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Promoting excellence in cleft care

CLEFT REGISTRY & AUDIT NETWORK



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## Cleft Registry and Audit NETWORK Database

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

### 2022 Annual Report

Results of the audit in England, Wales and Northern Ireland for children born with a cleft between January 2000 and December 2021

On behalf of the Cleft Development Group

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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](http://www.clapa.com) to find out more.

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2022 Annual Report: The supplementary tables (Appendices) (Excel Workbook)	December 2022
2022 Annual Report: Responses to outlier process (Appendices)	December 2022
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# Acknowledgements

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- Vanessa Hammond and Jo Shearer for their contribution towards the interpretation of the psychology outcome data at 5 years of age – on behalf of the Psychology CEN.

We would like to thank all at the UK NHS CDG for supporting the outlier process, which was piloted in 2021. In particular, we would like to recognise the extra time and effort it has taken for clinical colleagues to engage and respond so constructively to outlier communications since 2021. 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. The process has been adopted by the Cleft Development Group for use from 2022. It will be introduced in a staged manner up to and including the 2024 CRANE report to allow all teams the necessary time to put in place appropriate resources. The staged implementation also reflects the significant ongoing pressures experienced by clinical services during recovery from the various waves for the COVID-19 pandemic. It is planned that from 2025 the outlier policy will be implemented in full.

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 2022.

This work was funded by the Specialised Services National Business Intelligence and Information Team NHS England and NHS Improvement on behalf of England and Wales, and the Northern Ireland Specialist Services Commissioning Team. It was carried out by the project team of the CRANE Database, which is overseen by the UK NHS Cleft Development Group (CDG)<sup>1</sup>.

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<sup>1</sup> [The supplementary tables](#) 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

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As I sit at the beginning of December, considering the foreword to the third report during my tenure as CRANE Clinical Lead, it is difficult not to let one's mind wander forward to thoughts of the festive season. For many, this is time with family and friends and the potential for a little time to rest and recuperate. After the last few years I think we all deserve it!

I look forward to this time to indulge a love of reading. The bar that I set for measuring whether the festive season has delivered (what I was hoping for) is to read a least one piece each of fiction and non-fiction. While there is the satisfaction that comes with gaining knowledge, it is really getting wrapped up in the story being told in a novel that I enjoy the most.

While this report is 'non-fiction', in many ways it could be considered akin to a novel. Within each of its many strands there lies another chapter in the evolving story of cleft care in the UK. Each section of text, each point on the funnel chart, each bar in the chart, each table is a character or moment in the novel that you must get to know and understand if you are to really benefit from what the 'story' has to say.

I often return to novels I have really enjoyed multiple times. While I believe that I know a story after the first reading, each subsequent journey through the book offers me something else. Whether it be a greater understanding of a character, a richer appreciation of a place or time, a deeper appreciation of the emotions being experienced or, in the best novels, a life lesson learnt, each return has something useful to offer. The same could also be said to be true for the CRANE report. Despite having been involved in the writing of each of the last three reports, each time I return to them they offer me more – as do all those that came before my direct involvement with CRANE.

Recent work in engaging with cleft services and specialties has revealed that most colleagues access the CRANE reports for specific aims. I hope that by encouraging you to spend a little more time exploring other, less familiar parts of the report (and returning to previous reports), you too will develop a greater understanding of each service, a richer appreciation of the challenges and successes in each region, and a deeper understanding of the time, effort and passion that people put into delivering cleft care in the UK. As we all look forward to 2023, may I also ask you to consider looking back at how cleft care has developed in the UK over the last 20 years and reflect on how this has been documented (and hopefully helped) by subsequent CRANE reports.

I hope that this latest report continues to build on CRANE's rich history and influences cleft care both nationally and globally. As the single largest registry and audit of cleft care in the world, CRANE is well positioned not only to tell (with confidence) the true story of cleft care in the UK, but also to serve as a template for open, transparent reporting of cleft care processes and outcomes elsewhere in the world.

I hope you enjoy 'the story' as much as I do being a part of telling it.

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



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

# Executive summary








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. The registry information presented is for children born in 2018 to 2020. The audit outcomes presented are for children born in 2012-2014.



## Registry information

<b>22,724</b>	Children registered in CRANE, over the last 22 years, since 2000.		75% of babies with a cleft palate were diagnosed in the first 24 hours after birth, despite the impact of COVID-19 on services.
<b>834</b>	Registrations in 2021, the second year impacted by the COVID-19 pandemic.		84% of families were referred to a cleft team within 24 hours of birth and 95% of families received contact within 24 hours of referral.
<b>86%</b>	of babies with a cleft were born at term (>37 weeks). This compares to 92% in the general population.	<b>98%</b>	of families with verified consent status agreed to the collection of outcome data.

## Audit outcomes at 5 years of age

 <b>86%</b>	<b>Body mass index</b> of children with a cleft had a healthy BMI. Children most likely to be of normal weight are those with CP, UCLP or BCLP.	 <b>61%</b>	<b>Speech</b> 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.
 <b>61%</b>	<b>Dental health</b> of children with a cleft had no decayed, missing or filled teeth (dmft=0), compared to 77% of their non-cleft peers.	 <b>82%</b>	<b>Psychology</b> of children with a cleft had 'Strengths and Difficulties' scores* in the low/normal range, compared to 90% of their non-cleft peers. Furthermore, 96% of families were screened at least once before the target age of 6 years.
 <b>38%</b>	<b>Facial growth</b> of children with a complete unilateral cleft lip and palate (UCLP) had scores reflecting good dental arch relationships.		

## Development work

<b>Scientific research articles</b>	<b>Conference presentations</b>
We have produced <b>11</b> scientific research articles over the past decade.	We were involved in <b>9</b> different presentations at the International Congress of Cleft Lip, Palate and Related Craniofacial Conditions in 2022.
This work has helped improve the way CRANE conducts and reports on audit, as well as the treatment pathways and experiences of current and future patients.	Our findings were shared with health professionals and patient representatives.
	

For further information visit [www.CRANE-Database.org.uk](http://www.CRANE-Database.org.uk) |  @CRANE\_News



# Key findings and recommendations 2022

Findings	Recommendations	Source
<b>Registry</b>		
<b>Registrations in CRANE</b>		
<ul style="list-style-type: none"> <li>There were 834 registrations in 2021, the second year impacted by the COVID-19 pandemic.</li> <li>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) (24%), unilateral cleft lip and palate (UCLP) (22%) and bilateral cleft lip and palate (BCLP) (10%) (in 2018-20).</li> </ul>	<ol style="list-style-type: none"> <li>CRANE will engage with cleft services and the Nursing Clinical Excellence Network (CEN) to ensure optimum assessment and recording of registrations.</li> <li>CRANE will continue to record cleft births and validate case ascertainment using external datasets.</li> </ol>	Chapter 3, Section 3.1 <b>Indicator<sup>2</sup>: #1</b>
<b>Timely diagnosis</b>		
<ul style="list-style-type: none"> <li>Similar rates of antenatal diagnosis for CL, UCLP and BCLP were observed in 2021, the second year impacted by the COVID-19 pandemic, by comparison with rates in 2018-20.</li> <li>Significant improvement has been observed in the timely detection of CPs, particularly in 2018-20. However, almost 1 in 4 children with CP still have a diagnosis beyond 24 hours from birth.</li> </ul>	<ol style="list-style-type: none"> <li>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.</li> </ol>	Chapter 3, Section 3.2 & 3.3 <b>Indicator: #2</b>
<b>Gestational age and birth weight</b>		
<ul style="list-style-type: none"> <li>The average gestational age of babies born with a cleft in 2018-20 was 38.4 weeks.</li> <li>14% of babies with a known gestational age were born prematurely (&lt;37 weeks' gestation). This compares to 8% in the general population<sup>3</sup>.</li> <li>Children with cleft lip had a higher average birth weight (3,305 g) than those with a cleft affecting the palate (3,120g).</li> </ul>	<ol style="list-style-type: none"> <li>Cleft services should review procedures to ensure processes are in place to record and submit data on gestational age and birth weight. This information is required to track the association of prematurity and low birth weights with clefting.</li> <li>The research community should validate and further investigate the apparent association between cleft-affected pregnancies and prematurity at birth.</li> <li>CRANE will engage with the Royal College of Midwives (RCM) and Royal College of Obstetricians and Gynaecologists (RCOG) to communicate our findings.</li> </ol>	Chapter 3, Section 3.4
<b>Contact with cleft services</b>		
<ul style="list-style-type: none"> <li>84% of families were referred within 24 hours of birth. This rate varied significantly across cleft types (2018-20).</li> <li>95% of families were contacted by a cleft service within 24 hours of referral.</li> </ul>	<ol style="list-style-type: none"> <li>Cleft services should continue to work with referring obstetric, midwifery and neonatal units to improve timeliness of diagnosis and early referrals.</li> <li>Cleft services with high levels of referrals within 24 hours should share their best practice recommendations.</li> </ol>	Chapter 3, Section 3.5 <b>Indicators: #3 &amp; #4</b>

<sup>2</sup> CRANE core indicators are detailed in [the supplementary tables](#).

<sup>3</sup> Birth characteristics in England and Wales: 2019. Available at:

<https://www.ons.gov.uk/peoplepopulationandcommunity/birthsdeathsandmarriages/livebirths/bulletins/birthcharacteristicsinenglandandwales/2019> [Last accessed: 08/12/2022]



<b>CRANE Consent</b>		
<ul style="list-style-type: none"> <li>Consent status was verified for 90% of families registered in 2018-20. This meant they had agreed or declined to their child's outcome data being recorded in CRANE.</li> <li>The verified consent rate has fallen in recent years.</li> <li>Of the families who had reached a decision, rate of agreed consent was 98%.</li> </ul>	<p>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.</p> <p>10. CRANE will continue to work with cleft services and the Nursing CEN to improve consent status verification.</p>	<p>Chapter 3, Section 3.6</p> <p><b>Indicator: #5</b></p>
<b>Outcomes at 5 years</b>		
<b>Child growth</b>		
<ul style="list-style-type: none"> <li>Less than 50% of children have data available for this outcome measure.</li> <li>86% of children with growth data had a healthy body mass index (BMI).</li> <li>According to BMI categorisation, 2% were underweight, 7% overweight and 4% obese (vs 1%, 13% and 10% nationally).</li> </ul>	<p>11. Cleft services should aim to assess children's weight and height at age 5 and improve the reporting of these measures in the CRANE database. This will facilitate more meaningful comparisons between subgroups in the future.</p>	<p>Chapter 4, Section 4.1</p> <p><b>Indicator: #6</b></p>
<b>Dental health</b>		
<ul style="list-style-type: none"> <li>Dental decay was experienced by 39% of children (having at least one decayed, missing or filled tooth (dmft &gt;0)) and 14.5% of children were classified as having extensive caries (dmft &gt;5). These rates increased significantly as cleft type became more involved.</li> <li>The average Treatment Index (rate of treated disease) was 77%, 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.</li> </ul>	<p>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).</p>	<p>Chapter 4, Section 4.2</p> <p><b>Indicators: #7 &amp; #8</b></p>
<b>Facial growth (for children with complete UCLP)</b>		
<ul style="list-style-type: none"> <li>38% of children with a complete UCLP had 5-year-old Index scores reflecting 'good' dental arch relationships.</li> <li>Dental arch relationships have remained stable over time.</li> </ul>	<p>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 the national co-ordinator and assessed using the 5-year index, with results recorded on the CRANE Database.</p> <p>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.</p>	<p>Chapter 4, Section 4.3</p> <p><b>Indicator: #9</b></p>
<b>Speech (for children born with a cleft affecting the palate)</b>		
<ul style="list-style-type: none"> <li>61% achieved speech within the normal range.</li> <li>72% had speech without difficulties resulting from existing or previous structural anomalies.</li> <li>69% had speech without cleft-related articulation difficulties.</li> <li>18% of children had secondary surgery for speech purposes before the age of 5 years.</li> </ul>	<p>15. Information given to parents by cleft services about expected speech outcomes should take into account the child's cleft type.</p> <p>16. The research community should undertake to develop risk-adjustment models for analysing speech outcomes among children with a cleft.</p>	<p>Chapter 4, Section 4.4</p> <p><b>Indicators: #10, #11 &amp; #12</b></p>
<b>Psychology screening</b>		
<ul style="list-style-type: none"> <li>98% of families were screened at least once before the target age of 6 years.</li> <li>95% were seen by a psychologist and a psychosocial screen was completed at age 5 (Tiers of Involvement Measure 1 to 4, referred to as 1a+).</li> </ul>	<p>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).</p>	<p>Chapter 4, Section 4.5</p>

<ul style="list-style-type: none"> <li>18% of children born with a cleft had 'high' or 'very high' Strengths and Difficulties Questionnaire scores. These rates are higher than the population norms.</li> </ul>		<b>Indicators:</b> <b>#13 &amp; #14</b>
<b>All outcomes at 5 years</b>		
<p>An outlier process was applied to children's outcomes at 5 years of age. Cleft services were permitted to provide a response to being identified as a positive or negative outlier.</p>	<p>18. All cleft services should work together to explore reasons for variation in outcomes at 5 years of age.</p>	<p>Chapter 4</p>
<b>Database development work</b>		
<b>Peer-reviewed publications and presentations</b>		
<ul style="list-style-type: none"> <li>Three scientific articles were published over the last year.</li> <li>CRANE was involved in 9 different oral presentations at the International Congress of Cleft Lip, Palate, and Related Craniofacial Conditions in July 2022.</li> </ul>	<p>19. Collaboration is key to sharing CRANE data and facilitating research that informs clinicians, families and policy makers.</p>	<p>Chapter 5, Section 5.1</p>
<b>Improving accuracy and use of data</b>		
<ul style="list-style-type: none"> <li>Collection of data on syndromes and additional diagnoses via new data fields within the CRANE Database provides more flexibility when performing analyses.</li> <li>A collaborative study with the Cleft Collective found that concordance of cleft phenotype classification between two data sources (CRANE Database and the Cleft Collective database) decreases when more detail, such as laterality and completeness, is included.</li> </ul>	<p>20. Clinical Excellence Networks (CEN) should liaise with CRANE to determine which syndromes/additional diagnoses should potentially be included/excluded when reporting each cleft-related outcome.</p> <p>21. Further work to validate cleft classification using LAHSAL/LAHSAL code is required.</p>	<p>Chapter 5, Section 5.2</p>
<b>Developmental work to improve our understanding of clefting and determinants of cleft-related outcomes</b>		
<ul style="list-style-type: none"> <li>An analysis of CRANE-HES linked data revealed that 39% of children with a cleft have additional congenital malformations. This rate varies according to cleft type, and is highest among those with CP (53%) and lowest among those with CL (22%).</li> <li>Sex, cleft type and extent of hard palate involvement have a significant impact on speech outcomes at 5 years of age, particularly in terms of achieving 'normal' speech (Standard 1) and speech without significant cleft-related articulation difficulties (Standard 3).</li> <li>Cleft type, ethnicity and socio-economic status were associated with dental health and access to dental care and treatment.</li> <li>CRANE has explored educational outcomes in children born with non-syndromic cleft using linkage to the National Pupil Database. Children with a cleft had lower educational attainment than the general population and the educational attainment gap was consistent at age 5, 7 and 11 years.</li> </ul>	<p>22. The introduction of standardised screening for some cleft subgroups should be considered given the high rate of additional malformations.</p> <p>23. Cleft services should establish good links with paediatric and genetic services due to the high prevalence of additional malformations occurring alongside clefts.</p> <p>24. Evidence for risk adjustment based on patient factors has been provided for speech and dental outcomes to allow accurate comparisons across cleft services.</p> <p>25. Further work to understand which groups of children improve their educational attainment and which do not will enable targeting of appropriate resources to reduce the educational attainment gap.</p>	<p>Chapter 5, Section 5.3</p>
<b>Data quality – throughout report</b>		
<ul style="list-style-type: none"> <li>There was significant variation in data completeness for registrations and also for specialty-specific outcomes across the period analysed for this report.</li> <li>Recovery from the COVID-19 pandemic has significantly affected some specialties' ability to submit 5-year-old outcome data.</li> </ul>	<p>26. Cleft services should pay particular attention to assessing outcomes and reporting these to CRANE.</p> <p>27. Cleft services, Clinical Excellence Networks (CENs) and CRANE should work together to identify and overcome barriers to collecting and submitting data.</p>	<p>All</p>

# 1. Introduction

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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<sup>4</sup>. 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 2023 once GDPR-compliant data sharing agreements are in place. The geographical representation of the cleft services is detailed in [the supplementary tables](#).

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<sup>5</sup> for children with a cleft lip and/or palate born in England, Wales and Northern Ireland between 1 January 2000 and 31 December 2021, 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 2018-2020. Summary information on 2021 births is also provided. Data for 2020 and 2021 births should be interpreted with caution given the highly variable impact the COVID-19 pandemic has had on the ability of clinical services 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 2012-2014. Summary information on 2015 births is also provided. Data for 2014 and 2015 births should be interpreted with caution given the highly variable impact the COVID-19 pandemic has had on the ability of clinical services to complete clinical audit and process the data.
- **Database development work:** This section details the 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 [the supplementary tables](#).

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

<sup>5</sup> Registered in the CRANE Database by 27 June 2022.

## 2. Methods

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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. This chapter provides a summary of these two data sources. Full details of the methods employed are given in the supplementary [Methods document published alongside this report](#).

### 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 [the supplementary tables](#)).

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<sup>6</sup> to collect additional information, including cleft-related outcomes, 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 27 June 2022 are 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. For full details of the methods used within this report, please see the CRANE Annual Report Methods 2022 document, available online alongside this Annual Report (<https://www.crane-database.org.uk/reports/crane-database-2022-annual-report/>).

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<sup>6</sup> Confidential Advisory Group (CAG) Section 251 Approval <https://www.hra.nhs.uk/approvals-amendments/what-approvals-do-i-need/confidentiality-advisory-group/> Last accessed 12/12/2022.

## Missing data

Missing data have been excluded from the denominators presented in all tables, figures and supplementary tables of this report, with the exception of tables and figures relating to data completeness (see [the supplementary tables](#) 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 [the supplementary tables](#) 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

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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 2018 and 31 December 2020 (consistent with a rolling 3 years, as for other sections of this report).
- **Cohort 2.** Children born between 01 January 2021 and 31 December 2021; 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 are 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: 2018-2020. Plus summary information on 2021 births <sup>7</sup>
	Denominators	<ul style="list-style-type: none"> <li>• 2,838 (2018-2020) and 834 (2021) children registered.</li> <li>• 2,614 (2018-2020) and 775 (2021) children with a specified cleft type.</li> <li>• 2,806 (2018-2020) and 830 (2021) children with a record of sex.</li> </ul>
	Numerator	Number of children with each patient characteristic
	Data completeness	<ul style="list-style-type: none"> <li>• 92% of 2,838 registrations had a specified cleft type (2018-2020).</li> <li>• 99% of 2,838 registrations had a specified sex (2018-2020).</li> </ul>
	Countries	England, Wales and Northern Ireland
	Indicator	#1 <sup>8</sup>
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?	<ul style="list-style-type: none"> <li>• 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) (24%), unilateral cleft lip and palate (UCLP) (22%) and bilateral cleft lip and palate (BCLP) (10%), respectively (2018-2020).</li> <li>• 56% of registrations were boys. As expected from historical experience, they were more likely to have a CL, UCLP or BCLP than registered girls (2018-2020).</li> </ul>	
Recommendations	Cleft services should aim to: <ul style="list-style-type: none"> <li>• register all children with a cleft in the CRANE database,</li> <li>• specify cleft type and sex for every child registered.</li> </ul> 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 22,724 children born in England, Wales and Northern Ireland with cleft lip and /or palate over the last 22 years<sup>9</sup>. Among them, 2,838 children were registered between 2018 and 2020, and 834 children were registered in 2021<sup>10</sup>.

Figure 3.1 displays the number of CRANE registrations in 2021 for each cleft service – within the second year impacted by the COVID-19 pandemic – by comparison with the yearly average across 2018-20 and the national averages for both cohorts. Eleven out of thirteen cleft services registered a lower number of children born with a cleft in 2021 in comparison to the yearly average number of registrations across the 2018-20 period. Raw data in [the supplementary tables](#) show the distribution of registrations for each cleft service, in 2018-20 and 2021.

<sup>7</sup> Affected by the COVID-19 pandemic and the lockdown that ensued.

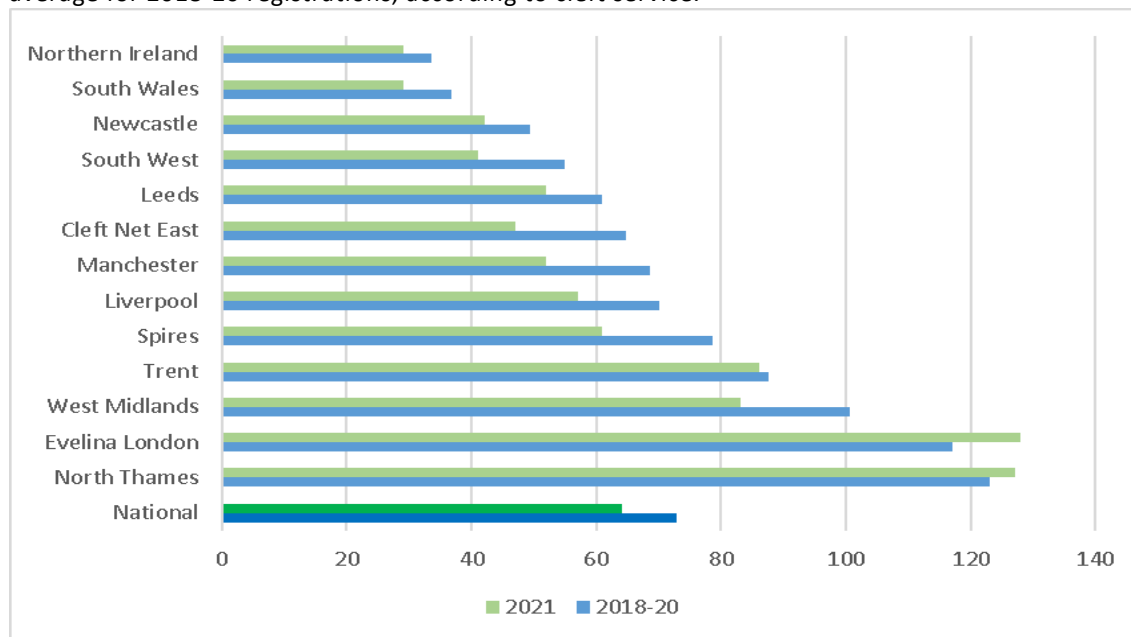
<sup>8</sup> CRANE core indicators are detailed in [the supplementary tables](#).

<sup>9</sup> Cohorts include all patients registered in the CRANE Database between 1 January 2000 and 31 December 2021, inclusive.

<sup>10</sup> Eleven children born and registered in the CRANE Database in 2018-20 were reported to have died within the first 12 months after birth; there were no deaths reported for children born and registered in 2021 (0.3% 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 children born in 2021 who were registered on CRANE, by comparison with the yearly average for 2018-20 registrations, according to cleft service.



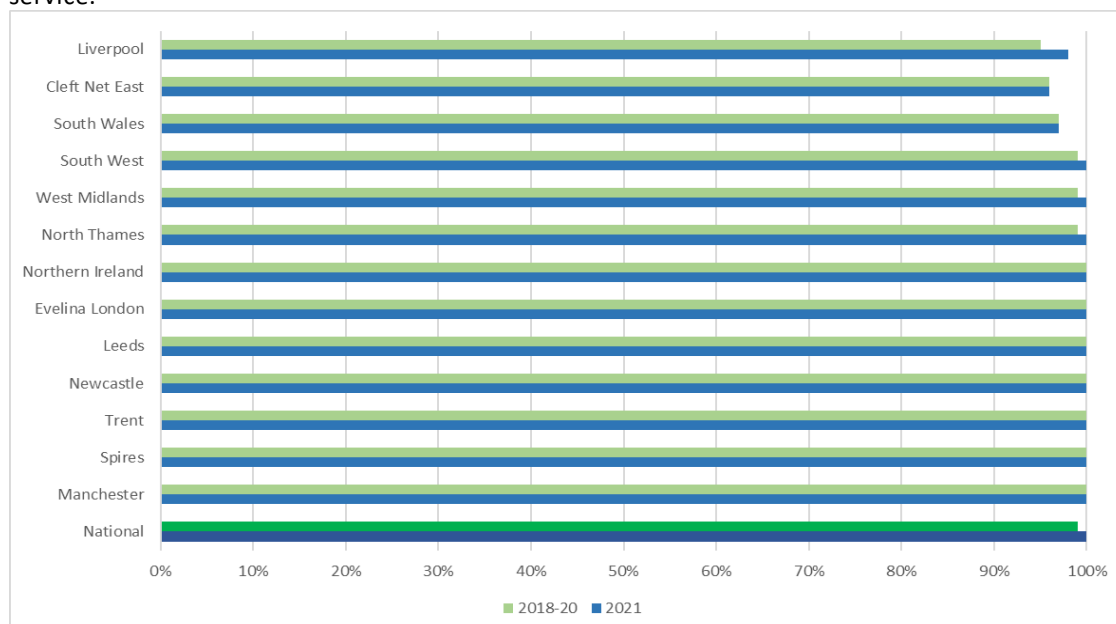
**Note:** Children registered in the CRANE Database between 1 January 2018 and 31 December 2021, by the 27 June 2022.

### 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 2018-20, compared to 2021 registrations and national averages for the two periods.

- Recording of sex data remains high nationally, with 99% of children in the 2018-20 cohort (95%-100% range across cleft services) and 2021 cohort (96%-100% range across cleft services) having their sex recorded in CRANE.
- 92% of all registrations in 2018-20 had their cleft type specified in CRANE (72%-100% range), compared to 93% among 2021 registrations (67%-100% range).

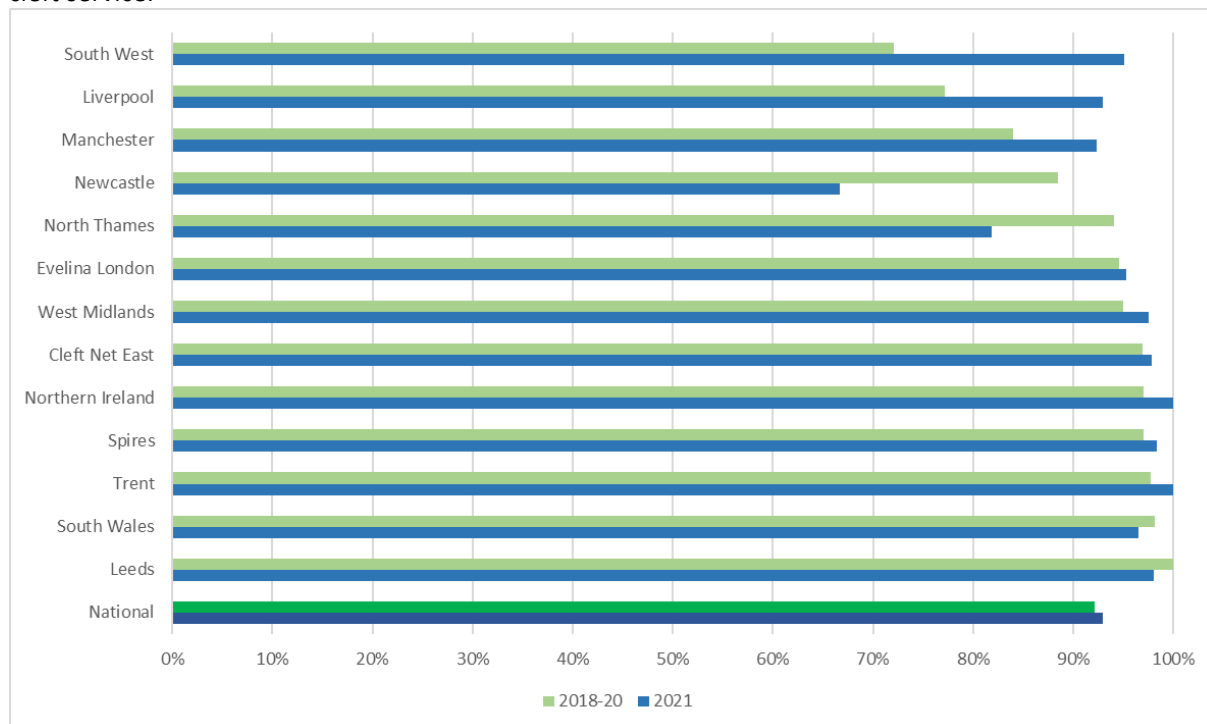
**Figure 3.2.** Percentage of CRANE-registered children with sex reported, according to birth cohort and cleft service.



**Note:** Children registered in the CRANE Database between 1 January 2018 and 31 December 2021, by the 27 June 2022.

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 Chapter 5 Database development work), cleft services should seek to report sex and cleft type for all children registered in the CRANE Database to ensure appropriate risk stratification / adjustment is possible in the future.

**Figure 3.3.** Percentage of CRANE-registered children with cleft type specified, according to birth cohort and cleft service.



**Note:** Children registered in the CRANE Database between 1 January 2018 and 31 December 2021, by 27 June 2022.

### 3.1.3. Registrations by cleft type and sex

Among children registered between 2018 and 2020, inclusive, with a recorded cleft type (n=2,614), CP was the most common of the four cleft types<sup>11</sup>, representing 44% of registrations, followed by CL (24%), UCLP (22%) and BCLP (11%). Raw data in [the supplementary tables](#) show the distribution of cleft type by each cleft service, for registrations with a specified cleft type, in 2018-20 and 2021.

Of the children registered and diagnosed with UCLP (n=565) or BCLP (n=276) in 2018-20, 46% and 56%, respectively, were complete clefts involving the hard and soft palate. For children diagnosed in 2021 with UCLP (n=184) and BCLP (n=63), 36% and 57%, respectively, were complete clefts involving the hard and soft palate.

Overall, 56% of children registered between 2018 and 2020 were boys. Raw data in [the supplementary tables](#) show the distribution of sex by cleft services for registrations in 2018-20, as well as for 2021 registrations. Among 2018-20 registrations, boys were significantly more likely to have a CL (66%), UCLP (67%) or BCLP (70%) than girls (p<0.001). For the same period, CP was significantly more prevalent among girls (58%) 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.

