



Prevalence and presentation patterns of enamel hypomineralisation (MIH and HSPM) among paediatric hospital dental patients in Toronto, Canada: a cross-sectional study

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Abstract

Purpose The purpose of the study was to determine the prevalence and presentation patterns of molar incisor hypomineralisation (MIH) and hypomineralised second primary molars (HSPM) in the Division of Paediatric Dentistry at The Hospital for Sick Children (SickKids) in Toronto, Canada.

Methods A cross-sectional study of 429 eligible participants was carried out by nine trained and calibrated examiners. The European Academy of Paediatric Dentists (EAPD) criteria for diagnosis and documentation of MIH and HSPM defects were used.

Results Molar incisor hypomineralisation and HSPM prevalence was 12.4% and 5.2%, respectively. Regarding MIH, the affected molars and incisors attributed to 5.6% of the total prevalence, the remaining having only molars affected. Demarcated white opacities were most common in MIH (60%) and HSPM (67%), followed by yellow/brown opacities (MIH 22%, HSPM 9%), post-eruptive breakdown (MIH 8%, HSPM 24%), atypical caries (MIH 6%, HSPM 0%), and atypical restorations (MIH 4%, HSPM 0%). In both MIH and HSPM, single surface hypomineralised lesions were significantly more common than multi-surface lesions ($p < 0.0001$). Most lesions affected buccal enamel (MIH 55%, HSPM 47%). Lesion extension was most frequently less than 1/3 of a tooth surface (MIH 58%, HSPM 67%) and this was significantly more common in teeth affected by HSPM than MIH ($p = 0.03$). Individuals with HSPM were more likely to present with two affected molars than individuals with MIH ($p = 0.03$). Hypomineralised second primary molars were not predictive for MIH.

Conclusions The prevalence of MIH and HSPM was within the range of published studies. The most common MIH and HSPM lesions affected single surface of a tooth, mostly on the buccal surface and were less than 1/3 of the surface area size.

Keywords MIH · HSPM · Hypomineralisation · Prevalence · Presentation

Introduction

According to the literature, molar incisor hypomineralisation (MIH) is a qualitative defect of enamel affecting one to four first permanent molars with possible incisor involvement (Weerheijm 2003). Hypomineralised second primary molars

(HSPM) are a common presentation of hypomineralisation in the primary dentition (Elfrink et al. 2008). Due to the shared period of enamel maturation, it was speculated that HSPM and MIH are related and consequently, HSPM was reported to be predictive for MIH (Taylor 2017).

Molar incisor hypomineralisation prevalence ranged from 3.6 to 44%. Hypomineralised second primary molars prevalence ranged from 4.9 to 9% (Elfrink et al. 2015). There are no epidemiological studies from North America. The lack of methodological and reporting standardisation of epidemiological studies hindered the ability to compare studies (Elfrink et al. 2015; Lygidakis et al. 2010; Ghanim et al. 2015). Therefore, to advance MIH/HSPM epidemiological knowledge, the EAPD recommended the use of a standardised data collection tool specifically developed for evaluation of enamel hypomineralisation. To our knowledge, this is the

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first study to provide insight into the prevalence and presentation patterns of MIH and HSPM in Canada.

The presentation patterns of MIH and HSPM vary within and among patients. Dental conditions of systemic origin, such as amelogenesis imperfect or fluorosis, have a generalised presentation pattern where lesions are of similar severity and affect all teeth. In MIH and HSPM, the presentation patterns are unique to the surface level of each affected tooth in terms of size and severity of lesions. Hence, epigenetic causes including localised alterations to enamel proteins during the perinatal period were postulated to explain the variable clinical presentations (Vieira and Kup 2016).

The aims of this investigation were to determine the prevalence and presentation patterns of MIH and HSPM among new patients in the Department of Dentistry at The Hospital for Sick Children (SickKids), Toronto. This was done utilising the data collection tool developed by Ghanim et al. (2015) to comply with the EAPD recommendation for standardised assessment of MIH/HSPM.

Materials and methods

Approval of the study protocol was obtained from the Research Ethics Board (REB) at SickKids (REB# 1000052479).

