Head and neck myiasis: a case series and review of the literature

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Objective. This study describes 9 cases of myiasis affecting the head and neck region and discusses the demographic distribution, treatment, clinical characteristics, and sequelae of the disease in light of the literature.

Study Design. The study was performed in 2 steps. In the first part, 9 cases seen over a period of 10 years at the Emergency Department of Hospital da Restauração in Brazil were studied. In the second part, a literature search was performed in PubMed for articles on head and neck myiasis published from 1975 to March 2017.

Results. The case series mainly consisted of male patients in their 30s. The palate was the most commonly affected site. Myiasis was caused by *Cochliomyia hominivorax* in all patients, who were treated by mechanical removal of the larvae and debridement of necrotic tissue, followed by oral ivermectin. All patients had sequelae resulting from bone destruction.

Conclusions. Head and neck myiasis generally affects individuals with poor hygiene habits, drug users, and individuals with neurologic and psychosocial disorders. The treatment of choice is mechanical removal of larvae and surgical debridement combined with oral ivermectin. This study provides information that could help clinicians in the diagnosis and management of this condition. (Oral Surg Oral Med Oral Pathol Oral Radiol 2017;124:e249–e256)

The first reports of myiasis in humans and animals in the Neotropical region date back to the sixteenth century.[1] Myiasis is the infestation of living tissues by the larvae of fly species within the order Diptera, which are mainly found in tropical and subtropical regions.[2,3] Human myiasis has a worldwide distribution, with involvement of more species and greater abundance in tropical and subtropical countries. In countries where it is not endemic, myiasis is a relevant condition and is the fourth most common travel-associated skin disease.[4,5] Myiasis is usually among the 5 most common dermatologic conditions, accounting for 7.3% to 11% of cases.[6]

Myiasis is classified according to the type of larva present in the wound: (1) Primary myiasis is caused by larvae that feed on living tissues (biophagous); and (2) secondary myiasis is caused by larvae that only feed on dead tissues (necrophagous).[2,7,8] Abdo et al.[9] classified myiasis as *obligatory* (when the larvae require living tissue to survive) and *facultative* (when the larvae develop in necrotic tissue).

In the head and neck region, the sites most commonly affected by myiasis are the ears, eyes, oral cavity, nose, paranasal sinuses, lymph nodes, mastoid region, and tracheostomy wound.[10] Factors that predispose to myiasis in this region include advanced age, low socioeconomic status, and medical comorbidities, such as a history of craniomaxillofacial trauma and malignant tumors.[7]

Because no standard protocol exists for the treatment of myiasis, different therapies are adopted. Initial treatment normally consists of mechanical removal of the larvae. According to Ribeiro et al.[11] and Gealh et al.,[12] ivermectin is increasingly being used for the treatment of head and neck myiasis. This antiparasitic agent of the macrolide family blocks nerve impulses at the nerve endings of endoparasites, causing paralysis and death of the larvae.[13] The sequelae of myiasis include blindness, hearing loss, and even death, and immediate treatment is thus essential.[14]

This report describes 9 cases of myiasis affecting the head and neck region and discusses the demographic distribution, treatment, clinical characteristics, and sequelae of the disease in light of the literature.

MATERIALS AND METHODS

Study design and ethical approval

This study was performed in 2 steps. In the first part, 9 cases of myiasis affecting the head and neck region were

Statement of Clinical Relevance

Head and neck myiasis is a disease that affects individuals with poor hygiene habits, drug users, and individuals with neurologic and psychosocial disorders. This article provides information that could help clinicians in the diagnosis and management of this condition.

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studied. In the second part, a literature review on head and neck myiasis was conducted in Medline (PubMed) of reports published from 1975 to March 2017. The study was approved by the local Ethics Committee (Approval No. 00670912.1.0000.5207) and was conducted in accordance with the guidelines of the Declaration of Helsinki.

