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Dental intrinsic green pigmentation from primary to mixed dentition: clinical and histological findings

ABSTRACT

Aim The aim of this report is to present a case of a child with green pigmentation of the primary dentition caused by bilirubin elevation due to choleostasis during neonatal life, and the 5-year follow-up.

Case report The case presented initially with bands of green pigmentation of all primary teeth in a pattern that followed the time of their calcification, with those formed earlier being more severely affected. Fading of the green pigmentation was detected during the follow-up, while erupted lower permanent incisors were normal. Histological findings of an exfoliated primary incisor showed a green line at the enamel-dentine junction with the external surface of the dentine showing a band of variable width and irregularly arranged tubules.

Conclusion Bilirubin green pigmentation of primary teeth follows a chronological pattern and its intensity fades with time. Overlying enamel in affected areas may appear thinner.

Keywords Choleostasis; Green pigmentation; Hyperbilirubinaemia; Primary dentition.

Introduction

Teeth intrinsic green pigmentation is an unusual condition, first described in 1912 [Langmead, 1912], that can affect both primary and permanent dentitions.

It is mainly associated with increased levels of bilirubin, a haemoglobin degradation end-product in the serum and must be differentially diagnosed from other intrinsic stains caused by medication, such as tetracycline.

Deposited bilirubin can cause intrinsic pigmentation if serum concentration exceeds 30 mg/100 ml [Amaral et al., 2008]. Pigment is permanently trapped in the dental hard tissues as they lose their metabolic activity after maturation and discolouration is produced by bilirubin oxidation [Watanabe et al., 1999; Guimares and Silva, 2003; Alto et al., 2004; Naudi et al., 2008]. Pigmentation reported varies from dark green to yellow [Watanabe et al., 1999; Chamber et al., 2012], with the extent and the intensity directly related to the duration and severity of the associated underlying disease [Watanabe et al., 1999; Alto et al., 2004; Chamber et al., 2012].

Reports of the literature are not in agreement on where intrinsic pigmentation deposits, enamel or dentine. Boyle and Dinnermarn [1941] suggest that pigmentation is incorporated in enamel caps, while according to Naudi et al. [2008] pigment is deposited in dentine during its formative stages. Recently, Guimaraes and Silva [2003] suggested that prenatally formed enamel was not itself discoloured but it scattered the light through the stained tissue of developing dentine, and Monte Alto et al. [2004] that bilirubin is incorporated during the pre-eruptive enamel development as mature enamel is virtually impermeable to intrinsic stains.

The aim of this report is to present a case of a child with severe green pigmentation of the primary dentition caused by bilirubin elevation due to choleostasis during neonatal life and the 5-year follow-up until the eruption of permanent teeth. Macroscopic and microscopic findings of an exfoliated primary tooth are also presented.

Case report

A 16-month old Caucasian male was referred to the Department of Paediatric Dentistry from his paediatrician for diagnosis and management of severe green discolouration of his primary teeth.

His medical history revealed premature birth (1540 g) at 31 weeks of gestation. Due to bowel rupture, he underwent surgery just after birth, which was complicated with Klebsiella Pneumoniae infection. He developed choleostasis and direct hyperbilirubinaemia at day 11 and had elevated bilirubin levels from day 19 (unconjugated 33,7 and conjugated 21,5) until resolution of choleostasis at day 39.

Intraoral findings

At initial clinical examination (age 16 months) all erupted primary teeth showed an unusual green



FIG. 1 Initial examination at the age of 16 months. Extensive green pigmentation at the cervical part of all primary teeth.



FIG. 4 Macroscopic photograph of the tooth after decalcification and bisection. Notice a green band of dentine on its external surface following the contour of its crown.



FIG. 2 Examination at the age of 3 years. Green pigmentation following a chronological pattern with incisors having a dull and rough enamel and canines showing thinning of the enamel on affected parts of the tooth.



FIG. 3 Examination at the age of 6.5 years. Primary teeth presented with enamel attrition, dull surface and canines showed thinning of the enamel on green affected areas. Erupting permanent teeth showed no signs of discolouration.



FIG. 5 Microscopic examination. No green pigment is evident, but the external band of dentine is more intensely eosinophilic (A). Notice the abrupt change in direction of the dentine tubules (B) (hematoxylin and eosin stain, A x200, Bx400).

pigmentation in bands, mainly located at the gingival third of the crown, fading towards the middle and presenting an opaque white color incisally (Fig. 1). The oral soft tissues were within normal limits, without any evidence of green pigmentation. Immediate management of the case involved consultation with the parents about the aetiology of the discolouration and alleviation of their aesthetic concerns and anxiety regarding the occurrence in permanent dentition. The parents were advised to keep the patient in a 6-month recall to monitor the development of the primary and permanent dentitions.

