Promoting excellence in cleft care

Cleft Registry and Audit NEtwork Database

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

2021 Annual Report

Results of the audit in England, Wales and Northern Ireland for children
born with a cleft between January 2000 and December 2020

On behalf of the Cleft Development Group

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Registered charity no: 212808

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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)

Contents

Acknowledgements ..... **iv**

Foreword ..... **v**

Executive summary **7**

Key findings and recommendations 2021 ..... **8**

1. Introduction **11**

2. Methods **12**

2.1. CRANE 12

2.2. Hospital Episode Statistics (HES) 13

3. Registry Information **14**

3.1. Registrations in CRANE 15

3.2. Timing of diagnosis for all cleft types 18

3.3. Timely diagnosis of cleft palate 21

3.4. Gestational age and birth weight 24

3.5. Contact with cleft services 27

3.6. CRANE consent 30

4. Audit Outcomes at 5 years of age **33**

4.1. Child growth 34

4.2. Dental health 40

4.3. Facial growth 50

4.4. Speech 55

4.5. Psychology 64

5. Research **75**

5.1. Congenital malformations in children born alive with a cleft 76

5.2. Congenital malformations of the circulatory system and speech at 5 years 79

5.3. Is there a difference in cleft-related speech outcomes between girls and boys at 5 years? 84

6. Development of the CRANE Database and future directions **88**

6.1. Future development of the CRANE Database and website 88

6.2. Scotland 89

6.3. Outcome measures 89

6.4. Data sources and future analyses 89

6.5. Quality Dashboard 91

6.6. CRANE Communications 92
7. Conclusion 95
Glossary and abbreviations 96

Documents related to this product, for reference, published separately:

Document	Published
2021 Annual Report: Appendixes	December 2021
2021 Annual Report: Responses to outlier process	December 2021
2021 Annual Report: Methods	December 2021

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 (CDG) 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;
- Marie Pinkstone and Imogen Underwood for their help to understand the challenges associated with speech data collection during the COVID-19 pandemic, as detailed in their statement (included in the Speech Audit Outcomes at 5 years of age section of this report), submitted 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.

This year has seen the piloting of a new outlier process for CRANE. We would like to thank all at the UK NHS CDG for supporting this process. In particular would like to recognise the extra time and effort it has taken for clinical colleagues to engage and respond so constructively to outlier communications. The positive way in which the outlier process has been received and responded to is hugely encouraging for the future of national clinical audit of cleft care.

Special thanks go to our colleagues at the Clinical Effectiveness Unit (CEU) of the Royal College of Surgeons of England for their methodological and statistical input throughout 2021; including Melissa Gannon - Research Fellow / Methodologist, and Amundeep Johal - Statistician / Quantitative Analyst.

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.

This work was funded by the National Specialised Commissioning Group for England, the Wales Specialised Health Services Committee, and the Northern Ireland Specialist Services Commissioning Team, and it was carried out by the project team of the CRANE Database, which is overseen by the UK NHS Cleft Development Group (CDG)[1].

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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

'Control what you can and accept what you can't'

Or better still

*Control what you can and **seek the opportunity in what you can't***

COVID-19 is now part of everyday life. Cleft services and cleft care have not been immune from its effect. At the start of the pandemic many thought that there was very little that cleft services could do to alleviate the impact on services. After all cleft care is not life threatening and - when debating resource allocation in competition with emergency, trauma and cancer services - it is not easy for cleft care to prevail. That said, while cleft care may not be immediately life-saving it does have the potential to be hugely life improving and not just in the immediate future; benefits of the right care at the right time can last a lifetime!

While other services may have been and continue to be prioritised, it is incumbent upon those with expertise and access to data to demonstrate the benefit of 'right time right place care' and argue for the relative priority of cleft care. We should recognise that data is king in such discussions, and we should aim to demonstrate the immediate and prolonged clinical, social, psychological (and financial) benefit of the maintenance of clinical services. The data that the clinical community have collected and inputted into the CRANE Database is a paramount resource in this realm. It has allowed the UK cleft community to argue for continued access to in-patient and theatre resources; when many others were unable to demonstrate clinical benefit to the same extent. The patience and perseverance that the clinical community has had in collecting audit data is now beginning to show benefit. Cleft audit is not done, it is only just beginning!

Early in the pandemic lockdown CRANE looked at the opportunity it had, through data linkage, to examine the relationship between timing of palatal surgery and the quality of speech outcomes. Despite CRANE's relatively large dataset (in excess of 21,000 patients) modern speech results were only available for a cohort of children born from 2007 up to 2013. Interrogation of the data available strongly indicated that delay to completion of primary cleft palate repair beyond 13 months was associated with significantly poorer speech outcomes. This knowledge, garnered from many years work in collecting and assessing speech outcomes by our clinical colleagues, was instrumental in allowing the cleft community to argue at a national level for resource maintenance to continue to deliver against the current cleft pathway. Without this analysis there was the risk that palate repair would have potentially not been a priority during the pandemic until children approached 2 years of age.

Speech is not an outcome in isolation. CRANE has also shown that school absences, including time taken off for medical appointments, negatively impacts on the educational outcomes of children with a cleft. Combining what we know about speech and educational outcomes for children with a cleft, we may infer that not only are speech outcomes be improved by operating at the right time but educational outcomes may also be improved through reduction in time off school for further surgery / Speech therapy appointments As a result, not only are speech outcomes prioritised by timely surgery so potentially is the educational outcome for this group of children.

Cleft care is not just surgery. It is, in fact, so much more! A colleague often reminds me that it is only a small part of cleft care, I very much agree. One of the themes identified in responses provided by cleft services to the *findings of this report, is that while theatre time has been relatively in most areas of the country to allow continued delivery of primary cleft care pathways on protocol (palate repair by 13 months) there has been significant and ongoing impact on out-patient activity across all other aspects including nursing, speech therapy, paediatric dentistry, audiology, psychology and orthodontics. This has affected cleft services ability to deliver the largest parts of cleft care, and simultaneously affected the ability of cleft services to collect and record audit data; which supports clinical practice and reassures patients and carers of the quality of care and outcomes they are receive.*

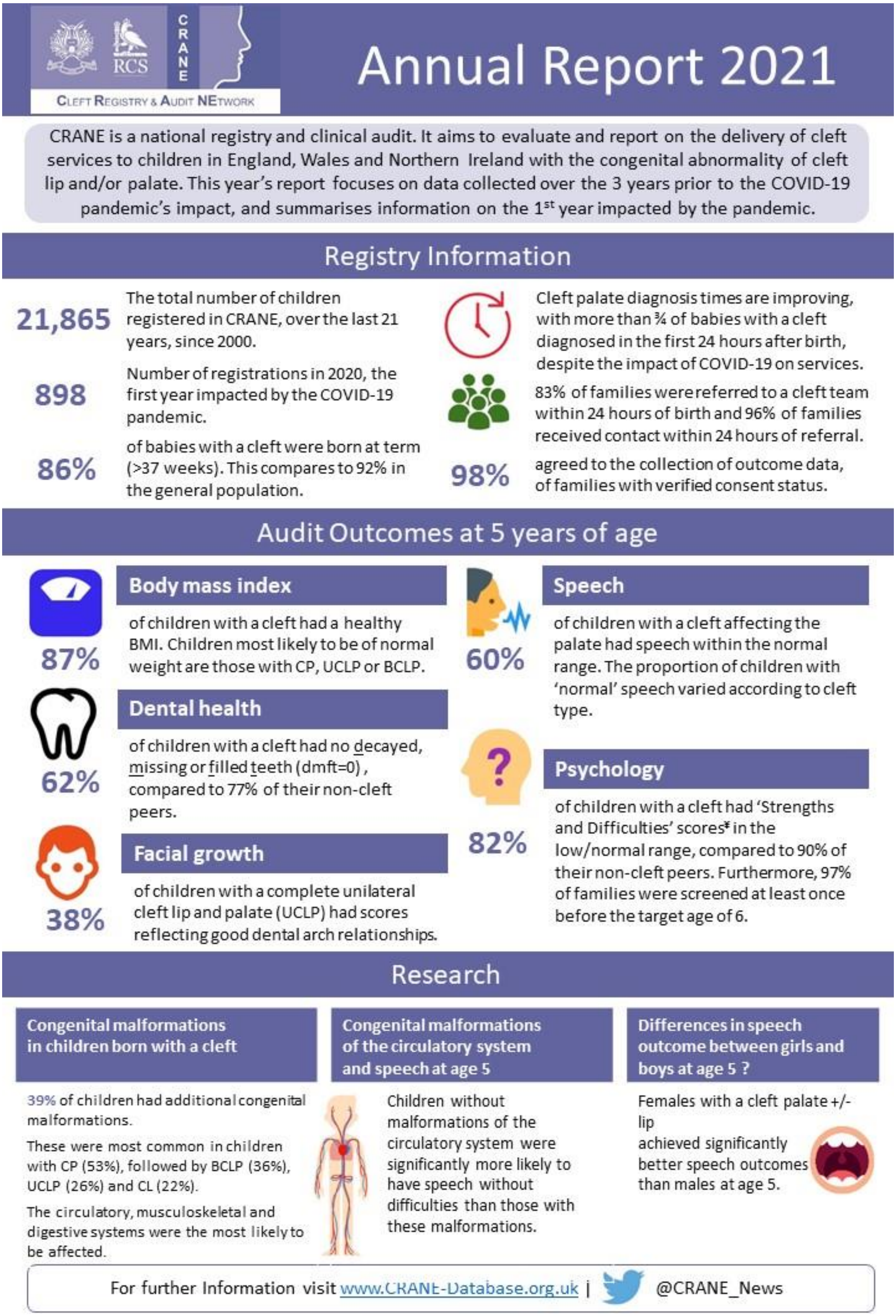
We want data to be available to inform and drive changes in the way that we deliver the highest quality out-patient care, in the same way that it has helped support the continued delivery of in-patient care. As we try to maintain outpatient activity, we need learn why some areas of the country have been able to continue with only small amounts of disruption while others have been brought to an almost complete standstill. Is it simply about which part of the country you are in and relative impact of COVID , or is it dependent on the type of setting those clinics are delivered from? While the reduction in the levels of care being delivered in some regions may have reduced the care burden in the short term, what has it done to the long-term well-being and life chances of persons affected by clefting? Time is needed before we can answer these questions but even then, it cannot happen without data.

Lessons from COVID may not be immediately available and we may have to wait at least 5 years until we can quantify the impact it has had full impact on early outcomes of cleft care, never mind longer term impacts. . In the meantime, we can continue to use the data we do have in meaningful ways to improve the current and near future prospects for the patients and families served by the cleft care community. Let's not just look to return 'to pre-COVID normals' but seek, in the opportunity of the pandemic, to make it better. Better for our patients, better for our families, better for our colleagues… In fact, let's just make it better for all!

With the best of season's greetings and wishing everyone a better new year.

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

Executive summary



CRANE

CLEFT REGISTRY & AUDIT NETWORK

Annual Report 2021

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 3 years prior to the COVID-19 pandemic's impact, and summarises information on the 1st year impacted by the pandemic.

Registry Information

21,865 The total number of children registered in CRANE, over the last 21 years, since 2000.

898 Number of registrations in 2020, the first year impacted by the COVID-19 pandemic.

86% of babies with a cleft were born at term (>37 weeks). This compares to 92% in the general population.

Cleft palate diagnosis times are improving, with more than ¾ of babies with a cleft diagnosed in the first 24 hours after birth, despite the impact of COVID-19 on services.

83% of families were referred to a cleft team within 24 hours of birth and 96% of families received contact within 24 hours of referral.

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

Audit Outcomes at 5 years of age

Body mass index

87% of children with a cleft had a healthy BMI. Children most likely to be of normal weight are those with CP, UCLP or BCLP.

Dental health

62% of children with a cleft had no decayed, missing or filled teeth (dmft=0), compared to 77% of their non-cleft peers.

Facial growth

38% 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 according to cleft type.

Psychology

82% of children with a cleft had 'Strengths and Difficulties' scores* in the low/normal range, compared to 90% of their non-cleft peers. Furthermore, 97% of families were screened at least once before the target age of 6.

Research

Congenital malformations in children born with a cleft

39% of children had additional congenital malformations.

These were most common in children with CP (53%), followed by BCLP (36%), UCLP (26%) and CL (22%).

The circulatory, musculoskeletal and digestive systems were the most likely to be affected.

Congenital malformations of the circulatory system and speech at age 5

Children without malformations of the circulatory system were significantly more likely to have speech without difficulties than those with these malformations.

Differences in speech outcome between girls and boys at age 5 ?

Females with a cleft palate +/- lip achieved significantly better speech outcomes than males at age 5.

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

Key findings and recommendations 2021

Findings	Recommendations	Source
Registry		
Registrations in CRANE		
• There were 898 registrations in 2020, the first year impacted by the COVID-19 pandemic. • Cleft Palate (CP) is the most common type of cleft, representing 44% of all cases with a known cleft type, followed by cleft lip (CL) (23%), unilateral cleft lip and palate (UCLP) (22%) and bilateral cleft lip and palate (BCLP) (11%) (in 2017-19[2]).	1. CRANE will engage with the nursing Clinical Excellence Network (CEN) to ensure optimum assessment and recording of registrations. 2. CRANE will continue to record cleft births and validate case ascertainment using external datasets.	Chapter 3, Section 3.1 **Indicator[3]: #1**
Timely diagnosis		
• Similar rates of antenatal diagnosis for CL, UCLP and BCLP been observed in 2020, the first year impacted by the COVID-19 pandemic, by comparison with rates in 2017-19[2]. • Significant improvement has been observed in the timely detection of CPs, particularly in 2017-19. 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.	Chapter 3, Section 3.2 & 3.3 **Indicator: #2**
Gestational age and birth weight		
• The average gestational age of babies born with a cleft in 2017-19 was 38.5 weeks[5]. • 14% of babies were born prematurely (<37 weeks' gestation). This compares to 8% in the general population. • Children with cleft lip had a higher average birth weight (3,297 g) than those with a cleft affecting the palate (3,087g).	4. Cleft services should review procedures to ensure processes are in place to record and submit data on gestational age and birth weight. To track association of prematurity and low birth weights with clefting. 5. The research community should validate and further investigate the apparent association between cleft-affected pregnancies and prematurity at birth. 6. CRANE will engage with the RCM and Royal College of Obstetricians and Gynaecologists (RCOG) to communicate our findings.	Chapter 3, Section 3.4
Contact with cleft services		
• 83% of families were referred within 24 hours of birth. This rate varied significantly across cleft types. In 2017-19[5]. • 96% of families were contacted by a cleft services within 24 hours of referral.	7. Cleft services should continue to work with referring obstetric, midwifery and neonatal units to improve timeliness of diagnosis and early referrals. 8. Cleft services with high levels of referrals within 24 hours should share their best practise recommendations.	Chapter 3, Section 3.5 **Indicators: #3 & #4**

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[2] Over the last three years prior to the impact of the COVID-19 pandemic.

[3] CRANE core indicators are detailed in Appendix 3.

CRANE Consent		
• Consent status was verified for 90% of families approached in 2017-19. This meant they had agreed or declined to their child's outcome data being recorded in CRANE[5]. • The verified consent rate has fallen in recent years. • Of the families who had reached a decision, rate of agreed consent was 98%.	9. Cleft services 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. 10. CRANE will continue to work with cleft services and the Nursing CEN to improve consent status verification.	Chapter 3, Section 3.6 **Indicator: #5**
Outcomes at 5 years		
Child growth		
• 87% had a healthy Body Mass Index (BMI). • The prevalence of overweight or obese children was higher among those with CL than among those with a cleft affecting the palate. • The prevalence of underweight children was higher among those with UCLP (3.1%) than among those with other cleft types.	11. Cleft services should aim to assess children's weight and height at age five and improve the reporting of these measures in the CRANE database. This will facilitate more meaningful comparisons between subgroups in the future.	Chapter 4, Section 4.1 **Indicator: #6**
Dental health		
• Dental decay was experienced by 38% 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 appropriate care at the earliest possible stage) was 70%. These rates decreased significantly as cleft type became more involved.	12. All children with a cleft should have a recommended care plan established by collaborative work between the families' General Dental Practitioner (GDP) and cleft services 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 services led).	Chapter 4, Section 4.2 **Indicators: #7 & #8**
Facial growth (for children with complete UCLP)		
• 38% of children with complete UCLP had 5 year old Index scores reflecting 'good' dental arch relationships. • Dental arch relationships remained stable over time.	13. Cleft Services should see all children age 5 with complete UCLP and take records of facial growth (impressions or photographs). Records should be shared with national co-ordinator and assessed using the 5-year index with results recorded on the CRANE Database. 14. The research community should undertake to compare UK facial growth outcomes with those in other countries and evaluate the predictive value of the 5 year old Index in the UK.	Chapter 4, Section 4.3 **Indicator: #9**
Speech (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. • 68% had speech without cleft-related articulation difficulties. • 18% of children had secondary surgery for speech purposes before the age of 5 years. • The percentage of children meeting the standards has not changed significantly over time.	15. Information given to parents by cleft services about expected speech outcomes should take into account the child's cleft type. 16. The research community should undertake to develop risk stratification models for analysing speech outcomes among children with a cleft.	Chapter 4, Section 4.4 **Indicators: #10, #11 & #12**

Psychology screening		
• 97% of families were screened at least once before the target age of 6 years. • 93% were seen by a psychologist and a psychosocial screen was completed at age 5 (TIM tiers 1 to 4, referred to as 1a+). • 18% of children born with a cleft had 'high' or 'very high' SDQ scores. These rates are higher than the population norms.	17. Cleft services 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).	Chapter 4, Section 4.5 **Indicators: #13 & #14**
All outcomes at 5 years		
An outlier process was applied to children's outcomes at 5 years of age, for the first time. All cleft services provided responses to being identified as positive or negative outliers.	18. All cleft services should work together to explore reasons for variation in outcomes at 5 years of age.	Chapter 4
Research		
Congenital malformations in children born with a cleft		
• 38.8% of children with a cleft had additional congenital malformations. • This was most common in children with CP (53%), followed by BCLP (33.5%), UCLP (26.3%) and CL (22.2%). • Body systems most likely to be affected by additional malformations were the circulatory system, musculoskeletal system and digestive system.	19. Given the frequency of certain structural malformations, clinicians should consider standard screening for children with a cleft 20. Establishing good links with paediatric and genetic services is recommended.	Chapter 5, Section 5.1
Congenital malformations of the circulatory system and speech at age 5		
• Children without congenital malformations of the circulatory system were significantly more likely to meet cleft speech standards than those with circulatory system malformations. • With adjustment for cleft type, children with malformations of the circulatory system had significantly reduced odds of achieving standard 2a and standard 3 than those without malformations of the circulatory system.	21. Surgical and Speech and Language Therapy CENs should agree whether to exclude children with congenital malformations from between-centre comparisons in speech outcome reporting in the future.	Chapter 5, Section 5.2
Is there a difference in cleft-related speech outcomes between girls and boys at age 5		
• Among children with CP and UCLP, a significantly higher percentage of girls met cleft speech standards 1 and 3 than boys. • Among children with BCLP, a significantly higher percentage of boys met cleft speech standard 2a. This was not significant for cleft speech standards 1 and 3. • With adjustment for cleft type, boys had significantly reduced odds of achieving cleft speech standard 1 and 3.	22. The child's sex should be factored into risk adjustment models when reporting cleft-related speech outcomes at 5 years.	Chapter 5, Section 5.3
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 2014.	23. Cleft services should pay particular attention to assessing outcomes and reporting these to CRANE. 24. Cleft services, Clinical Excellence Networks (CENs) and CRANE should work together to identify barriers to collecting and submitting data.	All

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[4]. 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 2022 once GDPR compliant data sharing agreements are in place. The geographical representation of the cleft services is detailed in **Appendix 4**.

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[5] for children with a cleft lip and/or palate born in England, Wales and Northern Ireland between 1 January 2004 and 31 December 2020, 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 services, and consent status for children born 2017-2019. Summary information on 2020 births is also provided. Data for 2020 births should be interpreted with caution given the highly variable impact the COVID-19 pandemic has had on clinical services ability to complete registrations and process the data.
- **Audit Outcomes at 5 years of age:** This section details cleft-related outcomes for children, registered in the CRANE Database, at 5 years of age and born 2011-2013. Summary information on 2014 births is provided but should be interpreted with caution given the highly variable impact the COVID-19 pandemic has had on clinical services ability to complete clinical audit and process the data.
- **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. It does so by reporting against process and outcome indicators, agreed by the UK NHS Cleft Development Group (CDG) and Clinical Excellence Networks (CENs), as detailed in **Appendix 3**.

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

[5] 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 service, and time of first contact between a patient and a cleft service. The CRANE Database also collects information about cleft-related treatment and outcomes. Each child born with a cleft in England, Wales and Northern Ireland should be referred to one of the 13 cleft services providing care in these countries shortly after having their cleft diagnosed (as listed in **Appendix 4**).

Since 2000, 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 services. In 2012 we sought and gained approval[6] to collect 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 services at some point between referral and the first primary repair. A coordinator within each cleft service 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 30 June 2021 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 years of available data:

- Registry Information is reported for children born between 1 January 2017 and 31 December 2019, as well as summary information on 2020 births. Data for 2020 births should be interpreted with caution given the highly variable impact the COVID-19 pandemic has had on clinical services ability to complete registration processes (Chapter 3).
- Audit Outcomes at 5 years of age are reported for children born between 1 January 2011 and 31 December 2013, as well as summary information on 2014 births. Data for 2014 births should be interpreted with

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[6] Confidential Advisory Group (CAG) Section 251 Approval https://www.hra.nhs.uk/approvals-amendments/what-approvals-do-i-need/confidentiality-advisory-group/ Last accessed December 2021.

caution given the highly variable impact the COVID-19 pandemic has had on clinical services ability to complete clinical audit and process the data (Chapter 4).

- CRANE Research is reported for children born between 1 January 2000 and 31 December 2014, 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 2021 document, available online alongside this Annual Report (https://www.crane-database.org.uk/reports/crane-database-2021-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 report Appendices 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 5** 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 services around the time of birth.

Timeframe: The most recent four years of data, presented in two cohorts of patients.

- **Cohort 1.** Children born between 01 January 2017 and 31 December 2019 (consistent with a rolling 3-years, as for other sections of this report).
- **Cohort 2.** Children born between 01 January 2020 and 31 December 2020; these are presented separately to allow comparison of the effect of the COVID-19 pandemic and the lockdown that ensued.

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	The CRANE Database
	Birth years	Three years: 2017-2019. Plus summary information on 2020 births[7]
	Denominators	• 3,007 (2017-2019) and 898 (2020) children registered. • 2,766 (2017-2019) and 806 (2020) children with a specified cleft type. • 2,970 (2017-2019) and 889 (2020) children with a record of sex.
	Numerator	Number of children meeting each patient characteristic
	Data completeness	• 92% of 3,007 registrations had a specified cleft type (2017-2019). • 99% of 3,007 registrations had a specified sex (2017-2019).
	Countries	England, Wales and Northern Ireland
	Indicator	#1[8]
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 44% of all cases with a known cleft type, followed by cleft lip (CL) (23%), unilateral cleft lip and palate (UCLP) (22%) and bilateral cleft lip and palate (BCLP) (11%), respectively (2017-2019). • 56% of registrations were boys. As expected from historical experience, they were more likely to have a CL, UCLP or BCLP than registered girls (2017-2019).	
Recommendations	Cleft services should aim to: • register all children with a cleft in the CRANE database, • specify cleft type and sex for every child registered. CRANE will continue to record cleft births and validate case ascertainment using external datasets.	

3.1.1. Number of registrations

The CRANE Database has registered a total of 21,865 children, born in England, Wales and Northern Ireland with cleft lip and /or palate over the last 21 years[9]. Among them, 3,007 children were registered between 2017 and 2019, and 898 children were registered in 2020[10].

Figure 3.1 displays the number of CRANE registrations in 2020 for each cleft service – during the first year impacted by the COVID-19 pandemic – by comparison with the yearly average across 2017-19 (pre-pandemic) and the national averages for both cohorts. Eleven out of thirteen cleft services registered a lower number of children born with a cleft in 2020 in comparison to the yearly average number of registrations across the 2017-19 period.

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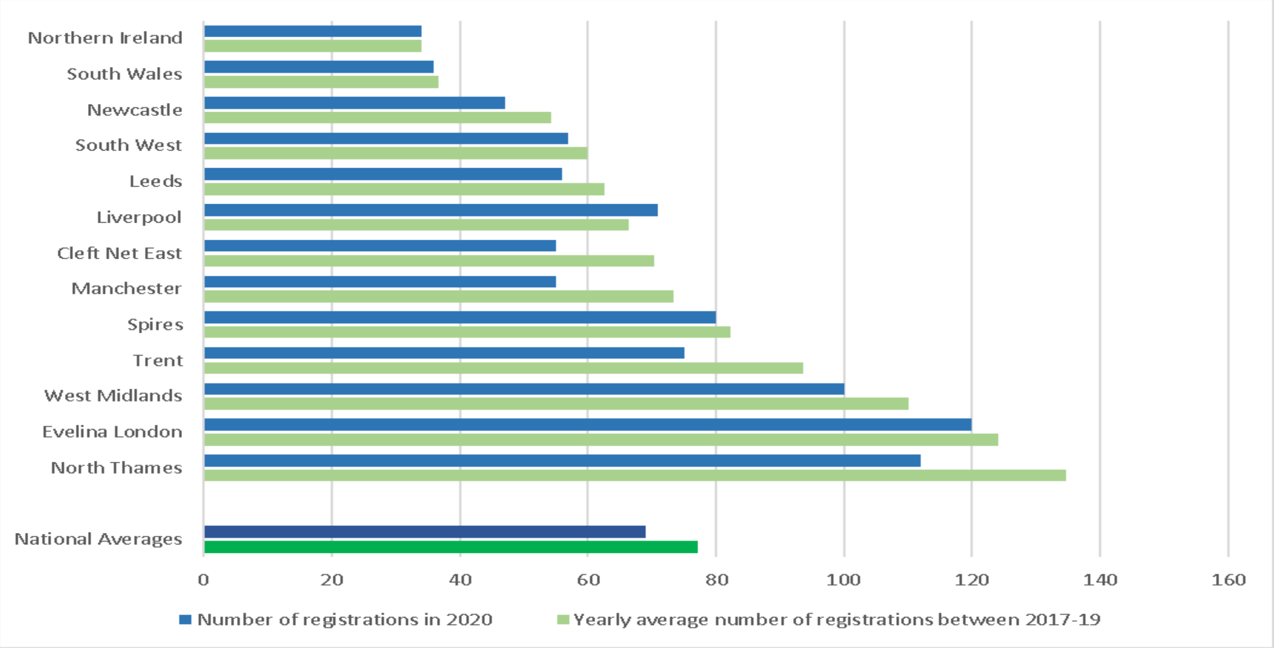
[7] Affected by the COVID-19 pandemic and the lockdown that ensued.

[8] CRANE core indicators are detailed in **Appendix 3**.

[9] Cohorts include all patients registered in the CRANE Database between 1st January 2000 and 31st December 2020, inclusive.

[10] Twelve children born and registered in the CRANE Database in 2017-19 were reported to have died within the first 12 months after birth; this was also the case for three children born and registered in 2020 (0.23% of registrations across those four birth years). It is not known from CRANE whether these children had additional anomalies or syndromes.

Figure 3.1. Number of CRANE-registered children in 2020, by comparison with the yearly average for 2017-19 registrations, according to cleft service.



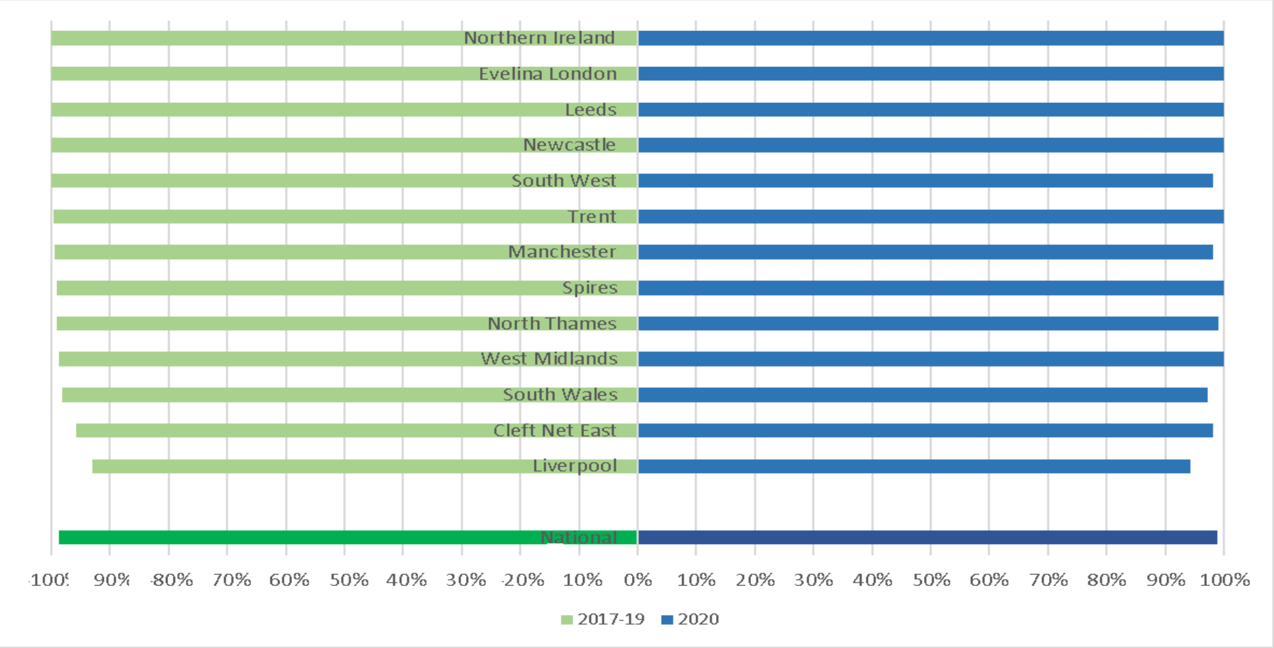
Note: Children registered in the CRANE Database between 1st January 2017 and 31st December 2020, by the 30 June 2021.

3.1.2. Data completeness for sex and cleft type

Participation in a national clinical audit, such as CRANE, means that all cleft services are asked to record the sex and cleft type of all children registered in the database. Figure 3.2 and 3.3 show the percentage of children with specified sex and cleft type for 2017-19, compared to 2020 registrations and national averages for the two periods.

- Recording of sex data remains high nationally, with 99% of children in the 2017-19 cohort (93%-100% range across cleft services) and 2020 (94%-100% range across cleft services), respectively, having their sex recorded in CRANE.
- 92% of all registrations in 2017-19 had their cleft type specified in CRANE (92%-99% range), compared to 90% among 2020 registrations (47%-100% range).

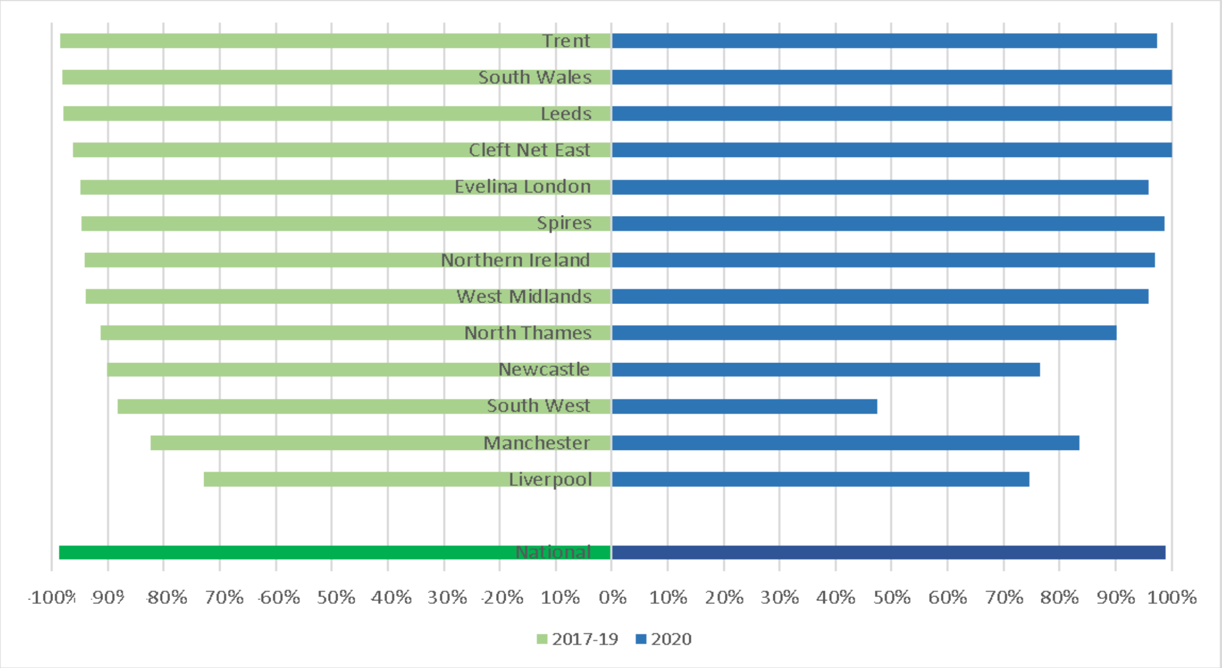
Figure 3.2. Percentage of children with sex reported in 2017-19, compared to national average for 2017-19 and 2020 registrations, by cleft services.



Note: Children registered in the CRANE Database between 1st January 2017 and 31st December 2020, by the 30 June 2021.

Given that both sex and cleft type are now recognised as determinants of speech outcomes and that cleft type is a known determinant of dental outcomes (see CRANE 2020 Annual Report), cleft services should seek to improve recording of sex and cleft type on the CRANE Database to ensure appropriate risk stratification / adjustment is possible in the future.

