

CASE REPORT

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# Mucosal leishmaniasis of the lips and cheeks: a first concomitant presentation of visceral and mucosal leishmaniasis in a patient living with HIV/AIDS in Monastir, Tunisia

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## Abstract

**Background** Visceral Leishmaniasis (VL) is the most severe and fatal disease if left untreated. In people living with HIV/AIDS (PLHA), VL is considered an emerging opportunistic infection. The aim of this manuscript was to report a first case in Tunisia of a concomitant presentation of visceral and oral leishmaniasis in a patient LHA. A systematic review of the literature was performed according to PRISMA guidelines, as well.

**Case presentation** The patient, a 43-year-old heterosexual man, treated for HIV/AIDS was referred for macrocheilitis of the upper and lower lips. A noticeable nodular and painless swelling extending to the cheeks' mucosa was noted. The patient's poor oral hygiene was evident due to the presence of multiple dental caries. Histological analysis of the biopsied lower lip sample revealed the presence of numerous *Leishmania* amastigotes. The diagnosis of VL was clinically confirmed by the presence of a mild splenomegaly and pancytopenia and biologically by the identification of the parasite using PCR Lei and the species *L. infantum* involved using RFLP-PCR and culture. The treatment consisted of an intravenous administration of liposomal Amphotericin B (Ambisome<sup>®</sup>, 40 mg/kg/weight) for a period of 6 weeks. A favorable outcome was noted after one year with the resolution of clinical symptoms and a negative *Leishmania* blood PCR test. After 2 years, the patient remained asymptomatic but showed a positive *Leishmania* blood PCR test. Dolutegravir<sup>®</sup> was introduced in the patient's ART regimen.

**Conclusions** To the best of our knowledge, this is the first case report in Tunisia of atypical VL diagnosed through an uncommon oral location in an HIV/AIDS co-infected patient. Since VL is a severe and potentially fatal disease, it is essential for dentists to perform a thorough clinical examination and adopt a multidisciplinary approach in order to ensure an early diagnosis and an effective treatment outcome.

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## Introduction

Leishmaniasis remains one of the world's most devastating neglected tropical diseases. The Americas, East Africa, North Africa, West Asia, and Southeast Asia are the main four regions in the world where Leishmaniasis is a major health problem. In 2018, 92 countries were considered endemic, and more than 1 billion people are at risk of being infected [1]. The disease is caused by protozoan parasites that are transmitted to humans through the bite of an infected female phlebotomine sandfly. In fact, more than 20 species of *Leishmania* are involved in the disease, and more than 90 species of sandfly are known to transmit the parasite [2, 3]. Over 70 species of wild and domestic animals have been identified as confirmed or potential reservoirs of *Leishmania* parasites, including bats, dogs, rodents, opossums, anteaters, sloths, hyraxes, and marsupials [4]. Three major forms of the disease are described: cutaneous Leishmaniasis (CL), the most prevalent form; visceral Leishmaniasis (VL), or Kala-Azar, the most severe and fatal disease if left untreated; and mucocutaneous Leishmaniasis, the most disfiguring form [1, 3]. Isolated oral mucosal forms of Leishmaniasis are rare and may occur in immunocompetent patients [5–7]. According to the World Health Organization (WHO), an estimated 30,000 and 1 million new cases of VL and CL, respectively, are reported each year [1]. However, this number could be underestimated. In the Middle East and North Africa (MENA) region, CL and VL are the most important infections and are caused by *L. infantum*, *L. donovani*, and occasionally *L. tropica* [8]. In Tunisia, four clinical conditions are described: VL, sporadic CL, chronic CL, and zoonotic CL. VL is caused by *Leishmania infantum*, and the dog is the main reservoir [4, 9]. Recent epidemiological data are lacking. In 2009, an annual incidence of VL varying between 50 and 100 cases has been reported. Children under 5 years old remain most commonly affected by the disease (80%). Annually 10 VL cases per 100,000 children were reported. Although rare in adults, VL cases are on the rise [10].

In people living with HIV/AIDS (PLHA), VL is considered an emerging opportunistic infection. Nearly 35 countries have reported cases of VL/HIV co-infection, with higher rates recorded in Brazil, India (Bihar), and Ethiopia. The risk of developing active VL is 100 to 2320 times higher in HIV patients [11]. In Tunisia, HIV/VL co-infection is rare. Data are lacking and are reported as case series [12].

