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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

2023 Annual Report

Results of the audit in England, Wales, Northern Ireland and Scotland for children born with a cleft between January 2000 and December 2022

On behalf of the Cleft Development Group

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

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This version (1.1) was released on 22 January 2024 and includes a correction of the x-axis label and key (corrected to 2020-22 births) for Figure 3.2, page 11 (Chapter 3: Registry Information).



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For further information, please refer to the website

<http://craniofacialsociety.co.uk/>

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

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

Document	Published
2023 Annual Report: The supplementary tables (Appendices) (Excel Workbook)	December 2023
2023 Annual Report: Responses to outlier process (Appendices)	December 2023

Acknowledgements

We would like to thank the following people and organisations:

- Children born with a cleft, their families, and the Cleft Services in England, Wales, Northern Ireland and Scotland for the collection and submission of data into the Cleft Registry & Audit NETwork (CRANE) Database;
- members of the UK NHS Cleft Development Group (CDG), Chaired by Simon Van Eeden, for their advice;
- Jackie Smallridge for her contribution towards the interpretation of the dental outcome data at 5 years of age – on behalf of the Dental Clinical Excellence Network (CEN);
- Lorraine Britton for her contribution towards the interpretation of the speech outcome data at 5 years of age – on behalf of the Lead Speech and Language Therapy group and CEN;
- Jo Shearer and Katherine Berlouse for their contribution towards the interpretation of the psychology outcome data at 5 years of age – on behalf of the Psychology CEN;
- Sinéad Davis and Victoria Parfect for their contribution towards the interpretation of the Newborn Hearing Screening Programme data – on behalf of the ENT and Audiology CEN;
- colleagues at the Clinical Effectiveness Unit (CEU) of the Royal College of Surgeons of England for their methodological and statistical input;
- Catherine de Blacam for the use of her medical drawings to illustrate the different cleft types.

We would like to acknowledge all at the UK NHS CDG and Cleft Services for supporting the outlier process, which was piloted in 2021 and implemented from 2022. In particular, we would like to recognise the extra time and effort it has taken for clinical colleagues to engage and respond so constructively to outlier communications since 2021. The positive way in which the outlier process has been received and responded to is hugely encouraging for the future of national clinical audit of cleft care. The process has been adopted by the CDG for use from 2022. It has been introduced in a staged manner in 2023, and will be done so up to and including the 2024 CRANE report to allow all teams the necessary time to put in place appropriate resources. The staged implementation also reflects the significant ongoing pressures experienced by clinical services during recovery from the various waves of the COVID-19 pandemic. It is planned that from 2025 the outlier policy will be implemented in full.

This work was funded by the Specialised Services National Business Intelligence and Information Team NHS England and NHS Improvement on behalf of England and Wales, the Northern Ireland Specialist Services Commissioning Team and Cleft Surgical Service for Scotland. It was carried out by the project team of the CRANE Database, which is overseen by the UK NHS CDG¹.

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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

“DO THE BEST YOU CAN UNTIL YOU KNOW BETTER. THEN WHEN YOU KNOW BETTER, DO BETTER” MAYA ANGELOU

It is an honour and a privilege to write the foreword for the CRANE 2023 Annual Report. The report summarises the data received from Cleft Services in England, Wales, Northern Ireland and Scotland and reports on their outcomes. The report also delineates current database development and references works that have successfully been published and those that are ongoing. Congratulations are due to the dedicated hardworking CRANE team and of course to all the cleft professionals and administration teams involved in data collection and submission. I commend this report to all interested parties and hope you find it as informative and illuminating as I have.

As we enter the festive period and time for celebration it is right to celebrate the success that CRANE is. Since the recognition in the Clinical Standards Advisory (CSAG) report for Cleft of a lack of meaningful data collection across the country and the statutory requirement for a national database, data collection and submission first to the Craniofacial Anomalies Register (CARE) and then to its successor CRANE by all cleft teams in England, Wales and Northern Ireland has resulted in the largest dataset for cleft in the world. Scotland has recently joined CRANE ensuring that CRANE now collects data for the whole of the UK. This is a significant achievement and has been made possible by the teams of dedicated cleft professionals around the UK and investment in CRANE.

Data collection is time consuming and requires foresight, planning and administrative support and is a challenge made worse by the recent COVID pandemic. The focus of cleft teams is and always has been to provide the best care for patients and it is easy in times of limited resource and increased pressure on services to shift the focus away from data collection. This, in my view, is short-sighted as it is this data that enables us to examine our outcomes, make meaningful comparisons and ultimately improve the care that is delivered to patients. Data completeness is critical for valid comparison and CRANE has played an essential role in emphasising the importance of data collection and data completeness by running feedback sessions and educational webinars, by providing easy to access resources for cleft teams on their website and through presentations to specialist Lead groups and Clinical Excellence Networks, the Quality Monitoring and Improvement Committee, and the Cleft Development Group. This has maintained the momentum of data collection despite all the challenges at this time of unprecedented pressure on the NHS.

While CRANE holds the largest longitudinal cleft database in the world it is one of the smaller audit projects housed in the Clinical Effectiveness Unit of the Royal College of Surgeons in England. As a relatively small audit, CRANE has struggled for some time to attract adequate funding to pursue the full possibilities inherent in modern data collection, linkage and analysis. Short-term funding has made data linkage to other databases, such as Hospital Episode Statistics (HES), challenging resulting in the need to prioritise some projects over others. Despite this though, important linkage work has continued with the linkage of CRANE data to the Newborn Hearing Screening Programme (NHSP) which helps for the first time, in such a large cohort, to elucidate the relationship between cleft type and hearing. CRANE has also continued to contribute to the scientific literature with four new publications and multiple conference presentations. It is also very pleasing to note from the case ascertainment work reported in this report that CRANE continues

to have a high case attainment rate when assessed against HES data and data from the Office for National Statistics. The collection of outcome data on a relatively large scale means that it is now possible to start developing risk adjustment models for different cleft outcomes - this work is still ongoing. The importance of all this work for the benefit of cleft patients cannot be emphasised enough and is quite simply the envy of the cleft world and any talk of funding loss for the CRANE Database should be robustly opposed.

Data collection is clearly important and essential as it enables meaningful data analysis which, on its own, has been shown to improve outcomes and raise standards. This, however, is not enough and focus over the last few years has shifted to looking at the variability in outcomes amongst cleft teams across the UK. The CRANE Outlier Policy and process while not fully operational is currently being used to inform cleft teams of their outcomes based on the funnel plots within this and previous annual reports. These funnel plots depict cleft team performance relative to the national mean and provide a very useful benchmark for cleft teams. The cleft community is now turning its focus to those teams performing well to try and understand why their outcomes are better so that all teams improve and in so doing decrease the year on year variability in outcomes which haven't improved significantly since the initial improvements seen after CSAG service centralisation. The factors for variability in outcome will be multifactorial but understanding these factors is what is required to ultimately ensure that the standard of care delivered across the country is the same for all cleft patients.

I refer you back to the opening quote from Maya Angelou, "do the best you can until you know better. Then when you know better, do better."

This, I think, succinctly sums up the ethos and aspiration of CRANE and all involved in cleft care in the UK. I am confident that cleft care in the UK will go from strength to strength in the years to come and I look forward to following its progress.

Wishing you all a safe and wonderful festive season and all you can hope for in the New Year!

Simon van Eeden

Chair of the Cleft Development Group

Executive summary



CRANE

CLEFT REGISTRY & AUDIT NETWORK

Annual Report 2023

CRANE is a national registry and clinical audit. It aims to evaluate and report on the delivery of cleft services to children in England, Wales, Northern Ireland and Scotland with the congenital abnormality of cleft lip and/or palate. The registry information presented is for children born in 2020 to 2022. The audit outcomes presented are for children born in 2014-2016.

Registry information

23,606 children have been registered in CRANE, over the last 23 years, since 2000.

896 children were registered in 2022.

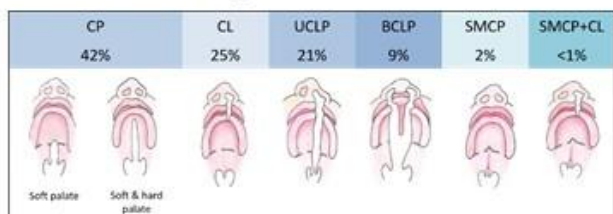


76% of babies with a cleft were diagnosed before or within the first 24 hours after birth.



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

Distribution of cleft type



81% of families had a verified consent status, 79% consented and 2% declined consent.

Audit outcomes at 5 years of age



Body mass index

82%

of children with a cleft had a healthy BMI. 4% were underweight, 10% 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

45%

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



Speech

60%

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



Psychology

84%

of children with a cleft had 'Strengths and Difficulties' scores* in the low/normal range, compared to 90% of their non-cleft peers. Furthermore, 93% of families were screened at least once before the target age of 6 years.

Development work

CRANE case ascertainment confirmed using ONS & Hospital Episode Statistics (HES) data

High case ascertainment levels in CRANE were confirmed using Office for National Statistics (ONS) and HES data for live births.

The overall estimate for the prevalence of cleft lip and/or palate (CL/P) was **15.1 per 10,000 live births** in CRANE, from 2009 to 2018 in England.

CRANE - Newborn Hearing Screening Programme (NHSP) data linkage

For the first time, CRANE-consented children were linked to the NHSP at the individual-level.



Where records were linked, **98%** of children had a complete newborn hearing screening assessment.

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

Key findings and recommendations 2023

Source	Findings	Recommendations
Registry		
Registrations: Cleft type, Robin Sequence and sex		
Chapter 3, Sections 3.1.1-3.1.3	<ul style="list-style-type: none"> • There were 2,640 CRANE-registered children born 2020-2022. • 42% had cleft palate (CP), 25% 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 0.5% had SMCP with CL. • 21% of children with CP were reported to have Robin Sequence. • 57% of registrations were boys and 43% were girls. 	<ol style="list-style-type: none"> 1. It is recommended that services ensure RS status is accurately reported for all children with a CP. 2. CRANE will engage with the Royal College of Midwives (RCM) and Royal College of Obstetricians and Gynaecologists (RCOG) to communicate our findings.
Gestational age and birthweight		
Chapter 3, Sections 3.1.4-3.1.5 Indicators²: #1 & #2	<ul style="list-style-type: none"> • 55% of CRANE-consented children had a recorded gestational age and birthweight. • The average gestational age of babies born with a cleft in 2020-22 was 38.6 weeks. • 12% of babies with a known gestational age were born prematurely (<37 weeks' gestation). This compares to 8% in the general population³. • The average birthweight was 3,213g. • Children with a BCLP and CP had significantly lower birthweights than those with CL. • 12% of babies with birthweight reported had a low birthweight (<2,500g). This compares to 6% in the general population². The rate among babies born at term (was 5%, while the corresponding rate in the general population is 3%. 	<ol style="list-style-type: none"> 3. 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. 4. 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.
Timing of diagnosis		
Chapter 3, Section 3.2 Indicator: #3 & #4	<ul style="list-style-type: none"> • 95% of CRANE-registered children had diagnosis time reported. • 76% of children (without SMCP) received a timely diagnosis; 78% of children with a cleft involving the lip were diagnosed antenatally, while 72% of children with CP were diagnosed before or within 24hrs of birth. Only 33% of children with SMCP were diagnosed before or within 24hrs of birth. • Despite improvement in the timely detection of CPs, more than 1 in 4 children still have a diagnosis beyond 24 hours from birth. 	<ol style="list-style-type: none"> 5. CRANE will seek to collaborate further with the clinical workforce to identify barriers to recording timing of diagnosis for all registrations in CRANE. 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. 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.

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

³ Birth characteristics in England and Wales: 2021. Available at:

<https://www.ons.gov.uk/peoplepopulationandcommunity/birthsdeathsandmarriages/livebirths/bulletins/birthcharacteristicsinenglandandwales/2021> [Last accessed: 04/12/2023]

Referral to and contact with Cleft Services

<p>Chapter 3, Section 3.3</p> <p>Indicators: #5 to #8</p>	<ul style="list-style-type: none"> 74% of CRANE-registered children had a recorded 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. 91% 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. 	<ol style="list-style-type: none"> 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. Cleft Services with high levels of referral and contact times within 24 hours should share their best practice recommendations.
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CRANE consent

<p>Chapter 4</p> <p>Indicator: #9</p>	<ul style="list-style-type: none"> Consent status was verified for 81% of children born 2020-22 and 90% of those born 2014-16. This meant they had given informed consent or declined consent for CRANE to collect outcome data. Consent verification rates varied significantly according to Cleft Service and birth year. 	<ol style="list-style-type: none"> Cleft Services should review their procedures to identify reasons for low rates of verified consent, and ensure that CRANE consent status is recorded for every child with a cleft. CRANE will continue to work with Cleft Services and the Nursing CEN to improve consent status verification.
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Outcomes at 5 years

Child growth

<p>Chapter 5, Section 5.1</p> <p>Indicator: #10 & #11</p>	<ul style="list-style-type: none"> 36% of CRANE-consented children had a recorded height and weight. 82% of children with growth data had a healthy body mass index (BMI). This compares to 77% in the general population of 5 year olds⁴. According to BMI categorisation, 4% were underweight, 10% overweight and 5% obese. Corresponding rates in the general population are 1%, 12% and 9%, respectively. 	<ol style="list-style-type: none"> 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. CRANE will continue to liaise with CDG members and the nursing CEN to encourage all services to collect this data.
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Dental health

<p>Chapter 5, Section 5.2</p> <p>Indicators: #12 to #16</p>	<ul style="list-style-type: none"> 36% of CRANE-consented children had recorded dmft scores. Dental decay was experienced by 38% of children (having at least one decayed, missing or filled tooth (dmft >0)) and 13% of children were classified as having extensive caries (dmft >5). The average Treatment Index (rate of treated disease) was 77%, and the average Care Index (having received the appropriate care at the earliest possible stage) was 70%. 	<ol style="list-style-type: none"> Cleft Services should aim for a calibrated specialist in paediatric dentistry to see all children for a dental assessment at 5 years of age 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.
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⁴ National Child Measurement Programme Tables, England 2019/20 School Year – Published 29th October 2020 – Available at <https://digital.nhs.uk/data-and-information/publications/statistical/national-child-measurement-programme/2019-20-school-year> [Last accessed: 12/12/2023]

Facial growth (for children with complete UCLP)

<p>Chapter 5, Section 5.3</p> <p>Indicator: #17 & #18</p>	<ul style="list-style-type: none"> • 45% of CRANE-children with a complete UCLP had 5-year-old Index scores reported. • Of these, 45% had scores reflecting ‘good’ dental arch relationships, 33% ‘fair’ and 22% ‘poor’. 	<p>16. Cleft Services should aim to take dental impressions or photographs of all children with a complete UCLP around the age of 5 years, to allow for an assessment using the 5-year-old Index.</p> <p>17. The research community should undertake to compare UK facial growth outcomes with those in other countries and evaluate the predictive value of the 5-year-old Index in the UK.</p>
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Speech (for children with CP, UCLP and BCLP)

<p>Chapter 5, Section 5.4</p> <p>Indicators: #19 to #22</p>	<ul style="list-style-type: none"> • 46% 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. • 60% met speech outcome standard 1: The achievement of speech with no evidence of a structurally related problem and no cleft speech characteristics requiring intervention. • 72% met speech outcome standard 2a: The achievement of speech without evidence of a structurally related speech difficulty. • 68% met speech outcome standard 3: The achievement of speech without evidence of significant cleft-related speech characteristics, which may require therapy or surgery. • 17% of children had secondary surgery for speech purposes before the age of 5 years. 	<p>18. Children with a cleft affecting the palate should have speech assessed and reported to CRANE.</p> <p>19. Where possible, Cleft Services should investigate possibilities of remote methods for consensus listening.</p> <p>20. Cleft Services should work together to explore reasons for variation in speech outcomes.</p>
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Psychology screening

<p>Chapter 5, Section 5.5</p> <p>Indicators: #23 to #26</p>	<ul style="list-style-type: none"> • 53% of CRANE-consented children had recorded TIM scores • Of these, 93% 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 4, referred to as 1a+). • 44% of CRANE-consented children had a Strengths and Difficulties Questionnaire (SDQ) score. • Of these, 16% had ‘high’ or ‘very high’ SDQ scores, indicating a greater level of difficulties, which may require psychological input or intervention. 	<p>21. 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).</p>
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All outcomes at 5 years

<p>Chapter 5</p>	<ul style="list-style-type: none"> • An outlier process was applied to children’s outcomes at 5 years of age. Cleft Services were permitted to provide a response to being identified as a positive or negative outlier. 	<p>22. All Cleft Services should work together to explore reasons for variation data completeness and outcomes at 5 years of age.</p>
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Data quality – throughout report

<p>Chapters 3 to 5</p>	<ul style="list-style-type: none"> • There was significant variation in data completeness for registrations and also for specialty-specific outcomes across the periods analysed for this report. • Recovery from the COVID-19 pandemic has significantly affected some specialties’ ability to submit 5-year-old outcome data. 	<p>23. Cleft Services identified as negative outliers for data completeness are encouraged to consider methods for improving the capture and reporting of these data.</p> <p>24. Cleft Services, Clinical Excellence Networks (CENs) and CRANE should work together to identify and overcome barriers to collecting and submitting data.</p>
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Database development work

Case ascertainment

Chapter 6, Section 6.1	<ul style="list-style-type: none"> Using live births from the Office for National Statistics⁵ and CRANE registrations for births from 2009 to 2018 in England, the overall estimate for the prevalence of cleft lip and/or palate (CL/P) was 15.1 per 10,000 live births. Using Hospital Episode Statistics (HES), the overall estimate for the prevalence of CL/P was also 15.1 per 10,000 live births. Although fluctuations and differences between the two data sources exist in annual prevalence rates, the similar overall rate indicates high case ascertainment levels in CRANE. 	25. The research community should undertake to explore whether regional differences in the prevalence of clefting exist across England.
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Newborn Hearing Screening Programme

Chapter 6, Section 6.2	<ul style="list-style-type: none"> For the first time, CRANE-consented children have been linked to the Newborn Hearing Screening Programme (NHSP) at the individual-level. 11,944 (95%) eligible children were linked to NHSP and 11,730 (98%) had a complete newborn hearing screening assessment. 24% of children had no clear response in one or both ears. Cleft type influenced both linkage rate and screening outcome significantly. 	26. Further investigations are needed to understand why linkage rate and screening outcome vary according to cleft type. In certain cases babies are referred directly for diagnostic assessment and miss out the 'screening' hearing assessment step. This would be more likely to occur in children with syndromes. Children with no clear response are referred for audiological diagnostic hearing screen assessments; this part of the process needs to be explored further.
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Peer-reviewed publications and presentations

Chapter 6, Section 6.3	<ul style="list-style-type: none"> Four scientific articles were published over the last year. <ol style="list-style-type: none"> Etoori D, Park MH, Blackburn RM, Fitzsimons KJ, Butterworth S, Medina J, et al. 2023 Number and timing of primary cleft lip and palate repair surgeries in England: whole nation study of electronic health records before and during the COVID-19 pandemic. <i>BMJ Open</i> 13(6):e071973. Park MH, Fitzsimons KJ, Deacon S, Medina J, Wahedally MAH, Butterworth S, et al. 2023 Longitudinal educational attainment among children with isolated oral cleft: a cohort study. <i>Archives of Disease in Childhood</i> 108:563-568. Butterworth S, Fitzsimons KJ, Medina J, et al. 2023 Investigating the Impact of Patient-Related Factors on Speech Outcomes at 5 Years of Age in Children With a Cleft Palate. <i>The Cleft Palate Craniofacial Journal</i>. 60(12):1578-1590 Fitzsimons K, Hamilton M, van der Meulen J, et al. 2023 Range and frequency of congenital malformations among children with cleft lip and/or palate. <i>The Cleft Palate Craniofacial Journal</i>. 60(8):917-927. CRANE was involved in four different oral presentations at the Craniofacial Society of Great Britain and Ireland Annual Conference in April 2023. 	27. Collaboration is key to sharing CRANE data and facilitating research that informs clinicians, families and policy makers.
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⁵ Office for National Statistics: Annual summary statistics on live births and stillbirths in England and Wales, 2019. Released July 2020 (checked October 2020). Available at: <https://www.ons.gov.uk/peoplepopulationandcommunity/birthsdeathsandmarriages/livebirths/datasets/birthsummarytables>

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 Wales⁶. 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 used to further examine treatment and outcomes for cleft lip and/or palate in England and, in the last year, we have linked with the Newborn Hearing Screening Programme in England.

The aims of the CRANE Database are:

1. to register birth, demographic and epidemiological data for all children born in the UK with the congenital abnormality of cleft lip and/or palate;
2. to record the treatment of children with a cleft lip and/or palate and the outcome of such treatment.

This Annual Report presents findings from data submitted to the CRANE Database⁷ 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 2020-2022.
- **Consent:** Consent levels for those born 2020-2022, reflecting recent registrations, and those born in 2014-2016, 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 2014-2016.
- **Database development work:** Development activity undertaken by the CRANE team over the last 12 months. This year we focus case ascertainment levels in the CRANE Database compared with Hospital Episode Statistics and we report on linkage between CRANE and the Newborn Hearing Screening Programme.

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](#).

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 4 July 2023.

2. Methods

2.1. Datasets

2.1.1. CRANE

CRANE is an online custom-built secure database that holds information on children born with a cleft lip and/or palate in England, Wales and Northern Ireland. Scotland joined in January 2023, making CRANE a UK-wide registry and audit. 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 approval⁸ 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 (where relevant). Broadly, timeframes are the most recent years of available data: 2020-2022 births for Registry information and 2014-2016 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, and (3) 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 <u>L</u> ip	Right <u>A</u> lveolus	<u>H</u> ard palate	<u>S</u> oft palate	Left <u>A</u> lveolus	Left <u>L</u> ip

⁸ Confidential Advisory Group (CAG) Section 251 Approval <https://www.hra.nhs.uk/approvals-amendments/what-approvals-do-i-need/confidentiality-advisory-group/> [Last accessed 23/11/2023]

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 cases⁹), cleft type is based on the type reported by the region/ unit registering the child. 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, with the exception of tables and figures relating to data completeness (see [the supplementary tables](#) for a breakdown of those reported for each outcome).

2.1.2. Hospital Episode Statistics (HES)

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

2.1.3. 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 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 Kruskal-Wallis test was used to compare differences between exposure categories.

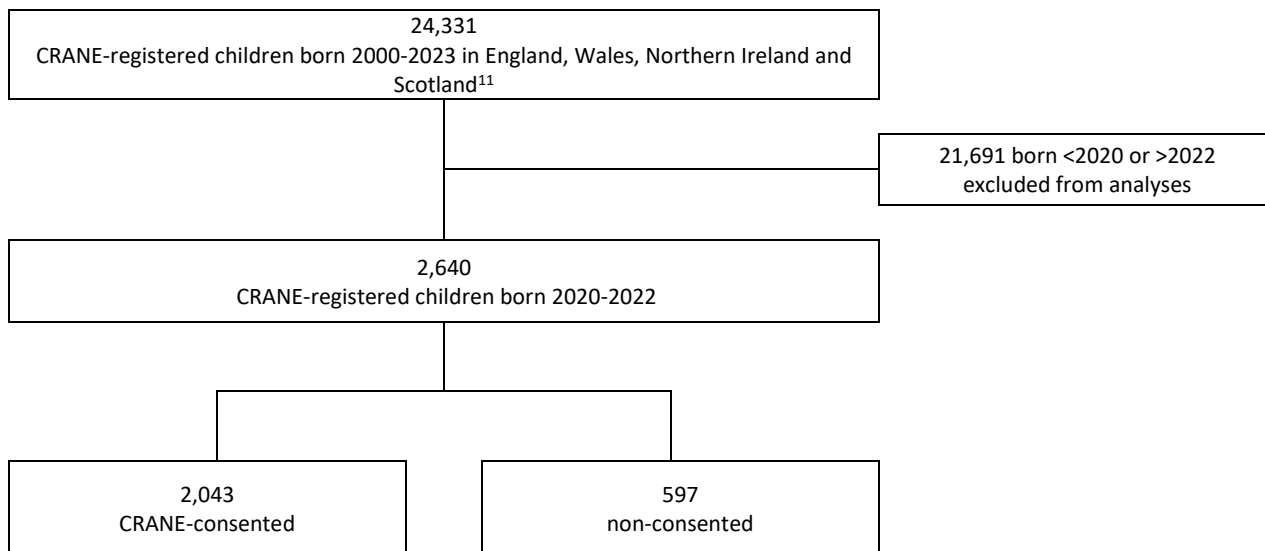
⁹ LAHSAL has not been reported for 7% of children registered between 01 January 2020 and 31 December 2022.

¹⁰ Overview of Newborn Hearing Screening Programme: <https://www.nhs.uk/conditions/baby/newborn-screening/hearing-test/> [Last accessed: 24/02/2023]

3. Registry information

This chapter details the characteristics of CRANE-registered children born with a cleft lip and/or palate in 2020-2022. 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=2,043 (77%)	CRANE non-consented N=597 (23%)
Cleft type	✓	✓
Robin Sequence	Cleft palate alone	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 is an exception to the Section 251 exemption and is collected for all children whose families have given informed consent to outcomes data collection by the CRANE Database.


