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Case Report

Evolution of Clinical Manifestations of Neck and Face due to Cutaneous Leishmaniasis Resulting In Diagnostic Errors

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Abstract

Background: Cutaneous leishmaniasis is the most common form of leishmaniasis caused by flagellate protozoa of the genus Leishmania transmitted by sand fly bites. Old World leishmaniasis is endemic in the Mediterranean Sea and the neighbouring countries. We believe, that this case is interesting by the fact that we had a very rear disease case that can be observed in nonendemic area. We present a case of a 22-year-old man with a cutaneous leishmaniasis in a localised form of ulcers on the right cheek and the right part of the neck. Histopathological examination showed diffuse dermal infiltrate predominantly of macrophages with admixture of few lymphocytes, eosinophils and plasma cells. In a very small number of macrophages amastigotes were seen. On their surface and occasionally extracellularly rod-shaped kinetoplasts were noticeable. It should be stressed that both clinical and laboratory data were not peculiar for this disease. Adults in endemic areas have stable immunity for protozoal infections. This made diagnostication and timely management of the disease very difficult. But clinical effect of drug therapy which is specific for cutaneous leishmaniasis treatment proved, in spite of the absence of ulcer soft tissues, blood and cerebrospinal puncture *Leishmania*, that our diagnosis was correct. The case, described by us, may be interesting for dermatologists, parasitologists, surgeons and other medical specialists. Because of higher rate of travel and work abroad increased number of sporadic cases of cutaneous leishmaniasis in non-endemic areas should be taken into account. Cutaneous leishmaniasis is a rare disease in Kazakhstan, especially in the north region. Because of higher rate of travel and work abroad increased number of sporadic cases of cutaneous leishmaniasis in non-endemic areas should be taken into account.

Keywords: Cutaneous leishmaniasis, Non-endemic areas, Kazakhstan

Background

According to the WHO data, today there are 12 million patients with cutaneous and visceral leishmaniasis in the world and 350 million at risk (1). The annual incidence is worldwide from 400.000 to 600 000 new cases (1, 2). More than 90% of cutaneous leishmaniasis worldwide can be found in Afghanistan, Iran, Saudi Arabia, Syria, Brazil and Peru (1, 2). Incidence of leishmaniasis in Kazakhstan is rare and mainly on the south of the country. Over the last 10 years, 497 cases of cutaneous leishmaniasis and 9 cases of visceral leishmaniasis, including 6 fatalities were registered there (3). Children up to 5 years of age with cutaneous leishmaniasis are the most common

among patients with this disease in endemic areas (1, 2). But some authors argue that the majority of cases of cutaneous leishmaniasis are found in adult men between 20 and 40 years (4). Tourists and workers from the endemic areas have an increased risk of cutaneous leishmaniasis. Labor migration to Kazakhstan from neighboring countries with different endemic tropical diseases results in registration of these diseases which are not peculiar for the region. This creates certain difficulties both in diagnosis and management of such pathology. Besides, leishmaniasis evolution reveals some changes in the disease clinical picture which is the cause of diagnostic errors (4-7).

Case Presentation

The patient of 22 years of age, a builder by profession, Uzbekistan citizen, was hospitalized to the maxillofacial surgical department in March 27, 2010. He got ill 14 days ago with a focus of inflammation on his right cheek but did not go to the doctor. He tried to manage the disease himself, used liniments, and made an attempt to press the purulent secretion out. His state worsened, pain became severe, temperature increased up to 39 ^{0}C. In March 24, 2010 he went to private medical center where the abscess was opened and some drugs (antibiotic ointments) were recommended. In dynamics his state worsened again, edema became greater and covered right parotid-masticatory area, pain became severe, movements to open mouth became limited, temperature increased, general fatigue and fever were observed. In March 27, 2010 he went to traumatologic center where he was examined by maxillofacial surgeon and urgently hospitalized with preliminary diagnosis: putrefractive-necrotic phlegmon of the right buccal and parotid-masticatory area (Fig. 1).
His general condition was grave: due to evident endotoxicosis the patient was adynamic with severe diffuse muscles hypotony, fever 39 ^{0}C, yellow pale color of skin, subicteritiousness of sclera. Pulmonary respiration within all fields was normal, no rales, respiration frequency 32 in 1 min. Heart tones were distinct with stable tachycardia up to 100 in 1 min., blood pressure 80/60, and abdomen was soft but its volume increased, liver palpated 2 cm lower rib edge. Spleen was 8 cm lower rib edge, dense and painful. The skin in buccal and parotid-masticatory right area was infiltrated, edematous, warm in palpation. There was an ulcer with irregular borders on the skin and necrotic mass was seen in its bottom.

