Disseminated leishmaniasis in a four-year-old child in Yaoundé, Cameroon

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Leishmaniasis is a disease caused by a protozoan parasite of the genus leishmania with worldwide distribution and is transmitted to man by phlebotomine sand flies. The clinical presentation could range from a single cutaneous ulcer to disseminated leishmaniasis. We report the case of a four-year-old boy admitted to our hospital with ulcers, wasting, progressively distending abdomen, and fatigue evolving for about two months. On admission, he was febrile and pale, with diffuse oozing wet ulcers on the limbs and face, hepatosplenomegaly, and enlarged inguinal lymph nodes. The complete blood count revealed pancytopenia with low reticulocyte count, and serum protein electrophoresis showed hypoalbuminemia and hypergammaglobulinemia. Skin biopsy revealed amastigotes in phagocytic cells. The above findings suggested cutaneous and visceral localization of the leishmania; however, the parents absconded with the boy just when treatment was instituted, believing that the child was bewitched. The outcome is expected to be fatal visceral involvement.

Key words: cutaneous ulcers, lymph nodes, splenomegaly, prolonged fever, leishmania.

Leishmaniasis is a disease caused by different species of a protozoan parasite belonging to the genus leishmania. The parasites are transmitted between mammalian hosts by phlebotomine sand flies. It was first recorded and described in 1901 by Leishman, who initially considered it as trypanosomiasis, but in 1903, Captain Donovan described the parasites as being new and different from trypanosomes. The link between these organisms and kala azar was eventually discovered by Major Ross, who used the name *Leishmania donovani*[1-3]. The disease affects all age groups and both sexes alike. About 12 million people are infected worldwide[2,4]. There are different clinical presentations of the disease, ranging from a single cutaneous ulcer to the disseminated form. The clinical entities usually occur separately and depend on the infecting species of the protozoan[1,5,6]. The visceral form is 100% fatal if not treated, while the cutaneous form can heal spontaneously [(1)]. We report the first case in Cameroon with both visceral and cutaneous lesions occurring in the same patient simultaneously.

Case Report

A four-year-old boy was referred from a rural hospital to our hospital (The Mother and Child Centre of the Chantal Biya Foundation, Yaoundé) for recurrent fever and diffuse skin eruptions (Fig. 1). He was reported to have been bitten by insects locally called "moot-moot" prior to the onset of fever. The insect bites left lesions that gradually developed into large ulcerated swellings on both legs and his face. He was unsuccessfully treated

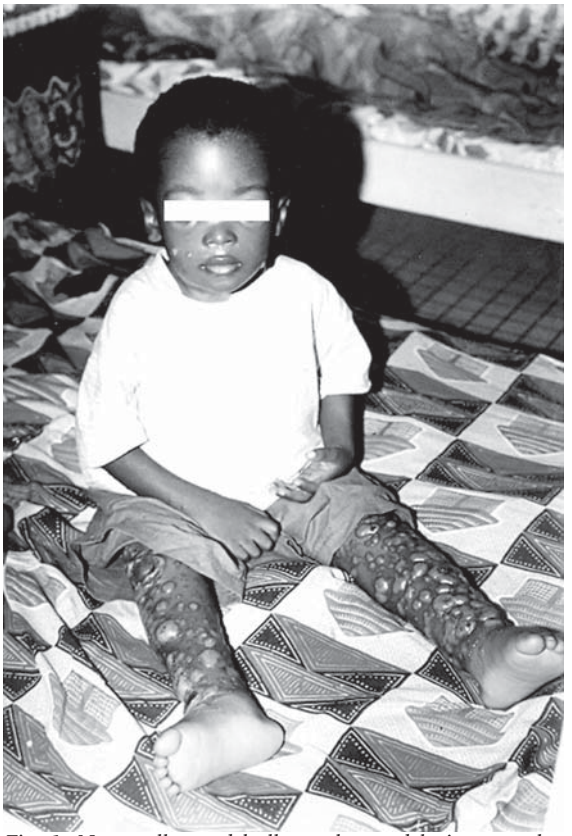

Fig. 1. Note pallor and bullous ulcerated lesions on the face and legs of the patient.

