

CASE REPORT

Manifestation of molar-incisor hypomineralisation in twins: clinical case reports

Manifestação da hipomineralização molar- incisivo em gemelares. relatos de casos clínicos

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ABSTRACT

Molar-incisor hypomineralisation is a qualitative defect of dental tissue of systemic origin affecting one or more permanent first molars and sometimes the permanent incisors as well. There are still no conclusive data on the aetiology of this hypomineralisation, however, systemic factors such as respiratory diseases and prenatal and perinatal complications are regarded as possible causes. The objective is to present three clinical cases of twins, one Monozygotic and two Dizygotic Twins with molar-incisor hypomineralisation, showing evidence of its manifestation as well as clinical the characteristics and aetiological factors involved. The clinical findings involving twins suggest that ameloblasts are specifically affected in their developmental phase, which includes a number of factors. Although prenatal and perinatal complications are not decisive in the development of molar-incisor hypomineralization, it is suggested a possible genetic susceptibility to the disease. Prospective observational studies using a population sample containing data on the last three months of gestation to the eruption of permanent teeth are needed to confirm the cause-effect relationships.

KEYWORDS

Pediatric dentistry; Tooth desmineralization; Twins.

RESUMO

A Hipomineralização molar-incisivo é um defeito qualitativo do tecido dental, de origem sistêmica, que afeta um ou mais primeiros molares permanentes e, por vezes, os incisivos permanentes. Ainda não há dados conclusivos sobre a etiologia desta hipomineralização, no entanto, os fatores sistêmicos, como doenças respiratórias e complicações pré-natais e perinatais são considerados como possíveis causas. O objetivo deste estudo é apresentar três casos clínicos de crianças gemelares, sendo um casal de gêmeos monozigóticos, e dois gêmeos dizigóticos com a alteração de hipomineralização molar-incisivo, além de mostrar evidências de sua manifestação, bem como as características clínicas e os fatores etiológicos envolvidos. Os achados clínicos envolvendo gêmeos mostram que ameloblastos são especificamente afetados em sua fase de desenvolvimento, que inclui uma série de fatores, como complicações pré-natais e perinatais, porém não decisivos no desenvolvimento da alteração e sugere ainda uma possível susceptibilidade genética para a esta doença. São necessários estudos observacionais prospectivos utilizando uma amostra da população, contendo dados sobre os últimos três meses de gestação até a erupção dos dentes permanentes para confirmar as possíveis relações de causa-efeito.

PALAVRAS-CHAVE

Odontopediatria; Desmineralização do dente; Gêmeos.

INTRODUCTION

Molar-incisor hypomineralisation (HMI) is a qualitative defect of dental tissue of systemic origin, affecting one or more permanent first molars and sometimes the permanent incisors as well. It is clinically identified by white, yellow or brown spots with smooth surface and normal enamel thickness. This stained enamel is porous and can fracture under masticatory forces, thus leaving the dentine without protection and favoring the development of carious lesion. This enamel defect manifests asymmetrically, suggesting that ameloblasts are affected during a very specific phase of their development [1].

There are still no conclusive data on the aetiology of this hypomineralisation. However, systemic factors such as respiratory diseases and prenatal complications are regarded as possible causes. Low birth weight in association with hypoxia (lack of oxygen for ameloblasts) is also reported, including calcium and phosphate metabolic disorders and childhood history of high-fever disease [2-3]. Some authors suggest that exposure to dioxin (environmental pollutant) during prolonged breast-feeding and antibiotics use can increase the risk of HMI [4]. Due to the difficulty in distinguishing the clinical picture in terms of disease, fever and antibiotics, one cannot state which factor indeed causes such an alteration [2].

Children having health problems in the first three years of life, which is a critical period for the crown formation of permanent of first molars and incisors, are more likely to suffer from HMI [3]. According to Whatling and Fearne [4], genetic studies are needed to assess the aetiology of HMI as the clinical practice suggests the possibility of a genetic susceptibility to this disease.