Co-infected patients may harbor a significant load of the parasite in their blood, making them a reservoir and source of infection for the vector. The parasite can also accelerate immune suppression through virus replication stimulation [11, 13]. As a result, the diagnosis may

be challenging due to the absence of common leishmaniasis symptoms such as fever and splenomegaly [11, 14, 15]. Atypical clinical presentations have been reported in unusual sites such as the oral and gastrointestinal mucosa, lymph nodes, skin, pleura, and respiratory tract [16, 17]. The treatment of VL in HIV co-infected individuals is based on the use of lipid formulations of Amphotericin B (LAB), which can be infused at a dose of 3–5 mg daily to a total dose of 40 mg/kg or given in 10 intermittent doses (on days 1–5, 10, 17, 24, 31, and 38) [18]. As reported, VL is more severe in HIV-infected individuals, with an outcome marked by relapse and death [11, 19]. The aim of this manuscript was to report the first condition, in Tunisia, of atypical VL diagnosed through an unusual oral involvement and discuss clinical, biological features and treatment outcomes, based on a review of the literature.

## Case presentation

The patient, a 43-year-old heterosexual man, from the region of Moknine (Monastir-Tunisia), living with HIV/AIDS and treated for a chronic schizophrenia (Haldol® and Largactil®), was referred in March 2016 by the psychiatric department, for a swelling of the right lower mandibular cheek. The diagnosis of cellulitis of dental origin was made, at that time, on the basis of a poor oral hygiene and the presence of multiple dental infectious foci. Systemic antibiotics ( $\beta$ -Lactams 2 g/day and Metronidazole 750 mg/day for 7 days) and pain killers were prescribed as well as a panoramic radiography. The patient didn't show up for teeth extractions. In June 2017, he was referred by the infectious diseases' department for a macrocheilitis of both upper and lower lips. As for his medical history, the patient was treated for hypothyroidism (L-thyroxine), iron-deficiency anemia, and HIV/AIDS co-infected with HBV since 1994. In Germany, he was put on Zidovudine® and Nevirapine®, with poor compliance with treatment. On his first consultation in the infectious diseases' department in 2004, his CD4 count was 625/mm<sup>3</sup> and viral load count was 2625 copies/mm<sup>3</sup>. He was treated with Zidovudine®/Lamivudine® and Efavirenz® changed to Zidovudine®/Lamivudine® and Indinavir® because of Efavirenz® intolerance. In 2008, he was put on Zidovudine®/Lamivudine and Lopinavir® with a poor compliance to treatment. In 2010, virological and immunological failure was diagnosed with a CD4 count of 255/mm<sup>3</sup> and a viral load count of 87.932 copies/mm<sup>3</sup>. Because of the occurrence of pulmonary tuberculosis, the treatment was shifted to Tenofovir®/Emtricitabine® and Reyataz®. The viral load was undetectable and the CD4 count was 81/mm<sup>3</sup>.

During the patient's oral examination, a history of a swelling of the lower mandibular cheek that appeared 12

months ago, spontaneously resolved, and reappeared on the upper lip was reported. No history of dental pain was recorded. A noticeable nodular swelling with multiple fissures of both the upper and lower lips was noted. The swelling was extending to the cheeks' mucosa. Upon palpation, the swelling was painless and appeared inflammatory. Additionally, small eruptions (2 to 3) were observed at the corner of the mouth (Fig. 1A–D). Facial palsy, fissured tongue, and cervical adenopathy were absent. The patient's poor oral hygiene was evident due to the presence of multiple dental caries and infected roots. However, there was no tooth mobility or periodontal pockets. Panoramic radiography confirmed the presence of dental infectious foci without any bone anomalies (Fig. 2). Since a dental cause for the swelling was ruled out; and various infectious and non-infectious diagnoses were considered, an incisional biopsy of the lower lip was performed under local anesthesia. The incision was very hemorrhagic, but homeostasis was achieved through a surgical wound suturing. The histological analysis revealed a significant lymphocyte inflammatory infiltrate with multiple round to oval bodies in the cytoplasm of macrophages consistent with *Leishmania* amastigotes (Fig. 3A and B). The patient returned to the infectious diseases' department for further exploration. In fact, a mild splenomegaly was detected during the clinical examination with no associated fever. The biological results showed pancytopenia, with a total white blood cell count of 900/



**Fig. 2** Panoramic radiography shows multiple dental caries, infected dental roots, and periapical lesions, related to root

