A Single Skin Lesion—An Unsual Presentation of Lepromatous Leprosy[1]

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Lepromatous leprosy is usually described as a generalized disease with numerous macules, papules, or nodules distributed symmetrically over the skin of the face, trunk, and extremities ([1]). Lepromatous leprosy presenting as a solitary lesion with a high bacterial count has not been reported in the literature and is a rare occurrence in most people's experience. In this paper, we describe a patient who presented with a single skin lesion showing the histopathological features of infiltrated or nodular lepromatous leprosy.

A CASE REPORT

Clinical presentation. The patient was a 52-year-old, light-skinned man who was born in Indonesia and in 1966 moved to the United States. Nine months prior to admission to Carville in 1977, a small lesion resembling an insect bite appeared above his left elbow. It persisted and enlarged, and after several months an attempt was made by his private physician to destroy it using dry ice. It did decrease in size with this treatment, but later enlarged again and was thought to represent a keloid. Plans were therefore made to have a plastic surgeon excise it and, prior to surgery, a biopsy was taken. A diagnosis of leprosy was made, and the patient was then referred to Carville. Examination here revealed only a 3 cm erythematous, hyperpigmented, horseshoe-shaped lesion just above the left elbow which had a cord-like consistency with some loss of light touch and pain sensation over and adjacent to it (Fig. 1). The left superficial radial cutaneous nerve was moderately enlarged and non-tender. The remainder of his skin appeared completely normal and sensory examination was otherwise normal. There was no motor loss noted. The patient had no known history of exposure to leprosy.

Slit-skin scrapings from the lesion showed a bacterial index (BI) of 5+ with morphological index (MI) of 0%. Skin scrapings at six additional sites (the ears, knees, and elbows) were all negative for acid-fast bacilli (AFB). The lepromin skin test was negative. BALB/c mouse foot pad culture was done and an inoculum of 5×10^3 organisms multiplied in eight months to 1.27×10^6 in control mice receiving no drugs. No growth was detected in animals continuously fed diets containing 0.0001% w/w dapsone, 0.001% clofazimine, or 0.01% rifampin, indicating that the bacilli were fully sensitive to dapsone, clofazimine, and rifampin. Löwenstein-Jensen medium was also inoculated with a suspension from the biopsy and showed no growth at 35°C–37°C in six weeks.

After the initial diagnosis, the patient was started on dapsone 100 mg daily, which he has continued to the present time. There have been no new lesions, and the initial lesion now appears inactive.

Histopathology. Biopsy sections showed a flat atrophic epidermis with marked hyperkeratosis. Underneath the flattened epidermis was a clear zone separating a deeper confluent inflammatory infiltrate (Fig. 2). The lesion was composed mainly of spindle-shaped macrophages with some showing foamy changes (Fig. 3). There was a diffuse sprinkling of mononuclear cells which can be identified as plasma cells and lymphocytes. No nerve bundles were seen. Sections with acid-fast stains showed clumps of bacilli in the macrophages, including the spindle-shaped ones (Fig. 4). A histopathological diagnosis of subpolar lepromatous leprosy with histoid features was made. A second biopsy taken nine months after the start of treatment showed again the flat epithelium with the clear zone. There was now organization of the granuloma, and the

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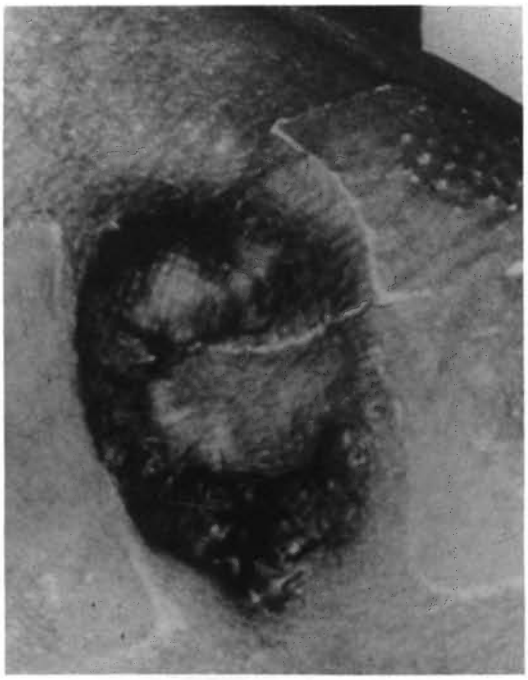

FIG. 1. The lesion on the elbow at the time of diagnosis.

