

EXTENDED ABSTRACT

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TWO SIMULTANEOUS CASES OF LEPROSY DEVELOPING IN TATTOOS¹

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There is still uncertainty concerning the transmission of leprosy, though it is one of the oldest known diseases. Close and prolonged association with leprosy persons has always been recognized as dangerous, yet minimal precautions seem adequate to prevent infection. Various theories of transmission have enjoyed popularity, including those of fish as intermediate hosts and insects as vectors, infection by sexual intercourse and by wearing contaminated clothing, and inoculation by person to person vaccination; also that leprosy is acquired through the nasal mucous membrane.

During the past century it has been a debated question whether or not infection can be induced by inoculation into the skin. Klingmüller (1) reviewed well the evidence for and against experimental inoculation. Jeanselme (2) cited the negative experiments of Danielssen and Boeck (3), Profeta (4) and Mouritz (5), and concluded that there was no adequate proof of transmission by inoculation. Rogers and Muir (6) accepted transmission by inoculation. They questioned the case of Keanu who was inoculated by Arning (7) and subsequently developed leprosy, but accepted as valid among others that of Marchoux (8) who, while operating on a leprosy patient, pricked the finger of his assistant who developed leprosy after several years. De

¹ The original article was published in the *American Journal of Pathology* 23 (1947) 805-817, which periodical by established policy does not permit identical reprinting. Republication in modified form was not agreed to by the authors on the ground that it might cause confusion in the literature. The report being regarded as of too much importance to be dismissed with an ordinary abstract this extended one, made with more liberties in arrangement than in phraseology, is run in this way. The plates are from the original cuts, loaned for the purpose.—EDITOR.

Langen (9) reported an accidental inoculation by a physician using for a hypodermic injection a syringe which had been used on a person with leprosy. Lagoudaky (10) was repeatedly injected with blood and developed cutaneous lesions.

Negative results are difficult to evaluate because of the long incubation period and the possibility of subclinical or dormant infections. Different persons, age groups, races and the sexes vary widely in susceptibility. Since leprosy tends to localize in scars and tattoos and to be activated by trauma, it is difficult to prove that an infection following inoculation actually resulted from it.

REPORT OF CASES

The remarkable coincidences which occurred in the two cases here presented offer very strong evidence favoring transmission by inoculation. The men, friends and residents of the same town in civilian life, were in the same unit of the U. S. Marine Corps; they were tattooed together in Melbourne, Australia by the same man on the same day in June 1943, they and the tattooer being inebriated; and they developed maculo-anesthetic leprosy in the tattoos about 2½ years later. A third man tattooed at the same place, but at a different time, shows no evidence of leprosy.

Case 1.—A white male, 24 years old, with nothing significant in the family or personal history except for malaria in 1942 and some weakness of the left arm ascribed to accidental chipping of the left scapula in 1945, complained chiefly of increased pigmentation and numbness of a tattooed area on the extensor surface of the lower left forearm. Nothing untoward had taken place after the tattooing in 1943 until March or April 1946, when the area and a zone about 1.5 cm. wide around it were noticed to have become pale red (Fig. 1) and to be insensitive to light touch and pain. A deliberate burn with a lighted cigarette caused no sensation.

On physical examination, the distal one of the two tattoos on the extensor surface on the left forearm (i.e., the one made in Melbourne) presented a uniform, pale, fawn-colored appearance over its entire tattooed area and a narrow zone around it. In this discolored area, the total diameter of which was 9 cm., there was loss of sensation to pain and light touch. The other tattoo showed no evidence of disease. A slight congestion of the nasal mucous membrane was noted, and a nontender swelling, about 0.5 x 4 cm., on the lateral surface of the juncture of the middle and lower thirds of the left upper arm. The axillary and inguinal lymph nodes were small, and there was no palpable evidence of lesions of the nerve trunks.

Routine laboratory examinations (hemoglobin, red and white cell counts, and urinalysis) gave normal results. The Kahn reaction was negative.

A biopsy specimen was removed (November 2, 1946) from the pigmented area outside the tattoo, without anesthetic and with no discomfort to the patient. Sections showed tuberculoid

tissue reaction, and occasional acid-fast bacilli were found, averaging about four per section. Smears from the nasal mucosa were negative. Intradermal injection of 0.1 cc. of O.T. (1:10,000) caused a positive reaction. In view of the history of microscopic findings, a provisional diagnosis of cutaneous tuberculoid leprosy was made.²

Case 2.—A white male, 25 years old, with nothing pertinent in his history except for malaria in 1942 and occasional malarial chills since then, complained chiefly of numbness and pigmentation of the tattooed area on the flexor surface of the left forearm, made in 1943. About January 1946, he noticed that that area and a zone about 1.5 cm. wide about it was becoming dusky red and numb, since when the color had gradually darkened (Fig. 2). Two new areas had appeared on the extensor surface of the upper left arm 7.5 cm. above the elbow, and had become confluent (Fig. 3); they were dark violaceous in color and numb, with no elevation of the surface.