Figure 3.3. Percentage of children with cleft type specified in 2017-19, compared to national average for 2020 registrations, by cleft services.



Note: Children registered in the CRANE Database between 1st January 2017 and 31st December 2020, by the 30 June 2021.

3.1.3. Registrations by cleft type and sex

Among children registered between 2017 and 2019, inclusive, with a recorded cleft type (n=2,766), CP was the most common of the four cleft types[11], representing 44% of all registrations, followed by CL (23%), UCLP (22%) and BCLP (11%). Raw data in **Appendix 6** shows the distribution of cleft type by each cleft service, for registrations with a specified cleft type, in 2017-19 and 2020.

Of the children registered and diagnosed with UCLP (n=618) or BCLP (n=301) in 2017-19: 67% and 55%, respectively, were complete clefts involving the hard and soft palate. For children diagnosed in 2020 with UCLP (n=165) and BCLP (n=83): 75% and 57%, respectively, were complete clefts involving the hard and soft palate.

Overall, 56% of children registered between 2017 and 2019 were boys. Raw data in **Appendix 6** shows the distribution of sex by cleft services for registrations in 2017-19, as well as for 2020 registrations. Among 2017-19 registrations, boys were significantly more likely to have a CL (63%), UCLP (67%) or BCLP (67%) than girls ($p<0.001$). For the same period, CP was significantly more prevalent among girls (56%) than boys ($p<0.001$).

Registration of all live births with a cleft lip and/or palate in the CRANE Database is a key performance indicator (#1) for cleft services.

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[11] Cleft type is defined according to reported LAHSAL codes or, where LAHSAL has not been reported (for 11% of children registered between 01 January 2017 and 31 December 2019), it is based on the cleft type reported by the cleft service registering the child.

3.2. Timing of diagnosis for all cleft types

Cohort summary	Data source	The CRANE Database
	Birth years	Three years: 2017 to 2019. Plus summary information on 2020 births[12]
	Denominator	2,897 (2017-2019) and 825 (2020) children with a recorded diagnosis time
	Numerator	Number of children diagnosed at each time point
	Exclusion	Children without a recorded diagnosis time
	Data completeness	96% of 3,007 CRANE-registered children had a recorded diagnosis time (2017-2019).
	Countries	England, Wales and Northern Ireland
	Indicator	#2[13]
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 (for clefts involving the lip) or within 24 hours of birth.	
What did we find?	**All cleft types** • Rates of **antenatal diagnoses** for all cleft types during the first year impacted by the COVID-19 pandemic (2020) approached rates in the three years prior (2017-19) – 45% in 2020 and 46% in 2017-19. • Rates of **timely diagnoses (antenatally or within 24 hours of birth)** of all cleft types during the first year impacted by the COVID-19 pandemic (2020) were the same to rates in the three years prior (2017-19) – 87% in both 2020 and in 2017-19. **Clefts involving the lip** • Rates of **antenatal diagnoses** for clefts involving the lip (CL, UCLP, BCLP), during the first year impacted by the COVID-19 pandemic (2020) approached rates in the three years prior (2017-19) – 78% in 2020 and 81% in 2017-19. • Rates of **timely diagnoses (antenatally or within 24 hours of birth)** for clefts involving the lip (CL, UCLP, BCLP), during the first year impacted by the COVID-19 pandemic (2020) were the same as rates in the three years prior (2017-19) – 98% in both 2020 and in 2017-19.	
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 3,007 children born and registered in CRANE between 2017 and 2019, 96% of them had a recorded diagnosis time (range: 90% - 100% between cleft services). Out of the 898 children born and registered in CRANE in 2020 – during the first year impacted by the COVID-19 pandemic – 92% (n=825) of them had a recorded diagnosis time (range: 23% - 100% between cleft services). Cleft services should be commended for their commitment to recording this information, particularly during the year impacted by the COVID-19 pandemic.

3.2.2. Timely diagnosis of all cleft types

The variation in diagnoses of all clefts by cleft service were explored and are presented in Figure 3.4. In 2017-19, 46% of clefts were diagnosed during antenatal screening and 41% were diagnosed at birth, leaving 13% diagnosed late according to the National Standard. Figure 3.4 also shows that the percentage of clefts diagnosed

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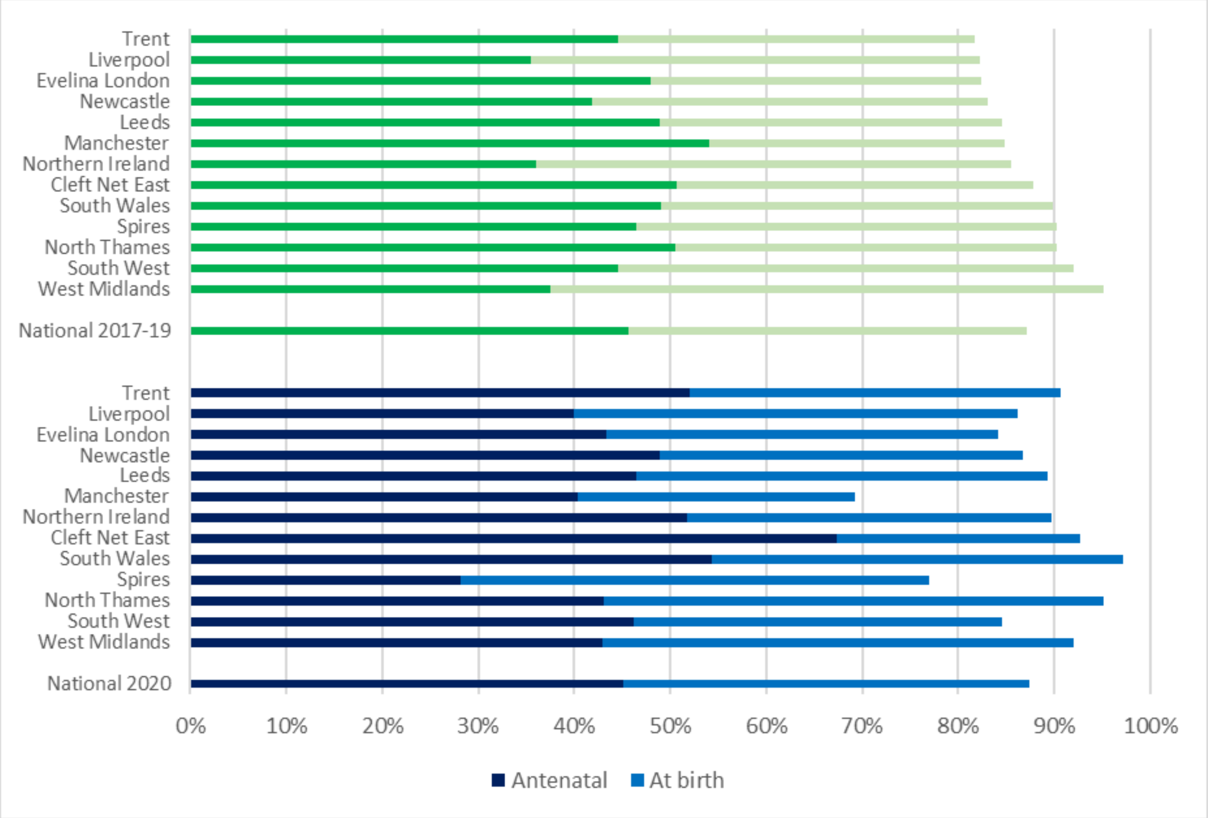
[12] Affected by the COVID-19 pandemic and the lockdown that ensued.

[13] CRANE core indicators are detailed in **Appendix 3**.

in a timely manner in 2017-19 varied by cleft service, ranging from 82% (Trent, 45% before and 37% at birth) to 95% (West Midlands, 38% before and 57% at birth). This variation suggests that practice varies between maternity units, with some better that others at timely identification of clefts.

Figure 3.4 also shows that in 2020, 45% of clefts were diagnosed during antenatal screening and 42% were diagnosed at birth, leaving 13% diagnosed late according to the National Standard (as in 2017-19). The percentage of clefts diagnosed in a timely manner in 2020 varied by cleft service, ranging from 69% (Manchester, 40% before and 29% at birth) to 97% (South Wales, 54% before and 43% at birth). This variation suggests that practice varied considerably between maternity units, during 2020, with some achieving high rates of identification of clefts in a timely manner despite the challenges presented by the first year of the COVID-19 pandemic.

Figure 3.4. Percentage of timely diagnoses, including those made antenatally and at birth, among all CRANE-registered children, born 2017-19, according to cleft service – by comparison with children born 2020.



Note: Children missing diagnosis time are excluded. Includes unspecified cleft types.

Appendix 6 shows the breakdown of timing of diagnoses reporting for each cleft service, for 2017-19 as well as 2020 births.

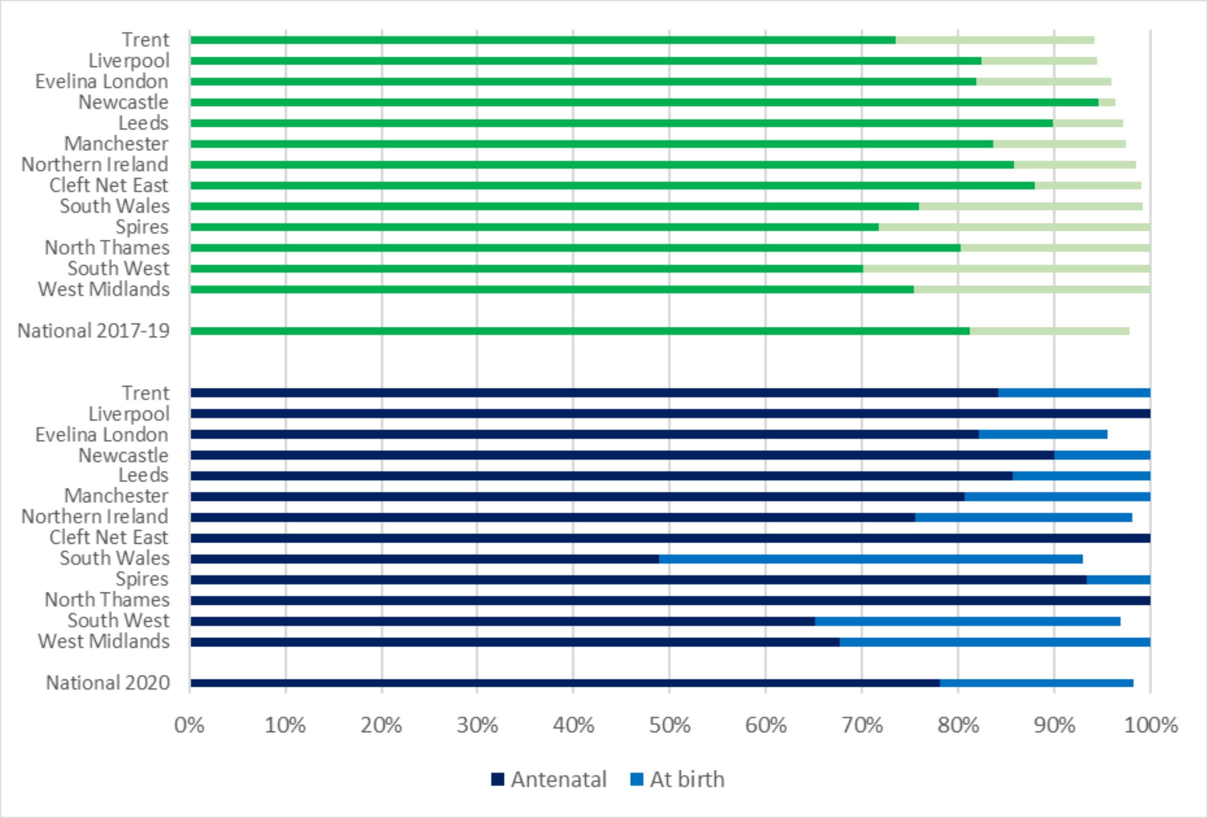
3.2.3. Timely diagnosis of clefts involving the lip (CL, UCLP and BCLP)

The variation in diagnoses of clefts involving the lip by cleft service were explored and are presented in Figure 3.5. In 2017-19, 81% of clefts involving the lip were diagnosed during antenatal screening and 17% were diagnosed at birth, leaving 2% diagnosed late according to the National Standard. Figure 3.5 also shows that the percentage clefts involving the lip diagnosed in a timely manner in 2017-19 varied by cleft service, ranging from 94% (Newcastle) to 100% (Liverpool, West Midlands, South West, and Northern Ireland).

Figure 3.5 also shows that in 2020, 78% of clefts involving the lip were diagnosed during antenatal screening and 20% were diagnosed at birth, leaving 2% diagnosed late according to the National Standard (as in 2017-19). The percentage of clefts involving the lip diagnosed in a timely manner in 2020 varied by cleft service, ranging from 93% (Spires) to 100% (for 9 of 13 cleft services).

These figures show that services achieved high rates of identification of clefts involving the lip in 2020, in a timely manner and consistent with 2017-19, despite the challenges presented by the first year of the COVID-19 pandemic.

Figure 3.5. Percentage of timely diagnoses, including those made antenatally and at birth, among CRANE-registered children with clefts involving the lip (CL, UCLP and BCLP), born 2017-19, according to cleft service – by comparison with children born 2020.



Note: Children missing diagnosis time are excluded. Includes unspecified cleft types.

Appendix 6 shows the breakdown of timing of diagnoses reporting for each cleft service, for 2017-19 as well as 2020 births.

Timely diagnoses of all clefts is a key performance indicator (#2) for cleft services and should be recorded for all registrations in the CRANE Database. Registering this information for cleft services ensures that children diagnosed with a cleft, receive the care and support that they and their families need, in a timely fashion.

3.3. Timely diagnosis of cleft palate

Cohort summary	Data source	The CRANE Database
	Birth years	• 10 years: 2011 to 2020 to explore trends over time • 3 years: 2017 to 2019 to explore regional differences
	Denominator	• For 2011-20 births: 4,153 children with cleft palate alone and a recorded diagnosis time • For 2017-19 births: 1,186 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 2011-20 with CP (n=4,422) had a recorded diagnosis time • 97% of children born 2017-19 with CP (n=1,217) had a recorded diagnosis time
	Countries	England, Wales and Northern Ireland
	Indicator	#2[14]
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 5 years between 2015 and 2019. 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.	

3.3.1. Timely diagnosis of cleft palate

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)[15]. Because of this, we continue to report on the timeliness of CP diagnoses.

Figure 3.6 shows that there have been significant improvements in the rates of timely diagnosis of CP (antenatally or within 24 hours of birth) over the last 10 years (p=0.005) with rates increasing from 66% for children born in 2012 to 75% 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'*[16] 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.

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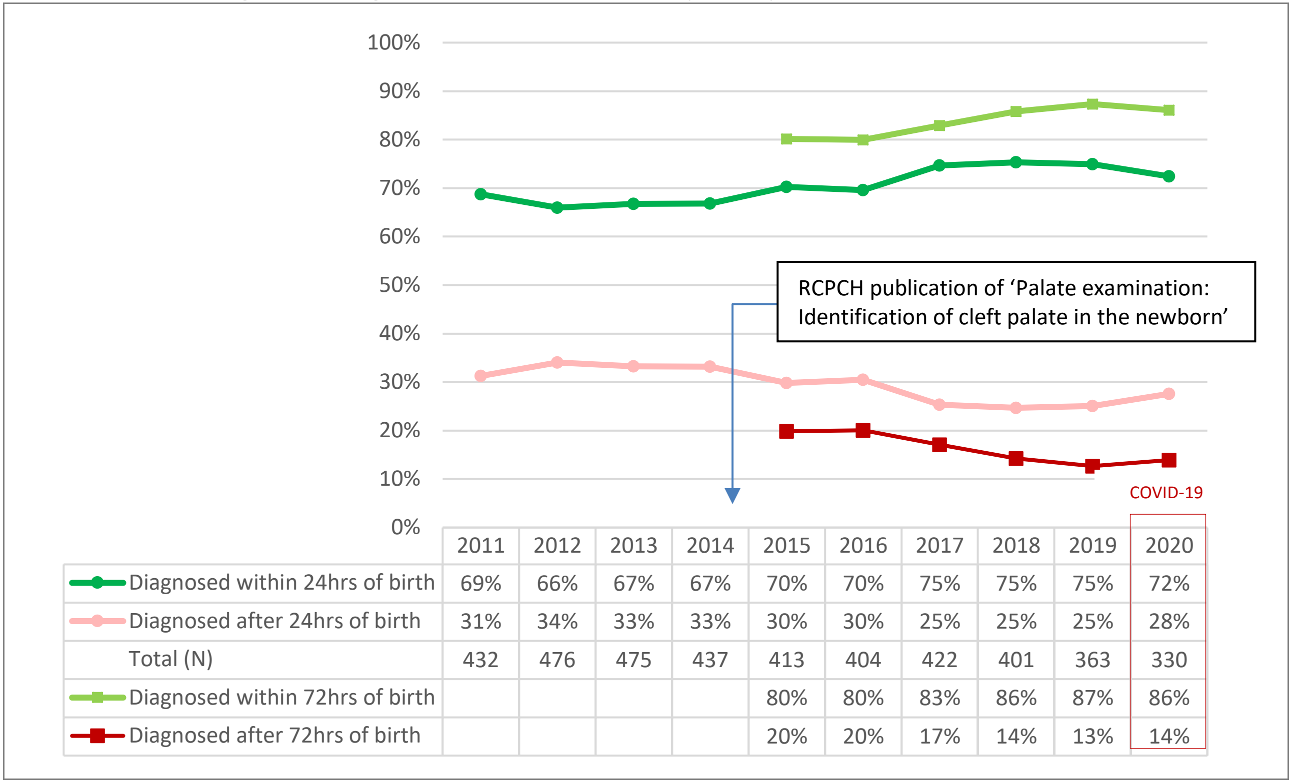
[14] CRANE core indicators are detailed in **Appendix 3**.

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

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

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. COVID has arrested the year on year improvement in timely diagnoses, this may reflect stresses on clinical services in light of the pandemic.

Figure 3.6. Rates of timely diagnosis (antenatal and within 24 hours of birth), as well as diagnoses within 72 hours of birth, among CRANE-registered children with CP, by birth year.



	2011	2012	2013	2014	2015	2016	2017	2018	2019	2020
Diagnosed within 24hrs of birth	69%	66%	67%	67%	70%	70%	75%	75%	75%	72%
Diagnosed after 24hrs of birth	31%	34%	33%	33%	30%	30%	25%	25%	25%	28%
Total (N)	432	476	475	437	413	404	422	401	363	330
Diagnosed within 72hrs of birth					80%	80%	83%	86%	87%	86%
Diagnosed after 72hrs of birth					20%	20%	17%	14%	13%	14%

Note: Children missing diagnosis time are excluded.

Current RCPCH guidance is that full newborn examinations should be undertaken within 72 hours of birth[17]. This now includes a complete palate examination as per the 2014 palate examination document.

Figure 3.6 (above) demonstrates a significant improvement in the rate of CP diagnosis within 72 hours[18] over the last six years (ranging from 87% in 2019 vs. 80% in 2015, p=0.015). This means that almost 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 5 years from 2015 to 2019 (inclusive) has been greater in the 72 hours data. 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.

It is worth noting that the impact of the COVID-19 pandemic on diagnosis times (and recording of this data) since March 2020 has yet to be fully quantified.

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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[17] 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

[18] The option to collect this information was added to the CRANE Database in May 2014.

3.3.2. Diagnosis times for children with a cleft palate by region

Table 3.1 shows that, overall, 2.4% of CPs were diagnosed during antenatal screening and 72.5% were diagnosed at birth, leaving 25.1% diagnosed late according to the National Standard. Clinically, the diagnosis of cleft palates antenatally is extremely difficult on standard 20-week anomaly ultrasound scans.

Table 3.1 also shows that, although a total of 75% of CPs were diagnosed in a timely manner (antenatal or at birth), rates varied by cleft service. Ranging from 65% (Evelina London, 4% before and 61% at birth) to 90% (West Midlands, 1% before and 89% at birth). Meaning the percentage of children diagnosed late according to the National Standard ranged from 35% to 10% between services. This wide and significant variation between rates of diagnoses within 24-hours of birth and after 24-hours of birth (p<0.001) suggests that practice varies considerably between maternity units, with some better that others at identifying a cleft of the palate during the new-born examination.

Table 3.1. Number (%) of CRANE-registered children born between 2017 and 2019 with a cleft palate, according to the timing of diagnosis and region / unit.

Cleft service	**Time of diagnosis in relation to birth n (%)**							
	Antenatal	**At birth**	**< 72 hours**	**≤1 week**	**≤1 month**	**≤6 months**	**>6 months**	**All**
Newcastle	1 1.7%	41 70.7%	2 3.4%	5 8.6%	3 5.2%	6 10.3%	0 0%	58
Leeds	1 1.3%	53 69.7%	9 11.8%	7 9.2%	2 2.6%	4 5.3%	0 0%	76
Liverpool	1 1.4%	48 67.6%	15 21.1%	2 2.8%	4 5.6%	1 1.4%	0 0%	71
Manchester	0 0.0%	48 68.6%	9 12.9%	6 8.6%	5 7.1%	1 1.4%	1 1.4%	70
Trent	1 0.8%	81 64.8%	16 12.8%	10 8%	4 3.2%	9 7.2%	4 3.2%	125
West Midlands	1 0.7%	123 89.1%	4 2.9%	4 2.9%	5 3.6%	1 0.7%	0 0%	138
Cleft Net East	0 0.0%	59 72%	12 14.6%	6 7.3%	3 3.7%	2 2.4%	0 0%	82
North Thames	9 6.0%	110 73.3%	9 6%	8 5.3%	4 2.7%	10 6.7%	0 0%	150
Spires	2 2.1%	73 76.8%	9 9.5%	2 2.1%	4 4.2%	3 3.2%	2 2.1%	95
South Wales	1 2.0%	43 84.3%	3 5.9%	0 0%	2 3.9%	2 3.9%	0 0%	51
South West	6 8.1%	57 77%	5 6.8%	4 5.4%	2 2.7%	0 0%	0 0%	74
Evelina London	6 4.0%	92 61.3%	24 16%	10 6.7%	11 7.3%	6 4%	1 0.7%	150
Northern Ireland	0 0.0%	32 69.6%	5 10.9%	1 2.2%	4 8.7%	0 0%	4 8.7%	46
All	**29 2.4%**	**860 72.5%**	**122 10.3%**	**65 5.5%**	**53 4.5%**	**45 3.8%**	**12 1%**	**1,186**

Note: Recording of 'timing of diagnosis' within 72 hours commenced in May 2014 to align CRANE data collection with NIPE standards[19].

It is worth noting that the percentage of children diagnosed within 72-hours of birth ranged from 76% (Newcastle) to 93% (West Midlands). This variation between rates of diagnoses within 72-hours of birth and after 72-hours of birth was also found to be statistically significant (p=0.013). Diagnosis time among CP patients born over the first year impacted by the COVID-19 pandemic, 2020, are presented in **Appendix 6**.

Overall, the most recent CRANE data show an encouraging trend with regards to timely detection of cleft palates. Nevertheless, 9% of children with a CP were not diagnosed until they were more than one week old, which is concerning given that the National Standard states that clefts should be diagnosed within 24 hours of birth to enable immediate referral to a specialist hospital. This helps to ensure the baby, and their family, receive appropriate care and support as soon as possible.

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

3.4. Gestational age and birth weight

Cohort summary	Data source	The CRANE Database (consented cases only)
	Birth years	Three years: 2017-2019. Plus summary information on 2020 births[20]
	Denominators	• 1,023 (2017-2019) and 489 (2020) children with a recorded gestational age • 1,017 (2017-2019) and 489 (2020) children registered with a recorded birth weight • 857 born at term (≥37 weeks) with a recorded birth weight
	Numerators	• children born prematurely (<37 weeks' gestation) • children born at term with a low birth weight (<2500 g) • 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	• 50% of 2,052 eligible consented children had a recorded gestational age and birth weight. • 0.2% had a reason why gestational age or birth weight were not collected. • 49.8% were missing 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). • Among babies born in 2019 in the general population of England & Wales, 8% of them were born prematurely, 7% of them had a low birth weight (LBW) and 11% of them had a high birth weight (HBW)[21].	
What did we find?	• The average gestational age was 38.5 weeks (2017-2019). • The percentage of premature births among children born with a cleft was 14% and is higher than in the general population (8% of premature births) (2017-2019). • The average birth weight was 3,136 g. Children with CL had a higher average birth weight (3,297 g) than those with a CP (3,087 g), UCLP (3,105 g) and BCLP (3,074 g) (2017-2019). • Among children born at term (≥37 weeks), 7% of them weighed <2500 g (LBW) and 8% of them weighed ≥4000 g (HBW) (2017-2019).	
Recommendations	• Cleft services should aim to improve the reporting of gestational age to allow us to report on the percentage of pre-term babies within the cleft population, and on service adherence to national timing of surgery guidance. • Cleft services should aim to improve the reporting of birth weight. This is important information that helps to monitor the percentage of babies with LBW and HBW among the cleft population. • The research community should validate and further investigate the higher percentage of premature births among children diagnosed with a cleft. • CRANE will engage with the RCM and Royal College of Obstetricians and Gynaecologists (RCOG) to communicate our findings.	

3.4.1. Data completeness for gestational age

Among children born in 2017-2019 whose families had consented to their children's data being collected in the CRANE Database (n=2,052), 50% children had gestational age reported to CRANE[22]. For children registered in

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[20] Affected by the COVID-19 pandemic and the lockdown that ensued.

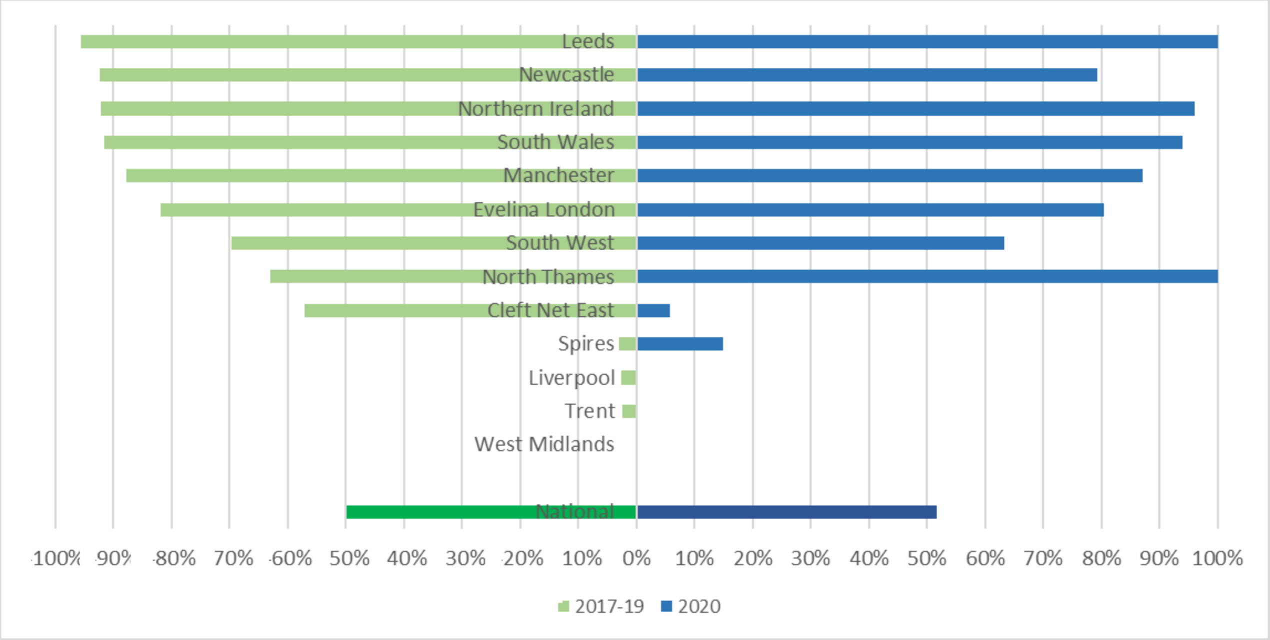
[21] Birth characteristics in England and Wales: 2019. Available at: https://www.ons.gov.uk/peoplepopulationandcommunity/birthsdeathsandmarriages/livebirths/bulletins/birthcharacteristicsinenglandandwales/2019 [Last accessed: 26 Oct 2021]

[22] Among the remaining 50% of consented children without gestation data in 2017-19, four of them did not have this outcome collected as they were transferred out or unavailable for data collection ('other reason' in CRANE).

2020 (n=489 children), the first year impacted by the COVID-19 pandemic, 52% had a recorded gestational age. Figure 3.7 shows the variation in the percentage of consented children with a recorded gestational age for 2017-19 (2%-96% range) and for 2020 (0%-100% range) for each Cleft Service.

It is important to record gestation data in the CRANE Database for all children diagnosed with a cleft because it is an essential data item required to monitor the percentage of children born prematureprematurely with a cleft, for risk adjustment of outcomes of cleft care; and to report on services' adherence to national timing of surgery guidance.

Figure 3.7. Percentage of CRANE-consented children with recorded gestational age in 2017-19 and 2020, by cleft services and national percentage for 2017-19 and 2020 registrations.



Note: Al children registered in CRANE between 01 January 2017 and 31 December 2020, by the 30 June 2021. Children without consent for data collection at birth are excluded. West Midlands cleft service submitted no gestational age data between 2017 and 2020.

3.4.2. Gestational age

Among babies born between 2017 and 2019 with known gestational age at birth (n=1,023), 14% were born prematurely (< 37 weeks' gestation), compared to 12% in the 2020 cohort. Raw data in **Appendix 6** shows the variation in the percentage of children born prematurely in the 2017-19 cohort, compared to 2020 registrations, for each cleft service. It is higher than the reported 8% national average reported for premature births in England and Wales[23] in 2019.The average gestational age for the 2017-19 cohort was 38.5 weeks (95% confidence interval: 38.3 to 38.6 weeks). 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

Among children born in 2017-2019 whose families had consented to their children's data being collected in the CRANE Database (n=2,052), 50% children had birth weight recorded[24]. For children registered in 2020 (n=489 children), the first year impacted by the COVID-19 pandemic, 51% had a recorded birth weight. Figure 3.8 shows

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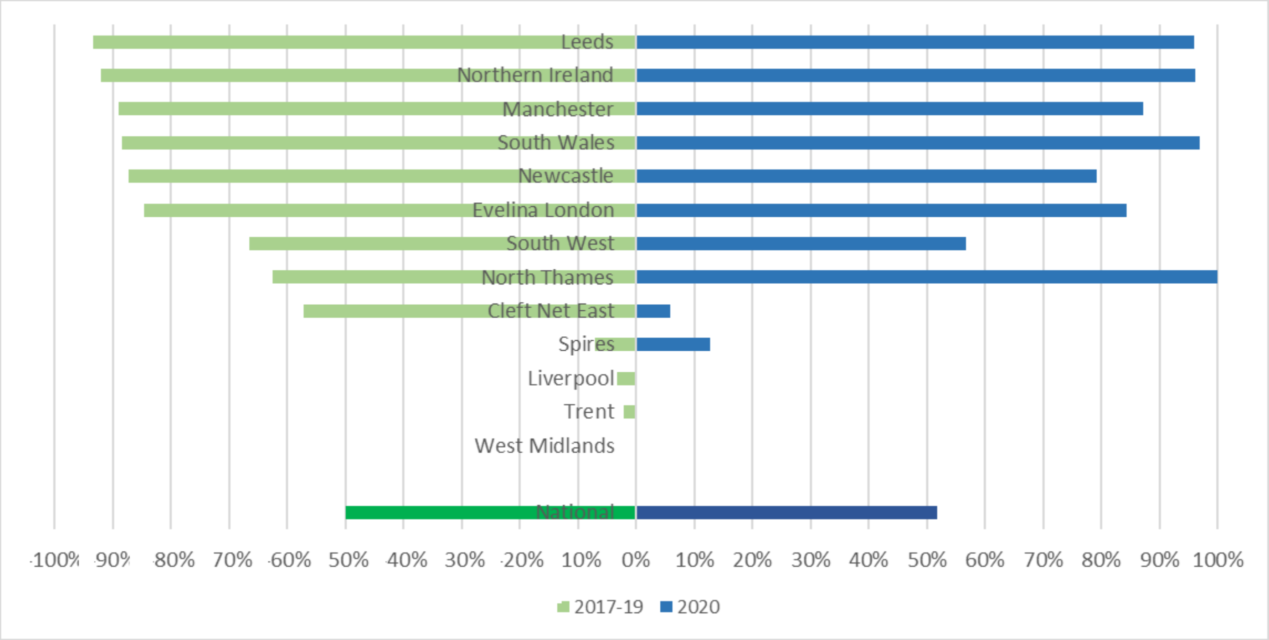
[23] Birth characteristics in England and Wales: 2019 - Available at: https://www.ons.gov.uk/peoplepopulationandcommunity/birthsdeathsandmarriages/livebirths/bulletins/birthcharacteristicsinenglandandwales/2019 [Last accessed: 15/10/2021]

[24] Among the remaining 50% of consented children without gestation data in 2017-19, four of them did not have this outcome collected as they were transferred out or unavailable for data collection ('other reason' in CRANE).

the variation in the percentage of consented children with a recorded birth weight for 2017-19 (2%-93% range) and for 2020 (0%-96% range), for each cleft service.

It is important to record birth weight data in the CRANE Database for all children diagnosed with a cleft. Cleft services should aim to improve the reporting of birth weight because it helps to monitor the percentage of babies born with a low birth weight (<2500 g) or with a high birth weight (≥4000 g).

Figure 3.8. Percentage of CRANE-consented children with recorded birth weight in 2017-19 and 2020, by cleft services and national percentage for 2017-19 and 2020 registrations.



Note: All children registered in CRANE between 01 January 2017 and 31 December 2020, by the 30 June 2021. Children without consent for data collection at birth are excluded. West Midlands cleft service submitted no birth weight data between 2017 and 2020.

