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Case report

A tricky nasal polyp

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Summary
Leishmaniasis is an infectious disease caused by a protozoon of the genus Leishmania. It can manifest as cutaneous, mucosal, localized lymphadenitis forms or it can result as multiorgan involvement. In this article we present the case of a 59-year-old male with a polypoid mass attached to the septum at the right nasal cavity and symptoms as nasal congestion, hyposmia and anterior rhinorrhea. Histological sections of the mass showed oval-shaped inclusions in the cytoplasm of histiocytes suspicious for Leishmania amastigotes. The diagnosis was confirmed by detection of Leishmania complex DNA with polymerase chain reaction (PCR).

Key words: mucosal leishmaniasis, leishmaniasis, leishmania, nasal polyp

Introduction

Leishmaniasis is an infection caused by protozoans of the genus Leishmania, which belongs to Kinetoplastida. In the cytoplasm of the parasite there is a mitochondrial organelle, the kinetoplast, which contains extranuclear DNA [1,2]. Leishmaniasis is caused by 20 different Leishmania species and over 90 sandfly species can transmit Leishmania parasites. Humans can be host reservoirs and transmission between humans may occur [1]. Leishmaniasis can manifest as cutaneous, mucosal, localized lymphadenitis forms or it may have a multiorgan involvement (called "kala azar") [2]. Conditions related to the parasite or to the host may play a crucial role in mucosal involvement: clinical manifestation of leishmaniasis depends on parasite factors, host resistance and immunological response [1]. Cutaneous and visceral leishmaniasis are endemic in Europe [2]. In 2018, 70 cases of cutaneous leishmaniasis and 72 cases of visceral leishmaniasis were reported in Italy by the World Health Organization.

A cutaneous lesion is typically the primary site of infection, but leishmania can involve the mucosa as well. This latter can represent the primary site of infection and the nose is usually the most affected site, but any site can be affected. Vessels in the anterior portion of the nasal septum (called Kiesselbach's zone) and lower temperatures in the nose, can promote the development of amastigotes.[1] Final diagnosis require demonstration of Leishmania parasites [2].

Here we report a case of mucosal leishmania presenting as a nasal polyp.

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Case report

A 59-year-old male presented to Head and Neck Surgery Service at San Raffaele Scientific Institute in March 2019 due to nasal congestion, hyposmia and anterior rhinorrhea. The patient lived in a small village near Trento, Trentino-Alto Adige, in the north of Italy. Clinical information was collected: the patient owned a dog and reported a tick bite two years earlier. Other skin lesions were neither clinically founded nor reported by the patient.

Endoscopic nasal examination revealed a polypoid mass attached to the septum at the right nasal cavity, without lymphadenitis. Due to the uncertain nature of the lesion, a computed tomography (CT) was carried out showing deflection of the nasal septum and right sinusopathy, particularly in the contest of maxillary and anterior ethmoidal sinuses. Surgery was recommended: septoplasty and FESS (Functional Endoscopic Sinus Surgery) were performed, and the specimen was sent to the Pathology Unit.

Macroscopically, the specimen presented as brown-greyish polyp of 2.5x1x1 cm. Hematoxylin and eosin (H&E) sections showed nasopharyngeal mucosa infiltrated by a huge number of mononuclear inflammatory cells as lymphocytes, plasma cells and histiocytes, with erosion and ulceration of the mucosa. At high power field, it was possible to appreciate oval-shaped inclusions in the cytoplasm of histiocytes suspicious for Leishmania amastigotes (Fig. 1A) and confirmed by special stains GIEMSA (Fig. 1B) and PAS (Periodic acid Schiff) and immunohistochemistry (IHC) for CD1a clone EP3622 (Ventana) (Fig. 1C). The diagnosis was validated by detection of Leishmania complex DNA with polymerase chain reaction (PCR). Leishmania antibodies were detected in the blood of the patient as well (34.7 U.A. – positive: > 11 U.A.).

After one year of follow-up the patient presented no local or systemic recurrence.

