SHORT COMMUNICATION

CUTANEOUS LEISHMANIASIS WITH UNUSUAL PRESENTATION

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Three cases of cutaneous leishmaniasis with unusual presentation seen at Central Hospital Tripoli, Libya are described. Case No.1 and 2 had 102 and 109 noduloulcerative lesions with generalized almost bilaterally symmetrical distribution, suggestive of dissemination. Case No.3 had large verrucous carcinoma but histopathology suggested lupoid (chronic cutaneous) leishmaniasis. Patient No. 2 developed hypersensitivity reaction to sodium stibogluconate after 10 days but responded well to the treatment. Case No.2 did not tolerate the above treatment and was treated successfully with combination of rifampicin and INH. Case No.3 was treated successfully with liquid nitrogen cryotherapy given by Cryosurg-Frigitonics, using special open probes.

Key Words: Cutaneous leishmaniasis, Lupoid (chronic cutaneous) leishmaniasis

Introduction

Old world cutaneous leishmaniasis is endemic in certain areas of Libya.[1] During 1978, Bhakta Viziam could see 23 cases at Central Hospital Tripoli.[2] Later we have seen 60 cases during 1981-83.[2] Since then number of cases at Tripoli is increasing constantly and some new endemic areas are discovered in Libya (although still there is no evidence of endemicity at Tripoli). Among a huge number of cases seen between 1984 and 1994, 3 patients showing unusual features are described.

Case 1

A 35-year-old Libyan woman came with multiple nodulo-ulcerative lesions of 2 months duration. Initially she developed a few papular and nodular lesions on face, exposed parts of forearms and legs. Within 2-3 weeeks she developed similar multiple lesions on trunk and unexposed parts of the extremities with almost bilaterally symmetrical distribution. Most of the lesions ulcerated within 2-3 weeks and the size varied between 1-3 cm (Fig.1 and 2). Total number of lesions was 102.

Fig. 1. Patient No. 1- Noduloulcerative lesions on face and forearms

Slit smear done from forearm, leg and trunk lesions revealed leishmania tropica bodies (LT bodies) in all. Blood smear stained with Giemsa stain was negative for LT bodies. Routine investigations on blood, urine, X-ray chest, ECG and ultrasound abdomen did not reveal any abnormality.

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Treatment was started with injection sodium stibogluconate 600 mg intramuscular daily. After 10th injection she developed itchy erythematous urticarial rash on gluteal region around the injection site. So the treatment was discontinued. By this time the lesions regressed well

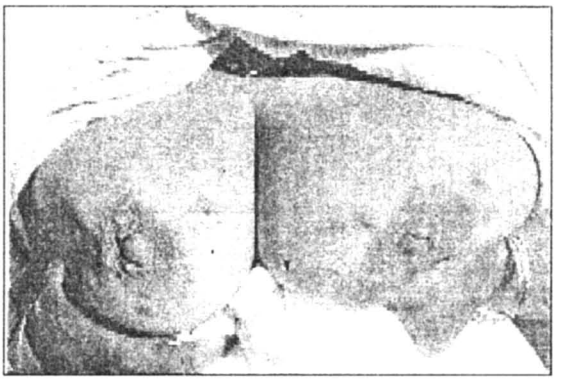

Fig.2. Patient No. 1 - Noduloulcerative lesions on breast

and healed completely with in a month.

Case 2

A 65-year-old Libyan man presented with 109 nodulo-ulcerative lesions of 2 months duration. This patient also noticed initial few lesions on exposed parts (face, hands and feet) and later developed multiple lesions on trunk and extremities with almost bilaterally symmetrical distribution. Most of the lesions were ulcerated and size varied from 1 to 4 cm.

Slit smear done from hand, feet and trunk lesions revealed multiple intra and extra-cellular LT bodies in all. Blood smear did not show LT bodies. Routine investigations including ECG, X-ray chest and ultrasound abdomen did not reveal any abnormality. The patient was a known hypertensive and was on betablockers for last 5 years.

We tried test dose (1 ml) of sodium stibogluconate but he felt precordial discomfort. So we started treatment with rifampicin 600 mg and INH 300 mg daily and continued for 2 months, by that time all lesions healed almost completely and no adverse effect of the treatment was noticed.

Case 3

A 70-year-old Libyan man has been gradually developing nodulo-ulcerative lesions for 2 years. At the time of assessment he had 8 large (3-12 cm), thick (1-4 cm) lesions with hypertrophic granulation tissue and verrucous appearance on right foot, shin and left calf (Fig.3). He also had multiple grouped nodular lesions (1-3cm) with diffuse infiltration on left elbow and forearm and dorsum of left hand (Fig.4).

