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PRIKAZ SLUČAJA
CASE REPORT

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Zeleno obojenje zuba povezano s razinom bilirubina

Green Teeth Related to Bilirubin Levels

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Sažetak

Svrha: Opisan je oblik zubne pigmentacije kod nedonoščadi uzrokovan taloženjem serumskog bilirubina tijekom kalcifikacije zuba. Razina bilirubina praćena je tri mjeseca nakon rođenja, u razdoblju dok se još uvijek razvijaju krune primarnih zuba. Takva je anomalija asimptomatska, ali je i razlog za veliku zabrinutost i tjeskobu u obitelji. **Prikaz slučaja:** Navedeni slučaj potvrđuje koliko je važna medicinska anamneza, a u ovom slučaju i testovi za dugotrajnu konjugiranu hiperbilirubinemiju kako bi se potvrdila dijagnoza i utješili roditelji. Estetska terapija nije poduzeta jer je dijete bilo premalo, a i njegovo stanje nije zahtijevalo takvu intervenciju. Plan liječenja uključivao je kontrolu svaka tri mjeseca kako bi se održavalo oralno zdravlje bez karijesa. **Zaključak:** Budući da ovo rijetko stanje utječe na zube, roditelji će najvjerojatnije zbog terapije potražiti doktora dentalne medicine koji zna o kakvoj je anomaliji riječ pa će odabrati multidisciplinarni pristup.

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Uvod

Zeleni zubi iznimno su neuobičajena abnormalnost koja može utjecati na mliječne ili trajne zube. Kada se pojavi prekomjerna hiperbilirubinemija u plazmi, ona uzrokuje reverzibilnu obojenost svih tkiva osim zuba (1), jer su žučni pigmenti trajno *zarobljeni* zbog gubitka metaboličke aktivnosti nakon sazrijevanja. Pigmentacija može biti od žute do tamnijih nijansi zelene (2). To je razlog za tjeskobu u obitelji koja često traži doktora dentalne medicine kako bi doznala dijagnozu. Korigiranje ove abnormalnosti može biti kompliciranо (3) i mora biti rezultat zajedničkog rada zubara i liječnika.

U ovom članku opisan je slučaj u kojemu je zelena pigmentacija mliječnih zuba bila uzrokovana hiperbilirubinemijom. Ovo je prvo izvješće u kojem se prikazuje razina bilirubina tijekom prva tri mjeseca života nedonoščeta, u razdoblju kada se još uvijek razvijaju krune mliječnih sjekutića.

Prikaz slučaja

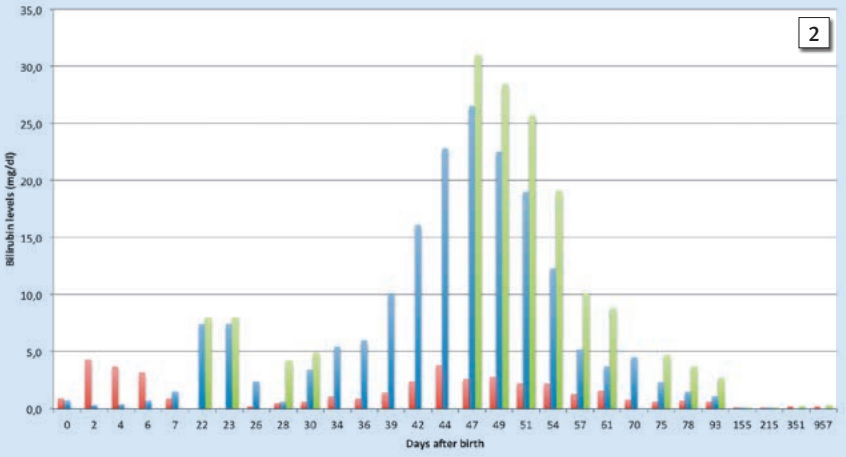
Trogodišnji dječak došao je u pratnji roditelja u ordinaciju dentalne medicine zbog zelene pigmentacije na maksilarnim sjekutićima. U medicinskoj anamnezi roditelji su naveli da je njihov sin rođen u gestacijskoj dobi od 27 tjedana i carskim rezom te je nakon rođenja težio 940 grama. U suradnji s rodilištem u kojem je dijete došlo na svijet prikupljeno je više informacija – novorođenče je imalo tešku respirator-

Introduction

Green teeth are an extremely uncommon abnormality that can affect both primary and permanent teeth. When excessive hyperbilirubinemia occurs in the plasma, it causes reversible staining of all tissues except for the teeth (1), because the bile-pigments are permanently trapped due to loss of metabolic activity after maturation. The pigmentation may vary from yellow to deep shades of green (2). It is a cause of anxiety to the family who often visit a dentist to obtain a diagnosis. The management of this abnormality may be complicated (3) and must be a result of a collaboration of a dentist and a physician. This paper reports a case in which green pigmentation of primary teeth is caused by hyperbilirubinemia. This is the first report demonstrating bilirubin levels during the first three months of life of a premature baby, period in which the crowns of deciduous incisors are still forming.