¹¹ Scotland joined in January 2023 so have one year (births in 2022) of registrations only.


3.1. CRANE registrations: patient and birth characteristics

The CRANE Database has registered more than 24,000 children born in England, Wales, Northern Ireland and Scotland with cleft lip and / or palate over the last 23 years. Cleft Services are asked to register all children born alive with a cleft in the UK (Scotland joined in 2023). For the most recent three-year reporting period (2020-2022 births), the total number of registrations ranged from 96 to 355 between Cleft Services (see ‘Registrations’ in Supplementary tables). This section describes the patient characteristics of these registrations.

Cohort summary	
Data source	The CRANE Database. Extract taken: 4 July 2023
Birth years	Three years: 2020 to 2022
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	No exclusions
Raw data	‘Patient characteristics’, ‘Gestation’ & ‘Birthweight’ in the supplementary tables

3.1.1 Cleft type

Data completeness: Cleft type specification	
Notes	Cleft type is based on the LAHSAL code provided. Where this is not available, services can select cleft type from a drop-down menu. LAHSAL code is important because it provides information on the laterality of the cleft lip and the completeness of the cleft.
Denominator	2,640 CRANE-registered children
What did we find? 	<ul style="list-style-type: none"> 93% of CRANE-registered children had their cleft type specified (Cleft Service range: 69%-100%, $p < 0.001$). This compares to 92% of those born 2018-2020. Reporting did not vary significantly according to birth year ($p = 0.202$).

Outcome: Cleft type distribution and cleft characteristics	
Denominator	2,459 children with cleft type reported
What did we find? 	<ul style="list-style-type: none"> 42% of children had a cleft palate alone (CP). 25% of children had a cleft lip alone (CL). Of these, 58% were left-sided, 33% were right-sided and 8% were bilateral. Among those with a unilateral CL, the left:right ratio was 63%:37%. 21% of children had a unilateral cleft lip and palate (UCLP). The left:right ratio was 63%:37%. Overall, 71% had a complete UCLP¹³. 9% of children had a bilateral cleft lip and palate (BCLP). 46% had a complete BCLP. 2% of children had a submucous cleft palate (SMCP) alone. <1% of children had a submucous cleft palate with cleft lip (SMCP+L) There were no significant differences in the distribution of the four main cleft types (CL, CP, UCLP & BCLP) across Cleft Services ($p = 0.576$); however, Northern Ireland registered a much higher proportion of children with SMCP (22% of their registrations) than the overall average (2% of registrations).

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

¹³ A complete UCLP was defined as LAHS or HSAL codes, indicating a complete cleft affecting all three components of the mouth on either the right or left side

3.1.2 Robin Sequence

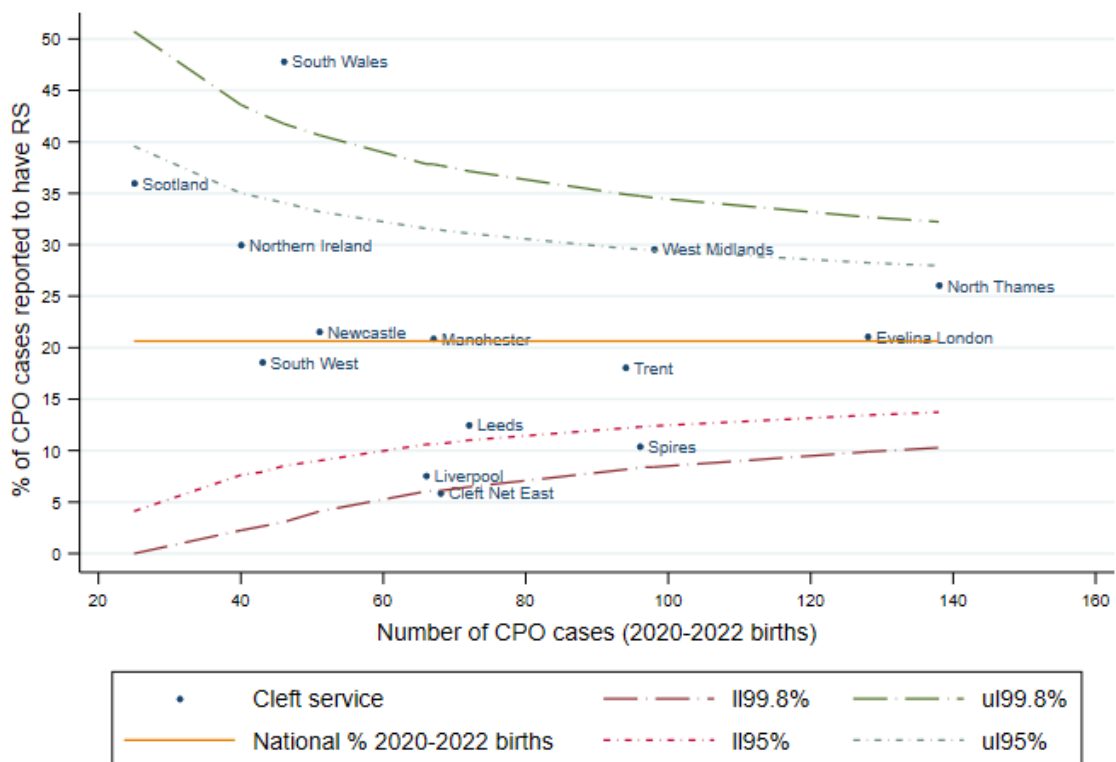
Data completeness: Robin Sequence status	
Notes	<ul style="list-style-type: none"> For children with a cleft palate alone, services are required to report Robin Sequence (RS) status (present or absent). RS is a congenital birth defect where micrognathia and glossoptosis appear together with cleft palate.
Denominator	1,032 CRANE-registered children with cleft palate alone (excludes those with SMCP)
What did we find?	88% of CRANE-registered children with cleft palate alone had their Robin Sequence status (present or absent) specified (Cleft Service range: 12%-100%, $p < 0.001$).




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	1,032 children with cleft palate
What did we find?	<ul style="list-style-type: none"> 21% of children with cleft palate alone were reported to have RS (Cleft Service range: 6%-48%, $p < 0.001$). The rate was 23% among those with a specified RS status (Cleft Service range: 10%-50%). These rates equate to an average of 71 children born each year with RS. 68% of children with cleft palate alone were confirmed as not having RS (Cleft Service range: 52%-90%³). A further 12% did not have their status confirmed and are therefore assumed not to have it.
Outliers	Positive: 1. South Wales (48%) Negative: 1. Cleft Net East (6%)




Figure 3.2. Funnel plot showing the percentage of CRANE-registered children with cleft palate alone, born 2020 to 2022, with Robin Sequence (RS), according to Cleft Service.




3.1.3 Sex

Data completeness: sex	
Denominator	2,640 CRANE-registered children
What did we find? 	99% of CRANE-registered children had their sex specified (Cleft Service range: 97%-100%, p=0.001). This has not changed compared to the last reporting period (births 2018-2020).

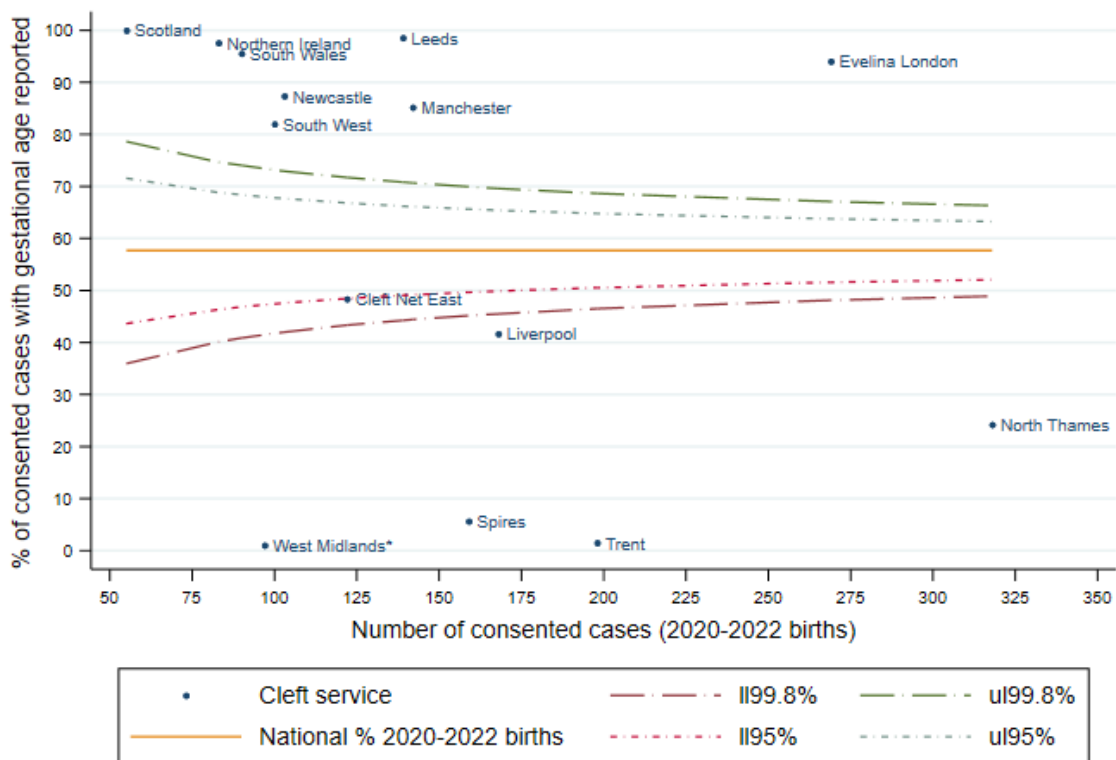
Outcome: Sex ratio	
Denominator	2,625 children with sex reported
What did we find? 	<ul style="list-style-type: none"> There were more boys (57%) than girls (43%) with a cleft (Cleft Service range for boys: 46%-64%, p=0.181). The sex ratio varied significantly according to cleft type (p<0.001). There were more boys than girls with clefts involving the lip (range: 63% in CL - 70% in CL+SMCP) and more girls (56%) than boys with clefts affecting only the palate.

3.1.4 Gestation

Data completeness: Gestational age	
Indicator	#1 - Gestational age recorded for all eligible children
Notes	<ul style="list-style-type: none"> Not subject to outlier policy but funnel plot provided to demonstrate variation in data completeness across services. Funnel plot is centred on the revised national rate after excluding data from services identified as negative outliers for consent verification (West Midlands).
Denominator	2,043 CRANE-consented children
What did we find? 	<ul style="list-style-type: none"> 55% of CRANE-consented children had a recorded gestational age (Cleft Service range: 1%-100%, p<0.001). This is similar to the 56% of those born 2018-2020. Reporting did not vary significantly according to birth year (p=0.706). <1% had a reason¹⁴ why gestational age data were not collected. 45% were missing data or a reason for not collecting data.
Outliers	<p>Positive: 1. Scotland (100%), 2. Leeds (99%), 3. Northern Ireland (98%), 4. South Wales (96%), 5. Evelina London (94%), 6. Newcastle (87%), 7. Manchester (85%), 8. South West (82%)</p> <p>Negative: 1. West Midlands (1%), 2. Trent (2%), 3. Spires (6%), 4. North Thames (24%), 5. Liverpool (42%)</p>

¹⁴ 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.

Figure 3.3. Funnel plot showing the percentage of CRANE-consented children, born 2020 to 2022, with gestational age data reported, according to Cleft Service.



Note: Funnel plot centred on the revised national percentage (57.7%) of children (born 2020-2022) with gestational age data reported.

Outcome: Gestational age	
Benchmarks	Among babies born in 2021 in the general population of England & Wales, 8% had premature births (<37 weeks' gestation) ¹⁵ .
Denominator	1,124 CRANE-consented children with gestational age reported
What did we find?	<ul style="list-style-type: none"> The mean gestational age was 38.6 weeks (95% CI 38.4-38.7 weeks). The percentage of premature births among children with gestational age reported was 12% (Cleft Service range 5.1%-22%¹⁶, p=0.622) and is higher than in the general population (8%). The percentage of premature births did not significantly differ according cleft type (p=0.076) or sex (p=0.638). It should be noted that the gestational age recorded in CRANE may not be representative of all babies born with a cleft as almost half of all consented children were missing information on this outcome.



¹⁵ Birth characteristics in England and Wales: 2021. Available at: <https://www.ons.gov.uk/peoplepopulationandcommunity/birthsdeathsandmarriages/livebirths/datasets/birthcharacteristicsinenglandandwales> [Last accessed: 19/11/2023]

¹⁶ among Cleft Services that submitted data for ≥10 cases.

3.1.5 Birthweight


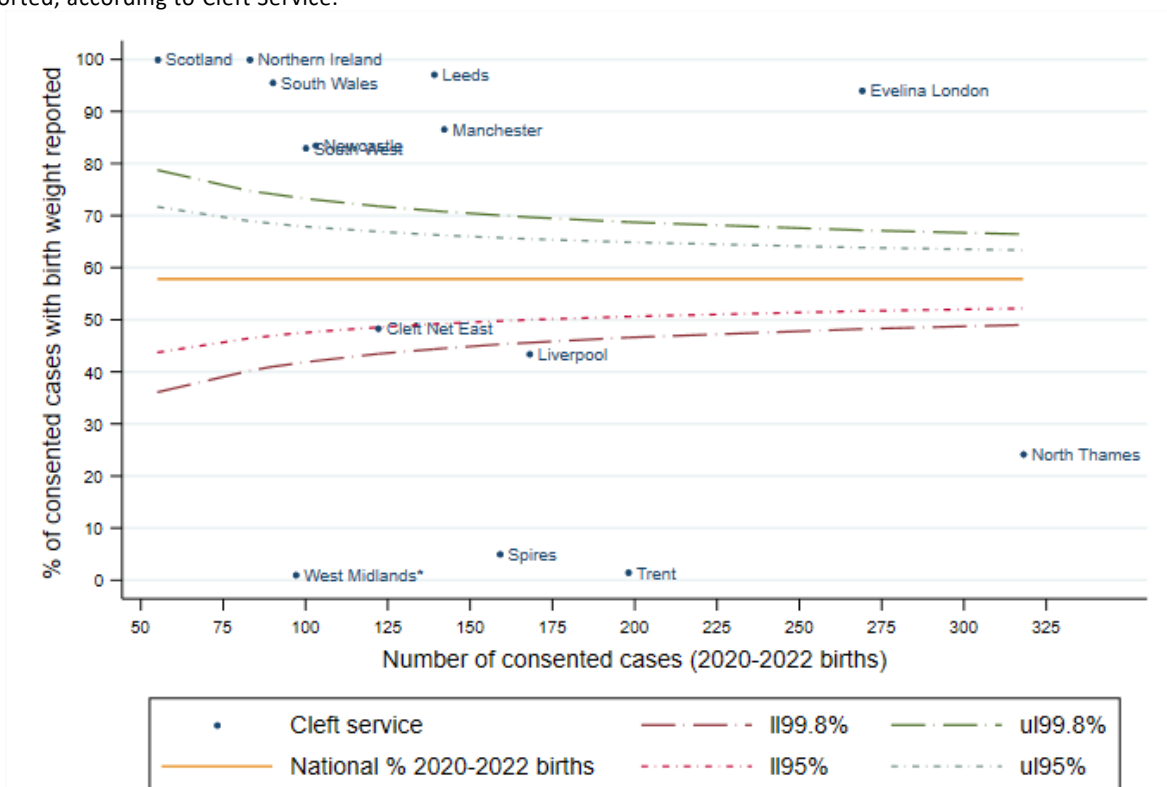

Data completeness: Birthweight	
Indicator	#2 - Birthweight recorded for all eligible children
Notes	<ul style="list-style-type: none"> Not subject to outlier policy but funnel plot provided to demonstrate variation in data completeness across services. Funnel plot is centred on the revised national rate after excluding data from services identified as negative outliers for consent verification (West Midlands).
Denominator	2,043 CRANE-consented children
What did we find?	 <ul style="list-style-type: none"> 55% of CRANE-consented children had a recorded birthweight (Cleft Service range: 1%-100%, $p < 0.001$). This is similar to the 56% of those born 2018-2020. Reporting did not vary significantly according to birth year ($p = 0.508$). <1% had a reason¹⁷ why birthweight data were not collected. 45% were missing data or a reason for not collecting data.
Outliers	<p>Positive: 1. Scotland (100%), 2. Northern Ireland (100%), 3. Leeds (97%), 4. South Wales (96%), 5. Evelina London (94%), 6. Manchester (87%), 7. Newcastle (84%), 8. South West (83%)</p> <p>Negative: 1. West Midlands (1%), 2. Trent (2%), 3. Spire (5%), 4. North Thames (24%), 5. Liverpool (44%)</p>

Figure 3.4. Funnel plot showing the percentage of CRANE-consented children, born 2020 to 2022, with birthweight data reported, according to Cleft Service.



Note: Funnel plot centred on the revised national percentage (57.8%) of children (born 2020-2022) with birthweight data reported.

¹⁷ 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.

Outcome: Birthweight	
Definitions	<ul style="list-style-type: none"> • Low birthweight (LBW) is defined as <2,500g • Healthy birthweight is defined as 2,500g-3,999g • High birthweight (HBW) is defined as ≥4,000g
Benchmarks	Among babies born in 2021 in the general population of England & Wales, 6% had a low birthweight (LBW). Among those born at term, the rate was 3% ¹⁸ .
Denominator	1,125 CRANE-consented children with birthweight reported. Of these, 973 were known to be born at term (≥37 weeks).
What did we find? 	<ul style="list-style-type: none"> • The mean birthweight was 3,213g (95% CI 3,175-3,250) among all children with birthweight reported. • Mean birthweight varied according to cleft type. Compared to children with CL (birthweight 3,315g), children with CP (3,174g, p=0.004) and BCLP (3,058g, p<0.001) had significantly lower birthweights, while those with UCLP (3,221g, p=0.098), CL+SMCP (3,504g, p=0.442) and SMCP alone (3,326g, p=0.941) had similar birthweights. • 12% of all children with birthweight reported had a LBW (Cleft Service range: 8%-24%¹⁹, p=0.343). The rate was 5% among those born at term (Cleft Service range: 3%-9%¹⁹, p=0.594). These rates are higher than those reported for the general population in 2021. • The percentage of children born at term with LBW did not differ significantly between cleft types (p=0.115) or sex (p=0.343). • It should be noted that the birthweight recorded in CRANE may not be representative of all babies born with a cleft lip and/or palate as almost half of all consented children were missing information on this and/or gestational age.

Recommendations: Patient characteristics
<ul style="list-style-type: none"> • The wide range in reported rates of RS between Cleft Services suggests that some are under-reporting this birth defect. It is recommended that services ensure RS status is accurately reported for all children with a CP. • 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. 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. • 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. • CRANE will engage with the Royal College of Midwives (RCM) and Royal College of Obstetricians and Gynaecologists (RCOG) to communicate findings.

¹⁸ Birth characteristics in England and Wales: 2021. Available at: <https://www.ons.gov.uk/peoplepopulationandcommunity/birthsdeathsandmarriages/livebirths/datasets/birthcharacteristicsinenglandandwales> [Last accessed: 19/11/2023]

¹⁹ Among Cleft Services that submitted data for ≥10 cases.

3.2 Timing of diagnosis

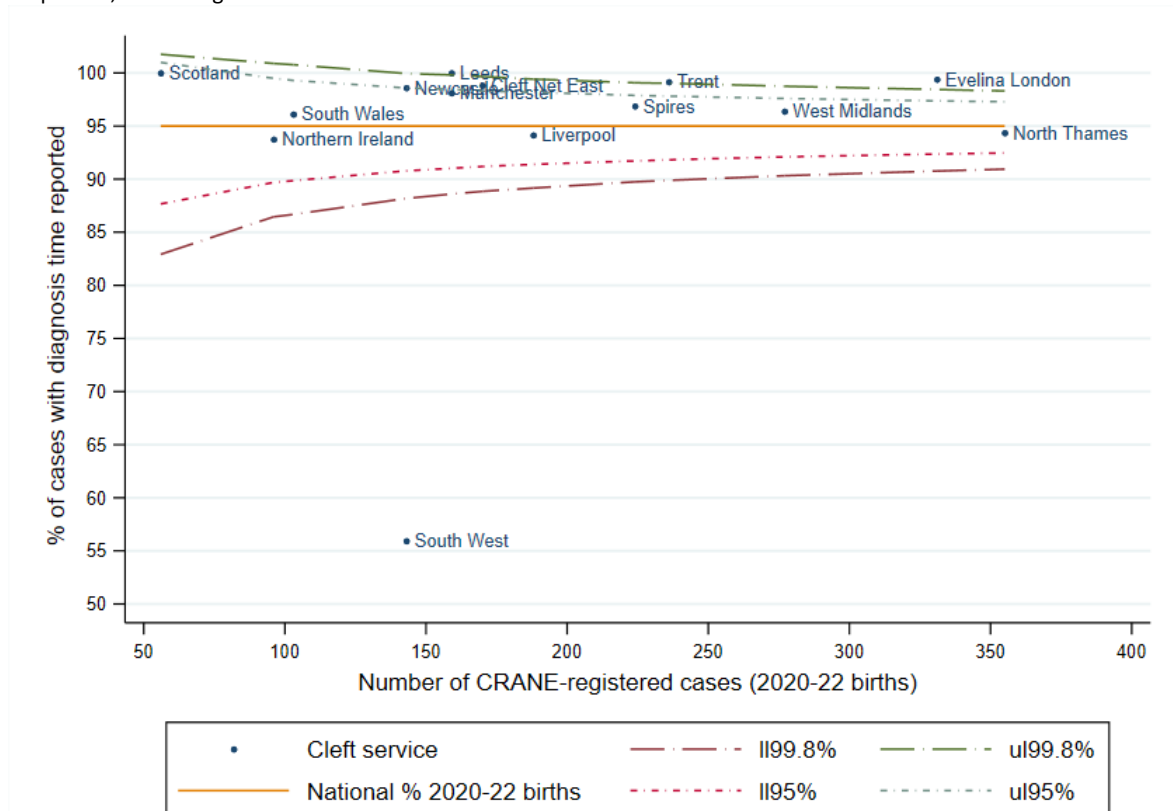
A cleft involving the lip is usually identified during a routine ultrasound scan at around 20 weeks' gestation. Clefts affecting only the palate can be difficult to detect by ultrasound and are usually diagnosed immediately after birth or during the newborn physical examination, performed within 72 hours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

Cohort summary	
Data source	The CRANE Database. Extract taken: 4 July 2023
Birth years	Three years: 2020 to 2022
Countries	England, Wales, Northern Ireland and Scotland
Inclusions	CRANE-registered children
Exclusions	None
Notes	<ul style="list-style-type: none">• Not subject to outlier policy but funnel plots provided to demonstrate variation in rates across services.• Data are not risk adjusted.
Raw data	'Diagnosis times' & 'Diagnosis times CPO' in the supplementary tables .

Data completeness: Diagnosis time	
Denominator	2,640 CRANE-registered children
What did we find?	<ul style="list-style-type: none"> 95% of CRANE-registered children had diagnosis time reported (Cleft Service range: 56%-100%, $p < 0.001$). This rate remains unchanged compared to the previous reporting period (births 2018-2020). The reporting of diagnosis time varied significantly according to birth year within the current cohort (2020: 92%, 2021: 96%, 2022: 97%, $p < 0.001$). Although reporting did not vary significantly according to known cleft type, only 118 out of 181 (65%) children with an 'unspecified' cleft type had diagnosis time reported.
Outliers	Positive: 1. Evelina London (99%) Negative: 1. South West (56%)



Figure 3.5. Funnel plot showing the percentage of all CRANE-registered children, born 2020 to 2022, who had diagnosis time reported, according to Cleft Service.



Note: Funnel plot centred on the national percentage (95.0%) of children (born 2020-2022) with diagnosis time reported.