Discussion

In humans, leishmaniasis occurs mainly as cutaneous leishmaniasis (CL), visceral leishmaniasis (VL) and mucosal leishmaniasis (ML), with the latter representing an uncommon manifestation of the disease. It has been suggested that mucosal involvement originates from hematogenous, or lymphatic spread of amastigotes from skin lesions or by contact of skin lesions with the mucosa; ML can occur many years after cutaneous manifestations [1,2]. However, the pathogenesis of mucosal lesions is not completely understood, considering that about 7% of patients with CL develop

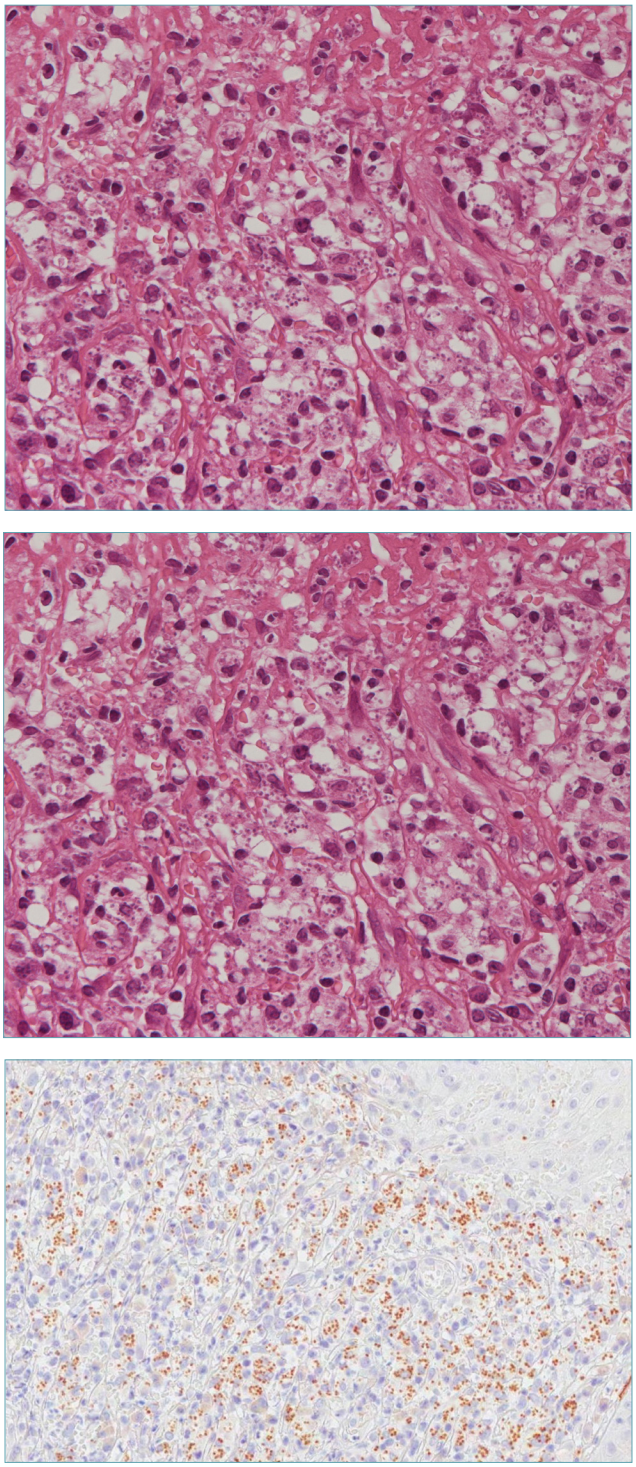

Figure 1. Polyp submucosa infiltrated by inflammatory cells, mainly lymphocytes and histocytes. In those latter, oval-shaped cytoplasmic inclusions can be appreciated (A, H&E 40X) which are positive for GIEMSA special stain (B, 20X) and CD1a immunohistochemistry (C, 20X) confirming the diagnosis of Leishmaniasis.

ML and some cases report mucosal lesions in the absence of cutaneous disease [1]. In our case, the patient did not refer any cutaneous lesion attributable to leishmania in his past medical history. We can speculate that our case corresponds to primary ML and the dog could have been the reservoir of the disease.

The first step for ML diagnosis is biopsy with histological evaluation. Microscopic sections usually show intense granulomatous inflammation made of lymphocytes, plasma cells, and histiocytes with tissue damage. Intracytoplasmic inclusions in the histiocytes seen in H&E staining and with GIEMSA may suggest the possibility of leishmania, but other organisms could present similarly. Moreover, in ML there are usually very few amastigotes that are not easily discernible in H&E sections, so diagnosis can be very challenging. Confirmation with other methods such as PCR, in vitro culture, inoculation into animals, skin test or serologic assay could be necessary [2].

