Case Report
Korean J Otorhinolaryngol-Head Neck Surg 2025;68(10):421-3 / eISSN 2092-6529
https://doi.org/10.3342/kjorl-hns.2025.00150

Auricular Cutaneous Leishmaniasis in South Korea: After Traveling to Mexico and the Galapagos Islands

Seunghyeok Lee[1], Sun Bean Kim[2], Gi Jung Im[1], and Euyhyun Park[1]

[1]*Department of Otorhinolaryngology-Head and Neck Surgery, Korea University College of Medicine, Seoul; and*
[2]*Division of Infectious Diseases, Department of Internal Medicine, Korea University College of Medicine, Seoul, Korea*

멕시코 및 갈라파고스 제도 여행 후 국내에서 진단된 이개 피부리슈마니아증 증례

이승혁[1] · 김선빈[2] · 임기정[1] · 박의현[1]

[1]고려대학교 의과대학 이비인후과학교실, [2]고려대학교 의과대학 내과학교실 감염내과

Received March 21, 2025
Revised April 15, 2025
Accepted April 17, 2025
Address for correspondence
Euyhyun Park, MD, PhD
Department of Otorhinolaryngology-Head and Neck Surgery,
Korea University
College of Medicine,
73 Goryeodae-ro, Seongbuk-gu,
Seoul 02841, Korea
Tel +82-2-920-6636
Fax +82-2-925-5233
E-mail peath@naver.com

Cutaneous leishmaniasis (CL) is a globally significant infectious disease. In Mexico, CL caused by *L. mexicana* affects the auricle and is commonly refered to as Chiclero's ulcer. In this particular case, a patient presented to our department with concerns of an ulcer and discharge on left external ear. Initially, our suspicions leaned towards perichondritis or cutaneous tuberculosis given the patient's age, sex and prevalence factors. However, after conducting a thorough history-taking and performing real-time polymerase chain reaction on a biopsy sample, we ultimately diagnosed it as CL. The wound improved dramatically after administration of meglumine antimoniate. This case underscores the challenges associated with diagnosing CL, particularly when it occurs in non-endemic regions. It also emphasizes considering overseas infections in patients with atypical diseases courses.

Korean J Otorhinolaryngol-Head Neck Surg 2025;68(10):421-3

Keywords Cutaneous leishmaniasis; Leishmania mexicana; Meglumine antimoniate; South Korea; Travel.

Introduction

Leishmaniasis is a protozoan disease of multiple clinical manifestations and caused by more than 20 *Leishmania* species.[1)] The auricle is rare site for cutaneous leishmaniasis (CL).[2)] In Mexico, CL affecting auricle is named as 'Chiclero's ulcer' after workers who gather latex gum. Clinicians may find it challenging to suspect this disease because its' clinical manifestations overlap with many other diseases. Here, we present a case of Chiclero's ulcer in South Korea initially suspected of various infectious disease but eventually successfully treated with meglumine antimoniate.

Case

On May 8th, 2023, a previously healthy 27-year-old female visited our office, complaining of a severe ulcerative lesion and pain on her left external ear that had persisted for one month. The upper half of the left pinna was edematous, with crust and yellowish discharge.

Due to our suspicion of acute perichondritis, she was hospitalized and received intravenous ceftazidime for 7 days. Inflammatory markers (white blood cell, erythrocyte sedimentation rate, C-reactive protein) in the initial lab results were not significantly elevated, but there was mild eosinophilia. The patient was applied mupirocin ointment targeting *Staphylococcus aureus*. After treatment and dressing, the wound showed slight improvement.

However, on the follow-up treatment date, her symptoms and the lesion were similar to the previous admission. A swab culture during admission revealed Methicillin-resistant *Staphylococcus capitis*, leading us to start oral TMP-SMX. We switched from mupirocin to fusidic cream based on swab culture results which indicated resistance to mupirocin ointment and susceptibility to fusidic-acid. As there was still no improvement, on May 28th, 2023, she was re-hospitalized and received intravenous vancomycin for 7 days. The wound did not improve even after receiving antibiotics covering gram-positive and negative organisms. Eventually, we suspected cutaneous tuberculosis.

Tuberculosis is endemic in South Korea, but there is limited data about the treatment of cutaneous tuberculosis. We performed a biopsy on the lesion, consulted the Infectious Diseases Department, and initiated the HERZ treatment regimen empirically (H-isoniazid, E-ethambutol, R-rifampin, Z-pyrazinamide).

