OSSEOUS FORMATION IN LUPUS ERYTHEMATOSUS *

WILLIAM B. TRIMBLE, M.D.

Professor of Dermatology and Syphilology, University and Bellevue College, New York University; Attending Physician, New York Skin and Cancer Hospital

NEW YORK

The following case seems to be sufficiently unusual and to have enough interesting features, both clinical and microscopic, to justify recording it.

REPORT OF CASE

History.—Recently, the patient presented himself at the college clinic and the following notes were made: The man was of medium weight and height and apparently in good physical condition; aged 42, born in Austria, occupation, painter. His family history had no bearing on the condition; his father's death was caused by asthma at 62; his mother died of pneumonia at 58. There were ten children in the family—six boys and four girls—all alive and well, except one brother whose death was caused by "stomach trouble," as the patient expressed it.

His past history was of little consequence. He had been in excellent health all his life with the exception of the skin affection. At the age of 37, five years previous to this writing, he had noticed a small scaly lesion just in front of the left ear; this lesion persisted, was soon followed by others, the characters of which were scaling, dusky redness and central atrophy. At the time of examination, he had flat superficial scars on both sides of the face, which were frankly those of erythematous lupus. Three years after the outbreak on the face, he noticed quite accidently a large node in his right buttock.

Physical Examination.—Examination of this region revealed an area approximately somewhat larger than a silver dollar, of dull redness, in the center of which was a group of small atrophic macules. The border of the erythema was imperceptible, shading off into the healthy skin; a slight scaling was noticeable and some of the atrophic spots had coalesced, until it was safe to say that the whole central area was undergoing or had undergone atrophy.

This atrophied center must have been confined to the uppermost layers of the skin as it was extremely superficial, though typical from a clinical standpoint, in that it was a dead ivory white in appearance. There were no telangiectases observed. On attempting to pick up the skin between the fingers, it was noticed that it was attached to the underlying tissues and further palpation brought out the fact that a brawny, deep seated induration existed underneath. The whole lesion when surrounded by the fingers was of boardlike hardness and seemingly larger than a hen's egg. The outline was irregular, though it was about 4 inches in one diameter and 3 in the other.

The urine examination revealed nothing of consequence and the complement fixation reaction was negative.

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* Read before the Forty-Second Annual Meeting of the American Dermatological Association, held at Atlantic City, N. J., June 16-18, 1919.

HISTOPATHOLOGY

The first biopsy was a failure; it was done with a cutaneous punch, and did not go deep enough to get more than the superficial layers of skin; the specimen was however cut and stained, though reported as mildly inflammatory tissue with no definite diagnosis.

