Case report/Kazuistyka

Green teeth resulting from neonatal hyperbilirubinemia: Report of a case

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A B S T R A C T

In general, cosmetic changes or painful disorders often lead parents to seek dental care for their children. Several systemic disorders in pediatric patients can produce dental alterations. One of the manifestations of these disorders is the elevated serum level of bilirubin (hyperbilirubinemia), a product of hemoglobin degradation, which is deposited in mineralized and soft tissues. The aim of this article is to report a case of green teeth affecting all deciduous teeth in a 3-year-old girl. The patient was taken to see the dentist by their parents due to the presence of green pigmentation in the teeth. During birth, the patient suffered a traumatic injury that resulted in hyperbilirubinemia. This kind of enamel pigmentation is permanent and occurred during the period of dental development.

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Introduction

Changes in the normal color of deciduous and permanent teeth may occur due to factors intrinsic or extrinsic. The intrinsic stains are changes in the color of the tooth related to systemic factors such as: genetic factors, birth defects, metabolic defects, prenatal care, infectious diseases, neurological disorders, endocrine, kidney, liver, nutritional deficiencies and poisoning that usually occurred during odontogenesis [1]. Moreover, the formation of the extrinsic stains on teeth occurs primarily from the remains of food, medicinal substances and bacteria, which represent deposits, adhered to the enamel [2].

The intrinsic dental pigmentation is associated with a chemical active change in the tooth structure and it cannot be removed without changing the structure of the tooth. It can be caused by erythropoietic porphyria, fetal erythroblastosis, amelogenesis imperfecta, dental fluorosis, hyperbilirubinemia, decomposition of red blood cells and medications (tetracycline) [3].

Bilirubin, a breakdown product of heme, is normally excreted in the bile or further catabolized before excretion in the urine. Approximately 80% of bilirubin is derived from free hemoglobin resulting from hemocatabolism. In this

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physiological process, senescent red cells with approximately 120 days are destroyed by macrophages in the spleen, liver and bone marrow. Hyperbilirubinemia is a clinical condition characterized by an abnormal increase of bilirubin in the blood, which may result in jaundice. The jaundice is the clinical sign caused by the elevation of serum bilirubin levels and its deposition in the skin, sclera, mucous membranes and some organs (liver and kidneys). The deposition of this pigment in these tissues and organs produces a color ranging from yellow to black, passing through various shades of green [4].

All healthy neonates develop unconjugated hyperbilirubinemia during the first week of life, but 50% of these patients have jaundice, benign and transient in character [5]. Jaundice occurs when the liver fails to eliminate bilirubin produced by the body [6]. Severe neonatal hyperbilirubinemia carries a potential for permanent neurological impairment [7]. Furthermore, hyperbilirubinemia is associated with the development of changes in color of the teeth, known as green teeth [8]. These teeth do not show changes in their morphology. However, the green coloration considerably affects the esthetics of the affected tooth. The appearance of the dentition is of concern to a large number of people seeking dental treatment and the color of the teeth is of particular cosmetic importance [9]. The purpose of this report is to present a case of green teeth of the primary dentition in a 3-year-old Brazilian girl.

**Report of a case**

A patient of 2 years was brought for consultation by the mother due to the presence of green teeth in the mouth. The intraoral examination showed the presence of anterior primary teeth with a green aspect. All deciduous teeth were affected. No other changes involving the oral soft tissues were observed. According to the story told by the mother, the teeth were erupting in the mouth with a discoloration. The pigment involved about 5/6 of the crown of the incisor teeth (Fig. 1). At 3 years of age, the child presented with the first primary molars with half of the crown pigmented (Fig. 2).

Furthermore, the tip of the cusp canine was also pigmented. The clinical crown of all teeth showed hypoplasia areas between the middle and incisal thirds (Fig. 2). The teeth of the lower arch also showed a green coloration on the crown (Fig. 3). At this time, the child has not had the eruption of the primary second molars. The upper and lower canines had green pigmentation in the incisal half of the crown. The pigmentation of the first primary molars reached ¾ of the crown. According to the presence of this green pigmentation in the crown of teeth, a diagnosis of green teeth associated with a systemic disorder has been established.

The past medical history revealed that the child’s mother was 28 years old and that she had consulted 4 times during the prenatal period. During pregnancy, the mother had a urinary tract infection that was treated with cephalaxin. The mother was admitted in premature labor and premature rupture occurred an hour earlier. The child was born by cesarean section with difficult extraction. The indication for this type of delivery was due to dystocia progression.

