A Report of 2 Cases of Green Pigmentation in the Primary Dentition Associated With Cholestasis Caused by Sepsis

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ABSTRACT

Green pigmentation of teeth is uncommon but, when it occurs it is a cause of anxiety to the child and family. It also causes the child to lose self-esteem and increases social issues for the family. The purpose of this paper was to present the management of 2 unrelated patients who presented with green primary teeth following sepsis-induced liver dysfunction and hyperbilirubinemia in infancy. (J Dent Child 2008;75:91-4) Received December 10, 2006 | Last Revision May 1, 2007 | Revision Accepted May 10, 2007.

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Intrinsic tooth discoloration is caused when pigmentation is incorporated into the tooth substance.[1,2] There are a wide range of causes, including dental caries, trauma, internal resorption, restorative materials, amelogenesis and dentinogenesis imperfecta, tetracyclines, fluorosis, and systemic illness during tooth formation. Systemic illnesses are commonly hemolytic disease of the newborn, hyperbilirubinemia, and congenital porphyria.[1-4] Other systemic conditions that have been associated with intrinsic discoloration are premature birth, neonatal respiratory dysfunction, internal hemorrhage, congenital hypothyroidism, biliary hypoplasia, metabolic diseases, hepatic dysfunction, and neonatal staphylococcal sepsis.[3,4]

Intrinsic green discoloration of teeth is considered to be rare.[3-5] There are 52 published cases of green teeth in the literature, and the majority of reports are related to the primary dentition.[4-7] There are fewer reports of green staining in the permanent dentition. All the published cases had a history of biliary atresia,[8-10] which is the most common cause of green discoloration in both dentitions reported in the literature. Other causes reported include respiratory distress, hemolytic disease, bile duct occlusion, absence of bile ducts, hemolysis in utero, biliary hypoplasia, cholestasis associated with sepsis, congenital cytomegalovirus infection, and erythroblastosis fetalis.[4-7] As the pediatric care of infants and young children improves, it is likely that more children with life threatening illnesses will survive. It is possible that the frequency of green teeth will increase as a consequence.

The pigment, deposited in the teeth during the formation of hard dental tissue in patients suffering from severe jaundice, has been shown to be bilirubin and not biliverdin, as previously thought.[11,12] Bilirubin is derived from senescent red blood cells and from turnover of other heme-containing proteins such as cytochromes. Bilirubin is poorly soluble in aqueous solutions at physiological pH values and, consequently, it is transported in plasma bound to serum albumin. In the liver, bilirubin is conjugated to form a diglucuronide. Conjugated bilirubin is much more water-soluble than unconjugated (free) bilirubin. Almost all the plasma bilirubin in the normal state is unconjugated.[13] Bilirubin is extensively deposited throughout the body in hyperbilirubinemia, causing discoloration that can vary from yellow to deep shades of green.[4]

Not all infants with hyperbilirubinemia have green teeth, however, and it could be that the level of "unconjugated" bilirubin is more important than "conjugated" bilirubin, but

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no reference to confirm this has been found in the literature to date. Following resolution of hyperbilirubinemia, the pigmentation disappears from the soft tissues due to high tissue turnover. Hard dental tissues lose their metabolic activity after maturation. Hence, the bilirubin is permanently trapped leading to intrinsic staining by bilirubin oxidation, which is clinically visible in erupted teeth.[4,5,11]

The purpose of this paper was to present 2 cases of green-pigmented teeth, resulting from hyperbilirubinemia caused by sepsis, to further increase the knowledge regarding the possible causes of green discoloration of the primary dentition

CASE 1

A 12-month-old boy was initially referred to our clinic with discolored teeth. Weighing 0.765 kg, he was born prematurely at 24 weeks gestation by spontaneous breech to a 42-year-old mother. At birth, he was diagnosed with a large pulmonary ductus arteriosus and a massively distended left heart. The ductus arteriosus was ligated 6 weeks after birth. Two weeks after birth, the child developed necrotizing enterocolitis with perforation sepsis—resulting in Candidal peritonitis, which was treated with drainage and antibiotics to good effect. He developed jaundice and hyperbilirubinemia, with a total bilirubin of 8.7 mg/dl (normal=0-1.2 mg/dl). The level of unconjugated bilirubin was 0.2 mg/dl (normal=0-1.2 mg/dl). He suffered a respiratory arrest with circulatory shock secondary to Klebsiella septicemia and was intubated for almost a month. During this time, he suffered consumptive coagulopathy and thrombocytopenia, acute renal and liver failure, and an intraventricular hemorrhage. He has retinopathy of prematurity and secondary blindness.

