Promoting excellence in cleft care

**CLEFT REGISTRY & AUDIT NETWORK**



# Cleft Registry and Audit NEtwork Database

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

# 2024 Annual Report

Results of the registry and audit for children born with a cleft lip and/or palate in England, Wales, Northern Ireland and Scotland

On behalf of the Cleft Development Group

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

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

Kate Fitzsimons, Senior Research Fellow
Jibby Medina, Programme Manager
Sophie Butterworth, Clinical Research Fellow
Abhishek Dixit, CEU Data Manager
Craig Russell, Clinical Project Lead
Jan van der Meulen, Clinical Epidemiologist

Clinical Effectiveness Unit
The Royal College of Surgeons of England
Website: www.CRANE-Database.org.uk
Email: crane@rcseng.ac.uk
Telephone: 020 7869 6610

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

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**CLAPA** Cleft Lip & Palate Association

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

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| Document related to this product, for reference, published separately | Published |
|---|---|
| 2024 Annual Report: The supplementary tables (Appendices) (Excel Workbook) | December 2024 |
| 2024 Annual Report: Responses to outlier process (Appendices) | December 2024 |

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We would like to acknowledge all Cleft Services for supporting the outlier process, which was piloted in 2021 and implemented from 2022. We 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 was adopted by the CDG in 2022 and was introduced in a staged manner in 2023 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 of the COVID-19 pandemic. From 2025, the outlier policy will be implemented in full.

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[1] **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

In preparing to write this foreword for the 2024 CRANE report, I took the opportunity to look back at what others have written in previous annual reports. I was struck by the positive accounts of collaboration, development, and progress towards fuller understanding of best practice in cleft care. However, I was also struck by the ongoing uncertainty about the future of CRANE, our UK-wide registry and audit, established in 2000 and with more than 25,000 children registered to date.

The renowned US mathematician John Allen Paulos wrote that 'Uncertainty is the only certainty there is, and knowing how to live with insecurity is the only security'. This has certainly been the experience of the CRANE team, and as a small, specialist registry the level of funding uncertainty for CRANE has reached a new level. As a specialist clinician who has been part of the cleft clinical community for the past 22+ years, I am conscious of the delicate balance between audit data collection and burden of care for children and families. We have sought to manage this in our multidisciplinary teams in order to better understand what good treatment and good outcomes look like. This endeavour is not without its practical challenges, and this is evidenced in the report of ongoing variation between Cleft Services, in data completeness and specialty outcomes. The 2024 CRANE report calls on Cleft Services, specialty Clinical Excellence Networks (CENs) and CRANE to work together to overcome these barriers as far as possible, to highlight and hopefully to mitigate the inequities facing children born with a cleft. If ever there was a time to recognise the value of CRANE and support its ongoing work, this is it.

It may seem unusual to begin a foreword with such talk of uncertainty and instability. However, this is the current situation, and it is essential that all are aware of the potential implications. The best way to illustrate this is to encourage you to take the time to read the 2024 report and reflect on what it contributes. The report contains details of the registry and audit in England, Wales, Northern Ireland, and Scotland for children born with a cleft between January 2021 and December 2023, and outcomes for 5-year-old children born with a cleft between January 2015 and December 2017. I hope you can find time to read the full report, but for the time poor amongst you, I would highlight the Key findings and recommendations, pages 2-6. These tables, with reference to the detail in relevant report chapters, 'do what they say on the tin' in setting out the main registry and outcome findings and proposing actions for stakeholders. The recommendations relate to improving data completeness and reducing variation in outcomes and are relevant to services across the UK. Central to progress in both areas is CRANE's ongoing, positive engagement with Cleft Services, CENs and Royal Colleges. In 2024 this has included several online engagement events as well as a preliminary report presentation and review.

Examples of variation in processes and outcomes are clear in the 2024 report, one being that despite improvement in the timely detection of cleft palate, more than 1 in 4 children still have a diagnosis beyond 24 hours from birth. Similarly, access to local NHS dental care has still not recovered to pre-pandemic levels, particularly in some regions of the country. However, Cleft Services have positively engaged with the outlier reporting process, piloted in 2020/21 and adopted into reporting practice from 2022-2024, with risk adjustment planned from the 2025 cycle.

Tackling inequalities has become a national priority, across many areas, including healthcare. CRANE's development work in this area includes postcode assigned deprivation scores, recognising that variation in

outcomes may be influenced by patient demographics as well as access to care provided. A further area of development is data linkage with the National Hearing Screening Programme. Behind the scenes therefore, the hard work of CRANE continues, with collaborations, presentations, and peer reviewed publications. This work extends to involvement in the development of an updated National Specification for Cleft Services, including delivery of a nationwide organisational audit.

I would like to thank CRANE for this report, and all the teams and individuals who have contributed to it. Data collection is by no means the sum of cleft care, but robust audit that contributes to continuous quality improvement for children and families, is something we can all get behind.

Wishing you all a happy festive season and a bright 2025!

GmPhippen

Ginette Phippen

Chair of the Cleft Development Group

# Executive summary

## Annual Report 2024

**RCS | CRANE — CLEFT REGISTRY & AUDIT NETWORK**

CRANE is a national registry and clinical audit. It evaluates and reports on the delivery of cleft services to children born with a cleft lip and/or palate in England, Wales, Northern Ireland and Scotland. Registry information is presented for children born in 2021-2023 and audit outcomes at 5 years of age are presented for children born in 2015-2017.

## Registry information

**24,877** children have been registered, over the last 24 years, since 2000.

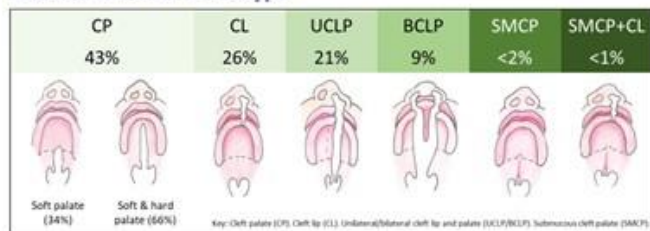**859** children were registered in 2023.

78% of babies with cleft lip were diagnosed antenatally, while 74% of babies with cleft palate alone were diagnosed within 24hrs of birth.

82% were referred to a cleft team within 24hrs of birth and 94% were contacted within 24hrs of referral.

**Distribution of cleft type**

| CP | CL | UCLP | BCLP | SMCP | SMCP+CL |
|----|----|------|------|------|---------|
| 43% | 26% | 21% | 9% | <2% | <1% |

Soft palate (34%)
Soft & hard palate (66%)

Key: Cleft palate (CP), Cleft lip (CL), Unilateral/bilateral cleft lip and palate (UCLP/BCLP), Submucous cleft palate (SMCP)

**88%** of families had a verified consent status, 85% consented and 3% declined consent.

## Audit outcomes at 5 years of age

**Body mass index**

**83%** of children with a cleft had a healthy BMI. 3% were underweight, 9% were overweight and 5% were obese.

**Dental health**

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

**Facial growth**

**52%** of children with a complete unilateral cleft lip and palate (UCLP) had scores reflecting good dental arch relationships.

**Speech**

**57%** of children with a cleft affecting the palate had speech comparable to their peers. Children with a cleft palate (CP) alone had better outcomes than those with a cleft affecting the lip and palate.

**Psychology**

**82%** of children with a cleft had 'Strengths and Difficulties' scores in the low/normal range, compared to 90% of their non-cleft peers. Furthermore, 92% of families were screened at least once before the target age of 6 years.

## Development work

**Deprivation according to postcode records**

For the first time, postcodes were used to assign deprivation scores to 88% of CRANE-consented children born 2014-2023. We found that babies with a cleft were no more likely to live in the most deprived areas than babies born in the general population. However, the percentage of children with a cleft living in the most deprived quintile areas varied between Cleft Services from **11% to 47%**.

**Newborn Hearing Screening Programme (NHSP) data linkage**

CRANE-consented children were linked to the NHSP at the individual-level to report on their diagnostic audiology assessments.

**61%** of children with a cleft had a diagnostic test:

**1 in 4** had conductive hearing loss, and

**1 in 25** had permanent hearing loss.

For further information visit www.CRANE-Database.org.uk      𝕏 @CRANE_News

# Key findings and recommendations 2024

| Source | Findings | Recommendations |
|---|---|---|
| **Registry** | | |
| **Patient characteristics: Cleft type, Robin Sequence and sex** | | |
| Chapter 3, Sections 3.1.1-3.1.3 | • There were 2,609 CRANE-registered children born 2021-2023.<br>• 43% had cleft palate (CP), 26% had cleft lip (CL), 21% had unilateral cleft lip and palate (UCLP), 9% had bilateral cleft lip and palate (BCLP), <2% had submucous cleft palate alone (SMCP) and <1% had SMCP with CL.<br>• 23% of all children with CP were reported to have Robin Sequence. This rate increased to 40% for those with a complete cleft of both the soft and hard palate.<br>• 56% of registrations were boys and 44% were girls. | 1. It is recommended that services ensure RS status is accurately reported for all children with a CP. |
| **Gestational age and birthweight** | | |
| Chapter 3, Sections 3.1.4-3.1.5<br><br>**Indicators[2]: #1 & #2** | • 74% of CRANE-consented children had a recorded gestational age and birthweight.<br>• The average gestational age of babies born with a cleft in 2021-23 was 38.5 weeks.<br>• 12% of babies with a known gestational age were born prematurely (<37 weeks' gestation). This compares to 8% in the general population[3].<br>• The average birthweight was 3,184g.<br>• Children with a BCLP and CP had significantly lower birthweights than those with CL.<br>• 12% of babies with birthweight reported had a low birthweight (<2,500g). This compares to 7% in the general population[2]. The rate among babies born at term was 4%, while the corresponding rate in the general population is 3%. | 2. Cleft Services should ensure that gestational age and birthweight are reported for all CRANE-consented cases, so that the data reported becomes more representative of children born with a cleft.<br>3. The research community should validate and further investigate the higher percentage of premature births and babies with low birthweight among children diagnosed with a cleft compared to rates in the general population.<br>4. CRANE will engage with the Royal College of Midwives (RCM) and Royal College of Obstetricians and Gynaecologists (RCOG) to communicate our findings. |
| **Timing of diagnosis** | | |
| Chapter 3, Section 3.2<br><br>**Indicator: #3 & #4** | • 97% of CRANE-registered children had diagnosis time reported.<br>• 78% of children with a cleft involving the lip were diagnosed antenatally, while 74% of children with CP were diagnosed before or within 24hrs of birth. Timely detection of CP increased to 84% when including diagnoses within 72 hours of birth. Only 21% of children with SMCP were diagnosed before or within 24hrs of birth.<br>• Despite improvement in the timely detection of CP, more than 1 in 4 children still have a diagnosis beyond 24 hours from birth. | 5. CRANE will seek to collaborate further with the clinical workforce to identify barriers to recording timing of diagnosis for all registrations in CRANE.<br>6. 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. |

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[2] CRANE core indicators are detailed in **the supplementary tables**.

[3] Birth characteristics in England and Wales: 2022. Available at: [Birth characteristics in England and Wales - Office for National Statistics (ons.gov.uk)](#) [Last accessed: 08/07/2024]

| | | 7. Cleft Services with lower levels of timely diagnoses should liaise with referring maternity services to notify them that they may be missing opportunities to detect clefts of the lip and palate in a timely manner.<br>8. Cleft Services should extract data from CRANE to identify late diagnoses and the Clinical Nurse Specialist or Clinical Lead should feed this information back to the relevant maternity services, ensuring open communication with Maternity Leads, NIPE leads and Neonatal Clinical Leads. |
|---|---|---|
| **Referral to and contact with Cleft Services** | | |
| Chapter 3, Section 3.3<br><br>**Indicators: #5 to #8** | • 82% of CRANE-registered children had a recorded postnatal referral time. Of these, 82% were referred to a Cleft Service within 24 hours of birth. This rate varied significantly according to Cleft Service and cleft type.<br>• 95% of registered children had a recorded contact time. 94% of families were contacted by a Cleft Service within 24 hours of receiving a referral. This rate varied significantly according to Cleft Service and cleft type.<br>• 84% of registered children were visited by a clinical nurse specialist within 24 hours of postnatal referral. This rate varied significantly according to Cleft Service and cleft type. | 9. Cleft Services should record the contact and referral time of all registrations by working with referring obstetric, midwifery and neonatal units to improve the capture of this information.<br>10. Regional variation in the percentage of children referred, contacted and visited 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. |
| **CRANE consent** | | |
| Chapter 4<br><br>**Indicator: #9** | • Consent status was verified for 88% of children born 2021-23 and 91% of those born 2015-17. This meant they had given informed consent or declined consent for CRANE to collect outcome data.<br>• Consent verification rates varied significantly according to Cleft Service but were consistent across birth years. | 11. Cleft Services with high consent rates should share their best practice recommendations.<br>12. Cleft Services with below average consent rates should review their procedures to identify opportunities to make improvements. |
| **Outcomes at 5 years** | | |
| **Child growth** | | |
| Chapter 5, Section 5.1<br><br>**Indicator: #10 & #11** | • 38% of CRANE-consented children born 2015-2017 had a recorded height and weight.<br>• 83% of children with growth data had a healthy body mass index (BMI). This compares to 77% in the general population of 5-year-olds[4].<br>• According to BMI categorisation, 3% were underweight, 9% overweight and 5% obese. Corresponding rates in the general population are 1%, 12% and 10%, respectively. | 13. 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.<br>14. CRANE will continue to liaise with CDG members and the nursing CEN to encourage all services to collect this data.<br>15. Research should explore reasons why the BMI distribution differs between the cleft and general population of 5-year-olds. |

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[4] National Child Measurement Programme Tables, England 2021/22 and 2022/23 School Years –Available at National Child Measurement Programme - NHS England Digital [Last accessed: 12/07/2024]

**Dental health**

| Chapter 5, Section 5.2 **Indicators: #12 to #16** | • 45% of CRANE-consented children had recorded dmft scores. <br>• Dental decay was experienced by 38% of children (having at least one decayed, missing or filled tooth (dmft >0)) and 15% of children were classified as having extensive caries (dmft >5). <br>• The average Treatment Index (rate of treated disease) was 75%, and the average Care Index (having received the appropriate care at the earliest possible stage) was 68%. | 16. Cleft Services should have at least 80% of all children with a cleft assessed at the age of 5 years by a calibrated specialist in paediatric dentistry, in line with paediatric dentistry CEN standards, and the dmft information should be recorded in the CRANE Database. <br>17. All children with a cleft should have a recommended care plan established by collaborative work between the family's local dental care provider and the specialist paediatric dentist in the Cleft Service. This should (a) treat the child as per the high-risk category of the dental health toolkit (Delivering Better Oral Health), (b) provide routine dental care within the general dental service, and (c) provide specialist level care including age-specific dental development assessment and treatment under inhalation sedation and general anaesthesia within the Cleft Service. <br>18. The COVID-19 pandemic had a significant impact on access to dental care in the early years for this population of patients, particularly those from more deprived areas. Anecdotal and local evidence suggests that access to NHS dental care has still not recovered to pre-pandemic levels particularly in some regions of the country.  All children with a cleft should have access to a local NHS dental provider by their 1st birthday to instigate early preventive advice and build a positive dental relationship. |
|---|---|---|

**Facial growth (for children with complete UCLP)**

| Chapter 5, Section 5.3 **Indicator: #17 & #18** | • 44% of CRANE-children with a complete UCLP had 5-year-old Index scores reported. <br>• 52% had scores reflecting 'good' dental arch relationships, 30% 'fair' and 18% 'poor'. | 19. Cleft Services should aim to take records of all children born with a complete UCLP before they turn 6 years of age to support an external facial growth assessment using the 5-year-old index. These records may take the form of study models or clinical photographs with a recording of the overjet *(the horizontal gap between the front teeth)*. Study models can be made from dental impressions with a bite record or digital scans of the teeth and bite. Photography guidance should be sought from the IMI Guide to Good Practice for Cleft Lip and Palate (template 2a) <br>20. 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. |
|---|---|---|

**Speech (for children with CP, UCLP and BCLP)**

| Chapter 5, Section 5.4 | • 54% of CRANE-consented children with a cleft affecting the palate had all 16 CAPS-A speech parameters reported. These are used to report on the national speech outcome standards. | 21. Children with a cleft affecting the palate should have speech assessed and reported to CRANE. <br>22. Cleft Services should work together to explore reasons for variation in speech outcomes. |
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| **Indicators: #19 to #22** | • 57% met speech outcome standard 1: The achievement of speech with no evidence of a structurally related problem and no cleft speech characteristics requiring intervention.<br>• 73% met speech outcome standard 2a: The achievement of speech without evidence of a structurally related speech difficulty.<br>• 65% met speech outcome standard 3: The achievement of speech without evidence of significant cleft-related speech characteristics, which may require therapy or surgery.<br>• 15% of children had secondary surgery for speech purposes before the age of 5 years. | |

**Psychology screening**

| | | |
|---|---|---|
| Chapter 5, Section 5.5<br><br>**Indicators: #23 to #26** | • 57% of CRANE-consented children had recorded TIM scores<br>• 92% were seen by a psychologist before the age of 6 years and a psychosocial screen was completed or psychological input arranged (Tiers of Involvement Measure 1 to 6 referred to as 1a+).<br>• 48% of CRANE-consented children had a Strengths and Difficulties Questionnaire (SDQ) score.<br>• Of these, 18% had 'high' or 'very high' SDQ scores, indicating a greater level of difficulties, which may require psychological input or intervention. | 23. Cleft Services should aim to see all children and families before their 6th birthday, undertake a psychological screen and ensure psychological support is provided if appropriate (to be recorded as a TIM score).<br>24. Cleft Services should begin to collect Cleft Q data for children born in 2015 who are 10 years of age. |

**All outcomes at 5 years**

| | | |
|---|---|---|
| Chapter 5 | • 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. | 25. All Cleft Services should work together to explore reasons for variations in data completeness and outcomes at 5 years of age. |

**Data quality – throughout report**

| | | |
|---|---|---|
| Chapters 3 to 5 | • There was significant variation in data completeness for registrations and for specialty-specific outcomes across the periods analysed for this report.<br>• Recovery from the COVID-19 pandemic has significantly affected some specialties' ability to submit 5-year-old outcome data. | 26. Cleft Services identified as negative outliers for data completeness are encouraged to consider methods for improving the capture and reporting of these data.<br>27. Cleft Services, Clinical Excellence Networks (CENs) and CRANE should work together to identify and overcome barriers to collecting and submitting data. |

**Database development work**

**Deprivation**

| | | |
|---|---|---|
| Chapter 6, Section 6.1 | • 88% of CRANE-consented children born 2014-2023 had multiple deprivation (MD) scores.<br>• The most deprived quintile was over-represented (27%) by cleft cases and the least deprived quintile was underrepresented (16%).<br>• The distribution of cleft cases across country-specific deprivation quintiles varied between UK nations, but there were no significant differences compared to the distribution of live births in the general population within each nation[5]. | 28. CRANE should assign an MD score to each registered patient with postcode available.<br>29. The relationship between IMD and cleft-related outcomes must be examined to determine whether MD should be incorporated into risk-adjusted models when comparing outcomes between Cleft Services. |

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[5] The distribution of live births in the general population across MD quintiles is not known for Northern Ireland.

| | | |
|---|---|---|
| | • The percentage of cleft patients living in the most deprived quintile areas varied between Cleft Services (11% to 47%). A clear North-South divide was evident, with Cleft Services in the North of England having higher rates of patients from the most deprived quintile compared to the South.<br>• The distribution of cleft cases across deprivation quintiles did not vary significantly between cleft types. | |

**Newborn Hearing Screening Programme (NHSP)**

| | | |
|---|---|---|
| Chapter 6, Section 6.2 | • 11,574 CRANE-consented children had linked NHSP records.<br>• 93% of children with cleft lip alone, who did not have a clear response on their newborn hearing screen had an audiology assessment.<br>• 72% of children with cleft palate +/- lip, regardless of the result of the newborn hearing screen had an audiology assessment. This is below the suggested benchmark of 100%.<br>Of the 7,116 children with an audiology assessment:<br>• 47% had satisfactory hearing in both ears.<br>• 38% had a hearing loss (HL) identified.<br>• 4% had a permanent HL identified. Of these 15% were conductive permanent, 25% were sensorineural, 27% were mixed, 19% were different HL in each ear, and 13% were unilateral loss only.<br>• 3% had a hearing aid offered or issued.<br>• 12% of children with a clear response in both ears on their newborn hearing screen were diagnosed with conductive temporary HL at audiological assessment. This was 2% in those with CL and 17% in those with CP+/-L.<br>• 15% had hearing status that 'had not yet been determined'. | 30. Further investigations are needed to understand why some children who should receive a diagnostic audiology assessment, according to NHSP protocol, do not have evidence of these taking place.<br>31. Referral of high-risk children for audiological assessment is recommended, particularly for those children with additional syndromes. |

**Peer-reviewed publications and presentations**

| | | |
|---|---|---|
| Chapter 6 | • Three scientific articles were published over the last year.<br>1. Butterworth S, Fitzsimons KJ, Britton L, et al. Investigating the Impact of Additional Congenital Malformations on Speech Outcomes at age Five in Children with a Cleft Palate. *The Cleft Palate Craniofacial Journal*. 2024;0(0). doi:10.1177/10556656241287759<br>2. Fell M, Fitzsimons KJ, Hamilton MJ, et al. Cleft lip Sidedness and the Association with Additional Congenital Malformations. The Cleft Palate Craniofacial Journal. 2024;0(0). doi:10.1177/10556656241261918<br>3. Fell M, Bradley D, Chadha A, et al. Sidedness in Unilateral Orofacial Clefts: A Systematic Scoping Review. The Cleft Palate Craniofacial Journal. 2023;0(0). doi:10.1177/10556656231221027<br>• CRANE was involved in five oral presentations at the Craniofacial Society of Great Britain and Ireland Annual Conference in April 2024 and gave two oral presentations at the European Cleft Palate Craniofacial Association Congress in June 2024. | 32. Collaboration is key to sharing CRANE data and facilitating research that informs clinicians, families and policy makers. CRANE should be adequately resourced to undertake continual development work and share information with key stakeholders at national and international meetings to ensure work has greatest impact for those born with a cleft in the UK and beyond. |

# 1. Introduction

The Cleft Registry & Audit NEtwork (CRANE) Database is a national register that was established in 2000 to collect information on children born alive with a cleft lip and/or palate in England and Wales[6]. Northern Ireland officially joined in 2015, and in January 2023 we welcomed Scotland. This means that CRANE is now a UK-wide cleft registry and audit. 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 periodically used to further examine treatment and outcomes for cleft lip and/or palate in England and, in 2022, we linked with the Newborn Hearing Screening Programme in England.

This Annual Report presents findings from data submitted to the CRANE Database[7] and has four main sections:

- **Registry Information:** Patient characteristics, diagnosis times, and early contact care information, including timing of referral to and contact by Cleft Services for children born 2021-2023.
- **Consent:** Consent levels for those born 2021-2023, reflecting recent registrations, and those born in 2015-2017, reflecting those eligible for 5-year outcome reporting.
- **Audit Outcomes at 5 years of age:** Cleft-related outcomes for CRANE-consented children at 5 years of age who were born 2015-2017.
- **Database development work:** Development activity undertaken by the CRANE team over the last 12 months. This year we describe the distribution of CRANE-consented children, born 2014-2023, across quintiles of multiple deprivation, and we report on the audiology assessments recorded as part of the Newborn Hearing Screening Programme for CRANE-consented children born 2006-2021.

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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[6] For further information on the background to the CRANE Database please visit https://www.crane-database.org.uk/
[7] Registered in the CRANE Database by 1 July 2024.

# 2. Methods

## 2.1. Datasets

### 2.1.1. CRANE

CRANE is an online custom-built secure database that holds registry and audit information on children born with a cleft lip and/or palate in the UK. 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 for those with consent. Each child born with a cleft in the UK should be referred to one of 14 Cleft Services (as listed in **the supplementary tables**), who are responsible for registering children on the CRANE Database.

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[8] to collect additional information on 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.

### CRANE cohort

The children and timeframes covered in each chapter, and sub-section, are indicated in the cohort summaries at the beginning of each section. Broadly, timeframes are the most recent years of available data: 2021-2023 births for registry information and 2015-2017 births for 5-year outcomes chapter.

Children whose parents have not consented to their data being used by CRANE have been excluded from the sections and tables on: (1) gestation and weight at birth, (2) 5-year outcomes, (3) deprivation, and (4) CRANE-NHSP analyses, as the data presented in these sections and tables are not collected for non-consenting cases.

### Cleft type

Cleft type was defined according to reported LAHSAL codes. The LAHSAL code is used to classify clefts, with each letter relating to one of the six parts of the mouth that can be affected by a cleft:

| **L** | **A** | **H** | **S** | **A** | **L** |
|---|---|---|---|---|---|
| Right Lip | Right Alveolus | Hard palate | Soft palate | Left Alveolus | Left Lip |

---

The code also indicates whether there is a complete cleft (upper case letter, e.g. L, A, H and/or S), an incomplete cleft (lower case letter, e.g. l, a, h and/or s), or no cleft (left blank). Where LAHSAL has not been reported (in 7% of all registered cases[9]), cleft type is either based on the type reported by the region/ unit registering the child or left as 'unspecified'. The four main cleft types include cleft lip (CL), cleft palate (CP), unilateral cleft lip and palate (UCLP) and bilateral cleft lip and palate (BCLP). The submucous tick box on the database is used to further classify clefts as submucous cleft palate (SMCP) alone or SMCP with cleft lip.

## Missing data

Missing data have been excluded from the denominators presented in all tables, figures and supplementary tables of this report, except for tables and figures relating to data completeness (see **the supplementary tables** for a breakdown of those reported for each outcome). When outcomes are not available, Cleft Services are requested to report a reason for this from a drop down: 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. Data completeness is described for each indicator presented throughout this report.

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

The Newborn Hearing Screening Programme (NHSP)[10], commissioned by the National Health Services for England (NHSE), is responsible for hearing loss detection among all English newborns. The NHSP database contains information on the screening assessment, usually performed within the first few weeks after birth, as well as referral status for audiological assessment, outcome of audiological assessment and type of hearing loss detected, if present. The CRANE-NHSP linked dataset contains individual-level data for children born with a cleft in England between 2006 and 2021, whose families consented to CRANE outcome data collection and/or linkage to health records.

## 2.2. Statistical analyses

Statistical analyses were tailored to the different types of outcome data. Proportions describing categorical outcomes (e.g. achieving or not achieving cleft speech standard 1) were compared across different exposure categories, such as year of birth and cleft type, using Chi-Square Tests. For normally distributed continuous outcome data (e.g. birthweight), linear regression was used to compare differences between exposure categories. For non-parametric continuous outcome data (e.g. Treatment Index and Care Index), a Kruskall-Wallis test was used to compare differences between exposure categories.

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[9] LAHSAL has not been reported for 4% of children registered between 01 January 2021 and 31 December 2023.
[10] Overview of Newborn Hearing Screening Programme: https://www.nhs.uk/conditions/baby/newborn-screening/hearing-test/
[Last accessed: 4/11/2024]

# 3. Registry information

Cleft Services should register all children born alive with a cleft in the UK. This chapter details the characteristics of CRANE-registered children born with a cleft lip and/or palate in 2021-2023. The timing of diagnosis, referral to Cleft Services and contact with families is also provided. This is key information for cleft care planning. Figure 3.1 details the children eligible for reporting.

**Figure 3.1.** CRANE cohort eligible for inclusion in this chapter.
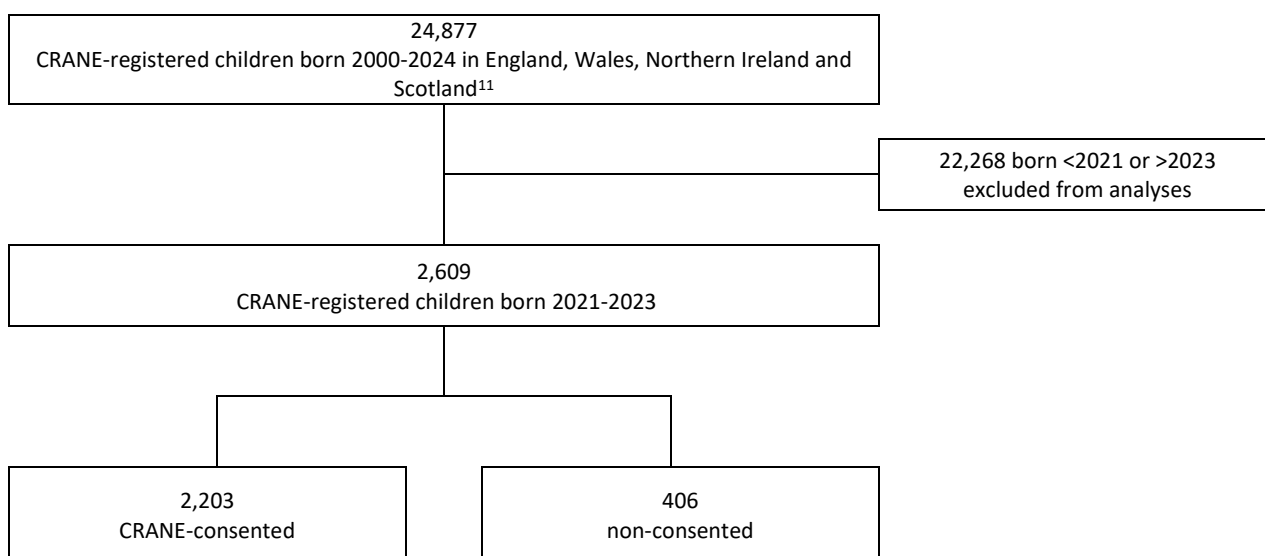


| Characteristic | CRANE-consented N=2,203 (84%) | CRANE non-consented N=406 (16%) |
|---|:---:|:---:|
| Cleft type | ✓ | ✓ |
| Robin Sequence (cleft palate alone) | ✓ | ✓ |
| Sex | ✓ | ✓ |
| Gestation* | ✓ | |
| Birthweight* | ✓ | |
| Diagnosis timing | ✓ | ✓ |
| Referral | ✓ | ✓ |
| Contact | ✓ | ✓ |

**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 birthweight are exceptions to the Section 251 exemption and are collected only for children whose families have given informed consent to outcomes data collection by the CRANE Database.