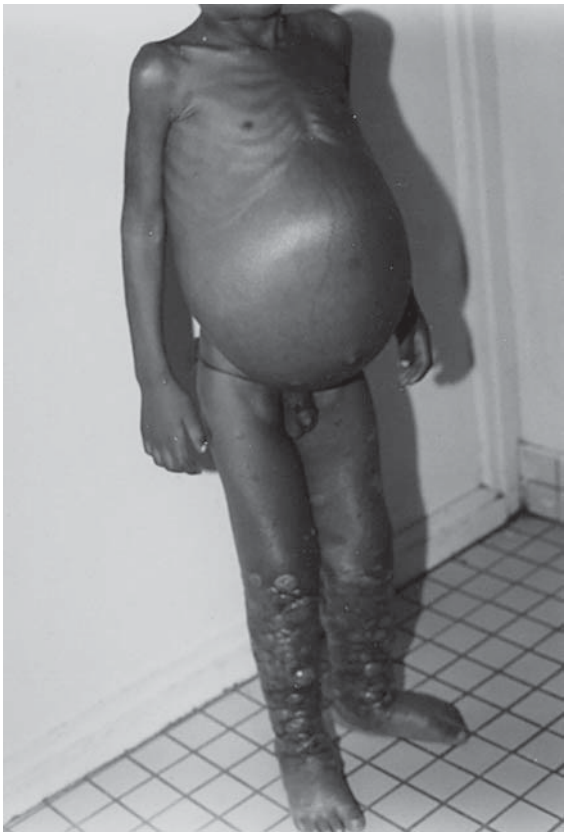

Fig. 2. Skin lesions on his exposed areas, distended abdomen with visible veins, and enlarged inguinal lymph nodes are seen.

with amodiaquine and cotrimoxazole and was then transferred to Yaoundé for appropriate management.

His past medical history was uneventful. On arrival in our hospital, he was febrile (body temperature: 38.7°C) and weighed 13 kg. He looked pale and was polypneic with mild nasal flaring and swollen eyes. There were ulcers on the face and prominent ribs, but the heart and lung examination was normal.

The abdomen was distended and tender with collateral circulation. The liver was enlarged, extending to about 15 cm below the right costal margin, and the spleen was also greatly enlarged, extending below the umbilical line (Fig. 2). The umbilicus was averted but there was no shifting dullness.

The inguinal lymph nodes were visibly enlarged. The lower limbs were covered with bullous eruptions of varying shapes and seizes, some taking the shape of a cauliflower, others volcano–like in appearance, with a "crater" on the top. Pus oozed from some of the ulcers while others were bleeding. There was pitting edema on both feet but the arms were thin and clear of any lesions.

The complete blood count showed: hemoglobin 4.8 g/dl, mean corpuscular volume 62 fl, and mean corpuscular concentration in hemoglobin 21.9 pg; white blood cell count 4000/mm^3; platelet count 83000/mm^3; and low reticulocyte count of 52060 = 1.9%.

No malignant cells were found on the blood smear. Blood cultures, blood parasites and the human immunodeficiency virus (HIV) serology were all negative; however, special stains for other blood parasites like leishmania and microfilaria were not done. Electrophoresis of plasma proteins showed hypoalbuminemia associated with hypergammaglobulinemia (total protein level of 65 g/L, albumin 15.7 g/L and gammaglobulin 36.1 g/L).

The biopsy from the skin lesion was stained with Giemsa and revealed histiocytes with large clear cytoplasm containing multiple

intracytoplasmic structures called Donovan's bodies with inflammatory infiltration (lymphocytes/plasmocytes) and a mixed bacteria flora. A hematoxylin & eosin (H&E) stain also revealed amastigotes in the cytoplasm of phagocytes. No malignant cells were found (Fig. 3).

This confirmed the diagnosis of leishmaniasis, and the child was placed on glucantime (20 mg/kg/day, gradually increasing the dose to a maximum of 60 mg/kg/day) and transfused 2 pints of whole blood. However, by the sixth day of this treatment, the child was taken out of the hospital by his parents with the belief that the disease was due to witchcraft and that modern conventional medicine could neither treat nor heal him. No further news of the child was obtained. The expected outcome is fatal without adequate treatment since there were signs of visceral involvement.

Discussion

Cases of cutaneous leishmaniasis have been reported in Chad, Central African Republic and northern Cameroon. Sporadic cases of visceral leishmania in children have been reported in southern Cameroon[4,7-11]. No case with the disseminated form of the disease has been reported so far. There is neither age nor sex predilection and anybody bitten by an infected sand fly can be affected[5,8]. The above case was reported to have been bitten by insects some months prior to the illness onset.

