Nasal Leishmaniasis Misdiagnosed With Intranasal Polyp in a Patient Candidate for Rhinoplasty

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Zakaria Zakariaei[1,2], Mahdi Fakhar[1], Simin Bari[1], Majid Derakhshani[1], Elham Sadat Banimostafavi[1,3] and Mostafa Soleymani[1]

[1]Iranian National Registry Center for Lophomoniasis and Toxoplasmosis, Imam Khomeini Hospital, Mazandaran University of Medical Sciences, Sari, Iran. [2]Toxicology and Forensic Medicine Division, Mazandaran Registry Center for Opioids Poisoning, Antimicrobial Resistance Research Center, Imam Khomeini Hospital, Mazandaran University of Medical Sciences, Sari, Iran. [3]Department of Radiology, Imam Khomeini Hospital, Mazandaran University of Medical Sciences, Sari, Iran.

ABSTRACT: Mucosal leishmaniasis (ML) is a chronic and rare form of leishmaniasis that causes malignant lesions in the mucosa of the nasal, pharyngeal, and laryngeal regions. We describe a 29-year-old woman who had been suffering from an intranasal polyp for 3 years. The polyp recurred annually after surgical removal, and was diagnosed as nasal leishmaniasis.

KEYWORDS: Intranasal polyp, mucosal leishmaniasis, nasal leishmaniasis, rhinoplasty

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CORRESPONDING AUTHOR: Mahdi Fakhar, Iranian National Registry Center for Lophomoniasis and Toxoplasmosis, Immam Khomeini Hospital, Mazandaran University of Medical Sciences, Farah-Abad Road, P.O Box: 48471- 91971, Sari 48166-3313, Iran. Email: mahdifakhar53@gmail.com

Introduction

Leishmaniasis is a zoonotic disease caused by different species of protozoans belonging to the genus *Leishmania*, and it is considered one of the most significant parasitic diseases transmitted by vectors worldwide. The vectors of this disease are the sandflies of the *Phlebotomus* genus in the Old World and *Lutzumia* in the New World.[1] Leishmaniasis affects over 12 million individuals in more than 100 countries.[2] Due to urbanization, climate change, and migration, the number of cases of this disease is rapidly increasing.[3] Living in crowded environments such as barracks, traveling to endemic areas, immune system deficiencies, and organ transplants are the most critical factors that increase the risk of developing this infection.[4]

Leishmaniasis is a disease that can be classified into cutaneous, mucocutaneous, and visceral forms, each with a wide range of clinical manifestations. However, new terms are now used to describe specific symptoms associated with *Leishmania* spp., infection. Mucosal leishmaniasis (ML) refers to the involvement of mucosal tissues by the parasite.[5] In particular, ML affects the mucous membranes in the upper respiratory tract (from the inner nostril wall to the larynx) and the oral cavity. Pharyngeal and laryngeal involvement usually occurs later as the disease progresses. The onset of ML is characterized by nonspecific inflammation symptoms such as nasal congestion, erythema, edema, serous rhinorrhea, and epistaxis. As the disease worsens, patients may experience dysphagia and dysphonia due to soft tissue deterioration in the nose, mouth, and throat, and the disease may spread to the lower respiratory tract.[6]

In Iran, leishmaniasis can present as cutaneous leishmaniasis, localized *Leishmania* lymphadenitis, visceral (kala-azar), and rarely ML.[7] ML is a less common form of the disease in Iran and is more prevalent in regions such as South America, Asia, Europe, and Africa. The Amazon region of Latin America is considered the most important endemic location for ML. However, due to the relatively low incidence of ML outside of this area, it may be less suspected and reported in other parts of the world.[6]

Several methods can be used to diagnose leishmaniasis, including needle biopsy, bone marrow aspiration, lymph node and spleen puncture, and cytology of skin and mucosal abrasions.[8] Polymerase chain reaction (PCR) is a sensitive and reliable test for detecting and identifying *Leishmania* parasites in clinical samples.[9] However, the diagnosis of mucosal leishmaniasis (ML) is primarily based on the identification of *Leishmania* amastigotes in mucosal lesions. The most common technique for visualizing these bodies is histological diagnosis, which involves the detection of amastigotes in mucosal samples stained with Giemsa or hematoxylin and eosin.[7]

ML is a relatively uncommon condition that occurs when *Leishmania* amastigotes spread from the skin to the mucosa through hematogenous or lymphatic diffusion. In some cases, ML can occur after a mosquito bite on the throat, nose, or other mucous membranes.[8] Given the frequent reports of mucosal involvement, an ear, nose, and throat (ENT) examination plays a crucial role in the differential diagnosis of ML. Conducting an ENT examination is often the first step in assessing suspected ML cases, which can prevent misdiagnosis and inappropriate treatment. Therefore, it's important for ENT specialists and other clinicians to be aware of this complication, particularly for patients with a positive travel history to endemic areas. Herein, we present an unusual case of ML in a young woman who had

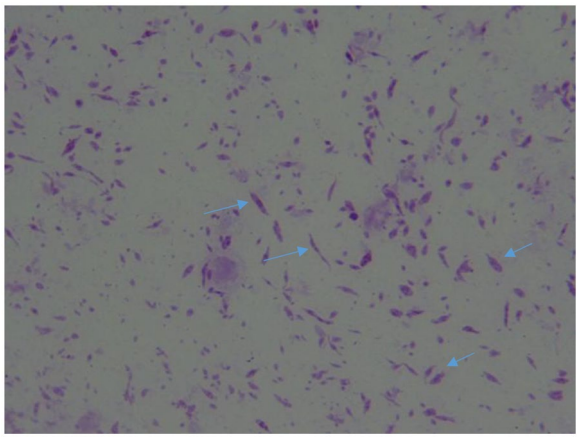

Figure 1. A direct smear of FNA sample taken from intranasal lesion stained with Gram stain showing a numerous developed and undeveloped promastigote of *Leishmania* parasite (arrowhead). (×1000).

a history of travel to a known endemic area for cutaneous leishmaniasis (CL). The patient was initially misdiagnosed with an intranasal polyp shortly after rhinoplasty.

