features. The condition in many respects resembled polyarteritis nodosa, but differed from it in that the vascular lesions were limited to the veins and in the presence of giant cells.

A full account of the clinical and laboratory findings is given and the differential diagnosis discussed.

The patient is alive and is being followed up.

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SPONTANEOUS MYXOEDEMA DEVELOPING AFTER THYROTOXICOSIS

BY

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Primary adult myxoedema most commonly occurs from idiopathic primary atrophy, but may also follow thyroidectomy, excessive radio-iodine dosage, or destructive thyroid disease such as lymphadenoid goitre (Hashimoto's thyroiditis). It is also frequently stated in textbooks that adult myxoedema may arise in patients who have previously had frank thyrotoxicosis ("burned out toxic goitre"), but it is exceedingly hard to discover records of such cases. Crotti (1938) describes one, but gives the impression that he has seen many more: he intimates that the progression from hyperthyroidism to hypothyroidism takes many years. McGavack (1951) describes the case of a patient who received only iodine treatment for his thyrotoxicosis but developed myxoedema some four years later. In view of the paucity of cases recorded, we are reporting one which was first treated for thyrotoxicosis with antithyroid drugs for 21 months; an active thyroid gland was still present at the end of this time. Twelve months later the patient was definitely myxoedematous.

Case Report

A married man aged 40 first presented in November, 1950, with an 18-months history of increasing fatigue, intolerance of hot weather, and 3 st. (19 kg.) loss of weight. He also complained of some dyspnoea on exertion and of some prominence of the eyes in the past two months. His appetite was very good. On examination at that time he was a thin, flushed, and anxious man (Fig. 1 A) with a fine tremor of the fingers, an increased peripheral circulation, and a slightly enlarged soft thyroid gland with a bruit. There was no true exophthalmos, lid retraction, or lid lag, but a definite "stare." There were no other physical signs. Investigation confirmed the diagnosis of hyperthyroidism (Fig. 2). The basal metabolic rate ranged from 140 to

160% of normal according to the Boothby standards (52 to 60 cal./square m./hr.). A radio-iodine test yielded only 10.7% excretion in 48 hours ("T" index of thyroid uptake = 105 compared with the normal of 3-13) (see Fraser *et al.*, 1953).

After a short course of an anti-thyroxine drug (butyl-4-hydroxy-3:5-diiodobenzoate) (Fraser and MacLagan, 1953), and after an episode of pyrexia which was found to be due to otitis media, the patient was discharged on methyl-thiouracil, 600 mg., and thyroid, 1 gr. (65 mg.), daily. He responded rapidly to treatment, gained weight, and remained well controlled on diminishing doses of thiouracil from January, 1951, to October, 1952. Antithyroid drug treatment was then stopped, and a radio-iodine test done a few days later showed that the gland was still active in that the 48-hour excretion was 16.7% (T=62). He was then given a short course of potassium iodide and re-tested in February, 1953, when the 48-hour radio-iodine excretion had risen to 77% (T=2.6). This was attributed to excessive ingestion of iodides as the patient appeared

Fig. 1.—Photographs taken in November, 1950 (A), and in January, 1954 (B).

	1950	1951	1952	1953	1954
	JAN.	DEC. JAN.	SEP. OCT.	FEB. MAY.	JAN.
B.M.R. %	+43 +45 +52				-27 -31 -33
Blood Cholesterol	177mg.%				391mg.%
48hr. ¹³¹ I Excretion.	10.6 %			16.7% 77%	79% 83 %
"t"	105			61.7 2.6	1.4 2.0
E.C.G.					
Weight in stone.	9.9	10.12	12.3	12.9 12.11	13.11
Anti-Thyroid Drug.					

1950

1951

1952

1953

1954

JAN.

DEC. JAN.

SEP. OCT.

FEB. MAY.

JAN.

Fig. 2.—Results of investigation.

euthyroid, but when the test was repeated three months later it yielded a similar result, and when he was seen again in October, 1953, he had all the features of myxoedema—full puffy facies, husky voice, and dry skin—and his weight had risen to 13 st. (82.5 kg.) (Fig. 1 B). On admission for reinvestigation it was found that his blood cholesterol had risen to 391 mg. per 100 ml., his basal metabolic rate had fallen to -28 to -33% (25-28 cal./square m./hr.), his protein-bound iodine was 2.8 µg. per 100 ml., and he had

electrocardiographic evidence of myxoedema. There was no evidence of pituitary failure: he had a normal 17-ketosteroid excretion, and a normal diuresis in response to a standard water load.