Training and calibration

Prior to participant enrolment into the study, nine paediatric dentists were trained on MIH/HSPM diagnosis and utilisation of the data collection tool from Ghanim et al. (2015). Y.W. held individual instruction sessions with each of the examiners. All examiners were calibrated using 20 intra-oral clinical images. The 20 images represented a range of clinical presentations of dental caries, MIH and HSPM as well as other developmental defects of enamel. Each examiner participated in two calibration sessions that were held 2 weeks apart. During each session, examiners assessed 20 images of enamel lesions displayed on an 11-inch MacBook Air® computer. Examiners scored each lesion in accordance with the grading system in Ghanim et al. (2015). Due to the inherent differences between a digital image and a clinical examination, verbal descriptions of the lesions were provided. Inter-rater and intra-rater kappa values were calculated for reliability of diagnosis of lesion type (MIH/HSPM vs. non-MIH/HSPM) and lesion extension.

Participants

SickKids Dentistry provides care to children with comorbid medical conditions, healthy children under 3 years of age with severe early childhood caries and children with dental

emergencies. The study sample consisted of new patients who presented to the Department of Dentistry at SickKids between May 30, 2016 and February 22, 2017. A new patient was defined as a patient who had no prior appointments in the Division of Paediatric Dentistry.

Patients were invited to participate in the study if they had at least one erupting second primary molar and/or first permanent molar for which at least 1/3 of the clinical crown was erupted. Individuals that did not have clinically visible first permanent molars, permanent incisors and/or second primary molars due to the individual's stage of dental development and not a result of extraction were excluded. All participants that were included in the study assented and/or their parents or guardians consented to their participation in the study (Fig. 1).

Examinations on each patient were performed in a dental operatory with a mirror and explorer. If it was deemed necessary to obtain an accurate diagnosis, examiners performed prophylaxis on teeth with a slow speed hand piece or toothbrush. Tooth surfaces were kept moist during examination. The scores for each tooth provided by the examiners were recorded by a dental assistant on a data collection form (Ghanim et al. 2015).

Statistical analysis

Based on reported prevalence values, a prevalence of 5% was assumed for enamel hypomineralisation. The estimated minimum sample size needed to produce a 95% confidence level with $\pm 2\%$ precision was calculated to be 429 participants.

All collected data were entered and stored in a RED-Cap™ database (Vanderbilt University, Nashville, Tennessee, USA) hosted at SickKids and analysed using Statistical Analysis Software, SAS® for Windows (SAS Institute, Cary, North Carolina, USA). Descriptive statistics were calculated for qualitative variables, represented by proportions. Quantitative variables were represented by means with confidence intervals. Significant differences between qualitative variables were calculated using χ^2 tests. Odds ratios between MIH and HSPM were used to determine if HSPM was prognostic for MIH. Predictive values, likelihood ratio and odds ratio were calculated for relationship between HSPM and MIH. The significance was set at $p < 0.05$.

Results

The training and calibration exercise was completed prior to assessment of any participants. For lesion type, examiners demonstrated substantial inter-rater agreement (0.65–0.73) and substantial intra-rater agreement (0.76). Inter-rater (0.4–0.5) and intra-rater agreement (0.56–0.76) were