Oral examination revealed that all the erupted primary anterior teeth were green in color. Subsequent dental reviews have confirmed that erupting posterior teeth also have green discoloration. The distribution of the discoloration was chronological with the deepest discoloration affecting the earliest formed dental hard tissues (Figure 1). The oral soft tissues were healthy and normal.

Dental treatment was initially restricted to preventive advice and advice on how the discoloration could be masked at a later date. At 3 years of age, the patient attended the day surgery unit for examination of the eyes under general anesthesia. The opportunity was taken to fissure seal the primary molar teeth and place direct veneers on the primary anterior teeth (Figure 2). The anterior teeth were aesthetically restored to mask the green discoloration using flowable compomer material directly bonded to acid-etched enamel using bonding agent.

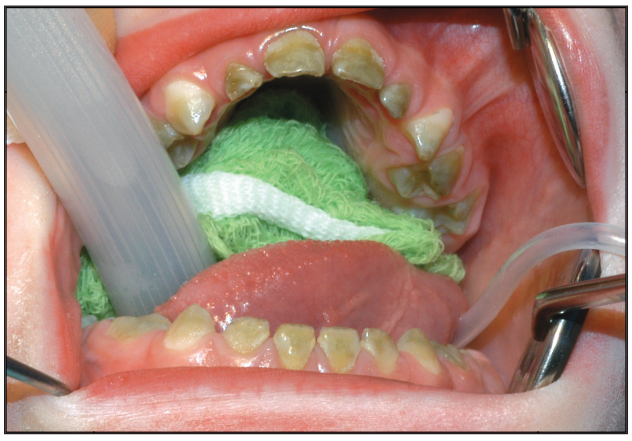

Figure 1. Clinical photograph showing chronological green dental discoloration in case 1.

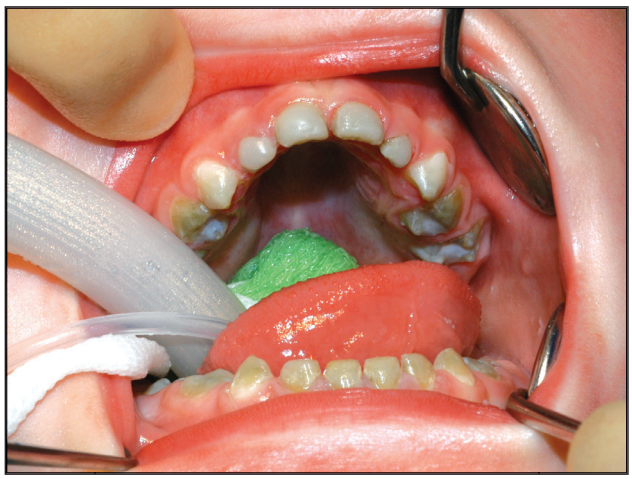

Figure 2. Clinical photograph after completion of dental treatment to mask green discoloration in case 1.

CASE 2

A 16-month-old boy was referred by his general practitioner for the treatment of discolored teeth. He was a twin born prematurely at 23 weeks gestation weighing 0.520kg, with a large patent ductus arteriosus, which was ligated 4 weeks after birth. He was subsequently diagnosed with necrotizing enterocolitis, resulting in perforation and Candida peritonitis. Additionally, he had coagulase-negative *Staphylococcus aureus* sepsis. He was jaundiced and had a total bilirubin of 19.3 mg/dl (normal=0-1.2 mg/dl). The level of unconjugated bilirubin was 2.6 mg/dl (normal=0-1.2 mg/dl). An abdominal ultrasound carried out at 4 months revealed no evidence of biliary atresia. Like the case 1 patient, he suffered an intraventricular hemorrhage and has retinopathy of prematurity, rendering him blind. In addition, he has language delay and difficulties with swallowing.

Oral examination revealed that all the erupted primary teeth had green discoloration, which was chronological in pattern. The green discoloration was mostly confined to the incisal half of the primary maxillary incisors, the incisal tip of the maxillary canines, and the incisal third of the first molars (Figures 3 and 4). The mandibular incisors seemed to be more severely affected, with green pigmentation extending over most of the clinical crown. The maxillary lateral incisors were also hypoplastic. No abnormalities were detected in the oral soft tissues.