3.4.4. Birth weight

Among consented cases with known birth weight, the overall average birth weight for 2017-2019 registrations (n=1,017) was 3,136 g (95% CI: 3,095 g to 3,178 g), compared to 3,242 g (95% CI: 3,164 g to 3,320 g) among 2020 registrations (n=251). For the same cohort, average birth weight was significantly lower among children diagnosed with a CP (3,087 g, 95% CI: 3,021 g to 3,152 g) (p-value<0.001), UCLP (3,105 g, 95% CI: 3,014 g to 3,197 g) (p-value=0.003) and BCLP (3,074 g) (p-value=0.004, 95% CI: 2,947 g to 3,202 g), when compared to average birth weight of those diagnosed with a CL (3,297 g, 95% CI: 3,221 g to 3,733 g). 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 51% of all consented children from 2017-19 registrations, were missing this information.

857 of 1,017 consented children with known birth weight between 2017 and 2019 were born at term (84%). Among them, 7% and 8% were born with low birth weight (<2,500 g) and high birth weight (≥4,000 g), respectively, compared to 7% and 11% of children with low birth weight and high birth weight, respectively, in the general population[25]. Raw data in **Appendix 6** shows the variation in the distribution of birth weight category among children born at term in the 2017-19 and 2020 period for each cleft service. 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 half of all consented children were missing information on their gestational age at birth.

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

3.5. Contact with cleft services

Cohort summary	Data source	The CRANE Database
	Birth years	Three years: 2017-2019 Plus summary information on 2020 births[26]
	Denominators	• 1,980 (2017-2019) and 450 (2020) children with a recorded referral time. • 2,571 (2017-2019) and 772 (2020) children with a recorded contact time.
	Numerators	• children referred to a cleft service within 24 hours of birth. • children contacted by cleft service within 24 hours of referral.
	Data completeness	• 66% of children had a recorded referral time. • 86% of children had a recorded time of first contact. • 34% of children were missing data on referral time, while 14% were missing data on time of first contact.
	Countries	England, Wales and Northern Ireland
	Indicators	#3 & #4[27]
Benchmark	• Children with a cleft should be referred to cleft services within 24 hours of birth. • Cleft services should contact families of children within 24 hours of receiving a referral.	
What did we find?	• 83% of families with a recorded referral time, were referred to a cleft service within 24 hours of birth (2017-2019). This percentage varied significantly across cleft types and sex. • 96% of families were contacted by a cleft service within 24 hours of referral (2017-2019).	
Recommendations	• Cleft services should aim to record the contact and referral time of all registrations by continuing to work with referring obstetric, midwifery and neonatal units to improve timeliness of diagnosis and early referrals. • Regional variation in the percentage of children referred and contacted within 24 hours demonstrate that some cleft services have high levels of referrals and contacts within 24 hours. They should share their best practise recommendations with cleft services with lower rates.	

3.5.1. Data completeness

Out of 3,007 children born and registered in CRANE between 2017 and 2019, 66% had a recorded *referral time* to a cleft service. Of the 898 children registered in 2020, the first year impacted by the COVID-19 pandemic, 50% had a recorded referral time. Figure 3.9 below shows the variation in data completeness rate for the recording of referral time in 2017-19 (range: 31% to 90%) and 2020 (range: 13% to 92%) registrations for each cleft service.

86% of 3,007 children born and registered in CRANE between 2017 and 2019 had a recorded *contact time* after referral. The same percentage (86%) of the 898 children registered in 2020 had their contact time recorded in the Database. Figure 3.10 shows the variation in data completeness rate for the recording of contact time in 2017-19 (range: 32% to 99%) and 2020 (range: 44% to 100%) registrations for each Cleft Service.

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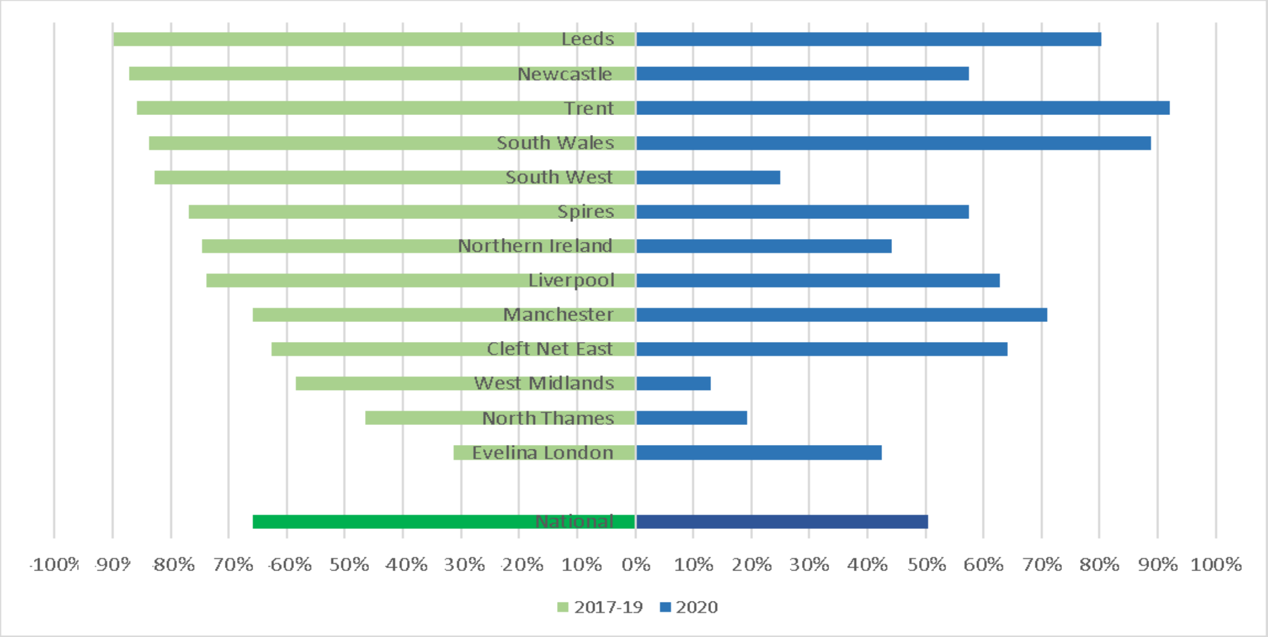
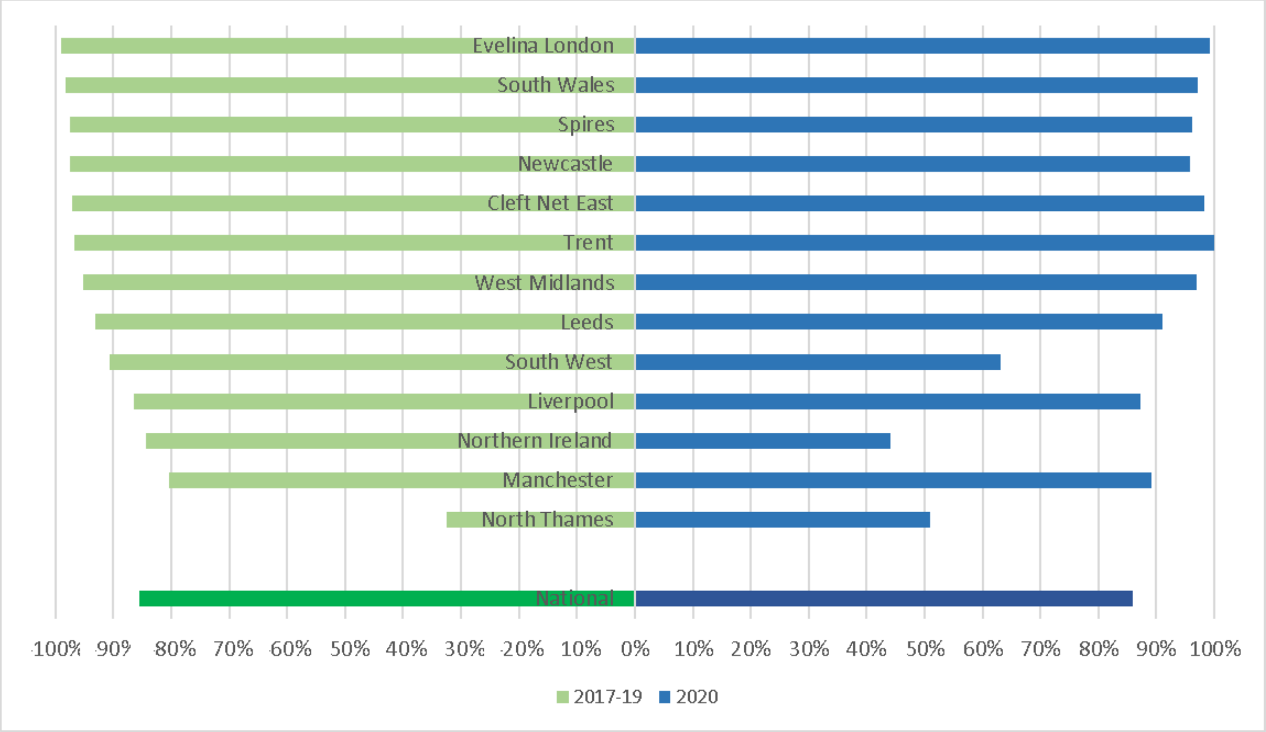
[26] Affected by the COVID-19 pandemic and the lockdown that ensued.
[27] CRANE core indicators are detailed in **Appendix 3**.

Figure 3.9. Percentage of CRANE-registered children with a recorded referral time in 2017-19 and 2020, by cleft services and national percentage for 2020 and 2017-19 registrations.



Note: All children registered in CRANE between 01 January 2017 and 31 December 2020, by the 30 June 2021. Excludes children who died within the first week after birth.

Figure 3.10. Percentage of CRANE-registered children with a recorded contact time in 2020, by cleft services and national percentage for 2020 and 2017-19 registrations.



Note: Data consists of all children registered in CRANE between 01 January 2017 and 31 December 2020, by the 30 June 2021. Excludes children who died within the first week after birth.

3.5.2. Referral and first contact

Out of 1,980 children with a recorded referral time in 2017-19, 83% of them were referred to a cleft service within 24 hours of birth, compared to 86% among the 450 children registered in CRANE in 2020 (the first year impacted by the COVID-19 pandemic).

This shows improvement in rate of referrals within 24 hours of birth over the last four years. Raw data in **Appendix 6** shows the variation in percentage of children referred within 24 hours of birth in 2017-19 (range: 74% to 88%) and 2020 (range: 67% to 94%) registrations, for each cleft service.

Furthermore, exploration of 2017-19 registrations showed that:

- Referrals also varied significantly by cleft type among the same cohort (p-value<0.001). Children diagnosed with CP had the lowest rate of early referrals within 24 hours of birth (70%), compared to CL (91%), 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.
- The percentage of children referred to a cleft service within 24 hours was significantly higher among boys (84%) than girls (81%) (p-value=0.034).

96% of 2,571 children with a recorded time of first contact with a cleft service in 2017-19 registrations were contacted within 24 hours of referral, compared to 94% among the 772 children registered in 2020 (the first year impacted by the COVID-19 pandemic). This small difference may reflect the challenges faced by cleft services during the COVID-19 period in 2020 to contact families of babies within 24 hours of referral. Raw data in **Appendix 6** shows the variation in percentage of children contacted within 24 hours of birth in 2017-19 (range: 84% to 100%) and 2020 (range: 80% to 100%) registrations, for each cleft service.

- 96% of boys and girls, respectively, were contacted within 24 hours of referral in 2017-19. 97% of children diagnosed with CL were contacted within 24 hours of referral in 2017-19, followed by those diagnosed UCLP (97%), BCLP (96%) and CP (95%).
- The percentage of children contacted by cleft services within 24 hours of referral in 2017-19 did not vary significantly by sex (p-value=0.624) or cleft type (p-value=0.318).

Recording of referral time and contact time are key performance indicators (#3 & #4) for cleft services and should be recorded for all registrations in the CRANE Database. Registering this information for cleft services ensures that children diagnosed with a cleft, receive the care and support that they and their families need, in a timely fashion.

3.6. CRANE consent

Cohort summary	Data source	The CRANE Database
	Birth years	Three years: 2011 to 2013 – key to running the 5-year outcomes outlier process Plus summary information on 2020 births[28]
	Denominator	3,261 children registered in CRANE
	Numerators	2,923 children whose families had made a decision about consent
	Data completeness	90% of 3,261 registered children had verified consent. This means they had agreed to or declined CRANE data collection.
	Countries	England, Wales and Northern Ireland
	Indicator	#5[29]
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?	• 90% 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%, consistent with previously reported high rates of positive consent.	
Recommendations	• Cleft services 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 services and the Nursing Clinical Excellence Network (CEN) to improve consent status verification.	

The families of 2,923 (90%) out of 3,261 children, born between 2017 and 2019, had made a decision to provide or decline consent (verified consent status). Of the families who had reached a decision, the rate of agreed consent was 98%, consistent with previously reported high rates of positive consent.

Figure 3.11 shows the variability in the verification of consent status according to the number of eligible cases within each cleft service. The funnel plot is centred on the adjusted national average of 90%.

The funnel plot shows that the rates of children with verified consent (consented or declined) varied by cleft service, ranging from 68.5% (Evelina London) to 99.5% (Leeds). The wide range may reflect the difficulty that some cleft services had in verifying consent during the COVID-19 pandemic.

Appendix 6 shows the breakdown of consent status, according to cleft services for 2011-13 births.

Completing this process of identifying outliers for this process indicator (#5), for the first time, has led to the identification of Evelina London as a negative outlier for the percentage of eligible cases with verified consent. This is because they fell below the lower 99.8% control limit, and were therefore excluded from the calculation of the overall average presented in the subsequent outcome funnel plots.

This process has also led to the identification of positive outliers such as Newcastle, Leeds, Manchester, and Northern Ireland cleft services.

See Box 1 and 2 for comments from cleft services on their outlier status.

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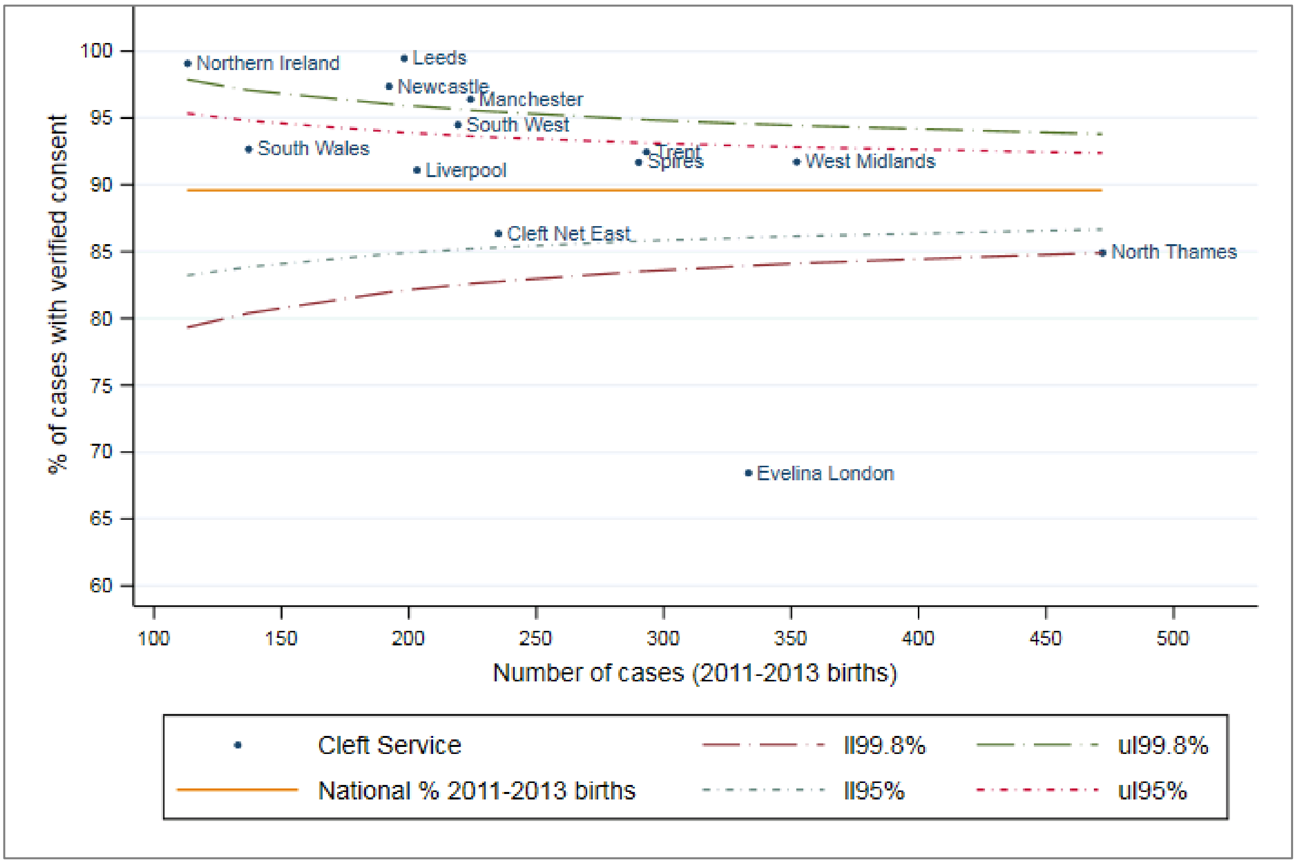
[28] Affected by the COVID-19 pandemic and the lockdown that ensued.

[29] CRANE core indicators are detailed in Appendix 3.

Figure 3.11. Funnel plot showing the percentage of CRANE-registered children (born 2011-2013) with verified consent, according to cleft service.



Note: Registered in CRANE by 30 June 2021. Funnel plot centred on the overall national percentage (89.6%) of children (born 2011-2013) with verified consent. See Appendix 6 for the raw data used to create this funnel plot.

Box 1. Summarised response to being a negative outlier for consent verification. **Full responses published in the "2021 Annual Report: Responses to outlier process" document.**

> "Upon investigation and performing a local audit, it was discovered that there are inconsistent multifactorial processes leading to low consent verification. Therefore...[an] action plan is proposed" to build consent verification into the existing nursing care pathway. In addition, cleft service team members will be accountable for the audit and the review of performance (for this indicator) on a monthly basis – often at team meetings – including the Lead CNS, the wider Directorate Management Team. Early implementation of this strategy "has already returned circa 80 retrospective consents to date".
>
> Evelina London cleft service, November 2021.

Box 2. Summarised responses to being positive outliers for consent verification. **Full responses published in the "2021 Annual Report: Responses to outlier process" document.**

> "Data collection was built into our service from the beginning and forms part of our care pathway…The final component of our 5 year data collection is the careful scheduling of 5 year audit clinics, with our audit coordinator identifying all patients for each cohort early every year, and then building specialist audit clinics around them over the 12 month period."
>
> Newcastle cleft service, November 2021.

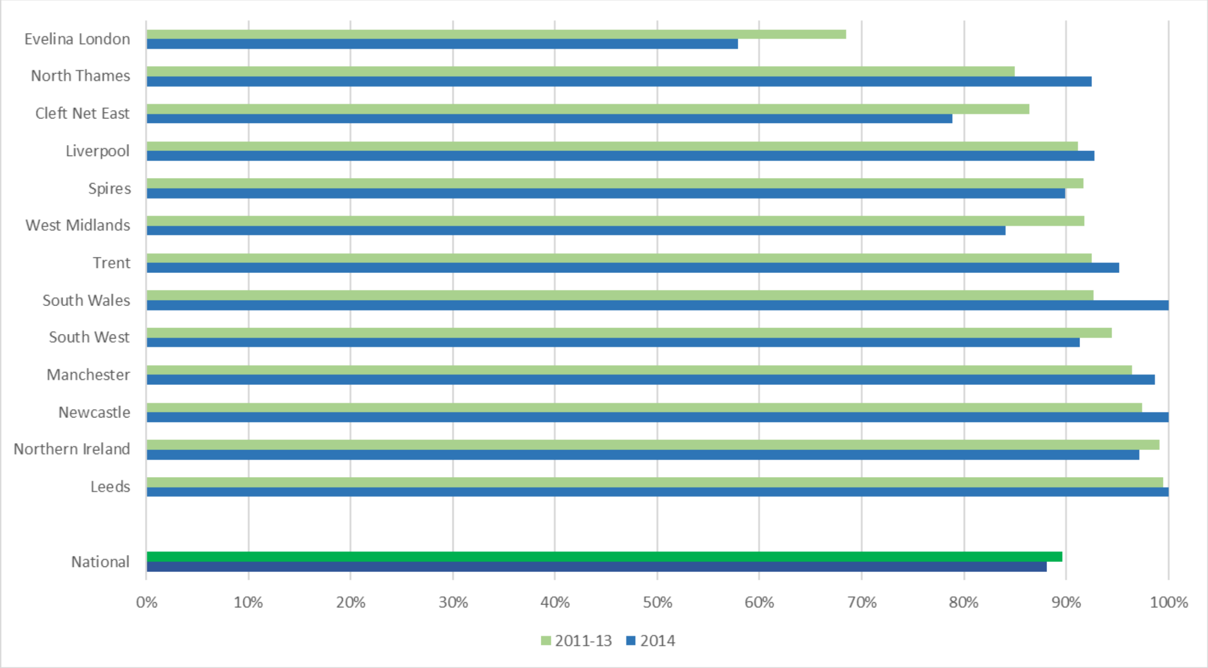
> "Our Cleft Team includes a dedicated Cleft Audit Coordinator….Much of the credit and responsibility for achieving these high levels of data submission onto CRANE is due to [Cleft Audit Coordinator]'s efforts and other members within the Cleft Administrative Team including … [the] Cleft Clinic Coordinator"
>
> Leeds cleft service, October 2021.

"We have reliable systems in place for consent verification. This is managed by the cleft coordinator who immediately updates CRANE when consent is given and highlights any patients attending clinic who need consent updating." Manchester cleft service, November 2021.
"CRANE have been clear that without initial consent at diagnosis then any further data cannot be included on CRANE, therefore the CNS is extremely diligent about discussing CRANE consent with families as soon as possible after making contact and it is appropriate to do so. She identifies and endeavours to follow up any families who have yet to provide consent by telephone. The CNS and Cleft coordinator continue to work tirelessly to identify any families who do not initially consent to inclusion on CRANE to ensure NI data can be as complete as possible". Northern Ireland cleft service, October 2021.

Anticipating challenges to consent verification due to the COVID-19 pandemic, Figure 3.12 summarises consent verification rates for children born in 2014 (88% achieved nationally), which will be reported on in the CRANE 2022 Annual Report, by comparison with consent verification rates in 2011-13.

Appendix 6 also shows the breakdown of consent status, according to cleft services for 2014 births.

Figure 3.12. Percentage of CRANE-registered children born 2014, with verified consent, according to cleft service – by comparison with children born 2011-13.



Note: Registered in CRANE by 30 June 2021. 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. See Appendix 6 for the raw data used to create this graph.

Seeking informed consent is a key performance indicator (#5) for cleft services, and the status of this should be recorded for all registrations in the CRANE Database. Registering this information for cleft services ensures that outcome information for children diagnosed with a cleft, can be audited in future years and reported on. For the benefit of patients and their families, as well the clinical community and commissioners.

4. Audit Outcomes at 5 years of age

This chapter details cleft-related outcomes for children at 5 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 services and are therefore excluded from reporting for all audit outcomes at 5 years of age. Submucous clefts comprised 3% of consented cases born between 1 January 2011 and 31 December 2013.

In previous years, we have reported on outcomes by individual cleft services. In 2020 and 2021, cleft services 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 three years of outcome data from children born 2011 to 2013 for child growth, dental health, facial growth, speech and psychology, and we provide summary data for 2014 births (as specified below).

Timeframe: The most recent four years of data, presented in two cohorts of patients.

- **Cohort 1.** Children born between 01 January 2011 and 31 December 2013 (consistent with a rolling 3-years, as for other sections of this report).
- **Cohort 2.** Children born between 01 January 2014 and 31 December 2014; these are presented separately to allow comparison of the effect of the COVID-19 pandemic and the lockdown that ensued.

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	The CRANE Database (consented cases only)
	Birth years	Three years: 2011 to 2013. Plus summary information on 2014 births[30]
	Denominator	1,253 Children with a recorded height and weight at 5 years
	Numerator	Number of children in each Body Mass Index (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[31] • Children without recorded height and weight at the age of 5 • Cleft services with consent verification outlier status (excluded from data completeness checks)
	Data completeness	• 44% of 2,850 eligible consented children had a recorded height and weight. • 0.5% of children only had a recorded height. • 1% of children only had a recorded weight. • 8% had a reason the child growth outcomes were not collected. • 46% were missing data or a reason for not collecting data.
	Countries	England, Wales and Northern Ireland
	Indicator	#6[32]
Benchmarks	• For 5-year-olds in the UK[33], 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. • National prevalence of underweight, overweight and obesity among 5 year olds was estimated at 1%, 13% and 10%, respectively[34].	
What did we find?	• 87% of children had a healthy BMI. • 2% of children were underweight, 7% overweight and 4% obese – according to BMI categorisation.	
Recommendations	Cleft services 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

Participation in a national clinical audit, such as the CRANE Database, requires all cleft services to submit growth data (height and weight) for all consented 5-year old children diagnosed with a cleft lip and/or palate. If growth data are not available, cleft services are asked to provide a reason for this (e.g. Patient transferred out of area or patient did not attend appointment). **Appendix 8** shows the breakdown of the percentage of children with reported height and weight at 5 years of age for each cleft service for the 2011-13 birth cohort.

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[30] Affected by the COVID-19 pandemic and the lockdown that ensued.

[31] Submucous cleft palate patients excluded from all 5 year outcomes as all/most teams do not audit these patients. Including cases with LAHSAL code identified as not possible, and likely submucous, when reviewed by cleft clinicians.

[32] CRANE core indicators are detailed in **Appendix 3**.

[33] According to the Royal College of Paediatrics and Child Health and UK-WHO growth charts – 2-18 years (Last accessed online: 4 November 2021) – Available at: https://www.rcpch.ac.uk/resources/uk-who-growth-charts-2-18-years

[34] National Child Measurement Programme Tables, England 2019/20 School Year – Published 29th October 2020 (Last accessed: 10 November 2021) – Available at https://digital.nhs.uk/data-and-information/publications/statistical/national-child-measurement-programme/2019-20-school-year

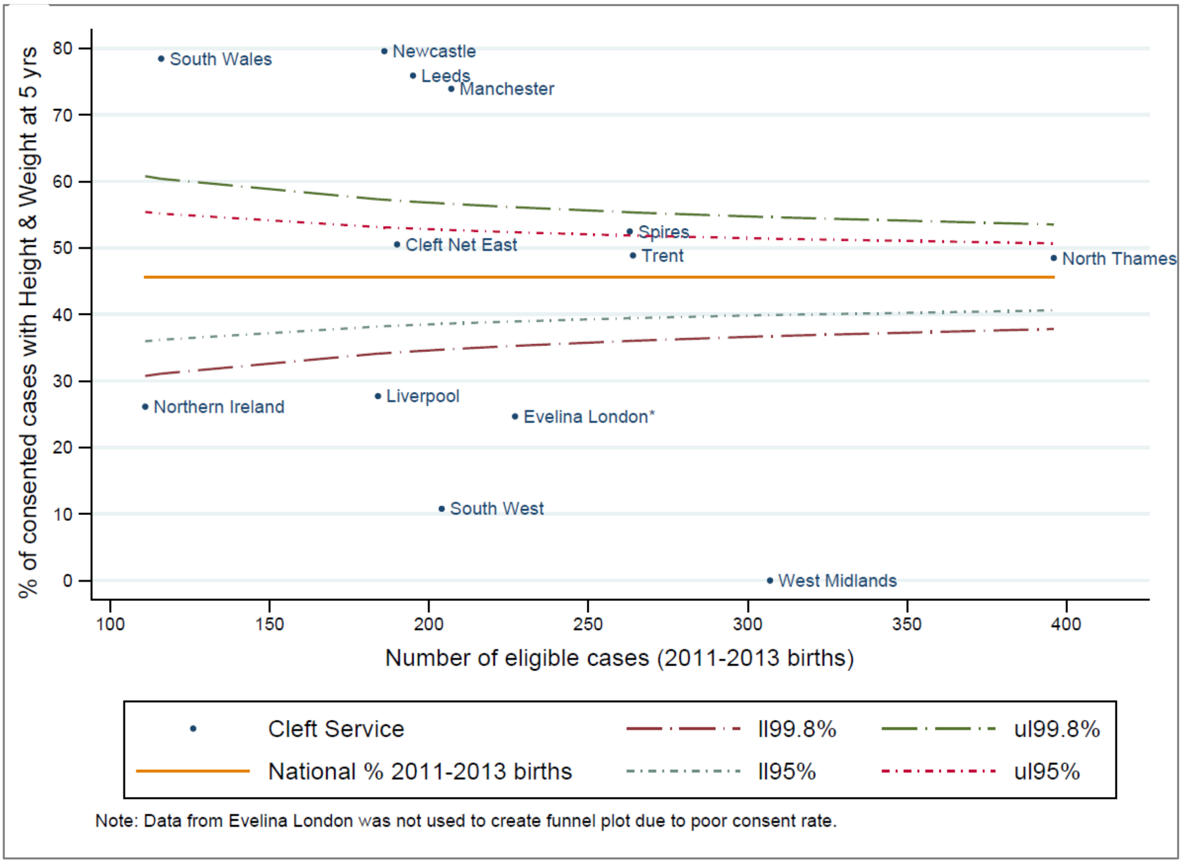
Of the 2,850 eligible 5-year old children born between 2011 and 2013, 44% (0% to 80% range between services) had a 5-year old height and weight reported. A further 2% of eligible cases had either only height or weight recorded. 8% had a reason why the outcome was not reported, and 46% were missing data or a reason.

Reasons for not collecting 5-year old child growth outcome data for 2011-13 births

Appendix 7 shows the breakdown of reasons reported for not collecting 5-year old child growth outcome data for 2011-13 births; the most common reason selected was 'Patient did not attend (DNA)' (35%).

Figure 4.1 shows the variability in the reporting of height and weight at 5 years of age according to the number of eligible cases in each cleft service. The funnel plot is centred on the adjusted national average of 46%[35]. The wide variability in the reporting of these 5 year old outcomes highlights challenges faced by some cleft services in recording the height and weight of 5 year old patients.

Figure 4.1 Funnel plot showing the percentage of CRANE-consented 5-year olds, born 2011-2013, with recorded child growth data submitted to CRANE, according to cleft service.



Note: Registered in CRANE by 30 June 2021. Funnel plot centred on the adjusted national percentage (45.6%) of children (born 2011-2013) with growth outcomes at the age of five years reported. See Appendix 8 for the raw data (and exclusions) used to create this funnel plot.

The funnel plot shows that Liverpool, West Midlands, South West, Northern Ireland and Evelina London are negative outliers for the percentage of eligible five years old with recorded growth data. This is because they are below the lower 99.8% control limit, and were therefore excluded from the calculation of the overall average presented in the subsequent healthy BMI outcome funnel. Completing this process of identifying outliers, for the first time, has also allowed the identification of positive outliers such as Newcastle, Leeds, Manchester, and South Wales. See Box 1 and 2 (below) for comments from cleft services on their outlier status.

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[35] The overall mean was 44.0% and adjusted mean was 45.6% (excluding cleft services with consent verification outlier status).

Box 1. Summarised responses to being negative outliers for '5 year old growth outcome data completeness at 5 years'. **Full responses published in the "2021 Annual Report: Responses to outlier process" document.**

"We have identified weaknesses in both resources and the process of collection of height and weight data for 5 year old patients attending audit clinics that account for the poor performance in this domain. Moving forward the process will be reviewed by the Liverpool cleft team to ensure better compliance with collection of height and weight data and entry of this data onto the CRANE Database."

Liverpool cleft service, November 2021.

There was not a robust system in place in 2018 – 2019 to collect the growth data and enter it onto CRANE due to a number of workforce issues and no data coordinator role within the team."

West Midlands cleft service, October 2021

"Our area covered by the South West Cleft Service is large: from Gloucester to the Isles of Scilly. We have a main hub in Bristol and run spoke clinics in five other centres (reduced from six previously). Measurements such as child growth and dental health are taken in these clinics. The pandemic response shut down our possibilities for face-to-face assessment of audit data set values."

South West cleft service, September 2021.

"Inconsistent process was revealed leading to incomplete data entry. In addition to creating a written policy and procedure and creating a systematic process for monitoring… Intake nurses"… will be given the authority and responsibility of taking and recording height and weight measurements.

Evelina London cleft service, October 2021.

"The data collection period was 2016 – 2018 for the birth cohort identified (2011-2013 births) and during this time we had 1 WTE Cleft Nurse Specialist (CNS), a combination of factors including staffing, patients failing to attend and children attending out of standard contributed to an incomplete data set. In order to address this issue in the future a second WTE CNS has been appointed and they will endeavour to ensure complete child growth audit outcomes are submitted to CRANE"

Northern Ireland cleft service, October 2021

Box 2. Summarised responses to being positive outliers for '5 year old growth outcome data completeness at 5 years'. **Full responses published in the "2021 Annual Report: Responses to outlier process" document,**

"Data collection was built into our service from the beginning and forms part of our care pathway…The final component of our 5 year data collection is the careful scheduling of 5 year audit clinics, with our audit coordinator identifying all patients for each cohort early every year, and then building specialist audit clinics around them over the 12 month period."

Newcastle cleft service, November 2021.

"Our Cleft Team includes a dedicated Cleft Audit Coordinator….Much of the credit and responsibility for achieving these high levels of data submission onto CRANE is due to [Cleft Audit Coordinator]'s efforts and other members within the Cleft Administrative Team including … [the] Cleft Clinic Coordinator"

Leeds cleft service, October 2021.

"We have reliable systems in place to collect and enter child growth data onto CRANE. The dental nurses collect and record height and weight data for 5 year old cleft patients attending audit clinics. There is a system in place to make sure this is not missed and appropriate equipment is available and accessible during cleft clinics. The cleft coordinator enters this data onto CRANE immediately after the audit clinics."

