Cleft Care India Study: A National Audit on Patients with Non-Syndromic Unilateral Cleft Lip and Palate Patients. Part 1: Background, Skeletal, Dental Arch, and Nasolabial Outcomes

Abstract

Background:

India has one of the world's largest burdens of cleft lip and palate (CLP), yet lacks a national outcomes registry or standardized system for cleft care evaluation. The *Cleft Care India Study (CCIS)* is a multicentric national initiative to assess current treatment outcomes for patients with non syndromic unilateral cleft lip and palate for 5, 12 and 20 year old age groups.

Methods:

Fourteen cleft care centers across India participated in this cross-sectional study. Standardized protocols for collecting data were formulated with input from the Study Steering Committee, validated through pilot testing, and employed for data collection. Data collected included intraoral and extraoral photographs, study models, radiographs, surgical information, oral health assessments, speech recordings, hearing evaluations, psychosocial measures, and the Intelligibility in Context Scale (ICS) questionnaire. In this paper, we present the dentofacial outcomes. Skeletal relationships were analysed using ANB, SNA, and SNB angles derived from cephalometric radiographs; dental arch relationships were evaluated using the 5-year or Modified GOSLON indices; and nasolabial aesthetics were assessed with the Asher–McDade index.

Results:

Data from 294 non-syndromic unilateral cleft lip and palate (UCLP) patients aged 5 (n=72), 12 (n=155), and 20 years (n=67) were analyzed. Reliability testing showed strong intra and inter-rater agreement for all indices and cephalometric data (ICC > 0.9). Nearly half of 12-year-olds (46.5%) and more than half of 20-year-olds (58.7%) demonstrated a Class III skeletal relationship (ANB < 0). Poor to very poor dental arch relationships (score > 3) were observed in 45%, 48%, and 43% of the 5-, 12-, and 20-year groups, respectively. Nasolabial aesthetic outcomes were rated poor to very poor in 24.3% of 5-year-olds, 25% of 12-year-olds, and 55.4% of 20-year-olds.

Conclusion:

This multicentric cleft outcomes audit in India demonstrated suboptimal skeletal, dental, and

nasolabial outcomes when benchmarked against international standards. These findings highlight an urgent need for the centralisation of cleft services, implementation of systematic national outcome monitoring, and adoption of standardised multidisciplinary care pathways to improve the quality and consistency of cleft management across India.

Keywords:
Cleft lip and palate, multicentric study, India, GOSLON index, skeletal relationship, nasolabial aesthetics, treatment outcomes.

Introduction:

Cleft lip and palate is one of the most common congenital anomaly. Approximately 28,000 to 33,000 children are born with cleft lip and/or palate annually in India.(1) These orofacial anomalies contribute significantly to the national disease burden, with an estimated 2.34 disability-adjusted life years (DALYs) lost and 3,821 years lived with disability (YLDs) per cohort, calculated up to 49 years of age.(2)The total unmet cleft treatment need in India is estimated at 72637 patients with cleft, with a range of 37% to 65% of cleft population.(3)(4)

Cleft treatment services in India remain predominantly urban-centric, with most comprehensive cleft care clinics situated in city-based hospitals.(1) In contrast, rural areas face significant disparities in access, with unmet treatment needs reported to be as high as 65%.(3) Studies have also identified a higher incidence of complete clefts among children in rural regions, where access to specialized care is limited.(5,6). The shortage of comprehensive cleft centers has resulted in a fragmented system of care delivery—characterized by limited availability of multidisciplinary teams, poor continuity of care, and inadequate follow-up. These deficiencies contribute to increased psychological distress among patients and families, as well as a higher incidence of revision surgeries.

India currently lacks a national cleft outcomes registry or any standardized system for data collection, a gap closely linked to the absence of centralized cleft care services. Centralization facilitates the development of national clinical guidelines, promotes uniform standards of care, and enhances the quality and consistency of treatment outcomes. Moreover, a centralized approach can significantly reduce the financial, psychological, and social burden on patients and their families by ensuring integrated, coordinated, and accessible care.

In the UK, cleft care was centralized further to the report from the Clinical Standard Advisory Group. This led to the closure of centres that failed to meet a minimum standard of care.(7) A

recent study pointed out that the centralisation in the UK has resulted in a huge improvement in the treatment outcomes and the care being provided to these patients. (8) Unfortunately, in India, there are no regulatory or surveillance organisations that can monitor the standard of care being rendered. Most of the cleft care services in India are unregulated with only 39% of cleft centres providing comprehensive care. Specialised speech and audiology consultation is available at less than 10% of the centres.(9)

The Cleft Care India Study (CCIS) was conceived as a nationwide, multicentric initiative to evaluate the standard of care provided to patients with non-syndromic unilateral cleft lip and palate across India. The study employed a cross-sectional design with prospective data collection, following standardized protocols for clinical and diagnostic record keeping. The primary objective was to assess outcomes at three key age points: 5, 12, and 20 years.