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[11] Scotland joined in January 2023 and have two years (births in 2022 and 2023) of registrations only.

## 3.1. Patient and birth characteristics

For the most recent three-year reporting period (2021-2023 births), total number of registrations ranged from 94 to 334 between Cleft Services (see 'Registrations 2021-23' in Supplementary tables). This section describes the patient characteristics of these registrations.

| Cohort summary | |
|---|---|
| Data source | The CRANE Database. Extract taken: 1 July 2024 |
| Birth years | Three years: 2021 to 2023 |
| Countries | England, Wales, Northern Ireland and Scotland |
| Inclusions | CRANE-registered children[12], including those without a specified cleft type and those with a submucous cleft palate, unless stated otherwise. |
| Exclusions | None |
| Raw data | 'Patient characteristics 2021-23', 'Gestation 2021-23' & 'Birthweight 2021-23' in **the supplementary tables** |

### 3.1.1 Cleft characteristics

| Data completeness: Cleft type specification | |
|---|---|
| Notes | Cleft type is based on the LAHSAL code provided. LAHSAL code also provides information on the laterality of the cleft lip and the completeness of the cleft. |
| Denominator | 2,609 CRANE-registered children |
| What did we find?  | • 96% of CRANE-registered children had their cleft type specified (Cleft Service range: 76%-100%, p<0.001). This compares to 93% of those born 2020-2022 (CRANE, 2023).<br>• Reporting did not vary significantly according to birth year (2021: 97%, 2022: 96%, 2023: 95%, p=0.071) or sex (female: 96%, male: 96%, p=0.525). |

| Outcome: Cleft type distribution and cleft characteristics | |
|---|---|
| Denominator | 2,505 children with cleft type reported |
| What did we find?  | • 43% of children had a cleft palate alone (CP). Of these, 33% had a cleft affecting only the soft palate and 27% had complete involvement of both the soft and hard palate.<br>• 26% of children had a cleft lip alone (CL). Of these, 55% were left-sided, 35% were right-sided and 10% were bilateral. Among those with a unilateral CL, the left:right ratio was 62%:38%. 12% of CL cases were reported to have a microform cleft lip.<br>• 21% of children had a unilateral cleft lip and palate (UCLP). The left:right ratio was 57%:43%. Overall, 70% had a complete UCLP, defined as LAHS or HSAL LAHSAL codes.<br>• 9% of children had a bilateral cleft lip and palate (BCLP). 58% had a complete BCLP.<br>• <2% of children had submucous cleft palate (SMCP) alone<br>• <1% of children had submucous cleft palate with cleft lip (SMCP+L)<br>• 3% of children were reported to have a syndrome. This rate ranged from 0% among those with SMCP+CL to 5% among those with CP.<br>• There were no significant differences in the distribution of the four main cleft types (CL, CP, UCLP & BCLP) across Cleft Services (p=0.431); however, Northern Ireland registered a much higher proportion of children with SMCP (25% of their registrations) than the overall average (3% of registrations). |

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[12] 11 children who died before reaching two years of age are included in this registration chapter.

### 3.1.2 Robin Sequence

| Data completeness: Robin Sequence status | |
|---|---|
| Notes | • For children with a cleft palate alone, services are required to report Robin Sequence (RS) status (present or absent). <br> • RS is a congenital birth condition characterised by micrognathia, glossoptosis and failure to thrive with or without a cleft affecting the palate. The classification of a child with RS varies between Cleft Services due to ongoing debate around the thresholds for diagnosis. Internationally there is an ongoing series of consensus meetings relating specifically to RS. CRANE contributes to the ongoing debates. |
| Denominator | 1,065 CRANE-registered children with cleft palate alone (CP) (excludes those with SMCP) |
| What did we find? | • 91% of CRANE-registered children with cleft palate alone had their Robin Sequence status (present or absent) specified (Cleft Service range: 30%-100%, p<0.001). This compares to 88% for the previous reporting period (births 2020-2022). <br> • Reporting did not vary significantly by birth year (2021: 90%, 2022: 90%, 2023: 94%, p=0.087) or by sex (female: 92%, male: 91%, p=0.383). |

| Outcome: Prevalence of Robin Sequence among children with a cleft palate alone | |
|---|---|
| Notes | Not subject to outlier policy but funnel plot provided to demonstrate variation in rates across services. |
| Denominator | 1,065 children with cleft palate alone (CP) (excludes those with SMCP) |
| What did we find? | • 23% of children with cleft palate alone were reported to have RS (Cleft Service range: 11%-60%, p<0.001). This rate equates to an annual average of 80 children born with RS between 2021-2023. <br> • 69% of children with CP were confirmed as not having RS (Cleft Service range: 18%-90%). A further 9% did not have their status confirmed and are therefore assumed not to have it. <br> • Prevalence of RS varied significantly according to extent of cleft involvement (LAHSAL code 's': 4%. 'S': 9%, 'Sh': 23%, 'SH': 40%, p<0.001) but not according to birth year (2021: 22%, 2022:21%, 2023: 25%, p=0.433) or sex (female: 24%, male: 20%, p=0.131). |
| Outliers | High: 1. South Wales (60%) <br> Low:  1. Liverpool* (11%), 2. Spires* (11%) <br> * Low alert status for two consecutive reporting periods and therefore classed as outlier. |

**Figure 3.2.** Funnel plot showing the percentage of CRANE-registered children with cleft palate alone, born 2021 to 2023, with Robin Sequence (RS), according to Cleft Service.
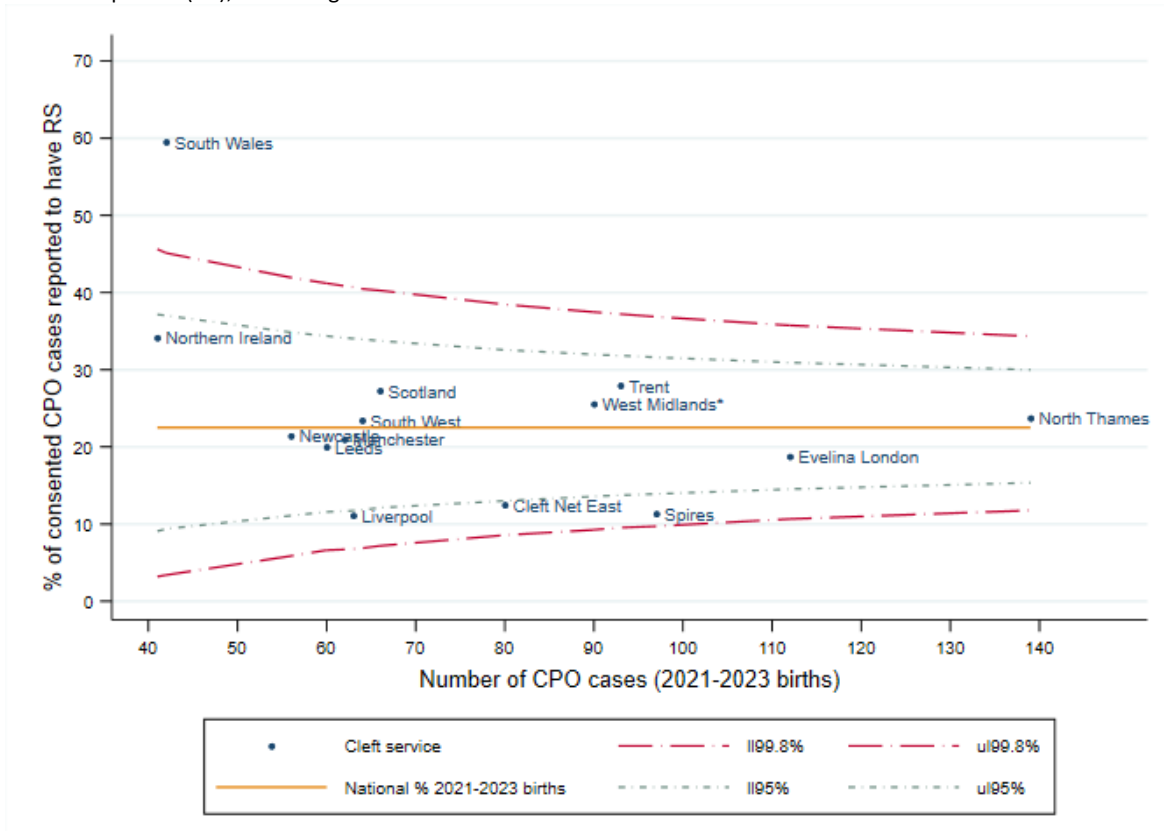


### 3.1.3 Sex

| Data completeness: sex | |
|---|---|
| Denominator | 2,609 CRANE-registered children |
| What did we find? | 99.6% of CRANE-registered children had their sex specified (Cleft Service range: 97%-100%, p<0.001). This has not changed compared to the previous reporting period (births 2020-2022). |

| Outcome: Sex ratio | |
|---|---|
| Denominator | 2,598 children with sex reported |
| What did we find? | • There were more boys (56%) than girls (44%) with a cleft (Cleft Service range for boys: 47%-62%, p=0.203).<br>• The sex ratio varied significantly according to cleft type (p<0.001). There were more boys than girls with clefts involving the lip (range: 60% in CL - 69% in UCLP) and more girls (55%) than boys with clefts affecting only the palate. |

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### 3.1.4 Gestation

| Data completeness: Gestational age | |
|---|---|
| Indicator | #1 - Gestational age recorded for all eligible children |
| Notes | <ul><li>Not subject to outlier policy but funnel plot provided to demonstrate variation in data completeness across services.</li><li>Funnel plot is centred on the revised national rate after excluding data from services identified as negative outliers for consent verification (West Midlands).</li></ul> |
| Denominator | 2,203 CRANE-consented children |
| What did we find? | <ul><li>74% of CRANE-consented children had a recorded gestational age (Cleft Service range: <1%-100%, p<0.001). This compares to 55% for the previous reporting period (births 2020-2022).</li><li>Reporting did not vary significantly according to birth year (2021: 74%, 2022: 72%, 2023: 77%, p=0.069), known cleft type (CL: 76%, CP: 76%, UCLP: 78%, BCLP: 71%, SMCP+CL: 89%, SMCP: 71%, p=0.390) or sex (female: 73%, male: 75%, p=0.196).</li><li><1% had a reason why gestational age data were not collected.</li><li>25% were missing data or a reason for not collecting data.</li></ul> |
| Outliers | Positive: 1. Northern Ireland (100%), 2. Liverpool (99%), 3. Scotland (99%), 4. Trent (99%), 5. Evelina London (97%), 6. South Wales (96%), 7. Leeds (95%), 8. South West (94%)<br><br>Negative: 1. West Midlands (<1%), 2. North Thames (24%), 3. Cleft Net East (46%) |

**Figure 3.3.** Funnel plot showing the percentage of CRANE-consented children, born 2021 to 2023, with gestational age reported, according to Cleft Service.
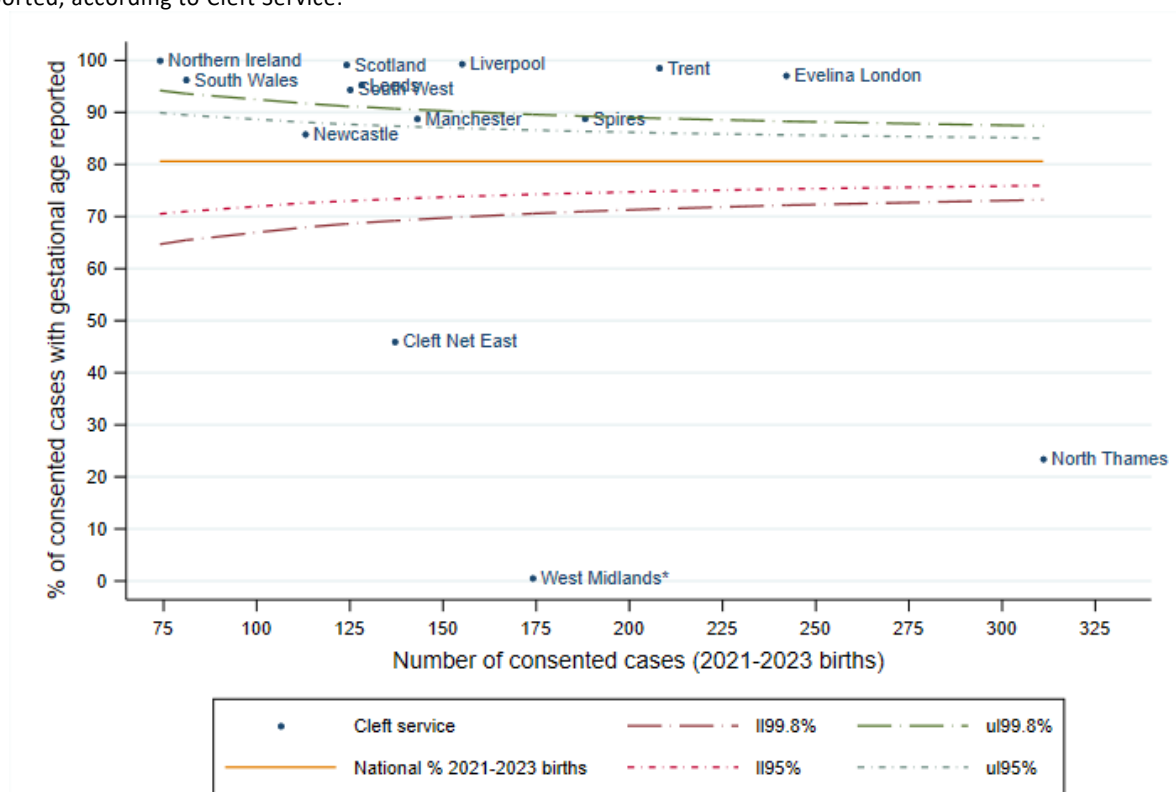


Note: Funnel plot centred on the revised national percentage (80.6%) of children (born 2021-2023) with gestational age data reported.

| Outcome: Gestational age | |
|---|---|
| Benchmarks | Among babies born in 2022 in the general population of England & Wales, 8% had premature births (<37 weeks' gestation)[13]. |
| Denominator | 1,637 CRANE-consented children with gestational age reported |
| What did we find? | • The mean gestational age was 38.5 weeks (95% CI 38.4-38.6 weeks). <br> • The percentage of premature births among children with gestational age reported was 12% (Cleft Service range 9%-18%[14], p=0.606) and is higher than in the general population (8%). <br> • The percentage of premature births did not vary significantly according to known cleft type (CL: 10%, CP: 14%, UCLP: 10%, BCLP: 14%, SMCP+CL: 7%, SMCP: 20%, p=0.248), sex (female: 12%, male: 12%, p=0.977) or birth year (2021: 11%, 2022:13%, 2023: 12%, p=0.684). <br> • There was no difference in the overall proportion of premature births when excluding data from Cleft Services identified as negative outliers for consent verification and/or gestational age data completion. This suggests that the preterm live birth rate of 12% is likely to be representative for children born with a cleft in the UK. |

[13] Birth characteristics in England and Wales: 2022. Available at: Birth characteristics in England and Wales - Office for National Statistics (ons.gov.uk) [Last accessed: 08/07/2024]
[14] among Cleft Services that submitted data for ≥10 cases.

## 3.1.5  Birthweight

| Data completeness: Birthweight | |
|---|---|
| Indicator | #2 - Birthweight recorded for all eligible children |
| Notes | • Not subject to outlier policy but funnel plot provided to demonstrate variation in data completeness across services.<br>• Funnel plot is centred on the revised national rate after excluding data from services identified as negative outliers for consent verification (West Midlands). |
| Denominator | 2,203 CRANE-consented children |
| What did we find? | • 74% of CRANE-consented children had a recorded birthweight (Cleft Service range: <1%-100%, p<0.001). This compares to 55% for the previous reporting period (births 2020-2022).<br>• Reporting did not vary significantly according to birth year (2021: 73%, 2022: 72%, 2023: 76%, p=0.190), known cleft type (CL: 74%, CP: 76%, UCLP: 78%, BCLP: 72%, SMCP+CL: 88%, SMCP: 75%, p=0.434) or sex (female: 73%, male: 75%, p=0.205).<br>• <1% had a reason why birthweight data were not collected.<br>• 26% were missing data or a reason for not collecting data. |
| Outliers | Positive:  1. Liverpool (100%), 2. Northern Ireland (100%), 3. Scotland (98%), 4. Trent (98%),<br>        5. South West (98%), 6. Evelina London (96%), 7. Leeds (95%), 8. South Wales (94%)<br>Negative: 1. West Midlands (<1%), 2. North Thames (24%), 3. Cleft Net East (47%) |

**Figure 3.4.** Funnel plot showing the percentage of CRANE-consented children, born 2021 to 2023, with birthweight reported, according to Cleft Service.
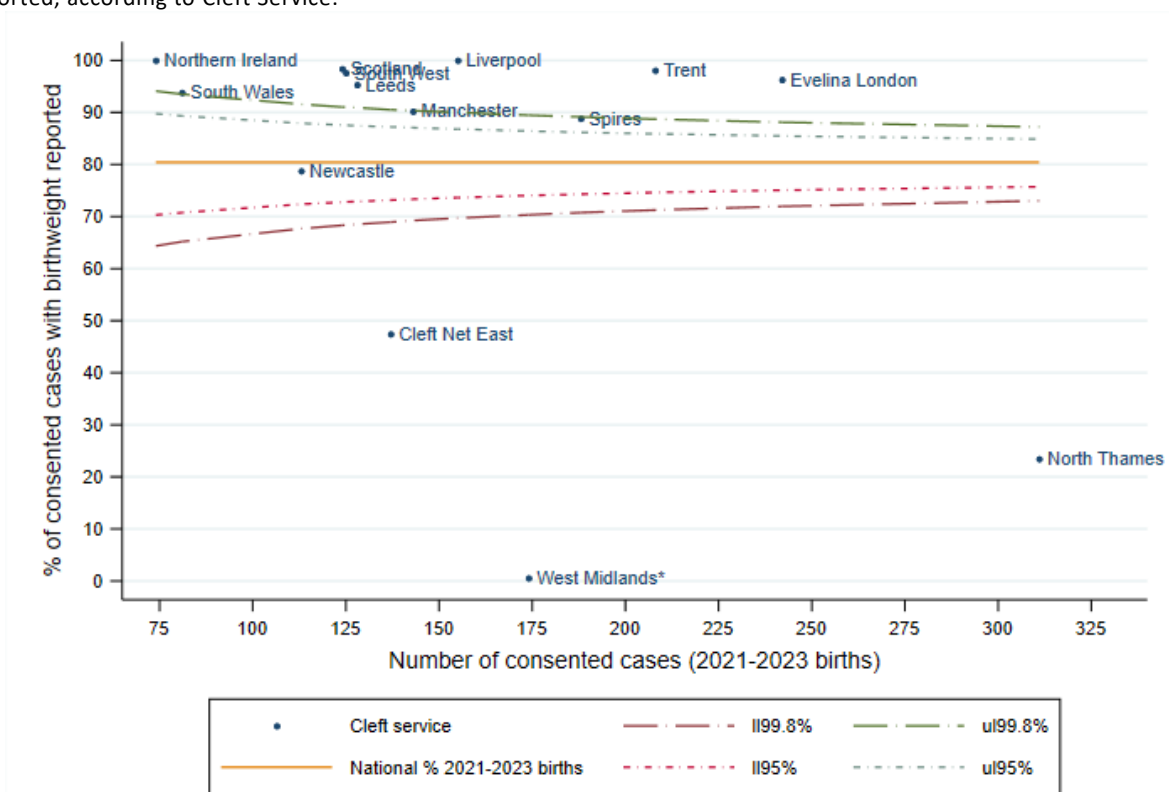


Note: Funnel plot centred on the revised national percentage (80.4%) of children (born 2021-2023) with birthweight data reported.

| Outcome: Birthweight | |
|---|---|
| Definitions | • Low birthweight (LBW) is defined as <2,500g<br>• Healthy birthweight is defined as 2,500g-3,999g<br>• High birthweight (HBW) is defined as ≥4,000g |
| Benchmarks | Among babies born in 2022 in the general population of England & Wales, 7% had a low birthweight (LBW). Among those born at term, the rate was 3%[15]. |
| Denominator | 1,632 CRANE-consented children with birthweight reported. Of these, 1,423 were known to be born at term (≥37 weeks). |
| What did we find? | • The mean birthweight was 3,184g (95% CI 3,152-3,216) among all children with birthweight reported, and was 3,329g (95% CI 3,302-3.355) among those born at term.<br>• Mean birthweight varied according to cleft type. Compared to children with CL (birthweight 3,239g), children with CP (3,148g, p=0.025) and BCLP (3,102g, p=0.030) had significantly lower birthweights, while those with UCLP (3,227g, p=0.793), CL+SMCP (3,255g, p=0.929) and SMCP alone (3,168g, p=0.626) had similar birthweights.<br>• 12% of all children with birthweight reported had LBW (Cleft Service range: 8%-19%[16], p=0.711). The rate was 4% among those born at term (Cleft Service range: 2%-6%[17], p=0.930). These rates are higher than those reported for the general population in 2022 (7% and 2%, respectively).<br>• The percentage of children born at term with LBW did not vary significantly by cleft type (CL: 4%, CP: 5%, UCLP: 4%, BCLP: 5%, SMCP+CL: 0%, SMCP: 6%, p=0.937), sex (female: 5%, male: 3%, p=0.058) or birth year (2021: 3%, 2022: 6%, 2023: 4%, p=0.154).<br>• There was no difference in the overall proportion of babies with low birth weight when excluding data from Cleft Services identified as negative outliers for consent verification and/or birthweight data completion. This suggests that the LBW rate of 4% is likely to be representative for children born with a cleft at term in the UK. |

| Recommendations: Patient and birth characteristics |
|---|
| • The wide range in reported rates of RS between Cleft Services suggests that there is potential under- and over-reporting of this condition. It is recommended that services work to agree consensus on clinical thresholds for classifying RS.<br>• Cleft Services identified as negative outliers for data completeness are encouraged to consider methods for improving the capture and reporting of these data items to CRANE.RS is a known determinant of outcome for speech and it is important that accurate RS status is recorded to allow appropriate risk adjustment of future speech results for each service.<br>• The research community should validate and further investigate the higher percentage of premature births and babies with low birthweight among children diagnosed with a cleft compared to rates in the general population.<br>• CRANE will engage with the Royal College of Midwives (RCM) and Royal College of Obstetricians and Gynaecologists (RCOG) to communicate findings. |

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[15] Birth characteristics in England and Wales: 2022. Available at: Birth characteristics - Office for National Statistics (ons.gov.uk) [Last accessed: 08/07/2024]

[16] Among Cleft Services that submitted data for ≥10 cases.

[17] among Cleft Services that submitted data for ≥10 cases.

## 3.2  Timing of diagnosis

A cleft involving the lip is usually identified during a routine ultrasound scan at around 20 weeks' gestation. Clefts affecting only the palate can be difficult to detect by ultrasound and are usually diagnosed immediately after birth or during the newborn physical examination, performed within 72 hours of birth. Once a diagnosis takes place, the local Cleft Service should be notified.

| Cohort summary | |
|---|---|
| Data source | The CRANE Database. Extract taken: 1 July 2024 |
| Birth years | Three years: 2021 to 2023 |
| Countries | England, Wales, Northern Ireland and Scotland |
| Inclusions | CRANE-registered children |
| Exclusions | None |
| Notes | • Not subject to outlier policy but funnel plots provided to demonstrate variation in rates across services.<br>• Data are not risk adjusted. |
| Raw data | 'Diagnosis times 2021-23' & 'Diagnosis times CPO 2021-23' in **the supplementary tables**. |

| Data completeness: Diagnosis time | |
|---|---|
| Denominator | 2,609 CRANE-registered children |
| What did we find?<br> | • 97% of CRANE-registered children had diagnosis time reported (Cleft Service range: 88%-100%, p<0.001). This compares to 95% for the previous reporting period (births 2020-2022).<br>• Reporting varied significantly according to known cleft type (CL: 99%, CP:98%, UCLP: 99%, BCLP: 99%, SMCP+CL: 100%, SMCP: 93%, p=0.003). Only 63% of children with an 'unspecified' cleft type had diagnosis time reported.<br>• The reporting of diagnosis time did not vary significantly according to birth year (2021: 97%, 2022: 97%, 2023: 97%, p=0.732) or sex (female: 97%, male: 97%, p=0.503). |
| Outliers | Positive:  1. Newcastle* (100%)<br>Negative: 1. West Midlands (88%)<br>* Positive alert status for two consecutive reporting periods and therefore classed as outlier. |

**Figure 3.5.** Funnel plot showing the percentage of all CRANE-registered children, born 2021 to 2023, who had diagnosis time reported, according to Cleft Service.
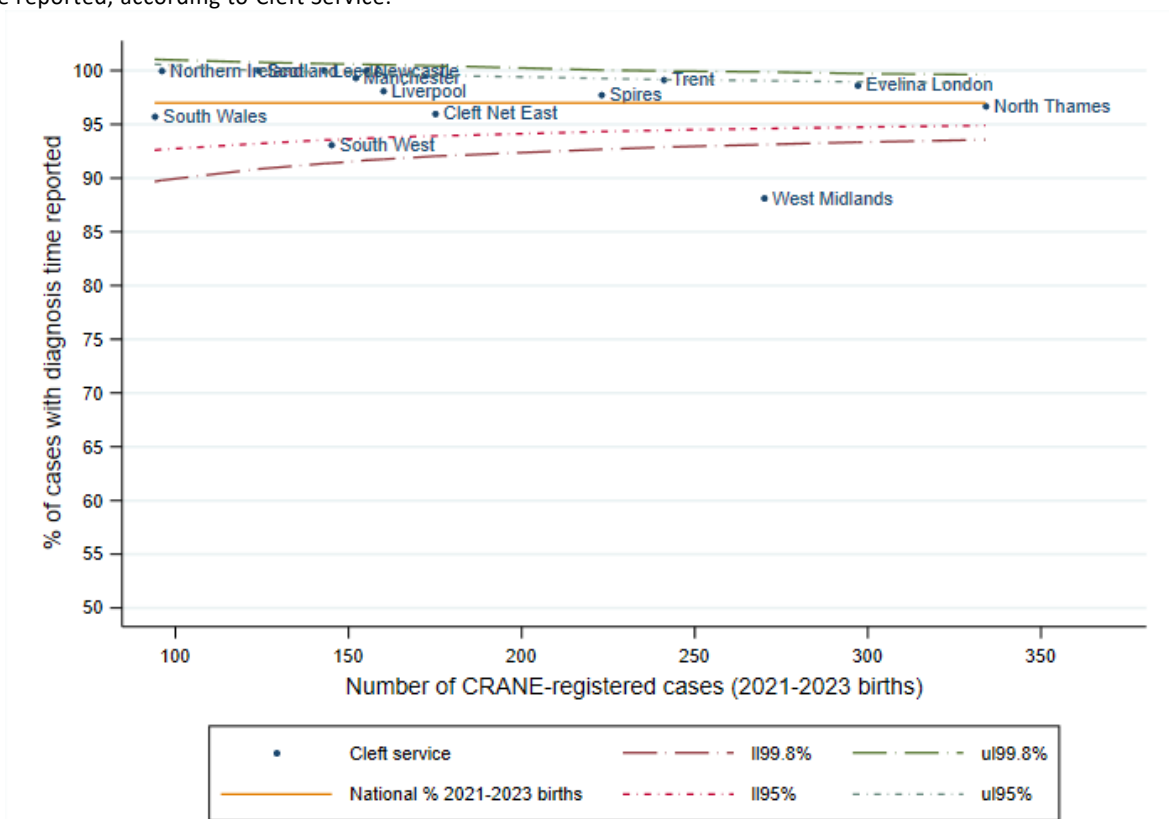


Note: Funnel plot centred on the national percentage (97.0%) of children (born 2021-2023) with diagnosis time reported.