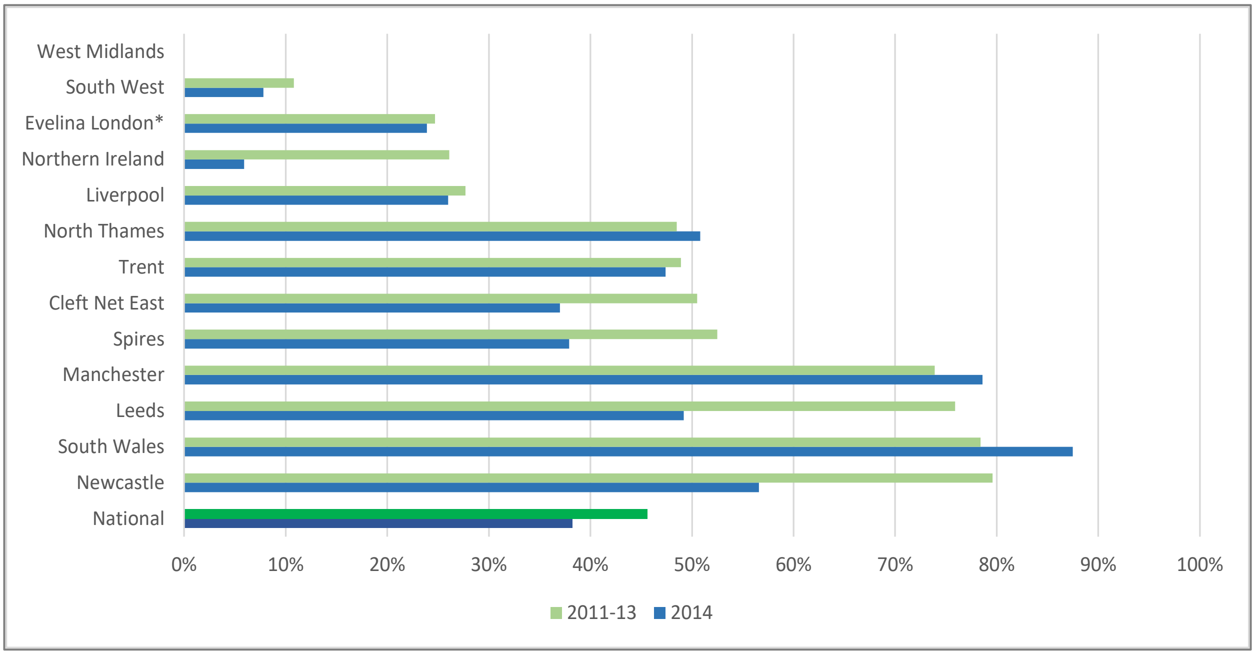
Manchester cleft service, November 2021.

> Good administration and dedicated clinics were quoted by this service as contributing to their positive outlier status. In addition, they "have the same dental nurse who takes height and weight measurements and also records the dmft scores and the reasons if not collected. This is documented on the clinic sheet and inputted by the cleft secretary when she is typing the clinic letters"
>
> South Wales cleft service, October 2021

Anticipating challenges to data completeness due to the COVID-19 pandemic, Figure 4.2 summarises data completeness for eligible children born in 2014 (reporting growth and height data); which will be reported on in the CRANE 2022 Annual Report. This is compared with 2011-2013 data to provide pre-pandemic context[36]. **Appendix 8 s**hows the breakdown of growth reporting for each cleft service, for 2011-13 as well as 2014 births.

As growth outcome data are not subject to the additional validation that other cleft care outcomes, such as speech and facial growth are subject to this may have been a factor in cleft services' ability to audit and submit 5 year old growth outcome data to the CRANE Database.

Figure 4.2. Percentage of CRANE-consented 5-year olds, born 2014, who had growth data submitted to CRANE, according to cleft service – by comparison with children born 2011-13.



Note: Registered in CRANE by 30 June 2021. See **Appendix 8** for the raw data (and exclusions) used to create this graph.

Overall, of the 872 eligible 5-year old children born in 2014, 38% had both height and weight reported (0%-88% range between services). 4 out of 13 cleft services were able to account for 50% or more of eligible cases.

Reasons for not collecting 5-year old child growth outcome data for 2014 births

Appendix 7 shows the breakdown of reasons reported for not collecting 5-year old child growth outcome data for 2014 births; the most common reason selected was 'Other' (47%). Comments provided to specify these 'Other' reasons, in the main, described clinics cancelled / data not collected due to the COVID-19 pandemic.

4.1.2. Height and weight

Analyses revealed that the average weight and average height among 5-year old children, born between 2011

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[36] A child born 31 December 2013, should have been audited for 5-year old outcomes before the age of 6. I.e. By the 31 December 2019 (pre-COVID-19 pandemic).

and 2013, with recorded growth data in the CRANE Database was 19.7 kg (95% CI: 19.5 kg to 19.9 kg) and 112.0 cm (95% CI: 111.7 cm to 112.3 cm), respectively.

These figures are consistent with UK national averages for 5-year-olds in the general population[37] for weight (range: 15 kg to 24 kg) and height (range: 99 cm to 119 cm). 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 (BMI)

For children born with a cleft between 2011 and 2013 and with recorded five year growth data, their 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[38]. Among consented children with 5-year old growth data recorded in the CRANE Database between 2011 and 2013, 87% had a healthy BMI, 7% were overweight, 4% were obese and 2% were underweight. The average BMI was 15.7 kg/m^2 (95% CI: 15.6 kg/m^2 to 15.7 kg/m^2).

4.1.4. Healthy BMI, by cleft service

Figure 4.3 shows the percentage of 5-year olds with healthy BMI, according to the number of eligible children with child growth data at 5 years of age (both height and weight) at each cleft service. The funnel plot is centred on the adjusted national average of 87%[39] (range 76%-90%).

Submitting this indicator of healthy BMI (#6) to the outlier process, for the first time, did not lead to the identification of any outliers for this outcome indicator.

Appendix 8 shows the breakdown of cases with healthy BMI at 5 years of age, as well as the other BMI categories, according to cleft service.

Anticipating challenges to recording height and weight before the age of 6, due to the COVID-19 pandemic: **Appendix 8** shows the breakdown of cases according to BMI categories, according to cleft service, for 2011-13 as well as 2014 births. Data for 2014 births will be reported on in the CRANE 2022 Annual Report.

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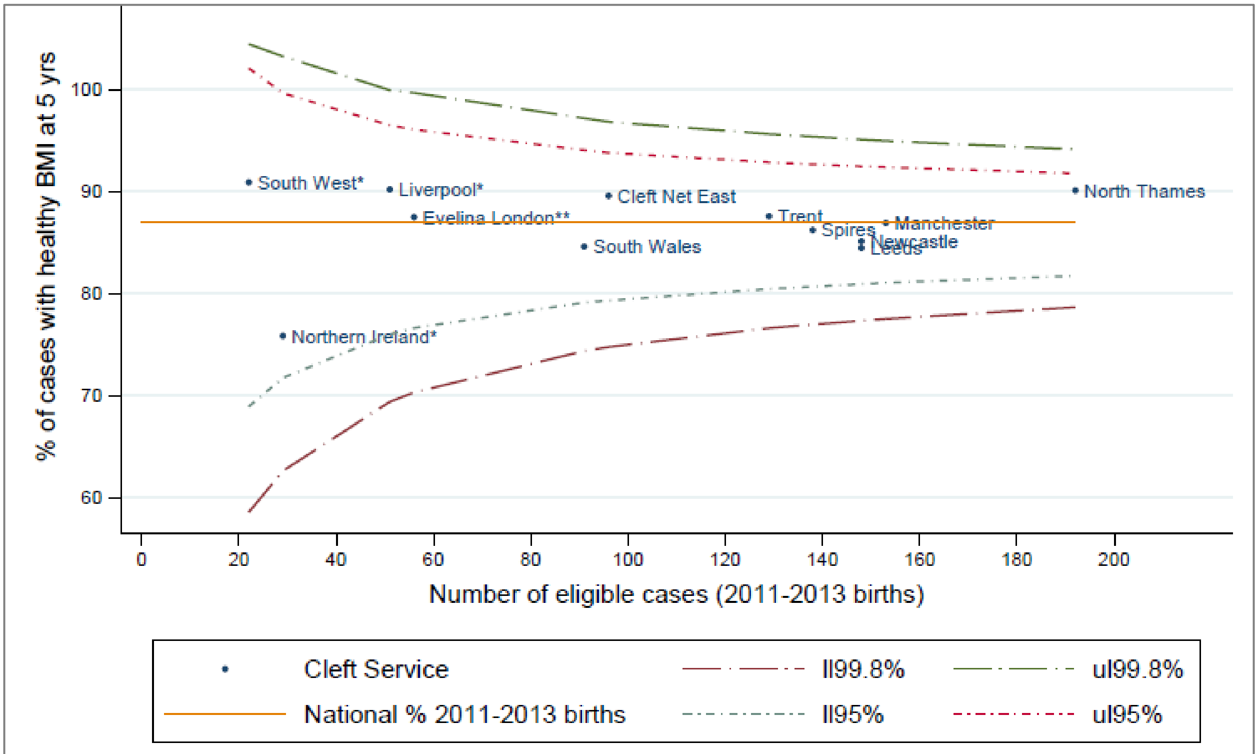
[37] According to the Royal College of Paediatrics and Child Health and UK-WHO growth charts – 2-18 years (Last accessed online: 4 November 2021) – Available at: https://www.rcpch.ac.uk/resources/uk-who-growth-charts-2-18-years

[38] According to the Royal College of Paediatrics and Child Health - 2-20 years Body Mass Index (Last accessed online: 4 November 2021). Available from: https://www.rcpch.ac.uk/resources/body-mass-index-bmi-chart

[39] Both the overall mean and adjusted mean (excluding cleft services with consent verification and data completeness outlier status) were 86.9%.

Figure 4.3 Funnel plot showing the percentage of CRANE-consented 5-year olds, born 2011-2013, with healthy BMI, according to cleft services



Note 1: Data from Evelina London was not used to create funnel plot due to poor consent verification rate.
Note 2: Data from Liverpool, West Mids, S West, Evelina & N.I. not used to create funnel plot due to poor data completion rates.
Note 3: West Midlands had no recorded Height & Weight (BMI) data.

Note: Registered in CRANE by 30 June 2021. Funnel plot centred on the adjusted national percentage (86.9%) of children (born 2011-2013) with healthy BMI at the age of five years reported. See Appendix 8 for the raw data (and exclusions) used to create this funnel plot.

4.2. Dental health

Cohort summary	Data source	The CRANE Database (consented cases only)
	Birth years	Three years: 2011 to 2013. Plus summary information on 2014 births[40]
	Denominators	• 1,586 5-year-old children with recorded decayed, missing or filled teeth (dmft) scores • 1,572 children with scores for the calculation of Treatment Index[41] • 1,575 children with scores for the calculation of Care Index[42]
	Numerators	• 602 children with at least one (>0) dmft • 221 children with extensive decay (dmft >5)
	Exclusions (not mutually exclusive)	• Children without consent to data collection • Children who died before the age of 5 years • Children with submucous cleft palates[43] • Children without a recorded dmft score at the age of 5 • Cleft services with consent verification outlier status (excluded from data completeness checks)
	Data completeness	• 56% of 2,850 eligible consented children had a recorded dmft score. • 10% had a reason dmft scores were not collected. • 34% were missing data or a reason for not collecting data.
	Countries	England, Wales and Northern Ireland
	Indicators	#7 & #8[44]
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[45]. • 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[46].	
What did we find?	• 38% 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 70%.	
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 services 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 services led). • Cleft services 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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[40] Affected by the COVID-19 pandemic and the lockdown that ensued.

[41] 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).

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

[43] Submucous cleft palate patients excluded from all 5 year outcomes as all/most teams do not audit these patients. Including cases with LAHSAL code identified as not possible, and likely submucous, when reviewed by cleft clinicians.

[44] CRANE core indicators are detailed in **Appendix 3**.

[45] 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

[46] 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 CRANE, means that all cleft services are asked to record dental health outcome data in the form of decayed, missing or filled teeth (dmft) scores for all consented 5-year-old children with a cleft lip and/or palate. If dmft score data are not available, cleft services are asked to report a reason for this (e.g. Patient transferred out of area or patient did not attend appointment, etc). Appendix 9 shows the breakdown of dental health reporting for each cleft service for the 2011-13 birth cohort.

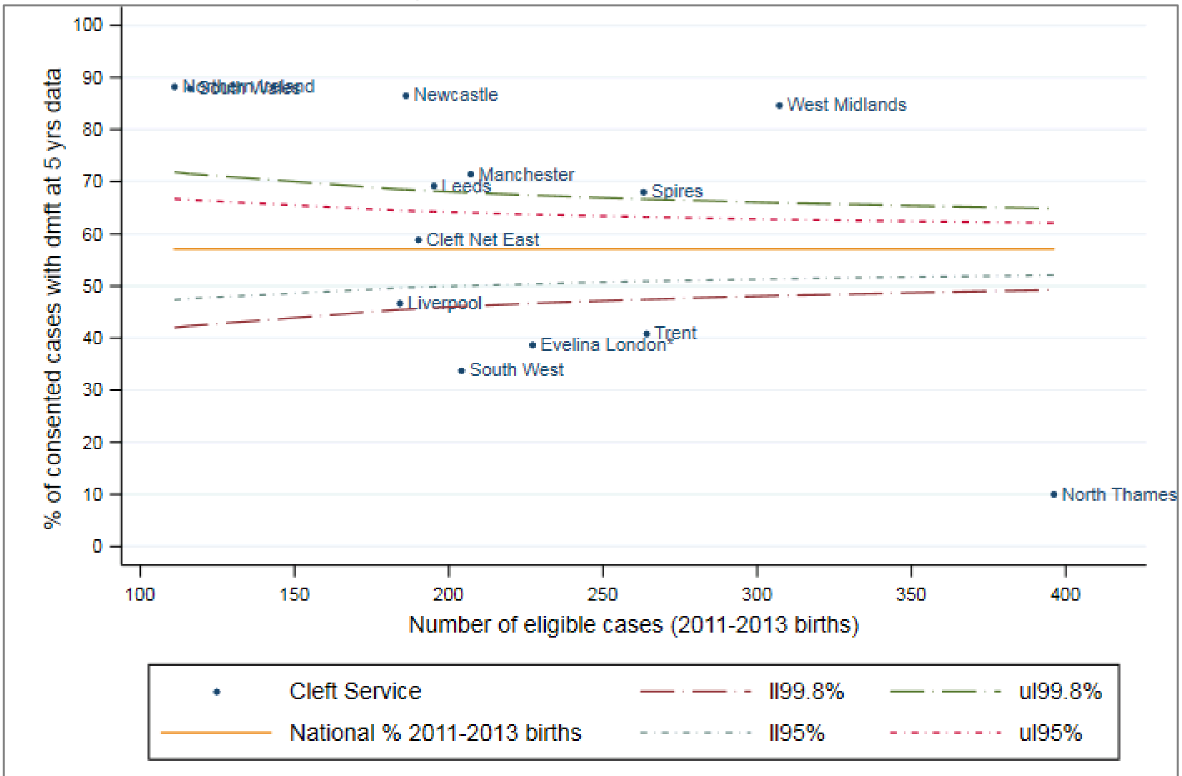
Of the 2,850 eligible 5-year old children born between 2011 and 2013, 56% had dmft scores[47] reported (10%-88% range between services). 10% of cases had a reason why the outcome was not reported, and 34% were missing data or a reason.

Reasons for not collecting 5-year old dental health outcome data for 2011-13 births

Appendix 7 shows the breakdown of reasons reported for not collecting dmft at 5 years of age for 2011-13 births; the most common reason selected was 'Lack of staff / facilities / equipment' (34%).

Figure 4.4 shows the variability in the reporting of dmft scores according to the number of eligible cases within each cleft service. The funnel plot is centred on the adjusted national average of 57%[48]. The wide range in reporting may reflect the difficulty that some cleft services had in performing dental health assessments during the COVID-19 pandemic; including difficulties due to some services having no dentists in post.

Figure 4.4. Funnel plot showing the percentage of CRANE-consented 5-year olds, born 2011-2013, who had dmft data submitted to CRANE, according to cleft service.



Note: Data from Evelina London was not used to create funnel plot due to poor consent rate.

Note: Registered in CRANE by 30 June 2021. Funnel plot centred on the adjusted national percentage (57.1%) of children (born 2011-2013) with dmft data reported. See Appendix 9 for the raw data (and exclusions) used to create this funnel plot.

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[47] This does not mean that the individual elements of the dmft score ('d' 'm' and 'f') were recorded in CRANE. 486 cases did not have a record of the scores for the individual elements, only a total dmft score. Cleft services will be liaised with to encourage completion of these data.

[48] The overall mean was 55.6% and adjusted mean was 57.1% (excluding cleft services with consent verification outlier status).

The funnel plot shows that Trent, North Thames, South West and Evelina London are negative outliers for the percentage of eligible cases with dmft data reported. This because they fell below the lower 99.8% control limit, and were therefore excluded from the calculation of the overall average presented in the subsequent outcome funnels. Completing this process of identifying outliers, for the first time, has also allowed the identification of positive outliers such as Newcastle, Leeds, Manchester, West Midlands, Spires, South Wales and Northern Ireland cleft services. See Box 3 and 4 (below) for comments from cleft services on their outlier status.

Box 3. Summarised responses to being negative outliers for 'dental health - data completeness at 5 years' – extracts. **Full responses published in the "2021 Annual Report: Responses to outlier process" document.**

"Trent Regional Cleft Network is not been staffed to provide a full paediatric dental service… the lack of clinical provision is reflected in the data completeness." They are "working to establish Service Level Agreements for attendance at the multidisciplinary cleft clinics with the Community Dental Services in each clinic area… [to] provide access to more specialist dental care in the community for cleft patients and provision of appropriate oral health advice" Trent cleft service, November 2021.
The recording of calibrated dmft data is "a long-standing problem and dates back to the unfortunate loss of 2 of our cleft calibrated paediatric dentists relatively close to one another...From a governance perspective, it is important to highlight for the sake of the commissioners that a dentally qualified clinician in the form of a paediatric dentist (staff grade/registrar) or an orthodontist has carried out a detailed dental examination at the time of the audit appointment. All cases where dental disease has been detected have been referred to the patient's own dentist or internally for dental treatment… the dental needs of our cleft patients are being managed but we were not in a position to make a calibrated score at their audit appointment." North Thames cleft service, October 2021.
"The pandemic has produced problems primarily of its own but also exposed weaknesses in the support that our service relies on. Maternity leave of the paediatric dental surgery consultant in the South West Cleft Service overlapped with the pandemic response. With only approximately 2.8 whole time equivalent consultant paediatric dentists in the South West of the UK at the time, and paediatric dentistry hard pushed with the work load of the region, a regular consultant replacement was not possible during this time. Therefore the dental health data will be incomplete." South West cleft service, September 2021.
"In reviewing our processes… for the birth cohort 2011 to 2013, it was noted that the data available at this time was collected by our then newly appointed part-time Consultant in Paediatric Dentistry… [who] had significant clinical backlogs to address which took up the majority of their working hours at that time." As well as "we do not have paediatric dental provision in our outreach clinics – an area where we do not have overarching control of local resources". Nevertheless, "new data collection processes were put in place…further staffing level increase(d)… increase(ing) our confidence that our service is now in a better position to gather data going forward." Evelina London cleft service, November 2021.

Box 4. Summarised responses to being positive outliers for ‘dental health - data completeness at 5 years’ – extracts. **Full responses published in the “2021 Annual Report: Responses to outlier process” document.**

“Data collection was built into our service from the beginning and forms part of our care pathway…The final component of our 5 year data collection is the careful scheduling of 5 year audit clinics, with our audit coordinator identifying all patients for each cohort early every year, and then building specialist audit clinics around them over the 12 month period.”

Newcastle cleft service, November 2021.

“Our…Team includes a dedicated Cleft Audit Coordinator…Much of the credit and responsibility for achieving these high levels of data submission…is due to [the Cleft Audit Coordinator]’s efforts and other members within the…Team including…[the] Cleft Clinic Coordinator”

Leeds cleft service, October 2021.

This service has “a consultant paediatric dentist 2 sessions per week and a dental therapist 4 sessions a week. There is always a member of the paediatric dental team present in the 5 year audit clinics and where possible this is a consultant paediatric dentist to comply with the CRANE requirements for audit data collection. There is an efficient system in place to ensure that dental health data is entered onto CRANE.”

Manchester cleft service, November 2021.

“The Consultant Dentist and the dental therapist are both calibrated in caries identification and enamel defects every two years to ensure the data collected is accurate and comparable nationally…our cleft coordinator, was given specific time out of her role to transfer the data from our Cleft database onto CRANE. This is very time consuming and not sustainable without having an identified data coordinator role for the cleft admin team.”

West Midlands cleft service, October 2021.

“In spite of not having a designated paediatric dentist on either the Salisbury or Oxford site our dental health data completeness is consistently good. We have a process for capturing data at the 5 year clinical & audit review appointment with administrative support for data input.”

Spires cleft service, November 2021.

Good administration and dedicated clinics were quoted by this service as contributing to their positive outlier status. In addition, they “have the same dental nurse who takes height and weight measurements and also records the dmft scores and the reasons if not collected. This is documented on the clinic sheet and inputted by the cleft secretary when she is typing the clinic letters”

South Wales cleft service, October 2021

“The Cleft Coordinator has always worked closely with the Consultant Dental Surgeon on submitting returns to CRANE, therefore they have a robust process in place which ensure high levels of data completeness. This involves cross checking and meeting regularly to upload CRANE records once completed.”

Northern Ireland cleft service, September 2021.

Anticipating challenges to data completeness due to the COVID-19 pandemic, Figure 4.5 summarises data completeness for eligible children born in 2014 (reporting of dmft data); which will be reported on in the CRANE 2022 Annual Report. This is compared with 2011-2013 data to provide pre-pandemic context[49]. **Appendix 9** shows the breakdown of dmft reporting for each cleft service, for 2011-13 as well as 2014 births.

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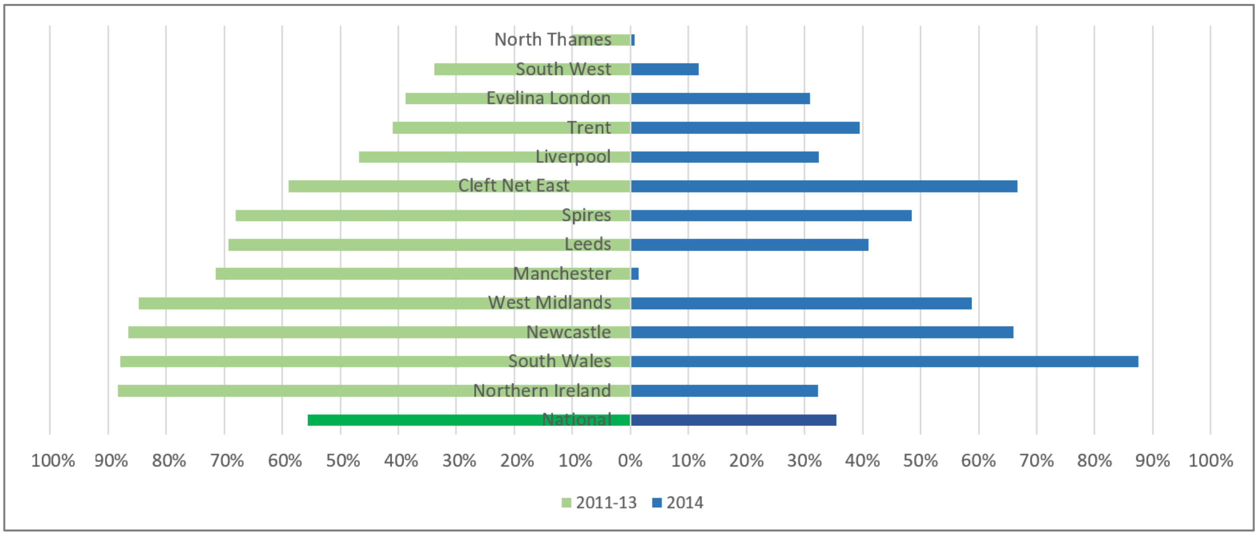
[49] A child born 31 December 2013, should have been audited for 5-year old outcomes before the age of 6. I.e. By the 31 December 2019 (pre-COVID-19 pandemic).

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, and the ongoing impact of the pandemic in 2021, may have impacted services' ability to audit and submit the most recent years of CRANE data collection.

Figure 4.5. Percentage of CRANE-consented 5-year olds, born 2014, who had dmft data submitted to CRANE, according to cleft service – by comparison with children born 2011-13.



Note: Registered in CRANE by 30 June 2021. See **Appendix 9** for the raw data (and exclusions) used to create this graph.

Overall, of the 872 eligible 5-year old children born in 2014, 35% had dmft data reported (1%-88% range between services). 4 out of 13 cleft services were able to account for more than 50% of eligible cases.

Reasons for not collecting 5-year old dental health outcome data for 2014 births

Appendix 7 shows the breakdown of reasons reported for not collecting dmft at 5 years of age for 2014 births; the most common reason selected was 'Other' (43%). Comments provided to specify these 'other' reasons, in the main, described clinics cancelled / data not collected due to the COVID-19 pandemic.

4.2.2. Decayed, missing and filled teeth (dmft) by cleft service

Among the 1,586 eligible children born in 2011-2013 with a reported dmft score, the mean number of dmft at 5 years was 2.05, with scores ranging from 0 to 20.

Overall, 38% of children had at least one dmft (>0).This is 5% higher than the general population average of 23%, 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 5% more of the CRANE cohort experienced dental decay, by comparison with the national population.

Furthermore, 14% of the CRANE cohort born in 2011-2013 had extensive decay (dmft >5) (as **defined below**).

The variation between cleft services by dental health outcomes dmft >0 and dmft >5 are presented below.

dmft >0 scores

Figure 4.6 shows the percentage of 5-year olds with at least one (>0) decayed missing or filled tooth, according to the number of children with valid dmft scores at each cleft service. The funnel plot is centred on the adjusted national average of 38%[50] (range 30%-50%).

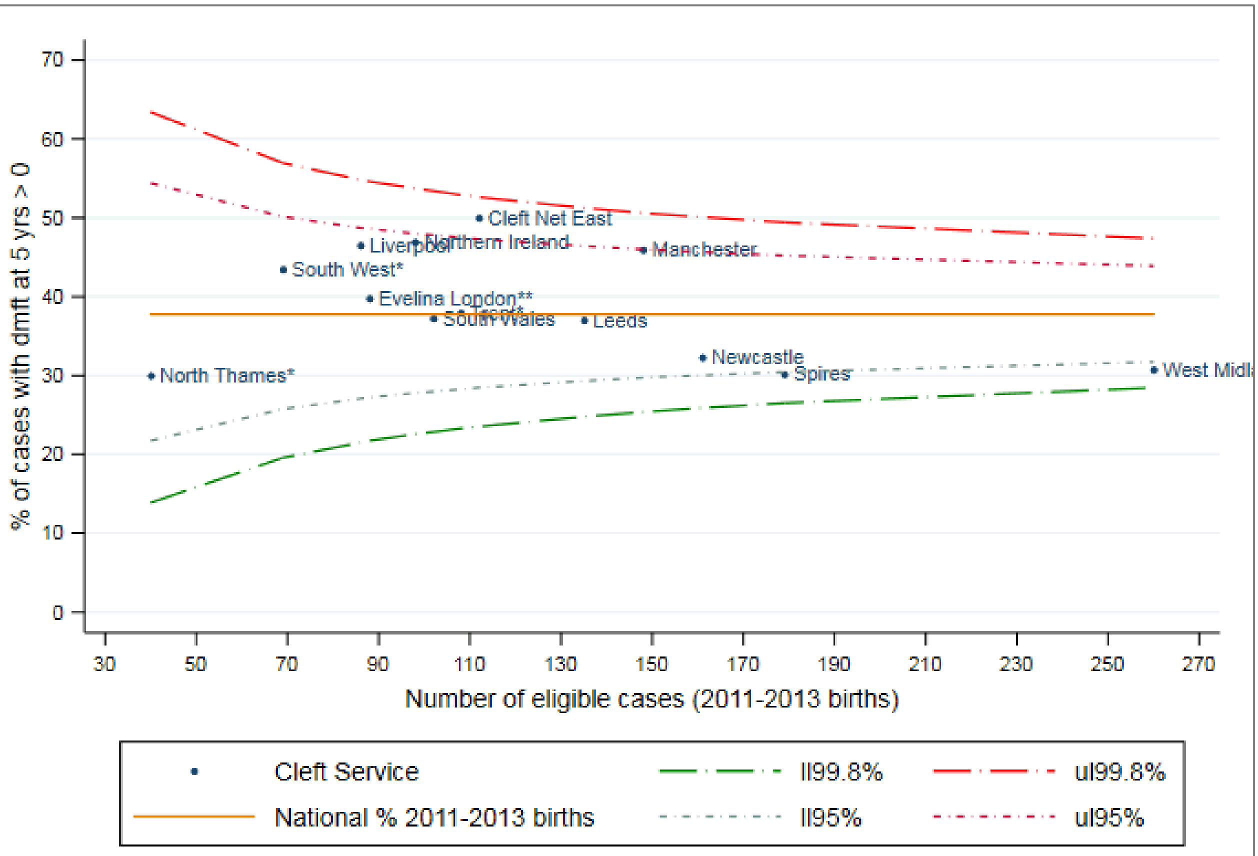
Submitting this indicator of dmft >0 (#7) to the outlier process, for the first time, did not lead to the identification of any outliers for this outcome indicator. **Appendix 9** shows the breakdown of cases with reported dmft >0 at 5 years of age, according to cleft service.

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[51].

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

Figure 4.6. Funnel plot showing the percentage of CRANE-consented 5-year olds, born 2011-2013, with dmft >0, according to cleft service.



Note 1: Data from Evelina London was not used to create funnel plot due to poor consent verification rate.
Note 2: Data from Trent, N Thames, S West & Evelina were not used to create funnel plot due to poor data completion rates.

Note: Registered in CRANE by 30 June 2021. Funnel plot centred on the adjusted national percentage (37.8%) of children (born 2011-2013) with dmft >0 reported. See **Appendix 9** for the raw data (and exclusions) used to create this graph.

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[50] The overall mean was 38.0% and adjusted mean was 37.8% (excluding cleft services with consent verification and data completeness outlier status) were 86.9%.

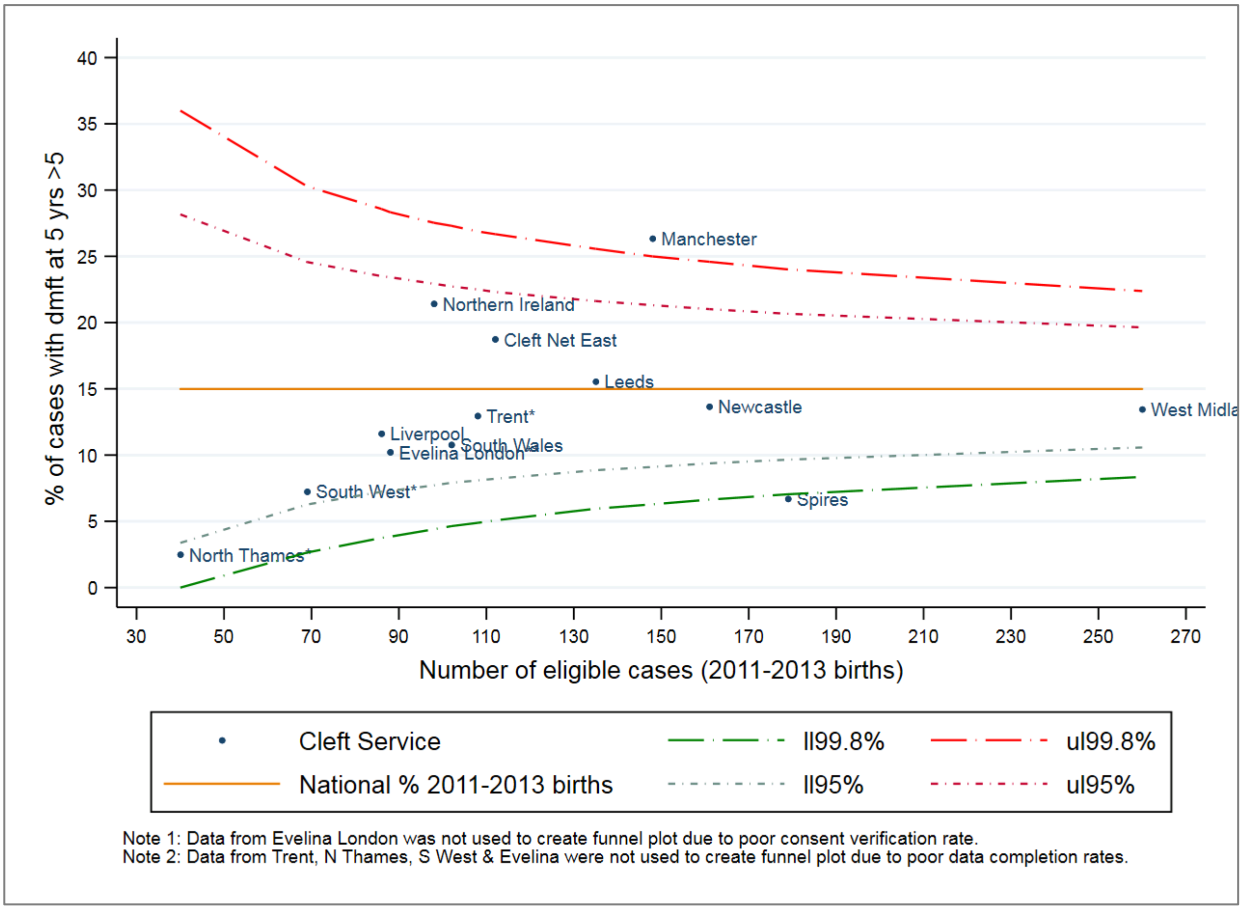
[51] (1) Al-Dajani M. Comparison of dental caries prevalence in patients with cleft lip and/or palate and their sibling controls. The Cleft Palate-Craniofacial Journal, 2009. 46(5): p. 529-531. (2) Britton, KF and Welbury, RR, Dental caries prevalence in children with cleft lip/palate aged between 6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

dmft >5 scores

Figure 4.7 shows the percentage of 5-year olds with more than five (>5) dmft, according to the number of children with valid dmft scores at each cleft service. The funnel plot is centred on the adjusted national average of 15%[52] (range 3%-26%).

