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PALEORADIOLOGIC STUDY OF A 17TH CENTURY CASE OF TREPONEMATOSIS (NYÁRLŐRINC, HUNGARY)

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Abstract

This study deals with a female skeleton in a good state of preservation found in grave no 330 in the medieval cemetery at Nyárlőrinc. The grave can be dated to the 17th century. Macroscopic morphologic and paleoradiologic studies (X-ray and computed-tomographic analyses) have been carried out. The individual has widespread, bilateral and symmetrical florid periostitis, especially of the humeri, tibiae and femora. Similar lesions can be detected on the forearm bones, clavicles, scapulae, fibulae and calcanei. Periosteal new bone formation is associated with osteolytic areas and cortical deformities in several locations. Osteolytic changes of the sphenoid are to be mentioned. Traces of clustered pits can be recognized on the frontal and right zygomatic, which recall the first stage of contiguous gummatous osteoperiostitis. Macroscopic, radiologic and CT investigations of the cranial and postcranial alterations support the diagnosis of treponemal disease. The differential diagnosis and geographic situation suggest that the individual may have been suffering from acquired syphilis. The aim of this work is to attract attention to the possibility of identification of syphilitic lesions in uncommon locations and to the usefulness of paleoradiologic investigations during the diagnosis of treponemal diseases in ancient bones. The publication of complete cases in a good state of preservation can help further paleopathologic studies.

Key words: paleopathology, paleoradiology, 17th century, treponematosis.

Introduction

Within the group of specific infectious diseases, treponemal and mycobacterial infections can produce relatively characteristic skeletal lesions (HACKETT, 1976; STEINBOCK, 1976). While tuberculosis and leprosy produce, in general, morphologically and radiologically distinguishable changes, a distinction between the different forms of treponematoses is more difficult and often impossible.

The three types of *Treponema*, which can produce osteoarticular alterations in man, already classified as independent species, are actually brought together as three subspecies: *Treponema pallidum ssp. pallidum* (causative agent of venereal syphilis);

Treponema pallidum ssp. pertenue (agent of yaws, non-venereal) and *Treponema pallidum ssp. endemicum* (bejel, endemic syphilis, non-venereal) (GRIMPREL, 1993; MARTIN, 1993). These agents belong among the few pathogenic bacterial species that cannot be cultured *in vitro*. It is important to mention the inability to distinguish between bejel, syphilis and yaws microbiologically, biochemically, histologically, immunologically, or even with sophisticated DNA techniques (NOORDHOEK et al., 1990; GRIMPREL, 1993). Therefore, the clinical and epidemiological contexts preserve major importance in the diagnosis.

The situation is perhaps more critical in paleopathology. As the great majority of cases provide only skeletal materials for the examination, among the different types of clinical manifestations only the osteo-articular changes can be taken into consideration during the retrospective diagnosis. Fortunately, skeletal changes due to treponemal infections are clearly described in the pathologic, radiologic or paleopathologic literature (e. g. HACKETT, 1976; STEINBOCK, 1976; ORTNER and PUTSCHAR, 1985; VICENS et al., 1987; RESNICK and NIWAYAMA, 1988; EDEIKEN et al., 1990; MARCOVE and ARLEN, 1992; BAHK, 1994; LAGIER et al., 1994). In typical acquired or late congenital syphilis, morphologic (macroscopic or microscopic) and X-ray analyses reveal disseminated periosteal reactions, with thickening of cortical bone (with most frequent involvement of the tibia), and gumma formation (frequently in the skull), either in the cortex or in the spongiosa, accompanied by extensive proliferation. In yaws, osteitis and periostitis resemble syphilitic changes of bone. Destructive gummatous changes result in bone absorption and even pathologic fractures (EIDEKEN et al., 1990). The bone lesions of bejel are identical to those found in venereal syphilis, differing only in the relative frequency of various types of lesions (STEINBOCK, 1976). The pathologic and radiologic literature indicate that, in general, apart from the typical skeletal symptoms of early congenital syphilis (NABARRO, 1954; PANUEL, 1994), the sole characteristic in venereal syphilis, we cannot distinguish the osseous manifestations of the three diseases in the osteologic samples (VICENS et al., 1987; DUTOUR et al., 1994; LAGIER et al., 1994).