This patient underwent examination of the eyes under general anesthesia at 18 months. As in case 1, the opportunity was taken to provide some dental treatment and direct adhesive veneers were placed on all the maxillary anterior teeth.

Figure 3. Clinical photograph showing green discoloration of primary maxillary anterior teeth in case 2.

Figure 4. Clinical photograph showing green discoloration of primary mandibular anterior teeth in case 2.

At the last dental review, the child was 3 years old. The anterior veneers had deteriorated, particularly on the primary lateral incisors which were hypoplastic. Dental treatment was once again carried out under general anesthesia in combination with an ophthalmic examination. The hypoplastic primary lateral incisors were extracted, all the remaining anterior teeth had new direct adhesive veneers constructed, and fissure sealants were placed on the posterior teeth (Figure 5).

Both patients have subsequently been reviewed and are enjoying good oral health and all the restorations and fissure sealants are intact.

Figure 5. Clinical photograph after completion of dental treatment to mask green discoloration and extraction of the primary maxillary lateral incisors in case 2.

DISCUSSION

Hyperbilirubinemia is defined as serum concentration of bilirubin greater than 1.5 mg/100 ml. Jaundice is usually manifested when the serum concentrations reach 7 mg/100 ml.[3,5] Neonatal hyperbilirubinemia occurs fairly commonly, resulting in visible jaundice.[3] Bilirubin is extensively deposited throughout the body during hyperbilirubinemia. It is permanently trapped in the hard tissue, but not in the soft tissue. Therefore, any teeth in the formative stages may also be affected by the excess bilirubin becoming intrinsically stained.[3] The diagnosis of bilirubin pigmentation is made if there is a clinical history of jaundice coupled with green tooth discoloration. Occasionally, patients may also have associated enamel hypoplasia.[5] To date, there only appears to be one case report in the literature of green teeth staining as a result of hyperbilirubinemia caused by intrahepatic cholestasis associated with sepsis by Candida species and *Klebsiella oxytoca*.[5] Herbert et al, however, describe a case where the patient had a Staphylococcal sepsis but did not include it as a possible cause of jaundice.[3,5] In case 1 of this report, the patient suffered from Candida peritonitis and Klebsiella septicemia. In case 2, the patient suffered from Candida peritonitis along with a coagulase negative *S aureus* sepsis.

Green pigmentation is visible through the translucent enamel[4] and can be very difficult to mask with restorative materials. It has been reported that pigmentation is brightest when the tooth first erupts and slowly fades with age. A possible explanation for this is the loss of enamel translucency as the child grows older, thus interfering with the transmission of the green coloration from the dentine.[5] Barta et al verified that tooth discoloration caused by blood incompatibility became less evident after 3 years of age.[14] Both cases described in this report appear to have a chronological pattern, which would indicate that discoloration is likely only in the primary dentition and in perhaps the cusps of the first permanent molars.

The appearance of a child's first tooth is often a developmental milestone for the family, and the appearance of a discolored tooth can be disturbing and a cause for anxiety.[2] As the child grows older and starts to attend school, the presence of discolored teeth can cause difficulties with peers.[2] In the present cases, the parents were anxious that every effort be made to prevent this from occurring. For this reason, physicians caring for children with a history of hyperbilirubinemia should be aware of the risk of green pigmentation in the primary and permanent dentition and should advise parents of the potential risk. As the child grows older, cosmetic treatment of these teeth becomes a priority to improve self-esteem and assist social integration.[2,5]

There are various treatment options, including composite veneers or crowns and possibly bleaching for the permanent dentition.[2,5] In the primary dentition, options are more limited and will include strip crowns or composite facings directly bonded to the anterior teeth. In the present cases, strip crowns would be the treatment of choice for anterior teeth. This option will be explored as the children become older.

CONCLUSIONS

Intrinsic green discoloration of teeth is not common, but can be expected to be seen with increasing frequency as a result of advances in pediatric medical care—including liver transplantation. It is important to reassure parents and discuss the cosmetic treatment available. Regular dental care with correct emphasis on prevention is necessary so that any cosmetic work that is possible can be done on a healthy dentition and periodontium.

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