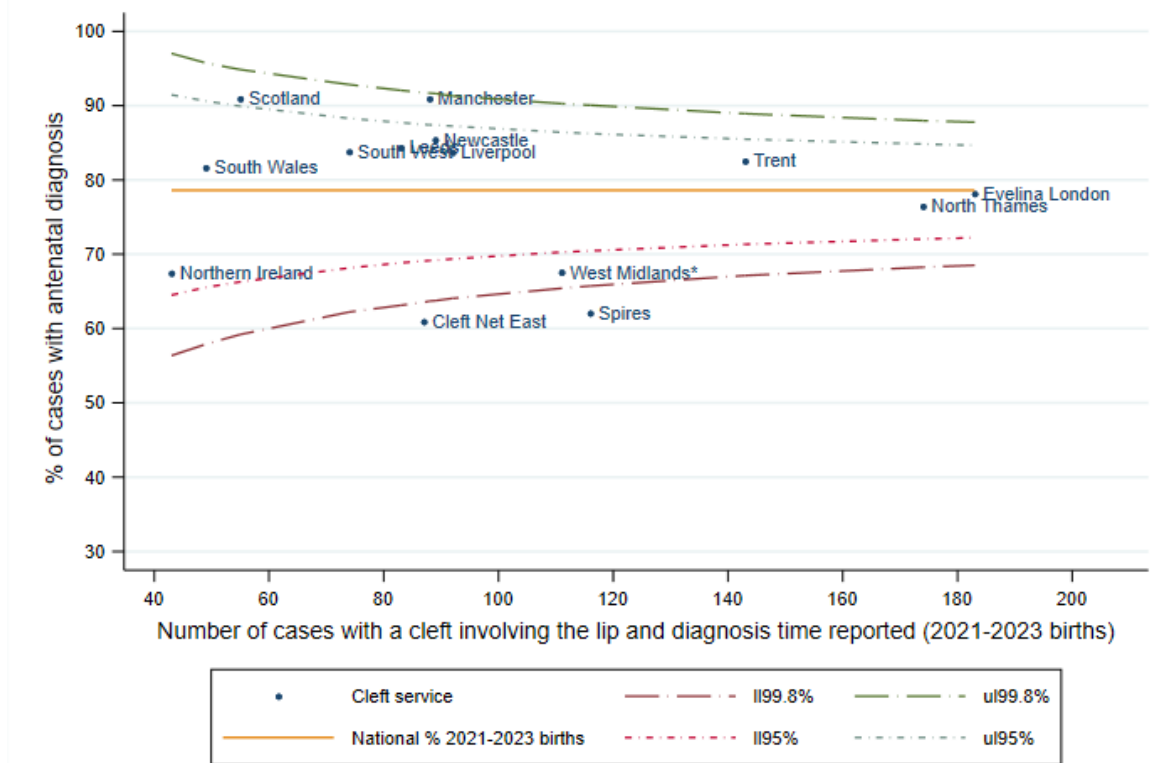| Outcome: Timing of diagnosis among all children with a cleft affecting the lip (CL, UCLP and BCLP) | |
|---|---|
| Indicator | #3 - Antenatal diagnosis for CL, UCLP and BCLP |
| Benchmark | Clefts involving the lip should be diagnosed antenatally. This was the case for 78% of children born 2020-2022. |
| Notes | • Outcome funnel plots are centred on the revised national rates after excluding data from services identified as negative outliers for timing of diagnosis data completeness (West Midlands).<br>• Outcome data reflect care provided by maternity services referring on to Cleft Services. |
| Denominator | 1,387 CRANE-registered children with a cleft affecting the lip and diagnosis time reported. |
| What did we find? | • 78% of children with a cleft affecting the lip were diagnosed antenatally (Cleft Service range: 61%-91%, p<0.001). This rate remains unchanged compared to the previous reporting period (2020-2022 births).<br>• Antenatal diagnosis rates varied significantly according to cleft type (CL: 69%, UCLP: 85%, BCLP: 88%, SMCP+CL: 68%, p<0.001) and sex (female: 75%, male: 80%, p=0.032), but not according to birth year (2021: 76%, 2022: 77%, 2023: 80%, p=0.350). |
| Outliers | Positive:  1. Manchester* (91%)<br>Negative: 1. Cleft Net East (61%), 2. Spires (62%)<br>* Positive alert status for two consecutive reporting periods and therefore classed as outlier. |

**Figure 3.6.** Funnel plot showing the percentage of CRANE-registered children with a cleft affecting the lip, born 2021 to 2023, who had an antenatal diagnosis, according to Cleft Service.


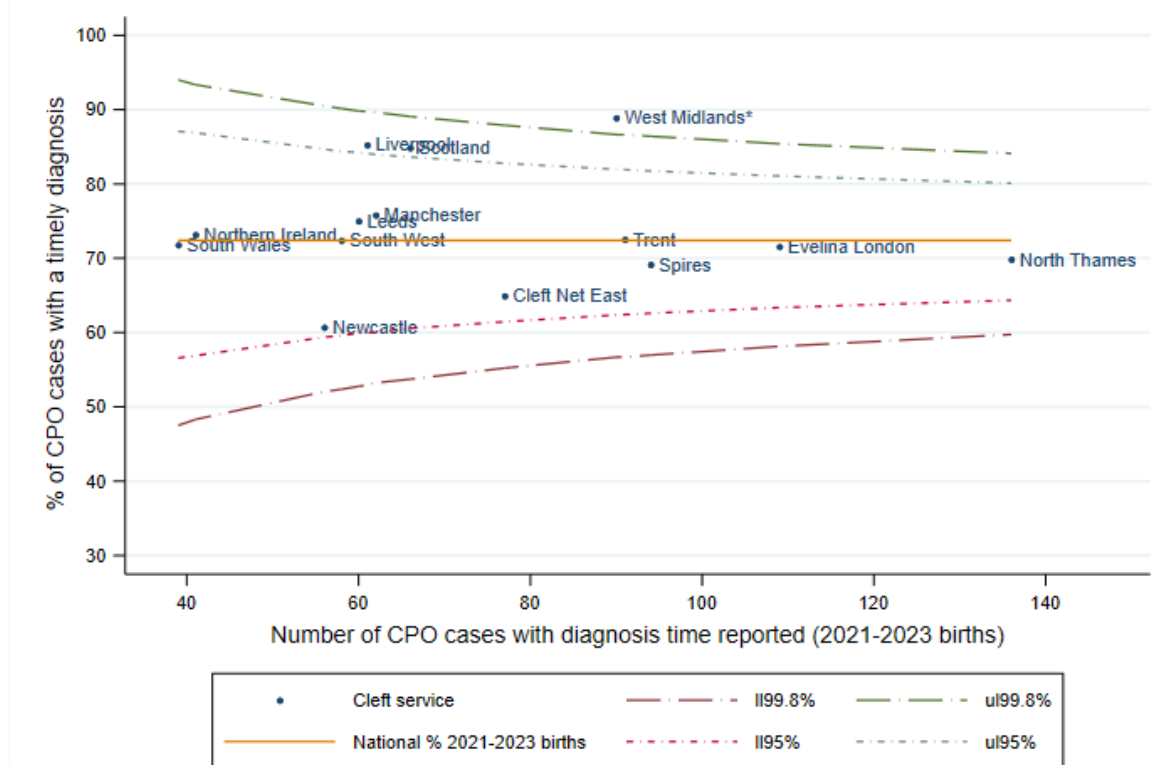
Note: Funnel plot centred on the revised national percentage (78.6%) of children (born 2021-2023) with an antenatal diagnosis.

| | |
|---|---|
| **Outcome: Timing of diagnosis among children with a cleft palate alone** | |
| Indicator | #4 - Timely detection of Cleft Palate (CP), within 24/72 hours from birth |
| Benchmark | Clefts affecting only the palate should be diagnosed before or within 24 hours of birth. This was the case for 72% of children born 2020-2022. This increased to 84% when including diagnoses within 72 hours of birth, which is the timeframe recommended for the NIPE newborn screening examination[18]. |
| Notes | • Outcome funnel plots are centred on the revised national rates after excluding data from services identified as negative outliers for timing of diagnosis data completeness (West Midlands).<br>• Outcome data reflect care provided by maternity and neonatal services referring on to Cleft Services. |
| Denominator | • 1,040 children with a cleft affecting the palate alone (excluding SMCP) and diagnosis time reported.<br>• 38 children with SMCP alone and diagnosis time reported. |
| What did we find?  | • 74% of children with a cleft affecting the palate alone (excluding SMCP alone) were diagnosed before or within 24 hours of birth (Cleft Service range: 61%-89%[19], p=0.005). This is a 2% increase compared to the previous reporting period (births 2020-2022).<br>• 24-hour diagnosis rates varied significantly according to extent of cleft involvement (LAHSAL codes 's': 57%, 'S': 70%, 'Sh': 73%, 'SH': 84%, p<0.001). The rate also varied according to birth year (2021: 74%, 2022: 70%, 2022: 78%, p=0.043) but not sex (female: 76%, male: 71%, p=0.095).<br>• Timely detection increased to 84% when including diagnoses within 72 hours of birth (Cleft Service range: 70%-92%, p=0.011). This remains unchanged from the previous reporting period (2020-2022 births).<br>• 72-hour diagnosis rates varied significantly according to extent of cleft involvement (LAHSAL codes 's':71%, 'S':80%, 'Sh':86%, 'SH':90%, p<0.001) and sex (female: 86%, male:81%, p=0.034), but not according to birth year (2021: 84%, 2022: 82%, 2023: 86%, p=0.478).<br>• 21% of children with a SMCP alone were diagnosed before or within 24 hours of birth. 18% were diagnosed 2-7 days after birth, 24% 7-28 days after birth, 18% 1-6 months after birth, and 18% >6 months after birth. |
| Outliers | Positive outlier: 1. West Midlands (89%)<br>Negative outlier: None |

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[18] Newborn and infant physical examination (NIPE) screening programme handbook - GOV.UK
[19] Note that West Midlands were a negative outlier for data completion so interpret this rate with caution. The highest rate among services not identified as negative outliers for consent or data completion was 85% (Liverpool).

**Figure 3.7.** Funnel plot showing the percentage of CRANE-registered children with a cleft affecting the palate alone (excluding SMCP), born 2021 to 2023, who were diagnosed before or within 24 hours of birth, according to Cleft Service.



Note: Funnel plot centred on the revised national percentage (72.4%) of children (born 2021-23) diagnosed antenatally or <24 hours after birth.

| Recommendations: Timing of diagnosis |
|---|
| • CRANE will seek to collaborate further with the clinical workforce to identify barriers to recording timing of diagnosis for all registrations in CRANE. |
| • CRANE will continue to monitor rates of antenatal and timely diagnoses to ensure issues are highlighted, as well as opportunities for learning and areas for improvement identified. |
| • Cleft Services with lower levels of timely diagnoses should notify referring maternity services that they may be missing opportunities to detect clefts of the lip and palate in a timely manner. |
| • Cleft Services should extract data from CRANE to identify late diagnoses and the Clinical Nurse Specialist or Clinical Lead should feed this information back to the relevant maternity services, ensuring open communication with Maternity Leads, NIPE leads and Neonatal Clinical Leads. |

## 3.3.   Referral to and contact with Cleft Services

Early referral to Cleft Services ensures that children diagnosed with a cleft receive the care and support that they and their families need, in a timely fashion.

| Cohort summary for referral and contact time | |
|---|---|
| Data source | The CRANE Database. Extract taken: 1 July 2024 |
| Birth years | Three years: 2021 to 2023 |
| Countries | England, Wales, Northern Ireland and Scotland |
| Inclusions | CRANE-registered children |

| Exclusions | Children who died before reaching 1 week of age[20] |
|---|---|
| Notes | • Not subject to outlier policy but funnel plot provided to demonstrate variation in rates across services. <br> • Data are not risk-adjusted. |
| Raw data | 'Referral 2021-23' and 'Contact & visit 2021-23' in **the supplementary tables**. |

### 3.3.1 Referral

| Data completeness: Postnatal referral time | |
|---|---|
| Indicator | #5 - Referral recorded for all eligible children |
| Denominator | 2,608 CRANE-registered children |
| What did we find? | • 82% of all CRANE-registered children had a recorded postnatal referral time (Cleft Service range: 56%-99%, p<0.001). This compares to 74% for the previous reporting period (births 2020-2022). <br> • The percentage of children with a recorded referral time varied significantly according to cleft type (CL: 85%, CP:82%, UCLP:88%, BCLP:89%, SMCP+CL: 84%, SMCP: 64%, p<0.001), but not according to birth year (2021: 80%, 2022: 83%, 2023: 82%, p=0.209) or sex (female: 83%, male: 81%, p=0.172). |
| Outliers | Positive:  1. Scotland (99%), 2. Liverpool (97%), 3. Manchester (94%), 4. Leeds* (90%), <br>           5. North Thames (89%) <br> Negative: 1. West Midlands (56%) <br> * Positive alert status for two consecutive reporting periods and therefore classed as outlier. |

**Figure 3.8.** Funnel plot showing the percentage of CRANE-registered children, born 2021 to 2023, who had postnatal referral time reported, according to Cleft Service.
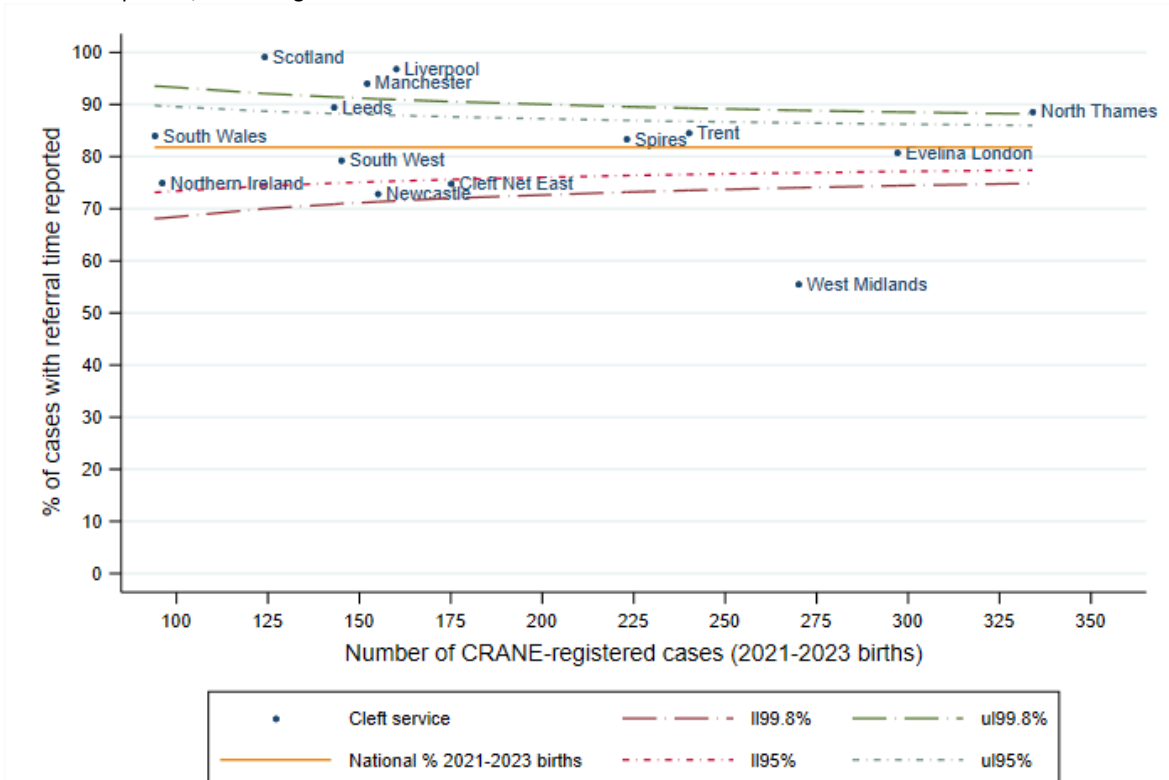


Note: Funnel plot centred on the national percentage (81.8%) of children (born 2021-2023) with referral time reported.

---

[20] Children dying within the first 6 days of life within this cohort (n=1) are excluded as referral may not be appropriate.

| Outcome: Referral to Cleft Service within 24 hours of birth | |
|---|---|
| Indicator | #6 - Referral to a Cleft Service within 24 hours of birth |
| Benchmarks | 82% of children born with a cleft in 2020-2022 were referred to Cleft Services within 24 hours of birth (CRANE, 2023). |
| Notes | As the percentage of CRANE-registered children with SMCP alone and unspecified cleft types vary between Cleft Services and that referral within 24hrs of birth is significantly lower among these children too, these children have been excluded from the rates presented in the funnel plot below. |
| Denominator | 2,134 CRANE-registered cases with referral time recorded |
| What did we find? | • 82% of children were referred to a Cleft Service within 24hrs of birth (Cleft Service range: 57%-90%, p<0.001). This remains unchanged from the previous reporting period (2020-2022 births).<br>• Referrals within 24hrs of birth varied according to cleft type (CL: 90%, CP: 68%, UCLP: 97%, BCLP: 96%, SMCP+CL: 81%, SMCP:25%, p<0.001) and sex (female: 78%, male: 84%, p=0.017). These findings are consistent with later diagnosis times for children with SMCP and CP, with the latter more prevalent among females.<br>• Referrals within 24hrs of birth did not vary significantly according to birth year (2021: 81%, 2022: 81%, 2023: 83%, p=0.512).<br>• When excluding from analyses children with SMCP alone or an unspecified cleft type, the overall referral rate within 24hrs of birth increased to 83% (Cleft Service range: 63%-91%, p<0.001). |
| Outliers | Positive: None<br>Negative: 1. Northern Ireland (63%) |

**Figure 3.9.** Funnel plot showing the percentage of CRANE-registered children, born 2021 to 2023, who had been referred to a Cleft Service within 24hrs of birth, according to Cleft Service. Excludes children with an SMCP alone and those with an unspecified cleft type.



Note: Funnel plot centred on the revised national percentage (82.4%) of children (born 2021-2023) referred within 24hrs of birth.

## 3.3.2 Contact

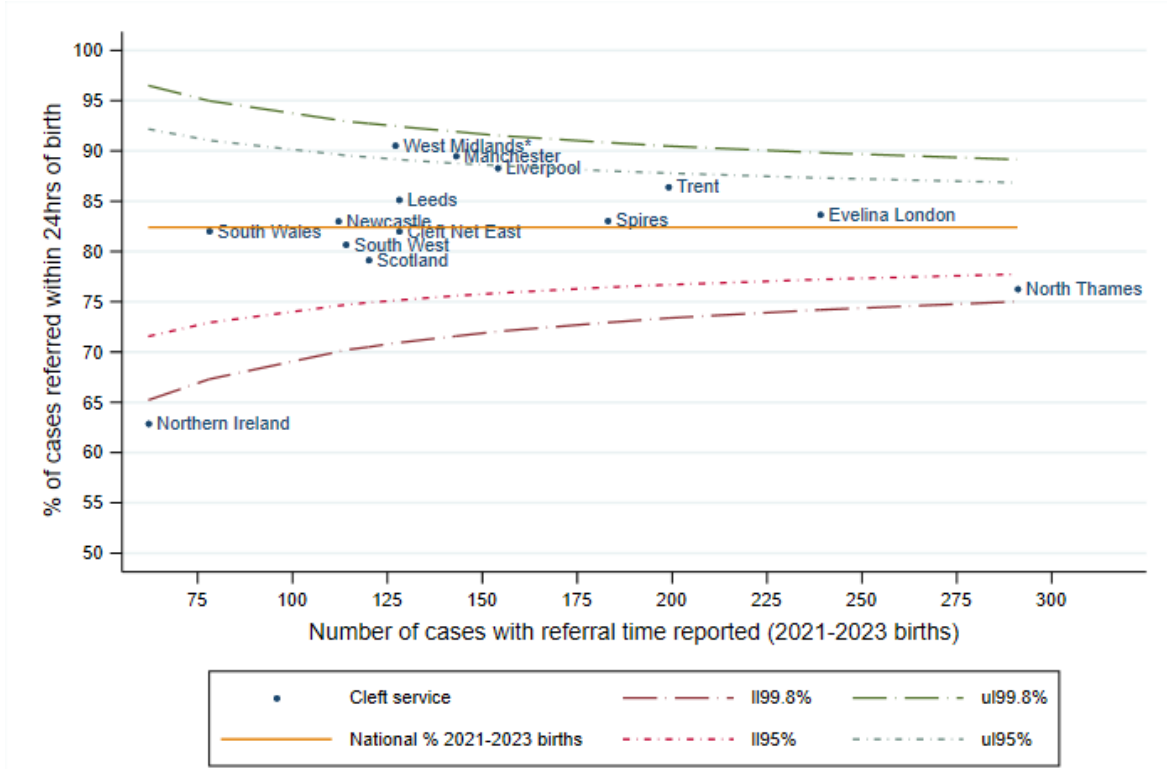| Data completeness: Contact time between Cleft Service and family after postnatal referral | |
|---|---|
| Indicator | #7 – Contact time between Cleft Service and family recorded for all eligible children |
| Denominator | 2,608 CRANE-registered children |
| What did we find? | • 95% of all CRANE-registered children had a recorded contact time (Cleft Service range: 84%-100%, p<0.001). This compares to 91% for the previous reporting period (births 2020-2022). |
| | • The reporting of contact time varied significantly according to birth year (2021: 92%, 2022: 96%, 2023: 95%, p=0.004) and cleft type (CL:97%, CP: 97%, UCLP: 97%, BCLP: 96%, SMCP+CL:100%, SMCP: 85%, p=0.004), but not by sex (p=0.947). |
| Outliers | Positive:  1. Leeds (100%), 2. Evelina London (99%), 3. Newcastle* (99%), 4. Manchester* (99%), 5. Liverpool* (98%)<br><br>Negative: 1. West Midlands (84%), 2. North Thames (89%)<br><br>* Positive alert status for two consecutive reporting periods and therefore classed as outlier. |

**Figure 3.10.** Funnel plot showing the percentage of CRANE-registered children, born 2021 to 2023, who had contact time between Cleft Service and family reported, according to Cleft Service.
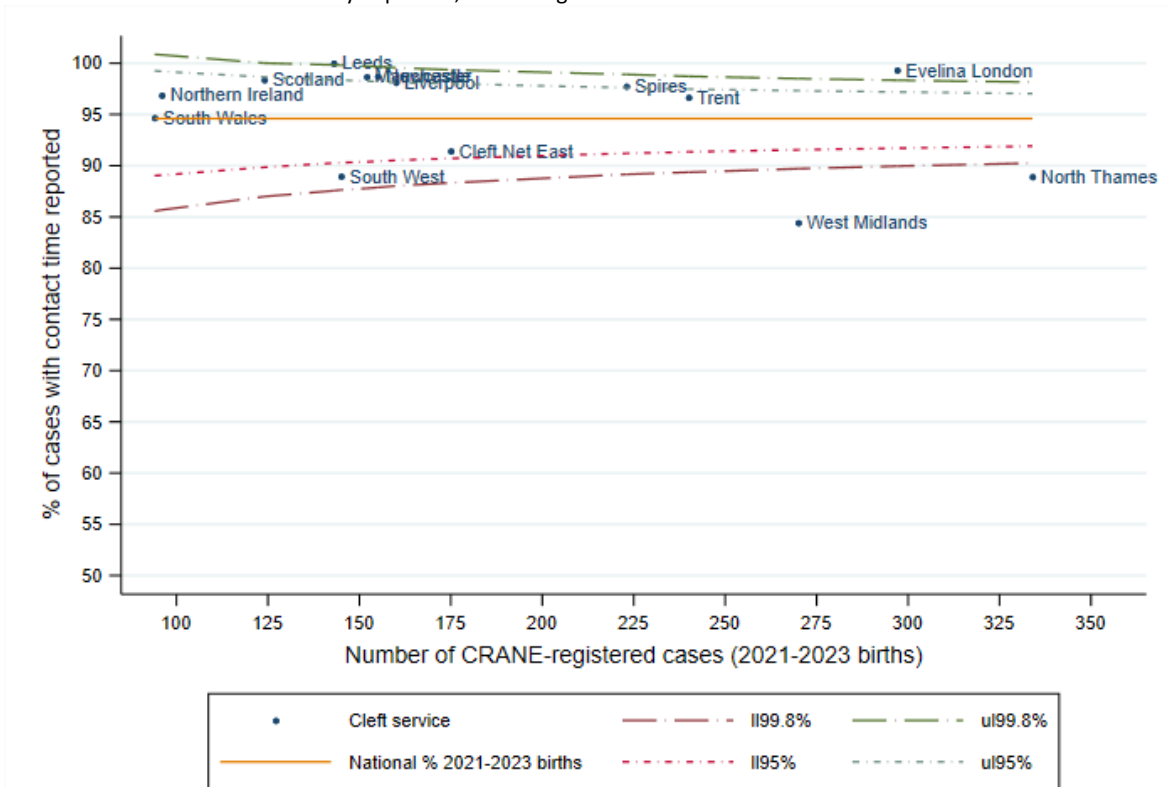


Note: Funnel plot centred on the national percentage (94.6%) of children (born 2021-2023) with contact time reported.

| Outcome: Contact between Cleft Service and family within 24 hours of postnatal referral | |
|---|---|
| Indicator | #8a - Contact between Cleft Service and family within 24 hours of postnatal referral |
| Benchmarks | 94% of children born with a cleft in 2020-2022 were contacted by their Cleft Service within 24 hours of the service receiving their postnatal referral (CRANE, 2023). |
| Denominator | 2,466 CRANE-registered cases with postnatal contact time reported |
| What did we find? | • 94% of children were contacted by Cleft Services within 24hrs of the service receiving a postnatal referral (Cleft Service range: 81%-100%, p<0.001). This is similar to the previous reporting period (2020-2022 births). <br><br> • The percentage of families receiving contact within 24hrs of referral varied significantly by cleft type (CL: 96%, CP: 94%, UCLP:95%, BCLP: 95%, SMCP:+CL: 89%, SMCP:71%, p<0.001), but not by birth year (2021: 95%, 2022: 94%, 2023: 95%, p=0.596) or sex (female:94%, male: 95%, p=0.700). |
| Outliers | Positive:  1. Trent (100%), Manchester* (100%) <br><br> Negative: 1. Northern Ireland (81%), 2. West Midlands (84%), 3. North Thames (90%), 4. Spires* (93%) <br><br> *Same alert status for two consecutive reporting periods and therefore classed as outlier. |

**Figure 3.11.** Funnel plot showing the percentage of CRANE-registered children, born 2021 to 2023, who had been contacted by a Cleft Service within 24hrs of postnatal referral, according to Cleft Service.



Note: Funnel plot centred on the revised national percentage (96.6%) of children (born 2021-2023) contacted within 24hrs of referral.

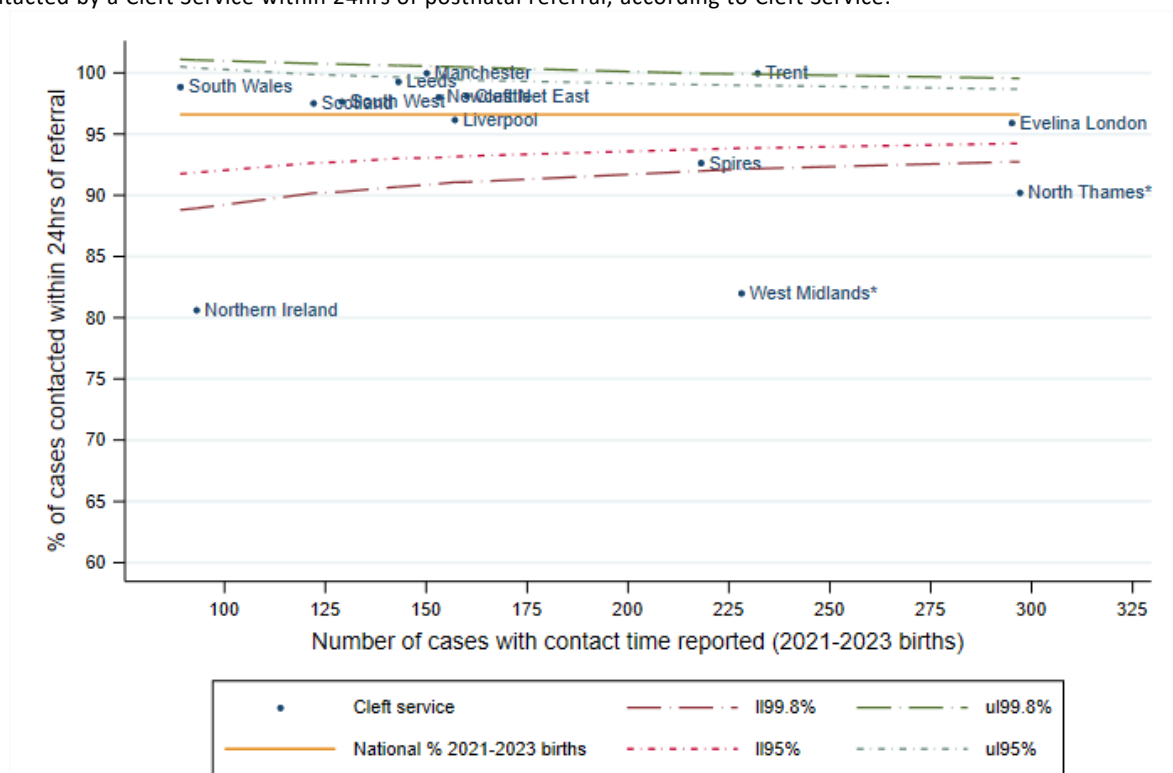| Outcome: Family visited by Clinical Nurse Specialist within 24 hours of postnatal referral | |
|---|---|
| Indicator | #8b– Visit by Clinical Nurse Specialist (CNS) within 24 hours of postnatal referral |
| Benchmarks | 81% of children born with a cleft in 2020-2022 were visited by a CNS within 24 hours of the service receiving their referral (CRANE, 2023). |
| Denominator | 2,608 CRANE-registered cases |
| What did we find?<br> | • 84% of children were visited by Cleft Services within 24hrs of the service receiving a postnatal referral (Cleft Service range: 35%-97%, p<0.001).<br>• The percentage of families visited by a CNS within 24hrs of postnatal referral varied significantly by known cleft type (CL:87%, CP: 83%, UCLP: 91%, BCLP: 86%, SMCP+CL:53%, SMCP: 44%, p<0.001) but not by birth year (2021: 82%, 2022: 83%, 2023: 86%, p=0.132) or sex (female: 84%, male: 84%, p=0.624). |
| Outliers | Positive: 1. Leeds (97%), 2. Liverpool (97%), 3. Evelina London (95%), 4. Scotland (95%), 5. Manchester (94%), 6. Trent (93%)<br>Negative: 1. Northern Ireland (35%), 2. West Midlands (66%), 3. South West (68%), 4. North Thames (71%) |

**Figure 3.12.** Funnel plot showing the percentage of CRANE-registered children, born 2021 to 2023, who were visited by a Clinical Nurse Specialist within 24hrs of postnatal referral, according to Cleft Service.