<sup>11</sup> Cleft type is defined according to reported LAHSAL codes or, where LAHSAL has not been reported (for 8% of children registered between 01 January 2018 and 31 December 2020), 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: 2018 to 2020. Plus summary information on 2021 births.
	Denominator	2,709 (2018-2020) and 804 (2021) children with a recorded diagnosis time
	Numerator	Number of children diagnosed at each time point
	Exclusion	Children without a recorded diagnosis time
	Data completeness	95% of 2,838 CRANE-registered children had a recorded diagnosis time (2018-2020).
	Countries	England, Wales and Northern Ireland
	Indicator	#2 <sup>12</sup>
Standard	<ul style="list-style-type: none"> <li>• 100% of children with a cleft should have a recorded diagnosis time.</li> <li>• 100% of clefts should be diagnosed in a timely manner, either antenatally (for clefts involving the lip) or within 24 hours of birth.</li> </ul>	
What did we find?	<p><b>All cleft types</b></p> <ul style="list-style-type: none"> <li>• Rates of <b>antenatal diagnoses</b> for all cleft types during the second year impacted by the COVID-19 pandemic (2021) approached rates in the three years prior (2018-20) – 43% in 2021 and 45% in 2018-20.</li> <li>• Rates of <b>timely diagnoses (antenatally or within 24 hours of birth)</b> of all cleft types during the second year impacted by the COVID-19 pandemic (2021) were similar to rates in the three years prior (2018-20) – 85% in 2021 and 87% in 2018-20.</li> </ul> <p><b>Clefts involving the lip</b></p> <ul style="list-style-type: none"> <li>• Rates of <b>antenatal diagnoses</b> for clefts involving the lip (CL, UCLP, BCLP) during the second year impacted by the COVID-19 pandemic (2021) approached rates in the three years prior (2018-20) – 76% in 2021 and 80% in 2018-20.</li> <li>• Rates of <b>timely diagnoses (antenatally or within 24 hours of birth)</b> for clefts involving the lip (CL, UCLP, BCLP) during the second year impacted by the COVID-19 pandemic (2021) were similar to rates in the three years prior (2018-20) – 97% in 2021 and 98% in 2018-20.</li> </ul>	
Recommendations	<ul style="list-style-type: none"> <li>• CRANE will seek to collaborate further with the clinical workforce to identify barriers to recording timing of diagnosis for all registrations in CRANE.</li> <li>• 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.</li> </ul>	

### 3.2.1. Data completeness

Of the 2,838 children born and registered in CRANE between 2018 and 2020, 95% had a recorded diagnosis time (range: 67% - 100% between cleft services). Of the 834 children born and registered in CRANE in 2021 – during the second year impacted by the COVID-19 pandemic – 96% (n=804) had a recorded diagnosis time (range: 59% - 100% between cleft services). Cleft services should be commended for their commitment to recording this information, particularly during the years impacted by the COVID-19 pandemic.

### 3.2.2. Timely diagnosis of all cleft types

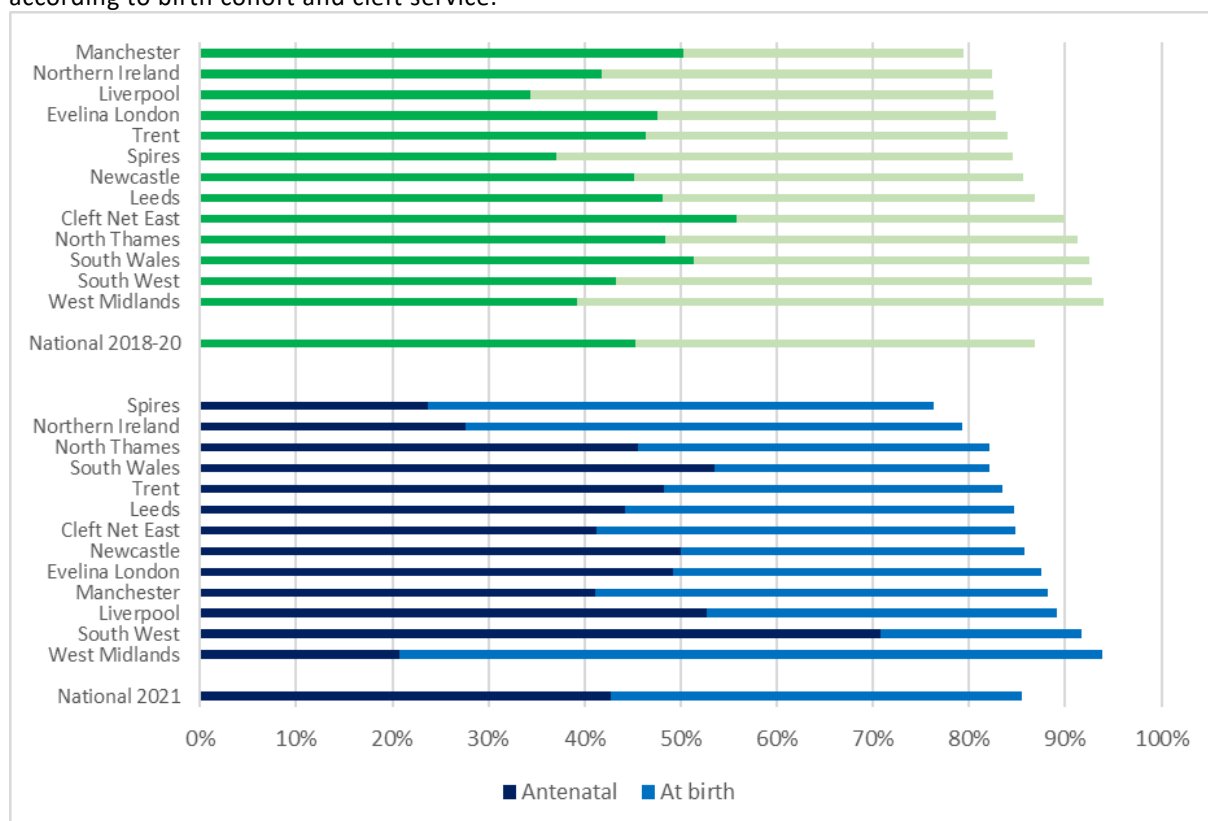
The variation in diagnoses of all clefts by cleft service was explored and is presented in Figure 3.4. In 2018-20, 45% of clefts were diagnosed during antenatal screening and 42% were diagnosed at birth, leaving 13% diagnosed late according to the National Standard. Figure 3.4 also shows that the percentage of clefts diagnosed

<sup>12</sup> CRANE core indicators are detailed in [Appendix 3](#).

in a timely manner in 2018-20 varied by cleft service, ranging from 79% (Manchester) to 94% (West Midlands). This variation suggests that practice varies between maternity units, with some better than others at timely identification of clefts.

Figure 3.4 also shows that in 2021, 43% of clefts were diagnosed during antenatal screening and 43% were diagnosed at birth, leaving 14% diagnosed late according to the National Standard (similar to 2017-19 rate). The percentage of clefts diagnosed in a timely manner in 2021 varied by cleft service, ranging from 76% (Spires) to 94% (West Midlands). This variation suggests that practice varied considerably between maternity units during 2021, with some achieving high rates of identification of clefts in a timely manner despite the challenges presented by the second year of the COVID-19 pandemic.

**Figure 3.4.** Percentage of CRANE-registered children who received a timely diagnosis (antenatal or at birth), according to birth cohort and cleft service.



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

The supplementary tables show the breakdown of timing of diagnoses reporting for each cleft service, for 2018-20 as well as 2021 births.

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

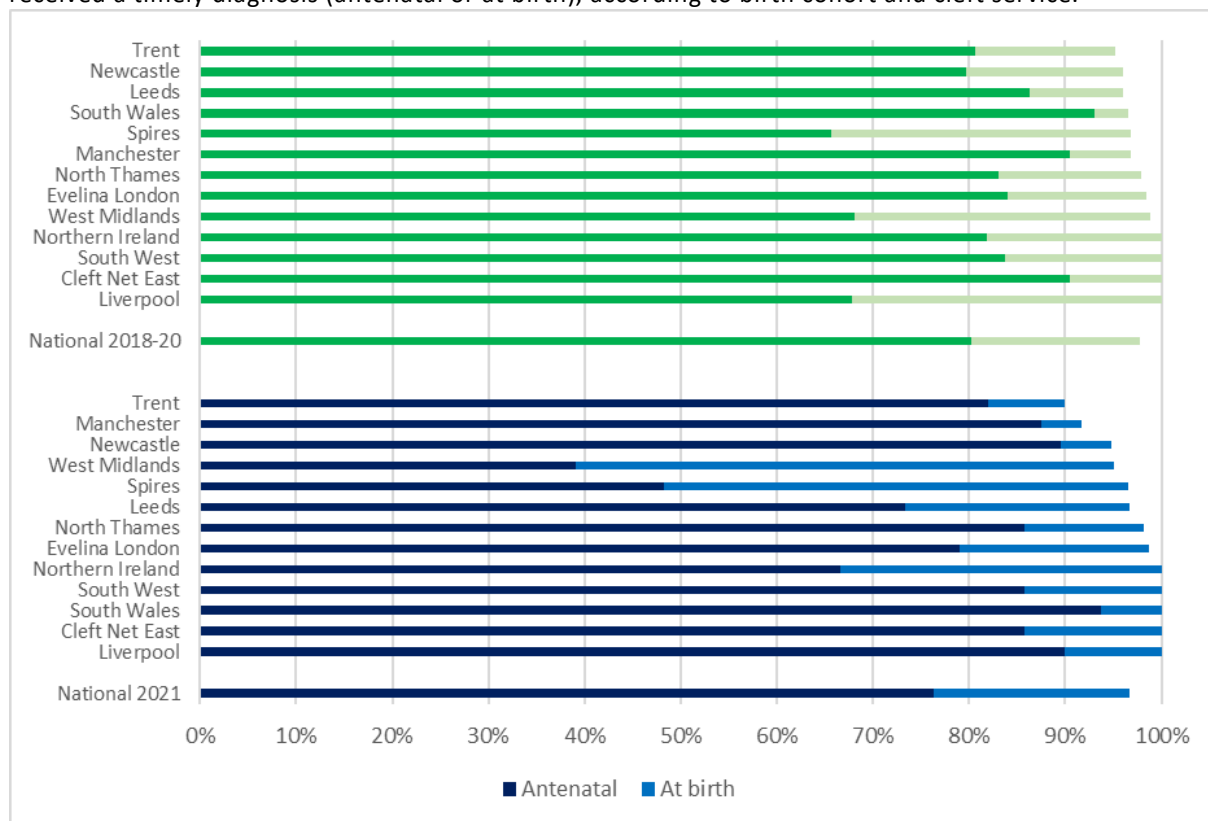
The variation in diagnoses of clefts involving the lip by cleft service were explored and are presented in Figure 3.5. In 2018-20, 80% of clefts involving the lip were diagnosed during antenatal screening and 18% were diagnosed at birth, leaving 2% diagnosed late according to the National Standard. Figure 3.5 also shows that the percentage of clefts involving the lip diagnosed in a timely manner in 2018-20 varied by cleft service, ranging from 95% (Trent) to 100% (Liverpool, Cleft Net East, South West, and Northern Ireland).

Figure 3.5 also shows that in 2021, 76% of clefts involving the lip were diagnosed during antenatal screening and 21% were diagnosed at birth, leaving 3% diagnosed late according to the National Standard (similar to 2018-20

rate). The percentage of clefts involving the lip diagnosed in a timely manner in 2021 varied by cleft service, ranging from 90% (Trent) to 100% (for 5 of 13 cleft services).

These figures show that most services achieved high rates of identification of clefts involving the lip in 2021, in a timely manner and consistent with 2018-20, despite the challenges presented by the second year of the COVID-19 pandemic. However, only 39% of clefts involving the lip were diagnosed antenatally in the West Midlands, compared to 68% for 2018 to 2020 births. Spire also had lower rates of antenatal diagnosis in 2021 (48%) compared to the preceding three years (66%).

**Figure 3.5.** Percentage of CRANE-registered children with a cleft involving their lip (CL, UCLP and BCLP) who received a timely diagnosis (antenatal or at birth), according to birth cohort and cleft service.



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

The supplementary tables show the breakdown of timing of diagnoses reporting for each cleft service, for 2018-20 as well as 2021 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. Prompt diagnosis ensures that children 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	<ul style="list-style-type: none"> <li>• 10 years: 2012 to 2021 to explore trends over time</li> <li>• 3 years: 2018 to 2020 to explore regional differences</li> </ul>
	Denominator	<ul style="list-style-type: none"> <li>• For 2012-21 births: 4,106 children with cleft palate alone and a recorded diagnosis time</li> <li>• For 2018-20 births: 1,113 children with cleft palate alone and a recorded diagnosis time</li> </ul>
	Numerator	Number of children diagnosed at each time point with cleft palate alone
	Exclusions (not mutually exclusive)	<ul style="list-style-type: none"> <li>• Children with CL, UCLP, BCLP or an unspecified cleft type</li> <li>• Children without a recorded diagnosis time</li> </ul>
	Data completeness	<ul style="list-style-type: none"> <li>• 95% of children born 2012-21 with CP (n=4,341) had a recorded diagnosis time</li> <li>• 97% of children born 2018-20 with CP (n=1,146) had a recorded diagnosis time</li> </ul>
	Countries	England, Wales and Northern Ireland
	Indicator	#2 <sup>13</sup>
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, at least 1 in 4 children with CP in 2021 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)<sup>14</sup>. 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 ( $p=0.011$ ), with rates increasing from 66% for children born in 2012 to 74% 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*'<sup>15</sup> in October 2014 (and its formal launch – along with a training module – in 2015). This guidance was drafted in response to the CRANE findings and in collaboration with key partners, including CRANE.

<sup>13</sup> CRANE core indicators are detailed in [the supplementary tables](#).

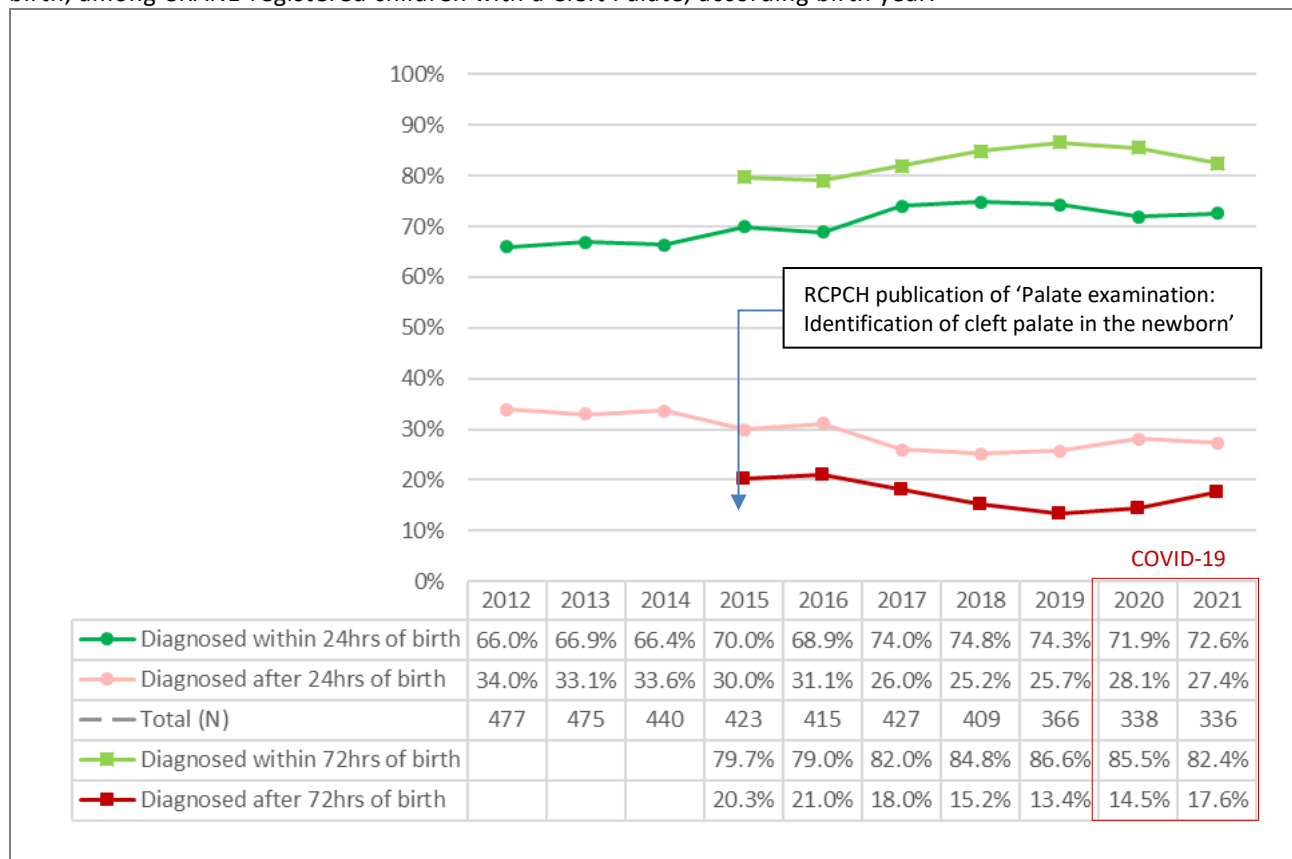
<sup>14</sup> Bannister P. Management of infants born with a cleft lip and palate. Part 1. *Infant*, 2008. 4(1): p. 5-8.

<sup>15</sup> Published October 2014 <https://www.rcpch.ac.uk/resources/palate-examination-identification-cleft-palate-newborn-best-practice-guide>

The last couple of years have seen a slight deterioration in the timeliness of CP diagnosis (71.9% and 72.6%, respectively). These are years affected by the COVID-19 pandemic. We will continue to monitor trends in CP diagnosis and hope that rates return to levels observed in 2019.

Despite the encouraging improvements in timely diagnosis rates, at least 1 in 4 children with CP continue to have a diagnosis beyond 24 hours from birth. The years of the COVID-19 pandemic have arrested the year--year improvement in timely diagnoses; this may reflect stresses on clinical services in light of the pandemic.

**Figure 3.6.** Rates of timely diagnoses (antenatal or within 24 hours of birth), and diagnoses within 72 hours of birth, among CRANE-registered children with a Cleft Palate, according birth year.



**Note:** Children missing diagnosis time are excluded.

Current RCPCH guidance is that full newborn examinations should be undertaken within 72 hours of birth<sup>16</sup>. 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<sup>17</sup> between 2016 and 2019, where rates increased from 79% to 87% (p=0.031). 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 (4% versus 7%). It is encouraging to see this improvement within the parameters of the formal full newborn examination; however, the cleft clinical community still supports an emphasis to improve earlier diagnosis within the first 24 hours from birth.

CRANE will continue to encourage work with partners involved in development of the original palate examination guidance to see if it is practical to include formal palate examination within current guidance on

<sup>16</sup> National Institute for Health and Care 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>

<sup>17</sup> The option to collect this information was added to the CRANE Database in May 2014.



infant feeding within the first 24 hours from birth.

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

Table 3.1 shows that, overall, 2% of CPs were diagnosed during antenatal screening and 71% were diagnosed at birth, leaving 27% diagnosed late according to the National Standard in the years 2018-20. 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 74% of CPs were diagnosed in a timely manner (antenatal or at birth), rates varied by cleft service, ranging from 58% (Manchester) to 90% (South Wales). This means the percentage of children diagnosed late according to the National Standard ranged from 10% to 42% 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 than others at identifying a cleft of the palate during the first 24 hours after birth, as advocated by cleft services nationally.

**Table 3.1.** Timing of diagnosis among CRANE-registered children born with a cleft palate in 2018 to 2020, according to cleft service.

Cleft service	Time of diagnosis in relation to birth n (%)							All
	Antenatal	At birth	< 72 hours	≤1 week	≤1 month	≤6 months	>6 months	
Newcastle	2 3.6%	38 69.1%	4 7.3%	2 3.6%	5 9.1%	4 7.3%	0 0%	55
Leeds	0 0.0%	61 75.3%	8 9.9%	4 4.9%	3 3.7%	3 3.7%	2 2.5%	81
Liverpool	1 1.3%	51 65.4%	15 19.2%	4 5.1%	5 6.4%	2 2.6%	0 0%	78
Manchester	0 0.0%	45 58.4%	17 22.1%	7 9.1%	6 7.8%	2 2.6%	0 0%	77
Trent	3 2.7%	76 67.9%	16 14.3%	5 4.5%	4 3.6%	5 4.5%	3 2.7%	112
West Midlands	0 0.0%	96 86.5%	6 5.4%	4 3.6%	3 2.7%	2 1.8%	0 0%	111
Cleft Net East	1 1.4%	50 71.4%	10 14.3%	7 10%	0 0%	2 2.9%	0 0%	70
North Thames	7 5.0%	109 77.9%	10 7.1%	5 3.6%	2 1.4%	7 5%	0 0%	140
Spire	2 2.0%	69 68.3%	15 14.9%	5 5%	2 2%	6 5.9%	2 2%	101
South Wales	2 4.1%	42 85.7%	2 4.1%	0 0%	1 2%	2 4.1%	0 0%	49
South West	2 3.9%	42 82.4%	4 7.8%	1 2%	2 3.9%	0 0%	0 0%	51
Evelina London	7 4.9%	87 60.4%	22 15.3%	10 6.9%	10 6.9%	7 4.9%	1 0.7%	144
Northern Ireland	0 0.0%	28 63.6%	3 6.8%	1 2.3%	4 9.1%	0 0%	8 18.2%	44
<b>All</b>	<b>27 2.4%</b>	<b>794 71.3%</b>	<b>132 11.9%</b>	<b>55 4.9%</b>	<b>47 4.2%</b>	<b>42 3.8%</b>	<b>16 1.4%</b>	<b>1,113</b>

**Note:** Recording of 'timing of diagnosis' within 72 hours commenced in May 2014 to align CRANE data collection with NIPE standards<sup>18</sup>.

It is worth noting that the percentage of children diagnosed within 72 hours of birth ranged from 71% (Northern Ireland) to 94% (South West). This variation between rates of diagnoses within 72 hours of birth and after 72 hours of birth was also found to be statistically significant ( $p = 0.011$ ). Diagnosis time among CP patients born over the second year impacted by the COVID-19 pandemic, 2021, are presented in [the supplementary tables](#).

Overall, the most recent CRANE data show an encouraging trend with regard 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.

<sup>18</sup> 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: 2018-2020. Plus summary information on 2021 births <sup>19</sup>
	Denominators	<ul style="list-style-type: none"> <li>• 1,120 (2018-2020) and 317 (2021) children with a recorded gestational age</li> <li>• 1,109 (2018-2020) and 316 (2021) children registered with a recorded birth weight</li> <li>• 943 (2018-2020) born at term (<math>\geq 37</math> weeks) with a recorded birth weight</li> </ul>
	Numerators	<ul style="list-style-type: none"> <li>• Children born prematurely (<math>&lt; 37</math> weeks' gestation)</li> <li>• Children born at term with a low birth weight (<math>&lt; 2500</math> g)</li> <li>• Children born at term with a high birth weight (<math>\geq 4000</math> g)</li> </ul>
	Exclusions (not mutually exclusive)	<ul style="list-style-type: none"> <li>• Children without consent to data collection</li> <li>• Children without a recorded gestational age</li> <li>• Children without a recorded birth weight</li> </ul>
	Data completeness	<ul style="list-style-type: none"> <li>• 56% of 1,990 eligible consented children had a recorded gestational age and birth weight.</li> <li>• 0.1% had a reason why gestational age or birth weight were not collected.</li> <li>• 44% were missing data or a reason for not collecting data.</li> </ul>
	Countries	England, Wales and Northern Ireland
Benchmark	<ul style="list-style-type: none"> <li>• 100% of eligible children should have a recorded gestational age and birth weight (or a valid reason it was not collected).</li> <li>• Among babies born in 2019 in the general population of England &amp; Wales, 8% were born prematurely. Of those born at term, 7% had a low birth weight (LBW) and 11% of them had a high birth weight (HBW)<sup>20</sup>.</li> </ul>	
What did we find?	<ul style="list-style-type: none"> <li>• The average gestational age was 38.4 weeks (2018-2020).</li> <li>• The percentage of premature births among children born with a cleft (with gestational age reported) was 14% and is higher than in the general population (8% of premature births) (2018-2020).</li> <li>• The average birth weight was 3,166 g. Children with CL had a higher average birth weight (3,305 g) than those with a CP (3,120 g), UCLP (3,128 g) or BCLP (3,141 g) (2018-2020).</li> <li>• Among children born at term (<math>\geq 37</math> weeks), 6% weighed <math>&lt; 2500</math> g (LBW) and 9% weighed <math>\geq 4000</math> g (HBW) (2018-2020).</li> </ul>	
Recommendations	<ul style="list-style-type: none"> <li>• 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.</li> <li>• 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.</li> <li>• The research community should validate and further investigate the higher percentage of premature births among children diagnosed with a cleft.</li> <li>• CRANE will engage with the Royal College of Midwives (RCM) and Royal College of Obstetricians and Gynaecologists (RCOG) to communicate our findings.</li> </ul>	

<sup>19</sup> Affected by the COVID-19 pandemic and the lockdown that ensued.

<sup>20</sup> Birth characteristics in England and Wales: 2019. Available at:

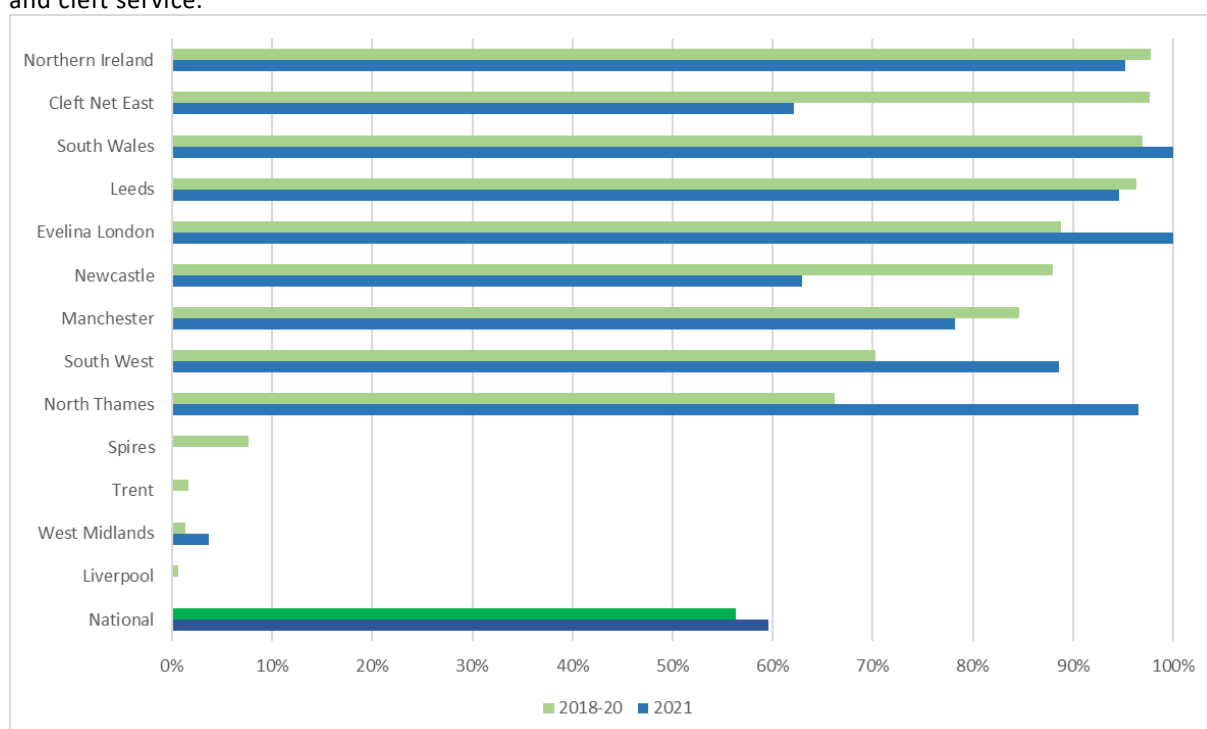
<https://www.ons.gov.uk/peoplepopulationandcommunity/birthsdeathsandmarriages/livebirths/bulletins/birthcharacteristicsinenglandandwales/2019> [Last accessed: 12/12/2022]

### 3.4.1. Data completeness for gestational age

Among children born in 2018-2020 whose families had consented to their children's data being collected in the CRANE Database (n=1,990), 56% had gestational age reported to CRANE<sup>21</sup>. For children registered in 2021 (n=532 children), the second year impacted by the COVID-19 pandemic, 60% had a recorded gestational age. Figure 3.7 shows the variation in the percentage of consented children with a recorded gestational age for 2018-20 (1%-98% range) and for 2021 (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 prematurely 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 gestational age reported, according to birth cohort and cleft service.



**Note:** All children registered in CRANE between 01 January 2018 and 31 December 2021, by 27 June 2022. Children without consent for data collection at birth are excluded.

### 3.4.2. Gestational age

Among babies born between 2018 and 2020 with known gestational age at birth (n=1,120), 14% were born prematurely (< 37 weeks' gestation), compared to 9% in the 2021 cohort. Raw data in [the supplementary tables](#) show the variation in the percentage of children born prematurely in the 2018-20 cohort, compared to 2021 registrations, for each cleft service. It is higher than the 8% national average reported for premature births in England and Wales<sup>22</sup> in 2019. The average gestational age for the 2018-20 cohort was 38.4 weeks (95% confidence interval: 38.3 to 38.6 weeks). It should be noted that the gestational age recorded in CRANE may not

<sup>21</sup> Among the remaining 44% of consented children without gestation data in 2018-20, two did not have this outcome collected as they were transferred out or unavailable for data collection ('other reason' in CRANE).

