NASCER E CRESCER
BIRTH AND GROWTH MEDICAL JOURNAL
year 2019, vol XXVIII, n.º 2

Nascer e Crescer - Birth and Growth Medical Journal
2019;28(2): 90-92. doi:10.25753/BirthGrowthMJ.v28.i2.14909

CASE REPORTS

CUTANEOUS LEISHMANIASIS - TWO CASES, TWO PERSPECTIVES

LEISHMANÍASE CUTÂNEA - DOIS CASOS, DUAS PERSPETIVAS

Inês Maio[I], Carla Teixeira[I], Catarina Prior[I], Susana Machado[II], Manuela Selores[II], Laura Marques[I]

ABSTRACT

Although endemic in some regions, as the Douro river valley, cutaneous leishmaniasis is a rare zoonosis in Portugal. Children are the most affected age group. Treatment remains controversial, given the lack of scientific evidence.

The goal of cutaneous leishmaniasis treatment is to avoid dissemination and progression to disfiguring lesions.

The option between no, systemic, or intralesional treatment should be carefully assessed.

Keywords: intralesional treatment; leishmaniasis; old-world leishmaniasis; parasitic infections

RESUMO

Embora endémica em algumas regiões, como a bacia hidrográfica do rio Douro, a leishmaníase cutânea é uma zoonose rara em Portugal. As crianças são o grupo etário mais afetado. O seu tratamento ainda é controverso, dada a falta de evidência científica. O objetivo do tratamento da leishmaníase cutânea é evitar a sua disseminação e evolução para lesões desfigurantes. A opção entre não tratar ou realizartratamento sistémico ou intralesional deve ser cuidadosamente avaliada.

Palavras-chave: tratamento intralesional; leishmaníase; leishmaníase do velho mundo; infeções parasitárias

INTRODUCTION

Leishmaniasis is a protozoan parasitic infection endemic in 98 countries that affects 0.7–1.3 million people per year. Sandflies transmit the infection to humans by inoculating *Leishmania* promastigotes into exposed skin, resulting in cutaneous, mucocutaneous, or visceral disease syndromes.[1-3] Clinical manifestations depend on the causative *Leishmania* species and underlying host immunity.[1,2,4,5]

Over 20 species cause human infection. Although individual species have unique transmission patterns, local reservoirs, and predominant clinical features, Leishmania are broadly subdivided into old-world and new-world species and, in general, the first cause a more indolent cutaneous disease.[1,3]

Cutaneous leishmaniasis (CL) is a rare zoonosis in Portugal.2Children are the most affected group. Its treatment remains controversial, given the lack of scientific evidence.6 Two cases in which different treatment options were adopted are herein presented.

I. Pediatrics Department, Centro Materno-Infantil do Norte, Centro Hospitalar Universitário do Porto. 4099-001 Porto, Portugal. inesmaiogoncalves@gmail.com; carlameitei@gmail.com; acprior@hotmail.com; laurahoramarques@gmail.com
II. Dermatology Department, Centro Hospitalar Universitário do Porto. 4099-001 Porto, Portugal. susanamlmachado@gmail.com; dermat@sapo.pt

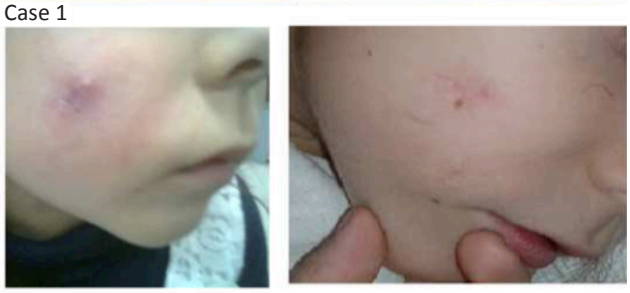
CASES DESCRIPTION

Case 1 – An 18-month-old toddler with no relevant past medical history developed erythematous, non-pruriginous, papulo-nodular lesions two weeks after returning from Morocco. Lesions initially presented in the face and subsequently in the back of the hands, trunk and abdomen (Figure 1).

Based on epidemiological history and lesion features, diagnosis of cutaneous leishmaniasis was established and subsequently histologically confirmed. The number of lesions, their potential for disfiguring scars, and the presence of disseminated papulo-nodular lesions in a young child lead us to opt for systemic treatment with intravenous amphotericin B. Treatment outcome was favorable, with complete lesion regression.