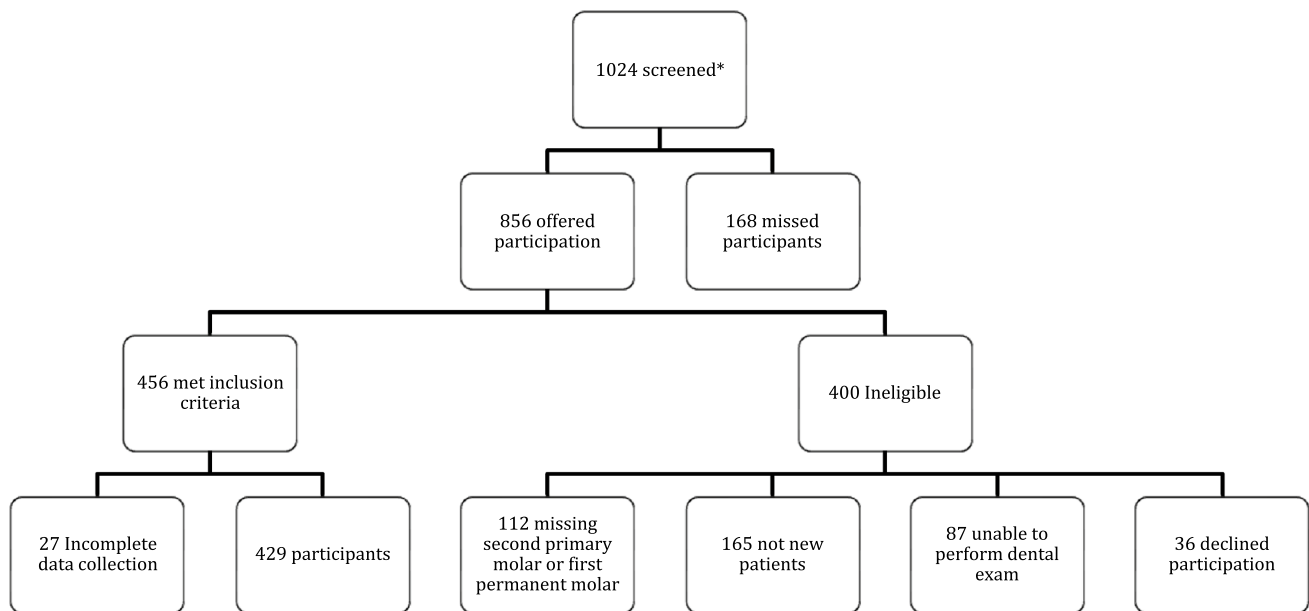


Fig. 1 Participant screening and enrolment as per inclusion and exclusion criteria. Asterisk: screened number of participants including all participants that were scheduled as “new patients” on daily clinic schedule

moderate for assessment of lesion extension (Landis and Koch 1977).

From the 856 screened patients, the final sample consisted of 120 healthy participants and 309 participants with at least one medical comorbidity. The sex distribution of participants was 248 males (58%) and 181 females (42%). Three hundred and sixty-five participants (85%) had at least one second primary molar and 233 (54%) had at least one first permanent molar (FPM). A total of 154 (36%) participants had at least one first permanent and second primary molar (SPM) present. Seventy-two (17%) of the patients have four first permanent molars, eight permanent incisors and four second primary molars.

Prevalence

Among the 429 participants, 48 patients presented with enamel hypomineralisation; 29 of them presented with MIH and 19 with HSPM. Three of the 48 children were diagnosed

with both MIH and HSPM. Among the participants diagnosed with MIH and/or HSPM, 29/48 patients (60%) were male and 19/48 (40%) were female. A total of 42 incisors, 116 FPMs and 36 SPMs, were hypomineralised (Table 1).

MIH

Among the patients with at least one FPM (233 participants), the prevalence of MIH was 12.4% (29/233 patients, 95% CI: 8.5–17.5) Thirteen of the 29 participants with MIH (5.6%, 95% CI: 2.6–8.5) had hypomineralisation defects on their FPMs and incisors (Table 1).

HSPM

Among the patients with at least one SPM (365 participants), the prevalence of HSPM was 5.2% (19/365 patients, 95% CI: 3.1–8.1) (Table 1).

Table 1 Prevalence based on selected sample groups

Sample group and no. of patients	Type of enamel hypomineralisation	Prevalence: percentage, 95% CI (no)
With at least one FPM (n = 233)	MIH (at least one molar affected)	12.4%, 8.5–17.4 (29)
With at least one FPM (n = 233)	MIH (molars and incisors affected)	5.6%, 2.6–8.5 (13)
With at least one SPM (n = 365)	HSPM	5.2%, 3.1–8.1 (19)
With at least one FPM and SPM (n = 154)	MIH and HSPM	1.9%, 0–4.1 (3)
All study participants (n = 429)	Other developmental defects of enamel	9.6%, 6.9–12.7 (41)
All study participants (n = 429)	MIH/HSPM and other enamel defects	0%

MIH and HSPM

Among the patients with at least one FPM and SPM (154 participants), the prevalence of HSPM and MIH was 1.9% (3/154 patients, 95% CI: 0–4.1) (Table 1).

