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UNIQUE ASPECTS OF WEST COAST TREPONEMATOSIS

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Skeletal populations from the western coast of North America clearly were afflicted with a treponemal disease very different from that previously documented elsewhere in North America. Six populations from west of the Sierra Cascades were compared with 5 sites east of the Cascades. A high population frequency (both in adults and subadults) of pauci-ostotic, periostitis was noted in the six western skeletal populations, identical to that reported previously with bejel in Negev Bedouins, Sudanese Nubians, and the Kit site from Iraq. Early populations, from east of the Cascades, had a very different polyostotic disease pattern, characteristic of yaws, and identical to that previously reported in Guam. Both patterns were clearly distinguished from syphilis, which appears to be a later development (mutation?). This study provides evidence that the treponematoses were transported to the New World by way of at least two migrations, one bringing yaws; the other, bejel. The population with bejel likely derived from a different population than that with yaws. Given the absence of treponemal disease variation in the very wide spectrum of environments represented by the bejel-afflicted populations, it is clear that environment is not the factor determining disease character. This study expands on animal studies documenting that the individual treponematoses are separate diseases and not simply climate-induced variation.

Key words: Paleopathology, bejel, yaws, syphilis.

Las poblaciones esqueletales de la costa occidental de Norteamérica fueron claramente afectadas por una treponematosis muy diferente a la previamente documentada en otras partes de Norteamérica. Seis poblaciones del Oeste de la Sierra Cascadas fueron comparadas con cinco poblaciones del Este de las Cascadas. Se registró una alta frecuencia (en adultos y subadultos) de periostitis pauci-ostotic en las seis poblaciones occidentales, idéntico a la periostitis anteriormente registrada con bejel en los beduinos de Negev, los nubianos de Sudan, y el sitio Kit de Iraq. Las poblaciones tempranas del Este de las Cascadas tenían un patrón muy distinto de la enfermedad poliostotic, característico de la pián e idéntico al previamente registrado en Guam. Ambos patrones fueron diferenciados de la sífilis, la cual aparece más tarde (mutación?). Este estudio provee evidencia de que las treponematosis llegaron al Nuevo Mundo a través de dos migraciones, una trayendo pian y la otra, bejel. La población con bejel probablemente derivó de una población diferente de la que tenía pián. Dada la ausencia de variación de treponematosis en un amplio espectro de ambientes representados por poblaciones afectadas con bejel, es claro que el ambiente no es el factor que determina el carácter de la enfermedad. Este estudio va más allá que los estudios de animales, documentando que las treponematosis individuales son enfermedades separadas y no simplemente variaciones inducidas por el medio ambiente.

Palabras claves: *Paleopatología, bejel, yaws, sifilis.*

The prepared mind sometimes sees only what it is prepared to see (what it expects) (Rose 1995). However, study of periosteal disease in populations from coastal North American archeological sites revealed an unexpected pattern. Baker and Armelagos (1988) speculated that venereal transmission was a post-Columbian event in North America. Others have not been so certain, instead identifying non-venereal treponemal disease (Bullen 1972; Neuman 1975; Powell 1995; Schermer et al. 1994). Such studies have speculated (on the basis of examination of "classic cases" that treponemal infection in North America was likely yaws or bejel. Validated criteria (Rothschild and Rothschild 1995b) allowed verification of their perspective, further clarifying yaws and later syphilis in such North American sites (Rothschild and Rothschild 1994a,b, 1995a; Rothschild et al. 1995). It was therefore astonishing to find a pattern in the Baja peninsula disparate from both treponematoses. Comparison of those findings revealed a pattern identical to that previously attributed to an alternative treponemal disorder (Hershkovitz et al. 1995; Rothschild and Rothschild 1995b). That unexpected finding led to a survey of sites west and north of the Sierra Cascades, which were contrasted with findings in sites east and north of the Cascades.

Syphilis in this manuscript refers to venereal as opposed to endemic syphilis. Anthropologic reports have occasionally used the latter term for all non-venereal (osseous-impacting) treponemal disease, while medical use (archaic) of endemic syphilis identifies bejel. This manuscript uses the term bejel, to distinguish the disease from yaws and (venereal) syphilis.