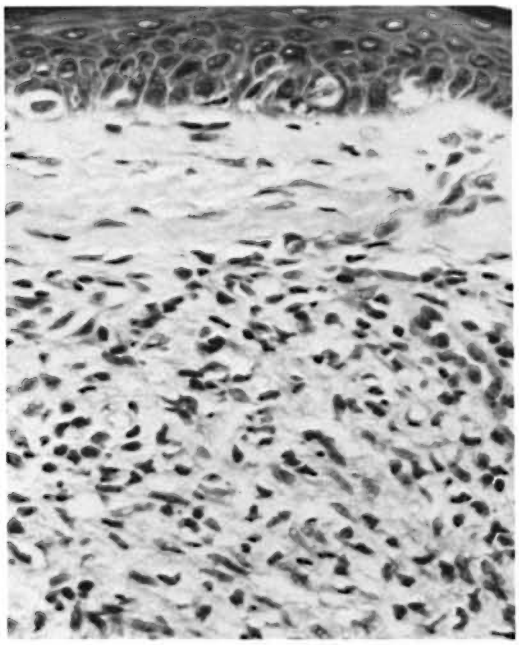

FIG. 2. Photomicrograph of the lesion before therapy, showing an atrophied epidermis. A clear area separates a band of spindle-shaped macrophages and a few scattered lymphocytes and plasma cells (H&E ×600).

macrophages were interspersed with numerous lymphocytes (Fig. 5). The macrophages were much fewer (Fig. 6) than in the previous biopsy and were packed with bacilli (Fig. 7). Significant resolution of the bacilli-filled macrophages as compared to the initial biopsy was noted. A final biopsy taken in 1983 showed no lesion in the epidermis. In the dermis there were collections of lymphocytes around capillaries (Fig. 8) and a few scattered foamy macropahges could be identified (Fig. 9). Acid-fast stain showed no bacilli. The picture is now consistent with inactive or healed lepromatous leprosy.

DISCUSSION

This patient had only a single, horseshoe-shaped lesion located near the left elbow. It was erythematous, hyperpigmented, and raised with a cord-like consistency. It is not surprising that it was mistaken for a keloid and that there was an attempt to treat it as such. However, the loss of sensation to touch and to pinprick is unusual and is helpful in suggesting that the lesion might be leprosy. Clinically, the nerves were not involved except for some enlargement of the left radial

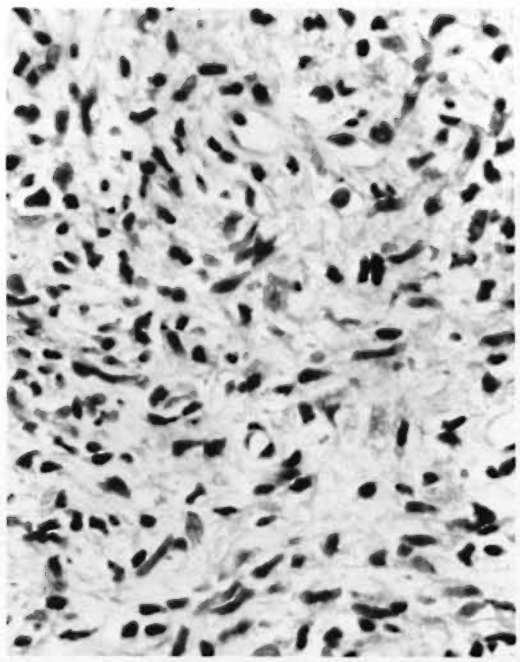

FIG. 3. Another microscopic field in the dermis showing clear, spindle-shaped macrophages and macrophages with foamy cytoplasms (H&E ×600).

