Case report

A child with nasal leishmaniasis cured with local therapy (cryotherapy plus intralesional antimony)

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Summary

Intralesional antimony is increasingly used to treat cutaneous leishmaniasis but the experience is limited on cartilaginous regions, especially in young children. We report a positive outcome in a 5-year-old child treated with cryotherapy plus intralesional antimony for a lesion of the nose.

KEY WORDS: leishmaniasis; antimony; cryotherapy; cartilage; pediatric.

Cutaneous leishmaniasis (CL) is a skin disease due to a protozoan called Leishmania sp., transmitted by sandflies. In Europe, autochthonous infection is reported in the Mediterranean area. Following recent WHO recommendations (1) and recent positive experience in travelers (2), pentavalent antimony has been increasingly used in CL with few lesions, generally in the form of intralesional meglumine antimoniate (Glucantime) with or without cryotherapy. Children can also benefit from this treatment approach although the practical constraints are different than in adults. Only very few experiences have been reported on the use of intralesional antimony in lesions on cartilaginous zones in children (Figure 1).

A 5-year-old boy was referred to the hospital for a non-healing ulceration on the tip of the nose that had appeared after a trauma 8 months before. The lesion had grown progressively and was covered by a necrotic scab, with an inflammatory border, in spite of the antiseptic treatment and antibiotics. Beneath the scab, the lesion was insensitive and the bleeding ulceration extended irregularly to the nostrils. Equally insensitive infracentimetric lymph nodes were found in the submandibular region. Considering the proximity to the alar cartilage and the depth of the ulceration, a MRI confirmed the integrity of the subcutaneous tissue (Figure 2). A biopsy was performed and revealed the presence of an epitheloid granuloma suggesting an infectious cause. The diagnosis of leishmaniasis was confirmed by the presence of typical amastigotes in the Giemsa-stained smear and typical promastigotes in culture.

A history of a trip to the suburbs of Tunis in Tunisia during the previous summer oriented towards infection with either Leishmania major or L. infantum. Systemic treatment with oral fluconazole (3 then 5 mg/kg/days for 6 weeks) was inefficient. A perfusion of liposomal amphothericin B (AmBisome, 5 mg/kg by slow intravenous infusion) led to an allergic reaction with dyspnea, malaise and cutaneous rash and was thus suspended. A local treatment was thus envisioned. Two intralesional injections of meglumine antimoniate, each following a short superficial cryotherapy, were performed 1 month apart under general anesthesia (Figure 3). A third injection was performed under local anesthesia 2 weeks after the 2nd. The lesion disappeared in 3 weeks and did not relapse after 9 months of follow-up (Figure 4).

Therapeutic decisions for this young child was difficult because of the inefficacy of the first line treatment [fluconazole (3, 4)] and the intolerance of the second line (2). Local treatment was then considered safer and possibly more effective than systemic options such as antimony or miltefosine. However, the depth of the lesion in a region so close to cartilages imposed a radiological analysis to assess the integrity of these structures. The tip of the nose being a sensitive zone and the border of the lesion being thickened, the first injection was performed under local anesthesia 2 weeks after the 2nd. The lesion disappeared in 3 weeks and did not relapse after 9 months of follow-up (Figure 4).

Some authors suggest using 1 ml of xylocaine at 1% with each injection, in order to reduce the pain (5). With this protocol, little pain was felt by the 13 patients of the study.
Although facial leishmaniasis lesions are quite frequent, the lesions are more frequently located on the cheek or the front. We found no published report of cutaneous leishmaniasis located on the tip of the nose treated intralesionally: such data is not clearly reported in the different articles (Table 1). Little data exists on the local toxicity on a growing juvenile cartilage. Pau reported 2 cases of endonasal leishmaniasis in adults (6), treated by intralesional meglumine; the treatment was well tolerated and did not lead to cartilage necrosis. Similarly, injection in the outer ear of 2 other adult patients also led to favorable outcome without local complications (7). In the present case, the absence of cartilaginous complication in a child encourages wider use of this treatment even in delicate cartilaginous zones.