The geographic and epidemiologic context and the skeletal pattern of the lesions can suggest a hypothesis concerning the type of the treponemal infection, but never certitudes in paleopathology. Accordingly, the recent attempt by ROTHSCHILD and ROTHSCHILD (1995; 1996) to distinguish the three forms of treponematoses in osteoarchaeologic samples seems hazardous. To be able to compare large samples of treponemal cases at a population level, we need hundreds of well-preserved and correctly diagnosed cases per population (i. e. thousands of well-preserved skeletons of individuals who could have lived in the same area and in approximately the same period). In fact, skeletal samples are not so abundant and the paleopathologist discovers relatively small numbers of cases, which are always incomplete and fragmentary, and where it is always difficult to establish the diagnosis of the treponemal disease. We consider that the publishing of complete osteoarchaeologic cases of treponematoses and their morphologic and radiologic diagnosis, can help researchers in studies and diagnoses of paleopathologic lesions in more fragmentary skeletal material.

Materials and methods

The subject of our observation is a well-preserved skeleton found in grave no 330 in the cemetery at Nyárlőrinc (central Hungary). The cemetery, which was excavated under the direction of GYÖRGY SZÉKELY (archeologist of the Katona József Museum in Kecskemét), was used between the 14th and 18th centuries (H. TÓTH, 1990). The grave in question can be dated to the 17th century approximately (SZÉKELY, personal communication).

We discovered the pathologic lesions of this skeleton during the storing and classification of the anthropological series in the collection of the Department of Anthropology at József Attila University. The sex and age at death of the skeleton were determined by means of traditional methods used in historical anthropology (KNUSSMANN, 1988). Following a macroscopic morphologic study, a detailed radiologic and computed-tomographic analysis of the skeleton was carried out at the Service d'Imagerie Médicale, CHU Nord, Université de la Méditerranée, Marseille. Both the clinical and the paleopathologic special literature (e. g. HACKETT, 1976; ORTNER and PUTSCHAR, 1985; VICENS et al., 1987; RESNICK and NIWAYAMA, 1988; EDEIKEN et al., 1990) were used for differential diagnosis.

Results and discussion

The young adult female skeleton (age at death: 20 to 30 years) displays multiple abnormal bone changes. These abnormalities consist of both proliferative and destructive lesions, with evidence of the body's attempts at repair and remodeling. The characteristics of the observed bone lesions on the skeleton indicate that the disease process was active at the time of death.

Fig. 1. Traces of clustered pits on the frontal and nasal bones (photo by GY. PÁLFI).

Traces of clustered pits can be recognized on the frontal (Fig. 1) and nasal bones, and the right zygomatic. The radiographic plain-film of the skull presents lytic lesions on the frontal bone (Fig. 2). The CT of the skull shows small lytic areas on the outer table of the frontal region. The inner table is intact (Fig. 3). As concerns the base of the skull, osteolytice changes of the sphenoid are to be mentioned. Periosteal reactions and areas of focal destruction are seen on the vertebral borders of both scapulae.