This paper presents the methodology of the CCIS along with an analysis of skeletal, dental, and nasolabial aesthetic outcomes in patients with unilateral complete cleft lip and palate. Additional outcome domains—including surgical interventions, speech and hearing, oral health, and psychological assessment—will be discussed in subsequent publications.

Subjects and Methods:

This study was approved by the XXXX Research Ethics Committee (approval no. 18/3/279/XXX/IEC/2020) on 18 March 2020. All participants provided written informed consent prior to enrolment in the study. This research was conducted ethically in accordance with the Declaration of Helsinki.

Data was collected according to the pre-set guidelines formulated for this study. Major cleft centers across India, whether from government organizations or external funding agencies like Smile Train, Akila Bharatha Mahila Seva Samaja (ABMSS) and Operation Smile, were invited for a face to face meeting and the study aims and objectives were explained. Centres that could not attend the meeting were later contacted by emails and phone calls and the interested centres were informed of the study aims and objectives through video conferencing. The participating centers were registered on a dedicated portal for the project (http://www.ccis.org.in/).

Development of Data Collection forms:

The first version of the data collection forms were formulated by the Study Steering Committee (SSC) with the help of the cleft collective UK team. The draft forms were discussed during the first face to face meeting between the SSC and the participating centre teams. The SSC members and centre teams that comprised the expert panelists included surgeons, orthodontists

and speech pathologists. Several rounds of discussions/controlled feedback were obtained before reaching a consensus/stability for each document.

Pilot testing:

The developed data collection forms, both online and paper version, were pilot tested from three centres. This allowed the clinicians and patients to provide feedback on the usability, understandability and comprehensiveness. Furthermore, the centres were requested to provide feedback on the online and paper version. Time taken to complete the forms were also noted. The findings from the pilot were considered and changes made as appropriate, with the final version of all the forms agreed by the SSC.

Data collection:

The pilot tested forms were made available on the Cleft Care India Site and all the included centres were requested to complete the forms online. In case of difficulty, the forms were available to be printed, and the completed forms could either be uploaded or sent to the DCC for entry on the site.

Each participating cleft care center was required to upload thirty case records of UCLP on the web portal. Fourteen cleft care centers provided data on patients with unilateral cleft lip and palate.

Study population:

The study population consisted of (i) non-syndromic patients with unilateral complete cleft lip and palate, including soft tissue band (Simonart's band) of less than 5 mm width and (ii) age group of 5, 12, or 20-years. Patients who had learning difficulties that prevent them from cooperating for data collection, and those who failed to provide consent were excluded.

Patients' ages had to closely align with each specified age group, with a margin of +6 months when presented at the centers. The specific age categories of 5, 12, and 20 were documented because, at 5 years, it was optimal for assessing surgical outcomes without any orthodontic intervention. At 12 years, the effectiveness of alveolar grafting procedures can be evaluated, and at 20, usually, all interventions are complete. Any patient reporting at the collaborating center and falling within the criteria below was included in the study. The data was recorded at the center irrespective of whether the patient was operated at that center or not.

Calibration:

Each center was assigned a unique center code, and none of the records included the names of the centers. One clinician from each center was trained to collect data. Training was carried out by the DCC and was held for all outcomes including photographs, impression taking, speech etc. However, no formal evaluation of the calibration exercise was conducted.

Data collection

Soft copies of a detailed booklet, specific for each age group was provided to the centres for data collection. The person recording the data, primarily orthodontists, worked at the center or was specially recruited for this task. The centers were supported with consumables or manpower for the smooth conduct of the project. The following records were taken of any patient who reported to the center and satisfying the inclusion criteria: Intraoral and extraoral photographs, study models, Radiographs (Lateral Cephalogram, OPG, Oblique Occlusal radiograph), surgical details form, oral health form, speech form, voice recordings, Intelligibility in context scale (ICS), and audiology form. All patients were recruited between December 2019 and June 2023. The records were uploaded on the web portal (www.ccis.org.in) in a standard format. The data collection training was conducted according to the protocol mentioned below.

Photographic record:

Seven extraoral facial photographs comprising frontal, frontal smile, left and right profile views, closeup nose, and lip, front whistling (to evaluate the functionality of the lips), and worm's eye view (to evaluate symmetry) were recorded. Six intra-oral photographs of frontal, right, and left buccal, maxillary occlusal view with cheek retractor, palatal view without retractor, and left canine view to capture the overjet were recorded with a DSLR Camera, macrolens and a ringflash. The photographs were standardised to comply with the recommended guidelines for photographs of children with cleft for audit purposes (10)

Radiographic Records:

Orthopantomogram (OPG), Lateral cephalogram, and oblique occlusal views were recorded for 12 and 20 year groups. Cephalograms were digitized and calibrated for digital cephalometric analysis. Lateral cephalograms were obtained to analyze sagittal skeletal, dento-alveolar, and dental relationships. OPG, being the essential diagnostic aid, was recorded to look at the number of teeth present and eruption status. An anterior oblique occlusal radiograph was taken to evaluate the cleft region for alveolar bone graft success.