On physical examination the skin of the entire tattooed area and a zone 1.5 cm. wide about it showed a violaceous discoloration (Fig. 2), with loss of sensation of pain and light touch. The two coalescent lesions above the elbow made an hour-glass-shaped area about 2.5 by 4 cm. (Fig. 3), with the same color and loss of sensation as the tattooed area. There was no palpable abnormality of the nerve trunk. In the left lumbar region at the waistline there was found a violaceous, flat lesion, 1 cm. in diameter, which had normal sensation and was not definitely related to the present illness. The axillary and inguinal lymph nodes were normal in size.

Routine laboratory examinations gave normal findings, including a negative Kahn reaction.

A biopsy specimen, removed (November 11, 1946) from the pigmented area outside the tattoo, without an anesthetic, was divided into two parts. One part was sent in saline solution to the Michigan Department of Health, which reported no acid-fast bacilli found in direct smears or in culture, and no evidence of tuberculosis after seven weeks in two guinea-pigs inoculated with the tissue. Sections of the other part presented the same microscopic appearance as in Case 1. Acid-fast bacilli were also demonstrated, but they were fewer, averaging only about one per section. Nasal smears were negative. The result of an intradermal inoculation with 0.1 cc. of O.T. (1:10,000) was negative. The tentative diagnosis was cutaneous tuberculoid leprosy.

DISCUSSION

Clinically, the lesions in these two cases could well be tuberculous. Microscopically, the presence of epithelioid tubercles with

² This first case was informally described to Dr. Claude Behn, of Detroit, who without seeing the patient made the original suggestion of leprosy as a probable diagnosis.

Langhans' giant cells, lymphocytic and plasma cell infiltration and occasional acid-fast bacilli is as characteristic of tuberculosis as of leprosy. The history, however, is more suggestive of the latter. The loss of sensation to pain and light touch, the negative tuberculin test, cultures and guinea-pig inoculation in Case 2, the presence of vacuolated cells, and the positive diagnosis by the U. S. Marine Hospital at Carville,³ established these cases as maculo-anesthetic or tuberculoid leprosy.

The long incubation period suggests resistance to the disease. The extensive traumatization of the skin incident to tattooing might favor its development. As noted by other observers, cinnabar (red mercuric sulphide) in the tattoos, which discourages spirochetal activity in syphilis of the skin, has no apparent effect on the bacillus of leprosy.

A single description suffices for both biopsy specimens. Each was taken from near the edge of the pigmented lesion, outside the tattoo. The microscopic picture (Figs. 4 to 8) so closely resembled tuberculosis of the skin as to be almost, if not quite, indistinguishable. The characteristic lesion consisted of a center of epithelioid cells with a rim of lymphocytes, a few plasma cells, and even fewer polymorphonuclear leucocytes, with occasional eosinophiles. These nodules were largely avascular. Many of them suggested Boeck's sarcoid, but occasional large ones showed an appreciable degree of caseous necrosis. The Langhans' giant cells were of all sizes, and in appearance and distribution they did not differ from those of tuberculosis; they occurred both in the epithelioid foci and scattered through the areas of lymphocytic infiltration.

These tuberculoid foci were present throughout the specimen, in both corium and subcutaneous fat, the process apparently extending beyond the depth of the excised tissue. The largest were in the deep layer of the corium, with smaller ones and extensive lymphocytic infiltration in the superficial layer. The foci in the subcutis were more discrete than those nearer the epithelial surface, and were smaller than those in the deep corium. There were no characteristic leprous foam cells, but there were occasional suggestive vacuolated cells.

The epidermis was irregularly atrophic, and there was flattening and partial loss of the dermal papillae, with lympho-

³ Unstained sections from both cases were submitted to the U. S. Public Health Service, and the Carville, Louisiana, leprosarium reported unequivocally "typical tuberculoid leprosy," though acid-fast bacilli were not demonstrated.

cytic invasion of the basal and prickle cell layers in some places. The hair follicles showed lymphocytic infiltration, the picture entirely compatible with the loss of hair characteristic of lepromas. There was a granulomatous involvement of the sweat glands, some of which had almost completely disappeared. The cutaneous nerves were involved, but not more so than other structures; there was no particular evidence of extension by way of the nerves. There was considerable involvement of the small vessels, but they did not show the swelling and proliferation of the endothelium found in syphilis.