Presentation patterns of MIH and HSPM

The frequency of first permanent molars and incisors and second primary molars affected by hypomineralisation are shown in Fig. 2. First permanent molars were most frequently affected, followed by maxillary permanent incisors. The maxillary left second primary molar was least likely affected by HSPM. In participants with MIH, the mean number of first permanent molars affected was 2.5, while in participants with HSPM, the mean number

of second primary molars affected was 1.5. First permanent molars were equally likely to be affected by MIH ($p = 0.51$). Similarly, there was no significant difference noted in the number of second primary molars involved in those with HSPM ($p = 0.25$). However, individuals with HSPM were significantly more likely to have two molars involved than individuals with MIH ($p = 0.03$). The presence or absence of hypomineralisation was assessed on three surfaces (buccal, occlusal/incisal, palatal) of each tooth. The majority of first permanent molars, incisors and second primary molars affected by MIH and HSPM had lesions affecting only one surface (see Fig. 3). Teeth with a single hypomineralised surface were significantly more common than teeth that had two or three surfaces affected ($p < 0.0001$). Additionally, HSPM was more likely to affect a single tooth surface than MIH ($p = 0.03$).

Fig. 2 Distribution of first permanent molars, incisors and second primary molars affected by MIH and HSPM

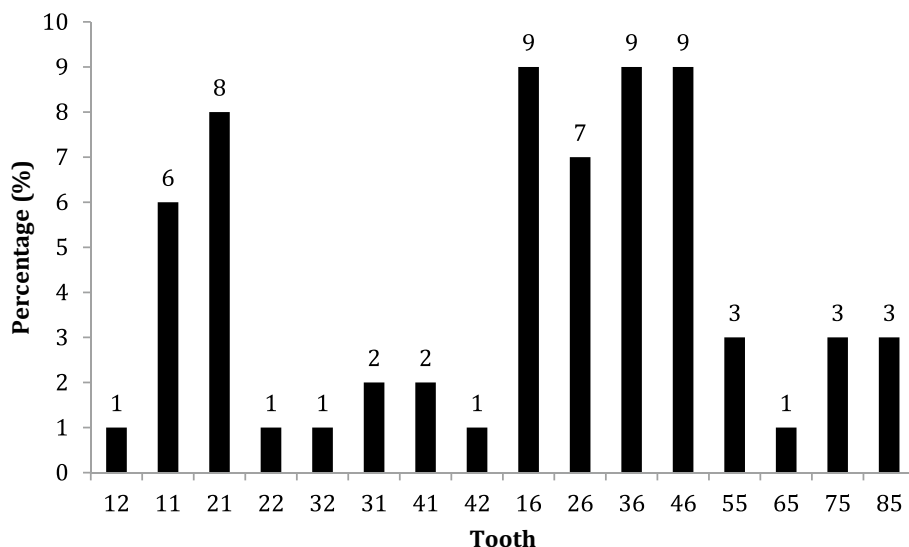
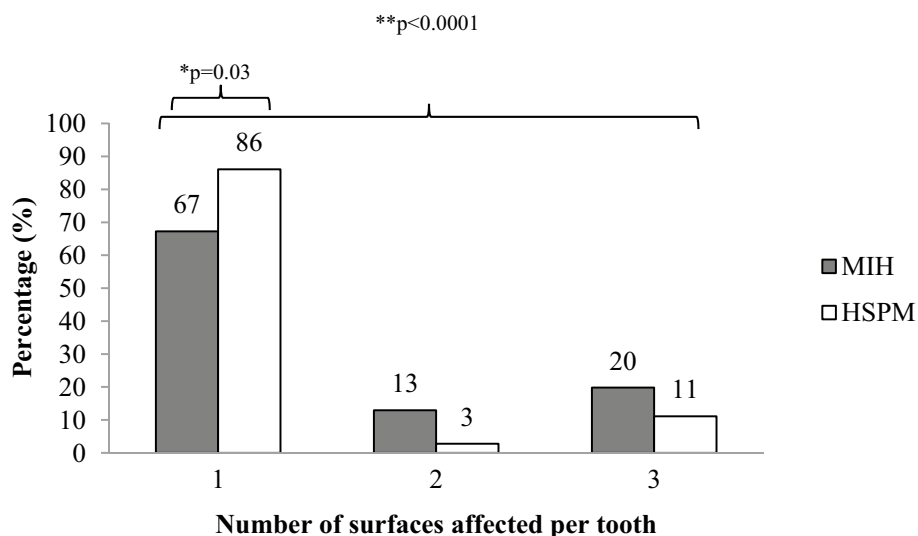