Study models:

Alginate impressions were made and casts were poured in plaster. All study models were coded for assessor blinding. The casts were posted to the DCC where bases were formed and coding was reconfirmed. Study models were rated based on five point scoring criteria to evaluate the

dental arch relationships using 5-year old Index (11), GOSLON index (12) and modified GOSLON index (13) for the 5, 12, and 20-year-old age groups, respectively.

Surgical details questionnaires:

A detailed questionnaire recording the previous pre-surgical orthopedics, surgical sequence, and questions pertaining to primary lip repair, palate repair, and secondary surgical procedures was completed with the help of the surgeon at that center. If the patient was not operated at the center, the information was obtained from the previous records if the patient had preserved them, and by clinical examination of the patient. Anticipating that many patients would not have any previous records or any knowledge of the treatment, an option of "No information available" for each question was provided.

Oral health status:

To assess the oral health status of patients, their past history of dental care and current oral health status were evaluated and scored at the time of the patient's visit. The reading were either recorded in the hardcopy of the form or directly on the portal.

Speech evaluation:

A novel method was used to collect the speech samples from the participating centres to overcome the challenge of unavailability of trained speech pathologists to collect the speech data at all centres. The speech sample to be elicited was pre-recorded by one of the investigators presenting the stimuli with appropriate time gaps for the participant to respond. This pre-recorded template served as the stimulus for eliciting the speech sample and was played using the speakers of the laptop/desktop while the speech sample was recorded. This method of eliciting and recording speech samples was pilot tested for its effectiveness in speech sampling before the actual data was recorded.

The speech sample was elicited using the tasks of conversation, syllable repetition, counting numbers from 1-10 and 60-70, and repetition of 3–4-word sentences loaded with pressure consonants and nasal consonants. The guidelines specified in the universal reporting parameters (14) were followed except for the use of single word repetition, owing to the unavailability of standard word lists in all the 5 languages tested. All the participating centres were instructed and trained in the guidelines for recording.

The recorded samples were independently rated by two trained speech language pathologists with experience in listening to cleft speech. The universal parameters for reporting speech outcomes in individuals with cleft (14) was used for perceptual evaluation of speech.

Intelligibility evaluation:

The intelligibility in context scale (ICS) is a rating scale that asks parents on how well they understand their child's speech during daily communication. This scale includes seven questions specifying different members including family and friends on the understanding of their child's speech. This tool has been recommended as a screening tool for patients who may have speech disorders.(15) This data was recorded for 5 and 12-year-olds.

Audiological evaluation:

A questionnaire assessed parents' perceptions of their child's (5 and 12 year old age group) hearing ability, including comprehension efficacy and incidence of ear pain or infection. In contrast, self-perception of hearing ability was documented for the 20 year old age group with UCLP.

Psychological evaluation:

The effect of appearance on self-perception and confidence was captured using a pre-validated questionnaire. History of teasing and bullying by peers, social integration of the child with cleft, signs of any anxiety, role of cleft care team in psychological support, and parent's satisfaction among many other factors were captured for 5, 12 and 20 year olds using a pre-validated questionnaire.(16)

Assessment of skeletal relationship

The skeletal relationship in the sagittal dimension was analyzed by measuring SNA, SNB, and ANB angles on the lateral cephalograms of patients in 12 and 20 year age groups. The radiographs were calibrated to a standardized dimension to obtain uniformity. If the lateral cephalogram was not clear, it was not evaluated. All the radiographs were coded and any identification marks were removed. To assess inter-rater and intra-rater reliability 10% of the radiographs were re-evaluated with a two-week interval. All the evaluations were carried out by two orthodontists with a minimum of 5 years of experience, recruited specifically for this purpose. The anatomical landmarks were manually marked on digital Cephalograms using

AutoCeph® Software and automated measurements were recorded. The outcomes were categorized into two subsets; patients with ANB ≤0 and ANB >0 in all age groups.

Assessment of Dental Arch Relationship

Two orthodontist assessors with more than 5 years of experience performed the assessment individually on study models that were coded. The dental arch relationship was analyzed using the five-point ordinal scale, 5 year old Index (11) for study models of 5 year olds, GOSLON index (12) for study models of 12 year olds, and modified GOSLON index (13) for study models of 20 year olds. Dental models were analysed in sagittal, vertical and transverse planes and were graded as either Grade 1: Excellent, Grade 2: Good, Grade 3: Fair, Grade 4: Poor, and Grade 5: Very Poor. The models were analysed independently in duplicate by both assessors. For intrarater reliability, the 10% of the study models were randomly coded and analysed at two different time points with a two week interval. A score >3 (Score 4+5) was considered as a requirement for orthognathic surgery to correct the dentoalveolar malocclusions.

