LEPROMATOUS MYOSITIS

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The ordinary affections of the skeletal muscles in leprosy, paralyses and atrophies, are such common and banal secondary effects of peripheral nerve involvement that not a great deal of interest has been taken in the changes in the muscles involved. It has not been established, and apparently in the past it was seldom even suspected, that in lepromatous leprosy the muscles of the extremities might be involved at all by the leprotic process itself. The object of this paper is to report our observations in various cases of lepromatous leprosy in which there was a direct involvement of the striated musculature as an apparently independent process.

For the older literature with bearing on this subject we are compelled to depend on Jeanselme (3). According to him, Y. Saijo and M. Takino pointed out in 1929 that not all trophic changes in the muscles are directly attributable to neuritis of the branch nerves which serve them, and that the leprotic process can invade and directly destroy the muscle fibers. This, the authors affirmed, would not be a rare phenomenon at all in the muscles of the tongue and larynx.

According to Jeanselme the leprosy bacillus has been observed intact in a granular state in muscle fibers by Babes, Fujinami, and Jeanselme himself, and in connective tissue between the atrophied fibers by Wnukow. The bacilli have been found particularly in the muscle fibers which are closely connected with mucous membranes exposed to leprotic infiltration. Leloir was the first to give a histologic and bacteriologic description of leprous glossitis, and his observations have been confirmed by other workers.

Nevertheless, it cannot be said that specific leprous muscle lesions have been observed frequently. Black and Denney (1) give them last place in the approximate order of frequency. K. Mitsuda (4) called attention to the finding of isolated lepra cells in the muscle fibers of the digestive tract. Uhlenhuth and Westphal have found the bacillus in the myocardium.

We have not found any publication that comes close to describing the muscular lesions which we have observed for some time, and of which we have made a detailed histologic investigation. We refer to

the progressive formation of a lepromatous granuloma within the large striated muscle, and not to the finding of isolated bacilli in muscular tissue, nor with the invasion of such tissue by bacilli from contiguous mucous membranes.

MATERIALS AND METHODS

The material for this study was taken from nodular lesions located deeply within the muscles affected. The sizes of the nodules varied from hazelnut to walnut. They were hard and slightly painful. Their exact location could be determined clinically only by careful palpation, which was necessary since no protuberance was visible on the surface on account of the depth at which they had been formed.

After making the incision in the aponeurosis, it was seen that the nodules were of a much lighter color than the apparently healthy tissue, and they were difficult to cut into because of their hardness. These peculiarities of color and consistence were enough to define their borders exactly.

The specimens of muscle tissue for examination, measuring about 2 x 3 cm., were removed under local anesthesia from the calf of the leg, or from the arm. They were fixed in formalin, and paraffin sections were stained by methods such as hematoxylin-eosin, ferric hematoxylin, van Gieson, PAS, Masson, and Fite (1947) for bacilli.

RESULTS

The histologic study revealed the presence of a specific granuloma in the connective-tissue muscle coverings (perimysium and endomysium), the ramifications of which it follows progressively, penetrating into the thick as well as the thin fasciae until it reaches the muscle fibers themselves. These become separated by its invasion of the epimysium. It is a creeping advance along the epimysium that envelopes the muscle fibers, and these become dissociated almost from the beginning.

In some zones there is a notable prevalence of histiocytes which have not yet become vacuolated in the manner characteristic of the Virchow cell, but are parasitized by morphologically slightly modified bacilli (Fig. 1). In other parts the histiocytes are seen to be clearly modified, forming atypical, different-sized foci of lepromatous granulomas. In these foci the bacilli often show notable morphologic changes, such as granulation and fragmentation, and they may even form amorphous acid-fast masses (Figs. 2 and 3). The lepromatous granuloma is so dense in some places that the fibers have become separated and, by a mechanism of compression, even ruptured (Fig. 4).

In spite of all this the muscle fibers, taken as a whole, do not show any great changes in the presence of the heavy lepromatous infiltration. Nevertheless, those fibers that are in close contact with dense granulomas have degenerated. They show loss of striation and appear frayed and partly destroyed. Within some zones of the lepromatous granuloma there may be seen remnants of fibers that have lost their structure entirely.