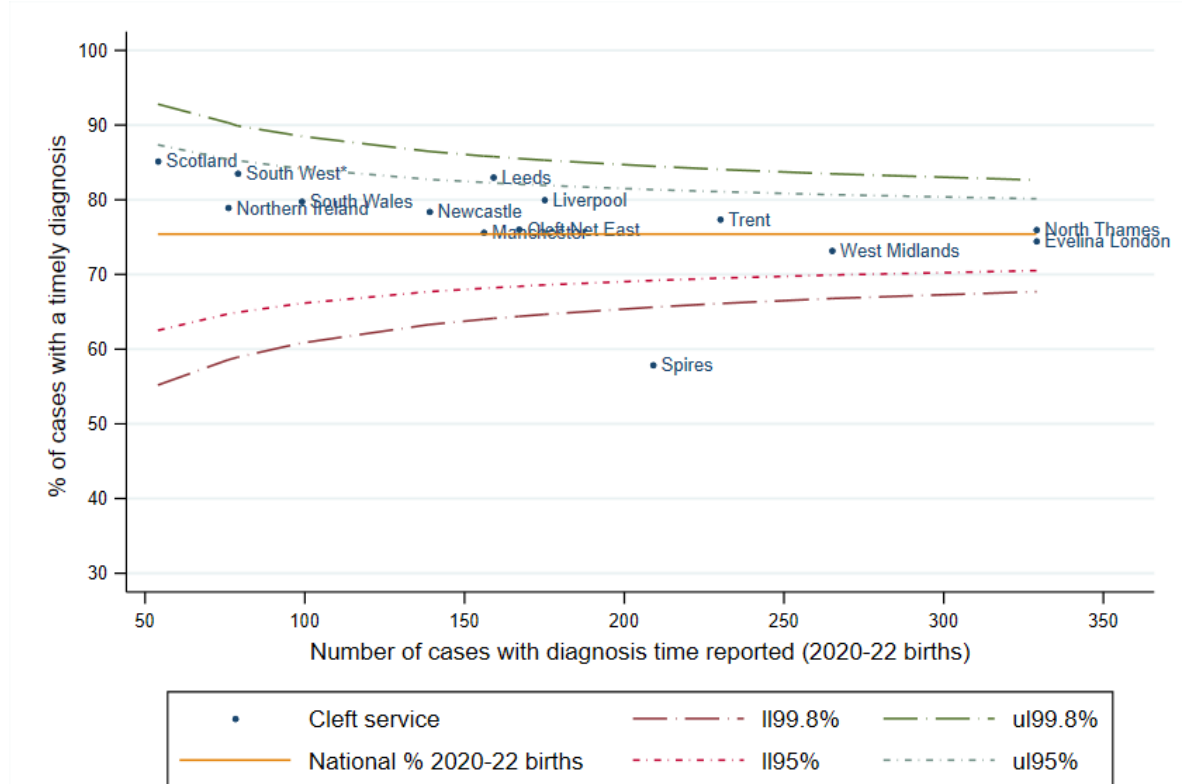
Outcome: Timing of diagnosis among all children with a cleft	
Indicators	#3 - Antenatal diagnosis for CL, UCLP and BCLP #4 - Timely detection of Cleft Palate (CP), within 24/72 hours from birth
Benchmark	Children with a cleft should have a timely diagnosis. This is antenatally for clefts involving the lip and before or at birth (<24 hours) for clefts affecting only the palate.
Notes	<ul style="list-style-type: none"> • 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 (South West). • Outcome data reflect care provided by maternity services referring on to Cleft Services. • Children with a submucous cleft palate (SMCP) alone are reported separately²⁰.
Denominator	2,466 CRANE-registered children with diagnosis time reported
What did we find?	 <ul style="list-style-type: none"> • 76% of all children with a cleft (excluding SMCP alone) received a timely diagnosis (antenatal for clefts involving the lip and antenatal or birth for clefts involving only the palate) (Cleft Service range: 58%-85%, $p < 0.001$). • The rate of timely diagnosis did not vary significantly according to birth year ($p = 0.925$).
Outliers	Positive: None Negative: 1. Spire (58%)

Figure 3.6. Funnel plot showing the percentage of all CRANE-registered children, born 2020 to 2022, who had a timely diagnosis, according to Cleft Service.



Note: Funnel plot centred on the revised national percentage (75.4%) of children (born 2020-2022) with a timely diagnosis.

²⁰ Diagnosis times are typically delayed for children with a submucous cleft palate alone, so these cleft types are reported separately from other clefts.


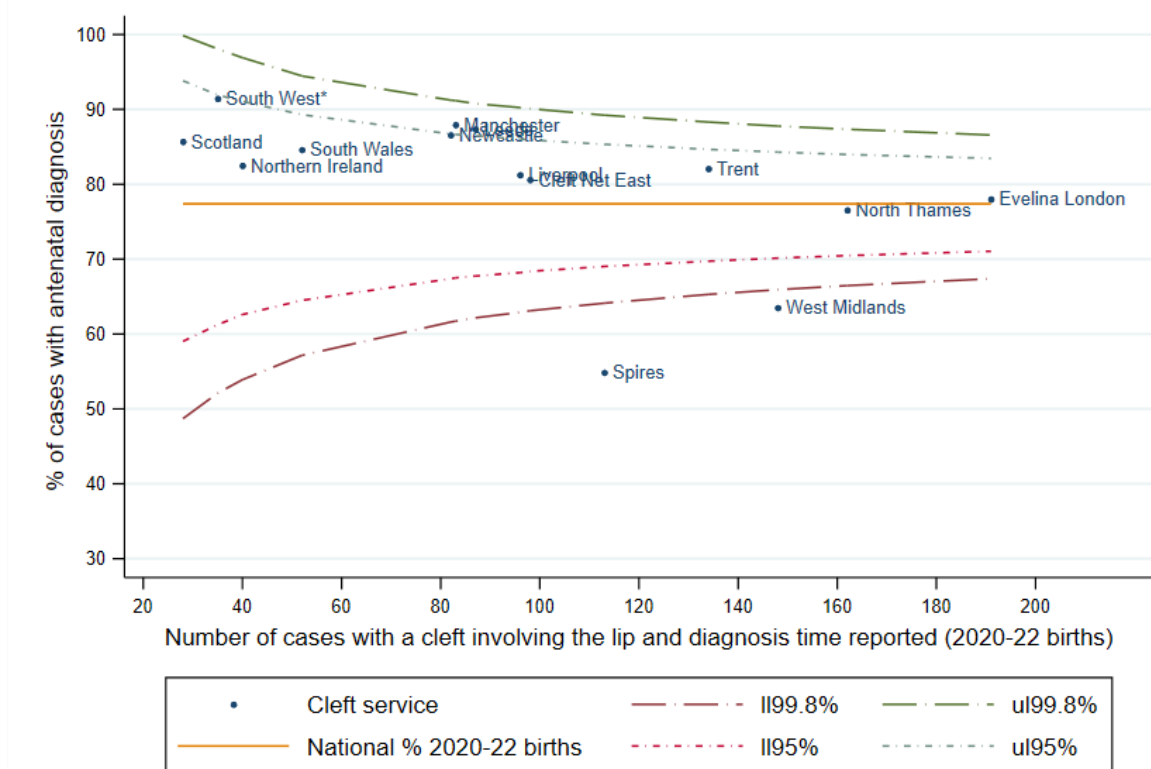
Outcome: Timing of diagnosis among all children with a cleft affecting the lip (CL, UCLP and BCLP)	
Indicator	#3 - Antenatal diagnosis for CL, UCLP and BCLP
Benchmark	Clefts involving the lip should be diagnosed antenatally. This was the case for 80% of children born 2018-2020.
Notes	<ul style="list-style-type: none"> • 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 (South West). • Outcome data reflect care provided by maternity services referring on to Cleft Services.
Denominator	1,349 CRANE-registered children with a cleft affecting the lip and diagnosis time reported.
What did we find?	 <ul style="list-style-type: none"> • 78% of children with a cleft affecting the lip were diagnosed antenatally (Cleft Service range: 55%-91²¹%, p<0.001). • Antenatal diagnosis rates varied significantly according to cleft type: 68% for CL, 75% for CL+SMCP, 84% for UCLP, and 88% for BCLP (p<0.001).
Outliers	Positive: None Negative: 1. Spires (55%), 2. West Midlands (64%)

Figure 3.7. Funnel plot showing the percentage of CRANE-registered children with a cleft affecting the lip, born 2020 to 2022, who had a timely diagnosis, according to Cleft Service.

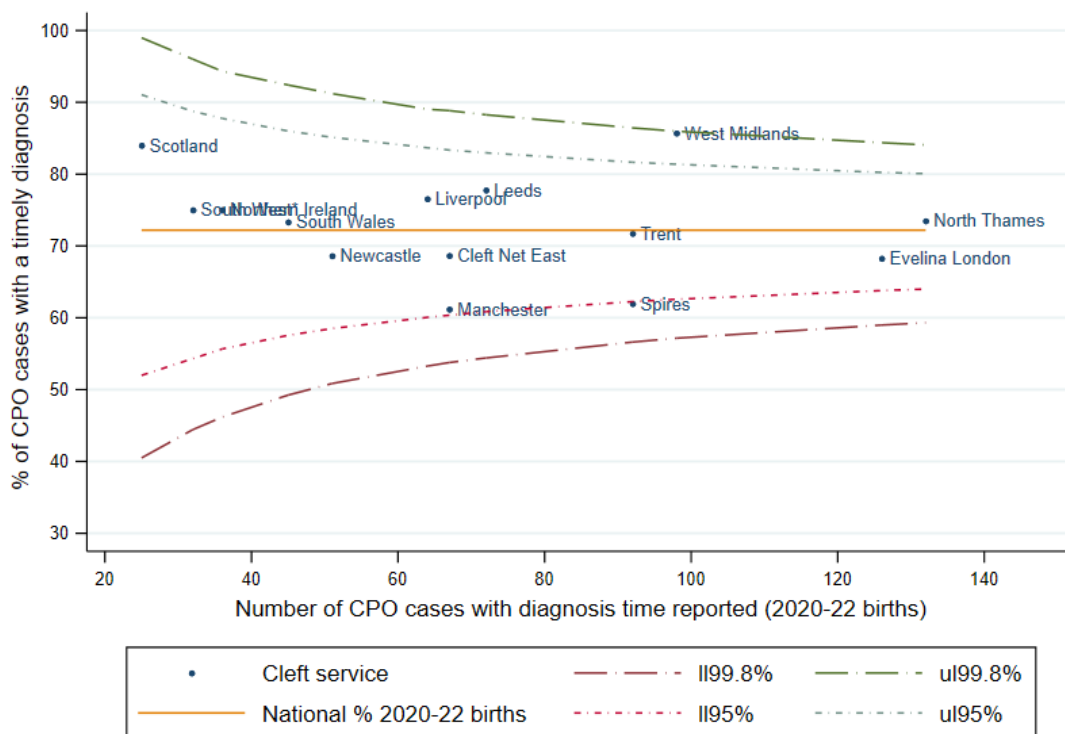


Note: Funnel plot centred on the revised national percentage (77.4%) of children (born 2020-2022) with an antenatal diagnosis.

²¹ Note that South West (91%) were a negative outlier for data completion so interpret this rate with caution. Second highest rate was 88% (Manchester).

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 at birth. This was the case for 74% of children born 2018-2020. This increased to 86% when including diagnoses within 72 hours of birth.
Notes	<ul style="list-style-type: none"> 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 (South West). Outcome data reflect care provided by maternity services referring on to Cleft Services.
Denominator	<ul style="list-style-type: none"> 999 Children with a cleft affecting the palate alone (excluding SMCP). 42 children with SMCP alone.
What did we find?	<ul style="list-style-type: none"> 72% 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%-86%, p=0.037). This proportion increased to 84% when including diagnoses within 72 hours of birth (Cleft Service range: 78%-92%, p=0.731). 33% of children with a SMCP alone were diagnosed before or within 24 hours of birth. 12% were diagnosed 2-7 days after birth, 12% 7-28 days after birth, 19% 1-6 months after birth, and 24% >6 months after birth.
Outliers	None

Figure 3.8. Funnel plot showing the percentage of CRANE-registered children with a cleft affecting the palate alone (excluding SMCP), born 2020 to 2022, who were diagnosed before or within 24 hours of birth, according to Cleft Service.



Note: Funnel plot centred on the revised national percentage (72.2%) of children (born 2020-22) diagnosed antenatally or <24 hours after birth.

Recommendations: Timing of diagnosis
<ul style="list-style-type: none"> 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 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.

3.3. Referral to and contact with Cleft Services

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

Cohort summary for referral and contact time	
Data source	The CRANE Database. Extract taken: 4 July 2023
Birth years	Three years: 2020 to 2022
Countries	England, Wales, Northern Ireland and Scotland
Inclusions	CRANE-registered children
Exclusions	Children who died before reaching 1 week of age ²²
Notes	<ul style="list-style-type: none">• Not subject to outlier policy but funnel plot provided to demonstrate variation in rates across services.• Data are not risk-adjusted.
Raw data	'Referral' and 'contact' in the supplementary tables .

²² Children dying within the first 7 days of life within this cohort: 0

3.3.1 Referral


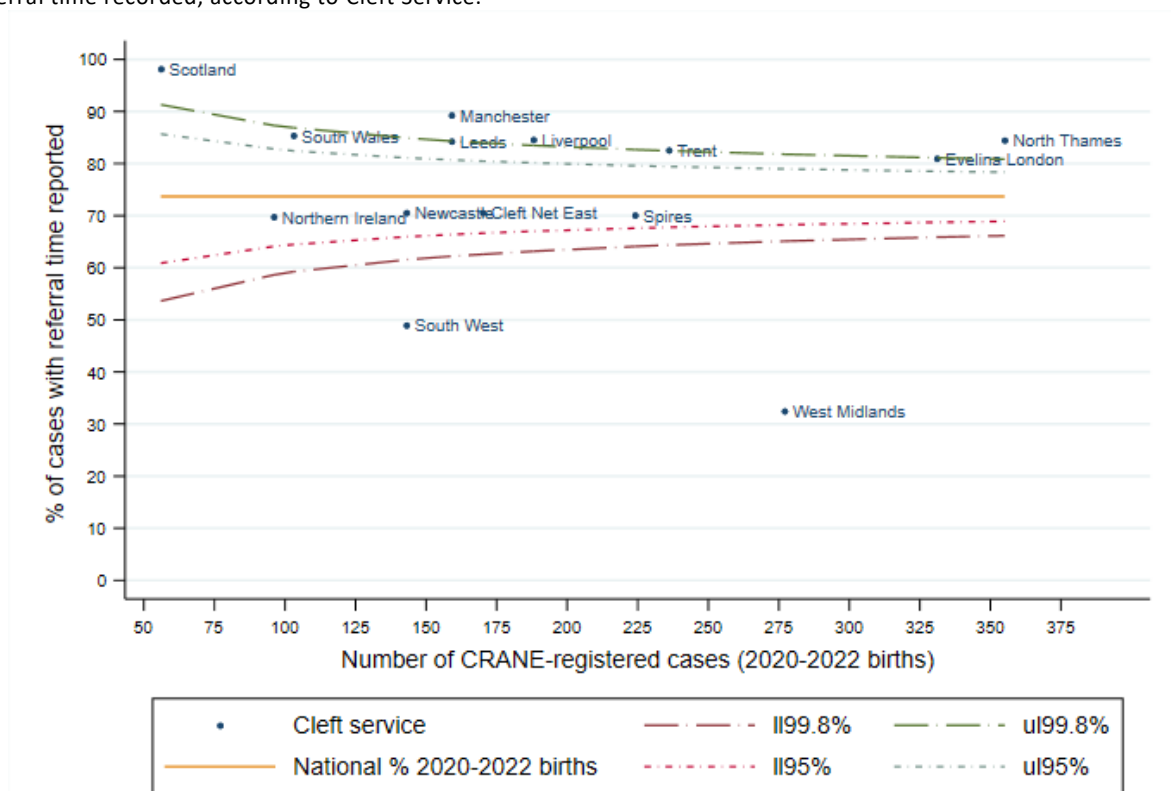
Data completeness: Referral time	
Indicator	#5 - Referral recorded for all eligible children
Denominator	2,640 CRANE-registered children
What did we find?	 <ul style="list-style-type: none"> 74% of all CRANE-registered children had a recorded referral time (Cleft Service range: 33%-98%, $p < 0.001$). This compares to 67% of those born 2018-2020. The reporting of referral time varied significantly according to birth year within the current cohort (2020: 67%, 2021: 77%, 2022: 78%, $p < 0.001$). The percentage of children with a recorded referral time varied significantly according to cleft type (range: 35% in those with unspecified cleft type-92% in those with LC+SMCP, $p < 0.001$).
Outliers	Positive: 1. Scotland (98%), 2. Manchester (89%), 3. North Thames (85%), 4. Liverpool (85%), 5. Trent (83%) Negative: 1. West Midlands (33%), 2. South West (49%)

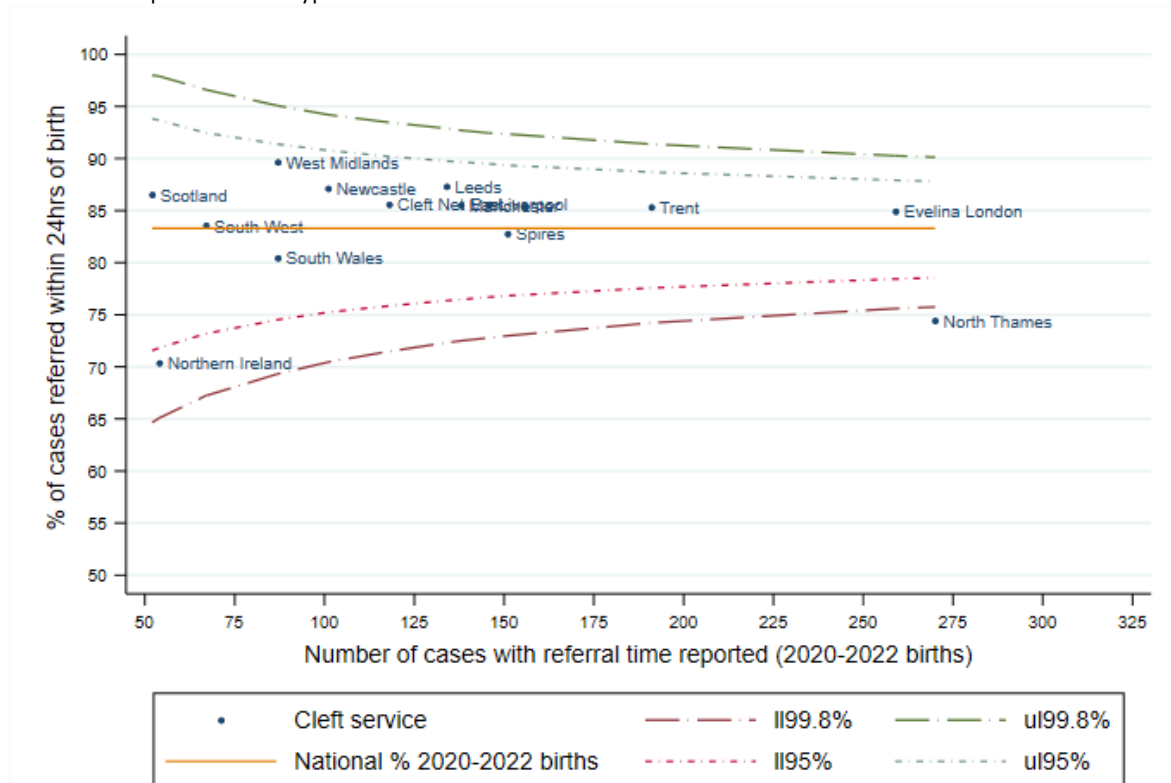
Figure 3.9. Funnel plot showing the percentage of CRANE-consented children, born 2020 to 2022, who had data on referral time recorded, according to Cleft Service.



Note: Funnel plot centred on the revised national percentage (73.7%) of children (born 2020-2022) with referral time reported.

Outcome: Referred to Cleft Service within 24 hours of birth	
Indicator	#6 - Referral to a cleft care team within 24 hours of birth
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.
Benchmarks	84% of children born with a cleft in 2018-2020 were referred to Cleft Services within 24 hours of birth.
Denominator	1,907 CRANE-registered cases with referral time recorded
What did we find?	<ul style="list-style-type: none"> 82% of children were referred to a Cleft Service within 24hrs of birth (Cleft Service range: 60%-89%, $p < 0.001$). This represents a 2% decrease on the previous reporting period (2018-2020 births). Children diagnosed with an SMCP alone had the lowest rate of referrals within 24hrs of birth (21%), followed by unspecified cleft types (65%) and then CP (68%). >90% of clefts involving the lip were referred within 24hrs of birth. These findings are consistent with later diagnosis times for children with SMCP, unspecified cleft types and CP. 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: 70%-90%, $p = 0.004$).
Outliers	Positive: None Negative: 1. North Thames (74%)

Figure 3.10. Funnel plot showing the percentage of CRANE-consented children, born 2020 to 2022, who had been referred to a Cleft Service within 24hrs after birth, according to Cleft Service. Excludes children with an SMCP alone and those with an unspecified cleft type.



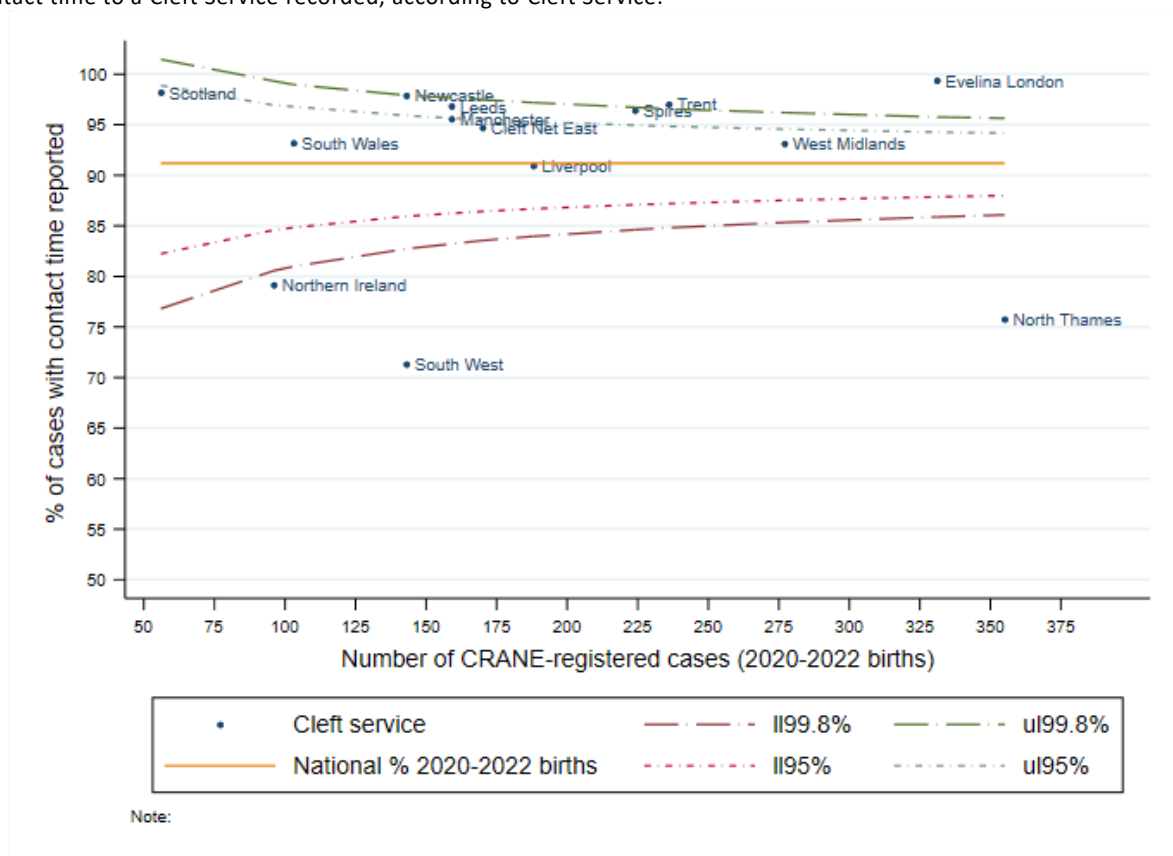
Note: Funnel plot centred on the revised national percentage (83.0%) of children (born 2020-2022) referred within 24hrs of birth.

3.3.2 Contact

Data completeness: Contact time between Cleft Service and family after referral	
Indicator	#7 - Contact recorded for all eligible children
Denominator	2,460 CRANE-registered children
What did we find?	<ul style="list-style-type: none"> 91% of all CRANE-registered children had a recorded contact time (Cleft Service range: 71%-99%, $p < 0.001$). This compares to 86% of those born 2018-2020. The reporting of contact time varied significantly according to birth year within the current cohort (2020: 86%, 2021: 92%, 2022: 96%, $p < 0.001$). Reporting of contact time did not vary significantly according to specified cleft types ($p = 0.231$); however, contact time was only reported for 64% for those with an unspecified cleft type.
Outliers	<p>Positive: 1. Evelina London (99%), 2. Trent (97%)</p> <p>Negative: 1. South West (71%), 2. North Thames (76%), 3. Northern Ireland (79%)</p>



Figure 3.11. Funnel plot showing the percentage of CRANE-consented children, born 2020 to 2022, who had data on contact time to a Cleft Service recorded, according to Cleft Service.



Note:

Note: Funnel plot centred on the revised national percentage (91.2%) of children (born 2020-2022) with contact time reported.