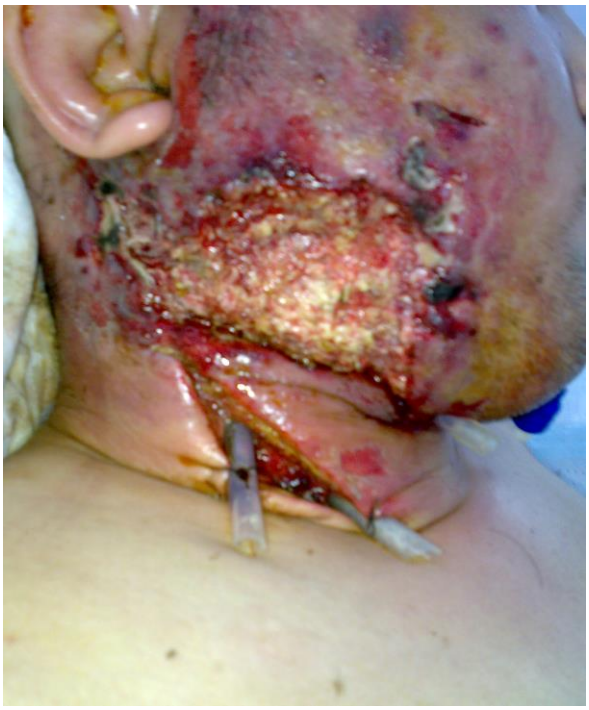

Fig. 1: Local picture in maxillofacial department arriving date (Original)

Hemogram in the patient's hospitalization showed anemia - Hb -105 g/l; erythrocytes - 3,7x 10^{12}/l, leukocytes 6,9x10^9/l, and accelerated blood-sedimentation test - 21 mm/h. Leucocytes formula with slight neutrophylis shift due to band forms (13%). Urinanalysis showed quantity - 100 mm, color - light-yellow, pH - 5, sugar, protein - 0,165 g/l, density - 1018, leucocytes - 4-2-5 in the field, and epithelial tissue 4-3-4 in the field.
The patient started to get nonspecific antibacterial, detoxication, analgetic and general health-improving therapy. Also we opened and drained cellular space of the right buccal and parotid-masticatory area. The material was sent to a histopathological

examination. Intensive therapy of the patient was not successful, general condition of the patient remained seriously bad due to the severe intoxication, intermitting fever and growing adynamic patient state (Fig. 2). Marked hepatosplenomegaly remained from the moment of the patient hospitalization. Pancytopenia, intensive anemia, poikilocytosis, erithrocytis anisocytosis and polychromatophilia progressed in blood analysis. Inflammatory infiltrate of the buccal and parotid-masticatory area expanded to the low submaxillary area and anterior-lateral right part of the neck, the area of the ulser itself increased. In two days after hospitalization the patient was operated on with general intratracheal ansthesia through the nose. The manipulation done was: "Opening and draining of low submaxillary area and lateral surface of the neck".