Assessment of Nasolabial Aesthetics

The standardised photographs of the patients uploaded on the portal were downloaded and saved in Microsoft PowerPoint with each slide containing the frontal and profile views of a patient. The photos were cropped with Adobe Photoshop C3 software. Frontal and profile photographs of patients were cropped in a trapezoidal manner so as to mask the surrounding features of the face, as in the previous studies.(17) The final photograph included the median canthi of both eyes and a small amount of sclera, and the trapezoid base was below the upper lip, including the commissure of the mouth and the full nose. Patient's photographs were assessed using Asher Mcdade's five-point Index.(17) Four parameters were assessed: Nasolabial form, symmetry of nose, shape of vermillion border, and nasal profile including upper lip. An average score was calculated and final rating assigned as very good (score 1), good (score 2), fair (score 3), poor (score 4), and very poor (score 5). The assessments were carried out by four orthodontists with a minimum of 5 years of experience. All the orthodontists were calibrated on scoring for nasolabial aesthetics prior to the assessment of study cases. Intra-rater reliability was calculated for all four assessors with 10% of the randomly picked images scored at two different time points two-weeks apart. For interpretation of outcome, the patients were divided into three categories based on average Nasolabial Score ≤ 2, >2 to 3, and >3 to 5.

Statistical analyses:

Data was descriptively reported for skeletal relationship, study model and Nasolabial index.

Skeletal Relationship: Mean (+SD) value of ANB, SNA, and SNB of all the patients in respective age groups (12- and 20- years) was calculated and categorised into ANB < 0 and ANB > 0.

Study model analysis: Mean (+SD) score of the 5-year index, GOSLON, and Modified GOSLON index for different age groups was calculated. The percentage distribution of patients with each score category and those with severity scores >3 were calculated for all groups.

Nasolabial index: The Nasolabial Index (Asher Mcdade et al)(17) has four parameters (Nasolabial form; Deviation of nose; Shape of Vermillion; and Nasal profile) and their scores in all age groups were calculated and compared using One-way ANOVA test. The average score of four parameters was calculated for each patient. The patients were graded into three categories (Score < 2; Score >2 to 3; Score >3 to 5)

Reliability analyses:

Goslon:

The Intra-class Correlation Coefficient (ICC) for test-retest reliability was based on two-way mixed effects, using means of multiple measurements and absolute agreement. Interpretation of ICC values <0.5 indicates poor reliability, 0.05 -.075 moderate reliability, 0.75 – 0.9 good reliability, >0.9 excellent reliability. Weighted Cohen's Kappa for inter-rater reliability of GOSLON Scoring of study models revealed strong agreement between the examiners while assessing the 5-year-olds (weighted kappa= 0.897) and almost perfect alignment while assessing the 12 and 20 year age groups (weighted kappa= 0.901 and 0.929) respectively.

Skeletal and dental:

Test-retest reliability results of SNA, SNB and ANB depicted excellent reliability (Cohen's Kappa for SNA= 0.995 C.I. 0.982 - 0.999; SNB= 0.973 C.I. 0.905 - 0.993; and ANB= 0.999 C.I. 0.995 – 1.00 respectively).

The Inter-rater reliability while assessing the dental arch relationship for 5-year-old was strong (kappa 0.897, SE 0.047), while assessing 12-year (kappa 0.901 SE 0.032) and 20 year age groups (kappa 0.929, SE 0.040) the examiners were in almost perfect agreement.

Results:

Fourteen cleft care centers from all over India participated in the study, and 294 patients were enrolled: 72 five-year-olds (Group I), 155 twelve-year-olds (Group II), and 67 twenty-year-olds (Group III).

Skeletal Relationship:

The mean SNA was 77.3° for 12-year-olds and 77.02^0 for 20-year-olds (Table 1). The mean SNB was 76.79° for 12-year-olds and 78.54^0 for 20-year-olds. ANB° values were negative (ANB≤0) in 46.5% of 12-year-olds, and 58.7% of 20-year-olds.(Table 2).

Table 1: Descriptive statistics of Cephalometric Parameters and Mean SNA, SNB and ANB of participants at all centres.

Cephalometric parameter	Group			
	12-year (N=142)		20-year (N=63)	
	Mean	S.D.	Mean	S.D.
SNA	77.3	4.81	77.02	5.48
SNB	76.79	3.9	78.54	4.2
ANB	0.55	4.17	-1.51	4.79

Table 2: Proportional Representation of Patients with Unilateral cleft lip and palate with ANB >0 and ANB <0.

