



Orbital myiasis

A case report and literature review

Yan-Ling Huang, MD^a, Lu Liu, MD^c, Hao Liang, MD^a, Jian He, MD^a, Jun Chen, MD^a, Qiao-Wen Liang, MD^a, Zhi-Yuan Jiang, MD^b, Jian-Feng He, MD^{a,*}, Min-Li Huang, MD^{a,*}, Yi Du, MD^{a,*}

Abstract

Rationale: Myiasis is a parasitic disease caused by fly larvae of the Diptera order that infest human and other vertebrate animal tissues. Orbital myiasis is a potentially destructive infestation of the orbital tissues, which may affect individuals with previous ocular diseases or disorders of consciousness.

Patient concerns: A 72-year-old man presented with a complaint of repeated pain for two years after trauma to his right eyelid and aggravated symptoms with larvae wriggling out for 2 days. An orbital computed tomography scan revealed right eyeball protrusion and periocular soft tissue edema. Two days later, magnetic resonance imaging showed that the shape of the right eyeball was changed and that the normal structure of the eyeball could not be identified.

Diagnoses: Due to the patient's symptoms and imaging examination results, the diagnosis of orbital mylasis was made.

Interventions: The patient was treated by exenteration of the right orbit, and all necrotic tissues and larvae were removed. The defect was repaired via reconstruction with a pedicled musculocutaneous flap from the forehead region. Antibiotics and tetanus toxoid therapy were utilized to prevent potential bacterial infection.

Outcomes: The patient recovered well postoperatively and was discharged uneventfully. During the 6-month follow-up period, the wound healed well.

Lessons: Advanced age and untreated eye trauma are risk factors for orbital myiasis. Timely removal of larvae and elimination of infections are important measures for protecting the eyeball.

Abbreviations: BCC = basal cell carcinoma, F = female, I.V. = intravenous, M = male, OD = right eye, OS = left eye, SCC = squamous cell carcinoma.

Keywords: Calliphoridae Diptera, Maggot Diptera: myiasis, ophthalmomyiasis, orbital myiasis

1. Introduction

Maggots are larvae of Diptera flies, most of which are mainly found in human and animal feces, garbage, decaying plants and

Editor: N/A.

Y-LH, LL, and HL contributed equally to this work and should be considered cofirst authors.

This study was supported by the National Natural Science Foundation of China (No. 81560162), the Guangxi Natural Science Foundation (No. 2016GXNSFAA380301, 2018GXNSFAA050052, 2016GXNSFBA380115). Self-funded research projects of Guangxi Health and Family Planning Commission (Z2016298).

The authors have no conflicts of interest to disclose.

^a Department of Ophthalmology, ^b Department of Hypertension division, the First Affiliated Hospital of Guangxi Medical University, ^c Guangxi Medical College, Nanning, Guangxi, China.

* Correspondence: Yi Du, Min-Li Huang, Jian-Feng He, Department of Ophthalmology, The First Affiliated Hospital of Guangxi Medical University, 6 Shuangyong Road, Nanning 530021, Guangxi, China (e-mail: duyi@gxmu.edu.cn, nnhml@163.com, hejianf@foxmail.com)

Copyright © 2020 the Author(s). Published by Wolters Kluwer Health, Inc. This is an open access article distributed under the Creative Commons Attribution License 4.0 (CCBY), which permits unrestricted use, distribution, and reproduction in any medium, provided the original work is properly cited.

How to cite this article: Huang YL, Liu L, Liang H, He J, Chen J, Liang QW, Jiang ZY, He JF, Huang ML, Du Y. Orbital myiasis: A case report and literature review. Medicine 2020;99:4(e18879).

Received: 8 July 2019 / Received in final form: 23 November 2019 / Accepted: 23 December 2019

http://dx.doi.org/10.1097/MD.000000000018879

animal carcasses and feed on feces and decaying organic matter. In cases of accidents or in certain specific species, they can infest vertebrate animals, including humans, and feed on living or dead tissue, as well as on body fluids, [1] leading to myiasis. Myiasis mainly occurs in animals such as cattle, goats and pigs but occasionally occurs in humans. [2] Advanced age, disease-ridden status, poor self-care, poor hygiene, and rural background are reported risk factors for human myiasis. A pastoral or rural background provides the conditions for the prevalence of myiasis because it is a zoonotic disease. Myiasis is mainly prevalent in tropical and subtropical regions, where a warm and humid climate prevails almost throughout the year, [1] or in developing countries with a large population density and poor sanitation.

Ophthalmomyiasis can involve the eye, orbit, and periorbital tissues. It is classified as external, internal or orbital in accordance with the site of the larvae infestation. [3] Limited superficial infestation of external ocular tissues such as the palpebra and conjunctiva is called external ophthalmomyiasis. When the larvae invade deeply and migrate into the subretinal space, internal ophthalmomyiasis occurs. Orbital myiasis is a more extensive infestation involving orbital tissue and is the most serious form. Once established, orbital myiasis progresses rapidly and can completely destroy the orbital tissues within days. [4] Fortunately, it is the least common form, [5] with only a few cases reported. Management of orbital myiasis ranges from simple manual removal of the maggots to destructive surgeries of the globe and orbit. [6]



Figure 1. A color photograph demonstrating that the right orbit was destroyed and several larvae were wriggling out.