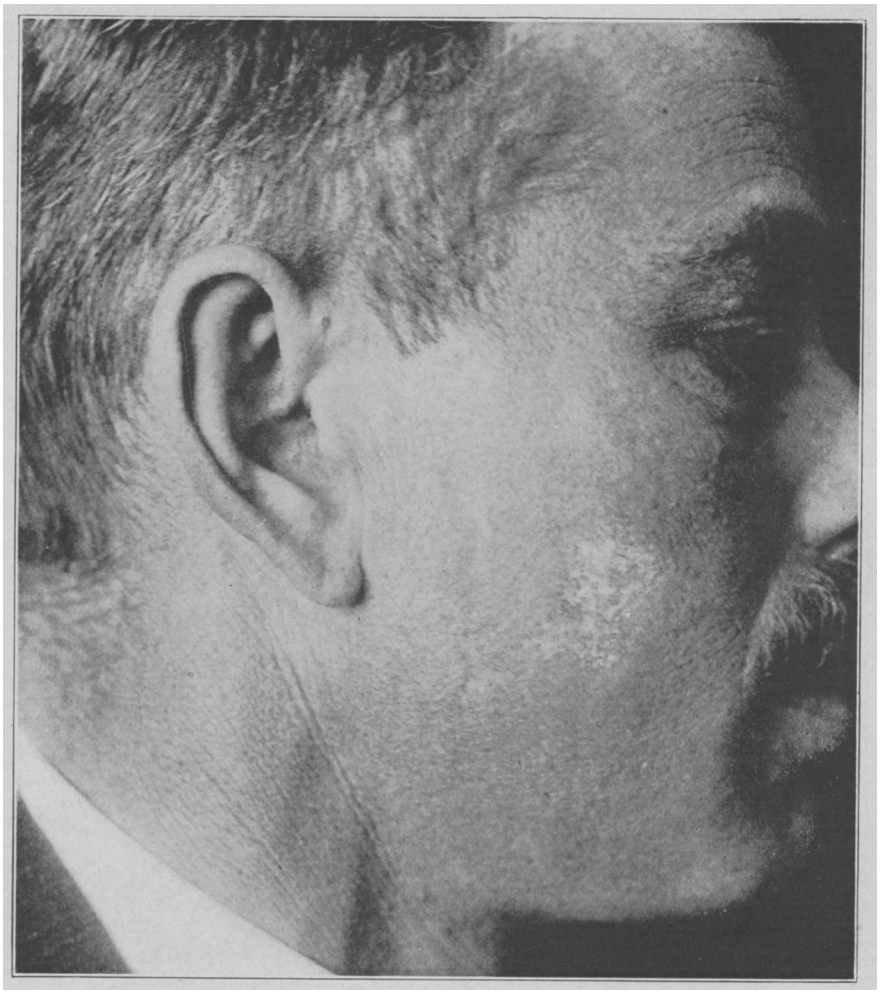

Fig. 1.—Lesion on right cheek. Superficial atrophy following healing of lupus erythematosus.

Another attempt was made to remove a piece of tissue by elliptical incision with a scalpel; this was more successful, though the incision was not as deep as desired, as the scalpel struck the hard mass, and no impression could be made on it.

The pathologic examination on this specimen was reported as chronic inflammatory tissue with calcareous deposits. The patient was eventually advised to have the whole mass removed by surgical operation; this was done in due time by Dr. George D. Stewart. The patient's recovery was uneventful.