Cranial pathology has long been explored as a potential technique for recognizing and distinguishing among the treponematoses (Ortner and Putschar 1985; Steinbock 1975). This perspective contradicts Hackett (1976). Hackett stated that he would not attempt to distinguish among the treponematoses, because skull samples were compromised by significant collection bias, and because he did not have documented examples of the various treponemal diseases.

Skull findings, like reports of selected or isolated cases (Bullen 1972; Powell 1995), do not lend themselves to epidemiologic differentiation of the treponematoses and therefore were excluded from discussion.

Dental stigmata would seem to be a good question yet hypoplasia results in infrequent observation in any syphilis-affected population. No one has been able to use dental stigmata or cranial changes to provide clear evidence of treponematosis variety. Use of skull and dental stigmata have not allowed distinguishing among the treponematoses.

Hacketts (1976) and Steinbocks (1975) speculation of possible variation in skeletal distribution of periosteal reaction stimulated us to explore such an approach based on examination of (treponemal disease) diagnosed populations. The resultant technique for distinguishing among the treponemal disorders has now been validated for use in population studies (Hershkovitz et al. 1995; Rothschild and Rothschild 1994a,b, 1995a,b, 1996; Rothschild et al. 1995). Standards were derived from study of skeletal populations/samples from populations with known disease (Hershkovitz et al. 1995; Rothschild and Rothschild 1995a,b). These were validated by comparison with other populations known to have the same disease (Hershkovitz et al. 1995; Rothschild and Rothschild 1995a). The findings were identical for each disease, although different among the different diseases.

The patterns are described in detail below. While there were differences in which bones were affected, the nature of the periosteal reaction (with the exception of sabre shin remodeling) was independent of the specific treponematosis. As spiculated, laminated and thickened periosteal reaction were equally represented in all treponematoses (Hershkovitz et al. 1995; Rothschild and Rothschild 1995a), they are therefore not further considered in distinguishing among the treponematoses. There was also no difference in frequency of proximal and distal distribution of periosteal reaction on a given bone.

There has been a misconception that the nature of periosteal reaction in a given bone has any special qualities. With the exception of "coffee-table" tibia and degree of sabre shin remodeling, there is no difference among the treponematoses. It is solely the pattern of disease that is diagnostic (from a differential perspective). Detailed description of periosteal reaction has no differential significance. The issue is the population pattern, not the character of individual lesions.

As only three patterns of osseous involvement have been noted in over 100 subsequent populations studied (Rothschild and Rothschild 1996a,b; Rothschild et al. 1995), the technique would appear to have merit. The three patterns are indistinguishable from those reported in clinical studies (as discussed below individually for each treponematosis), with perhaps one exception. Skeletal examination revealed unilateral tibial involvement only in syphilis. Skeletons document the lifetime impact of disease. This contrasts with Hacketts 1946 clinical report, which assessed only one point in time, the time of examination, and not the natural history of the disease.

Skeletal examination provides a window to the lifetime impact of disease.

Time course perspectives of disease epidemiology are dependent on modern distribution, the historical record, and paleopathologic evidence (Cockburn 1963). Yaws has been perceived as a tropical disease, because of its current distribution. Yet tropical climate is only one of the differences between the regions where yaws exists today and the catchment areas of the other treponematoses. It is unclear why these "pockets" have persisted. Determinants of contemporary disease distribution are quite complex. While one could speculate that climate is determinate, that hypothesis has not been validated. The preconceived notion of climate determination does not seem to fit with the paleo-epidemiologic findings. It seems appropriate to explore other explanations for the current catchment areas, "islands" where non-venereal treponematoses still exist. Perhaps if we understand the timing of their eradication in other areas of the world, we may have a tool to understand their contemporary persistance patterns.