Medical professionals are unfamiliar with orbital myiasis because it is such a rare disease, which may increase the difficulty of recognition and treatment. Here, we report a case of orbital myiasis and conduct a systematic literature review of cases previously reported in the literature, aiming to better outline the clinical features and therapeutic management of orbital myiasis. The patient himself consented to the publication of the study. This case report was approved by the ethics review committee of First Affiliated Hospital of Guangxi Medical University, (2019-KY-E-036), Nanning, China, and an informed consent form was signed by the patient himself.

2. Case report

A 72-year-old male patient presented to the emergency department on October 31, 2015, with a complaint of repeated pain for two years after trauma to his right eyelid and a 2-day history of symptoms aggravated by the wriggling out of larvae. The patient reported that his right upper lid had been injured by cane leaves 2 years prior, but no treatment was received. Then, he experienced repeated pain in his right eye, accompanied by gradually decreased vision until it was completely lost 1 year previously. His painful symptoms worsened 2 days before presentation, with bleeding, a crawling sensation and larvae wriggling out (Fig. 1). He denied a history of alcoholism, previous ocular surgery, or prolonged use of medications.

On ophthalmic examination, the visual acuity test revealed no light perception in his right eye. His right periorbital skin was red and edematous, and the eyelid was thickened. There was a large eyelid wound of approximately $4\,\mathrm{cm}*1\,\mathrm{cm}$ filled with numerous white larvae, some of which were crawling out. No abnormalities were found in the left eye or upon systemic examination. A computed tomography scan revealed that the right eyeball protruded and that the soft tissues around it were swollen (Fig. 2). Two days later, magnetic resonance imaging showed that the shape of the right eyeball was changed and that the normal structure of the eyeball could not be identified (Fig. 2). A diagnosis of orbital myiasis was made.

Considering potential infections, the patient received topical levofloxacin eye drops, intravenous ceftazidime and levofloxacin, and a tetanus antitoxin injection. In view of imaging evidence of total destruction of the globe caused by infiltration of the larvae, exenteration of the right orbit was performed in the patient. All necrotic tissues and nearly 100 larvae were removed. Then, the wound was closely observed for infections and possibly missed larvae. Within three days after surgery, there were still 3 larvae

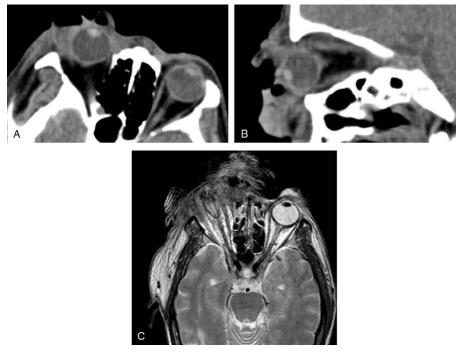


Figure 2. (A and B) An orbital computed tomography scan showed that right eyeball protrusion and periocular soft tissue edema. Two days later, (C) magnetic resonance imaging showed that the shape of the right eyeball was changed and the normal structure of the eyeball could not be identified.



Figure 3. The second week after orbit exenteration and frontal flap reconstruction.

crawling out of the orbit. On the ninth postoperative day, the defect was repaired via reconstruction with a pedicled musculocutaneous flap from the forehead region (Fig. 3). The patient recovered well postoperatively and was discharged uneventfully. During the 6-month follow-up period, the wound healed well, and the patient had no further complaints. Subsequently, he was lost to follow-up.



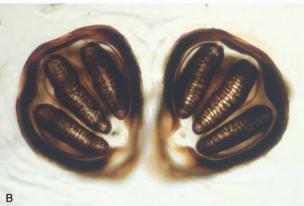


Figure 4. (A) Partial larvae extracted from necrotic tissue after surgery. (B) Posterior spiracle of the larva, 40× magnification.

The histopathological examination of the orbital contents revealed hyperplastic inflammatory granulation tissue, large areas of necrotic tissue and acute inflammatory exudates. The larvae were identified as the larvae of *Lucilia sericata* (Diptera: Calliphoridae) (Fig. 4A and B).

3. Literature review

A systematic literature review was performed through a search of the PubMed electronic database to identify all articles regarding human orbital myiasis published between January 1950 and December 2018. References from relevant articles were also included. The investigational strategy was based on an advanced search with the following terms: "orbit" or "orbital" and "myiasis." Only articles written in English were included.

Forty-six articles were found according to the abovementioned criteria, and their full text was assessed. Twenty-six reports of 27 patients with orbital myiasis were found. After screening the references, no articles needed to be added.

Finally, we identified 27 patients with orbital myiasis reported from 1950 to 2018 in the English-language literature (Tables 1 and 2).

4. Discussion

According to the literature review, orbital myiasis mainly affects elderly individuals, especially those whose ocular or periocular mucosal barrier is damaged, with causes including trauma, surgery, and ulcers. A rural background is also an important risk factor. The disorder is mainly manifested as inflammatory symptoms, in addition to the symptoms of preexisting diseases. Manual removal is useful for some patients, but half of the cases require surgery because of deep larval penetration and extensive destruction. Early use of ivermectin may avoid surgery, and even if not, such therapy can reduce the degree of damage and the scope of operative intervention.

4.1. Causes and risk factors

Diptera is a large order of insects with approximately 150,000 species in 10,000 genera and 150 families.^[7] According to the reported literature, there are 6 species of Diptera that cause orbital myiasis. The characteristics of these species are summarized in Table 3.