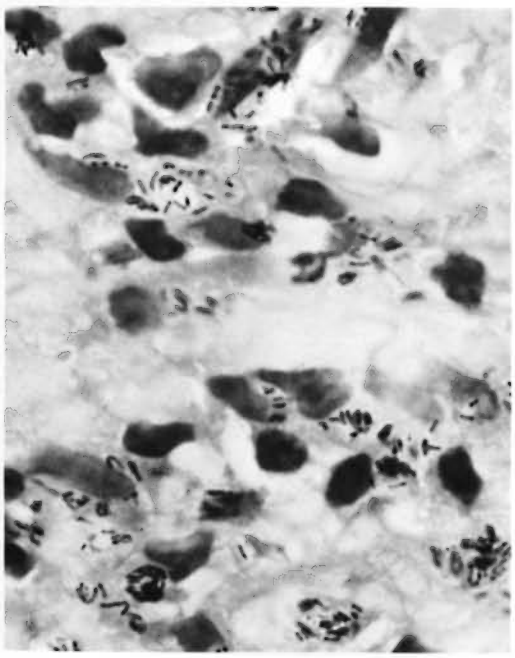

FIG. 4. Acid-fast stain of the same tissue as Figure 2, showing almost every cell in the field packed with bacilli (×1200).

cutaneous nerve. It is not possible, however, to rule out subclinical infection of the cutanous nerves and peripheral nerve trunks by the organism. There have been reports of single indeterminate lesions of the skin where acid-fast bacilli were detected in radial cutaneous nerves ([2]).

Histopathologically, many plasma cells and lymphocytes were present and the macrophages contained acid-fast bacilli. The lesion had a BI of 5+ and a MI of 0%. According to Ridley and Jopling, the very early stage of an active lepromatous leprosy lesion, which is not often seen except at the beginning of a relapse, shows a predominance of spindle-shaped cells that resemble fibrocytes in appearance although they ingest bacilli ([1]). It is also not uncommon to see plasma cells in well-established lesions of lepromatous leprosy. The scattered clumps of lymphocytes in the lesion, however, indicate that the patient had some localized immunity and that the disease should be classified as subpolar lepromatous disease.

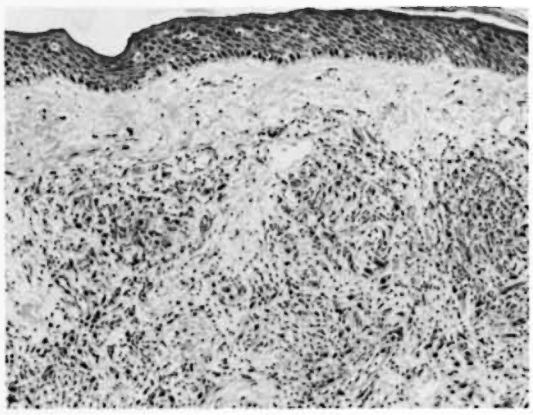

FIG. 5. Photomicrograph of the lesion 9 mos. after therapy, showing some atrophy of the epidermis. A clear area separates a large granuloma composed of a mixture of macrophages, lymphocytes, and plasma cells (H&E ×350).

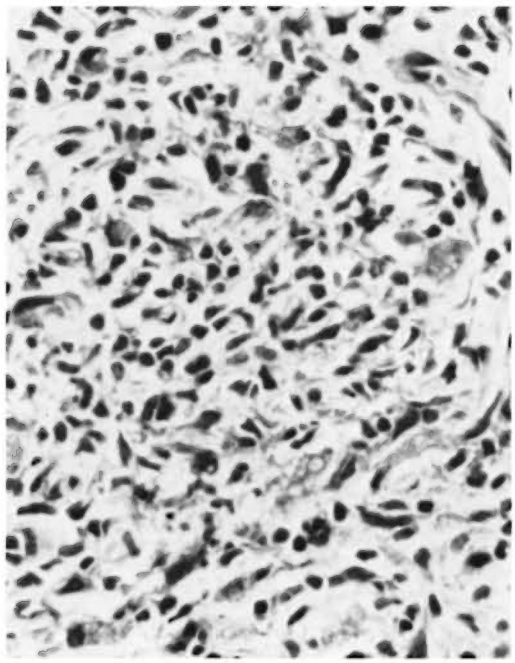

FIG. 6. High-power magnification of Figure 5, showing the persistent spindle-shaped macrophages and a few scattered foam cells (H&E ×600).

