

Deciduous molar hypomineralization in twins: A case report

Hipomineralização de molares decíduos em gêmeos: Um relato de caso

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ABSTRACT

Deciduous molar hypomineralization (DMH) is an enamel defect of multifactorial origin that affects the second deciduous molar and may be associated with canines and first deciduous molars. Clinically, these defects appear as white, yellow, or brown spots, which may progress to a posteruptive enamel breakdown, increasing sensitivity and caries risk. Children with DMH are more prone to develop MIH in permanent dentition. The aim is to present a clinical case of monozygotic twins with severe DMH, showing evidence of its manifestation as well as clinical characteristics and aetiological factors involved. Although DMH etiology remains unclear, clinical manifestations in twins suggest a possible genetic susceptibility to the disease, which may be associated with the shared environmental factors. Also, teeth affected with DMH require an increased need for restorative care. Aetiological risk factors and different treatments' knowledge regarding this condition are of substantial importance to improve patients' quality of life. More studies involving monozygotic and dizygotic twin pairs will be required to improve this case report data.

Keywords: Dental hypomineralization, deciduous, molar, case report.

RESUMO

A hipomineralização de molares decíduos (HMD) é um defeito do esmalte de origem multifatorial que afeta o segundo molar decíduo e pode estar associado a caninos e primeiros molares decíduos. Clinicamente, esses defeitos aparecem como manchas brancas, amarelas ou marrons, que podem progredir para uma fratura pós-eruptiva do esmalte, aumentando a sensibilidade e o risco de cárie dentária. Crianças com HMD são mais propensas a desenvolver HMI na dentição permanente. O objetivo é apresentar um caso clínico de gêmeos monozigóticos com HMD severa, mostrando evidências de sua manifestação, bem como características clínicas e fatores etiológicos envolvidos. Embora a etiologia da HMD permaneça obscura, as manifestações clínicas em gêmeos sugerem uma possível suscetibilidade genética à doença, que pode estar associada a fatores ambientais compartilhados. Além disso, os dentes afetados com HMD requerem uma necessidade maior de cuidados restauradores. O conhecimento dos fatores etiológicos de risco e dos diferentes tratamentos para esta condição são de importância substancial para melhorar a qualidade de vida dos pacientes. Mais estudos envolvendo pares de gêmeos monozigóticos e dizigóticos serão necessários para confirmar os dados deste relato de caso.

Palavras-Chave: Hipomineralização dental, decíduo, molar, caso clínico.

1 INTRODUCTION

Deciduous Molar Hypomineralization (DMH) is a qualitative defect of dental enamel development characterized by demarcated opacities that affects 1 to 4 second primary molars and may be associated with canines and first deciduous molars (Elfrink et al., 2008; Elfrink et al., 2012). Clinically, these demarcated opacities may be white, yellow, or brown (depending on the degree of severity), and may occur with or without destruction of the enamel by posteruptive fractures caused by the force of mastication due to the greater porosity of the enamel (Weerheijm, 2003). The global prevalence rate of DMH ranges from 4,6% to 21.8%, showing a considerable variation caused by the lack of standardized studies (Elfrink et al., 2008; Elfrink et al., 2009; Elfrink et al., 2015; Mittal and Sharma, 2015; Temilola et al., 2015; Owen et al., 2018).

Although the etiology of DMH remains unclear, authors agree that it is multifactorial, combining environmental factors from the prenatal and early life period and genetic and epigenetic factors (Jeremias et al., 2013; Jeremias et al., 2016; Vieira and Kup 2016; Teixeira et al. 2018). Twin studies have traditionally been used to investigate the contribution of genes (heritability) and environment on the etiology of conditions (Silva et al., 2019). It was observed that monozygotic twins share, on average, 100% of their genetic makeup, and a concordance between twin pairs can help explore the influence of shared, nonshared, and genetic factors to the variation in risk of HMD (Neale et al., 1994).

Besides, DMH is a useful predictor of molar-incisor hypomineralization (MIH) risk due to overlapping of the mineralization of the second primary molars and the first permanent molars during a period (Elfrink et al., 2012; Mittal and Sharma 2015; Negre-Barber et al., 2016; da Silva Figueiredo Sé et al., 2017; Garot et al., 2018) and contributes to pain and caries risk among young children (Elfrink et al. 2010; Gambeta-Tessini et al., 2019), which may interfere with a patient's quality of life. Children with HMD are six times more likely to develop MIH; so, as the second primary molars erupt four years before the permanent first molars, early diagnosis of HMD might allow the dentist to control recall intervals and advise parents of the importance of seeing the child as soon as the permanent first molar erupts (Negre-Barber et al., 2016; da Silva Figueiredo Sé et al., 2017).

Recognize shared environmental and non-environmental factors involved in DMH etiology, especially in monozygotic twins, as well its clinical manifestations will allow dentists to understand this enamel alteration implications and to implement early preventive care programs for this population, aiming to minimize their discomfort in deciduous and permanent dentition and to improve patients'/family's quality of life.

