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Factitious Lymphoedema, Secretan's Syndrome

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Abstract. Chronic lymphoedema can be self-inflicted in origin. Many different procedures can produce such chronic oedema. Three probable cases of factitious oedema are reported.

Key words: Pathomimia; Factitious oedema; Chronic lymphoedema; Secretan's syndrome

Secretan was a Swiss insurance physician who in 1901 described 11 patients suffering from persistent hard oedema of the dorsum of the hand (15). He noticed that in all his patients the oedema developed secondarily to a trauma which was not severe enough to produce a fracture. Furthermore the patients were covered by insurance. Since then several similar cases have been demonstrated to be of self-inflicted origin.

The purpose of the present paper is to draw attention to the diagnosis factitious lymphoedema by presenting a further three cases and a short review of the literature.

Patient 1

A 35-year-old female developed secondarily to a minor trauma of her right knee chronic painless lymphoedema of her right lowerleg. Furthermore she had severe excoriations on her lower legs and forearms. By physical investigation the lymphoedema was seen to be sharply demarcated proximally just below the knee by a circumferential furrow with erythema and excoriations (Fig. 1). She had had neither erysipelas nor deep thrombophlebitis. Lymphography, thorax X-ray and urography were normal. It was not possible to perform phlebography. A psychiatric investigation concluded that she suffered from a neurosis characterogenes.

Following the application of an occlusive protective casting, the lymphoedema regressed markedly. When this treatment was interrupted, exacerbation was noticed, with lymphoedema recurring initially proximally just below the demarcation ring.

Patient 2

A 48-year-old female was admitted because of a chronic painless oedema of her right hand and dorsum of the right forearm of 3 years' duration. The lymphoedema started in connection with a stay in hospital because of backache and myoses. Because of the lymphoedema she had been admitted to departments of orthopedic surgery and blood vessel surgery, but no treatment was suggested. For 7 or 8 years she had also had moderate eczema on her palms. Patch tests for chromate and nickel proved positive. A peroral provocation test with nickel chloride, 1 and 2 mg, proved negative. She had never had erysipelas or thrombophlebitis. For many years she had suffered from a paranoid psychosis and was treated with Perfenazine (Trilafone®, Schering), Chlorpromazine (Prozil®, Dumex) and Lysantine (GEA).

It was not possible to perform phlebography or lymphography. Mammography showed fibroadenomatosis bilateralis, which was also noticed at the physical examination. No lymph nodes could be palpated in her axillae.

Patient 3

A 32-year-old female was observed for 6 years, initially because of dyshidrotic eczema of her third left finger, subsequently also including the palms. An attack of lymphangitis of her left arm had been treated with Penicillin. During the last 5 years she has suffered from a constant, painless oedema of her left hand, dorsa of the fingers and the distal part of her forearm. Treatments with mitella, prednisone perorally and a protective plaster casting have been ineffective. Investigations for malignant tumours have been negative. Automutilation and referral to a psychiatric department have been mentioned to the patient but caused great aversion on her part. She is probably slightly mentally retarded. She has been awarded an invalidity pension.

DISCUSSION

In his paper in 1901, Secretan did not suggest a self-induced injury as the cause of the disease (15).

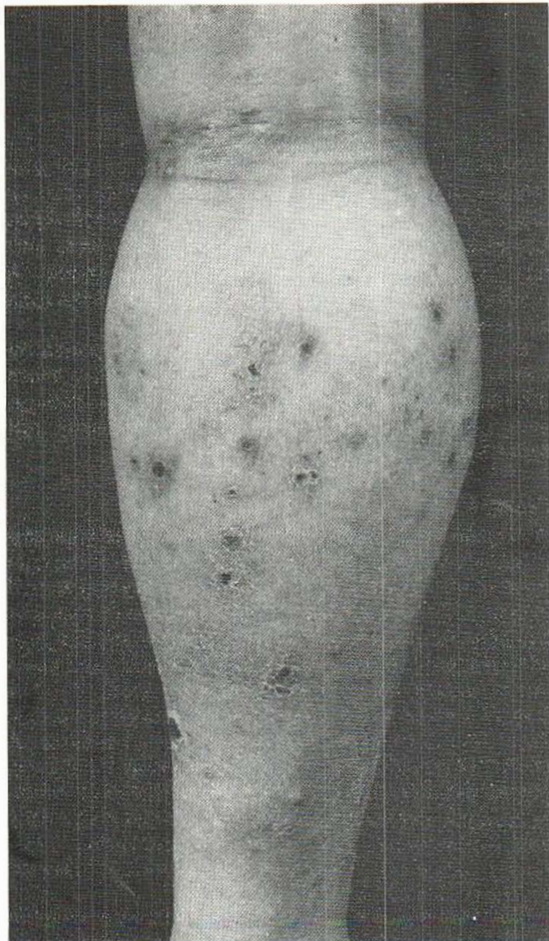


Fig. 1. Oedema and demarcation ring of the right lower leg, patient number 1.

