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Case presentation

Two small yellowish papules in a 1 year-old boy: cutaneous leishmaniasis

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Abstract

Cutaneous leishmaniasis (CL) is zoonosis with a spectrum of cutaneous manifestations caused by protozoan parasites of the genus *Leishmania*.

Manifestation varies according to the parasite virulence and the host immune response. Pentavalent antimonials (sodium stibogluconate and meglumine antimoniate) have been used as a first-line therapy for the last 70 years around the world.

We report a case of a 1-year-old boy with two small yellowish papules mimicking juvenile xantogranuloma diagnosed with cutaneous leishmaniasis after a biopsy. Patient underwent treatment with 2 sessions of intraleisonal (IL) meglumine antimoniate (Glucantime®) with complete clearance of both lesions.

Conclusion: Cutaneous leishmaniasis treatment is difficult to standardize; treatment options in children include wound care and watchful waiting, intraleisonal pentavalent antimonials, topical paramomycin, or oral miltefosine.

Keywords: Leishmania, *L. infantum*, Cutaneous leishmaniasis, antimonial.

Introduction

Cutaneous leishmaniasis (CL) is zoonosis with a spectrum of cutaneous manifestations caused by protozoan parasites of the genus *Leishmania*.

Manifestation of leishmaniasis vary according to parasite virulence and host immune response. Most cases in Spain are caused by *L. infantum*, which is considered endemic on the peninsula [1]. *L. infantum* is also endemic in the middle east, China, and central asia and manifests with atypical clinical features more frequently than other species [2]. Pentavalent antimonials (sodium stibogluconate and meglumine antimoniate) have been used as a first-line therapy for the last 70 years around the world [3].

Case synopsis

A 1-year-old boy, presented with a 3-month history of 2 crusted papules on the lower eyelid and on the right cheek. These were asymptomatic, but showed progressive growth over the 2 months.

Physical examination showed a 7 mm red yellowish papule on the lower right eyelid and smaller similar lesion on the right cheek (Figure1).
Figure 1. Two small red-yellowish papules on the lower right eyelid and the right submandibular area.

A 4 mm punch biopsy was performed; histologic findings are illustrated in Figures 2 and 3.

Figure 2. Skin biopsy showing a heavy dermal inflammatory infiltrate composed of lymphocytes, macrophages and several “Touton-like” giant cells.  Figure 3. Numerous parasites are present in the cytoplasm of macrophages (H&E)

Histological examination showed epidermal hyperkeratosis with focal parakeratosis. In the dermis, there was a heavy inflammatory infiltrate composed of lymphocytes, plasma cells, and many histiocytes, with sparse multinucleated giant cells. Numerous parasites were noted inside the cytoplasm of the histiocytes, which were enhanced with Giemsa stain.

A diagnosis of cutaneous leishmaniasis was rendered.

The patient underwent treatment with 2 sessions of intralesional (IL) meglumine antimoniate (Glucantime®) with complete clearance of both lesions. Glucantime® was used undiluted; 1 ml (300 mg) was injected around the lesion and we repeated the injection at the same dose after one week.

Discussion
In children with cutaneous leishmaniasis the most common site affected is usually the face and the classic clinical picture is a small single papule or plaque that is usually asymptomatic; it may have surface erosion with crusting [4]. The presence of multiple lesions is extremely rare [2].

Because of the pink-yellowish color in our case the differential diagnosis included juvenile xantogranuloma.

The diagnosis is made by identifying the parasite in the lesion; although a punch biopsy is the gold standard, a thick drop scraping of the base of the papule has a high sensitivity and good microscopic concordance when compared with a biopsy [5].

Polymerase chain reaction (PCR) to confirm the diagnosis can be done using the cytology or biopsy specimen in cases in which the biopsy is non-specific.

It is difficult to standardized treatment for cutaneous leishmaniasis because of many factors such as poor study design and the difficulty to define a “successful treatment” [3,6]. Treatment options include intralosomal or intramuscular pentavalent antimonials. The major problem with the injection of antimonials in children is the pain, which is greater when administered intralosionally [3]. Other options that are being used safely in children are summarized in Table 1. It is important to remark that in immunocompetent patients with less than 3 lesions, which are small in size < 30 mm and non-disfiguring, wound care and watchful waiting are safe options because of several studies reporting spontaneous healing after 8-12 weeks [7-9].

