2011	2012	2013	2014	2015	2016	2017	2018	2019
Recorded referral time	85.3%	77.9%	73.4%	77.8%	77.2%	73.1%	65.2%	63.6%	65.6%	60.8%
Recorded contact time	83.7%	71.4%	66.6%	80.0%	86.4%	88.2%	80.6%	83.5%	86.7%	86.2%
Total (N)	1,062	1,107	1,168	1,167	1,073	1,119	1,073	1,081	1,018	897
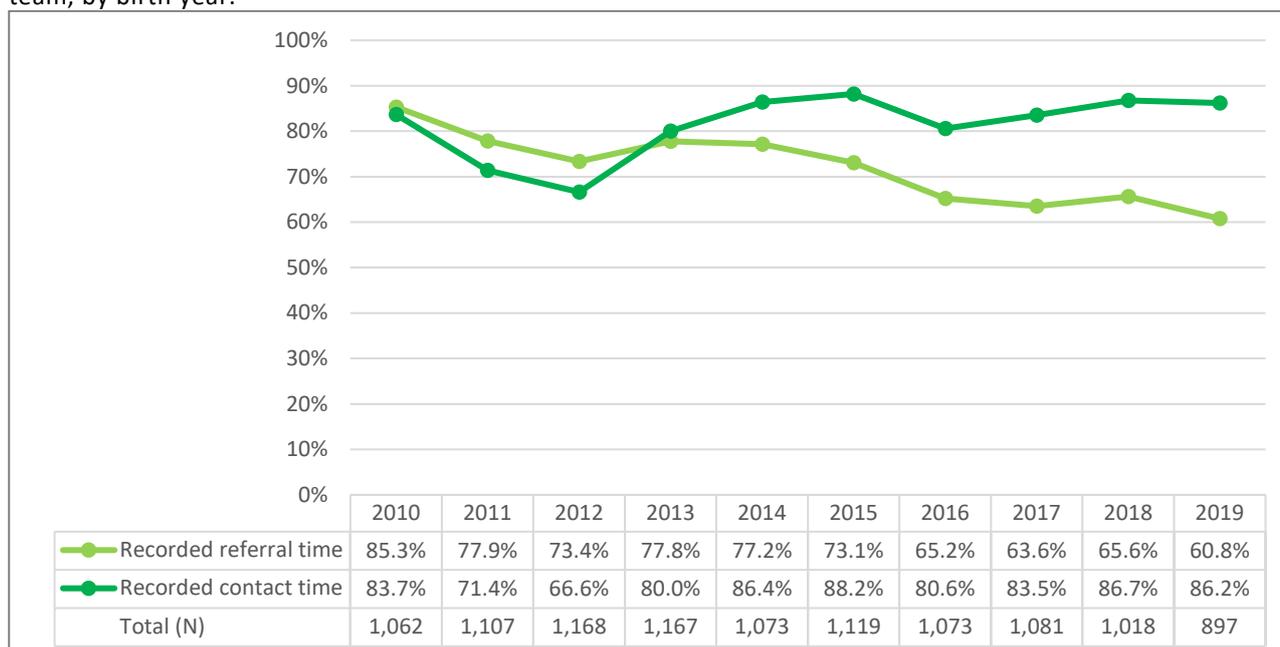
Over the same period of time, 81.1% of children had a recorded contact time. Data completeness for contact time dropped from 83.7% in 2010 to 66.6% in 2012 but then improved to 86.7% in 2018 (p<0.001), as shown in Figure 3.10. It is essential for cleft care teams to record the referral and contact time to enable the monitoring and improvement of timely referrals and first contact with families.

3.5.2. Referral and first contact

Among the 7,779 children born between 2010 and 2019 with a recorded referral time, 80% of children were referred to a Cleft care team within 24 hours of birth. As shown in Figure 3.11, this rate improved from 77.3% in 2011 to 85.2% in 2018 (p=0.003). The percentage of children referred to a cleft care team within 24 hours of birth varied significantly by cleft type (p=0.003). Children diagnosed with CP have the lowest rate of early referrals within 24 hours of birth (66%), compared to CL (88%), UCLP (95%) and BCLP (93%). This is consistent with later diagnosis times for children with CP. For more information on diagnosis times for children with CP, please refer to Section 3.3 of this report.

Figure 3.11. Timeliness of referral and first contact with a cleft care team for CRANE-registered children, by birth year.

	2010	2011	2012	2013	2014	2015	2016	2017	2018	2019
Referral < 24 hours	78.5%	77.3%	77.9%	80.6%	80.6%	82.3%	78.9%	82.1%	85.2%	82.0%
Contact < 24 hours	87.7%	92.0%	92.0%	89.9%	95.1%	97.4%	96.8%	96.3%	95.8%	96.2%
Total (N)	1,062	1,107	1,168	1,167	1,073	1,119	1,073	1,081	1,018	897
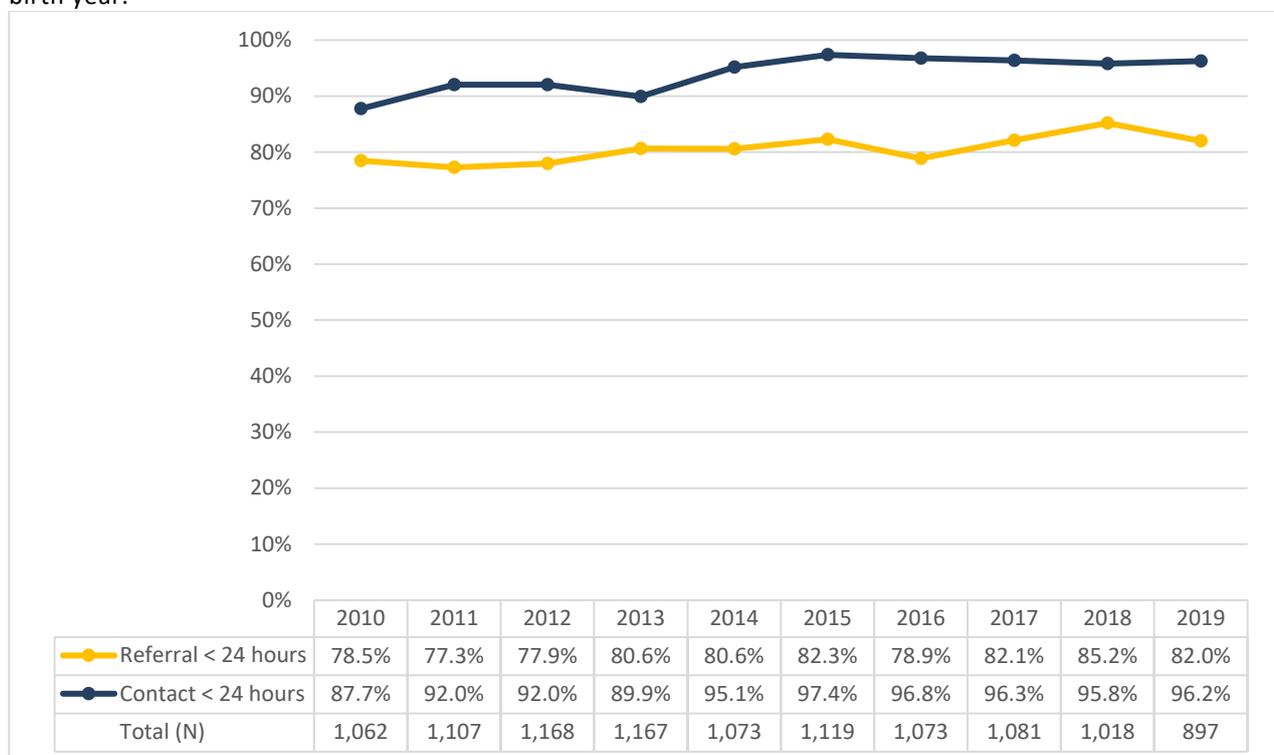
Note: Children missing referral and first contact time are excluded.

Of the 8,729 children with a recorded time of first contact with cleft care teams, 94% were contacted within 24 hours of referral. This rate increased from 87.7% in 2010 to 97.4% in 2015 (p<0.001), as displayed in Figure 3.11. Contact within 24 hours of referral has remained above 95% since 2015. This demonstrates the commitment of cleft care teams to ensure a timely response to new referrals of babies born with a cleft in order to help support these babies and their families in the important initial stages.

For the same cohort of patients, the percentage of cleft care teams establishing contact with parents within 24 hours of referral varied according to cleft type (p<0.001). Children with UCLP and BCLP had higher rates of contact within 24 hours (96.0% and 96.2%, respectively) than those with CP (92.7%) and CL (93.5%).

3.6.   CRANE consent

Cohort summary	Data source	CRANE Database
	Birth years	Ten years: 2010 to 2019
	Denominator	10,765 children registered in CRANE
	Numerators	8,544 children whose families had made a decision about consent
	Data completeness	79% of 10,765 registered children had verified consent. This means they had agreed to or declined CRANE data collection.
	Countries	England, Wales and Northern Ireland
Benchmark	100% of families of children with a cleft should be approached for consent verification (to provide informed consent or decline) CRANE outcome data collection.	
What did we find?	• 79% of families had reached a decision about agreeing or declining consent. • Of the families who had reached a decision, the rate of agreed consent was 98%, and this rate of positive consent remained consistently above 97% over the last 10 years. • The overall percentage of registered children with verified consent has declined over the last ten years.	
Recommendations	• Cleft care teams should review their procedures to identify reasons for low rates of verified consent, and ensure that CRANE consent status is recorded for every child with a cleft. • CRANE will continue to work with cleft care teams and the Nursing Clinical Excellence Network (CEN) to improve consent status verification.	

The percentage of children born between 2010 and 2019 whose families had made a decision to provide or decline consent (verified consent status) varied significantly by birth year (p<0.001). Overall, 8,544 (79.4%) out of 10,765 children had a verified consent status. Figure 3.12 shows that the percentage of children awaiting consent verification increased steadily from 2.6% for those born in 2010 to 35.3% for those born in 2019 (p<0.001). Deterioration in consent verification over time has been discussed with the UK NHS Cleft Development Group (CDG). Individual cleft care teams are examining their processes to address this issue.

Figure 3.12. Consent status distribution among CRANE-registered children, by birth year.

	2010	2011	2012	2013	2014	2015	2016	2017	2018	2019
Informed consent	95.2%	90.8%	83.6%	81.2%	81.7%	76.4%	70.7%	65.9%	67.8%	62.5%
Awaiting verification	2.6%	7.0%	13.5%	15.5%	14.7%	20.9%	28.2%	32.2%	30.5%	35.3%
Declined consent	1.7%	1.8%	1.5%	1.3%	2.4%	1.3%	0.7%	0.8%	1.0%	1.3%
Not possible to verify	0.5%	0.4%	1.5%	2.0%	1.1%	1.3%	0.3%	1.1%	0.8%	0.8%
Total (N)	1,062	1,107	1,168	1,167	1,073	1,119	1,073	1,081	1,018	897
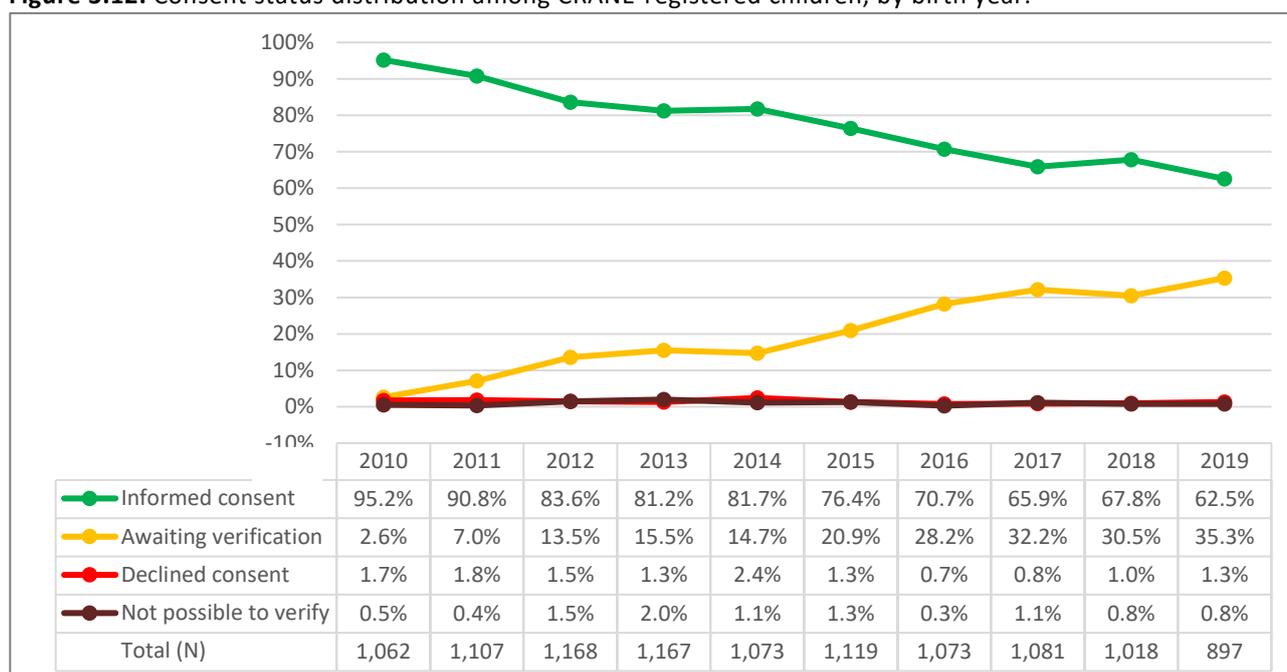
The rate of verified consent varied significantly by cleft type (p<0.001), and was highest among children born with UCLP (86%), followed by BCLP (84%), CL (82%) and CP (80%). Overall, cleft care teams reported that it was not possible to verify consent for 106 children (1.0%).

Although the percentage of children with informed consent has declined since 2010 (Figure 3.12) when calculated as a percentage of all registered cases, this is because of the increasing number of children whose consent status is yet to be verified.

Among the 8,544 families who had reached a decision to provide informed consent or decline consent (with verified consent), 8,394 (98.2%) agreed to their children's data being collected by CRANE. This rate did not vary significantly by birth year (p=0.261) or cleft type (p=0.464).

4. Audit Outcomes at 5 years of age

This chapter details cleft-related outcomes for children at five years of age, registered in the CRANE Database with a cleft lip and/or palate. Outcomes include children's growth, dental health, facial growth, speech and psychology at 5 years of age.

Submucous clefts are not audited by cleft teams and are therefore excluded from reporting for all audit outcomes at 5 years of age. Submucous clefts comprised 4% of consented cases born between 1 January 2004 and 31 December 2013.

In previous years, we have reported on outcomes by individual regions / cleft care teams. In 2020, cleft care teams could not be expected to complete and log assessments for all eligible children due to the impact of COVID-19 on cleft care services. Therefore, this year, we reflect on the national picture across the last 10 years for child growth, dental health, and facial growth data. Seven years of data are now available for speech and three years of data are available for psychology.

Timeframe: Broadly, the most recent ten years of data available – for children born between 1 January 2004 and 31 December 2013, with the exception of speech (2007-2013 births) and psychology (2011-2013 births) outcomes.

Legal basis for data collection and analysis: The data used for this section is collected for all children whose families have given informed consent to outcomes data collection by the CRANE Database.

4.1. Child growth

| Cohort summary	Data source	CRANE Database (consented cases only)
	Birth years	Ten years: 2004 to 2013
	Denominator	3,486 children with known height and weight at 5 years
	Numerator	Number of children in each BMI category (Underweight, Healthy weight, Overweight and Obese)
	Exclusions	• Children without consent to data collection • Children who died before the age of 5 years • Children with submucous cleft palates
	Data completeness	Weight: • 39% of 9,356 eligible children had a recorded weight at 5 years. • 8% had a reason the weight at 5 years was not collected. • 53% were missing weight data or a reason for not collecting data. Height: • 38% of 9,356 eligible children had a recorded height at 5 years. • 8% had a reason the height at 5 years was not collected. • 54% were missing height data or a reason for not collecting data.
	Countries	England, Wales and Northern Ireland
Benchmarks		• For 5-year-olds in the UK, healthy BMI ranges between 13.0 and 17.5 (2nd to 91st centiles). • Underweight is BMI <13.0 kg/m^2, overweight is BMI 17.5-19.0 kg/m^2 (92nd to 98th centiles), and obese is BMI >19.0 kg/m^2 [12]. • National prevalence of underweight, overweight and obesity among five year olds was estimated at 1%, 13% and 15%, respectively, among boys, and 2%, 11% and 11%, respectively, among girls [13].
What did we find?		• Overall, 86% of children had a healthy Body Mass Index (BMI). • The percentage of children who were underweight, overweight and obese was 1.5%, 7.0% and 3.1% in boys, respectively. The corresponding rates among girls were 2.6%, 9.2% and 4.4%, respectively. • The prevalence of overweight and obesity was found to be higher among children with CL than among those with a cleft affecting the palate (CP, UCLP and BCLP), while the prevalence of underweight was higher among children with CP (2.5%) than among those with other cleft types.
Recommendations		Cleft care teams should aim to assess children's weight and height around the age of five years and improve the reporting of these measures in the CRANE database. This will facilitate more meaningful comparisons between subgroups in the future.

4.1.1. Data completeness

Of the 9,356 children born in 2004 to 2013 whose families had consented to their children's data being collected by CRANE, 3,647 (39.0%) had a 5-year old weight reported to CRANE. Figure 4.1 shows that the percentage of children with weight information improved from 16.3% for 2004 births to 52.4% among 2013

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[12] According to the Royal College of Paediatrics and Child Health - 2-20 years Body Mass Index (checked October 2020). Available from: https://www.rcpch.ac.uk/sites/default/files/2018-03/boys_and_girls_bmi_chart.pdf - See also English National Child Measurement Programme's healthy weight calculator. Reviewed 15 October 2020 (checked October 2020). Available from: https://www.nhs.uk/live-well/healthy-weight/national-child-measurement-programme/

[13] National Child Measurement Programme Tables, England 2018/19 School Year – Published 10th October 2019 (Last accessed: October 2020) – Available at https://digital.nhs.uk/data-and-information/publications/statistical/national-child-measurement-programme/2018-19-school-year

births (p<0.001). Overall, there were 3,522 (37.6%) children with 5-year old height data reported to CRANE. Data collection for this has also improved over the last ten years, increasing from 15.9% for 2004 births to 51.5% among 2013 births (p<0.001) (Figure 4.2). In total 3,486 (37.2%) children had both weight and height data at 5 years of age. There were 714 (6.7%) children who had a valid reason reported for not collecting 5-year-old growth data. Overall, 53% and 54% of children were missing either data or a valid reason for the data not being collected for weight and height, respectively. Despite improvements in data reporting over the last ten years of data collection, efforts are required to better report weight and height to the CRANE Database.

