Leprosy in individuals unearthed near the Ermida de Santo André and Leprosarium of Beja, Portugal

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Abstract  Documentary sources refer to leprosy patients in the Portuguese territory since the first century AD, and in the Middle Ages around 70 leprosaria were established. However, prior to 2003 this historical evidence had not been confirmed by archeological findings. The excavation performed in monitoring the rehabilitation done by the Polis program in the area of the Ermida de Santo André (hermitage of Saint Andrew) allowed the exhumation of seven human skeletons, and commingled bones from at least three individuals, in the vicinity of the Beja leprosarium. The objective of this study is to present the paleopathological lesions relevant to the discussion of the differential diagnosis of leprosy. Macroscopic observation of the bones and scrutiny of lesions according to the paleopathological literature allowed the identification of a probable case of leprosy in an adult male, showing rhinomaxillary changes and concentric remodeling of hand and foot bones, and four possible cases (two young adults and two adults, all probably males), with a set of lesions in facial bones and skeletal extremities. The poor preservation of the bones precluded further confirmation of this diagnosis. According to historical data, the leprosaria functioned between the 14th and 16th centuries AD. The exact chronology of these findings was not determined either during the excavation or by radiocarbon dating because the bones presented poor collagen levels. In Portugal as a whole there are few osteological evidences of leprosy, and thus this study adds new information about this chronic infectious disease.

Key words: Hansen’s disease, leprosy, paleopathology, rhinomaxillary changes, destructive diaphyseal remodeling

Introduction

Leprosy has been present in Europe since at least the 4th–3rd centuries BC (Mariotti et al., 2005; Roberts and Manchester, 2005). For the following centuries the paleopathological and/or written records of this disease are rare. However, it is known that in the 5th century AD the first hospital for leprosy patients was established (Carvalho, 1932; Mira, 1947; Dueñas et al., 1973). The archeological excavation of medieval cemeteries associated with leprosaria has revealed an increased number of skeletons with lesions compatible with leprosy—for example, the cases of St. James and St. Mary Magdalene hospitals at Chichester (Magilton et al., 2008) and St. Mary Magdalene at Winchester (Roffey and Tucker, 2012), both in England, and St. Jørgen’s at Naestved (Møller-Christensen, 1952, 1953a, b; Andersen, 1969; Bennike, 1991, 2002) and St. Jørgen’s at Odense (Arentoft, 1999; Boldsen, 2001; Matos, 2009; Matos and Santos, 2013) in Denmark.

In Portugal the first written references to leprosy date from the 1st century AD and during the Middle Ages around 70 small leprosaria were founded in the country (Carvalho, 1932). According to this author, this disease was rare until the 17th century. The first paleopathological evidence of this chronic infection was found in 2003 during the excavation conducted along with the rehabilitation of the area surrounding the Ermida de Santo André (hermitage of Saint Andrew), in Beja, by the Polis programme (Antunes-Ferreira and Rodrigues, 2003).

The Ermida de Santo André in Beja dates from the 15th century AD (Viana, 1943; Espanca, 1992; Borrela, 1995; Goes, 1998) but according to written sources it may have been built in the 12th century AD (Cardoso, 1751; Viana, 1943; Borrela, 1995). Documentary research revealed the existence of a leprosarium (gafaria) in the surroundings of this hermitage (Espanca, 1992; Goes, 1998). However, its chronology was not fully established. Carvalho (1932) quoted a will dated from 1377 made in favor of the poor and leprosy victims from the Albergaria de Santa Anna which seems to correspond to the Beja leprosarium. Mestre (1991) stated that the leprosarium of Saint Anne was extinct in the 15th century and their belongings were incorporated in the hospital of Our Lady of Mercy (Nossa Senhora da Piedade) built in the 15th/16th centuries. This hospital is also called Mercy hospital (Hospital da Misericórdia) (Carvalho, 1932;
Mestre, 1991) and in 1932 it still preserved the old archives of the gafaria (Carvalho, 1932). The name of the patron of the leprosarium is not the same in the publications consulted, since Goes (1998) used the designation St. Lazarus leprosarium, reporting its activity at least between 1509 (Goes, 1998) and around the end of the 16th century (Espanca, 1992; Goes, 1998). This discrepancy might be related to the administrative change that happened after its incorporation in the Mercy hospital. Further references to this place were found in early 20th-century reports that the houses and other ruins from the leprosarium were demolished in 1939 and the hermitage was renovated under the supervision of the municipality and heritage office (DGEMN, Direcção-Geral dos Edifícios e Monumentos Nacionais) (Borrela, 1998; Borrela and Campaniço, 2004).

