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LEPROSY: INITIAL LESION AND SURGICAL CURE A CASE REPORT*

By

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Under this title I presented these two case reports in The Proceedings of the Staff Meetings of The Clinic in August 1937. Dr. N. E. Wayson had, however, previously submitted a report on the same cases which was published in The Archives of Dermatology and Syphilology, Vol. 36, No. 6, December 1937. It was from him that I obtained most of my information on one of the cases. I now try to bring them up to date, since both survive.

The first case occurred in a man who has been under the observation and care of so many of us in varied capacities, that I feared his case might, through misunderstanding, never be recorded and it is too important a one to be lost. Drs. Grover Batten, N. E. Wayson, and G. B. Tuttle have made most of the observations and have given most of the treatment but others of us have been associated. There was the danger of too many cooks spoiling the broth, by neglecting to record this history. Our patient was born in 1873 (he is now 71) and was raised and educated in France. His previous history is irrelevant but it is reasonable to assume that in France he was at no time in even casual contact with cases of leprosy; surely not a prolonged nor intimate contact. He himself is of that opinion.

En route to Hawaii he made a short stop in Tahiti, the first place he might probably have come in contact with leprosy. He stated, however, that he knew of no personal contact with lepers during his sojourn there. In April 1925 he went to Kalaupapa, Hawaii's leper settlement, for a period of about 6 months and returned to Honolulu. He subsequently returned to Kalaupapa in April 1927 and has remained there almost continuously since that date. At Kalaupapa he was, not by necessity but by his own volition, in intimate contact with the patients, though not nursing nor dressing them. He was an indefatigable worker and night-long vigils at the bedside of patients were not unusual; to the contrary, these vigils were so frequent and

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long that they were detrimental to his general health. He was a consummate musician and a scholarly student, reading being his chief diversion.

He spent many hours in study. It had become his habit to hold a book in one hand and abstractedly scratch his forehead with the nails of the other. This habit had become noticeable to his fellows.

In that area of his forehead which it had become his habit to scratch, he noticed, early in September 1932, a small, solitary pink macule. That was about 7 years after his first personal contact with Hansen's disease. It grew slowly larger and became slightly elevated. Shortly thereafter, in a contiguous area of the semi-bald forehead, two smaller but similar maculo-papular lesions made their appearance.

Six months after the appearance of the first lesion (*i.e.* on March 28, 1933) he came to Honolulu and presented himself to Dr. N. E. Wayson, senior leprologist at Kalihi Hospital and Receiving Station, for examination and it is to Dr. Wayson that I am indebted for the following record of the examination at that time.

"On examination there is an elevated, flat, reddish-pink lesion approximately 1½ x 2 cm. and two flat papules of similar appearance and approximately ½ cm. in diameter. These lesions are located on the forehead, posterior to a line which was the probable original hairline. (The patient is partially bald on the upper part of the scalp and there is no definite hairline over the forehead.) The infiltration of these lesions seems to involve the entire thickness of the skin.

"A superficial examination was made for sensory disturbances in other parts of the body and an inspection of the skin of the entire body was made and the various superficial nerve trunks commonly involved were palpated. No further evidence of probable leprous involvement was determined with the exception that there was questionable droop in the lower eyelid of the right eye. The patient's face was heavily bearded so that the inspection for paresis or paralysis of facial muscles was not entirely satisfactory.

"Two snips* were made from each of the two lesions. In one of these snips a number of acid-fast organisms, arranged in a manner typical of those found in leprosy were observed.

"The patient was advised to consult his family physician and have the lesions completely excised.This was done and the tissue excised was submitted by Dr. Batten.

"A portion of one of the lesions was ground, digested with antiformin, the suspension centrifuged and the sediment stained; a number of acid-fast organisms was found in these smears. The remainder of the sediment was used to make inocula on glycerine potato and egg yolk media, and for the

* Footnote: A "snip" in Hawaii means Wade's scraped incision. The corner of a stiff razor blade nicks a small mass of skin which is pinched tightly by two fingers to avoid bleeding. The blade is twisted as it is removed and the tissue juices so obtained on it are smeared on clean, new microscopic slides and properly stained for examination.

inoculation of guinea pigs. The remaining portion was used for histological sections."

The cultures and guinea pigs were not finally read unti June 30, 1933. This long period of observation (3 months) was necessitated by the resemblance of this one and only group of lesions in the case, histologically, to those of tuberculosis. During the study the health of the public was protected by the surgical excision of the lesion in question and by isolation of the patient.

On June 1, 1933, a Wassermann and a Presumptive Kahn were made; the results were negative. On June 30, 1933, neither the cultures nor the guinea pigs showed any evidence of the growth of tubercle bacilli. The histologic sections revealed a typical picture of tuberculoid leprosy, apparently of sometime standing. Giant cells were numerous as were typical acid-fast bacilli. The adjacent tissue was normal except for a blood vessel at some distance from the macular area, in the wall of which appeared a small granuloma.

The wound on the forehead, made by the excision of the lesions, healed smoothly by first intention, in due time. The skin about the scar had a slightly exaggerated pink blush but the appearance otherwise was normal.