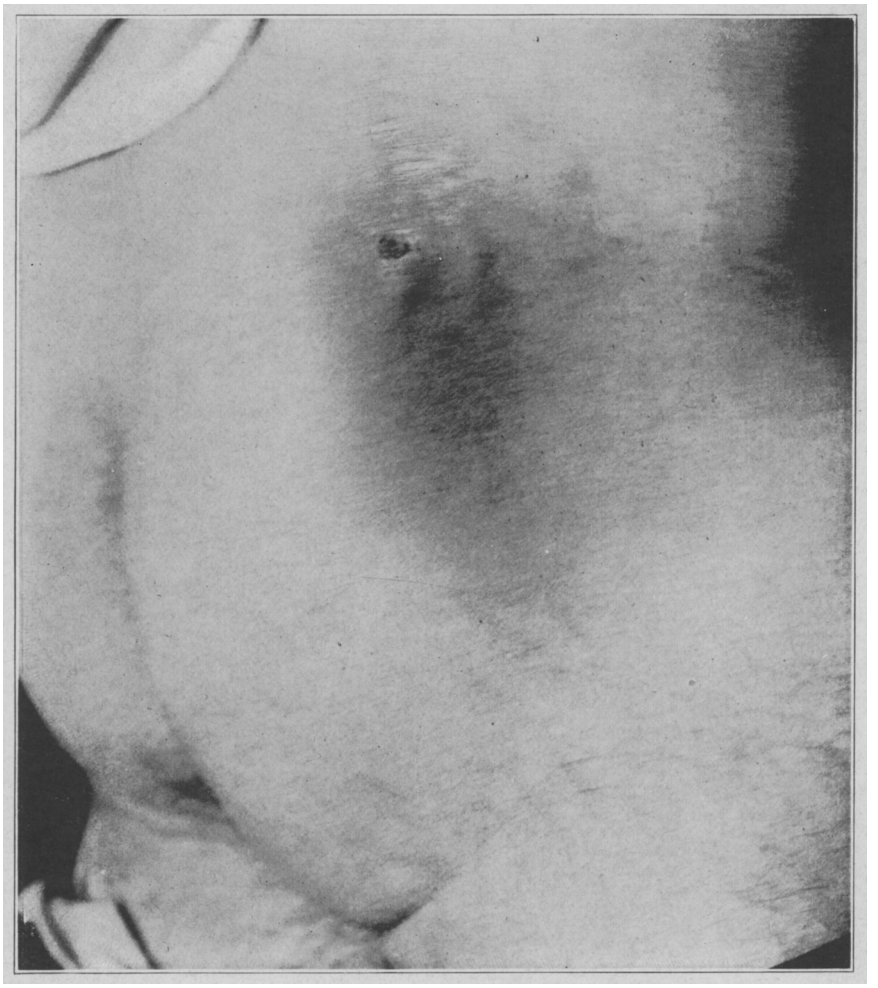

Fig. 2.—Original lesion on right buttock. Dark shadowed area represents dusky redness; superficial atrophy below. Small round crusted lesion marks biopsy punch.

The piece of tissue removed was about 4 by 3 inches in size and perhaps 2 inches deep. Viewed on the cut surface, one could see as well as feel the thin edge of what was apparently bony tissue. The bony layer, which was about on the level with the subcutaneous fat,

ran under the skin, throughout practically the whole specimen, and any attempt at puncture or incision from the top would strike this ledge.

Suitable pieces of this specimen were obtained for further study and a pathologic examination was made by Dr. D. S. D. Jessup.

MACROSCOPIC EXAMINATION

The gross specimen is a section from the skin with the underlying tissues measuring 10 by 8.5 by 1.5 cm. in depth. On section the tissue cuts with a gritty feeling, small areas of calcareous deposits being scattered through the deeper layers of the skin and subcutaneous fat.

MICROSCOPIC EXAMINATION

The epidermis presents a finely corrugated border in one portion, where the calcareous deposit lies deep in the corium and subcutaneous tissue. Where the deposit approaches nearer the surface the collagen takes a bluish stain and the epidermic border is slightly depressed; there is hyperplasia of the stratum mucosum and loss of the pegs in areas. In the areas of corrugated epidermis there is some hyperkeratosis; the depressions apparently corresponding to the mouths of the hair follicles, are filled with keratotic material. An occasional sweat gland duct is seen, but in the section examined, no hairs. The sweat gland coils appear normal in numbers; a few of the groups show edema of the cells.

The papillary bodies appear normal except that the elastic tissue fibers are decreased in number, and are practically absent where the pegs are compressed or missing. In the area above referred to, where calcareous material is found near the papillary layer, there is marked infiltration of the upper portion of the corium with round cells and cells which are probably necrotic. The supporting tissue is small in amount. The tissue immediately beneath shows irregular shaped islands of calcareous material located sometimes in dense fibrous tissue, sometimes in fibrous tissue of looser structure, more cellular, with fatty tissue in the immediate neighborhood, and sometimes in areas in which there is evidence of hemorrhage.

Beginning around the sweat coils and smaller blood vessels, through the entire specimen there is round cell exudation. This appears sometimes as small islands of round cells with a few blood vessels, the lumina of some of which are invisible. This round cell exudation does not bear a definite relation to the calcareous deposit. Elastic tissue fibrils are scattered throughout as are also collagen fibrils. No elastic tissue fibrils are found in the areas of round cell exudation.

CHEMICAL EXAMINATION

The chemical examination was made by Prof. John Mandel, who reported the tissue as bone formation. His method, briefly, consisted of allowing the specimen to soak for a long time in dilute hydrochloric acid to dissolve bone tissue.

