LUCIO'S SPOTTED LEPROSY (DIFFUSE LEPROMATOUS LEPROSY OF MEXICO) REPORT OF A CASE IN HAWAII

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The concept of "pure diffuse" lepromatous leprosy, a clinical entity familiar to Mexican physicians for nearly a century but relatively unfamiliar in other parts of the world, has been thoroughly clarified in a series of papers by Latapí and Chevez Zamora, of Mexico City (2-5). So distinct an entity is this form of leprosy that it seems unwise any longer to use the term employed by Lucio and Alvarado (6), who originally described it as "St. Lazarus' Disease," or, as it was colloquially known in Mexico up to at least 1885 (1), *lepra lazarina,* for the term "lazarine" has repeatedly been employed, by Pardo-Castelló (7, 8) and others, to designate any cases of leprosy characterized by the formation of bullae and ulcers—a phenomenon not confined to diffuse lepromatous leprosy of Mexico, or, as Wade (10) would call it, Lucio leprosy.

It is quite apparent from the literature, from personal communications from students of leprosy in other countries, and from a brief visit to parts of Central America made by both of us in 1948, that this form of leprosy is rare outside of central Mexico and Costa Rica (9). Neither of us had ever, to our knowledge, seen a case in Hawaii. In 1949, however, in the course of examining a group of candidates for temporary release from Kalaupapa Settlement, we were struck by the presence of conspicious depigmented atrophic scars, from 1 to 4 cm. in diameter and of irregular outline, on the legs and to a lesser extent on the arms of a young female patient. Inquiry revealed that she had never manifested any nodular lesions during the course of her illness, and that the scars we noticed were the sequelae of a recurring macular and vesicular eruption present almost from the beginning of her illness. Further investigation suggested strongly that she represented a healed case of diffuse lepromatous leprosy—and, since she was native-born, an indigenous one; and for that reason the case is reported here.

CASE REPORT

The patient, O. R., an 18-year-old Portuguese girl, was admitted to The Queen's Hospital in Honolulu on August 16, 1934, because of possible recurrent appendicitis and acute tonsillitis. She gave a history of good health, aside from recurrent episodes of lower abdominal pain, until about nine months prior to admission. Since that time she had had repeated crops of flaccid, nonitching vesicles, sometimes only one or two at a time, appearing on the arms and legs.

On examination her skin was noted to be generally of a dusky, slightly cyanotic hue, except for an indistinct pink flush over the lower portion of the face. The hemogram was normal except for mild leucocytosis, which disappeared soon after tonsillectomy. Wassermann and Kahn tests of the blood both gave strongly positive reactions on two occasions, with "over 400" and 800 Kahn units being found in the two specimens examined. A single forced sexual exposure was said to have occurred three months previously, but the introitus admitted only a fingertip owing to a small hymeneal ring. The duskiness of her skin aroused the suspicion of leprosy, and thermal and tactile anesthesia of the extremities was found. She was discharged from the hospital on August 31.

On September 21 she was admitted to the Kalihi Leprosy Receiving Hospital in Honolulu. There the duration of the skin lesions was given as one year. No known contact with leprosy was acknowledged. The patient stated that she had been born in Kalaheo, on the Island of Kauai. Her parents were living and well; both were of Portuguese ancestry. A brother 16, and two sisters, 15 and 22, were said to be nonleprous.

Examination by Dr. James T. Wayson disclosed a pink flush across the cheeks and nose, "bat-wing" in shape; cyanosis and atrophy of the hands, with early contracture of both fifth fingers; swelling and cyanosis of the legs; and "scattered pustules," not further described, over the body and hands. Leprosy bacilli were demonstrated in skin of the right buttock and right ear lobe, by Wade's scraped incision method.

