

MINERVA

PEDIATRICA

VOLUME 68 · No. 6 · DECEMBER 2016



EDIZIONI · MINERVA · MEDICA

Green teeth caused by neonatal hyperbilirubinemia

Dear Editor

Neonatal hyperbilirubinemia occurs in about 60% of newborns during the first week of life and causes accumulation of bilirubin pigment within the skin and mucous membranes which manifests as a yellow discoloration. When bilirubin levels remain elevated for several months, bilirubin pigments deposit throughout the body, including the teeth. There is no estimate but the prevalence of green teeth is low.¹ The major complication of an elevated serum bilirubin is classic neonatal neurotoxicity, kernicterus. Clinical studies have demonstrated the association of kernicterus and of jaundice with hypoplastic green-stained dentine. Histological evaluation of green-stained deciduous teeth from patients with hyperbilirubinemia has shown bilirubin deposits. We report a case of green teeth of the primary dentition in a patient with a history of neonatal cholestasis.

We present a case of prolonged conjugated hyperbilirubinemia associated with Alagille Syndrome.² Our patient, a female premature newborn, presented the following characteristics: monochorionic diamniotic twin pregnancy, delivery at 34 weeks of gestation and birth by urgent cesarean due to alterations in the cardiocardiographic trace. Advanced neonatal reanimation was required, with tracheal intubation. Apgar Score was 3 (1') and 5 (5') respectively. Umbilical arterial pH of 7.09. The physical dimensions at birth were as follows: weight 1870 g, length 44 cm and head circumference 30 cm.

During the first hours of life generalized hypotonia were observed together with hypotension, so ionotropic support with dopamine was required. The newborn had a tendency to metabolic acidosis (minimum serum bicarbonate levels 11.5 mmol/L). Facial abnormalities like hypertelorism, epicanthus and broad forehead were observed. Mechanical ventilation was performed during 28 days. From admission hematuria was observed (10350 cel/mcL) and subsequently anuria developed, the following analytical results were obtained: creatinine 274 mcM/L, urea 22.3 mmol/L, sodium 124 mEq/L, potassium 6 mEq/L, chloride 91 mEq/L, calcium 1.85 mmol/L, total protein 33 g/L. In consequence, peritoneal dialysis was required from the third day of life and maintained for 24 days. The renal Doppler ultrasound showed an increase in arterial resistance and an almost total absence of diastolic flow, indicative of peripheral and bilateral renal artery stenosis. The isotope renogram showed reduced cortical uptake. The coagulation study

was abnormal and a state of disseminated intravascular coagulation (DIC) required several infusions of packed cells, fresh plasma and platelets. A cardiological examination revealed the presence of a foramen ovale type atrial septal defect with left to right shunt, together with pulmonary artery stenosis. Prior to the withdrawal of peritoneal dialysis, progressive jaundice was observed, developing to a cholestatic syndrome during several months with direct bilirubin levels of 427.5 mcM/L (N: 0–3.4 mcM/L) and peak levels of serum ferritin of 6600 mcg/L (N: 25–200 mcg/L), although these levels fell after treatment with chelating agents and vitamins. The newborn developed *Enterobacter cloacae* sepsis at 24 days of life, with raised levels of bilirubin, GGT and factor V.

With the data at that moment available, the existence of neonatal hemochromatosis was proposed as a diagnostic possibility, and a total blood exchange transfusion was performed, together with the infusion of intravenous immunoglobulin (1 g/kg). Hepatic Doppler ultrasound showed mild intrahepatic biliary dilatation. The liver scan showed biliary elimination delay and the liver biopsy revealed bile duct paucity, but the possible presence of hemosiderin deposits was discounted. With respect to hydroelectrolytes, the levels of sodium, potassium and chlorine were normal, accompanied by hyperphosphataemia (3.68 mmol/L) and hypokalaemia (1.25 mmol/L), which improved after the oral administration of 1.25 hydroxycholecalciferol. Arterial hypertension (154/86 mmHg) required the administration of captopril (0.5 mg/kg/8 h) and losartan (0.3 mg/kg/day).

A genetic study for Alagille Syndrome confirmed a heterozygous duplication in the exon 6 area of the *JAG1* gene, located at 20p12.1-p11.23 with an autosomal dominant mode of inheritance. Currently the patient is 3 years old and has chronic renal insufficiency in predialysis stage, she is expecting a kidney transplant. Furthermore, she has a deep hypoacusia. The jaundiced skin had resolved but intraoral examination showed a dark green pigmentation of her fully erupted primary teeth (Figure 1).

