

# Back to the basics in the paleogenomic era: what about the diagnosis of tuberculoid leprosy in the paleopathological record?

VÍTOR MATOS [vmatos@antrop.uc.pt] | CIAS -Research Centre for Anthropology and Health | Department of Life Sciences, University of Coimbra, Portugal

## Introduction

The first paleopathological criteria for the diagnosis of leprosy were presented 65 years ago, at the VI International Leprosy Congress in Madrid, by the Danish physician V. Møller-Christensen (1953, 1967), who defined *facies leprosa*. Later the concept of *rhinomaxillary syndrome* (RS) became central to the retrospective diagnosis of leprosy. RS was proposed by Andersen and Manchester (1992: 122) who considered that “the presence of all components of the rhinomaxillary syndrome is pathognomonic of lepromatous or near-lepromatous leprosy”. Lepromatous (LL) and tuberculoid (TL) leprosy are the two clinical polar forms of leprosy according to the Ridley and Jopling (1966) classification. During the last six decades *facies leprosa*, RS, and appendicular destructive bone changes (Figure 1), have been widely used (Ortner, 2003), and sometimes misused, to characterize the chronological and geographic distribution of leprosy. However, a review of published paleopathological leprosy cases shows that TL is almost absent in the bioarcheological record. This gap must be addressed in order to improve our knowledge regarding the origin and paleoepidemiology of leprosy. This study aimed to collect and correlate clinical and paleopathological data on leprosy related bone changes to answer the following:

- Do LL and TL present distinct patterns of bone involvement?
- Can TL be diagnosed in archaeological skeletal material?

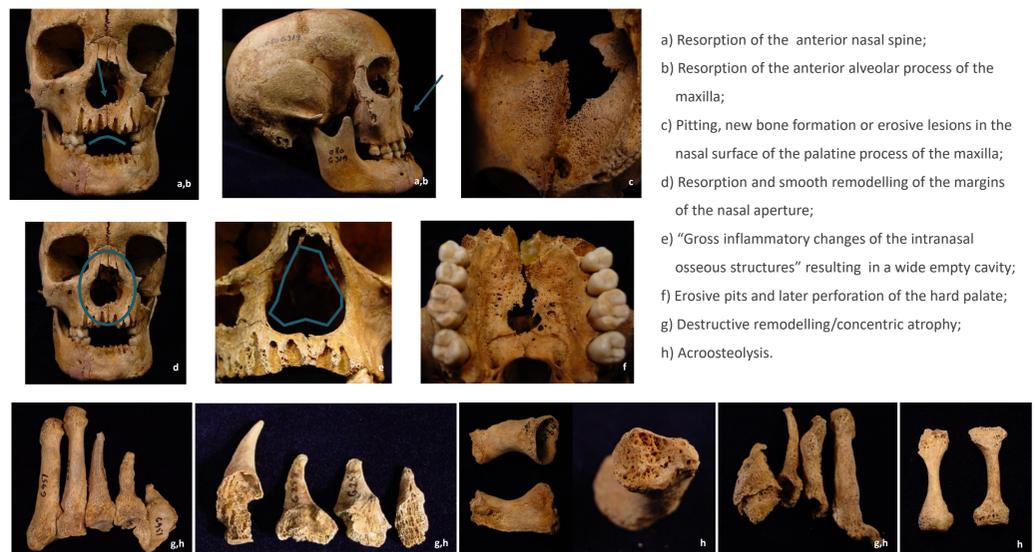


Figure 1 – Bone changes commonly occurring in leprosy patients according to paleopathological literature: rhinomaxillary area (a-f) and appendicular skeleton (g-h).

## Samples and Methods

### 1. Medical archives - Leprosarium Hospital-Colónia Rovisco Pais (HCRP), Tocha, Portugal

300 clinical files randomly selected (details in Matos, 2009):

- 150 files from each type of leprosy (LL and TL);
- 150 from each sex;
- 35 (11,9%) non adults and 258 adults (88,1%).

Screening: between 1947-1985 (median: 1958; s.d.: 8,57).

First admission: 4 to 93 years old (median: 37; s.d.: 17,6).

Only bone changes before antibiotherapy were collected.



Figure 2 - The HCRP was the Portuguese National Leprosarium between 1947 and 1996 (photo taken around 1947).

### 2. St. Jørgen's medieval leprosarium skeletons (13th – 16/17th cent.), Odense, Denmark

191 skeletons, housed at the University of Southern Denmark, with well preserved rhinomaxillary area were observed (details in Matos, 2009):

- 148 adults (77,5%) and 43 non adults (22,5%);
- 75 males, 63 females and 37 undetermined.