Step 1: sample

A case-series study that involved patients of both genders and different ages with head and neck myiasis was conducted. The sample was determined by spontaneous demand at the Emergency Department of Hospital da Restauração, Recife (PE, Brazil) between January 2006 and January 2016. The data were grouped according to (1) gender, age, location, etiology, presence of local and systemic disorders, duration of treatment, time since onset of the disease, length of hospital stay, and sequelae; (2) fly species involved; and (3) treatment.

In the case of oral myiasis, the oral conditions of the patients were recorded, and the following conditions were defined as a "poor oral hygiene": gingivitis, halitosis, and dental calculus.

The diagnosis of myiasis was based on clinical characteristics and the presence of larvae, which were identified by an entomologist.[2]

Step 2: literature review and case series

Articles on head and neck myiasis published from 1975 to March 2017 in all languages were analyzed and the cases were mapped according to continent of occurrence of the disease. The Medline (PubMed) database was searched using the following keywords: "oral myiasis," "nasal myiasis," "ophthalmomyiasis," "maxillofacial myiasis," "head and neck myiasis," "ear myiasis," "oral malignancy myiasis," "nasal malignancy myiasis," "ophthalmomyiasis malignancy," "maxillofacial malignancy myiasis," "head and neck malignancy myiasis," and "ear malignancy myiasis."

Case reports not related to human myiasis, duplicate studies, and articles without abstracts were excluded. The cases were counted and separated by continent for a better understanding of the distribution of the disease in the world.

RESULTS

Case series

The sample of this study consisted of 9 patients (6 men and 3 women). No association with any malignant tumor was observed. Myiasis was caused by larvae of the same fly species (*Cochliomyia hominivorax*) in all patients. The mean age of the patients was 38 years. With respect to anatomic location, the submandibular region was involved in 1 case, the orbital region in 2 (Figure 1), and the retroauricular region in 2. The palate alone was affected in 1 patient. In the 3 other patients, in addition to the palate, infestation occurred in the lip and alveolar ridge in 2 and in the alveolar ridge in 1. The patients had a low socioeconomic status and low education level. Regarding etiology, a poor oral hygiene was a cause in 3 patients, and 3 other patients exhibited mental retardation (1 case), drug use (1 case) and cerebral palsy (1 case) as local and systemic conditions.

Fig. 1. Clinical presentation of a 21-year-old female patient with myiasis in the orbital region.

First, the patients received surgical intervention under local or general anesthesia (Table I), which consisted of mechanical removal of the larvae, debridement of necrotic tissue, and primary closure of the wound. Second, all patients received a systemic antibiotic (cephalothin, 1 g, every 6 hours), anti-inflammatory agent (ibuprofen, 400 mg, every 6 hours), and analgesic (paracetamol, 500 mg, every 8 hours). This protocol was followed by oral administration of a single dose of ivermectin (150 µg/kg) in all cases. The time since the onset of the disease ranged from 3 to 8 months. The average length of hospital stay was 12 days, with a treatment duration of 9 days. The sequelae observed were bone destruction in all cases, hearing loss on the affected side (2 cases), blindness (2 cases), oroantral communication (1 case), and irreversible salivary gland involvement (1 case).

Literature review

In the second step, 814 articles were retrieved, and 482 were excluded. The remaining 325 studies (707 cases) were selected according to the inclusion criteria and are indicated on the map in Figure 2.

