Oral and maxillofacial myiasis: a case series and literature review

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Objective. The aim of this study was to describe a series of 10 cases of oral-maxillofacial myiasis, discussing its main features, demographic distribution, and treatment aspects.

Study design. A retrospective study was carried out involving male and female patients of any age with oral-maxillofacial myiasis. The sample was determined by spontaneous demand at the emergency ward of a hospital between January 2005 and January 2011 (6 years). After treatment of each case, data were gathered on the presence of associated systemic disorders, time elapsed since onset of the disease, and treatment established. A review of the literature on this topic was also carried out.

Results. The sample was made up of 10 patients, all treated with surgical debridement whether or not associated with the use of oral ivermectin. Mean time elapsed since the onset of the disease ranged from 4 to 36 months. The middle third of the face was the most frequently affected region (7 cases). Oral-maxillofacial myiasis predominantly affected the male gender (70%).

Conclusions. Oral-maxillofacial myiasis affects individuals with poor hygiene and neurologic and/or psychologic alterations. It occurs predominantly in countries near the tropics. The treatment of choice is surgical debridement.


Myiasis is the invasion of human or animal tissue by fly larvae that evolve into parasites.¹ This is a worldwide phenomenon related to latitude and the life cycle of various species of flies. Myiasis occurs predominantly in the tropics and subtropics in countries with inadequate hygiene, poor housing conditions (proximity to domesticated animals), and warm weather.²

Myiasis is classified as either primary or secondary. Primary myiasis is caused by larvae that feed on living tissue (biophagous). This form of myiasis is common in cattle and rare in humans. It is produced by Cochliomyia hominivorax ("varejeira" fly), which lays 20-400 eggs on exposed wounds, with larvae hatching within 24 hours.³ Secondary myiasis is caused by flies that feed on dead tissue (necrobiophagous). This is the more common type and attacks patients with lesions that have necrotic cavities.¹,⁴

Infestation is most often subcutaneous, producing furunculoid or boil-like lesions, and it can also occur in certain body cavities and wounds.⁵ The larvae obtain their nutrition from the surrounding tissue and burrow deep tunnels into soft tissue, separating the gums and mucoperiosteum from the bone.⁶

Oral-maxillofacial myiasis has been described in the literature since 1909 and is considered to be a rare occurrence.⁷ The cases described are mostly due to medical and anatomic conditions, such as neglected mandibular fracture,⁸ lip incompetence, cerebral palsy, mouth breathing, anterior open bite,⁹ patients undergoing mechanical ventilation,¹⁰ cancrum oris,⁸ and tooth extraction.¹¹

The aim of the present study was to describe a series of 10 cases of oral-maxillofacial myiasis, discussing its main features, demographic distribution, and treatment aspects and comparing the findings with earlier studies in the literature.
MATERIALS AND METHODS

A retrospective study was carried out involving male and female patients of any age with oral-maxillofacial myiasis. The sample was determined by spontaneous demand at the emergency ward of a hospital between January 2005 and January 2011 (6 years). After treatment of each case, data were gathered on gender, age, site, species of larva, the presence of associated systemic disorder, time elapsed since onset of the disease, and treatment established. In all cases, the larvae were sent for parasitologic analysis for the confirmation of the species.

All of the patients underwent operations soon after admission to the emergency ward. Under general anesthesia, the mechanical removal of the larvae was performed, along with debridement of the necrotic tissue and an attempt at the maximal possible primary closure of the wound, as exemplified in case 1 (Figs. 1-4). The patients were also treated with a systemic antibiotic (cephalotin, 1 g every 6 h), antiinflammatory, and analgesic. No specific medication protocol for ivermectin was instituted, and the decision to use this drug fell to the health care professional conducting each case.

Literature review

All papers published in the literature between 1963 and 2010 on oral-maxillofacial myiasis in all languages were surveyed to determine the mapping of cases per
country of occurrence of the disease. The Medline (Pubmed) database was used for this search, using the following descriptors were used: oral myiasis, nasal myiasis, ophtalmomyiasis, and maxillofacial myiasis.