Cases of orbital myiasis reported between 1950 and 2018 were mainly in Asia and South America, especially India and Brazil. These places are consistent with the distribution of flies mentioned in Table 3. These sites are located in the tropics or subtropics with a warm and humid climate throughout the year and are suitable for the growth and reproduction of flies. Myiasis is a zoonotic disease that is more common in pastoral or rural areas than in urban areas, especially in developing countries lacking basic sanitation and with inadequate garbage disposal. With the increased mobility of people across nations and continents; however, infection may be seen in nonendemic areas.^[5]

In addition to a rural background, orbital myiasis is mainly associated with malignant tumors, especially skin cancer around the eyes. [8–18] Eye injury without proper medical care is a potential risk factor. [6,10,11,16,19–22] In addition, orbital myiasis is also associated with previous eye surgery, such as evisceration. [23] All of these factors have a commonality of ulcerative or necrotic tissues. Therefore, orbital myiasis is considered to be a special

(continued)

Date	Author	Age/sex	Geographical Iocation	Species	Number of larvae	Risk factors	Laterality	Vision of the involved eye	Symptoms	Treatment	Outcome
2019	Present study	72/M	China	Lucilia sericata	Nearly 100	Rural background, a eyelid trauma without treatment	00	Lost 1 yr ago	Repeated pain, crawling sensation, bleeding, larvae wriggling out from the eye	I.V. Antibiotics, orbital exenteration and graft repair, injection tetanus foxnid	Healed well
1967	Mathur et al ^[32]	65/M	India	Hypoderma bovis	21	Rural background	QO	Not mentioned	Pain due to "worms" in the eye	Turpentine, manual removal, I.V. Antibiotic	Healed well, perception of
1970	Gupta et al ^[31]	9/09	India	Unknown	Numerous	Rural background	Bilateral	Not mentioned	Pain and swelling in the nose and eyes, larval and systemic infection	Irrigated with chloroform turpent- ine, manual removal	Refused further treatment
1970	Wood et al ^[25]	49/F	United States	Calliphora vomitoria	About 200	Stuporous condition	Bilateral	No light perception	Semiconscious and hallucinating, maggots were found in all body	Irrigated with normal saline and atropine, manual	Died
1986	Kersten et al ⁽¹⁹⁾	65/M	Saudi Arabia	Chrysomya bezziana	Numerous	Rural background, a trauma without treatment, stuporous condition	SO	No light perception	Pain, orbital soft tissue swelling and proptosis, decreasing visual acuity, multiple cutaneous ulcerations and layal infestition.	I.V. Antibiotics, orbital exenteration	Healed well
1990	Agarwal et al ^[8]	70/F	India	Unknown	About 100	BCC	SO	Lost	Pain, bleeding, a brown, fleshy, ulcerated, foul smelling mass all around the left orbit, maggots were seen	Turpentine, irrigated with potassium permanganate solution, manual removal, I.V. Antibiotics and analoesirs	Healed well
1990	Sachdev et al ^[33]	80/F	India	Chrysomya bezziana	About 70	Endophthalmitis after Iens extraction	00	Lost 1 yr ago	An ulcer over the right upper lid with blood stained discharge, worms crawling in the ulcer	Turpentine, xylocaine, irrigated with hydrogen peroxide, manual removal. I.V. Antihiotics	Healed well
1999	Rocha et al ^[9]	54/M	Brazil	Cochliomyia hominivorax	More than 100	Rural background, BCC, alcoholism	00	No light perception	Pain and swelling in right orbital region, worms can be seen	Orbital exenteration	Healed well
2001	Baliga et al ^{ro]}	72/M	India	Oestrus ovis	About 100	Rural background, BCC, a trauma	SO	Not mentioned	Painful, burning, maggot infested ulcer mimicking basal cell carcinoma	Turpentine, ether, chloroform, I.V. Antibiotics, enucleation and graft repair	Healed well, no signs in 20 mo
2003	Caca et al ⁽¹¹⁾	85/F	Turkey	Hypoderma bovis	71	BCC, a trauma, a history of eyelid surgery	00	No light perception	A wound in right eye for over 1 yr, larvae in the same eye for 1 wk	Orbital exenteration, total maxillectomy and graft renair	Healed well
2003	Balasubramanya et al ^[26]	72/M	India	Cochliomyia sp	More than 300	Rural background, stuporous condition, right hemiparesis, hypertensive, diabetic, alcoholic, smoke	QO	Lost	Dirninished vision in both eyes, with pain, redness, and discharge; the right orbital contents were thickened, indurated and full of maggots	Irrigated with hydrogen peroxide, manual removal, I.V. Antibiotics	Died
2004	Devoto et al ^[37]	10/M	Argentina	Cochliomyia hominivorax	20	A history of enucleation due to right retinoblastoma	00	Lost 8 yr ago	The orbit was inflamed and painful, the implant largely exposed, magnotis were seen	Extracting the implant, manual removal	Healed well
2004	De Tarso et al ^[27]	80/M	Brazil	Cochliomyia hominivorax	Not mentioned	Rural background, alcoholic, diabetic, hypertensive, smoke	SO	No light perception	Pain and swelling in left orbital region, maggots were seen	Oral ivermectin, I.V. Antibiotics, enucleation	Healed well
2005	Costa et al ⁽³⁸⁾	55/M	Brazil	Cochliomyia hominivorax	Not mentioned	Advanced carcinoma of ethmoidal sinus	SO	No light perception	Intense pain in left orbital region, a crawling sensation, and maggots coming out of the nose	Oral ivermectin, I.V. Antibiotics, orbital exenteration	Healed well
2006	Osorio et al ^[12]	M/62	Colombia	Cochliomyia hominivorax	Not mentioned	Rural background, BCC	SO	Not mentioned	Purulent secretions coming from the left orbital area, maggots were seen	Oral ivermectin and antibiotics, enucleation	Died from Atrioventricular block
2006		W/92	Colombia	Cochliomyia hominivorax	Multiple	Rural background, BCC	SO	Not mentioned			Healed well