At birth, the child had been in gestation for 31 weeks and was weighing 1660 g. The cause of preterm birth was...
premature rupture of membranes. After birth, the child proved to be hypotonic, cyanotic and did not cry. A cycle of positive-pressure ventilation and cardiac massage for recovery were made. In addition, she was intubated in the delivery room. The newborn had neonatal asphyxia and slow infusion. In addition, the child had tocotraumatism, anemia, bloating, and early sepsis. The child was treated with ampicillin, gentamicin, dopamine, furosemide, pipercillin, tazobactam, dopamine, dobutamine, fentanyl and metronidazole. A diagnosis of large lesion in subcapsular hepatic lobe was established and it was classified as grade II splenic laceration that was treated expectantly. Infection of the child was treated for 52 days and the child remained intubated (intubation) for 12 days due to apnea and surfactant administration.

During her stay in the Intensive Care Unit, volume globular and bilirubin levels were investigated (Table 1). Jaundice was diagnosed on the second day of life by yellowish discoloration of the skin during routine physical examination. The pathological jaundice was treated with phototherapy. The child had the same blood type (A positive) as the mother and received packed red blood cells. The child presented with hyperbilirubinemia for about 2 months.

The mother was instructed on the importance of diet and oral hygiene was reinforced. As the green pigmentation affected the deciduous dentition and there was a possibility it could also occur in the permanent dentition, the child’s mother was advised to return for follow-up dental. No cosmetic treatment has been established in affected tooth. As the child was premature, we observed a delayed eruption of deciduous teeth and consequently also in the permanent dentition.

**Discussion**

Bilirubin is produced mainly by degradation of the hemoglobin of red blood cells, the final product of the catabolism of heme [6]. The clinical jaundice in newborns occurs when bilirubin levels in blood plasma reaches values between 5 and 7 mg/dL. Severe neonatal hyperbilirubinemia, when unmonitored or untreated, can progress to acute bilirubin encephalopathy [10].

Neonatal jaundice is one of the most common problems for newborn infants during the first weeks of life, affecting approximately 60–70% of term babies and almost all premature babies [11]. According to Shrestha et al. [13], the risk factors associated with preterm birth are: inadequate antenatal checkup (52%), maternal age <20 years (34.7%), antepartum hemorrhage (23.4%) and pregnancy induced hypertension (13.1%). Morbidities more commonly observed in preterm infants are: clinical sepsis (66.7%), hyperbilirubinemia (58.8%), birth asphyxia (26.8%) and hyaline membrane disease (23.5%) [12]. The patient in this case report showed the major health problems related to prematurity, especially sepsis, neonatal jaundice, and apnea. In this case, bilirubin was considered pathologic due to lesion in the subcapsular lobe resulting from tocotrauma. The levels of total bilirubin reached 30.58 mg/dL in the 18th day of life.

According to Watanabe et al. [12], total bilirubin levels between 1.2 and 1.8 mg/dL are already capable of inducing pigmentation tooth. These authors have identified levels of bilirubin in deciduous teeth obtained from 2 patients with a history of severe liver dysfunction and hyperbilirubinemia. The teeth were histologically analyzed and bilirubin was extracted and quantified by spectrophotometry.

![Table 1 - Values of globular volume and levels of bilirubin of the patient](image)