Value of ANB^O (in degrees)	12-year (n) (%)	20-year (n) (%)
$ANB^O \leq 0$	66 (46.5%)	37 (58.7%)
$ANB^O > 0$	76 (53.5)	26 (41.3%)

Dental Arch Relationship:

The mean scores for the three age groups are described in Table 3. Score >3 depicts need for complex orthodontic treatment or orthognathic surgery to correct the dental arch

relationship. Nearly half of the patients with UCLP had a Score >3 in all three age groups (Table 4).

Table 3: Mean 5-year index, GOSLON, and Modified GOSLON scores for different 5, 12, and 20-year-old age groups, respectively.

Age	N	Mean	S.D.
5-year	69	3.22	1.096
12-year	147	3.44	0.923
20-year	65	2.94	1.273

Table 4: 5-year index, GOSLON, and Modified GOSLON scores$^{\alpha\beta\gamma}$ for 5-, 12-, and 20-year patients with UCLP respectively.

Scores	5-year	12-year$^{\beta}$	20-year$^{\gamma}$
Score 1	5	4	12
Score 2	13	16	13
Score 3	20	57	12
Score 4	24	54	23
Score 5	7	17	5
Score >3 (sum score 4+5)/N$^{\#}$	31/69 (44.92%)	71/148 (47.97%)	28/65 (43.07%)

$^{\alpha}$ 5-year-old index by Atack et al. ; $^{\beta}$ GOSLON Index; $^{\gamma}$ Modified GOSLON Index; # Severe malocclusion with poor prognosis indicating treatment by surgical intervention

Nasolabial Aesthetics:

The mean value of each parameter of Nasolabial Index is provided in Table 5. In the 5-year and 12-year age groups, around 25% of patients had a nasolabial index score of >3. In the 20-year-olds 55.4% had a score of >3 (Table 6).

Table 5: Comparison of mean values of each parameter of Asher McDade Nasolabial Index across the age group.

Group	N	Nasolabial form	Deviation of nose	Shape of Vermillion	Nasal profile
5-year	70	2.99 (1.04) $^{\alpha}$	2.54 (0.88)	2.54 (0.94)	2.51 (1.09)
12-year	152	3.01 (0.98) $^{\beta}$	2.34 (0.91) $^{\beta}$	2.57 (1.02) $^{\beta}$	2.74 (1.02)
20-year	65	3.48 (1.02) $^{\alpha,\beta}$	2.89 (0.98) $^{\beta}$	2.92 (0.95) $^{\beta}$	2.83 (1.16)

$^{\alpha,\beta}$ Similar super scripts for each feature denote a statistically significant difference among those age groups (One Way ANOVA)

Table 6: Average Nasolabial index scores for 5, 12, and 20-year olds with UCLP

Score range	5-year n (%)	12-year n (%)	20-year n (%)
Score ≤ 2	15 (21.4)	32 (21.1)	8 (12.3)
Score >2 to 3	38 (54.3)	82 (53.9)	21 (32.3)
Score >3 to 5	17 (24.3)	38 (25)	36 (55.4)
Total Patients analyzed (N)	70	152	65

Discussion:

Fourteen centers across India, mostly privately funded or aided by Non-governmental organizations, provided data for analysis. The results of the study showed that nearly half of the 5, 12 and 20 year olds have a poor to very poor (score 4 or 5) dental arch relationship and a quarter of 5 and 12 year olds and more than half of 20 year olds have poor to very poor (score 4 or 5) nasolabial aesthetics.

Each center was advised to provide 10 consecutive cases of the three specific age groups to avoid selection bias. However, due to the varied recall protocols, COVID issues, and restricted time for data collection, there were uneven submissions, with most centres providing more cases for the 12-year age group. Conventionally, patients' cleft protocols in India do not recall

patients until 7 years of age, resulting in a limited number of 5-year-olds. Similarly, most centre protocols tend to discharge patients by around 18 years of age or sooner if they decline orthognathic surgery.

Skeletal outcomes:

The skeletal relationship of jaws and the soft tissue convexity is crucial as it reflects on the quality of surgery performed. This study found that more than half (58%) of 20-year-olds and just under half (47%) of 12-year-olds had a Class III skeletal profile with an ANB of less than 0 degrees. A similar study in the United States, the Americleft study, evaluated 8-year-olds (ranging from 6 to12 years) showed that the ANB ranged from a minimum of +2.40 °at one centre to a maximum of +5.42 ° at another.(18) The difference in results could be because of the age difference at the time of evaluation where the Americleft study reported on a much younger population. The mean ANB in this study was 0.5^0 for 12 year olds and -1.5^0 for 20 year olds. Our results are similar to the CSAG study, which reported a mean ANB of 0.4^0, with 50% of patients having a marked class III and 70% of patients having a class III skeletal relationship.(19) The results from the Eurocleft study was similar to the Americleft with an maxillomandibular discrepancy (ss-n-sm) ranging from 1.6^0 to 4.4^0.(20)

Dentoalveolar outcomes:

