

Acta Medica International
2017, Volume 4(1):88-91.



Diffuse Cutaneous Leishmaniasis Presenting as Erythroderma

Entela Shkodrani¹, Ermira Vasili¹, Alert Xhaja¹, Silvan Frangaj², Amarda Cenko¹

¹Clinic of Dermato-Venerology, University Hospital Center "Mother Theresa", Tirana, ALBANIA.

²Hospital of Shkodra, Shkoder, ALBANIA

ABSTRACT

Introduction: Exfoliative Erythroderma syndrome is a serious, at times life-threatening reaction pattern of the skin characterized by generalized and uniform redness and scaling involving the entire skin and often associated with systemic toxicity, lymphadenopathy and fever. Diffuse Cutaneous Leishmaniasis observed in anergic patients with a low immune response. **Case report:** We are presenting a case of a 17-year-old patient diagnosed with Diffuse Cutaneous and Visceral recidival Leishmaniasis, which clinically presented Erythroderma and skin exfoliation. The most common causes of Erythroderma are Psoriasis, Allergic Dermatitis, Drug Reactions, Lymphoma and Pityriasis Rubra Pilaris. Conversely, Diffuse Cutaneous Leishmaniasis is characterized by the presence of non-ulcerative nodules which resemble Lepromatous Leprosy, scattered in every part of the body. To the best of our knowledge, the present clinical manifestation of Leishmaniasis has not yet been reported in the literature. **Conclusion:** In the correct clinical and epidemiological scenario, diffuse Cutaneous Leishmaniasis should be suspected in patients presenting Exfoliative Erythroderma and diffuse nodular lesions.

Key words: Exfoliative erythroderma, Cutaneous leishmaniasis, Visceral leishmaniasis.

DOI : 10.5530/ami.2017.4.16

Article History

Submitted: 26th Sep 2016

Accepted: 13th Dec 2016

Article Available online

www.actamedicainternational.com

Copyright

© 2016 AMI. This is an open-access article distributed under the terms of the Creative Commons Attribution 4.0 International license.

*Address for correspondence:

Entela Shkodrani,

Rr. Nikolla Jorga, Kulla 9, Ap.40,
Tirana, ALBANIA.

Ph no: 00355692081029

Email: entela.shkodrani@gmail.com



INTRODUCTION

The exfoliative erythroderma syndrome is a serious, at times life-threatening reaction pattern of the skin characterized by generalized and uniform redness and scaling involving the entire skin and often associated with systemic toxicity, lymphadenopathy and fever. Both stages, acute and chronic, merge one into the other. In the acute and subacute phases, there is a rapid onset of generalized vivid red erythema and fine branny scales. In the chronic exfoliative erythroderma syndrome, the thickening and scaling of the skin continues and becomes lamellar. About 50% of patients have a history of a pre-existing dermatosis, which can be diagnosed only in the acute and subacute stages. The most common cutaneous disorders (by frequency) are: psoriasis, allergic dermatitis, drug reactions, lymphoma and pityriasis rubra pilaris. Diffuse cutaneous leishmaniasis is observed in anergic patients with a low immune response. The infection is characterized by the presence of a primary element, which spreads and develops in different areas of the skin. The presence of non-ulcerative nodules which resemble lepromatous leprosy in every part of the

body is characteristic. Many cases of visceral leishmaniasis with the presence of hyperpigmented patches are reported. In some cases xerosis is present.

CASE REPORT

We are presenting a case of a 17-year-old patient diagnosed with diffuse cutaneous and visceral recidival leishmaniasis, which clinically presented erythroderma and skin exfoliation. A 17-year old male patient was hospitalized on 04/01/2011 with the diagnosis: Exfoliative erythroderma for determination. The objective examination of the patient demonstrated generalized erythema of the body accompanied by numerous scaling elements (Figure 1) and focal purulent erosive lesions, located mainly in hands and feet (Figure 2). In certain parts of the erythematous lesions, small, hard nodules are present, some of which eroded in the center. Nodules are asymmetrically distributed in certain parts of the body, back, arms and less on feet. The main subjective complaints of the patient are pruritus and purulent secretions discharge from the eroded lesions. The patient reports that the complaints started 2 weeks

