Archives of Oral Biology 60 (2015) 1554–1564

Contents lists available at ScienceDirect

Archives of Oral Biology

journal homepage: www.elsevier.com/locate/aob

Dental stigmata and enamel thickness in a probable case of congenital syphilis from XVI century Croatia

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ARTICLE INFO

Article history:
Received 4 February 2015
Received in revised form 3 June 2015
Accepted 3 July 2015

Keywords:
Enamel defects
Fournier canine
Hypoplasia
Mulberry molars
Micro-CT

ABSTRACT

Objective: To analyse the dental remains of an individual with signs of congenital syphilis by using macroscopic observation, CBCT and micro-CT images, and the analysis of the enamel thickness.
Design: Anthropological analysis of human skeletal remains from the 16th century archaeological site Park Grič in Zagreb, Croatia discovered a female, 17–20 years old at the time of death, with dental signs supportive of congenital syphilis: mulberry molars and canine defects, as well as non-specific hypoplastic changes on incisors. The focus of the analysis was on three aspects: gross morphology, hypoplastic defects of the molars, canines and incisors, as well as enamel thickness of the upper first and second molars.
Results: The observed morphology of the first molars corresponds to the typical aspect of mulberry molars, while that of the canines is characterised by hypomineralisation. Hypoplastic grooves were observed on the incisal edges of all incisors. The enamel of the first molars is underdeveloped while in the second molars a thick-enamelled condition is observed.
Conclusions: Our observations for the dental and skeletal evidence are supportive to a diagnosis of congenital syphilis for this specimen from XVI century Croatia. The use of CT imaging helped documenting the diagnostic features and quantifying the effect of the dental stigmata on first molars.

1. Introduction

Syphilis is a chronic disease that belongs to the group of treponematoses caused by the spirochetal bacterium *Treponema pallidum*. Based on mode of transmission it can be divided into two types: acquired, transmitted through sexual contact, and congenital, transmitted transplacentally to the developing foetus by an infected mother (Ortner, 2003)

Dental anomalies in patients with congenital syphilis have long been the subject of research (Hutchinson, 1857; Hutchinson, 1858; Fournier, 1884; Bauer, 1931; Sarnat & Shaw, 1942; Bauer, 1944; Putkonen & Paatero, 1961; Bernfeld, 1971), but in general, affected dental specimens are rare findings in archaeological contexts (Hillson, Grigson, & Bond, 1998; Gaul & Grossschmidt, 2014).

Dental signs of congenital syphilis include defects of permanent teeth or dental stigmata: Hutchinson's incisors, Fournier's canines, Moon's molars, mulberry molars (Bauer, 1944). All of them have their origin in the process of amelogenesis. Although amelogenesis is genetically controlled, it is very sensitive to different environmental disturbances (Souza et al., 2013). Treponemal infection of enamel organs damages or destroys the ameloblasts and disrupts the formation of tooth germs (Bauer, 1944). Burket (1937) considered also metaplasia and aplasia of the ameloblasts as possible conditions caused by the treponematosis. Inflammation around the tooth germ can alter enamel formation in two main ways (Bauer, 1944; Hillson et al., 1998): (a) as minor or moderate hypoplastic defects and (b) as hypoplastic disturbance with morphological distortions. The former can occur in many teeth and in any period of enamel formation resulting in non-specific hypoplastic defects, while the latter can occur due to stronger pressure on the early tooth bud and during tooth morpho-differentiation resulting in the alteration of its shape. The

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http://dx.doi.org/10.1016/j.archoralbio.2015.07.002
0003-9969/

inflammation around the tooth germ caused by congenital syphilis decreases with time, so the hypoplastic abnormalities affect only specific teeth at characteristic locations.

Hypoplastic changes caused by congenital syphilis can be divided into three main groups. The first group includes hypoplastic changes with clear and well-defined morphological features that are specific to congenital syphilis. These are Hutchinson's incisors and Moon's molars and they should be considered pathognomonic of this disease (Hutchinson, 1857; Hutchinson, 1858; Fournier, 1884; Hillson et al., 1998; Moon, 1877; Pflüger, 1924; Molnar, 1971). The inflammation due to *T. pallidum* during the morphological differentiation of the permanent teeth causes a disturbance in the formation of the enamel–dentine junction (Sarnat & Schour, 1941), which results in a typical appearance of the affected teeth. However, Hutchinson's incisors and Moon's molars occur only in a low percentage of subjects affected by congenital syphilis (Putkonen & Paatero, 1961) and with various degree of expression (Fournier, 1884).

The second group of hypoplasy includes Fournier's canines and mulberry molars that occur mostly, but not uniquely in congenital syphilis (Fournier, 1884; Jacobi, Cook, Corruccini, & Handler, 1992; Hillson et al., 1998). Therefore, Fournier's canines and mulberry molars are not defined as pathognomonic to congenital syphilis even though highly suggestive of this condition (Gaul & Grossschmidt, 2014).

