A Rare Case of Congenital Syphilis and a Supernumerary Fourth Molar in an Early 20th Century African American Woman

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ABSTRACT Congenital syphilis is a disease recognized for interfering with odontogenesis, producing specific dental characteristics including Hutchinson's incisor, Moon's molar, Fournier's molar and mulberry molar, while its past treatments including mercury are known to affect amelogenesis. Supernumerary teeth, mainly associated with syndromes, are not commonly found in cases of congenital syphilis. A rare case of congenital syphilis in an individual (P000707) treated with mercury and a mandibular left fourth molar with normal morphology is presented.

Materials and Methods: During a systematic examination of 28 skeletons with treponemal disease at the Smithsonian museum in Washington, DC, a supernumerary mandibular distomolar in one individual (P000707) was revealed.

Results: P000707 was an African American female, 26 years of age. Dentition showed severe enamel hypoplasia of the maxillary and mandibular incisors, left canine, and upper first molars, consistent with the effects of treatment of congenital syphilis by mercurial compounds. Crown of the left mandibular distomolar has typical molar morphology but is smaller in size than other permanent molars. Arrangement of grooves resembles the +4 pattern, but is complex due to crenulation. Oblique x-ray revealed that the fourth molar had one root with a pulp chamber extending towards the apex, suggesting taurodontism. No other distomolar teeth were present.

Conclusions: Congenital syphilis and treatment containing mercury may not influence the development of supernumerary teeth due to: (1) the age at which the development of the fourth molar takes place, (2) the stage of the infection at the time of development and (3) the age at which treatments containing mercury are administered to patients with congenital syphilis.

ous systems are affected. Pathological signs appear in birth affecting initial crown formation. However, these two stages of the disease. During the early stage, skele- dental abnormalities do not occur in all cases of congenochondritis, and osteomyelitis (Hira et al., 1985; ranges from 30 to 50% (Putkonen and Paatero, 1961), McLean, 1931) while during the late stage, signs can while changes in first permanent molars range between include frontal bossing, short maxilla, high palatal arch, 3 and 37% (Berfield, 1971). saddle nose, Higoumenakis's sign, diaphysitis, metaphysitis and sabre shins (Fiumara and Lessell, 1970; syphilis due to its antibacterial effects (Hutchinson, Rasool and Giovender, 1989). However, the disease is 1874, 1878; Warner, 1881). Even though mercury was most recognized for interfering with tooth formation seen to benefit infected individuals, it was also seen to (odontogenesis), producing certain characteristic teeth produce dental abnormalities that were different from including Hutchinson's incisors, Moon's molar, Fournier's molar and the mulberry molar (Fournier, 1886; Hutchinson, 1863; Karnosh, 1926; Moon, 1884). Even though these characteristic dental signs in congenital syphilis are seen in the permanent teeth (upper central incisors and first molars), which erupt approximately at

Congenital syphilis is a disease caused by the trans- 6-8 years of age, the dental abnormalities in these teeth mission of Treponema pallidum, from the mother to the are produced during the early stages of the disease, that fetus during pregnancy or at birth. In the neonate, vari- is, once the infection and fever set in around the time of tal manifestations include periosteal reactions, oste- ital syphilis. The incidence of Hutchinson's incisors

In the past, mercury was used to treat congenital

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42

those caused by the disease. Hutchinson recognized at 26 years of age. Occlusal and oblique X-rays of the that mercury affected amelogenesis resulting in se- mandible were taken using a Frankenstein unit to see vere enamel hypoplasia (Hutchinson, 1878). Treat- whether a fourth molar was present on the right side. ments containing mercury were given to infants soon Chemical analysis was performed to detect any levels after birth, the time which enamel formation in per- of mercury. A Bruker Tracer III-V handheld analyser manent teeth begins. First permanent molars and was used on hypoplastic portions of the central and incisors begin their formation around birth and this is lateral incisors. The initial analysis used an allwhen they are exposed to disease. Mercury used to elements setting. The settings for the following test treat syphilitic infants continued for months after were elevated to (0.001" Cu, 0.001" Ti, 0.012" Al filter birth, severely affecting other tooth formation, de- at 40 keV/16 micro amps for 300 seconds, without pending on the length of time the treatment was ad- vacuum) (Ioannou et al., In press). ministered (Hutchinson, 1878). The abnormalities produced by congenital syphilis can be combined with the effects of treatment containing mercury (severe hypoplastic effects) (Hutchinson, 1878; Moon, central and lateral incisors, canines, premolars and all 1884). Treatment with mercury was commonly used in cases of congenital syphilis until the early 20th century. The whole suite of changes caused by congenital syphilis and treatments containing mercury have been discussed in detail (Ioannou et al., 2016).

