

# **Otomyiasis: Systematic Review**

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## **Abstract**

**Introduction** Myiasis is a disease caused by fly larvae that grow in the tissues of animals and humans. It can cause a variety of local symptoms, like erythema or pain, depending on its location, and generalized symptomatology, such as fever and malaise. Myiasis can generate severe complications, for instance sepsis, or directly impact vital tissues. Its management varies depending on the location, and on the preferences of the doctor that faces this challenge. Myiasis usually occurs in tropical countries, and, in many places, it is not a rare condition. The cases are rarely reported, and there are no published management protocols. **Objective** To review the literature regarding the most common agents, the predisposing factors and the treatment alternatives for otic myiasis, a rare form of human myiasis caused by the infestation of fly larvae in the ear cavities.

# Data synthesis We present a systematic review of the literature. The search in five databases (Medline, Embase, Cochrane Database of Systematic Reviews, LILACS and RedALyC) led to 63 published cases from 24 countries, in the 5 continents. The ages of the patients ranged from newborn to 65 years old. The most common agents belong to the Sarcophagidae or Calliphoridae families. Chronic otitis media, previous otic surgical procedures, mental deficit, alcohol or drug abuse, sleeping outdoors, prostration, and malnutrition were predisposing factors. The treatment alternatives are herein discussed.

**Conclusion** The results highlight the need for monitoring, follow-up and standardization of medical approaches.

## **Keywords**

- ► diptera
- ear diseases
- larva
- myiasis
- parasites
- sarcophagidae

### Introduction

The word myiasis was first used by Hope, in 1840,<sup>1</sup> to refer to a parasitic disease caused by certain fly larvae during a particular stage of their development when they feed on animal and human tissues. The geographic distribution of the condition is almost exclusively limited to hot, tropical areas, and its etiological agent varies from one region to another. Depending on the affected organ, myiasis can be classified in cutaneous, enteric, ophthalmic, nasopharyngeal, urogenital, oral and, as in our case, the less common otic

myiasis. This article presents a specific systematic review of the published literature on otic myiasis.

#### **Review of the Literature**

#### Methodology

A systematic review of the literature was made in June of 2016 on five different databases (Medline, Embase, Cochrane Database of Systematic Reviews, LILACS and RedALyC). To widen the sensitivity of the search, we used free terms; ► Supplementary Material 1 (available online only) lists the

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search terms as well as the search constructs for the different databases. We conducted a systematic review by following the guidelines of the Preferred Reporting Items for Systematic reviews and Meta-Analyses (PRISMA)<sup>2</sup> (>Supplementary Material 2). No limits were applied for age, year of publication, or language. Articles without an abstract were excluded. The titles and summary lists were evaluated by two researchers simultaneously to evaluate exclusion criteria: those that were clearly irrelevant, as well as those focused on therapeutic treatment for larvae, non-human animal studies, forensics, entomologic studies, or patients with risk factors such as recent surgical wounds, cancer or ulcers. Information from the full-text remaining articles was collected in a predesigned Excel spreadsheet ► Supplementary Material 3. The quality of the publications was assessed using the Joanna Briggs Institute scale for case series,<sup>2</sup> and the Center for Evidence-Based Management (CEBMa) scale for individual case report studies.<sup>3</sup>

#### Results

From the 272 initial studies (**Fig. 1**),<sup>4</sup> 82 of them were selected for full text review, 29 of which could not be found (23 of these were published between 1918 and 1994). After a second exclusion analysis, 6 articles were rejected for being considered irrelevant, and 7 more for failing to meet the

quality criteria. The remaining 40 studies reported 63 cases from 24 countries (Argentina, Australia, Belize, Canada, Colombia, Costa Rica, Cuba, Dominican Republic, Germany, India, Iran, Italy, Malaysia, Morocco, Nigeria, Paraguay, Poland, Rumania, Saudi Arabia, South Korea, Spain, Thailand, Turkey, and United States). **– Table 1**<sup>5–44</sup> summarizes the information from these articles (**– Supplementary Material 4**).

