

LIPO-ATROPHY OF THE ANKLES AND ITS RELATION TO OTHER LIPO-ATROPHIES

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Abstract. Two typical cases of lipo-atrophy of the ankles are described, one case of atrophy of the ankles with associated atrophy of the whole extremity, and one case of localized atrophy of the knee. For comparison, we present one case of atrophy in a diabetic patient due to insulin injections but developing at distant sites, and in one case, a child in whom atrophy followed antibiotic injections. Primary inflammatory vascular changes in the subcutaneous tissue were demonstrated in atrophy of the ankles, as in the early period of insulin-induced lipo-atrophy. Attention is called to the possible relation of the described lipo-atrophies.

Key words: Localized lipo-atrophy; Insulin-induced lipo-atrophy; Lipo-atrophia annularis; Primary atrophy of the subcutaneous tissue; Lipo-atrophia semicircularis; Panatrophy Gowers'

Lipo-atrophy of the ankles was first described by Shelley & Izumi (7). Atrophic changes at the ankles were observed in a 6-year-old girl. They were preceded by a non-inflammatory swelling of the subcutaneous tissue.

In view of its annular appearance (with changes encircling one ankle in the form of a complete ring and an incomplete one around the other ankle), the lesions corresponded, in fact, to the primary atrophy of the subcutaneous tissue described under the name of lipo-atrophia annularis (3, 4).

In the three described cases (3, 4, 7) focal lesions of annular lipo-atrophy were present on the upper extremities unilaterally or bilaterally, exclusively in women. In some cases they were preceded by swelling of the subcutaneous tissue, associated with a red-cyanotic discoloration of the skin. The duration of the changes varied; in some cases they regressed and then recurred. Another three cases described as lipo-atrophia semicircularis (5) differed from annular lipo-atrophy since no complete rings were present and the focal lesions were more numerous, unilateral or bilateral, located mainly on the arms,

usually laterally. All cases were observed in young women and no provoking factors could be demonstrated.

The case of annular atrophy of the ankles reported by Shelley & Izumi differs from annular lipo-atrophy only in its location. Apart from this, however, the similarity is striking, i.e. the atrophy involves mainly the subcutis, while the overlying skin is intact.

The patients have no neurologic disorders and are in good general health. The disease causes neither contractures nor difficulty in walking.

The aetiological factors have not been elucidated. Finding marked inflammatory vascular changes, Ferreira-Marques has put forward a hypothesis that damaging external factors, especially pressure, may be responsible for the disease. Bruinsma believes that the disease may be associated with an abnormal vulnerability of the fatty tissue which responds to pressure or repeated trauma with an inflammatory reaction followed by sclerosis and atrophy of the subcutaneous tissue. Shelley considers that the atrophy may represent a cutaneous symptom of systemic disease—endocrinological, neurological or metabolic, with the possibility of latent diabetes. Geschwandter & Münzberger classify lipo-atrophia semicircularis as an idiopathic localized lipo-atrophy, in contrast to localized lipo-atrophies due to external factors—repeated trauma or injections of insulin, corticosteroids, etc.

It is the purpose of this report to describe further cases of lipo-atrophy of the ankles which is, probably, a not so exceedingly rare disease. In one case we had followed up and examined histologically primary changes preceding the atrophy. Another purpose was to demonstrate a relation of lipo-atrophy of the ankles to annular lipo-atrophy and lipo-atrophy due to insulin injections or trauma.



Fig. 1. Case 1. Lipo-atrophy of the ankles in a 15-year-old girl.

CASE REPORTS

Case 1

The patient was a 15-year-old girl with a history of rheumatic fever. Several months prior to examination, she noticed oedema of the ankles in a stocking-like arrangement, followed by atrophy. In the early stage, flat subcutaneous nodules appeared on the lower legs.

Annular atrophy of subcutis was pronounced, mostly on the distal parts of legs and on the feet (Fig. 1).

Laboratory investigations showed accelerated ESR (15 mm after 1 hr and 40 mm after 2 hours), antistreptolysin level 500 U, increased IgG and IgM in immunoelectrophoresis. The blood glucose curve and 24-hour glycaemic profile were normal. Vascular function tests, sensory chronaxy, and electromyography, failed to demonstrate any abnormalities.

