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# Green Pigmentation of Deciduous Teeth: Report of Two Cases

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#### **ABSTRACT**

The purpose of this paper was to present 2 cases of green pigmentation in primary teeth caused by hyperbilirubinemia, from different pathologies during the neonatal period. Medical history revealed systemic problems during the neonatal period of an infectious, neurological, renal, respiratory, and cardiological nature, as well as a high amount of bilirubin in both cases and hepatitis in the first. The staining in the teeth was due to hyperbilirubinemia, caused by these systemic conditions. The clinical characteristics of teeth may help in the diagnosis of present current or past systemic diseases. The reported cases confirm the relevance of past medical history in establishing the diagnosis of the etiology of green pigmentation as a result of high levels of bilirubin serum. (*J Dent Child.* 2004;71:179-182)

KEYWORDS: DENTAL ENAMEL, BILIOUS PIGMENT, BILIRUBIN, ODONTOGENESIS

The presence of hyperbilirubinemia during enamel and dentin formation results in dental pigmentation, due to accumulation of intrinsic staining by bilirubin oxidation. <sup>1-8</sup> Bilirubin is a breakdown product from red blood cells, which, at high levels, can cause jaundice and accumulate in the interstitial fluid, mucosa, and skin, resulting in discoloration that can vary from yellow to deep shades of green.<sup>7</sup>

Intrinsic pigmentation occurs during dental formation and has been frequently related to drug administration—particularly tetracycline<sup>5</sup>—and systemic alterations present at birth, such as hemolytic anemia in newborns.<sup>7,9,10</sup>

In newborns, other complications may be associated with discoloration of teeth, such as biliary atresia, premature birth, neonatal respiratory dysfunction, significant internal hemor-

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rhage, congenital hypothyroidism, biliary hypoplasia, metabolic diseases, hepatic dysfunction, and generalized hyperbilirubinemia. 4,6,7,9-11 Herbert and Delcambre reported the presence of iatrogenic anemia, direct and indirect hyperbilirubinemia, and neonatal staphylococcal sepsis, among other findings, in cases of green dental pigmentation. 5

Although hyperbilirubinemia has been reported in literature, green pigmentation of primary teeth has been rarely documented. Barta et al described primary teeth pigmentation due to blood incompatibility and verified that dental discoloration became less evident at the third year of life. Prevalence in these cases ranged from 20% to 25%, while hemolytic anemia occurs in a proportion of 1:10 births. Tank observed this same clinical aspect in 2 cases of persistent icterus, probably due to blood transfusions.

Morisaki et al<sup>6</sup> reported 7 cases, observing green pigmentation in primary teeth in all cases and the permanent teeth in 5 cases (direct bilirubin serum of more than 5 mg/dl). The gingiva, tongue, and buccal mucosa also showed discoloration. Histological findings suggested there was an accumulation of bile pigments in the dental hard tissues.<sup>6</sup>

The extent of discoloration and its intensity were directly related to the duration and severity of the pathology, respectively—in most cases occurring only in primary dentition.<sup>3,7,11,16</sup> This paper presents 2 cases of green pigmentation in primary teeth caused by hyperbilirubinemia, from different pathologies during the neonatal period.

#### CASE 1

#### **HISTORY**

A 4<sup>1</sup>/<sub>2</sub>-year-old white male, admitted to the Oral Pathology Department of a public dentistry school in Rio de Janeiro, Brazil for evaluation of his teeth's green coloration, was referred to the Department of Pediatric Dentistry. His mother was hypertensive, and her blood type was A positive. The child, born at term via caesarean section, weighed 3,070 g at birth, and his blood type was O positive. During the anamnesis, an anoxia at labor was caused by 2 umbilical cord loops, requiring resuscitation and the use of methyldopamine. Respiratory distress, asphyxia, and infection were diagnosed, as well as pallor of the pupil and hemorrhage of the retina, ischemichypoxia syndrome, serious pulmonary arterial hypertension secondary to asphyxia, and myocardial ischemia.

Fifteen days after birth, icterus was observed, with total bilirubin serum of 24.5 mg/dl. The patient received several blood transfusions due to microangiopathic anemia, probably secondary to sepsis caused by *Enterobacter sp.* After 5 days, the child showed homogeneous hepatosplenomegaly, biliary mass ("mud") and dilation of the renal pelves. After 70 days, the patient, weighing 3,350g, was discharged. The mother related that the child had pneumonia and several convulsions during hospitalization, and when he was 12 months old, hepatitis was diagnosed through specific serology. No treatment was performed, however, since the disease was already in the final stage.

The child's diet was uncontrolled (eating between meals), but oral hygiene methods were performed daily after lunch and before going to bed.

