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Review article

Human urogenital myiasis: A systematic review of reported cases from 1975 to 2017



Roghiyeh Faridnia^a, Masoud Soosaraei^a, Hamed Kalani^b, Mahdi Fakhar^{c,*},
Pikka Jokelainen^{d,e,f}, Reza Zolfaghari Emameh^g, Elham Sadat Banimostafavi^h,
Hajar Ziaei Hezarjaribi^c

^a Student Research Committee, Mazandaran University of Medical Sciences, Sari, Iran

^b Infectious Diseases Research Centre, Golestan University of Medical Sciences, Gorgan, Iran

^c Toxoplasmosis Research Center, Department of Parasitology, School of Medicine, Mazandaran University of Medical Sciences, Sari, Iran

^d Laboratory of Parasitology, Department of Bacteria, Parasites and Fungi, Infectious Disease Preparedness, Statens Serum Institute, Copenhagen, Denmark

^e University of Helsinki, Helsinki, Finland

^f Estonian University of Life Sciences, Tartu, Estonia

^g Department of Energy and Environmental Biotechnology, Division of Industrial and Environmental Biotechnology, National Institute of Genetic Engineering and Biotechnology, Tehran, Iran

^h Department of Radiology, Imam Khomeini Hospital, School of Medicine, Mazandaran University of Medical Sciences, Sari, Iran

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ABSTRACT

The public health importance of myiasis [infestation with dipterous (fly) larvae] remains unknown. This disease is spread worldwide in animals and humans, but baseline data on its prevalence are limited. In particular, knowledge on human urogenital myiasis (UGM) is scattered. As such, a systematic search was undertaken of five English and five Persian databases for publications describing UGM cases in English or Persian published between 1975 and 2017. In total, 45 papers reporting 59 UGM cases from various regions of the world are included in this review. All included papers were from the English databases. The age of patients ranged from 5 to 89 years, and the mean age was 40.6 years. Thirty-six of the patients were female and 19 were male. The highest number of cases ($n = 12$) was reported from Brazil. The most common genera causing UGM were *Psychoda* spp. (23.7%) and *Cochliomyia* spp. (11.8%). The vagina was the most commonly reported anatomical location of UGM for women, and the urogenital tract was the most commonly reported location for men. Thirteen cases were reported from rural areas and eight cases from urban areas; the location of other cases was not specified. The incidence of UGM is likely to be substantially underestimated when evaluated based on published case reports. Epidemiological studies, such as questionnaires to medical doctors, could help to gather the necessary baseline data on the occurrence of UGM.

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* Corresponding author at: Toxoplasmosis Research Center, School of Medicine, Department of Parasitology, Mazandaran University of Medical Sciences, Farah-Abad Road, P.O. Box 48471-91971, Sari, Iran.

E-mail address: mahdif53@yahoo.com (M. Fakhar).

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Introduction

Myiasis is a parasitic disease caused by the presence of dipterous (fly) larvae in living or dead tissues of live vertebrates (i.e. animals and humans). Entry of the larvae occurs through skin wounds or body cavities, such as the mouth, ears, eyes and urogenital tract. The larvae are able to pierce and penetrate both healthy and necrotic tissues; therefore, tetanus and secondary infection may occur as complications of myiasis [1,2]. In humans, the infestation is endemic in some areas and travel-related in other areas [2]. The disease is associated with socio-economically poor regions, certain cultural habits, and favourable weather conditions for flies [2]. The reported predisposing risk factors for human myiasis include poor hygiene, advanced age, diabetes, psychiatric illness, rural background, low socio-economic status and homelessness [2,3].

Based on the parts of the body involved, myiasis is classified into different types, including cutaneous, subcutaneous, nasopharyngeal, intestinal and urogenital (UGM) [2,3]. Based on the relationship between the host and the larvae, myiasis can also be classified into obligatory, facultative and accidental myiasis. Several fly families, genera and species can cause myiasis [2,4].