This site, along with the cemetery identified in 2009 in the ‘Leprosarium Valley’ in Lagos, dated to between the 15th and 17th centuries (Ferreira et al., 2013) are, so far, the only two archeological evidences of past leprosaria cemeteries in this country.

The aim of the current paper is to report the pathological lesions with relevance to the differential diagnosis of leprosy in individuals exhumed from the cemetery near the Ermida de Santo André.

### Materials and Methods

In 2003, during the Polis rehabilitation program in Beja, the surroundings of the Ermida de Santo André were excavated by the archeology enterprise Crivarque, Lda, having as anthropologist one of the authors (N.A.F.). In the fieldwork seven surveys were opened in the vicinity of the hermitage (five were located about 20 m north, one next to the north wall of the hermitage and one 5 m south), totaling an area of 42.56 m².

Human remains were found in survey number 6, located near the north wall of the hermitage. This area was originally 4 m × 3 m but was later extended to 5.8 m × 3.2 m (18.56 m²) due to the presence of ten graves. However, only seven were excavated because the remaining three (sk 2, 9, and 10) were outside the working area. This excavation also exposed a pavement (built of small blocks of granite, quartzite and quartz) placed directly over one of the burials; a small wall, partially destroyed, built in brick and close to the grave number 7, which possibly was from one of the houses destroyed in the early 20th century; and a drainage pipe placed by the DGEMN which also affected the burials. In short, skeleton 1 did not include the skull, cervical vertebrae, and scapulae; in skeleton 3 the right humerus was absent; skeleton 4 is the only one which is quite complete; skeleton 5 preserved only the skull, humeri, and the upper part of the trunk; skeleton 6 did not preserve the lower left limb bones and the right tibia, fibula, and foot bones; skeleton 7 did not preserve the distal portion of both femurs or any of the remaining lower limb bones; and skeleton 8 did not have the skull.

The graves did not show any delimitation structure with the four older ones having an oval shape built into the rock while the three more recent ones were placed over the previous. All the individuals were inhumed in dorsal decubitus position, with a NE–SW orientation, aligned with the hermitage wall, suggestive of Christian burials. Their upper limbs, when discernible, were placed along the body (n = 1), above the abdominal region (n = 2), or above the pelvis (n = 3). Beside the individuals in articulation, over the right lower limb of skeleton 3 were placed commingled bones from at least three individuals, two adults and one non-adult. Despite all efforts it was not possible to match these bones with the primary inhumations. However, this possibility cannot be completely discarded due to post-mortem fragmentation.

The doubts about the chronology of the leprosarium of Beja were not solved by the excavation because the stratigraphy points to a medieval/modern occupation. Thus, a bone sample from skeleton 3 was sent to the radiocarbon unit of Oxford University but the analysis “failed due to very low yield.”

In the laboratory, the human remains were observed macroscopically, through a naked eye observation using an artificial light and, whenever necessary, a 10× magnifier lens. Sex and age at death were estimated according to standard methods (Ferembach et al., 1980; Buikstra and Ubelaker, 1994; Bruzek, 2002).