Note: Funnel plot centred on the national percentage (84.4%) of children (born 2021-2023) visited by a CNS within 24hrs of postnatal referral.

| Recommendations: Referral and contact |
|---|

- Cleft Services should record the referral and contact time for all registrations by working with referring obstetric, midwifery and neonatal units to improve the capture of this information.
- Regional variation in the percentage of children referred, contacted and visited 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.

# 4. Consent

Cleft Services are responsible for approaching the families of all children born alive with a cleft to obtain consent for CRANE to collect outcome data. This section reports the consent status of children born 2021-2023, reflecting recent registrations, and 2015-2017, reflecting registrations of children who should have undergone their 5-year-old assessment of cleft-related outcomes. Consent verification is a key performance indicator and denotes a confirmed consent status, whereby families have either given informed consent or declined consent for CRANE to collect outcome data. In accordance with our Outlier Policy[21], data from any service identified as a negative outlier for consent verification will be excluded from revised totals and averages used to generate funnel plots of outcomes collected for consented children only. This ensures that results reflect patient populations only from services with acceptable levels of consent verification.

## 4.1. CRANE consent, 2021-2023 births

| Cohort summary | |
|---|---|
| Data source | The CRANE Database. Extract taken: 1 July 2024 |
| Birth years | Three years: 2021 to 2023 |
| Countries | England, Wales, Northern Ireland and Scotland |
| Inclusions | CRANE-registered children |
| Exclusions | • Children who died<br>• Children with submucous cleft palate[22] |
| Notes | • Consent verification is not subject to outlier policy for recent birth years<br>• Data are not risk-adjusted |
| Raw data | 'Consent 2021-23' in **the supplementary tables**. |

| Outcome: Consent status | |
|---|---|
| Indicator | #9 - Consent status verified for all CRANE-registered children. |
| Benchmarks | • 100% of CRANE-registered children should have their consent status verified (informed consent given or declined) regarding CRANE outcome data collection.<br>• 81% of children born 2020-2022 had a verified consent status (CRANE, 2023). |
| Denominator | 2,507 (2,502 CRANE-registered children + 5 non-consented/registered children in Scotland) |
| What did we find? | • 88% of eligible children had a verified consent status (Cleft Service range: 66%-100%, p<0.001). This rate increased by 7% compared to the previous reporting period (births 2020-2022).<br>• Consent verification varied significantly according to known cleft type (CL: 88%, CP: 88%, UCLP: 92%, BCLP: 94%, p=0.004) but not by sex (female: 90%, male: 88%, p=0.125) or birth year (2021: 88%, 2022: 89%, 2023: 88%, p=0.943).<br>• 86% had provided informed consent to outcome data collection in CRANE. Among those with verified consent status, 97% gave consent (Cleft Service range: 87%-100%, p<0.001).<br>• 3% declined consent.<br>• 11% were awaiting consent verification.<br>• <1% were not possible to verify.<br>• Of those with consent, 99% also consented to linkage of CRANE data with health and education data. |

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[21] CRANE Outlier Policy. https://www.crane-database.org.uk/content/uploads/2022/12/CRANE-Outlier-Policy_20Mar2023.pdf
[22] Patients with submucous cleft palate are excluded from outcomes.

| Outliers | Positive: 1. Liverpool (100%), 2. Manchester (99%), 3. Scotland (97%), 4. North Thames (97%) |
|---|---|
| | Negative: 1. West Midlands (66%) |

**Figure 4.1.** Funnel plot showing the percentage of CRANE-registered children, born 2021 to 2023, who had verified consent status, according to Cleft Service.
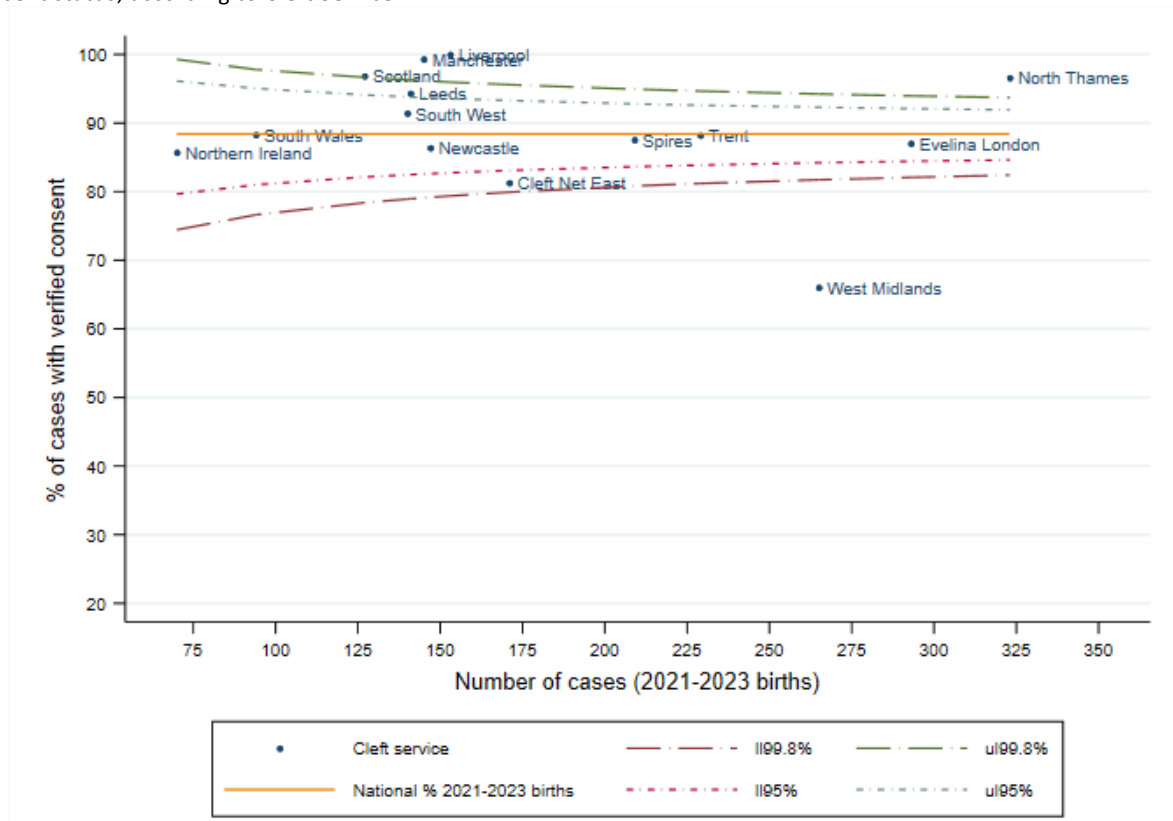


## 4.2. CRANE consent, 2015-2017 births

| Cohort summary | |
|---|---|
| Data source | The CRANE Database. Extract taken: 1 July 2024 |
| Birth years | Three years: 2015 to 2017 |
| Countries | England, Wales, Northern Ireland and Scotland. |
| Inclusions | CRANE-registered children |
| Exclusions | • Children who died before the age of 5 years<br>• Children with submucous cleft palate[23] |
| Notes | • Subject to outlier policy.<br>• Data are not risk-adjusted.<br>• Scotland joined CRANE in 2023 and has retrospectively entered data for 2016-2017 births to be included in analyses. Legally, Scotland can only register on CRANE consented cases, and therefore must provide us with their denominator separately. |
| Raw data | 'Consent 2015-17' in **the supplementary tables**. |

---

[23] Patients with submucous cleft palate are excluded from outcomes.

| Outcome: Consent status | |
|---|---|
| Indicator | #9 - Consent verification status verified for all CRANE-registered children |
| Benchmarks | • 100% of CRANE-registered children should have their consent status verified (informed consent given or declined) regarding CRANE outcome data collection.<br>• 90% of 5-year-old children born 2014-2016 had a verified consent status (CRANE, 2023). |
| Denominator | 3,249 (3,191 CRANE-registered children + 58 non-consented/registered children in Scotland) |
| What did we find?<br> | • 91% of eligible children had a verified consent status (Cleft Service range: 66%-100%, p<0.001). This rate is similar to the previous reporting period (2014-2016 births).<br>• Consent verification varied significantly according to known cleft type (CL: 91%, CP: 93%, UCLP: 95%, BCLP: 97%, p<0.001) but not by sex (female: 93%, male: 93%, p=0.915) or birth year (2015: 92%, 2016: 93%, 2017: 92%, p=0.521).<br>• 88% had provided informed consent to outcome data collection in CRANE. Among those with verified consent status, 98% gave consent (Cleft Service range: 94%-100%, p<0.001).<br>• 2% declined consent.<br>• 8% were awaiting consent verification.<br>• <2% were not possible to verify. |
| Outliers | Positive:  1. Leeds (100%), 2. Newcastle (98%), 3. North Thames* (95%)<br><br>Negative: 1. Scotland (66%)<br><br>*Positive alert for two consecutive reporting periods and therefore classed as outlier. |

**Figure 4.2.** Funnel plot showing the percentage of CRANE-registered 5-year-olds, born 2015 to 2017, who had verified consent status, according to Cleft Service.



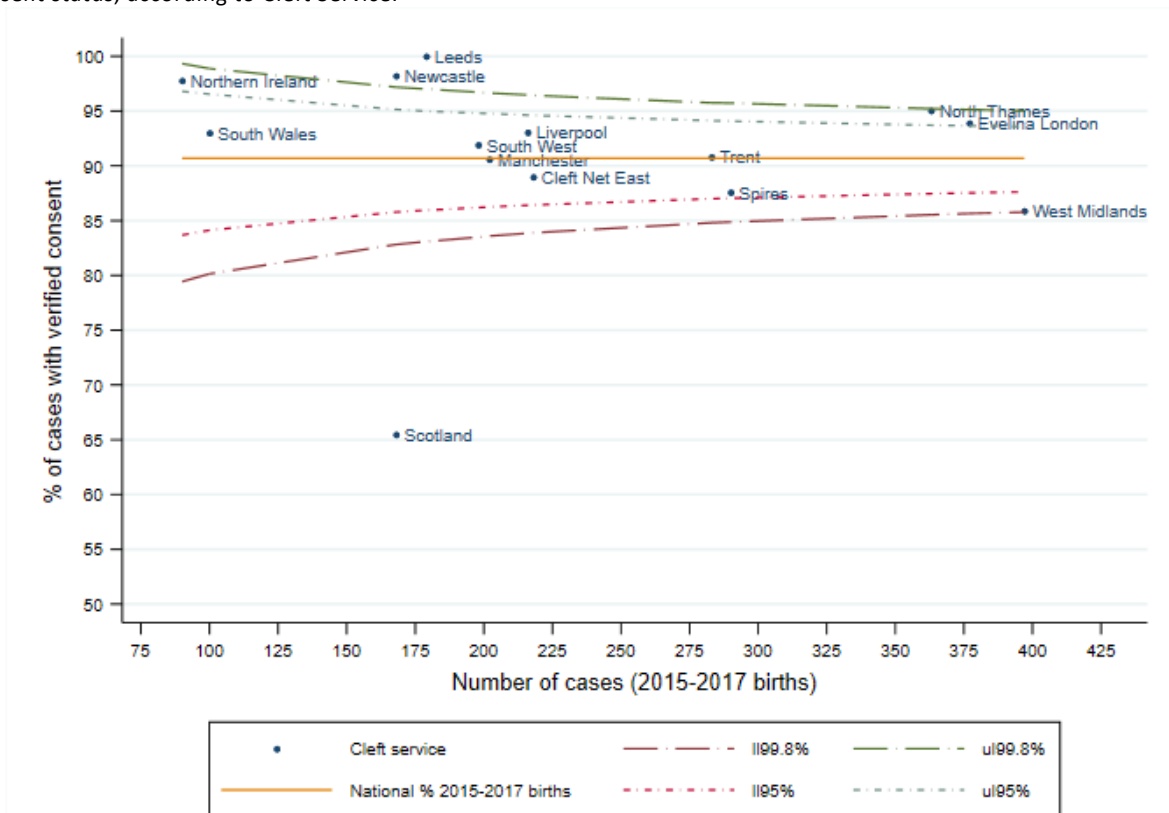| Recommendations: Consent |
|---|
| • Cleft Services with high consent rates should share their best practice recommendations.<br>• Cleft Services with below average consent rates should review their procedures to identify opportunities to make improvements. |

# 5. Audit outcomes at 5 years of age

This chapter presents cleft-related 5-year outcomes for CRANE-consented children, born 2015 to 2017.

**Figure 5.1.** CRANE cohort eligible for 5-year outcome reporting.

```
┌─────────────────────────────────────────┐
│                  3,374                   │
│  CRANE-registered children born 2015-2017│
│  in England, Wales, Northern Ireland and │
│              Scotland                    │
└─────────────────────────────────────────┘
              │           ┌──────────────────────────────────────┐
              ├───────────┤  407 children without consent excluded│
              │           └──────────────────────────────────────┘
┌─────────────────────────────────────────┐
│                  2,967                   │
│        CRANE-consented children          │
└─────────────────────────────────────────┘
              │           ┌──────────────────────────────────────┐
              ├───────────┤ 20 children who died before 5 years   │
              │           │ excluded                              │
              │           └──────────────────────────────────────┘
┌─────────────────────────────────────────┐
│                  2,947                   │
│   CRANE-consented children alive at      │
│            5 years                       │
└─────────────────────────────────────────┘
              │           ┌──────────────────────────────────────┐
              ├───────────┤ 75 children with submucous cleft palate│
              │           │ (SMCP) excluded as these cases are not │
              │           │ audited by Cleft Services             │
              │           └──────────────────────────────────────┘
┌─────────────────────────────────────────┐
│                  2,872                   │
│  children eligible for outcome reporting │
│            at 5 years                    │
└─────────────────────────────────────────┘
```

| Outcome | CLO N=669 (23%) | CPO N=1,219 (42%) | UCLP N=646 (23%) | BCLP N=290 (10%) | Non-specified N=48 (2%) |
|---|---|---|---|---|---|
| Child growth | ✓ | ✓ | ✓ | ✓ | ✓ |
| Dental health | ✓ | ✓ | ✓ | ✓ | ✓ |
| Facial growth | | | Complete UCLP only | | |
| Speech | | ✓ | ✓ | ✓ | |
| Psychology | ✓ | ✓ | ✓ | ✓ | ✓ |

**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.

## 5.1. Child growth

All Cleft Services are required to submit growth data (height and weight) for all consented 5-year-old children diagnosed with a cleft lip and/or palate.

| Cohort summary | |
|---|---|
| Data source | The CRANE Database (consented cases only). Extract taken: 1 July 2024 |
| Birth years | Three years: 2015 to 2017 |
| Countries | England, Wales, Northern Ireland and Scotland |
| Inclusions | CRANE-consented children, including those without a specified cleft type |
| Exclusions | • Children who died before the age of 5 years<br>• Children with submucous cleft palate |
| Notes | • Subject to outlier policy.<br>• Data are not risk-adjusted.<br>• Funnel plots are centred on the revised national rates after excluding data from services identified as negative outliers for consent verification (Scotland).<br>• Cleft Services with <10 cases are not shown on funnel plot due to insufficient data. |
| Raw data | 'Child growth 2015-17' in **the supplementary tables**. |

| Data completeness: Height and weight | |
|---|---|
| Indicator | #10 - Child growth at 5 years recorded for all eligible children. |
| Denominator | 2,872 CRANE-consented children |
| What did we find? | • 38% of eligible consented children had a recorded height and weight (Cleft Service range: 3%-82%, $p<0.001$). This rate compares to 36% for the previous reporting period (births 2014-2016).<br>• <1% of children only had a recorded height.<br>• <1% of children only had a recorded weight.<br>• 35% had a reason the child growth outcomes were not collected.<br>• 25% were missing data or a reason for not collecting data.<br>• The proportion of children with height and weight measures varied significantly according to birth year (2015: 22%, 2016: 42%, 2016: 50%, $p<0.001$) but not by cleft type (CL: 36%, CP:39%, UCLP:40%, BCLP: 43%, $p=0.163$) or sex (female: 38% male:38%, $p=0.818$). |
| Outliers | Positive:  1. South Wales (82%), 2. Northern Ireland (73%), 3. Newcastle (64%), 4. Manchester (63%),<br>         5. Cleft Net East (56%), 6. North Thames (52%)<br>Negative: 1. West Midlands (3%), 2. South West (9%), 3. Evelina London (15%), 4. Scotland (22%),<br>         5. Spires (29%) |

**Figure 5.2.** Funnel plot showing the percentage of CRANE-consented 5-year-olds, born 2015 to 2017, with growth data reported, according to Cleft Service.
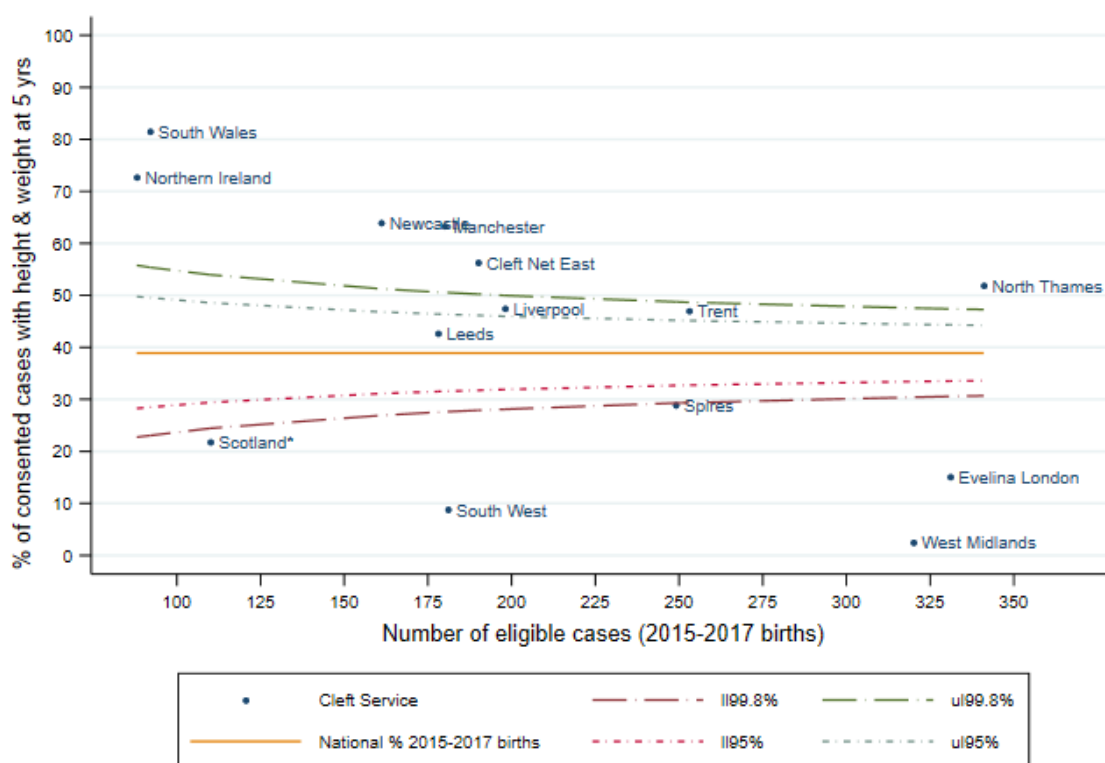


Note: Funnel plot centred on the revised national percentage (38.9%) of children (born 2015-2017) with child growth outcomes reported.

| Outcome: Healthy BMI | |
|---|---|
| Indicator | #11 - Healthy Body Mass Index (BMI) at 5 years of age. |
| Benchmarks | • Prevalence of underweight, healthy weight, overweight and obesity among 5-year-olds in the general population is estimated at 1%, 77%, 12% and 10%, respectively[24]. <br> • 82% of eligible children born 2014-2016 were reported to have a healthy BMI (CRANE, 2023). |
| Notes | • Body mass index (BMI) at 5 years of age was calculated as weight (kg)/height (m)$^2$. <br> • For 5-year-olds in the UK[25], Underweight is BMI <13.0 kg/m2, healthy BMI 13.0-17.5 (2nd to 91st centiles), overweight is BMI 17.5-19.0 kg/m2 (92nd to 98th centiles), and obese is BMI >19.0 kg/m$^2$. <br> • Funnel plot is centred on the revised national rate after excluding data from services identified as negative outliers for consent verification (Scotland) and child growth data completeness (West Midlands, Spires, Evelina London, South West and Scotland). West Midlands is not plotted due to insufficient BMI data. |
| Denominator | 1,099 CRANE-consented children with a recorded height and weight |

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[24] National Child Measurement Programme Tables, England 2021/22 and 2022/23 School Years –Available at National Child Measurement Programme - NHS England Digital [Last accessed: 12/07/2024]

[25]According to the Royal College of Paediatrics and Child Health and UK-WHO growth charts – 2-18 years – Available at: https://www.rcpch.ac.uk/resources/uk-who-growth-charts-2-18-years [Last accessed: 12/12/2022]

| | |
|---|---|
| What did we find?  | - Average weight, height and BMI for those with reported data was 20.4kg (95%CI 20.2-20.6kg), 113.6cm (95%CI 113.2-113.9cm) and 15.7 kg/m$^2$ (95% CI: 15.6 kg/m$^2$ to 15.8 kg/m$^2$), respectively. |
| | - 83% of children had a healthy BMI (Cleft Service range: 73% to 96%[26], p=0.332). This represents minimal change from the previous reporting period and is higher than the 77% reported for the general population. |
| | - 3% of children were underweight, 9% overweight and 5% obese. The proportion of children within each BMI category varied significantly according to cleft type (p=0.048[27]), sex (female: underweight: 6%, healthy weight:79%, overweight: 11%, obese: 4%; Male: underweight: 1%, healthy weight: 86%, overweight: 7%, obese: 5%, p<0.001), but not birth year (p=0.431). |
| Outliers | None. |

**Figure 5.3.** Funnel plot showing the percentage of CRANE-consented 5-year-olds, born 2015 to 2017, with a healthy BMI, according to Cleft Service.



Note: Funnel plot centred on the revised national percentage (82.2%) of children (born 2015-2017) with a healthy BMI.

| Recommendations: Child growth |
|---|
| - 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 cleft subtypes in the future. |
| - CRANE will continue to liaise with CDG members and the nursing CEN to encourage all services to collect this data. |
| - Research should explore reasons why the BMI distribution differs between the cleft and general population of 5-year-olds. |

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[26] Note that Scotland (96%) were a negative outlier for consent verification and child growth data completeness so interpret this rate with caution. The highest rate among services not identified as negative outliers for consent or data completion was 90% (Leeds).

[27] See supplementary tables for breakdown by cleft type.

## 5.2. Dental health

5-year-old children with all cleft types are eligible for an assessment of their dental health and have this recorded as a decayed, missing or filled teeth (dmft) score. A dmft score is a measure of oral health and reflects the total number of teeth that are decayed, missing or filled. A dmft >0 indicates experience of dental decay and dmft >5 indicates experience of extensive dental decay. 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[28].

| Cohort summary | |
|---|---|
| Data source | The CRANE Database (consented cases only). Extract taken: 1 July 2024 |
| Birth years | Three years: 2015 to 2017 |
| Countries | England, Wales, Northern Ireland and Scotland |
| Inclusions | CRANE-consented children, including those without a specified cleft type |
| Exclusions | • Children who died before the age of 5 years<br>• Children with submucous cleft palate |
| Notes | • Subject to outlier policy.<br>• Data are not risk-adjusted.<br>• Cleft Services with <10 cases are not shown on funnel plot due to insufficient data.<br>• Funnel plots are centred on the revised national rates after excluding data from services identified as negative outliers for consent verification (Scotland). |
| Raw data | 'Dental health 2015-17' in **the supplementary tables** |

| Definitions for measures of intervention | |
|---|---|
| Treatment Index | 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[29]. 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 caries free dentition. |
| Care Index | Care Index reflects the extent to which dental decay has been successfully treated by restorative techniques (e.g. 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[30]. 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 some cases, a tooth may be so malformed that it cannot be restored even if identified early and extraction may be the only option |

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[28] (1) Al-Dajani. 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):529-531. (2) Britton and Welbury, 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):236-241.

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

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

| Data completeness: dmft scores | |
|---|---|
| Indicators | #12 – dmft at 5 years recorded for all eligible children |
| Denominator | 2,872 CRANE-consented children |
| What did we find? | • 45% of eligible consented children had recorded dmft scores (Cleft Service range: 3%-80%, p<0.001). This rate compares to 36% for the previous reporting period (births 2014-2016). <br> • 32% had a reason dmft scores were not collected. <br> • 23% were missing data or a reason for not collecting data. <br> • The proportion of children with dmft scores varied significantly according to birth year (2015: 31%, 2016: 47%, 2017: 57%, p<0.001). Data completeness also varied according to cleft type (CL: 41%, CP: 45%, UCLP: 50%, BCLP: 47%, p=0.016) but not by sex (female: 45%, male: 45%, p=0.944). |
| Outliers | Positive: 1. South Wales (80%), 2. Newcastle (74%), 3. Scotland (74%), 4. Northern Ireland (71%), 5. Cleft Net East (70%), 5. Manchester (66%) <br><br> Negative: 1. North Thames (3%), 2. Evelina London (31%), 3. West Midlands (34%) |

**Figure 5.4.** Funnel plot showing the percentage of CRANE-consented 5-year-olds, born 2015 to 2017, who had decayed, missing or filled teeth (dmft) scores reported, according to Cleft Service.
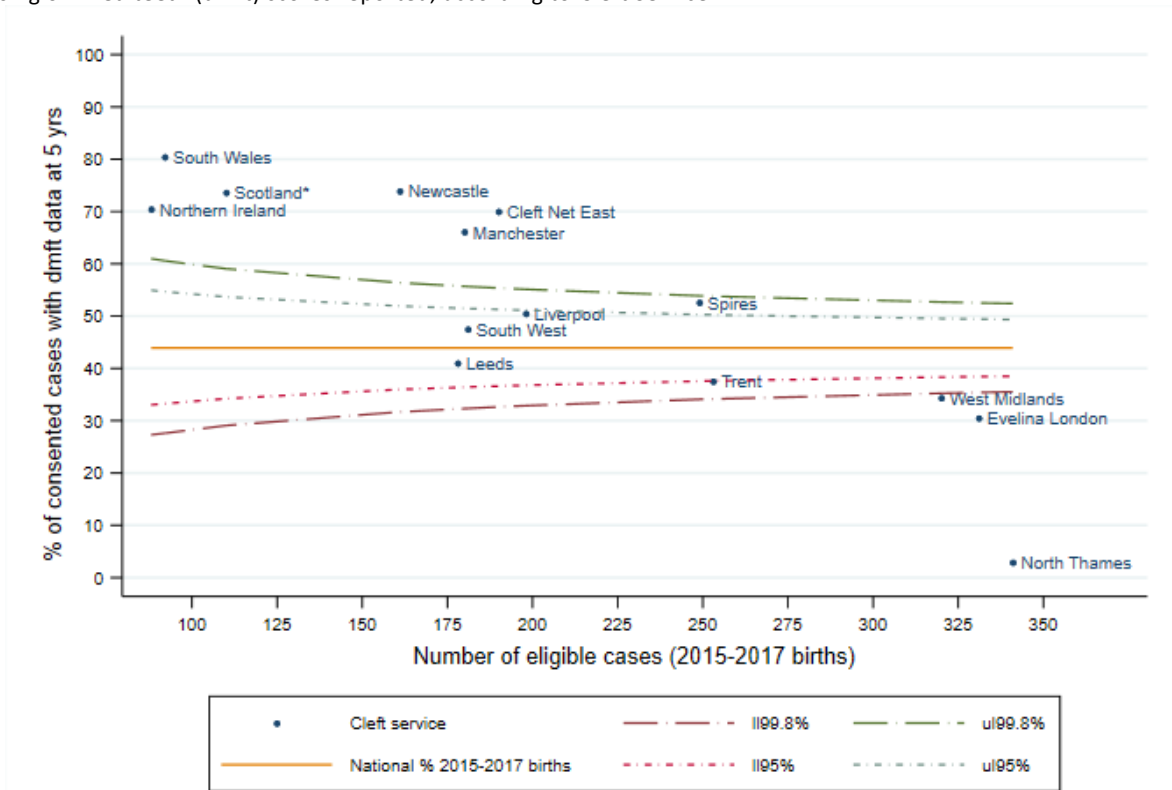


Note: Funnel plot centred on the revised national percentage (43.9%) of children (born 2015-2017) with dmft scores reported.

## 5.2.1. Dental decay

| | |
|---|---|
| **Outcome: dmft >0 scores** | |
| Indicator | #13 – Dental decay at 5 years of age |
| Benchmarks | • The Oral Health Survey of 5-year-old children in 2022 reported that 29% of children in the general population of England had dental decay, with at least one (>0) dmft[31]. <br> • 38% of eligible children born 2014-2016 were reported to have dmft >0 scores (CRANE, 2023). |
| Notes | Funnel plots are centred on the revised national rates after excluding data from services identified as negative outliers for consent verification (Scotland) and dmft data completeness (North Thames, Evelina London, and West Midlands). |
| Denominator | 1,294 CRANE-consented children with a recorded total decayed, missing or filled teeth (dmft) score |
| What did we find? | • The mean number of dmft at 5 years was 2.13, with scores ranging from 0 to 21. <br> • 38% of children with a cleft had at least one (>0) dmft (Cleft Service range: 10%[32]-49%, p=0.001). <br> • The proportion of children with >0 dmft varied significantly between cleft types (CL: 32%, CP: 37%, UCLP: 42%, BCLP: 45%, p=0.014) but not by birth year (2015: 36%, 2016: 38%, 2017: 39%) or by sex (female: 38%, male: 38%, p=0.965). |
| Outliers | Positive: 1. West Midlands (21%), 2. Spires* (35%) <br> Negative: None <br> * Positive alert status for two consecutive reporting periods and therefore classed as outlier. |

**Figure 5.5.** Funnel plot showing the percentage of CRANE-consented 5-year-olds, born 2015 to 2017, with experience of dental decay (dmft >0), according to Cleft Service.



