Pathology

Two-mm punch biopsies were removed from her trunk on the third postnatal day and prepared for light and electron microscopy by routine procedures.

Light microscopy

The dermis contained aggregated cell infiltrates consisting mainly of histiocytes and, among them, Touton giant cells and intermingling lymphocytes and eosinophilic granulocytes. No foam cells were seen. No structural changes were seen in the epidermis.

Electron microscopy

Infiltrating histiocytes contained markedly invaginated nuclei, a granular endoplasmic reticulum and lysosomes (Fig. 2). Touton-type giant cells showed identical cell organelles and nuclei, but a greater abundance of lysosomes and endoplasmic reticula. The nuclei were extremely invaginated, presenting bizarre shapes (Fig. 3).

No fat droplets were present in the cytoplasm, but cholesterol clefts were seen in the cytoplasm (Fig. 3). No Langerhans cell granules could be detected.

DISCUSSION

The finding of histiocytes and Touton-type giant cells indicates that the patient was suffering from juvenile xanthogranuloma, although no fat vacuoles could be demonstrated in the skin biopsy. The disease has been separated from histiocytosis X by the lack of Langerhans cell granules in the infitrating cells (7). No such granules were found in the present case. In previous electron microscopical studies, fat droplets and lysosomes were reported to be a common finding in the infiltrating histiocytes and Touton cells. Esterly et al. (2) were unable to detect an enclosing bounding membrane around the fat droplets, whereas Kjaerheim et al. (3) and Wolff et al. (7) showed such a structure to be present. None desribed cholesterol clefts in the ground cytoplasm of the infiltrating cells, such as is reported here. The problem concerning the origin of fat droplets being either metabolic products or lysosomal material, i.e. either non-enclosed or enclosed by a bounding membrane, is still a matter of debate. Cholesterol clefts might indicate

Fig. 2. Electron micrograph of histiocytes (H) with conconvoluted nuclei, lysosomes (L) and a granular endoplasmic reticulum (thin arrows). ×3900.

Fig. 3. Electron micrograph of Touton-type giant cell shows bizarre nuclei, numerous lysosomes (L) and granular endoplasmic reticulum (thin arrows). Cholesterol clefts are indicated by thick arrows. ×9900.

that the fat metabolization in the histiocyte has changed in some way. Numerous primary lysosomes may possibly develop into vacuoles of the cytoplasm.

Serial sectioning has demonstrated that multiple nuclei of Touton-type giant cells were the result of cutting the strongly invaginated nuclei (3). Fusion of cells due to the complexity of interdigitating protrusions of the cytoplasm has been suggested (4). In the present study no remarkable cytoplasmic protrusions could be found. Clinically, xanthogranuloma juvenile may bear some resemblance to urticaria pigmentosa (5) or histiocytosis X (8). Since the disease regresses spontaneously, and malignant transformation is unlikely, no treatment is advisable.

REFERENCES

- Crocker, A. C.: Skin xanthomas in childhood. Pediatrics 8: 573-597, 1951.
- Esterly, N. B., Sahiti, T. & Medenica, M.: Juvenile xanthogranuloma. Arch Dermatol 105: 99–102, 1972.
- Kjaerheim, A. & Stokke, T.: Juvenile xanthogranuloma of the oral cavity. Oral Surg 38: 414–425, 1974.
- 4. Seifert, H. W.: Membrane activity in juvenile xanthogranuloma. J. Cut Pathol 8: 25–33, 1981.
- 5. De Villez, R. L. & Limmer, B. L.: Juvenile xanthogranuloma and urticaria pigmentosa. Arch Dermatol 111: 365-366, 1975.
- 6. Webster, S. B., Reister, H. C. & Harman, L. E.: Juvenile xanthogranuloma with extracutaneous lesions. Arch Dermatol 93: 71–76, 1966.
- Wolff, H. H. & Braun-Falco, O.: Zur Diagnostik und Therapie des Morbus Hand-Schüller-Christian. Hautarzt 23: 163-169, 1972.
- 8. Wolff, H. H., Vigl, E. & Braun-Falco, O.: Juveniles Xanthogranulom und Organmanifestationen. Hautarzt 26: 268-272, 1975.

Leprosy in Vietnamese Refugees: A Case Report

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Abstract. A 41-year-old Vietnamese refugee was found to have tuberculoid leprosy. Dapsone treatment cleared her skin lesion, but did not remove the paraesthesia in



Fig. 1. The pictures show the skin changes of the patient at the first clinical investigation.

the right ulnar nerve. Immunological investigations revealed a strongly positive lepromin test and a significant change in the ratio of T-y and T- μ lymphocytes in peripheral blood. The incidence of leprosy in Vietnamese refugees in Scandinavia is discussed.

Key words: Dapsone; Lepromin test: Leprosy; T lymphocytes; Vietnamese refugees

Endemic leprosy disappeared from Scandinavia in the middle of this century. Occasionally, it is found in immigrants (6). We report here a case of leprosy in a Vietnamese refugee.

Table 1.

Date	Ratio of T-γ/T-μ		Lep- romin stimu-	Tuber- culin stimu-
	Patient	Control	lation index	lation index
04.02.80	38.0	0.8	1.5	8.2
15.02.80* 29.02.80* 28.05.80 15.09.80	1.9 7.7 5.1 3.9	0.5 0.6 0.5 0.6	7.0 1.4 1.7 3.2	9.5 0.9 n.d. 15.1

Stimulation index = The ratio between the DNA synthesis in cultures stimulated with antigen and without antigen. An index ≥ 2.0 indicates the presence of lymphocytes with specificity for the added antigen.