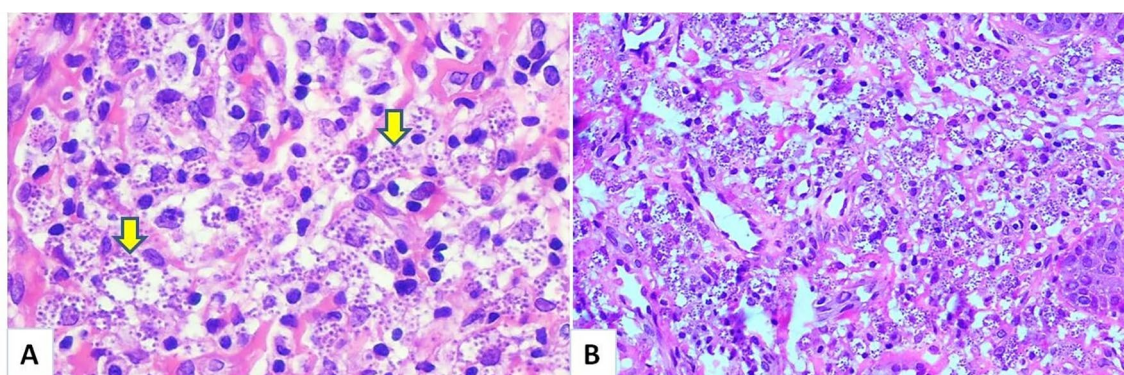
$\text{mm}^3$ , neutrophils of  $600/\text{mm}^3$ , hemoglobin of  $7.2 \text{ g/dL}$ , and a platelet count of  $62,000/\text{mm}^3$ . The CD4 count was  $80 \text{ cells}/\text{mm}^3$ . No other opportunistic infections were noted. The diagnosis of VL with an unusual oral manifestation was made.

Several methods were used to detect, isolate and identify *Leishmania* species involved in the present case. Microscopic observation of *Leishmania* was positive after May Grunwald-Giemsa staining (MGG) of concentrated leukocytes isolated from the patient's blood sample (Fig. 4A). Immunoprinting using *Leishmania* Western Blot IgG LDBIO Diagnostics<sup>®</sup> yielded positive results and showed the presence of antibodies



**Fig. 1** A–C Clinical views showing diffuse swelling of both upper and lower lips extending to the mucosa of the cheeks. Note the presence of fissures in the lower lip and cheeks. D Poor oral hygiene





**Fig. 3** **A, B** Histological finding of the biopsied lower lip show a significant lymphocyte inflammatory infiltrate with multiple round to oval bodies in the cytoplasm of macrophages, which were consistent with *Leishmania* amastigotes (hematoxylin and eosin staining,  $\times 400$  magnification)

(IgG) to two *Leishmania* protein bands: p14 and p16 (Fig. 4B). The identification of the parasite's genus from the blood sample and from the histological slices of the biopsied lower lip was performed by PCR, using a single pair of primers (Lei70L and Lei70R) to amplify a conservative region of the 18S rRNA (345 base pairs or bp) [20] (Fig. 4C). The PCR–RFLP was used to differentiate between the species *L. infantum*, *L. major* and *L. killicki* using the ITS1 locus (Internal Transcribed Spacer 1, 300–350 bp), which is flanked by two highly conserved regions (18S rRNA et 5.8S). This locus is characterized by its polymorphism among *Leishmania* species [21, 22]. In fact, 3 reference species were used, based on parasites' species involved in Leishmaniasis in Tunisia [9]: *L. infantum* MON-24 (MHOM/DZ/82/LIPA59), *L. major* MON-25 (MHOM/MA/81/LEM265) and *L. killicki* MON-8 (MHOM/TN/80/LEM163) (Reference National Center for Leishmaniasis, Montpellier, France) (Fig. 4D). The confirmation of *L. infantum* was done through culture using the appropriate medium for samples obtained from the blood and small eruptions scraped from the corner of the mouth (Fig. 4E).