The standard population from which the diagnostic critera for yaws was a historically-diagnosed population (Kurashina 1992; Rothschild and Heathcote 1993; Stewart and Spoehr 1952). Yaws is a high population frequency (20-40% phenomenon, achieving full population penetrance by the second decade of life (Helfet 1944; Hunt and Johnson 1923; Moss and Biegelow 1922, Rothschild and Heathcote 1993, Rothschild and Rothschild 1994a,b, 1995a,b; Rothschild et al. 1995). It is polyostotic in distribution, frequently affecting all long bones and commonly affecting hand and foot bones. The average number of affected bone groups [e.g., tibia(e) = one bone group; fibula(e), a second, femora(e) a third bone group) affected is always three or greater (Rothschild and Heathcote 1993; Rothschild and Rothschild 1994a,b, 1995a,b; Rothschild et al. 1995). Symmetry is common with tibial involvement always (in all sites examined to date) bilateral. Sabre shin deformity, when present, has only limited surface remodeling. The residual surface periosteal reaction is always recognizable.

If yaws is considered a disorder of high population penetrance, the opposite is true for syphilis. Osseous lesions in syphilis afflict only 5-13% of the population (Freedman and Meschan 1943; Jostes and Roche 1939; Rothschild and Rothschild 1994a,b, 1995a,b; Rothschild et al. 1995) and even that is limited in its osseous impact. Tibial involvement is commonly (1/8-1/3) unilateral and the average number of bone groups affected is only 1-2.5 (Rothschild and Rothschild 1994a,b, 1995 a,b; Rothschild et al. 1995). While upper extremity, hand and foot involvement are rare, the degree of tibial involvement may so efface surface evidence of periosteal reaction that the resultant sabre shin tibia appears completely smooth.

An unanticipated observation has been the infrequency of osseous, or even dental evidence of congenital syphilis in the archaeologic record. Hypoplasia, reducing chances of tooth survival, probably explains the rarity of documentation of pathognomonic dental changes (El-Najjar et al. 1978; Mansilla and Pijoan, personal communication; Pinborg 1970; Sullivan 1986).

Observation of periosteal reaction in a subadult actually is much more common in yaws, and actually quite rare in syphilis (Rothschild and Rothschild 1994a,b, 1995 a,b, 1996a, 1997; Rothschild et al. 1995). Review of the clinical literature revealed that periosteal reaction from congenital syphilis is a short-lived event, usually with complete remodeling (such that it is no longer recognizable within 3 months of occurrence) (Levin 1970; McLean 1931). Thus, its archeological record rarity (less than 5% of subadults affected) is not surprising.

The third treponemal disease which affects bone is bejel. Bejel is a high population frequency disorder (Hershkovitz et al. 1995; Hudson 1958; Rothschild and Rothschild 1995b; Spirov 1991), affecting 20-40%, similar to that noted in yaws. In contrast to yaws, bejel is pauci-ostotic, with an average of two bone groups affected (Hershkovitz et al. 1995; Rothschild and Rothschild 1995b). Contrasted with syphilis, however, tibial involvement has been invariably (in all sites examined to date) bilateral in bejel and sabre shin remodeling is never so extensive as to remove all surface evidence of periosteal reaction.

The specificity of these findings for bejel, fully reproducible in disparate populations [e.g., Negev Bedouin, Sudanese Nubians, and the Kish site from Iraq, dated from 150-15,000 years before present (ybp)] (Hershkovitz et al. 1995; Rothschild and Rothschild 1995b, 1996b), validates the criteria allowing its recognition as distinct from syphilis and yaws. These findings are easily distinguished from other causes of periosteal reaction, even if one ignores their extreme rarity (as population phenomena) (Resnick and Niwayama 1988; Rothschild 1982; Rothschild and Martin 1993).

Methods

Six populations from west of the Sierra Cascades were compared with 5 sites east of the Cascades, one from the Aleutians, and one from Alaska. 83 adult individuals from Los Palmos (Baja peninsula, dated at 500-800 ybp), 88 from CCo295 (San Francisco Bay area, dated at 2250 ybp), 13 from Baldwin, 6 from Parizeau Point and 26 from Lachane (all from Prince Rupert Harbor, British Columbia, dated at 3200 ybp) and 27 from Crescent Beach (costal British Columbia, dated at 3000-4000 ybp) were examined (Table 1). Individuals examined from east of the Cascades included 89 from Pottery Mound (Colorado Plateau, New Mexico, dated at 550-650 ybp), 15 from Chinchera (Mogollan tradition, New Mexico, dated at 1300 ybp), 173 from Grasshopper (Mogollan tradition, dated at 750 ybp), 5 from White Dog Cave (Colo rado Plateau, New Mexico, dated at 2000 ybp), 8 from Olmos Dam (San Antonio, Texas, dated at 1100-1800 ybp) (Table 1). Additionally, 431 individuals from Point Hope (Alaska, 300-600 ybp) and 16 from Amaknak Island (Aleutians, 1900 ybp) were examined (Table 1).