Note: Funnel plot centred on the revised national percentage (41.1%) of children (born 2015-2017) with >0 dmft.

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[31] National Dental Epidemiology Programme (NDEP) for England: Oral health survey of 5 year old children 2022 Data on children in the general population in Wales and Northern Ireland were not available at the time of producing this report.
[32] Note that North Thames (10%) was a negative outlier for data completeness so interpret this rate with caution. The lowest rate among services not identified as negative outliers for consent or data completeness was 31% (Spires).

## 5.2.2. Extensive dental decay

| | |
|---|---|
| Outcome: dmft >5 scores | |
| Indicator | #14 – Extensive dental decay at 5 years of age |
| Benchmarks | • The Child Dental Health Survey in 2013 reported that 13% of children in the general population of England, Wales and Northern Ireland had extensive dental decay (dmft >5) at 5 years of age[33].<br>• 13% of eligible children born 2014-2016 were reported to have dmft >5 scores (CRANE, 2023). |
| Notes | Funnel plots are centred on the revised national rates after excluding data from services identified as negative outliers for consent verification (Scotland) and dmft data completeness (North Thames, Evelina London, and West Midlands). |
| Denominator | 1,294 CRANE-consented children with a recorded total decayed, missing or filled teeth (dmft) score |
| What did we find? | • 15% of children with a cleft had extensive dental decay (dmft >5) (Cleft Service range: 0%[34]-23%, p=0.007).<br>• The proportion of children with dmft >5 varied significantly between cleft types (CL: 12%, CP: 15%, UCLP: 14%, and BCLP: 26%, p=0.001) but not by birth year (2015: 13%, 2016: 14%, 2017: 17%, p=0.199) or by sex (female: 16%, male: 15%, p=0.620). |
| Outliers | Positive: 1. West Midlands (6%), 2. Spires* (8%)<br><br>Negative: None<br><br>* Positive alert status for two consecutive reporting periods and therefore classed as outlier. |

**Figure 5.6.** Funnel plot showing the percentage of CRANE-consented 5-year-olds, born 2015 to 2017, with extensive dental decay (dmft >5), according to Cleft Service.



Note: Funnel plot centred on the revised national percentage (16.5%) of children (born 2015-2017) with >5 dmft.
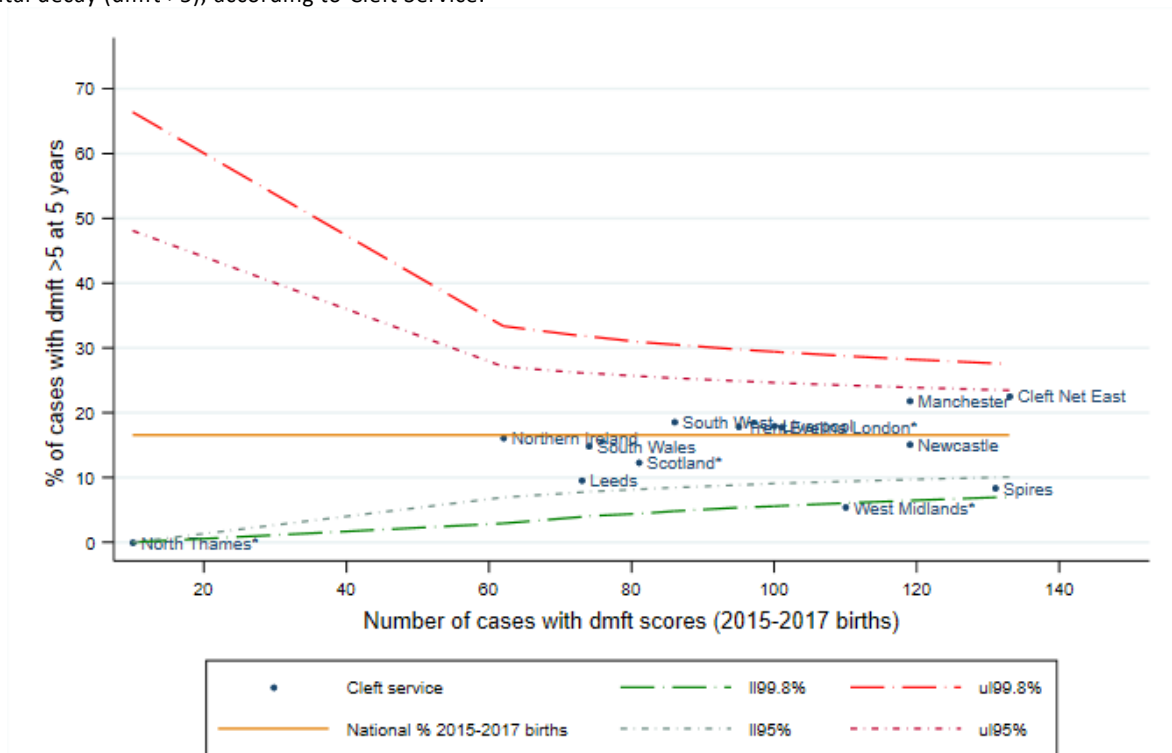
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[33] Child Dental Health Survey 2013, England, Wales and Northern Ireland - https://digital.nhs.uk/data-and-information/publications/statistical/children-s-dental-health-survey/child-dental-health-survey-2013-england-wales-and-northern-ireland

[34] Note that North Thames (0%) was a negative outlier for data completeness so interpret this rate with caution. The lowest rate among services not identified as negative outliers for consent or data completeness was 8% (Spires).

### 5.2.3. Dental health measures of intervention

| Treatment Index | |
|---|---|
| Indicator | #15 – No untreated disease, as measured by the dental treatment index at 5 years of age |
| Benchmarks | • The Oral Health Survey of 5-year-old children in 2022 reported that the average Treatment Index for children is 14% in the general population in England[35].<br>• The average Treatment Index for eligible children born 2014-2016 was 77% (CRANE, 2023). |
| Notes | • High mean treatment index scores indicate that children have high levels of treated dental disease.<br>• Not subject to outlier policy but funnel plots provided to demonstrate variation in rates across services.<br>• Funnel plots are centred on the revised national rates after excluding data from services identified as negative outliers for consent verification (Scotland) and dmft data completeness (North Thames, Evelina London, and West Midlands). |
| Denominator | 1,294 CRANE-consented children with scores for the calculation of Treatment Index[36] |
| What did we find? | • The average Treatment Index for these children was 75% (Cleft Service range: 63%-100%[37], p=0.007)[38].<br>• Treatment Index did not vary significantly between cleft types (CL: 76%, CP: 76%, UCLP: 74%, and BCLP: 76%, p=0.871), by birth year (2015: 77%, 2016: 74%, 2017: 75%, p=0.853), or by sex (female: 76%, male: 74%, p=0.491). |
| Outliers | Positive:  1. Cleft Net East (85%)<br>Negative: None |

**Figure 5.7.** Funnel plot showing the average dental Treatment Index percentage for CRANE-consented 5-year-olds, born 2015 to 2017, according to Cleft Service.



Note: Funnel plot centred on the revised national Treatment Index percentage (72.7%) for children (born 2015-2017).

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[35] National Dental Epidemiology Programme (NDEP) for England: Oral health survey of 5 year old children 2022
[36] 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).
[37] Note that North Thames (100%) was a negative outlier for data completeness so interpret this rate with caution. The highest rate among services not identified as negative outliers for consent or data completion was 85% (Cleft Net East).
[38] A Kruskall-Wallis test was used to compare the mean rank of Treatment Index scores between subgroups.

| Outcome: Care Index | |
|---|---|
| Indicator | #16 – No untreated disease, as measured by the dental care index at 5 years of age |
| Benchmarks | • The Oral Health Survey of 5-year-old children in 2022 reported that the average Care Index for children is 7% (100% is the desirable outcome) in the general population in England[39].<br>• The average Care Index for eligible children born 2012-2014 was 70% (CRANE, 2023). |
| Notes | • High mean care index scores indicate that children have received the appropriate care at the earliest possible stage.<br>• Not subject to outlier policy but funnel plots provided to demonstrate variation in rates across services.<br>• Funnel plots are centred on the revised national rates after excluding data from services identified as negative outliers for consent verification (Scotland) and dmft data completeness (North Thames, Evelina London, and West Midlands). |
| Denominator | 1,294 CRANE-consented children with scores for the calculation of Care Index[40] |
| What did we find? | • The average Care Index for these children was 68% (Cleft Service range: 56%-90%[41], p=0.0020).<br>• Care Index did not vary significantly between cleft types (CL: 73%, CP: 69%, UCLP: 65%, and BCLP: 63%, p=0.083), by birth year (2015: 71%, 2016: 66%, 2017: 68%, p=0.626), or by sex (female: 69%, male: 67%, p=0.591). |
| Outliers | Positive:  1. West Midlands (81%)<br>Negative: 1. Manchester* (56%)<br>* Negative alert status for two consecutive reporting periods and therefore classed as outlier. |

**Figure 5.8.** Funnel plot showing the average dental Care Index percentage for CRANE-consented 5-year-olds, born 2015 to 2017, according to Cleft Service.



Note: Funnel plot centred on the revised national Care Index percentage (65.4%) for children (born 2015-2017).

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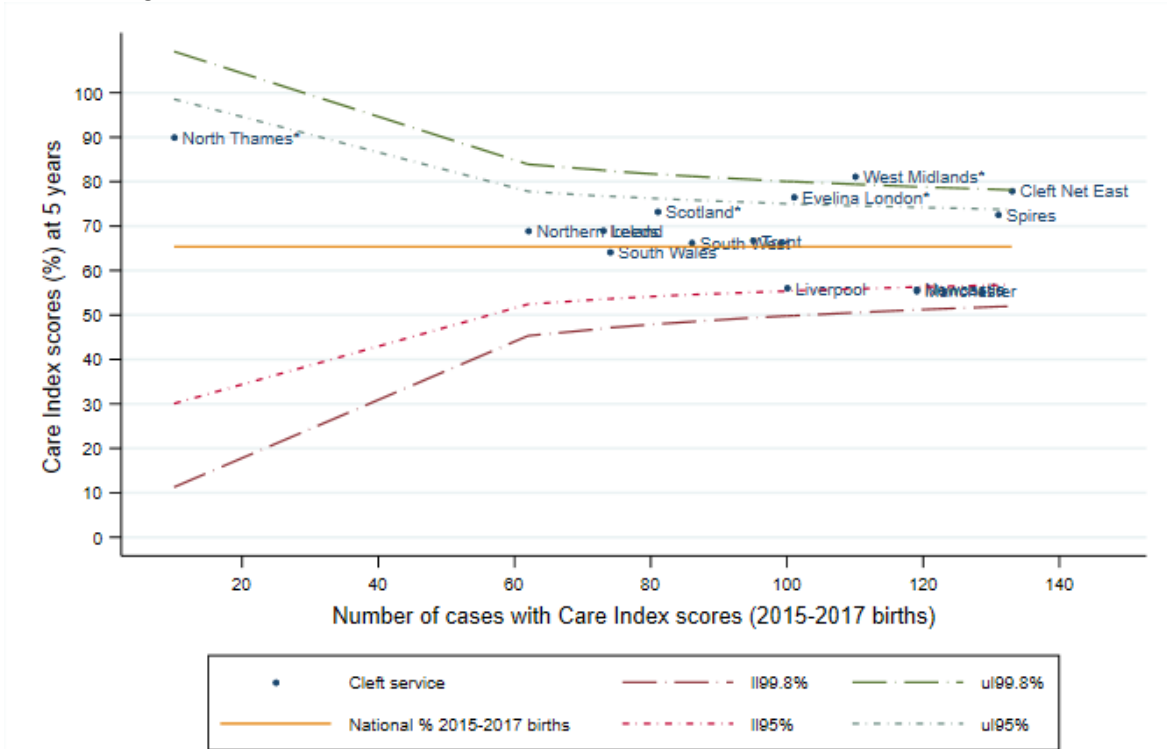[39] National Dental Epidemiology Programme (NDEP) for England: Oral health survey of 5-year-old children 2022
[40] Care Index calculated using: data on filled teeth (f) and dmft scores; or a dmft score of 0 (equating to a Care Index = 1).
[41] Note that North Thames (90%) was a negative outlier for data completeness so interpret this rate with caution. The highest rate among services not identified as negative outliers for consent or data completion was 78% (Cleft Net East).

| Recommendations: Dental health |
| --- |

- Cleft Services should have at least 80% of all children with a cleft assessed at the age of 5 years by a calibrated specialist in paediatric dentistry, in line with paediatric dentistry CEN standards, and the dmft information should be recorded in the CRANE Database.

- All children with a cleft should have a recommended care plan established by collaborative work between the family's local dental care provider and the specialist paediatric dentist in the Cleft Service. This should (a) treat the child as per the high-risk category of the dental health toolkit (Delivering Better Oral Health), (b) provide routine dental care within the general dental service, and (c) provide specialist level care including age-specific dental development assessment and treatment under inhalation sedation and general anaesthesia within the Cleft Service.

- The COVID-19 pandemic had a significant impact on access to dental care in the early years for this population of patients, particularly those from more deprived areas[42]. Anecdotal and local evidence suggests that access to NHS dental care has still not recovered to pre-pandemic levels particularly in some regions of the country.  All children with a cleft should have access to a local NHS dental provider by their 1st birthday to instigate early preventive advice and build a positive dental relationship.

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[42] Aminu AQ, McMahon AD, Clark C, Sherriff A, Buchanan C, Watling C, Mahmoud A, Culshaw S, Mackay W, Gorman M, Braid R, Edwards M, Conway DI. Inequalities in access to NHS primary care dental services in Scotland during the COVID-19 pandemic. Br Dent J. 2023 May 24:1–6. doi: 10.1038/s41415-023-5856-z. Epub ahead of print. PMID: 37225842; PMCID: PMC10208681.
O´Connor, R., Landes, D. & Harris, R. Trends and inequalities in realised access to NHS primary care dental services in England before, during and throughout recovery from the COVID-19 pandemic. Br Dent J (2023). https://doi.org/10.1038/s41415-023-6032-1
Stennett, M., Tsakos, G. The impact of the COVID-19 pandemic on oral health inequalities and access to oral healthcare in England. Br Dent J 232, 109–114 (2022). https://doi.org/10.1038/s41415-021-3718-0

## 5.3   Facial growth

Records of facial growth (impressions or photographs) from 5-year-old children with a complete UCLP are 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 (e.g. orthodontics / alveolar bone grafting), which may influence growth[43]. 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[44]. The 5-year-old Index has, therefore, been used to compare treatment outcomes between Cleft Services. 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. Recently, data have been published showing that, whilst 'good' scores at 5 years of age have strong predictive value in terms of predicting the long-term (mid to late teenage years) value of dental arch relationships, 'fair' and 'poor' 5-year-old Index scores have limited predictive value[45]. As such, 'fair' and 'poor' scores should be interpreted with caution at 5 years of age.

| Cohort summary | |
|---|---|
| Data source | The CRANE Database (consented cases only). Extract taken: 1 July 2024 |
| Birth years | Three years: 2015 to 2017 |
| Countries | England, Wales, Northern Ireland and Scotland |
| Inclusions | • CRANE-consented children<br>• Complete UCLP |
| Exclusions | • Children who died before the age of 5 years |
| Notes | • Subject to outlier policy.<br>• Data are not risk-adjusted.<br>• Funnel plots are centred on the revised national rates after excluding data from services identified as negative outliers for consent verification (Scotland).<br>• Cleft Services with <10 cases are not shown on funnel plot due to insufficient data. |
| Raw data | 'Facial growth 2015-17' in **the supplementary tables**. |

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[43] 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.

[44] 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.

[45] 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

| Data completeness: 5-year-old Index scores | |
|---|---|
| Indicator | #17 – Facial growth at 5 years recorded for all eligible children. |
| Denominator | 496 CRANE-consented children eligible for 5-year-old Index scores |
| What did we find? | • 44% of eligible children had recorded 5-year-old Index scores (Cleft Service range: 0%-86%, p<0.001). This compares to 45% for the previous reporting period (births 2014-2016). |
| | • 187/218 (86%) scores were externally validated[46]. |
| | • 31% had a reason the facial growth scores were not collected. |
| | • 25% were missing data and a reason for not collecting data. |
| | • The proportion of children with 5-year-old index scores varied significantly according to birth year (2015: 24%, 2016: 59%, 2017: 49%, p<0.001), but not according to sex (female: 56%, male: 56%, p=0.994). |
| Outliers | Positive:  None |
| | Negative: 1. West Midlands (0%), 2. South West (15%) |

**Figure 5.9.** Funnel plot showing the percentage of CRANE-consented 5-year-olds, born 2015 to 2017, with a complete UCLP who had facial growth data reported, according to Cleft Service.
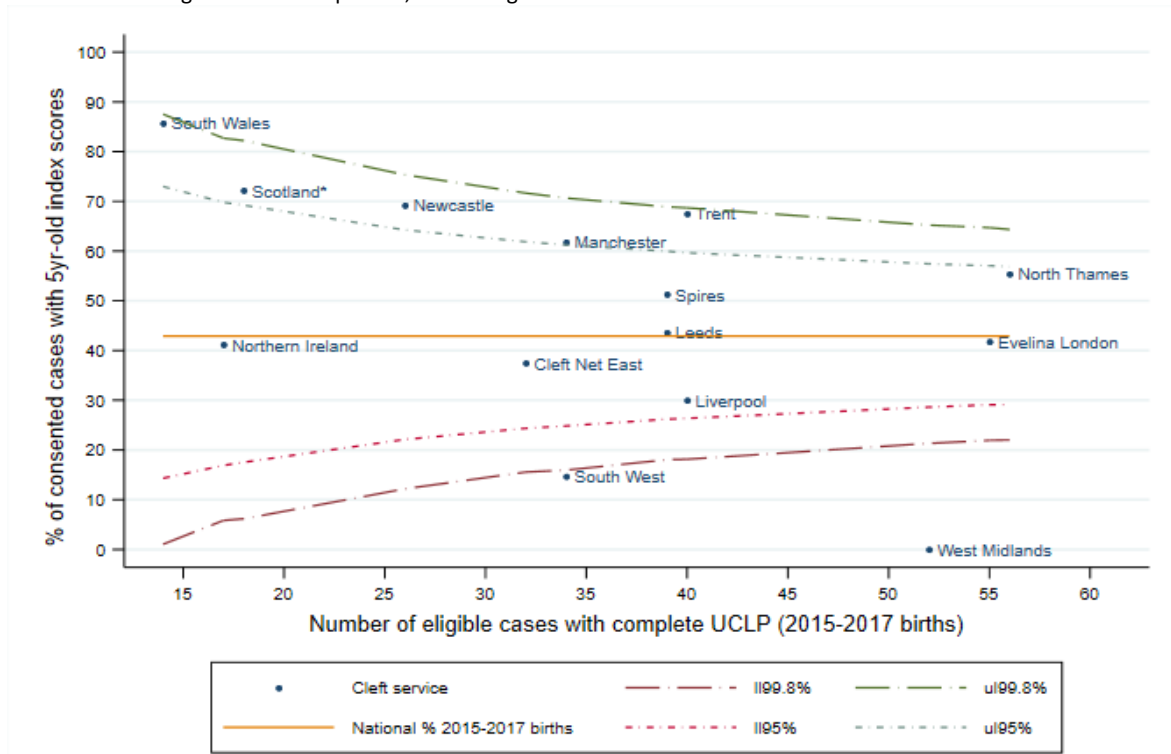


Note: Funnel plot centred on the revised national percentage (42.9%) of children (born 2015-2017) with facial growth data reported.

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[46] Some units score the models of children treated in their unit (internal scores) before they are sent off to be scored externally (external scores) by a blinded process undertaken by calibrated examiners. For this report we have analysed externally validated scores where available; where these were unavailable, internal scores are included in the analyses.

| Outcome: 5-year-old index scores | |
|---|---|
| Indicator | #18 – Children with Five-Year-Old Index scores reflecting 'good' dental arch relationships. |
| Benchmarks | • Cleft Care UK reported in 2015 that 53%, 28% and 19% of children with UCLP had 'good', 'fair' and 'poor' dental arch relationships, respectively[47]. <br> • 45% of eligible children born 2014-2016 were reported to have 'good' scores (CRANE, 2023). |
| Notes | • Funnel plot is centred on the revised national rate after excluding data from services identified as negative outliers for consent verification (Scotland) and facial growth data completeness (West Midlands and South West). <br> • West Midlands, South West, and Northern Ireland are not plotted due to insufficient data (n<10). |
| Denominator | 218 CRANE-consented children with 5-year-old index scores |
| What did we find? | • 52% of children had scores reflecting 'good' dental arch relationships at 5 years old (Cleft Service range: 40%-67%[48], p<0.001). This represents a 7% improvement on the previous reporting period (births 2014-2016) and is similar to the percentage reported in the Cleft Care UK study. <br> • 30% of children had scores reflecting 'fair' dental arch relationships at 5 years old. <br> • 18% of children had scores reflecting 'poor' dental arch relationships at 5 years old. <br> • Differences in the proportion of children with 'good' dental arch relationships were not statistically significant for birth year (2015: 35%, 2016: 56%, 2017: 55%, p=0.06) or sex (female: 45%, male: 55%, p=0.165). |
| Outliers | None. |

**Figure 5.10.** Funnel plot showing the percentage of CRANE-consented 5-year-olds, born 2015 to 2017, with a complete UCLP who had good facial growth scores, according to Cleft Service.
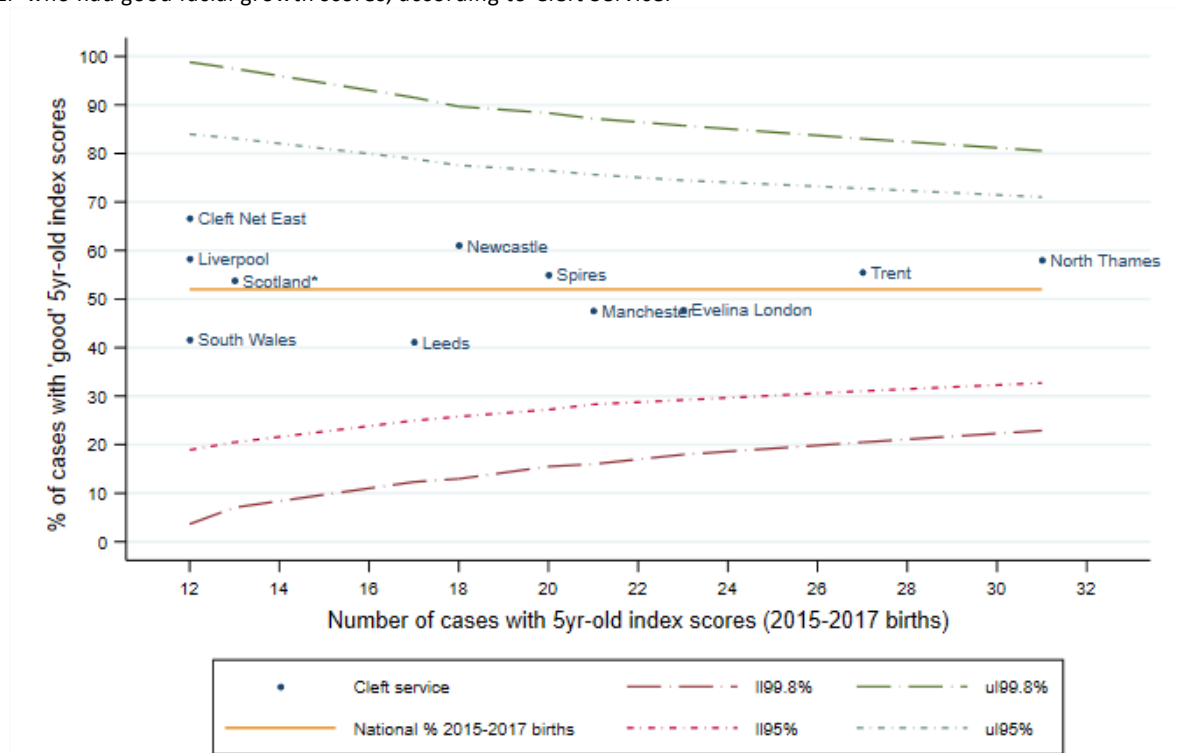


Note: Funnel plot centred on the revised national percentage (52.0%) of children (born 2015-2017) with 'good' facial growth outcomes.

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[47] Al-Ghatam, Jones, Ireland, Atack, Chawla et al. 2015 Structural outcomes in the Cleft Care UK study. Part 2: dento-facial outcomes. Orthodontics & Craniofacial Research 18(Suppl. 2): 14–24.
[48] among Cleft Services that submitted data for ≥10 cases.

| Recommendations: Facial growth |
|---|
| <ul><li>Cleft Services should aim to take records of all children born with a complete UCLP before they turn 6 years of age to support an external facial growth assessment using the 5-year-old index. These records may take the form of study models or clinical photographs with a recording of the overjet *(the horizontal gap between the front teeth)*. Study models can be made from dental impressions with a bite record or digital scans of the teeth and bite. Photography guidance should be sought from the IMI Guide to Good Practice for Cleft Lip and Palate (template 2a).</li><li>The research community should undertake to:<ul><li>compare UK facial growth outcomes with those in other countries, and</li><li>evaluate the predictive value of the 5-year-old Index in UK populations.</li></ul></li></ul> |

## 5.4. Speech

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

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

| Cohort summary | |
|---|---|
| Data source | The CRANE Database (consented cases only). Extract taken: 1 July 2024 |
| Birth years | Three years: 2015 to 2017 |
| Countries | England, Wales and Northern Ireland |
| Inclusions | <ul><li>CRANE-consented children</li><li>Children with a cleft affecting the palate (CP, UCLP, BCLP)</li></ul> |
| Exclusions | <ul><li>Children who died before the age of 5 years</li><li>Children with submucous cleft palate</li><li>Children with a diagnosed syndrome[49] entered onto the CRANE Database</li></ul> |
| Notes | <ul><li>Subject to outlier policy.</li><li>Data are not risk-adjusted.</li><li>All funnel plots are centred on the revised national rates after excluding data from services identified as negative outliers for consent verification (Scotland).</li><li>Speech standard outcome funnel plots are centred on the revised national rates after also excluding data from services identified as negative outliers for data completion (West Midlands and South West).</li></ul> |
| Raw data | 'Speech 2015-17' & '16-CAPS-A speech paramts' in **the supplementary tables**. |

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[49] 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).

| Data completeness: CAPS-A scores | |
|---|---|
| Indicator | #19 – Speech scores at 5 years recorded for all eligible children |
| Denominator | 1,811 CRANE-consented children eligible for CAPS-A assessment |
| What did we find? | • 54% of children had all 16 CAPS-A speech parameters reported (Cleft Service range: 28%-80%, p<0.001). This compares to 46% for the previous reporting period (births 2014-2016). Of those with speech data, 11% were assessed by an external CAPS-A trained listener (Cleft Service range: 0%-100%). |
| | • 1% had some but not all 16 CAPS-A speech parameters reported. |
| | • 41% had a reason the speech outcomes were not collected. |
| | • 4% were missing data and a reason for not collecting data. |
| | • The proportion of children with complete speech data varied significantly according to birth year (2015: 28%, 2016: 64%, 2017: 68%, p<0.001) but not by cleft type (CP: 53%, UCLP: 57%, BCLP: 49%, p=0.055) or by sex (female: 44%, male: 48%, p=0.150) |
| Outliers | Positive:  1. South Wales (80%), 2. Trent (74%), 3. Scotland (73%), 4. Newcastle* (66%) |
| | Negative: 1. South West (28%), 2. West Midlands (37%) |
| | *Positive alert status for two consecutive reporting periods and therefore classed as outlier. |

**Figure 5.11.** Funnel plot showing the percentage of CRANE-consented 5-year-olds, born 2015 to 2017, with speech outcomes reported, according to Cleft Service.
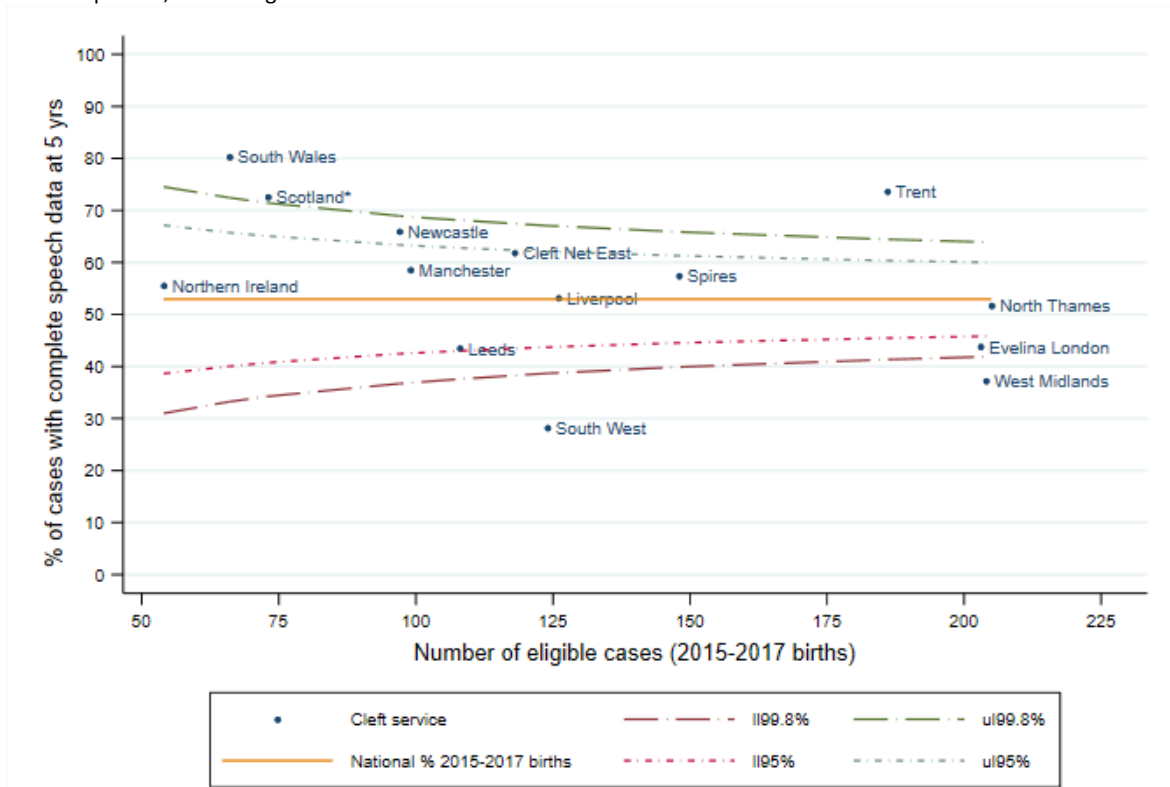


Note: Funnel plot centred on the revised national percentage (52.9%) of children (born 2015-2017) with speech outcomes reported.

| Outcome: National Speech Standard 1 | |
|---|---|
| Indicator | #20 – The achievement of speech with no evidence of a structurally related problem and no cleft speech characteristics requiring intervention: This standard is achieved when patients have green ratings across all 16 CAPS-A speech parameters. |
| Benchmarks | 60% of children born 2014-2016 met speech outcome standard 1 (CRANE, 2023). |
| Denominator | 973 CRANE-consented children with all 16 CAPS-A scores. |
| What did we find? | • 57% of children met speech outcome standard 1 (Cleft Service range: 37%[50]-72%, p<0.001). This is 3% lower than in the previous reporting period (births 2014-2016).<br>• 15% of children within this cohort had secondary surgery for speech purposes before the age of 5 years. This is lower than the 17% of children born 2014-2016.<br>• The percentage of children meeting standard 1 varied significantly according to cleft type (CP: 70%, UCLP: 48%, BCLP: 31%, p<0.001) and sex (female: 61%, male: 54%, p=0.047), but not by birth year (2015: 65%, 2016: 58%, 2017: 54%, p=0.052). |
| Outliers | Positive: 1. Trent (72%)<br><br>Negative: 1. North Thames* (44%), 2. Cleft Net East* (45%)<br><br>* Negative alert status for two consecutive reporting periods and therefore classed as outlier. |

**Figure 5.12.** Funnel plot showing the percentage of CRANE-consented 5-year-olds, born 2015 to 2017, meeting speech outcome standard 1, according to Cleft Service.