Submitting this indicator of dmft >5 (#8) to the outlier process, for the first time, led to the identification of outliers for this outcome indicator.

Figure 4.7. Funnel plot showing the percentage of CRANE-consented 5-year olds, born 2011-2013, with dmft >5, according to cleft service.



Note 1: Data from Evelina London was not used to create funnel plot due to poor consent verification rate.
Note 2: Data from Trent, N Thames, S West & Evelina were not used to create funnel plot due to poor data completion rates.

Note: Registered in CRANE by 30 June 2021. Funnel plot centred on the adjusted national percentage (15%) of children (born 2011-2013) with dmft >5 reported. See **Appendix 9** for the raw data (and exclusions) used to create this graph.

The funnel plot shows that the Manchester cleft service are negative outliers for the percentage of cases with dmft >5 reported. This because they fell above the upper 99.8% control limit. Completing this process of identifying outliers for this outcome indicator (#8), for the first time, has also allowed the identification of the Spires cleft service as a positive outlier. See Box 5 and 6 (below) for comments from cleft services on their outlier status; as well as Box 7 for a statement from the Paediatric Dentistry CEN on the interpretation of dmft outcomes.

Appendix 9 shows the breakdown of cases with reported dmft >5 at 5 years of age, according to cleft service.

Anticipating challenges to collecting dmft scores, due to the COVID-19 pandemic: **Appendix 9** also shows the breakdown of cases meeting each of the above two outcome indicators (#7 and #8), according to cleft service, for 2011-13 as well as 2014 births. Data for 2014 births will be reported on in the CRANE 2022 Annual Report.

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[52] The overall mean was 13.9% and adjusted mean was 15.0% (excluding cleft services with consent verification and data completeness outlier status) were 86.9%.

Box 5. Summarised responses to being negative outliers for ‘dmft >5 indicating experience of extensive dental decay’. **Full responses published in the “2021 Annual Report: Responses to outlier process” document.**

“We regularly review the dental health data within the unit at Manchester and with the Paediatric Dental CEN. We always compare our data with the regional dmft data from the NHS Dental Epidemiology Programme review of 5 yr olds. Their data from 2017 shows that Greater Manchester has the highest dmft in the country… As you have highlighted we are a positive outlier for completeness of data collection for dental health and we had a consultant in Paediatric dentistry start in 2016 so would expect a greater rate of detection of decay… We have been auditing our 3 yr olds’ dmft and picking up high caries risk patients and providing dental treatment, preventive advice and reviewing them within the cleft unit more frequently (every 3-6 months).”

Manchester cleft service, November 2021.

Box 6. Summarised responses to being positive outliers for ‘dmft >5 indicating experience of extensive dental decay’. **Full responses published in the “2021 Annual Report: Responses to outlier process” document**

“We also have a focus on dental health information and support that begins with our clinical nurse specialists at birth and continues through all early years and children’s clinics and contacts with the multidisciplinary team, including liaison with GDP and Special Care dentistry colleagues. On the whole we have excellent primary care salaried dental device support across the region…We are completely open to discussions with other centres about our results and our service model.”

Spires cleft service, November 2021.

Box 7. Statement from Paediatric Dental CEN.

As not all units have a Cleft Calibrated Paediatric Dentist differences in dmft outcomes between units should be interpreted with caution where not all data which has been submitted fulfils the specific criteria outlined; namely that it is collected by a Specialist in Paediatric Dentistry who has successfully completed appropriate calibration within the preceding 24 months. Differences in the collection of data as well as completeness of data collected should therefore be borne in mind when comparing units against each other.

Joanna May, Lucy Burbridge, and Jacqueline Smallridge, December 2021.

4.2.3. Dental health measures of intervention by cleft service

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[53]. 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[54]. 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.

Table 4.1 shows that the percentage of children achieving each dental health measure of intervention at 5 years (as **defined above**) varies by cleft service. Overall, the average Treatment Index was 76%, and the average Care Index was 70% for children with a cleft assessed at 5 years of age.

High mean index scores indicate that children have high levels of treated dental disease (as expressed by the Treatment Index) and receive the appropriate care at the earliest possible stage (as expressed by the Care Index). A Kruskall-Wallis test was used to compare the mean rank of Treatment Index and Care Index scores between cleft services; with no statistically significant differences identified between services.

Table 4.1. Average dental Treatment Index and average Care Index, according to cleft service for CRANE-registered consented children born 2011 - 2013.

Cleft service	Eligible cases with dmft reported	Mean Treatment Index		Mean Care Index	
	N	N	(%)	N	(%)
Newcastle	161	160	78.4%	161	70.8%
Leeds	135	132	78.2%	135	73.0%
Liverpool	86	83	70.4%	84	62.1%
Manchester	148	147	71.3%	147	63.1%
Trent	108	108	75.6%	108	70.0%
West Midlands	260	259	80.0%	259	75.1%
Cleft Net East	112	111	76.8%	111	69.9%
North Thames	40	40	75.8%	40	70.5%
Spires	179	177	77.4%	176	74.7%
South Wales	102	102	80.9%	102	70.5%
South West	69	68	71.5%	67	67.5%
Evelina London	88	87	73.4%	87	69.6%
Northern Ireland	98	98	74.6%	98	64.0%
Total	1,586	1,572	76.4%	1,575	70.2%

Note: Registered in CRANE by 30 June 2021. Exclusions (not mutually exclusive): Children who died before the age of 5 years and those with submucous cleft palates.

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

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

Anticipating challenges to providing and recording information on dental health interventions – as reflected by treatment and care indices – due to the COVID-19 pandemic: **Appendix 9** shows the information in Table above, according to cleft service, for 2014 births. Data for 2014 births will be reported on in the CRANE 2022 Annual Report.

The CRANE 2019 Annual Report presented findings on decayed missing and filled teeth (DMFT) at 10 years for 2007 and 2008 births. Exploration of the data in 2019 revealed a low number of data returns (14% in 2019). This has improved to 18% data completeness when explored in 2021 (for 2007, 2018 and 2009 births). 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 services 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 2022, 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	The CRANE Database (consented cases only)
	Birth years	Three years: 2011 to 2013. Plus summary information on 2014 births[55]
	Denominator	334 5-year-old children with complete Unilateral Cleft Lip and Palate (UCLP) and recorded 5 year old Index scores
	Numerator	The number of children classified as having 'good', 'fair' or 'poor' 5 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 palates56 • Children without a recorded 5 year old Index score • Cleft services with consent verification outlier status (excluded from data completeness checks)
	Data completeness	• 65.9% of 507 eligible consented children had recorded 5 year old Index scores. • 14.2% had a reason the facial growth scores were not collected. • 19.9% were missing data or a reason for not collecting data.
	Countries	England, Wales and Northern Ireland
	Indicator	#9[57]
Benchmarks	• 100% of eligible children with a complete UCLP should have a recorded 5 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[58].	
What did we find?	• 37.7% of children had scores reflecting 'good' dental arch relationships at 5 years old. • 38.3% of children had scores reflecting 'fair' dental arch relationships at 5 years old. • 24% of children had scores reflecting 'poor' dental arch relationships at 5 years old.	
Recommendations	• Cleft services 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 5 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 5 year old Index in UK populations.	

4.3.1. Data completeness

Participation in a national clinical audit, such as CRANE, means that all cleft services are asked to record 5 year old Index scores – used to evaluate the effects of primary surgery on the facial growth of children with complete UCLP before the use of any other interventions – for all consented 5-year-old children . If a 5 year old Index score is not available, cleft services are asked to report a reason for this[59] (e.g. Patient transferred out of area or

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[55] Affected by the COVID-19 pandemic and the lockdown that ensued.

[56] Submucous cleft palate patients excluded from all 5 year outcomes as all/most teams do not audit these patients. Including cases with LAHSAL code identified as not possible, and likely submucous, when reviewed by cleft clinicians.

[57] CRANE core indicators are detailed in **Appendix 3**.

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

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

patient did not attend appointment, etc). Appendix 10 shows the breakdown of facial growth reporting for each cleft service for the 2011-13 birth cohort.

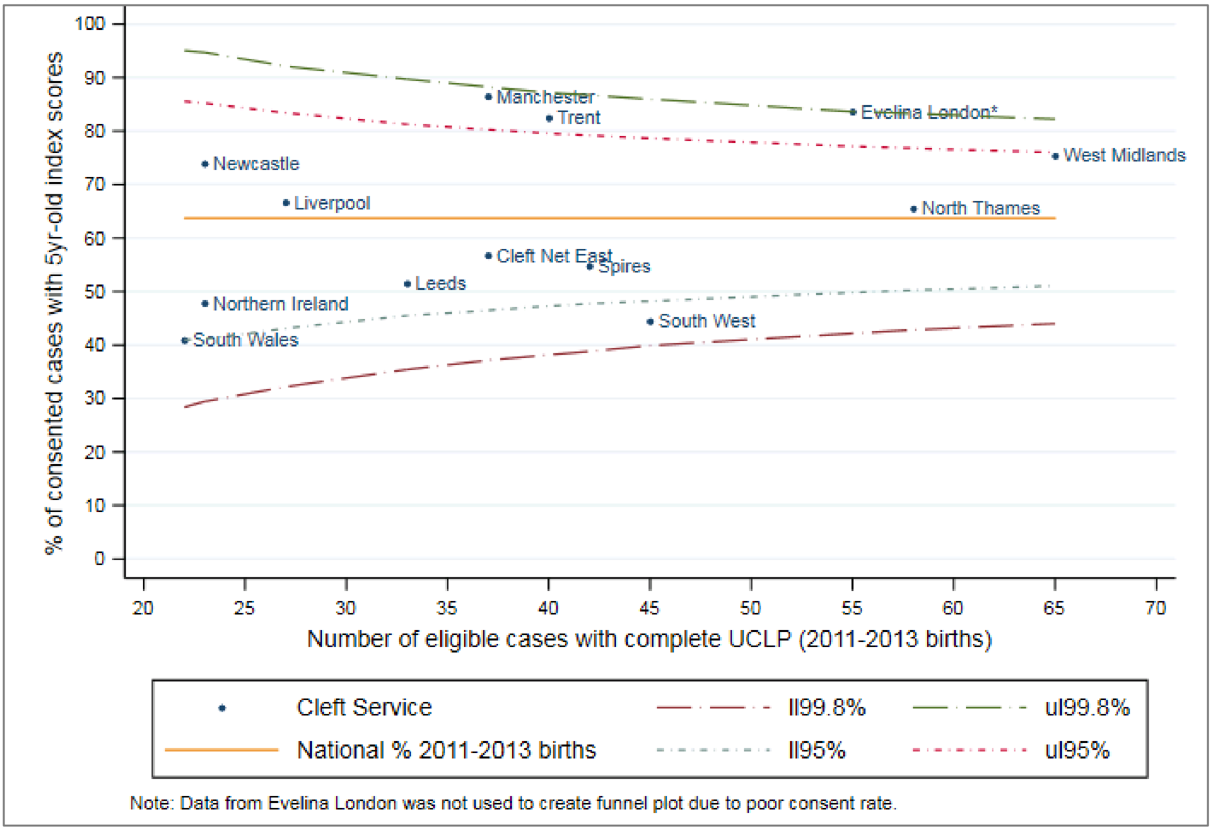
Of the 507 eligible 5-year old children, born between 2011 and 2013, 66% had a 5 year old Index score recorded in CRANE (40.9% -86.5% range between services). 14.2% of children had a documented reason why the outcome was not collected, and 19.9% of children were missing data or a documented reason.

Reasons for not collecting 5-year old facial growth outcome data for 2011-13 births

Appendix 7 shows the breakdown of reasons reported for not collecting 5-year old facial growth outcome data for 2011-13 births. The most common reason selected was 'Patient did not attend' (41.7%).

Figure 4.8 shows the variability in the reporting of facial growth data according to the number of eligible cases within each cleft service. The funnel plot is centred on the adjusted national average of 64%[60]. The wide range in reporting may reflect the difficulty that some cleft services had in performing facial growth assessments during the COVID-19 pandemic or lack of staff members within services.

Figure 4.8. Funnel plot showing the percentage of CRANE-consented 5-year olds, born 2011-2013, who had facial growth data submitted to CRANE, according to cleft service.



Note: Data from Evelina London was not used to create funnel plot due to poor consent rate.

Note: Registered in CRANE by 30 June 2021. Funnel plot centred on the adjusted national percentage (63.7%) of children (born 2011-2013) with facial growth outcomes at the age of five years reported. See **Appendix 10** for the raw data (and exclusions) used to create this funnel plot.

The funnel plot shows that there are no outliers for the percentage of eligible cases with facial growth data items reported. This means that no cleft service is above or below the 99.8% control limit that would warrant exclusion from overall average calculations presented in the subsequent outcome funnels.

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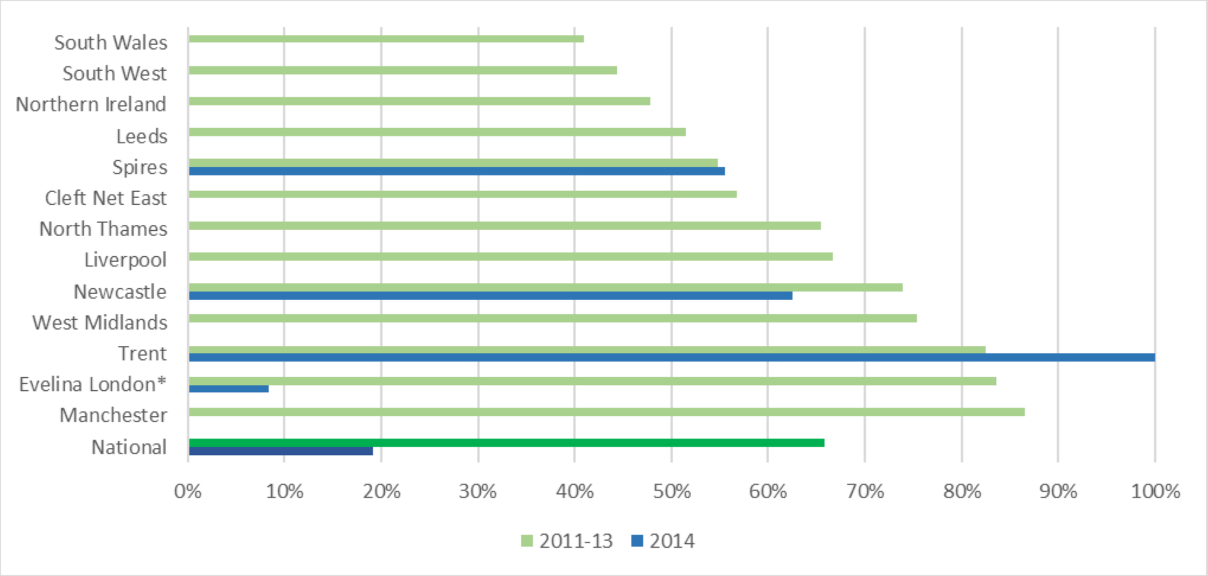
[60] The overall mean was 65.9% and adjusted mean was 63.7% (excluding cleft services with consent verification outlier status).

Anticipating challenges to data completeness due to the COVID-19 pandemic, Figure 4.9 summarises data completeness for eligible children born in 2014 (reporting 5 year old Index scores), which will be reported on in the CRANE 2022 Annual Report. This is compared with 2011-2013 data to provide pre-pandemic context[61]. **Appendix 10** shows the breakdown of facial growth reporting for each cleft service, for 2011-13 as well as 2014 births.

For the 2014 birth cohort no external validation of 5 year old Index scoring was possible, due to the COVID 19 pandemic, and therefore only internally validated scores were recorded in the CRANE Database.

Figure 4.9. Percentage of CRANE-consented 5-year olds, born 2014, who had facial growth data submitted to CRANE, according to cleft service – by comparison with children born 20111-13.



Note: Registered in CRANE by 30 June 2021. See **Appendix 10** for the raw data (and exclusions) used to create this graph.

Overall, of the 141 eligible 5-year old children born in 2014, 19% had a 5 year old Index score recorded in CRANE (0% - 100% range between services). 11% of children had a documented reason why the outcome was not collected, and 70% of children were missing data or a documented reason.

Reasons for not collecting 5-year old facial growth outcome data for 2014 births

Appendix 7 shows the breakdown of reasons reported for not collecting 5-year old facial growth outcome data for 2014 births; the most common reason selected was 'Other' (50%). Comments provided to specify these 'other' reasons, in the main, described clinics cancelled / data not collected due to the COVID-19 pandemic.

4.3.2. 5 year old index scores

Dental models of 5-year old children with a complete UCLP were assessed using the 5 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[62]. Dental arch relationships at 5 years have been thought to

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[61] A child born 31 December 2013, should have been audited for 5-year old outcomes before the age of 6. I.e. By the 31 December 2019 (pre-COVID-19 pandemic).

[62] 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.

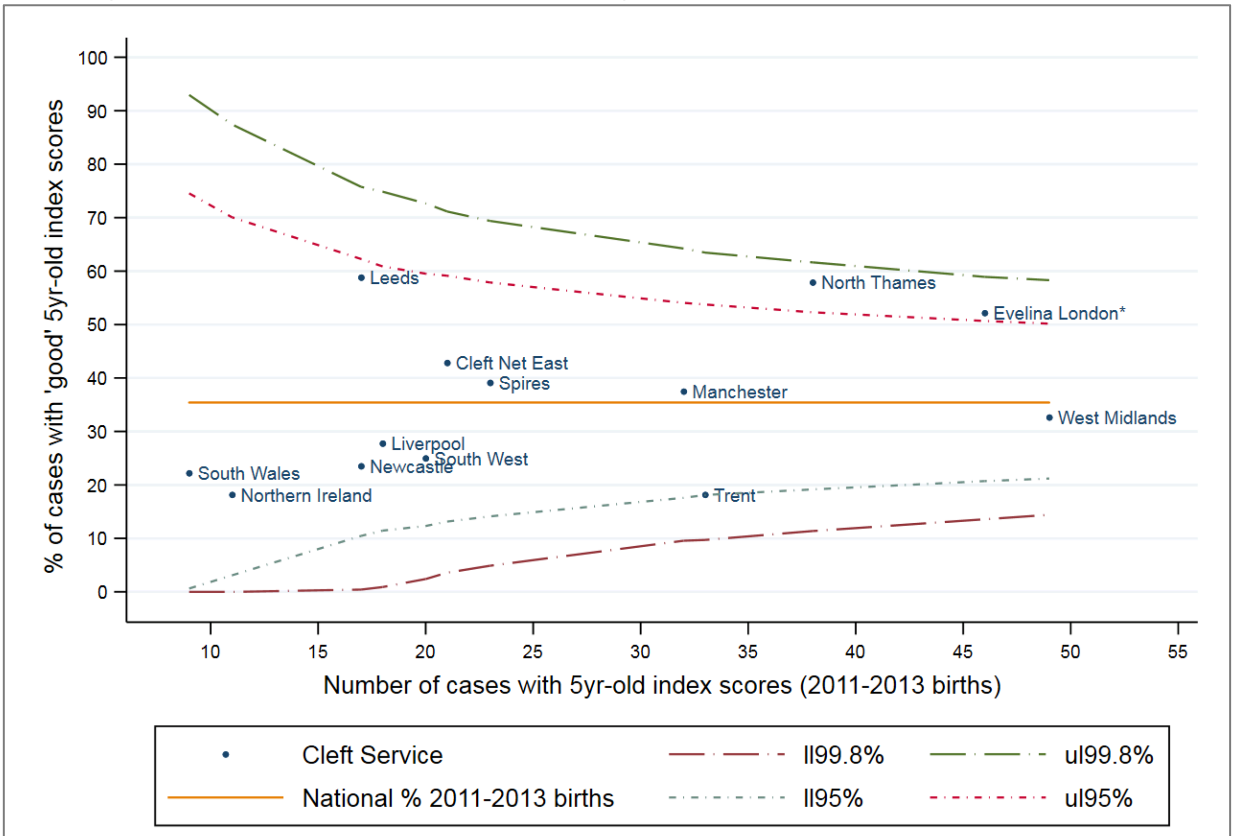
predict treatment outcome in terms of facial growth on a population basis rather than at the individual child level[63]. The 5 year old Index has, therefore, been used to compare treatment outcomes between cleft services and surgeons. Patients scoring ‘1’ and ‘2’ on the index are considered to have good outcomes, while those scoring ‘4’ and ‘5’ are thought to have poor outcomes in terms of facial growth.

Among the 334 children with reported 5 year old Index scores, 93% (312) were externally validated. Where an externally validated score was unavailable, internal scores were included in the analyses. Overall, 38% had 5 year old Index scores of ‘1’ or ’2,’ reflecting ‘good’ dental arch relationships, 38% had a ‘fair’ score, while 24% of children had scores of ‘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 of age in 1998[64].

4.3.2. Good 5 year old index scores, by cleft service

Figure 4.10 shows the percentage of 5-year olds with 5 year old Index scores reflecting ‘good’ dental arch relationships, according to the number of eligible children with facial growth scores at 5 years of age at each cleft service. The funnel plot is centred on the adjusted national average of 35%[65] (range 18%-59%). All services fall within the 99.8% control limits.

Figure 4.10. Funnel plot showing the percentage of CRANE-consented 5-year olds, born 2011-2013, who had good facial growth scores submitted to CRANE, according to cleft service.



Note: Data from Evelina London was not used to create funnel plot due to poor consent rate.

Note: Registered in CRANE by 30 June 2021. Exclusions (not mutually exclusive): Children who died before the age of 5 years, with submucous cleft palates, and those born with an incomplete UCLP. Funnel plot centred on the adjusted national percentage (35.4%) of children (born 2011-2013) with ‘good’ facial growth outcomes reported.

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[63] 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.

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

[65] The overall mean was 37.7% and adjusted mean was 35.4% (excluding cleft services with consent verification and data completeness outlier status) were 86.9%.

Submitting this indicator of 'good' dental arch relationships (#9) as measured by the 5 year old Index to the outlier process, for the first time, did not lead to the identification of any outliers for this outcome indicator. **Appendix 10** shows the breakdown of cases with Good, Fair and Poor scores at 5 years of age, according to cleft service.

The distribution of Good, Fair and Poor scores continues to be variable between cleft services; as shown graphically in **Appendix 10**. This highlights an area that would benefit from discussion and further research within the UK to actively investigate the reasons for this. Comparison with contemporaneous cohorts from elsewhere in Europe may provide insight as to whether further improvement in facial growth is possible.

Anticipating challenges to conducting facial growth scoring before the age of 6, due to the COVID-19 pandemic: **Appendix 10** shows the breakdown of cases meeting each facial growth process or outcome standard, according to cleft service, for 2011-13 as well as 2014 births. Data for 2014 births will be reported on in the CRANE 2022 Annual Report.

Last year we highlighted that Swedish facial growth data had shown that 'fair' and 'poor' 5 year old Index scores at 5 years of age had limited predictive value in terms of predicting the long-term (mid to late teenage years) value of dental arch relationships[66]. As such, 'fair' and 'poor' scores should be interpreted with caution5 year old5 years of age5 year old. This is why the CRANE 2021 Annual Report recommended that further research be undertaken to see whether the results from the Swedish cohort are replicated in the UK and/or other countries. In order to do this, the Orthodontic Clinical Excellence Network (CEN) are engaging in discussions to perform a pilot study in the UK, which will begin in due course.

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[66] 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

4.4. Speech

Cohort summary	Data source	The CRANE Database (consented cases only)
	Birth years	Three years: 2011 to 2013. Plus summary information on 2014 births[67]
	Denominator	1,317 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 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[68] • Children with a diagnosed syndrome[69] entered onto the CRANE database • Children without all 16 CAPS-A scores • Cleft services with consent verification outlier status (excluded from outcome data completeness checks)
	Outcome data completeness	• 69.5% of 1,896 eligible children had all 16 CAPS-A speech parameters reported. • 2.1% had some but not all 16 CAPS-A speech parameters reported. • 22.5% had a reason the speech outcomes were not collected. • 5.9% were missing data or a reason for not collecting data.
	Countries	England, Wales and Northern Ireland
	Indicators	#10, #11 & 12[70]
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)[71]. • Speech without difficulties resulting from existing or previous structural anomalies. • Speech without significant cleft-related articulation difficulties.	
What did we find?	• 60.1% achieved speech within the normal range. • 71.0% had speech without difficulties resulting from existing or previous structural anomalies. • 67.5% had speech without cleft-related articulation difficulties. • 17.7% of children had secondary surgery for speech purposes before the age of 5 years.	
Recommendations	• All children with a cleft affecting the palate should have their speech assessed and reported to CRANE. • Where possible, cleft services should investigate possibilities of remote methods for consensus listening. • All cleft services should work together to explore possible reasons for variation in speech outcomes. • The research community should undertake to develop risk stratification models for analysing speech outcomes among children with a cleft.	

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[67] Affected by the COVID-19 pandemic and the lockdown that ensued.

[68] Submucous cleft palate patients excluded from all 5 year outcomes as all/most teams do not audit these patients. Including cases with LAHSAL code identified as not possible, and likely submucous, when reviewed by cleft clinicians.

[69] Cases flagged as syndromic are excluded, with the exception of children with a recoded (named) congenital malformation of the circulatory system, or congenital malformation of the nervous system (e.g. micrognathia / spina bifida). This is a (new) refinement to the methodological approach to reporting speech outcomes.

[70] CRANE core indicators are detailed in **Appendix 3**.

[71] 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 Outcome data completeness

All cleft services are requested to report to CRANE all 16 CAPS-A parameters for each consented 5-year old child with a cleft affecting the palate. If speech outcomes are not available, cleft services are asked to report a reason for this[72] (e.g. Patient transferred out of area or patient did not attend appointment). Appendix 11 shows the breakdown of speech reporting for each cleft service, for the 2011-13 birth cohort.

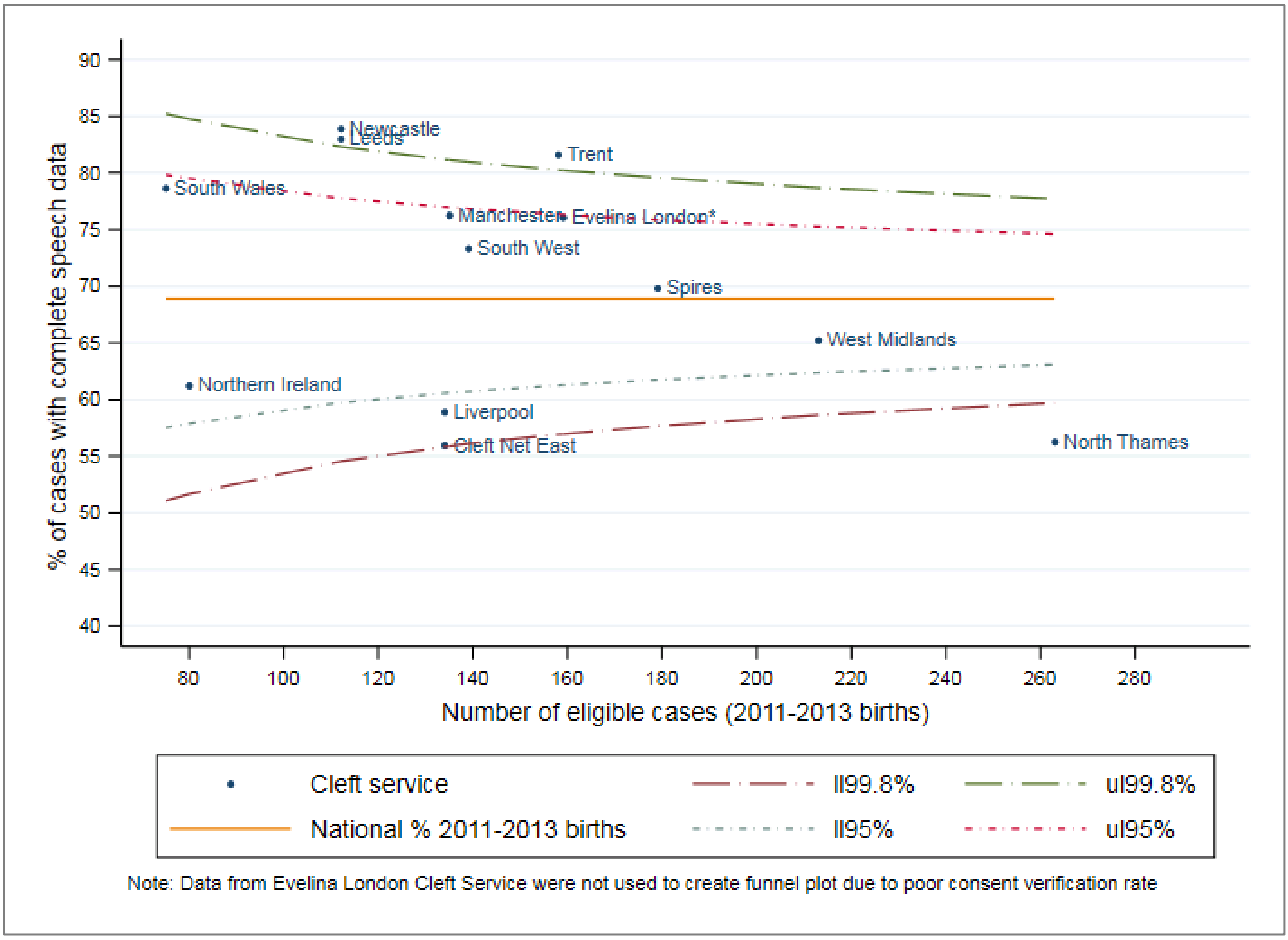
Of the 1,896 eligible cases, 70% (56%-84% range between services) had all 16 CAPS-A parameters reported. A further 2% of cases had some but not all 16 CAPS-A parameters reported. 23% of cases had a reason why the outcome was not reported, and 6% were missing data or a reason.

Reasons for not collecting 5-year old speech outcome data for 2011-13 births

Appendix 7 shows the breakdown of reasons reported for not collecting 5-year old speech outcome data for 2011-13 births; the most common reason selected was 'Patient DNA' (29%).

Figure 4.11 shows the variability in the reporting of all 16 CAPS-A parameters at 5 years of age, according to the number of eligible cases in each cleft service. The funnel plot is centred on the adjusted percentage meeting standard of 69%[73].

Figure 4.11. Funnel plot showing the percentage of CRANE-consented 5-year olds, born 2011-2013, with speech outcomes reported, according to cleft service.



Note: Data from Evelina London Cleft Service were not used to create funnel plot due to poor consent verification rate

Note: Registered in CRANE by 30 June 2021. Funnel plot centred on the adjusted national percentage (68.9%) of children (born 2011-2013) with speech outcomes reported. See Appendix 11 for the raw data (and exclusions) used to create this funnel plot.

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

[73] The unadjusted national average was 69.5% and adjusted percentage with speech outcome data was 68.9% (excluding cleft services with consent verification outlier status).

The wide variability in the reporting of these 5 year old outcomes highlights challenges faced by cleft services in recording the speech data for 5 year old patients.

The funnel plot shows that North Thames is a negative outlier for the percentage of eligible cases with all 16 CAPS-A parameters reported. This is because they fell below the lower 99.8% control limit, and were therefore excluded from the calculation of the overall average presented in the subsequent outcome funnels. Results from this region should therefore be interpreted with caution. Completing this process of identifying outliers, for the first time, has also allowed the identification of positive outliers such as Newcastle, Leeds and Trent cleft services. See Box 8 and 9 (below) for comments from cleft services on their outlier status.

Box 8. Summarised response to being a negative outlier for 'speech – outcome data completeness at 5 years'. **Full responses published in the "2021 Annual Report: Responses to outlier process" document.**

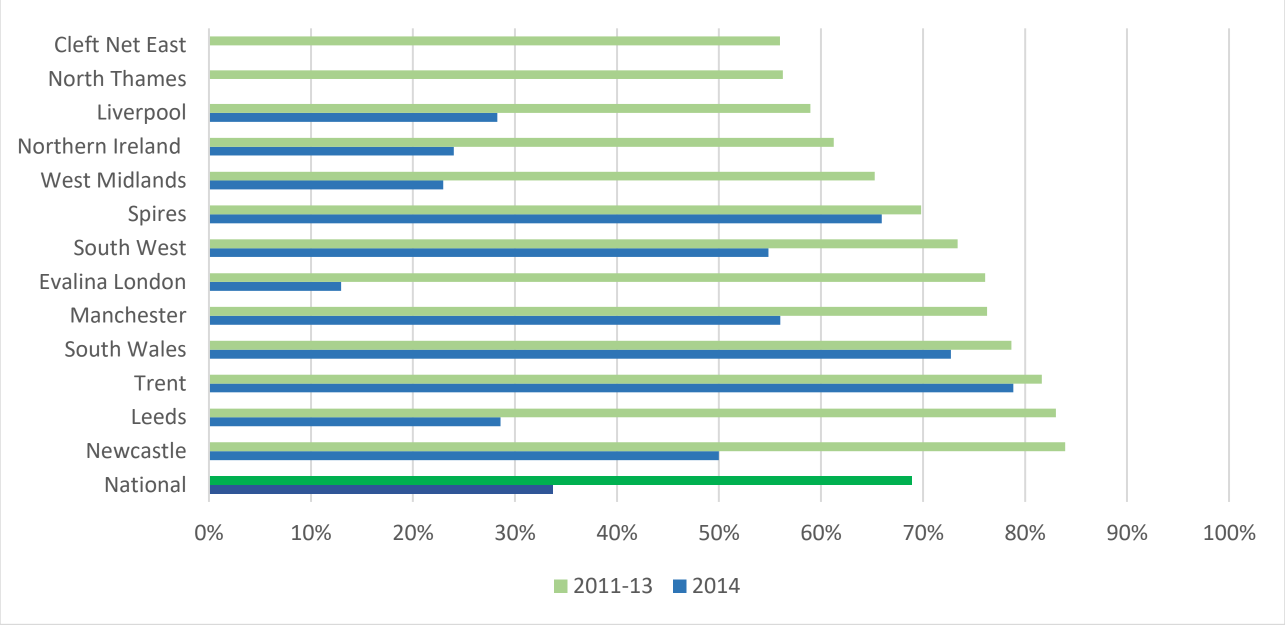
"The cleft data coordinator is relatively new to the role and unfortunately a short time after her appointment her direct supervisor left for a period maternity leave. A successor to this supervisory role has not yet been appointed. In addition, the cleft service has been without a substantive service manager for the last 3 months. Unfortunately, this person helps monitor and coordinate data collection exercises and their absence has made this process more complicated…The main issue with missing data seems to be technicalities around the recording and recovery of speech samples, rather than patients not being recorded at their 5-year audit appointment. This was at a time when we were moving from video tapes to digital recording. We feel we have now eliminated this problem. In 2011 and 2012 we had a large number of DNA/cancellations, but this improved for the 2013 cohort." North Thames cleft service, October 2021.