Symptoms of UGM vary according to the organ involved and the severity of infection [5,6]. The only definitive treatment of UGM is to remove the larvae, and in such cases, the symptoms will disappear thereafter [7]. Consequently, correct diagnosis is necessary in order to avoid unnecessary treatment [8].

Currently, the real burden of myiasis is unknown, and baseline data on its prevalence are limited and scattered. For this reason, UGM can be considered as a neglected disease. Moreover, UGM diagnosis may be confused because no specific symptoms associated with this infestation are known, so awareness of this infestation is of major importance in order to be reported correctly by physicians. As such, a systematic review of publications reporting cases of UGM was undertaken to clarify the latest status of this disease in the literature.

Materials and methods

Study design

This systematic review was not preregistered. The search process, screening and selection was systematically designed and performed as follows.

Information sources and search strategy

Ten databases were included as information sources in the search for reports on UGM. Five were English databases (PubMed, Google Scholar, ScienceDirect, Scopus and Web of Science) and five were Persian databases [Magiran, Irandoc, ELM net, Barakat Knowledge Network System (formerly Iran medex) and Scientific Information Database]. No further searches (e.g. from reference lists of the papers) were performed.

The search was performed using the following terms: myiasis, screwworm, maggot, human, urogenital, genitalia, vulva, vagina, vulvovaginal, penis, penile, urethra and urinary tract, in combination with each other. Any written papers in English or Persian, published between 1975 and 2017 (42 years), reporting case(s) of UGM were considered for screening, regardless of study design.

Inclusion of publications and data collection

In total, 361 records were identified, 96 of which were duplicates (Fig. 1). Papers reporting the same findings and papers that were not about humans were also excluded. Four reviewers (RF, MS, HK and HZH) screened the remaining papers independently for inclusion in the review. Disagreements were resolved by MF.

The following data were extracted from the papers included in the review: year of publication, first author, age and sex of patient (s), country, rural or urban place of residence, organ involved, and species of fly. There was no contact with the authors of the papers. Risk of bias of individual studies was not formally assessed; publication bias was expected.

Statistical analysis

Data were analysed using SPSS v16 (IBM Corp., Armonk, NY, USA) and presented as proportions (%). Moreover, Chi-squared test and two-tailed *t*-test were utilized to compare proportions in each group, and $p < 0.05$ was considered to indicate significance.

Results

Overall, from the 10 databases searched, 45 papers were included in the review (Fig. 1) [5–49]. All 45 papers were from the English databases. All the papers were UGM case reports or UGM case series, and reported a total of 59 cases of UGM from various regions of the world (Appendix 1 in supplementary material).

Analysis of the cases based on the extracted data is shown in Table 1. The age range of patients was 5–89 years old, and the mean age was 40.6 years. Nine patients were under 18 years of age. Thirty-six females and 19 males were included, and the patient's sex was not reported in four studies. Of the 59 cases, most were from Asian countries ($n = 19$, 10 of which were from India), followed by American countries ($n = 16$, 12 of which were from Brazil), European countries ($n = 13$ cases) and African countries ($n = 8$). The place of residence was reported for 21 patients: 13 patients lived in rural areas and eight patients lived in urban areas. The most frequently reported common fly species causing UGM were *Psychoda* spp. (23.7%) and *Cochliomyia* spp. (11.8%) (Appendix 2 in supplementary material). The vagina was the most commonly reported anatomical location of UGM for women, and the urogenital tract was the most commonly reported location for men. The infestation rate was higher in women (61%) than men (32.2%).

