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INTERNATIONAL JOURNAL OF LEPROSY

VOLUME 31, NUMBER 3

JULY-SEPTEMBER 1963

LEPROUS ORCHITIS IN REACTIONAL BORDERLINE CASES

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Testicular lesions in leprosy of the lepromatous type are common and well known. On the other hand, lesions in testes in nonlepromatous cases seem to be extremely rare and are not well documented in the literature. In this paper is presented a detailed account, including the histopathologic study, of three leprosy cases belonging to the borderline group which showed tuberculoid granulomas of the testes.

MATERIAL AND METHODS

A study of the testes of leprosy patients willing to undergo biopsy of that organ was undertaken. The patients were inpatients of the Schieffelin Leprosy Research Sanatorium, Karigiri. Three such cases which showed definite tuberculoid granulomas in the testicular tissue were chosen for this study. In each instance a detailed history was taken, and a thorough physical examination was made, with routine examinations of blood, urine and stools. A skin smear was made by the usual scraped-incision method, and biopsy specimens were taken—of the skin to confirm the classification of the disease, and one from the testes for detailed study.

CASE HISTORIES

CASE 1 (S.L.R.S. 4288).—An unmarried male, aged 21 years, reported to the outpatient department with the complaint of an erythematous plaque on his back since two years before. Toward the latter part of this period the disease began to spread, and multiple patches gradually appeared on other parts of the body. His left breast began to increase in size, becoming painful and tender. There was no history of any contact with a known leprosy patient.

Examination.—Examination at the time of admission revealed multiple patches of different sizes on the trunk, extremities and face (Figs. 1 and 2). These patches had raised edges which sloped away from the centers, and they were erythematous and somewhat scaly. In some areas, especially on the back of the trunk and the extremities, the patches seemed to coalesce together to give an appearance of gross

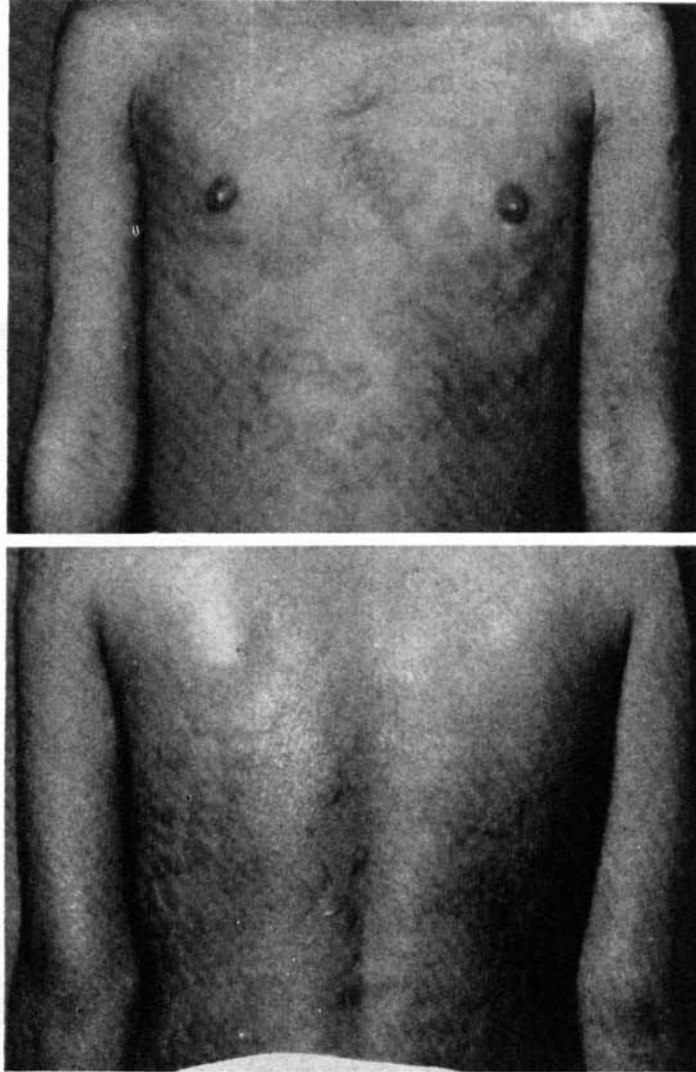


FIG. 1.—Anterior aspect of the body of Case 1, showing multiple ring-shaped lesions (tuberculoid) on the chest, but more diffuse by marginate plaques on the upper arms.