Note: Funnel plot centred on the revised national percentage (57.4%) of children (born 2015-2017) meeting speech outcome standard 1.

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[50] Note that South West (37%) was a negative outlier for data completeness so interpret this rate with caution. The lowest rate among services not identified as negative outliers for consent or data completeness was 44% (North Thames).

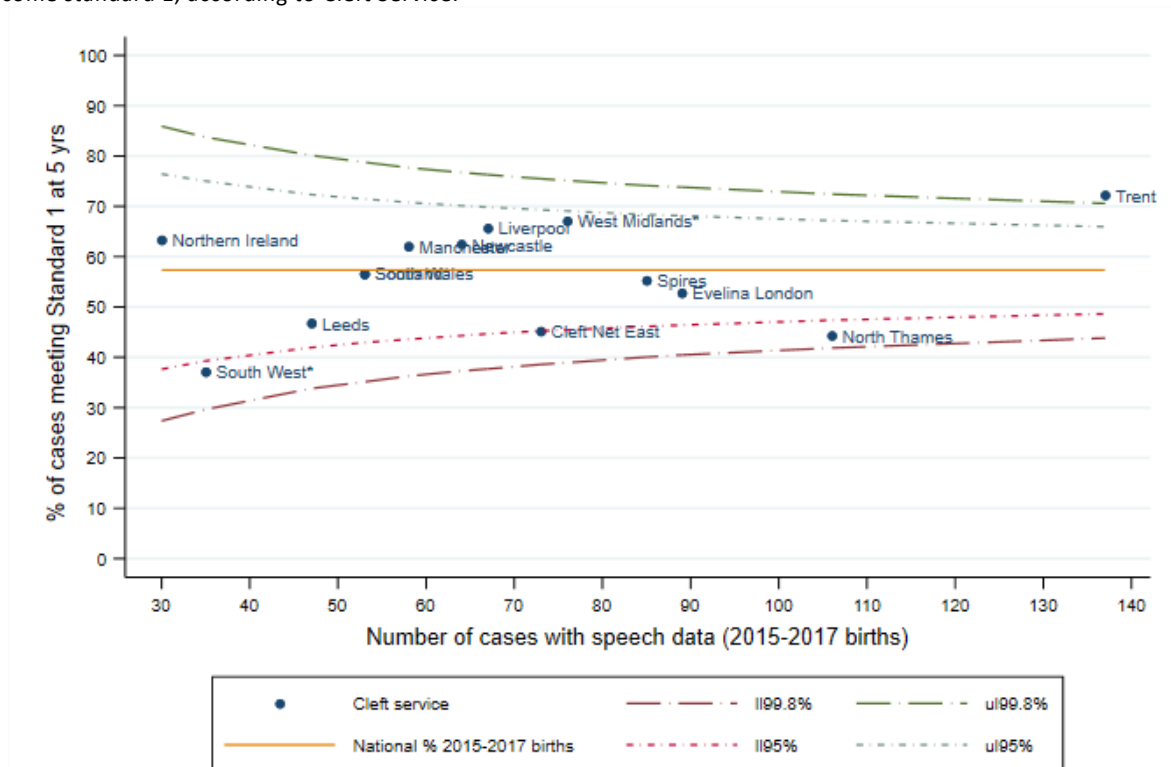| Outcome: National Speech Standard 2a | |
|---|---|
| Indicator | #21 – The achievement of speech without evidence of a structurally related speech difficulty. This standard is achieved when patients have no reported history of velopharyngeal surgery or fistula repair for speech purposes and have green 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. |
| Benchmarks | 72% of children born 2014-2016 met speech outcome standard 2a (CRANE, 2023). |
| Denominator | 973 CRANE-consented children with all 16 CAPS-A scores. |
| What did we find? | • 73% of children met speech outcome standard 2a (Cleft Service range: 55%-87%, p<0.001). This represents minimal change from the previous reporting period (2014-2016 births).<br>• The percentage of children meeting standard 2a did not vary significantly according to cleft type (CP: 74%, UCLP: 73%, BCLP: 66%, p=0.178), birth year (2015: 75%, 2016: 75%, 2017: 70%, p=0.251), or sex (female: 70%, male: 75%, p=0.068). |
| Outliers | Positive: 1. Trent (87%)<br>Negative: 1. Leeds* (55%)<br>* Negative alert status for two consecutive reporting periods and therefore classed as outlier. |

**Figure 5.13.** Funnel plot showing the percentage of CRANE-consented 5-year-olds, born 2015 to 2017, meeting speech outcome standard 2a, according to Cleft Service.



Note: Funnel plot centred on the revised national percentage (72.6%) of children (born 2015-2017) meeting speech outcome standard 2a.

49

| Outcome: National Speech Standard 3 | |
|---|---|
| Indicator | #22 – The achievement of speech without evidence of significant cleft-related speech characteristics (on sentence repetition), which may require therapy and/or surgery: This standard is achieved when patients have 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). |
| Benchmarks | 68% of children born 2014-2016 met speech outcome standard 3 (CRANE, 2023). |
| Denominator | 973 children with all 16 CAPS-A scores. |
| What did we find? | • 65% of children met speech outcome standard 3 (Cleft Service range: 40%[51]-78%, p=0.002). This is 3% lower than in the previous reporting period (2014-2016 births).<br>• The percentage of children meeting standard 3 varied significantly according to cleft type (CP: 77%, UCLP: 55%, BCLP: 39%, p<0.001) and sex (female: 69%, male: 61%, p=0.006), but not according to birth year (2015: 70%, 2016: 64%, 2017: 63%, p=0.253). |
| Outliers | Positive: 1. Trent (78%)<br>Negative: None |

**Figure 5.14.** Funnel plot showing the percentage of CRANE-consented 5-year-olds, born 2015 to 2017, meeting speech outcome standard 3, according to Cleft Service.
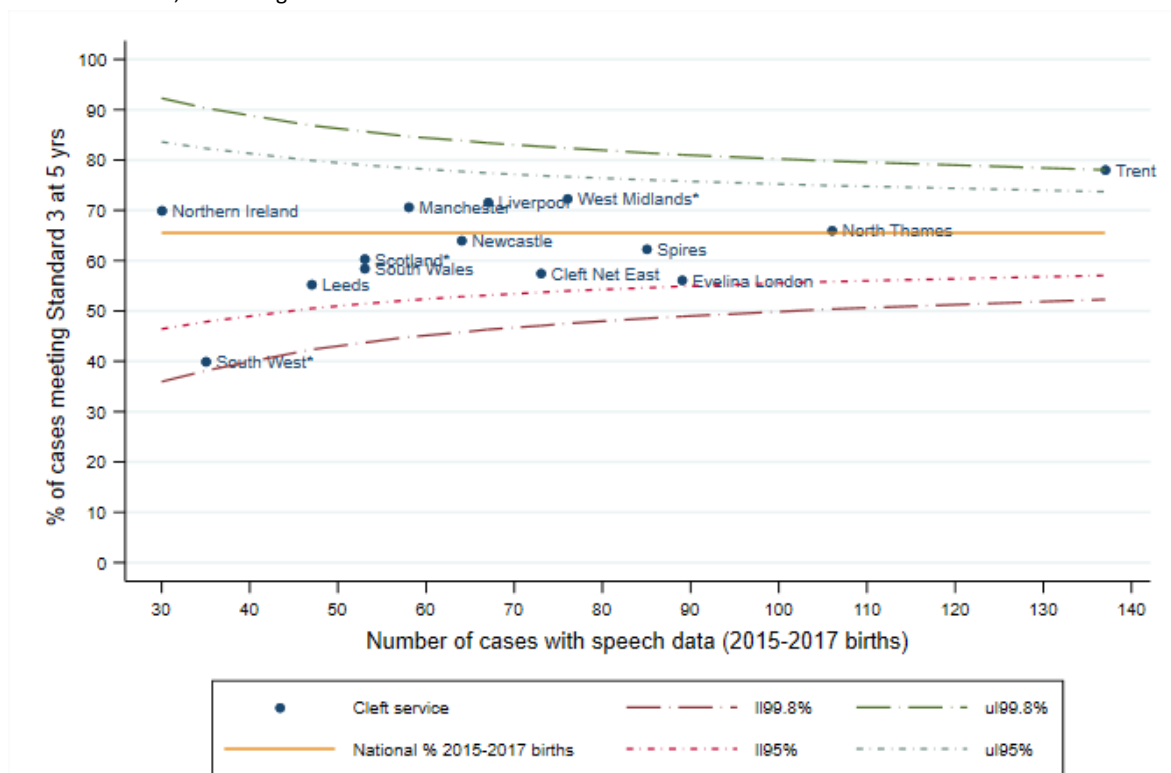


Note: Funnel plot centred on the revised national percentage (65.5%) of children (born 2015-2017) meeting speech outcome standard 3.

| Recommendations: Speech |
|---|
| • Children with a cleft affecting the palate should have speech assessed and reported to CRANE.<br>• Cleft Services should work together to explore reasons for variation in speech outcomes. |

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[51] Note that South West (40%) is a negative outlier for data completeness so interpret this rate with caution. The lowest rate among services not identified as negative outliers for consent or data completeness was 55% (Leeds).

| Statement written by Imogen Underwood, Chair of Lead Cleft Speech and Language Therapy (SLT) group, December 2024. |
| --- |
| 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's Annual Report contains speech data from the 2015 and 2016 birth cohorts impacted by the pandemic. This means data are not directly comparable across centres and the 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 and submission of all available audit data. |

| Statement of intention from CRANE |
| --- |
| In recent years, CRANE has investigated the impact of patient-related factors on speech outcomes at 5 years of age in children with cleft palate+/-lip[52]. This development work found that sex, cleft type, and extent of hard palate involvement have a significant impact on speech outcome. <br><br> This year, we further developed this work and found that the presence of two or more additional congenital anomalies (in the absence of a known syndrome) significantly reduced the odds of achieving all three cleft speech standards[53]. Whilst CRANE collects information on syndromes and some specific additional anomalies (affecting the circulatory system and nervous system), complete additional anomaly information is dependent on up-to-date linkage to Hospital Episode Statistics (HES), Patient Episode Database for Wales (PEDW), and Scottish Morbidity Records (SMR), which is currently not possible due to funding restrictions. Similar data for Northern Ireland are not available for linkage. Ideally, complete additional anomaly information would be included in speech risk-adjustment models, but in the absence of data linkage, CRANE intends to incorporate patient characteristics that are recorded in the CRANE Database (sex, cleft type, extent of hard palate involvement, Robin Sequence and anomalies of the circulatory system) into risk-adjustment models for service-level speech outcome reporting from 2025 onwards. |

[52] Butterworth S, Fitzsimons KJ, Medina J, et al. Investigating the Impact of Patient-Related Factors on Speech Outcomes at 5 Years of Age in Children With a Cleft Palate. The Cleft Palate Craniofacial Journal. 2023;60(12):1578-1590. doi:10.1177/10556656221110094

[53] Butterworth S, Fitzsimons KJ, Britton L, et al. Investigating the Impact of Additional Congenital Malformations on Speech Outcomes at age Five in Children with a Cleft Palate. The Cleft Palate Craniofacial Journal. 2024;0(0). doi:10.1177/10556656241287759

## 5.5. Psychology

All children with a cleft should be seen by a psychologist before their 6th birthday. The **Tiers of Involvement Measure (TIM)** records the tier (level) of involvement by a psychologist from the Cleft Multi-Disciplinary Team (MDT). Tiers range from 0 to 6 (see box 'TIM Scores').

> **TIM Scores**
> 0. Patient not seen by Psychologist
> 1. Psychological input not needed
> 2. Psychological input provided during the clinic
> 3. Psychological input provided during the clinic (tiers 3-6)

The **Strengths and Difficulties Questionnaire (SDQ)** is a brief behavioural screening questionnaire designed for use with 3-16-year-olds. These questionnaires should be completed by the child's parents/guardians and the scores submitted for all CRANE-consented 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[54]. Total scores are then categorised into four groups (See box ''Total difficulties' SDQ score groups'). Low scores, indicating no concern, are classified as being in the 'close to average' range. Scores in the 'high' and 'very high' range indicate a greater level of difficulties, which may require psychological input or intervention.

> **'Total difficulties' SDQ score groups**
> 1. Close to average
> 2. Slightly raised,
> 3. High
> 4. Very high.

| Cohort summary | |
|---|---|
| Data source | The CRANE Database (consented cases only). Extract taken: 1 July 2024 |
| Birth years | Three years: 2015 to 2017 |
| Countries | England, Wales, Northern Ireland and Scotland |
| Inclusions | CRANE-consented children, including those without a specified cleft type |
| Exclusions | • Children who died before the age of 5 years<br>• Children with submucous cleft palate |
| Notes | • Subject to outlier policy.<br>• Data are not risk-adjusted.<br>• Funnel plots are centred on the revised national rates after excluding data from services identified as negative outliers for consent verification (Scotland). |
| Raw data | 'Psychology 2015-17' in **the supplementary tables** |

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[54] 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

## 5.5.1. Tiers of Involvement Measure

| Data completeness: Tiers of Involvement score | |
|---|---|
| Indicators | #23 – TIM scores recorded for all eligible children |
| Denominator | 2,872 CRANE-consented children |
| What did we find? | • 57% of eligible children had recorded TIM scores (Cleft Service range: 13%-92%, p<0.001). This compares to 53% for the previous reporting period (births 2014-2016).<br>• 29% had a reason[55] TIM scores were not collected.<br>• 14% were missing data and a reason for not collecting data.<br>• The percentage of children with TIM scores varied significantly according to birth year (2015: 44%, 2016: 56%, 2017: 71%, p<0.001) and cleft type (CL: 57%, CP: 55%, UCLP: 62%, and BCLP: 62%, p=0.014), but not according to sex (female: 43%, male: 43%, p=0.649). |
| Outliers | Positive:  1. South Wales (92%), 2. Scotland (87%), 3. Newcastle (80%), 4. Liverpool (76%),<br>    5. Cleft Net East (74%), 6. Northern Ireland (73%), 7. North Thames (70%),<br>    8. Spires (70%), 9. Manchester (68%)<br>Negative: 1. West Midlands (13%), 2. Trent (20%), 3. South West (39%) |

**Figure 5.15.** Funnel plot showing the percentage of CRANE-consented 5-year-olds, born 2015 to 2017, who had Tiers of Involvement Measure (TIM) scores reported, according to Cleft Service.
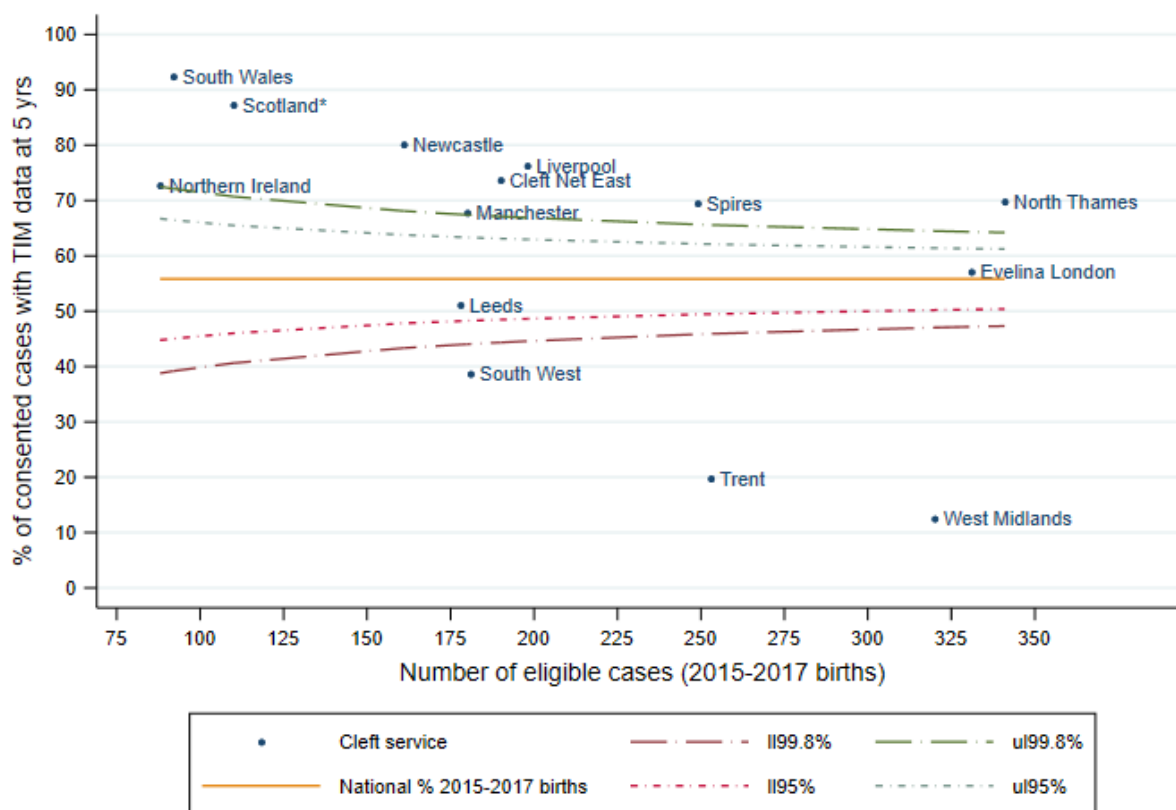


Note: Funnel plot centred on the revised national percentage (55.8%) of children (born 2015-2017) with TIM scores reported.

---

[55] Additional reasons specific to psychology data collection: Screen only partially completed; Not completed due to language barriers; Parents declined to complete; Not appointed before 6 years.

| Outcome: TIM scores of 1a+ (TIM tiers 1 to 6) | |
|---|---|
| Indicator | #24 – All eligible children seen by a psychologist before the age of 6 years. |
| Benchmarks | • 100% of children should be seen by a psychologist and have a TIM assessment<br>• 93% of eligible children born 2014-2016 were reported to have TIM scores of 1a+ (CRANE, 2023). |
| Notes | Funnel plot is centred on the revised national rates after excluding data from services identified as negative outliers for consent verification (Scotland) and TIM data completeness (Trent, West Midlands and the South West). |
| Denominator | 1,638 CRANE-consented children with TIM scores |
| What did we find? | • 92% of those with a TIM score were seen by a psychologist before the age of 6 years and a psychosocial screen was completed or psychological input arranged (TIM tiers 1 to 6, also referred to as TIM tier 1a+) (Cleft Service range: 70%[56]-100%, p<0.001).<br>    o 30% were assessed as not needing psychological input (TIM score 1).<br>    o 52% received psychological input during the MDT clinic (TIM score 2).<br>    o 10% required further psychological action (TIM scores 3-6).<br>• 8% were not seen by a psychologist (TIM score 0).<br>• The percentage of children with TIM scores of 1a+ did not vary significantly according to cleft type (CL: 90%, CP: 93%, UCLP: 94%, BCLP: 93%, p=0.159), birth year (2015: 90%, 2016: 94%, 2017: 92%, p=0.065), or sex (female: 92%, male: 93%, p=0.495). |
| Outliers | Positive:  1. Scotland (100%), 2. Newcastle (100%), 3. Cleft Net East (100%), 4. Leeds (99%)*,<br>             5. Spires (98%), 6. Manchester (98%)*<br>Negative: 1. West Midlands (70%), 2. Liverpool (74%), 3. North Thames (83%)<br>*Positive alert status for two consecutive reporting periods and therefore classed as outlier. |

**Figure 5.16.** Funnel plot showing the percentage of CRANE-consented 5-year-olds, born 2015 to 2017, with TIM scores of 1a+, according to Cleft Service.



Note: Funnel plot centred on the revised national percentage (92.0%) of children (born 2015-2017) with TIM 1a+ scores.

---

[56] Note that West Midlands (70%) was a negative outlier for data completeness so interpret this rate with caution. The lowest rate among services not identified as negative outliers for consent or data completion was 74% (Liverpool).

| Recommendations: Tiers of Involvement |
| --- |

- TIM scores should be recorded for all CRANE-consented children.
- Cleft Services should aim to see all children and families before the age of 6 years and ensure that psychological support is provided if appropriate.

### 5.5.2. Strengths and Difficulties Questionnaire

| Data completeness: Strengths and Difficulties Questionnaire (SDQ) score | |
| --- | --- |
| Indicators | #25 – SDQ scores recorded for all eligible children |
| Denominator | 2,872 CRANE-consented children |
| What did we find? | • 48% of eligible children had recorded SDQ scores (Cleft Service range: 6%-85%, p<0.001). This compares to 44% for the previous reporting period (births 2014-2016).<br>• 37% had a reason[57] SDQ scores were not collected.<br>• 16% were missing data or a reason for not collecting data.<br>• The proportion of children with SDQ scores varied significantly according to birth year (2015: 32%, 2016: 48%, 2017: 63%, p<0.001) but not according to cleft type (CL: 47%, CP: 47%, UCLP: 52%, BCLP: 49%, p=0.144) or sex (female: 52%, male: 53%, p=0.547). |
| Outliers | Positive: 1. South Wales (85%), 2. Newcastle (76%), 3. Cleft Net East (72%), 4. Scotland (70%), 5. Northern Ireland (69%), 6. Spires (64%), 7. Liverpool (60%), 8. North Thames (57%)<br><br>Negative: 1. West Midlands (6%), 2. Trent (17%), 3. South West (27%), 4. Evelina London (41%)*<br><br>* Negative alert status for two consecutive reporting periods and therefore classed as outlier. |

**Figure 5.17.** Funnel plot showing the percentage of CRANE-consented 5-year-olds, born 2015 to 2017, who had Strengths and Difficulties Questionnaire (SDQ) scores reported, according to Cleft Service.
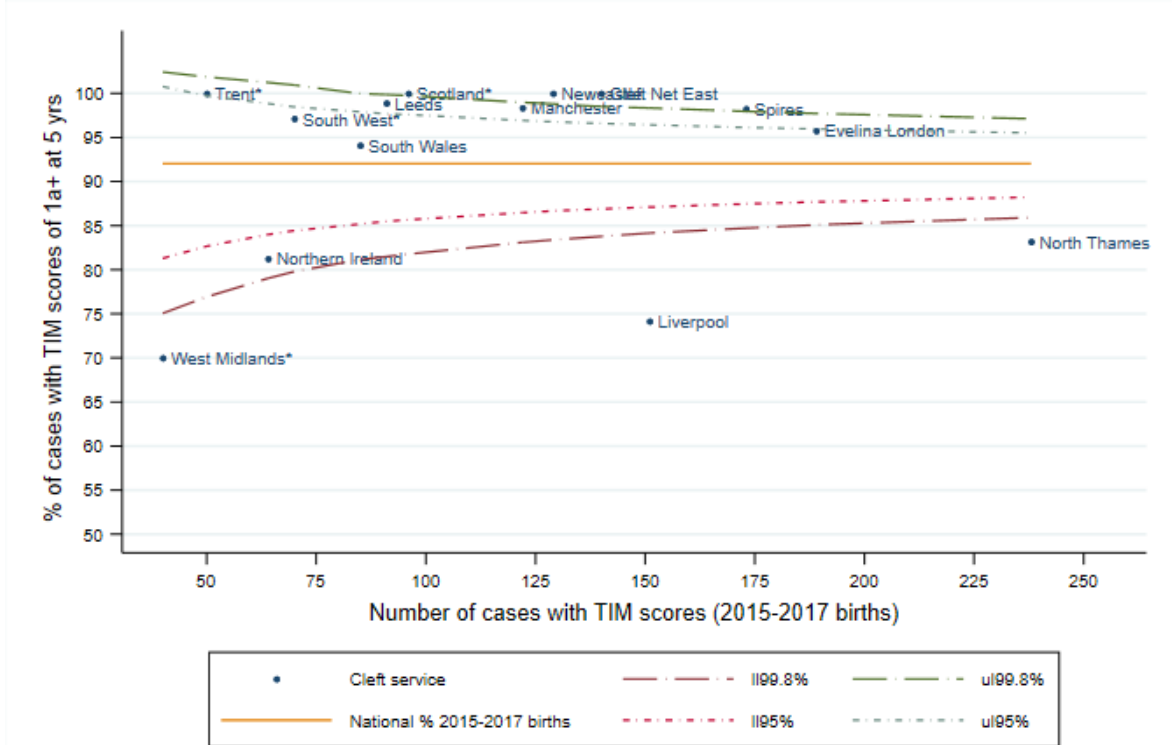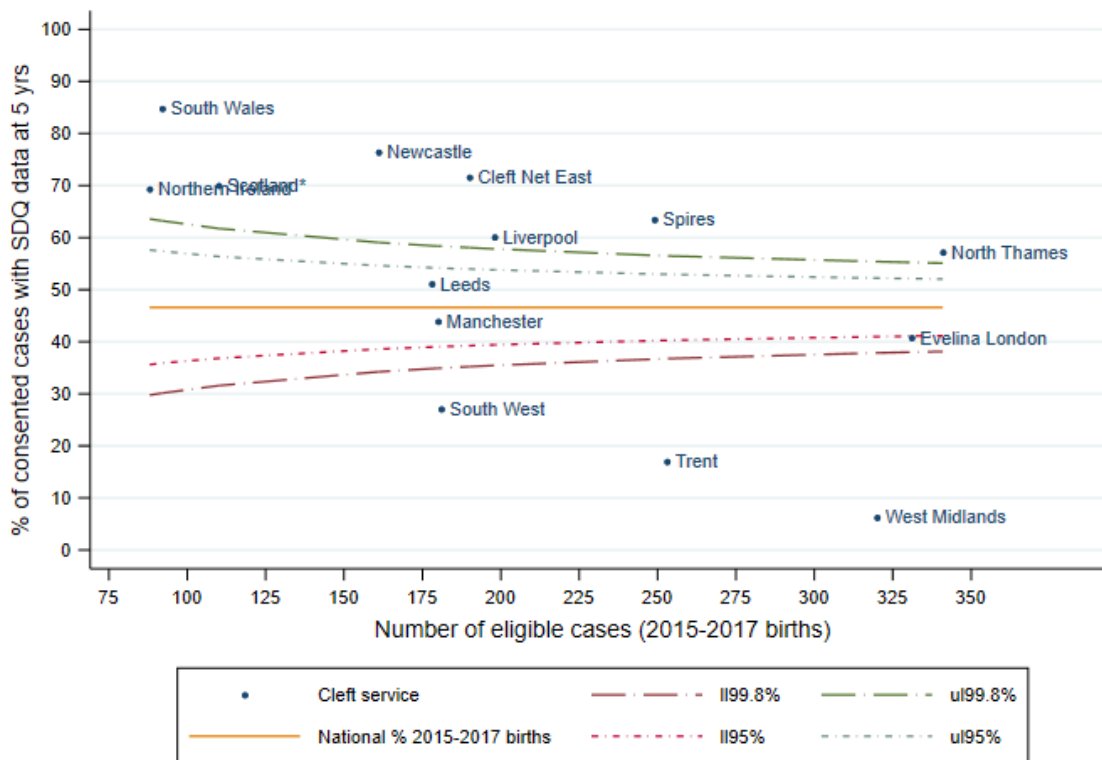


Note: Funnel plot centred on the revised national percentage (46.6%) of children (born 2015-2017) with SDQ scores reported.