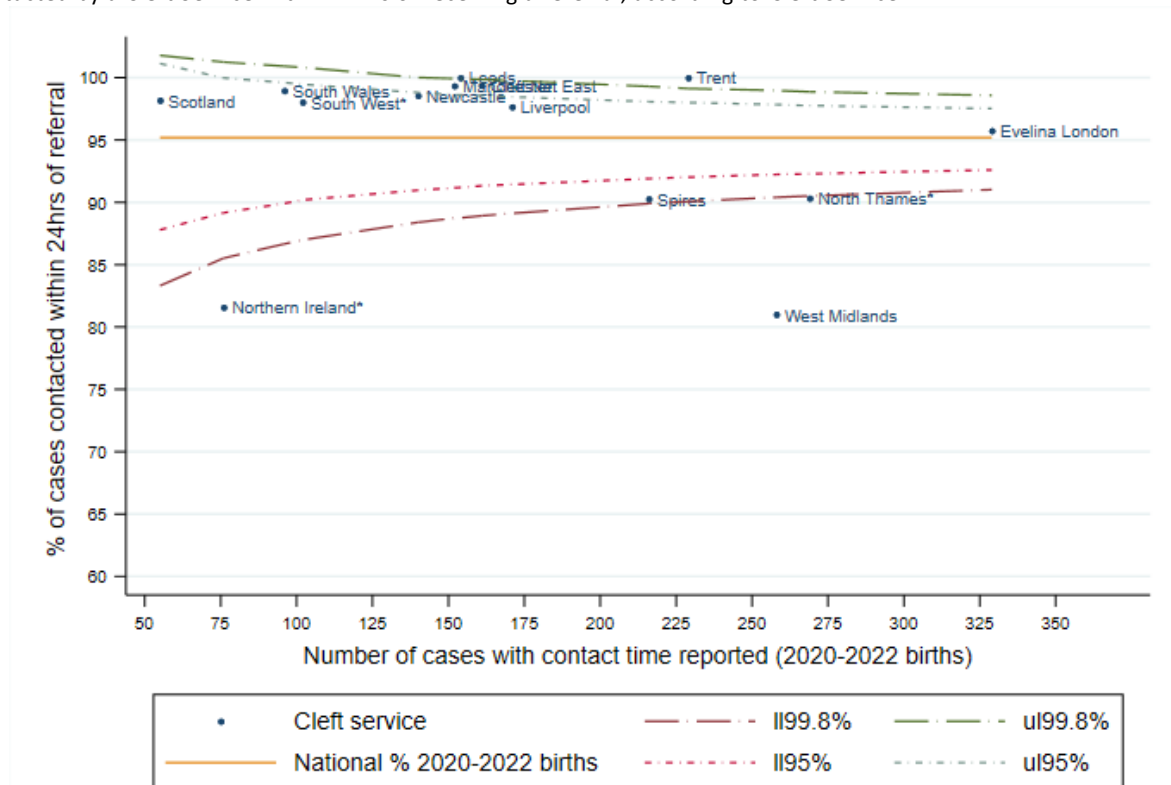
Outcome: Contacted family within 24 hours of referral	
Indicator	#8 - Contact with a cleft care team within 24 hours of referral
Benchmarks	94% of children born with a cleft in 2018-2020 were contacted by their Cleft Service within 24 hours of the service receiving their referral.
Denominator	2,408 CRANE-registered cases with contact time reported
What did we find?	 <ul style="list-style-type: none"> 94% of children were contacted by Cleft Services within 24hrs of the service receiving a referral (Cleft Service range: 81%²³-100%, p<0.001). This is consistent with the previous reporting period (2018-2020 births). The percentage of families receiving contact within 24hrs of referral varied by cleft type (p<0.001). Children with an SMCP alone had the lowest rate (74%), followed by CP (93%). >95% of other cleft types were contacted within 24hrs of referral.
Referral outliers	Positive: 1. Leeds (100%), 2. Trent (100%) Negative: 1. West Midlands (81%), 2. Northern Ireland (82%), 3. North Thames (90%)

Figure 3.12. Funnel plot showing the percentage of CRANE-consented children, born 2020 to 2022, who had been contacted by a Cleft Service within 24hrs of receiving a referral, according to Cleft Service.



Note: Funnel plot centred on the revised national percentage (95.2%) of children (born 2020-2022) contacted within 24hrs of referral.

Recommendations: Referral and contact
<ul style="list-style-type: none"> 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. Regional variation in the percentage of children referred and contacted within 24 hours demonstrates that some Cleft Services have high levels of referrals and contacts within 24 hours. They should share their best practice recommendations with Cleft Services with lower rates.


²³Note that West Midlands (81%) were a negative outlier for consent verification so interpret this rate with caution. The lowest rate among services not identified as negative outliers for consent or data completion was 90% (Spires).

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. As a key performance indicator for Cleft Services, a child's consent status should be recorded for all registrations in the CRANE Database. This section reports the consent status of children born 2020-2022, reflecting recent registrations, and 2014-2016, reflecting registrations of children who should have undergone their 5-year-old assessment of cleft-related outcomes. Consent verification denotes a confirmed consent status, whereby families have either given informed consent or declined consent for CRANE to collect outcome data. Consent verification rates are reported according to Cleft Service. In accordance with our Outlier Policy²⁴, data from any service identified as a negative outlier (where their rate falls below the expected range) for consent verification will be excluded from revised totals and averages used to generate funnel plots reported in the 5-year outcome section of this report. This ensures that the audit results reflect patient populations only from services with acceptable levels of consent verification.

4.1. CRANE consent, 2020-2022 births

Cohort summary	
Data source	The CRANE Database. Extract taken: 4 July 2023
Birth years	Three years: 2020 to 2022
Countries	England, Wales, Northern Ireland and Scotland
Inclusions	CRANE-registered children
Exclusions	<ul style="list-style-type: none"> Children who died Children with submucous cleft palate²⁵
Notes	<ul style="list-style-type: none"> Not subject to outlier policy. Data are not risk-adjusted.
Raw data	'Consent' in the supplementary tables .

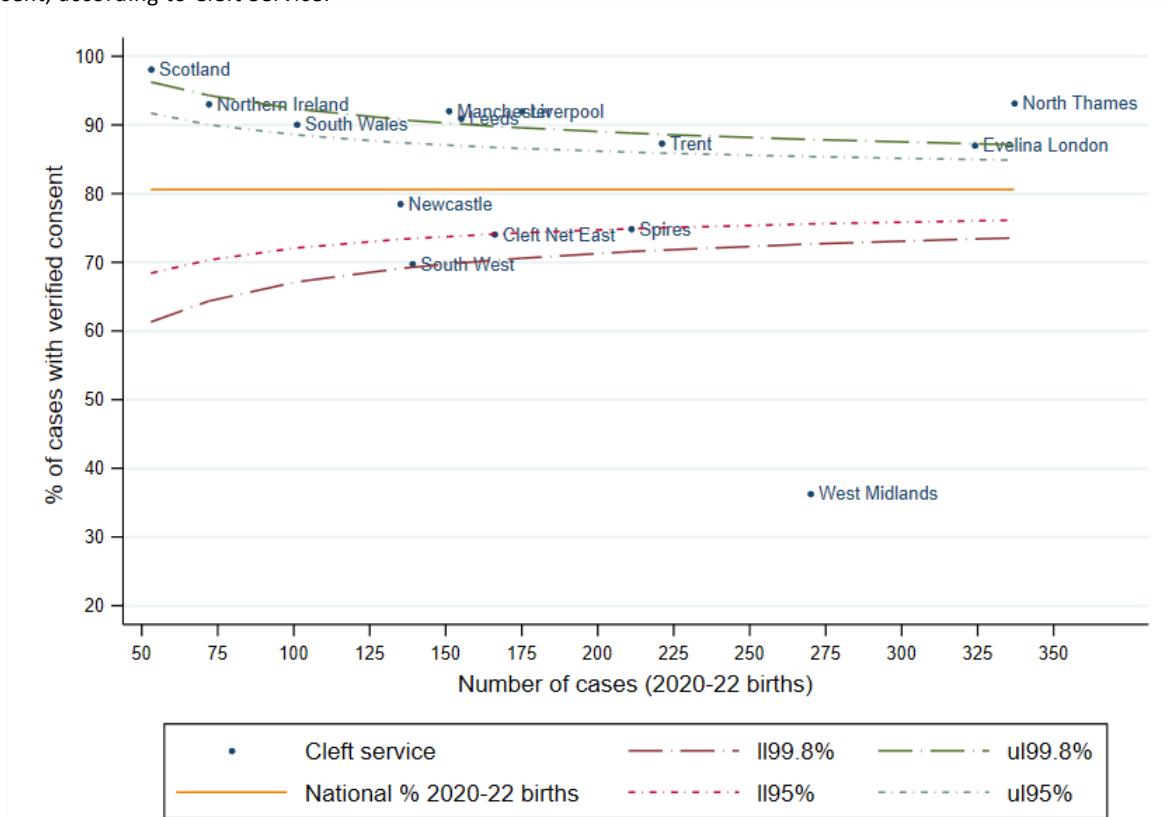
Outcome: Consent status	
Benchmarks	100% of families of children with a cleft should be approached for consent verification (provide informed consent or decline) regarding CRANE outcome data collection.
Indicator	#9 - Consent verification undergone with all families of children with a cleft lip and/or palate recorded in the CRANE Database. Full details in the supplementary tables .
Denominator	2,510 CRANE-registered children
What did we find? 	<ul style="list-style-type: none"> 81% of eligible children had a verified consent status (Cleft Service range: 36%-98%, p<0.001). This rate varied significantly according to birth year (2020: 76%, 2021: 84%, 2022: 82%, p<0.001); 79% had provided informed consent to outcome data collection in CRANE; 2% declined consent; 19% were awaiting consent verification; 1% were not possible to verify. Of those with consent, 97% also consented to linkage of CRANE data with health and education data.

²⁴ CRANE Outlier Policy. https://www.crane-database.org.uk/content/uploads/2022/12/CRANE-Outlier-Policy_20Mar2023.pdf

²⁵ Patients with submucous cleft palate are excluded from outcomes, including cases with a LAHSAL code identified as implausible, and likely submucous, when reviewed by cleft clinicians.

Outliers	Positive: 1. Scotland (98%), 2. North Thames (93%), 3. Liverpool (92%), 4. Manchester (92%), 5. Leeds (91%) Negative: 1. West Midlands (36%)
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Figure 4.1. Funnel plot showing the percentage of CRANE-registered children, born 2020 to 2022, who had verified consent, according to Cleft Service.



4.2. CRANE consent, 2014-2016 births

Cohort summary	
Data source	The CRANE Database. Extract taken: 4 July 2023
Birth years	Three years: 2014 to 2016
Countries	England, Wales and Northern Ireland.
Inclusions	CRANE-registered children
Exclusions	<ul style="list-style-type: none"> Children who died before the age of 5 years Children with submucous cleft palate²⁶
Notes	<ul style="list-style-type: none"> Subject to outlier policy. Data are not risk-adjusted. Scotland joined CRANE in 2023 and therefore had insufficient data for 2014-2016 births to be included in analyses.
Raw data	'Consent' in the supplementary tables .

²⁶ Patients with submucous cleft palate are excluded from outcomes, including cases with a LAHSAL code identified as implausible, and likely submucous, when reviewed by cleft clinicians.


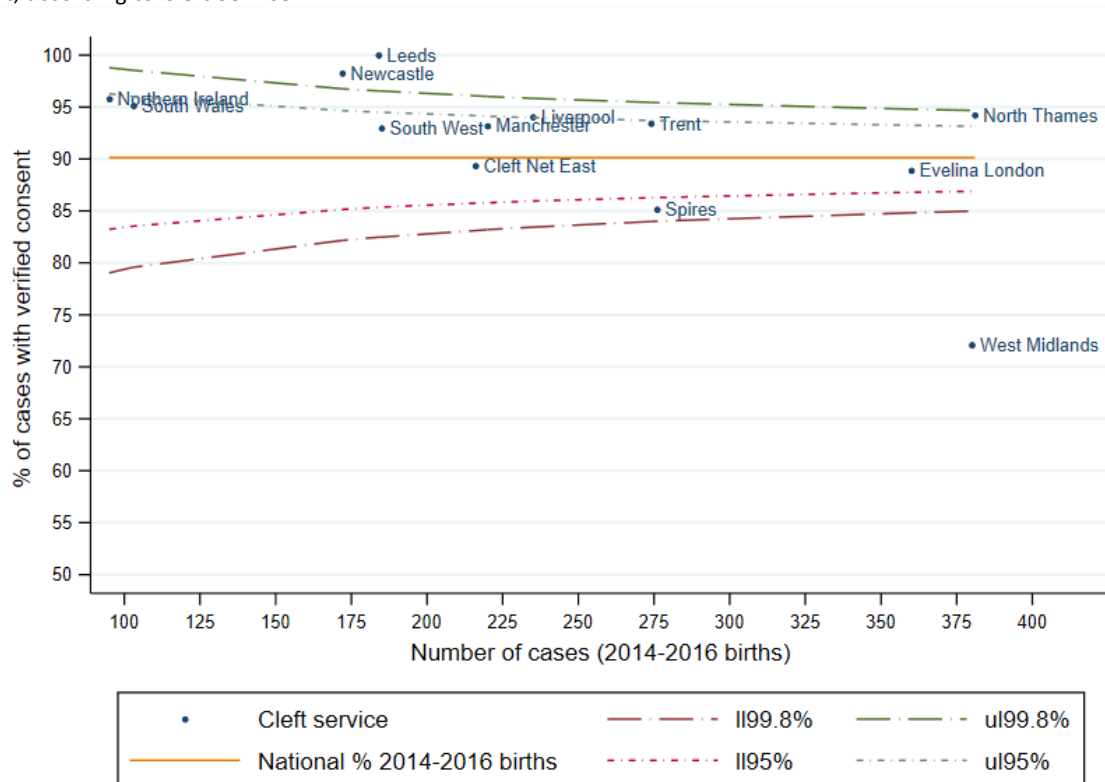
Outcome: Consent status	
Benchmarks	<ul style="list-style-type: none"> 100% of families of children with a cleft should be approached for consent verification (provide informed consent or decline) regarding CRANE outcome data collection. 90% of 5-year-old children born 2012-2014 had a verified consent status.
Indicator	#9 - Consent verification undergone with all families of children with a cleft lip and/or palate recorded in the CRANE Database. Full details in the supplementary tables .
Denominator	<ul style="list-style-type: none"> 3,081 CRANE-registered children
What did we find?	 <ul style="list-style-type: none"> 90% of eligible children had a verified consent status (Cleft Service range: 72%-100%, $p < 0.001$). This rate remains unchanged compared to the previous reporting period (2012-2014 births), but varied significantly according to birth year within the current cohort (2014: 92%, 2015: 90%, 2016: 88%, $p = 0.01$). 88% had provided informed consent to outcome data collection in CRANE. 2% declined consent. 9% were awaiting consent verification. 1% were not possible to verify.
Outliers	Positive: 1. Leeds (100%), 2. Newcastle (98%) Negative: 1. West Midlands (72%)

Figure 4.2. Funnel plot showing the percentage of CRANE-registered 5-year-olds, born 2014 to 2016, who had verified consent, according to Cleft Service.

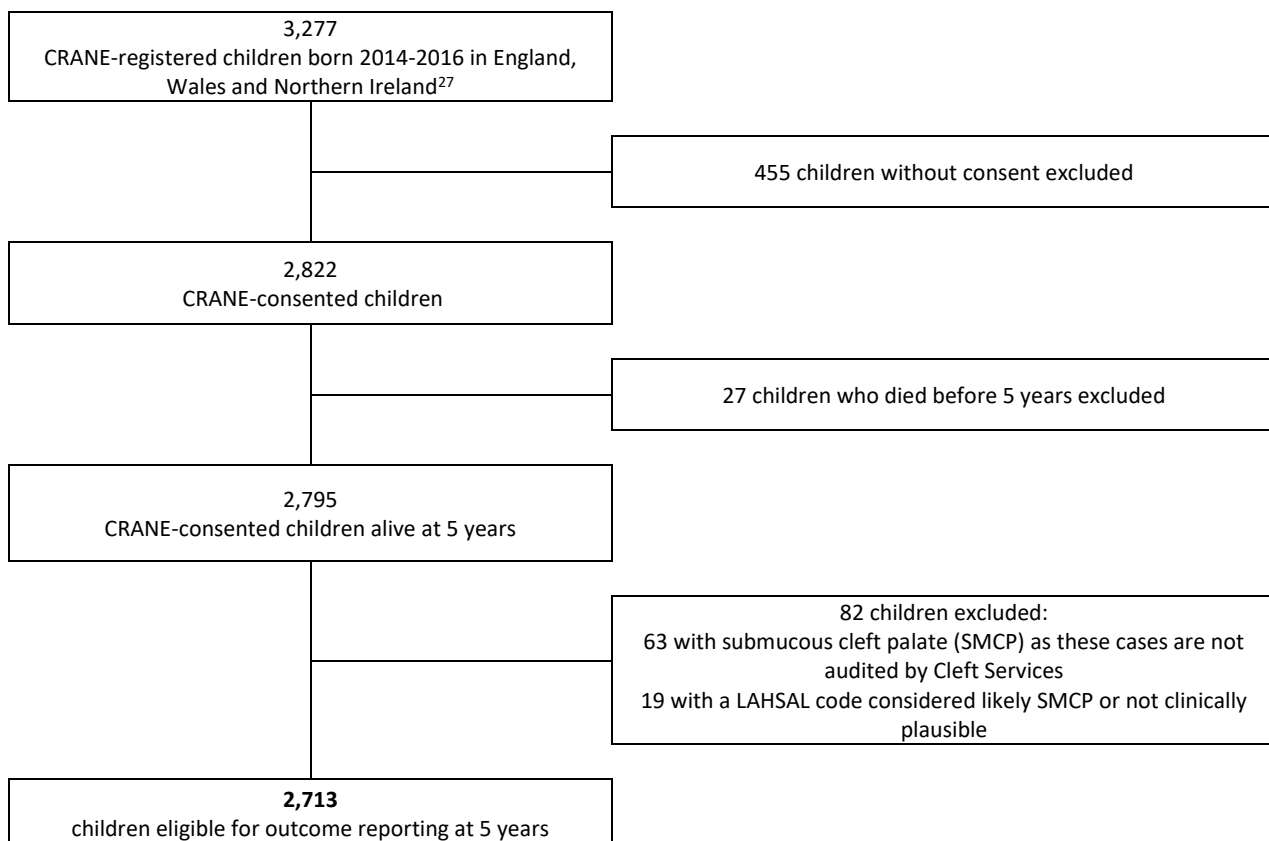


Recommendations: Consent
<ul style="list-style-type: none"> Cleft Services should review their procedures to identify reasons for low rates of verified consent, and ensure that CRANE consent status is recorded for every child with a cleft. CRANE will continue to work with Cleft Services and the Nursing Clinical Excellence Network (CEN) to improve consent status verification.

5. Audit outcomes at 5 years of age

This chapter presents cleft-related 5- year outcomes for CRANE-consented children, born 2014 to 2016. Figure 5.1 details the eligible cohort and the outcomes collected.

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



Outcome	CLO N=683 (25%)	CPO N=1,149 (42%)	UCLP N=587 (22%)	BCLP N=261 (10%)	Non-specified N=33 (1%)
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.

²⁷ Scotland joined CRANE in January 2023 and therefore have no eligible cases for outcome reporting.

5.1. Child growth

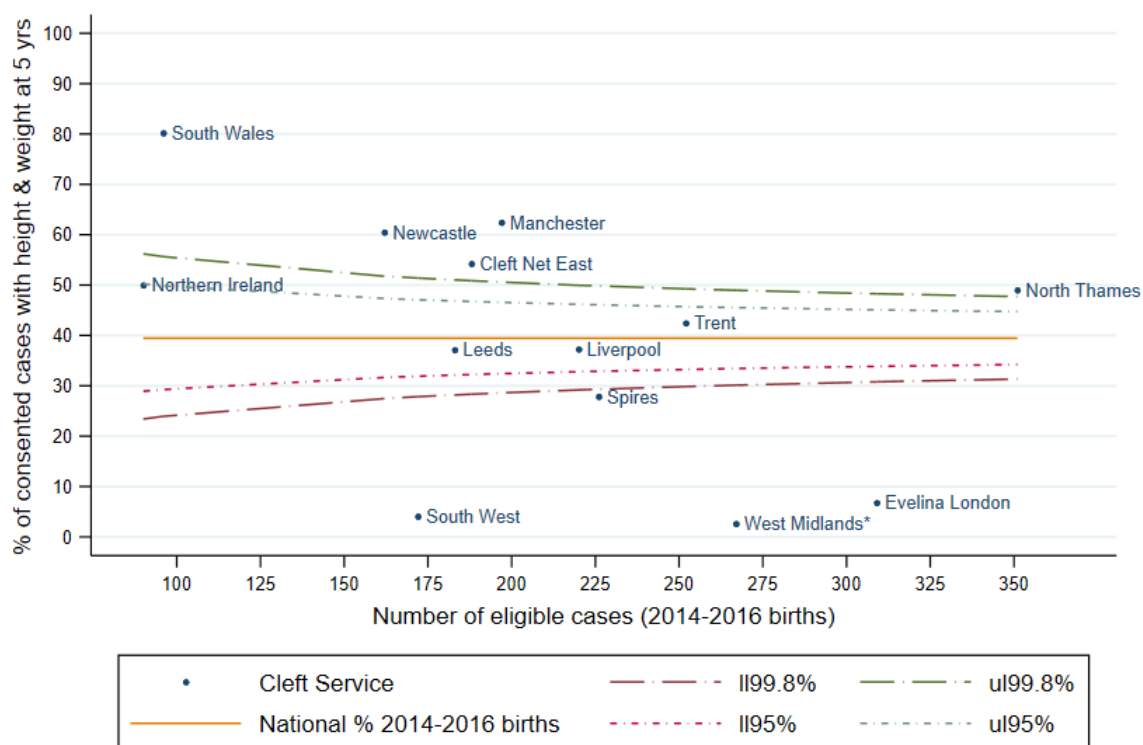
All Cleft Services are required to submit growth data (height and weight) for all consented 5-year-old children diagnosed with a cleft lip and/or palate. If growth data are not available, Cleft Services are asked to provide a reason for this (e.g. patient transferred out of area or patient did not attend appointment).

Cohort summary	
Data source	The CRANE Database (consented cases only). Extract taken: 4 July 2023
Birth years	Three years: 2014 to 2016
Countries	England, Wales and Northern Ireland
Inclusions	CRANE-consented children, including those without a specified cleft type
Exclusions	<ul style="list-style-type: none"> Children who died before the age of 5 years Children with submucous cleft palate²⁸
Notes	<ul style="list-style-type: none"> Subject to outlier policy. Data are not risk-adjusted. Funnel plots are centred on the revised national rates after excluding data from services identified as negative outliers for consent verification (West Midlands). Cleft Services with <10 cases are not shown on funnel plot due to insufficient data.
Raw data	'Child growth' in the supplementary tables .

Data completeness: Height and weight	
Indicator	#10 - Child growth at 5 years recorded for all eligible children.
Denominator	2,713 CRANE-consented children
What did we find? 	<ul style="list-style-type: none"> 36% of eligible consented children had a recorded height and weight (Cleft Service range: 3%-80%, $p < 0.001$). This compares to 46% of those born 2012-2014. <1% of children only had a recorded height. <1% of children only had a recorded weight. 21% had a reason the child growth outcomes were not collected. 42% were missing data or a reason for not collecting data. The proportion of children with height and weight measures varied significantly according to birth year (2014: 40%, 2015: 22%, 2016: 46%, $p < 0.001$).
Outliers	Positive: 1. South Wales (80%), 2. Manchester (62%), 3. Newcastle (61%), 4. Cleft Net East (54%), 5. North Thames (49%) Negative: 1. West Midlands (3%), 2. South West (4%), 3. Evelina London (7%), 4. Spire (28%)

²⁸ Patients with submucous cleft palate are excluded from all 5-year outcomes, including cases with a LAHSAL code identified as not possible, and likely submucous, when reviewed by cleft clinicians.

Figure 5.2. Funnel plot showing the percentage of CRANE-consented 5-year-olds, born 2014 to 2016, with growth data reported, according to Cleft Service.



Note: Funnel plot centred on the revised national percentage (39.5%) of children (born 2014-2016) with child growth outcomes reported.