Fig. 3.—Linear scar following excision operation.

This solution was treated with ammonium oxalate, which causes a precipitate of calcium oxalate. Ammonium molybdate added to the original solution will prove the presence of calcium phosphate by precipitation, and calcium carbonate by effervescence.

All of these tests were positive, consequently his report.

COMMENT

Several diagnoses were taken into consideration at the original clinical examination. They were: organization, fibrosis, etc., following old inflammatory changes; deep seated sarcoid; localized scleroderma, and lupus erythematosus. All of these ideas, with the exception of the first, were based merely on visualizing the lesion, taking into consideration the color, scaling and atrophy. The first — fibrosis and organization — seemed a reasonable diagnosis, as such changes could even account for the superficial atrophy from pressure on the skin capillaries; the patient, however, denied emphatically any injury or intramuscular injection, which caused the supposition to be discarded. Sarcoid was brought to mind on account of the depth of the lesion and the color, though mainly because the writer had seen several cases of the Darier-Roussy type, with typical lesions of erythematous lupus on the face; it was not given very serious consideration, as a single lesion of such ivory-like hardness in that locality was not especially characteristic.

The density and sclerotic nature of the lesion with the epidermic atrophy pointed strongly to the diagnosis of localized scleroderma; it was impossble, however, to get away from the fact that inflammation existed, and scleroderma is an atrophy and not an inflammation. As already mentioned, the skin over the induration was dusky red, scaly and atrophied, so the remaining alternative was to believe all the existing lesons — that is, those of the face and buttock — to be the same; and this was the ultimate conclusion. Of what the indurated mass consisted remained in obscurity until a rather elaborate study had been made, the result of which has already been given. Whether to look on all the lesions as one process, or whether to consider those of the face as lupus erythematosus and the one on the buttock as an entirely different entity, was the question to be solved.

This has been solved in part by later developments in the case yet to be described. The patient was kept under observation and after a lapse of some months the lesions on the face began to take on renewed activity, new ones rapidly appeared, spread to the neck, shoulders, arms and hands and in a very short time the case developed into one of typical erythematous lupus of the so-called disseminate variety. Simultaneously with this new outbreak, inflammatory action again appeared in and around the scar on the buttock, and the integument in that location assumed the same appearance as the other lesions, with subacute inflammation and scaling.

This phenomenon seemingly was fairly good clinical evidence that the buttock lesion was erythematous lupus; it could not be proved pathologically, although another section of skin was removed following

the recurrence; after all, the microscopic picture of lupus erythematosus is not so characteristic that it could furnish absolute proof.

No attempt was made to review the literature; if any one has reported a similar case, priority will not be contested.