Box 9. Summarised responses to being positive outliers for 'speech – outcome data completeness at 5 years'. **Full responses published in the "2021 Annual Report: Responses to outlier process" document.**

"This clearly represents hard work by every member of our team and in particular the diligence and thoroughness of our data co-ordinator. It is supported by us having a fully staffed and very stable team…5 year data collection is the careful scheduling of 5 year audit clinics, with our audit coordinator identifying all patients for each cohort early every year, and then building specialist audit clinics around them over the 12 month period." Newcastle cleft service, October 2021.
"Our Cleft Team includes a dedicated Cleft Audit Coordinator…. Who is dedicated to her role and is assiduous in terms of ensuring maximal data completeness and CRANE consent verification wherever possible….As a Cleft Service, we always prioritise the importance of Audit Clinics and plan for these well in advance … to ensure that patients attend ….within the patient age time frame, such as before age 6" Leeds cleft service, October 2021.
"In Trent we have a rolling system of collecting and analysing speech audits. The specialist SLTs in the team are proactive in collecting speech audit recordings for their area and as Lead SLT I am proactive in identifying any cases who have not been seen and chasing this. We hold monthly listening days throughout the year with 2 CAPSA trained listeners ... Our stable listening group and rolling system of listening also helped our performance." Trent cleft service, October 2021.

Anticipating challenges to outcome data completeness due to the COVID-19 pandemic, Figure 4.12 summarises outcome data completeness for eligible children born in 2014 (reporting of all 16 CAPS-A speech parameters), which will be reported on in the CRANE 2022 Annual Report. This is compared with 2011-2013 data to provide pre-pandemic context[74]. **Appendix 11** shows the breakdown of speech reporting for each cleft service, for 2011-13 as well as 2014 births.

Figure 4.12. Percentage of CRANE-consented 5-year olds, born 2014, who had all 16 CAPS-A speech parameters submitted to CRANE, according to cleft service – by comparison with children born 2011-13.



Note: Registered in CRANE by 30 June 2021. See **Appendix 11** for the raw data (and exclusions) used to create this graph.

Overall, of the 587 eligible 5-year old children born in 2014, 33.7% had all 16 CAPS-A speech parameters reported (0%-78.8% range between services). 6 out of 13 cleft services were able to account for 50% or more of eligible cases.

Reasons for not collecting 5-year old speech outcome data for 2014 births

Appendix 7 shows the breakdown of reasons reported for not collecting 5-year old speech outcome data for 2014 births; the most common reason selected was 'Other' (76%). Comments provided to specify these 'Other' reasons, in the main, described clinics cancelled / data not collected due to the COVID-19 pandemic.

4.4.2. Speech outcomes

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

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

The distribution of scores across the individual 16 CAPS-A speech parameters for those born 2011-2013 are presented in **Appendix 11**.

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[74] A child born 31 December 2013, should have been audited for 5-year old outcomes before the age of 6. I.e. By the 31 December 2019 (pre-COVID-19 pandemic).

Resonance and Nasal Airflow

In terms of resonance, 6% of children had moderate or severe hypernasality i.e. nasal sounding speech[75]. This is indicative of velopharyngeal dysfunction (VPD), which is when the palate is unable to close off the nasal airway during speech. In addition, results of the Cleft Speech Characteristics show that 3% of children had 'weak and or nasalised consonants' and 2% 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.

Overall, 84% (1,108/1,317) of children had ratings indicating that no structural problems existed in relation to resonance and nasal airflow by their 5 year assessment[76].In order to achieve good speech, 234 out of 1,317 children (18%) with reported surgical data[77] had undergone secondary surgery for speech purposes before the age of 5 years. Of these, 161 (69%) had resonance and nasal airflow ratings that indicated there were no structural problems that existed in relation to these parameters, suggesting that the fact that these children had undergone secondary surgery had resolved these speech issues in time for starting school.

Cleft Speech Characteristics (CSCs)

Out of the 1,317 children (born 2011-13) with reported ratings for all 12 cleft speech characteristics, 65.5% had ratings indicating they did not exhibit any CSCs[78]. 'Palatalisation / Palatal' anterior oral CSCs were the most commonly occurring CSC, affecting 23.9% of children (11.1% with ratings of one or two consonants affected (light green ratings) and 12.8% with three or more consonants affected (amber ratings)). These CSCs can vary in severity and may affect speech acceptability more than speech intelligibility. The cleft speech characteristics which are more likely to affect speech intelligibility are the posterior, non-oral and passive CSCs (see **Appendix 11** 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 percentage of 5-year olds meeting each of the following three nationally agreed Speech Outcome Standards[79]:

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).

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[75] With a hypernasality score of '3' or '4' (red ratings).

[76] All green ratings of '0' or '1'.

[77] VP surgery/fistula repair data was reported for 2954/2980 (99.1%) eligible children.

[78] All green ratings of 'A' and in selected cases of 'B' – as per **Appendix 11**.

[79] 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.

4.4.3. Speech outcomes by cleft service

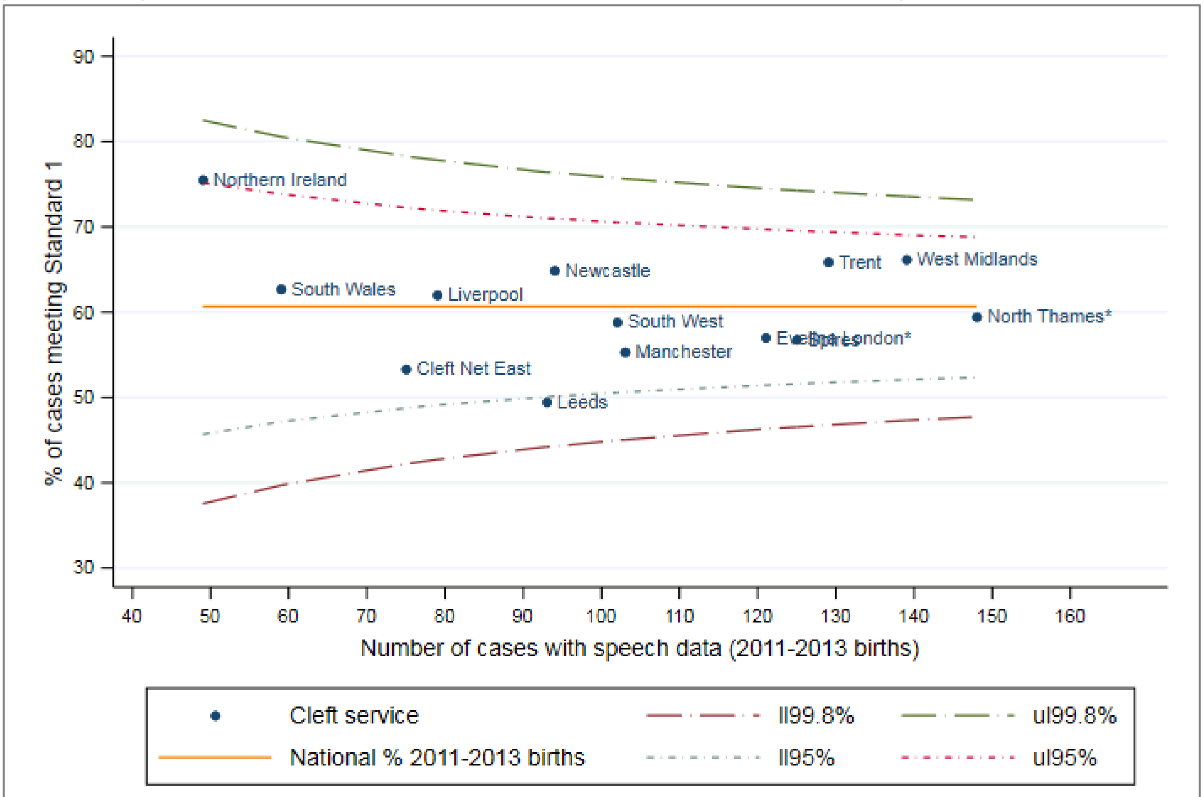
Normal speech

Out of the 1,317 children (born 2011-2013) with reported ratings for all 16 CAPS-A speech parameters, 60.1% of children across all units achieved the National Speech Outcome Standard 1 (this is an unadjusted national average). *They had normal (green) ratings across all 16 CAPS-A speech parameters.*

In line with the Outlier process being implemented for the first time in 2021, the adjusted percentage meeting the standard was 60.6% (excluding services with consent and outcome data quality rates not sufficient for inclusion), as presented in Figure 4.13. This means that both the unadjusted and adjusted results are in line with the National Speech Outcome Standard 1 benchmark of 61%, reported in the CRANE 2019 Annual Report.

The funnel plot[80] in Figure 4.13 (see **Appendix 11** for raw data) shows the percentage of children (born in 2011-2013) achieving *normal speech* according to the number of auditable children within each cleft service with ratings for all 16 CAPS-A speech parameters (more information on funnel plots can be found in the Glossary). The funnel plot is centred on the adjusted percentage meeting standard of 60.6% (range 49.5%-75.5%).

Figure 4.13. Funnel plot of 5-year olds (born 2011-2013) with ratings suggesting speech within the normal range, according to the number of children in each cleft service with CAPS-A ratings.



Note 1: Data from Evelina London Cleft Service were not used to create funnel plot due to poor consent verification rate.
Note 2. Data from North Thames Cleft Service were not used to create funnel plot due to poor data completion rates.

Note: Registered in CRANE by 30 June 2021. Funnel plot centred on the adjusted national percentage (60.6%) of children (born 2011-2013) with speech rated within the normal range. See Appendix 11 for the raw data (and exclusions) used to create this funnel plot.

The funnel plot shows that most (11/13) cleft services had a rate of normal speech that fell within the 95% control limits of the funnel plot (i.e. within one standard deviations of the adjusted percentage meeting standard). Northern Ireland had a normal speech rate of 75.5% while the corresponding figure in Leeds was 49.5%. The funnel plot shows that these units fall just outside of the upper and lower 95% control limits,

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

respectively. It should be noted that Northern Ireland had the fewest patients and that only 61% of their eligible cases had outcomes reported (see Figure 4.13). Leeds reported outcomes for 83% of eligible cases, which was one of the highest percentages out of all cleft services.

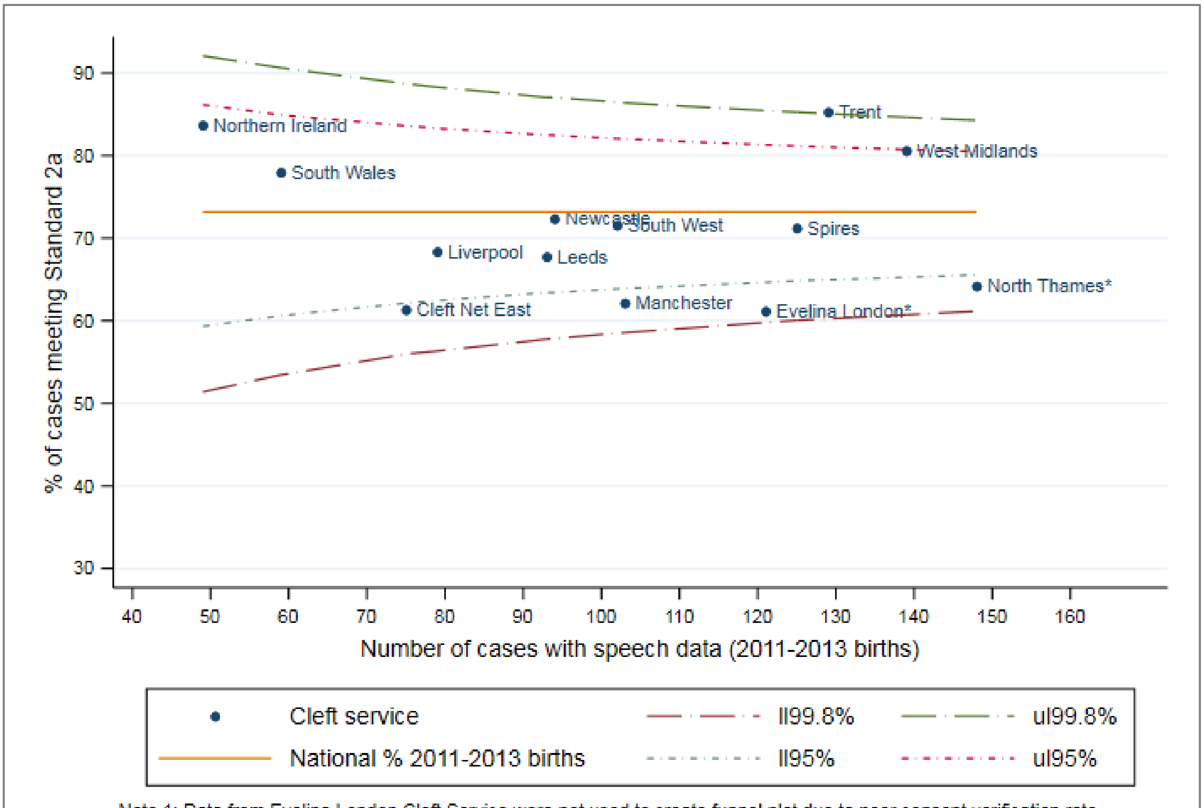
Absence of structurally-related speech difficulties

Out of the 1,317 children (born 2011-2013) with reported ratings for all 16 CAPS-A speech parameters, 71% of children across all services achieved the National Speech Outcome Standard 2a (this is an unadjusted national average). *They had no reported history of surgery for speech purposes and normal (green) ratings across the following six CAPS-A speech parameters: Hypernasal resonance, both nasal airflow parameters (audible nasal emission and nasal turbulence), and all three Passive CSCs.*

In line with the Outlier process being implemented for the first time in 2021, the adjusted percentage meeting the standard was 73.2% (excluding services with consent and outcome data quality rates not sufficient for inclusion), as presented in Figure 4.14. This means that both the unadjusted and adjusted results are in line with the National Speech Outcome Standard 2a benchmark of 71.5%, reported in the CRANE 2019 Annual Report.

Figure 4.14 (see **Appendix 11** for raw data) shows the percentage of 5-year olds with speech ratings that suggest they do not have structurally-related speech difficulties [81], according to the number of children at each service with CAPS-A ratings. The funnel plot is centred on the adjusted percentage meeting standard of 73% (range 61%-85%).

Figure 4.14. Funnel plot of 5-year olds (born 2011-2013) with ratings suggesting no structurally-related speech difficulties, according to the number of children at each service with CAPS-A ratings.



Note 1: Data from Evelina London Cleft Service were not used to create funnel plot due to poor consent verification rate.
Note 2. Data from North Thames Cleft Service were not used to create funnel plot due to poor data completion rates.

Note: Registered in CRANE by 30 June 2021. Funnel plot centred on the adjusted national percentage (73.2%) of children (born 2011-2013) without structurally-related speech difficulties. See Appendix 11 for the raw data (and exclusions) used to create this funnel plot.

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

The funnel plot shows that there is a lot of variability between services in the percentage of children without structurally-related speech difficulties. In particular, the 85.3% in Trent was on the upper 99.8% control limit, and the care and service provision offered by this service may be worth investigating for best practice recommendations.

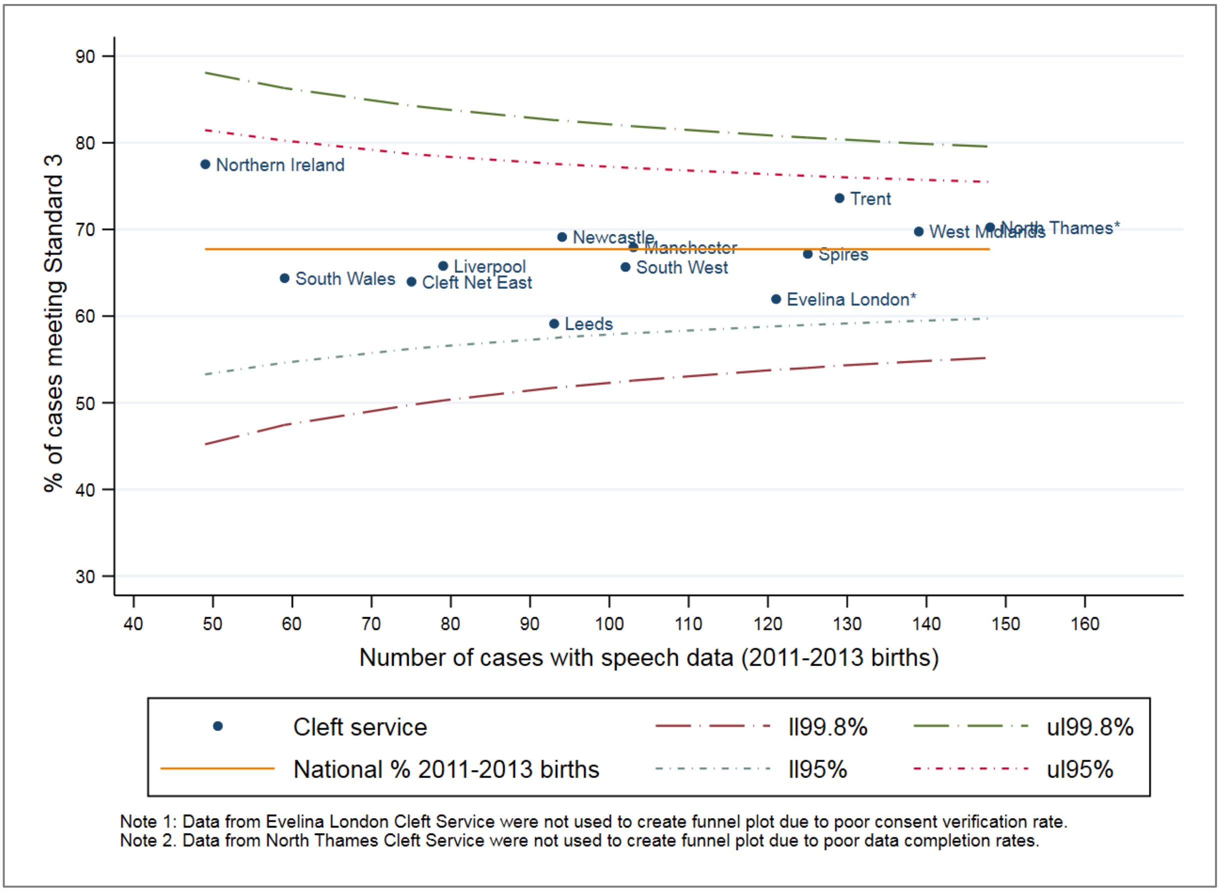
Absence of cleft-related articulation difficulties

Out of the 1,316 children (born 2011-2013) with reported ratings for all 16 CAPS-A speech parameters, 67.5% of children across all cleft services achieved the National Speech Outcome Standard 3(this is an unadjusted national average). *They had normal (green) ratings across the following 10 CSCs: All three Anterior Oral CSCs, both Posterior Oral CSCs, all four Non Oral CSCs, and gliding of fricatives (a Passive CSC).*

In line with the Outlier process being implemented for the first time in 2021, the adjusted percentage meeting the standard was 67.7% (excluding services with consent and outcome data quality rates not sufficient for inclusion), as presented in Figure 4.15. This means that both the unadjusted and adjusted results are in line with the National Speech Outcome Standard 3 benchmark of 68%, reported in the CRANE 2019 Annual Report.

Figure 4.15 (see Appendix 11 for raw data) shows the percentage of 5-year olds with speech ratings that suggest they do not have cleft-related articulation difficulties[82], according to the number of children within each service with CAPS-A ratings. The funnel plot is centred on the adjusted percentage meeting standard of 68% (range 59%-77%). All services fall within the 95% control limits.

Figure 4.15. Funnel plot of 5-year olds (born 2011-2013) with ratings suggesting no cleft-related articulation difficulties, according to the number of children at each service with CAPS-A ratings.



Note 1: Data from Evelina London Cleft Service were not used to create funnel plot due to poor consent verification rate.
Note 2. Data from North Thames Cleft Service were not used to create funnel plot due to poor data completion rates.

Note: Registered in CRANE by 30 June 2021. Funnel plot centred on the overall national percentage (67.7%) of children (born 2011-2013) without cleft-related articulation difficulties. See Appendix 11 for the raw data (and exclusions) used to create this funnel plot.

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

Despite the considerations around missing data and the lack of formal adjustment for potential confounding factors, as described above, presenting the data in Figures 4.13, 4.14 and 4.15 as funnel plots centred on adjusted national percentages is the most straightforward method (at this time[83]) of checking whether or not any services deviate significantly from the expected standards. However, the results should be interpreted with caution in those services where there is a high level of missing speech data. Nevertheless, it is recommended that differences in outcomes between services without a high volume of missing data should be explored further to determine whether there are patient characteristics or interventional factors that are associated with more favourable speech outcomes.

Box 10. Statement written by the Speech and Language Therapy Lead Therapists Group.

The Covid-19 pandemic has had a significant impact on each regional cleft service as they have had to respond to varying clinical demands on their service. This has led to wide variability in each of the services being able to collect, review and report speech data including missing data. This year, therefore, the data is not complete for some centres and is not comparable across centres. In addition, outlier data should be interpreted with caution. The Cleft SLT Lead group continue to drive for excellence across the UK and remain committed to the provision & submission of all available audit data.

Marie Pinkstone and Imogen Underwood, October 2021.

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

4.5. Psychology

Cohort summary	Data source	The CRANE Database (consented cases only)
	Birth years	Three years: 2011 to 2013. Plus summary information on 2014 births[84]
	Denominators	• 2,622 5-year old children eligible for psychology audit • 1,825 children with recorded date of face-to-face psychosocial screen (either at the 5-year audit or at an earlier screening)[85] • 1,639 children with recorded Tiers of Involvement Measure (TIM) scores • 1,472 children with recorded Strengths and Difficulties Questionnaire (SDQ) scores
	Numerators	• 1,423 5-year old children with all 8 psychology 5-year-audit data items • 1,765 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[86] • Children with a diagnosed syndrome entered onto the CRANE Database[87] • Children without a recorded date of first face-to-face psychosocial screening • Cleft services with consent verification outlier status (excluded from data completeness checks)
	Data completeness	• 54% of 2,622 eligible children had all psychology data items reported (complete data) • 11% had some but not all 8 scores reported (incomplete data) • 21% had a reason psychology data was not collected • 14% were missing data or a reason for not collecting data
	Countries	England, Wales and Northern Ireland
	Indicators	#13 & #14[88]
Benchmarks	• 100% of eligible children should have recorded psychology 5-year-audit data (or a recorded reason outcome not provided). • 100% of eligible children should be screened at least once before the age of 6 years. • 100% of children should be seen by a psychologist and have a TIM assessment at 5 years of age[38]. • SDQ population norms: 10% of children aged 5 to 10 years old have SDQ scores that are 'high' or 'very high'.	
What did we find?	• 97% of families (with recorded date of screen, either at the 5-year audit or at an earlier screening) were screened at least once before the target age of 6 years. • 93% were seen by a psychologist at age 5 and a psychosocial screen was completed or psychological input arranged (TIM tiers 1 to 4, also referred to as TIM tier 1a+). • 18% of children with a documented SDQ score had 'high' or 'very high' scores. These percentages are higher than the 10% SDQ population norms.	
Recommen-dation	Cleft services 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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[84] Affected by the COVID-19 pandemic and the lockdown that ensued.

[85] Dates of 'psychological screening at age five' were prioritised. Where unavailable, 'date of first face-to-face screening' used.

[86] Submucous cleft palate patients excluded from all 5 year outcomes as all/most teams do not audit these patients. Including cases with LAHSAL code identified as not possible, and likely submucous, when reviewed by cleft clinicians.

[87] Following guidance by the Psychology CEN, the 2022 report will implement an amendment to the methodology for reporting of psychology outcomes by CRANE; whereby syndromic cases will not be excluded from outcome reporting from 2022 onwards.

[88] CRANE core indicators are detailed in **Appendix 3**.

4.5.1. Data completeness

Participation in a national clinical audit, such as CRANE, means that all cleft services are asked to record psychology scores for all consented 5-year-old children with a cleft lip and/or palate. If psychology outcome data are not available, cleft services are asked to report a reason for this[89] (e.g. Patient transferred out of area or patient did not attend appointment, etc). Appendix 12 shows the breakdown of psychology reporting for each cleft service for the 2011-13 birth cohort.

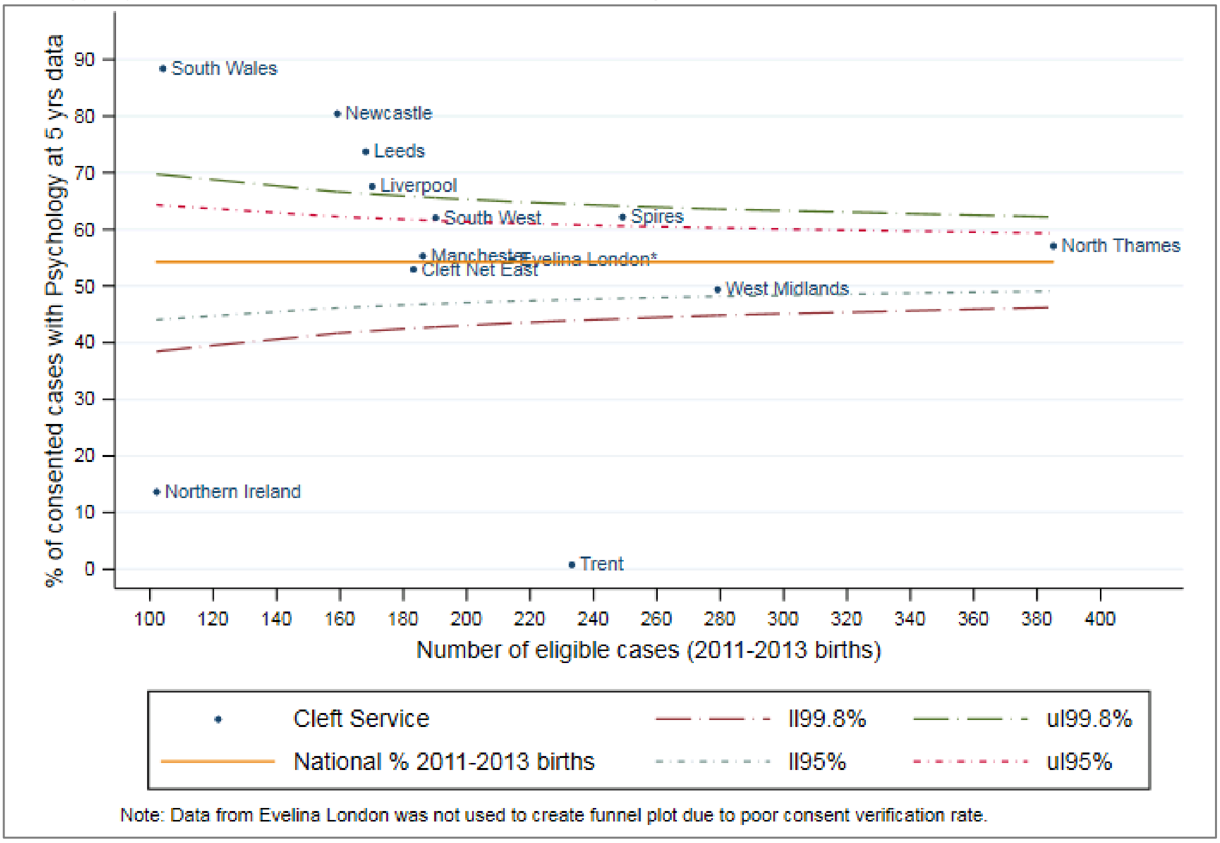
Of the 2,622 eligible 5-year old children born between 2011-13, 54% had all 8 psychology 5-year-audit[90] data items reported (1%-89% range between services). A further 8% of cases had some but not all 8 psychology screening data items reported. 24% of cases had a reason why the outcome was not reported, and 14% were missing data or a reason.

Reasons for not collecting 5-year old psychology outcome data for 2011-13 births

Appendix 7 shows the breakdown of reasons reported for not collecting 5-year old psychology outcome data for 2011-13 births; the most common reason selected was 'Lack of staff / facilities / equipment' (46%).

Figure 4.16 shows the variability in the reporting of all 8 psychology 5-year-audit data items according to the number of eligible cases within each cleft service. The funnel plot is centred on the adjusted national average of 54%[91].

Figure 4.16. Funnel plot showing the percentage of CRANE-consented 5-year olds, born 2011-2013, who had all psychology 5-year-audit data submitted to CRANE, according to cleft service.



Note: Data from Evelina London was not used to create funnel plot due to poor consent verification rate.

Note: Registered in CRANE by 30 June 2021. Funnel plot centred on the adjusted national percentage (54.2%) of children (born 2011-2013) with psychology outcomes reported. See Appendix 12 for the raw data (and exclusions) used to create this funnel plot.

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

[90] Terminology clarified since 2020 report, because one psychology data item is now captured earlier than at the 5-year-audit. 'Date of first face-to-face screening', separate to the date of the 5-year-audit can now be collected from birth (since October 2020).

[91] The overall mean was 54.3% and adjusted mean was 54.2% (excluding cleft services with consent verification outlier status).

The wide range in reporting may reflect the difficulty that some cleft services had in performing psychology assessments during the COVID-19 pandemic; including difficulties due to some services having no psychologist funded for MDT care.

The funnel plot shows that Trent and Northern Ireland are negative outliers for the percentage of eligible cases with all 8 psychology 5-year-audit data items reported. This because they fell below the lower 99.8% control limit, and were therefore excluded from the calculation of the overall average presented in the subsequent outcome funnels. Completing this process of identifying outliers, for the first time, has also allowed the identification of positive outliers such as Newcastle, Leeds, Liverpool and South Wales cleft services. See Box 11 and 12 (below) for comments from cleft services on their outlier status

Box 11. Summarised responses to being negative outliers for 'psychological wellbeing - data completeness at 5 years'. **Full responses published in the "2021 Annual Report: Responses to outlier process" document.**

This service "has not been staffed to provide a full cleft psychology service…limited clinical service is provided at the point that the Lead Team perceives the greatest clinical need which is across the adolescent and young adult group …The limited clinical service [available for 5-year-olds] is purely at the point of clinical need in a minority of the cleft population of our region, and therefore collects no audit data, and therefore no data is currently submitted to CRANE". The position of the Trent regional Cleft Network for Clinical Psychology provisions reflects a wider systems issue that Clinical Psychology is underprovided across Nottingham Children's Hospital in a variety of areas, this is a recognised as one of the major clinical risks held on the Trust's risk register, and is subject of a business case written to address the whole Nottingham Children's Hospital Clinical Psychology needs including those of the Trent Regional Cleft Network."

Trent cleft service, November 2021.

"The NI Cleft Palate team have had staffing issues in the past however this is being addressed and staff are committed to ensuring we have 100% data completeness for all areas moving forward. We have put in place a system where the Cleft Coordinator will now meet with each discipline and internal cross checking and quality control processes will be completed prior to submitting data to CRANE."

Northern Ireland cleft service, October 2021.

Box 12. Summarised responses to being positive outliers for 'psychological wellbeing - data completeness at 5 years'. **Full responses published in the "2021 Annual Report: Responses to outlier process" document.**

"Data collection was built into our service from the beginning and forms part of our care pathway…The final component of our 5 year data collection is the careful scheduling of 5 year audit clinics, with our audit coordinator identifying all patients for each cohort early every year, and then building specialist audit clinics around them over the 12 month period."

Newcastle cleft service, November 2021.

"Our Cleft Team includes a dedicated Cleft Audit Coordinator….Much of the credit and responsibility for achieving these high levels of data submission onto CRANE is due to [Cleft Audit Coordinator]'s efforts and other members within the Cleft Administrative Team including … [the] Cleft Clinic Coordinator"

Leeds cleft service, October 2021.

"Following the publication of the CRANE 2020 Annual Report, we were concerned about the level of possible missing data/data without a valid reason for missing/incorrect data for our service. We therefore undertook a review of all our records…and re-submitted these to CRANE prior to this year's report. This process was very labour intensive and involved considerable resource commitment from the clinical psychology team. We confirm our willingness to receive external review to learn from good practice (as per…the Outlier Policy)."

Liverpool cleft service, October 2021.