Outcome: Healthy BMI	
Indicator	#11 - Healthy Body Mass Index (BMI) at 5 years of age.
Benchmarks	<ul style="list-style-type: none"> Prevalence of underweight, healthy weight, overweight and obesity among 5 year olds in the general population is estimated at 1%, 77%, 12% and 10%, respectively²⁹. 87% of eligible children born 2012-2014 were reported to have a healthy weight (CRANE, 2022).
Notes	<ul style="list-style-type: none"> Body mass index (BMI) at 5 years of age was calculated as weight (kg)/height (m)². For 5-year-olds in the UK³⁰, 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/m². Funnel plot is centred on the revised national rate after excluding data from services identified as negative outliers for consent verification (West Midlands) and child growth data completeness (West Midlands, Spires, Evelina London and South West).
Denominator	972 CRANE-consented children with a recorded height and weight

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

³⁰According 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]


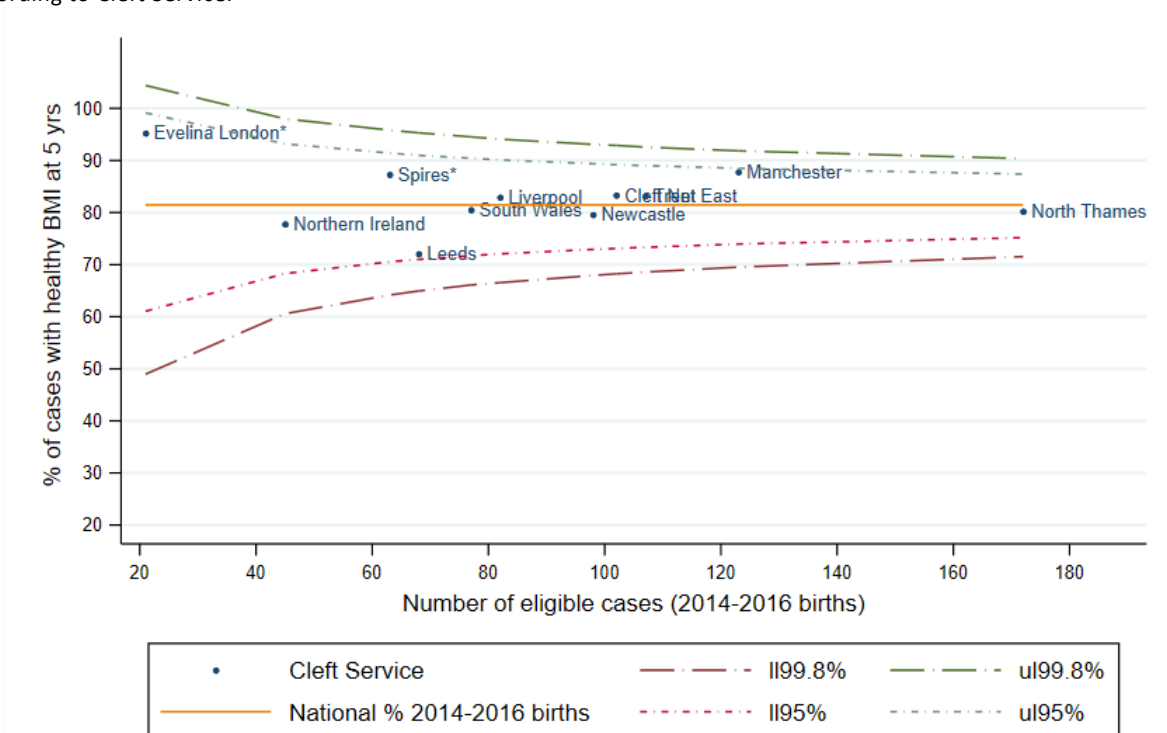
<p>What did we find?</p> 	<ul style="list-style-type: none"> • Average weight, height and BMI for those with reported data was 20.3kg (95%CI 20.1-20.5kg), 113.3cm (95%CI 113.0-113.7cm) and 15.7 kg/m² (95% CI: 15.6 kg/m² to 15.8 kg/m²), respectively. • 82% of children had a healthy BMI (Cleft Service range: 72%³¹ to 95%³², p=0.314). This represents a 5% decrease from the previous reporting period, but is still higher than the 77% reported for the general population. • 4% of children were underweight, 10% overweight and 5% obese. • BMI category distribution varied by sex p<0.001 (more females were underweight, more males were a healthy weight) but there was no significant difference according to known cleft type (p=0.099).
<p>Outliers</p>	<p>None.</p>

Figure 5.3. Funnel plot showing the percentage of CRANE-consented 5-year olds, born 2014 to 2016, with a healthy BMI, according to Cleft Service.



Note: Funnel plot centred on the revised national percentage (81.5%) of children (born 2014-2016) with a healthy BMI.

<p>Recommendations: Child growth</p>	
<ul style="list-style-type: none"> • 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. 	

³¹ among Cleft Services that submitted data for ≥10 cases.

³² Note that Evelina London (95%) were a negative outlier for 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 88% (Manchester).

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 cleft³³.

Cohort summary	
Data source	The CRANE Database (consented cases only). Extract taken: 4 July 2023
Birth years	Three years: 2014 to 2016
Countries	England, Wales and Northern Ireland
Inclusions	CRANE-consented children, including those without a specified cleft type
Exclusions	<ul style="list-style-type: none"> Children who died before the age of 5 years Children with submucous cleft palate³⁴
Notes	<ul style="list-style-type: none"> Subject to outlier policy. Data are not risk-adjusted. Cleft Services with <10 cases are not shown on funnel plot due to insufficient data. Funnel plots are centred on the revised national rates after excluding data from services identified as negative outliers for consent verification (West Midlands).
Raw data	'Dental health' 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 ³⁵ . 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 ³⁶ . Care indices with a value close to 1 (100%) indicate that there are high levels of care provided by fillings (not extraction or no treatment), which is the desired outcome. In instances where a tooth is very poorly formed, extraction may be the treatment of choice.

³³ (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.

³⁴ Patients with submucous cleft palate are excluded from all 5-year outcomes, including cases with a LAHSAL code identified as implausible, and likely submucous, when reviewed by cleft clinicians.

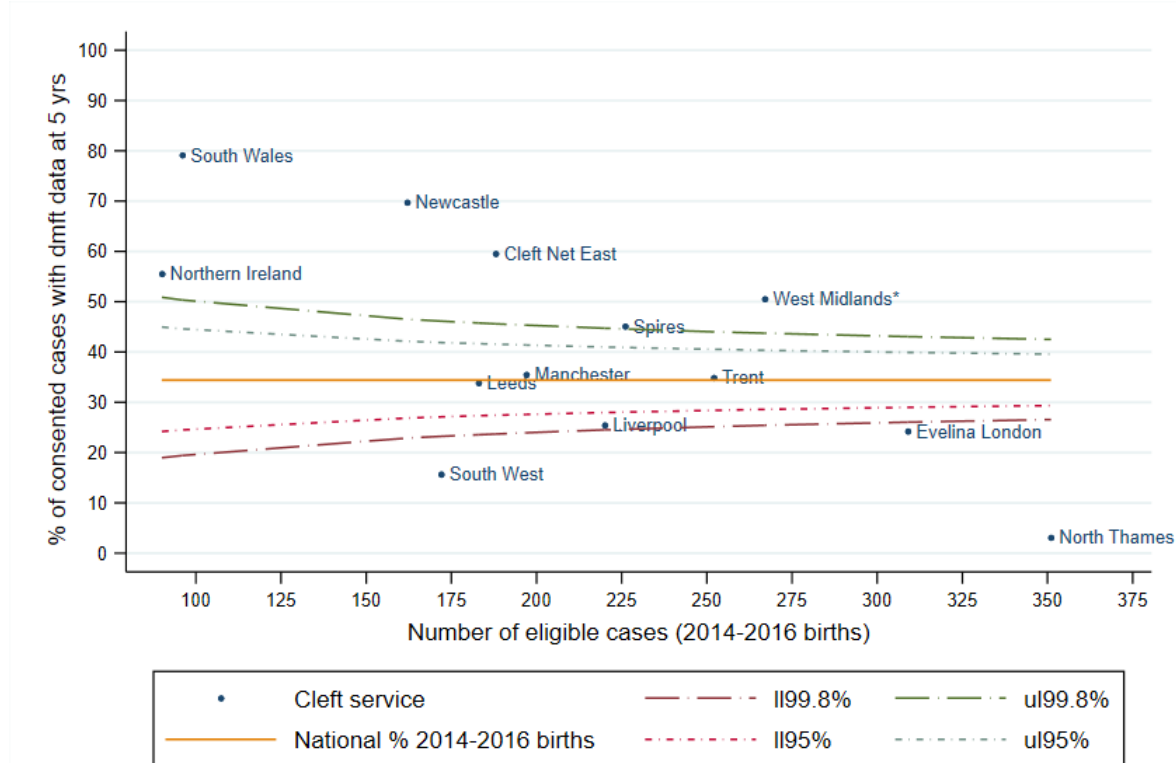
³⁵ If a dmft score for an individual is 0 then the treatment index is 1 (100%) as there is no untreated dental disease.

³⁶ 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	2,713 CRANE-consented children
What did we find?	<ul style="list-style-type: none"> 36% of eligible consented children had recorded dmft scores (Cleft Service range: 3%-79%, $p < 0.001$). This compares to 48% of those born 2012-2014. 27% had a reason³⁷ dmft scores were not collected. 37% were missing data or a reason for not collecting data. The proportion of children with dmft scores varied significantly according to birth year (2014: 35%, 2015: 31%, 2016: 42%, $p < 0.001$) and cleft type (CL: 67%, CP: 66%, UCLP: 58%, and BCLP: 61%, $p = 0.004$).
Outliers	<p>Positive: 1. South Wales (79%), 2. Newcastle (70%), 3. Cleft Net East (60%), 4. Northern Ireland (56%), 5. West Midlands (51%), 6. Spires (45%)</p> <p>Negative: 1. North Thames (3%), 2. South West (16%), 3. Evelina London (24%), 4. Liverpool (26%)*</p> <p>*Negative alert for 2 consecutive reporting periods</p>



Figure 5.4. Funnel plot showing the percentage of CRANE-consented 5-year-olds, born 2014 to 2016, with a cleft who had decayed, missing or filled teeth (dmft) scores reported, according to Cleft Service.



Note: Funnel plot centred on the revised national percentage (34.4%) of children (born 2014-2016) with dmft scores reported.

³⁷ 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.

5.2.1. Dental decay


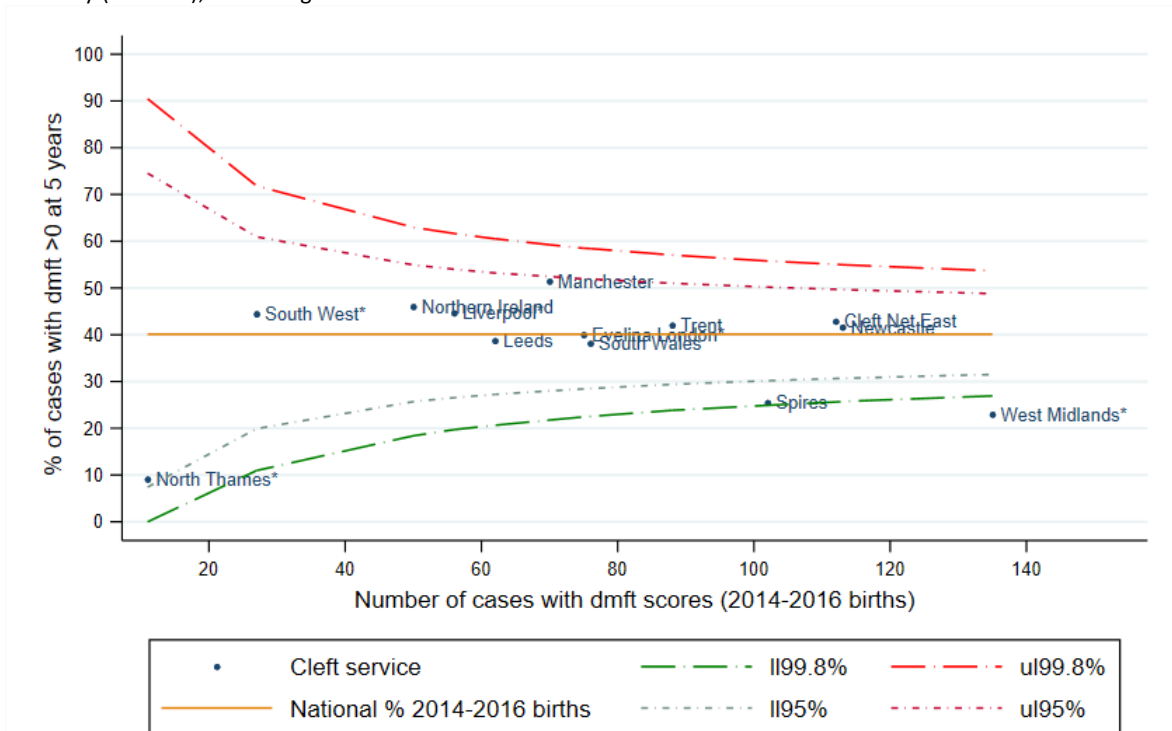
Outcome: dmft >0 scores	
Indicator	#13 – Dental decay at 5 years of age
Benchmarks	<ul style="list-style-type: none"> The Oral Health Survey of 5 year old children in 2022 reported that 29% of children had dental decay, with at least one (>0) dmft, in the general population of England³⁸. 39% of eligible children born 2012-2014 were reported to have dmft >0 scores (CRANE 2022).
Notes	Funnel plots are centred on the revised national rates after excluding data from services identified as negative outliers for consent verification (West Midlands) and dmft data completeness (South West, Evelina London, North Thames, and Liverpool).
Denominator	977 CRANE-consented children with a recorded total decayed, missing or filled teeth (dmft) score
What did we find?	 <ul style="list-style-type: none"> The mean number of dmft at 5 years was 1.94, with scores ranging from 0 to 20. 38% of children with a cleft had at least one (>0) dmft (Cleft Service range: 9%³⁹-51%, p=0.001). Differences between cleft types in the percentage of children with >0 dmft were statistically significant: 30% in CL, 38% in CP, 43% in UCLP and 42% in BCLP (p=0.020).
Outliers	<p>Positive: 1. West Midlands (23%), 2. Spires (26%)*</p> <p>* Positive alert for 2 consecutive reporting periods</p> <p>Negative: None</p>

Figure 5.5. Funnel plot showing the percentage of CRANE-consented 5-year olds, born 2014 to 2016, with experience of dental decay (dmft >0), according to Cleft Service.



Note: Funnel plot centred on the revised national percentage (40.1%) of children (born 2014-2016) with >0 dmft.

³⁸ National Dental Epidemiology Programme (NDEP) for England: Oral health survey of 5 year old children 2022 - <https://www.gov.uk/government/statistics/oral-health-survey-of-5-year-old-children-2022/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.

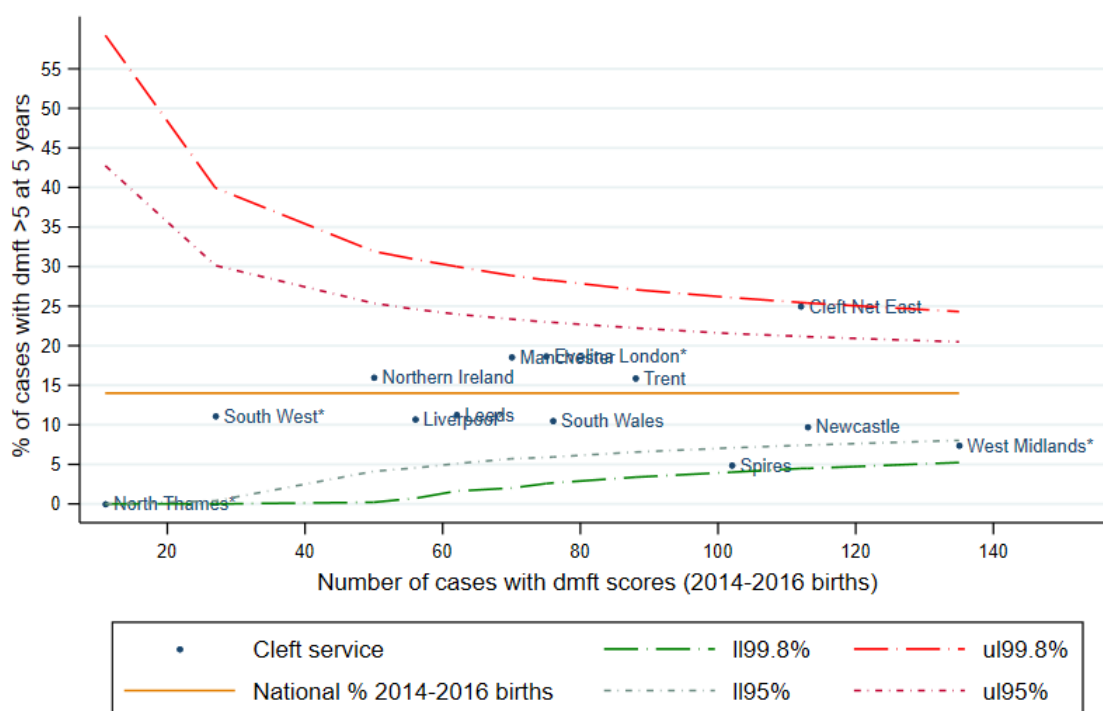
³⁹ Note that North Thames (9%) were a negative outlier for data completeness so interpret this rate with caution. The lowest rate among services not identified as negative outliers for consent or data completeness was 26% (Spires).

5.2.2. Extensive dental decay

Outcome: dmft >5 scores	
Indicator	#14 – Extensive dental decay at 5 years of age
Benchmarks	<ul style="list-style-type: none"> The Child Dental Health Survey in 2013 reported that 13% of children had extensive dental decay (dmft >5) in the general population of England, Wales and Northern Ireland at 5 years of age⁴⁰. 14% of eligible children born 2012-2014 were reported to have dmft >5 scores (CRANE 2022).
Notes	Funnel plots are centred on the revised national rates after excluding data from services identified as negative outliers for consent verification (West Midlands) and dmft data completeness (South West, Evelina London, North Thames, and Liverpool).
Denominator	977 CRANE-consented children with a recorded total decayed, missing or filled teeth (dmft) score
What did we find?	<ul style="list-style-type: none"> 13% of children with a cleft had extensive dental decay (dmft >5) (Cleft Service range: 0%⁴¹-19%, $p=0.001$). Differences between cleft types in the percentage of children with dmft >5 were not statistically significant: 9% in CL, 14% in CP, 13% in UCLP, and 18% in BCLP ($p=0.148$).
Outliers	None



Figure 5.6. Funnel plot showing the percentage of CRANE-consented 5-year olds, born 2014 to 2016, with extensive dental decay (dmft >5), according to Cleft Service.



Note: Funnel plot centred on the revised national percentage (14.0%) of children (born 2014-2016) with >5 dmft.

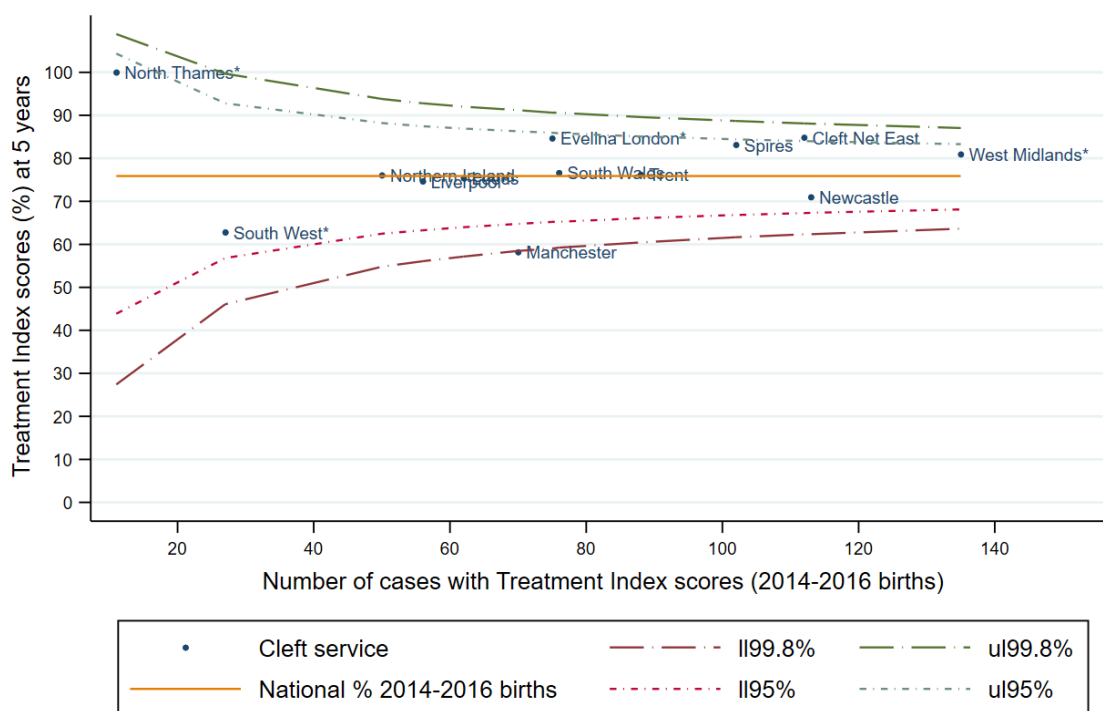
⁴⁰ Child Dental Health Survey 2013, England, Wales and Northern Ireland - <https://digital.nhs.uk/data-and-information/publications/statistical/children-s-dental-health-survey/child-dental-health-survey-2013-england-wales-and-northern-ireland>

⁴¹ Note that North Thames (0%) were a negative outlier for data completeness so interpret this rate with caution. Second lowest rate was 5% (Spire).

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	<ul style="list-style-type: none"> 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 England⁴². The average Treatment Index for eligible children born 2012-2014 was 77% (CRANE 2022).
Notes	<ul style="list-style-type: none"> High mean treatment index scores indicate that children have high levels of treated dental disease. Not subject to outlier policy but funnel plots provided to demonstrate variation in rates across services. Funnel plots are centred on the revised national rates after excluding data from services identified as negative outliers for consent verification (West Midlands) and dmft data completeness (South West, Evelina London, North Thames, and Liverpool).
Denominator	962 CRANE-consented children with scores for the calculation of Treatment Index ⁴³
What did we find?	<ul style="list-style-type: none"> The average Treatment Index for these children was 77% (Cleft Service range: 58%-100%⁴⁴). A Kruskal-Wallis test was used to compare the mean rank of Treatment Index scores between Cleft Services, which identified no statistically significant differences between services ($p=0.0744$).
Outliers	Positive: None Negative: 1. Manchester (58%)

Figure 5.7. Funnel plot showing the average dental Treatment Index percentage for CRANE-consented 5-year olds, born 2014 to 2016, according to Cleft Service.



Note: Funnel plot centred on the revised national Treatment Index percentage (75.9%) for children (born 2014-2016).

⁴² National Dental Epidemiology Programme (NDEP) for England: Oral health survey of 5 year old children 2022 - <https://www.gov.uk/government/statistics/oral-health-survey-of-5-year-old-children-2022/national-dental-epidemiology-programme-ndep-for-england-oral-health-survey-of-5-year-old-children-2022>.

⁴³ 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).

⁴⁴ Note that North Thames (100%) were 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% (Spires and Cleft Net East).


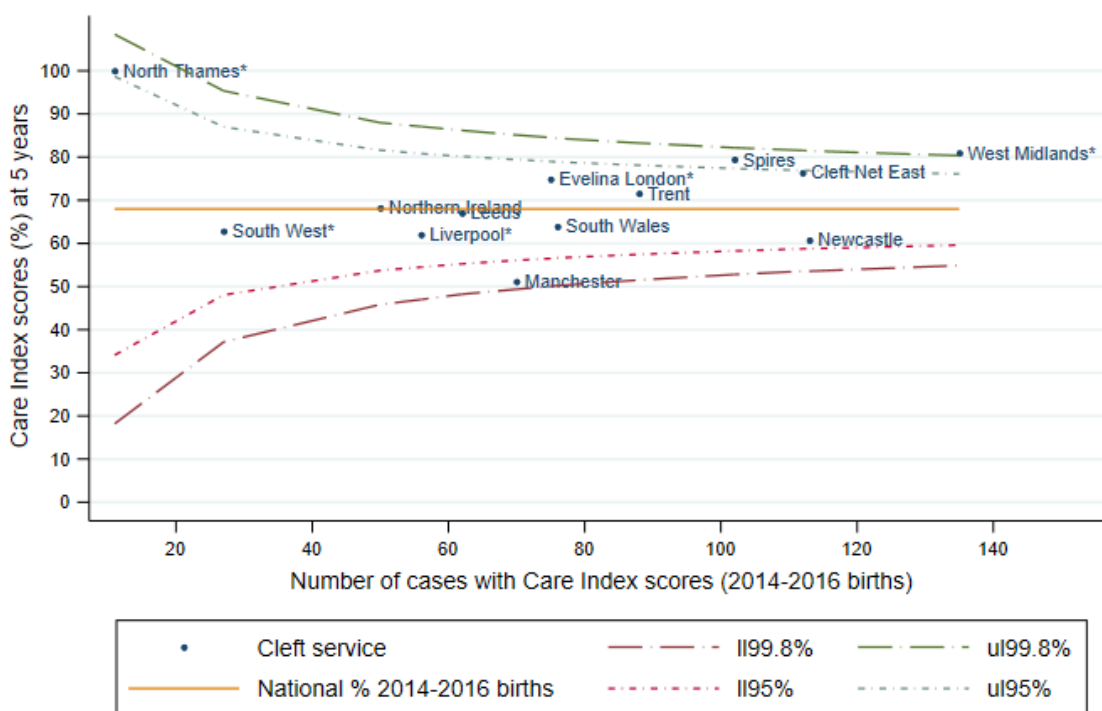
Outcome: Care Index	
Indicator	#16 – No untreated disease, as measured by the dental care index at 5 years of age
Benchmarks	<ul style="list-style-type: none"> The Oral Health Survey of 5 year old children in 2022 reported that the average Care Index for children is 7% (100% is the desirable outcome) in the general population in England⁴⁵. The average Care Index for eligible children born 2012-2014 was 70% (CRANE 2022).
Notes	<ul style="list-style-type: none"> High mean care index scores indicate that children have received the appropriate care at the earliest possible stage. Not subject to outlier policy but funnel plots provided to demonstrate variation in rates across services. Funnel plots are centred on the revised national rates after excluding data from services identified as negative outliers for consent verification (West Midlands) and dmft data completeness (South West, Evelina London, North Thames, and Liverpool).
Denominator	962 CRANE-consented children with scores for the calculation of Care Index ⁴⁶
What did we find?	 <ul style="list-style-type: none"> The average Care Index for these children was 70% (Cleft Service range: 51%-100%⁴⁷). A Kruskal-Wallis test was used to compare the mean rank of Care Index scores between Cleft Services, which identified a statistically significant differences between services (p=0.006).
Outliers	Positive: 1. West Midlands (81%) Negative: None

Figure 5.8. Funnel plot showing the average dental Care Index percentage for CRANE-consented 5-year olds, born 2014 to 2016, according to Cleft Service.



