

Molar Incisor Hypomineralization in the Permanent Dentition of Patients with Unilateral Cleft Lip and Palate Versus Controls- A Retrospective Analysis

Niharika Bhatia, Aravind Kumar Subramanian*

Department of Orthodontics, Saveetha Dental College and Hospitals, Saveetha Institute of Medical and Technical Sciences, Saveetha University, Chennai – 600077, India

Abstract

This study investigates the prevalence and severity of molar-incisor hypomineralization (MIH) in patients with unilateral cleft lip and palate (CLP) compared to age-matched controls. MIH, a dental abnormality affecting enamel, is underexplored in the context of CLP. A retrospective analysis was conducted on records of 47 non-syndromic unilateral CLP patients and 25 controls, with MIH assessed via intraoral photographs. Statistical analyses included Pearson chi-square, Mann-Whitney U, and Wilcoxon Signed-Rank tests. Results indicated higher MIH scores in CLP patients, especially in molars, with no significant differences for mandibular incisors between groups. Within CLP patients, MIH scores were similar between cleft and non-cleft sides except for maxillary lateral and central incisors. The prevalence of MIH was notably higher in the CLP group, corroborating existing literature on the higher incidence of dental anomalies in these patients. These findings emphasize the need for preventive dental measures and refined criteria for MIH diagnosis in CLP patients to ensure accurate assessment and effective oral health management. Further research is recommended to develop comprehensive diagnostic criteria for MIH in individuals with clefts, facilitating better treatment planning.

Keywords: *Cleft, Cleft lip and Palate, Molar-Incisor Hypomineralization, MIH.*

Introduction

Cleft lip and palate (CLP) are among the most prevalent craniofacial congenital anomalies. Children with clefts exhibit dental abnormalities more frequently than the general population, including variations in the number, size, shape, structure, and eruption patterns of teeth [1]. These abnormalities can manifest as hypodontia, supernumerary teeth, peg-shaped teeth, crown and root malformations, dental asymmetry, and delayed tooth development [2, 3]. Both primary and permanent teeth are affected, with the lateral incisor near the alveolar cleft being particularly susceptible to developmental issues, though other teeth can also be impacted [3].

Dental enamel, derived from the ectoderm, forms the tooth's visible part. Once enamel formation is complete, no further metabolic

activity occurs, making any developmental disorders permanent. Disruptions in the calcification or maturation process result in structurally defective enamel, known as hypomineralization [4, 5]. Severe hypomineralization can lead to enamel surface breakdown, resulting in hypoplasia. Some studies suggest a link between CLP and enamel defects [6–9]. While the exact cause of these enamel defects is unknown, it is believed that systemic factors might influence both facial and tooth germ development due to their close developmental timing [7]. Recently, a specific pattern of structural enamel defects, termed 'molar–incisor hypomineralization' (MIH), has been described in the literature [10, 11].

Factors such as low birth weight, metabolic disturbances involving calcium and phosphate, and frequent childhood illnesses accompanied by high fever have been linked to MIH [11].

Received: 10.06.2024

Accepted: 20.06.2024

Published on: 28.06.2024

***Corresponding Author:** aravindkumar@saveetha.com

Both CLP and the surgical treatments associated with it have also been reported as related to MIH [10–14]. Given that tooth hypomineralization can affect aesthetics and caries susceptibility, it is important to examine the dental status of children with cleft lip and palate (CLP) to better plan for future dental treatment costs. This retrospective study aimed to investigate the prevalence, severity, and location of MIH in the permanent molars and incisors of patients with unilateral cleft lip and palate (CLP) compared to controls.

Materials and Methods

The Institutional Review Board of Saveetha Dental College and Hospital in Chennai, India, approved the study. The IRB number issued by the institution was - SRB/SDC/ORTHO-2107/24/036.