Figure 4.1. Percentage of 5-year old CRANE-consented children who have recorded weight data, a reason for missing weight data or missing data without a reason, according to birth year.
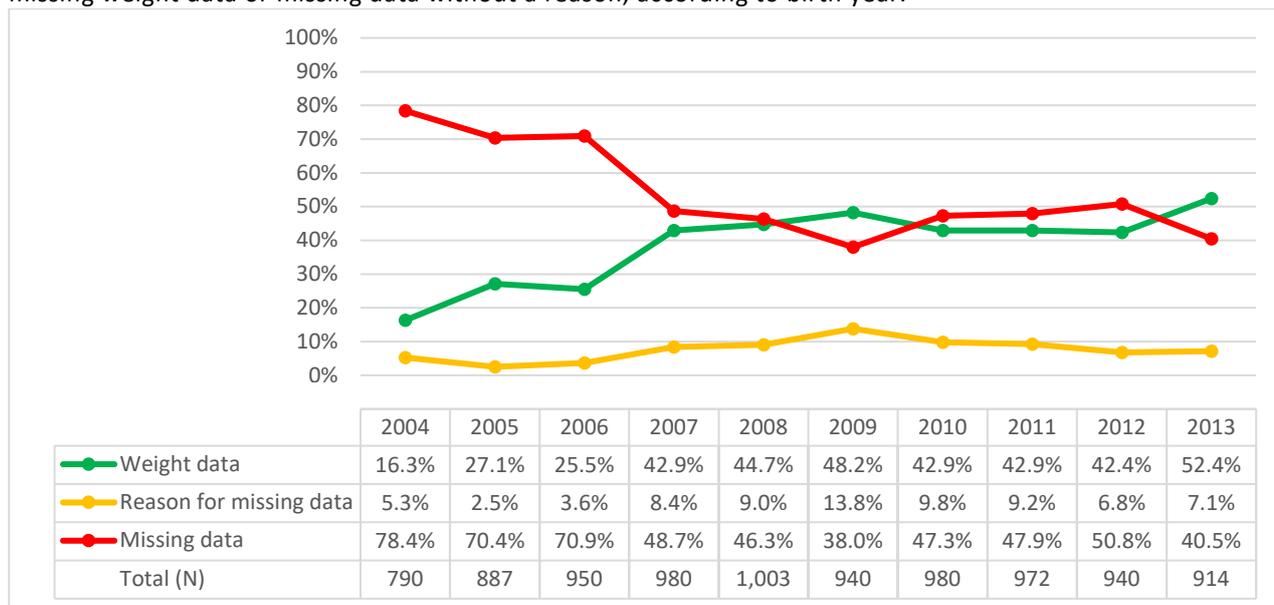
	2004	2005	2006	2007	2008	2009	2010	2011	2012	2013
Weight data	16.3%	27.1%	25.5%	42.9%	44.7%	48.2%	42.9%	42.9%	42.4%	52.4%
Reason for missing data	5.3%	2.5%	3.6%	8.4%	9.0%	13.8%	9.8%	9.2%	6.8%	7.1%
Missing data	78.4%	70.4%	70.9%	48.7%	46.3%	38.0%	47.3%	47.9%	50.8%	40.5%
Total (N)	790	887	950	980	1,003	940	980	972	940	914

Note: Registered in CRANE by 13 July 2020. Exclusions (not mutually exclusive): Children who died before the age of 5 years and children with submucous cleft palates

Figure 4.2. Percentage of 5-year old CRANE-consented children who have recorded height data, a reason for missing height data or missing data without a reason, according to birth year.
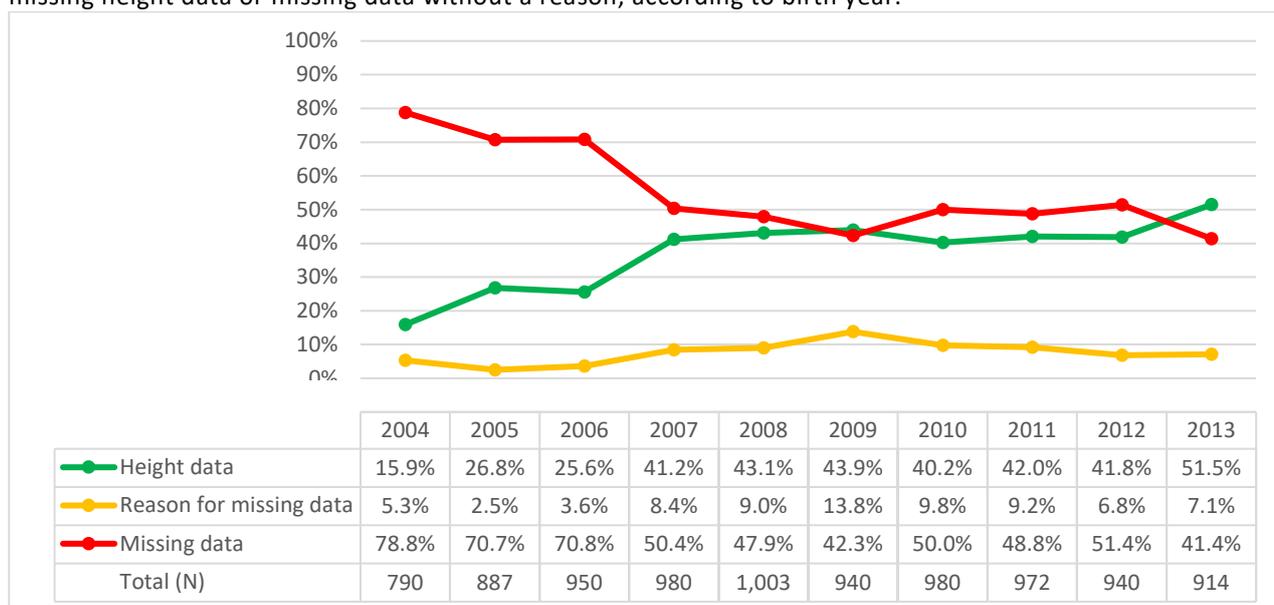
	2004	2005	2006	2007	2008	2009	2010	2011	2012	2013
Height data	15.9%	26.8%	25.6%	41.2%	43.1%	43.9%	40.2%	42.0%	41.8%	51.5%
Reason for missing data	5.3%	2.5%	3.6%	8.4%	9.0%	13.8%	9.8%	9.2%	6.8%	7.1%
Missing data	78.8%	70.7%	70.8%	50.4%	47.9%	42.3%	50.0%	48.8%	51.4%	41.4%
Total (N)	790	887	950	980	1,003	940	980	972	940	914

Note: Registered in CRANE by 13 July 2020. Exclusions (not mutually exclusive): Children who died before the age of 5 years and children with submucous cleft palates

There was significant variation in data completeness of 5-year old weight and height data by cleft type (p<0.001 in both cases). Data completeness was higher among children with UCLP (47.0% W, 45.8% H) and BCLP (46.2% W, 43.8% H), compared to children with CP (36.8% W, 35.3% H) and CL (34.4% W, 33.6% H).

4.1.2. Height and weight

Analyses revealed that the mean weight for 5-year old children, born between 2004 and 2013, was 19.7 kg (19.9 kg for boys and 19.6 kg for girls) and mean height was 111.8 cm (112.4 cm for boys and 111.0 cm for girls). These figures are consistent with UK national averages for 5-year-olds in the general population for weight (range: 15 kg to 22 kg)[14] and height (range: 101cm to 116cm)[14]. Whilst this may appear encouraging considering the potential problems with feeding that some children with a cleft might experience in early life, 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 since almost two thirds of eligible children were missing this information.

4.1.3. Body Mass Index

For children born with a cleft between 2004 and 2013, body mass index (BMI) at 5 years of age was calculated [weight (kg) / height (m) ^{2}] and categorised into weight status categories (underweight, healthy weight, overweight and obese), based on age- and sex-specific cut-offs defined by the Royal College of Paediatrics and Child Health[15]. Among the 5-year old children registered in CRANE who had available growth data, the mean BMI was 15.7 kg/m^2, (15.8 kg/m^2 among boys and 15.7 kg/m^2 among girls), which did not vary significantly over the ten-year period. Mean BMI did vary significantly by cleft type (p<0.001). It was higher among children with CL (16.1 kg/m^2) when compared to children with CP (15.6 kg/m^2), UCLP (15.6 kg/m^2) and BCLP (15.7 kg/m^2).

### 4.1.4. Prevalence of overweight and obesity

Among those children with 5-year old growth data, 2% were underweight, 8% were overweight, and 4% were obese. The fluctuations in the prevalence of different BMI groups over time, as shown in Figure 4.3, should be interpreted with caution due to the high level of missing growth data.

There was significant variation (p=0.001) in BMI group distribution between boys and girls with recorded growth data. The percentage of children with a healthy BMI was higher among boys (88.4%) than girls (83.8%), while fewer boys were underweight (1.5%) than girls (2.6%). Similarly, fewer boys were overweight and obese compared to girls (7.0% vs 9.2% for overweight; 3.1% vs. 4.4% for obese).

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[14] According to the Royal College of Paediatrics and Child Health and UK-WHO growth charts – 2-18 years(checked October 2020) – Available from: https://www.rcpch.ac.uk/resources/uk-who-growth-charts-2-18-years
[15]According to the Royal College of Paediatrics and Child Health - 2-20 years Body Mass Index (checked October 2020). Available from: https://www.rcpch.ac.uk/sites/default/files/2018-03/boys_and_girls_bmi_chart.pdf

Figure 4.3. BMI group distribution among 5-year old CRANE-consented children, by birth year.
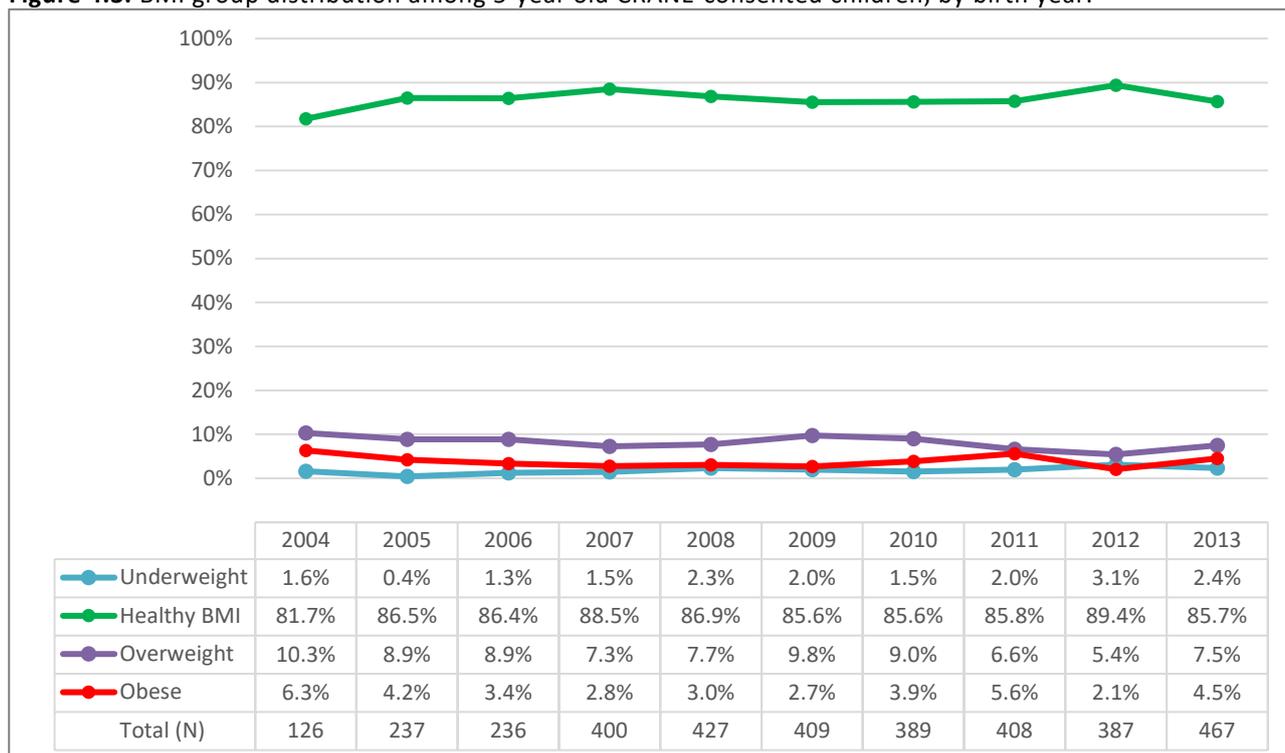
	2004	2005	2006	2007	2008	2009	2010	2011	2012	2013
Underweight	1.6%	0.4%	1.3%	1.5%	2.3%	2.0%	1.5%	2.0%	3.1%	2.4%
Healthy BMI	81.7%	86.5%	86.4%	88.5%	86.9%	85.6%	85.6%	85.8%	89.4%	85.7%
Overweight	10.3%	8.9%	8.9%	7.3%	7.7%	9.8%	9.0%	6.6%	5.4%	7.5%
Obese	6.3%	4.2%	3.4%	2.8%	3.0%	2.7%	3.9%	5.6%	2.1%	4.5%
Total (N)	126	237	236	400	427	409	389	408	387	467

Note: Registered in CRANE by 13 July 2020. Exclusions (not mutually exclusive): Children who died before the age of 5 years, children with submucous cleft palates, and children missing 5-year old growth data

The prevalence of overweight and obesity was found to be higher among children with cleft lip (CL) than among those with a cleft affecting the palate – with cleft palate (CP), unilateral cleft lip and palate (UCLP) or bilateral cleft lip and palate (BCLP). While the prevalence of underweight was higher among children with CP (2.5%) than among those with other cleft types (Table 4.1).

Table 4.1. Percentage of 5-year old CRANE-consented children with growth data who are underweight, healthy weight, overweight or obese, according to cleft type.

Cleft type	All eligible cases	Eligible cases with weight and height reported		Underweight		Healthy weight		Overweight		Obese	
	N	N	(%)	N	(%)	N	(%)	N	(%)	N	(%)
CL	2,216	729	(32.9)	9	(1.2)	597	(81.9)	75	(10.3)	48	(6.6)
CP	4,028	1,384	(34.4)	36	(2.6)	1,208	(87.3)	94	(6.8)	46	(3.3)
UCLP	2,136	960	(44.9)	17	(1.8)	841	(87.6)	77	(8.0)	25	(2.6)
BCLP	923	391	(42.4)	5	(1.3)	350	(89.5)	27	(6.9)	9	(2.3)
Total	9,303	3,464	(37.2)	67	(1.9)	2,996	(86.5)	273	(7.9)	128	(3.7)
p-value		$p<0.001$				$p<0.001$					

Note: Registered in CRANE by 13 July 2020. Exclusions (not mutually exclusive): Children who died before the age of 5 years, children with submucous cleft palates, and children with a non-specified cleft type. p-values are for differences between cleft types.

4.2. Dental health

Cohort summary	Data source	CRANE Database (consented cases only)
	Birth years	Ten years: 2004 to 2013
	Denominators	• 5,439 5-year-old children with recorded decayed, missing or filled teeth (dmft) scores • 5,257 children with scores for the calculation of Treatment Index[16] • 5,272 children with scores for the calculation of Care Index[17]
	Numerators	• 2,201 children with at least one (>0) dmft • 768 children with extensive decay (dmft >5) • Number of children diagnosed with each cleft type
	Exclusions (not mutually exclusive)	• Children without consent to data collection • Children with an unspecified cleft type • Children who died before the age of 5 years • Children with submucous cleft palates[18] • Children without a recorded dmft score at the age of 5
	Data completeness	• 59% of 9,176 eligible children had a recorded dmft. • 10% had a reason dmft was not collected. • 31% were missing data or a reason for not collecting data.
	Countries	England, Wales and Northern Ireland
Benchmark		• 100% of eligible children should have recorded dmft outcome data at the age of 5 years (or a valid reason it was not collected). • 23% of children had at least one (>0) dmft and 13% had extensive decay (dmft >5) in the general population of England, Wales and Northern Ireland at 5 years of age[19]. • The average Treatment Index reported for children is 25% (100% indicates no untreated disease and is the desirable outcome) and the average Care Index reported for children is 10% (100% is the desirable outcome) in the general population in England[20].
What did we find?		• 41% of children with a cleft had at least one (>0) dmft and 14% had six or more dmft (>5). • The average Treatment Index was 76%, and the average Care Index was 69%. • Rates of dmft >0 and dmft >5 increased significantly as cleft type became more involved. • Treated Index scores and Care Index scores decreased significantly as cleft type became more involved.
Recommendations		• All children with a cleft should have a recommended care plan established by collaborative work between the families' General Dental Practitioner (GDP) and cleft care teams to: (a) treat the child as per the high-risk category of the dental toolkit (delivering better oral health), (b) provide local dental care (GDP led), and (c) provide age-specific dental development assessments and advice (cleft care team led). • Cleft care teams 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 CRANE database.

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[16] Treatment Index calculated using: Data on missing teeth (m), filled teeth (f), and dmft scores; or a dmft score of 0 (equating to a treat. index = 1).

[17] Care Index calculated using: Data on filled teeth (f) and dmft scores; or a dmft score of 0 (equating to a Care Index = 1).

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

[19] 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

[20] National Dental Epidemiology Programme for England: oral health survey of five-year-old children 2019 (results) - https://assets.publishing.service.gov.uk/government/uploads/system/uploads/attachment_data/file/873492/NDEP_for_England_OH_Survey_5yr_2019_v1.0.pdf. Data on children in the general population in Wales and Northern Ireland were not available at the time of producing this report.

4.2.1. Data completeness

Participation in a national clinical audit, such as the CRANE audit, means that all regions / cleft care teams are asked to record decayed, missing or filled teeth (dmft) scores for all consented 5-year-old children with a cleft lip and/or palate. If dental outcome data - in the form of dmft scores - are not available, cleft care teams are asked to report a reason for this[2] (e.g. Patient transferred out of area or patient did not attend appointment).

Overall, dmft data have been provided for 5,439 children, representing 59.3% of 9,176 children who were eligible. 9.5% had a recorded valid reason why their dental health could not be audited. 31.2% of those eligible for dental health audit were missing data or a valid reason. Appendix 5 shows the breakdown of reasons reported for not collecting 5-year old dental data; the most common reason selected was 'Lack of staff / facilities / equipment' (37%).

Figure 4.4 shows the variation in dmft data reported by birth year, over the last ten years. Rates of recording of dmft data have shown particular and significant deterioration, from 64.1% among those born in 2009 to 54.5% for those born in 2013 (p<0.001). There could be many reasons for this change over the period examined, including challenges relating to staffing. It is recommended that the paediatric dental CEN discuss this further and work in collaboration with CRANE to reverse the trend.

Dental outcome data are not subject to the additional validation that some other cleft care outcomes are subject to, such as speech and facial growth. Nevertheless, the onset of the COVID-19 pandemic in early 2020 may have impacted teams' ability to audit and submit the most recent year of CRANE data.

Figure 4.4. Percentage of 5-year old CRANE-consented children born 2004-13 who have dmft data, a reason for missing dmft data or missing data without a reason, according to birth year.
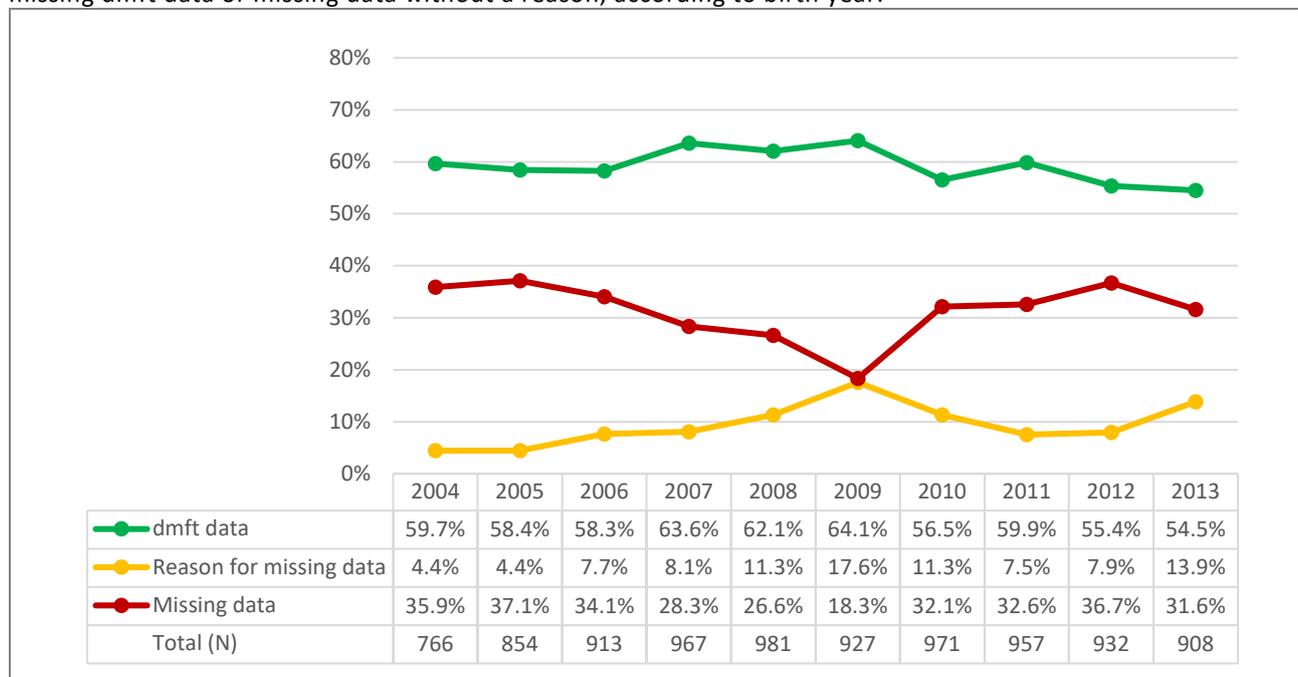
	2004	2005	2006	2007	2008	2009	2010	2011	2012	2013
dmft data	59.7%	58.4%	58.3%	63.6%	62.1%	64.1%	56.5%	59.9%	55.4%	54.5%
Reason for missing data	4.4%	4.4%	7.7%	8.1%	11.3%	17.6%	11.3%	7.5%	7.9%	13.9%
Missing data	35.9%	37.1%	34.1%	28.3%	26.6%	18.3%	32.1%	32.6%	36.7%	31.6%
Total (N)	766	854	913	967	981	927	971	957	932	908

Note: Registered in CRANE by 13 July 2020. Exclusions (not mutually exclusive): Children who died before the age of 5 years, children with submucous cleft palates, and children with a non-specified cleft type.

There was also significant variation in completeness of dmft data by cleft type (p<0.001). Data completeness was higher for children with UCLP (68.4%) and BCLP (67.5%), compared to children with CP (56%) and CL (53.1%).

4.2.2. Dental health by birth year

Among the 5,439 eligible children with a reported dmft score, the mean number of dmft at 5 years was 2.05, with scores ranging from 0 to 20.

Overall, 40.5% of children had at least one dmft (>0), and 14.1% had extensive decay (dmft >5) (as **defined right**).

The fluctuations between birth years in the dental outcomes are presented in Figure 4.5. This figure shows that the percentage of 5-year-old children with dmft>0 has reduced significantly across the ten birth years (p=0.024), and remained consistent for dmft>5 (p=0.62). Appendix 6 shows the breakdown of outcome reporting for each year.


Dental health outcomes at 5 years defined

dmft score describes the dental caries an individual has experienced and is a measure of oral health. It reflects the total number of teeth that are decayed, missing or filled. The risk of dental caries is thought to be higher among children with a cleft lip and/or palate compared with children without an oral cleft[21].

- A dmft >0 indicates experience of dental decay.
- A dmft >5 indicates experience of *extensive* dental decay.