In view of what has been observed above, the objective of the present work is to present three clinical cases of twins with an enamel defect known as molar-incisor hypomineralisation, showing evidence of its manifestation as well

as the clinical characteristics and aetiological factors involved.

CLINICAL CASES

Case 1 – Monozygotic Twins

Monozygotic male twins aged 11 years old were referred to the Child Dentistry Clinic of the Araraquara Dental School, UNESP, because they were complaining of sensitivity in their permanent molars. On intra-oral examination, it was observed the presence of moderate HMI in the first twin, ACS, whose teeth 11, 31, and 41 had white opacity. Tooth 16 also presented opacity, whereas tooth 26 had an atypical but satisfactory restoration and a yellow-brown opacity. Atypical restorations were also present in teeth 36 and 46 (Figure 1A). The second twin, GCS, also had moderate HMI with white-yellow opacity in tooth 21 and atypical but satisfactory restorations in teeth 16, 26, 36 (Figure 1B). According to their mother, she had taken antibiotics during pregnancy and the twins were delivered at term by caesarean section. The twins had been daily fed with feeding-bottle since then presented episodes of amygdalitis associated with high fever, followed by use of antibiotics. Only the first twin experienced respiratory problems.

Case 2 – Dizygotic twins

Dizygotic male twins aged 8 years old were diagnosed with HMI during epidemiological survey and then they were invited for dental treatment in the Child Dentistry Clinic of the Araraquara Dental School, UNESP. The first twin, VTM, had a HMI characterised by white-yellow opacity in teeth 21 and 46, including mild structural loss in tooth 16 (Figure 2A). The second twin, NTM, had only a white opacity in tooth 26 (Figure 2B). According to their mother, the twins were delivered at term by Caesarean section, and they were breastfed for 3 months and then fed with feeding-bottle for 3 years. Only the second twin took antibiotics and had “chicken pox”.

Case 3 – Dizygotic twins

Dizygotic twins aged 8 years old, one male and other female, were referred to the Child Dentistry Clinic of the Araraquara Dental School, UNESP, because their permanent teeth were “crumbling”. On intra-oral examination, it was observed the presence of severe HMI

with structural loss in teeth 16, 26, 36 and 46 in both children (Figure 3A and 3B). According to their mother, the twins were born at term by natural delivery and since then had received only feeding-bottle for 3 years. Both twins had “chicken pox” and influenza, taking antibiotics before the age of 3 years old. Only the first twin had respiratory problems.

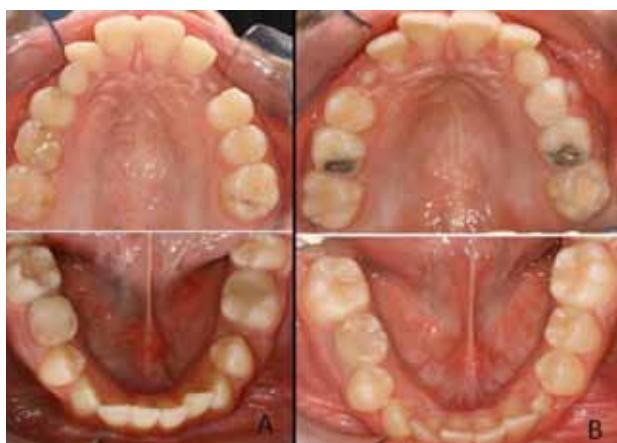


Figure 1 – (A) Twin ACS with atypical restoration and brown opacity in tooth 16, white opacity in tooth 26, and atypical restorations in teeth 36 and 46 (B) Twin GCS with atypical restorations in teeth 16, 26, and 36.



Figure 2 – (A) Twin VTM with mild structural loss in tooth 16 and white-yellow opacity in teeth 21 and 46 (B) Twin NTM with white opacity in tooth 26.