FIG. 2.—Back of Case 1, largely covered by plaque-like lesions, mostly with indefinite borders.

infiltration of the skin. Edema of hands and feet was present. The ulnar, lateral popliteal, and posterior tibial nerves on both sides were thickened. The left breast showed some enlargement and was tender. The right testis was slightly swollen, but not tender.

Routine examination of the urine and stools showed no significant abnormality. The hemoglobin was 13.75 gm. per cent. A skin smear gave a bacteriologic index of 1.12.

Clinical diagnosis.—The patient's condition was diagnosed as borderline leprosy in reaction.

Histopathology.—The skin biopsy specimen showed atrophy of the epidermis in areas, and in some places the rete pegs were flattened out. In the corium there was a large collection of infiltrate cells consisting of epithelioid and giant cells, macrophages, and lymphocytes (Fig. 3). They almost formed a band beneath the epidermis, separated from it by a clear zone. Infiltrate-cell collections were also present in the deeper corium. Nerve bundles showed both perineural and intraneural inflammation. The Ziehl-Neelsen stain for acid-fast bacilli showed many broken-up bacilli inside nerve bundles. The histopathologic picture was quite consistent with borderline leprosy.

The biopsy specimen of the testes showed, grossly, a wedge-shaped piece of tissue, grayish-white in color and firm in consistency. No normal tubules could be picked up with forceps.

On microscopic examination the tunica vaginalis was found to be markedly thickened with fibrous tissue. Hyalinization of the fibrous tissues was extensive. The inner lining of the thickened tunica was infiltrated with areas of granulomatous inflammation consisting of collections of epithelioid and giant cells and lymphocytes. The seminiferous tubules showed atrophy, and there was no spermatogenesis. Only Sertoli cells were seen lining the tubules, and the basement membranes were markedly thickened. The interstitial tissue was diffusely infiltrated by epithelioid and giant cells, macrophages, lymphocytes, and plasma cells (Figs. 4 and 5). In areas the seminiferous tubules were also infiltrated by inflammatory cells and were destroyed (Fig. 6). There was proliferation of Leydig cells in focal areas. In one such area the Leydig cells were infiltrated by inflammatory cells and were being destroyed.

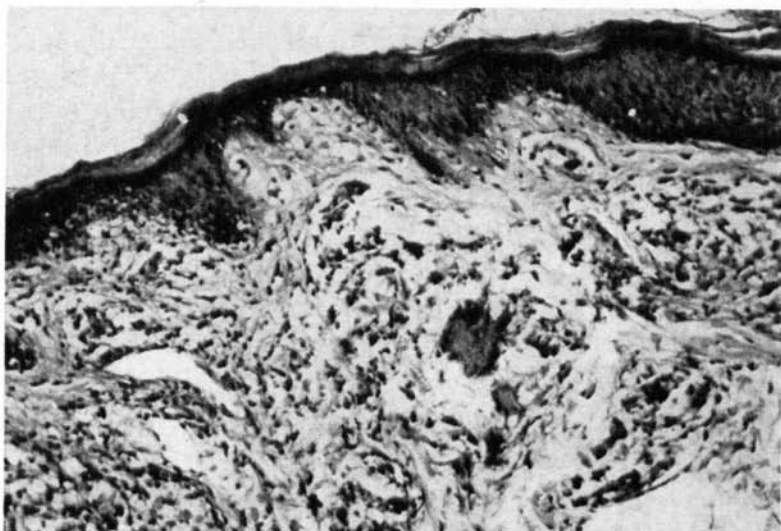


FIG. 3.—Section of skin lesion, showing epithelioid-cell collections, giant cells, and lymphocytes. (H & E, 150X.)