Of the 63 cases reported, 34 were men (62%) and 21 were women (in 8 cases, no gender was reported). The ages ranged from 2 days to 65 years old, with an average of 26. Chronic otitis media, previous otic surgical procedures, mental deficit, alcohol and drugs use, sleeping outdoors, prostration, malnutrition, jaundice and previous episodes of otomyiasis were predisposing factors. In 30 patients, no risk factors were mentioned. As for the etiological agents, the parasite was identified in 44 cases, 23 corresponded to the *Sarcophagidae* family, 20 to the *Calliphoridae* family, and 1 to the *Oestridae*. Eleven cases were reported in Latin America (2 in Argentina, 2 in Colombia, 2 in the Dominican Republic, 2 in Paraguay and one each from Belize, Costa Rica, and Cuba); 7 of them informed the agent, which was always from the *Calliphoridae* family.

Complications were reported in 23 of the 63 cases, describing tympanic perforation, chronic otitis media, temporal bone extension, cartilaginous destruction, cellulitis and perichondritis. The use of extraction with clamp, either

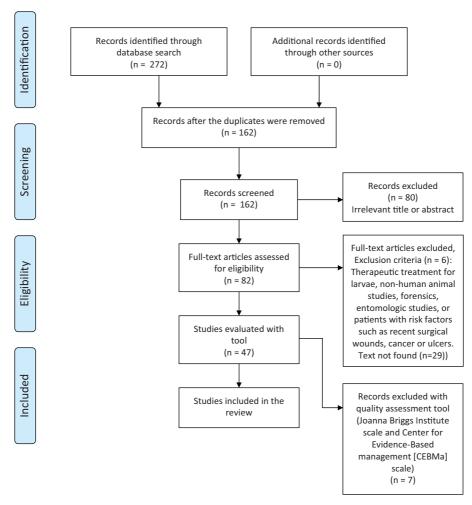


Fig. 1 Prisma diagram of the search strategy.<sup>4</sup>

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 Table 1
 Characteristics of patients with otomyiasis

Poforonco	Country	5	ΔΩΦ	Candar	Clinical characteristics	Etiologic agent
2	Turkey	: 9	2=7 vr	3 M 3F		Wohlfahrtia maanifica
9	Colombia	2	33, 23 yr	M, M	Drug addiction, otologic surgery in childhood due to otitis media	
7	Malaysia	-	2 d	Σ	1	Sarcophagidae
8	Turkey	-	8 yr	Σ	Chronic otitis media	W. magnifica
6	Turkey	1	37 yr	M	Previous otomyiasis	W. magnifica
10	Saudi Arabia	-	12 yr	Σ	I	W. magnifica
11	Cuba	-	25 d	ı	ı	Calliphoridae phaenicia
12	Costa Rica	-	9 yr	Σ	Chronic otitis media	Cochliomyia hominivorax
13	Spain	1	55 yr	M	Alcoholic	C. phaenicia
14	Spain	-	5 mth	Σ	I	W. magnific
15	Turkey	-	12 d	Ш	Jaundice, thyroid dysgenesis	Lucilia sericata
16	Poland	2	44, 57 yr	F, M	Carcinoma of middle ear	L. sericata
17	Thailand	1	2 d	Ī	_	Parasarcophaga (Liosarcophaga) dux (Thomson)
18	South Korea	1	54 yr	M	_	L. sericata
19	USA	1	7 wk	M	_	Calliphoridae
20	Canada	1	60 yr	Μ	1	L. sericata
21	Spain	1	65 yr	F	-	C. bezziana
22	Dominican Republic	1	44 yr	F	-	_
23	India	1	3 yr	F	Otitis chronic media and mental retardation	Musca domestica
24	Turkey	1	57 yr	F	1	W. magnifica
25	Dominican Republic	1	26 yr	F	1	C. hominivorax
26	Malaysia	1	10 yr	F	1	Chrysomya megacephala
27	Italy	1	52 yr	F	Mental retardation	Sarcophaga hemorrhoidalis.
28	Paraguay	1	28 yr	M	Otorrhea bilateral	_
59	Belize	1	ı	-	_	C. hominivorax
30	Argentina	2	11, 9 yr	M, F	Chronic malnutrition	C. hominivorax
31	Australia	1	16 yr	Μ	Mental retardation	Sarcophaga
32	Paraguay	1	37 yr	Μ	1	C. hominivorax
33	Nigeria	_	4 mth	ட	I	1

