Green teeth associated with hyperbilirubinemia: a case report

ABSTRACT

Background Green stain is an uncommon clinical condition associated with deposition of bilirubin in the dental hard tissues. The staining of the teeth is due to hyperbilirubinemia caused by systemic conditions.

Case report The purpose of this report is to present a case of green teeth of the primary dentition in a 17-month old girl associated with hydrocephalus and Hirschprung's disease.

Conclusion The clinical characteristics of the teeth may help in the diagnosis of current or past systemic diseases.

Keywords Bilirubin; Green teeth; Hyperbilirubinemia; Pigmented teeth.

Introduction

The developing tooth germ is sensitive to a wide range of systemic disturbances and is unable to recover once it is damaged, and the tooth often acts as a repository of information on systemic insults received during development. For the primary teeth, this record begins at about the 4th month of intra-uterine life and continues through the first year of life. Thus, defects of the primary teeth might have the potential for identification of children who have undergone certain systemic insults during prenatal and early postnatal life [Bhat.M and Nelson. K.B, 1989]. Hyperbilirubinemia defined as a serum concentration of bilirubin greater than 1.5 mg/100 ml, and jaundice is clinically visible when serum concentration become greater than 7.0 mg/100 ml. Hyperbilirubinemia in neonatal life can result from a variety of problems experienced during this period. It can be due to infections or metabolic, toxic, genetic and anatomic abnormalities. Prolonged conjugated hyperbilirubinemia, or neonatal cholestasis, is caused by a defect in the excretion of ciliary salts, which leads to a decrease in the secretion of bile salts, which in turn leads to a decrease in the secretion of bile constituents, including bilirubin [Guimarães et al., 2003]. Bilirubin is extensively deposited throughout the body during hyperbilirubinemia, although it disappears from soft tissues after remission, nevertheless in hard tissues bilirubin is permanently trapped, because after maturation these tissues lose metabolic activity. The presence of hyperbilirubinemia during enamel and dentin formation results in dental green pigmentation, due to accumulation of intrinsic staining by bilirubin oxidation [Guimarães et al., 2003].

The prevalence of green teeth is low, and literature provides data of 48 cases. Although hyperbilirubinemia has been reported in literature, green pigmentation of primary teeth has been rarely documented. Considering that green staining of dental hard tissues is such an uncommon condition, the purpose of this article is to present such a case [Luciane et al., 2004].

Case report

A 17-month old male patient was referred to the Department of Paedodontics KLE VK Institute of Dental Sciences, Belgaum (India) from the Paediatric Department of KLE’s Dr Prabhakar Kore Hospital, for the unusual green pigmentation of his deciduous teeth.

History

The medical history of the child revealed that he had been treated for hydrocephalus and Hirschprung disease (congenital aganglionic megacolon). The child was delivered with caesarian section and was a full-term birth, however he was unable to pass stools for 9 consecutive days after birth. The abdominal sonography showed the presence of multiple dilated bowel loops which is a characteristic feature of Hirschprung disease. The laboratory findings showed presence of E-coli and Citrobacter freundii complex indicating the presence of sepsis. Increased levels of urea and creatinine were seen, the total bilirubin and direct bilirubin levels were also assessed at different time intervals for one month (Table 1), showing progressive increase of the values. The child was surgically treated for the hydrocephalus and enlarged colon and as a consequence his medical condition improved. The child also had a total hearing loss in the right ear and partial loss in the left ear. On subsequent follow-ups the paediatrician noted a greenish discoloration of the erupted primary teeth, hence the patient was referred to our department.
Dental findings

Intraoral examination showed no soft tissue abnormalities. Hard tissue examination showed erupted deciduous maxillary central and lateral incisors, first molars and right canine. In the mandibular region central and lateral incisors, first molars and right canine were erupted. All the erupted teeth exhibited a greenish discoloration of the cervical and middle third of the crown surface (Fig. 1). The incisal third of anterior teeth and occlusal one third of posterior teeth showed no discoloration. The morphology of these teeth was unaltered. The crown surface had a rough texture compared to the smoothness of the normal dentition. There were no hypoplastic or hypocalcific regions. The teeth presented no mobility.

Scratching test of the teeth with a scaler did not remove the discoloration which confirmed that the color was intrinsic in nature. Radiographic finding showed a large coronal and radicular pulp space with thinner enamel, dentine and cementum lining (Fig. 2, 3). Histologic sectioning was not done as the teeth were not extracted. Based on the history and clinical findings the dental diagnosis was chlorodontia or green teeth due to hyperbilirubinemia. As aesthetics was not a major consideration for the parents and the child was very young no treatment was undertaken. The patient is kept on a regular follow-up schedule.

Discussion

Clinical and experimental studies have indicated that serum bilirubin, a degradation product of haemoglobin, may be deposited during the formation of hard dental tissues and entail discoloration. Therefore, bilirubin discoloration can be clinically visible after eruption [Watanabe et al., 1999]. Full-term infants with hyperbilirubinemia during the neonatal period were reported to have intrinsic staining, low frequency of enamel hypoplasia, and enamel hypocalcification [Grahnen and Granath, 1962]. This finding correlates with the present case. Fortunately, neonatal management can now usually prevent dangerous elevations of bilirubin [Bhat and Nelson, 1989]. There is no estimate, but the prevalence of green teeth is low. The literature provides data for 48 cases and all are associated with systemic conditions. Green-stained teeth have been reported in infants and children, with an age range from newborn to 11 years.