Slit smear done from forearm lesion was positive for LT bodies but negative in leg lesion. Histopathology of left shin lesion revealed hyperkeratosis, irregular marked acanthosis, with diffuse and granulomatous infiltration through out the dermis consisting of lymphocytes, plasma cells, epithelioid cells and giant cells. Giemsa staining of histopathologic section revealed few small LT bodies in one corner of the section.

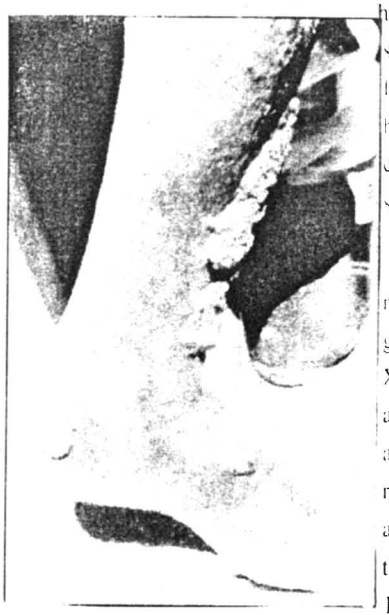

Fig.3. Patient No. 3 - Thick verrucous lesions

Although routine investigations including X-ray chest, ECG and ultrasound abdomen did not reveal any abnormality, since there was past history of MI, we did not try sodium stibogluconate. This patient was treated with liquid nitrogen cryotherapy given by Cryosurg - Frigitonics using special open probes. Two to four fortnightly treatments were given with freeze time 45-60 seconds according to thickness of various lesions. All lesions regressed considerably in two months and healed completely with mild atrophy within 3 months.

Discussion

In our previous study[1] most of the Libyan patients had single lesions, although one of them had 20 lesions on exposed parts of the body. The present cases 1 and 2 had 102 and 109 lesions respectively with almost bilaterally symmetrical distribution and involvement of unexposed sites of the body. These facts reveal that the classical categories of cutaneous leishmaniasis are undergoing considerable change in light of clinical evidence suggesting that the organisms may not be as localized as previously thought but may be haematogenously spread through out the body. Although we could not detect LT bodies in the blood smears of these patients, in light of clinical evidence we cannot exclude the possibility of dissemination in case 1 and 2. Kubba et al,[3] have noticed dissemination in 20% of their Saudi Arabian cases but we observed definite phenomenon of dissemination only in these two cases among a huge number of cases seen between 1981 and 1994. Visceral infection due to leishmania tropica has also been reported[4-6] but we did not observe this phenomenon in our patients.

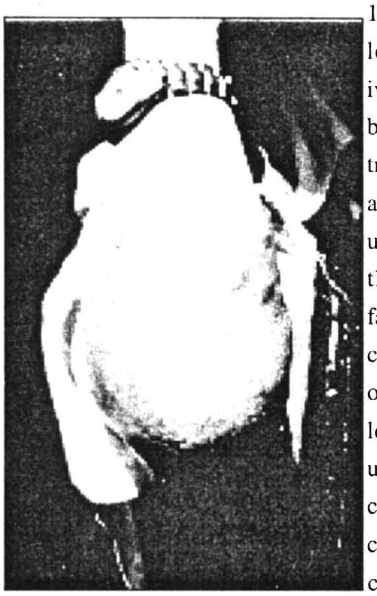

Fig.4. Patient No. 3-Grouped nodular lesions with infiltration

Clinical features of patient no. 3 suggest the diagnosis of rare variant- the lupoid leishmaniasis which has been previously described.[7-9] The leg lesions of this patient had clinical similarity with that of verrucous carcinoma but the histopathology suggested chronic cutaneous leishmaniasis.

Patient no.1 developed localized delayed hypersensitivity reaction to sodium stibogluconate. We have observed similar reaction previously in another patient.

Patient no.2 was managed successfully with alternative treatment, a combination of rifampicin and INH. Similar results have been recorded previously.[2,10] This combination is superior to that of monotherapy with rifampicin.[2]

Patient no.3 was treated successfully with cryotherapy using liquid nitrogen through special probes. We have also treated successfully 12 typical cases with 1 to 3 lesions by only single similar cryotherapeutic procedure and depigmentation of the treatment site was observed in one patient (unpublished data). Previously we treated 4 patients by applying liquid nitrogen with cotton swab with slow improvement but al-Majali et al[12] reported good results with this form of therapy. Gindan et al[13] have also utilized cryotherapy as first choice therapy for cutaneous leishmaniasis.

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