Histological examination (biopsy from a flat nodule): abundant inflammatory infiltrations in the subcutis, composed of lymphocytes, histiocytes, numerous fibroblasts and macrophages; inflammatory lesions of small arterioles and venules; numerous newly formed blood vessels; young connective tissue replacing fat lobules (Fig. 2). No evident abnormalities were found in the corium.

Course of the disease. The nodules resolved but atrophy progressed slowly. Procaine penicillin, penicillamine (1.5 g daily) for one month, and vitamin E (300 mg daily), had no effect.

Case 2

The patient was a 35-year-old woman with a history of intervertebral disc prolapse 6 years prior to examination. Atrophic changes began about 1 year later and spread gradually, involving both lower extremities.

Laboratory investigations demonstrated no abnormalities.

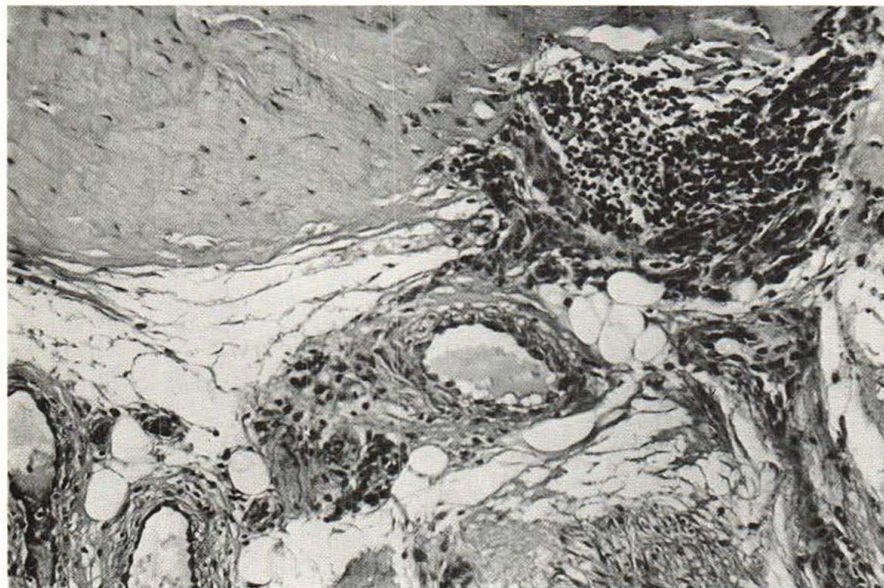


Fig. 2. Case 1. Histology of early flat nodule. Inflammatory infiltrations and inflammatory lesions of small vessels in the subcutaneous tissue. H + E, $\times 180$.

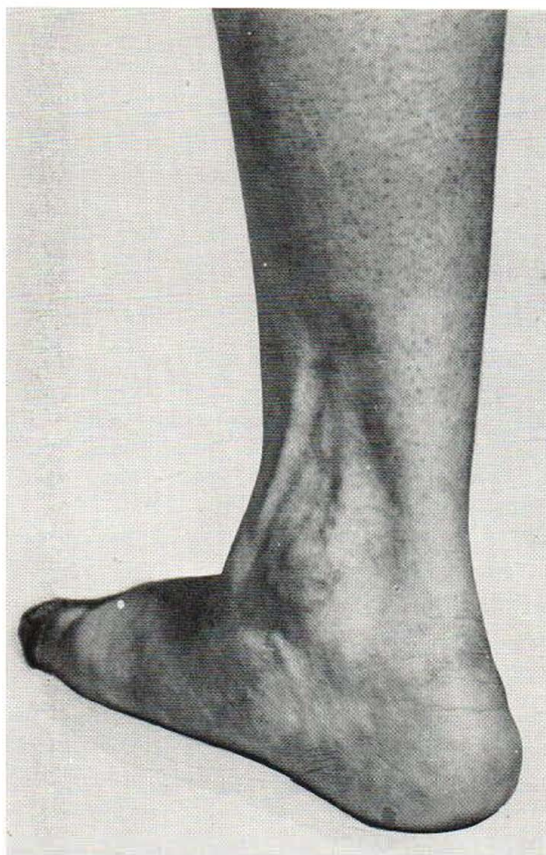


Fig. 3. Case 2. Lipo-atrophy of the ankles in a 35-year-old woman.

Radiograms of the spinal column, EMG, vascular function tests, and sensory chronaxy, were normal.