The observations of Dr. R. K. Maddock, P. A. Surgeon, U. S. P. H. S., of the Leprosy Investigation Station at Kalihi Hospital, were as follows:

"The lesions on this patient are for the most part very vague and indistinct. Involving most of the face below the eyes is a large pink macule which extends laterally and also involves the left ear and the lower half of the right ear. This macule also extends a short way down the neck. On both upper extremities, chest, back and buttocks are numerous very vague hypopigmented macules of various sizes. On the hands, forearms, legs and feet there are small vesicular lesions and numerous small hypopigmented scars resulting from these lesions. The four extremities are cyanotic.

"There is a slight droop to the right lower lid. The right hand is slightly weaker than the left (patient is right handed) and the thenar and hypothenar eminences are very slightly atrophied. The skin of the right palm is drier than the left. The right ulnar nerve was slightly tender, no nerve enlargement was found. Tactile anesthesia was found along the inner border of the right hand, over the left elbow, in the center of the large macule on the right buttock, and over the right tendo Achilles. Thermal dysesthesia was found over the right forearm extending down over the inner surface of the posterior aspect of the right hand, on the inner surface of the left forearm, both legs, and the left foot.

"Diagnosis—Cutaneous 1, Neural 1." [1]

The rest of the physical examination disclosed nothing remarkable. Photographs taken at this time (Figs. 1-2) show the absence of circumscribed nodules, the early contracture of the fingers, a number of dark, irregularly oval macules on the legs. The diffuse dusky suffusion noted at the Queen's Hospital evidently persisted, for in a photograph taken a year later it was manifest by contrast with the blanching caused. by pressure of the physician's hand. (Fig. 3.)

Wassermann and Kahn reactions of the blood serum were positive, the latter in a titer of 800 units, on admission, six months later, and again after two and one-half years. Four tuberculin tests over a two-year period were all negative. Tissue fluid obtained by the scraped incision procedure was positive in four different locations on three occasions in the two and one-half year period. Neither a lepromin test nor a biopsy was performed during this admission.

Various treatments were employed during her hospital stay, viz., "bismuth" (kind and route not stated), chaulmoogra ethyl esters, bacterial antigen, and "contrast hydro baths."

A summary of her progress, recorded in January 1947, is as follows: "Very little change in her condition during the year with the exception of marked increase in her herpetiform lesions, which may or may not be leprous, in June." These herpetiform lesions are elsewhere described as vesicular. No mention is made, oddly enough, of the fact that they were leaving scars when they healed. The patient was transferred to Kalaupapa Settlement in June 1937, for custodial care.

The record made on admission to the Settlement, dated August 7, 1937, is as follows: "On admission there were numerous pustular blebs about 1 cm. in diameter on the anterior and medial surfaces of both thighs, with a few scattered on the legs and arms. During the first two weeks here, these increased in number and there were systemic evidences of toxic absorption. A regime of prompt opening of blebs was inaugurated. Neoarsphenamine was given weekly, 0.3 grams, beginning July 19th, with improvement apparent within one week...."

In September 1946 the patient was examined as a candidate for promin therapy. Her general health was good. There were no nodules or macules, but there was diffuse thickening of the skin, notably of the forearms and brow and chin, and numerous thin atrophic scars chiefly over the extremities. There was generalized thermal anesthesia, sparing only the face and an area above the symphisis pubis. Interosseous and thenar and hypothenar atrophy was noted. No enlarged nerves were felt, and no paralysis was found. The patient volunteered the information that her legs did not sweat. The eyebrows and eyelashes were absent. The eyes were normal

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[1] This form of diagnosis was evidently based on a strictly topical or anatomical interpretation of the Leonard Wood Memorial Conference classification which had been adopted at Manila three years earlier, in 1931. Virtually every patient at Kalihi Hospital was diagnosed as both "cutaneous" and "neural," (i.e., mixed), from about 1931 until about 1941. It seems significant that a survey of positive serologic tests for syphilis made by Badger during this period disclosed exactly the same incidence of such reactions—about 30 per cent—in primarily "cutaneous" and primarily "neural" cases.

except for slight irregularity of the right pupil; vision was 20/20 in each eye. The nasal septum had been destroyed. Nothing remarkable about the mouth and throat. Liver palpable at the costal margin.