* The lepromin test was strongly positive at these dates. Treatment started 7.2.82.

CASE REPORT

A 41-year-old Vietnamese woman came with an eruption of red papules on the ulnar side of her right lower arm (Fig. 1). She had had a few papules for years, whereas the present eruption occurred approximately 3 months before her first visit in the clinic and 6 or 7 months after her arrival in Denmark. She complained of paraesthesia and reduced sensitivity of the fourth and fifth right fingers. Her right ulnar nerve was enlarged and tender under the medial epicondyle. Histological examination of a papule revealed pronounced perineural epithelioid granulomatous formation going deeply into the corium. No acid-fast bacilli were found. A lepromin test proved positive, with both the early (Fernandez) and the late (Mitsuda) reaction. A tuberculin skin test proved negative (1 unit of tuberculin).

Treatment with dapsone was given for 18 months, giving clearance of the skin after 9 months. Her paraesthesia persists. The initial dapsone dosage was 25 mg daily, later increased to 50 mg daily, without side effects or complications.

Immunological investigations revealed normal quantities of lymphocytes and T lymphocytes in the blood (E-AET, E-4 and E-active techniques were used). However, the ratio between T cells with Fc receptors for IgG and IgM (T- γ and T- μ respectively) was reversed before vs. during therapy (Table I). Following the lepromin skin test we observed a transient positive lepromin reactivity of her lymphocytes in vitro and later a reduced tuberculin reactivity in vitro (Table I). The mitogen reactivity of lymphocytes was normal (PHA, Con A, PWM).

DISCUSSION

The major medical problems in Vietnamese refugees are caries, parasitic infections, skin infections and hepatitis B carrier state (2, 5). A survey of 880 refugees in Sweden (2) revealed two cases

of tuberculoid leprosy. Recently, another case has been reported (4). From 1975 to 1982, 3 397 Vietnamese refugees have arrived in Denmark. Up to the present, leprosy has been discovered only in our patient and in a 61-year old man with tuberculoid leprosy (Prof. Finn T. Black, personal communication).

The immune response of the patient was normal and consistent with the clinical diagnosis of tuberculoid leprosy. Using monoclonal antibodies, Bach et al. (1) found no significant change in T lymphocyte subpopulations in peripheral blood in leprosy. In our patient it is likely that the significant change in the ratio between T-γ and T-μ lymphocytes and her lymphocyte reactivity *in vitro* is a reflection of her resistance towards the infection.

The incidence of leprosy in Vietnam is considered to be approximately 1% (5). Apparently, the incidence in the refugees is much lower. However, Scandinavian doctors should be aware of its existence.

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REFERENCES

- Bach, M.-A., Chatenoud, L., Wallach, D., Tuy, F. P. D. & Cottenot, F.: Studies on T cell subsets and functions in leprosy. Clin Exp Immunol 44: 491, 1981.
- Castor, B., Frolov, I. & Hansen, C.: Infection panorama among Vietnamese boat people. Läkartidningen 78: 1659, 1981. (In Swedish.)
- Jensen, J. R., Cramers, M. & Thestrup-Pedersen, K.: Subpopulations of T lymphocytes and non-specific suppressor cell activity in patients with atopic dermatitis. Clin Exp Immunol 45: 118, 1981.
- Olcén, P. & Fredlund, H.: Lepra hos bătflykting. Report no. Vecka 41, 1981, from the National Bacteriological Laboratory, Department of Epidemiology, S-10521 Stockholm, Sweden.
- Prag, J., Nørredam, K. & Hardvendel, U.: The health of 150 Mon-Khmers during the first 30 months in Denmark. Ugeskr Log 141: 3022, 1979. (In Danish.)
- Schmidt, H.: A case of leprosy diagnosed in Denmark. Ugeskr Læg 134: 1169, 1972. (In Danish.)

White Sponge Nevus: Successful Treatment with Penicillin

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Abstract. White sponge nevus is a rare, benign, inherited disorder of the mucous membranes, affecting mainly the oral mucosa. A case is reported of white sponge nevus, which showed a definite improvement following penicillin administration.

Key words: White sponge nevus; Leukoderma exfoliativum mucosae oris; Penicillin

Since Cannon first reported 3 cases of white sponge nevus (WSN) of the oral mucosa in 1935 (1), fewer than 80 cases of this autosomal dominant disorder have been recorded in the literature.

The treatment of WSN is considered unrewarding, with the exception of penicillin, whose administration was noted to be associated with remission in 2 patients (3, 2). The purpose of this report is to describe a case of WSN in which the oral lesions almost completely disappeared during treatment with penicillin.

REPORT OF A CASE

A healthy 23-year-old medical student was seen in the Skin Clinic of the University of Parma in September 1981 because of diffuse, white, folded, asymptomatic lesions confined to the oral mucosa. The onset was noted in infancy on the right side of the tongue, followed by bilateral involvement of the tongue and cheeks within 2 years. Afterwards the condition remained stationary despite several therapeutic trials.

In 1978 he was treated with vitamin A orally (Arovit, Roche, Italy) for one year and then with nystatin (Mycostatin, Squibb, Italy) for 2 months, unsuccessfully. In January 1980 the histologic diagnosis of leukoplakia was made and treatment with liquid nitrogen was given, though without any effect.

The family history revealed that the patient's mother and grandmother had similar white spongy lesions of the mouth, whereas one brother and two aunts were not affected. Oral examination showed soft, creamy-white, folded, irregular plaques with spongy appearance, located on both sides of the tongue and cheeks (Fig. 1). No lesions were found on the palate, peritonsillar areas or lips. Moreover, conjunctivas, genitalia and anal mucosa were not affected.

The following investigations were carried out and were