Course of the disease: the atrophy spread very slowly and remained limited practically to the region of the ankles (Fig. 3).

Case 3

A 19-year-old girl who noticed, since the age of 7 years, atrophies on the right leg, without previous indurations.

We found atrophy of the whole right leg from the knee downwards with changes in the ankle region, of the same type as the atrophy of the ankles (Fig. 4).

Laboratory investigations, radiography of leg bones, vascular function tests, sensory chronaxy and electromyography: normal.

Case 4

Localized panatropy. A 39-year-old woman noticed atrophies of the right thigh around the knee, which had been slowly progressing for the past 10 years but which were not preceded by indurations (Fig. 5).

Laboratory investigations failed to disclose any abnormalities. Radiography of the vertebral column demonstrated a narrowed intervertebral space between L₄ and L₅ with

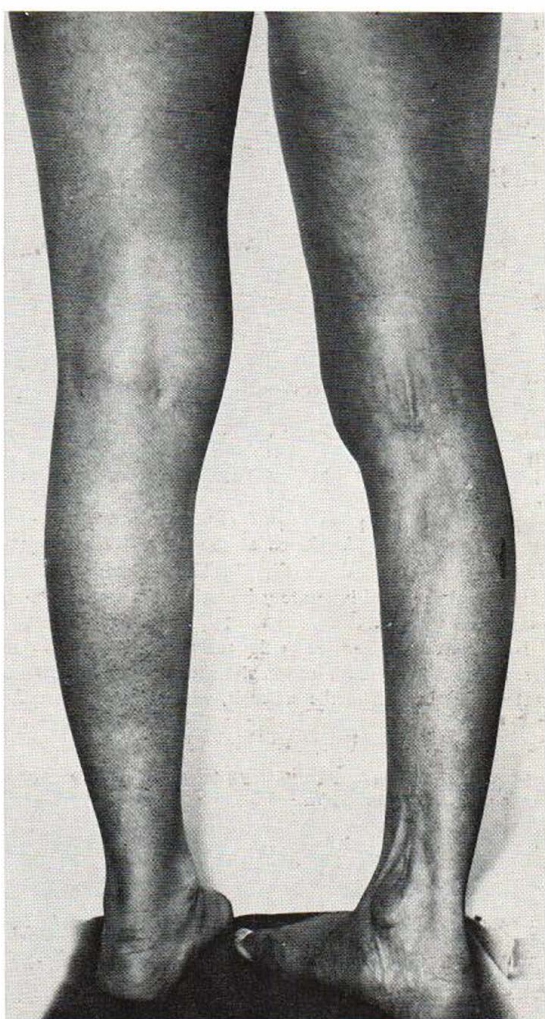


Fig. 4. Case 3. Atrophy of the right leg with marked atrophy of the ankles in a 19-year-old girl.

suspected herniation of the nucleus pulposus. No neurological abnormalities were found. Vascular function tests were normal.

Case 5

Localized lipo-atrophy. A 4-year-old girl, who at the age of two had had several injections of antibiotics. Some months afterwards, atrophy developed in the gluteal region (Fig. 6). Atrophies were not preceded by inflammation or induration, and spread slowly. The general health of the girl was good.

Laboratory investigations were normal. No evidence of latent diabetes was found in the child or her family.

Case 6

Diabetic lipo-atrophy. The patient was a woman aged 30 years with a history of diabetes since the age of 18. Post-

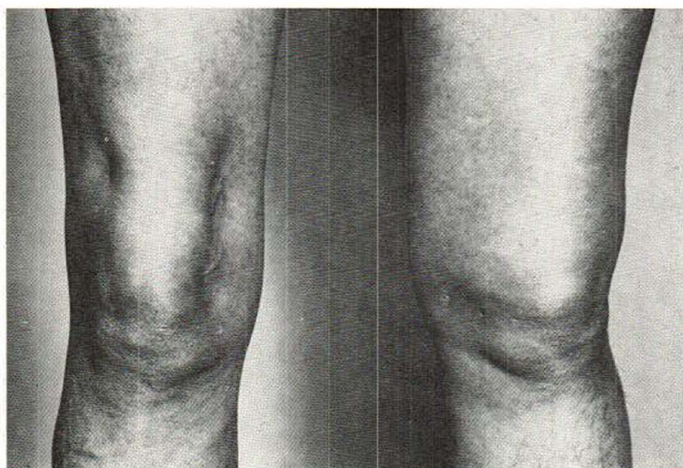


Fig. 5. Case 4. Localized panatrophy of the right thigh around the knee in a 39-year-old woman.

insulin lipo-atrophy developed at the site of insulin injections and in distant areas (Fig. 7). The general health was good. The results of laboratory investigations were normal, apart from diabetes.