In agreement with this, several case reports have been published (4, 6, 14). Prolonged disability and recurrent episodes have been typical for these patients. In other reports the oedema has been found to be of factitious origin (2, 3, 5) either because it resolved following the application of a protective plaster dressing (13) or because the patients were observed when they produced their lymphoedema (16).

Many different causes of factitious lymphoedema have been described. In his most impressive series of 22 patients, Smith (16) observed that the lymphoedema subsided initially following hospitalization. Later, recurrence was seen. He found the cause to be periodical application of tourniquet to the limb in 12 of the 22 patients. 7 patients repeated-

ly contused the dorsum of their hands. One girl admitted striking the hand against desks, chairs, tables and lockers during the day in order to produce the swelling. 3 patients repeatedly irritated the hand by scratching it or inserting small foreign bodies beneath the skin; 9 of these 22 patients were males. Recently a case of self-inflicted oedema of the dorsum of the left hand has been described in a young girl who later also developed atrophic scars on the dorsum of her left hand and excoriated acne on the cheeks (1).

In an experimental work on monkeys, Omer et al. (12) found that a similar disease could be produced only by such combinations of traumas: trauma plus local injection of autologous blood, lymphatic-venous obstruction plus trauma or lymphatic-venous obstruction plus trauma plus injection of autologous blood. Isolated trauma from surgical incision, blunt injury or injection of blood did not result in significant histologic changes. However, recurrent lymphatic and venous obstruction alone could produce significant histologic changes.

Only in the first patient reported in this paper, has the self-inflicted origin of the lesion been proven. However, in the other 2 patients, investigations for other conditions that can cause chronic lymphoedema have been negative. None of the patients complained of pains which may have been present in other cases (16). The most important differential diagnoses are lymphatic aplasia, lymphoedema praecox, mamma carcinomas and other tumours, surgery, irradiation, filariasis, infections, venous obstruction, and sympathetic dystrophy. A factitious etiology should especially be suspected particularly in patients with recurrent or chronic unilateral lymphoedema which is limited proximally by a well demarcated ring or sulcus of circumferential discoloration.

The condition is neither easy to diagnose nor easy to treat. Various methods of surgery have been recommended by several authors (8, 10, 11), while others have found no cure following surgery in larger series (13, 14, 16). Protective splinting and casting devices often result in immediate improvement, but recurrent episodes can be expected. Psychopathology should be suspected in all cases of self-inflicted oedema (13, 16). Psychotherapy has been suggested by some authors (7, 9). The achievement of insight should not be the chief objective of treatment. A triumphant confrontation is not

productive and may even worsen the situation. Prolonged disability is usual, but repeated supportive visits to a therapist may improve the condition.

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"Keratosi Palmoplantar Striata" (Brunauer-Fuhs Type)

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Abstract. Keratosis palmoplantar striata (Brunauer-Fuhs type), in three generations. The clinical, etiopathogenic and therapeutic aspects of this rare keratoderma are discussed.

Key words: Keratosis palmoplantar striata; Etiopathogenic aspects; Retinoid acid

In the group of palmoplantar keratoderma, according to Franceschetti & Schnyder's classification (7), the form which stands out, due to its exceptionality, is the one described by Brunauer (4), and Fuhs (8), which because of the linear appearance of the keratotic elements on the palms, was given the name "keratosis palmaris striata" by Siemens (10) 5 years later. This keratoderma is defined by the following characteristics: (a) dominant autosomal inheritance, (b) appearance in puberty or adulthood, (c) brought on by mechanical trauma, in general, of an occupational nature, (d) linear keratotic elevations on the palms, (e) elements of these same characteristics, but arranged in small islets, "areata form", on the soles.

Recently, we have had the opportunity, in the Department of Medical-Surgical Dermatology and Venereology at the University of Seville, to study a family affected by this process, and we will now go on to explain our case "princeps", and to discuss briefly the aspects which merit greater attention.

CASE REPORT

M. R. R., male, 35 years of age, a cooper, born in Dos Hermanas (Seville), first consulted us in May 1982, because since the age of 23, he had suffered from the appearance of keratotic elements on the palms. The areas in which the complaint was particularly more evident, were those subject to friction and pressure. The condition improved when the patient abstained from work. He also had a similar, though very painful condition on the soles of his feet. Both his hands and feet were submitted to a process of topical treatment and surgery, but the condition always reappeared. His father, brother and daughter were affected by the same process (Fig. 1).