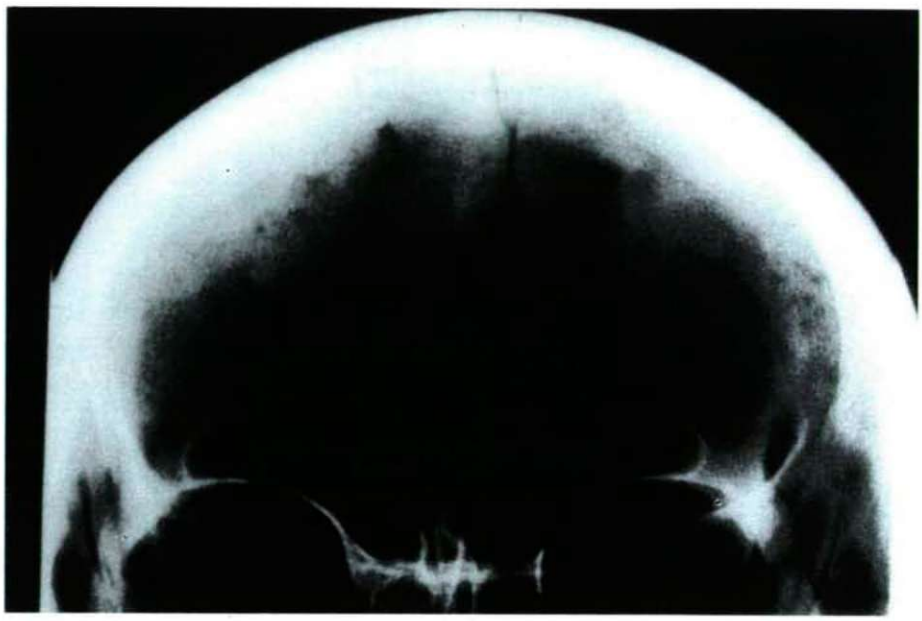

Fig. 2. X-ray picture of the skull presenting small lytic lesions on the frontal bone (X-ray by M. PANUEL).

The individual has widespread, bilateral and symmetric florid periostitis, especially of the humeri, tibiae and femora. Similar lesions can be detected on the forearm bones, clavicles, scapulae, fibulae and calcanei. Figure 4 presents the periosteal appositions and small areas of focal destruction on the distal diaphyseal and metaphyseal regions of the humeri. Radiological investigations of the humeri reveal subtle lytic areas associated with cortical thickening of the middle-third of the diaphysis. The periosteal appositions and cortical remodeling are well presented by the lateral view radiograph of the right humerus (Fig. 5).

Both femora exhibit extensive osteoproliferative lesions. CT examination of the femurs has been carried out (Fig. 6). The transverse CT scan of the affected region reveals a small round lytic area and a "double-layer" appearance, with a thick periosteal reaction and relative thinning of the cortex. The AP radiograph shows cortical thickening with periostitis and some radiolucent foci of cortical destruction (Fig. 7).

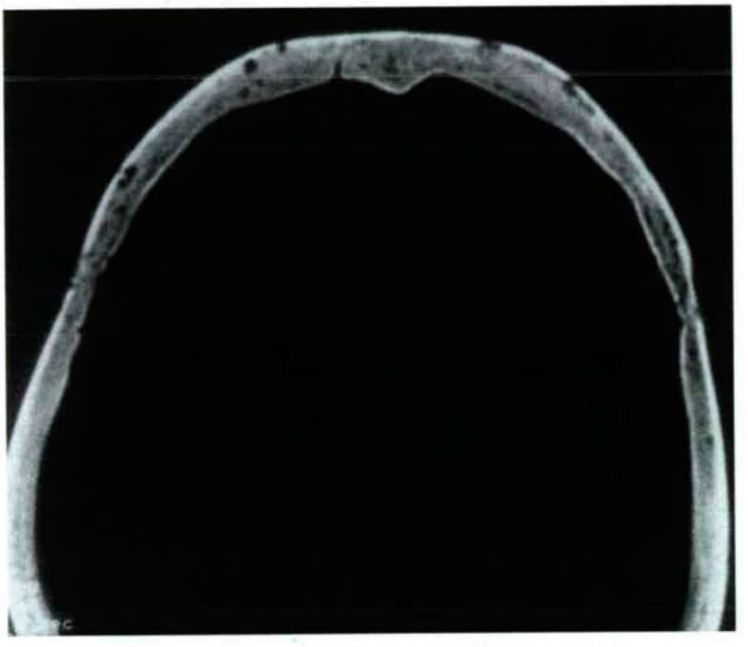

Fig. 3. CT picture of the skull: small lytic areas can be observed on the outer table of the frontal region (CT by M. PANUEL).

Fig. 4. Periosteal appositions and small areas of focal destruction on the distal diaphyseal and metaphyseal regions of the humeri (photo by GY. PÁLFI).

Fig. 5. Lateral view radiograph of the right humerus: the periosteal appositions and cortical remodeling are to be mentioned (X-ray by M. PANUEL).