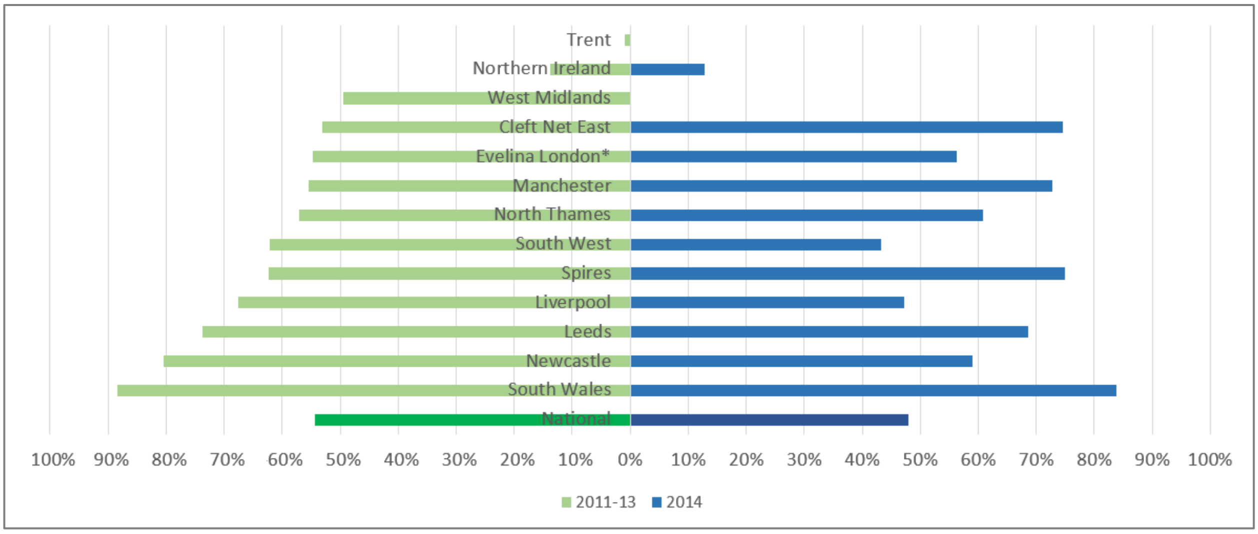
The South Wales cleft service quote the following as being key to achieving their positive outlier status: "Good administration support"; "a clear Was Not Brought policy which helps patients to attend for MDT appointments"; "designated clinics"; "a Clinical Psychologist (CP) and an Assistant Psychologist (AP) attend our clinics"; and having "the same dental nurse who takes height and weight measurements and also records the dmft scores and the reasons if not collected". "In terms of inputting, it is helpful only to have one or two staff inputting the data so it is consistent. We have identified our team secretary to do this who is now very familiar with the CRANE website… input(ing) information clinic by clinic".

South Wales cleft service, September 2021.

Anticipating challenges to data completeness due to the COVID-19 pandemic, Figure 4.17 summarises data completeness for eligible children born in 2014 (reporting of all 8 psychology 5-year-audit data items); which will be reported on in the CRANE 2022 Annual Report. This is compared with 2011-2013 data to provide pre-pandemic context[92]. **Appendix 12** shows the breakdown of psychology reporting for each cleft service, for 2011-13 as well as 2014 births.

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, and the ongoing impact of the pandemic in 2021, may have impacted services' ability to audit and submit the most recent year of CRANE data collection.

Figure 4.17. Percentage of CRANE-consented 5-year olds, born 2014, who had all psychology 5-year-audit data submitted to CRANE, according to cleft service – by comparison with children born 2011-13.



Note: Registered in CRANE by 30 June 2021. See **Appendix 12** for the raw data (and exclusions) used to create this graph.

Overall, of the 818 eligible 5-year old children born in 2014, 41% had all 8 psychology 5-year-audit data items reported (0%-84% range between services). 6 out of 13 cleft services were able to account for more than 50% of eligible cases.

Reasons for not collecting 5-year old psychology outcome data for 2014 births

Appendix 7 shows the breakdown of reasons reported for not collecting 5-year old psychology outcome data for 2014 births; the most common reason selected was 'Other' (48%). Comments provided to specify these 'other' reasons, in the main, described clinics cancelled / data not collected due to the COVID-19 pandemic.

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[92] A child born 31 December 2013, should have been audited for 5-year old outcomes before the age of 6. I.e. By the 31 December 2019 (pre-COVID-19 pandemic).

4.5.2. Face-to-face psychosocial screening before age 6 by cleft service

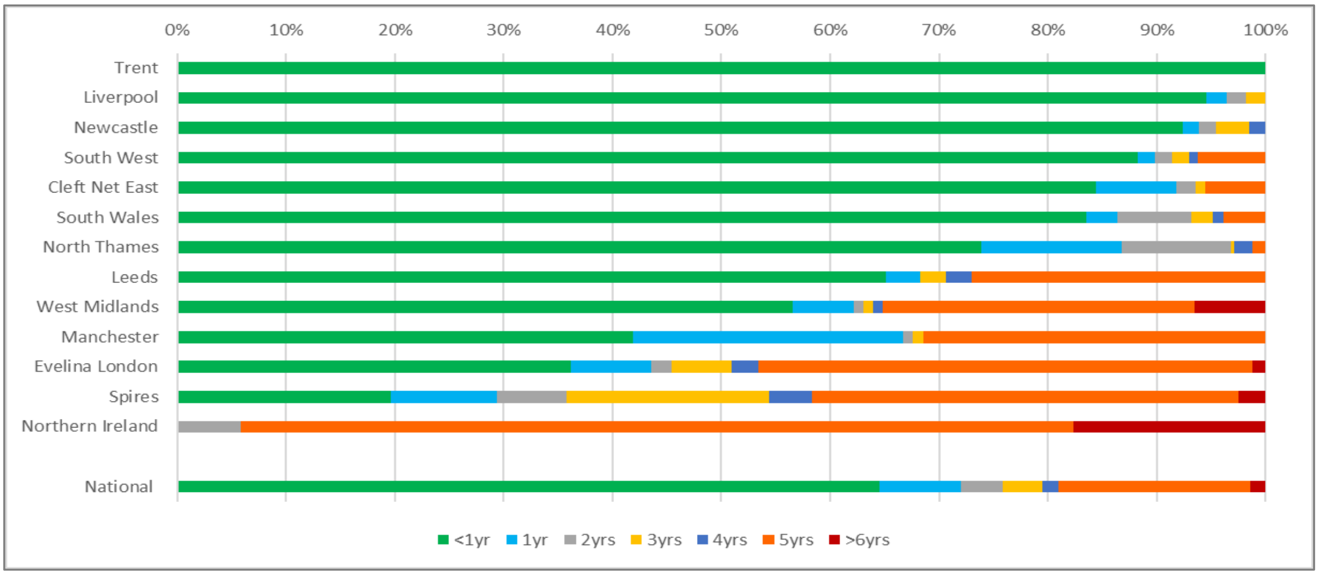
Although clinical psychologists have a target to see all children and families born with a cleft for a face-to-face psychosocial screen before the age of six years[93], families would ideally be seen earlier than this, where services' resources and structures allow. This is in order to: (a) Introduce and normalise psychology as part of the cleft service 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. Therefore this subsection of the report summarises:

- Information on children with 'date of first face-to-face screening'. The 'date of first face-to-face screening' data item is no longer collected in the 5-year-audit section of the CRANE Database; it is collected from birth (since October 2020); and
- information on children with dates of 'psychological screening at age five'.

The 'date of first face-to-face screening' was recorded for 1,795 children that went on to receive a psychology 5-year-audit. Where this date was unavailable, date of 'psychological screening at age five' was included in the analyses (adding information for 30 cases).

As shown in Figure 4.18, 65% of families of children born in 2011-13 were seen before their child's first birthday (this ranged from 0% to 100% according to cleft service). Overall, 8% were seen at age two, 4% were seen at age three, and 4% were seen at age four.

Figure 4.18. Percentage of CRANE-consented 5-year olds, born 2011-13, who were has face-to-face psychosocial screening by age, according to cleft service.



Note: Registered in CRANE by 30 June 2021. 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.

Dates of 'psychological screening at age five' were recorded for 1,626 5-year old children eligible for psychology audit. Where this date was unavailable, 'date of first face-to-face screening' was included in the analyses (adding information for 199 cases). Based on this information, 1,765 children were confirmed as having at least one face-

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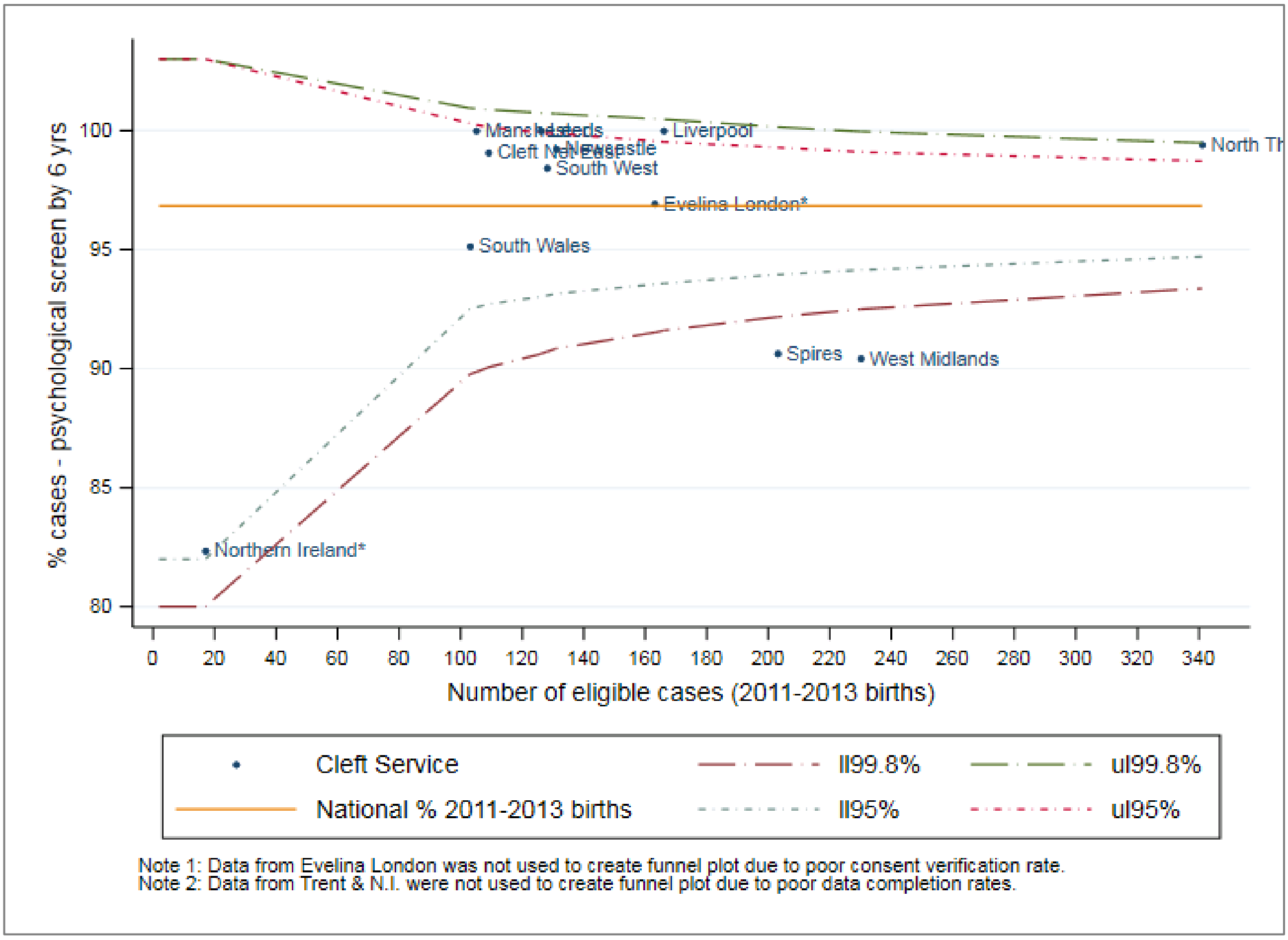
[93] Clinical Psychological & Counselling Services Standards Core Standard (#38). NHS Standard Contract - Cleft lip and / or palate services including non-cleft velopharyngeal dysfunction (all ages). NHS Commissioning Board, 2013. https://www.england.nhs.uk/specialised-commissioning-document-library/service-specifications/

to-face psychosocial screen before the age of 6 years (97% of the total 1,825[94] children who had been screened at the 5-year-audit or earlier)[95].

Figure 4.19 shows the percentage of 5-year olds with at least one psychology screen before the age of six, according to the number of eligible children at each cleft service with the funnel plot centred on the adjusted national average of 97%[96] (range 82%-100%).

Submitting this indicator of having at least one psychology screen before the age of six (#13) to the outlier process, for the first time, led to the identification of outliers for this outcome indicator.

Figure 4.19. Funnel plot showing the percentage of CRANE-consented 5-year olds, born 2011-2013, with at least one psychology screen before the age of six, according to cleft service.



Note: Registered in CRANE by 30 June 2021. Funnel plot centred on the adjusted national percentage (96.7%) of children (born 2011-2013) with at least one psychology screen before the age of six. See Appendix 12 for the raw data (and exclusions) used to create this funnel plot.

The funnel plot shows that the West Midlands and Spires are negative outliers for the percentage of eligible cases with at least one psychology screen before the age of six. This is because they fell below the lower 99.8% control limit on the funnel plot. Appendix 12 shows the breakdown of cases with reported psychology data at 5 years of age, meeting each psychology process or outcome standard, according to cleft service. See Box 13 (below) for comments from cleft services on their outlier status.

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[94] Dates of 'psychological screening at age five' were recorded in CRANE (n=1,626 for 2011-13 births). Where this date was unavailable, 'date of first face-to-face screening' – which can be collected any time point after birth – was included in the analyses (n=199 for 2011-13 births). **Caveat:** 'Date of first face-to-face screening' was not included in the 8 data items that contribute to data completeness at the point of the 5-year audit.

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

[96] The overall mean was 96.7% and adjusted mean was 96.8% (excluding cleft services with consent verification and data completeness outlier status).

Box 13. . Summarised responses to being negative outliers for 'face-to-face psychosocial screening before age 6'. **Full responses published in the "2021 Annual Report: Responses to outlier process" document.**

A number of factors contributed to this outlier status: "Significant staffing related issues across the Health in Mind team (maternity, sickness, vacancy, recruitment difficulties) which has reduced the dedicated support to the Cleft Service to 0.5 wte Clinical Psychologist, with only ad hoc support from Assistant Psychologists. Paediatric Psychology staffing levels are currently logged on the Hospital Trust risk register." Also "in addition to the screening contacts, the psychologist (and assistants) would also be seeing those who have requested psychological support or responding to clinical need identified by the team; clinical need will always be prioritised, sometimes resulting in less flexibility during the time when families are present in clinic"

West Midlands cleft service, October 2021.

"We note that the funnel plot for this measure is very narrow so that a small difference in case numbers could result in outlier status whilst still achieving a relatively high percentage of screening, as in this case. Prior to your letter, we were made aware of issues with documentation of psychological screening for some patients born in the years [reported on]…Some data has already been amended and this work will now be completed…in the light of your notification." "Workforce constraints" were also flagged as a challenge.

The Spires cleft service, October 2021.

Anticipating challenges to conducting psychological screening before the age of 6, due to the COVID-19 pandemic: Appendix 12 shows the breakdown of cases meeting each psychology process or outcome standard, according to cleft service, for 2011-13 as well as 2014 births. Data for 2014 births will be reported on in the CRANE 2022 Annual Report.

4.5.3 Tiers of Involvement Measure (TIM) at 5 years of age, by cleft service

Overall, 93% of the 1,639 eligible 5-year old children with TIM scores of 0 to 4 had TIM scores of 1a+, i.e. they were seen by a psychologist and a psychosocial screen was completed or psychological input arranged as required.

Psychological input provided in clinic can be preventative or in response to a concern raised by the family. Examples of preventative input include advice on talking to

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. Child and family seen by a psychologist and a psychosocial screen carried out with no further psychological input required (for the purposes of this report, TIM scores 1a and 1b are included in this category[97]).
2. Psychological input provided in clinic (for the purposes of this report, TIM scores 2a, 2b and 2c are included in this category[98]).
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[99]).

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[97] 1a is psychosocial screen undertaken and no concerns identified, 1b is where psychosocial screen undertaken and needs are being met by another service or agency.

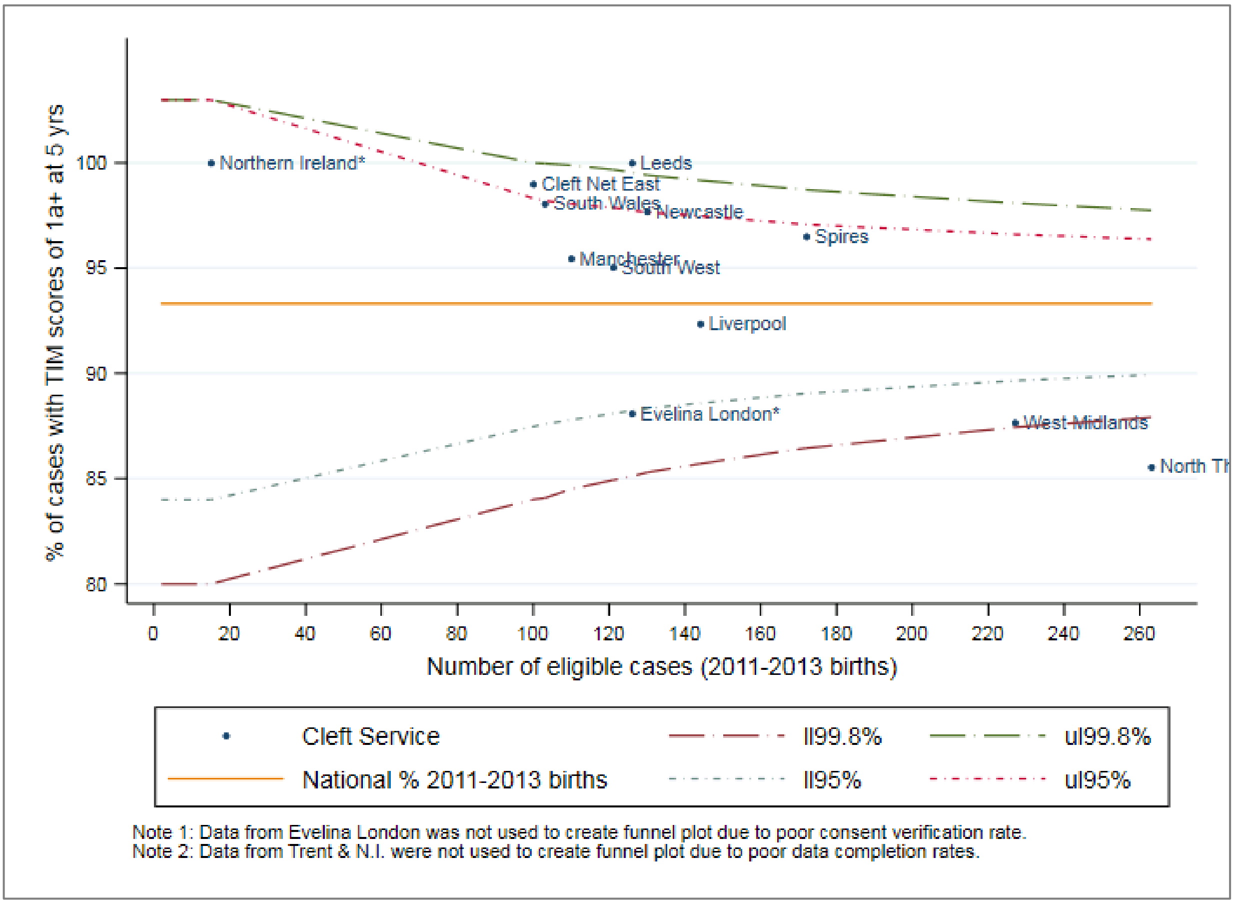
[98] 2a preventative psychological input provided, 2b psychological input provided in response to a concern, 2c psychological input provided including both preventative and input in response to a concern.

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

your child about their cleft, helping children and parents to prepare for potential comments and questions about their cleft. Examples of input in response to a concern include managing difficult behaviour and concerns about confidence or anxiety.

Figure 4.20 shows the percentage of 5-year olds with a TIM score of 1a+, according to the number of eligible children at each cleft service. The funnel plot is centred on the adjusted national average of 93%[100] (range 86%-100%).

Figure 4.20. Funnel plot showing the percentage of CRANE-consented 5-year olds, born 2011-2013, with a TIM score of 1a+, according to cleft service.



Note: Registered in CRANE by 30 June 2021. Funnel plot centred on the adjusted national percentage (93.3%) of children (born 2011-2013) with a TIM score of 1a+. See Appendix 12 for the raw data (and exclusions) used to create this funnel plot.

The funnel plot shows that North Thames is a negative outlier for the percentage of eligible cases with a TIM score of 1a+ (seen by or having had input from a psychologist). This is because they fell below the lower 99.8% control limit on the funnel plot. Completing this process of identifying outliers, for the first time, has also allowed the identification of Leeds as a positive outliers.

TIM scoring is currently undergoing calibration by the psychology CEN. Because of this exercise, this outcome is not included in the list of core indicators, found in Appendix 3.

Appendix 12 shows the breakdown of cases with reported psychology data at 5 years of age, meeting each psychology process or outcome standard, according to cleft service for 2011-13 births. See Box 14 and 15 (below) for comments from cleft services on their outlier status

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[100] The overall mean was 93.0% and adjusted mean was 93.3% (excluding cleft services with consent verification and data completeness outlier status).

Box 14. Summarised responses to being negative outliers for 'low rates of children identified as having a TIM score of 1a+'. **Full responses published in the "2021 Annual Report: Responses to outlier process" document.**

This cleft service conducted an extensive and thoroughly detailed review of their data, in response to this outlier process. Some "missing scores were entered in wrong clinical database (due to) human error". This cleft service also quoted trying to enter TIM data at a time that a 'reason outcome was not collected' had been recorded for SDQ scores, which would then render CRANE "unable to hold the (TIM) data".

Note: The way in which Psychology data is captured by the CRANE Database has been updated, as requested by the Psychology CEN, to address the latter challenge flagged by this service (October 2020).

North Thames cleft service, October 2021.

Box 15. Summarised responses to being positive outliers for 'high rates of children identified as having a TIM score of 1a+'. **Full responses published in the "2021 Annual Report: Responses to outlier process" document.**

"Our Cleft Team includes a dedicated Cleft Audit Coordinator….Much of the credit and responsibility for achieving these high levels of data submission onto CRANE is due to [Cleft Audit Coordinator]'s efforts and other members within the Cleft Administrative Team including … [the] Cleft Clinic Coordinator"

Leeds cleft service, October 2021.

Anticipating challenges to collecting TIM scores, due to the COVID-19 pandemic: **Appendix 12** shows the breakdown of cases meeting each psychology process or outcome standard, according to cleft service, for 2011-13 as well as 2014 births. Data for 2014 births will be reported on in the CRANE 2022 Annual Report.

4.5.4 High / very high Strengths and Difficulties Questionnaire (SDQ) scores

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[101].

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.

Overall, 18% of the 1,472 eligible 5-year old children with SDQ scores had high/very high SDQ scores.

This is higher than the rate of 10% of 5-year old children in the general population (i.e. the population norm) in Great Britain with high or very high SDQ scores (last sampled in the general population in 2000)[102].

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[101] 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

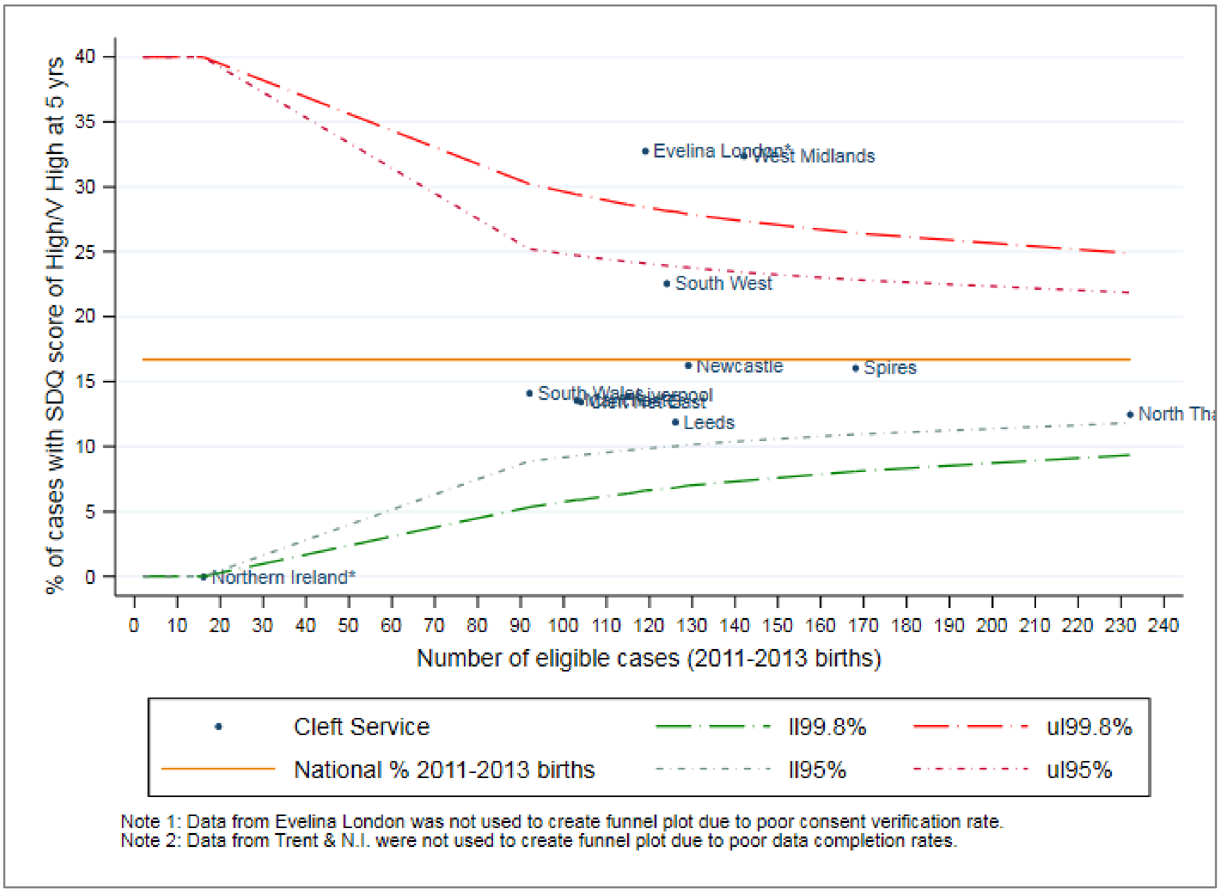
[102] The sample are described in more detail in: Meltzer, H., Gatward, R., Goodman, R., and Ford, F. (2000) Mental health of children and adolescents in Great Britain. London: The Stationery Office.

Figure 4.21 shows the percentage of 5-year olds with a TIM score of 1a+, according to the number of eligible children at each cleft service. The funnel plot is centred on the adjusted national average of 17%[103] (range 0%-33%).

The funnel plot shows that the West Midlands and Evelina London are negative outliers with a high percentage of eligible cases with high/very high SDQ scores; falling outside of the upper 99.8% control limit on the funnel plot.

Appendix 12 shows the breakdown of cases with reported psychology data at 5 years of age, meeting each psychology process or outcome standard, according to cleft service. Identifying outliers for this process indicator (#14), for the first time, did not lead to the identification of positive outliers. See Box 16 (below) for comments from cleft services on their outlier status.

Figure 4.21. Funnel plot showing the percentage of CRANE-consented 5-year olds, born 2011-2013, with a high/very high SDQ score, according to cleft service.



Note 1: Data from Evelina London was not used to create funnel plot due to poor consent verification rate.
Note 2: Data from Trent & N.I. were not used to create funnel plot due to poor data completion rates.

Note: Registered in CRANE by 30 June 2021. Funnel plot centred on the adjusted national percentage (16.7%) of children (born 2011-2013) with a high/very high SDQ score. See Appendix 12 for the raw data (and exclusions) used to create this funnel plot.

Box 16. Summarised responses to being negative outliers for 'high/very high SDQ total scores indicating difficulties'. **Full responses published in the "2021 Annual Report: Responses to outlier process" document.**

This service outline challenges associated with not being staffed appropriately, including "Paediatric Psychology staffing levels are currently logged on the Hospital Trust risk register". They reflected that being an outlier for children scoring higher levels of symptoms on the SDQ "may suggest higher levels of need, often requiring more time, liaison, etc."

West Midlands cleft service, October 2021.

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[103] The overall mean was 17.8% and adjusted mean was 16.7% (excluding cleft services with consent verification and data completeness outlier status).

This cleft service “identified that significant errors were made in the calculation of SDQ scores – particularly for the 2011 and 2012 cohorts - and that when the raw data (questionnaire scores) is recalculated correctly, the correct data appears to be in line with national averages”. They have “undertaken a review of our local practice in respect to the calculation of SDQ scores and will ensure that, in future, we perform a range of checks on our data to ensure that no further errors of these types occur. We have already checked all the original SDQ forms and rescored them.”

Evelina London cleft service, October 2021

Anticipating challenges to collecting SDQ scores, due to the COVID-19 pandemic: **Appendix 12** shows the breakdown of cases meeting each psychology process or outcome standard, according to cleft service, for 2011-13 as well as 2014 births. Data for 2014 births will be reported on in the CRANE 2022 Annual Report.

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 type and frequency of additional congenital malformations in children born with a cleft in England, (2) differences in speech outcomes at 5 years between those with and without congenital malformations of the circulatory system, and (3) differences in speech outcomes at 5 years between girls and boys.

Timeframe: (1) Additional congenital malformation rates were calculated using data for children born between 1 January 2010 and 31 December 2012. (2) Differences in speech outcomes at 5 years between those with and without congenital malformations of the circulatory system are reported for children born between 1 January 2006 and 31 December 2012. (3) Differences in speech outcomes at 5 years between girls and boys are reported for children born between 1 January 2006 and 31 December 2014.

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

5.1. Congenital malformations in children born alive with a cleft

Cohort summary	Data sources	The CRANE Database (consented cases only) linked with Hospital Episode Statistics (HES)
	Birth years	13 years: 2000 to 2012* *Linkage between HES and CRANE data only available for births up until 2012.
	Denominator	9,403 CRANE-consented children linked to HES records with cleft type agreement between both datasets.
	Numerators	3,653 children with additional congenital malformations, identified using ICD-10 codes Q00-Q99
	Countries	England
What did we find?	• Overall, 38.8% of children with a cleft had additional congenital malformations. • The presence of additional congenital malformations was most common in children with CP (53.0%), followed by those with BCLP (33.5%), UCLP (26.3%), and then CL (22.2%). • Body systems most likely to be affected by additional malformations were the circulatory system, musculoskeletal system and digestive system.	
Recommendations	• Given the frequency of certain structural malformations, clinicians should consider standard screening for children with a cleft. • Establishing good links with paediatric and genetic services is recommended.	

5.1.1. Methods

Data sources

These retrospective analyses 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).

Study cohort

10,483 children born alive between 1 January 2000 and 31 December 2012 who were diagnosed with a cleft affecting the lip and/or palate and were registered in CRANE were successfully linked to HES records. Of these, 1,080 were excluded because there was either no agreement on cleft type between the two data sources (n=239) or cleft type was missing from one of the datasets (n=841). In total 9,403 children were included in the analyses.

Of the children included, 2,114 (22.5%) were classified as having cleft lip only (CL), 4,509 (48.0%) had cleft palate only (CP), 1,896 (20.2%) had unilateral cleft lip and palate (UCLP), and 884 (9.4%) had bilateral cleft lip and palate (BCLP).

Diagnoses of additional congenital malformations

The International Classification of Diseases 10th revision (ICD-10) was used to identify congenital malformations in the study cohort. All congenital malformations, deformations and chromosomal abnormalities are classified using codes Q00-Q99 (see Appendix 13). HES records for any single admission contain at least 14 diagnosis code fields. The presence of any one of the ICD-10 diagnostic codes (Appendix 13) representing congenital malformations and chromosomal abnormalities, in any of these diagnosis fields of a HES record was used to identify a child as having a congenital malformation in addition to their cleft. These malformations were categorised according to the body or organ system they affected (e.g. circulatory system).

Analysis

The number and percentage of children with each ICD-10 code representing congenital malformations listed in **Appendix 13** was calculated. The number and percentage of children with at least one malformation affecting a particular body system was also determined. Finally, the overall number and percentage of children with at least one additional congenital malformation (the presence of any of the ICD-10 codes listed in **Appendix 13**) in their HES records were calculated. These rates were further calculated for each of the four cleft-type subgroups. The Chi-square test was used to assess variations in percentages across cleft types. A p value <0.05 was considered statistically significant.

5.1.2. Results

Overall, 3,653 (38.8%) children had diagnoses of additional congenital malformations in their HES records. These rates varied significantly according to cleft type (p<0.001). Additional malformations were most common among those with CP (53.0%), followed by those with BCLP (33.5%), UCLP (26.3%), and then CL (22.2%) (Table 5.1). These rates roughly translate to 1 in 2 with CP, 1 in 3 with BCLP, 1 in 4 with UCLP, and 1 in 5 with CL.

Table 5.1 Number and percentage of children, born 2000 to 2012, with a cleft and additional congenital malformation, according to cleft type.

	Full cohort	Children with additional malformations	
Type of cleft	**N**	**n**	**(%)**
Cleft lip	2,114	470	(22.2)
Cleft palate	4,509	2,388	(53.0)
Unilateral cleft lip and palate	1,896	499	(26.3)
Bilateral cleft lip and palate	884	296	(33.5)
Full study cohort	9,403	3,653	(38.8)

Table 5.2 shows the prevalence of malformations affecting each body system, as identified by different ICD-10 codes. Over 10% of the study cohort had at least one malformation of the circulatory system. The predominant malformations were those affecting the cardiac septa and those of the great arteries, which were identified in 7.6% and 5.2% of the cohort, respectively. Musculoskeletal malformations were also identified in over 10% of the study cohort. Deformities of the feet were most prevalent, affecting 3.5% of children, followed by malformations of the skull and facial bones, which were identified in 2.8% of children. Whilst 8.3% of the cohort were identified as having malformations of the digestive system, these were primarily attributed to malformations of the tongue, mouth and pharynx (6.1% of children).