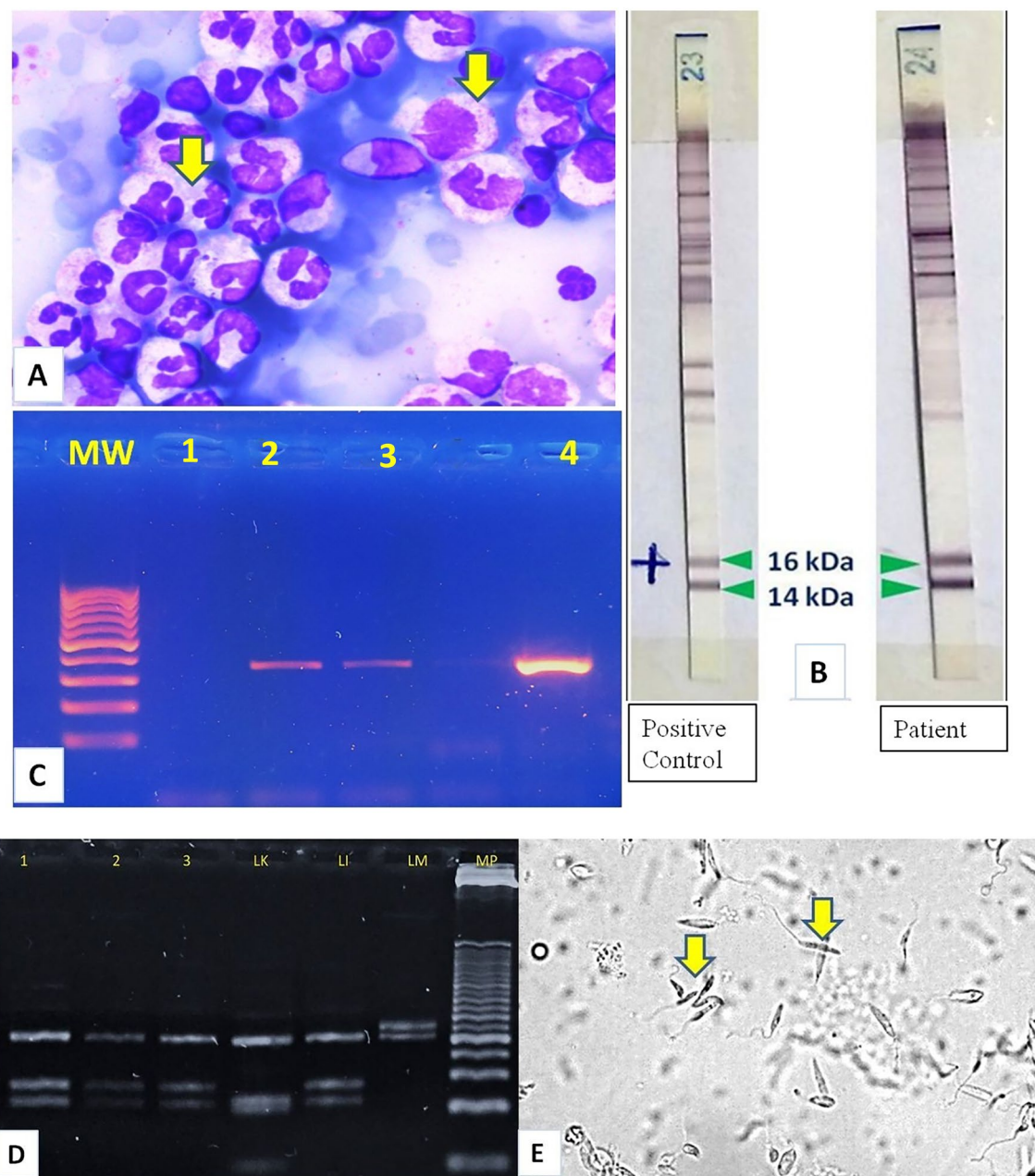
The treatment consisted of intravenous administration of LAB (Ambisome<sup>®</sup>, 40 mg/kg/weight) for 6 weeks. Favorable clinical and biological outcomes were recorded. The swelling of the lips and cheeks has resolved after 6 months with a follow-up period of five years (Fig. 5A and B). The blood PCR test for Leishmaniasis was negative. In June 2020, the PCR test turned positive and the serology test was negative. The patient was asymptomatic. In October 2023, a new ART regimen including Dolutegravir<sup>®</sup> was initiated with Tenofovir<sup>®</sup>, Lamivudine<sup>®</sup>. Currently, the patient is still monitored; he remains asymptomatic with a positive blood PCR test.

## Review and discussion

The review was independently conducted by two authors, LB and I.K. Only adult PLHA presenting concomitant mucosal oral and visceral leishmaniasis were included. HIV positive and negative adults and children with isolated VL, or isolated oral, nasal, laryngeal and pharyngeal locations were excluded from the review. No restrictions regarding the year or the language were considered. The literature review was conducted using MEDLINE via its interface PubMed and Scopus databases with the following MeSh/Key words “HIV”, “AIDS”, “Visceral Leishmaniasis”, “Atypical” “Oral Leishmaniasis”, “Lip Leishmaniasis”, “Ulcer”, “gingival Leishmaniasis” and their combinations using the Boolean equations “AND” “OR”. The PRISMA flow chart shows the selection process of studies by titles, abstracts and full texts. Other references were searched from the reference lists of full text-publications (Fig. 6). References obtained from the database search were managed using Mendeley Desktop 1.19.5. Data from the eligible 8 full texts, retrieved from 1994 to 2023, were extracted using a Microsoft Excel customized data sheet. Table 1 provides information on patients' characteristics, medical history, oral lesions, biological, clinical features, and treatment outcomes.

## Demographic data and medical history

The age of the patients ranged from 28 to 65 years, 6 patients were males and 2 were females [17, 26]. Among them, one was a MSM [16] and 1 man was bisexual [24]. Most patients were from Europe (France, Belgium, Italy, Spain,  $n=6$ ), while the other two were from India [26] and Africa (Ethiopia) [17], respectively. The present patient is a heterosexual adult man from an endemic area in the Sahel of Tunisia (Moknine). Medical history was available for 7 patients and not mentioned for one patient [16]. Six of the individuals were known to