Collections were chosen for examination, on the basis of adequacy of population size for recognition of pathology, preservation, and absence of obvious collection bias. Pueblo II and III samples were not included because that period is intermediate in time between those sites already documented as consistent. The purpose of the study was to identify the time of transition. Further samples prior to the documented transition time were problematic to locate. As they would not contribute to recognizing the transition time, their absence should not compromise interpretation. Skeletal remains were subjected to visual examination of all articular and cortical surfaces to identify all occurrences of bony alterations throughout each skeleton, specify the types of bony alterations at each occurrence, and map the distribution of occurrences in each skeleton. Metaphyseal and diaphyseal cortical and periosteal alterations were also assessed. All variation from normal smooth cortical surfaces was noted.

Treponemal disease was specifically recognized on the basis of periosteal reaction and osteitis (Freedman and Meschan 1943; Gann 1901; Goff 1967; Hunt and Johnson 1923; Jostes and Roche 1939; Moss and Bigelow 1922; Rothschild and Heathcote 1993; Rothschild and Rothschild 1994 1995a,b; Rothschild and Turnbull 1987; Rothschild et al. 1995).

Criteria for Recognition of Specific Treponematoses Syphilis, as a population phenomenon, produces recognizable periosteal reaction phenomenon in 5-14% of affected populations (Freedman and Meschan 1943; Jostes and Roche 1939; Rothschild and Rothschild 1994a,b, 1995a,b, 1996; Rothschild et al. 1995). By the time remodeling occurs sufficiently to produce a sabre shin phenomenon, the periosteal layer has usually been so re-worked as to remove all surface signs of periosteal reaction in most sabered tibiae (Rothschild and Rothschild 1994a,b, 1995a,b, 1996; Rothschild et al. 1995). Distribution of bone involvement is somewhat limited in syphilis. Tibial involvement may be unilateral and is typically associated with involvement of only one or two other bone groups (e.g., femora(e) is/are one bone group, fibula(e) another). While a single individual may have poly-ostotic disease, the average number of bone groups affected is usually 2, but always less than three (Rothschild and Rothschild 1994a,b, 1995a,b, 1996; Rothschild et al. 1995). Involvement of the radius and ulna is infrequent and more distal involvement (hand or foot) is quite rare. As osseous involvement in congenital syphilis remodels and disappears so quickly (Levin 1970; McLean 1931), juvenile skeletons often miss the narrow "window of opportunity" in which periosteal changes might be recognizable (Rothschild and Rothschild 1994a,b, 1995a,b, 1996; Rothschild et al. 1995). Thus, recognition of syphilitic osseous involvement in juveniles is infrequent. Recognition of fetal osseous involvement would be pathognomonic, but is observed too rarely for practical use in rejecting a syphilis diagnosis (in the absence of fetal involvement). Presentation of syphilitic hypoplastic incisors (Hutchinson) and molars (Mulberry) is apparently usually so poor (El-Najjar et al. 1978; Pinborg 1970; Sullivan 1986), as to preclude use of their absence as evidence of non-syphilitic origin of disease. Specificity of these dental phenomena has been claimed for syphilis, although we have been unable to find any studies documenting that specificity.