<table>
<thead>
<tr>
<th>Date</th>
<th>Globular volume</th>
<th>Direct bilirubin</th>
<th>Indirect bilirubin</th>
<th>Total bilirubin</th>
</tr>
</thead>
<tbody>
<tr>
<td>June 18</td>
<td>15%</td>
<td>N</td>
<td>N</td>
<td>N</td>
</tr>
<tr>
<td>June 19</td>
<td>32.9%</td>
<td>N</td>
<td>N</td>
<td>N</td>
</tr>
<tr>
<td>June 20</td>
<td>33.6%</td>
<td>3.27 mg/dL</td>
<td>9.29 mg/dL</td>
<td>12.56 mg/dL</td>
</tr>
<tr>
<td>June 21</td>
<td>37.5%</td>
<td>3.27 mg/dL</td>
<td>9.29 mg/dL</td>
<td>12.56 mg/dL</td>
</tr>
<tr>
<td>June 22</td>
<td>35.2%</td>
<td>Absence of bilirubin in the partial examination of urine</td>
<td>8.11 mg/dL</td>
<td>20.08 mg/dL</td>
</tr>
<tr>
<td>June 23</td>
<td>38.3%</td>
<td>11.97 mg/dL</td>
<td></td>
<td></td>
</tr>
<tr>
<td>June 25</td>
<td>32.2%</td>
<td>Jaundice++/4+</td>
<td></td>
<td></td>
</tr>
<tr>
<td>June 28</td>
<td>30.1%</td>
<td>11.55 mg/dL</td>
<td>23.25 mg/dL</td>
<td>11.70 mg/dL</td>
</tr>
<tr>
<td>June 30</td>
<td>41.0%</td>
<td>N</td>
<td>N</td>
<td>N</td>
</tr>
<tr>
<td>July 1</td>
<td>37.4%</td>
<td>N</td>
<td>N</td>
<td>N</td>
</tr>
<tr>
<td>July 2</td>
<td>36.0%</td>
<td>N</td>
<td>N</td>
<td>N</td>
</tr>
<tr>
<td>July 3</td>
<td>34.4%</td>
<td>N</td>
<td>N</td>
<td>N</td>
</tr>
<tr>
<td>July 4</td>
<td>34.0%</td>
<td>20.25 mg/dL</td>
<td>8.08 mg/dL</td>
<td>28.33 mg/dL</td>
</tr>
<tr>
<td>July 5</td>
<td>46.0%</td>
<td>21.12 mg/dL</td>
<td>9.46 mg/dL</td>
<td>30.58 mg/dL</td>
</tr>
<tr>
<td>July 9</td>
<td>43.9%</td>
<td>N</td>
<td>N</td>
<td>N</td>
</tr>
<tr>
<td>July 11</td>
<td>34.9%</td>
<td>N</td>
<td>N</td>
<td>N</td>
</tr>
<tr>
<td>July 13</td>
<td>N</td>
<td>10.6 mg/dL</td>
<td>4.02 mg/dL</td>
<td>14.68 mg/dL</td>
</tr>
<tr>
<td>July 17</td>
<td>33.2%</td>
<td>N</td>
<td>N</td>
<td>N</td>
</tr>
<tr>
<td>July 20</td>
<td>30.9%</td>
<td>N</td>
<td>N</td>
<td>N</td>
</tr>
<tr>
<td>July 21</td>
<td>N</td>
<td>9.14 mg/dL</td>
<td>1.67 mg/dL</td>
<td>10.81 mg/dL</td>
</tr>
<tr>
<td>July 27</td>
<td>26.0%</td>
<td>10.46 mg/dL</td>
<td>5.35 mg/dL</td>
<td>15.18 mg/dL</td>
</tr>
</tbody>
</table>

N = sample not collected.
analysis revealed a green line parallel to the incremental lines of dentin. These authors analyzed the deciduous canine of a child with a history of bilary atresia and congenital hyperbilirubinemia at birth that showed that the tooth were colored brown in the middle of the crown to half of the root. The other case was a boy of 7 years of age with a history of hemolytic anemia and hyperbilirubinemia at 18 months of life showed total bilirubin levels 20-60 mg/dL. The upper central incisors and lateral incisors had a change of color of green. Furthermore, this color change reached half incisal of the dental crown. These findings were similar to those observed in primary teeth of children of this report.

The normal color of teeth is determined by the shades of blue, green and pink of the enamel and is reinforced by shades of brown or yellow dentin below. Metabolic diseases and systemic factors can affect the development of teeth and cause tooth discoloration, such as: alkaptonuria, erythropoietic porphyria congenital, amelogenesis imperfecta, dentinogenesis imperfecta, tetracycline stain, and congenital hyperbilirubinemia. However, the teeth do not show morphological changes [9]. The patient in this case report had green teeth and areas of enamel hypoplasia. Despite these changes, the teeth remained preserved in its form.

The areas of enamel hypoplasia observed in the crown of the anterior teeth may be associated with systemic factors and intubation, because the child remained intubated for 12 days. The literature reveals that tracheal intubation in the neonatal period can cause enamel hypoplasia [14, 15].

The occurrence of hyperbilirubinemia in childhood is an important fact because the consequences of this systemic disorder can seriously compromise the color of teeth. The first report of case about green teeth associated with jaundice was performed by Weyers [16] in 1956. Since then, the dental literature shows some reports of patients who developed green teeth associated to neonatal hyperbilirubinemia [17-28].

It is known that the location and extent of pigmentation tooth corresponds to the period of tooth formation [28]. In this case report, the patient presented dental pigmentation in the crown of all deciduous teeth. These dental changes corresponded to the period when she was admitted to the Intensive Care Unit. Normally, deciduous tooth calcification process begins in the 4th month of intrauterine life and finishes 11 months after birth. The dental crown of incisors is completely formed 1 month after birth and the canines and molars after 6 months [29]. According to current knowledge, enamel mineralization proceeds in 2 steps. At first, ameloblasts secrete a protein matrix in which hydroxyapatite crystals are deposited up to a concentration of about 25%. In a second step, the enamel maturation, enamel matrix proteins are degraded and resorbed almost completely, while hydroxyapatite crystals grow in thickness, until the enamel attains a mineral content of about 95%. Consequences of this formation in 2 stages are also evident in the wisdom teeth showing crown pigmentation [30].