As a last but not least important fact, we want to point out that sometimes the leprosy bacillus were found morphologically intact within apparently undamaged muscle fibers.

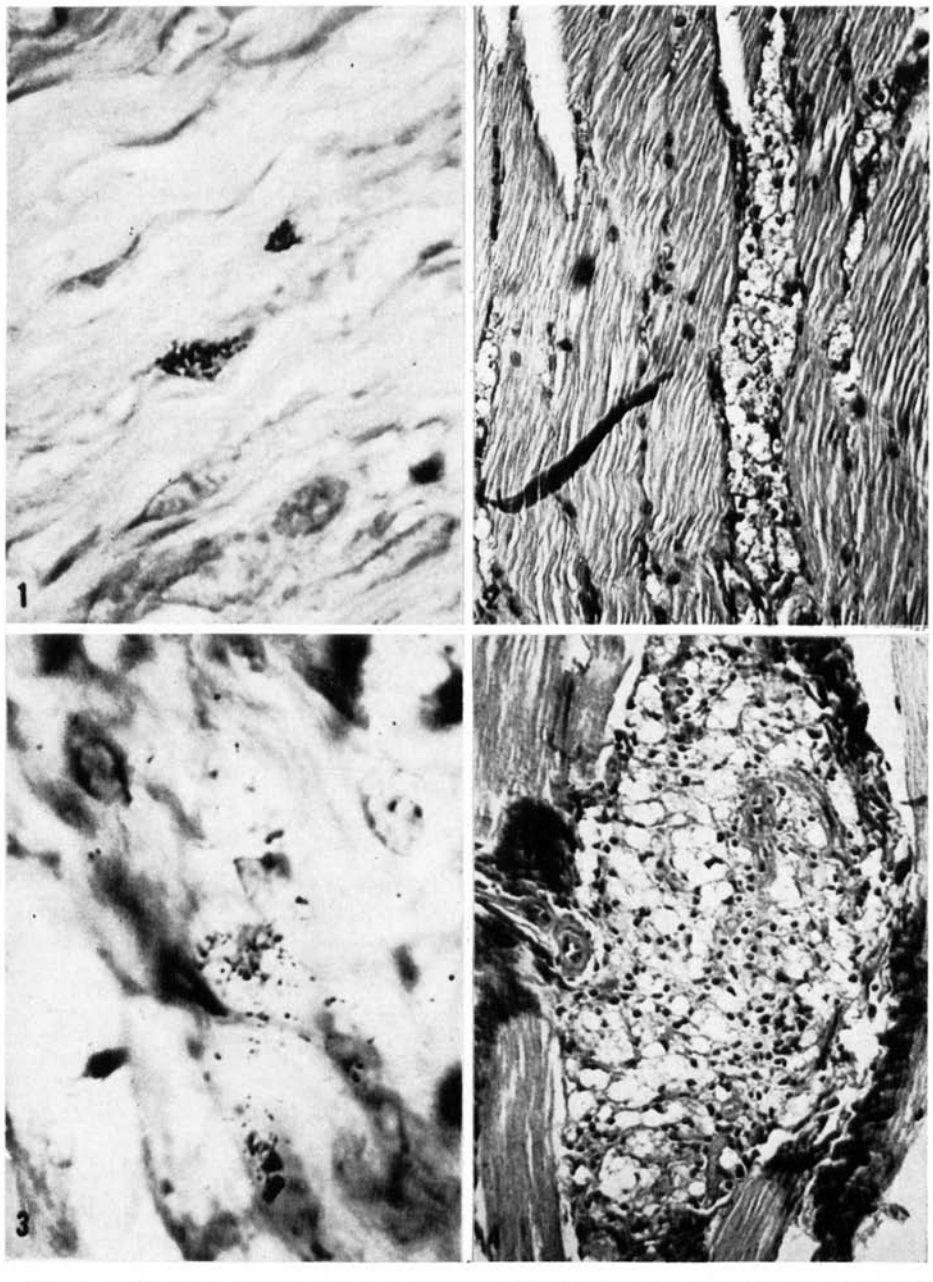

FIG. 1. Infiltration along internal perimysium with a histiocyte not yet vacuolized in the manner of the Virchow cell, but filled with bacilli, are practically unchanged. Stain: Fite-Faraco.

