Pinta in Austria (or Cuba?)

Import of an Extinct Disease?

Ingrid Woltsche-Kahr, MD; Bruno Schmidt, PhD; Werner Aberer, MD; Elisabeth Aberer, MD

Background: Pinta, 1 of the 3 nonvenereal treponematoses, is supposed to be extinct in most areas in South and Central America, where it was once endemic. Only scattered foci may still remain in remote areas in the Brazilian rain forest, and the last case from Cuba was reported in 1975.

Observation: A native Austrian woman, who had lived for 7 years in Cuba and was married to a Cuban native, developed a singular psoriasiform plaque on her trunk and several brownish papulosquamous lesions on her palms and soles during a visit to her home in Austria. Positive serological findings for active syphilis and the detection of spirochetes in the trunk lesion indicated early secondary syphilis, but an extensive case history and the clinical appearance fulfilled all criteria for pinta.

Conclusion: The acquisition of a distinct clinical entity, pinta, in a country where it was formerly endemic but now is believed to be extinct raises the question of whether the disease is in fact extinct or whether syphilis and pinta are so similar that no definite distinction is possible in certain cases.

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The nonvenereal treponematoses yaws, endemic syphilis (bejel), and pinta are caused by an organism that is morphologically and antigenically identical to the causative agent of venereal syphilis, Treponema pallidum. However, they differ significantly from syphilis in their mode of transmission, epidemiology, and clinical manifestation.

Pinta, which is endemic only in the western hemisphere, is the most benign spirochetal disease, showing only skin manifestations and occasionally mild systemic symptoms (Table). The lesions of pinta are characterized by first hyperkeratotic and later dyschromic eruptions. Like venereal syphilis, pinta chronically relapses. The primary lesion appears as a small papule or erythematous macule 7 to 70 days after inoculation and extends within several weeks to an erythematous infiltrated plaque of up to 12.5 cm in diameter. Several months to years after appearance of the initial lesion, small scaly papules that enlarge to psoriasiform plaques, “pintids,” may develop. These highly infectious secondary lesions may be sparse or numerous and, after healing, leave areas of gray, brown, or slate blue hyperpigmentation or depigmentation. In the late stage of pinta III (months to 10 years after the appearance of pintids), lesions marked by vitililolike depigmentation are the leading feature. These lesions are not believed to be infectious. Histopathological investigations show moderate acanthosis, spongiosis, sometimes hyperkeratosis and hypergranulosis, and liquefaction generation of the basal layer in the epidermis, and a perivascular dermal infiltrate composed of lymphocytes, plasma cells, and neutrophils in the upper dermis. The main reservoir of pinta is young adults, who have chronic skin lesions. Direct skin-to-skin contact is the likely mechanism of transmission.

Several reports of cases of yaws and bejel imported to Europe as well as the ever-increasing ease of travel to the world’s remotest areas legitimate the demand to consider a diagnosis of nonvenereal treponematoses in certain clinical situations. Lack of evidence in a recent case that pinta is completely eradicated in areas of Central America where it was once endemic seemed to justify this diagnosis in a case that fulfilled all historical, clinical, and serological criteria of this disease.

REPORT OF A CASE

A 28-year-old white Austrian woman, who was married to a native Cuban and had
lived for the past 7 years in an urban area of Cuba, developed a solitary erythematosquamous plaque on the left side of her trunk. This lesion had lasted for almost 1 month when she was examined at our outpatient clinic. She did not remember any fever, tick bites, changes in oral or genital mucous membranes, joint pain, or lymphadenopathy. On questioning, she told us that her husband had observed a similar lesion in his right subaxillary region about 4 months before but that it had disappeared spontaneously; a routine test of her husband’s serum before minor surgery in Cuba had disclosed a positive serological test result for syphilis, and the husband had then been treated with penicillin. Both our patient and her spouse denied having had any extramarital relations in the recent past.

Clinical inspection of the patient disclosed a solitary erythematous, hyperkeratotic annular plaque of 2.5 cm in diameter in the left thoracolumbar area, resembling tinea corporis, a psoriatic plaque, granuloma annulare, or hyperkeratotic lichen planus (Figure 1 and Figure 2). Furthermore, several discrete erythemosquamous lesions on the palms and soles (Figure 3), each about 1 cm in diameter, were found; the patient had not been aware of them. The remaining skin as well as oral and genital mucous membranes showed no abnormalities; the lymph nodes were not enlarged, and no alopecia or any other signs of secondary syphilis could be detected.

### Differential Diagnosis of Pinta and Venereal Syphilis

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<th>Characteristics</th>
<th>Pinta</th>
<th>Venereal Syphilis</th>
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<tr>
<td>Transmission</td>
<td>&gt;95% Nonsexually (extragenital pattern)</td>
<td>&gt;95% Sexually; hematogenous; diaplacental (3-9 mo)</td>
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<td>Clinical manifestations</td>
<td>Stage I: Extragénital; erythematous infiltrated plaque</td>
<td>Genital/extragénital; indolent ulcer; regional lymphadenopathy</td>
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<td>Stage II: Pintids (small, scaly papules that enlarge to psoriasiform plaques with peripheral accentuation); no systemic symptoms</td>
<td>Fever, joint pain, lymphadenopathy, exanthema, condylomata lata, corona veneris, “plaques muqueuses,” angina specifica, alopecia areolaris specifica, and luetic leukoderma</td>
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<td></td>
<td>Stage III: Vitiligilike depigmentation; no involvement of other organs</td>
<td>No clinical manifestation</td>
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<td>Histopathology</td>
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<td>Epidemiology</td>
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<td>Therapy</td>
<td>≤2-y duration: penicillin G benzathine, 2.4 million U intramuscularly, 1 application; &gt;2-y duration: penicillin G benzathine, 2.4 million U intramuscularly, 3 times in 1 wk (in case of allergy to penicillin: doxycycline, erythromycin)</td>
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*From Koff and Rosen.† TPHA indicates Treponema pallidum hemagglutination; FTA-ABS, fluorescent treponemal antibody absorption test; and ELISA, enzyme-linked immunosorbent assay.