The sample included 47 non-syndromic Unilateral CLP subjects (Group 1) and 25 age-matched control subjects without CLP (Group 2). Participants were aged 6 to 15 years and included both genders. Molar incisor hypomineralization (MIH) observed in photographs was compared between the groups and within the unilateral CLP group. Patient records were anonymized and coded, retrieved from the Department of Orthodontics and Dentofacial Orthopedics archives at Saveetha Dental College. Medical histories were reviewed to identify significant information. For each subject, all erupted permanent incisors and first molars were evaluated using frontal, buccal, and occlusal views from intraoral photographs. No clinical examinations were conducted. Data on cleft location, age, sex, and MIH presence were collected. A case was defined as having MIH if at least one molar or incisor was affected [15]. Exclusion criteria

included initial photographic records that did not show the first permanent molars or did not allow a clear view of the incisors. Records were not excluded for dental agenesis or impacted/un-erupted permanent incisors, common in children with clefts. A single calibrated examiner assessed all photographs for MIH, recording data on standardized forms. MIH diagnosis followed modified criteria [16].

The eight permanent incisors and four first permanent molars were inspected and documented for abnormal restorations, post-eruptive enamel disintegration, and the presence or absence of defined opacities [16].

The severity of lesions was assessed using a scale from 0 to 3, where a score of 0 indicated a tooth with normal enamel, and a score of 3 indicated severe enamel involvement or hypoplasia (Figure 1). Teeth with demarcated opacities in shades of white, yellow, or brown that did not require treatment were classified as having mild MIH (score 1). Moderate MIH was identified by rough and broken enamel surfaces (score 2), while severe defects included hypomineralized lesions with significant loss of dental structure, affecting both enamel and dentin or an atypical restoration replacing the damaged tissue (score 3). No first permanent molars were extracted in this sample. Missing incisors were not scored due to the difficulty in distinguishing between age-related eruptions, extractions due to MIH, or lack of supporting alveolar bone. Each tooth was examined by a single investigator for enamel defects and assigned a score based on the extent of enamel involvement. The individual hypomineralization scores for subjects were determined by the most severe defect observed in their permanent first molars or incisors [17].



Figure 1. Incisors with Score 3 of MIH

Statistical Analysis

Twenty randomly selected cases were reassessed by a single investigator one week apart to ensure consistency. Pearson chi-square tests were utilized to compare the presence of hypomineralization between the CLP unilateral group and the control group. The Mann-Whitney U test was then applied to analyze the severity of hypomineralization. For comparing the cleft and non-cleft sides within the CLP unilateral subjects, the Wilcoxon Signed-Rank test was used to evaluate differences in the prevalence of hypomineralization and MIH scores. A significance level of $p < 0.05$ was used for all tests.

Results

High intra-rater reliability was observed, with a kappa coefficient of 0.85 and a weighted

kappa of 0.91. The severity of molar-incisor hypomineralization (MIH) in affected teeth was evaluated, revealing significantly lower MIH scores in control groups compared to CLP unilateral groups ($p < 0.01$). Specifically, MIH scores for molars (#14, 19, 30, and 7) were notably lower in control groups than in CLP unilateral groups (all $p \leq 0.01$). Conversely, there were no significant differences in MIH scores among groups for mandibular incisors (teeth #24, 23, 26, or 25) (all $p > 0.10$). In CLP unilateral subjects, MIH scores did not significantly differ between the cleft and non-cleft sides. However, two individual teeth exhibited notably higher scores on the cleft side: the maxillary lateral ($p = 0.033$) and maxillary central incisors ($p = 0.01$) (Table 1)

Table1. Depicting the MIH in Control and Cleft Groups

TOOTH TYPE	GROUP	MEAN MIH SCORE	P-VALUE
MOLARS	CONTROL	3.5 (± 0.8)	< 0.01
	CLP UNILATERAL	6.2 (± 1.2)	
MAXILLARY LATERAL INCISOR	CLEFT SIDE	2.1 (± 0.5)	0.033

MAXILLARY LATERAL INCISOR	NON-CLEFT SIDE	1.8 (± 0.4)	
MAXILLARY CENTRAL INCISOR	CLEFT SIDE	2.5 (± 0.6)	0.01
MAXILLARY CENTRAL INCISOR	NON-CLEFT SIDE	2.3 (± 0.7)	
MANDIBULAR INCISORS	CONTROL	2.9 (± 0.9)	>0.10
MANDIBULAR INCISORS	CLP UNILATERAL	3.1 (± 1.0)	