Case 2 – A 6-year-old child was observed at the Dermatology outpatient clinic because of a single, gradually increasing papulo-nodular face lesion present for the past eight months, appearing after holidays in the Douro valley region (Figure 1). Diagnosis of cutaneous leishmaniasis was suspected and histologically confirmed, but molecular identification was not performed, since the collected specimen was not suitable for analysis. Considering size and location of the single lesion, intralesional treatment with meglumine antimoniate was selected. Treatment outcome was favorable, with complete lesion regression within four weeks and no residual scar.

Case 1



Case 2 - Pre and post-treatment

Figure 1

FINAL COMMENTS

Treatment goal of cutaneous leishmaniasis is to avoid dissemination, late mucosal involvement, and progression to disfiguring lesions, all of which were achieved in both cases presented. Cases fulfilled criteria for complex CL and required treatment.[1,3,6-8]

A systematic review of new-world cutaneous leishmaniasis treatment efficacy in adults favored treatment with systemic or intralesional drugs over watchful waiting; the most recent literature is debating which treatment should be used.[1,9]

Criteria to choose between systemic versus intralesional therapy must be assessed on a case-by-case basis, considering lesion number and location and individual patient characteristics.3,6 Intralesional treatment is preferred in single lesions, when the risk of dissemination is very low.[1,6]

Evaluating the best CL treatment is difficult because several factors may influence drug efficacy, including lesion size, number, and appearance, and disease duration prior to treatment.[9] Treatment efficacy also depends on the causative *Leishmania* species, so an effective treatment in one geographical area for a given organism may not be effective in a different geographical area or for a different organism in the same location.[9]

Differences in immune response, drug clearance, and overall drug exposure in pediatric patients have contributed to a higher proportion of treatment failure in children compared with adults.[7] A large systematic review of cutaneous leishmaniasis treatment in pediatric patients demonstrated high efficacy and tolerability of topical antimonial agents after a median number of five sessions.[7] Although amphotericin was not included in this review, the authors discussed that it can be used as an off-label medicine in children under the age of two, as in the first case here reported.

At present, better and more comprehensive information regarding old-world leishmaniasis treatment and the effectiveness of these drugs in children is lacking, especially in very young children.[7]

Increasing availability of molecular diagnostic tools, including PCR-based assays, will enable a shift towards species-directed treatment. When fully available, it will allow individualized treatment choices for patients with cutaneous leishmaniasis.

REFERENCES

1. Cota GF, De Sousa MR, Fereguetti TO, Saleme PS, Alvarisa TK, Rabello A. The cure rate after placebo or no therapy in American cutaneous leishmaniasis: A systematic review and meta-analysis. PLoS One. 2016; 11:1–15.
2. World Health Organization. Surveillance of leishmaniasis in the WHO European Region, 2016 Leishmaniasis. WHO Wkly Epidemiol Rec. 2018; 40:521–30.
3. Aronson N, Herwaldt BL, Libman M, Pearson R, Lopez-Velez R, Weina P, *et al*. Diagnosis and treatment of leishmaniasis: Clinical practice guidelines by the infectious diseases society of America (IDSA) and the American Society of tropical medicine and hygiene (ASTMH). Am J Trop Med Hyg. 2017; 96:24–45.
4. Showler AJ, Boggild AK. Cutaneous Leishmaniasis in Travellers:

a Focus on Epidemiology and Treatment in 2015. Curr Infect Dis Rep. 2015; 17:489.
5. Oliveira LF, Schubach AO, Martins MM, Passos SL, Oliveira RV, Marzochi MC, *et al*. Systematic review of the adverse effects of cutaneous leishmaniasis treatment in the New World. Acta Trop. Elsevier B.V. 2011; 118:87–96.
6. Sundar S, Chakravarty J. An update on pharmacotherapy for leishmaniasis. Expert Opin Pharmacother. 2015; 16:237-52.
7. Uribe-Restrepo A, Cossio A, Desai MM, Dávalos D, Castro M del M. Interventions to treat cutaneous leishmaniasis in children: A systematic review. PLoS Negl Trop Dis. 2018; 12:e0006986.
8. Gradoni L, López-Vélez R, Mokni M. Manual on Case Management and Surveillance of the Leishmaniases in the WHO European Region. WHO Reg Off Eur. 2017.
9. Heras-Mosteiro J, Monge-Maillo B, Pinart M, Pereira PL, Garcia-Carrasco E, Cuadrado PC, *et al*. Interventions for Old World cutaneous leishmaniasis (Review). Cochrane Database Syst Rev. 2017. 12: CD005067.

CORRESPONDENCE TO

Inês Maio
Pediatrics Department
Centro Materno-Infantil do Norte
Centro Hospitalar Universitário do Porto
Largo Prof. Abel Salazar
4099-001 Porto
Email: inesmaiogoncalves@gmail.com

Received for publication: 10.08.2018
Accepted in revised form: 05.02.2019