Tropical Biomedicine 40(2): 250-252 (2023)
https://doi.org/10.47665/tb.40.2.017

RESEARCH ARTICLE

A case report of an uncommon presentation of cutaneous leishmaniasis: A nose lesion

Alamin, A.A.[1*], Gebreyesus, M.W.[2], Mohamed, I.[3]

[1]Department of Pathology, College of Medicine, Taif University, P.O. Box 11099, Taif 21944, Saudi Arabia
[2]Department of Biology, Mai Nefhi College of Science, Mai Nefhi, Eritrea
[3]Department of Microbiology, College of medicine, Tail University, P.O. Box 11099, Taif 21944, Saudi Arabia
*Corresponding author: amakki@tu.edu.sa

ARTICLE HISTORY

Received: 6 May 2023
Revised: 2 June 2023
Accepted: 3 June 2023
Published: 30 June 2023

ABSTRACT

Leishmaniasis is a widely spread zoonotic disease caused by the bite of infected sandflies, particularly in developing countries. Cutaneous leishmaniasis can have a diverse range of presentations, ranging from minor skin nodules to significant mucosal damage. However, nose involvement is infrequent. Our report highlights a 15-year-old female patient with a persistent skin lesion on her nose for three months, which is a rare manifestation of cutaneous leishmaniasis. The lesion started as a raised spot with a brownish-red color and a crust but eventually developed into an ulcer that spread over the entire lobe of the nose and even moved toward the eye. Microscopic examination revealed the presence of *Leishmania* amastigotes, and a biopsy confirmed a diagnosis of cutaneous leishmaniasis. The patient received daily intravenous sodium stibogluconate doses of 9 mg/kg for 20 days, and three weeks later, there was a significant clinical improvement, with the ulcer beginning to heal and no more amastigotes visible on microscopic examination. It is crucial to keep cutaneous leishmaniasis in mind as a possible diagnosis for patients with skin lesions, even in regions where the condition is not prevalent.

Keywords: Leishmaniasis; cutaneous leishmaniasis; skin lesion; sodium stibogluconate; Eritrea.

INTRODUCTION

Leishmaniasis is caused by various types of *Leishmania* parasites and is transmitted by sandflies infected with the parasite as documented by Akhoundi *et al*. (2016). The disease can manifest in different forms, such as cutaneous, mucocutaneous, and visceral, with the specific species responsible and clinical presentation varying depending on the geographical location as demonstrated by Burza *et al*. (2018).

Leishmaniasis is prevalent in multiple regions worldwide, including Africa, Asia, and South America, as reported by the World Health Organization (WHO, 2023). According to Alvar *et al*. (2012), approximately 1.3 million new cases of this disease are documented each year. Their study highlighted *Leishmania major* and *Leishmania tropica* as the primary causes of cutaneous leishmaniasis in North Africa. On the other hand, in West Africa, *Leishmania donovani* is the prevailing cause of both visceral and mucocutaneous forms of the illness, as noted by Greenwood *et al*. (1984). Recent reports by Alamin (2020) indicate that cases of leishmaniasis have emerged in the capital city of Asmara, Eritrea, despite the lack of comprehensive documentation on the disease's incidence. These findings suggest that leishmaniasis poses a significant health concern in the country.

CASE REPORT

A 15-year-old female student, previously in good health, residing in Abardea, Asmara, presented herself at Halibet Referral Hospital with a two-week history of nasal congestion, a runny nose, swelling of the nasal area, and a persistent skin lesion located on the tip of her nose. The skin lesion initially appeared in September 2022 as a raised spot with a brownish-red color. Over time, it gradually increased in size and formed a crust, as shown in Figure 1.

Despite undergoing treatment with fusidic acid cream, applied 3 or 4 times a day for a duration of 10 days, the lesion on the patient's nose worsened, progressing into an ulcer accompanied by redness in the surrounding area. Over a period of three months, the skin lesion continued to enlarge, eventually extending to encompass the

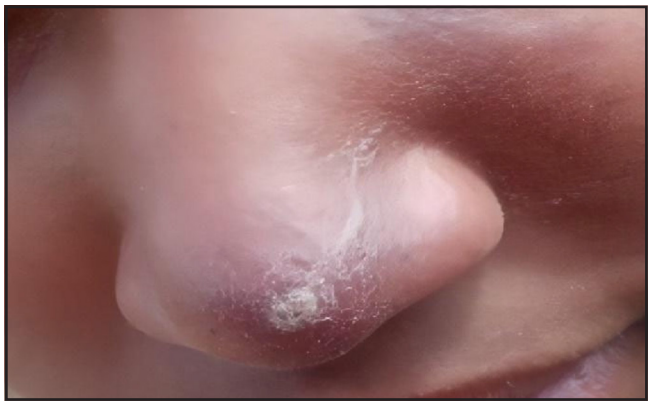

Figure 1. Nose lesion.

entire nasal lobe. It is important to note that the patient has not traveled, nor does she have a history of insect bites or exposure to sandflies. She resides in a densely populated household.