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[57] Additional reasons specific to psychology data collection: Screen only partially completed; Not completed due to language barriers; Parents declined to complete; Not appointed before 6 years.

| Outcome: High/Very High SDQ scores | |
|---|---|
| Indicator | #26 – Psychological concerns identified at the age of 5 years. |
| Benchmarks | • SDQ population norms: 10% of children aged 5 to 10 years old have SDQ scores that are 'high' or 'very high'[58]. <br> • 16% of eligible children born 2014-2016 were reported to have 'high' or 'very high' SDQ scores (CRANE, 2023). |
| Notes | Funnel plots are centred on the revised national rates after excluding data from services identified as negative outliers for consent verification (Scotland) and TIM data completeness (Trent, West Midlands, South West, and Evelina London). |
| Denominator | 1,364 CRANE-consented children with SDQ scores |
| What did we find? <br>  | • 18% of children with a documented SDQ score had 'high' (7%) or 'very high' (11%) scores (Cleft Service range: 10%-31%[59], p=0.089). This rate has increased by 2% compared to the previous reporting period (births 2014-2016). <br> • 82% had other SDQ scores. The majority had 'close to average' (71%) or 'slightly raised' (11%) SDQ scores. <br> • The percentage of children with 'high' or 'very high' SDQ scores varied significantly according to sex (female: 14%, male: 22%, p=0.001), but not according to cleft type (CL: 15%, CP: 18%, UCLP: 21%, BCLP: 22%, p=0.179) or birth year (2015: 19%, 2016: 16%, 2017: 20%, p=0.162). |
| Outliers | None |

**Figure 5.18.** Funnel plot showing the percentage of CRANE-consented 5-year-olds, born 2015 to 2017, with high/very high SDQ scores, according to Cleft Service.



Note: Funnel plot centred on the revised national percentage (17.5%) of children (born 2015-2017) with high/very high SDQ scores.

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[58] 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. 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.
[59] Note that South West (31%) was a negative outlier for data completeness so interpret this rate with caution. The highest rate among services not identified as negative outliers for consent or data completion was 23% (Northern Ireland).

| Statement of ambition from the Psychology Clinical Excellence Network (CEN) |
| --- |
| The Psychology CEN is comprised of a specialist group of Clinical Psychologists who work within Cleft Services in the UK. As a group, a decision was made in 2023 to retire the SDQ (see statement in 2023 CRANE Annual Report) and to replace it with a measure that is cleft-specific, valid and reliable. As of 1 January 2025, the CEN have agreed to utilise the Psychological and Social sub-sections of the CLEFT-Q for patients aged 10. The CLEFT-Q is a rigorously developed patient-reported outcome measure (PROM) that can be used internationally to collect and compare evidence-based outcomes from patients aged 8 to 29 years with cleft lip and/or palate[60]. The CLEFT-Q has 12 independently functioning scales that measure three overarching domains: Appearance, Facial Function and Health-related Quality of Life (H-RQoL). The Psychological Functioning scale and the Social Functioning scale within the H-RQoL domain will be administered either face-to face or via video with children who are 10 years of age. For more information about the introduction of the CLEFT-Q across UK Cleft Services, see the Appendix. |
| | The CLEFT-Q data will be submitted to the CRANE Database in due course, once the Database has been updated for this purpose. Cleft Services will continue to collect the Tiers of Involvement Measure (TIM) data for 5-year-old patients for the time being, as a process measure. |
| Timeline for CRANE Database data collection |
| In line with the above statement: The CRANE Database is not anticipating any additional SDQ data collection and will be removing the ability to submit SDQ data by early 2025. Early psychological screening (at age 1) and Tiers of Involvement (TIM, at 5-years-of-age) will continue to be collected by the CRANE Database and reported on as a process indicator. Furthermore, the CRANE Database will be adapted to allow entry of data collected using the specified two sub-sections of the CLEFT-Q data for those born from 1 January 2015. The timeline for this will be confirmed in 2025. |

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[60] CLEFT-Q | Q-Portfolio - MEASURING WHAT MATTERS TO PATIENTS: https://qportfolio.org/cleft-q/

# 6. Database development work

This chapter presents development work undertaken over the last year and includes peer-review publications, conference presentations, a focus on deprivation among CRANE-consented children, and the hearing status of CRANE-consented children, born 2006 to 2021 and linked with the Newborn Hearing Screening Programme.

Publications in 2024:

1. Butterworth S & Fitzsimons KJ (Joint First Author), Britton L, et al. (2024) Investigating the Impact of Additional Congenital Malformations on Speech Outcomes at age Five in Children with a Cleft Palate. *The Cleft Palate Craniofacial Journal*. doi:10.1177/10556656241287759

2. Fell M & Fitzsimons KJ (Joint First Author), Hamilton MJ, et al. (2024) Cleft lip Sidedness and the Association with Additional Congenital Malformations. *The Cleft Palate Craniofacial Journal*. doi:10.1177/10556656241261918

3. Fell M, Bradley D, Chadha A, et al. Sidedness in Unilateral Orofacial Clefts: A Systematic Scoping Review. The Cleft Palate Craniofacial Journal. 2023;0(0). doi:10.1177/10556656231221027

Conference contributions in 2024:

1. Cleft Registry and Audit NEtwork linkage with the Newborn Hearing Screening Programme (European Cleft Congress, June 2024)

2. The impact of additional congenital malformations on speech outcome at age 5 in children born with cleft palate +/- lip. (European Cleft Congress, June 2024)

3. Cleft case ascertainment in the CRANE Database compared with Hospital Episode Statistics data in England (CFSGBI, April 2024)

4. Post-centralisation hospital care for children with clefts in England: An analysis of Hospital Episode Statistics (CFSGBI, April 2024)

5. Cleft Registry and Audit NEtwork linkage with the Newborn Hearing Screening Programme (CFSGBI, April 2024)

6. Are left sided clefts a different entity to right sided clefts? Progress from an international research collaboration (CFSGBI, April 2024)

## 6.1. Distribution of CRANE-consented patients born between 2014 and 2023 across multiple deprivation (MD) quintiles

| Cohort summary | |
|---|---|
| Data sources | 1. CRANE Database<br>2. The English Indices of Deprivation 2019[61]<br>3. Welsh Index of Multiple Deprivation (WIMD) 2019[62]<br>4. Northern Ireland Multiple Deprivation Measures 2017[63]<br>5. Scottish Index of Multiple Deprivation 2020[64]<br>6. Office for National Statistics (ONS)[65]<br>7. Public Health Scotland[66] |
| Birth years | Ten years: 2014 to 2023 |
| Countries | England, Wales, Northern Ireland and Scotland |
| Inclusions | CRANE-consented children with a valid postcode of residence linked to a multiple deprivation (MD) score. |
| Notes | • The multiple deprivation (MD) measure used within each of the four nations varies and prevents scores from being directly comparable. Additionally, data are collected at different times for each index.<br>• Scotland joined CRANE in 2023. 96% of their included cases were born 2022-2023. |
| Denominator | 7,390 children with MD scores |
| What did we find? | • 88% of eligible children had an MD score.<br>• There was an inverse relationship between deprivation quintile and cleft cases. The most deprived quintile was over-represented (27%) by cleft cases and the least deprived quintile was underrepresented (16%).<br>• The distribution of cleft cases across country-specific deprivation quintiles varied between UK nations, but there were no significant differences compared to the distribution of live births in the general population within each nation[67].<br>• The percentage of cleft patients living in the most deprived quintile areas varied between Cleft Services (11% to 47%). A clear North-South divide was evident, with Cleft Services in the North of England having higher rates of patients from the most deprived quintile compared to those in the South.<br>• The distribution of cleft cases across deprivation quintiles did not vary significantly between cleft types. |
| Recommendations | • CRANE should assign an MD score to each registered patient with postcode available.<br>• The relationship between IMD and cleft-related outcomes must be examined to determine whether MD should be incorporated into risk-adjusted models when comparing outcomes between Cleft Services. |
| Raw data | 'Multiple deprivation' in **the supplementary tables** |

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[61] The English Indices of Deprivation 2019. URL: https://www.gov.uk/government/collections/english-indices-of-deprivation; Index of Multiple Deprivation (December 2019) Lookup in EN. URL: https://geoportal.statistics.gov.uk/maps/ad50773cd40e4907a450c5d8954a9d26

[62] Welsh Index of Multiple Deprivation (full Index update with ranks): 2019 | GOV.WALES; Index of Multiple Deprivation (December 2019) Lookup in WA. URL: https://geoportal.statistics.gov.uk/datasets/4386e6a924de4a9b9a73be94792916ce

[63] Northern Ireland Multiple Deprivation Measures 2017 - Technical report. URL: https://www.nisra.gov.uk/publications/nimdm17-results; (December 2017) Lookup in NI. URL: https://geoportal.statistics.gov.uk/datasets/f303360fcb79465eaf412d6b3e9ef12c

[64] Scottish Government. Scottish index of multiple deprivation 2020. URL: Scottish Index of Multiple Deprivation 2020 - gov.scot; Index of Multiple Deprivation (December 2020) Lookup in SC. URL: https://geoportal.statistics.gov.uk/datasets/b57e427cb9254a42b1598c851dfc909e

[65] Office for National Statistics: Annual summary statistics on live births and stillbirths in England and Wales, 2017: Live births, stillbirths and the stillbirth rate by Index of Multiple Deprivation (IMD) and country of usual residence (checked October 2024).

[66] Public Health Scotland: Births in Scotland: All live births by year, SIMD, and NHS Board of Treatment.

[67] The distribution of live births in the general population across IMD quintiles is different for Scotland and was not known for Northern Ireland.

## Introduction

Where cleft-related outcomes are compared between Cleft Services, the outcomes may not only be influenced by the care provided, but also by the demographics of the patient population. Determining the socioeconomic deprivation of patients registered on CRANE is necessary to explore whether: (1) children with a cleft are more likely to live in deprived areas than the general population, (2) differences in deprivation exist between Cleft Services' populations, and (3) differences in deprivation exist between different cleft types.

The Office for National Statistics (ONS) provides Index of Multiple Deprivation (IMD) as the official measure of relative socioeconomic deprivation for small administrative areas in England. The IMD measure comprises a weighted combination of seven parameters: income, employment, education, health, crime, barriers to housing and services, and the living environment. Wales, Scotland and Northern Ireland also provide their own deprivation measures, with a slight variation in their use of seven parameters. The variation in the definition of the deprivation measures between the four nations prevents the MD measure from being directly comparable.

Small administrative areas that provide indices of relative deprivation are called Lower layer Super Output Areas (LSOAs) in England and Wales, Data Zones (DZs) in Scotland and Super Output Areas (SOAs) in Northern Ireland. We refer to LSOAs and their equivalents (DZs and SOAs) as LSOAs. The UK has 42,619 LSOAs, of which England has 32,844, Wales has 1909, Scotland has 6976 and Northern Ireland has 890.

## Methods

### Data sources

The CRANE Database acts as a national registry of all live births affected by a cleft in the UK. It contains registrations of births from 2000 onwards, which are submitted by regional Cleft Services. CRANE data were extracted on 1 July 2024. The availability of patients' postcode in the CRANE Database is dependent on CRANE consent and is necessary to link their residence area to their deprivation measure.

MD datasets were obtained for all four UK nations. The datasets describe a one-to-one relationship between country-specific LSOAs and their respective MD measures. Each MD measure is divided into five equal groups (quintiles) as shown in the Supplementary tables. The first group represents the most deprived, while the fifth group represents the least deprived quintile.

### Included cases

CRANE-consented children, born 1 January 2014 to 31 December 2023[68], with a valid postcode of residence were included in this feasibility study. Children were grouped according to their country of residence, the Cleft Service providing their care, and according to cleft type: cleft lip (CL), cleft palate (CP), unilateral cleft lip and palate (UCLP), and bilateral cleft lip and palate (BCLP). Cleft type classifications were defined according the reported LAHSAL codes in the CRANE Database.

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[68] Scotland joined CRANE in 2023 and 96% of their included cases were born in 2022-2023.

For comparison, the MD quintile distribution was obtained for live births in England and Wales occurring in 2015 to 2022. Corresponding rates for live births in Scotland were also obtained for births occurring in the 2022/23 financial year to reflect the birth period of included cleft cases[68]. MD information was not available for babies born in the general population in Northern Ireland.

## Analyses

A two-step process was used to determine the relative deprivation level of the patients' postcodes. Firstly, postcodes were validated by determining their LSOA code through a publicly available dataset that has a one-to-one correlation between UK postcodes and their respective LSOA[69]. Secondly, LSOA codes were linked to their respective MD quintiles from each nation. Descriptive statistics were calculated as number (%) of cleft cases within each MD quintile. This was reported for each country of residence, Cleft Service and cleft type. Chi-Square analyses were used to determine whether the distribution of cleft cases across quintiles of deprivation differed significantly to live births in the general population within each country.

## Results

There were 8,381 CRANE-consented patients who were born between 2014 and 2023 with cleft lip and/or cleft palate in the United Kingdom. 867 (10.3%) children were missing postcode information, 95 (1.1%) had an invalid postcode and 24 (0.3%) children were from the Channel Islands, which are not connected to any of the four MD measures. A total of 7,390 (88.2%) children were successfully linked to an LSOA code, allowing an MD score to be assigned. Of those with an MD measure, 88% were from England, 6% from Wales, 4% from Northern Ireland and 2% from Scotland.

**Figure 6.1.** Distribution of CRANE-consented cases (born 2014-2022*) and general population (GP) births (born 2015-2021**) across Multiple Deprivation (MD) quintiles, according to country of residence and country-specific deprivation measures.
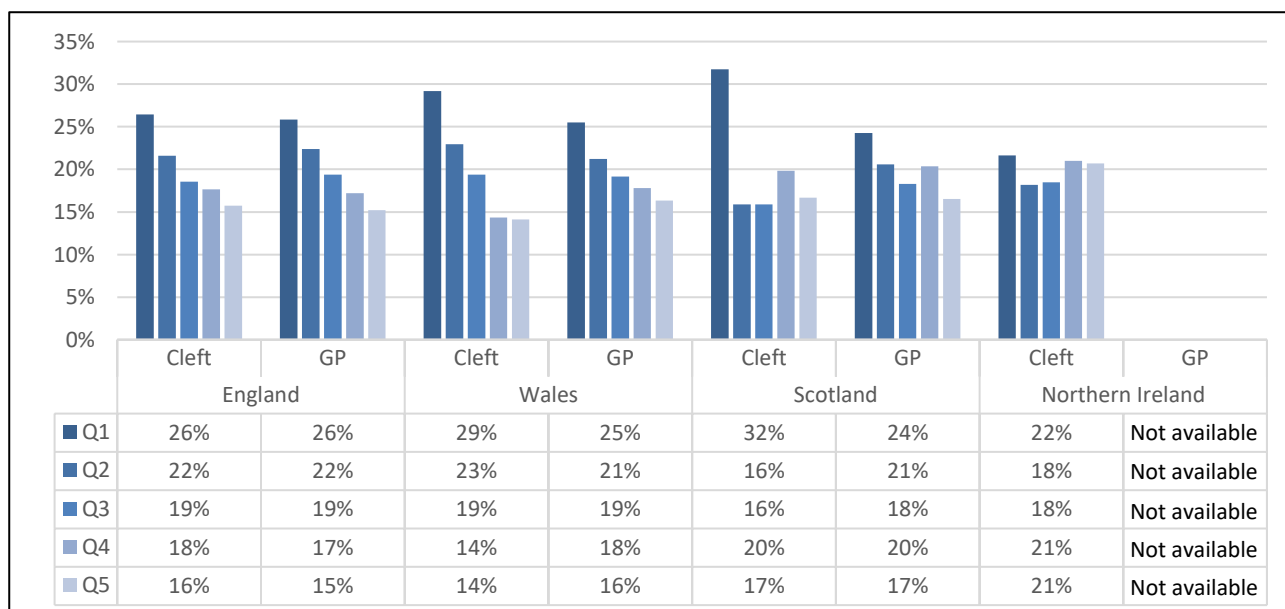


|  | England | | Wales | | Scotland | | Northern Ireland | |
|---|---|---|---|---|---|---|---|---|
|  | Cleft | GP | Cleft | GP | Cleft | GP | Cleft | GP |
| Q1 | 26% | 26% | 29% | 25% | 32% | 24% | 22% | Not available |
| Q2 | 22% | 22% | 23% | 21% | 16% | 21% | 18% | Not available |
| Q3 | 19% | 19% | 19% | 19% | 16% | 18% | 18% | Not available |
| Q4 | 18% | 17% | 14% | 18% | 20% | 20% | 21% | Not available |
| Q5 | 16% | 15% | 14% | 16% | 17% | 17% | 21% | Not available |

 * 121/126 children with MD scores in Scotland were born 2022-2023 as Scotland only officially joined CRANE in 2023. Interpret distribution with caution due to small numbers. **General population births in Scotland for 1 May 2022-31 April 2023. General population births across deprivation quintiles not available for Northern Ireland.

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[69] Postcode to OA (2011) to LSOA to MSOA to LAD (November 2022) Best Fit Lookup in the UK. URL: https://www.arcgis.com/sharing/rest/content/items/9c5ebee4163d435aa4defdaf348ba3c2/data

Figure 6.1 shows the distribution of cleft cases across deprivation quintiles according to country of residence. Corresponding rates for live births in the general population are provided for context and comparison. The distribution of cleft cases across deprivation quintiles did not vary significantly from the distribution of live births occurring in the general population in England (p=0.134), Wales (p=0.142) or Scotland (p=0.322). The skew to quintiles representing lower socio-economic deprivation reflects the fact that young families typically live in more deprived areas compared to people in later life stages. The distribution of live births in Northern Ireland across their deprivation quintiles is not available for comparison.

Figure 6.2 shows that the percentage of cleft patients living in the most deprived quintile areas varied between Cleft Services (range: 11% to 47%, p<0.001). A clear North-South divide was evident for services in England, with those in the North of England having higher rates of patients from the most deprived quintile compared to services in the South of England.

**Figure 6.2.** Funnel plot showing the percentage of CRANE-consented children, born 2014 to 2023*, living in areas within the most deprived quintile, according to Cleft Service.
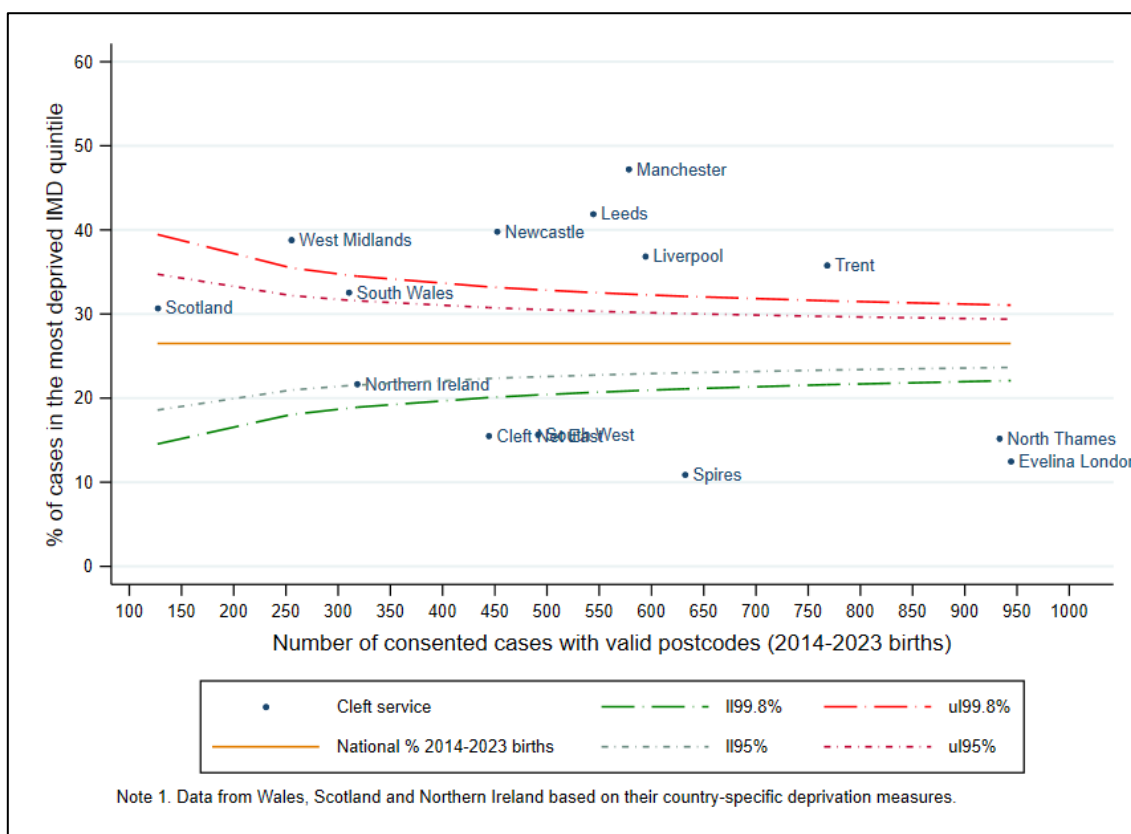


\* 121/126 children with MD scores in Scotland were born 2022-2023 as Scotland only officially joined CRANE in 2023.
Note that Wales, Scotland and Northern Ireland have their own individual country-specific IMD measures, which are not directly comparable with England. 18% of Liverpool's cases were resident in Wales and their IMD quintile is derived from the Welsh Index of Multiple Deprivation.

Figure 6.3 shows the distribution of CRANE-consented cases across the quintiles of deprivation for individual cleft types. The distribution across quintiles did not vary significantly between cleft types (p=0.120), and the most deprived quintile consistently had the highest percentage of children, while the least deprived had the lowest percentage of children.

**Figure 6.3.** Distribution of CRANE-consented children, born 2014 to 2023 in England, across Index of Multiple Deprivation (IMD) quintiles according to cleft type.



| | CL | CP | UCLP | BCLP |
|---|---|---|---|---|
| Q1 (most deprived) | 26% | 26% | 28% | 27% |
| Q2 | 21% | 22% | 20% | 24% |
| Q3 | 20% | 19% | 17% | 18% |
| Q4 | 16% | 17% | 19% | 17% |
| Q5 (least deprived) | 16% | 16% | 16% | 14% |
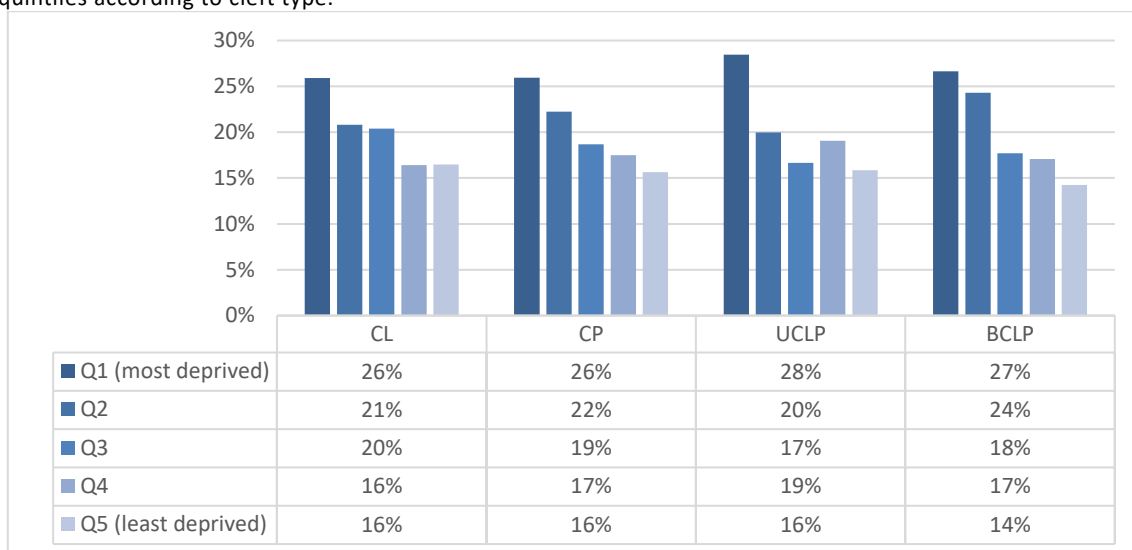
## Summary

An MD score was obtained for 88% of eligible CRANE-consented children born with a cleft in 2014-2023. Although 27% of the children analysed were assigned the most deprived quintile, the distribution of cleft cases across all deprivation quintiles did not differ significantly to the distribution of live births occurring in the general population. The results describe trends based solely on deprivation quintiles. Areas within the same deprivation level can have distinct health outcomes due to factors like healthcare access, community resources, and environmental influences. Future work should explore whether deprivation is associated with cleft-related outcomes and, where appropriate, deprivation scores should be included in risk-adjustment models.

## 6.2. Audiology assessment in children born with a cleft in England: Results from linkage with the Newborn Hearing Screening Programme (NHSP)

| Cohort summary | |
|---|---|
| Data sources | 1. CRANE Database <br><br> 2. Newborn Hearing Screening Programme (NHSP) Database |
| Birth years | 14.75 years: 01 March 2006 to 31 December 2021 |
| Countries | England |
| Inclusions | • CRANE-consented children with consent to data linkage <br> • Children with CL, CP, UCLP or BCLP (children with a submucous cleft palate excluded) |
| Raw data | 'Newborn Hearing Screening Prog' in **the supplementary tables** |

| Audiology assessment | |
|---|---|
| Benchmark | • 100% of children with cleft lip alone, who did not have a clear response on their newborn hearing screen should have an audiology assessment. <br> • 100% of children with cleft palate +/- lip should have an audiology assessment, regardless of the result of their newborn hearing screen. |
| Denominator | 11,574 English newborns registered in the CRANE Database had a CL/P and were linked to NHSP. <br> • 2,911 had CL alone <br> • 8,663 had CP+/-L (CP, UCLP, BCLP) |
| What did we find? | 7,116 (61%) children with CL/P had an audiology assessment recorded. <br> • 93% of children with cleft lip alone, who did not have a clear response on their newborn hearing screen had an audiology assessment. This is below the suggested benchmark of 100%. <br> • 72% of children with cleft palate +/- lip, regardless of the result of the newborn hearing screen had an audiology assessment. This is below the suggested benchmark of 100%. <br> • The proportion of children receiving an audiology assessment varied significantly according to cleft type (CL: 29%, CP: 69%, UCLP: 76%, BCLP: 79%, p<0.001) and syndromic status (non-syndromic: 59%, syndromic: 78%, p<0.001), but not sex (p=0.749). |

| Audiology outcome | |
|---|---|
| Denominator | 7,116 children with CL/P had an audiology assessment and outcome reported. |
| What did we find? | • 3,351 (47%) had satisfactory hearing in both ears. <br> • 2,721 (38%) had a hearing loss (HL) identified. Of these, 69% had conductive temporary HL, 1% had conductive permanent HL, 2% had sensorineural HL, 3% had mixed HL, 2% had different HL in each ear, and 23% had unilateral HL. <br> • 268 (4%) had a permanent HL identified. Of these 15% were conductive permanent, 25% were sensorineural, 27% were mixed, 19% were different HL in each ear, and 13% were unilateral loss only. <br> • 212 (3%) had a hearing aid offered or issued. <br> • 12% of all children with a clear response in both ears on their newborn hearing screen were diagnosed with conductive temporary HL at audiological assessment. This rate was 2% in those with CL and 17% in those with CP+/-L. <br> • 1,044 (15%) had hearing status that 'had not yet been determined'. |

| Recommendations | |
|---|---|
| | • Further investigations are needed to understand why children who should receive a diagnostic audiology assessment, according to NHSP protocols, do not have evidence of these taking place. <br> • Referral of high-risk children for audiological assessment is recommended, particularly those children with syndromes. |

## Introduction

Congenital hearing loss is one of the most common chronic conditions among children[70]. The Newborn Hearing Screening Programme (NHSP)[71], commissioned by the National Health Services for England (NHSE), is responsible for hearing loss detection among English newborns. It aims to identify babies who have permanent hearing loss as early as possible. Early detection of hearing loss and timely intervention among newborns can reduce delays in speech and language skills development. It provides long-standing beneficial effects on their social and emotional development, and quality of life from an early age[72]. It is recognised within the NHSP that certain risk factors can influence which pathway a child may need to follow from a screening, audiological diagnostic and ongoing surveillance perspective. Craniofacial anomalies, described by the NHSP as including a (noticeable) craniofacial anomaly such as cleft palate (excluding cleft lip only, minor pits or ear tags), are one of these risk factors. Children with these conditions must be reviewed for a targeted audiology assessment with audiology following the newborn hearing screen due to a higher potential risk of hearing loss. With CRANE-NHSP linked data, it is possible to quantify the proportion of children born with a cleft in England who experience hearing loss.

### Objectives

1. To determine the number of children with cleft lip/palate who received an audiology assessment.
2. To determine the number and percentage of children with cleft lip/palate who (a) had a permanent hearing loss identified at the time of their audiology assessment and (b) if this varied according to patient factors (sex, cleft type, syndromic status).
3. To determine the number and percentage of children with cleft lip/palate who (a) had a conductive temporary hearing loss identified at the time of their audiology assessment and (b) if this varied according to patient factors (sex, cleft type, syndromic status).

## Methods

### Data sources

The CRANE Database acts as a national registry of all live births affected by a cleft in the UK. It contains registrations of births from 2000 onwards, which are submitted by regional Cleft Services who usually receive referrals from maternity services upon the identification of a cleft lip and/or palate. CRANE registrations take place once the child has been born and the cleft has been confirmed.