Discussion

Publications reporting UGM are few in number and unevenly distributed geographically. Between 1975 and 2017, cases of UGM were mainly reported from Asia and South America, particularly from India and Brazil. These and other tropical and subtropical regions are considered typical for myiasis, whereas the disease is rare and considered as travel-related in developed countries with good healthcare systems, especially in Central Europe [2]. However, the Brazilian and Indian healthcare systems are more active to report interesting clinical findings [50]. Considering the reviewed studies, physicians are not usually expert to identify myiasis because cases of this disease are rare and the symptoms are not specific. Infestations are reported from both developed and

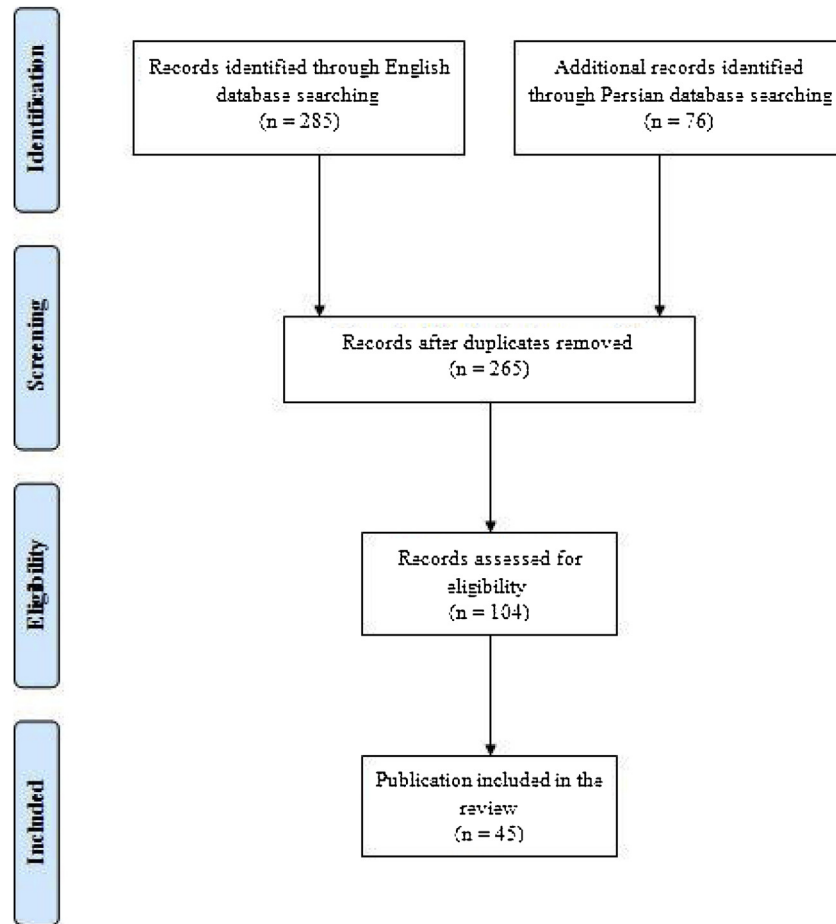


Fig. 1. PRISMA flow diagram showing the selected articles.

Table 1
Analysis of human urogenital myiasis according to the extracted variables.

p-value	n (%)	Category	Variable
<0.05	6 (10.1)	1975–1995	Year
	53 (89.8)	1996–2017	
>0.05	15 (25.4)	≤20	Age (years)
	16 (27.1)	>20 ≤ 40	
	8 (13.5)	>40 ≤ 60	
	12 (20.3)	>60 ≤ 80	
	4 (6.7)	>80	
<0.05	4 (6.7)	NR	Gender
	36 (61)	Female	
>0.05	19 (32.2)	Male	Country
	4 (6.7)	NR	
	12 (20.3)	Brazil	
	10 (16.9)	India	
	7 (11.8)	Turkey	
>0.05	7 (11.8)	Egypt	Living area (urban/rural)
	5 (8.4)	Iran	
	15 (25.4)	Other	
	3 (5)	NR	
	8 (13.5)	Urban	
>0.05	13 (22)	Rural	Organ involved
	38 (64.4)	NR	
	20 (33.8)	Urogenital	
	11 (18.6)	Vagina	
	5 (8.4)	Labia	
>0.05	8 (13.5)	Urethra	Other
	6 (10.1)	Vulva	
	4 (6.7)	Vulvovagina	
	7 (11.8)	Other	

NR, not reported.

developing countries, as low-income individuals are also susceptible to infestation in developed countries [49].