Enamel formation can be disrupted during the course of a systemic disorder, and all the teeth that form in this period may be marked by bands of enamel malformed. After healing of illness, the formation of normal enamel is restored again. Disorders induced by tetracycline or bilirubin showed clinically the time of injury to the tooth germ by the incorporation of these substances to the enamel [12, 31].

In this case, the patient report presented different levels of pigmentation of deciduous teeth. The crowns of the central and lateral incisors were more affected as half of their crowns were affected. The pigmentation was observed involving only half of the crown of the molars. Moreover, canine teeth were the least affected. As calcification of the permanent dentition has its beginning from birth, the possibility of dental pigmentation affecting the permanent teeth, especially the first molar, has not been ruled out.

High serum bilirubin levels favor the deposition of the product of hemoglobin degradation in many tissues, including soft tissue and mineralized tissues. When hyperbilirubinemia occurs during dental development, the teeth can develop a green color due to the bilirubin deposition. This intrinsic pigment keeps teeth permanently pigmented. The blood vessels of the dental follicle are the source of nutrients for the ameloblasts to synthesize the tooth enamel [32]. Thus, the pigmentation of the tooth is most likely the result of the pigment passing through the bloodstream to ameloblasts and, then, to the tooth enamel. Bilirubin is a yellow pigment. However, when this pigment undergoes oxidation it becomes a green pigment otherwise called biliverdin. In the past, it was believed that the accumulation of this pigment was responsible for the green teeth associated with hyperbilirubinemia. However, a study developed by Shibata et al. [33] proved that the green color comes from the accumulation of bilirubin and not of biliverdin.

The correct diagnosis for the cause of dental discoloration is important as, invariably, it has a profound effect on treatment outcomes. According to Shibata et al. [33], the diagnosis of bilirubin pigmentation is usually based on a clinical history of jaundice combined with the green coloration. Therefore, it would seem reasonable that health care providers have an understanding of the etiology of tooth discoloration in order to diagnose and recommend a more appropriate treatment.

Actually, cosmetic dentistry is presented as a source to meet the esthetic and functional wishes of patients and professionals. The tooth whitening is a technique used to solve the esthetics of discolored teeth. It can be performed for both vital and non-vital teeth. Various chemicals such as 35% hydrogen peroxide, carbamide peroxide and sodium perborate and others are employed [34]. In general, the esthetic result of whitening of natural teeth has a better result when compared to the restoration or resin crowns. In this case, the natural appearance of the enamel is more aesthetic in relation to restorative materials. The first reports of tooth whitening in primary teeth indicate the use of carbamide peroxide 10% for treatment of dental fluorosis. This technique requires a high level of adherence of the patient and the esthetic result does not always guarantee long treatment time due to the limited life of these teeth [35].

Moreover, some adverse effects of the tooth whitening may occur, such as: the external root resorption and dentin sensitivity [36]. According to Lee et al. [37], the use in children and adolescents of some dental bleaching agents
containing carbamide shows that: (1) 1 in every 2-3 patients may experience tooth sensitivity and/or gingival irritation after bleaching treatment, which may be more traumatic an experience for children than adults; (2) depending on dose, duration, frequency, and route, studies indicate excessive exposure to peroxide can be potentially harmful; (3) degree of potential toxicity and harmful outcomes increases in those who overuse whiteners – a concern in teenagers; (4) careful case selection using stringent criteria is suggested for primary teeth whitening; (5) whitening in healthy adolescents is a case-by-case determination that must include the weighing of risks (oral health and age) vs benefits (improved esthetic perception). However, there are some options of treatment for green teeth in the permanent dentition, such as: composite veneers, crowns, esthetic facings and possibly bleaching [18, 19, 27].

Authors' contributions/Wkład autorów

According to order.

Conflict of interest/Konflikt interesu

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Ethics/Etyka

The work described in this article has been carried out in accordance with The Code of Ethics of the World Medical Association (Declaration of Helsinki) for experiments involving humans; EU Directive 2010/63/EU for animal experiments; Uniform Requirements for manuscripts submitted to Biomedical Journals.

REFERENCES / PIŚMIENNICTWO