ago and he didn't receive any medication. He was hospitalized for diagnosis determination and treatment. The patient's anamnesis shows that he suffered from the same pathology with the same clinical signs 2 years ago. He has been hospitalized in the Dermatology Clinic where he has been undergoing the routine examinations as well as the cutaneous biopsy, in order to determine one of the following diagnoses: ichtiosis linearis circumflexa, atopic dermatitis and pityriasis rubra pilaris. The biopsy didn't confirm any of these diagnoses. The patient was treated with systemic and local corticosteroids and he has not noticed any cutaneous elements during a 2-year period. He reports sporadic gland swallowing and ulceration in the body folds, which were treated with systemic antibiotics by the family doctor. Family members reported that the patient was diagnosed with visceral leishmaniasis 7 years ago in his city and was treated with Pentavalent antimony solution 5 ml IM for 3 weeks over a period of 3 months. Physical examination: active posture; afebrile state; cor with rhythmic tones, no pathologic murmurs heard; regular heart rate; on auscultation clear, no vesicular breath sounds; axillar, inguinal and laterocervical big, hard palpable lymph nodes. There are neither cutaneous signs of hepatic insufficiency, nor collateral circulation. The liver is palpable below costal margin, not hard; the spleen is not palpable. There are generalized lymph nodes (Figure 3.). There is no indication of hepatic chronic pathology. There are no indications of any connective tissue systemic pathology. The patient performed a lymph node echo, an abdomen CT and a lymph node biopsy, in order to rule out a cutaneous lymphoma, morbus hodgkin and visceral leishmaniasis. The retroauricular (Figure 4) and neck region echo revealed several hypoechogenic lymph nodes of different sizes. The abdominal CT with an 8 mm cut and venous contrast revealed enlarged hepatic and splenic. The spleen had a diameter of 14.1 cm, the pancreas was homogenous, with no structural changes. Both kidneys appeared to be normal. Aortic and pericaval lymph nodes of a diameter of 1.1 cm were seen. Urinary bladder was normal, minimal liquid was seen in pelvis. The digestive transit was normal. No



Figure 1: Generalized erythema and scaling elements.

changes were seen in the peritoneal layer. No bone changes were seen. Mesenteric lymph nodes with a diameter smaller than 1 cm were seen. Cutaneous biopsy of the small nodular lesions was performed and the following histologic diagnoses were asked: psoriasis vulgaris generalisata, ichtiosis vulgaris, pityriasis rubra pilaris and cutaneous leishmaniasis. The lymph node biopsy, nr. 659, performed in the University Hospital Center, Tirana, Department of Pathologic Anatomy, described as follows: a histiocytic proliferation in the interfollicular layers and cords as well as separated in the germinative centers. It didn't confirm any of the diagnoses listed above. The cutaneous biopsy, nr. 251, performed in the University Hospital Center, Tirana and the American Hospital in Tirana, confirmed the presence of a lympho-histiocytic, chronic, perivascular and perifollicular inflammation, as well as that of basophilic cells which coincide with the diagnoses of cutaneous leishmaniasis (Figure 5).

After the lymph node biopsy results, the examination was oriented towards the bone marrow aspirate, in order to confirm the visceral leishmaniasis. The bone marrow was rich in all 3 series of elements. Megakaryocytic series elements were found. Leishmania was not found.

The direct microscopic parasitologic examination from a nodus, located on the back of the patient resulted positive for cutaneous leishmaniasis (Figure 6, 7). Laboratory examinations revealed: LDH 363 U/L, IGG 907 mg/dl, IGA 328 mg/dl, 245 mg/dl, ELISA/At anti-Leishmania 24.7 UI/mL, ALP 173 U/L, LDH 414 U/L, total protein 7.6g/dl, FIB 224 mg/dl, PCR 7.7 mg/dl, Glicemia 138 mg/dl, Creatinine 0.83 mg/dl, AST 24 U/L, ALT 17 U/L, Wright test/Antibrucella totale 0, HIV/AIDS negative, VDRL negative, FTA-Abs negative, ANCA screen 0.34, ANA light positive, Urine complete normal, Complete blood count: WBC 5.2 K/ul, RBC 5.25 m/ul, HGB 14.1 g/dl, HCT 41.5%, MCV 79.0 fl, MCH 26.9 pg, MCHC 34.0 g/dl, RDW 14.9 %, PLT 224 K/ul, MPV 10.0 fL. HBsAg negative, Radiography of thorax, free phrenico-costal sinuses, normal mediastinum Echography of the testicles was requested. Testicles echography: both testicles of normal size and structure. The cutaneous biopsy was taken again from the hyperkeratotic elements located in the arm of the patient. The biopsy results from Istituto dermatologico dell'Immacolata



Figure 2: Plantar hyperkeratosis.