From October 21, 1946, to July 1, 1948, the patient received promin, 5 grams intravenously daily, 6 days per week, 1 week off and 2 weeks on, to a total amount of 1,728 grams. Eight specimens of tissue fluid taken by the scraped incision method between January and July, 1948, showed no bacilli. The patient was granted "temporary release" from the Settlement on August 13, 1948, at which time she showed only nummular scars, chiefly on the extremities, extensive anesthesia, and madarosis.

Biopsy of the skin of the right forearm was reported at this time as follows: "The epidermis is normal. There are numerous streaks and foci of infiltrate scattered throughout the corium and upper hypoderm, made up chiefly of lymphocytes but also containing a few foam cells. There is definite involvement of cutaneous appendages, vessels and nerve trunks. Acid-fast bacilli 1-plus. Diagnosis: Lepromatous leprosy."

Biopsy of the region of the right knee at this time was reported as follows: "The epidermis shows rather marked acanthosis and hyperkeratosis. The granular layer is prominent and the rete pegs are blunted, or missing entirely. In the upper corium very small banal-appearing foci of infiltrate are present, arranged around small vessels. Some of the latter are dilated. The lower corium shows nothing remarkable. These histologic changes are in no way suggestive of leprosy, and no bacilli could be found after acid-fast staining."

COMMENT

It seems probable that this patient had had the Lucio type of leprosy, despite the omission of a lepromin test, the lack of detailed description of the course pursued by her vesicular lesions, the lack of a biopsy prior to the thirteenth year of her illness, and the unusually long and mild course of the disease prior to the beginning of treatment. The diffuse skin involvement, without circumscribed lesions except for malar flushing; the symmetric and widespreead but slight nerve involvement; the complete loss of eyebrows and eyelashes; the destruction of the nasal septum without involvement of the eyes or larynx; the multiple recurrent vesicular lesions on the extremities, with atrophic scarring (Fig. 4); the positive serologic tests for syphilis; and the lepromatous histologic structure when a biopsy was finally performed, are all consistent with this diagnosis.

SUMMARY AND CONCLUSIONS

A case of Lucio's spotted leprosy (diffuse lepromatous leprosy of Mexico) occurring in a Hawaiian-born Portuguese girl and healing under promin therapy, is reported.

So far as is known, this is the only case of this type of leprosy that has been observed in Hawaii.

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DESCRIPTION OF PLATE

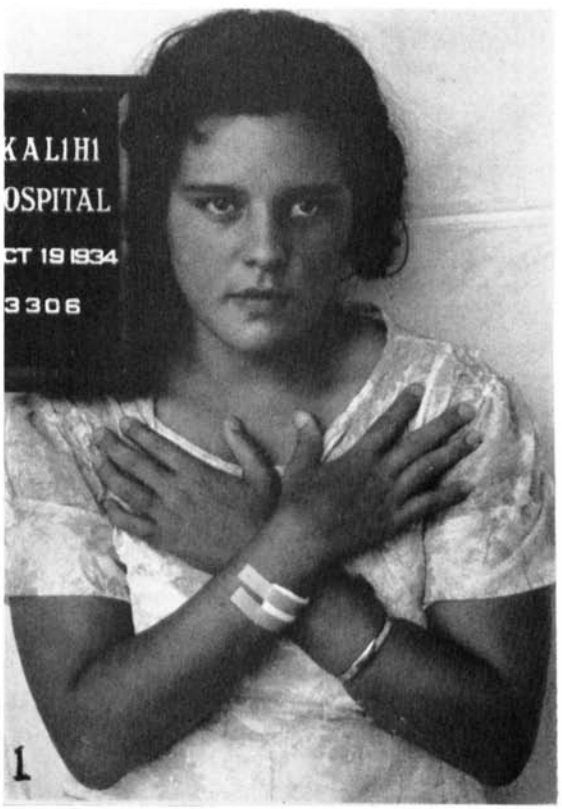
PLATE 3.

FIG. 1. Photograph of patient taken in the first year of illness, shortly after admission to the Kalihi Hospital in 1934, showing diffuse infiltration of the skin and minimal contraction of the fifth fingers of both hands.