The results of the present study for dentoalveolar outcomes are in accordance with the results from the Americleft study. The Americleft study reported a mean GOSLON score ranging from 2.63 to 3.66 for 6 to12-year-olds.(21) In the CSAG study, 37% of 5 year-olds and 39% of 12-year-olds reported poor or very poor dental relationship, compared to 45-48% in the present study. However, the CCUK study had the significantly less percentage of poor or very poor relationships (19%), indicating the improvement with inter-centre comparisons over time.(22) (23) In the Eurocleft study, the mean scores varied from 2.5-3.5 for 9 year old, from 2.1 to 3.2 for 12 year olds and from 1.7 to 3.4 for 20 year olds.(24)

Although measuring dentoalveolar outcomes is not a true measure of the skeletal arrangement and the need for future midface surgery, previous studies have pointed out that there is a good correlation between the dentoalveolar features and the craniofacial presentation. From the results of the study, although in accordance with the Americleft and CSAG reports, it is evident that there is an urgent need to improve the outcomes for cleft patients. The results from 20 year olds is worse for Indian patients with cleft compared with the American and European cleft centre outcomes.

Nasolabial Aesthetics:

There was a substantial discrepancy between the nasolabial aesthetic outcomes observed in the present study and those reported in previous cleft outcome evaluations. The initial UK-based Clinical Standards Advisory Group (CSAG) audit from 1998 reported that 27.6% of patients were rated as having poor or very poor facial appearance. A more recent audit of the same population (CCUK) showed a modest improvement, with 21.6% of five-year-olds receiving similar poor ratings for facial appearance.(25) In the CCUK only the frontal photographs were evaluated, whereas the present study evaluated both the frontal and profile views, offering a more comprehensive analysis. Among these, NL form emerged as the most adversely affected parameter, particularly in the 12- and 20-year-old cohorts. Similar findings were noted in the Eurocleft study, which reported worse nasolabial scores in older age groups, particularly among 17-year-olds. Consistently, across the Eurocleft and Americleft studies, nasolabial form has been rated as the most negatively affected feature in cleft outcomes.

Challenges and Methodology in Cleft Outcome Auditing Across Indian Centers

A major challenge encountered in this project was the general reluctance of Indian cleft teams to participate, largely due to limited interest in research and outcome evaluation. India has over 200 cleft care centers; however, only a small proportion of these are equipped with multidisciplinary facilities. The majority are supported by non-governmental organizations (NGOs), while only a few receive public funding.

Although many NGOs require basic record-keeping as part of their operational support, the overall standard of documentation remains low. Furthermore, there is significant variation in both the timing and the nature of the data collected, rendering inter-center comparisons impractical. A considerable number of centers are under-resourced or unaware of the importance of systematic auditing for quality assurance.(refer our questionnaire paper) Critically, there is no national registry or centralized database for reporting cleft outcomes in India.

The poor results obtained in this study, combined with the absence of a national database and limited clinician engagement with data-driven audit, highlight the urgent need to establish a national audit framework for cleft care in India.

In this study, data were collected prospectively. Participating centers were instructed to enroll consecutive patients who met the inclusion criteria at the time of presentation. Since no center routinely followed up with patients at 5 and 20 years of age, these patients were retrospectively identified through hospital records and invited to attend for the purpose of outcome auditing.

The objective of this study mirrored that of the Eurocleft, Americleft, and CCUK collaborations: to assess cleft outcomes and benchmark them against international standards. However, inter-center comparisons were not attempted due to two limiting factors, variable sample sizes across centers and the small overall number of cases, which precluded meaningful statistical analysis.

Nevertheless, the findings offer valuable insight into the current state of cleft care outcomes across India. A larger, future study involving more centers and greater sample sizes would facilitate inter-center comparisons and empower individual units to assess their performance and revise clinical protocols as needed.

Conclusion:

1. Fourteen centers across India, mostly privately funded or aided by Non-governmental organizations, provided data for analysis.
2. The results of the study showed that nearly half of the 5, 12 and 20 year olds have a poor to very poor (score 4 or 5) dental arch relationship.
3. Nearly half of 12 year olds (46%) and more than half of 20 year olds (58%) have an ANB of less than or equal to zero degrees (Class III skeletal relationship).
4. A quarter of 5 and 12 year olds and more than half of 20 year olds have poor to very poor (score 4 or 5) nasolabial aesthetics.
5. Further audits involving a greater number of centers across India and larger sample sizes are necessary to enable meaningful intercenter comparisons and to inform clinicians, service providers, and policymakers in the development of evidence-based, standardised cleft care pathways.

Declarations:

Ethical approval and consent:

This study was approved by the XXXX Research Ethics Committee (approval no. 18/3/279/XXX/IEC/2020) on 18 March 2020. All participants provided written informed consent prior to enrolment in the study.