This man was officially declared a leper on July 10, 1933; he had waived examination by a Board, but such an examining Board was nevertheless convened to make assurance doubly sure. The Board consisted of Drs. Gaspar, Batten, and Halford, all keen students of leprosy. He was, at his own request, returned directly to Kalaupapa, instead of being admitted to Kalihi Hospital, which is customary, this time as a patient, where he took up his residence in an institution for patients and resumed such of his activities and duties as brought him into contact with patients only.

I made the trip to Kalaupapa on the old S. S. Hawaii, that took the patient there. I seemed to be more affected by our arrival than did he. I went to "Staff Quarters" while he went to the Baldwin Home, newly reconstructed from the old Hospital. There I found him, an hour later, all alone in a large, bare, empty room. At my suggestion, three or four husky young patients and a truck quickly brought the patient's piano from the rectory to his new room. In a few moments he was lost in a Chopin's Nocturne—he had already made a happy mental adjustment.

Urged by his superiors and his physicians, he was now more cautious, his habits of life became more regular, his nutrition was adequate, he secured more rest and sleep and his general condition improved in spite of the fact that he received no so-called specific medication or treatment. Chaulmoogric esters were already definitely on the wane.

The pinkness in the excision scars slowly faded to a normal color and no new lesions made their appearance. Subsequent to the excision of the one group of lesions on the forehead and in the absence of the clinical history it would have been impossible for even an expert in leprosy to have made a

diagnosis of this disease, since the clinical picture was utterly negative and even repeated bacterioscopic examinations of intra-nasal scrapings and snips taken from the forehead, ear, nostrils, etc., failed to yield acid-fast organisms.

After the case had been clinically quiescent for several months and the number of bacterioscopic examinations required by the policy of the Leprosy Commission had failed to discover bacilli, the man was, after examination by an official board of physicians, consisting of Drs. Batten, Halford, and Luckie, placed on temporary release (on parole, in the local vernacular) on which status he had all the rights and privileges of a well person, save that he had to report monthly for detailed examination. On this status he resumed all his usual duties.

All active patients and, as well, those on parole are wholly and legally under the control of the Board and remain so until they are formally declared to be cured and fully released. Since 1925 no patient had been declared cured and therefore fully released.

No examination since the date of the excision of the lesion (March 1933) has yielded any signs or symptoms of leprosy. On October 8, 1934, I myself examined him. The patient was in excellent physical condition; the questionable lagophthalmos in the right eye was unchanged; the skin was pink and resilient; there were no cutaneous lesions that even remotely resembled leprosy; sensation everywhere was normal, and snips from nasal mucosa and ear lobe, on prolonged search, showed no lepra bacilli.

Subsequently, on May 25, 1935 I again examined him thoroughly at Kalaupapa and found him in excellent physical condition and nowhere could I find any evidence of leprosy. The scar on his forehead was hardly discernible, quite faded out. All snips up to that time had been negative for *M. leprae.*

On March 10, 1937, just four years after the diagnosis was made, Dr. Tuttle made a thorough examinantion of the patient and found no dermatological nor neurological abnormalities that pointed in any way toward leprosy. He prepared snips from the region about the surgical scar and from the ears. These I myself examined long and thoroughly, but could find no *M. leprae.* He had remained apparently "cured" for four years.

In January 1939, for a variety of reasons, the custom since 1925 was disregarded and this patient was examined by an official Board of Examining Physicians, consisting of Drs. Geo. B. Tuttle, Louis Gaspar, and the author, as a candidate for full discharge. After a thorough examination, no evidence of Hansen's disease could be found and the doctors recommended full discharge of the patient to the Board. According to the law, the decision of the examining board is final. The patient has remained since then on duty at Kalaupapa where today (September, 1944) he continues in good health; just twelve years after the first detected evidence of the disease or eleven years and six months after excision of the lesions.

This case of leprosy is unique from several standpoints: (1) It has occurred in an elderly European whose contact with leprosy has extended over a period of, at most, only about 7 years. The usual case in Hawaii occurs in an individual with a racial and familial predisposition, with a history of long contact, usually during childhood, with the appearance of the first lesions at or before puberty. (2) The location of the only skin lesions observed is unusual. It was formerly a dictum that leprosy always spares the scalp. (3) Unless the indefinite lagophthalmos was due to leprosy, findings referable to the nervous system were absent. (4) No other leprous lesions could be found and one might be justified in concluding that the bacilli had entered the body by way of the skin of the forehead and that this point of entrance presented an inoculation primary lesion. (5) The solitary group of lesions on the forehead was completely and widely excised and there was reason to hope that the elimination of this focus of infection might enable the patient to combat successfully the further development of the disease.

In this case, however, it was noted in the sections of the excised tissue, which were quite typical of tuberculoid leprosy, that, at a distance away from the major process (the papule) there appeared a comparatively large blood vessel whose wall had been invaded by what seemed to be an actively growing granuloma. While bacteria were not demonstrated in this granuloma, its presence in the blood vessel wall, outside the area of greatest activity, seemed to militate against the hope that *all* source of infection had been surgically removed. However, 11 years and 6 months after excision, there is no evidence of leprosy in or near the surgical field nor in the patient anywhere.