Vieira, 2008; Millhon and Stafne, 1941; Panjwani et alcanti et al., 2011; Harris and Clark, 2008). The morphology of supernumerary teeth can vary in each surface. individual from normal in shape and size, normal normal in shape and reduced in size (Harris and Clark, 2008; Kumar and Gopal, 2013; Rahnama et al., 2014).

This paper presents a case of congenital syphilis in an African American woman dating from the early 20th century with a fourth mandibular molar. A focus will be made on the development of the fourth molar in the presence of a disease, which primarily affects dental development.

MATERIALS AND METHODS

During a systematic examination of 28 skeletons held at the Smithsonian museum in Washington, DC, whose documentation stated that they had "treponemal or treponemal congenital" disease, a case of a supernumerary mandibular distomolar in one individual (P000707) was revealed. This individual was an African American female, who was born in 1903 and died of pulmonary tuberculosis in 1929,

RESULTS

All maxillary permanent teeth were present, the three molars. The enamel of the central incisors from the incisal third to the middle third of the crown appears mottled and thin (Figure 1). The incisal third of the lateral incisors and left canine demonstrate the same mottled appearance and pitted enamel hypo-Supernumerary teeth have been associated with plasia. Deep pits are apparent toward the middle various syndromes and disorders including Down's third of the crown of the central incisors and incisal and Gardner's, cleidocranial dysostosis, and cleft lip third of the lateral incisors and canines. In addition to and palate (Kumar and Gopal, 2013; Menezes and signs caused by mercury on the incisors and canines, other teeth display isolated hypoplastic pits. Maxilal., 2011; Sandler, 1951); however, they have not been lary premolars are not affected. First permanent modescribed in detail in cases of congenital syphilis. lars have abnormal occlusal surfaces, with cusps re-Supernumerary teeth are observed when more than duced in size and pitting hypoplasia, which is also 20 deciduous or 32 permanent teeth are present in an consistent with the side effects of mercury (Figure 2). individual. They can erupt, remain unerupted, or Diseased enamel is clearly demarcated from the become impacted (Kara et al., 2012; Mali et al., 2012). healthy enamel on the cervical third of the crown. Their appearance can be unilateral, bilateral, as a sin- The morphology of the second and third permanent gle tooth or in multiples (Brinkmann et al., 2012; Cav- maxillary molars is normal with normal groove patterns, but there is some enamel pitting on the occlusal

Mandibular permanent teeth include the central shape and reduced in size, conical in shape and ab- and lateral incisors, left and right canines, first and second premolars, second and third molars and the



Figure 1. The maxillary central and lateral incisors and left canine display hypoplastic enamel seen in patients with congenital syphilis treated with mercury. Signs include thin enamel, pitted enamel hypoplasia (in some places very deep), and distinct demarcation separating diseased from healthy enamel.



Figure 2. Occlusal view of the maxilla. First permanent molars have abnormal surfaces with small cusps and pitting hypoplasia

distomolar. The first permanent molars were lost ante-mortem, possibly by extraction and their alveoli are completely healed. All mandibular incisors have mottled enamel (Figure 3). The left and right second molars and the third left molar do not display severe hypoplasia, save for minor pitting. Their occlusal surfaces are crenulated. The third permanent molar on the right side is represented by its roots only. The crown has broken off probably after its destruction by dental caries. On the left side, in the mandible, there is a fully erupted fourth molar (distomolar). Its crown has normal molar morphology, but is smaller in size in comparison to the other permanent molars present. The arrangement of groves resembles the +4 pattern. However, the groove pattern is complex because of crenulation. Entoconid, metaconid, hypoconid and protoconid are present, and it appears that there may be a narrow metaconulid, but crenulations make it difficult to determine (Figure 4). An oblique

X-ray of the mandible shows that the distomolar only has one root with a large pulp chamber extending far down towards its apex, suggesting a taurodont condition (Figure 5). The third molar on the left is large and crowded between the distomolar and adjacent second molar. Its crown is rotated approximately 10 degrees and tilted mesially. Inspection of the X-ray



Figure 3. The anterior view of the mandibular incisors displaying enamel defects

does not reveal the presence of the antimeric distomolar (Figure 6). All molar crowns appear crenulated.