Table 1 (Continued)

Reference	Country	u	Age	Gender	Clinical characteristics	Etiologic agent
34	Romania	1	ı	-	Previous petromastoidectomy	L. sericata
35	Italy	-	44 yr	Σ	Farmer, chronic otitis media	W. magnifica
36	Malaysia	-	46 yr	Σ	Mental retardation	C. bezziana
37	India	14	1–35 yr	I	Chronic otitis media, ulcer, otitis externa and perichondritis.	ı
38	Iran	-	55 yr	M	ı	C. bezziana
68	Morocco	3	5,12,14	I	Chronic otorrhea	W. magnifica
40	Turkey	-	31 yr	M	Radical mastoidectomy secondary to gunshot wound	W. magnifica
41	Germany	1	49 yr	M	Alzheimer and tetraplegia	Sarcophaga
42	Iran	-	62 yr	LL.	Pulmonary edema and decompensated heart failure	L. sericata
43	Turkey	1	5 yr	M	-	W. magnifica
44	Spain	1	2 yr	M	Accompanied parents in field work	W. magnifica
Abbreviations: d day	Abbreviations: d days: F female: M male: mth months: wk weeks: yr years	nths wk we	Pks. Vr vears			

with or without anesthesia, was reported, and, in some cases, with the aid of suction and complementary otic lavage with different types of substances, such as saline, lidocaine, alcohol, and hydrogen peroxide. In 39 cases, antibiotics were mentioned, topical, oral or parenteral.

#### **Discussion**

Myiasis is a relatively common public health problem in developing countries, mainly in hot, tropical areas. <sup>45</sup> There are, however, no clinical practice guidelines for diagnosis or treatment of this disease. Otomyiasis occurs when the female fly, perhaps attracted by bad odor, deposits its larvae in the auditory meatus. <sup>46</sup> Chronic otitis media or others otic pathologies are, therefore, predisposing factors. A recent review article, with 45 cases of otomyiasis, <sup>47</sup> considers that sanitary conditions play an important role, but other triggers are: low socioeconomic status, swimming in stagnant waters, diabetes mellitus, alcoholism, prostration and suppurative chronic otitis media. Our review shows chronic otitis media as a main risk factor that favor larvae growth.

Taxonomic division establishes several families derived from the order *Diptera*: *Oestridae*, *Calliphoridae* and *Sarcophagidae* are the most important. The species *Wohlfahrtia magnifica*, which belongs to the latter family, is a must-type larva, meaning that it requires living in a host tissue to complete its development. This species is the predominant etiologic agent worldwide, <sup>47</sup> but *Sarcophagidae* is the predominant family in Europe, and in the Middle East. In Latin America, all the cases that reported the larvae belonged to the *Calliphoridae* family.

After having diagnosed myiasis through physical examination and, in some cases, through diagnostic images, the treatment must begin as soon as possible, including direct or surgical extraction of the larva. To achieve that goal, the literature shows different treatments, which include antiparasitic therapy with oral ivermectin<sup>49,50</sup> or topic permethrin. The occlusion of the lesion is used to force the larvae to exit, thus facilitating its manual extraction; this can be done using olive oil or petroleum jelly. Another popular treatment, commonly used in rural areas, involves placing a piece of bacon to attract the larvae, as reported in the Villamizar case.

The treatment should also include extraction using alligator forceps under micro-otoscopy vision, in some cases anesthesia, suction and ear washes with saline solution 0.9%. Systemic antibiotic therapy is also used when there is chronic otitis media, as discussed by Yuca et al<sup>5</sup> The location of the infection and its proximity to the brain are a concern, and authors have discussed the risk of myiasic meningitis and eventually death of the patient, but we did not find this complication reported in the literature.

#### **Final Comments**

Otomyiasis is a parasitic condition that tends to affect vulnerable subjects in tropical countries. Improving personal

care would reduce the risk. Training health providers is important for early diagnosis and treatment.

#### Note

All authors contributed equally to this work.

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