Figure 1. A single nummular plaque in the left thoracolumbar region.

Figure 2. Close-up view showing a hyperkeratotic annular plaque.
Wound serum was harvested from the hyperkeratotic plaque on the trunk after mechanical erosion of the plaque. On dark-field microscopy, multiple typical treponemes could be observed performing their characteristic bending and rotational movements.

Histopathological examination was done on a 4-mm punch biopsy specimen from the plaque after formalin fixation and paraffin embedding. Hematoxylin-eosin staining showed a psoriasislike picture with parakeratosis, acanthosis, spongiosis, and an infiltration of lymphocytes, eosinophils, and abundant neutrophils in the epidermis forming intraepidermal abscesses. There was edema of the papillary dermis and an obscured epidermal-dermal interface. In the dermis there was a patchy perivascular and interstitial infiltrate of lymphocytes, plasma cells, neutrophils, and some eosinophils. Blood vessels showed dilation but no substantial endothelial swelling. Several melanophages were present in the dermis. Furthermore, abundant treponemes were detectable in the epidermis by silver staining and by an immunoperoxidase staining on cryostat sections with the use of a polyclonal anti–Treponema pallidum antibody in a 1:200 dilution (Biodesign International, Kennebunk, Me) (Figure 4).

The following specific and nonspecific serological test results for Treponema were positive in our patient: T pallidum hemagglutination test, 1:5120; fluorescent treponemal antibody absorption test for IgM and IgG; Captia-Syphilis-M IgM enzyme-linked immunosorbent assay (Biotech; Gull Laboratory, Bad Homburg, Germany); and VDRL test, 1:32. Western blot analysis with a T pallidum IgM and IgG test kit (Marblot; VIRAMED, Munich, Germany) showed bands of 15.6, 17, 47, 61, and 83 kd in the IgG blot and 17, 37, 41, and 47 kd in the IgM blot, a pattern typical of Treponema (Figure 5). As controls, serum samples from 8 patients with syphilis and 1 healthy person were investigated. Serum from our patient (lane 1) demonstrated bands nearly identical to those of serum from patients with syphilis I and II.

The patient was treated with 2.4 million U of penicillin G benzathine according to the World Health Organization regimen for the treatment of early syphilis. The lesions disappeared within a few days, and markers of active syphilis (VDRL) became negative.

COMMENT

Nonvenereal treponematoses differ significantly from venereal syphilis in their mode of transmission, epidemiological characteristics, and clinical appearance, but not in their serological test results and treatment recommendations. The typical clinical manifestations of one of the nonvenereal treponematoses, pinta, however, closely resemble those of syphilis (Table). Treponema carateum, the cause of pinta, is morphologically and antigenically indistinguishable from T pallidum. In fact, despite advanced technical developments in the last decade, no differences between the subtypes of treponema can be detected by Western blot or Southern blot hybridization techniques. Treponema carateum cannot be cultured in vitro but can be transmitted to chimpanzees. Noordhoek et al4 reported that the T pallidum subspe-
cies pertenue and pallidum differed by a single nucleotide; these findings, however, could not be replicated by others. It has been suggested that the variations in clinical expression are entirely caused by divergent environmental conditions, such as temperature.5,6

Since 1975, pinta has seemed to be extinct worldwide except in a few scattered areas in the Brazilian rain forest.2,6,7 Yet, any textbook on dermatovenereology still describes this disease, and the medical student has to learn the differential diagnosis.

Our patient fulfills all the criteria of pinta stage I to II, with the typical primary skin lesion on the thoracolumbar area in the form of an annular hyperkeratotic plaque, and secondary papulosquamous lesions on the palms and soles, spirochetes within the lesion, and serological findings indicative of second-stage syphilis. In contrast, in syphilis II, symmetrical lesions are present, and on histological examination, swelling and proliferation of endothelial cells and no melanophages but epithelioid and giant cells are seen. Neutrophilic microabscesses are more often described in yaws. The patient, like her husband, obviously acquired her disease in an area in which pinta was once endemic. Yet, neither the patient nor her husband suffered from the typical signs of stage I syphilis. In such a case the closest diagnosis is pinta. We therefore conclude that if a disease such as pinta still does exist in Cuba, our patient suffered from that disease and did import it to Europe. If, however, the disease is extinct—except perhaps for a few scattered foci in the South American rainforest—why do we still burden our medical knowledge with such an outdated historical term?

Nonvenereal treponematoses constituted a major health problem for many underdeveloped countries before mass treatment programs were initiated in the 1950s.8,9 No further cases of pinta have been reported to the World Health Organization from Mexico or Colombia since 1979.10 The Centers for Disease Control and Prevention in Atlanta, Ga, and the Pan American Health Organization in Washington, DC, have received no reports of pinta from Cuba in the past 20 years (G. P. Schmid, MD, MSc, Centers for Disease Control and Prevention, e-mail communication, May 11, 1998). Moreover, according to G. Kouri, MD, of the Instituto Pedro Kouri Ministerio de Salud Publica in Havana, Cuba, pinta has not been diagnosed in Cuba for many years (e-mail communication, May 11, 1998).

There is, however, no proof of pinta’s extinction, since patients with pinta may remain contagious for prolonged periods, may harbor subclinical disease, and do not develop lifelong immunity. Because the ease of worldwide travel can aid in their dissemination, nonvenereal treponematoses should therefore still be considered in differential diagnoses.

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REFERENCES