On examination, a single plaque with a crater-like ulcer, measuring 2×3 cm, was observed on the tip of the nose. The lesion was covered in scabs and had a moist, smooth, and shiny ulcer floor. Additionally, it displayed a well-defined raised erythematous margin.

Prior to admission, the complete blood count (CBC) test revealed an elevated total white blood cell (WBC) count, specifically in the neutrophil and monocyte populations. However, after a three-month period, a follow-up CBC test was conducted, indicating a return to normal WBC count.

Approximately 6 µL of blood was collected from the nose lesion and directly placed onto a pre-cleaned and labeled slide to create a thick smear for detecting the cutaneous *leishmania* parasite. The blood was spread on the slide and allowed to air-dry. After air-drying, the blood film was fixed with methanol and then stained using a 10% Giemsa stain solution for 15 minutes. The film was differentiated in buffered water (pH 6.8), rinsed, and air-dried before examination under an ordinary compound light microscope.

The microscopic examination revealed the presence of *Leishmania* amastigotes, (Figure 2) and a biopsy confirmed a diagnosis of cutaneous leishmaniasis.

The histological findings revealed the presence of *Leishmania* parasites within the affected tissue. The infected macrophages exhibited disrupted cellular integrity and loss of normal cellular morphology (Figure 3).

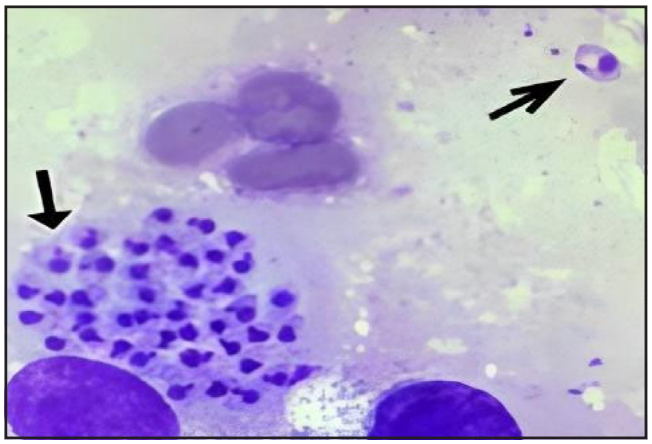

Figure 2. Smear of skin lesion demonstrating amastigotes.

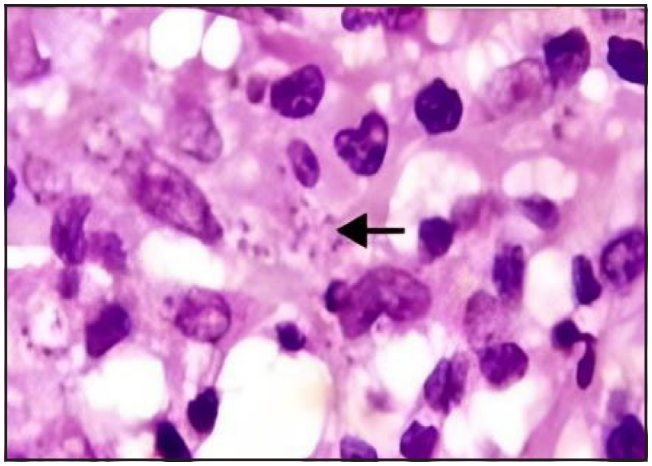

Figure 3. Histology of cutaneous leishmaniasis H & E-stain.

The patient received treatment with a daily intravenous dose of 9 mg/kg of sodium stibogluconate for 20 days, and after three weeks, the patient exhibited significant clinical improvement. The ulcer began to heal, and no more amastigotes were visible on microscopic examination.

DISCUSSION

Leishmaniasis is a neglected tropical disease prevalent in many parts of the world, particularly in developing countries. As reported by Hotez *et al*. (2012) the epidemiology of the disease is complex, with different species and forms of the disease prevailing in different regions.

Rhajaoui *et al*. (2012) identified three distinct *Leishmania* species, namely *Leishmania major*, *Leishmania tropica*, and *Leishmania infantum*, as the causative agents of cutaneous leishmaniasis. However, Masmoudi *et al*. (2007) reported that each species has its own distinct clinical manifestation.

Peter *et al*. (2009) reported that various species of *Leishmania*, including *Leishmania major*, *Leishmania tropica*, and *Leishmania aethiopica*, are responsible for causing cutaneous leishmaniasis in East Africa.

The clinical presentation of leishmaniasis is not only determined by the specific species of *Leishmania* causing the infection but also influenced by the host's immune response, primarily regulated by cellular immunity. As a result, a wide range of symptoms can occur even within a limited geographical area. In the study conducted by (Bari, 2009), it was found that various factors can impact the immune response to *Leishmania* infection. These factors include the parasite's resistance to macrophages, dysfunction of macrophages, reduced production of interferon, an imbalance between T helper cell type 1 and T helper cell type 2 cells, impaired recruitment of immune cells, and heightened delayed hypersensitivity. Additionally, the number of parasites introduced, the location of infection, the host's nutritional state, advanced age, menopause, oral steroid use, and wound contamination with inorganic materials can also impact the clinical outcome.