Attack of the part	100)	(continued).										
Fig. United States Unicovom A massive Scott Fig. Be light percection Scott S	Date	Author	Age/sex	Geographical location	Species	Number of larvae	Risk factors	Laterality	Vision of the involved eye	Symptoms	Treatment	Outcome
Figure 10 10 10 10 10 10 10 10		2								Swelling, erythema and pain in left orbital area, larvae coming out of the swollen area	Oral ivermectin and antibiotics, surgical debridement	
Signal or aligned Signal	2007	Jain et al ^[13]	65/F	United States	Unknown	A massive infestation	200	00	No light perception	Warmth, redness, oedema, decreased vision and restricted ocular motility OD, maggots were seen	Manual removal	Refused furti surgical interventi
Again Line of a signature of the control of	2009	Raina et al ^[14]	20/M	India	Family Calliphorid- ae	Multiple	Rural background, BCC	SO	Lost	A brown, flesty, ulcerated, and foul- smelling lesion, maggots were seen	Turpentine, xylocaine, manual removal, orbital exenteration and graft repair, I.V. Antibiotics and analoesics	Healed well
Victability of the contribution of the cont	5000	Abalo-Lojo et al ⁽¹⁵⁾	85/M	Spain	Lucilia sericata	About 70	BCC, mental illness, disoriented, dehydrated, cachectic, in poor hygienic condition	SO	Lost	Pain and burning in the left orbital region, worms crawling	Irrigated with diluted metronidazole, manual removal, I.V. Antibiotics and analgesics, orbital exenteration	Died
Kritationine et al ³⁴¹ 87F Iran Obsezione Open 150 Some lege of soul control Obt mentioned Leit cusier was gain and necrotical. Other describeration Other describeration Open 100 Run mentioned Observation No mentioned Integration in principle of an object of a principle of a principl	2010	Yeung et al ⁽¹⁶⁾	90/F	Hong Kong, China	Chrysomya bezziana	Numerous	SCC, a trauma, bed- bound, chronic obstructive airways disease, dementia	QO	Not mentioned	Blood-stained discharge coming from right eye, "worms" can be seen	Orbital exenteration	Died from systemic disease
Manya et al ²⁰¹ 1.5M india Wichitahria 35 A trauma of eyelid 05 Not mentioned Sweling, liching, foul smelling of the find many care al ²⁰¹ 1.5M india with many care at al ²⁰¹ 1.5M india with many care a	2011	Khataminia et al ^[4]	87/F	Iran	Chrysomya bezziana	Over 150	Some type of skin cancer, a history of eyelid surgery, bedbound	SO	Lost	Left ocular was pain and necrotized, with several live larvae	Orbital exenteration, injection tetanus toxoid	Healed well
Putthran et al ^[21] 70/F India Unknown Over 100 Rural background, a loss the eye had been or Pairrut, sweling of the right face, the removed 8 no ago presence of wormy creatures where seen removed 8 no ago presence of wormy creatures where seen removed 8 no ago presence of wormy creatures where seen removed a languiser and entropy of the right perception. Not mentioned Misra et al ^[22] 80/M India et al ^[23]	2012	Maurya et al ^[20]	1.5/M	India	Wohlfahrtia magnifica	35	A trauma of eyelid	SO	Not mentioned	Swelling, itching, foul smelling discharge, worms crawling	Turpentine, xylocaine, manual removal, topical, and I.V. Antibiotics	Healed well
Kamel et al ¹⁷¹ 24M India Family Calliphorid- are Not mentioned pignentiosa SCC, xeroderma pignentiosa OB (scharge, magods were seen analgasis, orbital per analgasis, orbit	2012	Puthran et al ^[21]	70/F	India	Unknown	Over 100	Rural background, a trauma, a history of right eye remove	QO	Lost, the eye had been removed 8 mo ago	Painful, swelling of the right face, the presence of wormy creatures	Topical antibiotics and ivermectin, manual removal, oral ivermectin, injection tetanus toxoid	Healed well
Misra et al ^[23] 80/M India Lucilia sericata Numerous Rural background, a o o or trauma without teatment teatm	2012	Kamal et al ^[17]	24/M	India	Family Calliphorid- ae	Not mentioned	SCC, xeroderma pigmentosa	SO	No light perception	Pain, blood tinged and Foul smelling discharge, maggots were seen	I.V. Antibiotics and analgesics, orbital exenteration	Healed late
Kalamkar et al ^[23] G5/F India Chrysomya 12 Rural background, a 0D Lost 10 yr ago the right upper eyelid, maggots manual removal, topical evisceration and transfer and swelling burlier sylocaine, and 1.1 Antibiotics, oral evisceration. Around 100 Rural background, 0S Lost Pandey et al ^[18] 73.5M Nepal Unknown Around 100 Rural background, oral surgical in left eyelid Raeley et al ^[8] 90/F India Obestrus ovis 27 A trauma of eyelid, 0S Not mentioned public in general control in the corresponding self-care and maggots wrigilion of public control in the corresponding son manual removal, robital removal, robital removal, robital removal, robital removal, robital removal, robital repair repair self-care and robital self-care repair public disconding son manual removal, robital removal rem	2013	Misra et al ^[22]	80/M	India	Lucilia sericata	Numerous	Rural background, a trauma without treatment	QO	Not mentioned	Pain and swelling of right upper lid, maggots were seen	Turpentine, xylocaine, manual removal, I.V. Antibiotics and analgesics	Healed well
Pandey et al ¹¹⁸ 73/M Nepal Unknown Around 100 Rural background, OS Lost Pain, itching, crawling sensation, foul- Manual removal, oral Hesperich Surgical in left eyelid smelling purulent discharge, analyseiscs and ivermectin, orbital eventeration and graft repair repair repair repair self-care and level alone, poor lateral canthus, expulsion of Antibiotics, injection pur and maggosts and tenderness oral ivermectin, I.V. over lateral canthus, expulsion of Antibiotics, injection pur and maggosts and tenderness oral ivermectin, I.V. over lateral canthus, expulsion of Antibiotics, injection purs and maggosts and tenderness oral vermeetin, I.V. over lateral canthus, expulsion of Antibiotics, injection purs and maggosts and tenderness oral vermectin, I.V. over lateral canthus, expulsion of Antibiotics, injection purs and maggosts and tenderness oral vermectin, I.V. over lateral canthus, expulsion of Antibiotics, injection purs and maggosts and tenderness oral vermectin, I.V. over lateral canthus, expulsion of Antibiotics, injection purs and maggosts and tenderness oral vermectin, I.V. over lateral canthus, expulsion of Antibiotics, injection purs and maggosts and tenderness oral vermectin, I.V. over lateral canthus, expulsion of Antibiotics, injection purs and maggosts and tenderness oral vermectin, I.V. over lateral canthus, expulsion of Antibiotics, injection purs and maggosts and tenderness oral vermectin, I.V. over lateral canthus, expulsion of Antibiotics, injection purs and maggosts and tenderness oral vermectin, I.V. over lateral canthus, expulsion of Antibiotics, injection purs and maggosts and tenderness oral vermectin, I.V. over lateral canthus, expulsion of Antibiotics, injection pursuance and tenderness oral vermecting and tendern	2016	Kalamkar et al ^[23]	65/F	India	Chrysomya bezziana	12	Rural background, a history of evisceration	QO	Lost 10 yr ago	Pain, redness, pruritis and swelling of the right upper eyelid, maggots were seen	Turpentine, xylocaine, manual removal, topical and I.Y. Antibiotics, oral ivermectin	Healed well
Kaeley et al ^{16]} 90/F India <i>Destrus ovi</i> s 27 A trauma of eyelid, OS Not mentioned Left upper lid swelled up, peri-orbital Manual removal, topical and He. puffiness, redness, and tenderness oral ivermectin, I.V. self-care self-care training to an injection pus and maggots tetanus toxold	2016	Pandey et al ^[18]	73/M	Nepal	Unknown	Around 100	Rural background, BCC, a history of surgical in left eyelid	SO	Lost	Pain, ltching, crawling sensation, foul- smelling purulent discharge, maggots wriggling out	Manual removal, oral antibiotics, analgesics and ivermectin, orbital exenteration and graft repair	Healed well
	2017	Kaeley et al ^[6]	90/F	India	Destrus ovis	27	A trauma of eyelid, lived alone, poor self-care	SO	Not mentioned	Left upper lid swelled up, peri-orbital puffiness, redness, and tenderness over lateral canthus, expulsion of pus and maggots	Manual removal, topical and oral ivermectin, I.V. Antibiotics, injection tetanus toxoid	Healed well, restoration vision to I perception

