

Congenital syphilis in the skeleton of a child from Poland (Radom, 18th–19th century AD)

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ABSTRACT: An incomplete skeleton of a 3-year-old child with suspected congenital syphilis was found in the Radom area of Poland. Squama frontalis and zygomatic bones are characterized by significant bone loss. Radiographic pictures show a geographic destructive lesion of a serpiginous shape surrounded by a zone of reactive osteosclerosis in the squama frontalis. The radiographic findings included a slight widening and contour irregularities of the distal humeral metaphyses. The appearance of teeth did not suggest Hutchinson teeth, but the examination of the permanent molars showed signs of mulberry molars. Two teeth were tested for the presence of mercury. Chemical analysis did not indicate mercury accumulation (enamel: 0.07 $\mu\text{g/g}$, dentine: 0.14 $\mu\text{g/g}$, bone: 0.11 $\mu\text{g/g}$). Mercury values obtained for the examined samples were similar to those that are typical of healthy teeth in today's individuals.

KEY WORDS: paleopathology, congenital syphilis, Poland

Introduction

Although syphilitic symptoms are well described in the clinical literature, in terms of paleopathological research, there are relatively few unequivocally diagnosed cases of this disease, especially in very young individuals (Jacobi et al. 1992; Mitchell 2003; Erdal 2006; von Hunnis et al. 2006; Harper et al. 2011;

Nystrom 2011). This fact is connected with diagnostic doubts since other diseases (e.g., tuberculosis, Paget's disease or osteomyelitis) can present a similar macroscopic picture. In addition, the fragmentary nature of archaeological material limits diagnostic credibility (Jacobi et al. 1992; Rothschild and Rothschild 1997; von Hunnis et al. 2006; Ortner 2008; Marden and Ortner 2011; Gaul

and Grossschmidt 2014). In the case of children, the most observable defects attributed solely to venereal syphilis are the metaphyseal destruction of the long bones and/or malformations of the teeth. However, the literature most often describes cases of children or juvenile individuals with already erupted permanent teeth, showing possible signs of Hutchinson, Moon teeth, and increased enamel hypoplasia as well as dental caries (Hillson et al. 1998; Jacobi et al. 1992; Henneberg and Henneberg 1994; Erdal 2006; Hutchinson and Richman 2006; Harper et al. 2011). Assessment of possible syphilitic changes in deciduous dentition is far more difficult and defects are considerably less common in such teeth. One of the most often quoted cases is the 6/7-year-old child from Virginia (NMNH 379177), in whom unusually intensive hypoplastic defects on all anterior deciduous teeth of the maxilla and mandible were observed. However, the visible left tooth germ of the medial incisor did not have features typical of Hutchinson teeth. Another case of congenital syphilis was seen in a three-year-old child from the Fisher site, Virginia (NMNH 385786). Here, too, many of the deciduous teeth showed very advanced hypoplasia, however, there were no morphological changes in the tooth germs of permanent incisors and molars (Ortner 2003). The first probable case of congenital syphilis from Central Europe (Austria) was published by Gaul and Grossschmidt (2014). The grave (FH-206) belongs to 6-year-old child. However, the material was exclusively represented by dental remains.

Interesting is the problem of diverse frequency of damaged teeth (deciduous or permanent). In the 19th century, Jonathan Hutchinson noted that tooth deformation in individuals suffering

from syphilis could be caused not by the disease itself but rather by mercury therapy administered until the 1940s, when it was replaced by penicillin (Putkonen 1963; Powell and Cook 2005; Rassmusen et al. 2008). Mercury was used mainly in the Middle Ages and in the early modern period as a preventive measure in case of individuals suffering from a venereal disease and leprosy (Dracobly 2004; Rassmusen et al. 2008). We know that this treatment led to the atrophy of the kidneys, loss of teeth and hair, and finally resulted in the destruction of the brain (Dracobly 2004). In view of the above information we could test if the teeth of individual from Radom were damaged due to mercury therapy.

The aim of our study was not only to present the case of a 3-year-old child with possible congenital syphilis, but also pay special attention to examine the possible presence of mercury in the teeth and bone of this individual.