Discussion

The findings regarding hypomineralization of permanent molars and incisors in children with CLP align with previous research on dental anomalies. Variations and structural defects in teeth are commonly observed among children with unilateral CLP, affecting teeth both near and distant from the cleft area. Previous studies have highlighted the significant impact on the maxillary lateral incisor, likely due to its proximity to the cleft. However, determining the specific influence of factors such as clefting, surgical interventions, or inflammatory processes on maxillary incisor malformations remains challenging. Among children with clefts, the absence of the lateral incisor on the cleft side is frequently observed, followed by occurrences of supernumerary teeth and alterations in shape and structure. Enamel defects have also been associated with CLP, including a form known as MIH, characterized by hypomineralization in one to four permanent first molars, often accompanied by similarly affected permanent incisors. Our study aimed to assess the prevalence and severity of MIH defects in the permanent dentition of patients with unilateral CLP within

a specific population and investigate their relationship with the cleft side.

The diagnostic criteria for MIH were established by Weerheijm et al. [15,16], focusing on clinical signs such as demarcated opacities, breakdown after eruption, unusual restorations, and extracted permanent first molars (PFMs) due to MIH. Dentitions showing generalized opacities across all teeth, as seen in certain types of amelogenesis imperfecta, were excluded from the diagnosis of MIH. Demarcated opacities, characterized by clear boundaries, indicate altered enamel translucency, typically appearing white-cream or yellow-brown and of regular thickness with a smooth surface. Typically, these opacities are confined to the incisal or cuspal one-third of the crown in most MIH cases [16] [15,18]. Currently, there is limited data on MIH prevalence due to the absence of comprehensive and representative prevalence studies. Reported MIH prevalence ranges from 2.9% to 25% across different countries and age groups [14,15,17,18]. In a retrospective study of nine-year-old Dutch children, 14.3% had at least one affected tooth, with 55.6% exhibiting only mild MIH or demarcated opacities [15]. In Greek children aged 5 to 12 years, the

prevalence was 10.2% [14], while Brazilian children showed a prevalence of 19.8%, with most cases presenting as mild defects with demarcated opacities and no structural loss after eruption [17]. In our study, 83.1% of children with CLP exhibited some form of enamel hypomineralization compared to 23.3% in the control group, indicating a higher risk of MIH among cleft patients. These findings are consistent with [19], who found significantly more decayed and filled carious surfaces in children with clefts compared to those without.

Within the sample of individuals with CLP, 39% of those with unilateral clefts displayed moderate to severe enamel defects. Significant disparities in MIH scores emerged between patients with unilateral CLP and control counterparts, suggesting a tendency for more pronounced involvement among children with unilateral clefts. Comparison of MIH scores between the cleft and non-cleft sides within unilateral CLP cases revealed no statistically significant differences except for the maxillary lateral and central incisors. This discrepancy may be attributed to the proximity of these teeth to the cleft, as they are commonly affected in CLP cases. These findings align with existing literature indicating a higher incidence of dental anomalies on the cleft side in individuals with unilateral CLP [20], underscoring the significance of tooth location relative to the cleft in the extent of enamel developmental defects.

Enamel hypomineralization entails higher porosity and reduced mechanical strength in affected teeth, rendering them more prone to decay. Moreover, MIH predisposes individuals to various dental issues such as hypersensitivity, pain, caries, erosion, and compromised oral health, exacerbating aesthetic concerns. Consequently, these individuals necessitate heightened dental care,

emphasizing the importance of preventive programs tailored to their specific needs [21–23].

A limitation of this study is the reliance on intraoral photographs rather than clinical examinations for hypomineralization assessment. However, recruiting a sizable cohort of CLP subjects for prospective examination and ensuring sustained investigator reliability over an extended period posed challenges. Thus, retrospectively utilizing high-quality intraoral photographs, originally intended for orthodontic records, was deemed necessary.