Figure 4. Occlusal surface of the mandible. The first permanent molars were lost ante-mortem. Both second molars, the left third molar and left fourth molar are present. The right third molar is represented by its root only. The fourth molar displays normal molar morphology

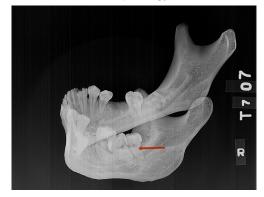


Figure 5. Oblique X-ray image of the mandible shows that the distomolar has only one root and that there is no antimeric distomolar. Note the large extent of the pulp cavity in the distomolar, suggesting it is a taurodont molar.



Figure 6. X-ray of the occlusal view of the mandible does not show any evidence of a right fourth molar

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teal reaction were observed on the long bones includfemora, as well as the lateral surface of the left ilium. than the central incisors. The left femur had lytic destruction along the lateral striated periosteal reaction is not noticeable.

DISCUSSION

a supernumerary distomolar in an African American woman. Although this condition is very rare during this time, it is probable, as one other case has been documented (Jacobi et al., 1992). However, in this case, the dental abnormalities in P000707 indicate that she was treated with mercury soon after birth. dental abnormalities characteristic of the disease. Changes in the morphology of the central maxillary incisors and left canine have enamel malformations that are compatible with dental abnormalities observed by Hutchinson in patients with congenital syphilis administered treatment containing mercury (Figure 7). Crown formation of the central permanent incisors begins at approximately three to four months postnatally and is complete at approximately 4 to 5 years of age (Nelson and Ash Jr, 2010). The specific changes in enamel caused by mercury are seen in one third of the crown, therefore, treatment would have started in the middle of the first year of life and

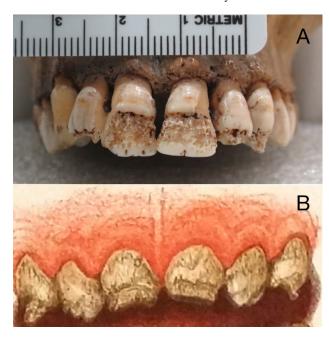


Figure 7. (A) Anterior teeth of P000707 (B) Patient treated with mercury as presented by Hutchinson in 1878 (16 year old boy). Both (A) and (B) display similarities in enamel abnormalities that occur as a result of treatments containing mercury. Mercury would have been administered at a somewhat older age in P000707 than in Hutchinson's patient. Hutchinson, (1878) p. 53, Plate VI, Items I (A)

post cranial skeleton, limited areas of nodular perios- ceased at approximately 2 years of age. Similarly, severe enamel malformations are observed on the ing the right tibia, fibula, humeri, radius, ulnae, and lateral incisors and canines that start forming later

The morphology of the maxillary first permanent border of the head in the anterior aspect, "classic" molars demonstrates a normal groove pattern towards the mesial end of the crown, while the rest of crowns' occlusal surfaces are reduced in size and hypoplastic. As the incisal third of the central incisors Here we present a case of congenital syphilis with and a portion of the occlusal surface of the first permanent molars appears to be normal, the rest of the crown is affected, which may be an indication that the onset of the infection was late in relation to tooth development.

> Congenital syphilis is known to produce specific However, it has been noted that in some cases of congenital syphilis, the classic dental changes that are usually observed such as Hutchinson incisors, Moon's molar and Fournier's molars do not occur (Švejda, 1952). Hutchinson also observed and described certain dental abnormalities that occurred as an effect of treatments containing mercury (Hutchinson, 1878). The dental abnormalities produced by the disease itself and treatments containing mercury were so distinct that Hutchinson deemed it worthy to document and illustrate both as separate entities. It is worth noting that the crescentic notch that occurs in the maxillary central incisors of congenital syphilis patients is not observable if they were treated with mercury (Hutchinson, 1878). The features observed in this P000707 are typical signs of teeth treated with mercury in patients with congenital syphilis (Hutchinson, 1878; Ioannou et al., 2016).

> While the results of the chemical analysis detected no levels of mercury, this neither confirms nor disproves that mercury was administered to this individual. Various explanations could be considered. It is possible that the low levels of mercury in the enamel could not be detected by the equipment. Another possible explanation for the lack of mercury detected could be due to the quick turnover rate of mercury in the body. The half-life of mercury ranges from 58 days for elemental mercury, 1-2 months for mercuric mercury (e.g. HgCl₂), to 70-80 days for methylmercury (National Research Council (US) 2000). Taking into account that this individual was treated with mercury for congenital syphilis in the early stages of life and died at 26 years of age, it is not abnormal to find extremely low levels of mercury. As indicated by Hutchinson, if 648 mg (10 grains) of mercury were introduced in a body of a young individual, after 20 years only a minute quantity of mercury would remain (2.13*10-25mg). Thus, it is more likely that a majority of the mercury would be cleared out, making it undetectable.