However, even after taking anti-tuberculosis medication for one week, there was no change in the lesion. Real-time polymerase chain reaction (PCR) of *Mycobacterium tuberculosis* and non-tuberculosis mycobacteria on the biopsy revealed negative results.

Upon thorough history-taking, the patient reported visiting Mexico and the Galapagos Islands during the Christmas season. Although she did not remember being bitten by any insects, we suspected CL considering this travel history.

She was re-hospitalized and underwent debridement and biopsy under local anesthesia on June 15th. The sample was stained using Giemsa's method and immuno-histochemical staining using with CD1a antibodies. But pathology and swab reports done in our hospital showed no leishmania amastigote. We sent samples to the Korea Disease Control and Prevention Agency for real-time PCR of *Leishmania* spp. Sequencing targeting ITS1 was conducted, and *L. mexicana* was finally diagnosed.

The first-line treatment for Leishmaniasis is pentavalent antimoniate, and fortunately, meglumine antimoniate (Glucantime®, Sanofi Aventis) was available at the National Medical Center. From June 30th, she received an intramuscular injection of meglumine antimoniate at 20mg/kg for 20 days and underwent daily dressing. After 3 weeks of using meglumine antimoniate, a skin rash and pain developed along the left buttock which resolved upon discontinuation of the drug. Fig. 1 shows the clinical progression of the auricular lesion from initial presentation through treatment. The lesion showed a dramatic improvement one month after the end of anti-leishmanial treatment.

Ethics Committee Approval for the study was obtained from the Institutional Review Board of the Medical Center (IRB Number: 2020AN0488).

Discussion

Leishmaniasis is a vector-borne infection caused by the protozoan parasite of the genus *Leishmania*. The vectors are female sandflies (*Phlebotomus* and *Lutzomyia*). Depending on the *Leishmania* species, the disease can cause three main clinical manifestations: 1) localized CL characterized by cutaneous ulcers, 2) muco-cutaneous leishmaniasis involving mucosa and underlying connective tissue such as cartilage structures; and 3) visceral leishmaniasis (VL) affecting internal organs like liver spleen and bone marrow.[3)]

In Mexico and Central America, localized CL involving the ear is known as 'Chiclero's ulcer.' The term is derived from workers who gather chicle latex for gum. These workers lived in the forest for long durations, increasing their exposure to the sandfly vector.[4)]

In South Korea, there have been only 29 cases of leishmaniasis including this case. 5 cases were VL and except these

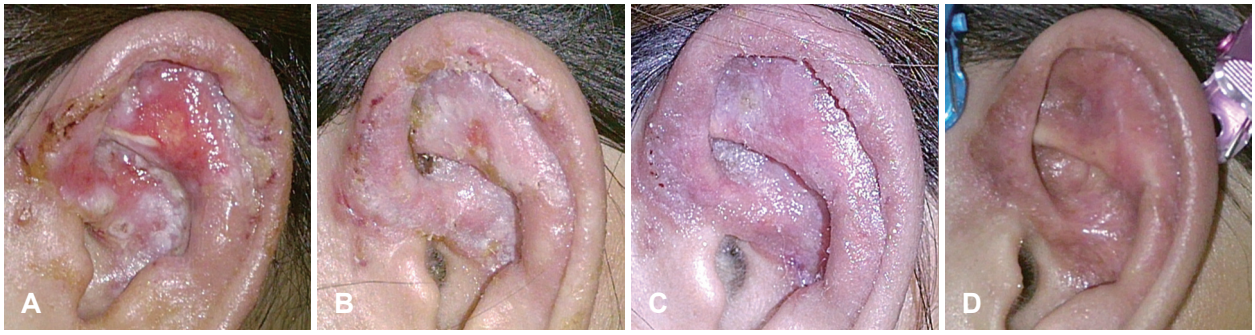

Fig. 1. Improvement of the auricular lesion after anti-leishmanial treatment. Before treatment (A), after 1 week (B), after 2 weeks (C), after 3 weeks (D) of intra-muscular injection of meglumine antimoniate.

cases are CL. But there is no evidence to indicate the occurrence of leishmaniasis vectors in the Republic of Korea, thereby limiting the risk of introduction and autochthonous transmission.[5)]

Chiclero's ulcer may manifest as an erythematous-edematous auricle or ulcer with discharge and rolled border. Differential diagnoses include perichondritis or carcinoma.[3)] In our case, we initially suspected perichondritis, which is more common according to age and sex.