BCC=basal cell carcinoma, F=female, I.V.=intravenous, M=male, OD=right eye, OS=left eye, SCC=squamous cell carcinoma.

Table 2

Summary of the 28 cases with orbital myiasis.

Characteristics	N (%)
Sex (n = 28)	
Male	17 (61)
Female	11 (39)
Age, $yr (n=28)$	
< 65 yr old	8 (29)
≥ 65 yr old	20 (71)
Species (n=23)	
Family Calliphoridae	18 (64)
Cochliomyia hominivorax	7 (25)
Chrysomya bezziana	5 (18)
Lucilia sericata	3 (11)
Calliphora vomitoria	1 (4)
Family <i>Oestridae</i>	4 (14)
Hypoderma bovis	2 (7)
Oestrus Ovis	2 (7)
Family Sarcophagidae	1 (4)
Wohlfahrtia magnifica	1 (4)
Geographical location (n=28)	10 (40)
India	13 (46)
Brazil	3 (11)
China Colombia	2 (7)
America	2 (7)
	2 (7)
Argentina Iran	1 (4) 1 (4)
Nepal	1 (4)
Saudi Arabia	1 (4)
Spain	1 (4)
Turkey	1 (4)
Risk factors (n=28)	. (.)
Advanced age (age ≥65 yr old)	20 (71)
Rural background	15 (54)
Cancer	14 (50)
Basal cell carcinoma	8 (29)
Squamous cell carcinoma	4 (14)
Some type of skin cancer	1 (4)
Advanced carcinoma of ethmoidal sinus	1 (4)
History of eye trauma	9 (32)
History of eye surgery	7 (25)
Stuporous condition	4 (14)
Alcoholism	3 (11)
Bed-bound	2 (7)
Treatment $(n=28)$	
Manual removal	16 (57)
Surgery	14 (50)
Enucleation	3 (11)
Exenteration	11 (39)
Turpentine	9 (32)
Ivermectin	8 (29)
Antibiotics	20 (71)
Tetanus toxoid	4 (14)
Outcome (n=26)	
Perception of light	2 (7)
Healed well	21 (75)
Died	5 (18)