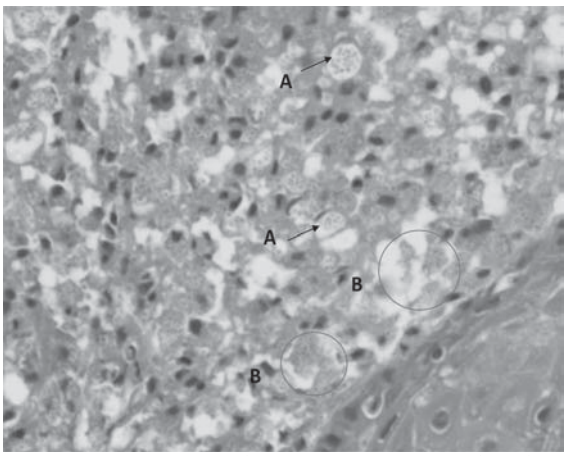

Fig. 3. H&E x40: Leishmania amastigotes within the cytoplasm of macrophages (A); others are diffused in the tissue after bursting of macrophages (B).

It is believed that the different clinical forms are caused by different species of the parasite [1,12], but the above case had both clinical presentations occurring simultaneously, although we could not identify the species because we were unable to do typing either by polymerase chain reaction (PCR)-ELISA or isoenzyme electrophoresis. The classical presentations are: cutaneous forms with a few skin ulcers on exposed areas of the body or diffuse cutaneous leishmaniasis presenting with disseminated, chronic skin lesions resembling lepromatous leprosy[2,4,5]. Our patient presented with both cutaneous lesions and signs of visceral involvement (fever, hepatosplenomegaly, enlarged peripheral lymph nodes, pallor, and wasting).

Skin lesions usually begin a few weeks or months after a sand fly bite. However, it takes months to years for the visceral form to manifest after the insect bite[13]. The skin lesions start as one or more sores on the exposed parts of the body, which can then change in size and appearance over time as could be seen on the legs of the case presented. The nodes draining the affected parts of the body are swollen. It is thought that some diffuse cutaneous forms can progressively extend to involve the viscera, resulting in fever, weight loss, and enlarged spleen and liver, as well as bone marrow invasion responsible for the bleeding and pallor observed in these patients. The liver involvement and malnutrition are believed to be responsible for the low protein level and the inflammation process revealed by the gammaglobulin peak.

Leishmaniasis is increasingly reported as coinfection in patients with HIV/acquired immunodeficiency syndrome (AIDS)[10], but this patient was free of HIV infection.

The diagnosis in the patient was suspected from the clinical presentation, pancytopenia with low reticulocyte count, hypoalbuminemia, and hypergammaglobulinemia, and was confirmed by histology of the skin lesions. The diagnosis is guided by the clinical presentation and confirmed by isolation of the parasite from the lesion[1,14].

Indirect findings that can support the diagnosis are complete blood count showing pancytopenia and a low reticulocyte count and serum protein electrophoresis revealing a peak of

gammaglobulin and hypoalbuminemia[14]. All these abnormal findings were noted in this case. However, the gold standard for diagnosis of visceral leishmaniasis is demonstration of the parasite in the bone marrow aspirate[14]. This was not done in our patient due to financial constraints.

The classical treatment is based on pentavalent derivatives of antimonates and diamidines and more recently with amphotericin B, but the side effects can be very severe[2,3,5,13]. Metronidazole has been used successfully for the cutaneous form of leishmania[4]. We used the antimonite derivative glucantime, but unfortunately the evolution of the disease with this treatment could not be evaluated since the patient was taken from the hospital before completion of the treatment. The parents considered the disease to be due to witchcraft and went for traditional treatment. Bouratbine et al.[15] reported similar difficulties in Tunisia, where traditional beliefs retard treatment, and in some cases, there is total refusal of conventional treatment, with disastrous consequences to the child.

The visceral form of the disease is lethal if not treated; however, the cutaneous form can heal spontaneously after several months to years, leaving scars[2,5,15].

In conclusion, although this parasitic infection is not very common in Cameroon, prolonged fever, skin lesions, and enlarged lymph nodes, liver and spleen should lead to suspicion of disseminated leishmaniasis in the differential diagnosis; consequent management measures should be taken because it is fatal if left untreated.

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