Several authors have reported the efficacy of local treatment in the management of cutaneous and mucosal leishmaniasis (8). Intralesional meglumine antimoniate presents numerous advantages: fewer toxic systemic side-effects due to lower dosing, high in site concentration of the active drug, rapid response and less scaring. Qasmi (5) reports a positive response in all cases with a scarless healing in 61% of the cases, a hypopigmented scar in 15% and an atrophic scar in 15%. However, no straightforward protocol exists: intralesional injections (2-5 ml/injection) are usually done at the 4 cardinal points, once a week, over 3 to 5 weeks. Some authors adjoin intramuscular injection, the number of which and the delay with the intralesional injections being highly variable; this protocol is proposed in case of periorificial lesions. A recent study reported the successful management of 9 lid cutaneous leishmaniasis lesions with intralesional injection (9). Some authors also adjoin cryotherapy during 10-25 s, which seems to increase the percentage of

**Figure 1** - Initial aspect of the lesion: a necrotic scab covers the ulceration of the tip of the nose.

**Figure 2** - On the left, sagittal T1 MRI image showing the thickening of the soft tissue of the dorsum and the tip of the nose and the disappearance of the subcutaneous fat layer signal. On the right, axial T2 Flair sequence showing the ulceration of the tip of the nose and the subcutaneous infiltration.
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Figure 3 - Aspect of the lesion after retrieval of the scab at the time of injection.

Figure 4 - Healing aspect 6 months after first injection: a slightly heterochromous scar can be seen at the tip of the nose.

Table 1 - Publications of cutaneous leishmaniasis in children treated by Glucantime: little data is given on the number of children presenting with cartilaginous lesions of the nose or the ear and the administration of local treatment in these particular cases.

<table>
<thead>
<tr>
<th>Author</th>
<th>Number of children included in the study</th>
<th>Number of lesions of the ear or nose</th>
<th>Particularities</th>
</tr>
</thead>
<tbody>
<tr>
<td>Vargas-Gonzalez (1999) (12)</td>
<td>24 children &lt;15 years (131 patients)</td>
<td>No precise data</td>
<td>Systemic IM treatment: 1 ampule/ day</td>
</tr>
<tr>
<td></td>
<td></td>
<td>ear: 28% of the cohort face: 24% of the cohort</td>
<td>No local treatment</td>
</tr>
<tr>
<td>Mujtaba (1999) (13)</td>
<td>59 patients &lt; 20 years (total of 96 patients)</td>
<td>64 lesions in the population (no precise data on ear/nose in children)</td>
<td>IL Glucantime weekly vs fortnightly</td>
</tr>
<tr>
<td>Salmanpour (2001) (14)</td>
<td>96 patients (3-6 years)</td>
<td>1 child with nasal lesion</td>
<td>No local treatment</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>Oral ketoconazole</td>
</tr>
<tr>
<td>Kharfi (2004) (15)</td>
<td>106 children &lt; 16 years</td>
<td>7 nose lesions</td>
<td>Systemic IM meglumine antimoniate when cartilage involved</td>
</tr>
<tr>
<td>Layegh (2009) (16)</td>
<td>39 children &lt; 13 years</td>
<td>31 lesions of the face and neck (no precise data on ear/nose)</td>
<td>IL Glucantime</td>
</tr>
</tbody>
</table>
A complete cure at 6 weeks (10). Asilian also demonstrated that CO₂ laser could also be used as a first-line therapy for cutaneous leishmaniasis (11); however, this therapeutic was not indicated in the present case because of the high risk of necrosis of the underlying cartilage.

In conclusion, intralesional antimony can be used in children, even in lesions of cartilaginous areas, a safe and effective salvage option when more conventional approaches cannot be used.

References