Histological examination in the early stage of atrophy in areas distant from the injection site showed marked inflammatory infiltrations in the subcutis, with sparse giant cells,

fat replacement by fibrous connective tissue, and abundant newly formed capillaries, as well as inflammatory changes in arteries and small veins (Fig. 8).

DISCUSSION

Cases 1 and 2 are typical forms of atrophy of the ankles. In case 1 it was possible to demonstrate primary flat nodular infiltrations which histologically

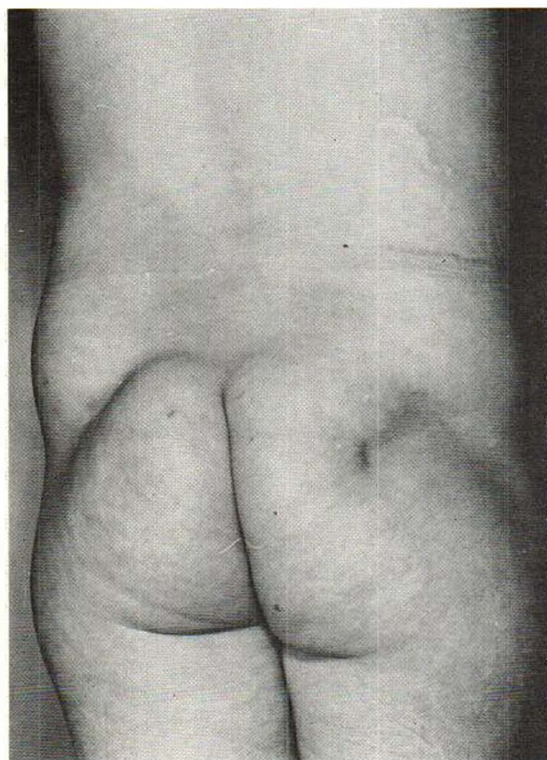


Fig. 6. Case 5. Localized lipo-atrophy of the buttocks in a 4-year-old girl.

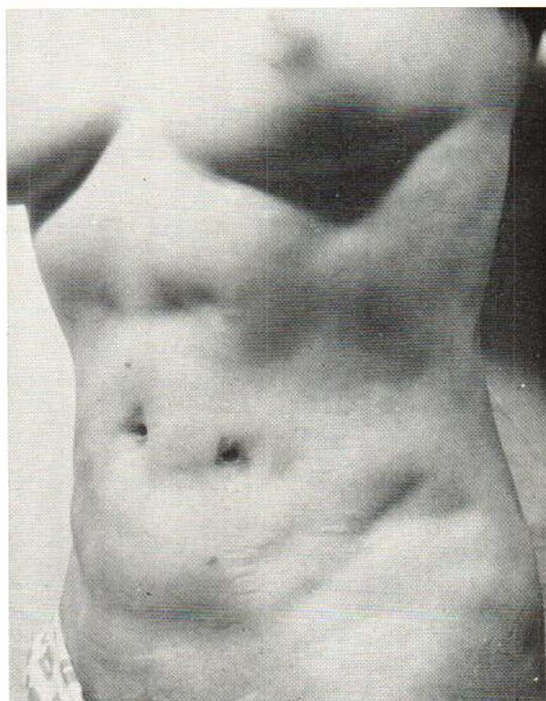


Fig. 7. Case 6. Diabetic lipo-atrophy in a 30-year-old woman.