Funding Statement:

This project was funded by the Scheme for Promotion of Academic and Research Collaboration (SPARC), Government of India and UK India Education and Research Initiative (UKIERI), United Kingdom (Grant No: P1025)

Conflict of Interest

NIL

References:

1. Mossey P, Little J. Addressing the challenges of cleft lip and palate research in India. Indian Journal of Plastic Surgery. 2009 Oct 1;42(SUPPL. 1).

2. Khajanchi MU, Shah H, Thakkar P, Gerdin M, Roy N. Unmet burden of cleft lip and palate in rural gujarat, india:A population-based study. World J Surg [Internet]. 2015 Jan 1 [cited 2025 Jun 30];39(1):41–6. Available from: /doi/pdf/10.1007/s00268-014-2628-6

3. Stewart BT, Carlson L, Hatcher KW, Sengupta A, Vander Burg R. Estimate of unmet need for cleft lip and/or palate surgery in India. JAMA Facial Plast Surg [Internet]. 2016 Sep 1 [cited 2025 Jun 30];18(5):354–61. Available from: /doi/pdf/10.1001/jamafacial.2016.0474?download=true

4. Balaji SM. Burden of orofacial clefting in India, 2016: A global burden of disease approach. Vol. 8, Annals of Maxillofacial Surgery. Wolters Kluwer Medknow Publications; 2018. p. 91–100.

5. Dvivedi J, Dvivedi S. A clinical and demographic profile of the cleft lip and palate in Sub-Himalayan India: A hospital-based study. Indian Journal of Plastic Surgery. 2012 Jan;45(1):115–20.

6. Upadhyaya D, Reddy G, … RM… of CLP and, 2017 undefined. Impact of educational and socioeconomic status of parents on healthcare access in cleft patients. journals.lww.comDN Upadhyaya, GP Reddy, RK Mishra, AK SinghJournal of Cleft Lip Palate and Craniofacial Anomalies, 2017•journals.lww.com [Internet]. [cited 2025 Jun 30]; Available from: https://journals.lww.com/cpcs/fulltext/2017/04020/impact_of_educational_and_socioeconomic_status_of.3.aspx

7. Sandy J, Williams A, Mildinhall S, Murphy T, Bearn D, Shaw B, et al. The Clinical Standards Advisory Group (CSAG) Cleft Lip and Palate Study. Br J Orthod [Internet]. 1998 [cited 2025 Jun 30];25(1):21–30. Available from: https://pubmed.ncbi.nlm.nih.gov/9547971/

8. Ness AR, Wills AK, Waylen A, Al-Ghatam R, Jones TEM, Preston R, et al. Centralization of cleft care in the UK. Part 6: a tale of two studies. Orthod Craniofac Res [Internet]. 2015 Nov 1 [cited 2025 Apr 3];18:56–62. Available from: https://onlinelibrary.wiley.com/doi/full/10.1111/ocr.12111

9. Batra P, Sybil D, Izhar A, Batra P, Thiruvenkatachari B. Standard of Care for Patients With Cleft Lip and Palate in India—A Questionnaire-Based Study. Cleft Palate Craniofacial Journal. 2023 May 1;60(5):536–43.

10. Jones M, Medicine MCJ of AM in, 2004 undefined. Implementation of standardized medical photography for cleft lip and palate audit. Taylor & FrancisM Jones, M CadierJournal of Audiovisual Media in Medicine, 2004•Taylor & Francis [Internet]. 2009 Dec [cited 2025 Apr 3];27(4):154–60. Available from: https://www.tandfonline.com/doi/abs/10.1080/01405110500038837

11. Atack N, Hathorn I, Mars M, Sandy J. Study models of 5 year old children as predictors of surgical outcome in unilateral cleft lip and palate. Eur J Orthod. 1997;19(2):165–70.

12. Mars M, Plint D, Houston W, … OBTC palate, 1987 undefined. The Goslon Yardstick: a new system of assessing dental arch relationships in children with unilateral clefts of the lip and palate. europepmc.orgM Mars, DA Plint, WJ Houston, O Bergland, G SembThe Cleft palate journal, 1987•europepmc.org [Internet]. [cited 2025 Apr 3]; Available from: https://europepmc.org/article/med/3479277

13. Mølsted K, Brattström V, Prahl-Andersen B, Shaw WC, Semb G. The Eurocleft study: Intercenter study of treatment outcome in patients with complete cleft lip and palate. Part 3: Dental arch relationships. Cleft Palate-Craniofacial Journal. 2005 Jan;42(1):78–82.

14. Henningsson G, Kuehn DP, Sell D, Sweeney T, Trost-Cardamone JE, Whitehill TL. Universal parameters for reporting speech outcomes in individuals with cleft palate. Cleft Palate-Craniofacial Journal. 2008 Jan;45(1):1–17.