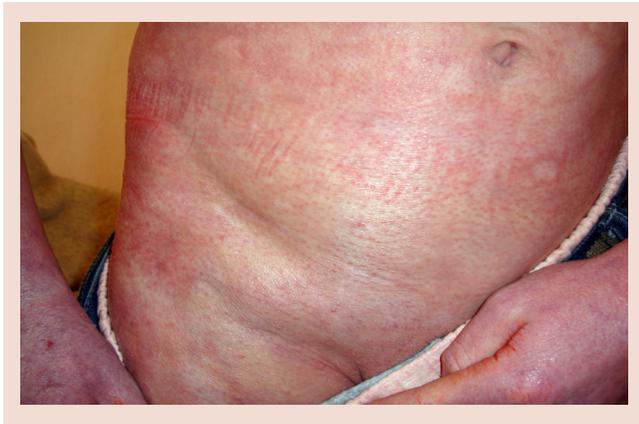


Figure 3: Cordon like lymphadenopathy.



Figure 4: Retroauricular Lymphadenopathy.

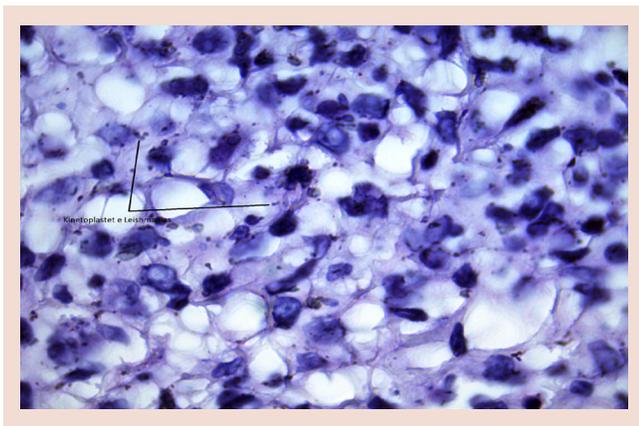


Figure 5: Macrophages situated in derma with kinetoplast(Arrow).

in Italy, date 23/03/2011, nr.003033 revealed: chronic lymphohistiocytic perivascular and perifollicular inflammation. Basophilic bodies that coincide with leishmaniasis are present. Based on the clinical, histopathological and parasitological results, the case was considered as Cutaneous Leishmaniasis with multiple elements, of the nodular dry variant. This form is very rare in Albania and the problem is further complicated by the fact that the patient has suffered visceral leishmaniasis 7 years ago and has been treated with pentavalent Anti-

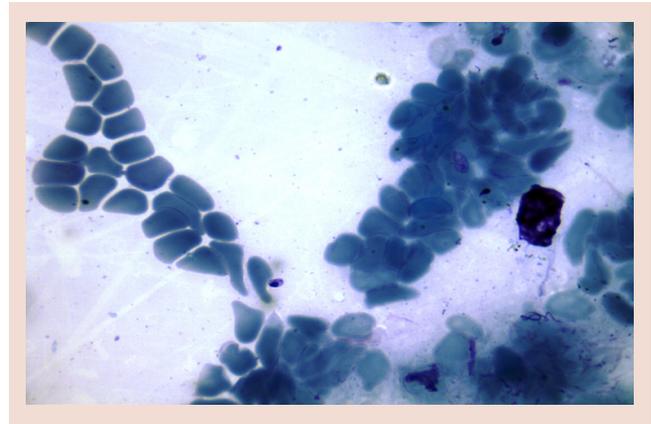


Figure 6: Presence of Leishmania in cutis.