Thus, the aim of this study was to present a clinical case of twins with DMH, showing evidence of its manifestation as well as the clinical characteristics, etiological factors and treatment plan involved.

2 CASE REPORT

Monozygotic female twins aged 6 years old were referred to the Pediatric Dentistry Clinic of Amazonas Federal University Dental School, UFAM, because their parents complained of dark discoloration on the posterior deciduous teeth, compromising the sensitivity and oral hygiene. This study was approved by the Research Ethics Committee (29449420.1.0000.5020), and the biological parents signed informed consent. A structured questionnaire addressing pregnancy issues and the child's early life was also administered (Figure 1). According to their mother, the twins were delivered

at term by cesarean section and had been daily fed with breastfeeding for two years and feeding-bottle until four years old. Also, she told us that his husband had a similar buccal condition until his adolescence what made him lose all the posterior teeth.

Figure 1: Summary of prenatal, perinatal, and postnatal medical history reported by each patient’s biological parents.

Medical History		
Prenatal	Perinatal	Postnatal
Recurrents episodes of urinary tract infections during fifth gestational month	Complicated birth caused by pre-eclampsia and hospitalization.	Recurrent acute respiratory tract infections (sinusitis, bronchitis, otitis)
Amoxicilin after the episodes of urinary tract infections	Twins’ mother did not remember the drugs used to treat pre-eclampsia	Rotavirus
Folic acid consumption in the first 10 weeks of pregnancy		Fever
Gestational hypertension and pre-eclampsia		Diarrhea
Placental abruption in sixth gestational month, which requires seven days of hospitalization		Amoxicilin and cephalixin
Candidiasis		

Dental examination revealed hypomineralization in the deciduous molars and canines of both twins, characteristic of severe DMH. In the first twin, FOS, white demarcated opacities were observed in the buccal surface of the lower right deciduous incisor and first molar. It was also seen yellow-brown opacities in teeth 53, 54, 55, 61, 62, 64, 65, 73, 74, 75, 83, and 85, with loss of tooth structure, dentinal sensitivity, and caries in teeth 54, 55, 64, 65, 75 e 85 (Figures 2a, b, and c). A non-surgical treatment focusing on controlling risk factors, such as carbohydrates intake and brushing with fluoridated toothpaste, will be implemented. In addition, dental prophylaxis followed by the application of fluoride varnish (Clinpro®, 3M do Brasil, Sumaré, SP, Brazil) will be part of this patient’s treatment plan. Teeth with loss of dental structure will be temporarily restored with resin-modified, glass ionomer cement Riva Light Cure® (SDI Brasil Indústria e Comércio Ltda., São Paulo, SP, Brazil) (teeth 54,55,64,65,84), steel crown (teeth 55 and 65) and resin composit Z200® (3M do Brasil, Sumaré, SP, Brazil) (tooth 85) following manufacturer’s instructions.

Figure 2: Twin FOS with severe DMH. (a) White and yellowish-brown opacities on teeth 81, 83, 84, 85, 73, 74 and 75 (b) loss of structure and caries on 54, 55, 64, 65. (c) Yellowish-brown opacities on teeth 53, 54, 55, 61, 62, 64, 65, with loss of structure and caries on teeth 75 and 85.



The second twin, JOS, presented negative behavior and also had severe DMH with yellowish-brown discoloration on teeth 53, 54, 63, 64, 65, 73, 74, 75, 83, 84, and 85, associated with severe loss of dental structure, dentinal sensitivity and caries in almost of them (especially teeth 55, 65, 74), except in tooth 83. She also presented caries in teeth 51 and 61. A yellowish-brown opacity in tooth 31, recently erupted, was observed (Figures 3a, b, c, and d). A non-surgical treatment, similar to that proposed for the other twin, will be carried out on this patient's treatment plan. Teeth with loss of dental structure will be temporarily restored with resin-modified, glass ionomer cement Riva Light Cure[®] (SDI Brasil Indústria e Comércio Ltda., São Paulo, SP, Brazil) (teeth 53,54,63,64,73,75, 84 and 85), steel crown (teeth 54 and 64), resin composit Z200[®] (3M do Brasil, Sumaré, SP, Brazil) (teeth 51 and 61) following manufacturer's instructions. An exodontia will be the chosen treatment for teeth 55, 65, and 74.

Figure 3: Twin JOS with severe DMH. (a) and (b) Yellowish-brown opacities on teeth 53, 54, 63, 64, 65, associated with severe loss of dental structure, dentinal sensitivity and caries (c) and (d) Yellowish-brown opacities on teeth 83, 84, 85, 73, 74 and 75, associated with loss of structure and caries on teeth 84, 85, 73, 74 and 75.