The literature does not indicate any sex predilection. In the present case, the green stain occurred because of hyperbilirubinemia caused by intrahepatic cholestasis associated with sepsis by E. coli and Citrobacter freundii complex. Jaundice has long been recognised as a clinical manifestation of infection in the newborn and during infancy. The jaundice associated with sepsis appears to be due to a partial injury of the liver cells, which retain at least some capacity for conjugating bilirubin, although the excretion of bile into the ductal system is impaired, as a result, the bile accumulates in the cells. The deciduous teeth of children who survive the brain damage of kernicterus are frequently stained and with a olive green color. Clinical studies have shown the association of kernicterus and of jaundice with hypoplastic green stained dentine [Tank, 1951] first correlated the variation in the intensity of green staining with the medical history. The deposition of pigment, its oxidation, and its distribution on the tooth surfaces increase and decrease at the same rate as the degree of icterus. However, green staining cannot be correlated directly with the duration and depth of jaundice. There are exceptions as reported by Marsland.

<table>
<thead>
<tr>
<th>AGE</th>
<th>TOTAL BILIRUBIN</th>
<th>DIRECT BILIRUBIN</th>
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<tr>
<td>3 days</td>
<td>5.3mg/dl</td>
<td>0.1 mg/dl</td>
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<tr>
<td>8 days</td>
<td>20.7 mg/dl</td>
<td>2.8 mg/dl</td>
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<tr>
<td>9 days</td>
<td>18.3 mg/dl</td>
<td>7.3 mg/dl</td>
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<tr>
<td>12 days</td>
<td>21.8 mg/dl</td>
<td>14.7 mg/dl</td>
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<tr>
<td>13 days</td>
<td>22.2 mg/dl</td>
<td>15.1 mg/dl</td>
</tr>
<tr>
<td>21 days</td>
<td>30.3 mg/dl</td>
<td>26.2 mg/dl</td>
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**TABLE 1** Bilirubin levels at various time intervals.
and Gerrard in which a child who had been jaundiced for only 1 week also developed green teeth [Miller et al., 1960], considered that to cause obvious green staining of teeth the bilirubin concentration must exceed 30 mg/100 ml which also correlates with the present case. Hypoplasia in green stained teeth occurred in the majority of cases in patients with a medical history of biliary atresia because of the malabsorption of fat soluble vitamins which does not occur in cases with sepsis which again correlates with the present case where the child showed no hypoplasia of the teeth.

The diagnosis of green teeth is usually based on a clinical history of jaundice combined with green tooth discoloration. The differential diagnosis of green teeth includes extrinsic discolorations caused by chromogenic bacteria or medicaments and intrinsic discoloration caused by dentinogenesis imperfecta, amelogenesis imperfecta, tetracycline and congenital erythropoietic porphyria [Robert et al., 1993; James et al., 1989]. In the present case the pigment could not be removed using a scaler, and the history and lab findings indicated hyperbilirubinemia due to sepsis. The microbiological analysis could not be done in this case. Hals and Nielsen et al. [1969] in their experiments on rats confirmed that green pigmentation was never seen in enamel but only in pre-enamel and predentine. They also observed irregular deposits of pre-enamel, incomplete mineralisation of enamel, disturbances of the mineralisation of dentine and inclusion of pulp tissue in the dentine. This explains the radiologic finding of the present case with large pulp chambers and deficient enamel and dentine. Since the hard tissue of the permanent teeth starts just after birth, if the subject suffers hyperbilirubinemia only during the neonatal period, the teeth are usually not affected. In the present case hyperbilirubinemia was present in the first month of the child’s life, so it would be interesting to know whether the permanent teeth will be affected, and therefore the patient is kept on a close follow-up. Since the child was 17 months old aesthetics was not a priority for the parents, they were educated about the various cosmetic options such as bleaching, composite veneers or polycarbonate crowns. The approach suggested by Giunta and Tsamtsouris [1978] involving the use of transillumination with ultraviolet light to accelerate the breakdown and dissipation of bilirubin products could also be tried.

The parents were counselled regarding the problems the child might encounter with peer teasing, this may hamper his social interaction and affect his self-esteem. In this case a cosmetic treatment could be done, but we preferred to wait [Zaia AA et al., 1993].

Conclusion

Today as result of preventive measures and recent advances in medical treatment, green teeth are considered a rarity. However, if such condition does occur, aesthetic corrections can be accomplished with the newer techniques in cosmetic dentistry. This becomes necessary especially when the child is teased by his peers resulting in limited social interaction. As pediatric dentists we are in a privileged position to diagnose, treat, and provide counseling to these children.

References

- Tank G. Two cases of green pigmentation of the deciduous teeth associated with hemolytic disease of newborn. JADA 1951;42:302-6.