Standard osteological and paleopathological methods were applied.

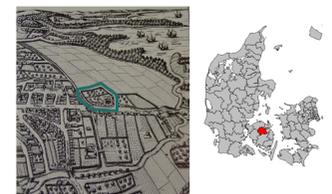
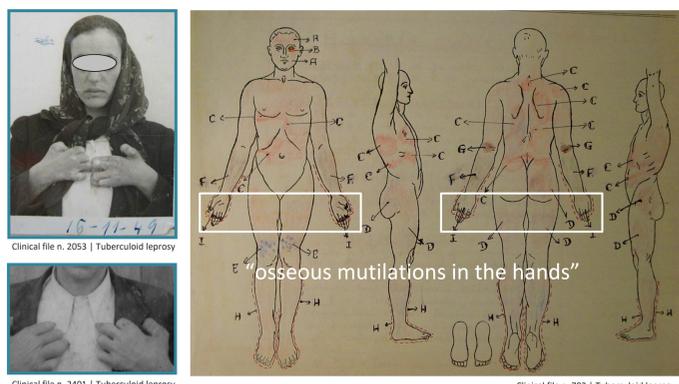


Figure 3 - Location of the St. Jørgen's leprosarium and cemetery in Odense, Denmark, around 1590 AD (in Arentoft, 1999: 31).

## Results and Discussion

### 1. HCRP leprosarium patients



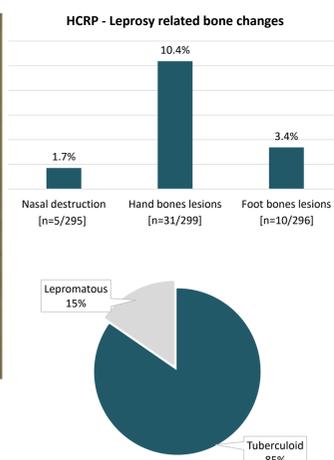
Overall prevalence of osseous lesions: **13,0%** (39/300).

Observed bone involvement patterns:

LL (**4,0%** [6/150]): Nasal destruction with or without hand and foot bone lesions.

LT (**22,0%** [33/150]): No nasal destruction and hands/feet bone lesions present.

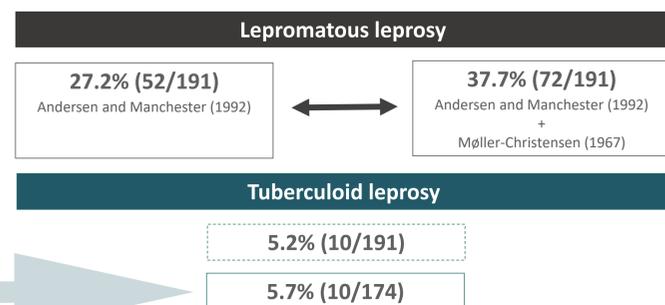
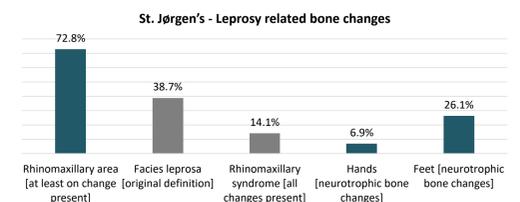
Bone changes more prevalent in TL than in LL patients (OR=8,18; IC95%=1,69-39,65; controlled for sex and time between onset and screening).



### 2. St. Jørgen's medieval leprosarium skeletons

**Lepromatous leprosy:** Frequency ranged between 27.2% (n=52) and 37.7% (n=72).

**Tuberculoid leprosy:** When the distinctive patterns of TL and LL found in the HCRP leprosarium archives were transposed to the St. Jørgen's sample, **ten (5.7%) possible cases of TL** were identified for the first time in paleopathological context.



- The paleopathological identification of TL should be considered when an adult skeleton without rhinomaxillary lesions presents acroosteolysis and destructive remodeling/concentric atrophy of hand and/or feet bones, and after the exclusion of other pathological conditions that must be considered on the differential diagnosis of these lesions, for example, diabetes, frostbite, psoriatic arthritis, Raynaud's syndrome, etc.
- The long time to affect the skeleton after the disease onset and the poor post mortem preservation of small and fragile bones, such as those from the nasal area, may be responsible for the few leprosy cases identified on human osteological remains, specially in relation to what would be expected from documentary sources.
- Further investigations are necessary concerning the retrospective diagnosis of leprosy. The knowledge on how the various forms of leprosy affected humankind would help a better understanding of leprosy evolution and history.

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FCT  
IF INVESTIGADOR FCT  
CIAS  
UNIVERSIDADE DE COIMBRA