Case reports that did not address human myiasis, those that appeared to be duplicated in the searches with different descriptors, and those carried out in countries that no longer exist were excluded. After the definition of the sample, the cases were counted and separated by country. Among a total of 570 papers found in the search, 490 were excluded owing to duplication in the searches using different descriptors and for reporting cases of myiasis in sites other than the oral-maxillofacial region. Two were excluded for the fact that they were from countries that no longer exist (Czechoslovakia) and one was excluded because the origin of the paper could not be identified. Papers that reported cases in animals were also excluded. Thus, a total of 493 papers were excluded and 77 papers (86 cases) fulfilled the inclusion criteria, as distributed on the map in Fig. 5.

RESULTS

The sample was made up of 10 patients, all of whom were treated with surgical debridement whether or not associated with the use of oral ivermectin. Mean time elapsed since the onset of the disease ranged from 4 to 36 months. The middle third of the face was the most frequently affected region (7 cases). Oral-maxillofacial myiasis predominantly affected the male gender (70%). These data are presented in Table I.

DISCUSSION

Oral-maxillofacial myiasis is a rare condition caused by flies of the order Diptera, which lay eggs or larvae in food, necrotic tissue, wounds, and intact mucosa. Classification is based on location in the host body [dermal, subdermal, nasopharyngeal, internal (organs), or urogenital]; in parasitologic terms, the condition is classified based on the parasite/host relationship (obligatory or facultative).12

The parasitologic examination of larval samples removed from the present patients determined that the etiologic agent in all cases was Cochliomyia hominivorax of the family Callipharidae, which is found mainly in Central and South America.13 Its larvae prefer warm-blooded animals or humans as hosts. The flies feed on exudates and deposit eggs in necrotic tissue. Each female is capable of laying 2,800 eggs, which are preferentially deposited in a recent wound, lesion, or simple scratch.14 In 12-24 days after being laid, the eggs hatch and the larvae begin to burrow into living tissue, including cartilage and bone.12 The complete cycle takes ~24 days.14

A number of authors state that myiasis occurs with greater frequency in regions with a warm climate in individuals with inadequate hygiene and poor housing conditions (proximity to domesticated animals).2,9,15 The mapping of the cases reported in the literature revealed a predominance of cases in tropical and subtropical regions. The countries with the largest number of cases were Brazil (21 cases), India (16 cases), and Turkey (11 cases). Among the 86 cases of oral-maxillofacial myiasis reported in the international literature, 21 (24.4%) occurred in Brazil, which is the country of origin of the present study. All patients exhibited inadequate hygiene and associated comorbidities.

A greater frequency of the disease is reported in patients with an altered psychologic profile or neurologic disease associated with predisposing clinical conditions, such as lip incompetence, mouth breathing, anterior open bite, under mechanical ventilation, or a wound that did not receive adequate care.8-11 In the cases reported, 6 patients has some type of psychologic or neurologic disorder, which, in theory, made them more susceptible to negligence regarding general health and progression of the disease.

The treatment of choice for oral-maxillofacial myiasis is surgical debridement, which may or may not be associated with the use of a systemic medication. Debridement has a curative function, but some foreign-body reactions may occur in cases in which larvae remain in the surgical wound owing to incomplete debridement.5 Certain drugs are suggested as adjuvants to treatment, such as ivermectin and nitrofurazone.16

Ivermectin is a semisynthetic agent from the family of macrolides, synthesized from natural substances, such as avermectin (obtained from actinomycetes). Ivermectin is a broad-spectrum antiparasitic drug for veterinary use, but with proven efficacy for some par-
asites that attack the human organism. It is generally administered in a single dose of 150-200 μg/kg of body weight. Ivermectin is absorbed quickly and reaches a high concentration in the blood within a relatively short period of time.14

The most common method for dealing with cutaneous myiasis is occlusion of the central ulcer to asphyxiate the larvae. Spontaneous expulsion of the larvae occurs soon after the administration of the asphyxiating agent, which brings larvae deposited in deeper tissue layers to the surface, thereby facilitating their surgical removal.17