The present case has been included in this table.

kind of wound myiasis to some extent.^[24] Other risk factors include a stuporous condition, ^[15,19,25,26] alcoholism, ^[9,26,27] and a bed-bound status. ^[4,16] Fetid conditions, including excessively long periods of sleep, poor self-care and poor hygiene, may attract flies. Yeung et al^[16] reported a case of orbital myiasis

occurring in a nursing home. Numerous cases of hospital-acquired myiasis have been reported, [28–30] although these infestations did not take place in the orbital area. The occurrence of these cases indicates that nosocomial infections must be monitored carefully. It is worth mentioning that advanced age is an important risk factor, as orbital myiasis is more common in middle-aged and older patients.

4.2. Clinical manifestations

Orbital myiasis is commonly present on only 1 side. It rarely involves bilateral sites. The 2 reported bilateral cases took place in patients who had a systemic infection and were semiconscious. [25,31] The number of infected larvae is usually large (dozens, or even hundreds), and this often indicates a pessimistic prognosis. In addition to the symptoms of tumor ulcers and traumatic wounds from preexisting conditions, the principal clinical manifestations are inflammatory symptoms of redness, swelling, heat, and pain caused by underlying diseases or larvae infection. The itching and crawling sensations caused by the larvae's peristalsis are also common complaints. The examination of most cases shows no light perception, perforation of the globe, and even destruction of the orbit in the diseased eyes. Maggots are seen in the orbital cavity.

4.3. Treatments and outcomes

When large numbers of maggots invade deeply into the orbit, they rapidly destroy the eyeball and orbital tissue and then invade the orbital bone, paranasal sinuses, and intracranial tissue. The invasion of the orbital apex most easily penetrates into the brain, causing fatal results. [26] Therefore, early identification and treatment are essential. The treatment principle is complete removal of larvae. Manual removal and surgery are available procedures that should be chosen according to the degree of larval invasion and the extent of tissue destruction, both of which can be used if necessary.

Manual removal should be utilized in the management of less extensive orbital myiasis. Maggots exhibit negative phototaxis. They penetrate deep into tissues to avoid light. [23] Since they can firmly clamp onto tissues by their hook-like structure, forceful removal may result in incomplete extractions leading to an inflammatory response, granuloma formation, and calcification. [21] Therefore, suffocating agents and anesthetic agents are recommended for use before manual removal to make the process easier. Suffocating agents, including turpentine oil, [8,10,14,20,22,23,31–33] petroleum jelly, [4,23] and liquid paraffin, [4,23] can block the larval breathing holes, forcing the aerobic maggots to migrate to the surface for air. It is worth noting that this method may also cause larval death due to asphyxiation in the tissues. Topical administration of anesthetic agents such as xylocaine [14,20,22,23,33] and cocaine [19] can paralyze the larvae and prevent them from penetrating deeper.

Ivermectin, a broad-spectrum antiparasitic drug, is considered a safe and noninvasive means of managing orbital myiasis. ^[6] The drug promotes the spontaneous emergence of larvae from deep in the orbital tissue and prevents the larvae from causing greater damage, thereby avoiding surgery and reducing the risk of death. Some authors believe that albendazole, butazolidin, and thiabendazole can also play the same role. ^[11,23]

Once the globe is penetrated, enucleation, and even exenteration, is inevitable. In cases where invasion is confined to the globe,

Table 3

Characteristics of the species reported to cause orbital myiasis.

Species	Common name	Family	Classification	Geographic distribution
Lucilia sericata	Green bottle blowfly, Sheep strike blowfly	Calliphoridae	Facultative*	Europe and North America
Calliphora vomitoria	Blue blowfly	Calliphoridae	Facultative	India, Japan, Central America, and Southern America
Chrysomya bezziana	Old World screwworm	Calliphoridae	Obligatory*	Tropical Africa, Asia, Papua New Guinea, and India
Cochliomyia hominivorax	New World screwworm	Calliphoridae	Obligatory	Central and South America
Hypoderma bovis	Cattle bot fly	Oestridae	Obligatory	All continents in the Northern Hemisphere
Oestrus ovis	Sheep nasal bot fly	Oestridae	Obligatory	Mediterranean countries, Southern Africa, Central America
Wohlfahrtia magnifica	Wohlfahrt wound myiasis fly	Sarcophagidae	Obligatory	Parts of Europe, Russia, China, the Middle East, and northern Africa

^{*} Based on the degree of host-parasite relationship, myiasis has been classified into three categories⁽³⁹⁾: (1) obligatory, the parasites depend on the host to complete their life cycle, they need living hosts and can penetrate through the intact skin or mucosa. (2) facultative, the parasites usually live freely in nature, but can invade the hosts initiate myiasis under certain circumstances. (3) accidental, the parasites usually live freely on dead and decaying organic matter, but when pregnant female flies accidentally in contact with the hosts initiate myiasis.

enucleation can effectively control the infection. [10,12,27] In cases in which invasion approaches the orbital apex or there is extensive destruction of ocular tissue, exenteration needs to be seriously taken into consideration. In our case, the patient's orbital tissues, including the eyeball, were destroyed by the larvae, and thus, exenteration was inevitable.