## **CLINICAL FINDINGS**

No caries was found, and a left unilateral crossbite was observed, as well as good hygiene oral and sound soft tissues. All the teeth displayed a green color, with the stain varying in degree and extension (Figure 1). The mandibular and maxillary central and lateral incisors showed a thin line of green pigmentation in the cervical third. The mandibular and maxillary canines and first and second molars revealed a green pigmentation at the middle third—in sharp contrast with the opalescent opaque white in the cervical third. Radiographic examination confirmed the absence of caries. Instructions were given on diet, and oral hygiene was reinforced. Light-cured filled resins on labial surfaces of the upper anterior teeth (canine to canine) and a fixed device to correct the post crossbite were included in the treatment plan. Treatment did not occur because the patient's mother had health problems.

#### CASE 2

#### **HISTORY**

A 22 month old, white female, was referred to the physician's office for evaluation of green coloration of her teeth. The mother said she had received prenatal care and that the pregnancy was incident-free. At 37 weeks and

4 days of gestation, she delivered via caesarean section, due to acute fetal tachycardia.

The newborn, presenting Apgar scores of 1/5/7, was taken to the neonatal intensive care unit immediately after birth, due to a hypovolemic shock and acute anemia, and remained there for 60 days. At birth, the child weighed 2,880 g, and presented the following: (1) respiratory discomfort; (2) slight pulmonary hypertension; (3) anuria during the first 36 hours; (4) oliguria for 3 days; and (4) diuresis with macroscopic hematuria and clots.

The newborn presented anasarca, and the ultrasound showed augmented kidneys. The neurological syndrome was hypertonia of the upper limbs, followed by hypotonia within 12 hours of life and myocardial ischemia on the first day. She received 7 units of eryhthrocyte concentration during hospitalization for anemia. The skull ultrasound was normal, evolving towards a slight increase of the left cerebellar hemisphere, in addition to areas of cortical necrosis of the left temporal area—observed by magnetic resonance of the skull at 17 days. At 12 days, the patient presented symptoms of cholestasis caused by sepsis, an accentuated increase of the transaminases (TGO and TGP), hypoalbuminemia, altered coagulogram, and low platelets. Maximum bilirubin was 27.6 mg/dl, decreasing to 3.6 mg/dl after 45 days. Antibiotic therapy began at 13 days, due to the infectious process. There was a slight loss of hearing in the right ear.

# **CLINICAL FINDINGS**

Clinical examination revealed green staining on all erupted teeth, while attached gingiva showed a yellow color. In the cervical third of anterior teeth and lower molars, the stains were less evident (Figure 2). No caries was found. At last examination (26 months of age), the primary first molars were present and pigmented. The patient will continue receiving private pediatric dental care.

# **DISCUSSION**

Green pigmentation may be seen through the translucent enamel and, presumably, is the result of a reaction of the developing tooth to the blood supply, which is rich in bilirubin serum. The degree of pigment deposition may be proportional to the serum concentration of bilirubin and may disappear gradually, <sup>1,3</sup> Figure 1 shows the interruption of the hyperbilirubinemia period, since the upper and lower canines and lower first primary molars in the cervical third do not show any green pigmentation.

The calcification process of primary teeth begins during the fourth month of fetal life and ends almost at the 11th month after birth on the primary second molars. <sup>16</sup> At birth, a significant portion of upper and lower central and lateral incisors exhibits more than half of the already formed crown. <sup>16</sup> Case 1 showed these elements exhibit discoloration only in the cervical third, with the upper and lower canines, lower first and second molars exhibiting in the middle third, and the upper molars in the cervical and middle third. This suggested pigmentation occurred at the postnatal period. The localization of the pigmented lines corresponded with medical history of



Figure 1. Case 1 shows variation intensity and green pigmentation distribution in the primary teeth of a  $4^{1/2}$ -year-old boy, demonstrating the relationship with tooth development stages.

the patient. This situation did not happen in case 2 because the teeth showed the total crown with discoloration.

The tooth formed after the hyperbilirubinemia period had complete resolution to normal color, with a sharp dividing line separating the green portions, as reported in the literature. <sup>1,7</sup>

The crowns of the first molars begin to form at birth, <sup>16</sup> and the hypothesis of permanent dentition being affected by icterus generated by erythroblastosis fetalis is very unlikely, except for these teeth. <sup>7,16</sup> Morisaki et al agree with these conclusions, having found green pigmentation of permanent first molars in 70% of the patients who presented icterus generated by congenital biliary atresia and/or neonatal hepatitis. <sup>6</sup> As these pathologies were present in the neonatal period in both cases, the pigmentation possibility of the permanent first molars should be considered, mainly in view of the stain localization of deciduous teeth and high bilirubin levels.

According to Ball, it is not possible to correlate the severity, intensity, and duration of the present pathology to the dental condition. According to Tank, Barta et al and Neville et al, however, the time to interruption may be estimated. Dase 1 showed that the cervical third of the lower canines and molars exhibited no green pigment. This suggests the normality of the bilirubin levels, which probably happened before the sixth month of life, the period during which the primary maxillary first and second molars already showed complete enamel formation. This period cannot yet be estimated for case 2, because all teeth present at the point of examination had green pigmentation.