Figure 4.5 also shows the rates of dmft >0 in the general population over the past ten years, as per the rates reported in the 'National Dental Epidemiology Programme for England: oral health survey of five-year-old children' for England. This shows that at least 10% more of the CRANE cohort experienced dental decay, on a yearly basis, by comparison with the national population.

Figure 4.5. Percentage of 5-year old CRANE-consented children born 2004-13 who have dmft data indicating at least one (>0), compared with known general population norms in England, and five or more dmft (>5).
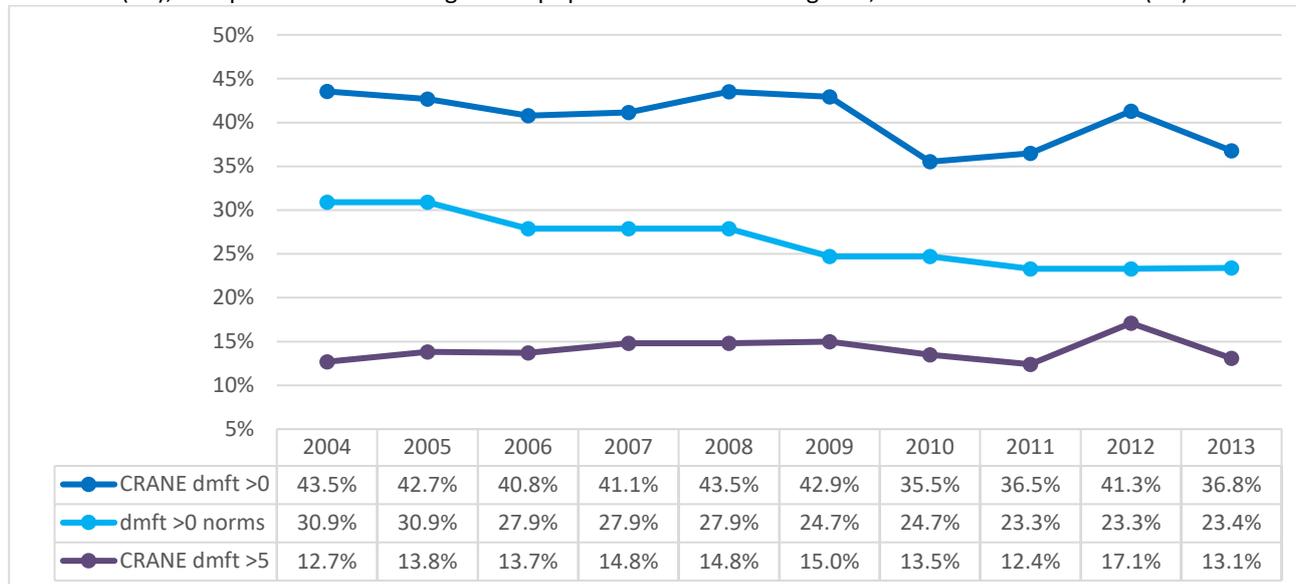
	2004	2005	2006	2007	2008	2009	2010	2011	2012	2013
CRANE dmft >0	43.5%	42.7%	40.8%	41.1%	43.5%	42.9%	35.5%	36.5%	41.3%	36.8%
dmft >0 norms	30.9%	30.9%	27.9%	27.9%	27.9%	24.7%	24.7%	23.3%	23.3%	23.4%
CRANE dmft >5	12.7%	13.8%	13.7%	14.8%	14.8%	15.0%	13.5%	12.4%	17.1%	13.1%

Note: Registered in CRANE by 13 July 2020. Exclusions (not mutually exclusive): Children who died before the age of 5 years, children with submucous cleft palates, and children born with a non-specified cleft type.

The population norm and the CRANE figures demonstrate an overall downward trend. Children with clefts are known to have a higher percentage of teeth with dentine or enamel anomalies making them susceptible to caries.

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[21] (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.

**Dental health measures of intervention defined**

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. dmft scores of 0 or scores for all three 'm', 'f' and 'dmft' data items are required for the calculation of Treatment Index. When calculated, treatment indices range from 0 to 1 and are usually expressed as a percentage[22]. 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.

Care Index reflects cases where children have experienced dental decay, identified at the earliest possible stage (which is preferable), and have been provided with care in the least invasive form possible, i.e. fillings. A dmft score of 0 or scores for both 'f' and 'dmft' data items are required for the calculation of the Care Index. When calculated, care indices also range from 0 to 1 and are usually expressed as a percentage[23]. 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. In instances where a tooth is very poorly formed extraction may be the treatment of choice.

Figure 4.6 shows that the percentage of children achieving each dental health measure of intervention at 5 years (as **defined above**) has fluctuated across the ten birth years. Appendix 6 shows the breakdown of outcome reporting for each year.

Figure 4.6. Average dental Treatment Index and average Care Index among 5-year old consented children born 2004-13, according to birth year.
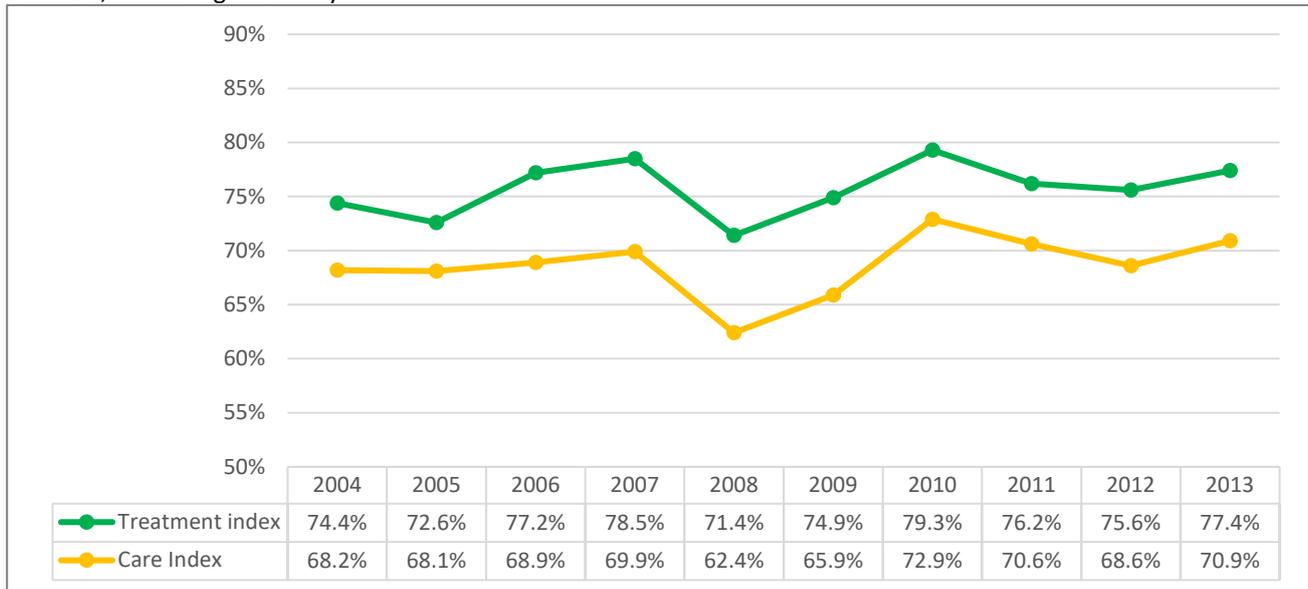
	2004	2005	2006	2007	2008	2009	2010	2011	2012	2013
Treatment index	74.4%	72.6%	77.2%	78.5%	71.4%	74.9%	79.3%	76.2%	75.6%	77.4%
Care Index	68.2%	68.1%	68.9%	69.9%	62.4%	65.9%	72.9%	70.6%	68.6%	70.9%

Note: Registered in CRANE by 13 July 2020. Exclusions (not mutually exclusive): Children who died before the age of 5 years, children with submucous cleft palates, and children born with a non-specified cleft type.

Overall, the average Treatment Index was 75.8%, and the average Care Index was 68.6% for children with a cleft assessed at 5 years of age.

4.2.3. Dental health by cleft type

When all birth years were combined, data completeness was found to vary between cleft types (p<0.001).

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

Table 4.2 shows that children with a cleft affecting both the lip and palate had the highest rates of data completeness compared to those with a cleft affecting either the lip or palate. This may reflect cleft care teams' focus on those at greatest risk. However, data suggest that while children with BCLP experience greater dental decay, they also have received significantly lower levels of timely dental intervention, as demonstrated by the Care Index.

Table 4.2. Number and % of 5-year old CRANE-consented children born 2004-13 with dmft data who had >0 dmft and >5dmft, and average Treatment Index and average Care Index, according to cleft type.

Cleft type	All eligible cases	Eligible cases with dmft reported		At least one (>0) dmft		dmft >5		Mean Treatment Index		Mean Care Index	
	N	N	(%)	N	(%)	N	(%)	N	(%)	N	(%)
CL	2,206	1,172	(53.1)	386	(32.9)	95	(8.1)	1,139	(78.4)	1,143	(74.9)
CP	3,955	2,213	(56.0)	872	(39.4)	354	(16.0)	2,136	(76.2)	2,140	(68.8)
UCLP	2,116	1,447	(68.4)	635	(43.9)	202	(14.0)	1,397	(73.8)	1,402	(66.5)
BCLP	899	607	(67.5)	308	(50.7)	117	(19.3)	585	(73.6)	587	(60.3)
Total	9,176	5,439	(59.3)	2,201	(40.5)	768	(14.1)	5,257	(75.8)	5,272	(68.6)
p-value		p<0.001		p<0.001		p<0.001		p=0.024		p<0.001	

Note: Registered in CRANE by 13 July 2020. Exclusions (not mutually exclusive): Children who died before the age of 5 years, children with submucous cleft palates, and children born with a non-specified cleft type. p-values are for differences between cleft types.

Table 4.2 shows that, as cleft type became more involved, rates of dmft >0 and dmft >5 increased significantly (p<0.001), while average Treatment Index and Care Index decreased significantly (p=0.024 and p<0.001, respectively). Further exploration of this data indicated that the presence of caries is variable for each cleft type over time.

The data reported for dmft, and the Treatment and Care Indices, at 5 years of age show that although dmft remains higher than the general population, the levels of Treatment Index are more than three times higher than in the general population, and the levels of Care Index are almost seven times higher than in the general population. These figures are encouraging and show that dental decay in cleft patients is being treated.

All cleft care teams should aim to follow the national recommendation for a dental check by one year of age, for all children with a cleft. At which time dental prevention advice can be given and a recommended plan to treat the child, as per the high-risk category of the dental toolkit (delivering better oral health), should be provided to the child's General Dental Practitioner (GDP), should it be needed. The GDP will be responsible for the Local dental care. The cleft care team will provide age-specific dental development assessments and advice, as well as a mechanism for referral to the cleft care team for advice and care if GDPs feel the care needed is beyond their competencies.

The CRANE 2019 Annual Report presented findings on decayed missing and filled teeth (DMFT) at 10 years for 2007 and 2008 births. This initial exploration of the data revealed a low number of data returns (14% in 2019). This has improved to 28% data completeness. A summary of the findings from this data has been made available to the Dental CEN for their interpretation and use. Nevertheless, given that fewer than 50% of cleft care teams collect this data at this time, there is insufficient data to accurately reflect on this outcome.

We will review data completeness for this outcome once again in 2021, and anticipate that we will be able to report on DMFT at 10 years in the coming years.

4.3. Facial growth

Cohort summary	Data source	CRANE Database (consented cases only)
	Birth years	Ten years: 2004 to 2013
	Denominator	1,103 5-year-old children with complete UCLP and recorded Five Year Old Index scores
	Numerator	Number of children classified as having 'good', 'fair' and 'poor' Five Year Old Index scores
	Exclusions (not mutually exclusive)	• Children without consent to data collection • Cases with incomplete UCLP • Children who died before the age of 5 years • Children with submucous cleft palates[24] • Children without a recorded Five Year Old Index score
	Data completeness	• 69% of 1,602 eligible[25] consented children had recorded Five Year Old Index scores. • 9% had a reason the facial growth scores were not collected. • 22% were missing data or a reason for not collecting data.
	Countries	England, Wales and Northern Ireland
Benchmark		• 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). • CSAG reported in 1998 that 29%, 34% and 37% of children with a complete UCLP had 'good', 'fair' and 'poor' dental arch relationships, respectively[26].
What did we find?		• 41.3% of children had scores reflecting 'good' dental arch relationships at 5 years old. • 33.5% of children had scores reflecting 'fair' dental arch relationships at 5 years old. • 25.2% of children had scores reflecting 'poor' dental arch relationships at 5 years old. • Dental arch relationships have remained stable over time.
Recommendations		• Cleft care teams should aim to take dental impressions or photographs of all children with a complete UCLP around the age of 5 years, to allow for an assessment using the Five Year Old Index. • The research community should undertake to compare UK facial growth outcomes with those in other countries. • The research community should undertake to evaluate the predictive value of the Five Year Old Index in UK populations.

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 has been used to evaluate 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[27]. Dental arch relationships at 5 years have been thought to predict treatment outcome in terms of facial growth on a population basis rather than at the individual child level[28]. The Five Year Old Index has, therefore, been used to compare treatment

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[24] Submucous cleft palate patients excluded from all 5 year outcomes as all/most teams do not audit these patients.
[25] Registered in the CRANE Database by the 13 July 2020.
[26] Clinical Standards Advisory Group, Clinical Standards Advisory Group. Report of a CSAG Committee on cleft lip and/or palate, 1998, The Stationery Office, London.
[27] 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.
[28] 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.

outcomes between centres and surgeons. Patients scoring '1' and '2' on the index are considered to have good outcomes, while those scoring '4' and '5' are thought to have poor outcomes in terms of facial growth.

4.3.1. Data completeness

Of the 1,602 children, born between 2004 and 2013, with a complete UCLP whose families had consented to their children's data being collected by CRANE, 1,103 (68.9%) had Five Year Old Index data recorded in CRANE. There were 140 (8.7%) children who had a valid reason why the outcome was not collected, and 359 (22.4%) eligible children were missing either data or a reason.

Figure 4.7. Percentage of 5-year old CRANE-consented children with a complete Unilateral Cleft Lip and Palate (UCLP) who have Five Year Old Index scores, a reason for missing data or missing data without a reason, according to birth year.
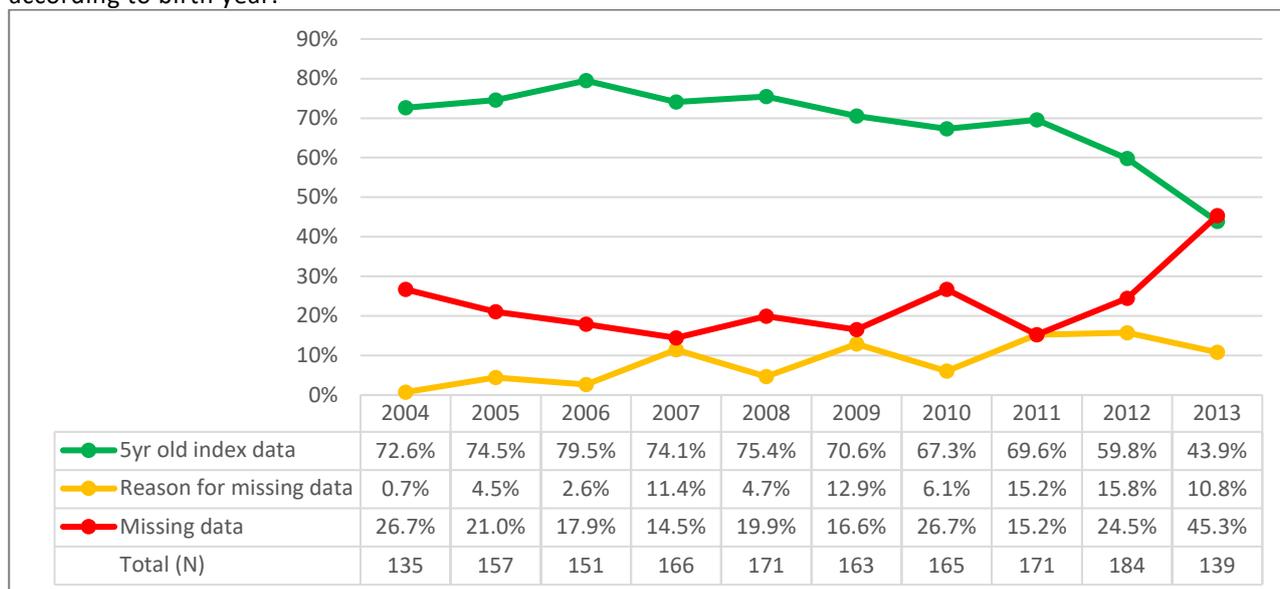
	2004	2005	2006	2007	2008	2009	2010	2011	2012	2013
5yr old index data	72.6%	74.5%	79.5%	74.1%	75.4%	70.6%	67.3%	69.6%	59.8%	43.9%
Reason for missing data	0.7%	4.5%	2.6%	11.4%	4.7%	12.9%	6.1%	15.2%	15.8%	10.8%
Missing data	26.7%	21.0%	17.9%	14.5%	19.9%	16.6%	26.7%	15.2%	24.5%	45.3%
Total (N)	135	157	151	166	171	163	165	171	184	139

Note: Registered in CRANE by 13 July 2020. Exclusions (not mutually exclusive): Children who died before the age of 5 years, children with submucous cleft palates, and children born with an incomplete UCLP. See Appendix 7 for the raw data.

Over the last 10 years (2004 to 2013 birth years), the proportion of eligible children with Five Year Old Index scores has fluctuated, with the best data submission year occurring for 2006 births (Figure 4.7). Since the 2011 birth year, rates of data submission have dropped significantly (p<0.001). For 2013 births, the proportion of children with missing data without a valid reason has more than doubled (45.3%) the average rate observed between 2004 and 2012 (20.2%). This is likely due to several centres being unable to provide any data for their eligible patients because of the onset of the COVID-19 pandemic occurring in early 2020. Whilst models and photos will have been taken by the end of 2019, these need to be scored by various clinicians before the data can be entered onto the CRANE Database. The fact that some centres were able to submit data for 100% of their eligible patients suggests that different audit processes are employed between the cleft centres.

Overall for 2004 to 2013 eligible births, 8.7% (140/1602) had a reason reported for the lack of 5-year old facial growth data. The breakdown of these reasons is shown in Appendix 5. The most common reasons were because the patient did not attend, cancelled, did not consent or did not cooperate. These accounted for 45% of those cases with a reason reported.

4.3.2. Five year old index scores

Among the 1,103 children with reported Five Year Old Index scores, 90% (993) were externally validated. Where an externally validated score was unavailable, internal scores were included in the analyses. Overall, 41.3% had Five Year Old Index scores of '1' or '2,' reflecting 'good' dental arch relationships, 33.5% had a 'fair' score, while 25.2% of children had scores '4' or '5', reflecting poor dental arch relationships. This represents an improvement on the CSAG finding that 29%, 34% and 37% (of 223 children with a complete UCLP) had 'good', 'fair' and 'poor' dental arch relationships, respectively, at 5 years old in 1998[29] (see Appendix 7 for information on children born between 2004 and 2013 with a complete unilateral cleft lip and palate, according to Five Year Old Index scores and birth year).

Figure 4.8 shows the annual fluctuations in the proportion of children classified according to collapsed Five Year Old Index scores. These annual fluctuations were not found to be statistically significant (p=0.315). Five year old facial growth scores have improved since the original CSAG study[30] but appear to have plateaued. Comparison with contemporaneous cohorts from elsewhere in Europe may provide insight as to whether further improvement in facial growth is possible.

Figure 4.8. Distribution of Five Year Old Index scores among 5-year old CRANE-consented children with a complete Unilateral Cleft Lip and Palate (UCLP), according to birth year.
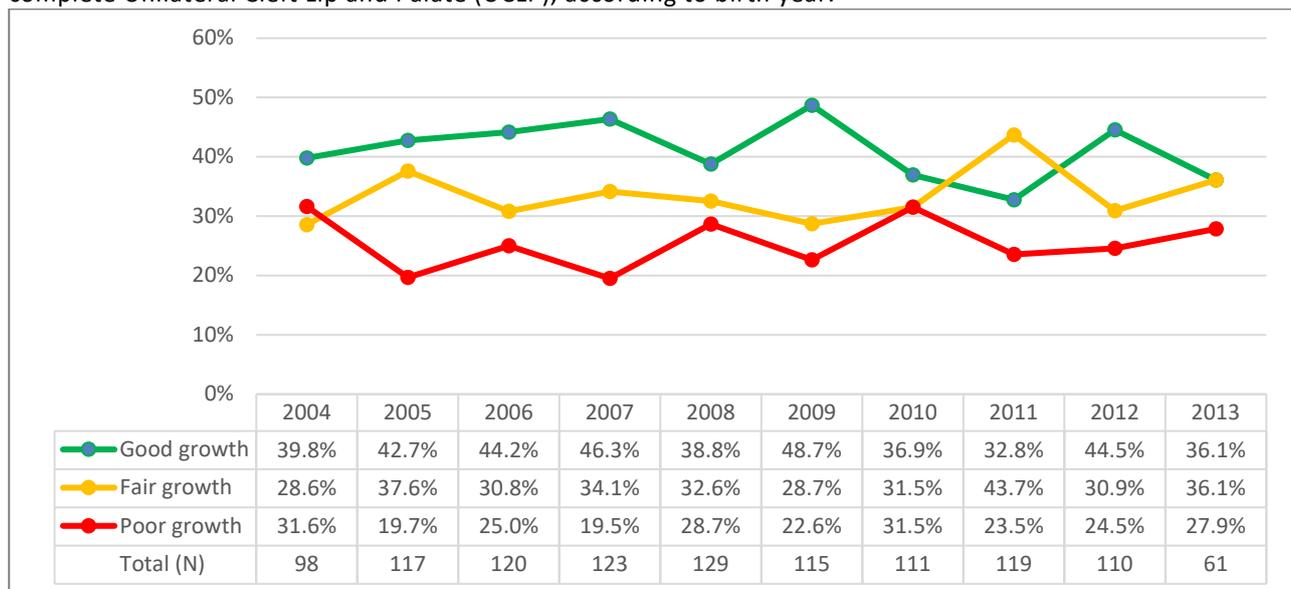
	2004	2005	2006	2007	2008	2009	2010	2011	2012	2013
Good growth	39.8%	42.7%	44.2%	46.3%	38.8%	48.7%	36.9%	32.8%	44.5%	36.1%
Fair growth	28.6%	37.6%	30.8%	34.1%	32.6%	28.7%	31.5%	43.7%	30.9%	36.1%
Poor growth	31.6%	19.7%	25.0%	19.5%	28.7%	22.6%	31.5%	23.5%	24.5%	27.9%
Total (N)	98	117	120	123	129	115	111	119	110	61

Note: Registered in CRANE by 13 July 2020. Exclusions (not mutually exclusive): Children who died before the age of 5 years, children with submucous cleft palates, children born with an incomplete UCLP, and children missing 5 Year Old Index data. See Appendix 7 for the raw data.