Material and Methods

In 2010, an incomplete skeleton of a child (No. 15) was found in Radom – Piotrkówka (Poland) (Fig. 1). The areas situated in the valley of the river Mleczna have their own long and diverse history. The earliest traces of human presence in this area are linked with communities of the early Middle Ages, further traces refer to the functioning production settlement of the 14th–15th centuries, and, later, to the parish cemetery from 1790–1812. The cemetery was located outside the town itself. Such a location was chosen, first of all, due to sanitary considerations, which became the main reason for locating cemeteries outside a densely built-up urban area starting from the 18th century. It was a significant change which put an end to



Fig. 1. Location map of the study area

the common custom of burying the dead near churches or in church vaults that were not very far from a densely built-up urban area (Piątkowski 2000; Sekulski and Stan 2009; Buko et al. 2010).

The skeleton was excavated at the parish cemetery, which dates to the period 1790–1812. The child's dental age was determined with the help of the method of tooth formation stages to be about 3 years (Hillson 2002; Liversidge et al. 2010). Surviving skeletal material was comprised of the cranium and mandible, clavicles, ribs, cervicothoracic spine, scapulae, humeri, left ulna, some phalanges and the left ilium. No lower limbs were found in the material as they had probably been destroyed during the exploration process. In the bones of the jaw on both sides there were erupted deciduous molar teeth (numeration according to FDI: 84, 85, 74, 75), a left canine (73) and both incisors (72, 71). Moreover, the presence of all permanent incisor germs (11, 12, 21, 22) and canine germs (13, 23) was found. In the arch of the mandible there were the first and second right molars (54, 55), as well as

the germs of both first molars (36, 46), both canine teeth (43, 33) and incisors (32, 31, 41, 42).

The macroscopic tests of the skeleton were carried out in the Laboratory of Biological Anthropology of the Cardinal Stefan Wyszyński University in Warsaw. Visual inspection was performed under direct surgical light. Microscopic examination of the lesions was performed using a Nikon SM 1500 microscope, TTL 2.5× dental magnifying glass and a sharp dental probe. Targeted X-ray photos were taken with a GE Medical System machine at 46 kV and 4 mA. All of the images were made in the anterior-posterior plane. Radiological studies were conducted at the Private Clinic in Siedlce (Poland) and Medical University of Warsaw (1st Department of Clinical Radiology). The choice of two independent research units gives the possibility to compare the obtained results, in order to eliminate a possible diagnostic error.

Chemical analyses were performed by the Institute of Anthropology of the Jagiellonian University (Cracow). A permanent medial incisor germ (11), a deciduous molar (84) tooth, and a fragment of finger bone were used in the tests for the presence of mercury (Hg). Preparation and analysis of tooth samples was performed in accordance with well-established procedures (Arany et al. 2004; Castro et al. 2010; Keça et al. 2012). The prepared samples were washed in deionised water obtained from Millipore Water Purification System (GEN-PURE System), in an ultrasound washer, which made it possible to remove impurities from the micro-cracks in the enamel. After that the mesial surface of the sectioned teeth was polished using a glass plate and deionised water. The samples prepared in this way were then dried

in a laboratory incubator at 60°C for 24 hours. Hg marking was done by means of the LA-ICP-MS laser ablation technique using an Nd laser: YAG, Macro, 266nm, NewWave (US) coupled with an Elan DRC-e Perkin Elmer spectrometer (US). Before the tests the measuring equipment was calibrated in accordance with the calcium phosphate controls (Standard Reference Materials 120c, Florida Phosphate Rock) soaked in mercury, from which a tablet was prepared using a laboratory press. Additionally, the reference material SRM NIST 612, commonly used for elemental marking along with the laser ablation technique, was used as a control. In order to obtain even more precise marking, the calcium isotope (^{43}Ca) was used as an internal pattern. The samples prepared for marking were placed in the laser chamber in such a way that the polished surface was parallel to the laser beam. Separate marking was used for tooth enamel and dentine.