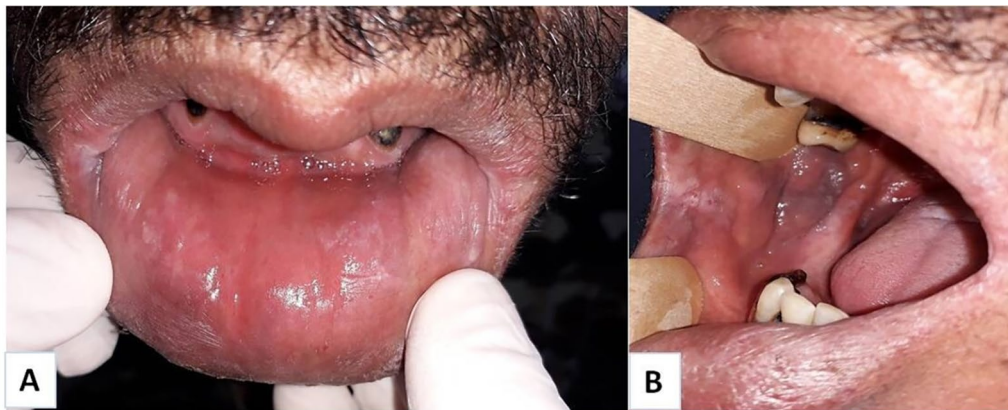


**Fig. 4** A–E Identification of *Leishmania*. **A** MGG from peripheral blood leucocytoconcentration shows intracellular *Leishmania* amastigotes. **B** Immunoprinting using *Leishmania* Western Blot IgG LDBIO Diagnostics® yielded positive results and showed the presence of two *Leishmania* protein bands: p14 and p16 (band 23: positive control and band 24: patients' results). **C** Agarose gel electrophoresis of 18S rRNA PCR product. Lane 1: negative control; Lane 2 patient blood, Lane 3: patient lip biopsy, Lane 4: positive control 343 bp; Lane MW: Molecular Weight marker (100 bp DNA ladder). **D** Restriction pattern of the amplified ribosomal Internal Transcribed Spacer 1 using HaeIII. Reference strains used were: *Leishmania infantum* MHOM/DZ/82/LIPA59 (LI) (three fragments of 187 bp, 72 bp and 55 bp), *Leishmania killicki* MHOM/TN/LEM163 (LK) (three fragments of 188 bp, 57 bp and 26 bp) and *L. major* MHOM/MA/81/LEM265 (LM) (two fragments of 206 bp and 132 bp). Lanes 1, 2, and 3: *L. infantum* identified from blood, lip biopsy and lip scraping product. MP: Molecular Weight marker (25-bp DNA ladder). **E** Culture using biphasic medium (blood horse based) from the peripheral blood leukocyte layer shows the promastigote form of *Leishmania*

be HIV positive, 3 patients were co-infected with HCV [23, 27, 28], and 2 patients were intravenous drug users (IVDU) [23, 28]. Three patients each received treatments

for laryngeal leishmaniasis [23], pulmonary leishmaniasis [24], and suspected abdominal tuberculosis [26]. One patient received prophylaxis against *pneumocystis*





**Fig. 5** A, B Clinical views after treatment showing resolution of the swelling of the lips and cheeks

*jirovecii* [27]. Oral lesions, such as candidiasis, and leishmaniasis were observed in 5 patients and were accompanied by symptoms such as fever, diarrhea, and vomiting [17, 24–26]. In one case, a patient with multiple gingival ulcers did not show any improvement after being treated with fluconazole and acyclovir [25]. The patient under study has been diagnosed with HIV since 1994 and was also co-infected with HBV. He has received treatment for iron-deficiency anemia, hypothyroidism and pulmonary tuberculosis. The patient experienced episodes of swelling in the right mandibular cheek. The diagnosis of cellulitis of dental origin was based on poor oral hygiene and the presence of multiple infected dental foci. Antibiotics were prescribed to treat the "cellulitis."