The Newborn Hearing Screening Programme (NHSP)[73], commissioned by the National Health Services for England (NHSE), is responsible for hearing loss detection among all English newborns. The NHSP database contains information on the screening assessment, usually performed within the first few weeks after birth, as well as the audiological assessment and type of hearing loss detected, if present. The CRANE-NHSP linked dataset contains individual-level data for children born with a cleft in England, whose families consented to CRANE outcome data collection and/or linkage to health records.

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[70] Tamsin Holland Brown, Childhood hearing impairment, Paediatrics and Child Health, Volume 30, Issue 1, 2020, Pages 6-13, ISSN 1751-7222, https://doi.org/10.1016/j.paed.2019.10.002.https://doi.org/10.1016/j.paed.2019.10.002.

[71] Overview of Newborn Hearing Screening Programme: https://www.nhs.uk/conditions/baby/newborn-screening/hearing-test/ [Last accessed: 11/11/2024]

[72] Korver AM, Smith RJ, Van Camp G, Schleiss MR, Bitner-Glindzicz MA, Lustig LR, Usami SI, Boudewyns AN. Congenital hearing loss. Nat Rev Dis Primers. 2017 Jan 12;3:16094. doi: 10.1038/nrdp.2016.94. PMID: 28079113; PMCID: PMC5675031.

[73] Overview of Newborn Hearing Screening Programme: https://www.nhs.uk/conditions/baby/newborn-screening/hearing-test/ [Last accessed: 11/11/2024]

### Patients

CRANE-consented cases, born 1 March 2006 to 31 December 2021, were included in our analyses if they had linked NHSP records. Children were grouped according to cleft type: cleft lip (CL), cleft palate (CP), unilateral cleft lip and palate (UCLP), bilateral cleft lip and palate (BCLP), submucous cleft palate with cleft lip (SMCP+CL) and submucous cleft palate alone (SMCP). These classifications were defined according to the reported LAHSAL codes[74] in the CRANE Database. For these analyses, children with an SMCP were excluded, as their detection may occur after the hearing screen. Sex and syndromic status were also obtained from the CRANE database.

### Outcome

According to the NHSP protocol[75], children with cleft lip alone are referred for audiological diagnostic assessment if their hearing screen does not provide a clear response in one or both ears or if they are targeted for follow-up due to the presence of certain risk factors. All children with a cleft involving the palate should be targeted for an audiological assessment regardless of the result of the hearing screen. Cleft lip alone is not considered a risk factor.

Outcomes following audiology assessment were defined as:

(a)  Satisfactory hearing or clear screen response in both ears;

(b)  Conductive temporary hearing loss in both ears;

(c)  Conductive permanent hearing loss in both ears;

(d)  Sensorineural hearing loss in both ears;

(e)  Mixed hearing loss in both ears;

(f)  Different hearing loss type in each ear;

(g)  Hearing loss any type in one ear;

(h)  Not yet determined or missing.

### Analysis

Completion of an audiological assessment and audiological outcomes were analysed according to cleft type, sex and syndromic status using Chi-Square tests. For all statistical tests, p-values <0.05 were considered significant.

## Results

### Children eligible for audiological assessment

A total of 11,574 CRANE-consented children born between 01 March 2006 and 31 December 2021 and residing in England were linked to an NHSP record. Of these, 2,911 had a cleft lip alone, and 8,663 had a cleft involving the palate +/- lip (CP, UCLP, BCLP) and should be targeted for audiological assessment.

### Audiological assessment

Overall, 7,116 (61%) children received an audiological assessment. The proportion of children with audiology assessments varied significantly according to cleft type (p<0.001) and were most likely to be recorded for children with a BCLP (79%), followed by children with UCLP (76%), CP (69%), and then CL (29%).

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[74] CDC Orofacial Clefts - https://www.cdc.gov/ncbddd/birthdefects/surveillancemanual/chapters4. Accessed 11/11/24.

[75] NHS public health functions agreement 2019-20. Service specification No.20, NHS Newborn Hearing Screening Programme, July 2019

**Figure 6.4:** Number and percentage of children who received a diagnostic audiology assessment according to newborn hearing screening response and risk factor classification. Cleft Lip without cleft palate and Cleft Palate +/- Cleft Lip reported separately.
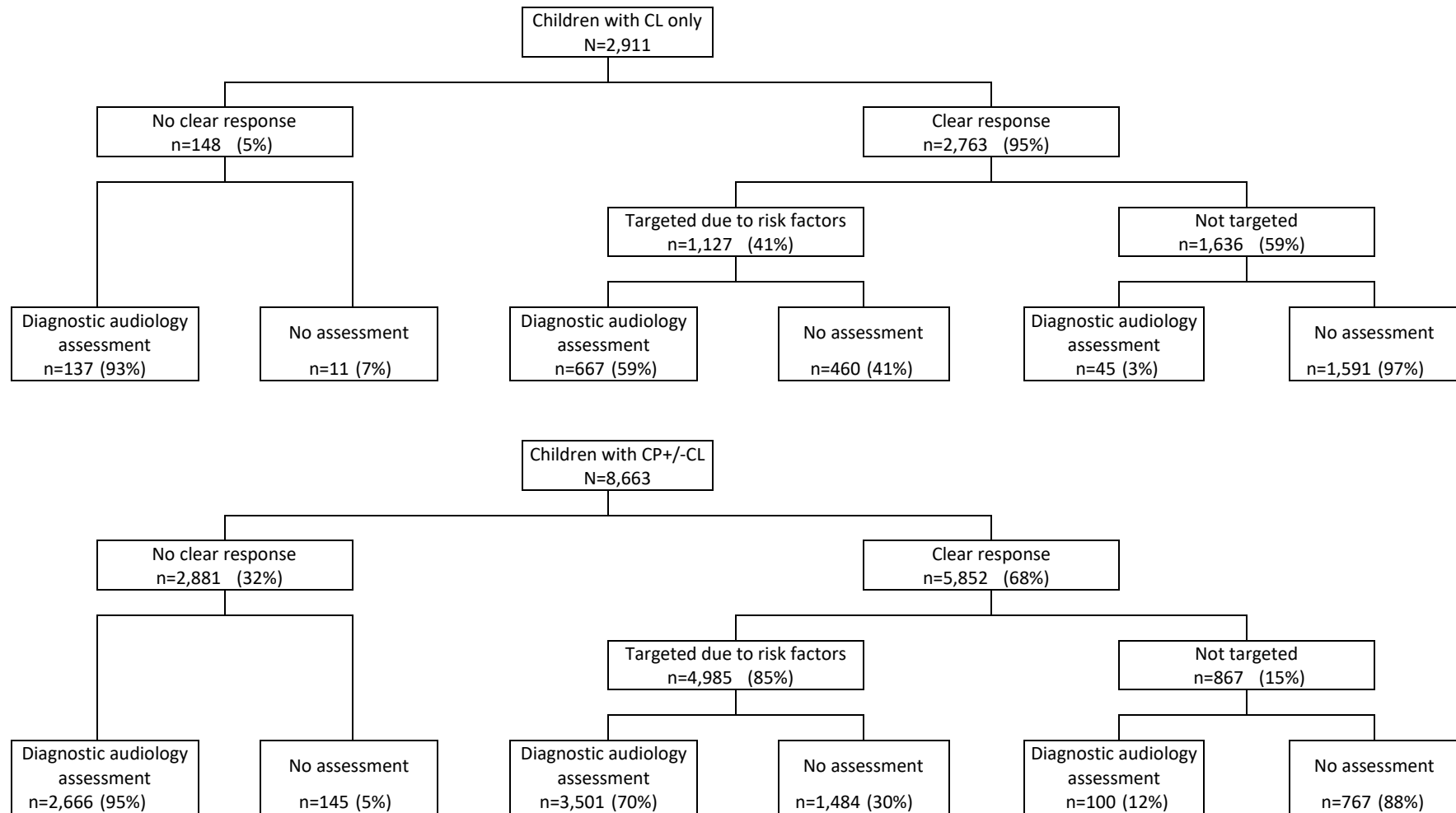
Figure 6.4 shows that among children with CL alone, 7% without a clear response on the newborn hearing screen had no evidence of diagnostic assessment. Of those with a clear response, 41% were classified as being targeted due to risk factors, but this may have been unnecessary, according to the NHSP protocol.

Among children with CP+/-CL, 72% received a diagnostic assessment, regardless of newborn hearing screening result, which is below the benchmark of 100%. Of those without a clear response on the newborn hearing screen, 5% had no evidence of diagnostic assessment. Of those with a clear response, 62% received an audiology assessment.

The proportion of children with a diagnostic assessment also varied significantly according to syndromic status (syndromic: 78%, non-syndromic: 59%, p<0.001) but not sex (p=0.749).

## Audiological Outcome

Of 7,116 children with a complete audiological assessment and outcome available, 3,351 (47%) had satisfactory hearing reported in both ears, 2,721 (38%) had a hearing loss identified and in 1,044 (15%) hearing status had not yet been determined.

**Figure 6.5:** Distribution of hearing loss type according to cleft type



| | CL | CP | UCLP | BCLP |
|---|---|---|---|---|
| Satisfactory hearing | 2,689 | 2,902 | 1,463 | 599 |
| Conductive temporary HL | 64 | 937 | 596 | 273 |
| Conductive permanent HL | 2 | 26 | 5 | 7 |
| Sensorineural HL | 7 | 43 | 11 | 6 |
| Mixed | 4 | 53 | 8 | 8 |
| Different HL each ear | 4 | 21 | 13 | 14 |
| Not yet determined | 102 | 626 | 322 | 150 |
| Unilateral HL | 39 | 295 | 203 | 82 |
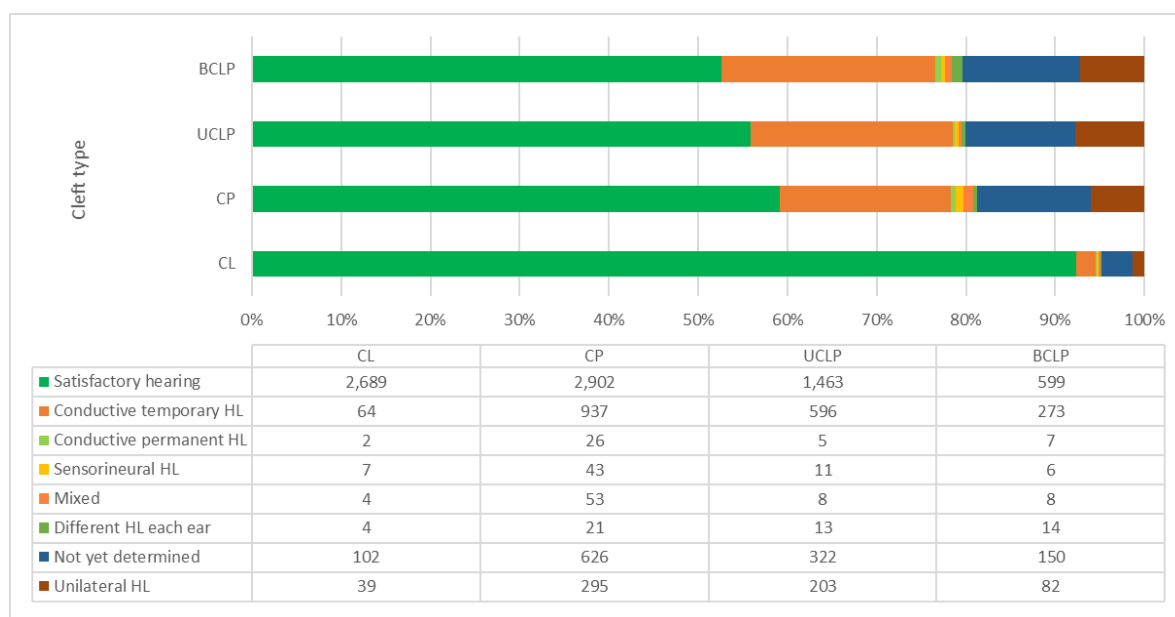
The proportion of children with satisfactory hearing varied significantly according to cleft type (CL 75%, CP 43%, UCLP 44%, BCLP 42%, p<0.001). Figure 6.5 shows the distribution of hearing status and includes those children who did not have an audiology assessment but did achieve a clear response in both ears on newborn screen. These children have been added into the 'satisfactory hearing' category and those not achieving a clear screen have been added to the 'not yet determined' category to give an overall picture of hearing status among children born with a cleft in England. It is possible that this method may underestimate hearing loss, particularly for conductive temporary hearing loss.

Of the 2,721 children with hearing loss identified;

- 69% had bilateral conductive temporary hearing loss (HL)
- 1% had bilateral conductive permanent HL
- 2% had bilateral sensorineural HL
- 3% had bilateral mixed HL
- 2% had different HL in each ear, and
- 23% had unilateral HL.

Permanent childhood hearing loss was identified among 268 (4%) children who had an audiology assessment. Of these, 15% had conductive permanent loss, 25% had sensorineural loss, 27% had mixed loss, 19% had different loss in each ear and 13% had unilateral permanent loss. There were 231 children on the permanent childhood hearing impairment (PCHI) register, representing 3.2% of those with a diagnostic assessment and 2.3% of the full study cohort.

212 children identified as having a permanent hearing loss had a hearing aid offered or issued. Presence of a permanent hearing loss varied significantly according to cleft type (range: 1% CL to 4% BCLP, $p < 0.001$) and syndromic status (11% syndromic vs. 1% non-syndromic, $p < 0.001$) but not according to sex ($p = 0.680$).

Bilateral conductive temporary hearing loss was identified among 1,870 (26%) children who had an audiology assessment. This varied significantly by cleft type (range: 8% in CL to 30% in UCLP and BCLP, $p < 0.001$) and sex (female: 31%, male: 34%, $p = 0.004$) but not according to syndromic status ($p = 0.257$).

1,035 (12%)) children who had a clear response on their newborn hearing screen went on to have a conductive temporary loss diagnosed at audiological assessment. This was 2% of children with CL and 17% of those with CP+/-L. If these children had not had a targeted audiological assessment this hearing loss type may not have been detected.

## Summary

This study describes hearing assessment among children born with a cleft in England. Over 90% of children with a cleft affecting only the lip were considered to have satisfactory hearing based on their audiology assessment or newborn hearing screening result. This was the case for just over half of those with a cleft involving the palate. Not all children with a cleft were referred appropriately for audiological assessment and further investigations are needed to understand why. Among those who were assessed, around 1 in 4 had conductive temporary hearing loss identified and 1 in 25 had permanent hearing loss identified. Given the importance of hearing in early speech development and communication abilities in early educational development, exploring the relationship between hearing loss, speech and educational outcomes among children with a cleft is strongly recommended.

We are grateful to CLEFT for funding this development work using the NHSP dataset.

# 7. Conclusion

The end of the year gives an opportunity for reflection on times past and also to look forward to the future with optimism and energy. Having prepared this report for you, it is difficult not to reflect on the progress made in the last five years. It seems no time at all since we all had to adapt to a global pandemic but this new year will be the fifth since those difficult days! Over the last five years the report has been systematised to provide a 'familiar' structure and look that hopefully all stakeholders are becoming comfortable with. It is not *exactly* the same and presentation has evolved and will continue to do so. But more of that later.

Looking back, everyone should be very proud of what has been achieved with CRANE data. While improvements in consent and data completeness may look modest to some, the small magnitude of improvement is due to the nature of our three-year rolling cohorts and the lag effect this may introduce. When you look at individual years there are significant improvements in the most recent years that see us as a community returning to and perhaps moving beyond pre-covid data quality. High levels of consent and data completeness are essential to having confidence in the conclusions that can be drawn from the data analysed. All this has been achieved in a time when there have been huge clinical, service and financial pressures and uncertainty locally and within the NHS as a whole. All involved - patients, parents /guardians, clinicians and CRANE team members - should be proud to be able to achieve this output during such time. With CRANE data collection/analysis and open publication, UK cleft care is at the forefront of cleft clinical audit on the international stage and long may this continue.

With that said, we should not rest on our laurels. Despite the improvements in data quality, there remains significant variation in consent levels, data completeness and outcomes between services, and we should continue to try to learn from beacons of good practice and not be afraid to acknowledge when we can do better. The cleft community in the UK is a hugely supportive family that acknowledges the limitations in the data we present including the lag between intervention and outcome that makes trusting the processes of change challenging. The Cleft Development Group has given full consent for the formal introduction of the outlier policy for next year's reporting cycle and that will include risk adjustment wherever possible. Introduction of risk adjustment for case mix allows for fairer comparison between services.

Assuming continuation of CRANE funding, aligned with appropriate resourcing and a longer-term contract, CRANE aspires to take steps towards real-time reporting. As we move into 2025. CRANE will engage in active discussion with all stakeholders to ensure this is done with the full understanding and support of the community.

Given our theme of reflection on the past and looking forward to the future, we would like to take this opportunity to publicly thank (on everyone's behalf) the contribution made to CRANE's development by a trusted colleague and friend. Jibby Medina (CRANE's former Research Fellow and current Programme Manager) has given 11 years of her professional life to the development, management and success of CRANE. Jibby has decide that now is a suitable time to move on in her career and we wish her every success with this. She should be proud of all she has contributed and the learning and experience that she can take to her future endeavours.

Wishing all of you the very best for the forth coming festive season and a happy and most productive new year.

Yours sincerely,

The CRANE Database team

# Glossary and abbreviations

| | |
|---|---|
| **Alveolus / alveolar** | The part of the jaw (gum) that supports the teeth and contains the tooth sockets. |
| **BCLP** | Bilateral cleft lip and palate |
| **CAPS-A** | Cleft Audit Protocol for Speech—Augmented |
| **Caries (dental)** | Dental caries are also known as tooth decay / dental decay or a cavity. |
| **CEN** | Clinical Excellence Network – previously referred to as Special Interest Group (SIG) |
| **CI** | Confidence interval |
| **CL** | Cleft lip only |
| **Cleft** | A failure of tissues to join during development. |
| **Cleft Development Group (CDG)** | NHS national group representing all stakeholders in cleft care that is responsible for the CRANE Database as well as oversight and guidance on all aspects of the delivery of reorganised cleft care. |
| **Cleft Services / regions** | These terms are used interchangeably throughout this report and refer to the hospital / multidisciplinary group that provides cleft surgery and care for children with a cleft; as well as submits data to the CRANE Database, sometimes as part of a wider cleft centre or network.<br>See **the supplementary tables** for further information on Regional Cleft Services. |
| **Cleft surgeon** | A surgeon undertaking cleft repair surgery in a region / Cleft Service. |
| **Clinical Standards Advisory Group (CSAG)** | A group established in 1991 to act as an independent source of expert advice on standards of clinical care for, and access to and availability of services to, NHS patients. |
| **Confidentiality Advisory Group (CAG)** | An independent statutory body established to promote, improve and monitor information governance in health and adult social care.<br>http://www.hra.nhs.uk/research-community/applying-for-approvals/confidentiality-advisory-group-cag/ |
| **CP** | Cleft palate only |
| **Craniofacial anomalies** | A diverse group of deformities in the growth of the head and/or face. |
| **Craniofacial Society of Great Britain and Ireland (CFSGBI)** | An inter-specialty group set up to study cleft lip and palate and other craniofacial anomalies. https://craniofacialsociety.co.uk/ |
| **CSCs** | Cleft Speech Characteristics |
| **Denominator (see also numerator)** | In mathematical terms, the bottom number in a fraction. Considering that a fraction represents a part of a whole, the denominator represents the total number of parts created from the whole, for example 100 in 70/100.<br><br>In the context of this report, we refer to the number of children in the cohort we are discussing that could meet certain criteria. For example, children with a Cleft Palate (CP) only. |
| **dmft** | Decayed, missing and filled teeth at 5 years of age |
| **ENT** | Ear, nose and throat |
| **Funnel plot** | A graph that identifies Cleft Services which are outliers, where the local situation might require closer inspection – either because an area is doing well or because there is some indication that it is performing poorly. In this report:<br>• Each point on the funnel plot represents a Cleft Service.<br>• Each funnel plot is for one outcome, with its values shown on the vertical/Y axis.<br>• The size of the Cleft Services' cohort is shown on the horizontal or X axis.<br>• The benchmark value or overall national percentage is shown as a horizontal line through the centre of the graph.<br><br>The graph shows two funnels that lie on either side of the benchmark and are called the control limits – similar to confidence intervals. |

- The inner lines show 2 standard deviations or 95% control limits. The outer lines represent 3 standard deviations or 99.8% control limits.
- The funnel shape is formed because the control limits get narrower as the population size increases.

The outer funnel is used to decide if an area is significantly different to the benchmark with 99.8% confidence. If a point lies within the funnel, then we conclude that it is not significantly different to the benchmark. If it falls outside the funnel then we can say the value is significantly 'better' or significantly 'worse' than the benchmark, depending on the direction of the indicator/outcome.

Funnel Plot Source: David Spiegelhalter, Medical Research Council Biostatistics Unit - http://www.erpho.org.uk/Download/Public/6990/1/INPHO%204%20Quantifying%20performance.pdf

| | |
|---|---|
| **General population** | In epidemiological terms, all individuals without reference to any specific characteristic.<br><br>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 birthweight in the general population of England & Wales, according to the Office for National Statistics (ONS) (as in the Registrations section of this report).<br><br>In some instances, the latest national figures are based on a random sample of children in the general population, which, again, may include children with a cleft or other health conditions. |
| **Hospital Episode Statistics (HES)** | A national database containing records on all admissions to NHS hospitals in England. |
| **LAHSAL** | A code used to classify cleft type. Each letter (LAHSAL) relates to one of the six parts of the mouth that can be affected by a cleft. |
| **MD** | Multiple Deprivation |
| **MDT** | Multi-disciplinary team. |
| **National Hearing Screening Programme (NHSP)** | The Newborn Hearing Screening Programme (NHSP)[76], commissioned by the National Health Services for England (NHSE), is responsible for hearing loss detection among all English newborns. The NHSP database contains information on the screening assessment, usually performed within the first few weeks after birth, as well as referral status for audiological assessment and type of hearing loss detected, if present. |
| **Numerator (see also denominator)** | In mathematical terms, the top number in a fraction. Considering that a fraction represents a part of a whole, the numerator represents how many parts of that whole are being considered, for example 70 in 70/100.<br><br>In the context of this report, we refer to the number of children meeting certain criteria. For example, receiving a certain type of care or meeting a standard. |
| **RS** | Robin Sequence is a congenital birth condition characterised by micrognathia, glossoptosis and failure to thrive with or without a cleft affecting the palate. |
| **SD** | Standard deviation |
| **SDQ** | Strengths and Difficulties Questionnaire |
| **SLT** | Speech and language therapy |
| **Submucous cleft palate (SMCP)** | The term submucous refers to the fact that the cleft is covered over by the lining (mucous membrane) of the roof of the mouth. This covering of mucosa makes the cleft difficult to see when looking in the mouth. |
| **TIM** | Tiers of Involvement Measure |
| **UCLP** | Unilateral cleft lip and palate |
| **WHO** | World Health Organization |

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[76] Overview of NHSP https://www.nhs.uk/conditions/baby/newborn-screening/hearing-test/ [Last accessed: 24/02/2023]

# Appendix. Introduction of the CLEFT-Q to CRANE

The rationale for changing what the Psychology Clinical Excellence Network (CEN) submits to CRANE was shared in the 2023 report so will not be repeated here.  A CRANE-CEN sub-group was set up in December 2023 comprising a senior Clinical Psychologist from each Cleft Centre across the UK.  From January 2024 the sub-group has met four times to decide what we collect and how we plan to collect it.

We have agreed to use the CLEFT-Q as our outcome measure. The CLEFT-Q was developed by Drs Anne Klassen and Karen Wong. The copyright is owned by McMaster University (Hamilton, Canada) and the Hospital for Sick Children (Toronto, Canada). The measure can be used for free for non-profit purposes, but users must sign a Licensing Agreement. For further information about this, please email the McMaster Liaison Office via milo@mcmaster.ca. This is the only validated and published cleft-specific measure of its kind to date.

To develop the measure, the authors used a concept driven approach: 138 concept elicitation interviews for children and young people with a cleft diagnosis aged eight to 29 years from six different countries were carried out. Content validity was established by conducting 69 cognitive interviews with feedback and advice obtained from 44 international experts.  It was then field tested in an international sample of 2,434 patients from 30 hospitals in 12 countries.  The authors of the CLEFT-Q state that "It represents a new generation of PROMS developed using a modern psychometric approach called the Rasch Measurement Theory".  The team followed internationally recommended guidelines to create the CLEFT-Q. A detailed description of the protocol has been published: https://bmjopen.bmj.com/content/10/3/e032332.

The CLEFT-Q has 12 independently functioning scales that measure three overarching domains, and one checklist.  Given the variety of scales, this provides clinicians with the flexibility to choose particular subsets of scales to measure their outcome of interest. The authors of the measure advise that the CLEFT-Q is included in ICHOM Standard Sets for craniofacial conditions to enable hospitals around the world to compare outcomes.  This sets the measure up as an ideal tool to use across the UK as a way of comparing our outcomes nationally. Our data could be compared internationally in the future. Please see the CLEFT-Q website for further details on its development. The CLEFT-Q Users Guide can be found here: CLEFT-Q-USERS-GUIDE.pdf (qportfolio.org)

Each of the three domains within the CLEFT-Q is composed of one or more independently functioning scales. The three domains are: Appearance, Facial Function and Health-related Quality of Life (H-RQoL). Within H-RQoL, there are four scales: psychological, social and school functioning and speech-related distress.   The CEN has agreed that we will utilise the Psychological and Social Functioning sub-scales to submit to CRANE, as there is a degree of over-lap with the school functioning sub-scale and we felt that speech-related distress was more specific to our Speech and Language Therapy colleagues.

Having reviewed our cleft cohort, it was also agreed that we would collect data for patients aged 10. We felt that this was a clinically beneficial time, given children would be transitioning to secondary school aged 11 and that attendance at clinic appointments aged 10 is optimal, as patients are also invited to meet with other members of the MDT at that age.  This means we are likely to obtain data from a representative sample of our patients. We also felt that seeing patients aged 10 reflected a more proactive intervention from Psychology, as issues can be identified as children also transition into adolescence, which we recognise can result in an increase in psychological distress.  We discussed meeting with 15- or 20-year-olds but identified that attendance is less optimal at these ages due to GCSE/work/further education commitments, which could impact outlier status for all and compromise the validity of our findings.  We all agreed that we want to ensure that we collect data that is meaningful and informative for Clinical

Psychology, our MDT colleagues and for our Patients. There was consensus that the use of these two CLEFT-Q sub-scales provides an optimal opportunity to do so.

We recognise that different Centres work differently and that the agreed changes would be more difficult to introduce in some centres than others. With that in mind, having confirmed the measure that we want to use, each Centre was tasked with conducting their own feasibility study from April 2024. Each Centre was asked to register to access the measure in order to sign their own Licensing Agreement and to consider their staffing, room availability and administrative support. To obtain a licensing agreement, please use the following link: https://research.mcmaster.ca/industry-investors/technologies-available-for-licensing/request-for-license/ Our CRANE representatives were made aware that they will need a licensing agreement in order to analyse the national data. Dr Jo Shearer contacted the CLEFT-Q team to request liaison with CRANE (see email dated 03.10.24).

The CLEFT-Q comes in multiple languages. The translation list can be found here: http://www.qportfolio.org/. It is the responsibility of each Cleft Centre to liaise with CLEFT-Q for access to any languages that they may require. Our Welsh colleagues are in liaison already about access to Welsh versions as this is not currently on the list. It is the responsibility of each Cleft Centre to liaise with CLEFT-Q about using electronic copies of the measure. Please review the User's Guide. The CLEFT-Q Computerized Adaptive Test (CAT) is available. It uses algorithms to select the most relevant items from each scale, based on the responses provided up to that point. There may be a small administration charge for using the CAT. The CEN do not feel this is required for the purposes of CRANE, as we will be using only two of the 12 sub-scales, which we believe will not be burdensome to our Patients.

We have agreed that a member of the Clinical Psychology Service will meet with individuals either face-to-face or via video. This can include pre-qualified staff, providing they receive adequate training beforehand and have access to supervision from a qualified Clinical Psychologist. In order to ensure equity of access, telephone consultations can be carried out if a family cannot attend in person and does not have access to technology to attend a video consultation.

We agreed that each Centre would contact Dr Jo Shearer (CEN CRANE Rep) and Dr Kat Berlouis (Locum CEN Chair) with any pressing concerns by September 2024. The time frame involves feedback to CRANE by October 2024 so that the database can be built, with a view to submitting data from January 2025. This will mean collecting data from January 2025 for those born in 2015. We recognise that we will need to collect data for three years before the data reaches maturity and that CRANE cannot analyse any data for the first 12 months. The CEN will continue to collect 5-year Tiers of Involvement (TIM) scores but hope to re-visit this once the CLEFT-Q data is mature and fully established. We are considering whether we collect 10-year TIM scores from January 2025 with a view to retiring the 5-year TIM once the data reaches maturity. This is yet to be confirmed, and Kat and Jo will liaise with the CEN for confirmation prior to January 2025. The TIM score will remain our Process Measure for the time-being. This will be discussed at the next CEN meeting in November 2024.

Whilst we have made every effort to future proof our decision, we recognise that we cannot predict changes in the longer term. We are also aware that with time new cleft-specific measures may become available that may be deemed more beneficial to our patients in terms of understanding their psychological outcomes. For now, we feel that the two sub-scales reflect many of the themes we come across within our clinical practise, the results of which will support us in understanding and therefore supporting our patients.

Dr Jo Shearer
Principal Clinical Psychologist
Lead Clinician for North Thames Cleft Lip/Palate Service, Maxillofacial & Dental Services
CRANE Representative for the Psychology CEN