Yaws, as a population phenomenon, produces recognizable periosteal reaction phenomenon in 20-40% of affected populations (Hackett 1946; Rothschild and Heathcote 1993; Rothschild and Rothschild 1994a,b, 1995a,b, 1996; Rothschild et al. 1995). Sabre shin reaction is associated with less complete surface remodeling than that noted in syphilis. Surface evidence of periosteal reaction is always present in Yaws (Heathcote and Rothschild 1993; Rothschild and Rothschild 1994a,b, 1995a,b, 1996; Rothschild et al. 1995). Distribution of bone involvement is more poly-ostotic than in syphilis. Tibial involvement is invariably bilateral and the mean number of bone groups affected is always 3 or greater. Humeri, radii and ulnae are commonly affected, as are hands and feet. As yaws is a disease acquired in early childhood, it is not surprising that it is frequently (20%+) recognized in subadult skeletons.

Bejel, as a population phenomenon, produces recognizable periosteal reaction as frequently as Yaws (Hershkovitz et al. 1995; Rothschild and Rothschild 1995b). Similar to Yaws, sabre shin reaction is frequent and associated with the same limited surface remodeling. In contrast to what has been observed in syphilis (Rothschild and Roths child 1994a,b, 1995a,b, 1996; Rothschild et al. 1995), Bejel is a pauci-ostotic disease (Hershkovitz et al. 1995; Rothschild and Rothschild 1995b). Although a few individuals in a population may have poly-ostotic disease, the average number of affected bone groups is two. Tibial involvement is bilateral. Disease acquisition in early childhood results in frequent (at times exceeding 20%) affliction of subadult skeletons.

Differential Diagnosis

Other disorders associated with periosteal reaction do not occur with sufficient population frequency (Resnick and Niwayama 1988; Rothschild 1982; McCarty and Koopman 1993) to merit serious consideration. Further, they do not cause sabre shin reaction and have very different patterns of skeletal affliction. Hypertrophic osteoarthropathy is predominantly a disease of distal diaphyses (Resnick and Niwayama 1988; Rothschild 1982; Rothschild and Martin 1993). It typically spares the epiphyses and often affects ribs and clavicles. Thyroid acropachy spares the proximal appendicular skeleton, predominantly producing hand and foot bone periosteal reaction (Resnick and Niwayama 1988; Rothschild 1982; Rothschild and Yoon 1982). Infantile cortical hyperostosis is a disorder afflicting clavicles, scapulae, and ribs (Resnick and Niwayama 1988). Hypervitaminosis A is predominantly an enthesial disease, and fluorosis produces highly characteristic trabecular alterations (Resnick and Niwayama 1988; Rothschild and Martin 1993; Seawright and English 1967). Chronic osteomyelitis is usually a monostotic phenomenon, usually with draining sinuses (Resnick and Niwayama 1988). As it affects any bone, without specific tibial predilection, it can usually be distinguished from treponemal bone disease.

Results

The pattern of treponemal disease on the west coast of North America was markedly different from either the early (yaws) or later pattern (syphilis) noted east of the Cascades (Table 1). The west coastal region from Canada to Mexico is characterized by a high population frequency (21-35%) of treponemal disease (both in adults and subadults), pauci-ostotic (less than 2 bone groups affected) in distribution, and invariably bilateral in its tibial distribution (Table 1). Sabre shin remodeling was always incomplete, with surface evidence of periosteal reaction easily recognized. This pattern was characteristic for bejel, and quite distinct from that noted for yaws and syphilis (Hershkovitz et al. 1995; Rothschild and Heathcote 1993, Rothschild and Rothschild 1994a,b, 1995a,b; Rothschild et al. 1995).

Table 1. Characteristics of pre and Post - transitional Treponemal Disease East, West and North of the Sierra Cascades,
Contrasted with Documented Bejel, Syphilis and Yaws

Early populations, from east of the Cascades and the Aleutians (1300-2000 ybp) had a very different pattern (Table 1). Although population frequency (20-27%), in both adults and subadults) was as high as that noted west of the Cascades, the skeletal distribution was quite different. It was polyostotic, frequently affecting hands and feet. The average number of affected bone groups was 3.5-6. That pattern was characteristic of yaws and quite different from that noted for syphilis and bejel (Hershkovitz et al. 1995; Rothschild and Heathcote 1993; Rothschild and Rothschild 19941,b, 1995a,b; Rothschild et al. 1995).