The paleopathological analysis followed the generic recommendations detailed in standard textbooks (Buikstra and Ubelaker, 1994; Aufderheide and Rodriguez-Martin, 1998; Ortner, 2003). Additionally, the identification of the specific bone lesions commonly considered relevant for the discussion of a leprosy diagnosis on dry skeletal material was based on the following: (1) rhinomaxillary changes were identified according to the original descriptions for facies leprosa made by Møller-Christsensen (1953b, 1961, 1967) and also using the later modifications brought by the rhinomaxillary syndrome concept proposed by Andersen and Manchester (1992); (2) bone changes of the hands and feet were researched following Møller-Christsensen (1961), Andersen (1969), Andersen and Manchester (1988), Andersen and colleagues (1992, 1994), Rothschild and Rothschild (2001), Ortner (2003, 2008a, b) and Rothschild and Behnam (2005); (3) periosteal reactions on long bone diaphyses were identified following Buikstra and Ubelaker (1994) and Ortner (2003) but also attending to the considerations made by Hackett (1976), Lewis et al. (1995), Matos and Santos (2006) and Weston (2008) regarding new bone formation classification, recording, and interpretation.

The differential diagnosis of the recorded lesions, solely or combined, and whether these are indicative, or not, of leprosy was discussed considering the works by Møller-Christsensen (1967), Andersen and Manchester (1992), Ortner (2003, 2008a, b), Waldron (2009) and Matos (2009).

### Results

Ten skeletons were identified in the area excavated. Of the seven exhumed, three were adolescent/young adults and four adults, probably males (Table 1). Three (sk 2, 9, and 10) were not excavated, remaining in situ, and the observation of pathological lesions was not possible during the fieldwork. The pathological observation of the exhumed individuals will be presented by anatomical region.
Rhinomaxillary changes

Rhinomaxillary bone changes were observed in three individuals (sk 3, 4, and 6), i.e. all the skeletons with this area preserved. The most conspicuous lesions were noticed in skeleton 4, showing pathological remodeling of the piriform margins (Figure 1A) and abnormal pitting on both anterior halves of the palate encompassing the incisive canal (Figure 1B). Both the anterior nasal spine and the posterior region of the palatine process were destroyed post mortem and thus not observable. In skeleton 6 only a small area from the right maxilla was preserved, showing new bone formation at the nasal floor, or nasal surface, of the palatine process. Additionally, this fragment shows that both anterior nasal spine and piriform margins were intact, i.e., without pathological changes (Figure 2). Skeleton 3 preserved a small fragment of the middle region of the palatine process, showing “inflammatory” changes, consisting of pitting and new bone formation in both nasal floor (Figure 3) and palate.

Despite the presence of nasal and palatal changes in these three individuals, the six classic signs of the rhinomaxillary syndrome (RMS) were not simultaneously present in the same individual. This could be either due to post-mortem destruction or because RMS changes are not always concomitant (as skeleton 6 demonstrates).

Postcranial changes

Concerning the lower limb bones, bilateral new bone formation on tibiae and fibulae was observed in skeletons 1, 3, 4, and 8 (Table 1). The most common lesions and location are exemplified in Figure 4. In skeleton 5, the two fragments of the left fibula recovered presented periosteal reaction while the right was not recovered. Skeleton 6 did not preserve these bones and skeleton 7 was poorly preserved, precluding the observation of bone surface; however, the neck of the femurs shows porosity also called cribra femoralis.

The most notorious lesions were found on skeleton 8, which presented bilateral and symmetrical extensive new...
bone formation on both tibiae and fibulae (Figure 5).

In the hand and foot bones, lesions were found in only three individuals (sk 1, 4, and 8). Interestingly, these changes affected simultaneously the hand and foot bones of the skeletons. Skeleton 4 showed the most striking postcranial lesions, all presenting bilateral distribution. In the hands, concentric diaphyseal destructive remodeling and acro-osteolysis was noticed in six left (Figure 6A) and one right phalange. The second right metacarpal showed evidence of acro-osteolysis and knife-edge diaphyseal remodeling (Figure 6B). As shown in Figure 7, similar pathological phenomena were recorded bilaterally and symmetrically on the second–fifth metatarsals. Additionally, the fifth left metatarsal presented an oval osteolytic lesion located at the
proximal epiphysis probably resulting from secondary infection (Figure 8). Phalanges from both feet (one left and two right) suffered diaphyseal concentric remodeling.