A recently published paper using Swedish facial growth data for 106 children with 19-year old follow-up examined the predictive value of dental arch relationship scores, assessed using the Five Year Old Index and Goslon Yardstick, from five years of age onwards[30]. Prediction rates were calculated as the proportion of models remaining in the same category (good - scores 1 and 2; fair - score 3; poor – scores 4 and 5) over time. If a person had been offered or received orthognathic surgery in their late teens, they were categorised into the 'poor' outcome group.

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[29] Clinical Standards Advisory Group. Report of a CSAG Committee on cleft lip and/or palate, 1998, The Stationery Office, London.
[30] Pegelow M, Rizell S, Karsten A, Mark H, Lilja J, Chalien MN, et al. Reliability and Predictive Validity of Dental Arch Relationships Using the 5-Year-Olds' Index and the GOSLON Yardstick to Determine Facial Growth. *The Cleft Palate-Craniofacial Journal* 2020

The results showed that 84% of children with Five Year Old Index scores of '1' and '2' remained in the same 'good' facial growth group at 19 years. For those with Five Year Old Index scores of '4' and '5', 40% were considered to have a poor facial growth outcome at 19 years of age. The predictive value of Five Year Old Index scores of 3 (fair) was very poor. While we do recommend that further research is undertaken to see whether these results are replicated in the UK and/or other countries, we do suggest that, in the meantime, fair and poor Five Year Old Index scores should be interpreted with caution, and that this should be considered when counselling parents and carers of children with a complete UCLP.

4.4. Speech

Cohort summary	Data source	CRANE Database (consented cases only)
	Birth years	Seven years: 2007 to 2013
	Denominator	2,980 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
	Exclusions (not mutually exclusive)	• Children without consent to data collection • Children with an unspecified cleft type or with a cleft affecting only the lip • Children who died before the age of 5 years • Children with submucous cleft palates[31] • Children with a diagnosed syndrome entered onto the CRANE database • Children without all 16 CAPS-A scores
	Data completeness	• 66% of 4,522 eligible children had all 16 CAPS-A speech parameters reported. • 2% had some but not all 16 CAPS-A speech parameters reported. • 22% had a reason the speech outcomes were not collected. • 11% were missing data or a reason for not collecting data.
	Countries	England, Wales and Northern Ireland
Benchmark/ Standards		• 100% of eligible children with a cleft affecting the palate should have all 16 CAPS-A scores reported to CRANE (or a valid reason they were not collected). • Speech within the normal range (see main text for a full description of these standards[32]). • Speech without difficulties resulting from existing or previous structural anomalies. • Speech without significant cleft-related articulation difficulties.

What did we find?		• The COVID pandemic has significantly affected some centres' ability to submit 5-year old speech data for children born in 2013.
		• For children born with a cleft affecting the palate between 2007 and 2013:
		   ○ 60% achieved speech within the normal range.
		   ○ 71% had speech without difficulties resulting from existing or previous structural anomalies. 18% of children had secondary surgery for speech purposes before the age of 5 years.
		   ○ 68% had speech without cleft-related articulation difficulties.
		   ○ These rates vary significantly according to cleft type. Children with a cleft affecting only the palate tend to fare better, while those with a bilateral cleft lip and palate tend to have the least favourable outcomes.
		• The proportion of children meeting the standards has not changed significantly over time.

Recommendations		• All children with an isolated cleft affecting the palate should have their speech assessed and reported to CRANE. • Where possible, cleft care teams should investigate possibilities of remote methods for consensus listening. • Information given to parents by cleft care teams about expected speech outcomes should take into account the child's cleft type. • The research community should undertake to develop risk stratification models for analysing speech outcomes among children with a cleft.

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[31] Submucous cleft palate patients excluded from all 5 year outcomes as all/most teams do not audit these patients.
[32] 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).

4.4.1. Data completeness

Since 2013, all regions / cleft care teams have been requested to report to CRANE all 16 Cleft Audit Protocol for Speech (CAPS-A) parameters for each consented child with a cleft affecting the palate. The CAPS-A assessment can be obtained at any point between the fifth birthday and the day before the sixth birthday. If speech outcomes are not available, regions / cleft care teams are asked to report a reason for this (see Appendix 5 for a breakdown of these reasons). Overall, complete speech data were provided for 2,980 children, representing 65.9% of the 4,522 children who were eligible. 1.7% had incomplete speech data reported and 21.8% had a reason why the speech could not be audited. 10.5% of those eligible for speech audit were missing data or a valid reason for not collecting data. Table A in Appendix 8 shows the breakdown of speech reporting for each year.

Over the seven birth years that 5-year old speech data have been collected and submitted to CRANE, the proportion of eligible cases with all 16 CAPS-A parameters reported ranged from 65.3% for 2007 births to 70.7% for 2012 births (see Figure 4.9) and did not vary significantly between these years (p=0.169). In 2020 however, as a result of the onset of the COVID-19 pandemic, there was a significant drop in the percentage of cases with full speech data and a significant increase in the percentage of cases without a reason for missing data (p<0.001). Whilst some centres were able to maintain their usual reporting levels for speech outcomes, there were a few centres who were missing data for the majority of their eligible cases. Although speech data will have been collected by 31 December 2019 for 2013 births, due to COVID-19 some centres were unable to consensus listen and submit data. The continuation of the pandemic through 2020 will have prevented many cleft care teams undertaking recordings from children born in 2014, and this will have implications for future reporting.

Figure 4.9. Percentage of 5-year old CRANE-consented children with a non-syndromic cleft affecting the palate who have full speech data, incomplete speech data, a reason for missing speech data or missing data without a reason, according to birth year.
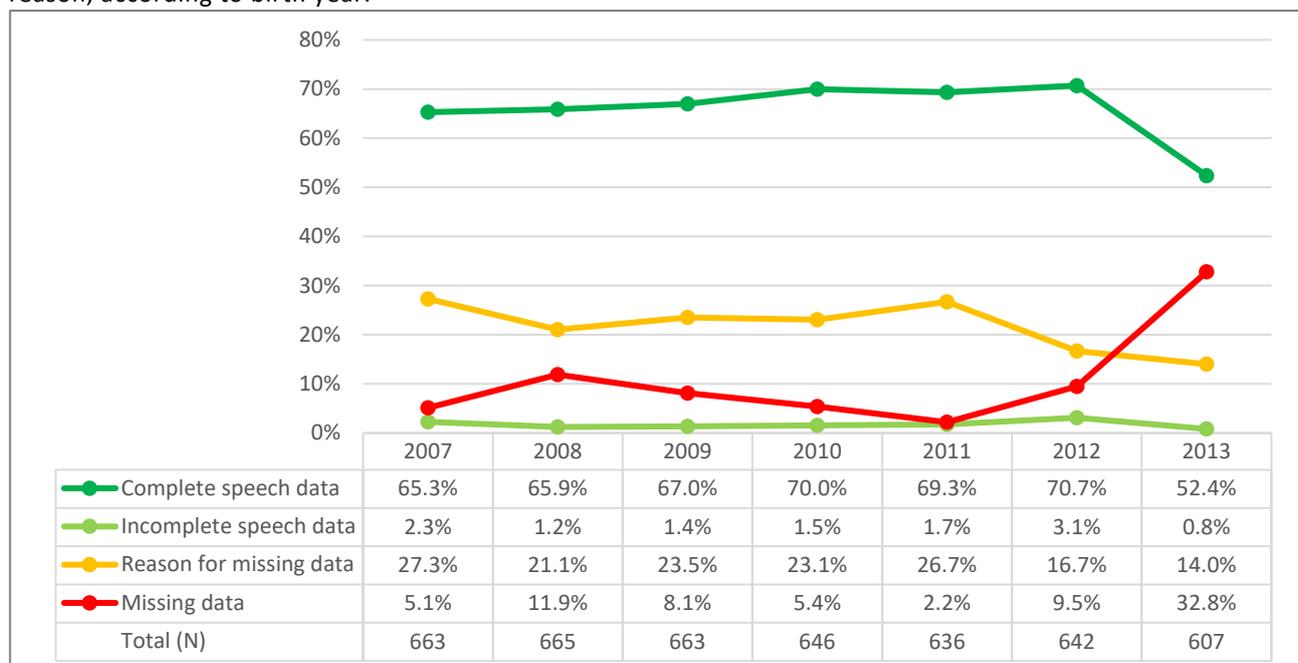
	2007	2008	2009	2010	2011	2012	2013
Complete speech data	65.3%	65.9%	67.0%	70.0%	69.3%	70.7%	52.4%
Incomplete speech data	2.3%	1.2%	1.4%	1.5%	1.7%	3.1%	0.8%
Reason for missing data	27.3%	21.1%	23.5%	23.1%	26.7%	16.7%	14.0%
Missing data	5.1%	11.9%	8.1%	5.4%	2.2%	9.5%	32.8%
Total (N)	663	665	663	646	636	642	607

Note: Registered in CRANE by 13 July 2020. Exclusions (not mutually exclusive): children who died before the age of 5 years, those with submucous cleft palates, those with either a CL or a non-specified cleft type, and those with a diagnosed syndrome entered into the CRANE Database. See Table A in Appendix 8 for raw data.

Overall, 21.8% (988/4522) had a reason reported for not collecting 5-year old speech data. The breakdown of these reasons is shown in Appendix 5. Currently, the Lead Speech and Language Therapy Group are working to standardise usage of reasons for not collecting speech data and guidance will be issued in due course.

4.4.2. Speech parameters and standards

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 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 2007-2013 are presented in Tables B and C in Appendix 8.

Resonance and Nasal Airflow

In terms of resonance, 5.2% of children had moderate or severe hypernasality i.e. nasal sounding speech[33]. 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 CSCs show that 3.2% of children had 'weak and or nasalised consonants' and 1.5% 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. These figures are shown in Table B of Appendix 8.

In order to achieve good speech, 549 out of 2,954 children (18.6%) with reported surgical data[34] had undergone secondary surgery for speech purposes before the age of 5 years. Of these, 391 (71.2%) had resonance and nasal airflow ratings that indicated there were no structural problems that existed in relation to these parameters, suggesting that the surgery resolved these speech issues in time for starting school.

Overall, 84.3% (2513/2980) of children had ratings indicating that no structural problems existed in relation to resonance and nasal airflow by their 5 year assessment[35]. Of these, 15.6% (391 children) had undergone secondary surgery.

Cleft Speech Characteristics (CSCs)

Out of the 2,980 children (born 2007-13) with reported ratings for all 12 CSCs, 66.1% had ratings indicating they did not exhibit any CSCs[36]. 'Palatalisation / Palatal' anterior oral CSCs were the most commonly

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[33] With a hypernasality score of '3' or '4' (red ratings).
[34] VP surgery/fistula repair data was reported for 2954/2980 (99.1%) eligible children.
[35] All green ratings of '0' or '1'.
[36] All green ratings of 'A' and in selected cases of 'B' – as per Appendix 8.

occurring CSC, affecting 23.7% of children (11.6% with ratings of one or two consonants affected (light green ratings) and 12.1% with three or more consonants affected (amber ratings)). These CSCs can vary in severity and may affect speech acceptability more than speech intelligibility. The CSCs which are more likely to affect speech intelligibility are the posterior, non-oral and passive CSCs (see Table C in Appendix 8 for rates of these). Therapy would often be indicated for these children, and/or further investigation of structure and possible surgery.

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[37]:

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 achievement of speech without difficulties resulting from 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 achievement of speech without 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).

Table 4.3. Number and % of 5-year old CRANE-consented children with a non-syndromic cleft affecting the palate who meet each of the three cleft speech standards, according to birth year.

Birth year	All eligible cases	Eligible cases* with all 16 CAPS-A parameters reported		S1. Normal speech		S2a. No structurally-related speech difficulties		S3. No cleft-related articulation difficulties	
	N	N	(%)	N	(%)	N	(%)	N	(%)
2007	663	433	(65.3)	258	(59.6)	306	(70.7)	290	(67.0)
2008	665	438	(65.9)	261	(59.6)	295	(67.4)	303	(69.2)
2009	663	444	(67.0)	269	(60.6)	322	(72.5)	304	(68.5)
2010	646	452	(70.0)	275	(60.8)	319	(70.6)	310	(68.6)
2011	636	441	(69.3)	272	(61.7)	322	(73.0)	296	(67.1)
2012	642	454	(70.7)	273	(60.1)	323	(71.1)	311	(68.5)
2013	607	318	(52.4)	182	(57.2)	220	(69.2)	206	(64.8)
Total	4,522	2,980	(65.9)	1,790	(60.1)	2,107	(70.7)	2,020	(67.8)
p-value		p<0.001		p=0.938		p=0.593		p=0.895	

Note: Registered in CRANE by 13 July 2020. *Exclusions (not mutually exclusive): children who died before the age of 5 years, those with submucous cleft palates, and those with a diagnosed syndrome entered into the CRANE Database. p-values are for differences between years.

Table 4.3 shows, out of the 2,980 eligible children who had full speech data available, the proportion of children achieving each cleft speech standard has remained fairly consistent across the seven birth years. Overall, around 60% of children with a non-syndromic cleft achieved normal speech at 5 years; 71% have

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[37] 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.

speech without any structurally-related speech difficulties, and 68% have speech without any cleft-related articulation difficulties.

Cleft type

When all birth years were combined, data completeness was found to vary between cleft types (p<0.001). Of the children with a non-syndromic CP, 60.9% had complete speech data available, while 71.8% and 72.7% of children with a non-syndromic UCLP and BCLP, respectively, had complete speech data at 5 years. A contributing factor is that more children with CP tend to have additional needs that may mean the assessment is clinically contraindicated.

Table 4.4 Number and % of 5-year old CRANE-consented children with a non-syndromic cleft affecting the palate who meet each of the three cleft speech standards, according to cleft type (birth years 2007-2013).

| Cleft type	All eligible cases*	Eligible cases* with all 16 CAPS-A parameters reported		S1. Normal speech		S2a. No structurally-related speech difficulties		S3. No cleft-related articulation difficulties	
	N	N	(%)	N	(%)	N	(%)	N	(%)
CP	2488	1,514	(60.9)	1,089	(71.9)	1,131	(74.7)	1,216	(80.3)
UCLP	1434	1,030	(71.8)	548	(53.2)	702	(68.2)	634	(61.6)
BCLP	600	436	(72.7)	153	(35.1)	274	(62.8)	170	(39.0)
Total	4,522	2,980	(65.9)	1,790	(60.1)	2,107	(70.7)	2,020	(67.8)
p value	p<0.001			p<0.001		p<0.001		p<0.001	

Note: Registered in CRANE by 13 July 2020. *Exclusions (not mutually exclusive): children who died before the age of 5 years, those with submucous cleft palates, and those with a diagnosed syndrome entered into the CRANE Database. p-values are for differences between cleft types.

Table 4.4 shows that the proportion of children achieving each speech standard varies considerably according to the type of cleft (p<0.001). Children with a cleft affecting only the palate have the most favourable results, while those with a BCLP have the least favourable results. The greatest differences between cleft types were observed for Speech standards 1 and 3, whereby the proportion of children with BCLP meeting the standards was approximately half that observed among children with CP. Given that many children with BCLP will have oral anatomy that predisposes to amber CSCs, this difference should be expected.

Significant differences also exist between cleft types in terms of the proportion of children undergoing secondary surgery for speech purposes (p<0.001). Of those with surgical data (99.1% of children with complete speech data), 15.2% of those with CP, 20.7% of those with UCLP and 25.5% of those with BCLP had undergone velopharangeal surgery or fistula repair before 5 years of age.

Figures 4.10 to 4.12 illustrate the proportion of children meeting each standard according to cleft type and birth year. These figures show that there are large differences between cleft types, especially for Standard 1 and Standard 3, but that for each cleft type, there are only minor fluctuations in the proportion of children meeting each standard by birth year. Children with BCLP appear to have the greatest fluctuations in the proportion of children meeting the standards by year; however, this, in part, may be due to the smaller numbers of children with speech data in this group, which ranged from 48 (for 2013 birth year) to 81 (for 2010 birth year).

Figure 4.10. Percentage of 5-year old CRANE-consented children who were considered to have speech within the normal range (Cleft Speech Standard 1), according to cleft type and birth year.


	2007	2008	2009	2010	2011	2012	2013
CP	69.9%	67.9%	73.2%	74.8%	75.2%	71.0%	71.2%
UCLP	56.5%	52.9%	55.4%	53.7%	51.7%	50.6%	51.3%
BCLP	35.7%	40.8%	31.9%	35.8%	33.9%	40.7%	27.1%
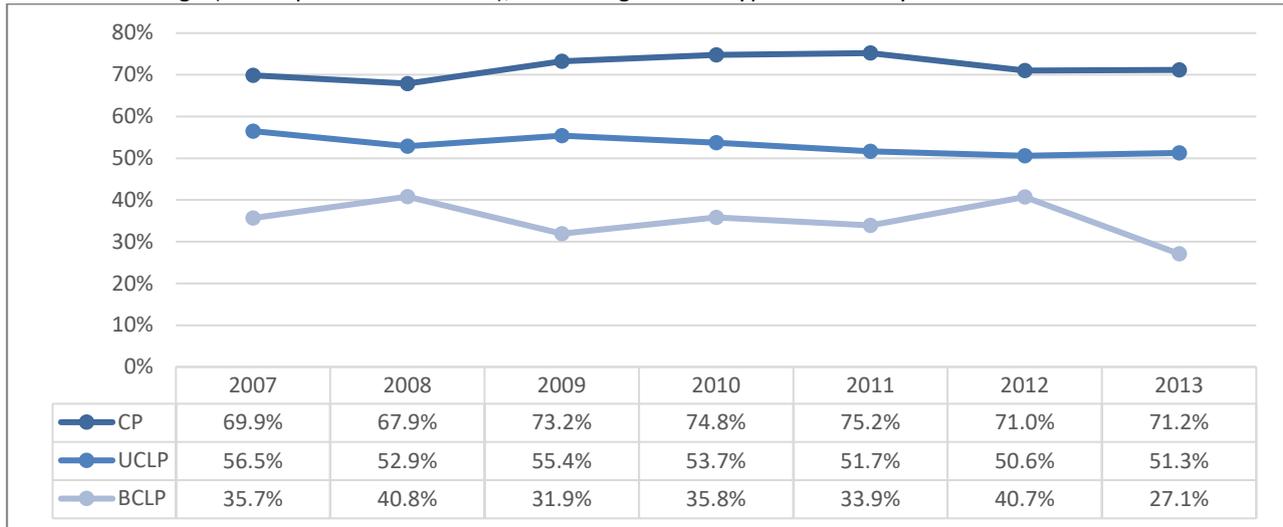
Note: See Table D in Appendix 8 for raw data

Figure 4.11. Percentage of 5-year old CRANE-consented children who did not have any structurally-related speech difficulties (Cleft Speech Standard 2a), according to cleft type and birth year.