Case report

The parents of a 3-year-old Caucasian male child accompanied their son to a dental office due to the presence of green pigmentation on his maxillary incisors. The medical history reported by the parents included that the male infant was born at 27 weeks and 1 day of gestation by Cesarean section and weighted 940 grams at birth. After having contacted the hospital in which the child was born, more information



Slika 1. Abnormalan klinički izgled kruna u mliječnoj denticiji
Figure 1 Abnormal clinical appearance of the crowns in the primary dentition.
Slika 2. Nekonjugirani bilirubin (crveni), konjugirani bilirubin (plavi) i ukupni bilirubin (zeleni) koncentracije (mg / dl) kod novorođenčadi odmah nakon rođenja (nulti dan) te 957 dana poslije
Figure 2 Unconjugated bilirubin (red), Conjugated bilirubin (blue) and Total Bilirubin (green) Concentrations (mg/dl) in neonate at birth (day 0) until 957 days after.

nu insuficijenciju zbog sindroma respiratornog distresa, jaku prijevremenu retinopatiju, periintraventrikularno krvarenje trećeg stupnja, bronhopulmonalnu displaziju te konvulzivni sindrom, a bila je potrebna i gastrostrostomija zbog poremećenog gutanja.

Fizikalni pregled bio je usredotočen na opći nutritivni status i na znakove bolesti jetara. Zbog poremećaja u gutanju novorođenče nije moglo sisati. Do četvrtog mjeseca bilo je hranjeno isključivo s pomoću nosnog katetera, a zatim mu je postavljena gastrična sonda. Zbog komplikacija tijekom prijevremenog porođaja, djetetu se tek poslije razvila neuropsihomotorika, te je zbog toga trebalo posebnu njegu, nutritivnu potporu, fizioterapiju i multidisciplinarno praćenje. Bilo je i na terapiji tetraciklinom te je kontinuirano dobivalo magnezijevo mlijeko, fenobarbiton, cefaleksin te vitamine A i D.

Osim zelenih zuba, intraoralni klinički pregled pokazao je normalan razvoj bez abnormalnosti gingive te normalnu strukturu i boju mekih tkiva. Uočena je normalna pigmentacija očnjaka i kutnjaka koji se formiraju šest mjeseci nakon rođenja. Roditelji su istaknuli da su njihovu sinu počeli nicati maksilarni sjekutići u dobi od 15 mjeseci. Također su rekli da se zelena pigmentacija na kruni maksilarnih sjekutića nalazila od trenutka kada su se zubi pojavili u ustima (slika 1.). Ti se zubi obično potpuno formiraju mjesec dana nakon rođenja. Budući da je pigmentacija bila na gornjim sjekutićima, ali ne i na očnjacima, možemo zaključiti da se ono što je uzrokovalo pigmentaciju dogodilo oko prvog mjeseca života.

Zbog sistemskih komplikacija uzorci krvi prikupljeni su tijekom prva tri mjeseca nakon rođenja te su izmjerene razine bilirubina (slika 2.). Plan dentalne terapije proveden je nakon što je roditeljima rečeno da se zbog zelenih zuba ne trebaju zabrinjavati jer ih uzrokuje komplikacija koja se dogodila prije. Jedina pritužba roditelja bila je estetika, simptoma nije bilo, dječak je pokazivao osobine djeteta s posebnim potrebama te još uvijek nije bio dovoljno zreo za estetsku terapiju. Zato je odlučeno pratiti ga svaka tri mjeseca kako bi se održavalo oralno zdravlje bez karijesa do promjene u trajnu denticiju.

was gathered: the newborn developed a severe respiratory failure due to the respiratory distress syndrome, severe retinopathy of prematurity, grade III peri-intraventricular hemorrhage, bronchopulmonary dysplasia, convulsive syndrome, and he needed gastrostomy due to a deglutition disturbance.

The physical examination focused on general nutritional status as well as on signs of liver disease. Due to the deglutition disturbance, the newborn could not be breastfed (nutritive suction). Up to the fourth month he had been fed exclusively through a nasal catheter and after that a gastric catheter was installed. Due to the prematurity complications, the child exhibited a severe neuro-psychomotor development delay, and, therefore, he needed a special care, nutritional support, physiotherapy and multidisciplinary approach. He had undergone a tetracycline therapy and was continuously using magnesium milk, phenobarbital, cephalexin and vitamins A and D.