Results

The squama frontalis exhibits a large area of bone loss (8.0×2.5 cm). The

floor of the lesion was perforated and the margins were rolled inwards toward the center of the lesion. An early-stage porous caries sicca lesion was observed above the glabella. An additional defect (1.0×0.4 cm) was detected on the side of the right orbit in the zygomatic region. Also, an abnormal periosteal bone reaction was noted on both parietal squamas in the region of the occipital margin as well as on the occipital squama in the region of the external occipital protuberance. Cortical bone suggested healing of a pathological process. Radiographic pictures show a geographic destructive lesion of a serpiginous shape surrounded by a zone of reactive osteosclerosis in the squama frontalis (Fig. 2).

Both humeri contained discrete periosteal bone reactions, which were limited to the lower parts of the diaphyses. However, both distal metaphyses of the humeral bone showed characteristic thickening of the cortical layer, which extended to the cancellous layer. The radiographic findings included a slight widening and contour irregularities of the distal humeral metaphyses, more pronounced on the left side, where a de-

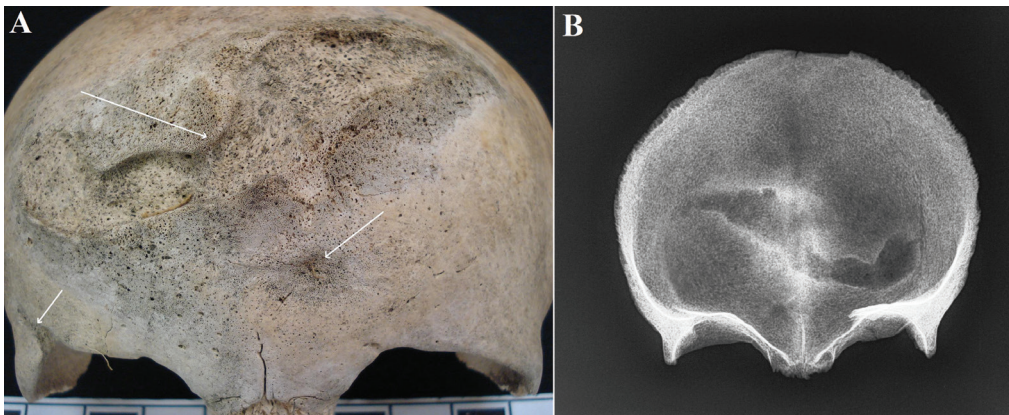


Fig. 2. Frontal squama with marked destructive lesions (A), and X-ray of the frontal squama (B)

structive lesion was present in the lateral aspect of the metaphysis accompanied by a subtle calcified linear periosteal reaction. There were no radiolucent bands in the metaphyseal areas. These findings were suggestive of a healed osteochondritis (Fig. 3).

The diaphysis of the left ulna was clearly enlarged, especially in the lower segment. In addition, there was extensive proliferative new bone formation and thickening of the compact layer. Radiological analysis confirmed the macroscopic observations. There is a geographic destructive lesion in the diaphysis of the left ulna accompanied by an exuberant lamellated (“onion skin”) periosteal reaction. These lesions were consistent with a chronic osteomyelitis (Fig. 4).

On all of the deciduous teeth both enamel hypoplasia and dental caries were observed, most often located in

atypical places (e.g. on the labial surfaces at the height of 1/3 of crowns or on tops of cusps). The pit-type hypoplasia primarily involved occlusal surfaces (tops of cusps and grooves) and smooth surfaces of crowns (Fig. 5). A delicate alveolar exposure of crown germs of the first permanent incisors of the maxilla revealed pitted hypoplasia on the labial surface below the incisal edge, close to the top of the three mamelons. Moreover, numerous opaque enamel spots were seen on the labial surfaces. However, the appearance of germs was not consistent with Hutchinson teeth (Fig. 6). Examination of the first lower permanent molar germs revealed signs of mulberry molars. There are topographic abnormalities of the occlusal surface, first of all, on the mesio- and the distolingual cusps. There are additional clear prominences in the grooves. It is a marked defect involving



Fig. 3. X-ray of the distal humeral metaphyses. Slight widening and contour irregularities. (arrows); subtle calcified linear periosteal reaction (arrowhead)



Fig. 4. Left ulna with an exuberant lamellated periosteal reaction: macroscopic (A) and X-ray picture (B)

enamel hypoplasia running around the base of all cusps (Fig. 7). There is no evidence of the changes typical for Moon's molars, where cusps resemble "islets" coated with an uneven layer of enamel and abnormally close spacing of cusps.