	2007	2008	2009	2010	2011	2012	2013
CP	71.8%	68.4%	81.3%	75.2%	78.2%	73.9%	73.9%
UCLP	71.4%	65.2%	62.8%	71.1%	70.3%	67.3%	69.2%
BCLP	65.7%	69.4%	65.3%	56.8%	59.7%	70.4%	54.2%
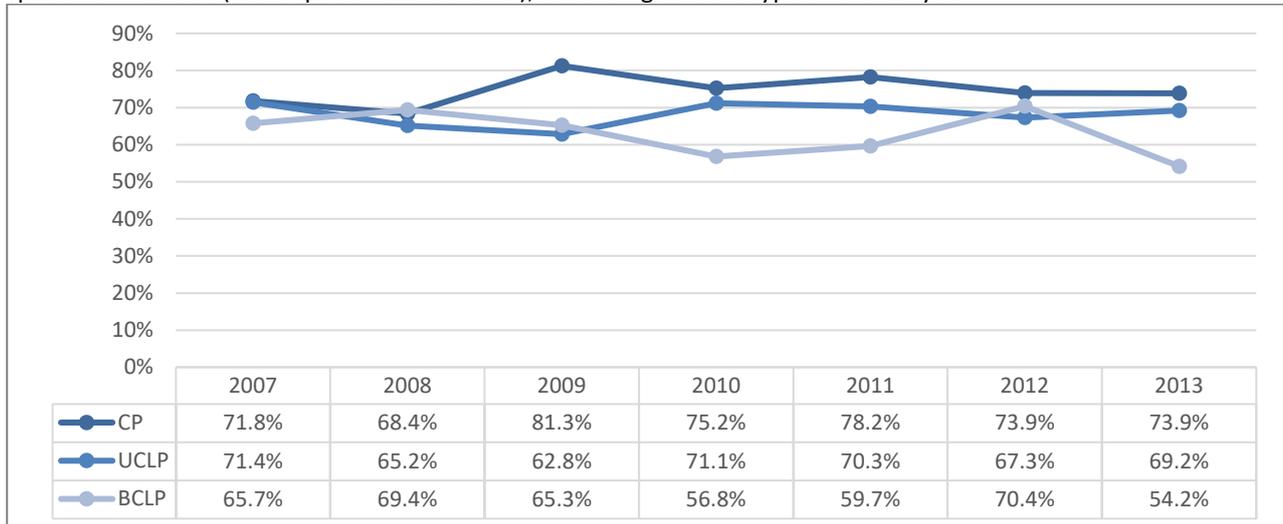
Note: See Table E in Appendix 8 for raw data

Figure 4.12. Percentage of 5-year old CRANE-consented children who did not exhibit any significant cleft-related articulation difficulties (Cleft Speech Standard 3), according to cleft type and birth year.

	2007	2008	2009	2010	2011	2012	2013
CP	77.5%	80.3%	81.3%	82.4%	80.3%	80.3%	79.7%
UCLP	64.9%	61.3%	64.2%	63.1%	58.6%	59.9%	58.1%
BCLP	40.0%	40.8%	37.5%	40.7%	37.1%	42.6%	33.3%
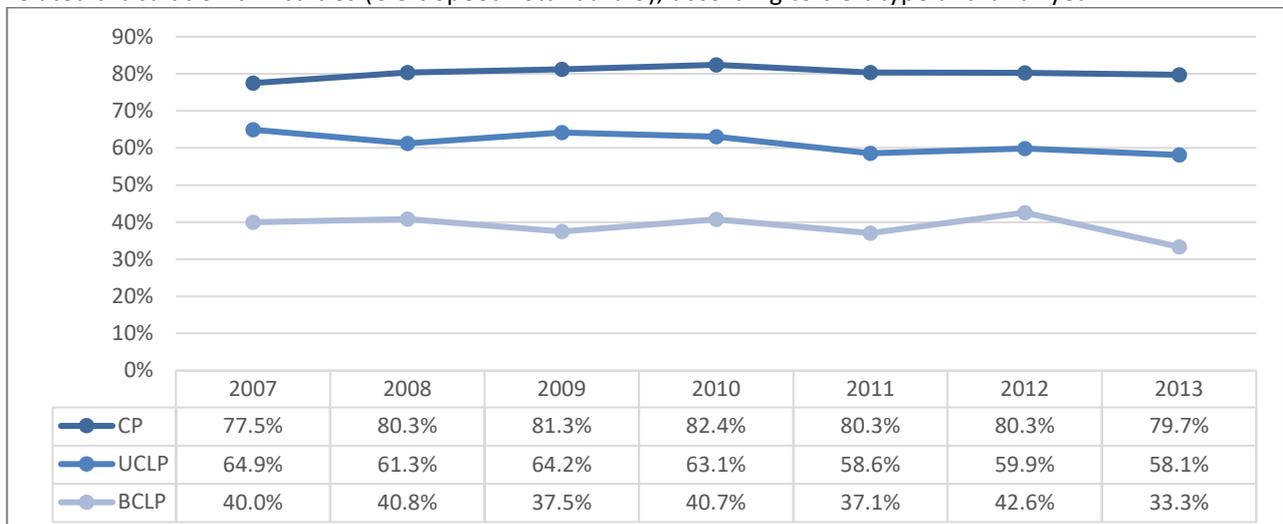
Note: See Table F in Appendix 8 for raw data

The differences in speech outcomes between different cleft types, as presented above, should be taken into account when counselling parents and carers on treatment and outcome expectations. CRANE is planning further work to explore whether the completeness of the cleft also affects speech outcomes.

4.5. Psychology

Cohort Summary	Data source	CRANE Database (consented cases only)
	Birth years	Three years: 2011 to 2013
	Denominators	• 1,739 5-year-old children with recorded psychology scores (with 1 to 9 psychology scores) • 1,490 children with recorded Tiers of Involvement Measure (TIM) scores • 1,463 children with recorded Strengths and Difficulties Questionnaire (SDQ) scores
	Numerators	• 1,678 children with at least one face-to-face psychosocial screen before age 6 • Number of children with a particular TIM level of psychological involvement • Number of children with a particular range of SDQ 'Total difficulties' scores
	Exclusions (not mutually exclusive)	• Children without consent to data collection • Children who died before the age of 5 years • Children with submucous cleft palates • Children with a diagnosed syndrome entered onto the CRANE Database • Children without a recorded date of first face-to-face psychosocial screening
	Data completeness	• 53% of 2,602 eligible children had all psychology scores reported (complete data). • 14% had some but not all 9 scores reported (incomplete data). • 22% had a reason psychology data was not collected. • 11% were missing data or a reason for not collecting data.
	Countries	England, Wales and Northern Ireland
Benchmarks		• 100% of eligible children should have recorded psychology screening data (or a recorded reason outcome not provided). • 100% of eligible children should be screened at least once before the age of 6 years[38]. • 100% of children should be seen by a psychologist and have a TIM assessment at 5 years of age[38]. • SDQ population norms[39]: 10% of children aged 5 to 10 years old have SDQ scores that are 'high' or 'very high'.
What did we find?		• 96% of families were screened at least once before the target age of 6 years. • 86% were seen by a psychologist and a psychosocial screen was completed (TIM tiers 1 to 4). Of these children, 49.1% received psychological support and had their needs met (incl. by other services, TIM tier 1b+), meaning 50.9% had no psychological concerns identified (TIM tier 1a). • 18% of children born with a cleft had 'high' or 'very high' SDQ scores. These percentages are higher than the 10% SDQ population norms. Of those children with a high or very high SDQ score, 57.3% received psychological support and had their needs met (incl. by other services, TIM tier 1b+), meaning 42.7% had no psychological concerns identified (TIM tier 1a).
Recommendations		Cleft care teams should aim to see all children and families at age 5, undertake a psychological screen and ensure psychological support is provided if appropriate (to be recorded as a TIM score).

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[38] 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/
[39] 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.

4.5.1. Data completeness

Participation in a national clinical audit, such as CRANE, means that all regions / cleft care teams are asked to record psychology scores for all consented 5-year-old child with a cleft lip and/or palate. If psychology outcome data are not available, regions / cleft care teams are asked to report a reason for this (e.g. Patient transferred out of area or patient did not attend appointment).

Overall, psychology data have been provided for 1,739 children, representing 66.8% of 2,602 children who were eligible. 22% had a recorded valid reason why the psychological screening could not be audited. 11.2% of those eligible for an audit of their psychological screening were missing data or a valid reason. Appendix 5 shows the breakdown of reason reported for not collecting 5-year old psychology outcome data; the most common reason selected was 'Lack of staff / facilities / equipment' (43.2%).

Over the three birth years that psychological screen data have been collected and submitted to CRANE, the percentage of eligible cases with psychological screen data reported ranged from 68.7% for 2012 births to 65.4% for 2013 births (see Figure 4.13) and did not vary significantly between these years (p=0.88).

Psychology screening outcome data are not subject to the additional validation that some other cleft care outcomes are subject to, such as speech and facial growth. Nevertheless, the onset of the COVID-19 pandemic in early 2020 may have impacted teams' ability to audit and submit the most recent year of CRANE data collection.

Figure 4.13. Percentage of 5-year old CRANE-consented children with a non-syndromic cleft affecting the palate who have full psychology data, incomplete psychology data, a reason for missing psychology data or missing data without a reason, according to birth year.
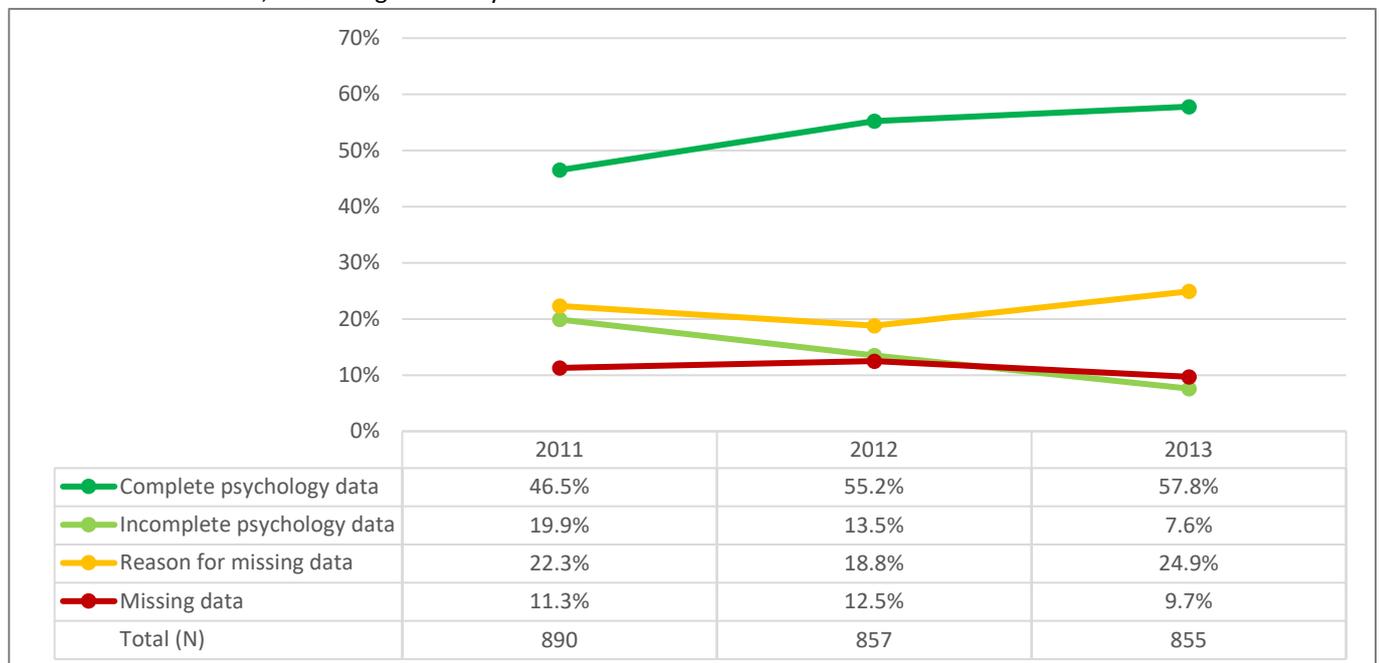
	2011	2012	2013
Complete psychology data	46.5%	55.2%	57.8%
Incomplete psychology data	19.9%	13.5%	7.6%
Reason for missing data	22.3%	18.8%	24.9%
Missing data	11.3%	12.5%	9.7%
Total (N)	890	857	855

Note: Registered in CRANE by 13 July 2020. Exclusions (not mutually exclusive): Children who died before the age of 5 years, children with submucous cleft palates, and those born with a diagnosed syndrome entered into the CRANE Database

4.5.2. Face-to-face psychosocial screening before age 6

Dates of 'psychological screening at age five' were recorded in CRANE. Where this date was unavailable, 'date of first face-to-face screening' was included in the analyses. Based on this information, 1,676 children

were confirmed as having at least one face-to-face psychosocial screen before the age of 6 years (96.5% of the total 1,737 children for whom psychology data have been provided)[40].

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: (a) Introduce and normalise psychology as part of the cleft care team early on, thereby increasing accessibility of psychology services to families, and (b) 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. Overall, 65.4% of families were seen before their child's first birthday. This ranged from 68.1% for 2012 births to 63.5% for 2013 births.

4.5.3. Psychological screening by birth year

Psychological screening outcomes at 5 years defined

The Tiers of Involvement Measure (TIM) records the tier (level) of involvement when a psychologist sees a patient / family in a Cleft Multi-Disciplinary Meeting (MDT) Clinic. Tiers range from 0 to 4 and are as follows:
 0. Patient not seen by Psychologist.
 1. Patient seen and psychosocial screen completed.
    1.a. No psychological concerns requiring cleft psychological input.
    1.b. Psychological support and/or needs met by other services e.g. Child and Adult Mental Health services.
 2. Psychological input provided in clinic.
    2.a. Preventative input only.
    2.b. Input in response to a problem/concern raised by family/child.
    2.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).

The Strengths and Difficulties Questionnaire (SDQ) is a brief behavioural screening questionnaire designed for use with 3-16 year olds. These questionnaires are completed by the parents of CRANE-registered children at 5 years of age.

The SDQ asks about 25 attributes, some positive and others negative, which are divided between scales on: Emotional symptoms, conduct problems, hyperactivity/inattention, peer relationship problems, and prosocial behaviour. The 'Total difficulties' score is calculated from the first four scales listed[41].

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; which may require psychological input or intervention.

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[40] Cases with recorded date of psychosocial screen prior to their date of birth were excluded from reporting.
[41] 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

Tiers of Involvement Measure (TIM) by birth year

Among the 1,490 children with a reported TIM assessment (85.7% of those with reported psychology screening data):

- 49.1% had been seen and had received psychological support and/or had their needs met (incl. met by other services) (TIM tier 1b+, of those with tier 1 to 4 scores). This meant that 50.9% had no psychological concerns identified (TIM tier 1a).
- 15.9% received psychological input in response to a promo/concern raised by family/child or both preventative input and input in response to problem (TIM tiers 2b and 2c only, of those with tier 1 to 4 scores).
- 24.8% needed and received input (excluding preventative input and that met by other services) (TIM tier 2b+, of those with tier 1 to 4 scores). This varied significantly across birth years, with 2011 births having experienced significantly more need for input than the other years audited (29.1%). This will be examined further as numbers of children involved, and years of data collection, grow.
- 4.5% were offered an additional psychology appointment (TIM tier 4, of those with tier 1 to 4 scores).

Typical themes for preventative psychological input provided in clinic would include: advice on talking to your child about their cleft, and helping children and parents to prepare for potential comments and questions about their cleft. Example themes for psychological input in response to a problem/concern raised by family/child include managing difficult behaviour, and concerns about confidence or anxiety.

The fluctuations between birth years of TIM scores are presented in Table 4.5. Of note is that a significant percentage of children audited in the most recent year of data collection, born in 2013, had a completed TIM assessment (93.7%), when compared to the first audit year of 2011 births (80.2%) (p<0.001).

Table 4.5. Percentage of 5-year old CRANE-consented children born 2011-13 who have psychology screening data indicating a specific TIM score, of those with tier 1 to 4 scores, according to birth year.

Birth year	All eligible cases	Eligible cases with psychology scores reported		TIM assessment (TIM tiers 1 to 4, of cases with psychology scores reported)		Input needed (incl. met by other services) (TIM tier 1b+, of 1 to 4)		Input needed in response to a concern, not including preventative input (TIM tiers 2b and 2c only, of 1 to 4)		Input needed (excl. preventative & met by other services) (TIM tier 2b+, of 1 to 4)		Offered additional psychology appointment (TIM tier 4, of 1 to 4)	
	N	N	(%)	N	(%)	N	(%)	N	(%)	N	(%)	N	(%)
2011	890	591	(66.4)	474	(80.2)	227	(47.9)	82	(17.3)	138	(29.1)	22	(4.6)
2012	857	589	(68.7)	492	(83.5)	229	(46.5)	69	(14.0)	99	(20.1)	14	(2.8)
2013	855	559	(65.4)	524	(93.7)	276	(52.7)	86	(16.4)	133	(25.4)	31	(5.9)
Total	2,602	1,739	(66.8)	1,490	(85.7)	732	(49.1)	237	(15.9)	370	(24.8)	67	(4.5)
p-value		*p=0.32*		*p<0.001*		*p=0.120*		*p=0.352*		*p=0.005*		*p=0.061*	

Note: Registered in CRANE by 13 July 2020. Exclusions (not mutually exclusive): Children who died before the age of 5 years, with submucous cleft palates, and those born with a diagnosed syndrome entered into the CRANE Database. p-values are for differences between birth years.

Strengths and Difficulties Questionnaire (SDQ) by birth year

Among the 1,463 children with reported SDQ Total scores (84.1% of those with reported psychology screening data) 17.9% had high/very high SDQ Total scores indicating difficulties. This percentage is higher

than the 10% population norm[42] (although consideration should be given to the fact that children at age 5 are at the younger end of the population norms sample of 5-10 year olds) [43].

Of the children with high/very high SDQ Total scores:

- 57.3% had been seen and received psychological support and/or had their needs met (incl. met by other services), as scored by TIM tier 1b+ (of those with tier 1 to 4 scores). This meant that 42.7% had no psychological concerns identified (TIM tier 1a). This varied significantly across birth years, with 2011 births having been seen and received support and/or had their needs met more than other audited birth years (in 70.6% of cases). Given the small numbers of children involved and with only three years of data, interpretation should be undertaken with caution.
- 36.3% needed and received psychological input (excluding preventative and input met by other services), as scored by TIM tier 2b+ (of those with tier 1 to 4 scores).

Compared to the wider group, a greater percentage of children scoring high or very high on the SDQ were identified by the psychologist in clinic as needing psychological input (both preventative and in response to a concern raised).

The fluctuations between birth years for these outcomes for children with high/very high SDQ Total scores are presented in Table 4.6. Of note is that a significant percentage of children audited in the most recent year of data collection, born in 2013, had a completed SDQ assessment (91.6%), when compared to the first audit year of 2011 births (78.2%) (p<0.001).

In addition, the variation in the percentage of children with high/very high SDQ scores and TIM tier 2b+ scores was statistically significant (p<0.001). Over half of children born in 2011 (52.9%) needed and received psychological input (TIM tier 2b+); at least 15% more that in the other birth years of 2012 and 2013. Given the small numbers of children involved and with only three years of data, interpretation should be undertaken with caution.

Table 4.6. Percentage of 5-year old CRANE-consented children born 2011-13 who have high/very high SDQ Total scores and psychological input as defined by TIM scores, according to birth year.

Birth year	All eligible cases	Eligible cases with psychology scores reported		SDQ Total scored (of cases with psychology scores reported)		High/very high SDQ Total scores		TIM tier 1b+ of children with high/very high SDQ Total scores		TIM tier 2b+ of children with high/very high SDQ Total scores	
	N	N	(%)	N	(%)	N	(%)	N	(%)	N	(%)
2011	890	591	(66.4)	462	(78.2)	68	(14.7)	48	(70.6)	36	(52.9)
2012	857	589	(68.7)	489	(83.0)	84	(17.2)	36	(42.9)	18	(21.4)
2013	855	559	(65.4)	512	(91.6)	110	(21.5)	66	(60.0)	41	(37.3)
Total	2,602	1,739	(66.8)	1,463	(84.1)	262	(17.9)	150	(57.3)	95	(36.3)
p-value		p=0.32		p<0.001		p=0.02		p=0.002		p<0.001	

Note: Registered in CRANE by 13 July 2020. Exclusions (not mutually exclusive): Children who died before the age of 5 years, with submucous cleft palates, and those born with a diagnosed syndrome entered into the CRANE Database. p-values are for differences between birth years.

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[42] 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.

[43] The percentage of children with high/very high SDQ Total scores indicating difficulties has increased significantly in recent years (ranging from 14.7% in 2011 to 21.5% in 2013, p<0.02). The significant improvement in reporting of SDQ Total scores could be a contributing factor. Given the small numbers of children involved and with only three years of data, interpretation should be undertaken with caution.

4.5.4. Psychological screening by cleft type

Tiers of Involvement Measure (TIM) by cleft type

When all birth years were combined, data completeness was found to vary significantly between cleft types (p<0.001). Data completeness was higher for children with UCLP (72.2%) and BCLP (71.1%), compared to children with CP (66.3%) and CL (62%).

Variation between cleft types for the four TIM outcomes considered are presented in Table 4.7. This was significant for children who had been seen and received psychological support and/or had their needs met (TIM tier 1b+) (p<0.01). Specifically, significantly fewer children with CP (at least 10%) only had psychological support needs identified and met (TIM tier 1b+), compared with children with CL only or cleft lip and palate (UCLP or BCLP). At age 5, children with CP only are less likely to have any visible features of their cleft, than those with a CL only or cleft lip and palate. Those with a cleft affecting the lip are more likely to receive preventative input, or have concerns, in relation to comments and questions from other people, which may account for this difference.

This was also significant for children that needed and received input (excluding preventative input and that met by other services) (TIM tier 2b+, of those with tier 1 to 4 scores) (p=0.016). Specifically, significantly fewer children with CL, compared with children with clefts involving the palate (CP, UCLP and BCLP), needed and received this type of input. This will be examined further as numbers of children involved, and years of data collection, grow.

