Transformation of Lymphocytoma cutis into a Malignant Lymphoma in Association with the Sign of Leser-Trelat

S. HALEVY and M. SANDBANK
Department of Dermatology, Beilinson Medical Center, Petah Tikva, and the Sackler School of Medicine, Tel Aviv University, Israel


A patient is described in which transformation of lymphocytoma cutis (LC) into a malignant lymphoma, diffuse, mixed small and large lymphocytes, occurred in association with the sign of Leser-Trelat (LT), which is a marker for internal malignancy. To the best of our knowledge, such an association has not been reported previously. (Received May 26, 1986.)

S. Halevy, Department of Dermatology, Beilinson Medical Center, Petah Tikva, 49 100, Israel.

Lymphocytoma cutis (LC) is essentially benign in nature and only rarely becomes malignant (1). We present a case in which malignant transformation of LC into a malignant lymphoma, diffuse, mixed small and large lymphocytes, was associated with the sign of Leser-Trelat (LT), which is a marker for internal malignancy (1, 2).

CASE REPORT

A 64-year-old male presented with erythematous papular lesions on the left side of the neck (Fig. 1). The diagnosis of LC was established based on the following histologic findings. The epidermis was unaffected. Below the epidermis there was a thin grenz zone, and in the upper and middle dermis there were foci of mononuclear cell infiltration, mainly in perivascular areas. The cellular infiltrate was composed mainly of nature lymphocytes with a few large lymphocytes and histiocytes. A few germinal centers were seen. There was small vessel proliferation with prominent swollen endothelial cells. The skin adnexa were preserved and did not show any cellular infiltration (Fig. 5). In addition to these, there were several pigmented seborrheic keratoses (histologically confirmed) on the trunk, with a predilection for the back.

Seven years later the patient reported having noticed a rapid increase in the number of the seborrheic keratoses as well as in the size of those already existing. The seborrheic keratoses on the back were seen to have a "splash effect" (Fig. 4) and there was an abundance of sebaceous hemangioma as well as "mixed lesions" composed of both seborrheic keratosis and hemangioma. Within 19 months the neck lesions were also observed to have changed markedly, with an accompanying pruritus. Now present were firm, non-tender nodules several centimeters in diameter with smooth overlying skin of a red-violaceous colour (Fig. 2). Histological examination of a nodule revealed findings compatible with malignant lymphoma, diffuse, mixed small and large lymphocytes. The epidermis was normal and thin grenz zone was seen beneath it. All of the dermis and subcutaneous fat was invaded by a dense homogenous cellular infiltration composed of small and large-sized lymphocytes. No germinal centers were found. No skin adnexa were found in the biopsy specimen. Reticulum stains showed thin, small broken reticulum fibers interspersed in the cellular...
Fig. 1. Erythematous papular lesions on the left side of the neck.

Fig. 2. Tumoral masses on the left side of the neck consisting of firm red-violaceous nodules several centimeters in diameter.

Fig. 3. Second biopsy: Lymphoma of skin = dense diffuse infiltration of the dermis by small and large sized lymphocytes (H & E. x315).

Fig. 4. Multiple seborrheic keratoses on the back giving a “splash effect”.

Fig. 5. First biopsy: LC = patchy infiltration with formation of germinal centers (H & E. x60).
infiltrate. The immunoperoxidase technique (PAP method) for the detection of light chains (kappa and lambda) as well as IgG and IgM was applied in the two biopsies and gave negative results. A thorough investigation of the patient revealed only enlargement of the liver and inguinal lymph nodes. Following radiotherapy for the skin lesions on the neck there was a complete regression of the tumoral masses, but no decrease in the number or size of the seborrheic keratoses.

DISCUSSION

LC belongs to the category of pseudolymphoma, a term applied to a group of benign dermatoses having histologic features that often make the distinction from lymphoma very difficult if not impossible.

In the present case the diagnosis of LC established in the first biopsy has been supported by the following: the presence of germinal centers, a paucity of medium-sized lymphocytes in comparison to both small and large lymphocytes, non-involvement of the deeper dermis, predilection of the infiltrate for perivascular areas, and vascular proliferation with endothelial swelling (3, 4).