Since mercury was administered as a treatment for syphilis at the beginning of the 20th century (Quétel 1990; Powell and Cook 2005; Rasmussen et al. 2008),

we decided to check whether the child from Radom or its mother had contact with this substance. Therefore, the deciduous molar (84), the permanent incisor germ (11), and a fragment of finger bone were used to determine the time of mercury treatment at different ontogenetic moments of the child's life. In all samples, mercury was at a low level (Table 1). Values of mercury concentration



Fig. 5. Deciduous upper canine with hypoplastic defects on the labial surface

obtained for the examined samples were similar to those typical for healthy teeth of today's individuals. The obtained results were compared with the material from Pień (14th–17th cent.), Strzelno (15th–16th

cent.), Płonkowo (16th–19th cent.), and Gniew (18th–19th cent.), where syphilis was also diagnosed, as well as with the teeth of healthy individuals from present times (Kepea et al. 2012).

Table 1. Mercury content (results in $\mu\text{g/g}$) in samples from Radom and other historical groups

	Radom* (I1)	Radom* (m1)	Pień* (M2)	Płonkowo* (M1)	Strzelno*	Gniew*	Modern individual 1	Modern individual 1
enamel	0.07	0.07	0.71	1.32	–	–	0.09	0.03
dentin	0.10	0.14	0.49	0.44	–	–	0.09	0.05
bone		0.11	–	2.30	2.66	0.93	–	–

*individuals with syphilitic lesions



Fig. 6. Permanent upper medial incisor with hypoplastic defects on the labial surface

Discussion

The osseous changes in the course of congenital syphilis may be divided into early and late (Teberg and Hodgman

1973; Rothschild and Rothschild 1997; Aufderheide and Rodríguez-Martín 2008; Harper et al. 2011). The spirochetes, after penetrating into the blood-



Fig. 7. Lower mulberry molars with marked hypoplastic areas

stream of the fetus, migrate to the perichondrium, periosteum, cartilage, and bone marrow. The sites of active endochondral ossification constitute a preferential target of invasion. The spirochetes cause the degeneration of osteoblasts, thus disturbing osteogenesis. Therefore, osteochondritis is – after periostitis – the second-most common presentation of skeletal involvement in congenital syphilis (Rasool and Govender 1989; Russo and Shryock 1945; Jacobi et al. 1992; Ortner 2003). Typically, symmetric involvement of sites of enchondral calcification may be observed in the epiphyseal-metaphyseal junction of tubular bones and costochondral regions. Provisional calcification zones are widened and metaphyseal contours are irregular and serrated. Within metaphyses, broad radiolucent bands may be observed. In more severe cases, destructive lesions caused by granulation tissue appear. The most typical site of erosion is the medial side of the proximal tibia (Wimberger's sign). Destructive lesions may progress into diaphyseal osteomyelitis. The epiphyseal-metaphyseal separation is another possible complication. Periostitis may be both reactive and infective (Aufderheide and Rodríguez-Martín 2008; Ortner 2003). The former accompanies the lesions of osteochondritis and osteomyelitis. The latter presents as a diffuse thickening of the periosteum, and is most visible along the shafts of the long tubular bones. The early manifestations of congenital syphilis generally regress in the first years of life, even in untreated individuals. Late osseous changes are related to the reactivation of spirochetes in young children and adolescents, and resemble those observed in acquired syphilis. The typical locations of osteomyelitis and periostitis in late congenital