The diagnosis of CL is the demonstration of the parasite by histology and culture or molecular analysis via PCR. Even though CL is self-limiting, irreversible disfiguring scar formation often occurs, so medical treatment is usually required.[3)] Parenteral pentavalent antimoniate and amphotericin B, oral azoles, and miltefosine have activity against CL.[6)] Although several antileishmanial drugs have been used during the past few decades, antimonials remain the first-line treatment for CL. Sodium stibogluconate and meglumine antimoniate are the two major formulations of antimonials in current use to CL.[7)] Meglumine antimoniate can cause local and general side effects. Local reactions included pain, burning sensation, edema, redness and skin reactions. The most important and serious side effect, which fortunately occurred the least frequently was anaphylactic shock.[8)] Amphotericin B is commonly used as an alternative drug with a broad spectrum of antiparasitic or antifungal activities with leishmaniasis resistance to antimonials.[7)] But amphotericin B has hepatotoxicity, so it should be used with caution. In South Korea, the National Medical Center stocks meglumine antimoniate.

Protection against sandfly bites, including the avoidance of outdoor activities from dusk to dawn and wearing appropriate clothing in endemic countries, is crucial for disease prevention.

This case highlights the importance for clinicians to consider overseas infectious diseases in patients refractory to conventional treatments. Although a diagnosis of leishmaniasis may not always be feasible in non-endemic regions, the risk can be mitigated by raising awareness of this emerging problem among physicians.[3)] Also, physicians should recognize that there is a Real-time PCR kit and meglumine antimoniate available in South Korea.

Acknowledgments

This research was supported by Basic Science Research Program through the National Research Foundation of Korea (NRF) funded by the Ministry of Education (NRF-2021R1I1A1A01052753), Ministry of Science and ICT (2022R1A4A1018869), and Korea University Research Fund (K2125741, K2211761). These funding sources provided only financial support and played no specific scientific role in this study.

Author Contribution

Conceptualization: Seunghyeok Lee, Euyhyun Park. Data curation: Seunghyeok Lee, Euyhyun Park. Funding acquisition: Euyhyun Park. Investigation: Euyhyun Park. Methodology: Seunghyeok Lee. Supervision: Sun Bean Kim, Gi Jung Im. Validation: Sun Bean Kim, Gi Jung Im. Writing—original draft: Seunghyeok Lee. Writing—review & editing: Euyhyun Park.

ORCIDs

Seunghyeok Lee	https://orcid.org/0009-0007-0451-6134
Sun Bean Kim	https://orcid.org/0000-0002-8830-1144
Gi Jung Im	https://orcid.org/0000-0002-9457-4253
Euyhyun Park	https://orcid.org/0000-0003-4373-6942

REFERENCES

1) Salman SM, Rubeiz NG, Kibbi AG. Cutaneous leishmaniasis: clinical features and diagnosis. Clin Dermatol 1999;17(3):291-6.
2) Robati RM, Qeisari M, Saeedi M, Karimi M. Auricular enlargement: an atypical presentation of old world cutaneous leishmaniasis. Indian J Dermatol 2011;56(4):428-9.
3) de Vries HJC, Schallig HD. Cutaneous leishmaniasis: a 2022 updated narrative review into diagnosis and management developments. Am J Clin Dermatol 2022;23(6):823-40.
4) Saito K, Murray HW. Chiclero's ulcer. Int J Infect Dis 2020;94:12-3.
5) Kim HJ, Kim EJ, Choi JW, Kim YC, Lee HI, Shin HI. A rare case of imported cutaneous leishmaniasis caused by Leishmania infantum in the Republic of Korea, 2021. Trop Med Infect Dis 2023;8(4):223.
6) Spinicci M, Zammarchi L, Gramiccia M, Di Muccio T, Bartolozzi D, Corsi P, et al. Effective meglumine antimoniate intralesional therapy for Chiclero's ulcer refractory to systemic liposomal amphotericin B. J Travel Med 2021;28(6):taaa169.
7) Madusanka RK, Silva H, Karunaweera ND. Treatment of cutaneous leishmaniasis and insights into species-specific responses: a narrative review. Infect Dis Ther 2022;11(2):695-711.
8) Esfandiarpour I, Farajzadeh S, Rahnama Z, Fathabadi EA, Heshmatkhah A. Adverse effects of intralesional meglumine antimoniate and its influence on clinical laboratory parameters in the treatment of cutaneous leishmaniasis. Int J Dermatol 2012; 51(10):1221-5.