FIG. 2. Lepromatous granuloma in the form of bands that separate the muscle fibers. Stain: Hematoxylin-eosin.

FIG. 3. Leprosy bacilli greatly modified by advanced granulation. Stain: Fite-Faraco.

FIG. 4. Lepromatous granuloma separating and partly lacerating muscle fibers. Stain: Fite-Faraco.

CASE HISTORIES

CASE 1. I.P., 2,453. A.R.A., male, mixed race, age 30 years. Moderately disseminated infiltrations with small nodules in the ears. Infiltrating lesions of the nasal mucous membrane. Sparseness of eyebrows, and irregular alopecia of the lanugo of the limbs. Thickening of the supraorbital, ulnar and external popliteal nerves. Sensory disturbances in the limbs.

Bacteriology, skin and nasal mucous membrane: Positive, 3+.
Immunology: Lepromin negative.
Histopathology, skin: Lepromatous granuloma, bacilli 3+.
Diagnosis: Lepromatous leprosy.
Treatment: Promin, DDS.

The course of the disease was characterized by frequent lepra reactions of the erythema-nodosum type. During these reactions it was possible to palpate several successively-occurring nodules in the fleshy masses of the gemelli, triceps brachii, quadriceps femoris, anterior and posterior tibialis, and in the musculature of the forearm.

A piece of tissue for biopsy was taken from a nodule located deeply below the aponeurosis in the gemelli muscles. Findings: lepromatous granuloma, 3+ positive for bacilli.

CASE 2. C.B., 3,134. M.M., female, Negro, age 44 years. Moderate, disseminated infiltration of the ears and arms. Ulnar nerves slightly enlarged. Amyotrophia in the hands. Sensory disturbances in the arms.

Bacteriology: Positive, 3+.
Immunology: Lepromin negative.
Histopathology, skin: Lepromatous granuloma, bacilli 3+.
Diagnosis: Lepromatous leprosy.
Treatment: Promin.

The patient suffered frequent lepra reactions of the febrile erythema-nodosum type, which lowered her general state of health. During one of these reactions a deep-seated, hard, ligneous, slightly painful mass of infiltration, about the size of a hazelnut, was palpated in the calf of the right leg.

A piece of tissue was removed from this nodule for biopsy. Findings: lepromatous granuloma located between the muscular fasciae with degenerative effects in the fibers. Bacilli 2+.

CASE 3. C.B. 3,033. R.J.N., male, brown-skinned, age 42. Diffuse infiltrating processes in a state of regression, with slight enlargement of the ulnar nerves and sensory disturbances in the forearms, elbows and knees.

Bacteriology: Positive, 3+.
Immunology: Lepromin negative.
Histopathology, skin: Lepromatous granuloma, bacilli 3+.
Diagnosis: Lepromatous leprosy.
Treatment: DDS.

After the patient had been hospitalized for two years, he was found to have several hard, painless nodules, each about the size of a hazelnut, within the muscular masses of the triceps, in the inner muscles of the right forearm, and in the soleus. When the patient was in the reactional state these nodules became at times harder and larger.

Biopsy of one of the nodules showed lepromatous granuloma between the fasciae, with degenerative processes in muscle fibers. Bacilli 1+.

CASE 4. C.B. 3,258. F.N.S., male, brown-skinned, age 22. The patient shows a generalized nodular infiltration, with lesions in the mucous membranes. There is enlargement of the nerves in the superficial cervical plexus. Sensory disturbances in the limbs.

Bacteriology, skin and nasal mucous membrane: Positive, 3+.
Immunology: Lepromin negative.
Histopathology, skin: Lepromatous granuloma, bacilli 3+.
Diagnosis: Lepromatous leprosy.
Treatment: Diphenylthiourea.

After 3 months of hospitalization the patient was found to have a firm, slightly

painful nodule, the size of a walnut, situated deep within the outer musculature of the left arm. According to the patient, he had been aware of the nodule for a year, and that its formation coincided with the appearance of his skin lesions. The nodule became painful and increased in size during lepra reactions of the erythema nodosum type.

Histologic study of the nodule removed from the muscular mass showed lepromatous granuloma between the fasciae, with degenerative processes in the fibers. Bacilli 2+.