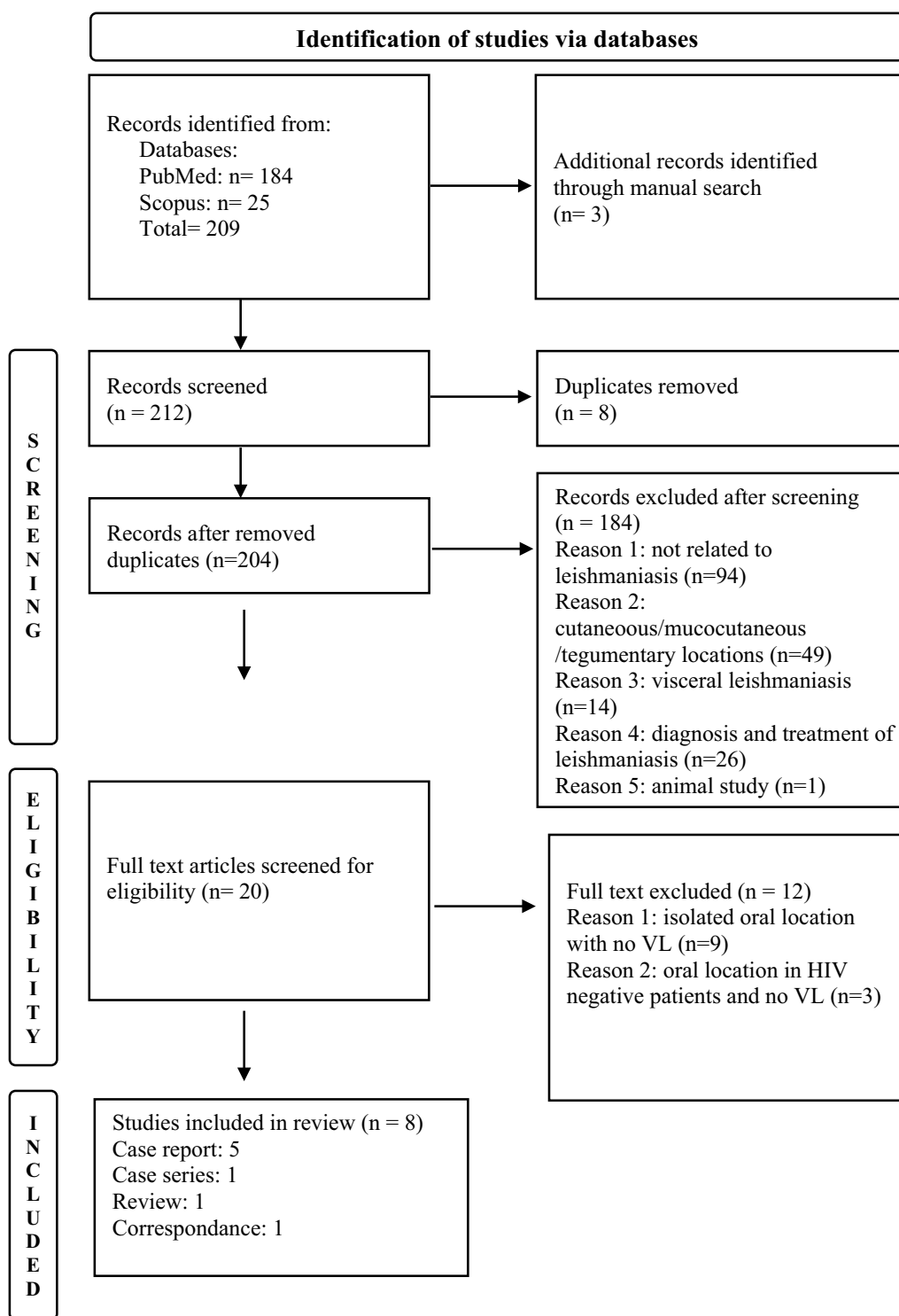
#### Oral lesions and systemic involvement

Regarding oral lesions' features, the palate [16, 17, 27] and the tongue [23, 26] were primarily affected, followed by the gingiva [25] and the lower lip [28]. In one case, the location was not specified (oral mucosa) [24]. All lesions were ulcerated and painful. Two cases reported bleeding on touch [17, 26]. The lesions were described as polypoid [16], nodular and exophytic [23], vegetative [27], and fungating [17]. Dysphagia and hoarseness were also reported [17, 23]. One patient had bilateral enlargement of submental and submandibular lymph nodes [26]. Fever was present in 7 cases, accompanied by weight loss [23, 25, 26, 28], hepatomegaly [17, 23, 26], or hepatosplenomegaly [24, 27] were also observed. Other mucosal involvements were noted in 2 cases, including the esophageal, gastric, and duodenal mucosa in one case [16] and the lung in another case) [24]. In the present case, the patient had mild splenomegaly with no associated fever. He presented with macrocheilitis of both upper and lower lips. The lesion was nodular and extended to the mucosa of the cheeks. According to Rosenthal et al. [29], the clinical

triad of fever, splenomegaly, and hepatomegaly was less frequently seen in patients with low CD4 counts. Considering the diverse clinical manifestations of oral leishmaniasis, it is important to rule out the following differential diagnoses for both granulomatous (such as tuberculosis, actinomycosis, sarcoidosis, deep fungal infections, syphilis, Melkersson-Rosenthal syndrome, and lymphogranuloma venereum) and non-granulomatous diseases (including allergy, chronic cellulitis, herpes infections, lymphoma, carcinoma, and Kaposi sarcoma) [30, 31].