Nitrofurazone is a synthetic nitrofuran classified as a topical broad-spectrum antiinfective agent used primarily in burn patients, skin grafts, and the prevention of urinary tract infection due to catheter use. Its specific administration for myiasis involves the liberal placement of the agent in the wound to allow the submerged larvae to come into contact with the offensive liquid substance, forcing them to exit the interior of the tissue and facilitating their removal. The topical administration of this agent occurs over 3 consecutive days.14

In the present study, 5 of the 10 cases were treated with oral ivermectin as an adjuvant to surgical debridement. Removal of the larvae was successful in all cases and no clinical difference was observed between the cases with and without the use of the drug. A number of authors recommend the use of ivermectin without surgical debridement.13,14 However, this treatment plan was not adopted in any of the cases reported here.

Based on the findings of the present study, oral-maxillofacial myiasis is a disease that affects individuals with poor hygiene, many of whom have neurologic and/or psychologic alterations. This condition occurs predominantly in countries near the tropics. The treatment of choice is surgical debridement for the removal of the cause, which may or may not be associated with a specific systemic medication.

REFERENCES


Table 1. Patient distribution

<table>
<thead>
<tr>
<th>Patient</th>
<th>Gender-age (y)</th>
<th>Site</th>
<th>Associated disorder</th>
<th>Causative agent</th>
<th>Duration</th>
<th>Treatment</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>M-72</td>
<td>Upper lip</td>
<td>Uncontrolled diabetes</td>
<td>Cochliomyia hominivorax</td>
<td>36 mo</td>
<td>Debridement</td>
</tr>
<tr>
<td>2</td>
<td>F-82</td>
<td>Hard palate</td>
<td>AIDS + mentally ill</td>
<td>Cochliomyia hominivorax</td>
<td>6 mo</td>
<td>Debridement + ivermectin</td>
</tr>
<tr>
<td>3</td>
<td>M-39</td>
<td>Preauricular</td>
<td>SAH + depression</td>
<td>Cochliomyia hominivorax</td>
<td>24 mo</td>
<td>Debridement + ivermectin</td>
</tr>
<tr>
<td>4</td>
<td>M-67</td>
<td>Retroauricular</td>
<td>Diabetes + mouth breather</td>
<td>Cochliomyia hominivorax</td>
<td>12 mo</td>
<td>Debridement</td>
</tr>
<tr>
<td>5</td>
<td>F-59</td>
<td>Maxilla</td>
<td>Squamous cell carcinoma</td>
<td>Cochliomyia hominivorax</td>
<td>24 mo</td>
<td>Debridement</td>
</tr>
<tr>
<td>6</td>
<td>F-78</td>
<td>Retroauricular</td>
<td>Uncontrolled diabetes</td>
<td>Cochliomyia hominivorax</td>
<td>8 mo</td>
<td>Debridement + ivermectin</td>
</tr>
<tr>
<td>7</td>
<td>M-26</td>
<td>Upper lip</td>
<td>Mental retardation</td>
<td>Cochliomyia hominivorax</td>
<td>3 mo</td>
<td>Debridement</td>
</tr>
<tr>
<td>8</td>
<td>M-34</td>
<td>Maxilla</td>
<td>Mentally ill</td>
<td>Cochliomyia hominivorax</td>
<td>18 mo</td>
<td>Debridement + ivermectin</td>
</tr>
<tr>
<td>9</td>
<td>M-37</td>
<td>Right orbita</td>
<td>Retinoblastome + depression</td>
<td>Cochliomyia hominivorax</td>
<td>6 mo</td>
<td>Debridement + ivermectin</td>
</tr>
<tr>
<td>10</td>
<td>M-40</td>
<td>Retroauricular</td>
<td>Depression</td>
<td>Cochliomyia hominivorax</td>
<td>4 mo</td>
<td>Debridement</td>
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</tbody>
</table>


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