Apart from the green teeth pigmentation, an intra-oral clinical examination revealed normal development, no gingival abnormalities, normal texture and color of the oral soft tissues. A normal pigmentation of canines and molars, which were formed 6 months after birth, was observed. The parents reported that the maxillary incisors erupted when the child was 15 months old. They also said that the green pigmentation was present in the crown of the maxillary incisors from the moment they appeared in the mouth (Figure 1). The maxillary incisors are usually completely formed one month after birth. As the pigmentation was present in the maxillary incisors but was not present in the canines, we concluded that what caused the pigmentation had occurred around the infant's first month of life.

Due to systemic complications, blood samples were collected during the first three months after birth, and bilirubin levels were measured (Figure 2). A dental treatment plan was elaborated after the family had been reassured that the green teeth represented no worries to the present situation of the child because it was caused by a complication that had hap-

Rasprava

Stalna zelena pigmentacija dentina glavna je promjena uzrokovana hiperbilirubinemijom i važan je pokazatelj doktorima dentalne medicine. Također je važno znati da je žutica uzrokovana mnogim procesima – od onih benignih do za život opasnih (4).

Zeleni zubi pojavljuju se u dentinu samo tijekom kalcifikacije zuba (5) – obojenost se može pojaviti i na mliječnima i na trajnima (6). Stadiji razvoja zuba već su dobro poznati, kalcifikacijsko razdoblje za maksilarne središnje sjekutiće počinje u 15-om tjednu intrauterino i kod dječaka završava mjesec dana nakon rođenja. Možemo pretpostaviti da se kod djeteta iz ovog prikaza hiperbilirubinemija pojavila u tom razdoblju. No teško je odrediti na kojoj se razini to dogodilo. Postoji samo sugestija korelacije između različitih stupnjeva zelenog obojenja u mliječnoj denticiji i ozbiljnosti pigmentacije (7). Područja zuba koja su bila kalcificirana nakon hiperbilirubinemije obično su normalne boje, no vidi se oštra crta koja razdvaja zeleni dio od zdravoga. U ovom slučaju nije bila vidljiva nikakva linija, što bi moglo upućivati na produljeno razdoblje hiperbilirubinemije (2, 8).

Hipoplazija cakline u oba maksilarna sjekutića mogla je biti uzrokovana promjenama organske matrice tijekom razvoja cakline koja nastaje zbog metaboličkih poremećaja, ali je vjerojatnije da je povezana s učincima osteopenije i ostalih poremećaja u metabolizmu kalcija i fosfata koji se pojavljuju u slučaju kronične bolesti jetara (4). Ovaj je pacijent uzimao i tetraciklin, a jedna od nuspojava toga lijeka jest njegova ugradnja u tkiva koja se kalcificiraju u razdoblju kada se primjenjuje. Ovakva trajna promjena boje može se pojaviti u spektru od žute ili sive do smeđe, ali nikada nije zelena kao kod ovog pacijenta. Štoviše, tijekom primjene tetraciklina krune mliječnih zuba kod ovog su pacijenta već bile kalcificirane.

Prevencija nakon svih prijevremenih porođaja mora biti prioritet – dijete iz ovog prikaza slučaja prolazilo je kroz dugotrajno razdoblje hiperbilirubinemije, rođeno je preuranjeno i bilo je pothranjeno, što upućuje na predisponirajuće rizične čimbenike za mnoge abnormalnosti, a među njima je i nicanje zelenih zuba u kasnom djetinjstvu (9). Uobičajeni uzroci teške neizravne hiperbilirubinemije su sepsa, preuranjeni porođaj, nepodudaranje krvne grupe i nedostatak G6PD-a (9). U ovom slučaju dogodili su se preuranjeno rođenje i nepodudarnost krvne grupe. U slučaju da dijete bude spremno za estetsku terapiju, ona će uključivati restauraciju kompozitnim smolama ili transiluminaciju ultraljubičastim svjetlom sa svrhom da se razgradi bilirubin (6).

U većini pregledane literature takvi su slučajevi samo opisani, te je bilo potrebno proučiti više tekstova kako bi se dobila informacija o prevalenciji, ozbiljnosti, etiologiji i

pened in the past. The parents' only complaint was the esthetics. There were no symptoms, the child had special needs and was still not mature enough to receive a cosmetic treatment. Therefore, it was decided to have regular checkups every 3 months to maintain oral health with no caries until the teeth exfoliation.

Discussion

The permanent green pigmentation of dentin tissues as the main alteration caused by hyperbilirubinemia is of interest to the dentist. It is also important to know that jaundice is caused by many disease processes ranging from benign to life threatening (4).