#### Biological investigations

All patients were positive for HIV, with CD4 counts reported for 7 patients: <100 cells/mm<sup>3</sup> for 4 patients (ranging from 10 to 71/mm<sup>3</sup>) [16, 17, 24, 26], >100/mm<sup>3</sup> for 2 patients [23, 28] and >200/mm<sup>3</sup> for one patient (334/mm<sup>3</sup>) with a mentioned viral load of 54,800 copies/ml [25]. Anemia was recorded in all cases. Pancytopenia [16, 17, 24], thrombocytopenia [27], and hyperglobulinemia were also noted [24, 25]. Oral biopsy, performed in all patients was positive in 7 cases and negative in one case [28]. In addition, oral Kaposi sarcoma was diagnosed in a patient with CD4 T cell counts of 140/mm<sup>3</sup> and a CD4/CD8 ratio of 0.338 [23]. Bone marrow aspiration was positive in 4 cases [16, 17, 24, 28], negative in 2 cases [23, 25], not mentioned in one case [27], and not done in another case [26]. Serology testing was positive in 4 cases [23–26], negative in one case [28], not performed in one case [16], and not mentioned in 2 cases [17, 27]. Regarding the identification of the parasitic species, *L. infantum* zimodeme MON1 was characterized in one case report using the NNN medium (Novy-MacNeil-Nicole) [24]. The identification was not done in 3 cases [16, 17, 28] and not mentioned in 4 cases [23, 25–27]. For the present patient, the diagnosis of leishmaniasis was initially based on histopathology findings from the lip biopsy.

**Fig. 6** PRISMA flowchart

**Table 1** Summary of clinical, biological features and therapeutic outcome of oral leishmaniasis in HIV-VL co-infected patients

Authors	Age/ Gender/ Origin	History	Location and Type of oral lesions	Hepatomegaly/ Splenomegaly	Other	Fever/ Weight loss	CD4/mm <sup>3</sup> /Viral Load cp/ mm <sup>3</sup>	Blood count	Oral biopsy/ Other	BMB/ Serology/ Other	Species	Treatment	Outcome
Michiels JF et al.1994 [16]	28, M/MSM France	NM	Tonsil/Pal- ate Ulcer/poly- poid	No No	Yes	Yes	50/ NM	Anemia Pancytopenia	+	+/No	ND	LAB/ ATB	D
Pineiro TV et al. 1998 [23]	34, M/het- erosexual Spain	IVDU, HCV, Genital herpes LLei/Gluc	Tongue Nodule Ulcer	Yes No	No	Yes Yes	140/ NM	Leucopenia	+/ KS	Neg/+	NM	NM	D
Nigro L et al 2003 [24]	46, M, bisexual Italy	VL/AIDS Pulmonary Lei HAART Oral Lei	Oral mucosa Ulcer	Yes Yes	Lung	Yes, asthe- nia	10/ NM	Anemia Pancytopenia hyperGlobulinemia	+	+/+	<i>L. infantum</i> MON1	Gluc /60mg/kg)	Sympt
Montineri A. et al.2007 [25]	48 M Italy	Gingival ulcers/ Candidiasis	Gingiva Ulcer	No No	No	Yes	334	54.800/MA /hyperGlobulinemia	+	Neg/+	NM	LAB	F/6 m
Kumar et al 2007 [26]	35 F India	Ulcer Tongue/ TTT for TB	Tongue Ulcer Bleeding	Yes No	No	Yes	12/CD4 18/CD8	Anemia	+FNA Lymph node	ND/+	NM	NM	D
Rodrigo L.M. et al.2007 [27]	65, M Spain	AIDS/HCV	Palate/ Uvula Ulcer	Yes Yes	No	Yes No	NM	Anemia /Thrombocytopenia	+	NM	NM	Gluc	F
Presmanes M.M. et al 2008 [28]	31, M Spain	IVDU, HCV/AIDS HAART,	Lower lip: Swelling Ulcer	No No	No	No Yes	160 NM	Anemia	Neg	+/Neg	ND	LAB	F
Diro E et al 2015 [17]	29, F Ethiopia	Systemic symptoms Oral lesion	Palate/ Mass Bleeding	Yes No	No	Yes No	71 NM	Anemia Pancytopenia	+	+	ND	LAB ATB	D
Present case 2017	43, M, het- erosexual Moknine- Tunisia	HIV/HBV/ TB/Anemia/ Hiv/S Oral swell- ing/ ATB	Lips/ Cheeks/ Nodules	No Yes	No	No Yes	81 Unde- tected	Anemia Pancytopenia	+	No/+/ Culture+ PCR+/ PCR RFLP+	<i>L. infantum</i>	LAB	F 2020: PCR+ Asympt

M: Man; MSM: Men who have sex with men; F: Female; IVDU: Intravenous drug user; HCV: hepatitis C virus HIV: LLei: Laryngeal Leishmaniasis; Gluc: Glucantime; VL: Visceral leishmaniasis; AIDS: Acquired immune deficiency syndrome; HAART: Highly Active Antiretroviral Treatment; TTT: Treatment; TB: Tuberculosis; BMB: Bone marrow biopsy; NM: Not mentioned; KS: Kaposi Sarcoma; +: Positive; Neg: Negative; FNA: Fine Needle Aspiration; ND: Not done; LAB: Liposomal amphotericin B; PCR RFLP: Polymerase chain reaction-Restriction fragment length polymorphism; D: Dead; Sympt: Symptomatic; F: Favorable; Asympt: Asymptomatic



Serology and leishmaniasis culture were positive. Indeed, the genus of *Leishmania* parasite and *L. infantum* species were identified using molecular methods (Lei PCR and RFLP-PCR) [31].