Note: Funnel plot centred on the revised national Care Index percentage (68.0%) for children (born 2014-2016).

⁴⁵ National Dental Epidemiology Programme for England: oral health survey of five-year-old children 2019 (results) - https://assets.publishing.service.gov.uk/government/uploads/system/uploads/attachment_data/file/873492/NDEP_for_England_OH_Survey_5yr_2019_v1.0.pdf. Data on children in the general population in Wales and Northern Ireland were not available at the time of producing this report.

⁴⁶ Care Index calculated using: data on filled teeth (f) and dmft scores; or a dmft score of 0 (equating to a Care Index = 1).

⁴⁷ Note that North Thames (100%) were 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 79% (Spire).

Recommendations: Dental health

- Cleft Services should aim for a calibrated specialist in paediatric dentistry to see all children with a cleft for a dental assessment at the age of 5 years, 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.

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 growth⁴⁸. 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 level⁴⁹. 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 value⁵⁰. 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: 4 July 2023
Birth years	Three years: 2014 to 2016
Countries	England, Wales and Northern Ireland
Inclusions	<ul style="list-style-type: none"> • CRANE-consented children • Complete UCLP
Exclusions	<ul style="list-style-type: none"> • Children who died before the age of 5 years
Notes	<ul style="list-style-type: none"> • Subject to outlier policy. • Data are not risk-adjusted. • Funnel plots are centred on the revised national rates after excluding data from services identified as negative outliers for consent verification (West Midlands). • Cleft Services with <10 cases are not shown on funnel plot due to insufficient data.
Raw data	‘Facial growth’ in the supplementary tables .

⁴⁸ 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.

⁴⁹ 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.

⁵⁰ 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


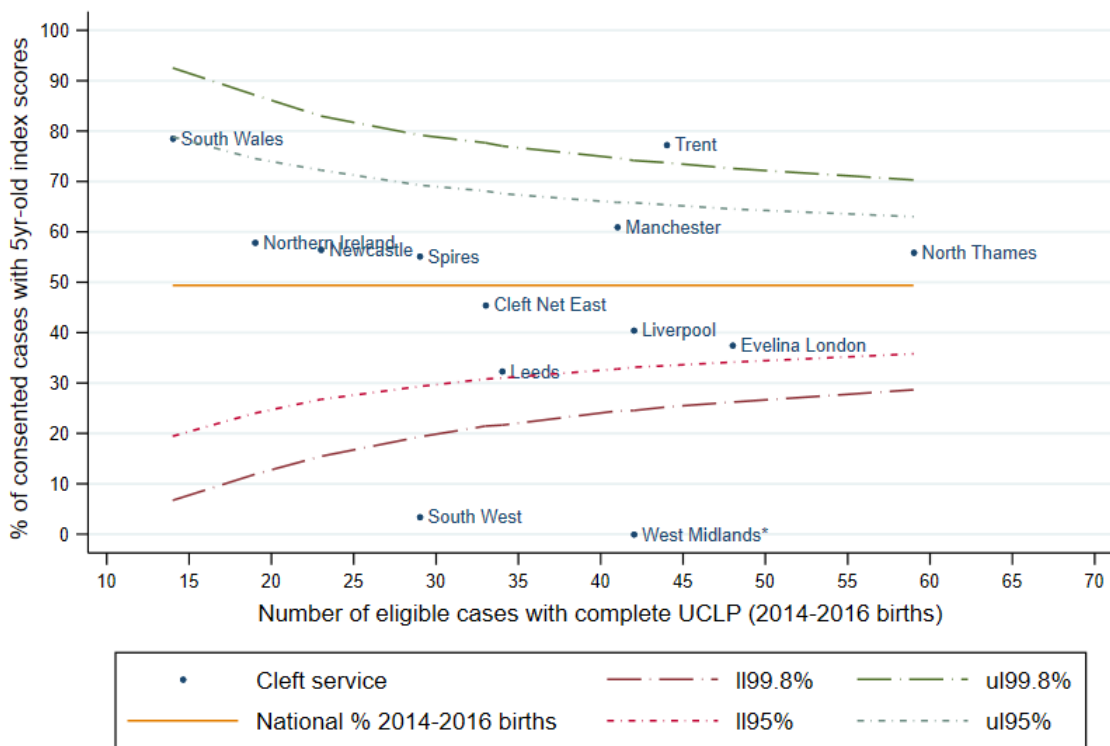
Data completeness: 5-year old Index scores	
Indicator	#17 – Facial growth at 5 years recorded for all eligible children.
Denominator	457 CRANE-consented children eligible for 5-year-old Index scores
What did we find?	 <ul style="list-style-type: none"> 45% of eligible children had recorded 5-year-old Index scores (Cleft Service range: 0%-79%, $p < 0.001$). This compares to 59% of those born 2012-2014. 167/205 (82%) scores were externally validated⁵¹. 29% had a reason⁵² the facial growth scores were not collected. 27% were missing data or a reason for not collecting data. The proportion of children with 5-year-old index scores varied significantly according to birth year (2014: 55%, 2015: 23%, 2016: 59%, $p < 0.001$).
Outliers	Positive: 1. Trent (77%) Negative: 1. West Midlands (0%), 2. South West (3%)

Figure 5.9. Funnel plot showing the percentage of CRANE-consented 5-year-olds, born 2014 to 2016, with a complete UCLP who had facial growth data reported, according to Cleft Service.



Note: Funnel plot centred on the revised national percentage (49.4%) of children (born 2014-2016) with facial growth data reported.

⁵¹ 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.

⁵² 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.


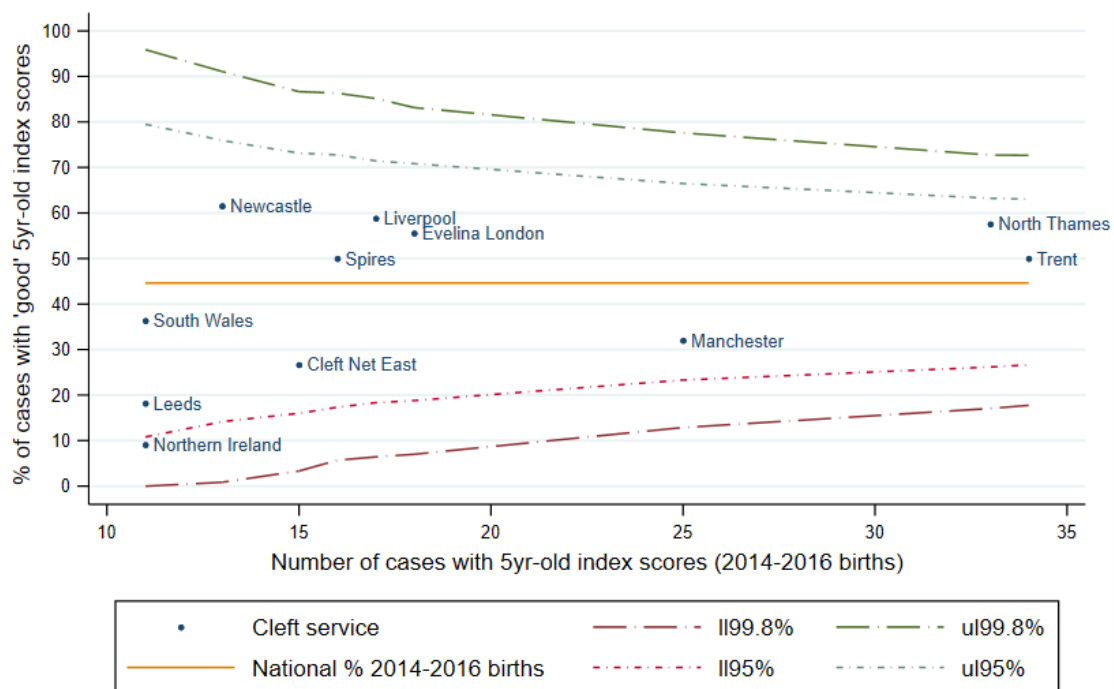
Outcome: 5-year-old index scores	
Indicator	#18 – Children with Five Year Old Index scores reflecting ‘good’ dental arch relationships.
Benchmarks	<ul style="list-style-type: none"> • Cleft Care UK reported in 2015 that 53%, 28% and 19% of children with UCLP had ‘good’, ‘fair’ and ‘poor’ dental arch relationships, respectively⁵³. • 38% of eligible children born 2012-2014 were reported to have ‘good’ scores (CRANE 2022).
Notes	Funnel plot is centred on the revised national rate after excluding data from services identified as negative outliers for consent verification (West Midlands) and facial growth data completeness (West Midlands and South West).
Denominator	205 CRANE-consented children with 5-year-old index scores
What did we find?	 <ul style="list-style-type: none"> • 45% of children had scores reflecting ‘good’ dental arch relationships at 5 years old (Cleft Service range: 9%-62%⁵⁴, $p < 0.001$). This represents a 7% improvement on the previous reporting period (2012-2014 births) but lower than the percentage reported in the Cleft Care UK study. • 33% of children had scores reflecting ‘fair’ dental arch relationships at 5 years old. • 22% of children had scores reflecting ‘poor’ dental arch relationships at 5 years old.
Outliers	None.

Figure 5.10. Funnel plot showing the percentage of CRANE-consented 5-year olds, born 2014 to 2016, with a complete UCLP who had good facial growth scores, according to Cleft Service.



Note: Funnel plot centred on the revised national percentage (44.6%) of children (born 2014-2016) with ‘good’ facial growth outcomes.

Recommendations: Facial growth
<ul style="list-style-type: none"> • Cleft Services should aim to take dental impressions or photographs of all children with a complete UCLP around the age of 5 years, to allow for an assessment using the 5-year-old Index. • The research community should undertake to: <ul style="list-style-type: none"> ○ compare UK facial growth outcomes with those in other countries, and ○ evaluate the predictive value of the 5-year-old Index in UK populations.

⁵³ 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.

⁵⁴ among Cleft Services that submitted data for ≥ 10 cases.

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: 4 July 2023
Birth years	Three years: 2014 to 2016
Countries	England, Wales and Northern Ireland
Inclusions	<ul style="list-style-type: none"> • CRANE-consented children • Children with a cleft affecting the palate (CP, UCLP, BCLP)
Exclusions	<ul style="list-style-type: none"> • Children who died before the age of 5 years • Children with submucous cleft palate⁵⁵ • Children with a diagnosed syndrome⁵⁶ entered onto the CRANE Database
Notes	<ul style="list-style-type: none"> • Subject to outlier policy. • Data are not risk-adjusted. • All funnel plots are centred on the revised national rates after excluding data from services identified as negative outliers for consent verification (West Midlands). • 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 (South West, Evelina London and Cleft Net East).
Raw data	‘Speech completeness’, ‘Speech standards’, ‘Speech standards by cleft type’ & ‘16-CAPS-A speech paramts’ in the supplementary tables .

⁵⁵ Patients with submucous cleft palate are excluded from all 5-year outcomes, including cases with a LAHSAL code identified as implausible, and likely submucous, when reviewed by cleft clinicians.

⁵⁶ 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).


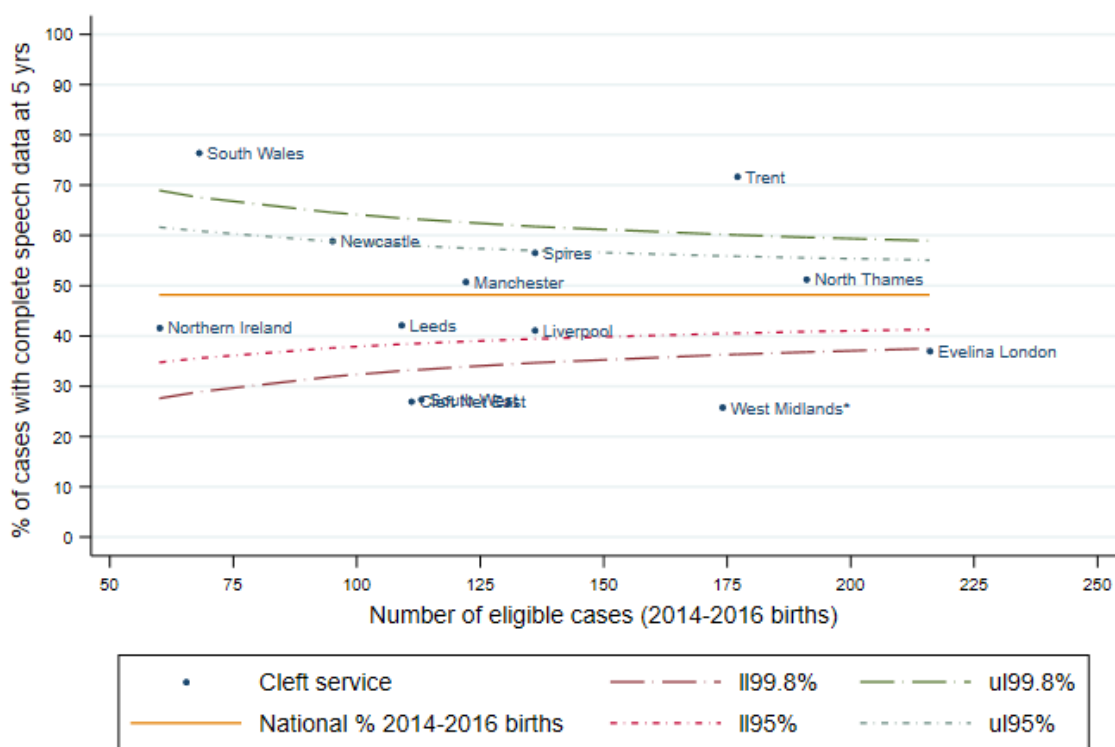
Data completeness: CAPS-A scores	
Indicator	#19 – Speech scores at 5 years recorded for all eligible children
Denominator	1,708 CRANE-consented children eligible for CAPS-A assessment
What did we find?	 <ul style="list-style-type: none"> 46% of children had all 16 CAPS-A speech parameters reported (Cleft Service range: 26%-77%, $p < 0.001$). This compares to 62% of those born 2012-2014. Of those with speech data, 13% 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. 48% had a reason⁵⁷ the speech outcomes were not collected. 5% were missing data or a reason for not collecting data. The proportion of children with complete speech data varied significantly according to birth year (2014: 52%, 2015: 27%, 2016: 59%, $p < 0.001$).
Outliers	Positive: 1. South Wales (77%), 2. Trent (72%) Negative: 1. West Midlands (26%), 2. Cleft Net East (27%), 3. South West (27%), 4. Evelina London (37%)

Figure 5.11. Funnel plot showing the percentage of CRANE-consented 5-year-olds, born 2014 to 2016, with speech outcomes reported, according to Cleft Service.



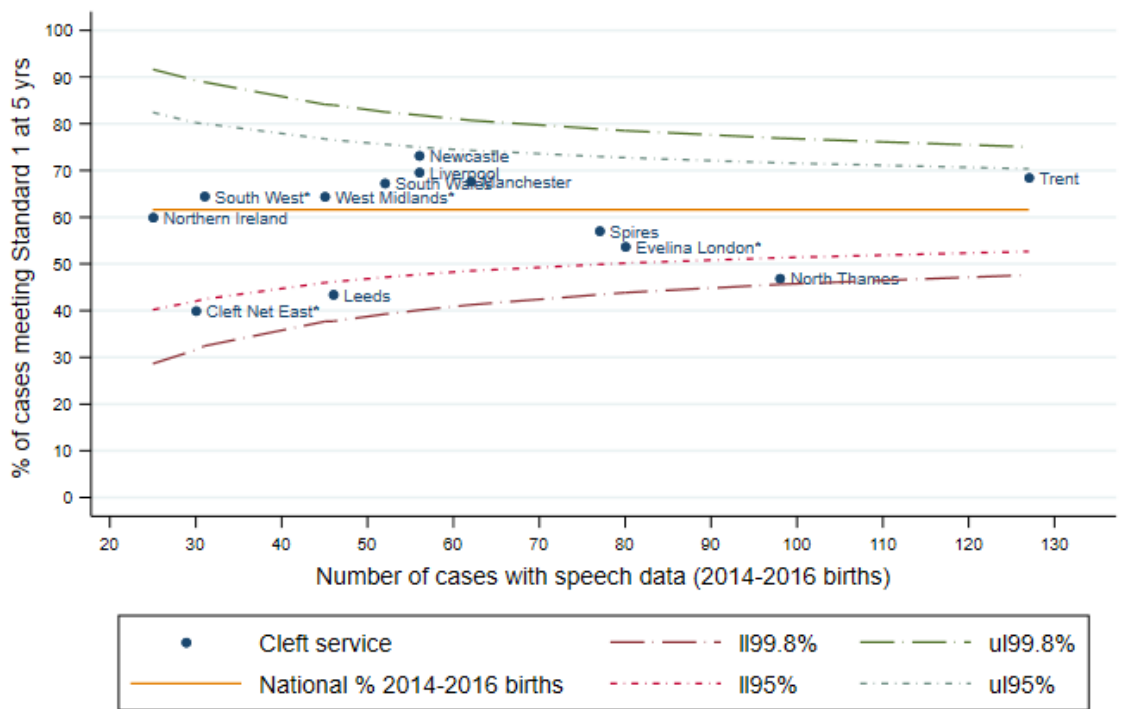
Note: Funnel plot centred on the revised national percentage (48.2%) of children (born 2014-2016) with speech outcomes reported.

⁵⁷ 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.

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	61% of children born 2012-2014 met speech outcome standard 1 (CRANE, 2022).
Denominator	785 CRANE-consented children with all 16 CAPS-A scores.
What did we find?	<ul style="list-style-type: none"> 60% of children met speech outcome standard 1 (Cleft Service range: 40%⁵⁸-73%, $p=0.001$). This is 1% lower than in the previous reporting period (2012-2014 births). 17% of children within this cohort had secondary surgery for speech purposes before the age of 5 years. This is similar to the 18% of children born 2012-2014. The percentage of children meeting standard 1 varied significantly according to cleft type: 72% in CP, 50% in UCLP and 37% in BCLP ($p<0.001$).
Outliers	None



Figure 5.12. Funnel plot showing the percentage of CRANE-consented 5-year olds, born 2014 to 2016, meeting speech outcome standard 1, according to the number of children in each Cleft Service with CAPS-A ratings.



Note: Funnel plot centred on the revised national percentage (61.6%) of children (born 2014-2016) meeting speech outcome standard 1.

⁵⁸ Note that Cleft Net East (40%) were a data completeness outlier so interpret this rate with caution. The lowest rate among services not identified as negative outliers for consent or data completion was 44% (Leeds).


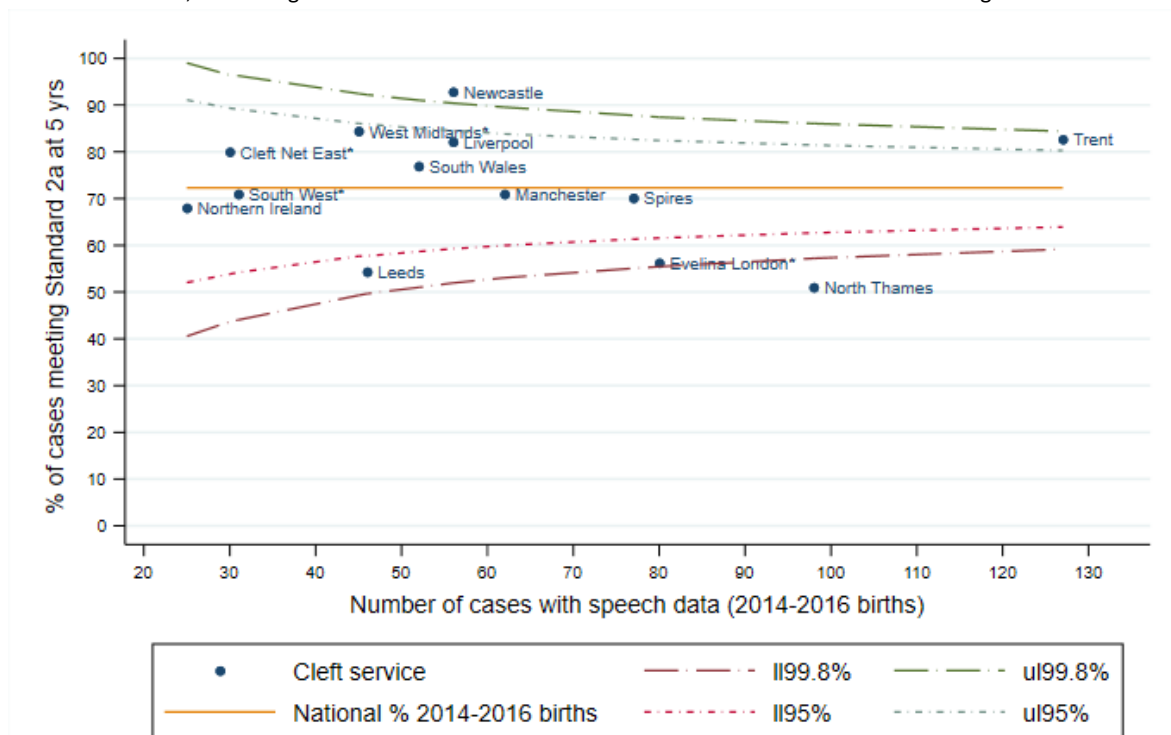
Outcome: National Speech Standard 2a	
Indicator	#21 – The achievement of speech without evidence of a structurally related speech difficulty. This standard is achieved when patients have no reported history of velopharyngeal surgery or fistula repair for speech purposes and have green ratings across 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 2012-2014 met speech outcome standard 2a (CRANE, 2022).
Denominator	785 CRANE-consented children with all 16 CAPS-A scores.
What did we find?	 <ul style="list-style-type: none"> 72% of children met speech outcome standard 2a. (Cleft Service range: 51%-93%, $p < 0.001$). This represents no change from the previous reporting period (2012-2014 births). Differences between cleft types in the percentage of children meeting standard 2a were not statistically significant: 74% in CP, 70% in UCLP and 65% in BCLP ($p = 0.110$).
Outliers	Positive: 1. Newcastle (93%) Negative: 1. North Thames (51%)

Figure 5.13. Funnel plot showing the percentage of CRANE-consented 5-year olds, born 2014 to 2016, meeting speech outcome standard 2a, according to the number of children in each Cleft Service with CAPS-A ratings.



Note: Funnel plot centred on the revised national percentage (72.3%) of children (born 2014-2016) meeting speech outcome standard 2a.


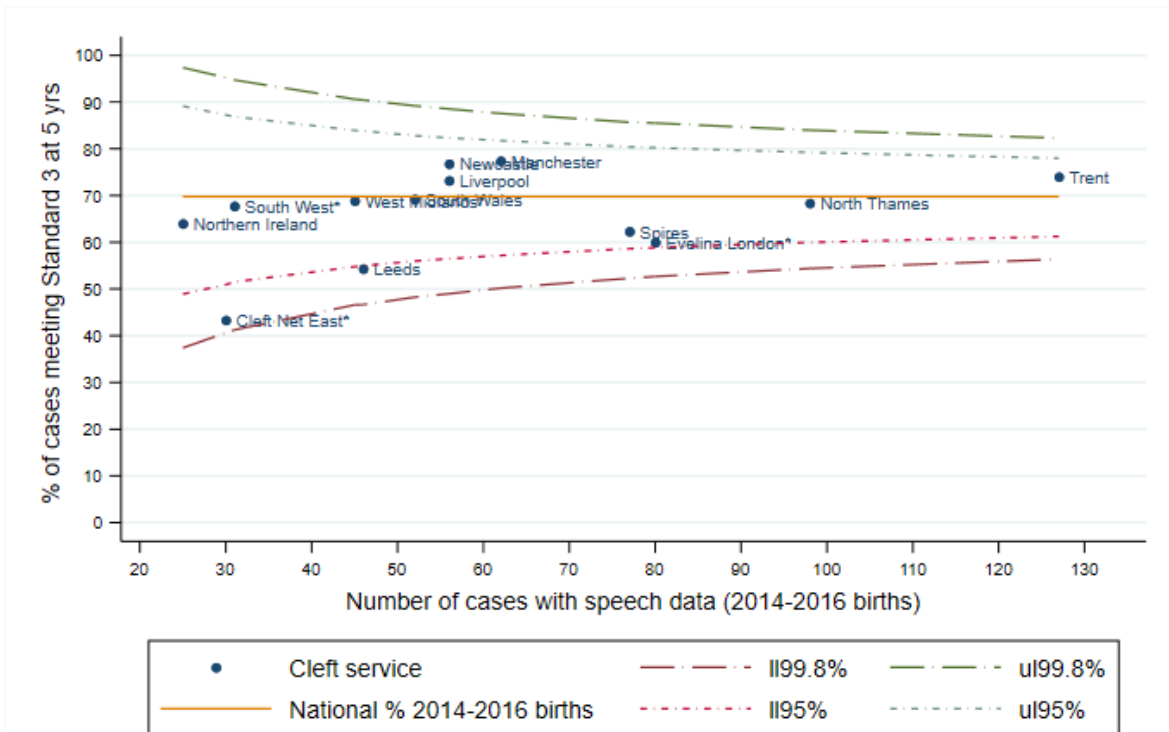
Outcome: National Speech Standard 3	
Indicator	#22 – The achievement of speech without evidence of significant cleft-related speech characteristics (on sentence repetition), which may require therapy and/or surgery: This standard is achieved when patients have green ratings across the following 10 CSCs: All three Anterior Oral CSCs, both Posterior Oral CSCs, all four Non Oral CSCs, and gliding of fricatives (a Passive CSC).
Benchmarks	69% of children born 2012-2014 met speech outcome standard 3 (CRANE, 2022).
Denominator	785 children with all 16 CAPS-A scores.
What did we find?	 <ul style="list-style-type: none"> 68% of children met speech outcome standard 3 (Cleft Service range: 43%⁵⁹-77%, p=0.026). This is 1% lower than in the previous reporting period (2012-2014 births). The percentage of children meeting standard 3 varied significantly according to cleft type: 80% in CP, 58% in UCLP and 41% in BCLP (p<0.001).
Outliers	None.