Since flies are biologic or mechanical vectors of protozoal, viral, bacterial or helminthic diseases, [11] myiasis may be accompanied by a bacterial infection. Antibiotics and tetanus toxoid therapy are utilized to prevent this complication. However, Puthran et al [21] and Kaeley et al [6] believed that systemic antibiotics were unnecessary because of the antibacterial activity of maggots. Many studies have demonstrated the potent antibacterial activity of larval excretions and secretions of L sericata and Lucilia cuprina against bacteria. [34–36] However, it is unclear whether other species have this antibacterial activity.

Inflammatory symptoms are relieved after removal of the larvae. Timely treatment can partially restore vision in patients with an intact eyeball. [6] However, the deep invasion of a large number of larvae can destroy the eyeball within a few days, leading to blindness. [27,33] Death occurs in patients who have systemic diseases or are in a stuporous condition at presentation. [12,15,16,25,26] All reported cases had no extraocular involvement.

4.4. Preventions

Orbital myiasis can be avoided through proper precautions. Popularizing knowledge is essential. The hazards of flies should be emphasized where flies are prevalent, so that not only myiasis but also other diseases transmitted by flies are prevented. Timely removal of garbage, spoilage plants, and animal carcasses in the environment, as well as a sterile insect technique, can notably reduce fly populations. Adequate nursing home and hospital sanitation and personal hygiene are crucial. Since female flies are strongly attracted by blood and secretions, [9,11,13] accumulated secretions should be cleansed from the eviscerated socket daily; eye trauma requires timely and appropriate medical care, and long-lasting ulcers need medical attention. Moreover, mosquito netting, insect repellents, and insecticides can prevent skin contact.

5. Conclusion

Orbital myiasis, a rapidly developing and highly destructive ocular parasitosis, is predominantly caused by the larvae of

family Calliphoridae flies, which are prevalent mainly in tropical and subtropical regions. Skin cancer around the eye, untreated eye trauma, and a history of eye surgery are important factors predisposing to orbital myiasis. Advanced age and a rural background are also common risk factors. Timely removal of all larvae can prevent deep larval penetration and protect the eyeball.

Author contributions

Conceptualization: Lu Liu, Hao Liang, Jian He, Jun Chen, Qiao-Wen Liang, Zhi-Yuan Jiang, Yi Du.

Data curation: Yan-Ling Huang, Lu Liu, Hao Liang.

Formal analysis: Yan-Ling Huang.

Funding acquisition: Yi Du.

Methodology: Jian He, Jun Chen, Qiao-Wen Liang, Zhi-Yuan Jiang, Yi Du.

Project administration: Yi Du. Resources: Lu Liu, Hao Liang.

Supervision: Hao Liang, Jian-Feng He, Min-Li Huang, Yi Du. Writing – original draft: Yan-Ling Huang, Lu Liu, Jian He, Jun Chen, Qiao-Wen Liang, Yi Du.

Writing – review & editing: Zhi-Yuan Jiang, Jian-Feng He, Min-Li Huang, Yi Du.

Yi Du orcid: 0000-0003-4879-5159.

References

- [1] Bhola N, Jadhav A, Borle R, et al. Primary oral myiasis: a case report. Case Rep Dent 2012;2012;734234.
- [2] Carvalho RW, Santos TS, Antunes AA, et al. Oral and maxillofacial myiasis associated with epidermoid carcinoma: a case report. J Oral Sci 2008: 50:103-5
- [3] Ozyol P, Ozyol E, Sankur F. External ophthalmomyiasis: a case series and review of ophthalmomyiasis in Turkey. Int Ophthalmol 2016;36:887–91.
- [4] Khataminia G, Aghajanzadeh R, Vazirianzadeh B, et al. Orbital myiasis. J Ophthalmic Vis Res 2011;6:199–203.
- [5] Padhi TR, Das S, Sharma S, et al. Ocular parasitoses: a comprehensive review. Surv Ophthalmol 2017;62:161–89.
- [6] Kaeley N, Kaushik RM, Rajput R, et al. Orbital myiasis with scalp pediculosis and buccal abscess-an uncommon presentation. J Clin Diagn Res 2017;11:OD01–2.
- [7] Francesconi F, Lupi O. Myiasis. Clin Microbiol Rev 2012;25:79-105.
- [8] Agarwal DC, Singh B. Orbital myiasis—a case report. Indian J Ophthalmol 1990;38:187–8.
- [9] Rocha EM, Yvanoff JL, Silva LM, et al. Massive orbital myiasis infestation. Arch Ophthalmol 1999;117:1436–7.
- [10] Baliga MJ, Davis P, Rai P, et al. Orbital myiasis: a case report. Int J Oral Maxillofac Surg 2001;30:83–4.