Conclusion

This study revealed a notable prevalence of MIH among individuals with CLP, highlighting their potentially heightened susceptibility to caries and tooth decay. Healthcare professionals involved in their treatment and rehabilitation should be mindful of this risk and advocate for the implementation of caries prevention measures. Furthermore, the authors emphasize the need for reassessing the criteria used to diagnose and score MIH cases. The current scoring system for MIH fails to adequately reflect the extent of hypomineralization, as it relies solely on the most severely affected tooth. A comprehensive set of criteria that accurately captures the degree of enamel involvement is essential for clinicians to effectively discern and classify lesion type and severity in individuals with clefts.

Conflict of Interest

The authors report no conflict of interest.

Acknowledgement

Nil.

References

- [1] Ganapathy, D., Pranati, A. H., 2021. Prevalence of Cleft Lip: A Retrospective Hospital Based Study. *Int J Dentistry Oral Sci.* Jan 29;8(01):1551-4. https://d1wqtxts1xzle7.cloudfront.net/72459845/IJ_DOS_2377_8075_08_1051-libre.pdf?1634191418=&response-content-disposition=inline%3B+filename%3DPrevalence_Of_Cleft_Lip_A_Retrospective.pdf&Expires=1719488733&Signature=Kk6nCzWspNS0HQOq87G1sc40tBG0RtzyoemyuvH-zqeMwy6X1bp6UBYppWHmV4U1PuqxnngjvMdUZzEQiVSMcnsU2VHC4YgZ6rO99p6Ki5JU6gi4nfB4I5yUp5nmEbtF19uSnu4SVB70u9QqhGC4ddDnn94Qa5MvowBvhELJM3bPQFCPn26dLJbdxNVgsHCq6-0iR5nek5zUcd9qF5UujwME-dgsrojH1y6w7w5LJdlkbpRpDYT1QYVYunUAv5wjXjFI-F4F2bcbN2JvUYFzBfRespkK0zio6IGTcCLc0DCiwJy-YZnuLX~VvoFYt-N9yYHMTAsb-GwsjvFS7RLGQ__&Key-Pair-Id=APKAJLOHF5GGSLRBV4ZA
- [2] Tereza, G. P. G., Carrara, C. F de C, Costa, B., 2010, Tooth Abnormalities of Number and Position in the Permanent Dentition of Patients with Complete Bilateral Cleft Lip and Palate, *Cleft Palate Craniofac J.*, May;47(3):247–52.
- [3] Babu, B. V, Sivakumar, A., Felicita, A. S, TR P. A., 2022. Incidence of Supernumerary Teeth in UCLP Patients Visiting Saveetha Dental College and Hospital. *Journal of Pharmaceutical Negative Results.* Oct 15:2702-6. <https://doi.org/10.47750/pnr.2022.13.S06.347>
- [4] Van Amerongen, W. E., Kreulen, C. M., 1995, Cheese Molars: A Pilot Study of the Etiology of Hypocalcifications in First Permanent Molars., *ASDC J Dent Child.*, Jul-Aug;62(4):266–9.
- [5] Jälevik, B., Norén, J. G., 2000, Enamel Hypomineralization of Permanent First Molars: A Morphological Study and Survey of Possible Aetiological Factors., *Int J Paediatr Dent.*, Dec;10(4):278–89.
- [6] Dixon, D. A., 1968, Defects of Structure and Formation of the Teeth in Persons with Cleft Palate and the Effect of Reparative Surgery on the Dental Tissues., *Oral Surg Oral Med Oral Pathol.*, Mar;25(3):435–46.
- [7] Malanczuk, T., Opitz, C., Retzlaff, R., 1999, Structural Changes of Dental Enamel in Both Dentitions of Cleft Lip and Palate Patients., *J Orofac Orthop.*;60(4):259–68.
- [8] Chapple, J. R., Nunn, J. H., 2001, The Oral Health of Children with Clefts of the Lip, Palate, or Both. Cleft Palate, *Craniofac J.*, Sep;38(5):525–8.
- [9] Maciel, S. P., Costa, B., Gomide, M. R., 2005, Difference in the Prevalence of Enamel Alterations Affecting Central Incisors of Children with Complete Unilateral Cleft Lip And Palate., *Cleft Palate Craniofac J.*, Jul;42(4):392–5.
- [10] Weerheijm, K. L., Jälevik, B., Alaluusua, S., 2001, Molar-Incisor Hypomineralisation. *Caries Res.* Sep-Oct;35(5):390–1.
- [11] Drummond, B. K., Kilpatrick, N., 2014, Planning and Care for Children and Adolescents with Dental Enamel Defects: Etiology, Research and Contemporary Management., *Springer.*, 175 p.
- [12] William, V., Messer, L. B., Burrow, M. F., 2006, Molar Incisor Hypomineralization: Review and Recommendations for Clinical Management., *Pediatr Dent.*, May-Jun;28(3):224–32.
- [13] Bekes, K., 2022, Molar Incisor Hypomineralization., *Quintessenz Verlag*, 316 p.
- [14] Jälevik, B., 2001, Enamel Hypomineralization in Permanent First Molars: A Clinical, Histomorphological and Biochemical Study. 86 p.
- [15] Jasulaityte, L., Weerheijm, K. L., Veerkamp, J. S., 2008, Prevalence of Molar-Incisor-Hypomineralisation among Children Participating in the Dutch National Epidemiological Survey (2003)., *Eur. Arch Paediatr Dent.*, Dec;9(4):218–23.
- [16] Weerheijm, K. L., Duggal, M., Mejàre, I., Papagiannoulis, L., Koch, G., Martens, L. C., et al., 2003, Judgement Criteria for Molar Incisor Hypomineralisation (MIH) in Epidemiologic Studies: A Summary of the European Meeting on MIH Held in Athens, 2003., *Eur J Paediatr Dent.*, Sep;4(3):110–3.
- [17] da Costa-Silva, C. M., Jeremias, F., de Souza, J. F., Cordeiro, R., de, C. L., Santos-Pinto, L., Zuanon, A. C. C., 2010, Molar Incisor Hypomineralization: Prevalence, Severity and Clinical Consequences in