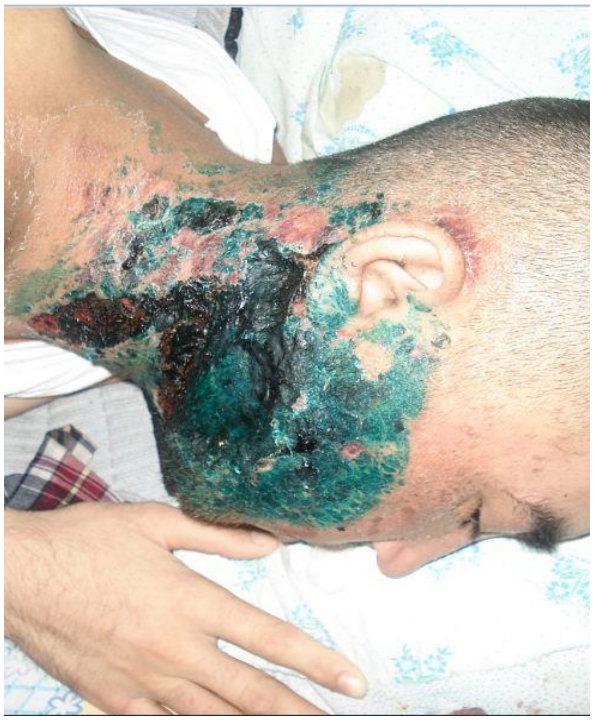

Fig. 2: Local picture after nonspecific therapy (Original)

The histopathological examination with staining by Romanovskiy-Giemza, examined under light microscope with high magnification (1000x) showed diffuse dermal infiltrate predominantly of macrophages with admixture of few lymphocytes, eosinophils and plasma cells. In a very small number of macrophages amastigotes were seen. On their surface and occasionally extracellularly rod-shaped kinetoplasts were noticeable.

Serologic examination for brucellosis, classical tiphus, enteric fever, yersiniosis, pseudotuberculosis was negative.

Antibodies to HIV were not discovered. Cutaneous leishmaniasis was suspected (for finding a very small amount of macrophages with amastigotes inside). For its verification biopsy of ulcer soft tissues, ulcer smears, blood smears and cerebrospinal puncture were done. But no leishmaniases were found (there was no possibility in our laboratory to do a PCR diagnostics or indirect immunofluorescence and ELIZA).

Cytomegalovirus, toxoplasmosis, malaria, AIDS, blood and oncological diseases, actinomycosis, pyodermia, dermatophytia, tuberculosis and sepsis were also excluded.

After discussion, on the medical consilium on the basis of epidemiological (he arrived from Uzbekistan in February 2010 which is endemic in cutaneous and visceral leishmaniasis), objective (hepatosplenomegalia, intermitting fever within the whole month in spite of intensive antibacterial therapy and ineffective surgery, severe anemia and progressive dystrophy) and laboratory data (the histopathological examination showed a very small number of macrophages with amastigotes inside, pancytopenia, anemia, proteinuria) patient's disease was diagnosed as cutaneous leishmaniasis.

Because of that specific therapy with glucantime (meglumine antimoniate) was started, according to the plan (18.6 ml (1506.6 mg) of glucantime in a 5% - 500 ml glucose solution intravenous drop infusions for 30 days) (3). This therapy gave a stable good clinical effect - temperature was normalized, hepatosplenomegaly decreased, endotoxicosis vanished, patient condition became better, hemogramm normalized. After 30 days the patient was discharged from the hospital with clinical improvement and final diagnosis: putrefactive-necrotic phlegmon of buccal and parotid-masticatory, low submaxillary area and anterior-lateral neck surface. Cutaneous leishmaniasis is a possible

case here (the diagnosis formulation in accordance with recommendations of the state sanitary-epidemiological surveillance) (3). In two years of observation we stated complete recovery confirmed by clinical and laboratory data.

Cutaneous leishmaniasis is a rare disease in Kazakhstan, especially in the north region (3). Because of higher rate of travel and work abroad increased number of sporadic cases of cutaneous leishmaniasis in non-endemic areas should be taken into account.

Conclusions

We believe that this case is interesting by the fact that we had a very rear disease case that can be observed in nonendemic area. It should be stressed that both clinical and laboratory data were not peculiar for this disease. Adults in endemic areas have stable immunity for protozoa infections. This made the diagnostic and timely management of the disease very difficult. But clinical effect of drug therapy which is specific for cutaneous leishmaniasis treatment proved, in spite of the absence of ulcer soft tissues, blood and cerebrospinal puncture *Leishmania*, that our diagnosis was correct.

Though endemic centers of cutaneous leishmaniasis are eradicated, the case described shows its relative frequency. The case, described by us, may be interesting for dermatologists, parasitologists, surgeons and other medical specialists.

Ethical considerations

Ethical issues (Including plagiarism, Informed Consent, misconduct, data fabrication and/or falsification, double publication and/or submission, redundancy, etc.) have been completely observed by the authors.

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