Figure 5.14. Funnel plot showing the percentage of CRANE-consented 5-year olds, born 2014 to 2016, meeting speech outcome standard 3, according to the number of children in each Cleft Service with CAPS-A ratings.



Note: Funnel plot centred on the revised national percentage (69.8%) of children (born 2014-2016) meeting speech outcome standard 3.

Recommendations: Speech
<ul style="list-style-type: none"> Children with a cleft affecting the palate should have speech assessed and reported to CRANE. Where possible, Cleft Services should investigate possibilities of remote methods for consensus listening. Cleft Services should work together to explore reasons for variation in speech outcomes.

⁵⁹ Note that Cleft Net East (43%) were a data completeness outlier so interpret this rate with caution. The lowest rate among services not identified as negative outliers for consent or data completion was 54% (Leeds).

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 listed⁶⁰. 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: 4 July 2023
Birth years	Three years: 2014 to 2016
Countries	England, Wales and Northern Ireland
Inclusions	CRANE-consented children, including those without a specified cleft type
Exclusions	<ul style="list-style-type: none"> • Children who died before the age of 5 years • Children with submucous cleft palate⁶¹
Notes	<ul style="list-style-type: none"> • Subject to outlier policy. • Data are not risk-adjusted. • Cleft Services with <10 cases are not shown on funnel plot due to insufficient data. • Funnel plots are centred on the revised national rates after excluding data from services identified as negative outliers for consent verification (West Midlands).
Raw data	‘Psychology data’ in the supplementary tables

⁶⁰ 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

⁶¹ Patients with submucous cleft palate are excluded from all 5-year outcomes, including cases with a LAHSAL code identified as implausible, and likely submucous, when reviewed by cleft clinicians.

5.5.1. Tiers of Involvement Measure


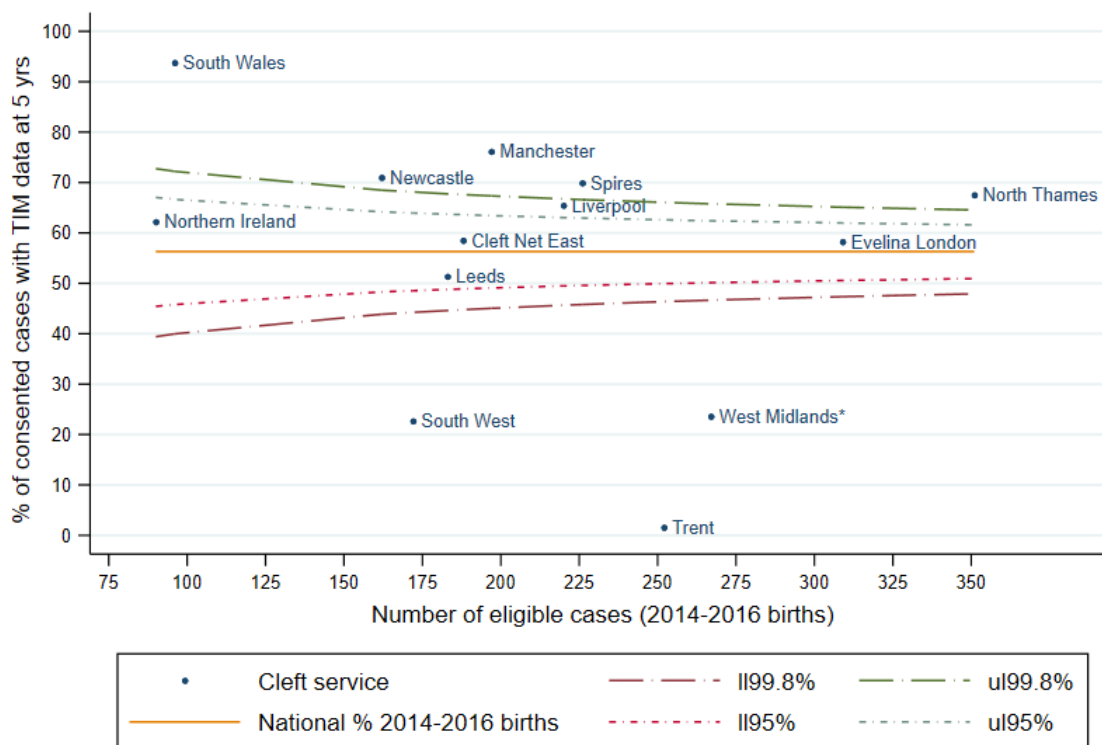
Data completeness: Tiers of Involvement score	
Indicators	#23 – TIM scores recorded for all eligible children
Denominator	2,713 CRANE-consented children
What did we find?	 <ul style="list-style-type: none"> 53% of eligible children had recorded TIM scores (Cleft Service range: 2%-94%, $p < 0.001$). 47% were missing data. The proportion of children with TIM scores varied significantly according to birth year (2014: 63%, 2015: 42%, 2016: 54%, $p < 0.001$).
Outliers	Positive: 1. South Wales (94%), 2. Manchester (76%), 3. Newcastle (71%), 4. Spires (70%), 5. North Thames (68%) Negative: 1. Trent (2%), 2. South West (23%), 3. West Midlands (24%)

Figure 5.15. Funnel plot showing the percentage of CRANE-consented 5-year-olds, born 2014 to 2016, with a cleft who had Tiers of Involvement Measure (TIM) scores reported, according to Cleft Service.



Note: Funnel plot centred on the revised national percentage (56.3%) of children (born 2014-2016) with TIM scores reported.


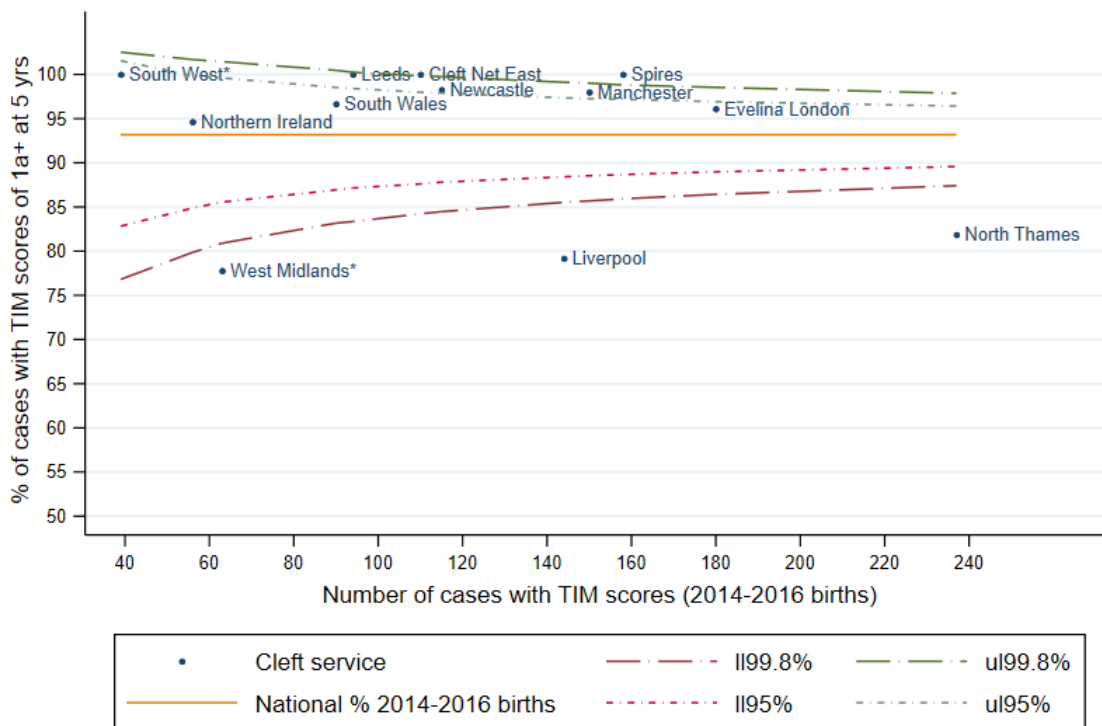
Key outcome: TIM scores of 1a+ (TIM tiers 1 to 6)	
Indicator	#24 – All eligible children seen by a psychologist before the age of 6 years.
Benchmarks	<ul style="list-style-type: none"> 100% of children should be seen by a psychologist and have a TIM assessment 93% of eligible children born 2012-2014 were reported to have TIM scores of 1a+ (CRANE 2022).
Notes	Funnel plots are centred on the revised national rates after excluding data from services identified as negative outliers for consent verification (West Midlands) and TIM data completeness (Trent, West Midlands and the South West).
Denominator	1,440 CRANE-consented children with TIM scores
What did we find?	 <ul style="list-style-type: none"> 93% 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: 78%⁶²-100%, p<0.001). <ul style="list-style-type: none"> 40% were assessed as not needing psychological input (TIM score 1). 47% received psychological input during the MDT clinic (TIM score 2). 6% required further psychological action (TIM scores 3-6). 7% were not seen by a psychologist (TIM score 0).
Outliers	<p>Positive: 1. Spireas (100%), 2. Cleft Net East (100%)*, 3. Leeds (100%)*</p> <p>*Positive alerts for 2 consecutive reporting periods</p> <p>Negative: 1. West Midlands (78%), 2. Liverpool (79%), 3. North Thames (82%)</p>

Figure 5.15. Funnel plot showing the percentage of CRANE-consented 5-year olds, born 2014 to 2016, with a TIM scores of 1a+, according to Cleft Service.



Note: Funnel plot centred on the revised national percentage (93.2%) of children (born 2014-2016) with TIM 1a+ scores.

Recommendations: Tiers of Involvement
<ul style="list-style-type: none"> 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.

⁶² Note that West Midlands (78%) were a negative outlier for consent verification so interpret this rate with caution. The lowest rate among services not identified as negative outliers for consent or data completion was 79% (Liverpool).

5.5.2. Strengths and Difficulties Questionnaire


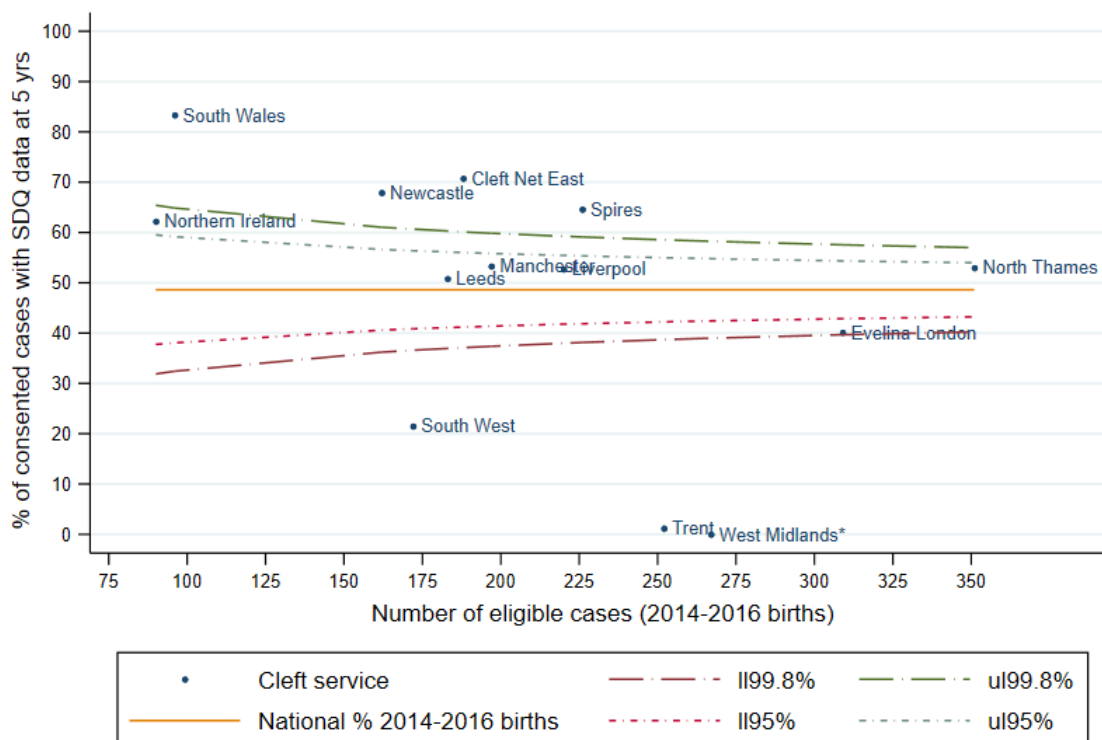
Data completeness: Strengths and Difficulties Questionnaire (SDQ) score	
Indicators	#25 – SDQ scores recorded for all eligible children
Denominator	2,713 CRANE-consented children
What did we find?	 <ul style="list-style-type: none"> 44% of eligible children had recorded SDQ scores (Cleft Service range: 0%-83%, $p < 0.001$). This compares to 55% of those born 2012-2014. 35% had a reason⁶³ psychology data were not collected. 21% were missing data or a reason for not collecting data. The proportion of children with SDQ scores varied significantly according to birth year (2014: 52%, 2015: 32%, 2016: 48%, $p < 0.001$).
Outliers	Positive: 1. South Wales (83%), 2. Cleft Net East (71%), 3. Newcastle (68%), 4. Spires (65%) Negative: 1. West Midlands (0%), 2. Trent (1%), 3. South West (22%)

Figure 5.16. Funnel plot showing the percentage of CRANE-consented 5-year-olds, born 2014 to 2016, with a cleft who had Strengths and Difficulties Questionnaire (SDQ) scores reported, according to Cleft Service.



Note: Funnel plot centred on the revised national percentage (48.6%) of children (born 2014-2016) with SDQ scores reported.

⁶³ 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.


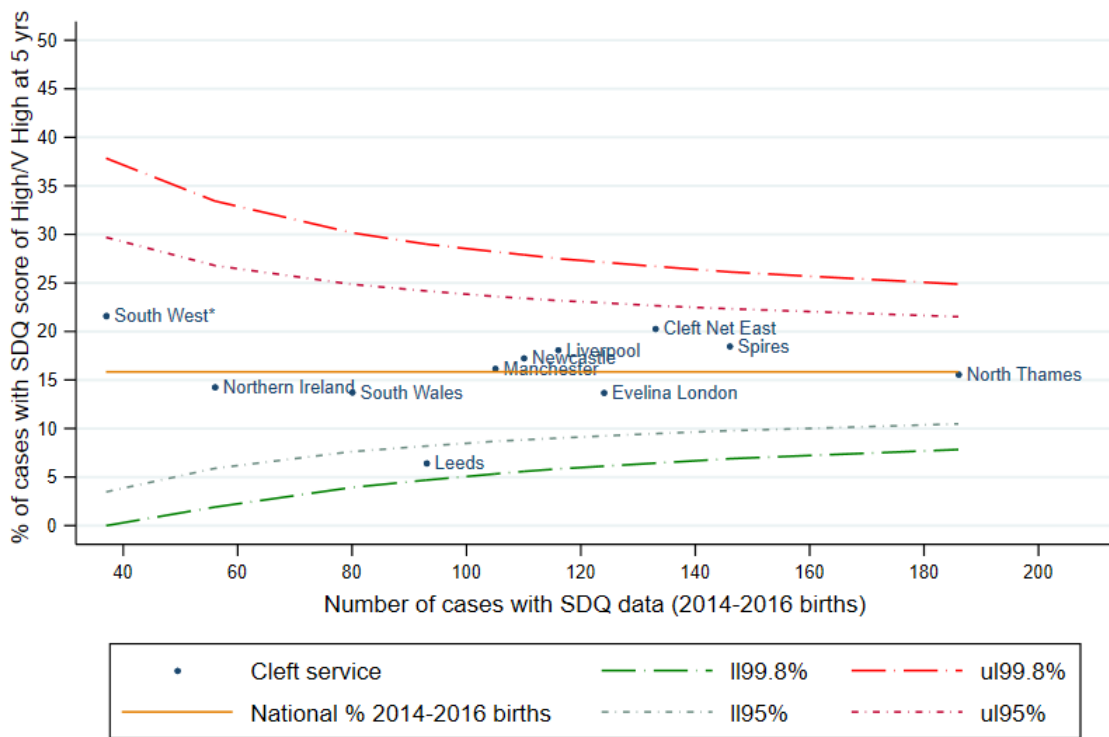
Key outcome: High/Very High SDQ scores	
Indicator	#26 – Psychological concerns identified at the age of 5 years.
Benchmarks	<ul style="list-style-type: none"> SDQ population norms: 10% of children aged 5 to 10 years old have SDQ scores that are 'high' or 'very high'⁶⁴. 18% of eligible children born 2012-2014 were reported to have 'high' or 'very high' SDQ scores (CRANE 2022).
Notes	Funnel plots are centred on the revised national rates after excluding data from services identified as negative outliers for consent verification (West Midlands) and TIM data completeness (Trent, West Midlands and the South West).
Denominator	1,189 CRANE-consented children with SDQ scores
What did we find?	 <ul style="list-style-type: none"> 16% of children with a documented SDQ score had 'high' (7%) or 'very high' (9%) scores (Cleft Service range: 7%-22%⁶⁵, p=0.385). This is 2% lower than the rate reported for 2012-2014 births. 9% had 'slightly raised' scores. The majority (75%) of children with SDQ scores had 'close to average' scores.
Outliers	None

Figure 5.17. Funnel plot showing the percentage of CRANE-consented 5-year olds, born 2014 to 2016, with high / very high SDQ scores, according to Cleft Service.



Note: Funnel plot centred on the revised national percentage (15.8%) of children (born 2014-2016) with high/very high SDQ scores.

⁶⁴ 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.

⁶⁵ among Cleft Services that submitted data for ≥10 cases.

Statement of ambition from the Psychology Clinical Excellence Network
<p>The Clinical Excellence Network is comprised of a specialist group of Clinical Psychologists who work within Cleft Services in the UK. As a group, we have reflected on the data that we submit to CRANE and believe that the Strengths and Difficulties Questionnaire (SDQ), which is our current Outcome Indicator, is not supporting us in understanding the impact of a cleft diagnosis on our patients. The SDQ is a measure of emotional wellbeing standardised on the general population. It is not cleft specific, the norms are outdated and are not organised into specific age categories. Furthermore, it cannot determine whether emotional wellbeing or difficulties are related to the patient’s cleft or not. To this end, we believe that continual collection of data using the SDQ lacks cleft-specific meaning. We have thus set up a CRANE CEN sub-group, which comprises a senior, qualified representative from each Cleft Service in the UK. We plan to arrange several meetings throughout 2024 with a key aim of utilising a new measure from January 2025. We believe that we need to replace our existing measure with something that is cleft-specific, valid, reliable and of interest to our patients, our colleagues and the wider NHS community. Our aim is to ensure that the information obtained can support us as a specialty in providing gold standard care to our patients as well as useful feedback to the wider multidisciplinary team. We are sharing our aims as a Statement of Ambition, recognising that we have given ourselves a year to find a measure, implement its use within each cleft centre and give our CRANE colleagues time to amend the database to reflect this change. We want to ensure that we future-proof our decision to minimise the risk of further adaptations in the near future. We recognise though that the needs of our patients change over time and we may need to adapt accordingly in time. We would like to commit to a new measure with the hope of truly understanding the psychological impact of a cleft diagnosis on our patients.</p>
Timeline of SDQ data collection
<p>CRANE will collect SDQ scores for children born up to 31 December 2017 only. Cleft Services should not enter SDQ scores onto the CRANE Database for children born from 1 January 2018 onwards. The final SDQ data submission will be reported in the CRANE 2024 Annual report⁶⁶.</p>

⁶⁶ <https://www.crane-database.org.uk/resources/data-collection-milestones-for-2024/>

6. Database development work

This chapter presents development work undertaken over the last year and includes peer-review publications, conference presentations and a focus on CRANE case ascertainment over a 10-year period and recent linkage with the Newborn Hearing Screening Programme for children born 2006 to 2021.

Publications in 2023:

1. Etoori D, Park MH, Blackburn RM, et al. Number and timing of primary cleft lip and palate repair surgeries in England: whole nation study of electronic health records before and during the COVID-19 pandemic. *BMJ Open* 2023;13:e071973.
2. Park MH, Fitzsimons KJ, Deacon S, et al. Longitudinal educational attainment among children with isolated oral cleft: a cohort study. *Archives of Disease in Childhood* 2023;108:563-568.
3. 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
4. Fitzsimons K, Hamilton M, van der Meulen J, et al. Range and frequency of congenital malformations among children with cleft lip and/or palate. *The Cleft Palate Craniofacial Journal*. 2023;60(8):917-927.

Conference contributions in 2023:

1. Additional congenital malformations in children with orofacial clefts are associated with poorer dental health outcomes at 5 years of age: Implications for reporting and clinical practice (CFSGBI, April 2023)
2. Timely Diagnosis of Cleft Palate and psychology outcomes at 1 and 5 years of age: A population-based cohort study using CRANE Database registrations for England, Wales and Northern Ireland (CFSGBI, April 2023)
3. The impact of congenital malformations of the circulatory and nervous systems on speech outcome at age 5 in children born with cleft palate +/- lip. Implications for annual UK cleft speech outcome reporting (CFSGBI, April 2023)
4. Evolution of cleft speech reporting in the UK. Incorporation of an early risk adjustment model does not substantially reduce the range in variability of speech outcomes (CFSGBI, April 2023)

6.1. Cleft case ascertainment in the CRANE Database compared with national administrative hospital data in England

Cohort summary	
Data sources	1. CRANE Database 2. Hospital Episode Statistics (HES), and 3. Office for National Statistics (ONS England) ⁶⁷
Birth years	Ten years: 2009 to 2018
Countries	England
Denominator	6,666,440 children born alive in the general population of England
Numerators	<ul style="list-style-type: none"> 10,090 children registered in CRANE with English Cleft Services 10,033 children identified in HES as having 1) a cleft diagnosis, and 2) a primary repair or dying before a primary repair would be performed according to UK guidelines.
What did we find?	<ul style="list-style-type: none"> Using CRANE registrations, the overall estimate for the prevalence of cleft lip and/or palate (CL/P) for the investigated period is 15.1 per 10,000 live births or 1 in 661 live births. Using HES records, the overall estimate for the prevalence of CL/P is 15.1 per 10,000 live births or 1 in every 664 live births. The prevalence of clefting has remained relatively stable over the birth years examined (range: 14.2 to 16.1 per 10,000 live births according to CRANE registrations; range: 13.8 to 15.9 per 10,000 live births according to HES records). Although fluctuations and differences between the two data sources exist in annual prevalence rates, overall rates are very similar, indicating high case ascertainment levels in CRANE.
Recommendations	The research community should undertake to explore whether regional differences in the prevalence of clefting exist across England.

Background

The CRANE Database requires Cleft Services to submit data on all children born alive with a cleft lip and/or palate in the UK (Scotland joined in 2023). This data is collected under a section 251 agreement for England, Wales and Northern Ireland but currently requires consent for children born in Scotland.

In our recent annual reports, a steady decline in the number of children being registered has been observed. Our aim was to determine whether this was in relation to a reduction in national live birth rate, a reduction in cleft incidence, or a reduction in children being registered on the CRANE Database, reflecting a decline in case ascertainment.

Our objectives were to:

1. compare the number of cleft registrations submitted to CRANE with the number of cleft cases identified in Hospital Episode Statistics, a national database of all National Health Service (NHS) hospital admissions in England, and
2. calculate the prevalence of live births affected by a cleft using CRANE and HES data sources and the national live birth rate in England, reported by the Office for National Statistics.