Table 4.7. Number and percentage of 5-year old CRANE-consented children, born 2011-13, with psychology data, according to Tiers of Involvement Measure (TIM) scores and cleft type.

Cleft type	All eligible cases	Eligible cases with psychology scores reported		TIM assessment (TIM tiers 1 to 4, of cases with psychology scores reported)		Input needed (incl. met by other services) (TIM tier 1b+, of 1 to 4)		Input needed in response to a concern, not including preventative input (TIM tiers 2b and 2c only, of 1 to 4)		Input needed (excl. preventative & met by other services) (TIM tier 2b+, of 1 to 4)		Offered additional psychology appointment (TIM tier 4, of 1 to 4)	
	N	N	(%)	N	(%)	N	(%)	N	(%)	N	(%)	N	(%)
CL	689	427	(62.0)	361	(84.5)	195	(54.0)	48	(13.3)	69	(19.1)	14	(3.9)
CP	1037	688	(66.3)	579	(84.2)	242	(41.8)	87	(15.0)	143	(24.7)	25	(4.3)
UCLP	609	440	(72.2)	388	(88.2)	204	(52.6)	70	(18.0)	109	(28.1)	23	(5.90
BCLP	239	170	(71.1)	149	(87.6)	84	(56.4)	32	(21.5)	44	(29.5)	5	(3.4)
Total	2,574	1,725	(67.0)	1,477	(85.6)	725	(49.1)	237	(16.0)	365	(24.7)	67	(4.5)
p-value		p<0.001		p=0.08		p<0.001		p=0.076		p=0.016		p=0.451	

Note: Registered in CRANE by 13 July 2020. Exclusions (not mutually exclusive): Children who died before the age of 5 years, with submucous cleft palates, and with syndromic diagnoses. p-values are for differences between cleft types.

Strengths and Difficulties Questionnaire (SDQ)

Outcomes for children with high/very high SDQ scores are presented by cleft type in Table 4.8. Of interest is that children with CL experienced significantly lower rate of high/very high SDQ scores, compared with children with clefts involving the palate (CP, UCLP and BCLP) (p=0.001). In fact the rate of high/very high SDQ scores in children with CL came close to the population norm of 10%. This suggests that the presence of CL only (repaired) adds little psychological burden for the child, *specifically at age 5*, as measured by the SDQ. However, the more individualised TIM score, which takes account of the psychological needs of the

child and the family, indicates that 13% of the CL only group received input in the clinic in response to a concern raised by the child or family, and a further 6% needed further input after the clinic.

Children with CP had the lowest rate of TIM tier 1b+. The higher rate of high and very high SDQ scores among those children with a cleft palate (with or without a cleft lip) may be linked to the increased likelihood of other difficulties, such as with hearing and speech, which are generally viewed as risk factors for more difficult behaviour in young children. This is consistent with lower rates of TIM tier 2b+ which indicates psychological input given in response to a concern raised.

The lower rates of TIM tier 1b+ for those children with CP only, may be linked to less preventative input provided around responding to other people's comments and questions, as mentioned above.

Table 4.8. Number and percentage of 5-year old CRANE-consented children, born 2004-13, with high/very high SDQ scores, according to Tiers of Involvement Measure (TIM) scores and cleft type.

Cleft type	All eligible cases	Eligible cases with psychology scores reported		SDQ Total scored (of cases with psychology scores reported)		High/very high SDQ Total scores		TIM tier 1b+ of children with high/very high SDQ Total scores		TIM tier 2b+ of children with high/very high SDQ Total scores	
	N	N	(%)	N	(%)	N	(%)	N	(%)	N	(%)
CL	689	427	(62.0)	361	(84.5)	41	(11.4)	29	(70.7)	13	(31.7)
CP	1037	688	(66.3)	571	(83.0)	112	(19.6)	54	(48.2)	39	(34.8)
UCLP	609	440	(72.2)	379	(86.1)	82	(21.6)	49	(59.8)	32	(39.0)
BCLP	239	170	(71.1)	144	(84.7)	24	(16.7)	15	(62.5)	9	(37.5)
Total	2,574	1,725	(67.0)	1,455	(84.3)	259	(17.8)	147	(56.8)	93	(35.9)
p-value	*p<0.001*			*p=0.562*		*p=0.001*		*p=0.065*		*p=0.863*	

Note: Registered in CRANE by 13 July 2020. Exclusions (not mutually exclusive): Children who died before the age of 5 years, with submucous cleft palates, and those born with a diagnosed syndrome entered into the CRANE Database. p-values are for differences between cleft types.

5. Research

This chapter details the research activity undertaken by the CRANE project team over the last 12 months.

The sections in this chapter report on (1) the estimated incidence of clefting across England, Wales and Northern Ireland, (2) speech outcomes at 5 years in relation to timing and number of primary palate repair surgeries, and (3) the relationship between dental health at 5 years and deprivation.

Timeframe: (1) Cleft incidence rates were calculated using data for children born between 1 January 2010 and 31 December 2019. (2) Speech outcomes at 5 years are reported for children born between 1 January 2006 and 31 December 2012. (3) Dental outcomes at 5 years are reported for children born between 1 January 2004 and 31 December 2012.

Legal basis for data collection and analysis: (1) The data used for the calculation of incidence is collected for all registered cases under a 'Section 251' exemption (of the NHS Act 2006 and its current regulations, the Health Service (Control of Patient Information (CPI) Regulations 2002), with approval from the Confidentiality Advisory Group (CAG) for the disclosure of CPI held by the CRANE Database. (2 & 3) The data used for analysing 5-year old outcomes is collected for all children whose families have given informed consent to outcomes data collection by the CRANE Database.

5.1. Incidence of cleft

Cohort summary	Data sources	CRANE Database, Office for National Statistics (ONS England & Wales)[44], Northern Ireland Statistics and Research Agency (NISRA)[45]
	Birth years	Ten years: 2010 to 2019
	Denominator	7,182,361 children born in the general population of England, Wales and Northern Ireland
	Numerators	• 10,765 children born with a cleft and registered in CRANE Database • 10,093 children with their cleft type specified in the CRANE Database • 10,595 children with their sex recorded in the CRANE Database
	Data completeness	• 94% of all CRANE-registered children (n=10,765) had their cleft type specified in CRANE. • 98%% of all CRANE-registered children (n=10,765) had their sex specified in CRANE.
	Countries	England, Wales and Northern Ireland
What did we find?		• The overall estimate for the incidence of cleft for the investigated period is 15.0 per 10,000 live births. • CP was the most common cleft type (Estimated incidence for the period 6.3 per 10,000 live births). • Incidence of each cleft type remained relatively stable throughout the period investigated. • Estimated Incidence of cleft was higher among girls than boys over the investigated period (17.0 per 10,000 live female births vs. 13.0 per 10,000 live male births).
Recommendations		The research community should undertake to validate these findings using the HES dataset and case ascertainment techniques.

5.1.1. Methods

Data sources

Analyses presented in this section used three sources of national data. First, the Cleft Registry and Audit Network (CRANE) database was used to identify all children born with a cleft in England, Wales, and Northern Ireland between 2010 and 2019. Second, data from the Office for National Statistics was used to calculate all live births in England and Wales during the specified time period. Third, data from Northern Ireland Statistics and Research Agency was used to calculate all live births in Northern Ireland.

Study cohort and patients

The study cohort consisted of 10,765 children born between 1 January 2010 and 31 December 2019 who were diagnosed with a cleft affecting the lip and/or palate and were registered in the CRANE Database by 13 July 2020. The population size for all live births in England, Wales and Northern Ireland across the same time period was estimated at 7,182,361 children.

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[44] Office for National Statistics: Annual summary statistics on live births and stillbirths in England and Wales, 2019. Released July 2020 (checked October 2020). Available at:
https://www.ons.gov.uk/peoplepopulationandcommunity/birthsdeathsandmarriages/livebirths/datasets/birthsummarytables
[45] Northern Ireland Statistics and Research Agency: Births, Deaths and Marriages in Northern Ireland. Released February 2020 (checked October 2020). Available at: https://www.nisra.gov.uk/publications/birth-statistics

Of the 10,765 children identified in CRANE, 93.8% (n=10,093) had their cleft type described as cleft lip (CL), cleft palate (CP), unilateral cleft lip and palate (UCLP), or bilateral cleft lip and palate (BCLP) in the database, while 98.4% (n=10,595) had their sex specified in the database.

Analysis

Incidence of cleft is estimated as the ratio of the number of children registered on the CRANE Database to the number of children born in England, Wales and Northern Ireland over the same period. Incident rates are estimated for each birth year between 2010 and 2019 to describe fluctuations in cleft incidence over time. Variation in incidence over time was compared by cleft type and by sex using the relative standard deviation (RSD) of the rates (calculated as the ratio of the standard deviation to the mean, and expressed as a percentage).

5.1.2. Results

Overall incidence of cleft

For the period covering 2010 to 2019, incidence of cleft in England, Wales and Northern Ireland is estimated at 15.0 per 10,000 live births. The incidence by year for the period varied from a peak of 16.0 in 2013 to a low of 13.5 in 2019 (see Figure 5.1 below). 2019 data should be interpreted with caution as late registrations often occur up to and after primary surgery which for those born in December 2019 with palatal involvement could be as late as January 2021 and hence after the data extract was taken to begin analysis and preparation of this report.

Figure 5.1. Incidence of cleft per 10,000 live births based on CRANE registrations and all live births from 1 January 2010 to 31 December 2019 in England, Wales and Northern Ireland, according to birth year.

	2010	2011	2012	2013	2014	2015	2016	2017	2018	2019
Annual incidence per 10,000	14.2	14.8	15.5	16.1	14.9	15.5	14.9	15.4	15.0	13.5
10-year mean incidence per 10,000	15.0	15.0	15.0	15.0	15.0	15.0	15.0	15.0	15.0	15.0
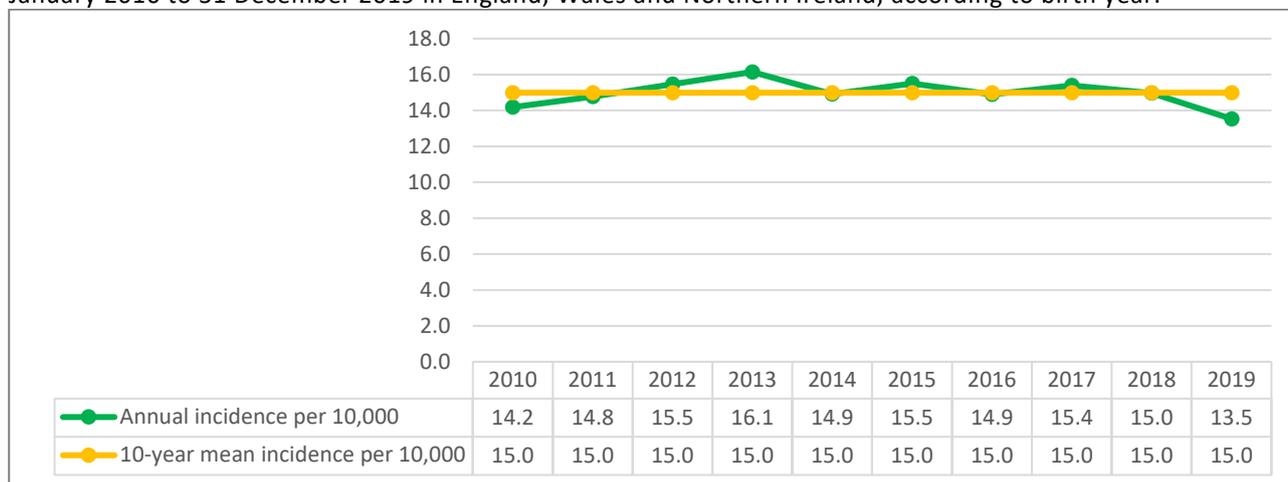
Females continue to have a higher incidence of clefting. The estimated incidence of clefting in female births is 17.0 per 10,000 live female births compared to 13.0 per 10,000 live male births for the same period. This difference reflects that cleft palates are the most common cleft type, constituting 45% of this cohort and that children born with cleft palate only are more likely to be female. (See Figure 5.2). Relative variability in incidence rates over time was similar in male and female births (RSD 5.1% in male births, 5.4% in female births).

Figure 5.2. Incidence of cleft per 10,000 live births based on CRANE registrations and all live births from 1 January 2010 to 31 December 2019 in England, Wales and Northern Ireland, according to birth year and sex.


	2010	2011	2012	2013	2014	2015	2016	2017	2018	2019
Incidence per 10,000 boys	12.4	12.7	13.5	13.5	11.7	12.7	12.6	13.0	12.8	11.5
Incidence per 10,000 girls	16.0	16.0	16.9	18.6	17.5	17.8	16.9	17.6	16.7	15.5
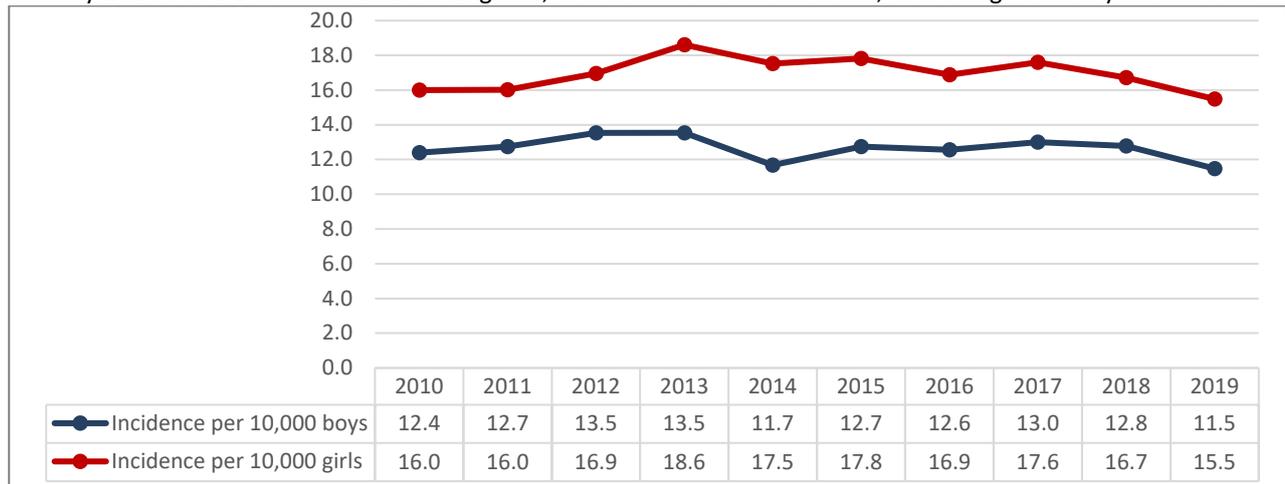
Overall incidence by cleft type

Of the 10,765 children registered in CRANE Database between 2010 and 2019, 94% of them (n=10,073) had a specified cleft type in the database. The estimated incidence of cleft for the period was highest among children born with CP (6.3 per 10,000 births), followed by CL (3.4 per 10,000 births), UCLP (3.1 per 10,000 births) and BCLP (1.4 per 10,000 births). The discrepancy between the addition of incidence for each cleft type and the overall estimated incidence of cleft represents the cases for which no cleft type was recorded and is equivalent to 0.9 per 10,000 live births (15.0 (overall estimate) vs. 14.1 (the sum of estimates for each cleft type)).

Figure 5.3 below shows the change over time in estimated incidence per 10,000 live births for each cleft type. Between 2010 and 2019, incidence of CL, CP, UCLP and BCLP per 10,000 live births in England, Wales and Northern Ireland fluctuated by a small amount year to year. Relative variability in incidence rates was greatest for BCLP (RSD 11.5%), followed by CL (8.5%), CP (7.2%) and UCLP (6.9%).

Figure 5.3. Incidence of cleft per 10,000 live births between 1 January 2010 and 31 December 2019, according to year and cleft type.

	2010	2011	2012	2013	2014	2015	2016	2017	2018	2019
CL	3.3	3.4	3.4	4.0	3.5	3.5	3.2	3.0	3.0	3.2
CP	6.2	6.5	6.7	7.1	6.3	6.3	5.9	6.2	5.9	5.4
UCLP	2.9	3.0	3.4	3.2	2.9	3.2	2.8	3.1	3.2	2.7
BCLP	1.5	1.2	1.3	1.5	1.4	1.2	1.3	1.6	1.6	1.2
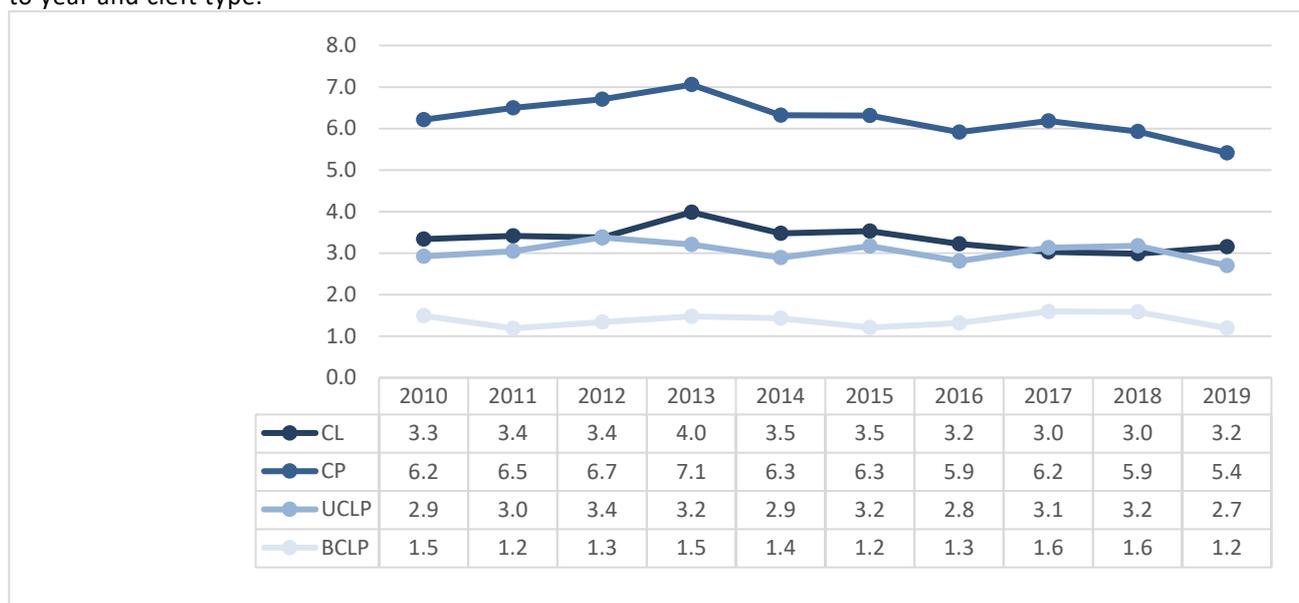
Note: Registered in CRANE by 13 July 2020. Children with a non-specified cleft type are excluded.

The data used in this incidence estimation represent live born children who are registered in CRANE under the care of a cleft team. CRANE does not register stillborn children who are identified as having a cleft or births of children who survive only for short times where the cleft team is not notified of the diagnosis as the child is not expected to live long enough for assessment / clinical input. Estimated relative incidence rates of clefting in this section are not absolute and should be interpreted carefully. The proportion of children for whom cleft type could be ascertained decreased over time (from 99% in 2010 to 92% in 2019), causing an underestimation of cleft type-specific incidence for the period.

Further research should be undertaken to validate cleft incidence over the last ten years. This case ascertainment is possible using HES data and we propose to do this at intervals to ensure that the CRANE dataset registrations continue to reflect all live born children with a cleft in the UK. Ongoing work in collaboration with the Cleft Collective will also allow us to validate cleft type for cases that are present in both data resources and where explicit consent for this linkage has been obtained by the Cleft Collective.

5.2. Timing and number of primary palate repair procedures and speech at 5 years

Cohort summary	Data source	CRANE Database (consented cases only) linked with Hospital Episode Statistics (HES)
	Birth years	Seven years: 2006 to 2012* *Linkage between HES and CRANE data only available up until 2012.
	Denominator	1,653 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
	Exclusions (not mutually exclusive)	• Children without consent to data collection • Children with an unspecified cleft type or with a cleft affecting only the lip • Children who died before the age of 5 years • Children with submucous cleft palates[46] • Children with diagnosed syndromes identified in their HES records • Children without a full set of CAPS-A parameters being reported • Children without a primary palate repair record in their HES history
	Countries	England
Aim		To explore the relationship between speech outcomes at 5 years of age and the timing of primary palate repair and the number of primary palate repairs undertaken to close the cleft.
Exposure		• Timing of the last primary palate repair surgery • Number of primary palate repair surgeries performed to close the cleft
Standard/outcomes		• The achievement of speech within the normal range (see text below for a full description of these standards[47]) • The achievement of speech without difficulties resulting from existing or previous structural anomalies • The achievement of speech without significant cleft-related articulation difficulties
What did we find?		The most favourable cleft-related speech outcomes were observed in children undergoing: • their last primary palate repair before 13 months of age, and • just one primary palate repair procedure on the palate. An increasing number of palate repair surgeries was associated with poorer outcomes.
Recommendations		• Children should have their palate repaired before they are 13 months old. • Where possible, surgeons should aim to repair the cleft palate in one procedure. • The research community should validate these findings whilst controlling for potential confounders.

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[46] Submucous cleft palate patients excluded from all 5 year outcomes as all/most teams do not audit these patients.
[47] 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).