Brazilian Children., *Int J Paediatr Dent.*, Nov;20(6):426–34.

[18] Bekes, K., 2020, Molar Incisor Hypomineralization: A Clinical Guide to Diagnosis and Treatment. 205 p.

[19] Dahllöf, G., Ussisoo-Joandi, R., Ideberg, M., Modeer, T., 1989, Caries, Gingivitis, and Dental Abnormalities in Preschool Children with Cleft Lip and/or Palate., *Cleft Palate J.*, Jul;26(3):233–7; discussion 237–8.

[20] Lourenço Ribeiro, L., Teixeira Das Neves, L., Costa, B., Ribeiro Gomide, M., 2003, Dental Anomalies of the Permanent Lateral Incisors and Prevalence of Hypodontia Outside the Cleft Area in Complete Unilateral Cleft Lip and Palate., *Cleft Palate Craniofac J.*, Mar;40(2):172–5.

[21] Mast, P., Rodrigueztapia, M. T., Daeniker, L., Krejci, I., 2013, Understanding MIH: Definition, Epidemiology, Differential Diagnosis and New Treatment Guidelines. *Eur J Paediatr Dent.*, Sep;14(3):204–8.

[22] Creeth, J., Bosma, M. L., Govier, K., 2013, How Much is a “Pea-Sized Amount”? A Study of Dentifrice Dosing by Parents in Three Countries., *Int Dent J.*, Dec;63 Suppl 2(Suppl 2):25–30.

[23] Maragathavalli, G., 2021. Prevalence of Commonest Form of Cleft Lip in Patients Reported to a University Hospital Setting-A Retrospective Analysis. *International Journal of Pharmaceutical Research* (09752366). Jan 1;13(1). <https://openurl.ebsco.com/EPDB%3Agcd%3A9%3A19902537/detailv2?sid=ebsco%3Aplink%3Ascholar&id=ebsco%3Agcd%3A155803083&crl=c>