Patient returned at the age of three years having green discolouration in all primary teeth with the distribution of the pigmentation following a chronological pattern (Fig. 2). Maxillary and mandibular primary incisors were more severely affected with the dark green pigmentation covering more than two thirds of the crown, while in primary canines and molars pigmentation was mainly located in the upper third of the crown with the cusps of the teeth being less affected. Enamel overlaying the pigmented areas was pitted in central incisors with irregular rough texture and appearing as thinner in canines. Oral hygiene was good and no caries were detected. Zirconia crowns for the affected anteriors were presented as a treatment

option to improve patient's aesthetics, but parents did not decide to follow this treatment.

At the final recall appointment at the age of six and a half years, pigmentation of all primary teeth was fading away, with its intensity being less evident and the color turning to a more yellow-green. Enamel surface texture was smooth and dull, with visible thinning of the outer enamel layer on the areas covering the green pigmentation. Permanent lower incisors had just erupted showing no evidence of discolouration or enamel defect at their incisal third (Fig. 3). No x-rays were taken throughout the follow-up due to parental concerns on radiation safety.

Histological examination

An exfoliated primary mandibular central incisor was subjected to histological examination, after fixation in 10% buffered formalin solution and decalcification in MicroDecfast (LaboSphera, Italy). Macroscopic examination of the tooth after bisection showed that its external surface was covered by a green band of dentine following the contour of its crown (Fig. 4). The staining was more intense at the junction of this band and the underlying dentine comprising the bulk of the tooth appeared normal. This staining band stopped abruptly, without becoming thinner in a gingival

direction. Microscopic examination of 5 µm-thick hematoxylin and eosin stained section did not reveal any green pigment but the external band of dentine, profoundly corresponding to the green band, was more intensely eosinophilic and the dentine tubules showed an abrupt change in direction (Fig. 5). The rest of the dentine was normal and no interglobular dentine was seen. The apical part of the pulp cavity was occupied by tertiary dentine, the cementum appeared normal and the radical surface of the tooth showed many dentinoclastic lacunae consistent with normal resorption.

Discussion

This is a rare case of a young child with a history of choleostasis and increased bilirubin levels early in neonatal life, presenting with intrinsic green pigmentation of his primary teeth and thinning of the enamel overlaying the pigmented incisal third of the canines. During the five years follow-up, fading of the green pigment to yellow green was seen while the incisal third of the lower permanent incisors appeared normal. Histology of an exfoliated primary incisor was consistent with this diagnosis.

Green pigmentation has been reported more often in primary dentition and the most common aetiological factor is biliary atresia [Amaral et al., 2008; Naudi et al., 2008; Sommer et al., 2010; Chamber et al., 2012; Rakaускаite et al., 2014]. Less common factors that have been reported are biliary hypoplasia, haemolytic disease, bile duct occlusion, cytomegalovirus infection, erythroblastosis fetalis and choleostasis as in the present case [Guimares and Silva, 2003; Naudi et al., 2008].

Diagnosis in the present case was intrinsic green pigmentation due to hyperbilirubinaemia caused by intrahepatic choleostasis due to sepsis in association with neonatal jaundice. Bilirubin is normally insoluble and is converted in the liver to a soluble conjugate in order to be eliminated from the body. In early days of life, liver cells are immature and therefore unable to successfully release the enzyme that conjugates bilirubin, increasing its unconjugated levels in plasma. Although in the literature it has been reported that discolouration occurs with either direct or indirect hyperbilirubinaemia [Guimares and Silva, 2003], a report by Sommer et al. [2010] classifies hyperbilirubinaemia according to the predominant type of bile pigment in the plasma. The present case cannot be clearly classified in any of the two categories: it was mainly associated with increased levels of unconjugated bilirubin due to overproduction of bilirubin and impaired bilirubin conjugation by glucuronyl transferase. However, due to the sepsis that influenced hepatocellular function it was also characterised by increased levels of conjugated bilirubin.

Bilirubin levels in this case were elevated until day 39

after birth, therefore involvement of only primary teeth was anticipated, with those developed earlier being more severely affected [Rakaускаite et al., 2014; Ball, 1964; AlQahtani et al., 2010]. Pigmentation of more than half of the crown of incisors, but less than one third of the crowns of the canines and molars with no involvement of the root is in accordance with the "chronological pattern" of the disease reported by most authors [Amaral et al., 2008; Tank, 1971; Borta et al., 1989; Neville et al., 1995]. Although in all cases there is a clear demarcation line separating pigmented and non-pigmented areas of the crown, localisation of the pigmented area differs with some reporting the incisal third [Sommer et al., 2010; Rakaускаite et al., 2014] and others the cervical third [Amaral et al., 2008; Chamber et al., 2012].