Reticulum staining showed the inflammatory granuloma with a typical appearance of borderline leprosy. In some areas reticulum fibers surrounded each cell, but in others such fibers were partly or completely absent. Epithelioid cells fusing together without any reticulum around individual cells were clearly seen (Fig. 7). P.A.S. stain showed thickening of the basement membrane of the seminiferous tubules. The acid-fast stain showed bacilli inside macrophages.

CASE 2 (S.L.R.S. 4755).—This patient, a male aged 45 years, reported first with a history of a hypopigmented patch on the back of his chest of three years duration. Subsequently, several lesions appeared on different parts of his face, trunk and extremities. He gave no history of contact with a leprosy patient before he acquired the disease.

Examination.—On examination, this patient was found to have on his face raised erythematous patches of different sizes but of indefinite borders, the surfaces scaly. On the front and back of the trunk, the buttocks, and the upper and lower extremities, there were multiple patches of different sizes symmetrically situated. All of these had a scaly and wrinkled surface. The borders of the lesions were indefinite, and seemed to coalesce one with another. The only patches that were anesthetic to light touch were those on the right thigh. The ulnar, lateral popliteal, posterior tibial, and greater auricular nerves were all enlarged, although there was no obvious paralysis. Both hands and feet were edematous.

Examination of the blood showed the hemoglobin level to be 12.25 gm. per cent. The urine showed no significant abnormality. Hookworm ova were found in the feces. The bacteriologic index was 1.87.

Clinical diagnosis.—The patient's condition was diagnosed as borderline leprosy in reaction, and he was admitted to the hospital.

Histopathology.—Biopsy specimens of the skin from the back of the chest showed marked atrophy of the epidermis, the rete pegs being flattened out. In the corium were focal collections of macrophages, epithelioid cells, and giant cells of the Langhans' type, collected mainly around skin appendages. Some nerve bundles showed intraneural infiltration by inflammatory cells. A few others showed perineural inflammation. The acid-fast stain showed bacilli inside macrophages and nerve bundles. The histopathologic picture of the skin lesions was entirely consistent with borderline leprosy.

The biopsy specimen of the testis showed fibrous thickening of the tunica vaginalis, which was infiltrated by focal collections of epithelioid cells and lymphocytes. The interstitial tissue was infiltrated by large numbers of these cells, and an occasional Langhans' giant cell was also seen. The inflammatory cells surrounded the tubules in some areas, and in a few other places there was intratubular infiltration