Table I. Data on patients with head and neck myiasis

Case	*Gender-age*	*Location*	*Etiology*	*Fly species*	*Type of anesthesia*	*Local or systemic disorder*	*Time since onset of disease*	*Treatment duration*	*Established treatment*	*Sequelae*
1	M-22	Lip, alveolar ridge, and palate	Poor oral hygiene	*Cochliomyia hominivorax*	General	None	3 months	10 days	Surgical debridement, analgesic, antibiotic, and oral ivermectin	Bone and soft tissue loss
2	F-50	Retroauricular region	Trauma	*Cochliomyia hominivorax*	Local	Intellectual disability	5 months	20 days	Surgical debridement, analgesic, antibiotic, and oral ivermectin	Bone and soft tissue loss; hearing loss on the affected side
3	F-45	Alveolar ridge and palate	Poor oral hygiene	*Cochliomyia hominivorax*	General	None	4 months	12 days	Surgical debridement, analgesic, antibiotic, and oral ivermectin	Bone and soft tissue loss
4	M-33	Retroauricular region	Trauma	*Cochliomyia hominivorax*	Local	None	3 months	8 days	Surgical debridement, analgesic, antibiotic, and oral ivermectin	Bone and soft tissue loss; hearing loss on the affected side
5	M-26	Lip, alveolar ridge, and palate	Trauma	*Cochliomyia hominivorax*	Local	None	3 months	10 days	Surgical debridement, analgesic, antibiotic, and oral ivermectin	Bone and soft tissue loss
6	M-57	Orbital region	Trauma	*Cochliomyia hominivorax*	General	Alcoholism	6 months	22 days	Surgical debridement, analgesic, antibiotic, and oral ivermectin	Blindness; bone and soft tissue loss
7	F-21	Orbital region	Trauma	*Cochliomyia hominivorax*	General	None	3 months	12 days	Surgical debridement, analgesic, antibiotic, and oral ivermectin	Blindness; bone and soft tissue loss
8	M-24	Palate	Poor oral hygiene	*Cochliomyia hominivorax*	General	Cerebral palsy	8 months	12 days	Surgical debridement, analgesic, antibiotic, and oral ivermectin	Bone and soft tissue loss; oroantral communication
9	M-65	Submandibular region	Trauma	*Cochliomyia hominivorax*	Local	None	3 months	8 days	Surgical debridement, analgesic, antibiotic, and oral ivermectin	Bone and soft tissue loss; loss of the salivary gland

F, female; *M*, male.

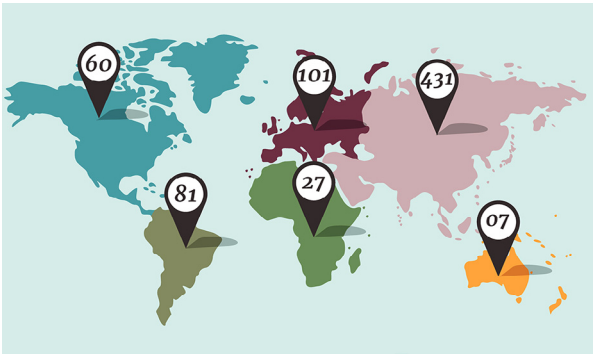

Fig. 2. World distribution of the 707 cases of head and neck myiasis reported in the literature.

A literature review associating myiasis with malignant neoplasms (Table II) identified 23 articles. The most significant malignant neoplasms were squamous cell carcinoma, basocellular carcinoma, Hodgkin lymphoma, and melanoma. The most affected countries were India in Asia, Tunisia in Africa, Germany in Europe, Brazil in South America, the United States in North America, and Australia in Oceania.

DISCUSSION

Myiasis is the infestation of human or animal tissues by larvae of fly species within the order Diptera,[2,3] which is intimately related to the life cycle of different species of these insects. The prevalence of the disease is high in tropical and subtropical regions.[2,7] Local factors associated with myiasis are poor oral hygiene, periodontal diseases, suppurative lesions, halitosis, mouth breathing, anterior open bite, lip incompetence, and trauma caused by physical aggression.[2,7,9,17-19] Systemic conditions that predispose to the development of myiasis are intellectual disability, cerebral palsy, epilepsy, alcoholism, and senility, conditions that are related to the socioeconomic profile of patients.[2,8,11,20-22] These conditions seem to play a crucial role in the development of the disease, as demonstrated by the cases reported so far. In the present case series, poor oral hygiene was observed in 3 cases, intellectual disability in 1, alcoholism in 1, and cerebral palsy in 1 patient.