After Ziehl-Neelsen staining of the sections, occasional acid-fast bacilli were found in the first case and rare ones in the second. They were found most often in or about the largest foci in the deep corium. Usually they occurred in pairs, or two single organisms in one oil-immersion field. They showed no significant variation from tubercle bacilli in either morphologic or staining characteristics.

SUMMARY

Two men from the same community, while serving in the U. S. Marine Corps, were tattooed by the same man on the same day in June 1943, in Melbourne, Australia. They both developed maculo-anesthetic or tuberculoid leprosy in their tattoos during the first half of 1946. One man had multiple tattoos but developed leprosy only in the one made in Melbourne the day when his friend was tattooed. A third Marine, tattooed at the same place but not on the same day, has shown no evidence of leprosy. These two cases provide strong evidence for the spread of leprosy by inoculation.

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DESCRIPTION OF PLATES

PLATE 33

FIG. 1. CASE 1. The distal tattoo, made in Melbourne, is the only one with pigmentation and anesthesia. The pigmentation is so light that it does not appear in the photograph. The dark spot by the star indicates the site where tissue was excised for biopsy.

FIG. 2. CASE 2. Tattoo on the left forearm showing the extent of the pigmentation. The skin suture is still present where tissue was taken for biopsy.

FIG. 3. CASE 2. Secondary lesions on the extensor surface of the left upper arm.

FIG. 4. CASE 2. Large Langhans' giant cell and a small epithelioid tubercle with lymphocytic infiltration about them. Hematoxylin and eosin stain. X 500.

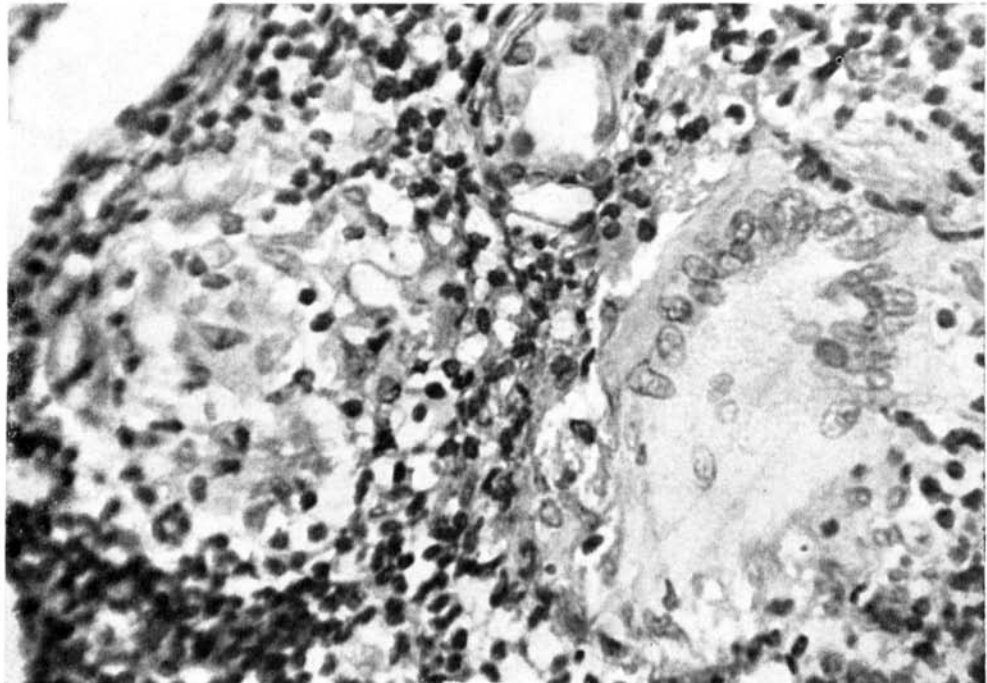


PLATE 33

PLATE 34

FIG. 5. CASE 1. Lepromatous reaction about a sweat gland. Hematoxylin and eosin stain. X 500.

FIG. 6. CASE 1. Lymphocytic infiltration of the epithelium and corium with a well formed Langhans' giant cell. Hematoxylin and eosin stain. X 500.