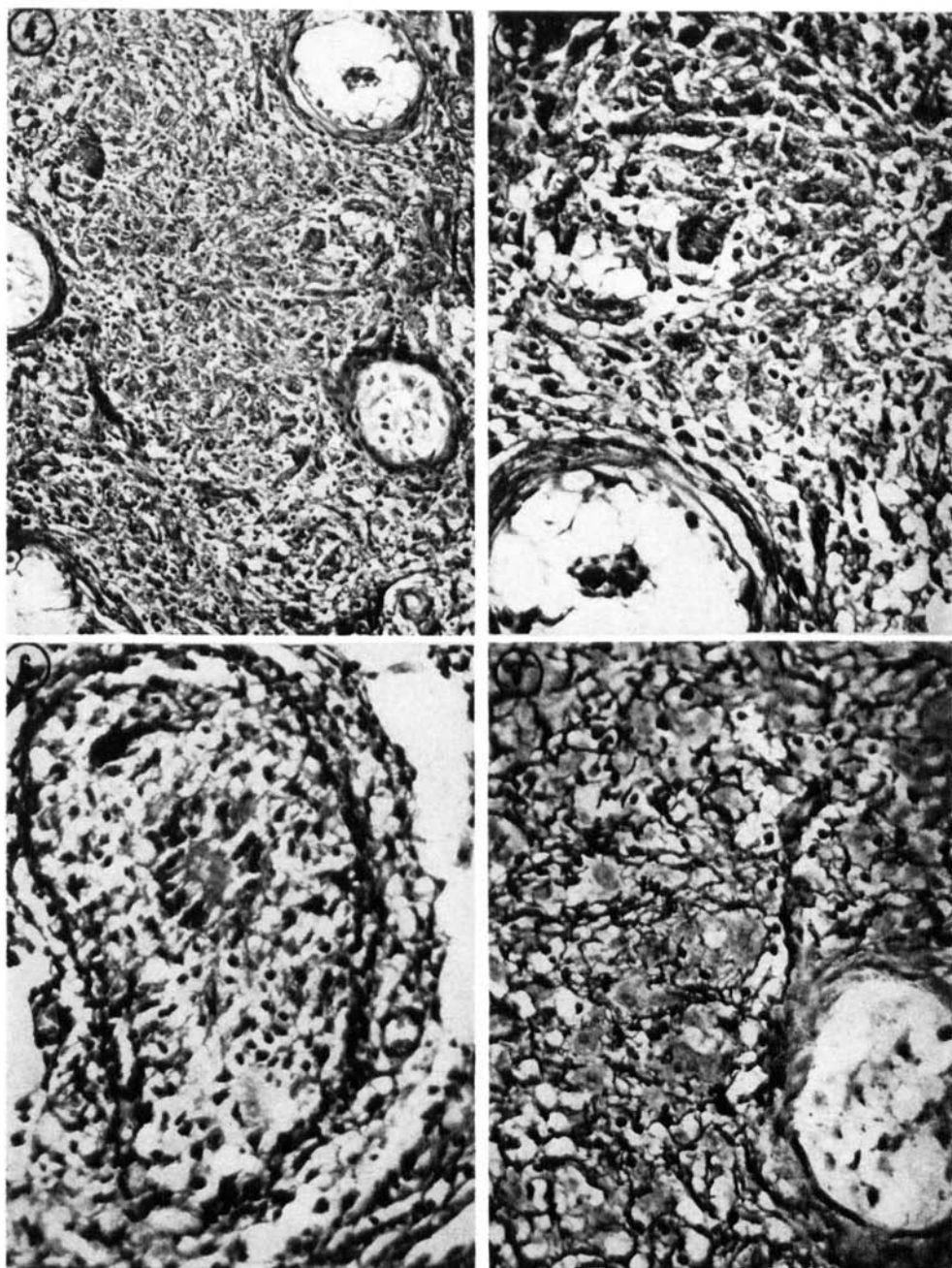


FIG. 4.—In the center of the field is an area of typical tubercloid change infiltrating the interstitial tissue. It is composed largely of epithelioid cells, with some lymphocytes and macrophages and a few poorly formed Langhans' giant cells. The few seminiferous tubules present, widely separated by the infiltrate, show atrophy and thickening of the basement membranes. (H & E, 150X.)

FIG. 5.—Higher magnification of an area from Fig. 4. Note the extensive vacuolation, usually fine-grained, of the epithelioid cells. (H & E, 300X.)

FIG. 6.—A seminiferous tubule filled by infiltrating epithelioid cells. A poorly-formed giant cell is seen in the center of the tubule. (H & E, 300X.)

FIG. 7.—Reticulum stain, showing preservation of reticulum fibers around macrophages and destruction of reticulum fibers in areas of epithelioid cell collections. This picture is typical of borderline leprosy. (Mag. 300X.)

resulting in complete destruction of the tubules. The tubules surrounded by inflammatory cells showed minimal thickening of the basement membrane. Others were apparently normal. There were many proliferating spermatogonia but no active spermatogenesis. Acid-fast staining showed bacilli inside macrophages and in the endothelium of blood vessels.

The reticulum stain showed reticulum fibers around macrophages, but in areas where there were epithelioid-cell collections and giant cells reticulum fibers were absent. The inflammatory-cell infiltrate was characteristic of borderline leprosy.

CASE 3 (S.L.R.S. 4691).—A male of 40 years reported to the hospital with a history of having developed swelling of both hands and feet 8 days after 5 days of oral treatment with hydnocarpus oil, given by an Ayurvedic physician. He had noticed raised patches on his left knee three months previously for which he had taken the Ayurvedic treatment. There was no history of contact.