5.2.1. Methods

Data sources, record linkage and study cohort

This retrospective cohort study used two national datasets, linked at an individual level using name, date of birth, postcode and National Health Service (NHS) number. The study cohort was identified in the Cleft Registry and Audit Network (CRANE) database. Children whose parents consent to CRANE linking their child's records to other data sources (~93% of CRANE registrations) had their records linked to the Hospital Episode Statistics (HES) database, which contains records on all admissions to NHS hospitals in England. Diagnostic information is coded using the *International Classification of Diseases 10th revision* (ICD-10) and procedure information is classified according to codes from the *Classification of Surgical Operations and Procedures 4th Revision* (OPCS-4).

Cohort

Speech data are available in CRANE for children with a non-submucous cleft affecting the palate who were born from 2006 onwards. The CRANE-HES linked dataset used for analyses contained records for children born up to 2012 and admissions up to 2015. We therefore restricted our study cohort to CRANE-consented children with cleft palate (CP), unilateral cleft lip and palate (UCLP) or bilateral cleft lip and palate (BCLP), who were born between 1 January 2006 and 31 December 2012, and who were still alive on their fifth birthday. Children were included in analyses if (1) their crane records were successfully matched with HES records (n=4,251), (2) they had no record of additional diagnoses or syndromes in their HES history (listed in Appendix 4), as these could influence speech outcomes (n=2,875), (3) both data sources showed agreement on cleft type, based on LASHAL code in CRANE and ICD-10 codes in HES (n=2,667), (4) complete five-year old speech data were available (n=1,755), and (5) there was a HES record of a primary palate repair (OPCS-4 F291) before the age of three years. In total, 1,653 children were included in analyses. Of these, 766 had CP, 620 had UCLP, and 267 had BCLP.

Primary palate repairs

Primary palate repairs were identified in HES using OPCS-4 code F291. Every episode containing this procedure was flagged and the start date of that hospital episode was used to calculate the time of repair in relation to the child's age. As some children undergo multiple primary palate repairs, the age of the last primary palate repair was used to reflect the age of the child at the time the palate repair had been completed.

Speech outcomes

Speech was evaluated using Cleft Audit Protocol for Speech – Augmented (CAPS-A) scores collected when the child was between 5 years 0 days and 5 years 364 days old.[1] The CAPS-A includes 16 parameters, which are listed in Appendix 8 (Table G), along with their potential scores. The CAPS-A scores are used to determine whether a child meets three nationally agreed cleft speech standards:[2]

1.   The achievement of normal speech. Children meeting this must achieve normal (green) scores across all 16 CAPS-A parameters.

2a. The achievement of speech without difficulties likely to be the result of existing or previous structural anomalies. Children meeting this must have no reported history of surgery for speech purposes and they must achieve normal (green) scores across hypernasal resonance, both nasal airflow parameters, and all three passive CSCs, as listed in Table G of Appendix 8.

3.   The achievement of speech without cleft-related articulation difficulties. Children meeting this standard must achieve normal (green) scores across all anterior oral CSCs, both posterior oral CSCs, all non-oral CSCs and gliding of fricatives, as listed in Table G of Appendix 8.

Analyses

Children were grouped according to when their last primary palate repair took place: <6 months, 6 to <13 months, and 13 to <36 months. We did not include primary palate repair records occurring beyond three years of age in case they were episodes of secondary surgery for speech purposes, incorrectly coded as a primary palate repair.

Percentages describing achievement of standards were compared between groups with chi squared tests. A p-value <0.05 was considered to indicate a statistically significant result. All statistical analyses were conducted using Stata V.15 (StataCorp, College station, Texas, USA).

Ethical Considerations

The study was exempt from NHS Health Research Authority ethics approval as it involves the analysis of a dataset that is collected for the purpose of service evaluation and is linked with parental consent to other datasets.[4]

5.2.2. Results

Timing of last primary palate repair procedure

Of the 1,653 children included in analyses, 13% had their palate repaired by the time they were 6 months old, 78% had their cleft palate repair completed between 6 months and <13 months of age, and 9% had their last primary palate repair between the age of 13 months and <36 months.

Figure 5.4 Percentage of children, born 2006 to 2012 with a non-syndromic cleft affecting the palate, meeting each Cleft Speech Standard according to the timing of their last primary palate repair.
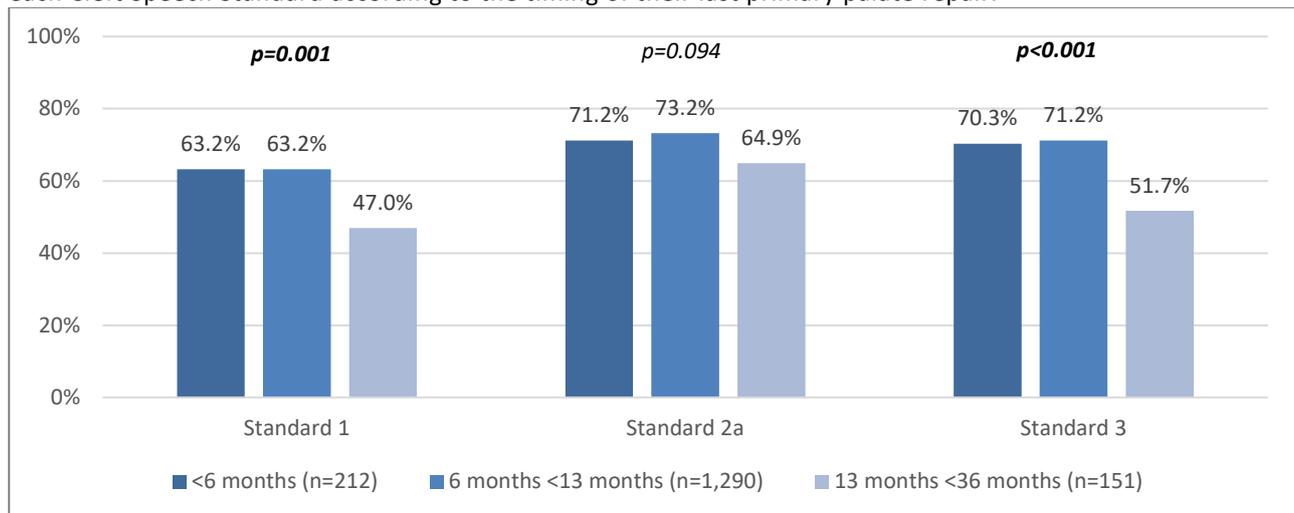
Figure 5.4 shows the percentage of children achieving each cleft speech standard according to their age at the time of their last primary palate repair. While there were no differences in percentages between those undergoing repairs <6 months and those undergoing surgery between 6 and 12 months, a significantly smaller percentage of children undergoing their last primary palate repair beyond 13 months of age achieved 'normal speech' (Standard 1) and achieved speech that was free of cleft-related articulation difficulties (Standard 3) (p<0.01). Differences in the percentage of children achieving speech that was free of structurally-related difficulties (Standard 2a) did not vary significantly according to the timing of repair (p=0.094).

Since we have previously reported differences in the achievement of cleft standards according to cleft type[48], we sought to determine whether the association between standards and timing of surgery was similar across the three cleft type subgroups.

For children with CP, significantly better outcomes were obtained when palate repairs were completed before 13 months and this was the case for all three speech standards (Figures 5.4 to 5.6). While differences did not reach statistical significance for UCLP and BCLP groups, there does appear to be a trend for better outcomes with Standard 1 (Figure 5.5) and 3 (Figure 5.7) when repairs were performed earlier rather than later. There is a weaker association between timing of repair and achieving Standard 2a for those with a cleft affecting both the palate and lip (Figure 5.6).

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[48] CRANE Project team on behalf of the Cleft Development Group (2019) *CRANE Database Annual Report 2019*. London: Clinical Effectiveness Unit, The Royal College of Surgeons of England.

Figure 5.5 Percentage of non-syndromic children, born 2006 to 2012, meeting Cleft Speech Standard 1 (normal speech), according to cleft type and timing of last primary palate repair.
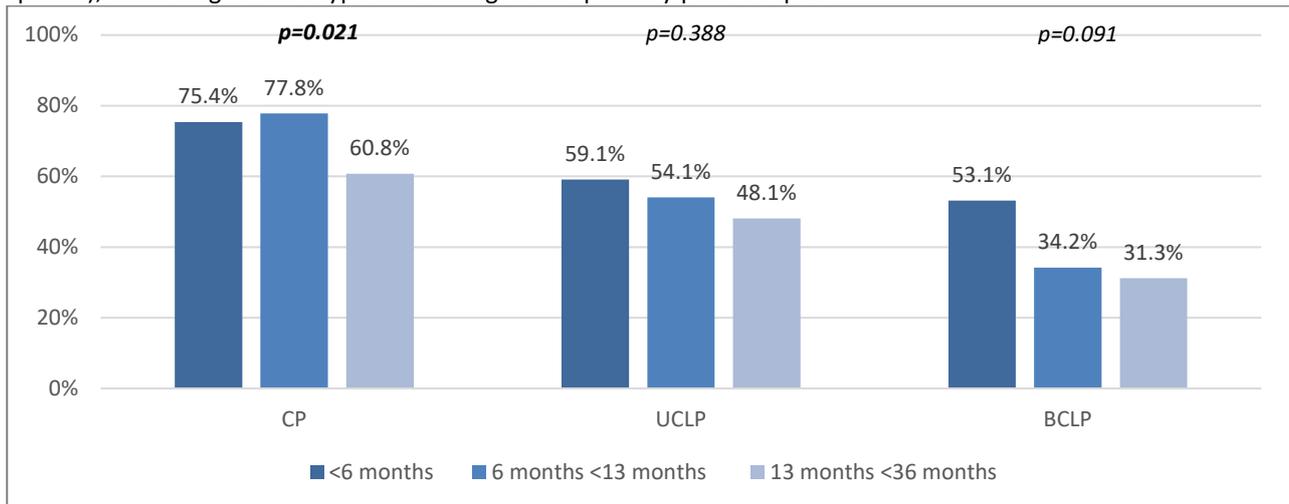

Note: See Table I in Appendix 8 for raw data

Figure 5.6. Percentage of non-syndromic children, born 2006 to 2012, meeting Cleft Speech Standard 2a (absence of structurally-related speech difficulties), according to cleft type and timing of last primary palate repair.
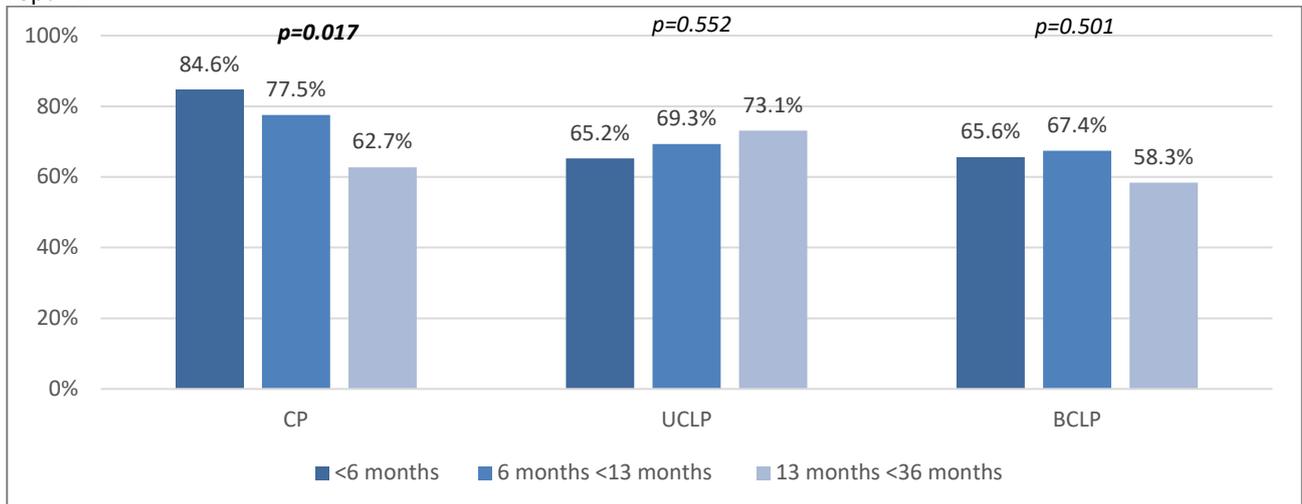
Note: See Table I in Appendix 8 for raw data

Figure 5.7. Percentage of non-syndromic children, born 2006 to 2012, meeting Cleft Speech Standard 3 (absence of cleft-related articulation difficulties), according to cleft type and timing of last primary palate repair.
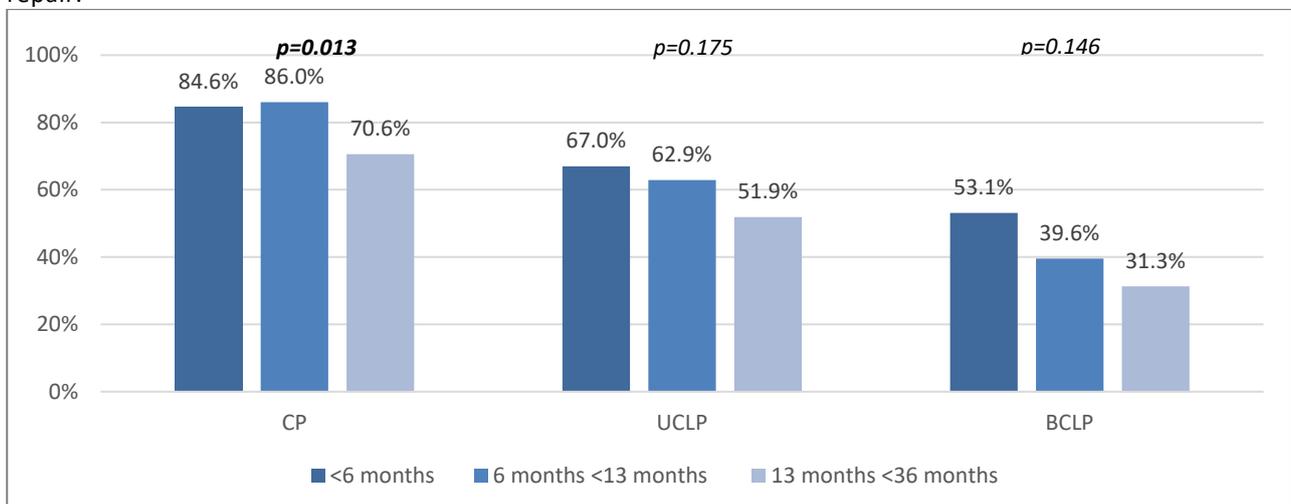
Note: See Table I in Appendix 8 for raw data

## Number of primary palate repair procedures

Of the 1,653 children included in analyses, 77% had just one primary palate repair, 22% had two primary palate repairs and just 0.2% had three primary palate repairs. Those undergoing three primary palate repairs also had a cleft affecting their lip. Table 5.1 shows that children undergoing just one primary palate repair were significantly more likely to achieve each cleft speech standard compared to those undergoing more than one repair.

Table 5.1. Number and % of children, born 2006 to 2012 with a non-syndromic cleft affecting the palate, meeting each Cleft Speech Standard according to the number of primary palate repairs they received before 36 months of age.

Number of primary palate repair within 24 months	Total N	STANDARD 1		STANDARD 2a		STANDARD 3	
		N	(%)	N	(%)	N	(%)
1 repair	1,264	848	(67.1)	942	(74.5)	940	(74.4)
2 repairs	365	169	(46.3)	238	(65.2)	201	(55.1)
3 repairs	24	<5	(12.5)	13	(54.2)	<5	(16.7)
Total	1,653	1,020	(61.7)	1,193	(72.2)	1,145	(69.3)
p value		***<0.001***		***<0.001***		***<0.001***	

Figure 5.8 shows the percentage of children meeting each cleft speech standard according to cleft type and number of primary palate repairs. In general, there was a trend for poorer speech outcomes with increasing number of primary palate repairs. Differences in the percentage of children meeting standard 1 according to the number of repairs reached statistical significance for those with UCLP only. For Standard 2a there were significant differences for those with CP and those with UCLP.

Figure 5.8. Percentage of non-syndromic children, born 2006 to 2012, meeting each of the Cleft Speech Standards, according to cleft type and number of primary palate repair procedures performed before 36 months of age.
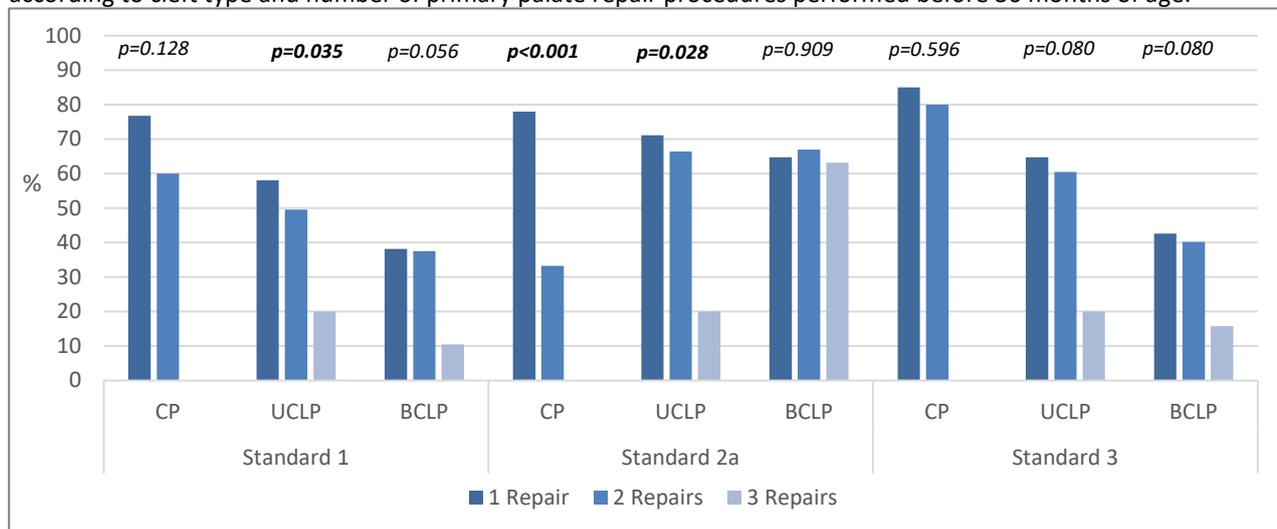
Note: Raw data are not presented in appendices due to General Data Protection Regulation (GDPR) requirements

Further work is being undertaken to explore the relationship between speech and the timing of repair and number of repairs whilst controlling for confounders such as the completeness of the cleft.

5.3. Relationship between dental health and deprivation at 5 years

Cohort summary	Data source	CRANE Database (consented cases only) linked with Hospital Episode Statistics (HES)
	Birth years	Nine years: 2004 to 2012* *Linkage between HES and CRANE data only available up until 2012.
	Denominators	• 4,083 children with recorded decayed, missing or filled teeth (dmft) scores and HES data • 3,926 children with scores for the calculation of Treatment Index[49] • 3,934 children with scores for the calculation of Care Index[50]
	Numerators	• 1,663 children with at least one (>0) dmft • 581 children with extensive decay (dmft >5) • Number of children within each deprivation quintile
	Exclusions (not mutually exclusive)	• Children without consent to data collection • Children with an unspecified cleft type • Children who died before the age of 5 years • Children with submucous cleft palates[51] • Children without a recorded dmft score and deprivation data at the age of 5
	Countries	England (Deprivation data were not available for children in Wales and Northern Ireland at the time of producing this report).
Aim		Explore the impact of deprivation on dental health among children with a cleft at 5 years of age
Exposure		Deprivation quintiles, assessed according to Indices of Multiple Deprivation (IMD)
Standard/outcomes		• Number of decayed, missing or filled teeth a child has at 5 years • Treatment Index and Care Index (see main text for full description of these)
What did we find?		• Rates of poor dental health (expressed by rates of dmft >0 and dmft >5) increased significantly as deprivation increased. • The proportion of treated dental disease and Care Index scores increased significantly as deprivation decreased.
Recommendations		Cleft care teams should have mechanisms in place to identify and target children from the most deprived areas to ensure they receive help, such as Sure Start, to facilitate their access to appropriate dental care and advice.

5.3.1. Methods

Data sources, record linkage and study cohort

This retrospective cohort study used two national datasets, linked at the patient-level using name, date of birth, postcode and National Health Service (NHS) number. The study cohort was identified in the Cleft Registry and Audit Network (CRANE) Database. Children whose parents consent to CRANE linking their child's records to other data sources (~93% of CRANE registrations) had their records linked to the Hospital Episode Statistics (HES) database, which contains records on all admissions to NHS hospitals in England.

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[49] Treatment index calculated using: Data on missing teeth (m), filled teeth (f), and dmft scores; or a dmft score of 0 (equating to a treat. index = 1).
[50] Care Index calculated using: Data on filled teeth (f) and dmft scores; or a dmft score of 0 (equating to a Care Index = 1).
[51] Submucous cleft palate patients excluded from all 5 year outcomes as all/most teams do not audit these patients.

Diagnostic information is coded using the *International Classification of Diseases 10th revision* (ICD-10) and procedure information is classified according to codes from the *Classification of Surgical Operations and Procedures 4th Revision* (OPCS-4).

