TWO SIMULTANEOUS CASES OF LEPROSY DEVELOPING IN TATTOOS

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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 leprous 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. Jeanelme (2) cited the negative experiments of Danielsen and Boeck (3), Profeta (4) and Mouriz (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 Keen 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 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

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1 The original article was published in the American Journal of Pathology 23 (1927), but, 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 phrasing, is run in this way. The plates are from the original cuts, boxed for the purpose. — Ed. T. O.T.
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 I.—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 slipping of the left scapula in 1943, 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 and to be insensitive to light touch and pain. A deluge of burn with a lighted cigarette caused no sensation.

On physical examination, the distal one of the two tattoos on the extensor surface of the left forearm (i.e., the one made in Melbourne) presented a uniform, pale, brown-coloured appearance over its entire tattooed area and a narrow zone around it. In this discoloured 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 mottled swell ing, about 0.3 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 loss of the nerve trunks.

Routine laboratory examination (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 anesthesia and with no discomfort to the patient. Section 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 the microscopic findings, a provisional diagnosis of cutaneous tuberculoid leprosy was made. 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.
Case 2.—A white male, 43 years old, with nothing pertinent in his
history except for malaria in 1941 and occasional chills since then.
(Comment) The first of randomness and pigmentation of the tattooed area on
the inner surface of the left forearm, made in 1941. About January 1945
he noticed that this area and a zone about 1.5 cm. wide about it was
becoming dusky red and numb, since when the color had gradually
declined. Two new areas had appeared on the external surfaces of the
upper left arm 7.5 cm. above the elbow, and had become confluent; 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 with loss of
sensation of pain and light touch. The two confluent lesions above the
elbow were an hour-glass shaped area about 2.5 by 1 cm. with the same
color and loss of sensation as the tattooed area. There was no palpable
abnormality of the nerves.

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 tuber-
culosus. Microscopically, the presence of epitheloid tubercles
with 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, 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 tattoo, 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 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 leukocytes, with occasional eosinophiles. These nodules were largely avascular. Many of them suggested Boeck's sarcoma, 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.

Fig. 1. Case 1. The distal tattoo, made in Melbourne, is the only one with pigmentation and anestheisia. 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 sutures are still present where tissue was taken for biopsy.

Fig. 3. Case 3. Secondary lesions on the extensor surface of the left upper arm.
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 lymphocytic 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.
REFERENCES

7. Arning, E. Appendix to the Report on Leprosy of the president of the Board of Health to the Legislative Assembly of 1886. P. C. Advertiser Steam Print, Honolulu, 1886, Appendix XXXVII-IV.