Fig. 3 Number of surfaces involved in teeth affected by MIH and HSPM. Lesions affecting a single surface are significantly more common than two or three surfaces ($p < 0.0001$). Single surface lesions were significantly more common in HSPM than MIH ($p = 0.03$)



Clinical presentations

White demarcated opacities were the most common presentations of MIH and HSPM (Fig. 4). Twelve of 29 patients with MIH (41%) had more than one clinical presentation on their first permanent molars and incisors. No participant with HSPM exhibited a combination of presentations. No participants with MIH or HSPM had extractions as a consequence of hypomineralisation. There were no statistically significant differences between the number of HSPM and MIH participants with white ($p=0.84$) and brown opacities ($p=0.32$). Significantly more surfaces affected by MIH were brown ($p=0.04$) compared to surfaces affected by HSPM. Significantly more surfaces affected by HSPM had PEB ($p=0.002$) compared to surfaces affected by MIH. Atypical caries or atypical restorations were not detected in this sample of individuals with HSPM.

In addition to location and number of surfaces involved, the lesion sizes were also documented. Hypomineralised defects were categorised as: I (< 1/3 of clinical crown

surface), II (1/3–2/3 of clinical crown surface) or III (1/3 < of clinical crown surface) (Fig. 5). Defects observed to be < 1/3 of clinical crown surface were the most common lesion size on first permanent molars, incisors and second primary molars ($p < 0.0001$). Size I lesions were significantly more commonly found on HSPM than MIH tooth surfaces ($p=0.0002$).

Hypomineralised second primary molars were not predictive for MIH (OR = 2.8, 95% CI: 0.76–10.12) ($p=0.11$).

Discussion

Many epidemiological investigations of MIH and HSPM have been published since the introduction of the EAPD Scale (Elfrink et al. 2015). Early studies originated primarily in Europe and Australia; however, in the last 5 years, interest in MIH arose in numerous regions including Africa and Asia (Elfrink et al. 2015). Nevertheless, inconsistencies in demographics, examination settings and study design for

Fig. 4 Clinical presentations of MIH and HSPM. Brown opacities are significantly more common in MIH than HSPM ($p=0.04$). PEB is significantly more common in HSPM than MIH ($p=0.002$). *Ares* atypical restoration, *Acar* atypical caries

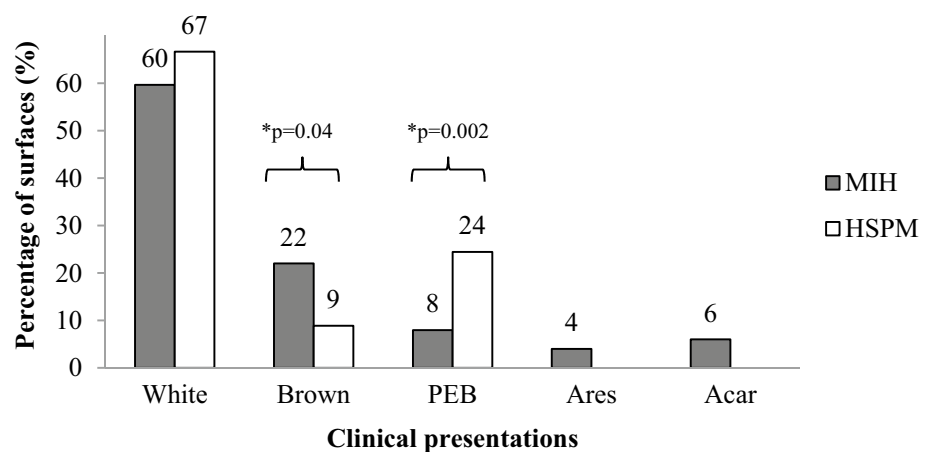
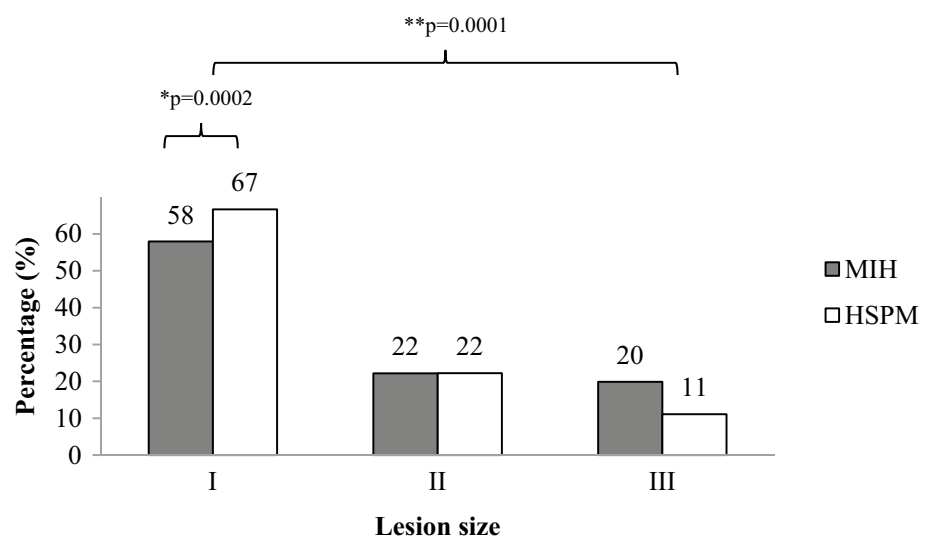