Later populations east of the Cascades (550-750 ybp) and from Alaska (400-600 ypb) had an even more disparate pattern (Table 1). They had a low population frequency (<13%) of treponemal disease (limited to adults), pauci-ostotic (less than 2 bone groups affected) in distribution, with occasional unilateral tibial involvement, and sabre shin remodeling so complete, as to efface all surface indications of periosteal reaction. Hands and feet were spared. This pattern is characteristic for syphilis and quite different from that for yaws and bejel (Hershkovitz et al. 1995; Rothschild and Heathcote 1993; Rothschild and Rothschild 19941,b, 1995a,b; Rothschild et al. 1995). It is labeled as "post-transitional" in Table 1, as it represents the treponematosis syphilis, that apparently derived from yaws in North America (Rothschild and Rothschild 1995, 1996).

Discussion

While yaws was clearly the original treponemal disease to enter central North America through Beringia, and was clearly present east of the Cascades (Rothschild and Rothschild 19941,b, 1995a,b, 1996; Rothschild et al. 1995), the western coast of North America was just as clearly host to a very different treponemal disease (Table 1). The sites (east of the Cascades) chosen for comparison were those, for which repatriation has not yet precluded adequate evaluation. Bejel was clearly present west of the Cascades (Table 1). Replacement by syphilis was noted in the Los Palmos, Baja region only after 1700 (Molto 1995), significantly later than observation east of the Cascades, and corresponding to the timing of European contact.

Treponemal disease in the west coastal region was characterized by a high population (both adult and subadult) frequency (21-35%), pauci-ostotic disorder with bilateral tibial involvement and only incomplete sabre shin remodeling. Such is the pattern documented for bejel (Hershkovitz et al. 1995; Rothschild and Heathcote 1993, Rothschild and Rothschild 1994a,b, 1995a,b, 1996 b; Rothschild et al. 1995. This pattern was markedly different from either the early (yaws) or later pattern (syphilis) noted east of the Cascades (Table 1). Treponemal disease from east of the Cascades and in the Aleutians initially was a high (in both adults and subadults) frequency (20-27%), polyostotic phenomenon, frequently affecting hands and feet. That is the pattern of yaws (Rothschild and Heathcote 1993, Rothschild and Rothschild 1994a,b, 1995a,b; Rothschild et al. 1995). Treponemal disease in later populations from east of the Cascades and Alaska was less frequent in adults (<13%), tended to spare subadults, was pauci-ostotic, with occasional unilateral tibial involvement and complete sabre shin remodeling noted. That is the pattern of syphilis (Rothschild and Heathcote 1993, Rothschild and Rothschild 1994a,b, 1995a,b; Rothschild et al. 1995.

Bejel, yaws, and syphilis are clearly separate disorders, rather than simply different manifestations of one disease [manifesting differently (e.g., according to climate). Climate may be easy to disallow. The affected west coast sites reflect a wide range of latitude and environment, some of which are quite similar to those east of the Cascades.

There is, however, other even more impressive evidence that the diseases are separate entities: Differential sensitivity of hamster strains and rabbits to the different treponemes provides evidence that they are different species. The LSH hamster strain is highly susceptible to Treponema endemicum (bejel) and Treponema pertenue (yaws), but minimally so to Treponemal pallidum (syphilis) (Schell et al. 1981). Rabbits are highly susceptible to Treponema pallidum (syphilis), while minimally to Treponema pertenue (yaws) (Larsen et al. 1995). While some diseases can manifest differently in different environments, susceptibility to treponemal disease was species, not strain dependent.

Is there something unique about the west coast of North America and/or the populations it supported, or was it the populations themselves? Did they represent the product of a migration from a bejel endogenous region, as has been suggested for introduction of yaws to the rest of North America (Rothschild and Rothschild 1994a, 1995a,b; Rothschild et al. 1995)? Bejel appears to have spread across the Asiatic steps, in a direct line to Japan (Froment, 1995). Did this continue across the Pacific Ocean?

This study provides evidence that treponematoses were transported to the New World by way of at least two migrations, one bringing yaws; the other, bejel. It additionally provides further evidence that the treponematoses are separate diseases, and not simply climate induced variations "on a theme".

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