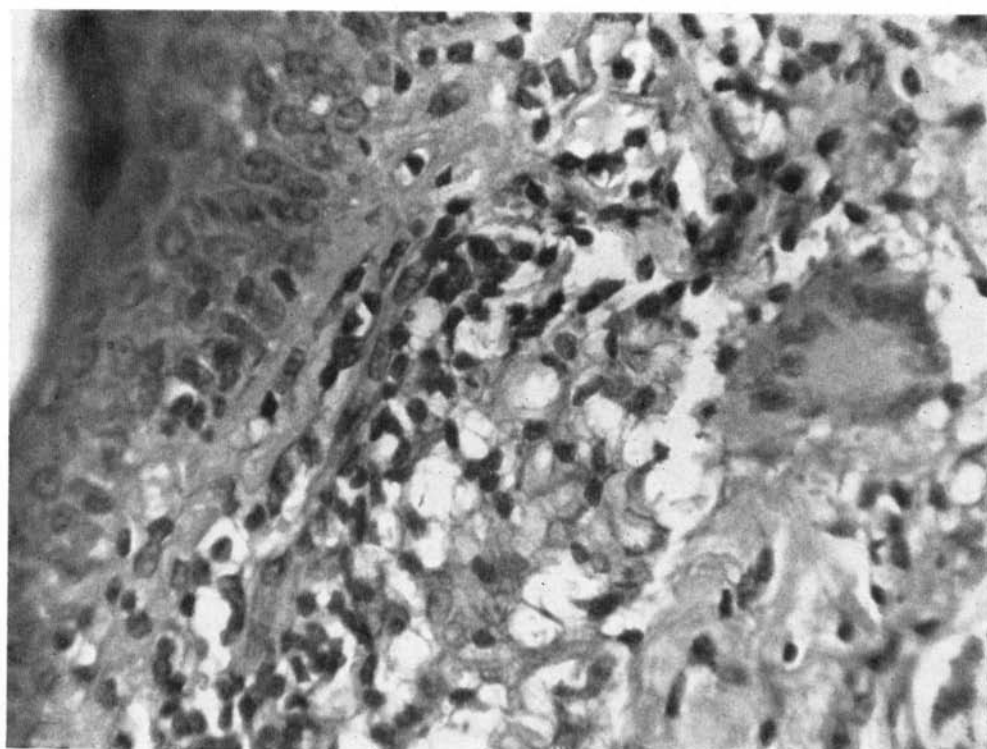
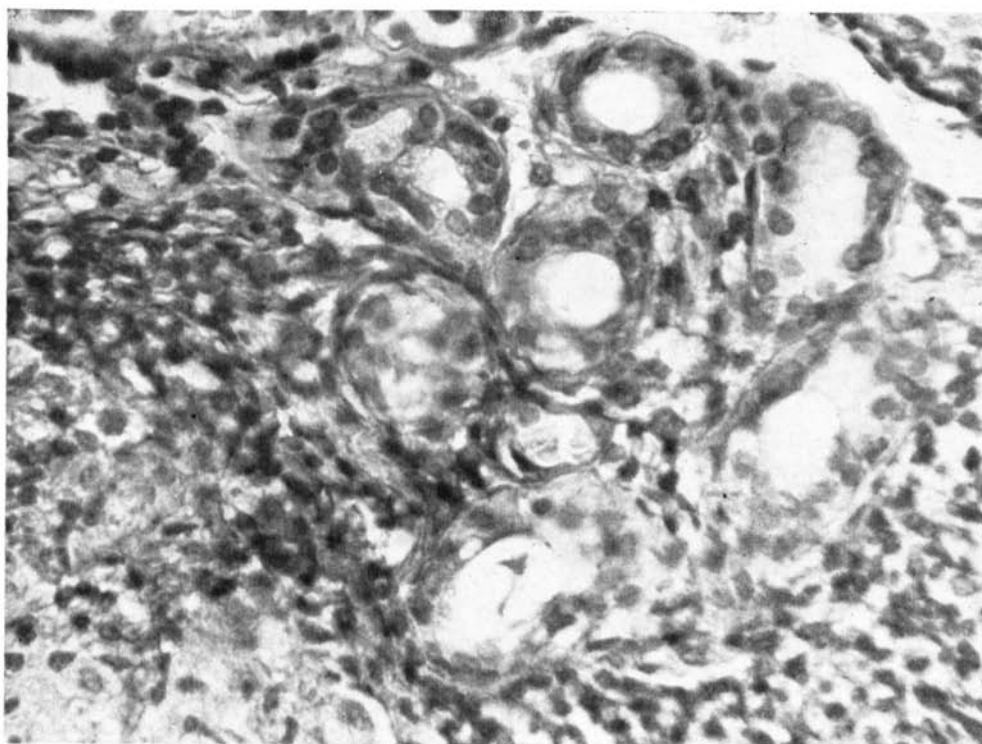


PLATE 35

FIG. 7. CASE 1. Lymphocytic infiltration in and about a hair follicle. Hematoxylin and eosin stain. X 500.

FIG. 8. CASE 2. Large epithelioid tubercle with caseous necrosis in the center. This lesion is microscopically indistinguishable from tuberculosis. Hematoxylin and eosin stain. X 180.