C. hominivorax, a fly species native to the Neotropical region that is common in Brazil,[23] is an obligate parasite of mammals, including humans. The larval stage represents the obligate parasite, whereas the other stages are harmless to the host. At this stage, the larvae feed fiercely on living tissue, including cartilage and bone,[7] initiating a feeding cycle that lasts approximately 3 days before migration from the wound.[23] In the present study, *C. hominivorax* was the species involved in all cases, which were classified as primary myiasis. The diagnosis of myiasis is clinical. However, identification of the larval species involved is certainly useful for epidemiologic surveys and, if not for eradication, at least for control of the disease through the cleanup of contaminated areas.

Although *C. hominivorax* larvae are the main agent of myiasis, other species, such as *Oestrus ovis*, *Wohlfahrtia magnifica*, *Chrysomya bezziana*, *Hypoderma bovis*, *Cordylobia anthropophaga*, *Hypoderma tarandi*, *Callliphora vicina*, *Musca nebulo*, *Musca domestica*, and *Lucilia sericata* have been reported in the literature as cases of this infestation.[2,17,20] In view of the complex and similar morphologies of different species, identification of the larval species should be performed by an experienced entomologist.[2]

The incidence of extraoral myiasis is higher than that of oral myiasis because tissues of the oral cavity are not permanently exposed to the external environment,[24] in agreement with the present study in which 5 of the 9 cases involved extraoral sites. The palate was the most commonly involved site in the oral cavity, and there were cases in which more than one site was affected. The correlation between anatomic location and wound etiology is an important factor in the development of myiasis, as are sanitary conditions and the presence of systemic disorders. The profile of patients with head and neck myiasis agrees with that in the study by Antunes et al.,[7] who evaluated 10 cases of oral and maxillofacial myiasis over a period of 6 years, and all of those 10 patients had some systemic disease.

A review of the English literature identified 31 cases of myiasis associated with malignant neoplasms involving the head and neck region. Treatment consisted of mechanical removal of the larvae, surgical debridement, and the administration of antibiotics. The association with a malignant tumor can be attributed to ulcerated and necrotic lesions exposed to the environment in patients with cancer and to inadequate disinfection.[25] However, cases of oral myiasis associated with malignant tumors are rare.[14-16,26] In general, the largest numbers of reports associated with malignancy are found in Asia, particularly India.

Several authors argue that myiasis occurs predominantly in countries with hot weather.[7] The countries most affected by this disease are India, Tunisia, Brazil, and Australia, which are the countries on their respective continents with this climate characteristic. Furthermore, India, Tunisia, and Brazil are underdeveloped or developing countries, thus demonstrating the social nature of the disease.

The standard treatment of myiasis focuses on antibiotic therapy concomitantly with mechanical removal of necrotic tissues.[7,27] This approach was adopted for the 9 patients of this case series, in which antibiotic therapy and mechanical debridement were effective. However, some foreign body–type reactions may occur in cases in which the larvae remain in the surgical wound because

