CASE REPORT

Indian erythrodermic postkala-azar dermal leishmaniasis

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SUMMARY

Postkala-azar dermal leishmaniasis (PKDL) is a complication of kala-azar or visceral leishmaniasis and is caused by *Leishmania donovani*. We describe an Indian male patient with the rarer erythrodermic form of PKDL and multiple unusual skin lesions viz. verrucous, annular and mucosal ulceration.

BACKGROUND

Postkala-azar dermal leishmaniasis (PKDL) is a complication of kala-azar (KA) or visceral leishmaniasis (VL) caused by *Leishmania donovani*, predominantly seen in the Indian subcontinent and Sudan. Erythrodermic PKDL is a rarer presentation of PKDL in India.

CASE PRESENTATION

A man aged 25 years had widespread erythrodermic infiltration along with erythematous, firm papulo-plaques since 10 years duration (figures 1 and 2). Some plaques were annular on the trunk and ulcerated and verrucous on the extremities (figure 1A, B). He also had palatal and genital mucosal lesions (figure 1C). He had a past history of KA.

INVESTIGATIONS

Skin biopsy showed numerous amastigote forms of *L. donovani*. Strip test based on the recombinant

Figure 2 (A) Diffuse erythrodermic infiltration of back along with numerous discrete papulonodular lesions. (B) Similar infiltration over the lower extremities with discrete papulonodular lesion over the scrotal area.

Figure 3 Results of PCR assay; lane M contains a 100 bp DNA ladder; lane 1 contains DNA from *Leishmania donovani* parasite (positive control); lane 2 contains DNA from dermal lesion biopsy; lane 3 contains DNA from healthy dermal biopsy (negative control).

Figure 1 (A) Widespread erythrodermic infiltration of the chest along with numerous annular plaques. (B) Close-up view showing diffuse background infiltration of the chest. (C) Ulceration of the palatal mucosa. (D) Massive infiltration of the ear lobe.

K39 antigen was positive. HIV serology was negative. The causative parasite in the skin biopsy was characterised as *L. donovani* using a species-specific kDNA-based PCR (figure 3). A diagnosis of erythrodermic PKDL was made.

DIFFERENTIAL DIAGNOSIS

Leprosy is the main differential diagnosis of classical PKDL because of the clinical and histopathological similarities. The small hypopigmented lesions, facial infiltration and madarosis in PKDL may simulate the clinical presentation of borderline and lepromatous leprosy. However, the centrofacial involvement, relative sparing of the ear lobes and the preferential involvement of midline back in PKDL favours this diagnosis. Histologically, PKDL can display leprosy pattern of infiltrate, but the

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Figure 4 Significant improvement post-treatment with flattening of annular plaques and reduction in generalised erythema.

presence of *L. donovani* bodies helps in establishing the diagnosis. The other differential diagnoses include pityriasis versicolor, vitiligo and secondary syphilis. The erythrodermic infiltration as seen in this case need to be differentiated from other causes of erythroderma, in particular cutaneous T-cell lymphomas.

TREATMENT

He was treated with miltefosine 50 mg three times a day for 3 months, but the response was partial. He was then administered intravenous amphotericin B 50 mg daily until a total of 1 g had been given.

OUTCOME AND FOLLOW-UP

The lesions completely resolved and there is no sign of recurrence 4 years after stopping amphotericin B (figure 4).

DISCUSSION

PKDL is an uncommon dermal complication seen as a sequel to KA or VL. PKDL is widely prevalent in Eastern India and Sudan. It is also reported from the neighbouring countries—Bangladesh, Nepal and Pakistan. Three main types of clinical lesions occur in Indian PKDL: erythematous induration in the centro-facial region and the butterfly area of the face with photosensitivity, symmetrical hypopigmented macules on the trunk and extremities, and papulonodular or plaque lesions clustered around the central part of the face with gradual involvement of other body sites.^{1–3} The unusual manifestations are xanthomatous, fibroid or warty lesions, annular plaques and only mucosal lesions. Concomitant VL and PKDL has also been reported from India.⁴

Our case had the extremely rare form of extensive erythrodermic involvement along with several unusual lesions viz. annular, verrucous, ulceration, crusting and palatal ulceration. There was diffuse infiltration with erythema involving the face, trunk and extremities, though this erythema was most marked around the individual papule and plaque. Such widespread involvement with these lesions has been described in Sudanese PKDL (grade 3 involvement).² In India, erythrodermic presentation of PKDL is rarely reported.^{5 6}

Recently, the advisory panel of WHO for *Leishmania* control recommended the use of antimonials or liposomal amphotericin B in East Africa, and amphotericin B deoxycholate or miltefosine in Bangladesh, India and Nepal, for the treatment of PKDL.⁷ In the Indian subcontinent, sodium antimony gluconate (SAG) is a less favoured drug due to its long duration of treatment and the adverse events, including local pain. Successful treatment of PKDL with miltefosine (50 mg three times daily for 60 days) has been reported from India.⁸ Another alternative to SAG for treatment of PKDL is amphotericin B. Experience with amphotericin B for treatment of PKDL in Sudan is promising, though literature on efficacy from the Indian subcontinent is lacking.⁹ The treatment response was slow in our case; hence, a combination therapy of miltefosine and amphotericin B was used.

Learning points

- ▶ Erythroderma is a rare presentation of postkala-azar dermal leishmaniasis (PKDL).
- ▶ It is important to recognise this variant of PKDL, as these patients are loaded with organisms and highly infective to the community.
- ▶ The treatment response may be slow as seen in our case. Hence, these kinds of cases should be closely monitored for resistance and if needed, they should be treated with combination therapy.

Contributors GS, NB, PS and VR drafted the manuscript. All four authors finally approved the manuscript. PS did PCR for the diagnosis of species and VR provided miltefosine for the treatment.

Competing interests None declared.

Patient consent Obtained.

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