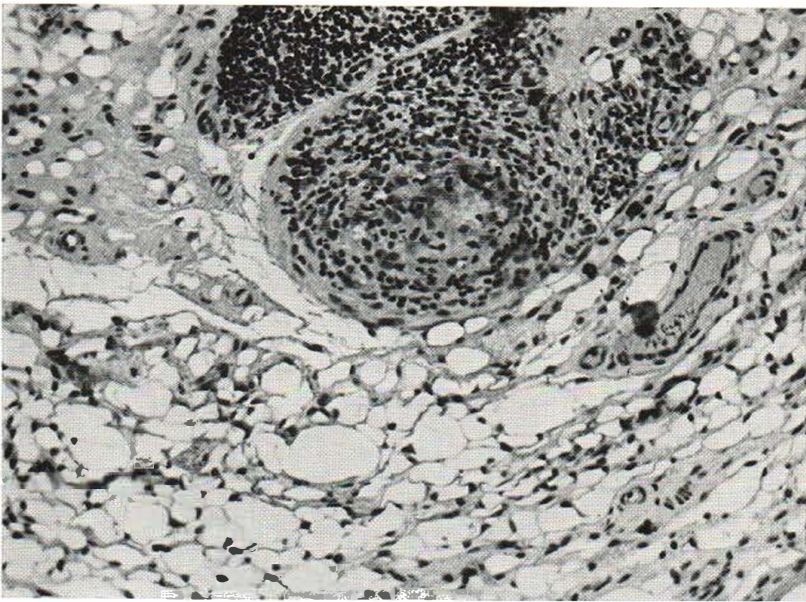


Fig. 8. Case 6. Histology of the diabetic lipo-atrophy (biopsy taken in a place distant from injection sites). Marked inflammatory infiltrations and vascular changes in the subcutaneous tissue.

corresponded to inflammatory vascular changes as reported by Ferreira-Marques (4), and, in sections with less marked inflammation, to those reported by Bruinsma (3).

It seems that the failure of Geschwandtner & Münzberger to demonstrate evident vascular involvement, and the far less intense inflammatory symptoms present in their cases, was due to the already complete atrophy.

The similarity of histological findings in our case to insulin-induced lipo-atrophy was striking and it seems that all lipo-atrophies reported here have primarily an inflammatory and vascular character. In the case of Shelley & Izumi (7) histological examination was not carried out. In our case 1, very early nodular lesions speak in favour of an inflammatory origin of this particular lipo-atrophy, as also do the remaining lipo-atrophies.

Case 3 shows that atrophy of the ankles may be associated with a more extensive atrophy involving the whole leg and the region of the knee.

Case 4 shows that the same form of lipo-atrophy, not necessarily circular, may affect the knee.

Cases 5 and 6 are lipo-atrophies related to injections, i.e. to trauma, although atrophies in the diabetic patient also developed in areas distant from injections as well, and in the child they spread far beyond the injection sites.

We have no photographic documentation but we observed several cases of incomplete annular changes

corresponding to lipo-atrophia semiannularis (5) localized either on arms or on thighs. These changes were—as in their cases—transient and in two cases it was possible to demonstrate the causative role of trauma (prolonged pressure during washing of linen). We consider that in semiannular lipo-atrophy as well as in atrophy of the ankles, the vulnerability of subcutaneous tissue as postulated by Bruinsma plays a significant role. In diabetes this vulnerability is particularly marked, probably due to vascular involvement. It is possible that certain metabolic disturbances and hormonal factors increase the susceptibility of the subcutaneous tissue, since changes of this type have so far only been reported in women. In our 5 cases, we found no diabetes in the patients or in their families. One patient had a history of rheumatic fever and in two, prolapse of nucleus pulposus was found. No neurological changes were demonstrated and sensory threshold and sensory chronaxy, as well as electromyography, failed to demonstrate any abnormalities. It must be remembered, however, that in cases reported by Bruinsma and Geschwandtner & Münzberger, rheumatic pains were present in patients or in their families.

Annular lipo-atrophy and atrophy of the ankles resemble Gowers' panatropy. The only difference is the coexistence of skin atrophy and the deeper location of changes in panatropy, since the lesions may also involve the muscles (hence the name

cutaneomuscular panatropy). However, electromyographic changes were found in one case of lipoatrophy reported by Bruinsma and, conversely Gowers' panatropy frequently fails to cause muscle involvement, and cutaneous changes may be minimal. Gowers' panatropy is a controversial disease entity (1, 2, 6) since, in a considerable part of reported cases, the atrophy was probably due to an involutionary morphea or subcutaneous morphea. By contrast, annular lipo-atrophy and atrophy of the ankles differ markedly from localized scleroderma and may be classified as panatropy only on the condition that the term "panatropy" is used as a general designation of deep atrophies related to idiopathic atrophies of unknown origin.

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Received August 9, 1974

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