### Table 1.
Current treatment options available for cutaneous leishmaniasis in children. Adapted from references 3,6-9.

<table>
<thead>
<tr>
<th>Drug</th>
<th>Class</th>
<th>Mechanism of action</th>
<th>Route of administration</th>
<th>Dose/ Frequency</th>
<th>Duration</th>
<th>Side effects</th>
<th>Comments</th>
</tr>
</thead>
<tbody>
<tr>
<td>Topical PR_MACL*</td>
<td>Aminoglycoside</td>
<td>Protein synthesis inhibitor, binds to 16S RNA</td>
<td>Topical</td>
<td>0.5D</td>
<td>10-20 days</td>
<td>Local inflammation</td>
<td>The association with Methylbenzenonium chloride has higher cure rates than with 10% area, for unknown mechanism.</td>
</tr>
<tr>
<td>Local Heat</td>
<td>Physical therapy</td>
<td>Cytokine mediated inflammation</td>
<td>Topical</td>
<td>50°C for 30 seconds</td>
<td>Single session</td>
<td>Local inflammation, Local pain</td>
<td>Requires special device (e., Thermomod Device)</td>
</tr>
<tr>
<td>Photodynamic therapy with antimonials</td>
<td>Photonsensitizer</td>
<td>Light-induced reaction, cell damaged</td>
<td>Topical</td>
<td>Single application/weekly</td>
<td>0-10 weeks</td>
<td>Local inflammation, Local pain</td>
<td>Apply a thick layer of photosensitizer for 30 min with fall, and then expose the area to sunlight for 2.5 h.</td>
</tr>
<tr>
<td>IL-AM**</td>
<td>Pentavalent antimony</td>
<td>Not yet completely elucidated</td>
<td>IL</td>
<td>300 mg/ml/weekly</td>
<td>Varies according to response. Up to 24 weeks</td>
<td>Local inflammation, Local pain</td>
<td>OWCL healing takes up to one month. Large ulcers may take longer. Can be combine with cryotherapy for better cure rates.</td>
</tr>
<tr>
<td>IL-Amb</td>
<td>Polyene</td>
<td>Interferes with ergosterol synthesis</td>
<td>IL</td>
<td>2 mg/ml/weekly</td>
<td>12 weeks</td>
<td>Phlebitis, Pain and fibrosis at the injection site.</td>
<td>Second-line therapy for resistant organisms.</td>
</tr>
<tr>
<td>Pentoxifylline</td>
<td>Xantine derivative</td>
<td>Not yet completely elucidated</td>
<td>Oral</td>
<td>400 mg/ 11s</td>
<td>10-20 days</td>
<td>Gastrointestinal, dizziness, Aseptic meningitis</td>
<td>Adjuvant therapy concomitant with IMAM</td>
</tr>
<tr>
<td>IM-AM**</td>
<td>Pentavalent antimony</td>
<td>Not yet completely elucidated</td>
<td>IM</td>
<td>20 mg/kg</td>
<td>10-20 days</td>
<td>Cardiac, hepatic, hematologic and pancreatic toxicities</td>
<td>NWCL, or several lesions or large ulcers in OWCL. Requires weekly labs and ECG.</td>
</tr>
<tr>
<td>S-Amb</td>
<td>Polyene</td>
<td>Interferes with ergosterol synthesis</td>
<td>IV</td>
<td>3 mg/kg/d</td>
<td>Days 1-5 and 10.</td>
<td>Renal toxicity, hypoplasemia</td>
<td>Requires weekly creatinine and potassium levels</td>
</tr>
<tr>
<td>Flucinazol</td>
<td>Triazol</td>
<td>Interferes with ergosterol synthesis</td>
<td>Oral</td>
<td>200 mg/BID</td>
<td>6 weeks</td>
<td>Renal and hepatic toxicity, Prolong QT (tendances de points)</td>
<td>Requires weekly CBC and hepatic enzymes.</td>
</tr>
<tr>
<td>Miltefosine</td>
<td>Alkylphosphonic acid</td>
<td>Protein kinase B (Akt) inhibitor</td>
<td>Oral</td>
<td>2.5-3 mg/mg/TID</td>
<td>28 days</td>
<td>Gastrointestinal, renal failure, hepatotoxicity</td>
<td>Patients not responding to ILAM. Requires weekly creatinine and hepatic enzymes.</td>
</tr>
</tbody>
</table>


**Conclusion**
Cutaneous leishmaniasis treatment is difficult to standardize. Treatment options in children include wound care and watchful waiting, intralosomal pentavalent antimonials, topical paramomycin, or oral miltefosine.

**References**