Case Presentation

On July 2019, a 29-year-old female with no history of immunodeficiency or underlying diseases presented to an ENT clinic in Mazandaran province, northern Iran. She had a 3-month history of an itching intranasal lesion on the right side of her nasal canal and recurrent nasal congestion each year. An ENT specialist diagnosed her with an intranasal polyp and recommended polypectomy and rhinoplasty. Three months after surgery, she returned to the specialist with swelling and inflammation in the nasal cavity, suggesting recurrence. To rule out bacterial infections as possible complications of polypectomy, the specialist performed a fine needle aspiration (FNA) from the intranasal lesion. The FNA fluid specimen was stored at ambient temperature for 3 hours before being delivered to the laboratory, where a direct smear was prepared, fixed, and stained using the Gram technique. Microscopic examination revealed spindle-shaped *Leishmania* promastigote-like structures without any amastigotes of the parasite (Figure 1).

To definitively confirm these identified promastigotes, an internal transcribed spacer1 (ITS1) -polymerase chain reaction (ITS1-PCR)-based assay was used.[10] To DNA extraction, the first step was to scrape the stained FNA smear using a sterile scalpel. The obtained scraping was then mixed with 100 µL of lysis buffer containing Tris-Hcl (50 mM pH7.6), EDTA (1 mm), Tween 20 (1%), and proteinase K solution (19 mg/mL) in the amount of 10 µL. Subsequently, the modified salting-out method was employed for DNA purification. Then, using ITS1-PCR, *Leishmania major* were identified.[10]

It makes clear that the delay in sample preparation and storage the FNA sample at room temperature (about 24°C–26°C) caused the amastigotes to transform into promastigotes, as seen in sandflies' midgut after feeding on blood. Also, in reviewing the history, the patient reported traveling to an endemic area of CL in Mashhad (eastern Iran) suburb area 3 months prior and noticing a bite in her nasal cavity. Finally, with this history and laboratory sample, intranasal ML was diagnosed, and the patient received glucantime treatment (20 mg/kg/day for 4 weeks). Six months later, there were no reported side effects, and the lesion improved. Written informed consent for publication of this case report was obtained from the patient. This study was conducted according to the Declaration of Helsinki Principles. Also, CARE guidelines and methodology were followed in this study.

Discussion

ML is a rare disease worldwide, even in endemic regions such as Iran,[7,11] although it is the most severe form of leishmaniasis due to its progressive lesions. These lesions can destroy not only the structure of the nose, but also the bony structures of the face, throat, and larynx.[12] In some patients with ML, an increase in the thickness of the nasal mucosa and sinuses may be observed.[13-15] While MCL is an uncommon clinical feature in Iran, there have been several diverse cases of patients with mucosal (ML) or MCL in various parts of the country over the past 50 years.[14]

Our patient was initially misdiagnosed with a polyp and underwent unnecessary polypectomy and rhinoplasty before ML was identified. Physicians in non-endemic areas may encounter patients with imported leishmaniasis if they have traveled to areas where CL is endemic. It's crucial for clinicians to be aware of ML's clinical features and the possibility of delayed onset, which can occur years after infection.[16] As such, physicians should always ask about travel history to endemic areas. The gold standard for diagnosing *Leishmania* amastigotes in mucosal lesions is histology, and staining techniques like Giemsa or hematoxylin-eosin can confirm its presence.

Diagnosing parasitic infections can be challenging since results from standard diagnostic tests like direct smear, culture, and PCR may not always detect the presence of the parasite. To increase diagnostic sensitivity, several simultaneous diagnostic procedures may be necessary. Computed Tomography (CT) is a useful tool for assessing early airway involvement and monitoring the response to treatment.[15,16] Clinicians must consider differential diagnoses such as Wegener granulomatosis, midline granuloma, lymphoproliferative disease, sarcoidosis, recurrent polychondritis, and non-keratinized squamous cell carcinoma when diagnosing ML. These conditions can have similar symptoms and findings, making it essential to rule them out before reaching a definitive diagnosis.

Conclusion

In any patient with a mucosal lesion on the head and neck, surgeons should not perform a resection immediately.

The patient's medical and travel history should be carefully considered. Mucosal sampling should be done for definitive diagnosis and to evaluate the differential diagnosis with ML because, in some cases, the patient's problem will be improved with drug treatment and non-invasive procedures. Therefore, it is important for ENT specialists and other clinicians to be aware of this complication, especially if they have a positive history of traveling to endemic areas. As a whole, ML should be ruled out in all patients who are candidates for polypectomy in endemic areas of CL to prevent severe complications.

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None.

Author Contributions
MF, ZZ, were involved in the interpretation and collecting of data and editing of the manuscript. ESB, MS involved in writing and preparing the final version of the manuscript. SB and MD were responsible for collecting data and submitting the manuscript. All authors reviewed the paper and approved the final version of the manuscript.

Ethical Approval
The study was approved by our local ethics committee.

Informed Consent
Written informed consent for publication of this case report was obtained from the patient.

Data Availability Statement
The data are available with the correspondence author and can be achieved on request.

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