Conclusion

Our data indicate that this patient had Graves's disease in 1950; that his thyroid gland was still functioning at the end of 1952, after treatment with thiouracil; and yet that one year later he had advanced primary myxoedema, which has since responded to thyroxine. We have been unable to find a record of any other case of myxoedema developing spontaneously so soon after thyrotoxicosis.

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SEZARY'S SYNDROME

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Disturbances of the reticulo-endothelial system associated with generalized erythrodermia are well known, and form a diverse group, from which Sézary and Bouvrain (1938) first described a distinct syndrome. This is characterized clinically by bizarre features, and pathologically by the appearance of peculiar monocytoid cells in the blood. Sézary (1949) reviewed all the four published cases (Sézary and Bouvrain, 1938; Sézary, Horowitz, and Maschas, 1938; Baccaredda, 1939; and Sézary and Bolgert, 1942), to which Wilson and Fielding (1953) have added one more—the first reported in Britain.

The purpose of this paper is to draw attention to this syndrome by recording the clinical and pathological features of two further cases, and to discuss their relation to the reticuloses.

Case 1

The patient, a widow aged 77, previously led an active, healthy life. She had had smallpox at the age of 11 years and pneumonia at 73. Seven sons were alive and well; one premature infant was stillborn. About eight years ago she first suffered from attacks of extensive erythrodermia "just like measles," with varying degrees of exfoliation, and intense pruritus, starting in the feet and spreading over the limbs and trunk. These attacks were transient initially and were preceded by malaise. Later all her skin assumed a mottled appearance. Her palms were occasionally affected so severely that she was unable to do her housework. The mucous membranes were not affected, nor was there any history of excessive sweating, although she felt the cold intensely.

Some twelve months ago she became aware of "hot aches" and stiffness of the fingers, with distortion and looseness of the nails—so much so that she was unable to relieve the intense pruritus by "having a good scratch." Her skin also showed scattered patches of both leucodermia and brown pigmentation. Alopecia was complete apart from

occasional tufts on the scalp. Recently oedema of the legs occurred, and infected excoriations have resulted in whitlows, and superficial ulcers on the ankles and elbows.

When first seen in 1950 she showed extensive erythema, fine desquamation, telangiectasia, and superficial lymphadenopathy. Various diagnoses were then considered, including parakeratosis variegata, pityriasis rubra pilaris, poikiloderma vasculare atrophicum, and lymphosarcoma. Skin and blood examinations at that time were equivocal. The presence of atypical white cells in the blood was noted, but their significance was not realized.

Clinical examination showed an elderly woman whose bizarre appearance is well illustrated by Fig. 1. The skin showed a generalized mottled erythrodermia with fine scaling and interspersed areas of slaty-brown pigmentation and leucodermia of varying sizes and indefinite distribution. Petechiae were maximal over the sternum and shins, the latter also showing erythema ab igne. The hair was absent apart from residual tufts on the scalp and a few remaining

FIG. 1.—Face and hands of Case 1.

hairs on the pubes. The eyelids showed chronic blepharitis and ectropion of the lower lids. The nose was shrunken and the nares were widened. The joints of the wrists and knees showed arthritic changes. There were oedema, ulnar deviation, and flexural contractures of the digits of both hands: full extension, active or passive, of the fingers of the right hand was impossible. The nails and terminal phalanges of the left hand were almost entirely absent; the thumb was normal. The changes were less marked in the right hand, although all fingers showed some degree of deformity. The lymph nodes in the axillae and groins were all enlarged, firm, rubbery, and discrete. There was one similar lymph node in the right cervical area. The mucous membranes were normal.

No significant lesion has been revealed in any other system. The spleen was not felt. The liver edge was palpable just below the costal margin. There was slight oedema of the legs. B.P. 170/80. Weight 7 st. 12 lb. (49.9 kg.). The temperature and pulse were normal. X-ray films of the skull, spine, pelvis, and chest were essentially normal. The mental condition was that of a well-adjusted euphoric elderly woman.

Case 2

The patient, a married woman aged 40, had suffered from eczema as a child. Her appendix had been removed at the