Fig. 5 Size of MIH and HSPM lesions. Size I lesions were significantly more common than sizes II and III ($p=0.0001$). HSPM size I lesions were significantly more common than MIH size I lesions ($p=0.0002$)



MIH and HSPM epidemiological research compromised the comparability of study outcomes. As a result, the EAPD promoted the need for a “worldwide standardised scoring and calibration system” to allow comparisons of MIH and HSPM prevalence values (Weerheijm 2008).

To date, representation from North America was absent. This study recorded a prevalence of 12.4% for MIH, 5.6% for MIH with incisor involvement and 5.2% for HSPM. These values fall within the range reported from existing publications and support that MIH and HSPM are worldwide and not a regional phenomenon (Weerheijm 2015). Our study was among the few that recorded details about the distribution of enamel hypomineralisation at the surface level and the only one to date that utilised a grading and data collection form endorsed by the EAPD (Mittal et al. 2014; Ghanim et al. 2015; Petrou et al. 2015).

Direct comparisons of the outcomes in this study in terms of MIH and HSPM distribution patterns with different outcomes from other investigations were difficult. In our study, the buccal enamel was most commonly affected in first permanent molars, incisors and second primary molars. In some studies, the occlusal surface of first permanent molars and buccal surface of incisors were commonly affected by hypomineralisation (Lygidakis et al. 2008; Ghanim et al. 2011; Mittal et al. 2014; Petrou et al. 2015). The average number of surfaces affected by MIH in our study (1.5) was lower than previous studies that reported greater than four surfaces affected (Mittal et al. 2014; Petrou et al. 2015). However, Petrou et al. examined all five surfaces of the tooth, unlike the present study’s protocol that reported three surfaces, which excluded the mesial and distal surfaces. The lingual/palatal, occlusal/incisal and buccal surfaces of clinically present teeth were examined because they were easily visualised and it was assumed that children had intact arches, limiting the ability to assess mesial and distal surfaces.

Previously, a statistically significant positive correlation between the number of surfaces involved and MIH severity was reported (Petrou et al. 2015). Petrou et al. defined severe lesions as those associated with a positive history of tooth sensitivity. In addition to patient symptoms, many investigators described mild cases as teeth with intact enamel or demarcated opacity and severe cases as teeth with compromised enamel including PEB, atypical restorations and atypical caries. Additionally, teeth that were extracted due to hypomineralisation were also categorised as severe MIH. Applying these criteria to our sample, only 18% of MIH and 24% of HSPM surfaces were characterised as severe. The significantly greater number of coronal surfaces with PEB seen in primary second molars compared to permanent molars ($p = 0.002$) could be explained by the smaller surface area of primary teeth. Elfrink and colleagues opined that hypomineralised defects on second primary molars were more likely to progress to PEB, atypical restorations,

atypical caries, and possible extractions (Elfrink et al. 2016). The use of SSCs for caries or large surface defects could explain the absence of atypical caries or restorations on second primary teeth. Our study did not consider SSC as atypical restoration as recommended in earlier studies because of the common use of SSC in primary dentition for treatment of early childhood caries. Characterisation of SSC as an atypical restoration would likely overestimate the prevalence of HSPM by masking the initial condition of the affected teeth.