According to Hosseinzadeh *et al*. (2012), making a diagnosis of cutaneous leishmaniasis can be challenging due to its rarity and limited diagnostic options in resource-limited settings. However, an accurate diagnosis can be achieved with careful clinical assessment and fine-needle aspiration cytology in endemic areas. Proper diagnosis helps determine the overall prevalence of the disease and allows for prompt treatment to prevent mucocutaneous complications and to perform clinical therapeutic trials.

The treatment of cutaneous leishmaniasis depends on the type of leishmaniasis, the species of *Leishmania*, and the patient's overall health. Available treatment options include pentavalent antimony, miltefosine, and amphotericin B. In the treatment of cutaneous leishmaniasis, especially in the Old World, topical application of specific medications such as paramomycin, ketoconazole, antimonials, azithromycin, and imiquimod has been utilized. In some cases, a combination of drugs may be used.

According to the World Health Organization (WHO, 2023), measures should be taken to reduce exposure to infected sandflies in order to prevent cutaneous leishmaniasis. This includes using insecticide-treated bed nets, wearing long-sleeved clothing, and avoiding outdoor activities during peak sandfly biting hours. Prompt and effective treatment of leishmaniasis cases can also help prevent the spread of the disease to others.

CONCLUSION

Uncommon presentations of cutaneous leishmaniasis are infrequent and can frequently cause a delay in diagnosis. It is thus crucial to consider uncommon manifestations of cutaneous *Leishmania* infection during clinical examinations, especially in individuals living in or visiting an endemic region, to ensure timely and appropriate treatment.

Funding
The authors declared that this study had received no financial support.

Competing interests
The authors declares that they have no competing interests.

Consent
Informed consent was obtained from the patient, allowing for the publication of this case report.

REFERENCES

Alamin, A.A. (2020). Visceral leishmaniasis in a non-endemic region of Eritrea. *Cureus* **12**: e11318. https://doi.org/10.7759/cureus.11318

Akhoundi, M., Kuhls, K., Cannet, A., Votýpka, J., Marty, P., Delaunay, P. & Sereno, D. (2016). A historical overview of the classification, evolution, and dispersion of *Leishmania* parasites and sandflies. *PLoS Neglected Tropical Diseases* **10**: e0004349. https://doi.org/10.1371/journal.pntd.0004349

Alvar, J., Vélez, I.D., Bern, C., Herrero, M., Desjeux, P., Cano, J., Jannin, J., den Boer, M. & WHO Leishmaniasis Control Team (2012). Leishmaniasis worldwide and global estimates of its incidence. *PLoS One* 7: e35671. https://doi.org/10.1371/journal.pone.0035671

Bari, A.U. (2009). Clinical spectrum of nasal leishmaniasis in Muzaffarabad. *Journal of the College of Physicians and Surgeons Pakistan* **19**: 146-149.

Burza, S., Croft, S.L. & Boelaert, M. (2018). Leishmaniasis. *Lancet* **392**: 951-970. https://doi.org/10.1016/S0140-6736(18)31204-2

Greenwood, B.M., Ajdukiewicz, A.B., Conteh, S., Hagan, P., Mabey, D.C. & Panton, L.J. (1984). Leishmaniasis in The Gambia. 3. Is its incidence increasing? *Transactions of the Royal Society of Tropical Medicine and Hygiene* **78**: 407-409. https://doi.org/10.1016/0035-9203(84)90135-4

Hotez, P.J., Savioli, L. & Fenwick, A. (2012). Neglected tropical diseases of the Middle East and North Africa: Review of their prevalence, distribution, and opportunities for control. *PLoS Neglected Tropical Diseases* **6**: e1475. https://doi.org/10.1371/journal.pntd.0001475

Hosseinzadeh, M., Omidifar, N. & Lohrasb, M.H. (2012). Use of fine needle aspiration cytology in the diagnosis of cutaneous leishmaniasis: A comparison with the conventional scraping method. *Tropical Doctor* **42**: 112-113. https://doi.org/10.1258/td.2011.110420

Masmoudi, A., Ayadi, N., Boudaya, S., Meziou, T.J., Mseddi, M., Marrekchi, S., Bouassida, S., Turki, H. & Zahaf, A. (2007). Clinical polymorphism of cutaneous leishmaniasis in centre and south of Tunisia. *Bulletin de la Société de Pathologie Exotique* **100**: 36-40.

Peter, K., Ngure, A., Zipporah, W., Ng'ang'a, G., Rukunga, W.K. (2009). A review of leishmaniasis in Eastern Africa. *Journal of Nanjing Medical University* **23**: 79-86. https://doi.org/10.1016/S1007-4376(09)60032-0

Rhajaoui, M., Sebti, F., Fellah, H., Alam, M.Z., Nasereddin, A., Abbasi, I. & Schönian, G. (2012). Identification of the causative agent of cutaneous leishmaniasis in Chichaoua province, Morocco. *Parasite* **19**: 81-84. https://doi.org/10.1051/parasite/2012191081

WHO. (World Health Organization) (2023). Leishmaniasis. https://www.who.int/news-room/fact-sheets/detail/leishmaniasis. Accessed 26 January 2023.