Respiratory disorders have been reported as a cause of neonatal icterus without interruption of the mechanism of keeping the ductus arteriosus functionally patent. This causes poor perfusion of the hepatic sinusoids and decreasing bilirubin excretion. <sup>5,7,9</sup> Both cases show similarities in respect to respiratory disorders, <sup>5,7,9</sup> anemia, icterus, <sup>9,10,13</sup> and high bilirubin levels exhibited in the neonatal period. <sup>1-8</sup>

To permit diagnosis and make a decision about the need for aesthetic treatment, it is important to evaluate whether pigmentation occurs in dentin and/or in enamel, is isolated or related to other problems, and is hereditary or not.



Figure 2. Case 2 shows clinical appearance of the primary teeth at 22 months of age.

Morphological defects were not observed in the cases mentioned, and no case of dental discoloration was found in the family history, suggesting the discoloration was due to systemic disorders occurring at birth.

Although the enamel formed before birth does not show green pigmentation, it may reflect the stained dentin tissues. <sup>1,3</sup> Rosenthal et al recommends cosmetic treatment for these cases, because the appearance of an infant's first tooth is a developmental milestone for the entire family and cause of parental anxiety and difficulties between children and their peers. <sup>10</sup> In case 1, the child had all primary teeth erupted, enabling light-cured filled composite on the facial surfaces of the anterior teeth. On the other hand, in case 2, only the anterior teeth and first molars were present, making it necessary for treatment to be delayed.

## **CONCLUSIONS**

The clinical characteristics of teeth may help in the diagnosis of present current or past systemic diseases. The reported cases confirm the relevance of past medical history in establishing the diagnosis of the etiology of green pigmentation as a result of high levels of bilirubin serum.

## REFERENCES

- 1. Tank G. Two cases of green pigmentation of the deciduous teeth associated with hemolytic disease of the newborn. *J Am Dent Assoc.* 1951;42:302-306.
- 2. Bevis DCA. Blood pigments in hemolytic disease of the newborn. *J Obstet Gynaec Brit Emp.* 1956;63:68-75.
- 3. Miller J, Forrester RM. Neonatal enamel hypoplasia associated with hemolytic disease and with prematurity. *Br Dent J.* 1959;106:93-104.
- Belanger GK, Sanger R, Casamassimo PS, Bystrom EB. Oral and systemic findings in biliary atresia: Report of 11 cases. *Pediatr Dent.* 1982;4:322-326.
- Herbert FL, Delcambre TJ. Unusual case of green teeth resulting from neonatal hyperbilirubinemia. *J Dent Child*. 1987;54:54-56.
- Morisaki I, Abe K, Tong LSM, Kato K, Sobue S. Dental findings of children with biliary atresia: Report of seven cases. *J Dent Child*. 1990;57:220-223.

- 7. Neville BW, Damm DD, Allen CM, Bouquot JE. Saunders C, eds. Oral & Maxillofacial Pathology. Philadelphia: 1995:56-57.
- 8. Shibata T, Watanabe K, Oda H, et al. Experimental bilirubin pigmentation of rat dentine and its detection by a qualitative analytical method. *Arch Oral Biol.* 1996;41:509-511.
- 9. Pindborg JJ. Pathology of the Dental Hard Tissues. Munksgaard, Copenhagen: 1970:443.
- Rosenthal P, Ramos A, Mungo R. Management of children with hyperbilirubinemia and green teeth. *J Pediatr*. 1986;108:103-105.
- 11. Watanabe K, Shibata T, Kurosawa T, Morisaki I, Kinehara M, Igarashi S, et al. Bilirubin pigmentation of human teeth caused by hyperbilirubinemia. *J Oral Pathol Med.* 1999;28:128-130.

- 12. Barta JE, King D, Jorgensen RL. ABO blood group incompatibility and primary tooth discoloration. *Pediatr Dent.* 1989;11:316-318.
- 13. Behrman RE, Kliegman RM. Disturbances of the blood. In: Behrman RE, Vaughan SC, eds. Textbook of Pediatrics. Philadelphia; 1983:383-388.
- 14. Barta JE, King D, Jorgensen RL. ABO blood group incompatibility and primary tooth discoloration. *Pediatr Dent.* 1989;11:316-318.
- 15. Scully C, Flint SR, Porter SR, edss. *Atlas Colorido de Doenças da Boca—Diagnóstico e Tratamento*. Rio de Janeiro: 1997:173.
- 16. Ball JS. Pigmentation of the primary dentition: Pathogenesis and diagnostic implications. *Pediatr Dent*. 1964;3:394-404.

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