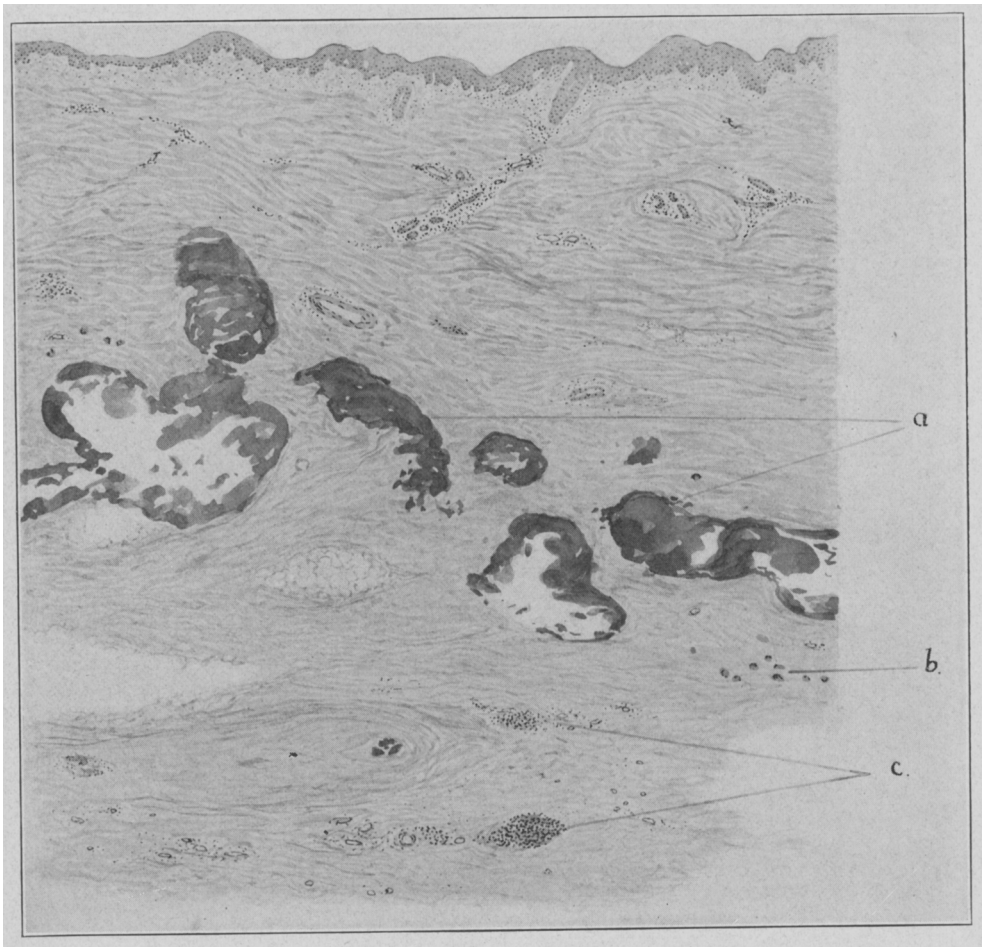

Fig. 4.—Calcareous deposits, *a;* scattered giant cells lying in the connective tissue near the calcareous deposits, *b;* areas composed of small round cells, *c.*

DISCUSSION

Dr. Pollitzer was of the opinion that the paper offered not the slightest proof of bone formation. He believed it was a case of the deposit of lime salts, which was very interesting as an observation though not very rare; but the title of the paper was misleading. The chemical examination simply proved the presence of a calcareous deposit and not the formation of bone. The microscopic examination was the only thing that could prove the formation of bone and that had not been presented. The formation of bone in the skin was very rare, but had been recorded in a small number of cases. A few years ago he had published a unique case of scleroderma in which there was

bone formation in the skin, clinically visible and microscopically demonstrated. Calcareous deposits in the skin occurred in many conditions; they should not be confused with bone formation.

DR. ZEISLER stated that he had only once come across anything that reminded him of the case which Dr. Trimble had presented. Several years ago he was consulted by a young woman who had a peculiar spot on her forehead. He could not say whether it was a cyst or something else, but began to pick at it and to his great amazement dug out a small bony mass. It was perfectly round and easy to take away from the surrounding tissue and was about the size of a three carat pearl. This was the only thing of the sort that he had ever seen.

DR. LITTLE thought that Dr. Pollitzer had made a very good correction of the title. He had seen cases of diffuse calcinosis, and that was probably what this case was.

DR. TRIMBLE believed that the tests he had related, taken with the clinical evidence, was enough to prove that the growth was bone. He was under the impression that calcareous deposits could be cut through with a strong, sharp knife. In this case, the knife made no impression on the growth. The chemist was positive in his opinion that he was dealing with bony tissue. The association with lupus erythematosus was to his mind the interesting feature. Whether it was directly connected with the pathologic process of that disease, or whether it was an independent lesion occurring in a patient with lupus erythematosus, was a question. He was firmly of the belief, however, after seeing and treating the case and having the various tests made, that it was bone.