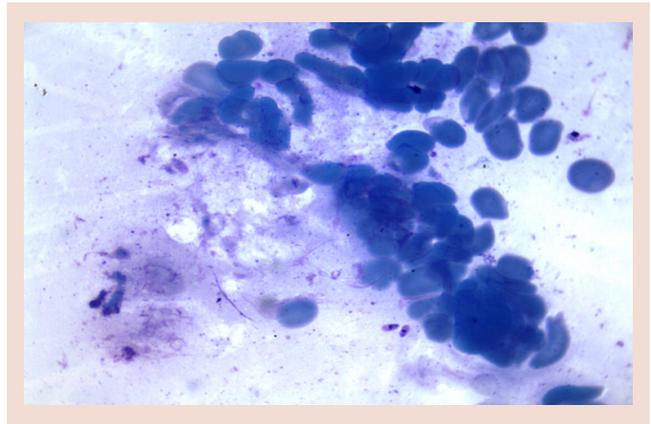


Figure 7: Presence of Leishmania in cutis.

mony solution. In this situation, a systemic treatment with Pentavalent Antimony accompanied by a careful monitoring of the biological parameters was recommended. Systemic treatment with Pentavalent Antimony 1.5g/5ml x2 / day was started for a period of 28 days.

DISCUSSION

The diagnosis of an exfoliative erythroderma is not easy and the history of a pre-existing dermatosis is the only key to conclude. The pathognomic signs and symptoms of the pre-existing dermatosis can help, for example the dark red color of the thick scales in psoriasis and their yellow color in pityriasis rubra pilaris, typical nail changes in psoriasis, lichenification, erosions and excoriations in atopic dermatitis, diffuse eczema, non-palmar hyperkeratosis with fissures in the T cell lymphoma, well confined plates within the erythroderma in pityriasis rubra pilaris, hyperkeratotic scales usually without hair fall in psoriasis etc. Visceral or cutaneous leishmaniasis is not mentioned in any case as a differential diagnosis of exfoliative erythroderma. In diffuse cutaneous leishmaniasis, the cutaneous elements are disseminated and resemble lepromatous leprosy. The condition generally starts with a primary lesion and later is further spread in different parts of the body. The lesions are ulcerated nodules full of parasites and mainly localized in feet, gluteal regions and face. In our patient, the

nodules were not ulcerated and were mainly localized in the arms, feet and back, on an erythematous skin. Unlike in lepromatous leprosy, there was no involvement of the peripheral nerves. On the other hand diffuse cutaneous leishmaniasis, does not invade the internal organs, responds partially to treatment alternatives and often relapses and chronicizes. The patient's family members refer that within a 3-4 year period, the patient has complained for continuous enlargement and ulceration of the lymph nodes, especially those inguinal and axillary, which would regress after the antibiotic therapy. The relapsing leishmaniasis is manifested by lesions located in the center or the periphery of the scar of previous nodules of leishmaniasis. The formation of new ulcers after primary infection can be noticed and usually cheekbones are affected. Over the years, these lesions tend to progress towards the center, forming a psoriasiform plaque. In visceral leishmaniasis, lymphadenopathy, characteristic nail changes and alopecia can be observed. Several cases are described in literature with the presence of the characteristic dark patches and, although rare, xerosis can also be seen, but there are no cases where a visceral leishmaniasis is manifested with generalized exfoliative erythroderma.

REFERENCES

1. Sehgal VN, Srivastava G. Erythroderma/generalized exfoliative dermatitis in pediatric practice: an overview. *Int J Dermatol.* 2006;45(7):831-9. Dermato-Venereology, Skin/VD Center, Sehgal Nursing Home, Panchwati, Azadpur, Delhi, India.
2. Yuan XY, Guo JY, Dang YP, Qiao L, Liu W, Erythroderma: A clinical-etiological study of 82 cases. *Eur J Dermatol* 2010;20(3):373-7.
3. Turchin I, Barankin B, Department of Family Medicine, University of Calgary, Alberta.
4. Dermacase. Erythroderma secondary to cutaneous T-cell lymphoma. *Can Fam Physician.* 2005;51:(963)971-3.
5. James, W D, Berger T G, *et al.* (2006). *Andrews' Diseases of the Skin: clinical Dermatology.* Saunders Elsevier. ISBN 978-0-7216-2921-6.
7. Singh N, Ramesh V, Arora VK, Bhatia A, Kubba A, Ramam M. Nodular post-kala-azar dermal leishmaniasis: a distinct histopathological entity. *J. Cutan. Pathol.* 1998;25(2):95-9.
8. Goulart LR, Goulart IM. Leprosy pathogenetic background: A review and lessons from other mycobacterial diseases. *Arch Dermatol Res.* 2009;301(2):123-37.
9. Gvozdenovic M, Nikulin E, Zec N, Kosoric D, Miladinovic Z. Kala Azar (Leishmaniasis visceralis) with muco-cutaneous lesions. *Acta Med Iugosl.* 1961;15:363-71.