The green teeth pigmentation is an alteration that occurs in the dentin only during its calcification (5), both for primary and permanent teeth (6). The teeth formation stages are already well known, the calcification period for the maxillary central incisors starts at 15 weeks *in utero* and is completed around one month after birth for males. We can then presume that for the child described in this case, hyperbilirubinemia occurred within this range. However, it was difficult to determine to what extent the bilirubin levels were elevated. There is only an assumption regarding a correlation between various degrees of green staining in the primary dentition and severity of pigmentation (7). Teeth areas that had been calcified after the hyperbilirubinemia period usually show normal color and a sharp dividing line is observed separating the green portion from the normal one. In this case, no line was visible, suggesting a more prolonged period of hyperbilirubinemia (2, 8)

Enamel hypoplasia in both maxillary incisors could have been caused by changes in the organic matrix of the developing enamel resulting from metabolic disturbances, but it is more likely that it is related to the effects of osteopenia and other disturbances of calcium and phosphate metabolism encountered in chronic liver disease (4). Also, this patient had undergone a tetracycline therapy. One of the side-effects of tetracycline is its incorporation into the tissues that are calcifying at the time of its administration. However, the permanent discoloration varies from yellow or gray to brown, but it is never green as it was in this patient. Moreover, during the tetracycline administration for this patient, the primary teeth crowns had already been calcified.

The prevention of all preterm births must be a priority. In this case, the child passed through a prolonged period of hyperbilirubinemia and was born as an extremely premature and underweight baby, which is one of the predisposing risk factors for many abnormalities such as the green teeth eruption in late infancy (9). More common causes of severe indirect hyperbilirubinemia are sepsis, prematurity, blood group incompatibility and G6PD deficiency (9). In the present case, the baby was born very preterm, at a very low birth weight and blood group incompatibility. In cases where the child is capable of receiving a cosmetic treatment, it involves composite resin restorations or transillumination with ultraviolet light aiming at bilirubin breakdown (6).

Most of the reviewed literature consisted of case reports. Further research is needed to provide information on

kliničkoj prezentaciji zelenih zuba. Iz opisanoga zaključujemo da se zdravstveni stručnjaci češće suočavaju sa slučajevima kada je potreban multidisciplinarni pristup. Zeleni zubi pokazuju da je pacijent doživio razdoblje hiperbilirubinemije tijekom razvoja zubne krune. Kako ovo nije česta abnormalnost, roditelji se uplaše te odlaze doktorima dentalne medicine kako bi riješili problem. Tijekom postavljanja konačne dijagnoze zelenih zuba doktor dentalne medicine trebao bi biti obaviješten o medicinskom uzroku i morao bi tražiti mišljenje doktora opće medicine. Provjera razine serumskog bilirubina u prvim mjesecima nakon rođenja može potvrditi dijagnozu zelenih zuba.

Sukob interesa

Ni jedan autor ovoga rada nije bio u sukobu interesa (Barbério, Zingra, Santos i Machado), a to se odnosi i na ostale suradnike.

the prevalence, severity, etiology and clinical presentation of green teeth. From this case, we concluded that health professionals should take a multidisciplinary approach when dealing with green teeth and similar abnormalities. There is some evidence to suggest that the patient experienced a period of hyperbilirubinemia during the formation of dental crown and the green pigmentation of the teeth is a rare consequence of neonatal hyperbilirubinemia. Since the green teeth pigmentation is not a common abnormality, families are often frightened and look for a dentist in order to solve the problem. When identifying green teeth, the dentist should be aware of medical causes e should look for a physician in order to establish a final diagnosis. Serum bilirubin levels must be checked in the first months after birth to confirm the diagnosis of green teeth.

Conflict of interests

There are no conflicts of interest between any authors of the manuscript (Barbério, Zingra, Santos and Machado) and other co-workers, companies or organizations that could inappropriately influence the authors' work.

Abstract

Objective: To describe a form of tooth pigmentation caused by serum bilirubin deposition during dental calcification in a premature child. The bilirubin levels during the three months after birth are presented. In this period the crowns of the primary teeth are still forming. Such anomaly does not have symptoms, but has a displeasing effect and great anxiety within the family. **Case report:** The case reported here highlights the relevance of past medical history, especially the diagnoses of prolonged conjugated hyperbilirubinemia, in this case, to confirm them and to comfort the family. In this case, no esthetic treatment had been undertaken because the child was still too young and not yet concerned. The dental treatment plan included regular checkups every 3 months for maintaining oral health to prevent caries. **Conclusion:** As this is a rare condition that affects the teeth, parents will most likely look for a dentist for treatment. Therefore, dentists must be aware or such abnormalities and take a multidisciplinary approach, thus making it possible to establish a final diagnosis.

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Key words
Tooth Discoloration; Infant, Premature; Hyperbilirubinemia, Neonatal; Bilirubin

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