The case reported seems to be one in which an accidental dermal inoculation caused leprosy with no neural findings and that excision of the lesions has prevented dissemination for 11½ years.

The other case is an even more interesting and important one. H.K.D., No. 1567, Hawaiian, was a female child of leprous parents. The father, aged 41, with anesthetic leprosy, had resided in the Settlement 20 years, and had fathered three other children by three other wives. None of the other children were leprous. The mother, also with neural leprosy, was 40 years old and had resided in the Settlement 16 years; she had had five other children by two former husbands; none of these children were leprous.

The small patient had been separated from her parents at the age of 6 hours, and placed in the "clean" nursery, where she was nursed by the "clean" wives of lepers. At the age of 19 months she presented on her right cheek a patch of leucodermia, which was not anesthetic, which yielded no acid-fast bacilli, and which they considered non-leprous. The infant patient presented on the flexor surface of the left forearm a red-brown nodule, 12 x 8 mm. and 2 mm. high, which had been there about two weeks. This lesion

was excised. When cut into, the nodule had the grayish-white appearance so commonly seen in leprous lesions. Smears from the juice of this nodule showed moderate numbers of acid-fast bacilli having the grouping characteristic of leprosy bacilli from tissue. Snips from the leucodermic patch on the cheek, and smears from both nostrils were negative.

The histological appearance of the nodule was entirely consistent with a leprous nodule, and acid-fasts were readily demonstrated in section. There was no necrosis.

Examined four months after the operation, the patient showed no progress of the disease, and the scar left by the excision of the nodule was free from acid-fast organisms.

This case was reported by Drs. Wm. J. Goodhue and Geo. W. McCoy in the January 1916 Public Health Bulletin, No. 75, entitled "Leprosy in a 19 Month Old Child." To them at that time, the case was worth recording because leprosy had appeared in a child so very young—it was at that time probably the earliest reported case. Leprosy was popularly supposed, at that time, to spare children under five years of age. It is difficult to understand now why the patient was not certified as leprous immediately and the records throw no light on this subject. Three months later, *i.e.* seven months after the excision, snips from the region of the excision scar showed a few atypical acid-fasts (the popular expression in Hawaiian medical circles was "leper dust"), and she was then declared a leprous patient and continued to reside in the Settlement, presumably with her leprous parents.

At the age of ten years she was again thoroughly examined by Drs. W. J. Goodhue and H. E. Hasseltine, of the U. S. Public Health Service, who could find no evidence of leprosy and they reported their findings in the U. S. Public Health Reports, 39, 1924, pp. 2680-2683.

At about the age of seventeen she married a patient, heavily infected with the lepromatous type of leprosy, with marked nodulation.

In 1937 her mother was still residing in the Settlement and from that fact one may deduce that hers was a case of tuberculoid leprosy.

A change in the economic status of parolees in Kalaupapa, giving them the same subsistence privileges and cash allowances as were given to the patients, brought this patient, as an applicant for parole, before a parole board consisting of Drs. Grover Batten, Geo. B. Tuttle, and the author on May 21, 1937. At that time there was no evidence of leprosy, past or present, and six snips at intervals between January 7 and May 21 had been negative for bacilli. The patient was not identified as the infant who had had leprosy at the age of nineteen months and the scar of excision was overlooked. She was recommended for parole, which was granted by the Board and she continued to live in the Settlement with her heavily leprous husband.

In January 1939 I identified this patient, re-examined her and found no evidence of Hansen's disease, but did find a transverse scar on the left fore-

arm, three and one-half inches below the antecubital crease, slightly elevated, one and a half inches long and three-sixteenths of an inch wide, tapering to a point at each end. The identification was established.

In August, 1944 when I visited her very sick husband in the Hospital I made a cursory examination of this patient. She still had the leucodermic, non-leprous spot on her cheek but I found no evidence of leprosy.

It is remarkable that she has remained free of Hansen's disease for nearly thirty years, for in the beginning the chances were very much against her because:

1. She was Hawaiian and that race seems quite susceptible to leprosy.
2. Both her parents were leprous, even though her mother probably had the tuberculoid form of this disease, and if there is an inherited, familial predisposition to the disease, she should have had it. (It seems generally agreed that the disease itself is not hereditary; only the predisposition is inherited from either or both parents.)
3. She *was* susceptible, as witnessed by the fact that she contracted the disease at the very early age of only nineteen months, probably the tuberculoid form.
4. She went through adolescence unscathed, though heavily exposed to infection.
5. She married a man with advanced lepromatous leprosy, who developed laryngeal lesions that necessitated a tracheal tube, and she was constantly exposed to heavy doses of infection.

Possibly the defense mechanism in these two cases is somewhat analogous to that aroused by B.C.G. vaccine in tuberculosis.

To my mind, these two case reports could be, to our people in Hawaii Nei, the most forceful propaganda for early case finding.