Pigmentation was more evident when primary teeth first erupted and faded slowly to a more yellow hue as the patient grew older, as reported previously [Tank, 1951; Borta et al., 1989]. This may be attributed to loss of enamel translucency as the patient grows interfering with the transmission of green pigment, directly affecting the reflection from discoloured dentinal tissues [Guimares and Silva, 2003; Naudi et al., 2008; Sommer et al., 2010] or may be attributed to some metabolic activity of the dentin.

In the present case pitted and dull enamel was observed at the labial surface of the incisors and thin at the buccal surface of the canines. This finding also underlines the chronological pattern of the disease. As enamel is formed from the inside to the outside, prenatally formed incisors are only slightly affected while postnatally formed canines present with evident thinning of the enamel outer layer. Although, due to the lack of parental compliance, it cannot be concluded whether thinning was present from the beginning or it can be attributed to physiological abrasion and masticatory function. In the literature there are no reports supporting the above mentioned hypothesis. Enamel hypoplasia has been reported in cases associated with biliary atresia [Guimares and Silva, 2003] and chronic liver disease [Morisaki et al., 1990; Watanabe et al., 1999; Amaral et al., 2008; Bimstein et al., 2011] and has been attributed to changes in the organic matrix of the developing enamel due to variations in hepatic efficiency and/or to the effects of osteopenia and other disturbances in the metabolism of calcium and phosphate [Morisaki et al., 1990; Watanabe et al., 1999; Guimares and Silva, 2003; Amaral et al., 2008; Bimstein et al. 2011; Chamber et al., 2012].

Macroscopically, the green line seen after tooth sectioning corresponds to bilirubin deposition in the dentin layer formed during the hyperbilirubinaemia phase [Watanabe et al., 1999]. Due to its colour the pigment deposited in dentine is most probably biliverdin, a metabolic end product of haemoglobin presented with increased levels in jaundice. Although

bilirubin is expected to give a yellow pigment once absorbed by the tissues, all evidence in the literature supports its presence attributing the pigmentation to its increased levels in the blood [Watanabe et al., 1999; Chamber et al., 2012].

The microscopic features of the tooth studied in the present case were in accordance with those previously reported [Watanabe et al., 1999; Amaral et al., 2008; Bimstein et al., 2011; Rakauskaite et al., 2014]. Enamel was removed during the decalcification process and therefore the presence of bilirubin in enamel could not be evaluated. We can assume though that the pigmented band corresponds to the outer dentin layer that was more intensely eosinophilic and characterised by a change in the direction of dentin tubules, i.e. representing dentin formed during the hyperbilirubinaemia phase [Watanabe et al., 1999; Bimstein et al., 2011; Chambers et al., 2012]. Collagen is more eosinophilic and this was seen in the pigmented band, while the change in the direction of odontoblasts, seen both in our case and in previous reports, is strongly reminiscent of tertiary dentine, the one produced as a response to certain stimuli (i.e. inflammation). If this represents an indication for altered function of the odontoblasts during the hyperbilirubinaemia phase, it should be further evaluated. No pigmented enamel or abnormal dentine tubules were seen [Watanabe et al., 1999], while the irregular cement-dentin junction reported by Bimstein et al. [2011] could not be observed, as the tooth root has been lost during normal exfoliation.

In this case, owing to the age of the subject, patient management was based on prevention, monitoring and parental counseling. It was important to explain to the parents about the green pigmentation chronological pattern and duration through the life of the tooth, to alleviate any concerns about potential harm to the function and health of the teeth and discuss the small likelihood of the pigmentation to be present to the permanent teeth. At the recall visit at age 3, parents were offered the option of restoring the anterior green teeth with zirconia crowns [Ashima et al., 2014] to mask the discolouration, but declined to follow this recommendation due to the patient's discomfort and possible high cost. In the literature, there are no cases reporting any restorative intervention in the primary dentition. Restoration of such discoloration in the permanent teeth has been reported with composite, porcelain and lab cured polymer resin veneers [Sommer et al., 2010; Chamber et al., 2012].

Conclusion

Bilirubin green pigmentation of primary teeth follows

a chronological pattern. Its intensity fades over time, while overlying enamel in affected areas becomes thinner. It may cause anxiety and discomfort to both parents and child.

Clinical significance

The case report adds to the literature information on a rare condition directly related to the patient's medical history in neonatal life that severely affects his dental hard tissues. The aetiological factors, clinical appearance, microscopic findings and possible treatment options are presented in great detail. It is interesting as the case is followed for a long time period and the progression of the condition is described until the eruption of first permanent teeth.

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