Cohort

Dental health data are available in CRANE for children with a non-submucous cleft affecting the palate born from 2004 onwards. The CRANE-HES linked dataset used for analyses contained records for children born up to 2012 and admissions up to 2015. Our analyses were restricted to CRANE-consented children with a specified cleft type – of cleft lip (CL), cleft palate (CP), unilateral cleft lip and palate (UCLP) or bilateral cleft lip and palate (BCLP), who were born between 1 January 2004 and 31 December 2012, and who were still alive on their fifth birthday. Children were included in analyses if their CRANE records were successfully linked with HES records and had complete dmft scores and deprivation data (n=4,083).

Deprivation

Deprivation quintiles were identified according to the Index of Multiple Deprivation (IMD) obtained from HES[52].

decayed, missing or filled teeth (dmft) scores

A dmft score describes the dental caries an individual has experienced and is a measure of oral health, reflecting the total number of teeth that are decayed, missing or filled. The risk of dental caries is thought to be higher among children with a cleft lip and/or palate compared with children without an oral cleft[53].

- A dmft >0 indicates experience of dental decay.
- A dmft >5 indicates experience of extensive dental decay.

Treatment Index

A 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.

dmft scores of 0 or scores for all three 'm', 'f' and 'dmft' data items are required for the calculation of Treatment Index. When calculated, treatment indices range from 0 to 1 and are usually expressed as a percentage[54].

Treatment indices with a value of 1 (100%) indicate that there is no untreated disease, which is the desired outcome.

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[52]https://digital.nhs.uk/data-and-information/data-tools-and-services/data-services/hospital-episode-statistics/hospital-episode-statistics-data-dictionary

[53] (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.

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

Care Index

A Care Index reflects cases where children have experienced dental decay, identified at the earliest possible stage (which is preferable), and have been provided with care in the least invasive form possible, i.e. fillings.

dmft scores of 0 or scores for both 'f' and 'dmft' data items are required for the calculation of the Care Index. When calculated, care indices also range from 0 to 1 and are usually expressed as a percentage[55].

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.

Analyses

Children were grouped according to deprivation quintiles. The percentage of children with dmft >0 and dmft >5 were compared between deprivation quintiles using chi squared tests. A Kruskall-Wallis test was used to compare the mean rank of Treatment Index and Care Index scores between deprivation quintiles. A p-value <0.05 was considered to indicate a statistically significant result. All statistical analyses were conducted using Stata V.15 (StataCorp, College station, Texas, USA).

Ethical Considerations

The study was exempt from NHS Health Research Authority ethics approval as it involves the analysis of a dataset that is collected for the purpose of service evaluation and is linked with parental consent to other datasets.[4]

5.3.2. Results

Analyses revealed that poor oral health (as expressed by rates of dmft >0 and dmft >5) was most prevalent among children living in the most deprived areas (quintile 1). These children were also least likely to have their dental disease treated (as expressed by the Treatment Index) or receive the appropriate care at the earliest possible stage (as expressed by the Care Index).

Table 5.2. Number and percentage of 5-year old CRANE-consented children born 2004-12 (in England) who had dmft>0 and dmft>5, and average Treatment Index and average Care Index, according to deprivation quintile.

Deprivation	Eligible cases with dmft reported	At least one (>0) dmft		dmft >5		Mean Treatment Index		Mean Care Index	
	N	N	(%)	N	(%)	N	(%)	N	(%)
Q1 - Most deprived	919	526	(57.2)	236	(25.7)	894	(63.1)	896	(52.2)
Q2	846	390	(46.1)	124	(14.7)	811	(72.3)	814	(64.9)
Q3	778	316	(40.6)	111	(14.3)	739	(77.6)	741	(69.6)
Q4	762	243	(31.9)	63	(8.3)	733	(80.5)	734	(75.9)
Q5 - Least deprived	778	188	(24.2)	47	(6.0)	749	(85.7)	749	(82.8)
Total	4,083	1,663	(40.7)	581	(14.2)	3,926	(75.3)	3,934	(68.4)
p-value		p<0.001		p<0.001		p<0.001		p<0.001	

Notes: (1) Registered in CRANE by 13 July 2020. Exclusions (not mutually exclusive): Children who died before the age of 5 years, submucous cleft palates, born with a non-specified cleft type, and not born in England. p-value for difference between deprivation quintiles.
(2) The heat map reflects direction of numbers and not whether the outcome is or is not desirable; the highest percentages are shown in the darkest colour while the lowest percentages are shown in the lightest colour.

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

Table 5.2. shows that rates of dental decay (as experienced by having had at least one decayed, missing or filled tooth (dmft >0)) and extensive caries (dmft >5) increased significantly as deprivation increased (p<0.001 for both outcome measures). While the proportion of treated dental disease and Care Index scores decreased significantly as deprivation increased (p<0.001 for both outcomes).

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 deprivation differences in the levels of dental disease will not only be affected by the dental care received by children. Dental 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.

Figure 5.9 illustrates the proportion of children with dmft >0 according to deprivation and birth year, and shows some change within deprivation quintiles.

Figure 5.9. Percentage of 5-year old CRANE-consented children who have dmft data indicating dmft >0, by deprivation quintile and birth year.
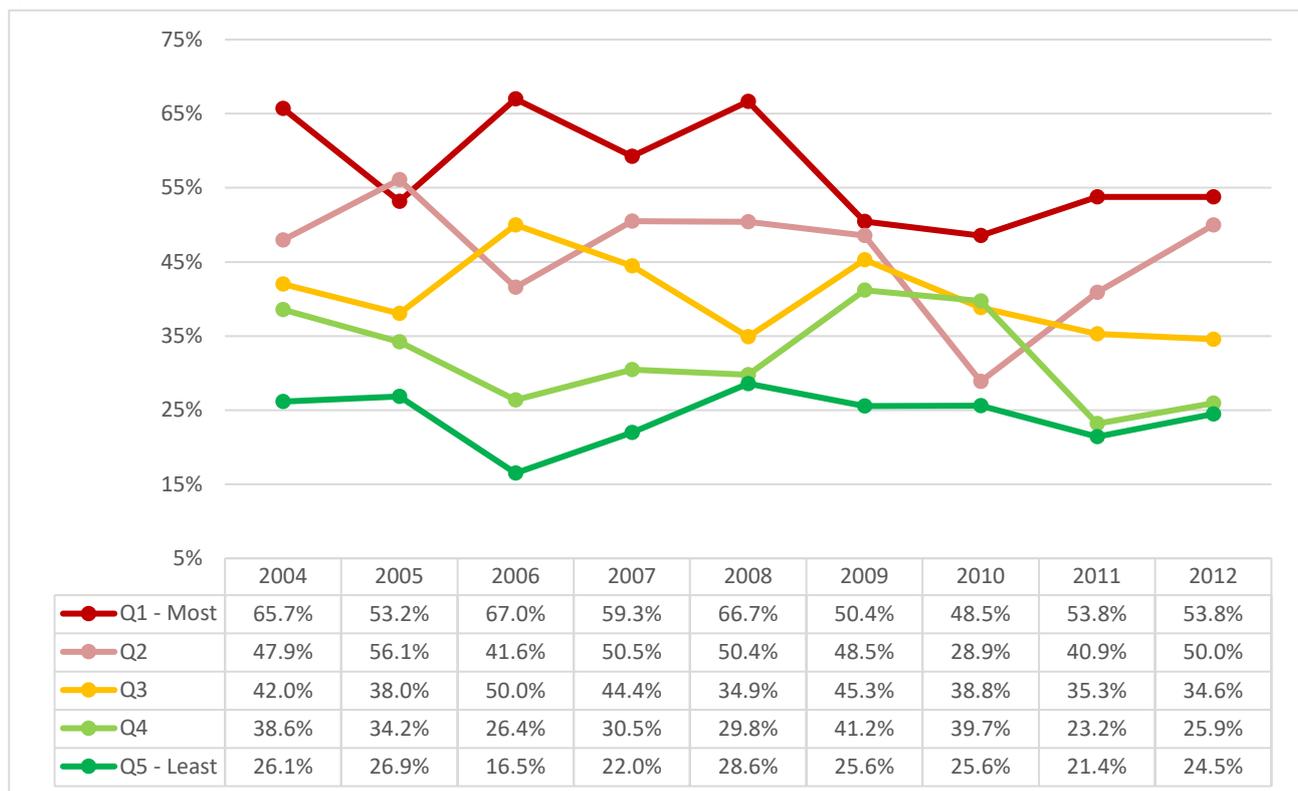
	2004	2005	2006	2007	2008	2009	2010	2011	2012
Q1 - Most	65.7%	53.2%	67.0%	59.3%	66.7%	50.4%	48.5%	53.8%	53.8%
Q2	47.9%	56.1%	41.6%	50.5%	50.4%	48.5%	28.9%	40.9%	50.0%
Q3	42.0%	38.0%	50.0%	44.4%	34.9%	45.3%	38.8%	35.3%	34.6%
Q4	38.6%	34.2%	26.4%	30.5%	29.8%	41.2%	39.7%	23.2%	25.9%
Q5 - Least	26.1%	26.9%	16.5%	22.0%	28.6%	25.6%	25.6%	21.4%	24.5%

Note: Registered in CRANE by 13 July 2020. Exclusions (not mutually exclusive): Children who died before the age of 5 years, submucous cleft palates, born with a non-specified cleft type, and not born in England.

As shown in Figure 5.9, the rate of dmft >0 in the most deprived quintile (Q1) dropped by 11.9% over the last 9 years, from 65.7% in for children born in 2004 to 53.8% for children born in 2012. The rate of dmft >0 in the least deprived quintile (Q5) has remained fairly constant over the last nine years, only dropping by about 2%. Large rates of fluctuation can be seen across all deprivation quintiles. The difference in rates of dmft >0 between the least and most deprived has narrowed in the last 9 years by 10.3% (39.6% difference between 26.1% (Q5) and 65.7% (Q1) in 2004 vs. 29.3% difference between 24.5% (Q5) and 53.8% (Q1)).

6. Development of CRANE Database and future directions

6.1. Future development of the CRANE Database and website

CRANE is continuing to negotiate with NHS England, Wales and Northern Ireland 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 with all UK regulations and laws post BREXIT.

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.
- More accurate data collection of syndromes
- Dental Defects of Enamel (DDE) section/items (at 5 and 10yrs) as proposed by the Paediatric Dental CEN of CFSGBI.
- Psychology data section as proposed by the Psychology CEN
- Surgical data collection. This will be taken forward in collaboration with the Surgical CEN of CFSGBI

6.2. Scotland

NHS Scotland management have reaffirmed their intention to submit data to the CRANE Database. COVID has delayed preparation in Scotland of contracts and data sharing agreements. The CRANE Database has been adapted to receive Scottish data and we remain positive that Scotland should begin inputting data early in 2021. 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.

6.3. Outcome measures

6.3.1 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. During 2019 and 20 work has begun to identify risk modulators in speech and dental outcomes. Initial analyses have identified four independent

modulators of speech and two independent modulators of dental health. Results of these investigations are being prepared for peer reviewed publication and dissemination. Moving forward, CRANE intends to develop a limited speech risk stratification model for speech and dental outcomes for inclusion in future reports. For the purposes of prospective speech outcome comparisons between cleft care teams, only patient characteristics would be included in these models.

6.3.2   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. While it was hoped to have this process complete for reporting in the 2020 report COVID has prevented face-to-face discussions by and with each of the clinical excellence networks. CRANE is attempting to complete this project through virtual contact but this remains challenging. Work will continue through 2020/21 and an update on progress will be made in next year's report.

6.4.   Data sources and future analyses

The General Data Protection Regulation (GDPR) coming onto force in 2018 has affected all data repositories and linkage is now a significantly more labour intensive (and thus costly) process. CRANE has identified a number of related data sources that would be advantageous to have regular linkage to facilitate both validation of CRANE held data and appropriate reporting of cleft related outcomes.

Data sources, to which regular CRANE linkage are being sought include; the National Pupil Database (NPD), the Hospital Episode Statistics (HES) database, and the Newborn Hearing Screening Programme (NHSP). Linkage projects require secure funding on each side of the linkage process to both ensure long-term sustainability of the data source but also safe holding of data transferred. The securing of a long-term funding contract for CRANE from NHS England is vital to this process.

6.4.1. 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.

Historic data linkage with this data source has seen CRANE contribute to the literature both to identify and describe the magnitude of the effect that being born with a Cleft lip and/or palate has on 5-year-old educational outcomes. More recently the CRANE team has published on the significant effect school absence has on educational attainment at age 7. Ongoing investigations utilising the historic linked educational records in the NPD are looking at the effect clefting has on longitudinal educational outcomes. We would plan to develop these initial analyses of longitudinal educational attainment among children with a cleft to allow tracking of educational attainment over time as part of CRANE outcome analyses. This is felt to be a more holistic method of demonstrating efficacy of cleft care delivery in the UK.

6.4.2. Hospital Episode Statistics (HES)

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.

Although unlinked HES data is a useful tool for some investigation it lacks the accuracy and flexibility that linkage of the CRANE dataset to the HES dataset would facilitate. Currently CRANE is only linked to HES for births from 2000-2012. New agreements are required to facilitate ongoing linkage required to deliver on the aim of producing risk stratified speech outcomes and other similar activity. Having this ability in place will reduce the already significant data collection burden on clinical teams. For example, timing of completion of palate repair data can be accessed thorough HES rather than asking teams to record every operation on CRANE.

6.4.3. Newborn Hearing Screening Programme (NHSP)

We have approval for linkage between our CRANE Database and the Newborn Hearing Screening Programme (NHSP)[56] 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.

6.4.4. Cleft Collective

Early 2020 saw the first data sharing activity between CRANE and the Cleft Collective, with Cleft Development Group (CDG) approval on behalf of NHS England (the main funder). This was CRANE's first experience, since the database launched 20 years ago, of sharing (providing) data. CRANE's experience prior to 2020 had only been as the receiver of data.

The team has learnt much around the legal responsibilities of providing data for the first time, with support from the RCS England's Data Protection Office, particularly in the context of the introduction of the GDPR in 2018 (and its associated challenges).

With the onset of COVID, the collaborative process between the Cleft Collective and CRANE slowed, as each team adapted to enforced changes in working practices. As we (hopefully) move to more recognisable working environments in 2021, work on this first data sharing project is planned to recommence. As we learn about this process and what benefits it can bring, we hope to be able to work more closely using the strengths of both datasets to answer questions important to patients and clinicians alike.

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

6.5. Quality Dashboard

The CRANE project team have submitted data on behalf of cleft care teams since the 2016/17 Quality Dashboard, up until the most recent Quality Dashboard year. 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 care 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 care 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[57]) – 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).

Methods initially suspended data requests due to the COVID pandemic and its effect on services. This meant that CRANE did not submit data on behalf of cleft care teams for Q4 of the 2019/20 Quality Dashboard (due May 2020). Data has now been requested on a voluntary basis and CRANE has engaged with cleft care teams to provided data when they have agreed (since Q1 of the 2020/21 Quality Dashboard data was due in August 2020). Ongoing data submission are being discussed at CDG; and CRANE will continue to be guided in its action by the CDG and the individual cleft care teams.

6.6. CRANE Communications

6.6.1 Dissemination of 2020 findings

- The report will be available on the CRANE website from December 2020.
- Publication of the Annual Report will be announced via the regular quarterly Newsletter.
- A Summary of Findings for Patients and Parents/Carers from this 2020 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

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[57] 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.

- 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](#))

6.6.2 Publications and Presentations related to the CRANE Database delivered in 2020

Peer reviewed Publications

- **School absence and achievement in children with isolated orofacial clefts.** Kate J Fitzsimons, Scott A Deacon, Lynn P Copley, Min Hae Park, Jibby Medina, Jan H van der Meulen**.** Kate Jane Fitzsimons, Lynn P Copley, Efrosini Setakis, Susan C Charman, Scott A Deacon, Lorraine Dearden, Jan H van der Meulen. Archives of Disease in Childhood. 2020; 0:1-6.
- **Reliability and predictive validity of dental arch relationships using the 5-Year-Old Index and the GOSLON Yardstick to determine facial growth.** Marie Pegelow, Sara Rizell, Agneta Karsten, Hans mark, Jan Lilja, Midia Najar Chalien, Mathias Lemberger, Petra Peterson, Kate Fitzsimons, Scott Deacon, Jibby Medina, Mary Calvert and Michael Mars. Cleft Plate -Craniofacial Journal. 2020. Accepted for publication (available online).

Presentations

- **Early school attainment: The impact of living with a cleft?** Scott Deacon on behalf of the CRANE Project Team. European Cleft and Craniofacial Initiative for Equality in Care (https://ecce.nu/). Conference & Meeting. 13 - 14 December 2019. Kristianstad, Sweden.
- **Discrepancy between nationally reported cleft dental outcomes and local experience: Getting to the root of the problem.** Jacqueline Smallridge, Jibby Medina and Craig Russell. International Association of Paediatric Dentists Conference. September 2020 (VIRTUAL ORAL PRESENTATION).
- **The influence of cleft type and socio-economic deprivation on caries experience in children with oronasal clefts. Evidence from 4,000 patient records on the UK CRANE Database.** Jacqueline Smallridge, Jibby Medina and Craig Russell. International Association of Paediatric Dentists Conference. September 2020 (VIRTUAL ORAL PRESENTATION).
- **National Impact of RCPCH Best Practice Guide on Timing of Detection of Cleft Palate - time to adopt successful local quality improvement initiatives.** Helen McElroy, Cathy Marsh, Rebecca Bailey, Joanne Leigh, Scott Deacon, Jibby Medina. The Royal College of Paediatrics and Child Health (RCPCH) Conference. July-December 2020 (VIRTUAL POSTER).

6.6.3 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 contact from the project team (e-mail/phone/Twitter) will further strengthen attempts to widen distribution of the Databases findings/ publications.

Scientific conferences to be attended in 2021:
- TBC dependent on impact of COVID-19 pandemic.

7. Conclusion

CRANE is continually looking to build on past successes and learning from feedback provided by the cleft community in the UK and abroad. The changes in the structure of this report were developed in response to feedback from busy colleagues who wanted to be able to focus on specific aspects of CRANE's activity before digesting the report as a whole. With sections on registry activity, 5-year clinical outcomes and research activity, we hope that we have been able to facilitate this.

Currently CRANE audits outcomes early on in life, in an attempt to inform with a proximity to interventions that stimulates debate and (hopefully) improvement with as little lag as possible. We are also conscious not to invest our relatively small resource in investigating measures that relate to care approaches of previous generations. At the same time, CRANE does appreciate that while 'short term' outcomes are our current focus, we also have to improve their utility and develop more specific and holistic measures of longer-term outcomes. These outcomes should be independent of the detail of early interventions while facilitating tracking of the effects of changes (and hopefully improvements) in care pathways through demonstration of psychological, physical and economic well-being.

CRANE is now close to being able to deliver on some of these aims. Established analyses of some short-term outcomes have sufficiently large datasets to allow for the development of meaningful risk-stratified comparison of treatment protocols offered by the various teams across the UK. For example, multiple independent risk modulators of both speech and dental outcomes have now been identified. Furthermore, the nature of the CRANE dataset and the team's experience in data linkage could allow for the potential utility of longer-term outcomes, such as educational attainment. It may be possible to investigate risk modulators of this outcome also in the near future

In summary, CRANE has been successful in providing robust information that facilitates more detailed outcome information for parents that is specific to their child's cleft characteristics. Furthermore, much has also been learned about the completeness of the dataset and this itself has stimulated significant positive interaction with clinical colleagues who all have seen the personal, professional and clinical utility of improving this aspect of the dataset.

Finally, CRANE has summarised our findings and recommendations in a short section after the introduction. We hope that this will encourage readers to engage with the full data included in the main report. We also hope that it will stimulate debate among those affected by cleft, their representative bodies, clinical and research colleagues, and also funders of research, such that all see the benefit of investing in cleft audit and research.

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

Best wishes,

The CRANE project team.

Glossary and abbreviations

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.
BCLP	Bilateral cleft lip and palate
CAPS-A	Cleft Audit Protocol for Speech—Augmented
Caries (dental)	Dental caries are also known as tooth decay / dental decay or a cavity.
CEN	Clinical Excellence Network – previously referred to as Special Interest Group (SIG)
CFSGBI	Craniofacial Society of Great Britain and Ireland
CI	Confidence interval
CL	Cleft lip only
Cleft	A failure of tissues to join during development.
Cleft care teams / cleft care teams / Regions / Units/ Services	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 care teams.
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 / cleft care team.
CLEFTSiS	The National Management Clinical Network for Cleft Service in Scotland
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/
CP	Cleft palate only
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
CRG	Clinical Reference Group
CSCs	Cleft Speech Characteristics
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.
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
FFT	Friends and Family Test
Funnel Plot	A graph that identifies regions / cleft care teams 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 / cleft care team. • Each funnel plot is for one outcome, with its values shown on the vertical/Y axis. • The size of the regions' /cleft care teams' cohort is shown on the horizontal or X axis. • The benchmark value or overall national percentage 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) (as in the Registrations section of this report).

	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.
ICD-10	International Classification of Disease 10th Revision
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.
MDT	Multi-Disciplinary Team
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.
OPCS-4	Classification of Surgical Operations and Procedures 4th Revision
Patient Episode Data Wales (PEDW)	A national database containing records on all admissions to hospitals in 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
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.
TIM	Tiers of Involvement Measure
UCLP	Unilateral cleft lip and plate
WHO	World Health Organization