Examination.—On examination, he was found to have multiple patches, symmetrically situated over the trunk and extremities. They were raised, irregular in size and shape, with borders that were vague and sloped away from the centers. Most of the lesions showed erythema, and some showed scaling and wrinkling. Some were anesthetic and some were not. There was pitting edema of both hands and feet. Both of the ulnar and the greater auricular nerves were thickened; the lateral popliteals and posterior tibials were not significantly enlarged. There was no obvious paralysis. The skin smear showed a bacteriologic index of 0.62. Hemoglobin was 12.80 gm. per cent.

Clinical diagnosis.—The patient's condition was diagnosed as borderline leprosy in reaction.

Histopathology.—The biopsy specimen of a representative skin lesion showed the epidermis to have undergone flattening of the rete pegs in most areas. In the corium were focal collections of epithelioid cells, macrophages and lymphocytes. The nerve bundles showed thickening of the perineurium, and perineural inflammation. The acid-fast stain showed bacilli inside nerve bundles. This histopathologic picture was quite consistent with the diagnosis of borderline leprosy.

A biopsy specimen of the testis showed a part of the tissue to be infiltrated by macrophages, epithelioid cells, lymphocytes and plasma cells. The tubules were infiltrated and completely destroyed. Remnants of them could be made out in the inflammatory-cell collections. There was some increase in vascularity. The remaining part of the testicular tissue showed only focal collections of inflammatory cells, mainly around blood vessels. A few tubules showed thickening of the basement membrane, while others showed no significant lesion. Active spermatogenesis was seen in some of the tubules. Acid-fast staining

showed numerous bacilli inside macrophages. Reticulum stain showed a borderline pattern.

DISCUSSION

In the 3 cases here reported, the inflammatory granuloma infiltrating the testis resembled histopathologically, the picture seen in the skin. There was a typical tuberculoid granuloma consisting of collections of epithelioid cells, giant cells, and lymphocytes infiltrating the testicular parenchyma. Macrophages containing a few acid-fast bacilli were also seen in every case.

The inflammation was mostly in the interstitial tissue, but the tubules were also invaded and destroyed. The tubules showed atrophy and thickening of the basement membranes, and some were replaced entirely by fibrous tissue. The Leydig cells in 1 case showed some hypertrophy and proliferation, but this condition was not significant in the other 2 cases. The tunica vaginalis was markedly thickened in all 3 cases, and was also infiltrated with epithelioid cells, lymphocytes and macrophages.

All the 3 patients of this study were clinically in reaction. It is reasonable to suggest that, previous to or during a reactive phase, there is a bacteremia. During this phase, bacteria are carried to different parts of the body through the blood stream. The testes are well-known to be an order of predilection for leprous lesions. It seems that during the reactive phase, when a large number of bacilli are in the blood circulation, the testes are infected in patients belonging to the tuberculoid type or to the borderline group of leprosy.

SUMMARY

Three cases of borderline leprosy in reaction, with tuberculoid granuloma of the testes, are presented. It is suggested that during the reaction phase, even in nonlepromatous cases, bacilli may be disseminated to different parts of the body including the testes, and may excite granulomatous inflammation where they locate.

RESUMEN

Son presentados tres casos de lepra limitrofe (borderline) en reacci3n, con granuloma tuberculoide de los testiculos. Se sugiere que durante la fase reaccional, aun en casos no lepromatosos, los bacilos pueden estar diseminados en diferentes partes del cuerpo incluso los testiculos, y pueden excitar donde est3n localizados una inflamaci3n granulomatosa.

RESUMÉ

Trois cas de lèpre border-line en r3action, avec accompagnement de granulomes tuberculoïdes des testicules, sont d3crits. La suggestion suivante est 3mise: durant la phase r3actionnelle, m3me chez les non-l3promateux, des bacilles peuvent 3tre dissimin3s dans diff3rentes parties de l'organisme y compris les testicules. Ces bacilles pourraient induire une inflammation granulomatense dans les endroits o3 ils parviennent suite 3 cette diss3mination.