Table II. Head and neck myiasis associated with malignancy

Author (Year)	*Country*	*Cases*	*Location*	*Neoplasm*	*Therapy*
David et al. (1996)[28]	India	1	Rhino-orbital	Non-Hodgkin lymphoma	Not mentioned
Caça et al. (2003)[29]	Turkey	1	Orbit, maxilla, nose, and ossa zygomatica	BCC	OE and total maxillectomy
Osorio et al. (2006)[30]	Colombia	2	Orbit; orbit and zygoma	SCC; BCC	OI, antibiotics, surgical debridement with enucleation of the eye; OI, antibiotics and surgical debridement
Hawayek & Mutasim (2006)[31]	USA	1	Parieto-occipital scalp	SCC	Application of petrolatum jelly in the region, wearing a shower cap, and surgical resection of the tumor
Bouwman et al. (2007)[32]	The Netherlands	1	Frontal	BCC	Resection of the tumor and surgical debridement of the dura
Gabriel et al. (2008)[14]	Brazil	1	Cervicofacial	SCC	MR and application of ether embedded cotton
Gopalakrishnan et al. (2008)[33]	India	4	Neck and pharynx; tracheal stoma; face; lip	Carcinoma of hypopharynx; carcinoma of larynx; cheek cancer; lip cancer	MR and application of turpentine oil; antibiotics and hematinics, MR and surgical debridement; not mentioned
Carvalho et al. (2008)[15]	Brazil	1	Midface	Epidermoid carcinoma	MR
Sesterhenn et al. (2009)[34]	Germany	1	Oropharynx	SCC	MR, antiseptics, and antibiotics
Wollina (2010)[35]	Germany	1	Scalp	SCC	MR, antiseptic
Yeung et al. (2010)[36]	China	1	Orbit	SCC	Emergency OE and surgical debridement
Pessoa & Galvão (2011)[16]	Brazil	1	Mandible	SCC	MR, antiseptics
Kamal et al. (2012)[37]	India	1	Orbit	SCC	Surgical debridement and emergency OE
Villwock & Harris (2012)[27]	USA	4	Ear; neck; nasal and facial; upper eyelid	SCC; melanoma; BCC; BCC	Antibiotics and MR
Bayindir et al. (2012)[38]	Turkey	1	Parotid gland	SCC	MR, saline solution, antiseptics
Robati et al. (2012)[39]	Iran	1	Auricle	SCC	MR, surgical debridement, antibiotics
Dharshiyani et al. (2012)[40]	India	1	Mandible	SCC	MR, antiseptics, anthelmintic, antibiotics
Tomy & Prabhu (2013)[41]	India	1	Orbit	Follicular thyroid carcinoma	Antibiotics, insulin doses, application of turpentine oil, and MR
Jan et al. (2013)[42]	USA	1	Nasal cavity and maxillary sinus	Intracranial meningioma	Surgical debridement and antibiotics
Rowicki & Pietniczka-Zaleska (2015)[43]	Poland	1	Neck	SCC	MR
Biradar et al. (2015)[44]	India	2	Buccal mucosa; buccal mucosa	SCC; BCC	MR, surgical debridement, antibiotics
Pandey et al. (2016)[45]	Nepal	1	Eyelid and orbit	BCC	MR, OI, and OE
Jorge et al. (2016)[46]	Brazil	1	Hard palate and nasal cavity	Extranodal NK/T-cell lymphoma	MR, OI, debridement and antibiotics

BCC, basal cell carcinoma; *OE*, orbital exenteration; *OI*, oral ivermectin; *MR*, Mechanical removal; *NK*, natural killer; *SCC*, squamous cell carcinoma.

Table III. Review of treatment options for myiasis in humans

Author (year)	*Management*	*Route of administration*
Ribeiro et al. (2001)[11]	Surgical removal (mechanical treatment) of the larvae and debridement of infected tissues	—
Antunes et al. (2011)[7] Villwock & Harris (2014)[27]	Antibiotic therapy concomitantly with mechanical removal of necrotic tissues	Intravenous
Hubler et al. (1974)[47] Felices & Ogbureke (1996)[48] Lata et al. (1996)[49] Messahel et al. (2010)[50]	Occlusion of the wound with an asphyxiating agent: Vaseline, heavy oil, bee wax, raw meat, mineral oil, varnish, adhesive tape, butter, chewed gum, turpentine oil, tobacco leaf, chloroform, and ether	Topical
Ribeiro et al. (2001)[11] Antunes et al. (2011)[7]	Ivermectin (single dose of 150 μg/kg)	Oral
Gealh et al. (2009)[12]	Nitrofurazone (over 3 consecutive days)	Topical

of incomplete removal of contaminated tissue.[2,17] As in oral myiasis, in which the mechanical removal of larvae from the necrotic area provides adequate outcomes, manual removal of the larvae and cleanup of necrotic tissues were also effective in this series of myiasis cases involving the head and neck region (i.e., superficial sites). Shinohara et al.[8] and Alves et al.[51] highlighted the need for immediate management of orofacial myiasis to avoid potential complications, such as extensive tissue destruction, palatal perforation, and cavernous sinus thrombosis.