- [11] Caca I, Unlu K, Cakmak SS, et al. Orbital myiasis: case report. Jpn J Ophthalmol 2003;47:412–4.
- [12] Osorio J, Moncada L, Molano A, et al. Role of ivermectin in the treatment of severe orbital myiasis due to Cochliomyia hominivorax. Clin Infect Dis 2006;43:e57–9.
- [13] Jain A, Desai RU, Ehrlich J. Fulminant orbital myiasis in the developed world. Br J Ophthalmol 2007;91:1565–6.
- [14] Raina UK, Gupta M, Kumar V, et al. Orbital myiasis in a case of invasive basal cell carcinoma. Oman J Ophthalmol 2009;2:41–2.
- [15] Abalo-Lojo JM, Lopez-Valladares MJ, Llovo J, et al. Palpebro-orbital myiasis in a patient with basal cell carcinoma. Eur J Ophthalmol 2009;19:683–5.
- [16] Yeung JC, Chung CF, Lai JS. Orbital myiasis complicating squamous cell carcinoma of eyelid. Hong Kong Med J 2010;16:63–5.
- [17] Kamal S, Bodh SA, Kumar S, et al. Orbital myiasis complicating squamous cell carcinoma in xeroderma pigmentosum. Orbit 2012; 31:137–9.
- [18] Pandey TR, Shrestha GB, Sitaula RK, et al. A Case of orbital myiasis in recurrent eyelid basal cell carcinoma invasive into the orbit. Case Rep Ophthalmol Med 2016;2016:2904346.
- [19] Kersten RC, Shoukrey NM, Tabbara KF. Orbital myiasis. Ophthalmology 1986;93:1228–32.
- [20] Maurya RP, Mishra D, Bhushan P, et al. Orbital myiasis: due to invasion of larvae of flesh fly (Wohlfahrtia magnifica) in a child; rare presentation. Case Rep Ophthalmol Med 2012;2012:371498.
- [21] Puthran N, Hegde V, Anupama B, et al. Ivermectin treatment for massive orbital myiasis in an empty socket with concomitant scalp pediculosis. Indian J Ophthalmol 2012;60:225–7.
- [22] Misra N, Gogri P, Misra S, et al. Orbital myiasis caused by green bottle fly. Australas Med J 2013;6:504–6.
- [23] Kalamkar C, Radke N, Mukherjee A. Orbital myiasis in eviscerated socket and review of literature. BMJ Case Rep 2016;2016: bcr2016215361.
- [24] Hall MJ, Wall RL, Stevens JR. Traumatic myiasis: a neglected disease in a changing world. Ann Rev Entomol 2016;61:159–76.
- [25] Wood TR, Slight JR. Bilateral orbital myiasis. Report of a case. Arch Ophthalmol 1970;84:692–3.
- [26] Balasubramanya R, Pushker N, Bajaj MS, et al. Massive orbital and ocular invasion in ophthalmomyiasis. Can J Ophthalmol 2003;38:297–8.

- [27] De Tarso P, Pierre-Filho P, Minguini N, et al. Use of ivermectin in the treatment of orbital myiasis caused by Cochliomyia hominivorax. Scand J Infect Dis 2004;36:503–5.
- [28] Hira PR, Assad RM, Okasha G, et al. Myiasis in Kuwait: nosocomial infections caused by lucilia sericata and Megaselia scalaris. Am J Trop Med Hyg 2004;70:386–9.
- [29] Jang M, Ryu SM, Kwon SC, et al. A case of oral myiasis caused by Lucilia sericata (Diptera: Calliphoridae) in Korea. Korean J Parasitol 2013; 51:119–23.
- [30] Mircheraghi SF, Mircheraghi SF, Ramezani Awal Riabi H, et al. Nasal nosocomial myiasis infection caused by Chrysomya bezziana (Diptera: Calliphoridae) following the septicemia: a case report. Iran J Parasitol 2016:11:284–9.
- [31] Gupta SK, Nema HV. Rhino-orbital-myiasis. J Laryngol Otol 1970; 84:453–5.
- [32] Mathur SP, Makhija JM. Invasion of the orbit by maggots. Br J Ophthalmol 1967;51:406–7.
- [33] Sachdev MS, Kumar H, Roop, et al. Destructive ocular myiasis in a noncompromised host. Indian J Ophthalmol 1990;38:184–6.
- [34] Teh CH, Nazni WA, Lee HL, et al. In vitro antibacterial activity and physicochemical properties of a crude methanol extract of the larvae of the blow fly Lucilia cuprina. Med Vet Entomol 2013;27: 414–20.
- [35] Teh CH, Nazni WA, Nurulhusna AH, et al. Determination of antibacterial activity and minimum inhibitory concentration of larval extract of fly via resazurin-based turbidometric assay. BMC Microbiol 2017;17:36.
- [36] Valachova I, Takac P, Majtan J. Midgut lysozymes of Lucilia sericatanew antimicrobials involved in maggot debridement therapy. Insect Mol Biol 2014;23:779–87.
- [37] Devoto MH, Zaffaroni MC. Orbital myiasis in a patient with a chronically exposed hydroxyapatite implant. Ophthalmic Plast Reconstr Surg 2004;20:395–6.
- [38] Costa DC, Pierre-Filho Pde T, Medina FM, et al. Use of oral ivermectin in a patient with destructive rhino-orbital myiasis. Eye (Lond) 2005; 19:1018–20.
- [39] Tomy RM, Prabhu PB. Ophthalmomyiasis externa by Musca domestica in a case of orbital metastasis. Indian J Ophthalmol 2013;61:671–3.