<sup>22</sup> Birth characteristics in England and Wales: 2019 - Available at: <https://www.ons.gov.uk/peoplepopulationandcommunity/birthsdeathsandmarriages/livebirths/bulletins/birthcharacteristicsinenglandandwales/2019> [Last accessed: 12/12/2022]

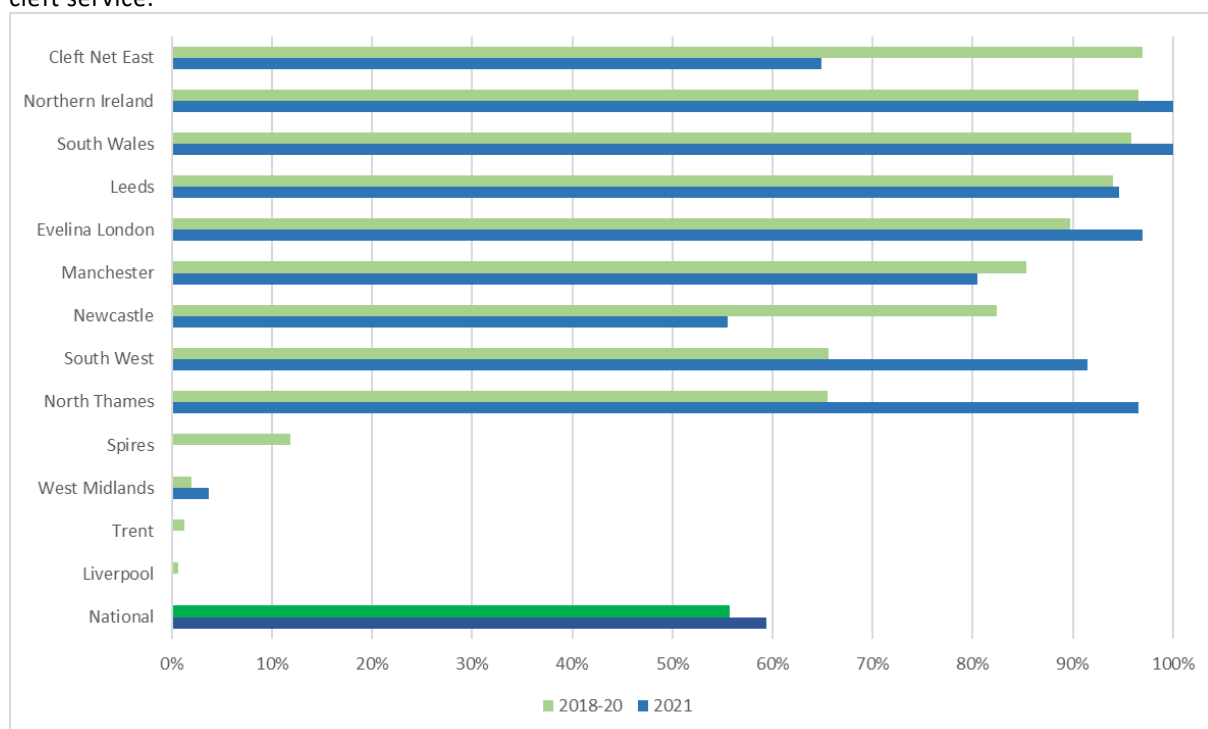
be representative of all babies born with a cleft lip and/or palate as almost half of all consented children were missing information on their gestational age at birth.

### 3.4.3. Data completeness for birth weight

Among children born in 2018-2020 whose families had consented to their children’s data being collected in the CRANE Database (n=1,990), 56% had birth weight recorded<sup>23</sup>. For children registered in 2021 (n=532 children), the second year impacted by the COVID-19 pandemic, 59% had a recorded birth weight. Figure 3.8 shows the variation in the percentage of consented children with a recorded birth weight for 2018-20 (1%-97% range) and for 2021 (0%-100% 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 birth weight reported, according to birth cohort and cleft service.



**Note:** All children registered in CRANE between 01 January 2018 and 31 December 2021, by 27 June 2022. Children without consent for data collection at birth are excluded.

### 3.4.4. Birth weight

Among consented cases with known birth weight, the overall average birth weight for 2018-2020 registrations (n=1,109) was 3,166 g (95% CI: 3,126 g to 3,206 g), compared to 3,307 g (95% CI: 3,238 g to 3,376 g) among 2021 registrations (n=316). For the 2018-2020 cohort, average birth weight was significantly lower among children diagnosed with a CP (3,120 g, 95% CI: 3,056 g to 3,184 g) (p-value<0.001), UCLP (3,128 g, 95% CI: 3,045 g to 3,212 g) (p-value=0.003) and BCLP (3,141 g, 95% CI: 3,020 g to 3,263 g) (p-value=0.024), when compared to average birth weight of those diagnosed with a CL (3,305 g, 95% CI: 3,230 g to 3,380 g). It should be noted that

<sup>23</sup> Among the remaining 44% of consented children without gestation data in 2018-20, two did not have this outcome collected as they were transferred out or unavailable for data collection ('other reason' in CRANE).

the birth weight recorded in CRANE may not be representative of all babies born with a cleft lip and/or palate as 44% of all consented children from 2018-20 registrations were missing this information.

A total of 943 of 1,109 consented children with known birth weight between 2018 and 2020 were born at term (85%). Among them, 6% and 9% were born with low birth weight (<2,500 g) and high birth weight ( $\geq$ 4,000 g), respectively, compared to 7% and 11% of children with low birth weight and high birth weight, respectively, in the general population<sup>24</sup>. Raw data in **the supplementary tables** show the variation in the distribution of birth weight category among children born at term in the 2018-20 and 2021 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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<sup>24</sup><https://www.ons.gov.uk/peoplepopulationandcommunity/birthsdeathsandmarriages/livebirths/bulletins/birthcharacteristicsinen glandandwales/2016/pdf>

### 3.5. Contact with cleft services

Cohort summary	Data source	The CRANE Database
	Birth years	Three years: 2018-2020. Plus summary information on 2021 births <sup>25</sup>
	Denominators	<ul style="list-style-type: none"> <li>1,897 (2018-2020) and 512 (2021) children with a recorded referral time.</li> <li>2,447 (2018-2020) and 772 (2021) children with a recorded contact time.</li> </ul>
	Numerators	<ul style="list-style-type: none"> <li>Children referred to a cleft service within 24 hours of birth.</li> <li>Children contacted by cleft service within 24 hours of referral.</li> </ul>
	Data completeness	<ul style="list-style-type: none"> <li>67% of children had a recorded referral time.</li> <li>86% of children had a recorded time of first contact.</li> <li>33% of children were missing data on referral time, while 14% were missing data on time of first contact.</li> </ul>
	Countries	England, Wales and Northern Ireland
	Indicators	#3 & #4 <sup>26</sup>
Benchmark	<ul style="list-style-type: none"> <li>Children with a cleft should be referred to cleft services within 24 hours of birth.</li> <li>Cleft services should contact families of children within 24 hours of receiving a referral.</li> </ul>	
What did we find?	<ul style="list-style-type: none"> <li>84% of families with a recorded referral time were referred to a cleft service within 24 hours of birth (2018-2020). This percentage varied by cleft service, cleft type and sex.</li> <li>95% of families were contacted by a cleft service within 24 hours of referral (2018-2020). This percentage varied by cleft service, cleft type and sex.</li> </ul>	
Recommendations	<ul style="list-style-type: none"> <li>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.</li> <li>Regional variation in the percentage of children referred and contacted within 24 hours demonstrates that some cleft services have high levels of referrals and contacts within 24 hours. They should share their best practice recommendations with cleft services with lower rates.</li> </ul>	

#### 3.5.1. Data completeness

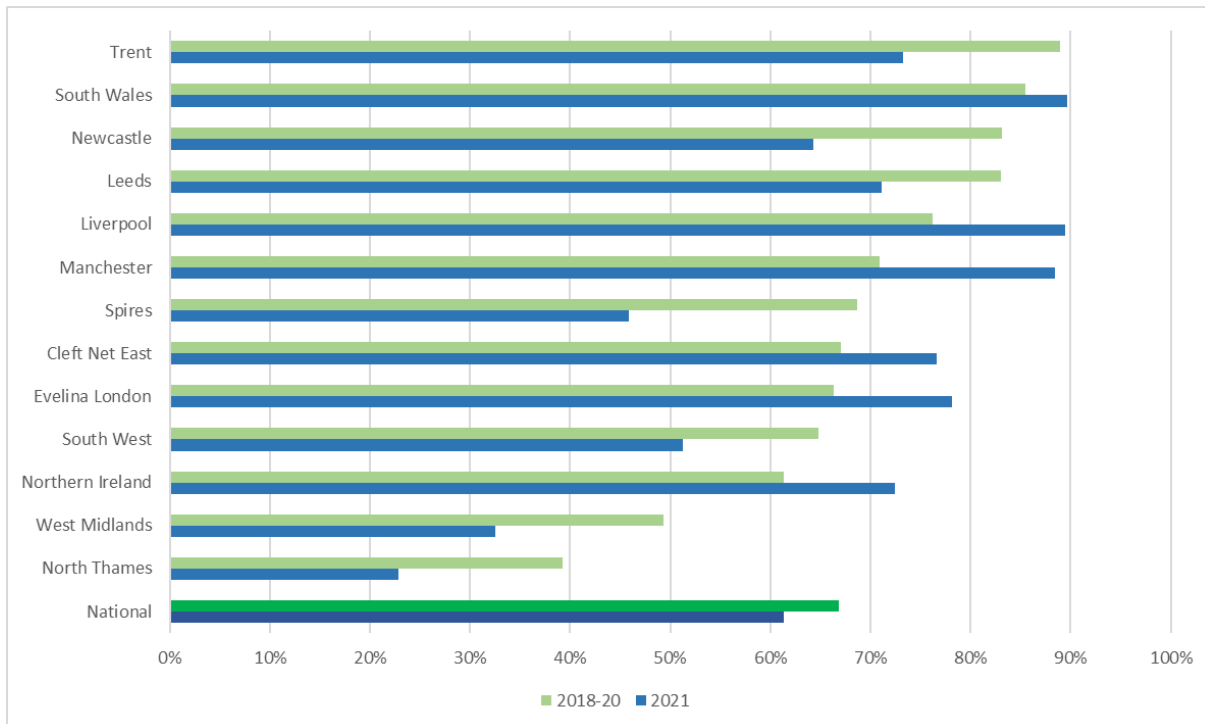
Out of 2,838 children born and registered in CRANE between 2018 and 2020, 67% had a recorded referral time to a cleft service. Of the 834 children registered in 2021, the second year impacted by the COVID-19 pandemic, 61% had a recorded referral time. Figure 3.9 below shows the variation in data completeness rate for the recording of referral time in 2018-20 (range: 39% to 89%) and 2021 (range: 23% to 90%) registrations for each cleft service.

Out of the 2,838 children born and registered in CRANE between 2018 and 2020, 86% had a recorded contact time after referral. The corresponding figure for 2021 births was 93%. Figure 3.10 shows the variation in data completeness rate for the recording of contact time in 2018-20 (range: 41% to 99%) and 2021 (range: 54% to 100%) registrations for each cleft service.

<sup>25</sup> Affected by the COVID-19 pandemic and the lockdown that ensued.

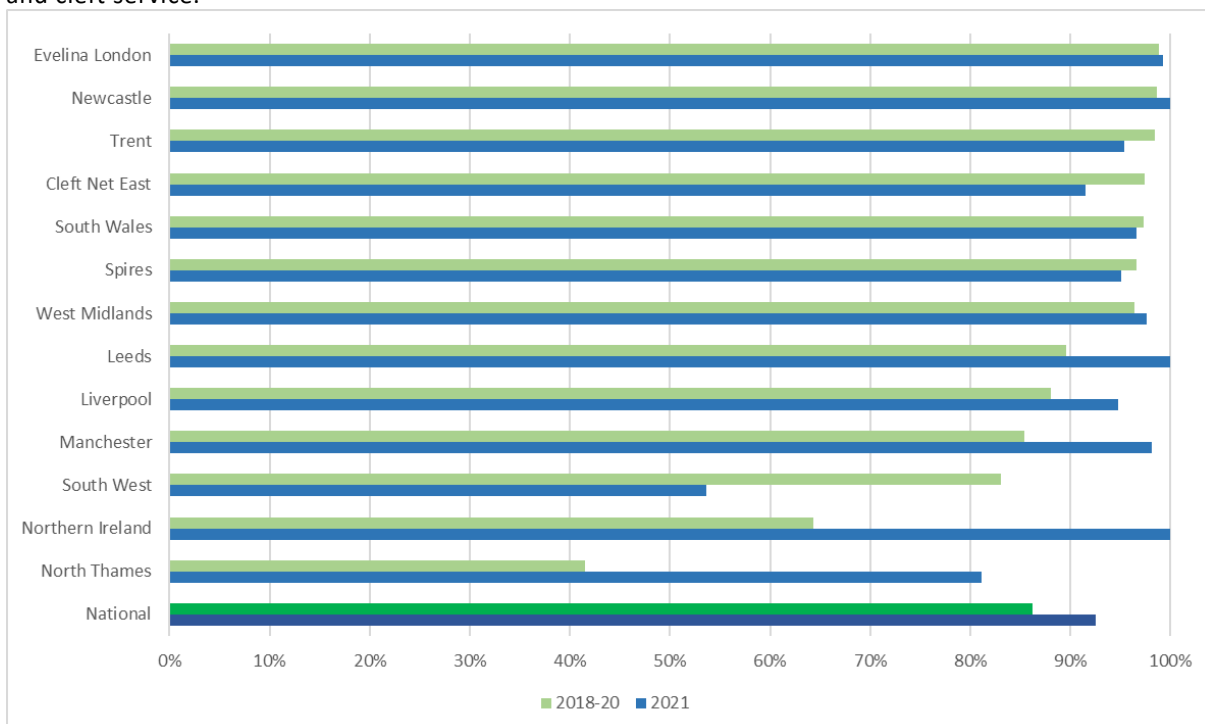
<sup>26</sup> CRANE core indicators are detailed in [the supplementary tables](#).

**Figure 3.9.** Percentage of CRANE-registered children with a recorded referral time, according to birth cohort and cleft service.



Note: All children registered in CRANE between 01 January 2018 and 31 December 2021, by 27 June 2022. Excludes children who died within the first week after birth.

**Figure 3.10.** Percentage of CRANE-registered children with a recorded contact time, according to birth cohort and cleft service.



Note: Data consist of all children registered in CRANE between 01 January 2018 and 31 December 2021, by 27 June 2022. Excludes children who died within the first week after birth.

### 3.5.2. Referral and first contact

Out of 1,897 children with a recorded referral time in 2018-20, and the 512 children registered in CRANE in 2021, 84% were referred to a cleft service within 24 hours of birth. This shows consistency in the rate of referrals within 24 hours of birth over the last four years. Raw data in [the supplementary tables](#) show the variation in



percentage of children referred within 24 hours of birth in 2018-20 (range: 76% to 88%) and 2021 (range: 52% to 93%) registrations, for each cleft service.

Furthermore, exploration of 2018-20 registrations showed that:

- Referrals also varied significantly by cleft type ( $p$ -value $<0.001$ ). Children diagnosed with CP had the lowest rate of early referrals within 24 hours of birth (71%), compared to CL (91%), UCLP (95%) and BCLP (95%). 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 slightly higher among boys (85%) than girls (83%), but this did not reach statistical significance ( $p$ -value=0.237).

Of the 2,447 children with a recorded time of first contact with a cleft service in 2018-20, 95% were contacted within 24 hours of referral. The same rate was observed for the 772 children registered in 2021. Raw data in [the supplementary tables](#) show the variation in percentage of children contacted within 24 hours of birth in 2018-20 (range: 82% to 100%) and 2021 (range: 79% to 100%), for each cleft service.

- The percentage of children contacted by cleft services within 24 hours of referral in 2018-20 varied significantly by cleft type ( $p$ -value=0.019). 95% of children diagnosed with CL were contacted within 24 hours of referral in 2018-20, followed by those diagnosed with UCLP (98%), BCLP (97%) and CP (94%).
- The percentage of children contacted within 24 hours of referral in 2018-20 was similar between boys (96%) and girls (95%) ( $p$ -value=0.625).

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: 2012 to 2014 – key to running the 5-year outcomes outlier process. Plus summary information on 2015 births <sup>27</sup>
	Denominator	3,200 children registered in CRANE
	Numerators	2,878 children whose families had made a decision about consent
	Exclusions	<ul style="list-style-type: none"> <li>Children who died before the age of 5 years</li> <li>Children with submucous cleft palates<sup>28</sup></li> </ul>
	Data completeness	90% of 3,200 registered children had verified consent. This means they had agreed to or declined CRANE data collection.
	Countries	England, Wales and Northern Ireland
	Indicator	#5 <sup>29</sup>
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?	<ul style="list-style-type: none"> <li>90% of families had reached a decision about agreeing or declining consent.</li> <li>Of the families who had reached a decision, the rate of agreed consent was 98%, consistent with previously reported high rates of positive consent.</li> </ul>	
Recommendations	<ul style="list-style-type: none"> <li>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.</li> <li>CRANE will continue to work with cleft services and the Nursing Clinical Excellence Network (CEN) to improve consent status verification.</li> </ul>	

The families of 2,878 (90%) out of 3,200 children born between 2012 and 2014 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 67.8% (Evelina London) to 100% (Northern Ireland). For information on the challenges experienced by Evelina London in obtaining consent, please see Box 3.1 below and refer to the [Response to outlier process](#) document for the full response.

[The supplementary tables](#) show the breakdown of consent status according to cleft services for 2012-14 births.

Identification of outliers for this process indicator (#5) has highlighted 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. See Box 3.1 (below) for comments from Evelina London on their outlier status.

This process has also led to the identification of positive outliers such as Newcastle, Leeds, Manchester and

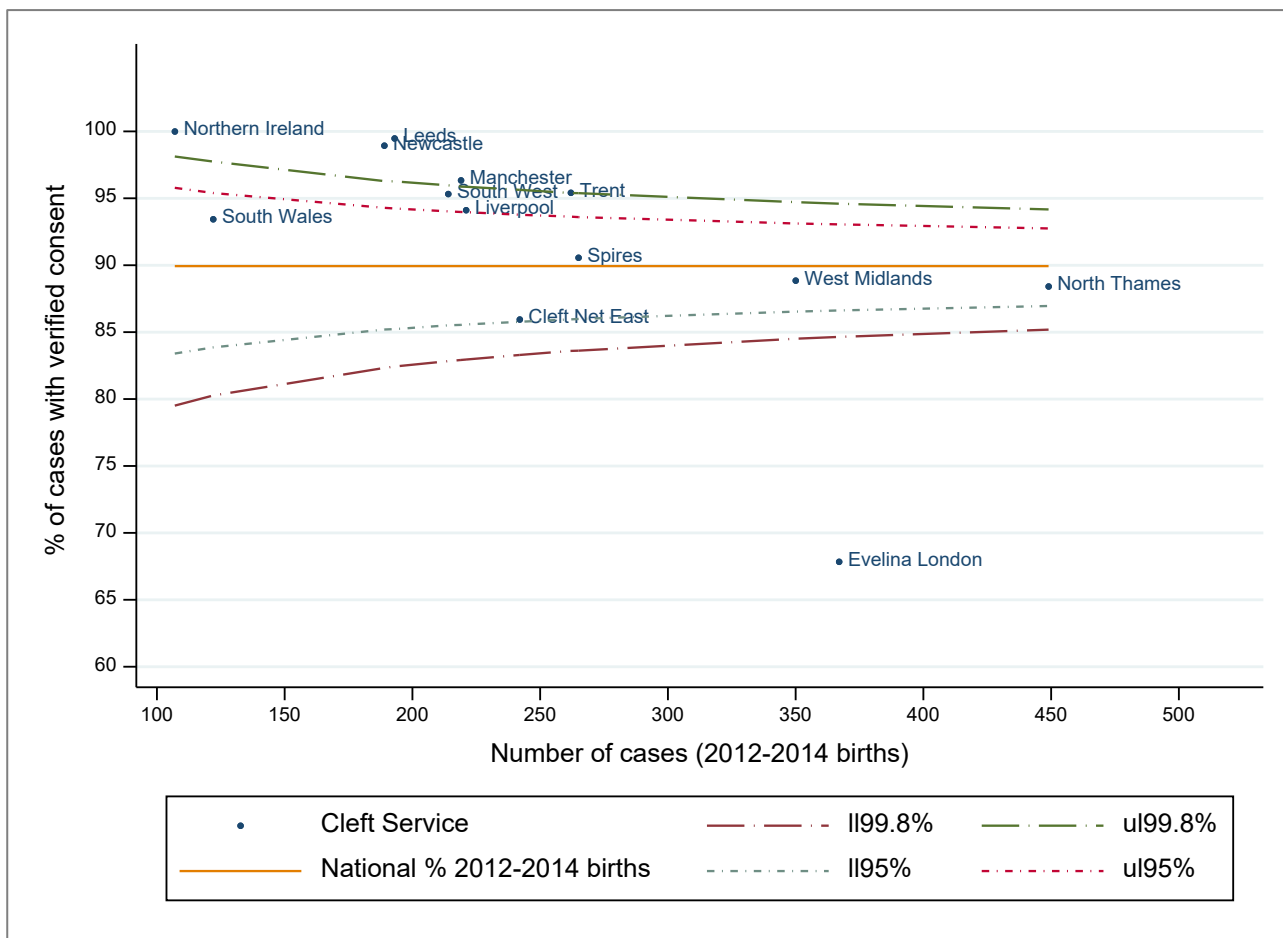
<sup>27</sup> Affected by the COVID-19 pandemic and the lockdown that ensued.

<sup>28</sup> 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.

<sup>29</sup> CRANE core indicators are detailed in [the supplementary tables](#).

Northern Ireland cleft services.

**Figure 3.11.** Funnel plot showing the percentage of CRANE-registered children, born 2012 to 2014, with verified consent, according to cleft service.



**Note:** Registered in CRANE by 27 June 2022. Funnel plot centred on the overall national percentage (89.9%) of children (born 2012-2014) with verified consent. See [the supplementary tables](#) for the raw data used to create this funnel plot.

**Box 3.1.** Summarised response to being a negative outlier for consent verification. Full responses published in the “2022 Annual Report: Responses to outlier process” document.

“For babies born in 2012-2014, our consent verification figure (the number of families where we have obtained their consent to be registered on the CRANE Database) is reported as 67% (246 out of 367 births). This is disappointingly low and our local investigations and audit demonstrate that there are a number of reasons why at the time when these children were born (between 8 and 10 years ago), consent was not collected. We are tackling this issue with both a retrospective and prospective strategy and are seeing some good results – certainly of the new processes put in place for gathering consent for babies born from 2021 onwards. Consent verification is now firmly built into the existing Cleft Clinical Nurse Specialist pathway with a goal of all relevant patients having had consent established (through contact with parents) within the first 6 months of life, and certainly by 12 months... We are confident that, over time, the Evelina London Cleft Service will no longer be a negative outlier in this respect ”

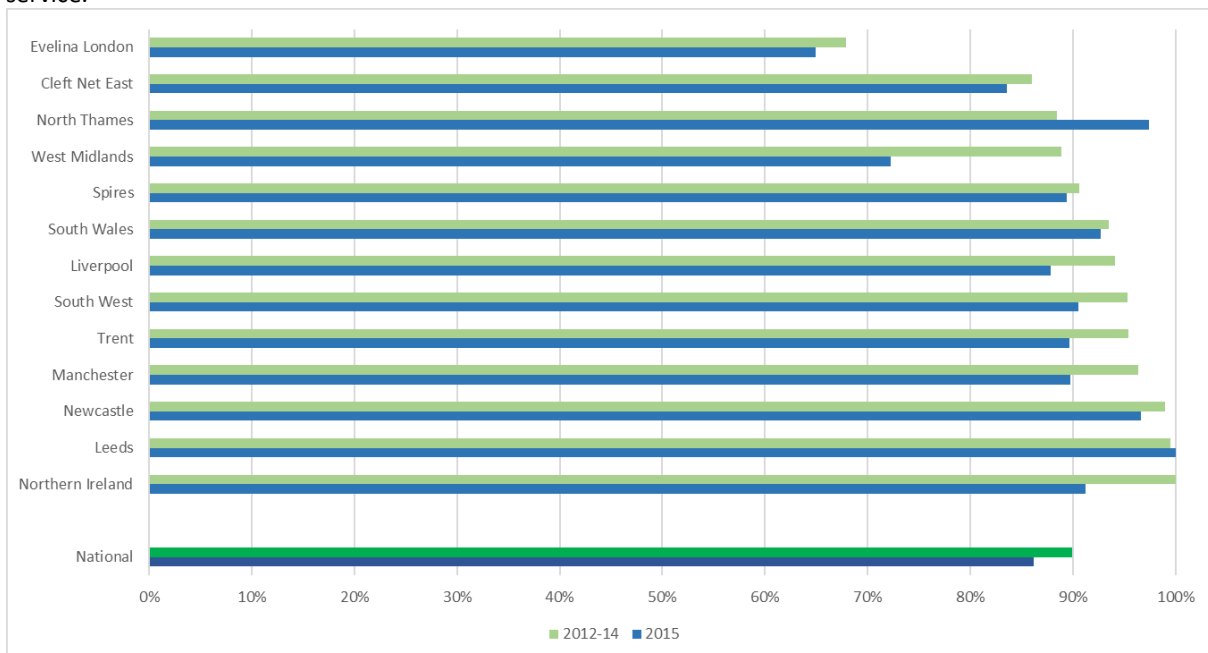
**Evelina London cleft service, November 2022.**

Read the full response in the [“2022 Annual Report: Responses to outlier process”](#) document published online.

Figure 3.12 summarises consent verification rates for children born in 2015 (86% achieved nationally), which will be reported in the CRANE 2023 Annual Report, by comparison with consent verification rates in 2012-14.

The supplementary tables also show the breakdown of consent status according to cleft service for 2015 births.

**Figure 3.12.** Percentage of CRANE-registered children with verified consent, according to birth cohort and cleft service.



**Note:** Registered in CRANE by 27 June 2022. Exclusions (not mutually exclusive): Children who died before the age of 5 years and those with submucous cleft palates. See the supplementary tables 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. This will benefit patients and their families, as well as the clinical community and commissioners.

## 4. Audit outcomes at 5 years of age

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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 2012 and 31 December 2014.

In previous years, we have reported on outcomes by individual cleft service. 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. This year, we reflect on three years of outcome data from children born 2012 to 2014 for child growth, dental health, facial growth, speech and psychology, and we provide summary data for 2015 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 2012 and 31 December 2014 (consistent with a rolling 3 years, as for other sections of this report).
- **Cohort 2.** Children born between 01 January 2015 and 31 December 2015; these are presented separately at the request of The Cleft Development Group.

**Legal basis for data collection and analysis:** The data used for this section are 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: 2012 to 2014. Plus summary information on 2015 births <sup>30</sup>
	Denominator	1,285 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	<ul style="list-style-type: none"> <li>Children without consent to data collection</li> <li>Children who died before the age of 5 years</li> <li>Children with submucous cleft palates<sup>31</sup></li> <li>Children without recorded height and weight at the age of 5</li> </ul>
	Data completeness	<ul style="list-style-type: none"> <li>46% of 2,812 eligible consented children had a recorded height and weight.</li> <li>&lt;1% of children only had a recorded height.</li> <li>1% of children only had a recorded weight.</li> <li>11% had a reason the child growth outcomes were not collected.</li> <li>41% were missing data or a reason for not collecting data.</li> </ul>
	Countries	England, Wales and Northern Ireland
Indicator	#6 <sup>32</sup>	
Benchmarks	<ul style="list-style-type: none"> <li>For 5-year-olds in the UK<sup>33</sup>, healthy BMI ranges between 13.0 and 17.5 (2<sup>nd</sup> to 91<sup>st</sup> centiles). Underweight is BMI &lt;13.0 kg/m<sup>2</sup>, overweight is BMI 17.5-19.0 kg/m<sup>2</sup> (92<sup>nd</sup> to 98<sup>th</sup> centiles), and obese is BMI &gt;19.0 kg/m<sup>2</sup>.</li> <li>National prevalence of underweight, overweight and obesity among 5 year olds was estimated at 1%, 12% and 10%, respectively<sup>34</sup>.</li> </ul>	
What did we find?	<ul style="list-style-type: none"> <li>87% of children had a healthy BMI.</li> <li>2% of children were underweight, 7% overweight and 4% obese – according to BMI categorisation.</li> </ul>	
Recommendations	Cleft services should aim to assess children’s weight and height around the age of 5 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). **The supplementary tables** show the breakdown of the percentage of children with reported height and weight at 5 years of age for each cleft service for the 2012-14 birth cohort.

<sup>30</sup> Affected by the COVID-19 pandemic and the lockdown that ensued.

<sup>31</sup> 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.

<sup>32</sup> CRANE core indicators are detailed in **Appendix 3**.

<sup>33</sup> According to the Royal College of Paediatrics and Child Health and UK-WHO growth charts – 2-18 years (Last accessed online: 12/12/2022) – Available at: <https://www.rcpch.ac.uk/resources/uk-who-growth-charts-2-18-years>

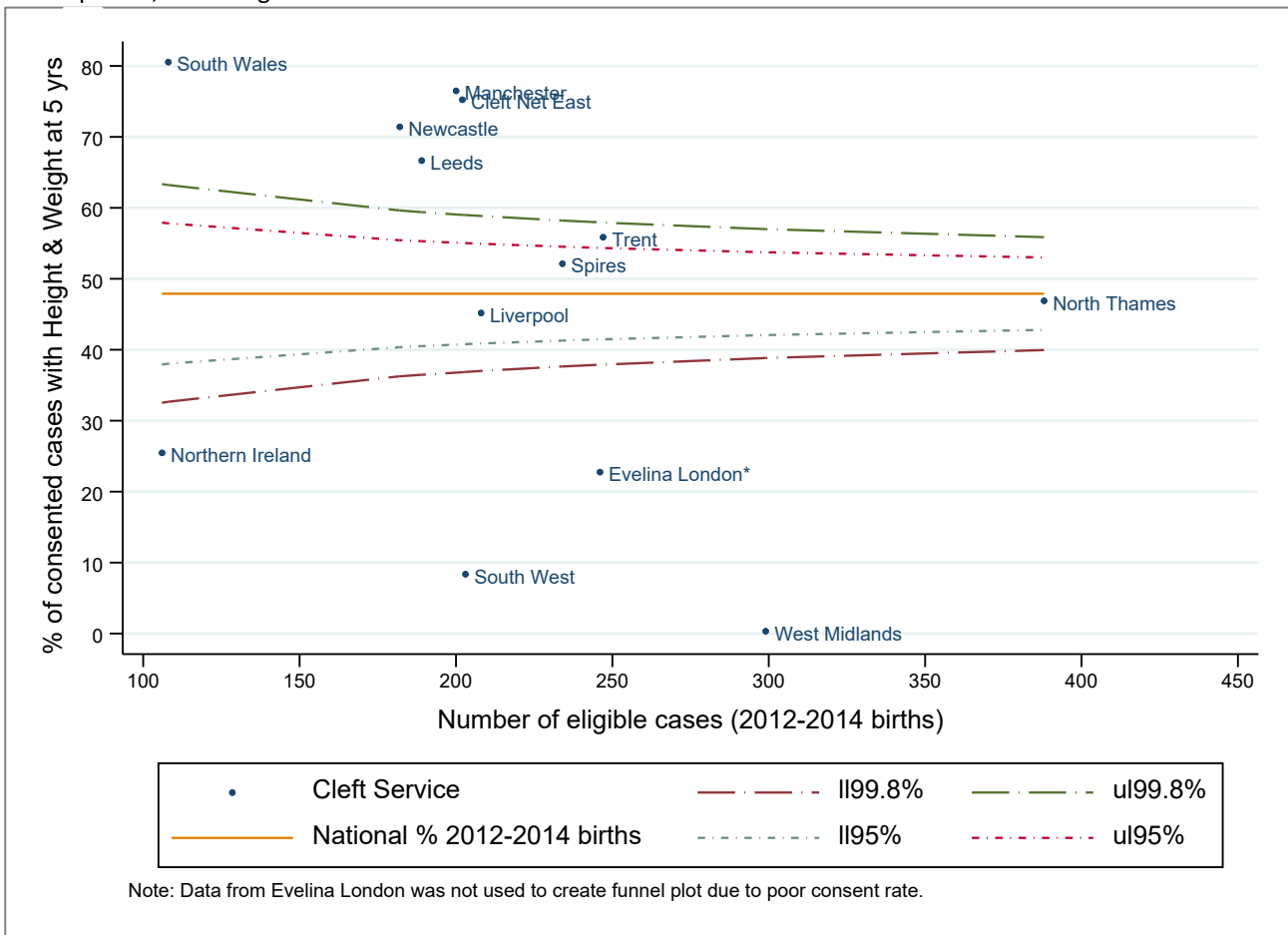
<sup>34</sup> National Child Measurement Programme Tables, England 2019/20 School Year – Published 29<sup>th</sup> October 2020 (Last accessed: 12/12/2022) – Available at <https://digital.nhs.uk/data-and-information/publications/statistical/national-child-measurement-programme/2019-20-school-year>

Of the 2,812 eligible 5-year old children born between 2012 and 2014, 46% (<1% to 81% 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. 11% had a reason why the outcome was not reported, and 41% were missing data or a reason.