Since dermal nerves with intraneural organisms were not seen and routine culture for acid-fast organisms was only done on one occasion on only Löwenstein-Jensen medium, one might wonder whether this lesion was caused by a cultivable acid-fast organism. Clinically there was enlargement noted of the left radial cutaneous nerve, and histologically there was foamy change of the macrophages containing bacilli. Further-

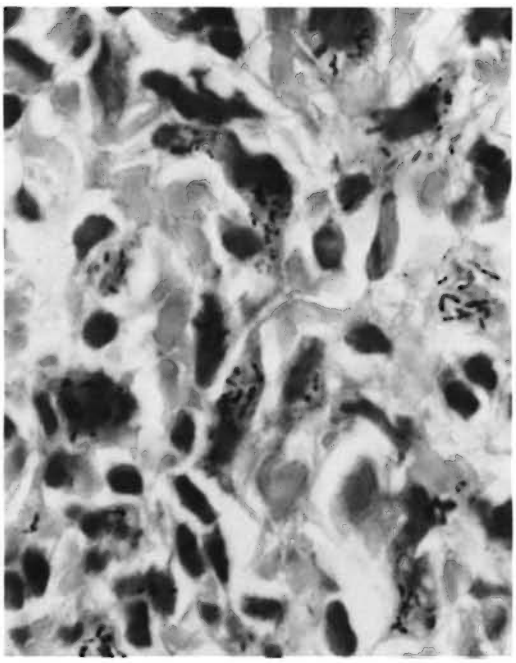

FIG. 7. Acid-fast stain of the same tissue as Figure 5, showing a few scattered macrophages which continue to have intracellular bacilli (×1200).

more, there has been a good response to dapsone and the growth of the organism in the foot pads of mice is characteristic of *Mycobacterium leprae* both as to time course and ceiling as well as drug sensitivity pattern. Therefore, it is highly unlikely that this lesion was caused by AFB other than *M. leprae.*

This patient is unusual in that he has a single lesion which histopathologically shows most, if not all, of the features of lepromatous leprosy but clinically resembles tuberculoid disease. The minimal loss of sensation can be explained by the fact that the lesion was at the elbow and there was marked hyperkeratosis of the skin in that area as seen histologically. Thus, we would note that a lesion clinically resembling polar tuberculoid leprosy can be immunologically and histopathologically lepromatous leprosy. This case emphasizes the importance of skin scrapings and histopathological identification and classification of all patients with unusual lesions. Without such information, this patient would have been considered tuberculoid or borderline tuberculoid and would have received limited treatment.

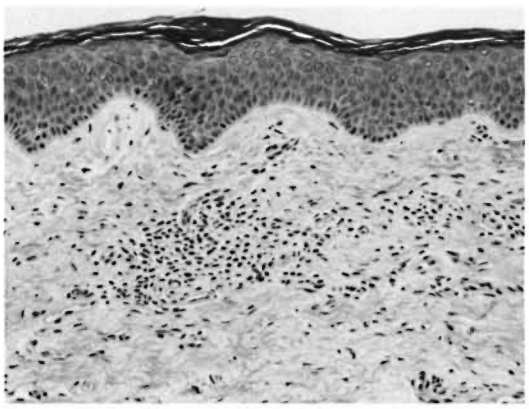

FIG. 8. Photomicrograph of the lesion after six years of therapy; the epidermis shows no significant abnormality. Small local collections of mononuclear cells are seen in the dermis (H&E ×350).

Finally, one can only speculate as to what factors may be involved in producing a situation such as this in which a lepromin-negative patient was, at least for the time interval in question, able to limit his disease to one skin lesion while bacilli were multiplying to a level of 5+ in that localized area. Minor trauma, lower skin temperature, and direct exposure of skin to *M. lep-*