The individual number 8 showed poor preservation of either the upper or lower extremities due to taphonomic constraints. Hand bone lesions were observed bilaterally. Both the fourth left metacarpal and an intermediate right phalange presented acro-osteolysis despite the diaphyseal destructive remodeling being absent. Additionally, a left proximal (?) phalange showed concentric destructive remodeling of the diaphysis and severe degenerative changes on the proximal joint. Concerning the feet it is highlighted that two metatarsals—the fifth right (?) and an unidentified one—presented extensive bone changes and acro-osteolysis. Additionally, the fifth metatarsal showed severe changes of the proximal epiphysis, probably resulting from secondary infection
(septic arthritis and osteomyelitis), where at least one draining canal (cloaca) is evident (Figure 9).

Skeleton 1 presented unilateral bone changes on the hands since only the left was affected. Ankylosis between an intermediate and distal left phalanx was noticed (Figure 10A). An additional incomplete left phalanx presented diaphyseal concentric remodeling. Bilateral foot lesions were observed. Two right metatarsals presented both acro-osteolysis, knife-edge concentric remodeling of the diaphysis, and degenerative changes at the proximal epiphysis (Figure 10B). Acro-osteolysis was further observed in two foot phalanges (one from each side). Additionally, dorsal exostoses were present on the left navicular (Figure 10C).

Discussion

The paleopathological analysis of the skeletons exhumed from the necropolis of the Ermida de Santo André at Beja provides the opportunity to improve our nascent knowledge regarding the presence of leprosy in the Portuguese territory in past times.

The main results obtained from the study of the seven individuals from the St. André church are summarized on Table 1.

Five out of the seven skeletons present lesions for which
leprosy could be considered the possible cause. It must be stressed that in the current state of knowledge there are no skeletal lesions that by themselves are pathognomonic, i.e. indicative of the presence of leprosy. In fact, Cook (2002: 82) emphasizes that “‘pathognomonic’ is rather strong language for a field with many limitations that we must recognize in paleopathology.” Differential diagnosis issues are challenging for paleopathologists since many pathological conditions share similar skeletal lesions. This difficulty is obvious in some recent publications (e.g. Phillips and Sivilich, 2006; Brothwell, 2010; Christensen et al., 2013) where leprosy was one of the clinical entities considered when performing differential diagnosis.

The pattern and combination of bony lesions are considered the key and, as Ortner (2008b: 206) notes, “the ability to diagnose leprosy in archaeological human skeletal remains ranges from problematic to highly likely.” Møller-Christensen in his pioneering works was aware of the puzzling nature of the paleopathological diagnosis of leprosy as revealed by his writings:

The degree of certainty of a diagnosis depends of course on the nature of the human remains, and how complete they were. If a cranium displaying facies leprosa was found, it was considered as a possible case of leprosy. If the tibiae and fibulae showed no pathological changes, or had not been preserved, the case was not regarded as being a sufficiently proven one of leprosy. Only when a cranium with facies leprosa was accompanied by tibiae and fibulae showing typical pathological changes, bilaterally and symmetrically, was a more firm diagnosis of the lepromatous type of leprosy made. Fairly certain was only possible when marked changes also occurred in preserved hand and foot bones (Møller-Christensen, 1967: 300).

More recently, the simultaneous presence of rhinomaxillary changes and acro-osteolysis and/or destructive remodeling of the hand and foot bones is considered highly suggestive of leprosy (Ortner, 2008a, b). However, this conservative approach is not always strictly followed and more flexible diagnostic criteria for the diagnosis of leprosy are often applied by researchers. For example, Andersen and Manchester (1992: 122) proposed that “the presence of all components of the rhinomaxillary syndrome is pathognomonic of lepromatous or near-lepromatous leprosy” and Waldron (2009: 101) suggests “operational definitions for leprosy,” these comprising “rhinomaxillary syndrome OR concentric loss of bone from phalanges of the feet or neuropathic change in the joints of the feet or ankles.” Thus, depending on the criteria adopted for the diagnosis of leprosy

Figure 10. Skeleton 1. (A) Dorsal view of ankylosed left hand intermediate and distal phalanges. (B) Dorsal view of two right metatarsals presenting acro-osteolysis, knife-edge diaphyseal remodeling and degenerative changes at the proximal epiphysis. (C) Dorsal view of the left navicular showing dorsal exostosis and porosity.
slightly different results, either at individual or population level, may be obtained (Matos, 2009).