Differential diagnosis includes malignant lesions (carcinoma or lymphoma), especially when associated with septum perforation, as well as granulomatous disease (sarcoidosis and Wegener's granulomatosis) and infectious causes (such as blastomycosis, paracoccidiodomycosis, leprosy, tuberculosis, syphilis, amebiasis) [1,2]. Clinically, the nasal mass can also be mistaken for rhinoscleroma, an infectious granulomatous process mainly involving the nasopharynx that is histologically characterized by large foamy mononuclear cells filled with gram-negative bacilli (called Mikulicz cells) surrounded by plasma cells, lymphocytes, and neutrophils. Final diagnosis is confirmed by isolation of *Klebsiella rhinoscleromatis* [3]. Inflammatory nasal polyps should also be considered in differential diagnosis. This type of polyp consists of respiratory epithelium (with areas of squamous or transitional epithelium) with mucous glands surrounded by hyaline or myxoid stroma; extracellular edema and inflammatory infiltration made of lymphocytes, plasma cells, mast cells, neutrophils and intense eosinophils are typically seen.

In our case, H&E sections revealed numerous oval-shaped inclusions in the cytoplasm of histiocytes, so we could first suggest leishmania as a causative agent. GIEMSA and PAS special stains and CD1a IHC (clone EP3622) all highlighted the inclusions as well. Amastigotes can be positive for CD1a IHC, with different specificity and sensibility depending on clones. MTB1 clone is characterized by high specificity and intermediate sensibility while clone O10 showed less accuracy in both sensitivity and specificity compared to the previous one. Clone EP3622 was occasionally investigated in CL, with good results in specificity and sensitivity [4]. In our case of mucosal leishmania, IHC for CD1a using EP3622 clone was positive. CD1a positivity was hypothesized to be related to the acquisition of the antigen by the amastigotes from the host dendritic cells or, in alternative, could be the result of homologous protein in amastigotes and Langerhans cells with antigens cross-reactivity. Variation in CD1a IHC sensitivity may depend on leishmania species, with higher sensitivity for the detection of Old World Leishmania comparing to New World ones [4,5].

IHC analysis using anti-Leishmania antibody was reported to be another tool for the identification of amastigotes with high sensitivity [4]. In our case we could not perform IHC anti-Leishmania because it is not available in our Department.

The presence of Leishmania was finally confirmed by detection of DNA by PCR and by the evidence of Leishmania antibodies in blood samples.

In conclusion, ML is a rare variant of leishmaniasis that is more frequent in the New World, and is usually secondary to a concurrent or previous cutaneous lesion. Here we presented a case of primary ML affecting a patient from the north of Italy. Even if uncommon, it is important to diagnose the disease early in order to remove and treat the lesion and to prevent local or systemic complications.

Conflict of interests statement

The authors declare no conflict of interest.

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Authors' contributions

FF and DF: manuscript writing, literature research and image acquisition. AV: manuscript writing and clinical data collection. SR: laboratory data collection and manuscript review. CD: manuscript review and supervision. FP: manuscript writing, review, and supervision.

References

1 Lessa MM, Lessa HA, Castro TWN et al. Mucosal leishmaniasis: epidemiological and clinical aspects. Braz J Otorhinolaryngol. 2007;73(6):843-847. https://doi.org/10.1016/S1808-8694(15)31181-2

2 Herwaldt BL, Leishmaniasis. Lancet. 1999;354(9185):1191-1199. https://doi.org/10.1016/S0140-6736(98)10178-2

3 Umphress B, Raparia K, Rhinoscleroma, Arch Pathol Lab Med 2018; 142:1533-1536. https://doi.org/10.5858/arpa.2018-0073-RA

4 Lopez-Trujillo M, Gonzàlez-Farreé M, Pujol RM et al. Diagnostic usefulness of immunohistochemical evaluation of CD1a antigen and polyclonal anti-Leishmania antibodies in cutaneous Leishmaniasis, Histol Histopathol 2021; 36: 567-576. https://doi.org/10.14670/HH-18-324

5 Sundharkrishnan L, North JP, Histopathologic features of cutaneous leishmaniasis and use of CD1a staining for amastigotes in Old World and New World leishmaniasis. 2017;(August):1005-1011. https://doi.org/10.1111/cup.13032