ABOUT AUTHORS



Entela Shkodrani: Dermatologist, Associated Professor at Department of Dermatology and Venereology, UHC "Mother Theresa", Tirana, Albania.

Graduated as Medical Doctor in 1992, at the Faculty of Medicine, University of Tirana. Three years of training 1995-1998 in Dermatology and Venereology, at the Clinic of Dermatology, University Hospital Center of Tirana. Master Degree in 1998 with the thesis "Atopic Dermatitis in Children, ISSAC study in Tirana", post University School of the Faculty of Medicine, University of Tirana. PhD Degree in 2003 with thesis "A clinical-epidemiological study on Atopic Dermatitis in children" Faculty of Medicine, University of Tirana. In 2015 graduated as Associate Professor in Medicine, University of Medicine, Tirana. Member and General Secretary of the Albanian Association of Dermatology and Venereology. Member of EADV, European Academy of Dermatology and Venereology Member of ISD, International Society of Dermatoscopy Phone: 00355692081029, email address; entela.shkodrani@gmail.com, Mailing address: Rr. Nikolla Jorga, Kulla 9, Ap.40, Tirana, Albania



Prof. Ermira Vasili: Dermatologist, Chief of Department of Dermatology and Venereology, UHC "Mother Theresa", Tirana, Albania In 1982–1984 she was specialized in Internal Medicine In 1984–1985 she was specialized as dermatologist at the Faculty of Medicine, University of Tirana. She is a lecturer in the Faculty of Medicine, department of Dermatology from 1993 till now. She is chief of dermatology department from 2009. Member and President of the Albanian Association of Dermatology and Venereology.



Alert Xhaja: Dermatologist, PhD, Department of Dermatology, UHC "Mother Theresa", Tirana, Albania Graduated as Medical Doctor in 1992-1998, at the Faculty of Medicine, University of Tirana. He completed a three-years postgraduate specialization in dermatology and venereology at the University Hospital Centre "Mother Tereza", 2000- 2004 . Master Degree in 2007-2008 post University School of the Faculty of Medicine, University of Tirana. PhD Degree 2010–2016 with the thesis "Risk factors and quality of life in psoriatic patients in Albania", Faculty of Medicine, University of Tirana. He is a lecturer at the Faculty of Nursing, University of Tirana, Albania from 2005. Member of the Albanian Association of Dermatology and Venereology. Phone: 00355(0)692067146, email address; axhaja@gmail.com, Tirana, Albania



Silvan Frangaj: Dermatologist, He was born in Shkoder, Albania in 1981. He was graduated from University of Tirana Faculty of Medicine in 2005 and worked for 5 years as a family physician. In 2014 he completed a three-years postgraduate specialization in dermatology and venereology at the University Hospital Centre "Mother Tereza" Department of Dermatology, Tirane, Albania. He currently works as a dermatologist at the Shkoder Regional Hospital, Shkoder, Albania. He is member of Albanian Association of Dermatology and Venereology.



Amarda Cenko: Resident in Dermatology and Venereology Department, UHC "Mother Theresa", Tirana, Albania She was born in May 29–th, 1983, in Elbasan, Albania. She studied Medicine in the Faculty of Medicine, University of Tirana, Albania during 2002 –2007. In the following period 2009 – 2011 she worked in the National Center of Continuing Education. She is currently attending the specialization school in Dermatology Department at University Hospital Center "Mother Theresa" in Tirana, Albania. She is full member of Albanian Order of Physician and Albanian Association of Dermatology and Venereology.

How to cite this article: Shkodrani E, Vasili E, Xhaja A, Frangaj S, Cenko A. Diffuse Cutaneous Leishmaniasis Presenting as Erythroderma. *Acta Medica International.* 2017;4(1):88-91.