### Treatments and outcomes

Different treatment regimens were prescribed. LAB was used in 5 cases [16, 17, 25, 27, 28], and antimonials were used in one case [24]. The drug was not specified in 2 cases [23, 26]. In one case, authors used intermittent doses of LAB on days 1–5, 10, 17, 31, and 38 [25], according to WHO guidelines [18]. LAB was associated with antibiotics (Ceftriaxone and Metronidazole) to prevent sepsis from the oral cavity in 2 cases [16, 17]. *N*-methylglucamine antimoniate (60 mg/kg) was prescribed in one case [24], and antimonials were substituted by LAB in one case [27]. The outcome was favorable or satisfactory in 3 cases [25, 27, 28], unfavorable with complications leading to death in 3 cases [16, 17, 26], and not specified in 2 cases [23, 24]. In the present case, the patient was treated with LAB at a total dose of 40 mg/kg for 6 weeks [18]. The outcome was favorable with resolution of biological and clinical symptoms, including swelling of the lips and cheeks. However, in 2020, as the patient discontinued his ART, the blood PCR test for Leishmaniasis was positive and the serology test was negative. Indeed, clinical symptoms were absent. In fact, to better control the parasite and prevent the recurrence of VL, a new ART regimen incorporating dolutegravir has recently been reintroduced. Some authors propose including dolutegravir to help the body eliminate the parasite, and suggest improving adherence and counseling services. Additionally, they recommend prolonging the duration of leishmaniasis treatment; implementing molecular monitoring and enhancing follow-up [32, 33]. In 2023, the patient remains asymptomatic with a positive PCR, despite ART treatment including dolutegravir. As reported by Molina R et al., there is no consensus to define “asymptomatic case”; HIV/VL coinfecting patients are easily infectious for sand flies, even those receiving prophylactic treatment for leishmaniasis. They represent an important source of VL transmission and spread of the disease, making coinfection prevention and control challenging. Therefore, the authors recommend to screen for VL in HIV infected patients in endemic and non-endemic areas [34].

### Conclusions

To the best of our knowledge, this is the first case report in Tunisia of unusual presentation of VL and it could be a concomitant VL/ML infection in a patient LHA. However, the nodular presentation of the oral condition is uncommon, compared to reports in the literature. Indeed this is the first report that investigates Leishmaniasis

using both molecular and non-molecular methods. The scarcity of these atypical locations may explain a lack of clinical suspicion. Therefore, as the oral cavity could be the first site of VL presentation, dentists play a crucial role in conducting a proper clinical examination and ruling out major infectious and non-infectious diseases that can mimic the oral condition. Finally, a multidisciplinary approach is mandatory to better treat VL-HIV co-infected patients and ensure a favorable outcome.

### Abbreviations

PLHA	People living with HIV/AIDS
VL	Visceral Leishmaniasis
LAB	Liposomal Amphotericin B
HBV	Hepatitis B virus

### Supplementary Information

The online version contains supplementary material available at <https://doi.org/10.1186/s12981-024-00660-1>.

Supplementary Material 1.

### Acknowledgements

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### Author contributions

LB performed oral and radiographic examinations, ruled out the dental origin of the lesion, indicated the lip biopsy and took pre and postoperative photos; AZ and SBH made the histological diagnosis from the lip biopsy; SB and HB performed biological analysis for the identification of *Leishmania* genus and species; IK, WM, AT and MC are the treating physicians who contributed to the treatment and monitoring of the patient. LB and IK made the review and wrote the manuscript. All authors took part in drafting and revising the manuscript and approved the publication of the final version of the article.

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### Availability of data and materials

No datasets were generated or analysed during the current study.

### Declarations

#### Ethics approval and consent to participate

The present case report is part of Latifa Berrezouga research project on PLHIV that was approved by the committee of ethics of the faculty of pharmacy, University of Monastir, Tunisia, under the reference CER-SVS/ISBM 013/2020. Professor Mohamed Chakroun is the Head of the Infectious Diseases' Department and Director of the Project. Informed consent from the patient's tutor was obtained with respect to the Helsinki declaration.

#### Consent for publication

Informed consent for publication related to clinical management data that are compliant with the infectious diseases' department was obtained from the patient's tutor.

#### Competing interests

The authors declare no competing interests.

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