**Reasons for not collecting 5-year old child growth outcome data for 2012-14 births**  
 The supplementary tables show the breakdown of reasons reported for not collecting 5-year old child growth outcome data for 2012-14 births. The most common reason selected was 'Lack of staff, facilities or equipment' (32%).

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 48%<sup>35</sup>. 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 2012 to 2014, who had growth data reported, according to cleft service.



**Note:** Registered in CRANE by 27 June 2022. Funnel plot centred on the adjusted national percentage (47.9%) of children (born 2012-2014) with growth outcomes at the age of 5 years reported. See the supplementary tables for the raw data (and exclusions) used to create this funnel plot.

The funnel plot shows that West Midlands, South West, Northern Ireland and Evelina London are negative outliers for the percentage of eligible 5-year-olds 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 has also allowed the

<sup>35</sup> The overall mean was 45.7% and adjusted mean was 47.9% (excluding cleft services with consent verification outlier status).

identification of positive outliers such as Newcastle, Leeds, Manchester, Cleft Net East and South Wales. See Box 4.1 (below) for comments from cleft services on their outlier status.

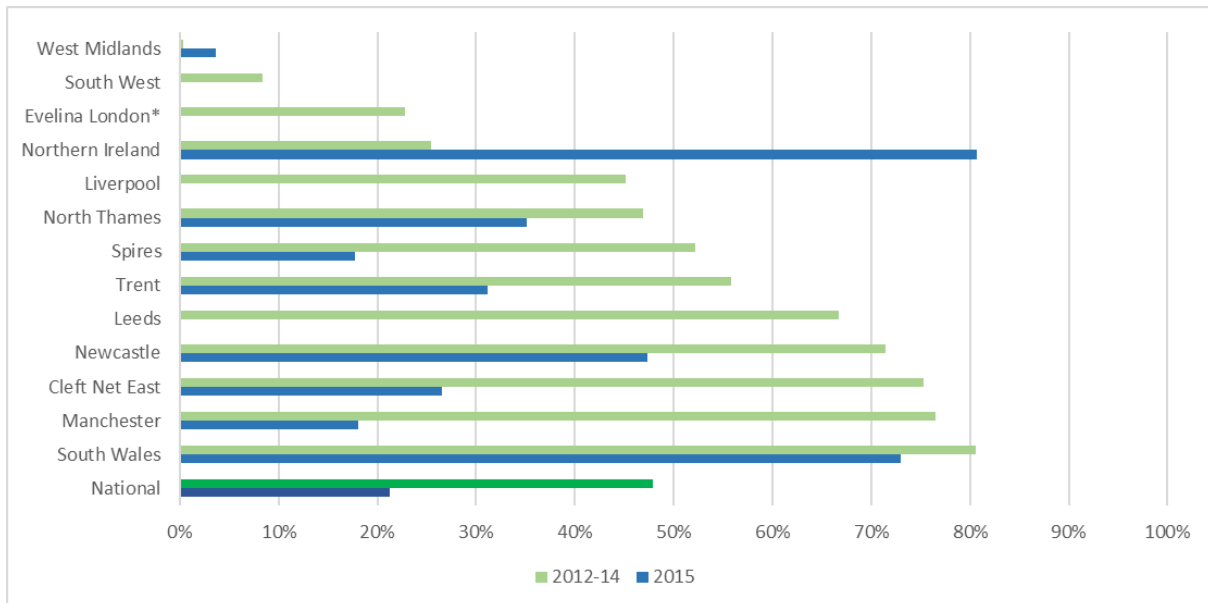
**Box 4.1.** Summarised responses to being negative outliers for ‘growth outcome - data completeness at 5 years’. Full responses published in the “2022 Annual Report: Responses to outlier process” document.

<p>“As a team we have worked hard to identify gaps in our data completeness and to rectify this. This has involved going back through the medical notes for some patients which has been time consuming. In order to avoid this for 2015 data and for the future, we have implemented a proforma at the designated 5 year old clinics.</p> <p>This has enabled us to record dental results and growth directly onto the proforma at the time of the clinic rather than searching through the medical note entries. If we are unable to obtain the information, a reason is given at the time. The data from the proforma is inputted onto the CRANE database by our designated secretary, responsible for recording the data. We hope this will continue to support data completeness for our unit.”</p> <p>South Wales cleft service, November 2022.</p>
<p>“This is a difficult metric to improve upon in the short term because these children turned 5 (the year in which the data should have been collected) between 2017 and 2019. We cannot retrospectively collect this data (it can only be collected when the child is 5 years old) and so cannot improve on this figure for previous years.</p> <p>However, our analysis of this issue for last year’s CRANE report found various reasons for the lack of data collection and we accept that our processes were inadequate at that time. We have thus improved on our protocol and now have a system in place whereby the dental nurses supporting our MDT clinics routinely take height and weight measurements for all child patients attending clinics in-person and certainly for those who are 5 years old. The heights and weights are logged on the patient’s electronic patient record and our Audit and Information Officer is notified of these so that the data can be uploaded to CRANE.”</p> <p>Evelina London cleft service, November 2022.</p>
<p>Read the full responses in the <a href="#">“2022 Annual Report: Responses to outlier process”</a> document published online.</p>

Anticipating ongoing challenges to data completeness due to the COVID-19 pandemic, Figure 4.2 summarises data completeness for eligible children born in 2015 (reporting weight and height data) compared with 2012-2014 data. **The supplementary tables** show the breakdown of growth reporting for each cleft service, for 2012-14 as well as 2015 births.



**Figure 4.2.** Percentage of CRANE-consented 5-year-olds who had growth data reported, according to birth cohort and cleft service.



**Note:** Registered in CRANE by 27 June 2022. See [the supplementary tables](#) for the raw data (and exclusions) used to create this graph.

Overall, of the 891 eligible 5-year-old children born in 2015, 21% had both height and weight reported (0%-81% range between services). Only two 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 2015 births**

[The supplementary tables](#) show the breakdown of reasons reported for not collecting 5-year-old child growth outcome data for 2015 births. The most common reason selected was ‘Patient DNA/cancelled/did not consent/ cooperate’ (35%).

**4.1.2. Height and weight**

Analyses revealed that the average weight and average height among 5-year-old children born between 2012 and 2014 with recorded growth data in the CRANE Database was 19.7 kg (95% CI: 19.6 kg to 19.9 kg) and 112.3 cm (95% CI: 111.9 cm to 112.6 cm), respectively.

These figures are consistent with UK national averages for 5-year-olds in the general population<sup>36</sup> for weight (range: 15 kg to 24 kg) and height (range: 99 cm to 119 cm). While 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 2012 and 2014 and with recorded 5-year growth data, their body mass index (BMI) at 5 years of age was calculated [weight (kg) / height (m) <sup>2</sup>] and categorised into weight status

<sup>36</sup> According to the Royal College of Paediatrics and Child Health and UK-WHO growth charts – 2-18 years (Last accessed online: 12/12/2022) – Available at: <https://www.rcpch.ac.uk/resources/uk-who-growth-charts-2-18-years>

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<sup>37</sup>. Among consented children with 5-year-old growth data recorded in the CRANE Database between 2012 and 2014, 86% had a healthy BMI, 8% were overweight, 4% were obese and 3% were underweight. The average BMI was 15.6 kg/m<sup>2</sup> (95% CI: 15.5 kg/m<sup>2</sup> to 15.7 kg/m<sup>2</sup>).

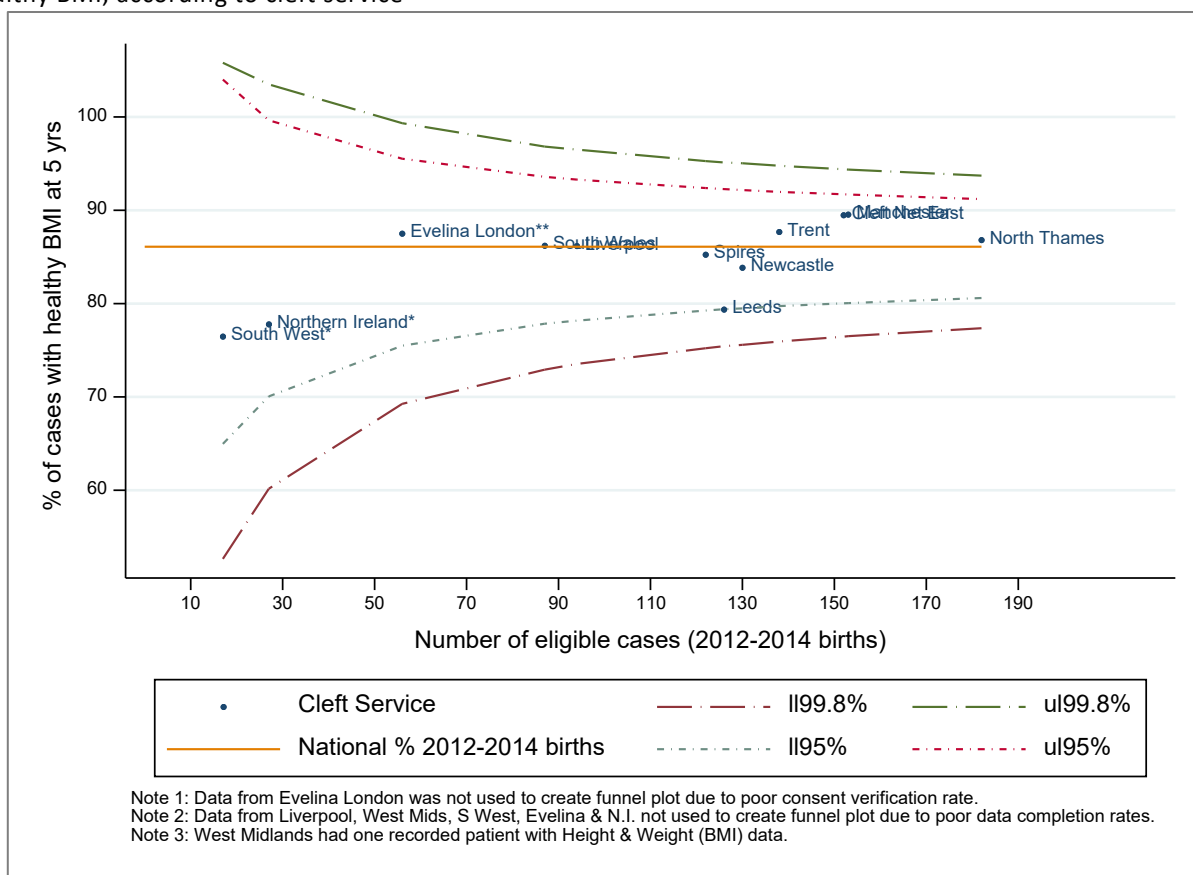
#### 4.1.4. Healthy BMI, by cleft service

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 86%<sup>38</sup> (range 77%-90%).

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

The supplementary tables show the breakdown of cases with healthy BMI at 5 years of age, as well as the other BMI categories, according to birth cohort and cleft service.

**Figure 4.3** Funnel plot showing the percentage of CRANE-consented 5-year-olds, born 2012 to 2014, with a healthy BMI, according to cleft service



**Note:** Registered in CRANE by 27 June 2022. Funnel plot centred on the adjusted national percentage (86.1%) of children (born 2012-2014) with healthy BMI at the age of five years reported. See the supplementary tables for the raw data (and exclusions) used to create this funnel plot.

<sup>37</sup>According to the Royal College of Paediatrics and Child Health - 2-20 years Body Mass Index (Last accessed online: 12/12/2022). Available from: <https://www.rcpch.ac.uk/resources/body-mass-index-bmi-chart>

<sup>38</sup> Both the overall mean and adjusted mean (excluding cleft services with consent verification and data completeness outlier status) were 86.9%.

## 4.2. Dental health

Cohort summary	Data source	The CRANE Database (consented cases only)
	Birth years	Three years: 2012 to 2014. Plus summary information on 2015 births <sup>39</sup>
	Denominators	<ul style="list-style-type: none"> <li>• 1,339 5-year-old children with recorded decayed, missing or filled teeth (dmft) scores</li> <li>• 1,330 children with scores for the calculation of Treatment Index<sup>40</sup></li> <li>• 1,331 children with scores for the calculation of Care Index<sup>41</sup></li> </ul>
	Numerators	<ul style="list-style-type: none"> <li>• 526 children with at least one (&gt;0) dmft</li> <li>• 189 children with extensive decay (dmft &gt;5)</li> </ul>
	Exclusions (not mutually exclusive)	<ul style="list-style-type: none"> <li>• Children without consent to data collection</li> <li>• Children who died before the age of 5 years</li> <li>• Children with submucous cleft palates<sup>42</sup></li> <li>• Children without a recorded dmft score at the age of 5</li> </ul>
	Data completeness	<ul style="list-style-type: none"> <li>• 48% of 2,812 eligible consented children had a recorded dmft score.</li> <li>• 17% had a reason dmft scores were not collected.</li> <li>• 35% were missing data or a reason for not collecting data.</li> </ul>
	Countries	England, Wales and Northern Ireland
	Indicators	#7 & #8 <sup>43</sup>
Benchmark	<ul style="list-style-type: none"> <li>• 100% of eligible children should have recorded dmft outcome data at the age of 5 years (or a valid reason it was not collected).</li> <li>• 23% of children had at least one (&gt;0) dmft and 13% had extensive decay (dmft &gt;5) in the general population of England, Wales and Northern Ireland at 5 years of age<sup>44</sup>.</li> <li>• 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<sup>45</sup>.</li> </ul>	
What did we find?	<ul style="list-style-type: none"> <li>• 39% of children with a cleft had at least one (&gt;0) dmft and 14% had extensive decay (dmft &gt;5).</li> <li>• The average Treatment Index was 77%, and the average Care Index was 70%.</li> </ul>	
Recommendations	<ul style="list-style-type: none"> <li>• All children with a cleft should have a recommended care plan established by collaborative work between the family's 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).</li> <li>• 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.</li> </ul>	

<sup>39</sup> Affected by the COVID-19 pandemic and the lockdown that ensued.

<sup>40</sup> 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).

<sup>41</sup> Care Index calculated using: data on filled teeth (f) and dmft scores; or a dmft score of 0 (equating to a Care Index = 1).

<sup>42</sup> 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.

<sup>43</sup> CRANE core indicators are detailed in [Appendix 3](#).

<sup>44</sup> 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>

<sup>45</sup> 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](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

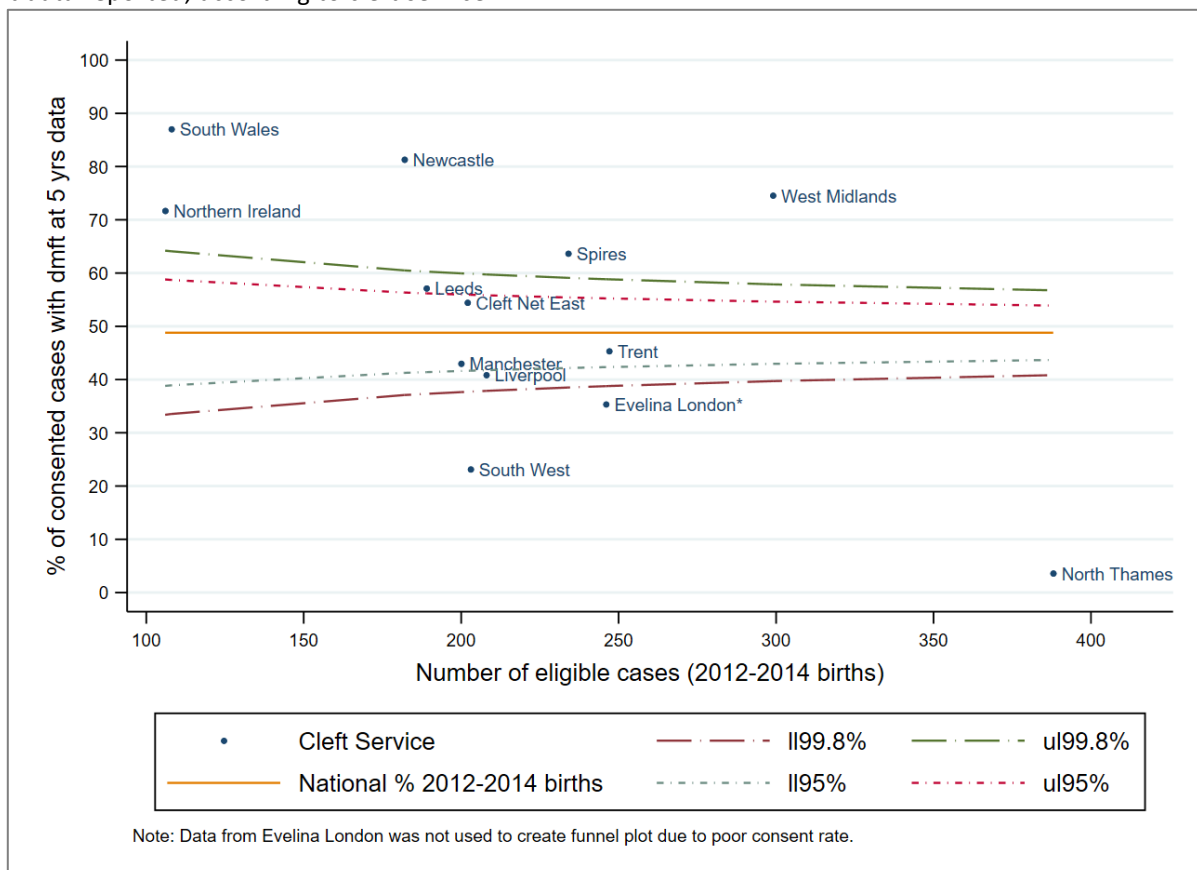
Participation in a national clinical audit, such as CRANE, means that all cleft services are asked to record dental health outcome data in the form of decayed, missing or filled teeth (dmft) scores for all consented 5-year-old children with a cleft lip and/or palate. If dmft score data are not available, cleft services are asked to report a reason for this (e.g. patient transferred out of area or patient did not attend appointment, etc). **The supplementary tables** show the breakdown of dental health reporting for each cleft service for the 2012-14 birth cohort.

Of the 2,812 eligible 5-year-old children born between 2012 and 2014, 48% had dmft scores reported (4%-87% range between services). 17% of cases had a reason why the outcome was not reported, and 35% were missing data or a reason.

**Reasons for not collecting 5-year old dental health outcome data for 2011-13 births**  
**The supplementary tables** show the breakdown of reasons reported for not collecting dmft at 5 years of age for 2012-14 births. The most common reason selected was 'Lack of staff facilities or equipment' (39%).

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 49%<sup>46</sup>. 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 2012 to 2014, who had dmft data reported, according to cleft service.



**Note:** Registered in CRANE by 27 June 2022. Funnel plot centred on the adjusted national percentage (48.8%) of children (born 2012-2014) with dmft data reported. See **the supplementary tables** for the raw data (and exclusions) used to create this funnel plot.

<sup>46</sup> The overall mean was 47.6% and adjusted mean was 48.8% (excluding cleft services with consent verification outlier status).

The funnel plot shows that North Thames, South West and Evelina London are negative outliers for the percentage of eligible cases with dmft data 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. Completing this process of identifying outliers also allowed the identification of positive outliers such as South Wales, Newcastle, West Midlands, Northern Ireland and Spires cleft services. See Box 4.2 and 3 (below) for comments from cleft services on their outlier status.

**Box 4.2.** Summarised responses to being negative outliers for ‘dental health - data completeness at 5 years’ – extracts. **Full responses published in the “2022 Annual Report: Responses to outlier process” document.**

“The problems with the provision of a cleft calibrated paediatric dentist within the team persist with just one person able to record the necessary outcomes at 5 years. The post of consultant in paediatric dentistry, specifically covering the cleft service, has been advertised 3 times but unfortunately any applicants applying have not been appointable. We are about to advertise for a fourth time and are hopeful that there are candidates interested in the post... within the last 12 months conditions within the department have changed so that our one calibrated clinician can now attend the audit clinics (previously they had a teaching commitment at another hospital). This has meant that for the last 12 months we have been recording calibrated DMFT outcomes, and we are hopeful this will be reflected in our outcomes moving forwards.”

**North Thames cleft service, November 2022.**

“We have taken considerable time to review our processes for gathering 5-year-old dental health outcome data...and note that the main reason for this not having been completed to a greater degree, was due to a lack of resource within our team ie an inadequate amount of Consultant Paediatric Dentist time within our service historically. Staffing level increases within cleft Paediatric Dentistry have been made and additional Consultant sessions were appointed to in late 2019. This has brought our establishment up to 1 WTE and increases our confidence that our service is now in a better position to gather data going forward.”

**Evelina London cleft service, November 2022.**

Read the full responses in the [“2022 Annual Report: Responses to outlier process”](#) document published online.

**Box 4.3.** Summarised responses to being positive outliers for ‘dental health - data completeness at 5 years’ – extracts. **Full responses published in the “2022 Annual Report: Responses to outlier process” document.**

“As a team we have worked hard to identify gaps in our data completeness and to rectify this. This has involved going back through the medical notes for some patients which has been time consuming. In order to avoid this for 2015 data and for the future, we have implemented a proforma at the designated 5 year old clinics. This has enabled us to record dental results and growth directly onto the proforma at the time of the clinic rather than searching through the medical note entries. If we are unable to obtain the information, a reason is given at the time. The data from the proforma is inputted onto the CRANE database by our designated secretary, responsible for recording the data. We hope this will continue to support data completeness for our unit.”

**South Wales cleft service, November 2022.**

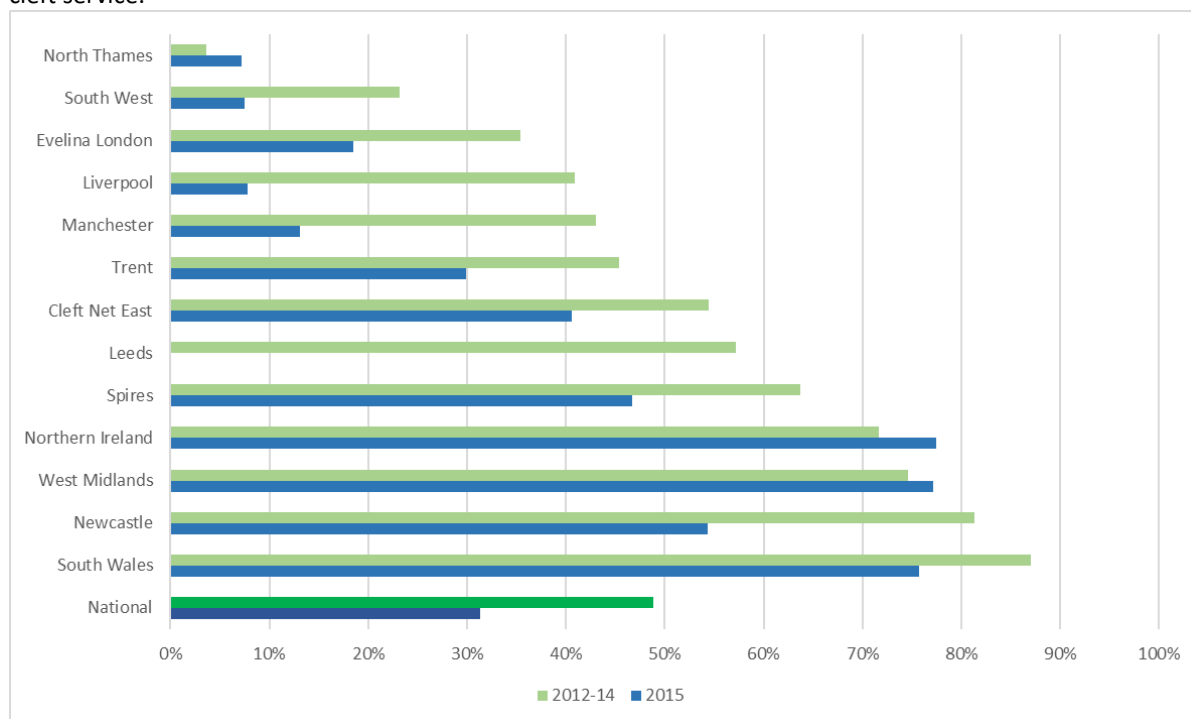
Read the full responses in the [“2022 Annual Report: Responses to outlier process”](#) document published online.

**The supplementary tables** show the breakdown of dmft reporting for each cleft service for 2012-14 as well as 2015 births.

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 the ability of cleft services to audit and submit the most recent years of CRANE data collection.

**Figure 4.5.** Percentage of CRANE-consented 5-year-olds who had dmft data reported, according to birth cohort and cleft service.



**Note:** Registered in CRANE by 27 June 2022. See [the supplementary tables](#) for the raw data (and exclusions) used to create this graph.

Overall, of the 891 eligible 5-year-old children born in 2015, 31% had dmft data reported (0%-77% range between services). Four 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 2015 births**

[The supplementary tables](#) show the breakdown of reasons reported for not collecting dmft at 5 years of age for 2015 births. The most common reason selected was ‘Other’ (56%). Comments provided to specify these ‘other’ reasons, in the main, described data not collected due to the COVID-19 pandemic and virtual clinics.

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

Among the 1,339 eligible children born in 2012-2014 with a reported dmft score, the mean number of dmft at 5 years was 2.07, with scores ranging from 0 to 20.

Overall, 39% of children had at least one decayed, missing or filled tooth (dmft >0). This is 16% 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<sup>47</sup>.

Furthermore, 14% of the CRANE cohort born in 2012-2014 had extensive decay (dmft >5) (as **defined below**). The corresponding rate for the general population is 13%<sup>50</sup>.

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

<sup>47</sup> 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>

## 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 39%<sup>48</sup> (range 21%-54%).

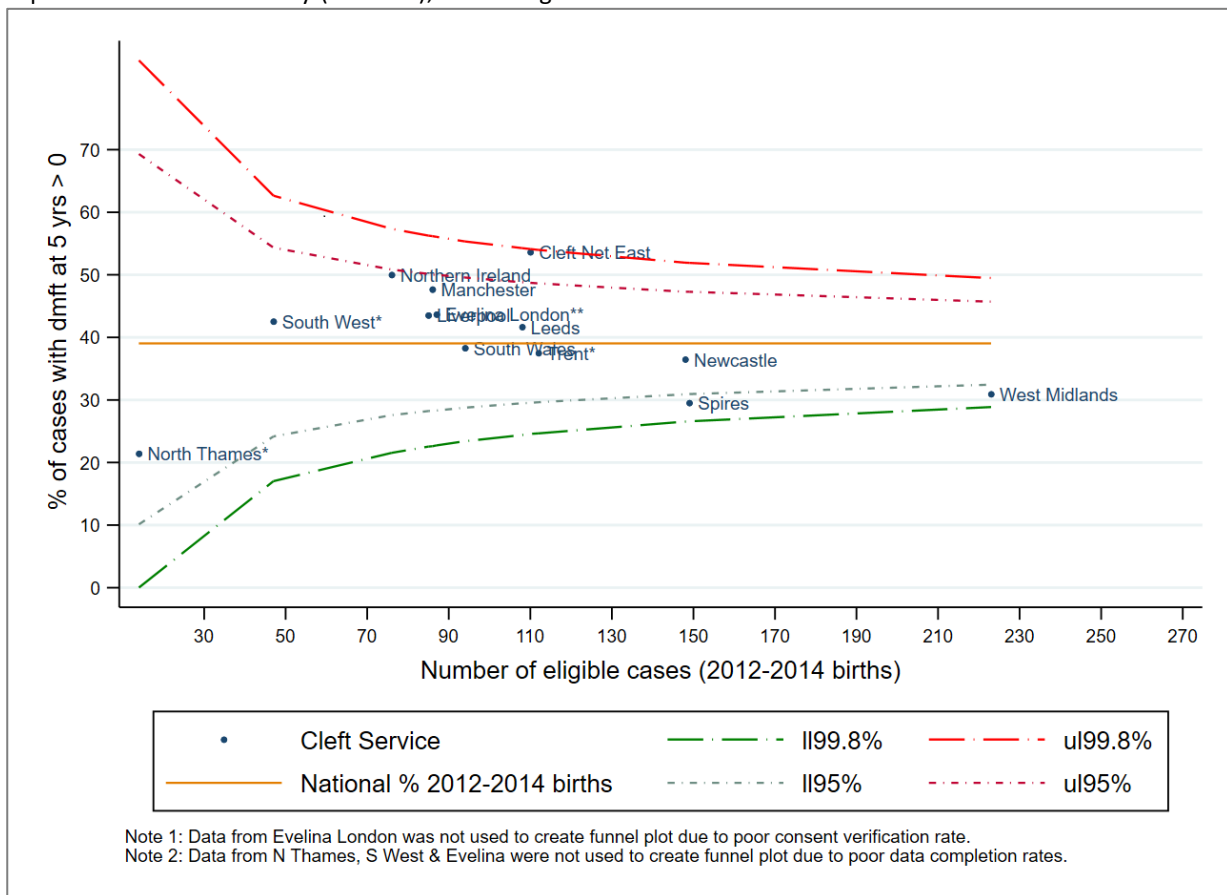
Submitting this indicator of dmft >0 (#7) to the outlier process did not lead to the identification of any outliers for this outcome indicator. **The supplementary tables** show the breakdown of cases with reported dmft >0 at 5 years of age, according to cleft service.