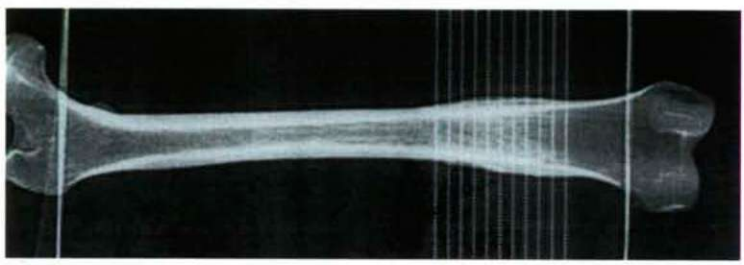

Fig. 6. CT analysis of the right femur (CT by M. PANUEL).

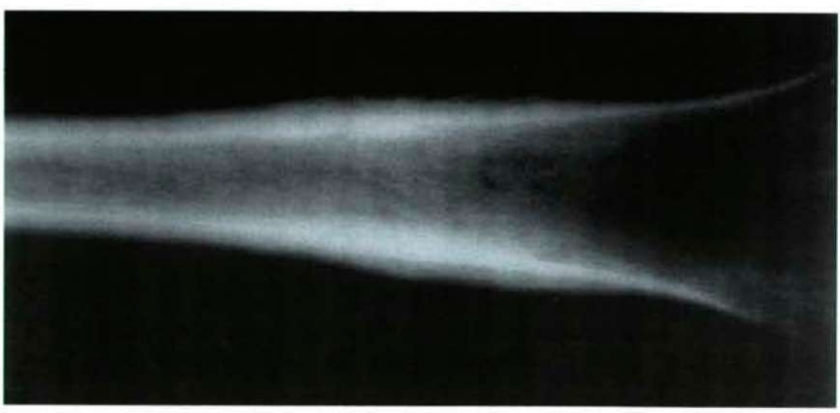

Fig. 7. AP radiograph of the right femur showing cortical thickening with periostitis (X-ray by M. PANUEL).

The tibiae and fibulae are both affected by the pathologic process. The macroscopic picture shows irregular periostitic hyperostosis (Fig. 8). The AP radiograph of the left tibia reveals cortical sclerosis of the diaphysis with periostitis (Fig. 9).

The positive lesions (periosteal appositions, cortical remodelling, etc.) must be regarded as indisputable *in vivo* processes. The paleopathologic interpretation of the osteolytic lesions is more difficult. However, these processes are, in all cases, associated in the same area of the bone with periostitic lesions and are characterized by sclerotic margins.

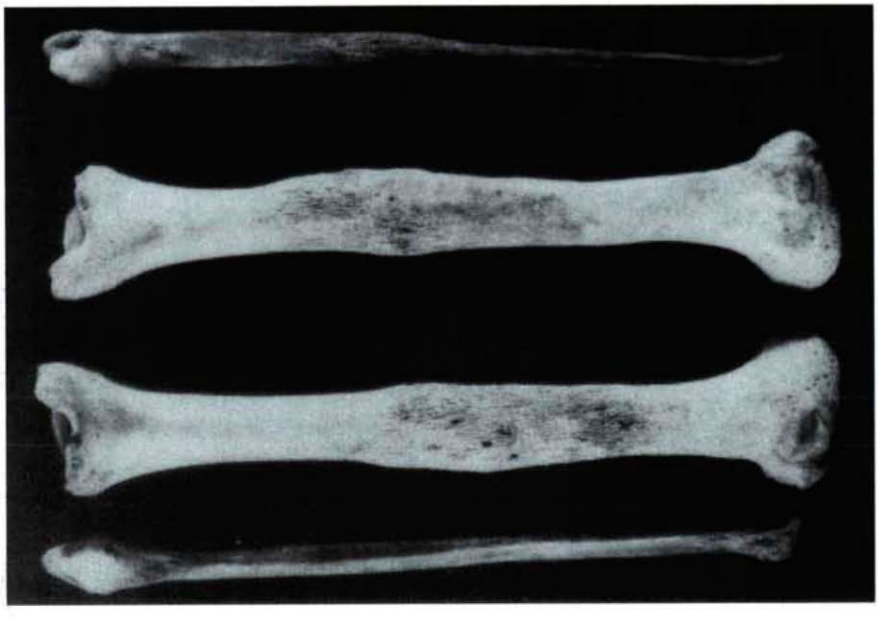

Fig. 8. Macroscopic picture of tibiae and fibulae, showing periostitic hyperostosis and osteolytic lesions (photo by GY. PÁLFI).