Documentation of lesion size is unique to the Ghanim scale (Ghanim et al. 2015). Most hypomineralisation defects observed in this study were less than 1/3 of the tooth surface. A positive correlation between lesion size and severity could not be analysed because 14 PEB surfaces were seen in only three participants, and of those, 12 surfaces originated from a single participant. Findings in our study further confirmed that severe cases of MIH/HSPM were rare whether this was defined by clinical presentation or lesion size.

The tool utilised in this investigation required the examination of all clinically present teeth and the recording of all developmental defects of enamel, not limited to hypomineralisation. This helped to clarify if hypomineralisation was localised to specific index teeth (i.e. MIH or HSPM) or presented on a more generalised spectrum of enamel hypomineralisation. For this part of the study, we did not report the prevalence of hypomineralisation on other non-index teeth. Symptoms are also associated with MIH and HSPM especially if lesions are considered to be severe. This was not part of the epidemiological scoring form, as diagnosis of MIH and HSPM are made purely based on clinical signs and not symptoms. The exhaustive nature of the assessment gave rise to additional issues. Examiners reported that the scale was overly complex and that the grading system was not intuitive. As a consequence, some examiners expressed confusion and frustration. Ideally, an epidemiological tool should be simple and easy to complete. Anything to the contrary potentially discourages its use or, worse, may compromise the integrity of the data should examiners complete the assessment inaccurately as a result.

The results of our study should be interpreted with awareness of its limitations. The sample size of the study, $n = 429$, comprised all participants who had at least one primary second molar or first permanent molar. Subsets of participants with at least one first permanent molar and second primary molar were used to determine the study’s prevalence of MIH and HSPM, respectively. The accuracy of the prevalence value is limited to the point in time of the examination and may not be an estimate of the true prevalence value, because many participants did not have all four of their first permanent molars or second primary molars present at the time of assessment. For this reason, the EAPD had recommended the age of MIH assessment to be 8 years. Implementing this specific age inclusion criteria was unrealistic

for collecting an adequate sample size within the study's timeline. In our study, patients with all four first permanent molars and second primary molars present had prevalence of MIH and HSPM 16.7% and 4.3%. These values were within the reported global ranges of MIH.

The accuracy of lesion assessment resides with individual examiners. Examiners in this investigation had better inter-rater agreement for lesion type than for lesion size.

Significantly higher prevalence of MIH in children with HIV than those without has been reported (Andrade et al. 2017). One may have speculated that a prevalence of MIH or HSPM greater than commonly reported values may have been reported for this sample due to the high rate of medical comorbidity among participants. While the aetiology of MIH/HSPM remaining unclear, perinatal events were hypothesised to disrupt amelogenesis in second primary molars, first permanent molars and incisors. The involvement of other systemic factors such as febrile illness, medication usage and local factors such as genetic mutations of ameloblast ion channels could explain the limited distribution of MIH/HSPM presentation patterns.

More studies that use the same methodology are needed to substantiate the findings of this study and to confirm that MIH and HSPM are distinct clinical entities. Study designs with a larger sample size reported a positive correlation between MIH and HSPM (Negre-Barber et al. 2016). Long-term studies that follow up a cohort of patients with HSPM until eruption of all primary first permanent molars will provide a more accurate relationship between HSPM and MIH. In addition to epidemiological research, the challenge continues to be in determining the right course of treatment, as the presentation patterns of MIH and HSPM vary greatly in terms of number of teeth, size, location and clinical presentation of hypomineralised defects.

Conclusions

- The prevalence values were 12.4% for MIH and 5.2% for HSPM; the prevalence value for MIH with HSPM was 1.9%.
- HSPM was not predictive for MIH.
- There was no significant difference in the number of second primary molars and first permanent molars affected by HSPM and MIH, respectively.
- Most teeth affected by HSPM and MIH have lesions involving a single surface; the lesions are less than 1/3 of tooth surface size and commonly found on buccal surfaces.
- The most common clinical presentation for HSPM or MIH was white demarcated opacity.

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Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest.

Ethical approval All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki Declaration and its later amendments or comparable ethical standards.

Informed consent Informed consent was obtained from all individual participants included in the study.

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