⁶⁷ Office for National Statistics: Annual summary statistics on live births and stillbirths in England and Wales, 2019. Released July 2020 (checked October 2020). Available at: <https://www.ons.gov.uk/peoplepopulationandcommunity/birthsdeathsandmarriages/livebirths/datasets/birthsummarytables>

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 Hospital Episode Statistics (HES) database (www.digital.nhs.uk) contains records on all diagnoses and treatments given during admissions to NHS hospitals in England. The NHS is a state-funded healthcare system that provides organized multidisciplinary care for all children born with a cleft in England. HES data are collected by professional health coders based in each NHS provider in England, primarily for the purpose of reimbursement. Records from each hospital episode are reviewed by coders and diagnostic information is captured using the International classification of Diseases—10th Revision (ICD-10, and procedure information is classified according to codes from the Classification of Surgical Operations and Procedures, 4th revision (OPCS-4). Full information on the HES data processing cycle and quality is publically available⁶⁸. The HES dataset analysed by CRANE contained all hospital admissions occurring between 1 January 2009 to 31 March 2020.

The total number of annual live births in England, between 2009 and 2018, was obtained from the Office for National Statistics⁶⁹ and was used to estimate the prevalence of live cleft births as a percentage of all live births.

Cleft cases

CRANE-registered cases, born 1 January 2009 to 31 December 2018, were included in our analyses if they had been registered by an English Cleft Service, in order to reflect the number of cases requiring care from English NHS trusts.

Patients in HES, born between 1 January 2009 and 31 December 2018, were identified as being a 'cleft' case if they had an ICD-10 code for cleft lip and/or palate (Q35, Q36 and Q37) and an OPCS code for a primary cleft repair (F031 and/or F291) occurring in their HES records. HES records up to 30 March 2020 were reviewed, providing a minimum follow-up period of 2 years and 3 months after birth to allow time for a primary repair to be performed. Children with a cleft diagnosis but without a primary repair record were also included if they had a cleft lip only (CLO) and died <6 months or a cleft palate ± lip and died <13 months, on the basis that they died before a repair may have been scheduled. Children with a 'Foreign' region of residence were excluded.

Analyses

The number of children born with a cleft was divided by the total number of reported live births occurring in England during the corresponding time period. Prevalence rates per 10,000 live births were calculated for both CRANE registrations and HES records and were estimated for each birth year between 2009 and

⁶⁸ [Hospital Episode Statistics. HES user guide: Health and Social Care Information Centre, 2010. www.hscic.gov.uk/hes](http://www.hscic.gov.uk/hes)

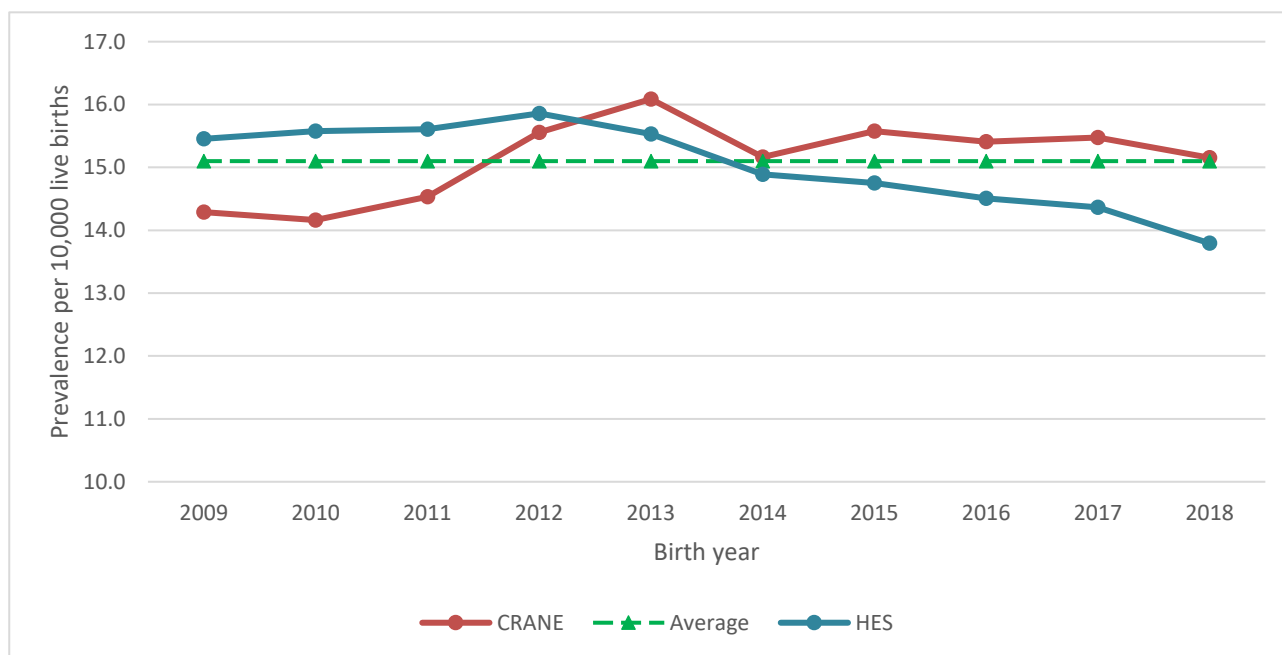
⁶⁹ <https://www.ons.gov.uk/peoplepopulationandcommunity/birthsdeathsandmarriages/livebirths/datasets/birthsummarytables>

2018 to describe fluctuations in cleft case ascertainment and prevalence over time. Analyses were performed in Stata V17.0.

Results

During the 10 birth years analysed, 6,666,440 children were born alive in England¹. Of these, 10,090 were registered in CRANE by English Cleft Services, corresponding to an overall cleft prevalence rate of 15.1 per 10,000 live births or a rate of 1 in 661 live births. For the same period, 9,613 children were identified in the HES dataset as having both a cleft diagnosis code and a primary repair procedure code. A further 420 children were identified as having a cleft diagnosis but died before a primary repair was performed. Therefore, in HES, a total of 10,033 children were identified as being born alive with a cleft, which corresponds to an overall cleft prevalence rate of 15.1 per 10,000 live births or a rate of 1 in 664. Figure 6.1 shows the annual prevalence rate for each data source and the overall average. Prevalence rates ranged from 14.2 (2010) to 16.1 (2013) per 10,000 live births based on CRANE registrations. In HES, rates ranged from 13.8 (2018) to 15.9 (2012) per 10,000 live births. Although HES rates suggest a downward trend in the prevalence of CL/P, this is not supported by prevalence rates based on CRANE registrations, and is likely the result of shorter follow-up times for these births.

Figure 6.1. Annual prevalence of live births affected by cleft lip and/or palate in England



In total, there were 57 more CRANE registrations than 'cleft' cases identified in HES over the 10 birth years. This corresponds to an annual difference of 1 per 117,000 live births. Although the overall average was similar between the two data sources, the annual fluctuations did vary between the two datasets. Since 2013, CRANE registrations have exceeded the number of cleft cases identified in HES. The inclusion of children with an untreated submucous cleft palate in CRANE could contribute to this difference, as these cases would not have met our HES 'cleft case' inclusion criteria. In CRANE, we identified 198 children who had a submucous cleft palate alone and were born during the birth years included in these analyses. Unfortunately, it is not possible to determine whether these children had undergone repair using CRANE data. If we were to exclude cases with a submucous cleft palate alone from CRANE registrations, the overall prevalence of an overt cleft would be 14.8 per 10,000 live births. It is likely that the rates described here

underestimate the true cleft prevalence as children dying shortly after birth may not even be referred to a Cleft Service. Equally, in HES, children with untreated clefts will not have been included unless they had a cleft diagnosis recorded in their records and died before a primary repair would be scheduled.

Summary

The overall number of CRANE registrations was very similar to the number of 'cleft' cases identified in the HES dataset over the 10-year period. This provides reassurance that CRANE has very high case-ascertainment levels and that registrations are representative of the true prevalence of clefting among live births in England, particularly among those that require cleft treatment.

6.2. CRANE linkage with the Newborn Hearing Screening Programme

Cohort summary	
Data sources	1. CRANE Database 2. Newborn Hearing Screening Programme (NHSP) Database
Birth years	14.75 years: 01 March 2006 to 31 December 2021
Countries	England
Inclusions	<ul style="list-style-type: none"> CRANE-consented children with consent to data linkage Children registered with an English Cleft Service and with an English postcode of residence
Raw data	'NHSP' in the supplementary tables

Data Linkage	
Benchmark	<ul style="list-style-type: none"> 100% of children should be linked to their newborn hearing screen data. The acceptable threshold for completion of a newborn hearing screen in England is $\geq 98\%$⁷⁰.
Denominator	12,566 English newborns registered in CRANE Database
What did we find?	<ul style="list-style-type: none"> 11,944 children out of 12,566 (95%) were linked to an NHSP record. Of these: <ul style="list-style-type: none"> 11,730 (98%) had a complete newborn hearing screening assessment 214 (2%) had an incomplete newborn hearing screening assessment Linkage rate varied according to cleft type ($p < 0.001$). Children with CL were more likely to be linked (97%), compared to those with UCLP (96%), BCLP (95%), SMCP+CL (95%), CP (94%) and SMCP (89%). Sex distribution did not vary between the linked and unlinked records but syndromic status did, with more children with a syndrome reported in the unlinked dataset ($p < 0.001$). Data linkage rate improved over time, from 92% in 2006 to 98% in 2021 ($p < 0.001$).

Screening Outcome	
Denominator	11,730 children with a complete newborn hearing screen assessment
What did we find?	<p>Of those with a complete newborn hearing screening assessment;</p> <ul style="list-style-type: none"> 8,901 (76%) children had a clear response bilaterally 1,165 (10%) had no clear response unilaterally 1,612 (14%) had no clear response bilaterally Screening outcome varied according to cleft type ($p < 0.001$). The percentage of children without a clear response in one or both ears ranged from 5% in those with CL to 36% in those with BCLP.

Recommendations	Further investigations are needed to understand why linkage rate and screening outcome vary according to cleft type. The newborn hearing screen cannot always be completed for children with significant additional medical needs. In certain cases babies are referred directly for diagnostic assessment and miss out the 'screening' hearing assessment step. This would be more likely to occur in children with syndromes. Children with no clear response are referred for audiological diagnostic hearing screen assessments; this part of the process needs to be explored further.
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⁷⁰ 2020/2021 Newborn Hearing Screening Programme (NHSP) standards reports (Accessed 02/12/2023)
<https://www.gov.uk/government/publications/202021-newborn-hearing-screening-programme-nhsp-standards-reports>

Introduction

Congenital hearing loss is one of the most common chronic conditions among children⁷¹. The Newborn Hearing Screening Programme (NHSP)⁷², 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 age⁷³. It is recognised within the NHSP that certain risk factors can influence which pathway a baby 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 hearing assessment following the newborn hearing screen due to a higher potential risk of hearing loss. Therefore it is essential to understand how the NHSP program is aiding early detection of hearing loss and the aetiology of hearing loss in children born with a cleft in England.

Objectives

1. To link consented children in the CRANE Database to their individual NHSP data.
2. To determine the percentage of children registered in the CRANE Database who (a) undergo a newborn hearing screening assessment and (b) have no clear response in one or both ears during their screening assessment.

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 centralised 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)⁷⁴, 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. 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.

⁷¹ 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>.

⁷² Overview of Newborn Hearing Screening Programme: <https://www.nhs.uk/conditions/baby/newborn-screening/hearing-test/> [Last accessed: 24/02/2023]

⁷³ 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.

⁷⁴ Overview of Newborn Hearing Screening Programme: <https://www.nhs.uk/conditions/baby/newborn-screening/hearing-test/> [Last accessed: 24/02/2023]

Patients

CRANE-registered cases, born 1 March 2006 to 31 December 2021, were included in our analyses if they had been registered by an English Cleft Service. Children with a non-English postcode of residence who were referred on to an English Cleft Service from a non-English maternity service (e.g. in Wales) were excluded.

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 codes⁷⁵ in the CRANE Database. Sex and syndromic status were also obtained from the CRANE database.

Outcome

Outcomes following newborn hearing screening assessment were defined as:

- (a) Clear response in both ears;
- (b) Clear response with targeted follow-up due to known risk factors recorded in the NHSP database;
- (c) No clear response in one ear only (unilateral);
- (d) No clear response in both ears (bilateral) and
- (e) For children with an incomplete assessment, a reason for this was provided.

According to the NHSP protocol⁷⁶, children 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.

Analysis

Data linkage rate between the CRANE Database and NHSP database was compared according to cleft type, sex and syndromic status. For all statistical tests, p-values <0.05 were considered significant.

Comparisons between the following subgroups were performed using a Chi-squared test.

- Percentage of children with linked records according to cleft type and year of birth,
- Percentage of children with a complete hearing screening assessment by cleft type, and
- Hearing screening outcomes by cleft type.

Results

Children eligible for data linkage

A total of 13,732 children, born between 01 March 2006 and 31 December 2021, were registered on the CRANE Database and provided consent for data linkage. 1,166 children, who were either registered with a non-English Cleft Service or from non-English postcodes but receiving their care from a Cleft Service based in England, were excluded. The cohort of CRANE-registered children eligible for data linkage therefore included 12,566 children (Figure 6.2).

⁷⁵ CDC Orofacial Clefts - <https://www.cdc.gov/ncbddd/birthdefects/surveillancemanual/chapters4>. Accessed 27 February 2023.

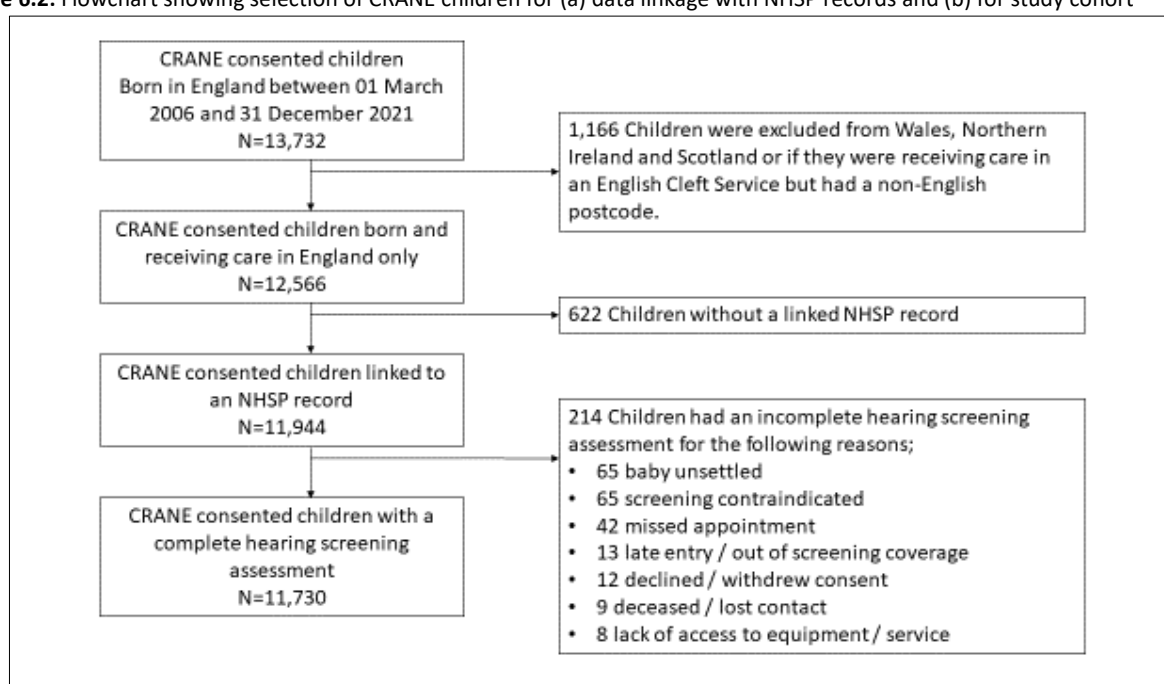
⁷⁶ NHS public health functions agreement 2019-20. Service specification No.20, NHS Newborn Hearing Screening Programme, July 2019

Data Linkage

11,944 out of 12,566 (95%) CRANE-consented children living in England were linked to an NHSP record, indicating a high data linkage rate between the two databases. The data linkage rate varied significantly by cleft type and year of birth ($p < 0.001$). It was highest among children with a cleft affecting the lip only (97%), followed by children diagnosed with UCLP (96%), BCLP (95%), SMCP+CL (95%), CP (94%) and SMCP (89%). Linkage for the full cohort increased from 92% for 2006 births to 98% for 2021 births, showing an improvement in linkage rate over time.

The patient characteristics of children in the linked and unlinked groups are reported in the Supplementary Tables. The distribution of girls and boys in the linked and unlinked cohort was not significantly different. The cleft type distribution and the syndromic status did vary. Children with a syndromic diagnosis were less likely to be linked to an NHSP record ($p < 0.001$) compared to those without a syndromic diagnosis.

Figure 6.2: Flowchart showing selection of CRANE children for (a) data linkage with NHSP records and (b) for study cohort



Hearing Screening Assessment

Out of 11,944 English children registered in the CRANE Database and linked to an NHSP record, 11,730 (98%) had a complete hearing screening assessment. The remaining 214 children (2%) had an incomplete hearing screening assessment for reasons explained in Figure 1 and were excluded from further analysis. 60% of screening assessments took place in hospital, 29% in Neonatal Intensive Care Units (NICU) and 10% in the community. The proportion of children within each outcome category was as follows;

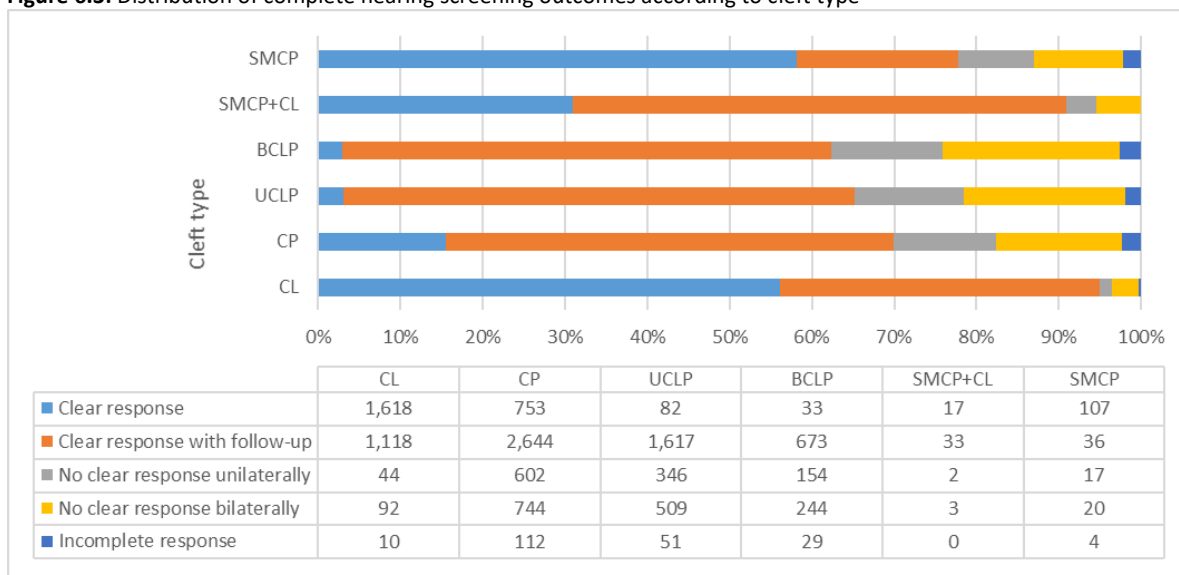
- (a) 23% Clear response bilaterally - no follow up;
- (b) 52% Clear response bilaterally - targeted follow up;
- (c) 10% No clear response unilaterally - referral;
- (d) 14% No clear response bilaterally - referral; and
- (e) 2% Incomplete assessment.

Out of 11,508 children with a complete hearing screening assessment and specified cleft type:

- 76% had a clear response in both ears whether or not they were targeted for follow-up
- 24% had no clear response in one or both ears.

The outcome of the hearing screening assessment varied significantly by cleft type ($p < 0.001$, see Figure 6.3). The proportion of children with a clear response in both ears decreased with increasing anatomical involvement of the four main cleft types (CL < CP < UCLP < BCLP), while the proportion of children with no clear response bilaterally increased with increasing anatomical involvement of the cleft.

Figure 6.3: Distribution of complete hearing screening outcomes according to cleft type



Screening rate among all CRANE registered children meets the acceptable national level for England but this was lower for children diagnosed with a cleft involving the palate.

Summary

A high proportion of children registered in CRANE have been linked to an NHSP record. A complete hearing screen was reported for 11,730 children. 24% of these children with a complete hearing screen did not have a clear response in one or both ears. Cleft type was found to significantly influence both linkage rate and the outcome of the hearing screen. Children born with an isolated cleft palate or SMCP may not have received this diagnosed at the time of their new born hearing screen. The process of triggering a targeted follow up audiology assessment relies on the craniofacial diagnosis (cleft) being added to the NHSP database at the time of entering the initial screen results. Further work needs to be undertaken to explore reasons for variation in linkage and screening outcome and to check if children with a cleft involving the palate were referred appropriately for targeted follow up audiological assessment. We are grateful to CLEFT for agreeing to fund further development work using the NHSP dataset in 2024.

7. Conclusion

In his foreword, retiring Cleft Development Group chair Simon van Eeden invokes the words of Maya Angelou and challenges us all **to do better** and with that take cleft care in the UK to the next level. Furthermore “the whole is greater than the sum of the parts” (Aristotle). With our stakeholders as individual parts – clinician, patient, parent, guardian or carer, manager, data scientist, commissioner or political leader – playing an important role in national clinical audit, by working together, we achieve an outcome greater than our constituent parts.

In many fields it has been clearly demonstrated that by sharing common goals, identifying together where improvements in process, pathways and performance can be made, agreeing how this is going to be achieved, and by when, that real meaningful change can be achieved. With small incremental gains in each area of practice or work, great things can be delivered.

Over the last 25 years, since the Clinical Standards Advisory Group (CSAG) report in 1998, we as a community have learnt a lot. In particular we have learnt a lot about data; how to collect and collate it, how to input it onto CRANE, how it can be linked, how it should be analysed, how variable and beautiful it can be, how it should be interpreted and what its limitations are. We have been on an information journey.

The CRANE 2020 Annual Report documented collated data from the previous 10 years. This current report shows that while most data had improved from that reported on in the 1998 CSAG report, the national picture both in terms of data completeness and outcomes was mostly static. Nevertheless, at service level there is considerable variability. This variability indicates that there is still much improvement that we, the whole UK cleft community, can achieve together.

We currently lead the world in our processes of annual data collection, analysis and publication of cleft care outcomes. We have delivered a more streamlined report that responds to feedback, encouraging all to read and benefit from the report in full, not just chapters relevant to an individual’s direct clinical activity.

Do not just think of the CRANE report as a one-off read. It is a resource that we can and should return to regularly in our quest for quality improvement. By reading and digesting, together, the stories told within each of the annual reports, we have the beginnings of a road map to the incremental gains that will define the future of cleft care in the UK.

What we have achieved together over the last 25 years is immense, but let’s not be complacent. Our community is now well-armed and informed by the outlier policy to drive improvement within teams and improvement across Cleft Care Services, particularly benefiting from shared learning. Now that we **know better**, let’s **do better TOGETHER!**

Wishing you all the very best for the festive season!

Yours most 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. 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. 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. In the context of this report, we refer to the number of children in the cohort we are discussing that could meet a certain criteria. For example, children with a Cleft Palate (CP) only.
dmft	Decayed, missing and filled teeth at 5 years of age
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: <ul style="list-style-type: none"> • Each point on the funnel plot represents a Cleft Service. • Each funnel plot is for one outcome, with its values shown on the vertical/Y axis. • The size of the Cleft Services’ cohort is shown on the horizontal or X axis. • The benchmark value or overall national percentage is shown as a horizontal line through the centre of the graph. <p>The graph shows two funnels that lie on either side of the benchmark and are called the control limits – similar to confidence intervals.</p>

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

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

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

General population	In epidemiological terms, all individuals without reference to any specific characteristic.
	In the context of this report, and to aid comparison, we sometimes refer to the latest national figures for children in the general population, which may also include children with a cleft or other health conditions. E.g. gestational age and birthweight in the general population of England & Wales, according to the Office for National Statistics (ONS) (as in the Registrations section of this report).
	In some instances, the latest national figures are based on a random sample of children in the general population, which, again, may include children with a cleft or other health conditions.
Hospital Episode Statistics (HES)	A national database containing records on all admissions to NHS hospitals in England.
ICD-10	International Classification of Disease 10th Revision
LAHSAL	A code used to classify cleft type. Each letter (LAHSAL) relates to one of the six parts of the mouth that can be affected by a cleft.
MDT	Multi-disciplinary team.
National Hearing Screening Programme (NHSP)	The Newborn Hearing Screening Programme (NHSP) ⁷⁷ , 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.
	In the context of this report, we refer to the number of children meeting a certain criteria. For example, receiving a certain type of care or meeting a standard.
OPCS-4	Classification of Surgical Operations and Procedures 4th Revision
RS	Robin Sequence
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

⁷⁷ Overview of NHSP <https://www.nhs.uk/conditions/baby/newborn-screening/hearing-test/> [Last accessed: 24/02/2023]