## 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<sup>49</sup>.

- 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 2012 to 2014, with experience of dental decay (dmft >0), according to cleft service.



**Note:** Registered in CRANE by 27 June 2022. Funnel plot centred on the adjusted national percentage (39%) of children (born 2012-2014) with dmft >0 reported. See **the supplementary tables** for the raw data (and exclusions) used to create this graph.

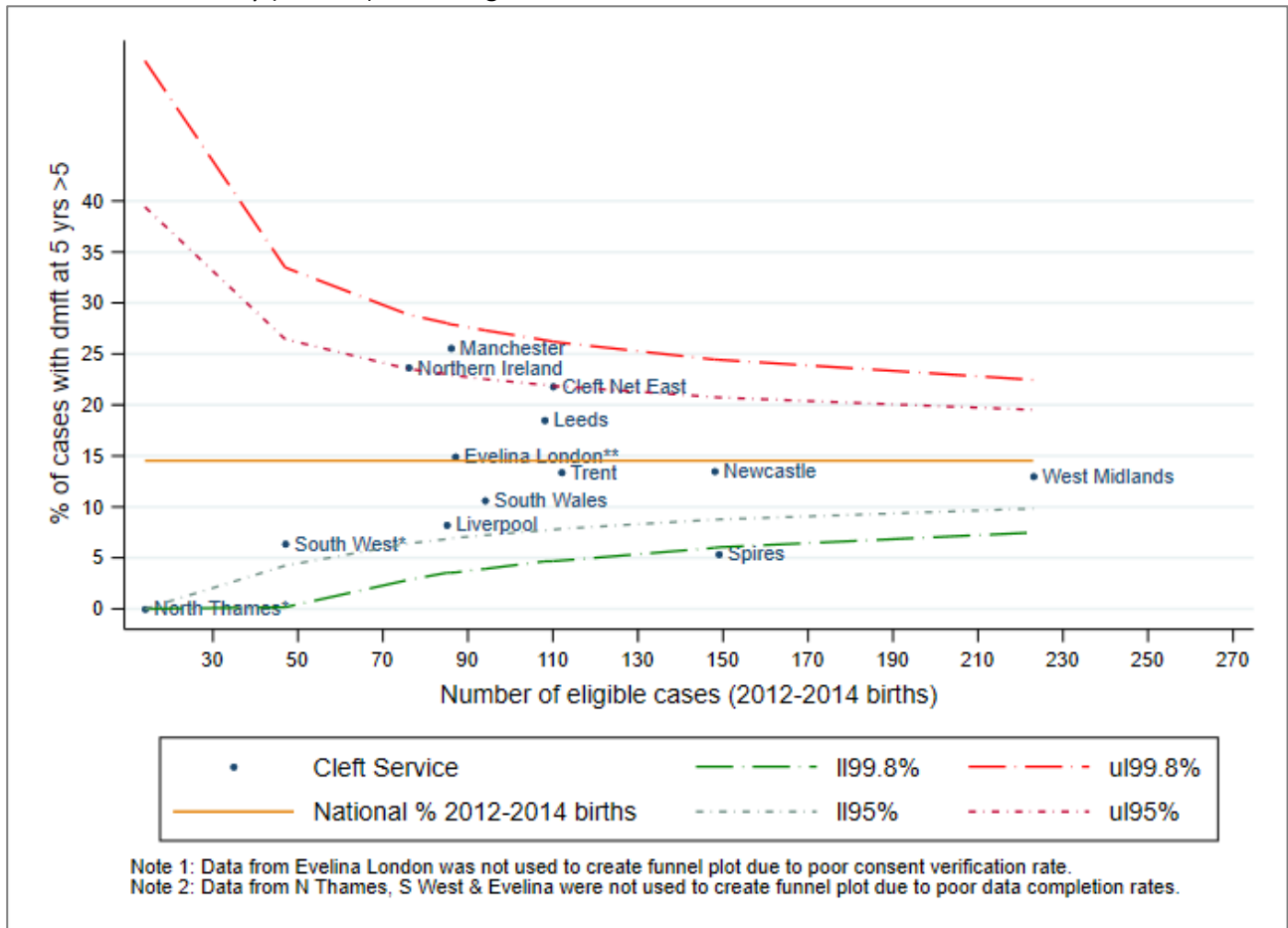
<sup>48</sup> The overall mean was 39.3% and adjusted mean was 39% (excluding cleft services with consent verification and data completeness outlier status).

<sup>49</sup> (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%<sup>50</sup> (range 0%-26%).

**Figure 4.7.** Funnel plot showing the percentage of CRANE-consented 5-year-olds, born 2012 to 2014, with extensive dental decay (dmft >5), according to cleft service.



**Note:** Registered in CRANE by 27 June 2022. Funnel plot centred on the adjusted national percentage (15%) of children (born 2012-2014) with dmft >5 reported. See the [supplementary tables](#) for the raw data (and exclusions) used to create this graph.

Completing this process of identifying outliers for this outcome indicator (#8) has allowed the identification of the Spires cleft service as a positive outlier. See Box 4.4 for a statement from the Paediatric Dentistry CEN on the interpretation of dmft outcomes.

The [supplementary tables](#) show the breakdown of cases with reported dmft >5 at 5 years of age for 2012-14 as well as 2015 births, according to cleft service.

<sup>50</sup> The overall mean was 14.1% and adjusted mean was 14.5% (excluding cleft services with consent verification and data completeness outlier status) were 86.9%.



**Box 4.4** Statement from Paediatric Dental CEN.

Cleft dental outcomes require assessment and scoring by a Cleft Calibrated Paediatric Dentist who has successfully completed appropriate cleft calibration in the preceding 24 months. However, not all units have a dentist who is so qualified. Therefore where dental outcomes are assessed and scored by persons other than a cleft qualified Specialist in Paediatric Dentistry they require interpretation with caution. Until such time as all services are able to provide specialist paediatric dental care to their patients inter-centre comparison of process and outcome data should also be undertaken with significant caution.

Furthermore significant geographical variations in dmft are known to occur within the general population. Regions where the local dmft is high (or low) are therefore more likely to sit towards the outer centiles of funnel plots. Within individual units the dental health of children with cleft lip and palate should be compared against both regional dental health outcomes as well as previous years to help evaluate changes in outcome.

Joanna May, Lucy Burbridge, and Jacqueline Smallridge, November 2022.

### 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. A dmft score of 0 or individual 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<sup>51</sup>. 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 provide 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<sup>52</sup>. 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 77%, 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 Kruskal-Wallis test was used to compare the mean rank of Treatment Index and Care Index scores between cleft services, which identified no statistically significant differences between services.

<sup>51</sup> If a dmft score for an individual is 0 then the treatment index is 1 (100%) as there is no untreated dental disease.

<sup>52</sup> If a dmft score for an individual is 0 then the Care Index is 1 (100%) as there is no dental disease.

**Table 4.1.** Average dental Treatment Index and average Care Index among CRANE-consented 5-year-olds, born 2012 to 2014, according to cleft service.

Cleft service	Eligible cases with dmft reported	Mean Treatment Index		Mean Care Index	
	148	148	73.3%	148	66.7%
Newcastle	108	108	74.3%	108	66.3%
Leeds	85	83	73.5%	84	62.0%
Liverpool	86	85	70.5%	85	63.1%
Manchester	112	112	79.4%	112	73.8%
Trent	223	222	78.9%	222	75.1%
West Midlands	110	109	79.8%	109	72.5%
Cleft Net East	14	14	85.7%	14	78.6%
North Thames	149	147	78.7%	147	75.0%
Spires	94	94	82.0%	94	68.8%
South Wales	47	45	73.0%	45	70.2%
South West	87	87	80.8%	87	72.1%
Evelina London	76	76	73.2%	76	62.1%
Northern Ireland	1,339	1,330	77.0%	1,331	70.0%
Total	148	148	73.3%	148	66.7%

**Note:** Registered in CRANE by 27 June 2022. Exclusions: Children with submucous cleft palates.

The supplementary tables show the information in the table above, according to cleft service, for 2015 births.

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 subsequent years, including in 2022, revealed a low number of data returns. A summary of the findings from these 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 currently, there are insufficient data to accurately reflect on this outcome.

We will review data completeness for this outcome once again in 2023, 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: 2012 to 2014. Plus summary information on 2015 births <sup>53</sup>
	Denominator	289 5-year-old children with complete unilateral cleft lip and palate (UCLP) and recorded 5-year-old Index scores (2012-2014)
	Numerator	The number of children classified as having ‘good’, ‘fair’ or ‘poor’ 5-year-old Index scores
	Exclusions (not mutually exclusive)	<ul style="list-style-type: none"> <li>• Children without consent to data collection</li> <li>• Cases with incomplete UCLP</li> <li>• Children who died before the age of 5 years</li> <li>• Children with submucous cleft palates<sup>54</sup></li> <li>• Children without a recorded 5 year old Index score</li> </ul>
	Data completeness	<ul style="list-style-type: none"> <li>• 59% of 489 eligible consented children had recorded 5-year-old Index scores.</li> <li>• 16% had a reason the facial growth scores were not collected.</li> <li>• 25% were missing data or a reason for not collecting data.</li> </ul>
	Countries	England, Wales and Northern Ireland
	Indicator	#9 <sup>55</sup>
Benchmarks	<ul style="list-style-type: none"> <li>• 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).</li> <li>• CSAG reported in 1998 that 29%, 34% and 37% of children with a complete UCLP had ‘good’, ‘fair’ and ‘poor’ dental arch relationships, respectively<sup>56</sup>.</li> </ul>	
What did we find?	<ul style="list-style-type: none"> <li>• 38% of children had scores reflecting ‘good’ dental arch relationships at 5 years old.</li> <li>• 37% of children had scores reflecting ‘fair’ dental arch relationships at 5 years old.</li> <li>• 25% of children had scores reflecting ‘poor’ dental arch relationships at 5 years old.</li> </ul>	
Recommendations	<ul style="list-style-type: none"> <li>• 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.</li> <li>• The research community should undertake to compare UK facial growth outcomes with those in other countries.</li> <li>• The research community should undertake to evaluate the predictive value of the 5-year-old Index in UK populations.</li> </ul>	

#### 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<sup>57</sup> (e.g. patient transferred out of area or patient did not attend appointment, etc). **The supplementary tables** show the breakdown of facial growth reporting for each cleft service for the 2012-14 and 2015 birth cohorts.

<sup>53</sup> Affected by the COVID-19 pandemic and the lockdown that ensued.

<sup>54</sup> Submucous cleft palate patients excluded from all 5-year outcomes. Including cases with LAHSAL code identified as not possible, and likely submucous, when reviewed by cleft clinicians.

<sup>55</sup> CRANE core indicators are detailed in **the supplementary tables**.

<sup>56</sup> Clinical Standards Advisory Group, Clinical Standards Advisory Group. Report of a CSAG Committee on cleft lip and/or palate, 1998, The Stationery Office, London.

<sup>57</sup> 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

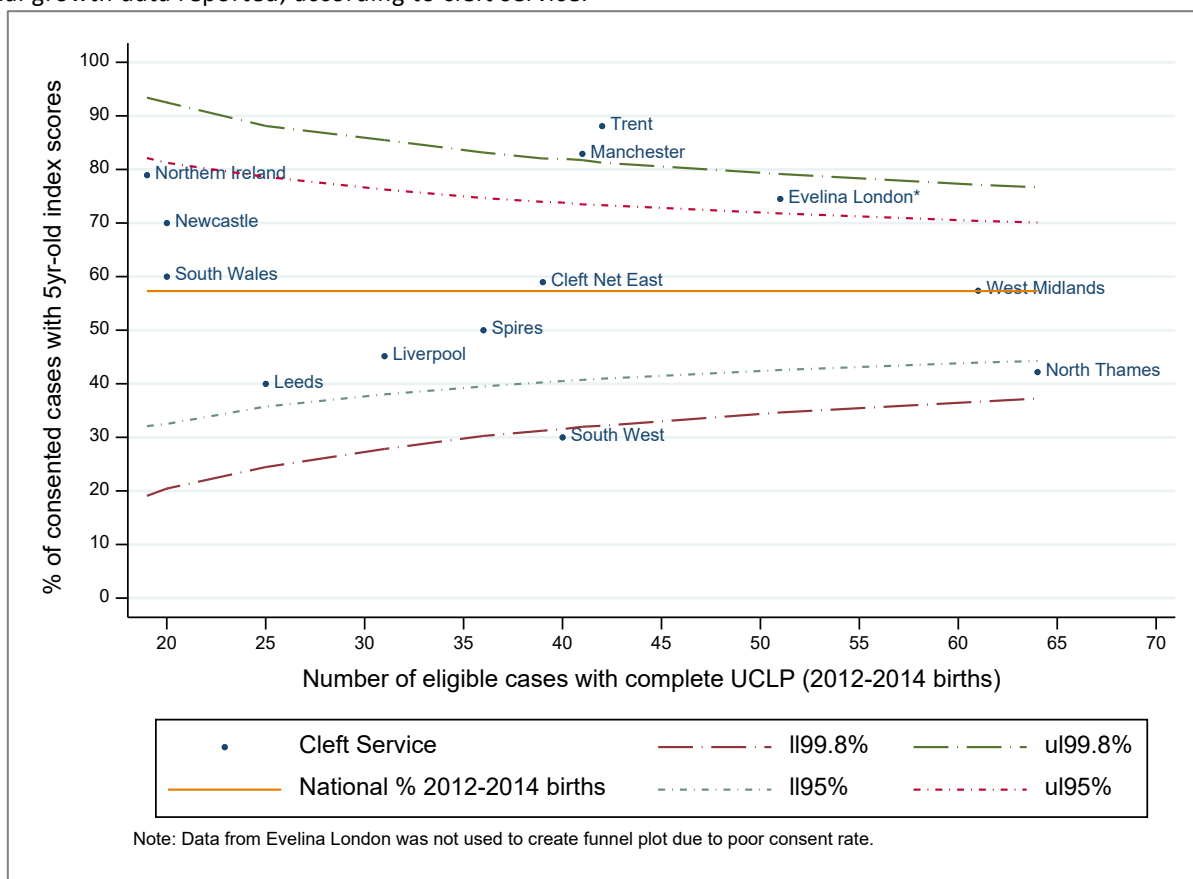
Of the 489 eligible 5-year-old children born between 2012 and 2014, 59% had a 5-year-old Index score recorded in CRANE (30% - 88% range between services). 16% of children had a documented reason why the outcome was not collected, and 25% of children were missing data or a documented reason.

**Reasons for not collecting 5-year-old facial growth outcome data for 2012-14 births**

The supplementary tables show the breakdown of reasons reported for not collecting 5-year old facial growth outcome data for 2012-14 births. The most common reason selected was 'Other' (34%). Comments provided to specify these 'other' reasons, in the main, described clinics cancelled / data not collected due to the COVID-19 pandemic.

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 57%<sup>58</sup>. 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 2012 to 2014, who had facial growth data reported, according to cleft service.



**Note:** Registered in CRANE by 27 June 2022. Funnel plot centred on the adjusted national percentage (57.3%) of children (born 2012-2014) with facial growth outcomes at the age of five years reported. See the supplementary tables for the raw data (and exclusions) used to create this funnel plot.

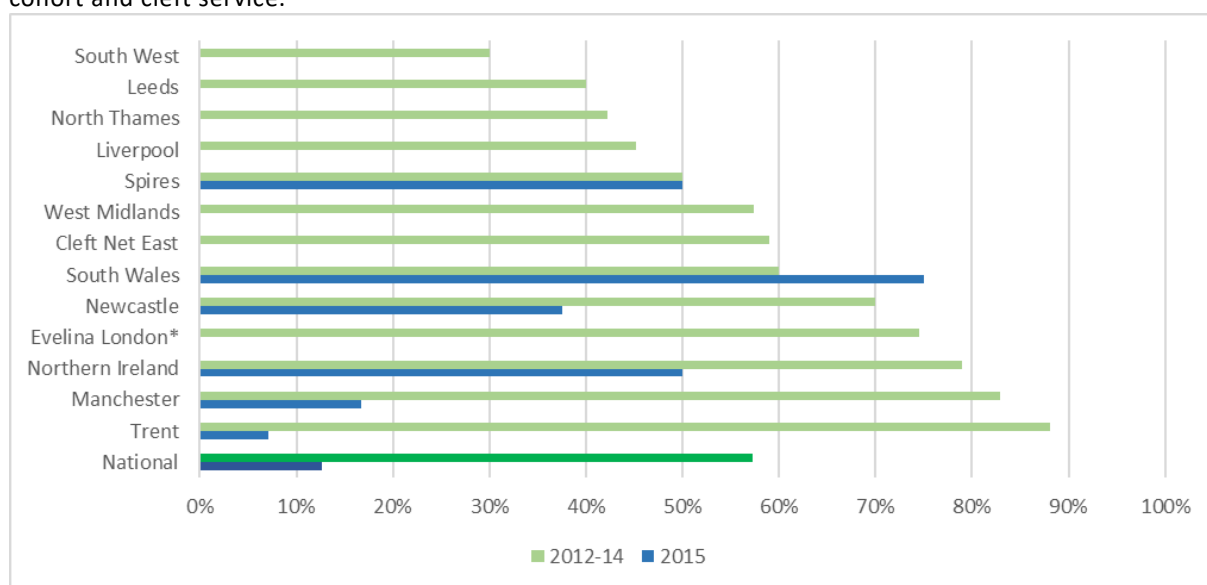
The funnel plot shows that South West is a negative outlier for the percentage of eligible cases with facial growth data items reported. This means that the South West cleft service is below the 99.8% control limit that would warrant exclusion from overall average calculations presented in the subsequent outcome funnels. Trent

<sup>58</sup> The overall mean was 59.1% and adjusted mean was 57.3% (excluding cleft services with consent verification outlier status).

and Manchester cleft services were positive outliers for the percentage of eligible cases with facial growth data items reported.

The supplementary tables show the breakdown of facial growth reporting for each cleft service, for 2012-14 as well as 2015 births. For the 2015 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 who had facial growth data reported, according to birth cohort and cleft service.



Note: Registered in CRANE by 27 June 2022. See the supplementary tables for the raw data (and exclusions) used to create this graph.

Overall, of the 150 eligible 5-year-old children born in 2015, 13% had a 5-year-old Index score recorded in CRANE (0% - 75% range between services). 17% 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 2015 births**

The supplementary tables show the breakdown of reasons reported for not collecting 5-year-old facial growth outcome data for 2015 births. The most common reason selected was 'Other' (65%). 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**

Records of facial growth (impressions or photographs) from 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<sup>59</sup>. Dental arch relationships at 5 years have been thought to predict treatment outcome in terms of facial growth on a population basis rather than at the individual child level<sup>60</sup>. The 5-year-old Index has, therefore, been used to

<sup>59</sup> 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.

<sup>60</sup> 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.

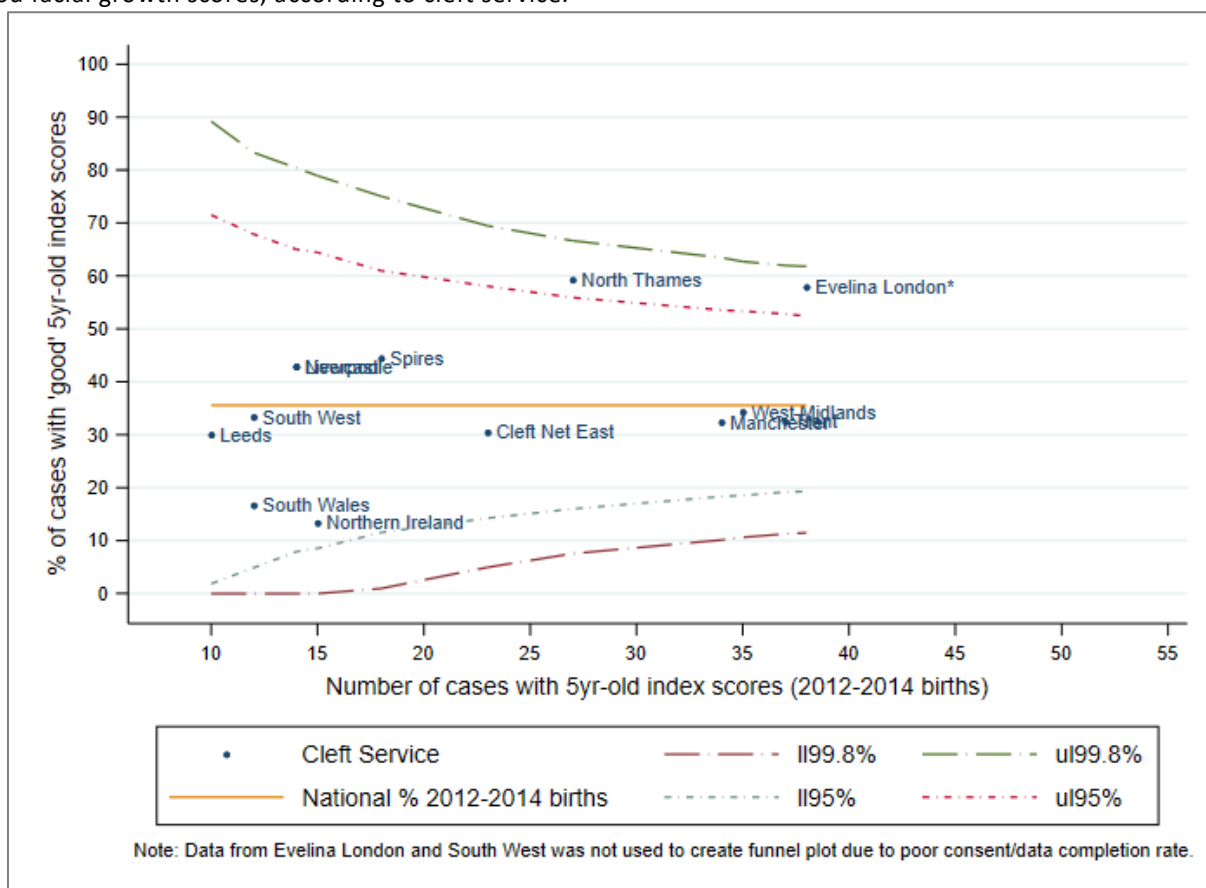
compare treatment outcomes between cleft services and surgeons. Patients scoring '1' and '2' on the index are considered to have good outcomes, while those scoring '4' and '5' are thought to have poor outcomes in terms of facial growth.

Among the 289 children with reported 5-year-old Index scores, 86% (249) 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, 37% had a 'fair' score, while 25% 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<sup>61</sup>.

### 4.3.3. 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 36%<sup>62</sup> (range 13%-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 2012 to 2014, who had good facial growth scores, according to cleft service.



**Note:** Registered in CRANE by 27 June 2022. Exclusions (not mutually exclusive): Children with submucous cleft palates, and those born with an incomplete UCLP. Funnel plot centred on the adjusted national percentage (35.6%) of children (born 2012-2014) with 'good' facial growth outcomes reported.

<sup>61</sup> Clinical Standards Advisory Group. Report of a CSAG Committee on cleft lip and/or palate, 1998, The Stationery Office, London.

<sup>62</sup> The overall mean was 38.4% and adjusted mean was 35.6% (excluding cleft services with consent verification and data completeness outlier status).

Submitting this indicator of ‘good’ dental arch relationships (#9) as measured by the 5-year-old Index to the outlier process did not lead to the identification of any outliers for this outcome indicator. **The supplementary tables** show the breakdown of cases with good, fair and poor scores at 5 years of age, according to cleft service for both 2012-2014 and 2015 cohorts.

The distribution of good, fair and poor scores continues to be variable between cleft services, as shown graphically in **the supplementary tables**. 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.

We have previously 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<sup>63</sup>. As such, ‘fair’ and ‘poor’ scores should be interpreted with caution at 5 years of age. This is why the CRANE 2020 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. The Orthodontic CEN of the CFSGB&I is currently looking at the feasibility of collecting and auditing dental relationship data prior to orthodontic intervention in the early adolescent period. It is hoped that a joint discussion with the Surgical CEN will happen at the CFSGB&I scientific conference in April 2023 to jointly agree a revised facial growth audit process to be put to CDG for agreement to be used in CRANE reporting.

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<sup>63</sup> 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: 2012 to 2014. Plus summary information on 2015 births <sup>64</sup>
	Denominator	1,141 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)	<ul style="list-style-type: none"> <li>• Children without consent to data collection</li> <li>• Children with an unspecified cleft type or with a cleft affecting only the lip</li> <li>• Children who died before the age of 5 years</li> <li>• Children with submucous cleft palates<sup>65</sup></li> <li>• Children with a diagnosed syndrome<sup>66</sup> entered onto the CRANE database</li> <li>• Children without all 16 CAPS-A scores</li> </ul>
	Outcome data completeness	<ul style="list-style-type: none"> <li>• 62% of 1,852 eligible children had all 16 CAPS-A speech parameters reported.</li> <li>• 2% had some but not all 16 CAPS-A speech parameters reported.</li> <li>• 29% had a reason the speech outcomes were not collected.</li> <li>• 7% were missing data or a reason for not collecting data.</li> </ul>
	Countries	England, Wales and Northern Ireland
	Indicators	#10, #11 & 12 <sup>67</sup>
Benchmark/ Standards	<ul style="list-style-type: none"> <li>• 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).</li> <li>• 61%<sup>68</sup> to have speech within the normal range (see main text for a full description of the standards)<sup>69</sup>.</li> <li>• 72%<sup>5</sup> to have speech without difficulties resulting from existing or previous structural anomalies.</li> <li>• 68%<sup>5</sup> to have speech without significant cleft-related articulation difficulties.</li> </ul>	
What did we find?	<ul style="list-style-type: none"> <li>• 61% achieved speech within the normal range.</li> <li>• 72% had speech without difficulties resulting from existing or previous structural anomalies.</li> <li>• 69% had speech without cleft-related articulation difficulties.</li> <li>• 18% of children had secondary surgery for speech purposes before the age of 5 years.</li> </ul>	
Recommendations	<ul style="list-style-type: none"> <li>• All children with a cleft affecting the palate should have their speech assessed and reported to CRANE.</li> <li>• Where possible, cleft services should investigate possibilities of remote methods for consensus listening.</li> <li>• All cleft services should work together to explore possible reasons for variation in speech outcomes.</li> <li>• The research community should undertake to develop risk stratification models for analysing speech outcomes among children with a cleft.</li> </ul>	

<sup>64</sup> Affected by the COVID-19 pandemic and the lockdown that ensued.

<sup>65</sup> Submucous cleft palate patients excluded from all 5-year outcomes as most teams do not audit these patients. Including cases with LAHSAL code identified as not possible, and likely submucous, when reviewed by cleft clinicians.

<sup>66</sup> Cases flagged as syndromic are excluded, with the exception of children with a recorded (named) congenital malformation of the circulatory system, or congenital malformation of the nervous system (e.g. microcephaly, spina bifida). This refinement to the methodological approach to reporting speech outcomes was introduced in January 2021.

<sup>67</sup> CRANE core indicators are detailed in [the supplementary tables](#).

<sup>68</sup> Benchmarks set using published speech outcome data from 2009-2011, CRANE Database 2018 Annual Report.

<https://www.crane-database.org.uk/reports/crane-database-2018-annual-report/>

<sup>69</sup> Britton L, Albery L, Bowden M, Harding-Bell A, Phippen G, and Sell D (2020) 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<sup>70</sup> (e.g. patient transferred out of area or patient did not attend appointment). **The supplementary tables** show the breakdown of speech reporting for each cleft service, for the 2012-14 birth cohort.

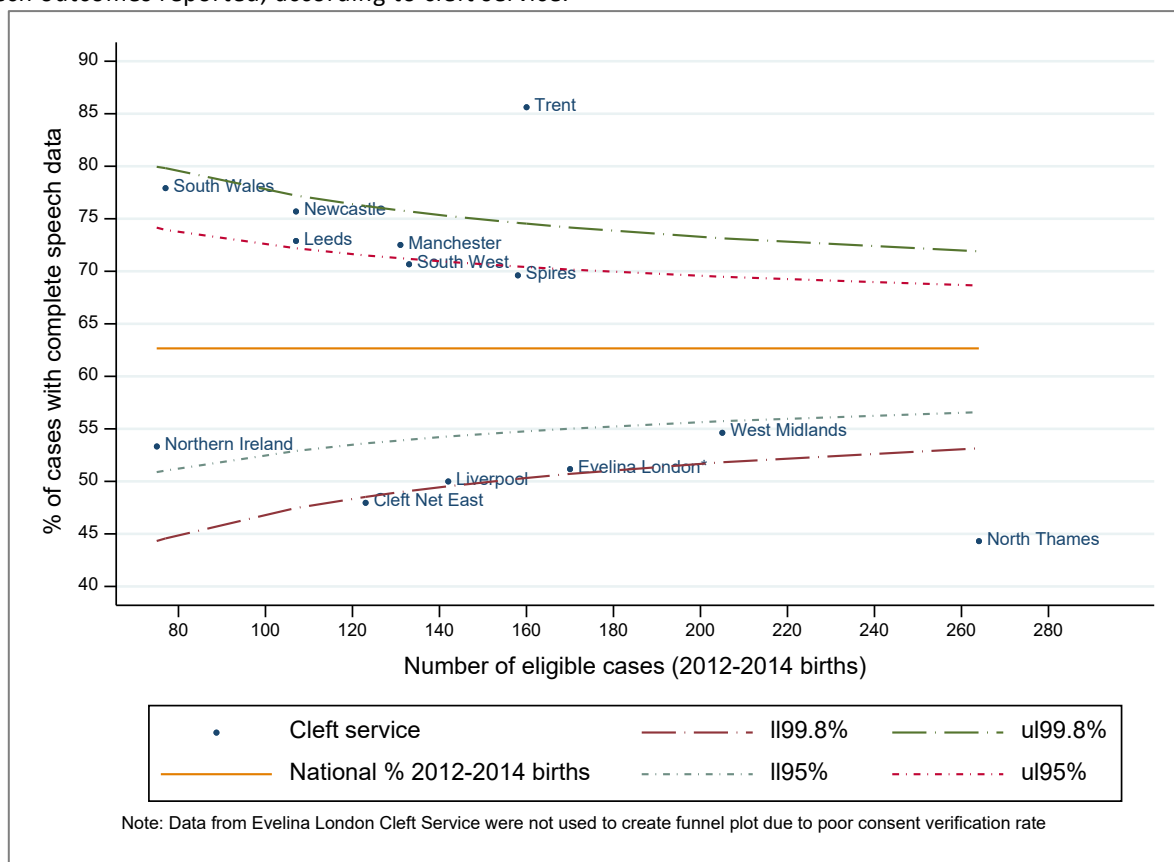
Of the 1,852 eligible cases, 62% (44%-86% range between services) had all 16 CAPS-A parameters reported. A further 2% of cases had some but not all 16 CAPS-A parameters reported. 29% of cases had a reason why the outcome was not reported, and 7% were missing data or a reason.