Figure 3 – (A) Twin JBC with mild structural loss in tooth 26, moderate structural loss in teeth 16 and 46, and severe structural loss in tooth 36 (B) Twin VBC with moderate structural loss in tooth 46 and severe structural loss in teeth 16, 26, and 36.

DISCUSSION

The formation of permanent first molars begins in the fourth month of intrauterine life and the early signs of mineralization appear close to birth. In the initial stages of enamel maturation, the ameloblasts are sensitive to systemic changes and cannot self-remodel [5].

Studies have pointed a relationship between HMI and medical problems during pregnancy [4], premature birth, twinning, Ceasarian delivery, and intercurrents during childbirth [6]; whereas others [4] found no relation with perinatal changes. In the above-mentioned cases, problems related to pregnancy and childbirth may have triggered the disease either in isolation or in conjunction. In the first and second cases, the twins were born by Ceasarian section, whereas in the third case the twins were born through natural delivery. Because the six children had manifested HMI, it was not possible to state that Ceasarian delivery, or even the premature birth in the second case, accounted for this dental alteration. Despite the presence of HMI, its intensity and localisation vary among siblings, even monozygotic twins. Such a difference is more evident in the second case, with twin VTM presenting spots and dental structural losses, whereas his brother had only one white spot.

The mineralization of permanent molars extends to the third year of life, and according to studies conducted in the past decade, systemic changes during this period of life are more commonly seen in patients with HMI. For Beentjes [2], patients with HMI are more likely to have medical problems in the first four years of life than those without such a disease. The same finding was reported by a study in which HMI was related to the presence of diseases in the first year of life. In addition, Tapias e Ledesma [7] found a relationship between diseases occurring in the first four years and enamel defects in permanent first molars. Chicken pox, as well as other diseases in the childhood, also seems to be associated with

HMI development [4]. Because of the limited retrospective information on aetiological factors provided by the caregivers, it is not possible to precisely define incidence and frequency of diseases affecting these children. In the second case, however, the less affected twin, NTM, had chicken pox and his brother had only dental structural loss.

According to the literature, respiratory problems like pneumonia [2] and asthma [8] are also associated with HMI, but in our study only two of the six children evaluated had respiratory changes in the early childhood.

Studies have also reported a relationship between HMI and childhood diseases, high fever, and antibiotics use [2], but due to the difficulty in distinguishing them clinically, no definitive conclusion can be drawn. Laboratory investigations are needed to determine whether fever, antibiotics or changes resulting from childhood diseases interfere with enamel structure in isolation or in conjunction, which would lead to the development of HMI. By investigating the fever as single factor, Tung [9] observed that induction of high fever in rats caused changes in their enamel structure.

With regard to amoxicillin, Laisi et al. [10] dissected mouse embryonic tooth germs at the beginning of the enamel matrix secretion and maintained them in cultures with and without amoxicillin for 10 days. They observed that teeth not exposed to or exposed to low doses of amoxicillin had normally evolved until the stage of dental maturation, whereas ameloblasts exposed to 4 mg/ml of amoxicillin remained elongated. However, a pioneer study on HMI was conducted in 1987 and showed the presence of dental changes in Swedish children even before the commercialisation of amoxicillin [5]. In the cases described in the present study, five of the six patients were given amoxicillin before they were 3 years old, and the one receiving no antibiotics (VTM) presented more severe symptoms compared to his brother, who had been exposed.

CONCLUSION

In sum, HMI has been drawing special attention in the clinical practice and aetiological data regarding this condition are of great importance. The clinical findings involving twins show that ameloblasts are specifically affected in their developmental phase, which includes a number of factors although prenatal and perinatal complications not decisive in the development of molar-incisor hypomineralization and suggest a possible genetic susceptibility to the disease. Prospective observational studies using a population sample containing data on the last three months of gestation to the eruption of permanent teeth are needed to confirm the cause-effect relationships.

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