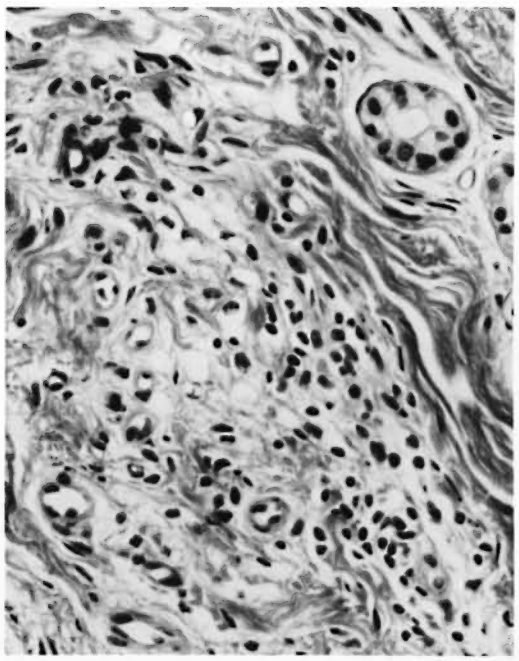

FIG. 9. Focal collections of mononuclear cells, some of which are skeletons of foamy macrophages, are seen in the deep dermis (H&E ×600).

rae might be possible factors involved in causing a lesion such as this.

SUMMARY

Lepromatous leprosy presenting as a solitary lesion with a high bacterial count is a rare occurrence. Such a case has been followed at the National Hansen's Disease Center, Carville, Louisiana, U.S.A., since 1977. The lesion was located on the left elbow and had been present for about nine months. The bacterial index in the lesion was 5+ and the morphological index was 0%, but slit-skin smears elsewhere were negative. The histopathology of the lesion was that of subpolar lepromatous leprosy, and the lepromin skin test was negative. Growth of the organism in the mouse foot pad was characteristic of *Mycobacterium leprae*, and the patient's response to dapsone monotherapy has been excellent. Therefore, this patient with a single skin lesion has lepromatous leprosy histologically, immunologically, and bacteriologically. This case illustrates the importance of slit-skin scrapings and biopsy in new cases with unusual lesions and, secondly, suggests that there are factors yet undetermined which play a significant role in determining host response to *M. leprae*.

RESUMEN

La lepra lepromatosa en la forma de una lesión solitaria y con una elevada cuenta bacteriana es un hallazgo muy raro. Un caso de este tipo ha sido estudiado en el Centro Nacional de la Enfermedad de Hansen en Carville, Luisiana, U.S.A., desde 1977. La lesión estuvo localizada sobre el codo izquierdo durante unos 9 meses. El índice bacteriológico en la lesión fue de 5+ y el índice morfológico fue de 0% pero los análisis de linfa en otros sitios fueron todos negativos. La histopatología de la lesión correspondió a la lepra lepromatosa subpolar y la prueba de la lepromina fue negativa. El crecimiento del microorganismo en el cojinete plantar del ratón fue característico del *Mycobacterium leprae* y la respuesta del paciente a la monoterapia con dapsona ha sido excelente. Por lo tanto, este paciente con una sola lesión es lepromatoso desde el punto de vista histológico, inmunológico y bacteriológico. El caso ilustra la utilidad de los raspados de ranuras dérmicas ("slit-skin-scrapings") y de la biopsia en el diagnóstico y clasificación de los casos con lesiones raras y secundariamente sugiere que hay factores todavía no bien estudiados que participan en la determinación de la respuesta del huésped al *M. leprae*.

RÉSUMÉ

Il est rare que l'on observe une lèpre lépromateuse se présentant sous forme d'une lésion solitaire, avec de nombreux bacilles. Un tel cas a été suivi au National Hansen's Disease Center, à Carville, en Louisiane, aux Etats-Unis, depuis 1977. La lésion était située sur le coude gauche, et a persisté pendant environ 9 mois. L'index bactériologique au niveau de la lésion était 5+; l'index morphologique était 0; des frottis cutanés de surface pratiqués ailleurs ont révélé des résultats négatifs. L'histopathologie de la lésion présentait des caractéristiques de lèpre lépromateuse infra-polaire; l'épreuve cutanée à la lépromine était négative. La croissance du microorganisme dans le coussinet plantaire de la souris était caractéristique de *Mycobacterium leprae*. La réponse du malade à la monothérapie par la dapsone à été excellente. On en conclu que ce malade présentant une lésion cutanée unique souffrait de lèpre lépromateuse, tant sur le plan histologique qu'au point de vue immunologique et bactériologique. Ce cas illustre l'importance des frottis cutanés et de la biopsie pour le diagnostic des nouveaux cas présentant des lésions inhabituelle. Il suggère de plus que certains facteurs encore non élucidés jouent un rôle notable pour déterminer la réponse de l'hôte à *M. leprae*.

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