FIG. 2. Active "Lucio spots" on the legs, and scars of previous lesions of that kind. Taken at the same time as Fig. 1.

FIG. 3. Illustrating the persistent diffuse suffusion and cyanosis of the skin of the back, in contrast with the blanched area produced by pressure of the examining physician's hand. Taken a year later than the preceeding photographs.

FIG. 4. Scars on the legs, the sequelae of the "Lucio spots," as seen in 1949.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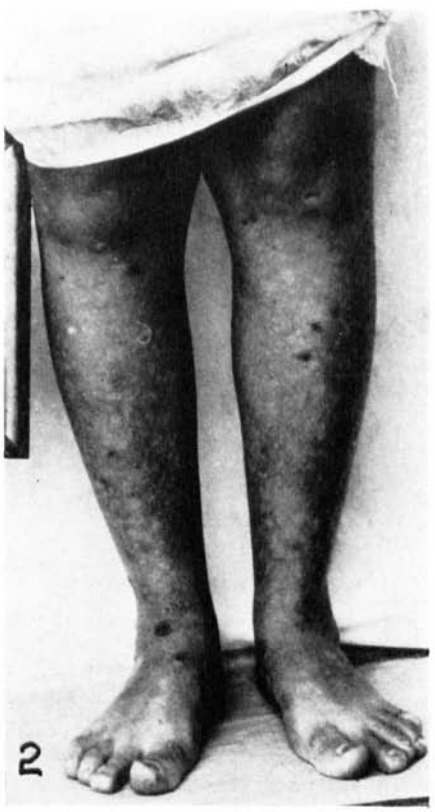
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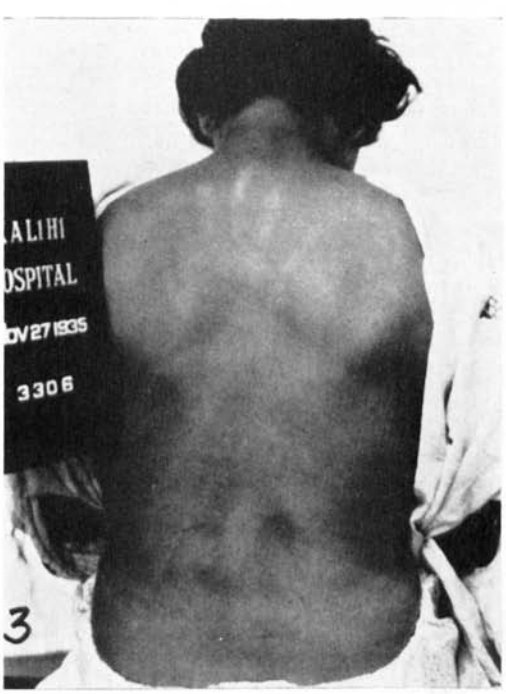
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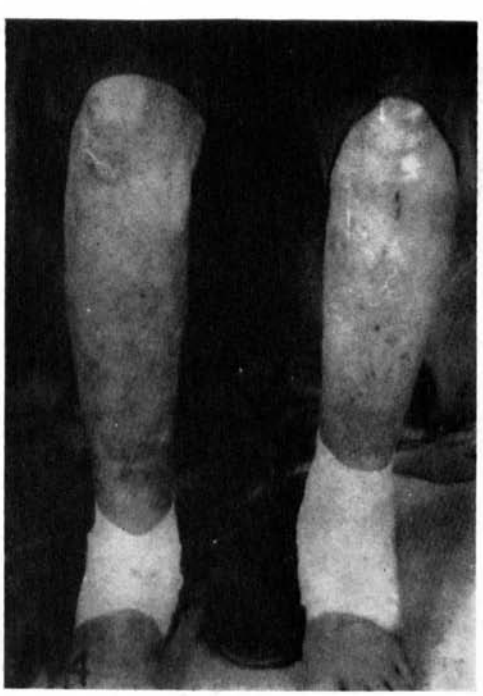

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