Upon oral examination of the twins' family members, only mild MIH was observed in the older sister. She presented only yellowish-white opacity on one of the lower first permanent molars' buccal surface with a small enamel fracture near the cuspid. Unfortunately, their father did not have any permanent molar in the mouth to allow possible enamel alteration diagnostic.

3 DISCUSSION

Enamel development of deciduous canines and second molars, permanent first molars, and incisors occur almost simultaneously, but the maturation phase of the permanent teeth is considerably longer than the primary teeth. During this overlapping period (before birth and between the first and third years of life), a risk factor might concurrently cause hypomineralization in the primary and permanent dentition (Elfrink et al., 2014; Temilola et al., 2015). Literature has shown a relationship between MIH and DMH (Elfrink et al., 2012; Mittal and Sharma, 2015; da Silva Figueiredo Sé et al., 2017; Garot et al., 2018), but the hypothesis of the DMH could be an essential predictor factor was contradictory for some authors (Elfrink et al., 2012; Ghanim et al., 2013). However, other studies showed that the presence of DMH is predictive for MIH (Negre-Barber et al., 2016; da Silva Figueiredo

Sé et al., 2017; Garot et al., 2018), and as the number of primary molars affected by hypomineralization increases, the greater the risk for developing MIH (Temilola et al., 2015). A real need of clinical and radiological follow-up for the children from this study may allow the dentist to provide family counseling and implement an early preventive program that minimizes posteruptive enamel breakdown and hypersensitivity in their permanent dentition.

The etiology of DMH and MIH is not fully understood, and the currently available scientific evidence is not enough to establish causality (Vieira and Kup, 2016). Literature has shown a relationship between MIH and DMH with a combination of medical problems during pre-, peri- and postnatal periods and genetic factors. Strong scientific evidence showed that shared environmental factors are more critical in DMH etiology than genetics (Silva et al., 2019). Children whose mothers were hypertensive, had delivery complications (Lopes-Fatturi et al., 2019), and began to consume folic acid in the first ten weeks of gestation (Elfrink et al., 2014) presented a higher prevalence of DMH, which agree with the maternal medical history of this case report. Alike seen in this report, in the perinatal period, twins and birth complications are significantly related to a higher prevalence of DDE (Vello et al., 2010) and DMH (Lopes-Fatturi et al., 2019). The postnatal exposures associated with DMH were the presence of otitis media and bronchitis, both related to a respiratory condition or infection. In a study adjusted for the multiple hierarchical approach, children with otitis media presented with 68% more prevalence of DMH than children who did not suffer from otitis media. The postnatal medical history related in the present case report showed recurrent episodes of otitis and bronchitis, with fever and the use of medicines. In the case described in this study, the second twin always presented more severe symptoms compared to her sister. Any fever (Elfrink et al., 2014; Pineda et al., 2015) and the use of antibiotics, especially amoxicillin (de Souza et al., 2016) in the child's first year of life could be associated with DMH. However, another study found no clear evidence that the use of drugs during pregnancy and during the first year of life is associated with second primary molar hypomineralization, emphasizing a need for more studies to analyze this relationship (Serna Muñoz et al., 2019). Identifying the presence of these factors in the anamnesis could enable identifying groups at risk for the hypomineralization of dental enamel (Lopes-Fatturi et al., 2019).

The presence of enamel hypomineralization in other family members from this case report reinforces the multifactorial idea of the DMH/MIH etiology (Jeremias et al., 2016).

Both MIH and DMH enamel lesions are classified as demarcated creamy-white or yellowish-brown opacities, which may be associated with a posteruptive breakdown, which facilitates dental development caries and extreme hypersensitivity that causes severe discomfort (Gambetta-Tessini et

al., 2019). Significant relationships were observed between the severity and number of DMH-affected teeth and the resulting hypersensitivity, oral hygiene, and dietary restrictions, demonstrating a need to desensitize hypomineralized teeth to allow adequate oral hygiene and thus improve the oral and systemic health of affected children (Ebel et al., 2018). In this case report, a fluoride varnish application that increases fluoride and calcium ion release was carried out, not only as a risk control measure for dental caries but also to reduce both twins' teeth sensitivity.

In addition, hypomineralized deciduous teeth require an increased need for restorative care. Several different treatment options and materials are available. Typically, patient- and tooth-related factors, like severity, symptoms, dental age, cost-benefit, patients'/parents' expectations, and patients' behavior, determine the optimum choice in each case. Besides, the dentist's own experience and skills may influence the choice of treatment (Wuollet et al., 2020). For severe DMH cases, health promotion and sugar control implementation are recommended, together with glass ionomer cement restoration, resin composites, steel crowns, and exodontia (Quintero et al., 2019).

More studies involving monozygotic and dizygotic twin pairs with genetic sequencing will be required to improve this case report data.

4 CONCLUSIONS

In sum, DMH represents a clinic challenge for all dentists, requiring special attention in clinical practice. The aetiological risk factors and different treatments' knowledge regarding this condition is of substantial importance to improve patients' quality of life.

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