DISCUSSION

With the exception of the descriptions of leprous glossitis, there has been no report made as yet, as far as we know, of the presence of lepromatous lesions in the striated, voluntary muscles, nor of the clinical and histopathologic characteristics of an interstitial leprous myositis, such as is found in our cases.[1]

The lesions have been found, deep within the muscular mass, as isolated firm nodules which have become painful and at times harder and larger during intercurrent lepra reactions. In some cases there has been only one nodule, in others several in succession, within the muscles of the limbs. Their function did not seem to be seriously impaired by the presence of these nodules.

As the nodules lie so deep and form no visible projection on the surface, it being necessary to cut through the aponeurosis to get to them, they can be discovered only by thorough palpation of the muscular mass. In one of our cases, in which there was a single nodule within the brachial triceps, it was difficult to find it by palpation when the muscle was relaxed, but quite easy when it was contracted. This shows that it is important to palpate the muscles thoroughly, not only in their flaccid but also in their contracted state. In another case a nodule, found in the deltoid, was so hard that it was difficult to get a needle into it in order to make an intramuscular injection.

The histologic study of these nodules has revealed a lepromatous granuloma between the muscular fibers, with the bacilli usually much broken down and often unrecognizable as such on account of their being reduced to an amorphous, acid-fast substance within the lepra cells. This rather interesting phenomenon may be due to the bacilli having been subjected to a higher temperature in the muscle tissue than they usually have in the skin ([2]).

In other parts of the granuloma, however, the morphology of the bacilli has suffered little or no change. We have also observed them, isolated or in groups, within the muscle fiber itself. We do not think that they could have been artifacts due to preparation, as they were found at different levels within the same fiber.

It seems to us that there is at first an interstitial process developing along the vessels of the perimysium and the endomysium, and that it is followed by destructive lesions in the muscle fiber. There is appar-

[1] The report entitled Myositis Interstitialis Leprosa, by Shigenori Ishihara [THE JOURNAL **27** (1959) 341-346], on four cases with lepromatous nodules in the calf of the leg, had not been published when the manuscript of this paper was prepared.—J.C.

ently a direct invasion of the muscle cells by the leprosy bacillus, which is found singly or in groups within the fiber itself. This may be an important aspect of the lepromatous myositis and should, we believe, be investigated thoroughly.

It is surprising how relatively intact the muscle cells remain in the presence of the lepromatous invasion, but we have observed that they are not conserved nearly as well when the patient has been through several lepra reactions.

It is our impression that these cases may prove to be not infrequent, when their existence becomes known and efforts are made to find them by thorough palpation in routine clinical examinations.

SUMMARY

Four cases of lepromatous leprosy are described in which there had been found, deep in the muscles of the limbs, nodular formations which increased in size and became painful during lepra reactions.

Histologic examination of the nodules revealed lepromatous granulomas that separated the muscle fibers. The bacilli within the granulomas were greatly changed in shape, and even at times appeared as amorphous acid- and alcohol-resistant masses within the lepra cells. The presence of intact bacilli, singly or in groups, in the muscle fiber is considered to be an important feature.

The impression has been gained that this localization of leprosy in muscle tissue may be more frequent than is realized, and it is suggested that a search for nodules be made systematically by palpation of the muscles in routine control.

RESUMEN

Descríbense aquí 4 casos de lepra lepromatosa en los que se encontraron, bien adentro de los músculos de los miembros, formaciones nodulares que aumentaban de tamaño y se volvían dolorosas durante las reacciones leprosas.

El examen histológico de los nódulos reveló granulomas lepromatosos que separaban las fibras musculares. Los bacilos del interior de los granulomas habían cambiado considerablemente de forma, y a veces hasta aparecían como masas amorfas ácido- y alcoholo-resistentes dentro de las células leprosas. Se considera que la presencia de bacilos intactos, ya aislados o en grupos, en la fibra muscular constituye una característica importante.

Se ha recibido la impresión de que esta localización de la lepra en el tejido muscular puede ser más frecuente de lo que se cree, y se propone que como comprobación habitual se haga sistemáticamente una pesquisa en busca de nódulos mediante la palpación de los músculos.

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