These two groups of dental defects present a distinct morphology and are exclusively or mostly found in association to congenital syphilis. Therefore, Hutchinson's incisors, Fournier's canines, Moon's molars and mulberry molars are widely known as dental stigmata (Condon, Becker, Condon, & Hoffman, 1994; Hillson et al., 1998). Importantly, the aforementioned dental stigmata are typical of permanent teeth, and manifestation on deciduous teeth were observed albeit more rarely than in permanent dentition (Fournier, 1884; Steinbock, 1976; Hutchinson & Richman, 2006). In such cases, circular grooves around the root and just below the crown of the second deciduous molars have been typically detected (Karnosh, 1926). Fournier (Fournier, 1884) mentioned also the possible occurrence of notched deciduous incisors.

Finally, the third group of hypoplastic defects collectively designated as general enamel hypoplasia can be caused by syphilis as well as other environmental factors (Curtin, 2005). They are widely used as non-specific health indicators reflecting general health, diet, and, more broadly, living conditions. While in congenital syphilis hypoplastic changes appear on the structures that calcify up to the first year of life, the disruptions causing general enamel hypoplasia often affect dental formation for a limited period of time (Putkonen, 1962, 1963). Hypoplastic defects may appear as linear grooves, depressions (especially labially) or pits visible on the outer surface. The enamel is hard but deficient in amount. Among the hypoplastic manifestations darker and greyish colour of the enamel should be taken into account. The presence of systemic disorders such as the anhidrotic form of hereditary ectodermal dysplasia, dental dysplasia, linear enamel hypoplasia and molar-incisor hypomineralization (MIH) might lead to a wrong diagnosis of congenital syphilis. However, if single manifestations may be misleading, the observations carried out on more teeth and the estimation of the time of appearance of the disturbance can be considered altogether for a correct diagnosis. Disturbances in amelogenesis due to congenital syphilis mostly appear around birth and decrease throughout time. This locates enamel defects on the first molars and permanent incisors whose crown mineralization starts around birth. Time and location specificity distinguishes hypoplastic defects in congenital syphilis from most other environmental disturbances that appear throughout a longer period of life.

Congenital syphilis in past populations is difficult to confidently diagnose due to several reasons. This is firstly affected by the 50% fatality rate of infected foetuses (Aufderheide, Rodríguez-Martín, 2011; Roberts & Manchester, 2005). Additionally, degree of skeletal and dental preservation, age at death, and recognition of specific pathological changes influence the diagnosis (Roberts & Manchester, 2005). Bone deformities and dental stigmata are currently the main way in which congenital syphilis can be diagnosed in archaeological human remains. Teeth are usually better preserved than bones therefore dental changes are more often used for diagnosis.

Frequency of congenital syphilis in archaeological populations is hard to estimate owing to several reasons. The first are high rates of foetal death caused by the infection. Secondly, infected children may not display any sign of the disease, especially on the skeleton. Thirdly, even if symptoms occur, dental stigmata are observed in only 30% of individuals with congenital syphilis (Aufderheide & Rodríguez-Martín, 2011; Larsen, 1997) Cook and Powell (2005) reported only 5.1% frequency of congenital syphilis in juveniles. The authors stated that the lesions used to identify the disease were less specific than those for adults. Rothschild and Rothschild (1997) concluded that only 5% of syphilitic children showed evidence of dental or osseous involvement. This rate is explained by quick remodelling of new bone that leaves no visible trace on the skeleton. In a study by Putkonen (1962), Hutchinson's incisors and Moon's molars were present, in 45% and 22%, respectively of children born with syphilis, compared to only 12% that showed skeletal evidence of the disease. However, during the life span, teeth can be lost owing to dental and periodontal diseases, and in any case, the dental stigmata can be obliterated by wear and attrition, which in hypoplastic teeth would occur even at a higher rate. Although they possess very specific morphological characteristics, syphilitic teeth are quite sporadic findings in archaeological samples. However, the analysis of human skeletal remains from the 16th century archaeological site Park Grič in Zagreb, Croatia revealed the presence of five individuals with skeletal changes indicative of syphilis (Mašić & Pantlik, 2008a,b). Additionally, individual from grave 55, thereafter PG55, had dental signs supportive of congenital syphilis (Vodanović, Lauc, Premužić, & Rajić Šikanjić, 2013; Lauc, Rajić Šikanjić, Premužić, Fornai, Mašić, & Vodanović, 2014).

The first aim of this paper is to provide a detailed description of PG55 dental remains and discuss the gross morphology and appearance of the affected teeth taking into account their outer enamel surface. Although it is known that the disturbance of the ameloblasts during the secretory phase may cause a reduction in enamel thickness (Souza et al., 2013) the enamel hypoplasia typical of teeth affected by congenital syphilis has been described in the literature only qualitatively, while no quantitative analyses have been performed up to now on syphilitic dental specimens. Also, little is known about enamel thickness and morphology of the dentine crown in symptomatic teeth of individuals affected by congenital syphilis. Therefore, we aim to investigate the 2D and 3D relative enamel thickness and dental tissue proportions in PG55 affected and unaffected maxillary molars in comparison to non-pathological molar teeth from recent modern humans.

2. Materials and methods

PG55 was a female aged 17–20 years at death based on morphological characteristics of the skeleton and dentition (Buikstra & Ubelaker, 1994; Scheuer & Black, 2004). The skeleton was very well preserved, with only some of the small bones and teeth missing owing to post-mortem damage (Fig. 1, right individual). Apart from the dental changes, there were additional lesions visible on the skeleton. Significant endocranial lesions were