Burg & Braun Falco (5) found B lymphocyte markers on the lymphocytes in LC whereas Van Hale & Winkelmann (6), using leukocyte monoclonal antibody staining, found that lymphocytoma was represented by nodular masses of B lymphocytes with peripheral and intervening zones of T cells. In the present case the morphological pattern of the lymphocyte infiltration was compatible with B lymphocyte infiltration (4) but no immunoglobulins were detected by the immunoperoxidase technique.

Transformation of LC into malignant lymphoma which occurs only rarely (1) was supported in the present case by the association with the sign of LT. The sign of LT refers to the sudden appearance and/or rapid increase in the number and size of seborrheic keratoses as a marker of internal malignancy (2). Among the various types of malignancy reported in association with the sign of LT are malignancies of the reticuloendothelial system, including mycosis fungoides (7, 8), Sezary syndrome (9), a well-differentiated lymphoma (7) and a poorly-differentiated lymphocytic lymphoma (10).

To the best of our knowledge, association of the sign of LT with malignant transformation of LC into a malignant lymphoma, diffuse, mixed small and large lymphocytes, as in our case, has not been reported previously. In the present case, as well as in another case of poorly-differentiated lymphocytic lymphoma associated with the sign of LT (10), the sign of LC preceded the discovery of the malignancy, and the clinical findings included a "splash effect" of the seborrheic keratoses and the abundance of hemangiomata and "mixed lesions". The mechanisms responsible for proliferation of seborrheic keratoses in association with malignancy and their possible regression following adequate therapy (7, 8) have not yet been clarified.

REFERENCES


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**Skin Tags: A Cutaneous Marker for Diabetes Mellitus**

MICHAEL KAHANA, EHUD GROSSMAN, ABRAHAM FEINSTEIN, MEIR RONNEN, MICHAEL COHEN and MIRIAM SCHEWACH MILLET

Department of Dermatology, *Department of Internal Medicine, The Chaim Sheba Medical Center, Tel Hashomer and Sackler School of Medicine, Tel Aviv University,* and Kupat Holim, Netanya, Israel


Two hundred and sixteen non hospitalized patients with skin tags (ST) were studied for the presence of diabetes mellitus (DM) and obesity. Overt DM was found in 57 (26.3%) patients and impaired glucose tolerance test was found in 17 (7.9%) patients. Sixteen new cases of DM were found among this group. All the diabetic patients in the study population had non-insulin dependent DM. Sixty-two (28.7%) of the patients were obese. No correlation was found between the localization, size, colour and number of the ST and the presence of DM. Our study indicates that ST are not associated with increased incidence of obesity compared to the general population. On the other hand, ST are associated with impaired carbohydrate metabolism, and may serve as a means for identifying patients at increasing risk of having DM. (Received September 2, 1986.)

Michael Kahana, Department of Dermatology, The Chaim Sheba Medical Center, Tel Hashomer, 52621, Israel.

Skin tags (ST) are small, soft, pedunculated, often pigmented lesions, usually occurring on the eyelids, neck and axillae. The condition is very common, particularly in middle-aged and elderly women. Obesity is a factor that has been associated with the development of ST. Aside from their unsightly appearance, these lesions were thought to bear little clinical meaning (1). Recently, an association between ST and acromegaly (2, 3) and colonic polyps (3-5) has been reported.

In 1976, Margolis & Margolis (6) reported the association between multiple ST and diabetes mellitus (DM). They examined prospectively 500 consecutive hospital admissions for the presence of ST. Approximately 75% of their 47 male patients with ST had elevated fasting and postprandial blood glucose in the diabetic range. Marglis & Margolis concluded that multiple, large, hyperpigmented, bilateral ST were predictive of DM in men. The association between ST and DM was briefly mentioned later (7, 8). However, no further studies confirming this relationship or relating it to the presence of obesity have been published. We therefore undertook our study to test these findings in non hospitalized population of both men and women.