Ivermectin belongs to the chemical group of avermectins and is a semisynthetic macrolide antibiotic. The drug is widely used for the treatment and control of worm infestations in large animals, such as cattle, horses, sheep, goats, and camelids. Ivermectin is generally administered as a single dose of 150 to 200 μg/kg body weight. Since the drug is rapidly absorbed and reaches high blood concentrations within a relatively short period, the larvae are quickly expelled from the wound.[12] The protocol described by Ribeiro et al.,[11] which recommends oral administration of ivermectin at a dose of up to 300 μg/kg (i.e., for patients weighing 40-60 kg: 2 tablets (12 mg); and for patients weighing 60-90 kg: 3 tablets [18 mg]) for 3 consecutive days does not cause any intercurrence and eliminates all larvae in cavitary myiasis.

Although Shinohara et al.[8] and Gealh et al.[12] recommend the use of ivermectin without debridement for patients with cutaneous myiasis, no standard protocol exists for the treatment of head and neck myiasis. The articles retrieved were analyzed with regard to treatment options for myiasis and routes of administration in humans (Table III). The 9 cases of the present study were treated by mechanical removal of the larvae and surgical debridement of necrotic tissues. In addition, the patients received oral ivermectin (a single dose of 150 μg/kg) as recommended by Antunes et al.[7] and Ribeiro et al.[11] It should be noted that the protocol adopted at our institution was effective in resolving the disease in the 9 study patients. This alternating or concomitant treatment with ivermectin is believed to exert a beneficial effect within a short period.

Most authors prefer topical or local infiltrative anesthesia for removal of the larvae and necrotic tissue.[7] In cases where the patient does not cooperate, general anesthesia is preferred for larval removal, which was performed in 5 patients of the present study. Three patients did not cooperate: 1 had cerebral palsy, and in 2 patients the site was difficult to access. After treatment, the larvae were completely eradicated in all cases after washing with physiologic saline or spontaneously eradicated (possibly after being swallowed or eliminated to the external environment).

The existence of a variety of treatments indicates the lack of a protocol; however, control measures of the fly population have emerged as an alternative strategy to prevent infestation. Basic sanitation and eradication of foci of adult fly proliferation are the first steps in combating this disease. Other measures that have been adopted include the application of ultraviolet light to attract flies and then eliminate them by electrocution and the use of boric acid for domestic elimination of this pest.[2,24,52] These findings highlight the social nature of the disease; its eradication requires the participation of the local community, the state, and the health care network because systemic problems should not be neglected.

The sequelae observed in this case series were severe and included blindness, hearing loss, and salivary gland involvement, in addition to the development of an oroantral communication and considerable bone and soft tissue loss. Similar or even more severe findings have been reported in the study by McGraw and Turiansky,[53] who found that myiasis can lead to massive tissue destruction, loss of vision and hearing, and even death. Establishment of the diagnosis and immediate treatment are therefore necessary to interrupt progression of the disease and to prevent more devastating sequelae.

The present results show that head and neck myiasis is associated with the socioeconomic profile of patients in underdeveloped countries and that the disease affects persons with poor oral hygiene and low socioeconomic status, homeless individuals, rural residents, drug users, and those with neurologic and psychosocial disorders.

In this respect, myiasis is mainly found to occur in developing countries and close to the tropics.

CONCLUSIONS

In summary, the treatment of choice for head and neck myiasis is surgical debridement to remove necrotic tissue, combined with a specific systemic medication, such as ivermectin. This study provides interesting information that could help clinicians in the diagnosis and management of this condition.

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