#### Reasons for not collecting 5-year-old speech outcome data for 2012-14 births

**The supplementary tables** show the breakdown of reasons reported for not collecting 5-year-old speech outcome data for 2012-14 births. The most common reason selected was 'Other' (42%). Comments provided to specify these 'Other' reasons, in the main, described clinics cancelled / data not collected due to the COVID-19 pandemic.

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 national percentage meeting the standard of 63%<sup>71</sup>.

**Figure 4.11.** Funnel plot showing the percentage of CRANE-consented 5-year-olds, born 2012 to 2014, with speech outcomes reported, according to cleft service.



**Note:** Registered in CRANE by 27 June 2022. Funnel plot centred on the adjusted national percentage (62.7%) of children (born 2012-2014) with speech outcomes reported. See **the supplementary tables** for the raw data (and exclusions) used to create this funnel plot.

<sup>70</sup> 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

<sup>71</sup> The unadjusted national percentage was 61.6% and adjusted percentage with speech outcome data was 62.7% (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, particularly during the COVID pandemic.

The funnel plot shows that North Thames and Cleft Net East are negative outliers 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 national percentage presented in the subsequent outcome funnels. Results from these regions should therefore be interpreted with caution. Completing this process of identifying outliers has also allowed the identification of positive outliers such as the Trent cleft service. See Box 4.5 and 4.6 (below) for comments from cleft services on their outlier status.

**Box 4.5.** Summarised response to being a negative outlier for ‘speech – outcome data completeness at 5 years’. **Full responses published in the “2022 Annual Report: Responses to outlier process” document.**

“Having reviewed the data set of the missing outcomes a number of patients attended for audit but were unable to co-operate or undertake a speech assessment (25 pts. 43%). In addition, a number of patients listed on CRANE were no longer known to the service with a closed referral (15%) or had transferred out of the North Thames Network. There is also a small number who DNA or declined assessment. Sadly, a number had passed away and this should have been updated on to the CRANE database. It is acknowledged that all these factors should have been recorded on the CRANE Database and we intend to update the CRANE dataset in the coming months.

The outlier report has provided us with an opportunity to review our systems across the North Thames Regional Network and we aim to provide 100% data completeness for the 2016 cohort. In addition, we will cross check our 2016 data set against the CRANE Database to ensure we are capturing all our patients within the Network. We have established a monthly meeting with our Data Manager and Audit Lead to ensure we continue to meet our obligations to CRANE and the patients we serve. Our thanks to CRANE for enabling us to reflect and seek solutions for the service.”

**North Thames cleft service, November 2022.**

Read the full responses in the [“2022 Annual Report: Responses to outlier process”](#) document published online.

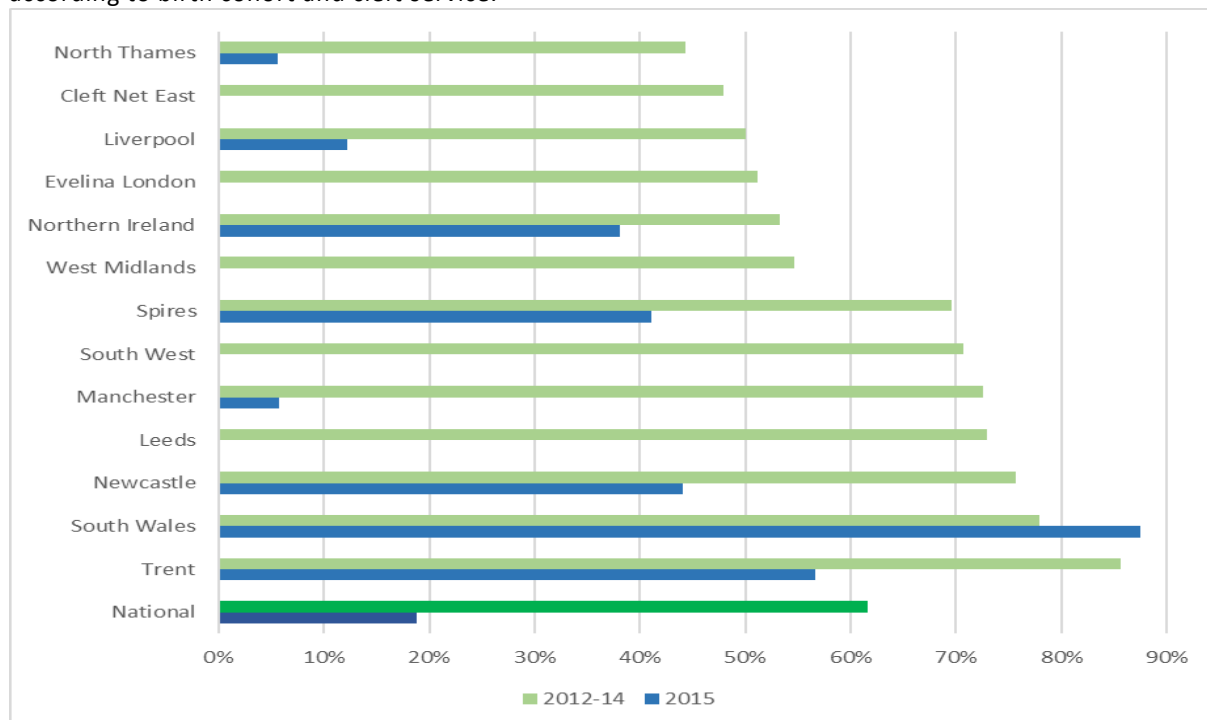
**Box 4.6.** Summarised responses to being positive outliers for ‘speech – outcome data completeness at 5 years’. **Full responses published in the “2022 Annual Report: Responses to outlier process” document.**

“In Trent we have a rolling system of collecting and analysing speech audits. The specialist speech and language therapists (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 two CAPSA trained listeners ... Our stable listening group and rolling system of listening also helped our performance.”

**Trent cleft service, November 2022.**

Anticipating ongoing challenges to outcome data completeness due to the COVID-19 pandemic, Figure 4.12 summarises outcome data completeness for eligible children born in 2015 (reporting of all 16 CAPS-A speech parameters) compared with 2012-2014 data. This information is also available in [the supplementary tables](#).

**Figure 4.12.** Percentage of CRANE-consented 5-year-olds who had all 16 CAPS-A speech parameters reported, according to birth cohort and cleft service.



**Note:** Registered in CRANE by 27 June 2022. See [the supplementary tables](#) for the raw data (and exclusions) used to create this graph.

Overall, of the 575 eligible 5-year-old children born in 2015, only 19% had all 16 CAPS-A speech parameters reported (0%-88% range between services). Only two 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 2015 births**

[The supplementary tables](#) show the breakdown of reasons reported for not collecting 5-year old speech outcome data for 2015 births. The most common reason selected was ‘Other’ (73%). 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 2012-2014 are presented in [the supplementary tables](#).

## Resonance and nasal airflow

In terms of resonance, 6% of children had moderate or severe hypernasality i.e. nasal sounding speech<sup>72</sup>. 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% (956/1,141) of children had ratings indicating that no structural problems existed in relation to resonance and nasal airflow by their 5-year assessment<sup>73</sup>. In order to achieve these ratings, 204 (18%) out of 1,122 children with reported surgical data<sup>74</sup> had undergone secondary surgery for speech purposes before the age of 5 years. Of these, 146 (72%) had resonance and nasal airflow ratings that indicated there were no structural problems that existed in relation to these parameters, suggesting that the secondary surgery had resolved these speech issues in time for starting school.

## Cleft Speech Characteristics (CSCs)

Out of the 1,141 children (born 2012-14) with reported ratings for all 12 cleft speech characteristics, 67% had ratings indicating they did not exhibit any CSCs<sup>75</sup>. ‘Palatalisation / Palatal’ anterior oral CSCs were the most commonly occurring CSC, affecting 24% of children (12% with ratings of one or two consonants affected (light green ratings) and 12% 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 [the supplementary tables](#) 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<sup>76</sup>:

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.

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<sup>72</sup> With a hypernasality score of ‘3’ or ‘4’ (red ratings).

<sup>73</sup> All green ratings of ‘0’ or ‘1’.

<sup>74</sup> VP surgery/fistula repair data was reported for 1122/1141 (98%) eligible children.

<sup>75</sup> All green ratings of ‘A’ and in selected cases of ‘B’ – as per [the supplementary tables](#).

<sup>76</sup> 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.

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).

In 2018 it was agreed with the Speech and Language Therapists that the benchmark for each of the three Speech Outcome Standards above would be set using speech data from the three preceding years. Therefore, for this year's benchmark we have used the speech outcomes achieved by 5 year olds born 2009 to 2011, published in the CRANE 2018 Annual Report<sup>77</sup>.

#### 4.4.3. Speech outcomes by cleft service

##### *Normal speech*

Out of the 1,141 children (born 2012-2014) with reported ratings for all 16 CAPS-A speech parameters, 60.6% of children across all units achieved the National Speech Outcome Standard 1 (this is the unadjusted national percentage meeting the standard). They had normal (green) ratings across all 16 CAPS-A speech parameters.

In line with the outlier process (implemented for the first time in 2021), the adjusted national percentage meeting the standard was 61.5% (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 2018 Annual Report.

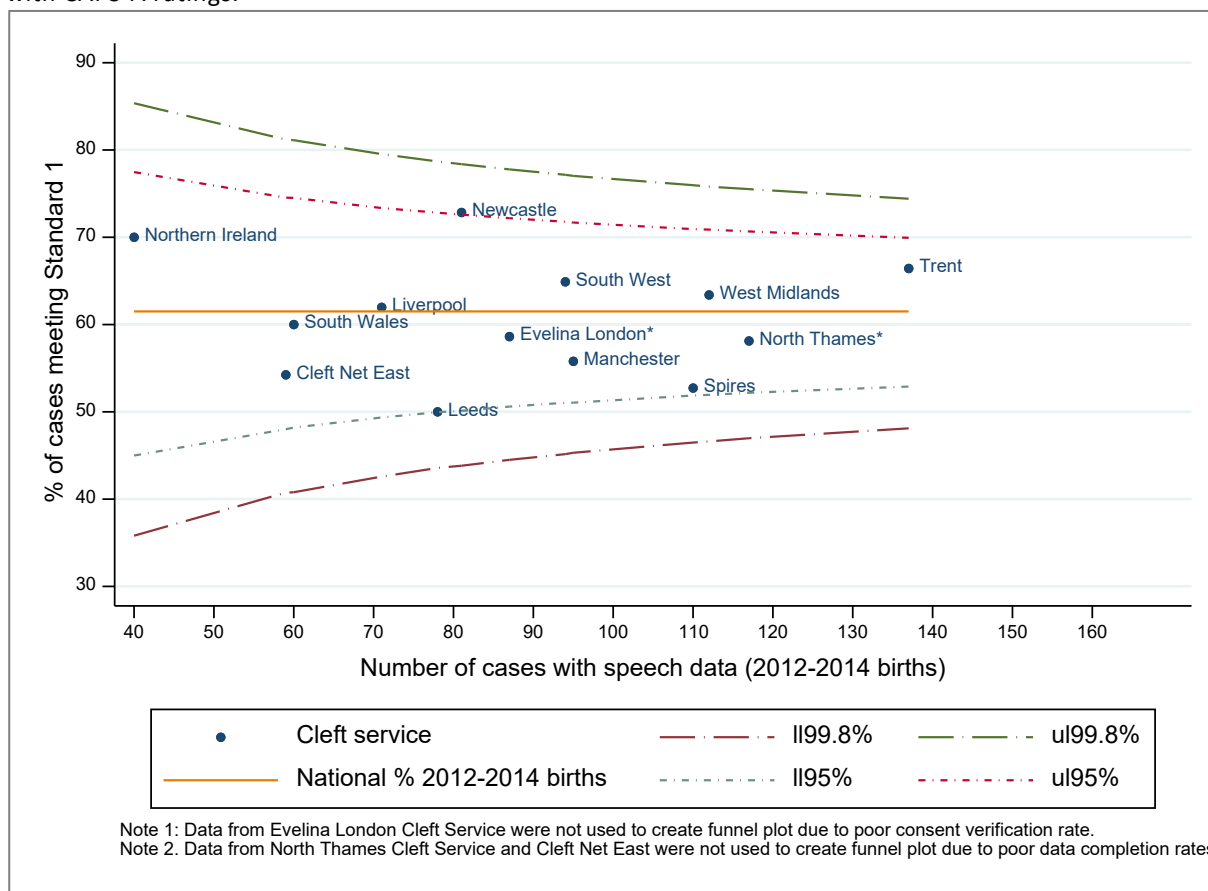
The funnel plot<sup>78</sup> in Figure 4.13 (see [the supplementary tables](#) for raw data) shows the percentage of children (born in 2012-2014) achieving normal speech according to the number of audited children within each cleft service with ratings for all 16 CAPS-A speech parameters (more information on funnel plots can be found in the glossary). The funnel plot is centred on the adjusted percentage meeting the standard of 61.5% (range 50%-72.8%).

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<sup>77</sup> CRANE Project team (2018) Cleft Registry and Audit Network (CRANE) Database 2018 Annual Report. London: Clinical Effectiveness Unit, The Royal College of Surgeons of England

<sup>78</sup> This funnel plot is calculated using valid data as denominators (not considering missing data) and is not adjusted (or risk adjusted) in any way.

**Figure 4.13.** Funnel plot showing the percentage of CRANE-consented 5-year-olds, born 2012 to 2014, with ratings suggesting speech within the normal range, according to the number of children in each cleft service with CAPS-A ratings.



**Note:** Registered in CRANE by 27 June 2022. Funnel plot centred on the adjusted national percentage (61.5%) of children (born 2012-2014) with speech rated within the normal range. See [the supplementary tables](#) 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).

### *Absence of structurally-related speech difficulties*

Out of the 1,141 children (born 2012-2014) with reported ratings for all 16 CAPS-A speech parameters, 70% of children across all services achieved the National Speech Outcome Standard 2a (this is an unadjusted national percentage). 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.

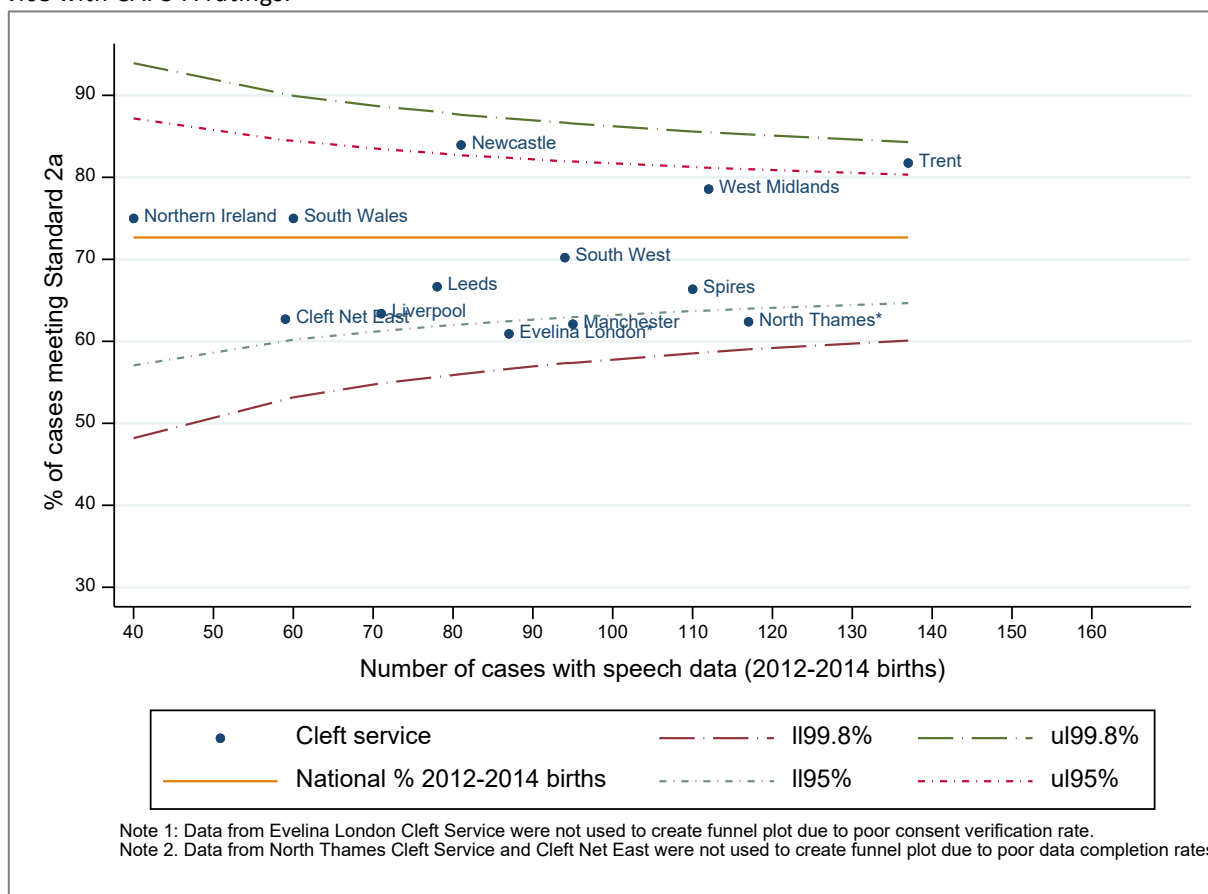
In line with the outlier process (implemented for the first time in 2021), the adjusted national percentage meeting the standard was 73% (excluding services with consent and outcome data quality rates not sufficient for inclusion), as presented in Figure 4.14. This means that both the unadjusted and adjusted results are in line with the National Speech Outcome Standard 2a benchmark of 72%, reported in the CRANE 2018 Annual Report.

Figure 4.14 (see [the supplementary tables](#) for raw data) shows the percentage of 5-year-olds with speech ratings that suggest they do not have structurally-related speech difficulties<sup>79</sup>, according to the number of

<sup>79</sup> 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.

children at each service with CAPS-A ratings. The funnel plot is centred on the adjusted national percentage of 73% (range 61%-84%) meeting the standard.

**Figure 4.14.** Funnel plot showing the percentage of CRANE-consented 5-year-olds, born 2012 to 2014, with ratings suggesting no structurally-related speech difficulties, according to the number of children at each service with CAPS-A ratings.



**Note:** Registered in CRANE by 27 June 2022. Funnel plot centred on the adjusted national percentage (72.7%) of children (born 2012-2014) without structurally-related speech difficulties. See [the supplementary tables](#) for the raw data (and exclusions) used to create this funnel plot.

The funnel plot shows that there is variability between services in the percentage of children without structurally-related speech difficulties. In particular, Newcastle and Trent are above the upper 95% control limit with reported outcomes for over 75% of their eligible cases (See [the supplementary tables](#)), and the care and service provision offered by these services may be worth investigating for best practice recommendations.

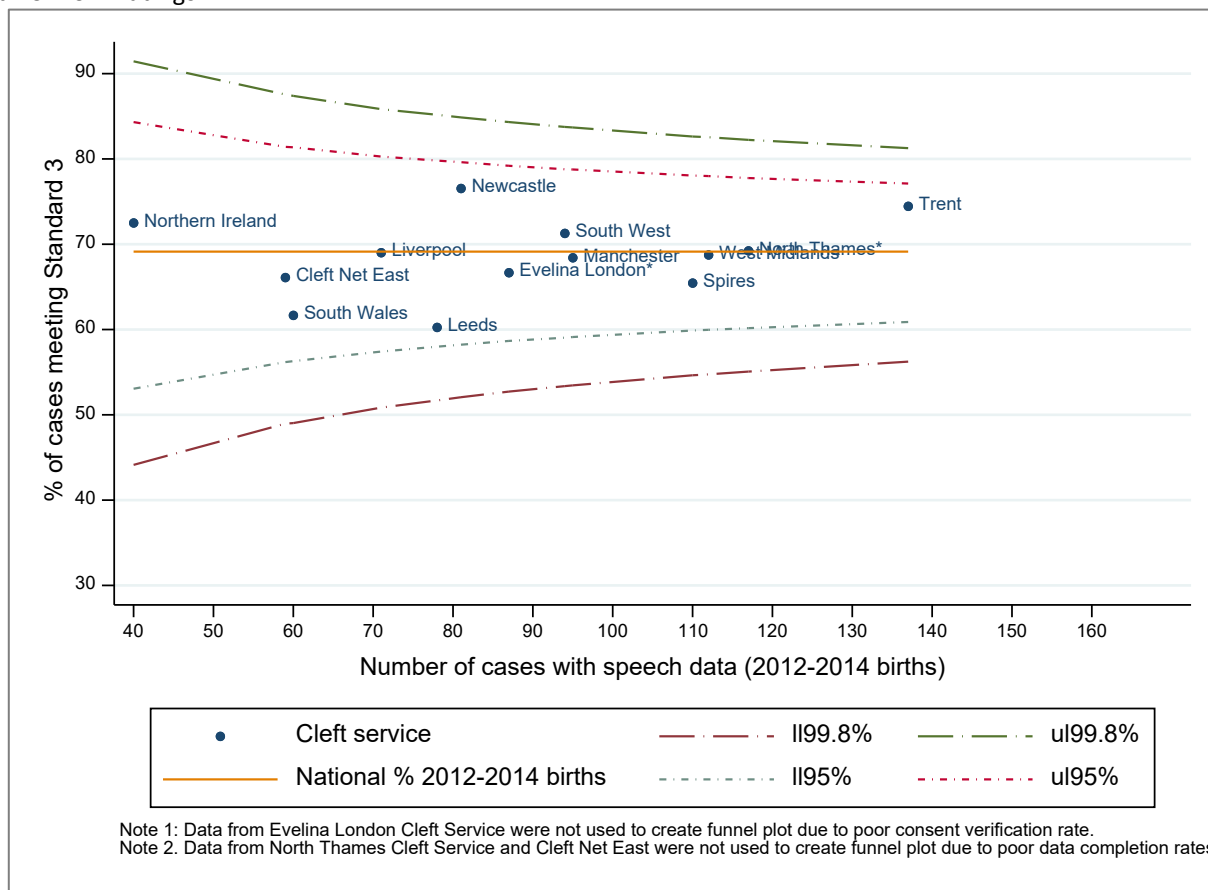
### Absence of cleft-related articulation difficulties

Out of the 1,141 children (born 2012-2014) with reported ratings for all 16 CAPS-A speech parameters, 68.8% of children across all cleft services achieved the National Speech Outcome Standard 3 (this is an unadjusted national percentage). 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 (implemented for the first time in 2021), the adjusted national percentage meeting the standard was 69.1% (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 2018 Annual Report.

Figure 4.15 (see [the supplementary tables](#) for raw data) shows the percentage of 5-year-olds with speech ratings that suggest they do not have cleft-related articulation difficulties<sup>80</sup>, according to the number of children within each service with CAPS-A ratings. The funnel plot is centred on the adjusted percentage meeting the standard of 69% (range 60%-77%). All services fall within the 95% control limits.

**Figure 4.15.** Funnel plot showing the percentage of CRANE-consented 5-year-olds, born 2012 to 2014, with ratings suggesting no cleft-related articulation difficulties, according to the number of children at each service with CAPS-A ratings.



**Note:** Registered in CRANE by 27 June 2022. Funnel plot centred on the overall national percentage (69.1%) of children (born 2012-2014) without cleft-related articulation difficulties. See [the supplementary tables](#) for the raw data (and exclusions) used to create this funnel plot.

Despite considerations relating to missing data and the lack of formal adjustment for known independent determinants of outcome, (see chapter 5, Database development work), current presentation of Figures 4.13, 4.14 and 4.15 as funnel plots centred on adjusted national percentages is currently the agreed method (at this time<sup>81</sup>) to check whether or not any service deviates significantly from the expected standards.

**Box 4.7.** 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, December 2022.

<sup>80</sup> No cleft type speech characteristics requiring SLT and/or surgery.

<sup>81</sup> 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: 2012 to 2014. Plus summary information on 2015 births <sup>82</sup>
	Denominators	<ul style="list-style-type: none"> <li>2,812 5-year-old children eligible for psychology audit</li> <li>2,077 children with recorded date of face-to-face psychosocial screen (either at the 5-year audit or at an earlier screening)<sup>83</sup></li> <li>1,834 children with recorded Tiers of Involvement Measure (TIM) scores</li> <li>1,586 children with recorded Strengths and Difficulties Questionnaire (SDQ) scores</li> </ul>
	Numerators	<ul style="list-style-type: none"> <li>1,549 5-year-old children with all eight psychology 5-year-audit data items</li> <li>Number of children with at least one face-to-face psychosocial screen before age 6</li> <li>Number of children with a particular TIM level of psychological involvement</li> <li>Number of children with a particular range of SDQ 'Total difficulties' scores</li> </ul>
	Exclusions (not mutually exclusive)	<ul style="list-style-type: none"> <li>Children without consent to data collection</li> <li>Children who died before the age of 5 years</li> <li>Children with submucous cleft palates<sup>84</sup></li> <li>Children without a recorded date of first face-to-face psychosocial screening</li> </ul>
	Data completeness	<ul style="list-style-type: none"> <li>55% of 2,812 eligible children had all psychology data items reported (complete data)</li> <li>11% had some but not all eight scores reported (incomplete data)</li> <li>25% had a reason psychology data was not collected</li> <li>9% were missing data or a reason for not collecting data</li> </ul>
	Countries	England, Wales and Northern Ireland
	Indicators	#13 & #14 <sup>85</sup>
Benchmarks	<ul style="list-style-type: none"> <li>100% of children should have recorded psychology 5-year audit data (or a recorded reason outcome not provided).</li> <li>100% of children should be screened at least once before the age of 6 years.</li> <li>100% of children should be seen by a psychologist and have a TIM assessment at 5 years of age<sup>38</sup>.</li> <li>SDQ population norms: 10% of children aged 5 to 10 years old have SDQ scores that are 'high' or 'very high'.</li> </ul>	
What did we find?	<ul style="list-style-type: none"> <li>96% 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.</li> <li>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+).</li> <li>18% of children with a documented SDQ score had 'high' or 'very high' scores. These percentages are higher than the 10% SDQ population norms.</li> </ul>	
Recommendation	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).	

<sup>82</sup> Affected by the COVID-19 pandemic and the lockdown that ensued.

<sup>83</sup> Dates of 'psychological screening at age five' were prioritised. Where unavailable, 'date of first face-to-face screening' used.

<sup>84</sup> 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.

<sup>85</sup> 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<sup>86</sup> (e.g. patient transferred out of area or patient did not attend appointment, etc). **The supplementary tables** show the breakdown of psychology reporting for each cleft service for both the 2012-14 and 2015 birth cohorts.

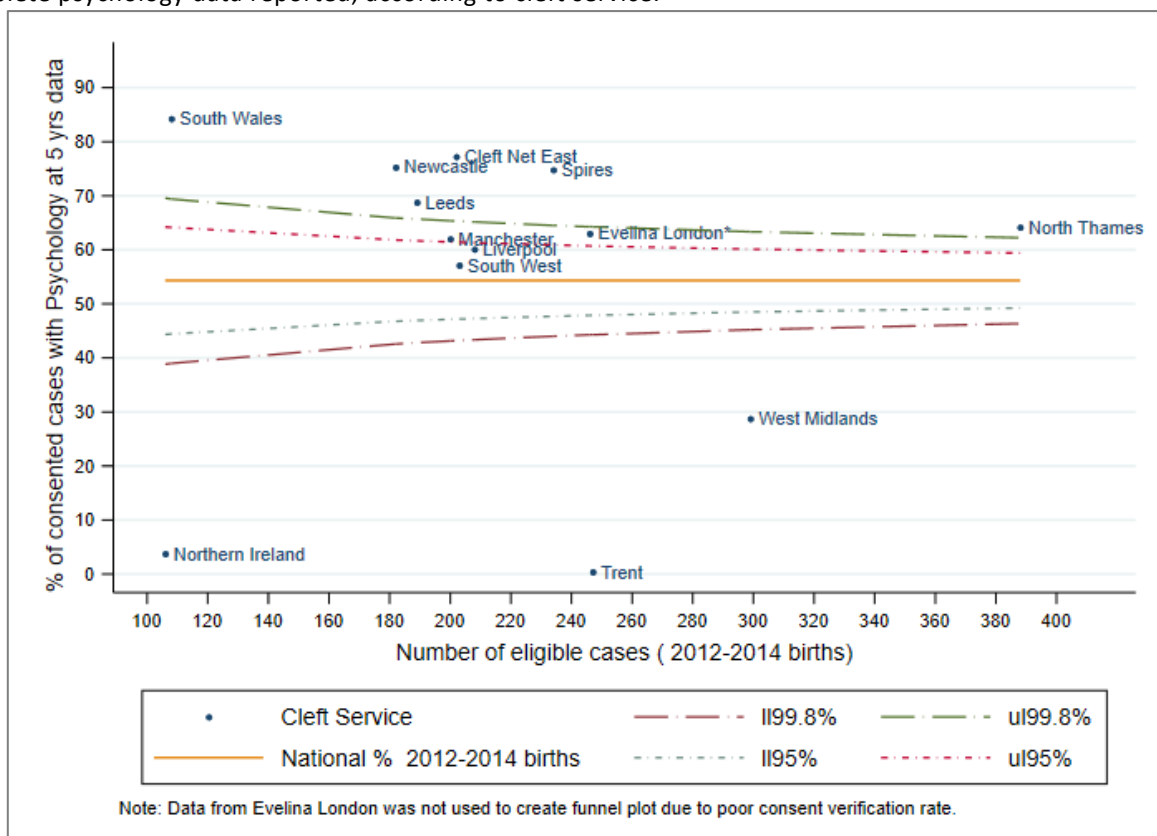
Of the 2,8122 eligible 5-year-old children born between 2012-14, 55% had all eight psychology 5-year audit<sup>87</sup> data items reported (0.4%-84% range between services). A further 11% of cases had some but not all eight psychology screening data items reported. Twenty-five percent of cases had a reason why the outcome was not reported, and 9% were missing data or a reason.