The most common system affected by malformations varied according to cleft type. Almost all types of malformations were most common in those with CP; however, one third of children with CP had a record of ‘other congenital malformations’ in their HES records. A further breakdown of these codes revealed 27.3% of children with CP had ‘congenital malformation syndromes predominantly affecting facial appearance’. This was much less common in all other cleft type subgroups (<1.6%).

Table 5.2. Number and % of children born with a cleft who have additional congenital malformations reported in their Hospital Episodes Statistics records, according to the type of malformation/diagnosis, and cleft type

ICD-10 codes	Description/system	CL N=2,114	CP N=4,509	UCLP N=1,896	BCLP N=884	Total N=9,403	
		n (%)	n (%)	n (%)	n (%)	n (%)	P value
Q00-Q07	Congenital malformations of the nervous system	20 (0.9)	250 (5.5)	33 (1.7)	32 (3.6)	335 (3.6)	<0.001
Q10-Q18	Congenital malformations of eye, ear, face and neck	50 (2.4)	333 (7.4)	64 (3.4)	67 (7.6)	514 (5.5)	<0.001
Q20-Q28	Congenital malformations of the circulatory system	82 (3.9)	648 (14.4)	139 (7.3)	87 (9.8)	956 (10.2)	<0.001
Q30-Q34	Congenital malformations of the respiratory system	112 (5.3)	238 (5.3)	97 (5.1)	40 (4.5)	487 (5.2)	0.816
Q38-Q45	Other congenital malformations of the digestive system	92 (4.4)	496 (11.0)	120 (6.3)	68 (7.7)	776 (8.3)	<0.001
Q50-Q56	Congenital malformations of the genital organs	84 (4.0)	249 (5.5)	86 (4.5)	61 (6.9)	480 (5.1)	0.002
Q60-Q64	Congenital malformations of the urinary system	28 (1.3)	141 (3.1)	28 (1.5)	28 (3.2)	225 (2.4)	<0.001
Q65-Q79	Congenital malformations and deformations of the musculoskeletal system	126 (6.0)	653 (14.5)	112 (5.9)	99 (11.2)	990 (10.5)	<0.001
Q80-Q89	Other congenital malformations	61 (2.9)	1,482 (32.9)	74 (3.9)	74 (8.4)	1,691 (18.0)	<0.001
Q90-Q99	Chromosomal abnormalities, not elsewhere classified	25 (1.2)	294 (6.5)	33 (1.7)	32 (3.6)	384 (4.1)	<0.001
	At least one congenital malformation in addition to a cleft	470 (22.2)	2,388 (53.0)	499 (26.3)	296 (33.5)	3,653 (38.8)	<0.001

Note: P value for difference in percentage of children with additional congenital malformations between cleft types.

Previous studies using CRANE-HES linked English data that report cleft-related care or outcomes have, on average, identified approximately 22% of all children with a cleft as having additional anomalies or syndromes[104]. In these previous studies, the list of congenital malformations and chromosomal abnormalities used to identify these children was different. The ICD-10 codes used were primarily restricted to the nervous system, circulatory system, and some syndromes frequently occurring among children with a cleft that were thought to influence the care or outcomes being reported. The current study is primarily focussed on reporting the range and frequency of all congenital malformations occurring among the cleft population. This highlights that reported rates of additional anomalies or syndromes will depend on the definition used and the purpose for which the malformations were detected.

Identifying the frequency and range of additional malformations occurring among children born with a cleft is important for counselling parents and for planning and commissioning cleft services. The implementation of routine screening for certain cleft phenotypes should be considered based on the high prevalence of additional malformations identified in the study. Good links with local genetic and paediatric services are recommended. Further research identifying the prevalence of other diagnoses, including intellectual disabilities and development disorders, would be beneficial.

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[104] Fitzsimons KJ, Copley LP, Deacon SA, et al. (2013) Hospital care of children with a cleft in England. *Archives of Disease in Childhood* 98: 970-974.
Fitzsimons KJ, Copley LP, Smallridge JA, et al. (2014) Hospital admissions for dental treatment among children with cleft lip and/or palate born between 1997 and 2003: an analysis of Hospital Episode Statistics in England. *International Journal of Paediatric Dentistry* 24: 200-208.
Fitzsimons KJ, Copley LP, van der Meulen JH, et al. (2017) Grommet surgery in children with orofacial clefts in England. *The Cleft Palate-Craniofacial Journal* 54: 80-89.

5.2. Congenital malformations of the circulatory system and speech at 5 years

<table>
<tr><td rowspan="6">Cohort summary</td><td>Data source</td><td>The CRANE Database (consented cases only) linked with Hospital Episode Statistics (HES)</td></tr>
<tr><td>Birth years</td><td>Seven years: 2006 to 2012*
*Linkage between HES and CRANE data only available up until 2012.</td></tr>
<tr><td>Denominator</td><td>2,511 5-year-old children born with a cleft affecting the palate (CP, UCLP, BCLP) who had all 16 CAPS-A speech parameters reported to CRANE
2,356 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</td></tr>
<tr><td>Numerator</td><td>The number of children with a particular speech outcome or the number meeting a standard</td></tr>
<tr><td>Exclusions
(not mutually exclusive)</td><td>• 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[105]
• Children without a full set of CAPS-A parameters being reported</td></tr>
<tr><td>Countries</td><td>England</td></tr>
<tr><td>Aim</td><td colspan="2">To explore whether there are differences in speech outcomes at 5 years of age between those with and without congenital malformations of the circulatory system.</td></tr>
<tr><td>Exposure</td><td colspan="2">• Congenital malformations of the circulatory system, identified using ICD-10 codes Q20-Q28.</td></tr>
<tr><td>Standard/outcomes</td><td colspan="2">• The achievement of speech within the normal range – Standard 1 (see text below for a full description of these standards[106])
• The achievement of speech without difficulties resulting from existing or previous structural anomalies – Standard 2a
• The achievement of speech without significant cleft-related articulation difficulties – Standard 3</td></tr>
<tr><td>What did we find?</td><td colspan="2">• Children without congenital malformations of the circulatory system were significantly more likely to meet cleft speech standards than those with these malformations.
• When restricting the analyses to children who were reported on CRANE to be non-syndromic, differences between those without and those with malformations of the circulatory system were only statistically significant for cleft speech standard 2a. Whilst a greater percentage of children without circulatory system malformations met standard 1 and standard 3 than those with these malformations, differences were not statistically significant.
• When restricting the analyses to children with cleft palate alone (CP) who were reported on CRANE to be non-syndromic, differences between those without and those with malformations of the circulatory system were statistically significant for all three speech standards.
• With adjustment for cleft type, children with malformations of the circulatory system had significantly reduced odds of achieving standard 2a and standard 3 than those without malformations of the circulatory system.</td></tr>
<tr><td>Recommendations</td><td colspan="2">• Surgical and Speech and Language Therapy CENs should agree whether to exclude children with congenital malformations from between-centre comparisons in speech outcome reporting in the future.</td></tr>
</table>

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

[106] 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 and record linkage

These retrospective analyses 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 cleft affecting the palate who were born from 2006 onwards. Children with a submucous cleft palate were excluded. 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. Children were included in the analyses if their crane records were successfully matched with HES records and they had complete 5-year old speech data available. In total, 2,511 children were included in these analyses. Of these, 1,318 (52.5%) had CP, 834 (33.2%) had UCLP, and 359 (14.3%) had BCLP. We also present results for children reported on CRANE as being non-syndromic (n=2,356). For these analyses, 1,199 (50.9%) had CP, 810 (34.4%) had UCLP, and 347 (14.7%) had BCLP.

Diagnoses of additional congenital malformations

The International Classification of Diseases 10th revision (ICD-10) was used to identify congenital malformations in the study cohort. All congenital malformations of the circulatory system are classified using codes Q20-Q28 (see Appendix 13). HES records for any single admission contain at least 14 diagnosis code fields. The presence of any one of the ICD-10 diagnostic codes (**Appendix 13**) representing congenital malformations of the circulatory system, in any of these diagnosis fields of a HES record was used to identify a child as having this type of malformation.

Speech outcomes

Speech was evaluated using Cleft Audit Protocol for Speech – Augmented (CAPS-A) scores collected at 5 years of age. The CAPS-A includes 16 parameters, which are listed in **Appendix 11**, along with their potential scores. The CAPS-A scores are used to determine whether a child meets three nationally agreed cleft speech standards:

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 **Appendix 11**.

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 **Appendix 11**.

Analyses

Children were grouped according to the absence or presence of congenital malformations of the circulatory system. Percentages describing achievement of standards were compared between the two groups with chi squared tests. Logistic regression was used to determine the odds of meeting each cleft speech standard according to the absence or presence of congenital malformations of the circulatory system, adjusted for cleft type. 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.

5.2.2. Results

Congenital malformations

Of 2,511 children with 5-year old speech outcome data submitted to CRANE and whose records were linked to HES, 199 (7.9%) had ICD-10 codes relating to congenital malformations of the circulatory system. Of these, 124 (62.3%) had CP, 52 (26.1%) had UCLP and 23 (11.6%) had BCLP. Of those with circulatory system malformations, 30 (15.1%) were reported on CRANE to have syndromes, and they would therefore usually be excluded from cleft speech outcome reporting.

Meeting the cleft speech standards

Table 5.3 shows a significantly higher percentage of children without malformations of the circulatory system met each cleft speech standard than those with malformations.

Table 5.3 Number and percentage of children meeting each cleft speech standard, according to the absence or presence of congenital malformations in the circulatory system.

		Meeting standard					
Malformations of the circulatory system	Total	S1. Normal speech		S2a. No structurally-		S3. No cleft-related	
	N	N	(%)	N	(%)	N	(%)
Absent	2312	1371	(59.3)	1638	(70.9)	1557	(67.3)
Present	199	102	(51.3)	125	(62.8)	120	(60.3)
Total	2511	1473	(58.7)	1763	(70.2)	1677	(66.8)
p value		***0.027***		***0.017***		***0.043***	

P value for difference in percentage of children meeting the standard between those without and those with malformations of the circulatory system

Table 5.4 includes only children reported to CRANE as being non-syndromic. All of these children would therefore usually be included in CRANE speech outcome reporting. Differences in percentages of those meeting the standard between the two groups are statistically significant for Standard 2a only.

Table 5.4 Number and percentage of children, reported to be non-syndromic on CRANE, meeting each cleft speech standard, according to the absence or presence of congenital malformations in the circulatory system.

		Meeting standard					
Malformations of the circulatory system	Total non-	S1. Normal speech		S2a. No structurally-		S3. No cleft-related	
	N	N	(%)	N	(%)	N	(%)
Absent	2187	1300	(59.4)	1546	(70.7)	1481	(67.7)
Present	169	92	(54.4)	106	(62.7)	106	(62.7)
Total	2356	1392	(59.1)	1652	(70.1)	1587	(67.4)
p value		*0.202*		***0.029***		*0.182*	

P value for difference in percentage of children meeting the standard between those without and those with malformations of the circulatory system

Table 5.5 has further restricted the analyses to those with a non-syndromic CP to aid a fairer comparison, given we have previously shown that speech outcomes vary significantly according to cleft type. The table shows that a significantly higher percentage of children without malformations of the circulatory system met each of the three standards than those with the malformations.

Table 5.5 Number and percentage of children with CP alone, reported to be non-syndromic on CRANE, meeting each cleft speech standard, according to the absence or presence of congenital malformations in the circulatory system.

		Meeting standard					
Malformations of the circulatory system	Total non-	S1. Normal speech		S2a. No structurally-		S3. No cleft-related	
	N	N	(%)	N	(%)	N	(%)
Absent	1099	785	(71.4)	822	(74.8)	886	(80.6)
Present	100	58	(58.0)	60	(60.0)	68	(68.0)
Total	1199	843	(70.3)	882	(73.6)	954	(79.6)
p value		***0.005***		***0.001***		***0.003***	

P value for difference in percentage of children meeting the standard between those without and those with malformations of the circulatory system

Table 5.6 shows the odds of achieving each standard according to the absence or presence of congenital malformations of the circulatory system, adjusted for cleft type, among children reported to CRANE as being non-syndromic. Those with these malformations had significantly lower odds of achieving cleft speech standard 2a and standard 3.

Table 5.6 Odds ratios, adjusted for cleft type, for meeting each cleft speech standard, according to the absence or presence of congenital malformations in the circulatory system among children reported to be non-syndromic on CRANE.

		Adjusted odds ratio of meeting standard					
Malformations of the circulatory system	Total non-	S1. Normal speech		S2a. No structurally-		S3. No cleft-related	
	N	OR	(95% CI)	OR	(95% CI)	OR	(95% CI)
Absent	2187	Ref		Ref		Ref	
Present	169	0.73	(0.53 to 1.01)	0.67	(0.49 to 0.93)	0.70	(0.50 to 0.98)
p value		*0.060*		***0.018***		***0.038***	

From January 2021 the CRANE Database changed the way it captured data on syndromes and additional diagnoses. Prior to January 2021, the ***Syndromes*** section of the CRANE database required cleft services to enter free text to describe named syndromes and descriptions of affected systems. This information is now captured by clear categories. These categories are based on analyses of free text entries on CRANE and analyses of additional diagnoses recorded in HES for children with a cleft. The nine most common syndromes/additional diagnoses for children with a cleft are now listed on the CRANE database and 'congenital malformations of the circulatory system' is one of these.

At the end of 2020, CRANE reviewed all CRANE records with syndrome information entered. Each record had this information re-coded according to the new categories. Any cases whose syndrome/additional diagnoses did not fit into one of the first nine categories was re-coded as ‘other’ and the free text entry was migrated to the new system.

The use of categories will allow CRANE greater flexibility with analyses, as different syndromes/significant additional diagnoses can now be included or excluded depending on the outcome of interest. Linkage with HES has enabled CRANE to identify those children with malformations of the circulatory system and to explore whether speech outcomes are different for those with these diagnoses. Surgical and Speech and Language Therapy CENs now need to agree whether or not to exclude children with circulatory system malformations from between-centre comparisons in speech outcome reporting in the future.

5.3. Is there a difference in cleft-related speech outcomes between girls and boys at 5 years?

Cohort summary	Data source	The CRANE Database (consented cases only)
	Birth years	Seven years: 2006 to 2014
	Denominator	3,178 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 with submucous cleft palates[107] • Children without a full set of CAPS-A parameters being reported • Children audited outside of the recommended time frame (5th year) • Children missing sex information
	Countries	England, Wales and Northern Ireland
Aim	To explore whether there are differences in speech outcomes at 5 years of age between boys and girls.	
Exposure	• Sex, as reported on CRANE.	
Standard/outcomes	• The achievement of speech within the normal range – Standard 1 (see text below for a full description of these standards[108]) • The achievement of speech without difficulties resulting from existing or previous structural anomalies – Standard 2a • The achievement of speech without significant cleft-related articulation difficulties – Standard 3	
What did we find?	• Among children with cleft palate and unilateral cleft lip and palate, a significantly higher percentage of girls met cleft speech standards 1 and 3 than boys. • Among children with BCLP, a significantly higher percentage of boys met cleft speech standard 2a. Differences between boys and girls with BCLP were not significant for cleft speech standards 1 and 3. • With adjustment for cleft type, boys had significantly reduced odds of achieving cleft speech standard 1 and 3. • Adjusted odds for achieving standard 2a were not statistically significant.	
Recommendations	• The child's sex should be factored into risk adjustment models when reporting cleft-related speech outcomes at 5 years.	

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

[108] 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).

5.3.1. Methods

Data sources

This retrospective analysis was based on data held in the Cleft Registry and Audit Network (CRANE) Database.

Cohort

Our analyses were restricted to CRANE-consented children born 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 2014. Children were included in analyses if their sex was recorded on CRANE and they had complete 5-year old speech audit data available.

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. The CAPS-A includes 16 parameters, which are listed in **Appendix 11**, along with their potential scores. The CAPS-A scores are used to determine whether a child meets three nationally agreed cleft speech standards:

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 **Appendix 11**.

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 **Appendix 11**.

Analyses

Children were grouped according to sex and cleft type. Percentages describing achievement of standards were compared between groups with chi squared tests. Logistic regression was used to determine the odds of meeting each cleft speech standard according to sex, adjusted for cleft type. 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.[4]

5.3.2. Results

Sex

Of 3,178 children with 5-year-old speech outcome data submitted to CRANE, 1,744 (54.9%) were male and 1,434 (45.1%) female. Of these, 1,640 (51.6%) had CP, 1,082 (34.0%) had UCLP and 456 (14.3%) had BCLP.

Meeting the cleft speech standards

Table 5.7 shows the number and percentage of females and males according to each cleft type. In line with the literature, cleft palate alone occurred more commonly in females and UCLP or BCLP occurred more commonly in males.

Table 5.7 Number and percentage of females and males according to cleft type.

	Total	Female		Male	
Cleft type	N	N	(%)	N	(%)
Cleft palate	1640	938	(57.2)	702	(42.8)
Unilateral cleft lip and palate	1082	368	(34.0)	714	(66.0)
Bilateral cleft lip and palate	456	128	(28.1)	328	(71.9)
Total	3178	1434	(45.1)	1744	(54.9)

Table 5.8 demonstrates the number and percentage of children meeting each speech standard. 60.2% of the full cohort achieved speech outcome standard 1 at age 5. Among children with cleft palate and unilateral cleft lip and palate, a significantly higher percentage of girls met cleft speech standards 1 and 3 than boys. Among children with BCLP, a significantly higher percentage of boys met cleft speech standard 2a. Differences between boys and girls with BCLP were not significant for cleft speech standards 1 and 3.

Table 5.8 Number (%) of children meeting each cleft speech standard, according to sex and cleft type.

			Meeting standard					
		Total	S1. Normal speech		S2a. No structurally-related speech difficulties		S3. No cleft-related articulation difficulties	
	Sex	N	N	(%)	N	(%)	N	(%)
All children		3178	1913	(60.2)	2252	(70.9)	2158	(67.9)
	Female	1434	954	(66.5)	1020	(71.1)	1064	(74.2)
	Male	1744	959	(55.0)	1232	(70.6)	1094	(62.7)
	p value		***<0.001***		*0.763*		***<0.001***	
Cleft palate		1640	1178	(71.8)	1228	(74.9)	1317	(80.3)
	Female	938	703	(75.0)	704	(75.1)	774	(82.5)
	Male	702	475	(67.7)	524	(74.6)	543	(77.4)
	p value		***0.001***		*0.850*		***0.009***	
Unilateral cleft lip and palate		1082	575	(53.1)	737	(68.1)	665	(61.5)
	Female	368	212	(57.6)	252	(68.5)	245	(66.6)
	Male	714	363	(50.8)	485	(67.9)	420	(58.8)
	p value		***0.035***		*0.854*		***0.013***	
Bilateral cleft lip and palate		456	160	(35.1)	287	(62.9)	176	(38.6)
	Female	128	39	(30.5)	64	(50.0)	45	(35.2)
	Male	328	121	(36.9)	223	(68.0)	131	(39.9)
	p value		*0.197*		***<0.001***		*0.346*	

P value relates to difference in the percentage of females and males meeting the standard.

Table 5.9 shows the odds of achieving each speech standard when adjusting for cleft type. Boys had significantly reduced odds of achieving cleft speech standard 1 and 3. The adjusted odds for achieving standard 2a were not statistically significant.

Table 5.9 Odds ratios and 95% confidence intervals (CI), adjusted for cleft type, for meeting each cleft-related speech standard, according to sex.

		Meeting standard					
	Total	S1. Normal speech		S2a. No structurally-related speech difficulties		S3. No cleft-related articulation difficulties	
Sex	N	aOR	(95% CI)	aOR	(95% CI)	aOR	(95% CI)
Female	1434	Ref		Ref		Ref	
Male	1744	0.78	(0.47 to 0.91)	1.09	(0.93 to 1.29)	0.78	(0.66 to 0.92)

It would be useful to compare these outcomes to the differences seen in speech and language acquisition in the non-cleft population, as it is known that in general boys are more likely than girls to have speech and language difficulties at this age.

6. Development of the CRANE Database and future directions

6.1. Future development of the CRANE Database and website

CRANE is continuing to work with NHS England, Wales and Northern Ireland to develop a new contract to sustainably support the project going forward. Current contract and funding arrangements covers the registry and audit function of the database as well as a limited amount of research time for database development. CRANE is committed to working with stakeholders and partners to develop the research potential of the data held and this is and will be done through collaborative research funding applications.

Work is ongoing with our IT provider, Crown Informatics Limited, to upgrade the database, in line with agreed developments – as proposed by our stakeholders:

- More accurate data collection of syndromes – this went live in January 2021.
- Dental Defects of Enamel (DDE) section/items (at 5 and 10yrs) as proposed by the Paediatric Dental CEN of CFSGBI – due to go live in January 2022.
- Psychology data section as proposed by the Psychology CEN – work is due to commence on this in early 2022.
- LAHSAL data collection items changed to collect LAHSHAL data to increase the phenotypic data available for analysis and linkage to other projects.
  Initial development work for LAHSAL to LASHAL conversion has been paused as recent collaborative work with the Cleft Collective demonstrated deteriorating concordance between the two datasets with increasing complexity of phenotypic description of Clefts. Work is ongoing to investigate the inter and intra-rator reliability of both LAHSAL and LAHSHAL to inform future direction and upon completion CRANE will discuss with the CDG an appropriate way forward.
- Surgical data collection. This will be taken forward in collaboration with the Surgical CEN of CFSGBI.
  CRANE has recently shared data with CDG and The Federation of Specialist Surgical Associations (FSSA) data pertaining to speech outcomes in relation to the timing of primary surgery. This has helped to define appropriate windows for primary lip and palate repair for recent RCS guidance on surgical priority. Lessons learnt in this exercise emphasise the importance of accurate coding of surgical activity. CRANE intends to engage the surgical CEN and utilise a list of key operative codes for use across the UK with the intention of linking to HES and related databases to collect operative information without burdening clinical colleagues.
- Hearing Experience. CRANE is working with the ENT/Audiology Clinical Excellence network to understand what data it is possible to collect, to document hearing experience of children with a cleft during early life (0-6 years of age).
  As part of this work CRANE is in the final stages of a data linkage exercise with the national Newborn Hearing Screening Programme (NHSP) to see if through linkage it is possible to identify rates of congenital deafness in cleft children and in hearing cleft patient's the presence (and level) of early hearing dysfunction.
- Primary Bone Grafting Outcomes and Orthodontic Care. CRANE has opened discussions with the Orthodontic CEN exploring the possibility of collection of data on both Bone graft assessment / outcome and also orthodontic care.

Should such data collection be supported for the first time, CRANE would collect information on all areas / specialties providing interventional primary clinical care for persons affected by clefting.

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 explore the potential for retrospectively entering data relating to children consented at 5-year-old audit. This will allow the project to achieve full UK coverage in a shorter time frame and is a significant step forward. Once data sharing agreements are in place for prospective registration of newborns affected by clefting in Scotland, CRANE will work with NHS Scotland on the detail of retrospective entry.

6.3. Outcome measures

6.3.1 Risk Adjustment

Despite significant improvement in both the volume and quality of data collection across the spectrum of cleft care, the lack of ability to risk adjust data continues to undermine potential for valid comparison of the outcomes achieved across the UK. A volume of data is now available within CRANE and is starting to be utilised to investigate risk stratification of cleft outcomes in the UK. Starting in 2019, work began to identify determinants of risk in speech and dental outcomes. Initial analyses have identified five independent determinants of speech (cleft type, extent of palatal involvement, sex, Robin sequence and presence of associated cardiovascular malformation) and two independent modulators of dental health (Cleft type and social background). Results of these investigations are being prepared for peer reviewed publication and dissemination. Moving forward, CRANE intends to develop a limited speech risk adjustment model for speech and dental outcomes for inclusion in future reports. For the purposes of prospective speech outcome comparisons between cleft services, 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. it was hoped to have this process complete for reporting in the 2020 and/or 2021 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 has continued through 2021/22 and an update on progress will be made in due course.

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.

The team has also encountered additional challenges in accessing data, such as from the NHSP, as a result of the transfer of the control of key healthcare datasets to NHS Digital from Public Health England (PHE). The transfer came about as part of the government's reforms to the public health system announced in March 2021.

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. Long term sustainable funding of CRANE is required to allow realisation of this aim.

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 the impact of the COVID-19 pandemic on rates of cleft surgery, as well as exploring geographic variations in adult cleft hospital activity over the last 10 years. This will 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 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 adjusted speech outcomes and other similar activity. Having this ability in place will reduce the already significant data collection burden on clinical teams. For example, cleft operative interventions and their timing data can be accessed through direct linkage with HES data rather than asking teams to record every operation on CRANE. Once ongoing HES linkage is achieved a pilot project looking at accuracy of HES held data with prospectively collected operative data will be required.

6.4.3. Newborn Hearing Screening Programme (NHSP)

We have approval for linkage between the CRANE Database and the Newborn Hearing Screening Programme (NHSP)[109] 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.

A contract to access this data was signed on 30 September 2021. Nevertheless, we have yet to receive clarity as to when the datasets will be made available to CRANE. This delay is a consequence of the transfer of the control of key healthcare datstest to NHS Digital from Public Health England (PHE). A challenge being experienced by other national clinical audits. The transfer came about as part of the government's reforms to the public health system announced in March 2021

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, but a presentation form the work was achieved at the delayed Craniofacial Society On-line scientific meeting in September of 2021. This work identified deteriorating concordance between the CRANE dataset and the Cleft collective data set with increasing complexity of phenotypic description (Cleft type, laterality of cleft type , LAHSAL code and LAHSHAL code) it intended that results of this collaborative work will be further disseminated in terms of a peer reviewed publication.

2021 saw further collaboration between researchers based at the Cleft collective and CRANE. Peer reviewed publications including data analysed at CRANE, or summary data shared by CRANE resulted in acceptance for peer reviewed publication, papers describing the effect of facial growth on speech outcomes and the effect of smoking on the incidence of cleft (detailed in section 6.6.2). As we (hopefully) move to more recognisable working environments in 2022, collaborative work will continue to develop enhancing the strengths of both datasets through synergistic working for the betterment of patients, clinicians and researchers alike.

6.5. Quality Dashboard

The CRANE project team have submitted data on behalf of cleft services since the 2016/17 Specialised Services 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).

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

- Measure Number CLP01: The number of Parents contacted by a Cleft services 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 services 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[110]) – 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 for the Specialised Services Quality Dashboard–speech data – is provided directly by the cleft services. 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).

From November 2021 (for the Q2 2021/22), dashboard data submissions were made directly to NHS England and NHS Improvement – as Methods no longer holds the contract to deliver these.

We have populated a web page with information on how six of the indicators are directly submitted by CRANE, to help cleft services better understand how each data point is calculated. Please see – https://www.crane-database.org.uk/resources/specialised-services-quality-dashboard-indicators-submitted-by-crane/

All other indicator data is provided directly by cleft services to NHS England and NHS Improvement (no longer Methods). Queries about other indicators (not provided by CRANE) can be raised directly with qcrs.externaldatagroup@england.nhs.uk

6.6. CRANE Communications

6.6.1 Dissemination of 2021 findings

- This report will be available on the CRANE website from December 2021.
- Publication of the Annual Report will be announced via the regular quarterly Newsletter.
- A Summary of Findings for Patients and Parents/Carers from this 2021 Annual Report will be produced in collaboration with CLAPA. CRANE aims to publish this contemporaneously with the main report. The summary will also be made available on the CRANE website.
  A Twitter feed for the project (@CRANE_News, active since August 2019) helps highlight and share activity, developments and outputs throughout the year.

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[110] 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.

6.6.2 Publications and Presentations related to the CRANE Database delivered in 2021

Peer reviewed Publications

- **The impact of changing cigarette smoking habits and smoke-free legislation on orofacial cleft incidence in the United Kingdom: evidence from two time-series studies.** Matthew Fell, Craig Russell, Jibby Medina, Toby Gillgrass, Shaheel Chummun, Alistair R.M. Cobb, Jonathan Sandy, Yvonne Wren, Andrew Wills, Sarah J. Lewis. PLOS ONE 2021 - doi: https://doi.org/10.1101/2021.06.25.21259517. *Accepted for publication October 2021 - pre-printed on medRxiv.*
- **The relationship between maxillary growth and speech in children with a unilateral cleft lip and palate at 5 years of age.** Matthew Fell, Jibby Medina, Kate Fitzsimons, Miriam Seifert, Anne Roberts, Craig Russell, Scott Deacon. Published 23 April 2021, The Cleft Palate-Craniofacial Journal.

Presentations

- **Comparing Apples and Pears – Identifying And Quantifying Differences In Speech Outcomes For Different Cleft Types.** Sophie Butterworth, Kate Fitzsimons, Jibby Medina, Scott Deacon, Hussein Wahedally, Jan van der Meulen, Craig Russell. Annual Conference for the Craniofacial Society of Great Britain and Ireland (CFSGBI). September 2021 (VIRTUAL ORAL PRESENTATION).
- **Pierre Robin Sequence Cleft Patients Are At Increased Risk of Poorer Speech Outcomes When Compared to Non Syndromic Cleft Palate But Not Cleft Lip and Palate Patients**. Craig Russell, Sophie Butterworth, Kate Fitzsimons, Jibby Medina, Min Hae Park, Hussein Wahedally, Jan van der Meulen. Annual Conference for the Craniofacial Society of Great Britain and Ireland (CFSGBI). September 2021 (VIRTUAL ORAL PRESENTATION).
- **Craniofacial Conference CRANE Update.** Craig Russell, Sophie Butterworth, Kate Fitzsimons, Jibby Medina, Min Hae Park, Hussein Wahedally, Jan van der Meulen. Annual Conference for the Craniofacial Society of Great Britain and Ireland (CFSGBI). September 2021 (VIRTUAL ORAL PRESENTATION).
- **Validation of the cleft classification in the Cleft Collective Cohort Studies.** Amy Davies, The Cleft Collective Team, The CRANE Database Project Team and The Cleft Multidisciplinary Collaborative. Annual Conference for the Craniofacial Society of Great Britain and Ireland (CFSGBI). September 2021 (VIRTUAL ORAL PRESENTATION).
- **Patient Reported Experience Measures (PREM) Feasibility Study.** Vanessa Hammond, J. Simmonds, C. Setters, J. Cartwright, Scott Deacon, Jibby Medina, South Wales Cleft Service, The Welsh Centre for Cleft, South West Cleft Service and CRANE. Annual Conference for the Craniofacial Society of Great Britain and Ireland (CFSGBI). September 2021 (VIRTUAL POSTER PRESENTATION).
- **The impact of changing cigarette smoking habits and smoke-**free legislation on orofacial cleft incidence in the UK. Matthew Fell, Craig Russell, Jibby Medina, Toby Gilgrass, Shaheel Chummun, Alistair Cobb, Jonathan Sandy, Yvonne Wren, Andrew Wills, Sarah Lewis, Cleft Collective, Scottish Cleft Service, CRANE, South West Cleft Service, University of Bristol.

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 2022:

- 4th International Congress of cleft lip, palate & related Craniofacial Anomalies, Edinburgh, Scotland, UK. 11-15 July 2022.

7. Conclusion

As we move towards a second Christmas period affected by yet another new variant of COVID-19 it is understandable people are frustrated about the ongoing nature of the pandemic and its effects. one could look at this year's activity report with a glass half empty given reductions in accrued data available for analysis, we prefer to consider it fortunate to have any data at all from this period. In fact we are amazed at the actual amount of data received.

We therefore wish to extend a huge thank-you to all the patients who attended clinics (when they could easily have decided not to), to all the clinicians who continued to assess patients and collect data for audit purposes (when they could have been easily distracted by other pressures in hospitals), to admin and co-ordination teams who collated and entered the collected data on to the database (when hospital admin services were under pressure from change in working environments and had increased workloads due to the challenges of ever changing clinic and admission practices), and to clinical leads who supported ongoing audit (when it would have been easier to batten down the hatches and deliver only essential acute clinical care). Thank-you one and all.

As we look forward to a new year and the opportunities it may bring we would especially like colleagues (clinical and research) to reflect on the key findings and recommendations section at the start of this document. As we read through this section we see that the statements made in each row could stimulate whole areas of research (each in their own right) that would further inform cleft care and (potentially) lead to improvements for the patients and families affected by clefting. We would encourage all to consider what questions these key findings (and associated detailed data) stimulate.

CRANE runs on a relatively small budget and it involves just a few people (each working only part time on the project) therefore there is a limit to what can be done on our own. We do however recognise the potential of the data set held. We believe that the potential (and the experience of the CRANE team) could be hugely multiplied through collaborative working. So as you read and reflect please ask yourselves this question...

'Do you have a question, the interest , the expertise, the time or resource access and/or the enthusiasm to contribute to CRANE maximising its utility for patients?'

If the answer is yes (to any part of the above) then please get in contact.

CRANE is continually looking to build on existing interaction and collaboration with the cleft community in the UK ***and*** abroad for the betterment of all.

With the very best of wishes.

Yours sincerely,

The CRANE project team

Glossary and abbreviations

Alveolus / alveolar	The part of the jaw (gum) 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 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 services / Regions	These terms are used interchangeably throughout this report and refer to the hospital / multidisciplinary group that provides cleft surgery and care for children with a cleft; as well as submits data to the CRANE Database, sometimes as part of a wider cleft centre or network. See **Appendix 4** for further information on Regional cleft services.
Cleft surgeon	A surgeon undertaking cleft repair surgery in a region / cleft services.
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
ENT	Ear, nose and throat
ESQ	Experience of Service Questionnaire
FFT	Friends and Family Test
Funnel Plot	A graph that identifies cleft services 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 cleft service. • Each funnel plot is for one outcome, with its values shown on the vertical/Y axis. • The size of the cleft services' 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

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