The clinical symptoms of UGM are variable; some patients remain asymptomatic and others display severe symptoms. General symptoms include abdominal pain, nausea, itching, rectal bleeding, vomiting and side pain [5]; and specific symptoms include dysuria, pollakiuria, haematuria, and larvae in urine [51]. Urethral involvement as an obstacle to urine flow has been reported in both women and men [10,12]. Involvement of the external parts of the female genitalia, especially the lamina minora and lamina majora, is often accompanied by symptoms such as tenderness, erythema and inflammation [17,19,33]. The symptoms of UGM are very variable when the vagina is involved, leading to misdiagnosis, and often include the aforementioned general symptoms [6]. Such symptoms may also be observed when the internal parts of the penis are involved [18]. When the glans penis is involved, lesions may be observed as ulcers [24].

Myiasis is often mistakenly diagnosed because the disease is very rare and its symptoms are not specific [7], and incorrect diagnosis can lead to unnecessary treatment [8]. UGM can be misdiagnosed as an obstructing ureteral stone [44]. Endoscopy can be used to diagnose and treat internal UGM [2]. Although biochemical examination is not beneficial for diagnosis, microhaematuria, albuminuria and leukocyturia may be observed in patients with myiasis [6,39]. According to the reviewed articles, the best way to diagnose cases where the larvae are not visible is to take a history from the patient [2].

Treatment of UGM varies according to the localization of larvae and the severity of the symptoms. There is no specific treatment as

larvae are insensitive to drugs. Often, initial treatment with antibiotics is useless due to misdiagnosis [24]. Mechanical removal of larvae, if they can be reached, is the best and only definitive treatment of UGM, and the use of antibiotics with anti-inflammatory drugs, if the symptoms are severe, is a therapeutic option [2,3,15,52]. In some patients, additional treatment may also be needed [53].

This review found no significant difference between rural and urban areas ($p > 0.05$); however, myiasis occurs mainly in rural areas, and there is an indirect relationship between self-perceived poor health, education level and myiasis [37]. Myiasis is more common in individuals with mental health problems, diabetes, immunodeficiency and low socio-economic status [24]. When parts of the urogenital system are exposed to flies, they are attracted to lay eggs or larvae in the genital cavities due to the unpleasant odour. Furthermore, individual habits, such as lying on the ground naked and poor hygiene, increase the risk of myiasis [54].

This review also found that infestation rates in women were higher than in men, and the most common organ involved in women was the vagina. Genital myiasis may occur through fly eggs transferring through dusty clothes to the vulva or glans penis [24]. It has been shown previously that some villagers dry washed their clothes on the ground, and flies lay eggs in the material leading to larvae attacking the host [33]. In addition, flies that lay eggs in genital cavities may have been attracted by the scent brought about by poor cleanliness and genital co-infections, and individuals that do not wear underwear outside or after or during intercourse may also have more infections [36].

Symptoms will be relieved after removing the maggots. In some cases, there are secondary bacterial and or *Trichomonas vaginalis* infections that should be treated with broad-spectrum antimicrobial agents [55].

This systematic review indicates that UGM is common in several tropical countries, mainly in Asia and South America. The authors believe that UGM cases worldwide are more common than the literature suggests, as there are likely to have been cases that have not been reported by physicians. In addition, in some of the articles reviewed in the current study, the genera of the larvae were not identified, and this important issue should be considered by researchers and physicians. As a whole, regular monitoring of individuals is required to improve public health education, particularly personal health care and environmental sanitation in regions with a high incidence of infestations, which can reduce sociopsychiatric obstacles caused by UGM in the community.

Conclusion

This systematic review found that UGM has been observed in several countries, mainly from Asia and South America. The incidence of UGM is likely to be substantially underestimated when evaluated based on published case reports.

Conflict of interest

None declared.

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Appendix A. Supplementary data

Supplementary material related to this article can be found, in the online version, at doi:<https://doi.org/10.1016/j.ejogrb.2019.02.008>.

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