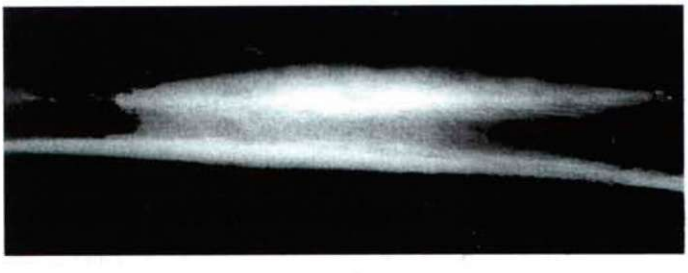

Fig. 9. AP radiograph of the left tibia, which reveals cortical sclerosis of the diaphysis with periostitis and focal areas of bone destruction (X-ray by M. PANUEL).

The picture of the above lesions is that of a generalized skeletal infection. The cranial lesions recall the first stage of contiguous gummatous osteoperiostitis (HACKETT, 1976). The macroscopic, radiologic and CT investigations of the cranial and

postcranial alterations support the diagnosis of a treponemal disease (ORTNER and PUTSCHAR, 1985; VICENS et al., 1987; LAGIER et al., 1994).

Conclusions

Our 17th century subject suffered from a disseminated treponemal disease. On the basis of the pathologic and radiologic literature, the skeletal pattern of the involvement suggests that the young adult woman may have been affected by acquired syphilis (EIDEKEN et al., 1990; MARCOVE and ARLEN, 1992; BAHK, 1994). As concerns the geographical context, the temperate climatic zone seems to be more favorable for the venereal form of infection than for a non-venereal one. On the other hand, we already have some medical historical and paleopathologic data from the Hungarian Plain prior to this period (MARCSIK, 1994). Acquired syphilis appears to be the most probable hypothesis in our case, but an endemic form cannot be discarded *a priori*.

It was mentioned in the introduction of this paper that traces of reliable treponemal cases in osteoarchaeology are very rare. To our knowledge, following the reports by FERENCZ and JÓZSA (1990) and MARCSIK (1994), the presented case is the third reported paleopathologic case of treponematosis from Hungary, in spite of the thousands of skeletons examined previously. This underlines the importance of the publication of these data, for diagnostic and also paleoepidemiologic reasons.

The scarcity of the discoveries is more characteristic - and problematic - in more ancient historical periods. This lack of data (especially in the Old World) has generated much controversy regarding the origins of treponemal disease and, in particular, acquired syphilis (e. g. HUDSON, 1958; COCKBURN, 1961; HACKETT, 1963; GRMEK, 1983; BAKER and ARMELAGOS, 1988; LIVINGSTONE, 1991; ROTHSCHILD and ROTHSCHILD, 1996; BRUN, 1996).

Fortunately, recent discoveries in this field (e. g. STIRLAND, 1991; HENNEBERG and HENNEBERG, 1992; PÁLFI et al., 1992; BLONDIAUX and ALDUC-LE BAGOUSSE, 1994; ROBERTS, 1994; ROTHSCHILD et al., 1995; HADJOUIS, 1996), have, at least partially, resolved the problem. Ever more scientists accept that treponematosis (or treponematoses) was (were) present in both hemispheres before the discovery of America, and that we have to search for the ancestor of *Treponema pallidum* in the same area as one of the ancestors of the genus *Homo*: in Africa (e. g. DUTOUR et al., 1994; FROMENT, 1994; POWELL, 1994; ROTHSCHILD et al., 1995; BRUN, 1996). However, the interpretation of the origin of the venereal syphilis is still controversal (ANDRÉ, 1994; ROTHSCHILD et al., 1995; BRUN, 1996; DUTOUR et al., 1996; ROTHSCHILD and ROTHSCHILD, 1996).

Some questions are resolved, others are just appearing: the definitive scenario of the history of treponematoses is still a long way off. It seems absolutely certain, however, that human paleopathology and particularly paleoradiology, will contribute largely in future researches on this topic.

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