Amongst the skeletal material unearthed near the Ermida de Santo André necropolis, only in skeleton 4 can a probable diagnosis of leprosy be established. This individual presents concomitant destructive remodeling in the rhinomaxillary region, metacarpals, metatarsals, and hand and foot phalanges. These lesions were bilateral in both hand and feet, and symmetrical in the left. It must be stressed that this is the best-preserved skeleton from the sample and, as noted by Pinhasi and Bourbou (2008), the paleopathological diagnosis of leprosy is always conditioned by the skeletal elements available.

The incomplete preservation due to taphonomic constraints of the remaining individuals made the diagnosis of leprosy either impossible, such as in the case of skeletons 5 and 7, or very difficult, namely in skeletons 1, 3, 6, and 8, which are considered possible (but not probable) cases of leprosy since a definitive diagnosis was unattainable. These four individuals can be grouped as follows:

1. Skeletons 1 and 8 do not preserve the skull but present destructive remodeling on hand and foot bones and bilateral new bone formation on the lower leg bones, whereas skeleton 6 did not preserve the lower limb bones. These individuals present poor cranial preservation and a clear diagnosis of leprosy cannot be established based on the observed lesions on the fragmentary rhinomaxillary area. Andersen and Manchester (1992) consider that “rhinomaxillary syndrome” is pathognomonic of lepromatous leprosy only when the full spectrum of lesions is present, but this is not the case of either skeleton 3 or 6. Even if this were the case the pathognomonic value of this syndrome is not consensual because this anatomical region may be involved in many other disease processes such as mucocutaneous leishmaniasis (Herwaldt, 1999; Manchester, 1994; Auderheide and Rodriguez-Martín, 1998; Ortner, 2003; Malekpour and Esfandbod, 2010; Marsteller et al., 2011), neoplasms (Hackett, 1976; Auderheide and Rodriguez-Martin, 1998; Ortner, 2003; Eggesbø, 2012; Koivunen et al., 2012), rhinoscleroma (Becker et al., 1981; Pontual et al., 2008), rhinosporidiosis (Bonifaz et al., 2011), sarcoidosis (Manchester, 1994; Mrówka-Kata et al., 2010), systemic mycosis (Zargari and Elpens, 2009; Bonifaz et al., 2011), treponematosis (Hackett, 1976; Manchester, 1994; Auderheide and Rodriguez-Martín, 1998; Ortner, 2003; Cook, 2002; Ortner, 2003; Cook and Powell, 2005, 2012), tuberculosis (lupus vulgaris) (Manchester, 1994; Ortner, 2003, 2008a; Roberts and Buikstra, 2003; Garg et al., 2010), and Wegener’s granulomatosis (Chauhan and Cruz, 2007). It must be noted, however, that according to Manchester (1994: 80), in what concerns the interpretation of rhinomaxillary lesions only leprosy, tuberculosis (lupus vulgaris) and treponematosis “are of practical significance in paleopathological differential diagnoses.”

Final Comments

In an area of 18.56 m² excavated near the Ermida de Santo André and also documented as the location of Beja leprosarium seven skeletons were unearthed, plus at least three individuals in commingled bones. Three more skeletons were identified but remained in the soil. From this assemblage one adult male presents lesions compatible with a probable case of leprosy while in four others (two young adults and two adults, all probably males) the poor bone preservation only allows the recording of possible cases.

Unfortunately neither the excavation nor the attempted radiocarbon dating of one skeleton allowed the determination of an exact chronology of these individuals. Nevertheless,
these findings reinforce the information from documentary sources that place the Beja leprosarium to have been active at least between the 14th and 16th centuries AD, in the area surrounding the Ernida de Santo André. Due to the importance of this site to the history of leprosy in the country, further excavation in this area would be beneficial.

Acknowledgments


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