#### Reasons for not collecting 5-year old psychology outcome data for 2012-14 births

**The supplementary tables** show the breakdown of reasons reported for not collecting 5-year old psychology outcome data for 2012-14 births. The most common reason selected was 'Lack of staff facilities or equipment' (42%).

Figure 4.16 shows the variability in the reporting of all eight psychology 5-year audit data items according to the number of eligible cases within services. The funnel plot is centred on the adjusted national average of 54%<sup>88</sup>.

**Figure 4.16.** Funnel plot showing the percentage of CRANE-consented 5-year-olds, born 2012 to 2014, who had complete psychology data reported, according to cleft service.



**Note:** Registered in CRANE by 27 June 2022. Funnel plot centred on the adjusted national percentage (54.3%) of children (born 2012-2014) with psychology outcomes reported. See **the supplementary tables** for the raw data (and exclusions) used to create this funnel plot.

<sup>86</sup> 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

<sup>87</sup> 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).

<sup>88</sup> The overall mean was 51.1% and adjusted mean was 54.3% (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, the West Midlands and Northern Ireland are negative outliers for the percentage of eligible cases with all eight psychology 5-year audit data items 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. Completing this process of identifying outliers has also allowed the identification of positive outliers such as Newcastle, Leeds, Cleft Net East, Spires and South Wales cleft services. See Box 4.8 (below) for comments from cleft services on their outlier status.

**Box 4.8.** Summarised responses to being positive outliers for ‘psychological wellbeing - data completeness at 5 years’. **Full responses published in the “2022 Annual Report: Responses to outlier process” document.**

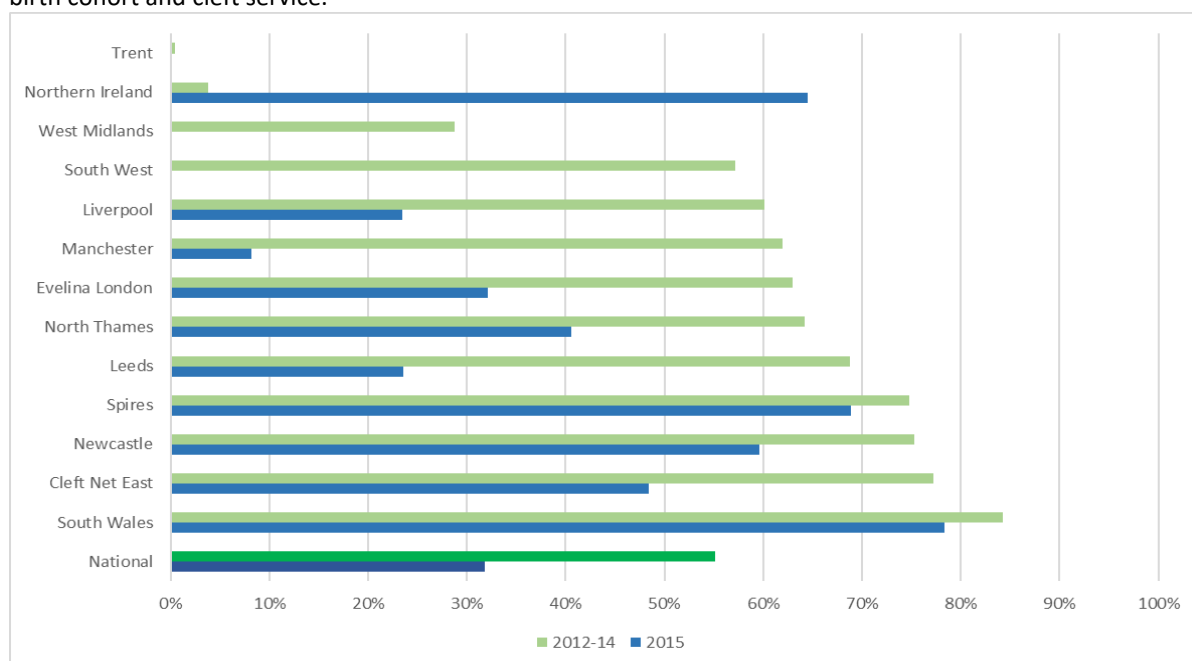
“As a team we have worked hard to identify gaps in our data completeness and to rectify this. This has involved going back through the medical notes for some patients which has been time consuming. In order to avoid this for 2015 data and for the future, we have implemented a proforma at the designated 5 year old clinics... The data from the proforma is inputted onto the CRANE database by our designated secretary, responsible for recording the data. We hope this will continue to support data completeness for our unit.”

**South Wales cleft service, November 2022.**

Anticipating ongoing challenges to data completeness due to the COVID-19 pandemic, Figure 4.17 summarises data completeness for eligible children born in 2015 (reporting of all eight psychology 5-year audit data items) compared with 2012-2014 data. [The supplementary tables](#) show the breakdown of psychology reporting for each cleft service for the two cohorts.

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 the ability of services to audit and submit the most recent year of CRANE data collection.

**Figure 4.17.** Percentage of CRANE-consented 5-year olds who had complete psychology data reported, according to birth cohort and cleft service.



**Note:** Registered in CRANE by 27 June 2022. See [the supplementary tables](#) for the raw data (and exclusions) used to create this graph.

Overall, of the 891 eligible 5-year-old children born in 2015, 32% had all eight psychology 5-year audit data items reported (0%-78% range between services). Only four 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 2015 births**

The supplementary tables show the breakdown of reasons reported for not collecting 5-year old psychology outcome data for 2015 births. The most common reason selected was 'Other' (51%). Comments provided to specify these 'other' reasons, in the main, described clinics cancelled / data not collected due to the COVID-19 pandemic and virtual appointments.

#### **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<sup>89</sup>, 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'.

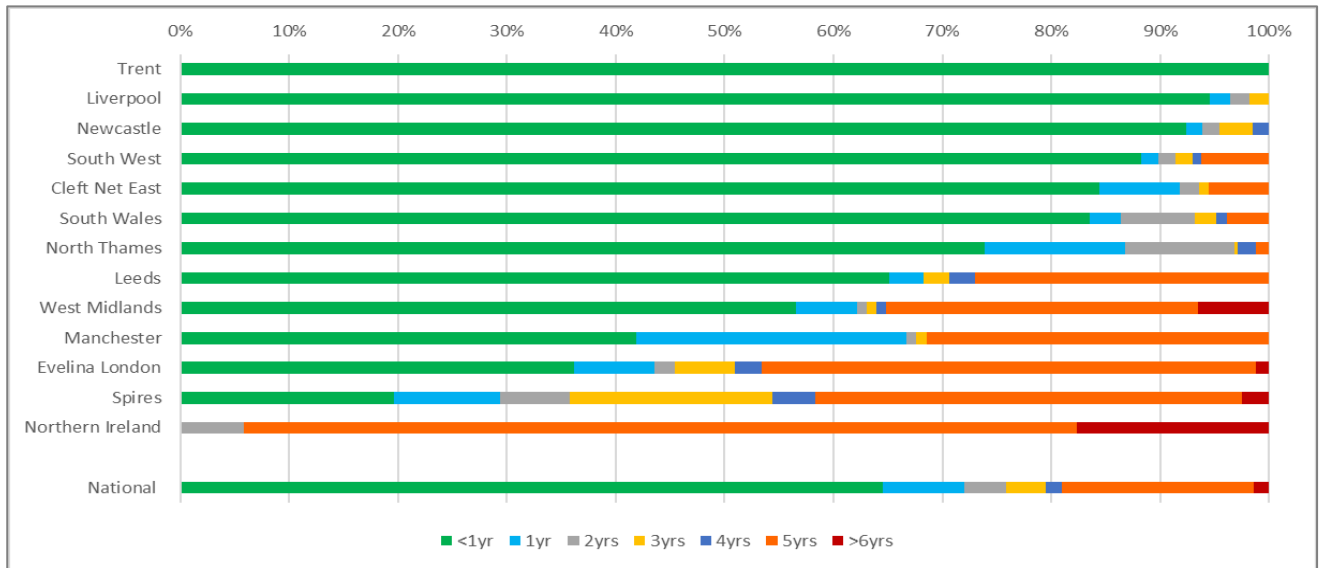
The 'date of first face-to-face screening' was recorded for 1,884 children. Where this date was unavailable, date of 'psychological screening at age five' was included in the analyses (adding information for 193 cases).

As shown in Figure 4.18, 63% of families of children born in 2012-14 were seen before their child's first birthday (this ranged from 0% to 100% according to cleft service). Furthermore, 7% were seen at age one, 3% were seen at age two, 4% were seen at age three, and 1% were seen at age four. The remaining 21% were first seen at the age of 5 or older.

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<sup>89</sup> 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/>

**Figure 4.18. Age of first face-to-face psychological screen received by CRANE-consented 5-year olds, born 2012 to 14, according to cleft service.**



**Note:** Registered in CRANE by 27 June 2022. Exclusions (not mutually exclusive): Children who died before the age of 5 years and those with submucous cleft palates.

**Dates of ‘psychological screening at age five’** were recorded for 1,771 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 306 cases). Based on this information, 97% children were confirmed as having at least one face-to-face psychosocial screen before the age of 6 years<sup>90</sup>.

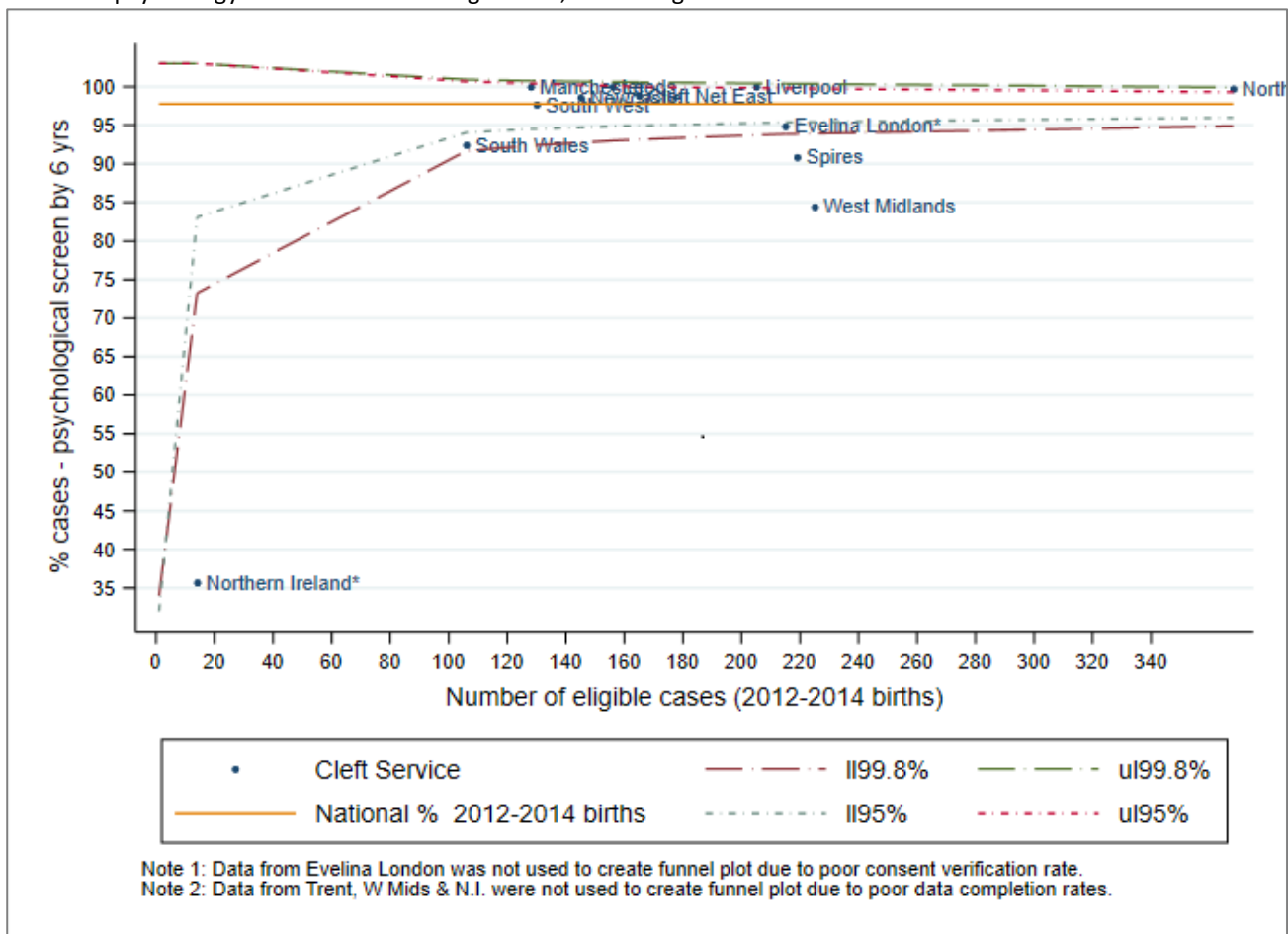
Figure 4.19 shows the percentage of 5-year-olds with at least one psychology screen before the age of six, according to the number of eligible children at each cleft service, with the funnel plot centred on the adjusted national average of 97%<sup>91</sup> (range 36%-100%).

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

<sup>90</sup> Cases with recorded date of psychosocial screen prior to their date of birth were excluded from reporting.

<sup>91</sup> The overall mean was 96.7% and adjusted mean was 96.8% (excluding cleft services with consent verification and data completeness outlier status).

**Figure 4.19.** Funnel plot showing the percentage of CRANE-consented 5-year-olds, born 2012 to 2014, with at least one psychology screen before the age of six, according to cleft service.



**Note:** Registered in CRANE by 27 June 2022. Funnel plot centred on the adjusted national percentage (96.8%) of children (born 2012-2014) with at least one psychology screen before the age of six. See [the supplementary tables](#) for the raw data (and exclusions) used to create this funnel plot.

The funnel plot shows that the West Midlands, Spires and Northern Ireland are negative outliers for the percentage of eligible cases with at least one psychology screen before the age of six. This is because they fall below the lower 99.8% control limit on the funnel plot. [The supplementary tables](#) show the breakdown of cases with reported psychology data at 5 years of age, meeting each psychology process or outcome standard, according to cleft service. The information is presented for birth 2012-14 and 2015 birth cohorts.

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

Overall, 93% of the 1,834 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.

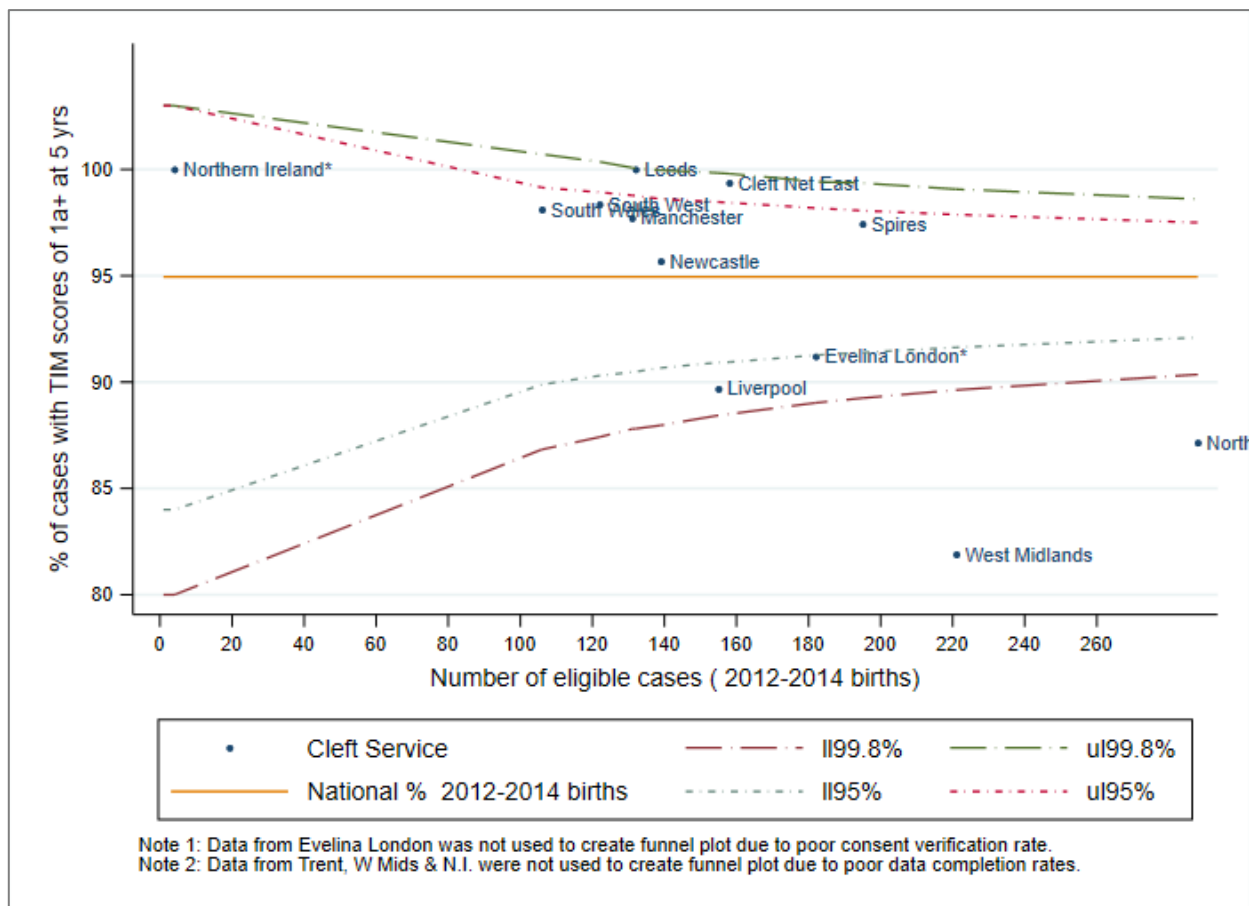
Psychological input provided in clinic can be preventative or in response to a concern raised by the family. Examples of preventative input include advice on talking to your child about their cleft, helping children and parents to prepare for potential comments and questions about their cleft. Examples 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 95%<sup>92</sup> (range 82%-100%).

**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<sup>93</sup>).
2. Psychological input provided in clinic (for the purposes of this report, TIM scores 2a, 2b and 2c are included in this category<sup>94</sup>).
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<sup>95</sup>).

**Figure 4.20.** Funnel plot showing the percentage of CRANE-consented 5-year-olds, born 2012 to 2014, with a TIM score of 1a+, according to cleft service.



**Note:** Registered in CRANE by 27 June 2022. Funnel plot centred on the adjusted national percentage (95%) of children (born 2012-2014) with a TIM score of 1a+. See the [supplementary tables](#) for the raw data (and exclusions) used to create this funnel plot.

<sup>92</sup> The overall mean was 93.0% and adjusted mean was 95% (excluding cleft services with consent verification and data completeness outlier status).

<sup>93</sup> 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.

<sup>94</sup> 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.

<sup>95</sup> 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.

The funnel plot shows that the West Midlands and North Thames are negative outliers 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.

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 [the supplementary tables](#).

[The supplementary tables](#) show 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 2012-14 births as well as 2015 births. See Box 4.9 (below) for comments from cleft services on their outlier status.

**Box 4.9.** Summarised responses to being negative outliers for ‘low rates of children identified as having a TIM score of 1a+’. **Full responses published in the “2022 Annual Report: Responses to outlier process” document.**

“We have identified unforeseen absences in staff and incorrect data entry as the main culprits. The introduction of new members of staff to the team can occasionally be delayed due to trust processes. This has led to occasional gaps in the provision of psychology support which is evident from the above. Secondly, we recognise that sense checking the data is important as we have identified some mistakes in the data entered. Since first recognising this, we have introduced quality assessment of each birth year to ensure the diagnoses are correct for each of our babies and that they have been entered correctly on CRANE. As we move into the successive years we were expecting to see the benefit of these quality improvements.”

**North Thames cleft service, November 2022.**

#### 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<sup>96</sup>.

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 | 3. High      |
| 2. Slightly raised  | 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 1,586 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)<sup>97</sup>.

<sup>96</sup> 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](http://www.sdqinfo.com)

<sup>97</sup> 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.

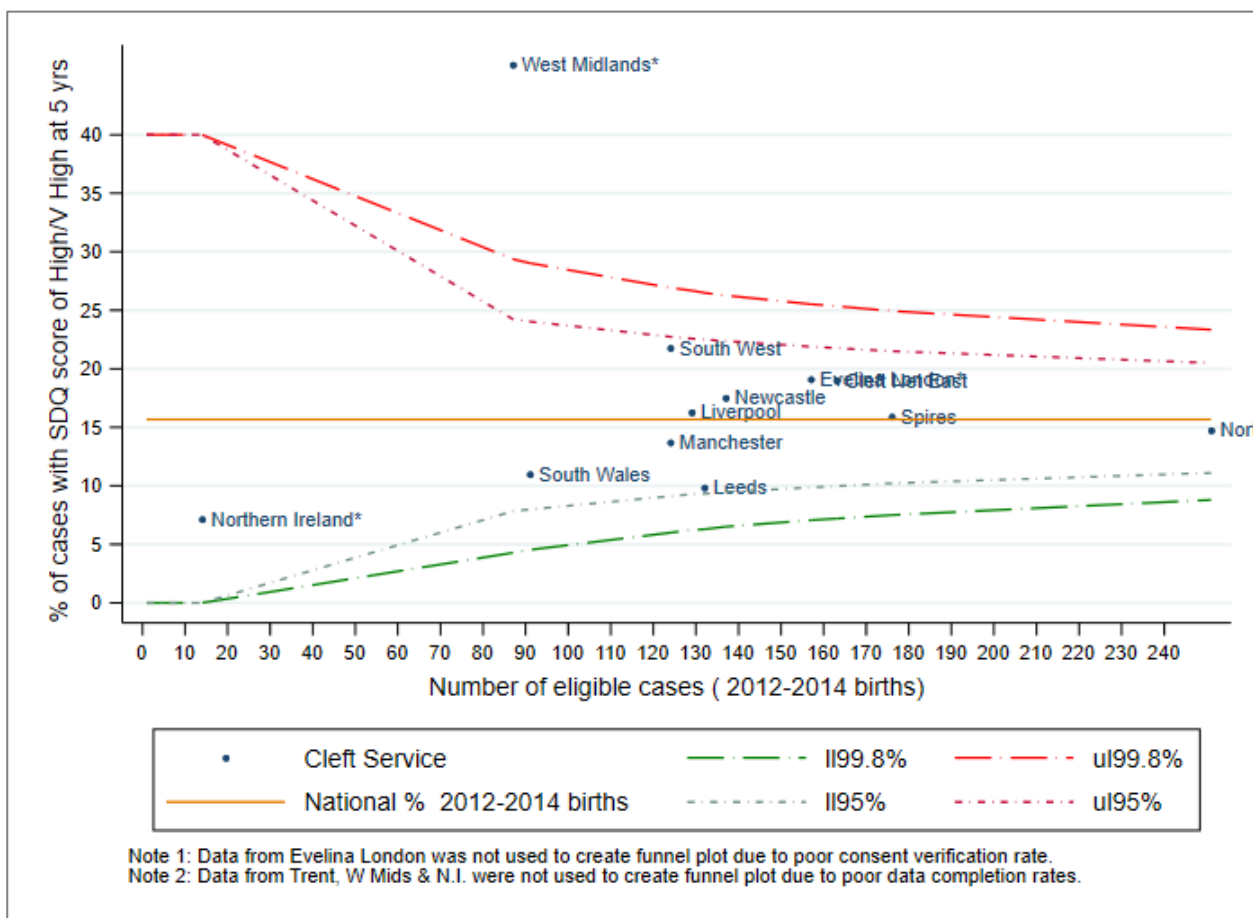


Figure 4.21 shows the percentage of 5-year-olds with a TIM score of 1a+, according to the number of eligible children at each cleft service. The funnel plot is centred on the adjusted national average of 16%<sup>98</sup> (range 0%-46%).

The funnel plot shows that the West Midlands is a negative outlier, 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.

The supplementary tables show 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) did not lead to the identification of positive outliers.

**Figure 4.21.** Funnel plot showing the percentage of CRANE-consented 5-year olds, born 2012 to 2014, with a high/very high SDQ score, according to cleft service.



**Note:** Registered in CRANE by 27 June 2022. Funnel plot centred on the adjusted national percentage (15.7%) of children (born 2012-2014) with a high/very high SDQ score. See the supplementary tables for the raw data (and exclusions) used to create this funnel plot.

<sup>98</sup> The overall mean was 17.8% and adjusted mean was 16.7% (excluding cleft services with consent verification and data completeness outlier status).

# 5. Database development work

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In this chapter, we provide information on recent work undertaken by CRANE that has furthered our understanding of clefting and its related outcomes. We highlight peer-reviewed publications and conference presentations that have either been published or taken place in the last year. We also provide information on recent work aimed at developing the Database and its functionality for patients, clinicians and care providers.

## 5.1 Publications and presentations

### 5.1.1 Peer-reviewed publications

**Investigating the impact of patient-related factors on speech outcomes at 5 years of age in children with a cleft palate.** Sophie Butterworth, Kate J Fitzsimons (Joint first author), Jibby Medina, Lorraine Britton, Stephanie Van Eeden, Muhammad Ally Hussein Wahedally, Min Hae Park, Jan H van der Meulen and Craig JH Russell. Published online 23 Jun 2022, The Cleft Palate-Craniofacial Journal (CPCJ).

<https://journals.sagepub.com/doi/abs/10.1177/10556656221110094>

**Range and frequency of congenital malformations among children with cleft lip and/or palate.** Kate J Fitzsimons, Mark J Hamilton, Jan H van der Meulen, Jibby Medina, Muhammad Ally Hussein Wahedally, Min Hae Park, Craig JH Russell. Published online 5 April 2022, The Cleft Palate-Craniofacial Journal (CPCJ).

<https://journals.sagepub.com/doi/abs/10.1177/10556656221089160>

**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. Published 24 November 2021, PLoS One. <https://doi.org/10.1371/journal.pone.0259820>

### 5.1.2 Conference presentations

International Congress of Cleft Lip, Palate and Related Craniofacial Anomalies, Edinburgh, July 2022

1) **Identifying determinants of speech outcome for children born with cleft palate +/- lip in England, Wales and Northern Ireland.** Sophie Butterworth, Kate J Fitzsimons, Lorraine Britton, Simon Van Eeden, Jibby Medina, Muhammed HA Wahedally, Min Hae Park, Jan H van der Meulen, Craig JH Russell

2) **Educational attainment among children with non-syndromic clefts: a longitudinal study.** Min Hae Park, Kate J Fitzsimons, Scott Deacon, Jibby Medina, Muhammed HA Wahedally, Sophie Butterworth, Craig JH Russell, Jan H van der Meulen

3) **Improvement in speech outcomes in England, Wales and Northern Ireland (2001-2012).** Lorraine Britton, Melanie Bowden, Kate Fitzsimons, Jibby Medina, Ginette Phippen, Marie Pinkstone, Craig Russell, Lucy Southby

4) **Timely Diagnosis of cleft palate in newborns: A population-based cohort study using CRANE Database registrations for England, Wales and Northern Ireland.** Megan Anderson, Jibby Medina, Kate J Fitzsimons,

Sophie Butterworth, Muhammed HA Wahedally, Min Hae Park, Jan H van der Meulen, Craig JH Russell, Scott Deacon

**5) Laterality of orofacial clefts and child growth, dental health, facial growth, speech and psychology in relation to outcomes at 5 years of age: Findings from the CRANE Database for England, Wales and Northern Ireland.** Jibby Medina, Kate J Fitzsimons, Matthew Fell, Sophie Butterworth, Muhammad HA Wahedally, Min Hae Park, Jan H van der Meulen, Craig JH Russell, David Chong

**6) Orofacial cleft laterality and additional congenital anomalies. Results of analysis of linked national databases.** Matthew Fell, Kate J Fitzsimons, Mark J Hamilton, Jibby Medina, Sophie Butterworth, Muhammed HA Wahedally, Min Hae Park, Jan H van der Meulen, David Chong, Craig JH Russell

**7) Range and frequency of additional congenital malformations in children born with cleft lip and/or palate.** Kate J Fitzsimons, Mark J Hamilton, Jan H van der Meulen, Jibby Medina, Muhammad HA Wahedally, Min Hae Park, Craig JH Russell

**8) What is the optimal timing of palate repair for speech outcomes? Analysis of two linked national databases in England.** Craig JH Russell, Kate J Fitzsimons, Lorraine Britton, Simon Van Eeden, Sophie Butterworth, Muhammed HA Wahedally, Min Hae Park, Jibby Medina, Jan H van der Meulen

**9) Identifying the impact of patient factors on dental health outcomes of cleft care at 5 years of age.** Jibby Medina, Jackie Smallridge, Kate J Fitzsimons, Sophie Butterworth, Muhammed HA Wahedally, Scott Deacon, Kate Walker, Min Hae Park, Jan H van der Meulen, Craig JH Russell

[American Cleft Palate and Craniofacial Association \(ACPA\) Conference, Texas, March 2022:](#)

**10) Comparing apples and pears – Identifying and quantifying differences in speech outcome by sex, cleft type and Robin Sequence. The first step towards utilisation of risk adjustment in service-level outcome reports.** Sophie Butterworth, Kate J Fitzsimons, Jibby Medina, Muhammed HA Wahedally, Min Hae Park, Jan H van der Meulen, Craig JH Russell

## 5.2 Improving the accuracy and use of collected data

Data collection can represent a burdensome exercise for patients, clinicians and care providers. CRANE continuously works to ensure that the data being collected are essential, relevant and useful for the various stakeholders interested in cleft care and its related outcomes. Where possible, data linkage exercises are carried out to collect information from other sources to reduce the burden of data collection for cleft service staff. These linkage exercises involve working with partners such as the Cleft Collective, Hospital Episode Statistics (HES) database<sup>99</sup>, National Hearing Screening Program (NHSP) and the National Pupil Database to share data that are mutually beneficial to all stakeholders.