syphilis are the tubular bones, particularly the upper two-thirds of the tibial shafts, the flat bones, and the cranium. Involvement of the skull is particularly characteristic with the frontal, parietal, and nasopalatine areas affected most commonly (Bauer and Caravati 1967; Dismukes et al. 1976; Jacobi et al. 1992; Mitchell 2003; von Hunnis et al. 2006). At first, lytic lesions display a moth-eaten or permeative pattern and later progress towards geographic lesions. The radiographic picture of diaphyseal osteomyelitis in the long bones does not differ from changes found in acquired syphilis. The involvement of the proximal tibia with reactive osteosclerosis and anterior bowing (“saber-like tibia”) is virtually pathognomonic. The findings observed in our specimen require differential diagnosis with other bacterial osteomyelitis and tuberculosis. Diaphyseal involvement in tuberculosis in children is possible (spina ventosa), although rare. The preferential sites of involvement in tuberculosis are the joints and the spine, which was completely normal in our specimen. Moreover, the biphasic character of lesions (healed osteochondritis and active osteomyelitis) observed in our specimen strongly points to syphilis. The other infection that was common at the time is smallpox. Variola osteomyelitis may also involve metaphyseal-epiphyseal junctions – however, the destructive lesions are usually more severe, leading in the latter stages of disease to growth and function impairment, which is rather unusual in congenital syphilis (Cockshott and MacGregor 1958). Overall, even though the bony parts of the lower extremities, where most typical changes of congenital syphilis are usually found, were not available for analysis, the skeletal lesions observed in our specimen

should be considered specific of congenital syphilis.

Both molar's germs have the appearance of mulberry molars. This examination showed topographic abnormalities of the occlusal surface. This was a rare opportunity to observe 'the mulberry germs'. Usually, we can find studies on erupted teeth. This defect is related to hypoplastic defects cutting into the bases of the cusps. In many papers, this defect was associated with Moon's molars, which are described as smaller and more dome-shaped than usual (Hillson et al. 1998; Hutchinson and Richman 2006). However, it is known that Moon's molars do not always accompany syphilis; on average they occur in one out of three patients, which means that in only 30–50% of cases of syphilis do the teeth show considerable deviation from the normal state (Horne 1953; Putkonen and Paatero 1961; Bernfeld 1971). Mulberry molars indicate serious disorders of the amelogenic organ as early as in the prenatal or perinatal periods (Hillson et al. 1998; 2002; Ortner 2003; Erdal 2006).

Although the "classical" shape of Hutchinson teeth is not always observed, the change of the size of cusps on the incisal edges may indicate crown formation disorder due to *Treponema pallidum* infection (Putkonen 1962). This concerns the uneven formation of the three mamelons on the incisor crown – a slight domination of the mesial and distal mamelons in relation to the central one. This results from the fact that the formation of dentine and enamel matrices on incisors in the central part has been disrupted, which leads to the weakening of the area. Consequently, this part of the crown is likely to wear out faster, and a character-

istic depression devoid of enamel may appear in the central part of the incisal edge (Jacobi et al. 1992; Hillson et al. 1998; Erdal 2006; Nystrom 2011). But this fact was not observed in the study sample. The germs of the first permanent incisors of the maxilla showed that the sizes of central mamelons are slightly higher than the mesial and distal mamelons. On the other hand, enamel hypoplasia in these areas, on permanent incisors indicates serious functional disorders of the amelogenic organ.

Mercury concentration values obtained for the examined samples were at a low level. This means that concentrations of mercury were similar to that typical of healthy teeth and bones in today's individuals (Powell and Cook 2005; Rasmussen et al. 2008; Keça et al. 2012). This means that observed topographic disturbances of posterior teeth (both smooth and occlusal surfaces), enamel surface and internal tooth structure could not be due to mercury, as Hutchinson suggested.

Conclusions

There are few publications of congenital syphilis in past populations. However, only a few described cases of children with diagnosed syphilis. The analysed case of congenital syphilis in the 3-year-old child shows characteristic changes in the skull, the metaphyses of the long bones and malformations of the deciduous and permanent teeth. In our case we observed the 'mulberry germs'. It is a rare opportunity to observe these lesions in such early ontogenetic stage. The mercury concentration in enamel and dentine were similar to those that are typical of healthy teeth in modern individuals.

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Author contribution

JT initiated the research, examined skeletal material, wrote first version of the manuscript. HMP reviewed literature for research. PP conducted radiological analysis. DOK and JT took photos of dental malformations and gave interpretation.

Conflict of interests

The authors declare that there is no conflict of interests.

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