The prevalence of green-stained teeth associated with hyperbilirubinemia is unknown. About 50 cases of green dental discoloration have been reported, mostly in the dental literature. Most of these cases occurred in children with hyperbilirubinemia subsequent to biliary atresia. Perhaps because conjugated bilirubin is more water-soluble and is able to be incorporated into enamel and dentin during matrix formation of the developing dentition.

In a review of 48 children with green-stained teeth secondary to hyperbilirubinemia, the mean duration of jaundice was 24.6 weeks and the maximum serum total bilirubin level was 20 to 90 mg/dL.¹ However, green staining cannot be correlated directly to the duration and depth of jaundice, it seems that other factors may determine which patients develop green teeth and which patients do not. This might be related to the density of the matrix, or to individual disturbances in the absorp-



Figure 1.—Patient's green teeth.

tion of calcium, phosphate and fat-soluble vitamins. Extreme preterm birth or extremely low birth weight may be additional risk factors.³ The primary teeth appear to be most affected, although staining of permanent dentition has been reported mostly in patients with a medical history of biliary atresia who can also develop hypoplasia of enamel.⁴

Stained primary dentition will eventually be replaced with normal permanent teeth whereby most experts do not recommend treatment of primary teeth. As children age, treating the teeth can help to improve self-esteem. Treatment considerations for green-stained teeth include restorative composite crowns and resin restorations as well as bleaching techniques and the use of transillumination with ultraviolet light to accelerate the breakdown and dissipation of bilirubin products.

José UBEROS *, Mariam TRIBAK,
María P. TORTOSA PINTO, Aida RUIZ LÓPEZ
Pediatric Service, San Cecilio Clinical
Hospital, Granada, Spain

*Corresponding author: José Uberos, Pediatric Service, San Cecilio Clinical Hospital, Calle Dr. Oloriz, 16, 18012, Granada, Spain. E-mail: juberos@ugr.es

References

1. Guimaraes LP, Silva TA. Green teeth associated with cholestasis caused by sepsis: a case report and review of the literature. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2003;95:446-51.
2. Uberos J, Moreno L, Muñoz-Hoyos A. Hypertension and Biliary Ductopenia in a Patient with Duplication of Exon 6 of the *JAG1* Gene. *Clin Med Insights Pediatr* 2012;6:61-6.
3. Battineni S, Clarke P: Green teeth are a late complication of prolonged conjugated hyperbilirubinemia in extremely low birth weight infants. *Pediatr Dent* 2012;34:103-6.

4. Sommer S, Magagnin K, Kramer PF, Tovo MF, Bervian J: Green teeth associated with neonatal hyperbilirubinemia caused by biliary atresia: review and case report. *J Clin Pediatr Dent* 2010;35:199-202.

Conflicts of interest.—The authors certify that there is no conflict of interest with any financial organization regarding the material discussed in the manuscript.

Manuscript accepted: April 29, 2015. - Manuscript revised: April 3, 2015. - Manuscript received: January 20, 2015.

(Cite this article as: Uberos J, Tribak M, Tortosa Pinto MP, Ruiz López A. Green teeth caused by neonatal hyperbilirubinemia. *Minerva Pediatr* 2016;68:502-3)

© 2015 EDIZIONI MINERVA MEDICA
Online version at <http://www.minervamedica.it>
Minerva Pediatrica 2016 December;68:503-5

Children's drawings as a window into creativity: ratings of disrupted emotionality in children's drawings predict creativity in narrative play construction

Dear Editor,

Children's drawings are an underused source material in the research and clinical practice of contemporary academic child psychiatry. Their underutilization is likely a product of their past frequent application in unsystematic and theory-driven fashions. Applying systematic research to children's drawings offers a means to gather reliable data on emotional development through an easily administered and quick test that may bring enjoyment to the child and serve as a stimulant for further inquiry into the child's life.

One systematic approach to addressing emotional development in children's drawings is that of Koppitz (1968).¹ Koppitz systematically evaluated hundreds of children's human figure drawings among healthy school-aged children and those in a clinical population to determine statistically-significant group differences. Koppitz found thirty items which frequently appeared in children's drawings that represented the drawing's accuracy, detail and complexity. These were termed developmental indicators (DIs). Additionally, Koppitz found thirty items which were found significantly more frequently in the clinical population than in the control population, and were hypothesized to be indicative of disturbed emotionality. These were termed emotional indicators (EIs). Through a coding manual with demonstrated inter-rater reliability, contemporary studies are capable of using Koppitz' procedure to develop insights into childhood emotional development.¹