changes or malformations in enamel development. In relation to changes on the post cranial skeleton of cal changes could be due to treponemal infection.

mal, unaffected by the disease, nor by treatments containing mercury. The smaller size of the distomoand becomes the tertiary stage after several weeks. syphilis. Tertiary syphilis does not affect tooth development. The development of the third permanent molar begins at approximately 7 to 10 years of age and the tooth is fully erupted between the ages of 17 and early 20s (Liversidge, 2015). It is possible that the fourth distomolar could have developed at the same age or even later. If the fourth molar had developed soon after the third molar, P000707 would have been in the could be asymptomatic and would not have affected congenital syphilis. amelogenesis or odontogenesis of the supernumerary fourth molar. However, it is possible that the fourth molar developed sooner. Studies have shown that The authors thank Dr. David Hunt for taking the Xfourth molars can appear between the ages of 11 and 2000; Orhana et al., 2006; Vlaykov et al., 2015).

It also appears common that distomolars demonstrate a single root, unlike the multiple roots observed in the other permanent molars (Ceperuelo et Alvesalo L, Varrela J. 1991. Taurodontism and the al., 2015; Ohata et al., 2013; Rahnama et al., 2014). However, root formation can vary among individuals (complete with closed apex or incomplete) (Ceperuelo et al., 2015; Kokten et al., 2003; Ohata et Andersson E-M, Axelsson S, Gjolstad L-F, Storhaug al., 2013). Since the distomolar in this case is taurodontic, it is not possible to determine whether it had

Other elements considered in the differential diag- fused multiple roots or a single root because no sepanosis include lead, zinc, copper and cadmium. High rate root canals can be seen. At least formally, the levels of lead can cause a decrease in microhardness root is a single unit. The cause of taurodontism is unof enamel (Gerlach et al, 2002) but cannot cause mal- clear. It has been associated with various syndromes formations of the enamel (Gerlach et al, 2002; (Andersson et al., 2013; Keeler, 1973; Rajić and Mes-Youravong et al, 2005). Fosse and Berg-Justesen trović, 1998) and multiple theories have been sug-(1977, 1978, 1978) and Tvinnereim et al. (1999) exam-gested in the literature (Alvesalo and Varrela, 1991; ined concentrations of zinc, copper, and cadmium in Witkop Jr et al., 1988). In this case, it should be conteeth and bone in humans and mice and recorded the sidered that the proportions of the root to crown size difference in concentration of these elements between and pulp cavity to root canal volumes may have deenamel, dentin, and bone, but did not record any veloped abnormally in the supernumerary, thus not normal, tooth without any special causes.

The development of extra teeth is not fully under-P000707, since the individual died of tuberculosis, it stood, although multiple theories have been suggestis difficult to say which of those described pathologi- ed such as hyperactivity within the dental lamina, and dichotomy of the tooth germ and these may be The crown morphology of the distomolar is nor- linked to genetic factors (Kokten et al., 2003; Kumar and Gopal, 2013). For instance, Martínez-González et al. (2012) found them in 0.96%, Shahzad and Roth lar is unlikely to be caused by congenital syphilis. (2012) in 2.2% and Kara et al. (2012) in 0.33%. It has Clinical studies have shown that distomolars can been found that supernumerary molars were also demonstrate normal molar morphology, have as more prevalent in African Americans (6.4%), than in many as three to seven cusps and be reduced in size, European Americans (0.9%) (Shahzad and Roth, in comparison to the other permanent molars (Asrani 2012). It has been suggested that African Americans et al., 2006; Ceperuelo et al., 2015; Kumar and Gopal, exhibit extra teeth more often than European Ameri-2013; Ohata et al., 2013; Shahzad and Roth, 2012). The cans (Harris and Clark, 2008), which may be related normal crown morphology in this case may be due to to African Americans having larger dental arches and the time at which the development of the fourth mo- greater crown and root dimensions. This would inlar began. It is the early stage of the disease that af- crease a probability of the appearance of the distomofects dental development. It occurs soon after birth lar in an African American suffering from congenital

CONCLUSION

A systemic infection such as congenital syphilis and its treatment with mercury may not influence the development of supernumerary teeth due to: (1) the age at which the development of the fourth molar takes place, (2) the stage of the infection at the time of development and (3) the age at which treatments tertiary stage of the disease; therefore, the disease containing mercury are administered to patients with

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