15. McLeod S, Crowe K, Shahaeian A. Intelligibility in context scale: Normative and validation data for English-speaking preschoolers. Lang Speech Hear Serv Sch. 2015 Jul 1;46(3):266–76.

16. Waylen R Ness A K Wills M Persson N Rumsey J R Sandy AA, Waylen A, Ness AR, Wills AK, Sandy JR, R Sandy PJ, et al. Cleft Care UK study. Part 5: child psychosocial outcomes and satisfaction with cleft services. Wiley Online LibraryA Waylen, AR Ness, AK Wills, M Persson, N Rumsey, JR SandyOrthodontics & Craniofacial Research, 2015•Wiley Online Library [Internet]. 2015 Nov 1 [cited 2025 Apr 4];18:47–55. Available from: https://onlinelibrary.wiley.com/doi/abs/10.1111/ocr.12113

17. Asher-Mcdade C, Roberts C, Shaw WC, Gallager C. Development of a Method for Rating Nasolabial Appearance in Patients with Clefts of the Lip and Palate. The Cleft Palate-Craniofacial Journal. 1991 Oct;28(4):385–91.

18. Daskalogiannakis J, Mercado A, Russell K, Hathaway R, Dugas G, Long RE, et al. The americleft study: An inter-center study of treatment outcomes for patients with unilateral cleft lip and palate part 3. Analysis of craniofacial form. Cleft Palate-Craniofacial Journal [Internet]. 2011 May [cited 2025 Apr 21];48(3):252–8. Available from: https://journals.sagepub.com/doi/full/10.1597/09-185.1

19. Williams MsAC, Bearn MrD, Mildinhall MsS, Murphy MsT, Sell DrD, Shaw DrWC, et al. Cleft Lip and Palate Care in the United Kingdom—The Clinical Standards Advisory Group (CSAG) Study. Part 2: Dentofacial Outcomes and Patient Satisfaction. The Cleft Palate-Craniofacial Journal [Internet]. 2001 Jan [cited 2025 Apr 21];38(1):24–9. Available from: https://journals.sagepub.com/doi/abs/10.1597/1545-1569_2001_038_0024_clapci_2.0.co_2

20. Brattström V, Mølsted K, Prahl-Andersen B, Semb G, Shaw WC. The Eurocleft Study: Intercenter Study of Treatment Outcome in Patients with Complete Cleft Lip and Palate. Part 2: Craniofacial Form and Nasolabial Appearance. The Cleft Palate-Craniofacial Journal [Internet]. 2005 Jan [cited 2025 Apr 21];42(1):69–77. Available from: https://journals.sagepub.com/doi/abs/10.1597/02-119.2.1

21. Hathaway R, Daskalogiannakis J, Mercado A, Russell K, Long RE, Cohen M, et al. The americleft study: An inter-center study of treatment outcomes for patients with unilateral cleft lip and palate part 2. Dental arch relationships. Cleft Palate-Craniofacial Journal [Internet]. 2011 May [cited 2025 Apr 21];48(3):244–51. Available from: https://journals.sagepub.com/doi/full/10.1597/09-181.1

22. Al-Ghatam E M Jones A J Ireland N E Atack O Chawla S Deacon L Albery A R M Cobb J Cadogan S Leary A Waylen A K Wills B Richard H Bella A R Ness J R Sandy RT, Al-Ghatam R, M Jones TE, Ireland AJ, Atack NE, Chawla O, et al. Structural outcomes in the Cleft Care UK study. Part 2:

dento-facial outcomes. Orthod Craniofac Res [Internet]. 2015 [cited 2025 Apr 21];18:14–24. Available from: https://onlinelibrary.wiley.com/doi/10.1111/ocr.12109

23. Williams MsAC, Bearn MrD, Mildinhall MsS, Murphy MsT, Sell DrD, Shaw DrWC, et al. Cleft Lip and Palate Care in the United Kingdom—The Clinical Standards Advisory Group (CSAG) Study. Part 2: Dentofacial Outcomes and Patient Satisfaction. The Cleft Palate-Craniofacial Journal [Internet]. 2001 Jan [cited 2025 Apr 21];38(1):24–9. Available from: https://journals.sagepub.com/doi/abs/10.1597/1545-1569_2001_038_0024_clapci_2.0.co_2

24. Mølsted K, Brattström V, Prahl-Andersen B, Shaw WC, Semb G. The Eurocleft Study: Intercenter Study of Treatment Outcome in Patients with Complete Cleft Lip and Palate. Part 3: Dental Arch Relationships. The Cleft Palate-Craniofacial Journal [Internet]. 2005 Jan [cited 2025 Apr 21];42(1):78–82. Available from: https://journals.sagepub.com/doi/abs/10.1597/02-119.3.1

25. Morrant DG, Shaw WC. Use of Standardized Video Recordings to Assess Cleft Surgery Outcome. The Cleft Palate-Craniofacial Journal. 1996 Mar;33(2):134–42.