### 5.2.1 Collection of data on syndromes and additional diagnoses

Prior to January 2021, the 'Syndromes' section of the CRANE Database required cleft teams to enter free text to describe named syndromes and descriptions of affected systems. Since January 2021, this information is now

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<sup>99</sup> Hospital Episode Statistics (HES) data have been re-used with the permission of NHS Digital. All rights reserved. Copyright © 2013

captured by clear categories. The Database now offers a drop-down menu that includes the nine most commonly occurring syndromes / additional diagnoses and an 'other' option that allows free text entries for less commonly occurring conditions. This update to the Database provides much more flexibility when performing analyses and will allow CRANE to liaise with each Clinical Excellence Network (CEN) to determine which syndromes/additional diagnoses from the list should potentially be included/excluded when reporting each cleft-related outcome.

### **5.2.2 Cleft phenotypes classification – LAHSAL or LAHSHAL**

At the request of colleagues within the Craniofacial Society of Great Britain and Ireland (CFSGBI), the CRANE Database has been asked to consider modifying the phenotypic description it collects for patients diagnosed with a cleft by moving from the LAHSAL classification to the LAHSHAL classification system for cleft type. LAHSHAL is thought to be a more clinically-friendly and research-sensitive instrument with greater phenotypic information<sup>100</sup>.

Since 2021, CRANE has collaborated with the Cleft Collective group to investigate concordance of cleft classification between the CRANE and Cleft Collective datasets. The level of agreement between the two data sources was compared using inter-rater reliability, calculated using Krippendorff's Alpha. It was found that agreement between the two sources decreased when the amount of detail on cleft type increased. The inter-rater reliability score dropped from 96% for records with the least detailed cleft classification (cleft lip only/cleft palate only/cleft lip and palate) to 74% for records with more detail provided, such as cleft laterality or the completeness of the cleft. Increasing phenotypic description led to deteriorating rates of concordance. For cases that were identified as having discordant cleft type information between the two data sources, cleft teams were asked to verify the LAHSAL code in the CRANE Database and to make relevant amendments where appropriate.

Accurate phenotyping is key to the development of our understanding of the epidemiology and aetiology of clefts, and is essential for studies that involve the analysis of phenotype and genotype information. The CRANE Database and the Cleft Collective are continuing this collaboration to investigate intra- and inter-rater reliability of LAHSAL and LAHSHAL classification systems in different clinical groups. This work will inform future recommendations to the Cleft Development Group as to whether CRANE continues to collect phenotypic information on clefts based on the LAHSAL classification system or migrates to LAHSHAL.

## **5.3 Developmental work to improve our understanding of clefting and determinants of cleft-related outcomes**

### **5.3.1 Influence of smoking on orofacial cleft aetiology**

Data collected as part of the registry function of the CRANE Database can aid in analysing factors that may impact the causality and incidence of clefts. One such example is the use of the CRANE registry data by collaborative partners at the Cleft Collective to understand the role of smoking on causality of clefting. This work did not show any link between active smoking and incidence of clefting. However, the results showed a reduction in incidence of clefting following the introduction of smoke-free legislation in England, Wales and Northern Ireland. This suggests that passive smoking may have a causal role. This research was published in the

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<sup>100</sup> McBride, Andrew & McIntyre, Grant & Carroll, Kris & Mossey, Peter. (2015). Sub-phenotyping and Classification of Orofacial Clefts: Need for Orofacial Cleft Sub-phenotyping Calls for Revised Classification. *The Cleft palate-craniofacial journal: official publication of the American Cleft Palate-Craniofacial Association*. 53. 10.1597/15-029.

PLoS One journal article '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' in November 2021 (see full reference listed above) and was presented at the International Congress of Cleft Lip, Palate and Related Craniofacial Anomalies. This collaboration highlights the importance of being able to share CRANE data for the purpose of facilitating research that informs clinicians, families and, importantly, public health policy makers.

### 5.3.2 Additional congenital malformations

As part of the work undertaken to improve the collection of syndrome and additional diagnoses information on CRANE, an extensive search of a CRANE-HES linked dataset was undertaken to determine which syndromes and additional diagnoses most commonly occur in children born with a cleft. This led to analyses published in the CPCJ article 'Range and Frequency of congenital malformations among children with a cleft lip and/or palate' (see above for full reference) and presented orally at the International Congress of Cleft Lip, Palate and Related Craniofacial Anomalies (see presentations 6 and 7 above). The study is the largest to date, and included 9,403 children born between 2000 and 2012 in England. It found that 38.8% of children had additional congenital malformations documented in their hospital admission records. The prevalence of additional congenital malformations was greatest among children with CP (53.0%), followed by those with BCLP (33.5%), UCLP (26.3%) and then CL±A (22.2%). Among those with UCLP, children with right-sided clefts were more likely to have additional malformations than those with left-sided clefts (31.6% vs 23.0%). Further analyses exploring the relationship between laterality and additional malformations have been performed and a manuscript is currently being prepared for submission. Recommendations from this work include the potential introduction of standardised screening for some cleft subgroups and the establishment of good links between cleft teams and paediatric / genetics services.

### 5.3.3 Determinants of cleft-related outcomes

Quality of healthcare-related outcomes is not simply the result of the quality of medical care. A significant part of the difference in health outcomes can be attributed to patient characteristics and social, economic and environmental factors, known as determinants of health. Identifying such determinants depends on having access to a volume of high-quality data that facilitates appropriately-powered statistical analyses. Analysis of the continuously growing CRANE dataset for the identification of independent determinants of outcomes is the first stage in moving towards the reporting of risk-adjusted outcomes. Risk-adjusted reporting provides the potential to accurately compare regional, national and international results, ensuring that we can more efficiently learn from best practice.

CRANE is actively working to identify determinants of outcomes collected in its expanding database. The volume of data varies between the various outcomes recorded in the database. At present, identifying determinants for speech and dental outcomes is favoured due to the high volume of children with these outcomes recorded. Analyses that significantly impacted on these outcomes are reported in this section.

## Speech

### Patient-related factors

Using CRANE data, the project team has been analysing the relationship between patient-related factors (sex, cleft type, cleft extent, Robin Sequence [RS]) and speech outcomes among 5 year old children with a cleft in England, Wales and Northern Ireland. The findings of this work are published in the CPCJ article 'Investigating the impact of patient-related factors on speech outcomes at 5 years of age in children with a cleft palate'. The

analyses were based on 3,157 children born 2006 to 2014 who had complete Cleft Audit Protocol for Speech – Augmented (CAPS-A) scores at 5 years. These ratings were used to determine whether children met each of the three national cleft speech standards<sup>101</sup> (See Speech section 4.4 for a description of these standards). It was found that sex, cleft type, and extent of hard palate involvement have a significant impact on speech outcome at 5 years of age, particularly in terms of achieving ‘normal’ speech (Standard 1) and speech without significant cleft-related articulation difficulties (Standard 3). In general, boys, those with a more anatomically involved cleft and those with a more extensive cleft palate were less likely to achieve the cleft speech standards. Incorporating these factors into risk-adjustment models for future service-level outcome reporting is recommended. Although children with RS were less likely to meet the speech standards than those with CP without RS, odds ratios were not statistically significant once adjusting for sex and cleft extent. As well as being published in the CPCJ, the findings from this work have also been shared across a number of oral presentations (1, 10, and 11 listed above).

### Cleft laterality

In a separate analysis, CRANE explored whether cleft laterality was associated with speech outcomes using CRANE data for 5-year-old children in England, Wales and Northern Ireland. The findings showed that among children with complete unilateral cleft lip and palate, fewer children with right-sided clefts compared to left-sided clefts had structurally-related speech difficulties or a history of velopharyngeal surgery / fistula repair for speech. This finding indicates that cleft laterality may be a risk factor that should be taken into account when analysing speech outcomes. This work was presented orally at the International Congress of Cleft Lip, Palate and Related Craniofacial Anomalies (See presentation 5 listed above).

### Timing of cleft palate repair

With the onset of COVID, CRANE explored the relationship between timing of palate repair and speech outcomes among children born with non-syndromic palatal clefts in England. This work was undertaken to guide decision making in the UK on relative prioritisation of cleft surgery at a time of limited resource. Analyses were performed using CRANE records linked to NHS England’s HES database, which allowed us to identify the time at which each child had their primary palate repair completed. A total of 1,653 children with a non-submucous cleft affecting the palate, born 2006 to 2012, with no record of additional diagnoses or syndromes (International Classification of Disease - 10 codes Q00-Q99) in their HES history were included in the analyses. Hospital records up to and including 2015 were reviewed and the Classification of Surgical Operations and Procedures 4th Revision (OPCS-4) was used to identify primary palate repairs using the code F291. Complete Cleft Audit Protocol for Speech – Augmented (CAPS-A) scores at 5 years were used to determine whether children met each of the three national cleft speech standards<sup>102</sup> (See Speech section 4.4 for a description of these standards).

The findings showed that there was no difference in speech outcome for all standards among children who had their palate repair completed prior to six months or between six and 12 months from birth, after adjusting for cleft type and extent of hard palate involvement. Children undergoing repair beyond 13 months of age were less likely to achieve normal speech (Standard 1) and speech that was free of cleft-related articulation difficulties (Standard 3) than those undergoing repair before 13 months.

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<sup>101</sup> Britton L, Albery L, Bowden M, Harding-Bell A, Phippen G, and Sell D (2020) National (UK) standards for speech for children born with cleft palate (+/-cleft lip /alveolus).

<sup>102</sup> Britton L, Albery L, Bowden M, Harding-Bell A, Phippen G, and Sell D (2020) National (UK) standards for speech for children born with cleft palate (+/-cleft lip /alveolus).

This project highlights that timing of cleft palate repair may influence speech outcomes among children with a cleft palate diagnosis and shows that early intervention, before 13 months of age, is associated with better articulatory outcomes for children born with clefts affecting the palate. This research was presented orally at the International Congress of Cleft Lip, Palate and Related Craniofacial Anomalies (see presentation 8 listed above) and we plan to prepare a manuscript for journal publication over the next year.

## Dental health

Previous studies have shown that children born with a cleft are at increased risk of poor dental health outcomes compared to the general population<sup>103</sup>. Under the scope of improving and modernising CRANE reporting of dental outcomes, the project team investigated which patient-related and socio-economic factors (sex, cleft type, ethnicity, and index of multiple deprivation) were associated with dental outcomes among children with a cleft. CRANE-HES linked data on 5-year-old children diagnosed with a non-submucous cleft, and born between 2004 and 2012, were analysed. Dental outcomes were defined using decayed, missing and filled teeth scores (dmft), where a dmft score greater than 0 indicates experience of dental decay and a dmft score greater than 5 (dmft>5) indicates experience of extensive dental decay.

Analyses of these linked records showed that risk factors for increased dental disease and reduced access to treatment/care include (1) having a more complex cleft type, such as bilateral cleft lip and palate, (2) being from a non-white ethnic background, and (3) living in the most deprived areas. There was no difference in dental outcomes between boys and girls. These findings were presented at the International Congress of Cleft Lip, Palate and Related Craniofacial Anomalies (see presentation 9 listed above).

Our findings provide useful insights on how dental health outcomes vary by patient and socio-economic factors and should be considered as potential factors to adjust for when reporting dental health outcomes among children with a cleft. Understanding the variations in dental outcomes benefits clinicians and care providers by providing evidence on which groups of patients may benefit most from focused dental surveillance as well as preventative dental treatment and care.

## Educational achievement

Our previous studies<sup>104, 105</sup> using CRANE data linked to the National Pupil Database (NPD) found that children born with non-syndromic cleft lip and/or cleft palate in England had lower academic achievement than the general population at age 5 years, and that, at age 7 years, absence from school had a dose-response relationship with educational attainment. These data suggest that the intensive process of cleft care and the time this requires away from early years education may impact on the educational development of children born with a cleft. To improve our understanding of educational attainment among children with a non-syndromic cleft, CRANE has used the CRANE-NPD linked data to explore and describe:

1. differences in educational attainment between children born with non-syndromic cleft and the general population at ages 5, 7 and 11 years; and

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<sup>103</sup> Dental Caries in Children with Cleft Lip and Palate, Wilawan Weraarchakul and Wiboon Weraarchakul, 2017. J Med Assoc Thai 2017; 100 (Suppl. 6): S131-S135. Full text. e-Journal: <http://www.jmatonline.com>

<sup>104</sup> K J Fitzsimons, L P Copley, Efrosini Setakis, Susan C Charman, S A Deacon, Lorraine Dearden, J H van der Meule/n. Early academic achievement in children with isolated clefts: a population-based study in England. *Archives of Disease in Childhood*. 2018;103:356-362.

<sup>105</sup> Fitzsimons KJ, Deacon SA, Copley LP, et al. School absence and achievement in children with isolated orofacial clefts. *Archives of Disease in Childhood* 2021;106:154-159.



2. longitudinal changes in educational attainment among children with a cleft during primary school.

The findings from this project showed that children with a non-syndromic cleft had lower educational attainment than the general population across all ages (5, 7 and 11 years) and all subjects (English, mathematics and science). Furthermore, across all ages and all subjects, the attainment gap was largest among children with cleft palate only and smallest among children with cleft lip only. The analyses revealed that approximately half of the children with low attainment at age 5 achieved normal attainment in all subject areas at age 11. The results of these analyses were presented orally at the International Congress of Cleft Lip, Palate and Related Craniofacial Anomalies (see presentation 2 listed above).

Low educational attainment can have long-lasting adverse effects on vocational, social and physical health outcomes<sup>106</sup>. The above findings, and previous publications from CRANE, suggest an ongoing stable attainment gap in the population of children born with a non-syndromic cleft. However, parents, carers and cleft clinicians should be reassured that attainment is not fixed and children with low attainment at age 5 can progress to expected levels by age 11. However, for every child that improves their attainment, others experience deterioration, such that the overall attainment gap persists between non-syndromic children with a cleft and age-matched peers in the general population.

Further work needs to be undertaken to understand which groups of children improve their attainment levels and which ones experience deterioration. This will help to better understand determinants of these changes such that recommendations can be proposed on how to target appropriate resources that will help all children with a non-syndromic cleft to bridge the current educational attainment gaps.

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<sup>106</sup> Davies NM, Dickson M, Smith GD, Van Den Berg GJ, Windmeijer F. The causal effects of education on health outcomes in the UK Biobank. *International Journal of Epidemiology*, Volume 49, Issue 4, August 2020, Pages 1282–1293, <https://doi.org/10.1093/ije/dyaa047>



## 6. CRANE Database future directions

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### 6.1. Future development of the CRANE Database and website

CRANE is continuing to work with NHS England, Wales and Northern Ireland to develop our contract to sustainably support the project moving forward. Current contract and funding arrangements running to the end of the financial year 2022-23 cover the registry and audit function of the Database as well as a limited amount of time for Database development. For the first time in 15 years, CRANE received a small inflationary up lift in April 2022. However, with current inflation levels being significantly higher, already stretched resources are being tested to their limits. CRANE is committed to working with stakeholders and partners to develop the full potential of the data held and linkage through collaborative activity.

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

- Dental defects of enamel (DDE) section/items (at 5 and 10 years), as proposed by the Paediatric Dental CEN of CFSGBI – this went live in May 2022.
- Psychology data section as proposed by the Psychology CEN – work is due to commence in 2023.
- 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 LAHSHAL 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/intra-rater reliability of both LAHSAL and LAHSHAL. These data will inform the future direction of phenotypic data collection. 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. Work undertaken by CRANE during COVID demonstrated the utility of linking CRANE data with Hospital Episode Statistics data to investigate the effect of timing of palate repairs on speech outcomes. This experience has demonstrated what can be achieved through linkage and how it can reduce the burden of data collection. Linkages, however, are not without cost. They require both the finances available to 'purchase' the linkage and also adequate within-organisation person resource to apply for, manage and maintain the linkage as well as to investigate linked data, analyse results and agree reporting strategies. CRANE is in discussion with funders to develop linkage potential through adequate funding arrangements.
- Hearing experience - CRANE is working with the ENT/Audiology Clinical Excellence Network to understand what data could be collected to document hearing experience of children with a cleft during early life (0-6 years of age).

CRANE has recently linked to the national Newborn Hearing Screening Programme (NHSP) to see if, through linkage, it is possible to identify rates of congenital deafness in children born with a cleft and the presence (and level) of early hearing dysfunction in the remainder of children with a cleft.

Primary bone grafting outcomes and orthodontic care - CRANE has opened discussions with the Orthodontic CEN exploring the possibility of data collection on both bone graft assessment/outcome and 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. The CRANE Database has been adapted to receive Scottish data. Final contract details are being worked out and we remain positive that Scotland will formally join the CRANE family and begin inputting data early in 2023.

NHS Scotland Management have also indicated a wish to explore the potential for consent from the families and guardians of children attending the 5-year-old audit. This would allow the project to achieve full UK 'audit' coverage in a shorter time frame and would be a significant step forward for the project. Once data sharing agreements are in place for prospective registration of newborns affected by clefting in Scotland, CRANE will work with NHS Scotland to gain consent for data collection of older children and define the detail of retrospective data entry.

## 6.3. Outcome measures

### 6.3.1 Risk adjustment

As outlined in Chapter 5, 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 the 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 determinants of cleft outcomes in the UK.

Starting in 2019, work began to identify patient-related factors that influence 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 cardiovascular malformations) and two independent determinants of dental health (cleft type and socio-economic status). The results of these investigations have and are being prepared for peer-reviewed publication and dissemination.

Moving forward, CRANE intends to develop a limited risk-adjustment model for speech and dental outcomes for inclusion in future reports. In doing so, CRANE has agreed to have an independent academic report on the validity of the proposed risk-adjustment process prepared for the Cleft Development Group to facilitate discussion and agreement prior to any adoption of its use in annual reporting.

### 6.3.2 Young people and adult outcomes

The Clinical Directors group of the CFSGB&I previously asked CRANE to lead on a project to agree an outcome set for young people and adults, via a multiphase Delphi consultation using different methods of engagement to develop consensus. COVID interruptions on work and current resource limitations prevented this work moving beyond the second round of consensus development. However, with the consent of the CDG Chair and CFSGB&I President at time of request, CRANE now has permission to share this information with researchers working in this area at the University of Bristol to ensure the learning achieved to date is not lost, but is utilised to inform the future development of young people and adult outcomes.

## 6.4. Data sources and future analyses

The General Data Protection Regulation (GDPR) coming into force in 2018 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, facilitating both validation of CRANE data and appropriate reporting of cleft-related outcomes.

Data sources to which regular CRANE linkage is 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 ensure both long-term sustainability of the data source and safe holding of the data transferred. The securing of a long-term adequate funding contract for CRANE is vital to this process. Negotiations are ongoing with NHS England/Wales and Northern Ireland to provide a funding arrangement that can allow CRANE to fully deliver in this way. For practical reasons (volume of available data for investigation and analysis), development activity is currently being undertaken with linkages to English data to allow proof of utility. Once demonstrated, it is incumbent on a pan UK national audit to link to the relevant datasets in each of the devolved nations to ensure that standard and risk-adjusted reporting can happen for all.

#### **6.4.1. National Pupil Database (NPD)**

CRANE continues its work with NPD and CRANE-HES-linked data and is seeking to maintain current linkage for the purposes of completing work on longitudinal follow-up of educational attainment in children and young persons affected by clefting. GDPR legislation requires contracts for data sharing that are time-limited, and CRANE is currently working on an extension to our current contract to support publication of this work.

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

#### **6.4.2. Hospital Episode Statistics (HES)**

CRANE, through the Clinical Effectiveness Unit, 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 hospitals in England with similar activity for non-cleft patients. CRANE will use this access and its experience with HES data to investigate the impact of the COVID-19 pandemic on registrations and rates of cleft surgery.

Work needs to be undertaken to better understand the differential effects that severe impacts on health service provision (such as the consequences of the COVID pandemic) have on delivery of cleft care. Such work should include investigation of the differential impact on primary and secondary cleft surgical interventions, as well the effects on outpatient therapeutic cleft services including audiology, speech and language and dentistry. This will help inform patients, clinicians and commissioners as to what care has taken place across NHS England over the recent past. These data have the potential to provide a resource for future planning of services.

Although unlinked HES data is a useful tool for some investigations, it lacks the accuracy and flexibility that linked data would facilitate. Currently, CRANE is only linked to HES for births from 2000 to 2012. New agreements to facilitate ongoing linkage are required to deliver on the aim of producing risk-adjusted speech

and dental outcomes and other similar activity. Having this in place will reduce the already significant data collection burden on clinical teams. For example, data on cleft operative interventions and their timing 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 the accuracy of HES data with prospectively collected operative data will be required.

### 6.4.3. Newborn Hearing Screening Programme (NHSP)

The last month has seen the completion of seven years' work to achieve linkage between the CRANE Database and the Newborn Hearing Screening Programme (NHSP)<sup>107</sup> data, initially with Public Health England (PHE) and more recently NHS Digital. The purpose of linkage is to look at the relationship between clefts and permanent childhood hearing impairment (PCHI).

### 6.4.4. Cleft Collective

2022 has seen ongoing collaboration between CRANE and the Cleft collective. GDPR remains a challenge for all to navigate and this is no different for either CRANE or the Cleft Collective. While CRANE is not able to legally share data for unrestricted onward sharing we are actively discussing how best to make the use of both resources for the purposes of improving our knowledge of cleft and the outcomes of care.

## 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).
- 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 a 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<sup>108</sup>) – 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:

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<sup>107</sup> <http://hearing.screening.nhs.uk/>

<sup>108</sup> 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.

- Measure Number CLP07: The number of 5-year-old children with all 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. CRANE will continue to provide these data for NHS England in 2023.

We have populated a webpage 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](mailto:qcrs.externaldatagroup@england.nhs.uk)

## 6.6. CRANE communications

### 6.6.1 Dissemination of 2022 findings

- This report was published on the CRANE website in December 2022.
- Publication of the Annual Report will be announced via the regular quarterly Newsletter.
- A Summary of Findings for Patients and Parents/Carers from this 2022 Annual Report was produced in collaboration with CLAPA. The summary is available on the CRANE website.
- A one page summary using infographics is also 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.

### 6.6.2 Publications and presentations related to the CRANE Database delivered in 2022

These are described in detail in Chapter 5 – on Database development work.

### 6.6.3 Public interaction

CRANE has decided to continue to actively participate in scientific conferences; this will include 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 (email/phone/Twitter) will further strengthen attempts to widen distribution of the Database’s findings and publications.

Scientific conferences to be attended in 2023:

- Craniofacial Society of Great Britain and Ireland (CFSGBI) Conference, Cardiff, 19-23rd April 2023.

## 7. Conclusion

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Although 2022 was supposed to be the year where we began to move beyond COVID, we unfortunately see evidence daily that this is not the case. COVID and its management has left huge financial pressures on governments, institutions and the public. Management of the pandemic has also had secondary effects in healthcare. The stresses of working in pressured clinical environments has resulted in many choosing to retire or seek alternative employment opportunities. This has resulted in less staff availability to return to pre-pandemic levels of clinical activity. Furthermore, two years of relative isolation and social distancing seem to have led to waning immune education. Children are increasingly experiencing respiratory viruses and other infections, and this is placing significant burdens on healthcare services. It is also worth noting the wider global economic and political context and the associated increase in costs of living, which are likely to add further pressures on secondary healthcare as people try to balance budgets and health.

So as we move towards 2023, one could look at this year's activity report with a glass half empty given ongoing reductions in accrued data available for analysis. We, however, prefer to continue to consider ourselves fortunate to have any data at all from this period. The volume of data received is testament to the dedication of the patients and their families who attended clinics (when they could easily have decided not to), 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), the administrative staff and co-ordination teams who collated and entered the data on to the Database (when hospital administrative services were under pressure from changes in working environments and had increased workloads due to the challenges of ever changing clinic and admission practices), and the clinical leads who supported ongoing audit (when it would have been easier to batten down the hatches and deliver only essential 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, in both clinical and research fields, 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 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 dataset held. We believe that the potential, and the experience of the CRANE team, could be hugely multiplied through collaborative working. We look forward to publishing a Data Access Requests (DAR) policy<sup>109</sup>, taking into consideration the post-GDPR landscape, and a clear DAR governance process in 2023.

If you have a question that you feel could be answered using CRANE data, then please get in contact with the CRANE team once our DAR policy and process is in place. 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.

Finally, we would like to reflect on experiences had at this year's International Cleft Congress in Edinburgh. This congress usually only happens every four years, although COVID necessitated a 5-year hiatus since our last meeting in Orlando, Florida. We were able to share much of the development work that we have undertaken

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<sup>109</sup>This will be signed off by the Specialised Services National Business Intelligence and Information Team, at NHS England and NHS Improvement (NHSE/I), who now directly manage the main CRANE contract on behalf of English and Welsh services. The Cleft Development Group (CDG) will continue to review applications for data release and collaboration, with NHSE approval for data release.

over this time with the wider cleft scientific community. In so doing we were also able to meet old friends and make many new ones. What was clear from all our conversations and discussion is how envious the rest of the world is for what we have. We have structured regional cleft centres (covering the whole of the UK) that all work to agreed similar pathways and with agreed processes and outcome measures, with data being centrally collated, analysed in agreed ways and openly and transparently shared annually with users and providers of services, commissioners and the general public. While the last few years have been challenging we, as the cleft community in the UK, should be very grateful for what we have and not rest on our laurels, but continue to invest in CRANE, work together to fully realise the opportunity we have and share our experience and findings for the betterment of cleft care both here in the UK and with our friends and partners across the world.

With the very best of wishes for the New Year.

Yours sincerely,

The CRANE Project Team

# Glossary and abbreviations

<b>Alveolus / alveolar</b>	The part of the jaw (gum) that supports the teeth and contains the tooth sockets.
<b>BCLP</b>	Bilateral cleft lip and palate
<b>CAPS-A</b>	Cleft Audit Protocol for Speech—Augmented
<b>Caries (dental)</b>	Dental caries are also known as tooth decay / dental decay or a cavity.
<b>CEN</b>	Clinical Excellence Network – previously referred to as Special Interest Group (SIG)
<b>CFSGBI</b>	Craniofacial Society of Great Britain and Ireland
<b>CI</b>	Confidence interval
<b>CL</b>	Cleft lip only
<b>Cleft</b>	A failure of tissues to join during development.
<b>Cleft Development Group (CDG)</b>	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.
<b>Cleft services / regions</b>	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 <a href="#">the supplementary tables</a> for further information on Regional cleft services.
<b>Cleft surgeon</b>	A surgeon undertaking cleft repair surgery in a region / cleft service.
<b>CLEFTSiS</b>	The National Management Clinical Network for Cleft Service in Scotland.
<b>Clinical Standards Advisory Group (CSAG)</b>	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.
<b>Confidentiality Advisory Group (CAG)</b>	An independent statutory body established to promote, improve and monitor information governance in health and adult social care. <a href="http://www.hra.nhs.uk/research-community/applying-for-approvals/confidentiality-advisory-group-cag/">http://www.hra.nhs.uk/research-community/applying-for-approvals/confidentiality-advisory-group-cag/</a>
<b>CP</b>	Cleft palate only
<b>Craniofacial anomalies</b>	A diverse group of deformities in the growth of the head and facial bones.
<b>Craniofacial Society of Great Britain and Ireland (CFSGBI)</b>	An inter-specialty group set up to study cleft lip and palate and other craniofacial anomalies. <a href="http://www.cfsgb.org.uk">www.cfsgb.org.uk</a>
<b>CRG</b>	Clinical Reference Group
<b>CSCs</b>	Cleft Speech Characteristics
<b>Denominator (see also numerator)</b>	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.



<b>dmft</b>	Decayed, missing and filled teeth at 5 years of age
<b>DMFT</b>	Decayed, missing and filled teeth at 10 years of age
<b>DfE</b>	Department for Education
<b>DoH</b>	Department of Health
<b>ENT</b>	Ear, nose and throat
<b>ESQ</b>	Experience of Service Questionnaire
<b>FFT</b>	Friends and Family Test
<b>Funnel plot</b>	<p>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:</p> <ul style="list-style-type: none"> <li>• Each point on the funnel plot represents a cleft service.</li> <li>• Each funnel plot is for one outcome, with its values shown on the vertical/Y axis.</li> <li>• The size of the cleft services’ cohort is shown on the horizontal or X axis.</li> <li>• The benchmark value or overall national percentage is shown as a horizontal line through the centre of the graph.</li> </ul> <p>The graph shows two funnels that lie on either side of the benchmark and are called the control limits – similar to confidence intervals.</p> <ul style="list-style-type: none"> <li>• The inner lines show 2 standard deviations or 95% control limits. The outer lines represent 3 standard deviations or 99.8% control limits.</li> <li>• The funnel shape is formed because the control limits get narrower as the population size increases.</li> </ul> <p>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.</p> <p>Funnel Plot Source: David Spiegelhalter, Medical Research Council Biostatistics Unit - <a href="http://www.erpho.org.uk/Download/Public/6990/1/INPHO%204%20Quantifying%20performance.pdf">http://www.erpho.org.uk/Download/Public/6990/1/INPHO%204%20Quantifying%20performance.pdf</a></p>
<b>General population</b>	<p>In epidemiological terms, all individuals without reference to any specific characteristic.</p> <p>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 &amp; Wales in 2016, according to the Office for National Statistics (ONS) (as in the Registrations section of this report).</p> <p>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.</p>
<b>Hospital Episode Statistics (HES)</b>	A national database containing records on all admissions to NHS hospitals in England.
<b>ICD-10</b>	International Classification of Disease 10th Revision

<b>LAHSAL</b>	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.
<b>Managed Clinical Network (MCN)</b>	A formally organised network of clinicians.
<b>MDT</b>	Multi-disciplinary team.
<b>National Pupil Database (NPD)</b>	A database containing records on all pupils in England as they progress through primary and secondary education.
<b>Numerator (see also denominator)</b>	<p>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.</p> <p>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.</p>
<b>OPCS-4</b>	Classification of Surgical Operations and Procedures 4th Revision
<b>Patient Episode Data Wales (PEDW)</b>	A national database containing records on all admissions to hospitals in Wales.
<b>PRS</b>	Pierre Robin Sequence
<b>RCPCH</b>	Royal College of Paediatrics and Child Health
<b>SCG</b>	Specialised Commissioning Group
<b>SD</b>	Standard deviation
<b>SDQ</b>	Strengths and Difficulties Questionnaire
<b>SLT</b>	Speech and language therapy
<b>Submucous cleft palate</b>	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.
<b>TIM</b>	Tiers of Involvement Measure
<b>UCLP</b>	Unilateral cleft lip and plate
<b>WHO</b>	World Health Organization