

Fly Larvae Induce Urinary Myiasis with no Urinary Symptoms but with Neurological Compromise

Retracted

Abstract

Objective: A 13-year-old patient presented at the pediatric intensive care unit with confusion, disorientation, bradypsychia, bradilalia, hallucinations and delusions. To address her condition, she was promptly induced into a barbiturate coma and assessed for bacterial or viral infection.

Methods: The corresponding laboratory studies carried out in accordance with the clinical practice guidelines ruled out serious bacterial disease, the CSF was compatible with viral encephalitis and no viral infection was detected. The patient did not show changes suggestive of myiasis, but she stopped urinating and bladder catheterization revealed fly larvae in the urine.

Results: The results of the taxonomic analysis showed that the larvae belonged to the fly species *Clogmia albipunctata*. Like this, the result of the diagnosis was myiasis induced by parasitic infestations.

Conclusions: The antiparasitic treatment was successful and the patient was discharged.

Keywords: Myiasis; Bladder; Larva; *Telmatoscopus*; *Albipunctata*

Introduction

Myiasis are parasitic infestations in humans or animals by fly larvae that infest skin, necrotic tissues and natural cavities [1-3]. This term was first coined in 1840 by Hope, who proposed the term "myiasis" (from the Greek myia: Fly) to describe the infestation by larval stages of various species of flies [4]. Myiasis is classified according to the location of the larvae in the human body and is generally defined as cutaneous, cavitory and/or intestinal [5]. In humans, various factors predisposing to infestation are required, including previous injuries, such as wounds, recent surgical episodes, basal cell carcinomas, lipedema, poor hygiene, immune-deficiencies, close coexistence

of humans with animals, malnutrition, poor lifestyle habits, population and climate [6,7].

The *Clogmia albipunctata* fly, also named *Telmatoscopus albipunctata*, is a primitive *Nematocera* of the *Psychodidae* family, *Psychodinae* subfamily, with cosmopolitan distribution [8]. Adult *Clogmia albipunctata* insects are of minor medical importance, having been implicated in cases of inhalant allergy in South Africa [9] and as a possible vector mechanism for bacterial pathogens associated with nosocomial infections in infested German hospitals [9,10]. However, *Clogmia albipunctata* larvae have been reported as a cause of human nasopharyngeal myiasis [8,11], testicular myiasis and urinary myiasis [12,13].

Although urinary myiasis of any type is rare, it can occur in all parts of the world, but its prevalence is higher in tropical and subtropical areas, particularly in warm climates [14]. Therefore, myiasis is reported more frequently in developing countries. As far as we know, there are only four reported cases worldwide [15-18]. However, its incidence is probably underestimated since most cases occur in rural areas [19,20]. Because urinary tract invasion tends to occur primarily through inoculation with contaminated water sources, it affects women more than men [1-3]. To our knowledge, this is the first near-fatal case of urinary myiasis in a healthy, immunocompetent and non-instrumented woman in Mexico.

Case Presentation

A 13-year-old girl belonging to a Scout group visited the region of Tepoztlán, Mexico, where she swam in a river in the state of Cuernavaca. Two weeks later, she was admitted to the emergency department due to sleepiness after 4 days of inactivity without eating. She was found incoherent and disoriented, with bradypsychia and bradialia, hallucinations and delirium. When admitted to the (PICU) Pediatric Intensive Care Unit, she had a Glasgow score of 14, increased deep tendon reflexes and positive bilateral Babinsky. Laboratory tests ruled out serious bacterial disease and the CSF was compatible with viral encephalitis. PCR for enterovirus, adenovirus and HSV-1, HSV-2, HSV-6 was negative. Magnetic resonance reported extra-axial leptomeningeal enhancement, a finding compatible with meningitis. Diffusely slow EEG was observed without irritant activity. Supportive treatment improved the neurological outcome. Since there are no previous pathologies of this nature associated with myiasis, none of these symptoms led to suspect that the cause was a urinary tract infection by fly larvae.

Investigation and Treatment

Follow-up of the patient in the PICU required the implantation of a bladder catheter. Replacement of this catheter on the second day of hospitalization revealed the presence of abundant larvae. Urine analysis showed macroscopically visible pupae. The sample was sent to the "Instituto Nacional de Ciencias Médicas y Nutrición Salvador Zubirán" where the larvae were identified as *Clogmia albipunctata*. A cystoscopic evaluation in the operating room allowed visualization of two larvae. Bladder cleansing was performed and biopsies were obtained from the base of the

trigone, where macroscopically inflammatory changes characterized by one-centimeter red macules were observed. The anatomopathological exam revealed no larvae but showed chronic inflammatory changes. Epidemiological screening of the family was performed with negative results. A medical evaluation of the scout group involved in the trip revealed no additional positive cases. The patient was treated with three doses of Ivermectin (200 mcg/day) for 15 days. Following this treatment, the patient denied the passage of larvae. She also denied symptoms of dysuria or incontinence. A cystoscopy was performed a few weeks later, which showed normal bladder mucosa and orthotropic ureteral orifices with no evidence of larvae or other acute or previous infections (Figure 1). These findings show that the patient made a full recovery. However, a urine culture and urinalysis will be performed in one year's time.



Figure 1: Motile larva of *Telmatoscopus albipunctata* visualized by cystoscopy.

Results and Discussion

The *Clogmia albipunctata* fly is a nematocyst known as the drain or toilet fly. Its distribution is rural and cosmopolitan, often inhabiting places with stagnant water [13]. Myiasis infestations affecting the urinary tract are considered the rarest forms. In addition to affecting organs or cavities, there are other more frequent clinical forms, such as infected skin or wounds. When it affects the bladder, the infestation is considered facultative or opportunistic; however, predisposing factors have been described that condition its progression in the host. Some of them are previous injuries, immuno-deficiencies, close contact with animals, very poor hygienic habits, constant exposure to larvae in humid and hot places and even some factors associated with the larval life cycle [13,21].

The clinical presentation of bladder infestation is very varied and symptoms are very non-specific; occasionally, there is urinary dysuria and urgent and frequent urination, which often depends on the number of larvae and the time of contact with the bladder surface. Diagnosis requires documentation of larval outgrowth during micturition or on placement of a Foley catheter [22]. In our case, our patient's exposure to river water in a subtropical area of Mexico was the main predisposing factor. However, she reported no urinary symptoms prior to catheterization. However, she reported no urinary symptoms prior to catheterization, but neurological compromise due to coma epilepticus secondary to barbiturates. Undoubtedly, symptoms that do not help to diagnose a myiasis problem, but

could be explained by the release of neurotransmitters from the *Diptera*. The decision to perform a diagnostic cystoscopy with biopsies allowed us to verify the absence of previous lesions in the urethra and bladder and obtain bladder tissue to determine the degree of reaction with lymphocytes and neutrophils, eosinophils, giant and plasma cells, as well as larval cross-sections. In terms of therapy, there is no standard treatment regimen. Treatment is usually initiated with Ivermectin (0.2 mg per kg), an oral anthelmintic agent with known activity against arthropods such as flies and ticks. The drug is metabolized in the liver and its metabolites are largely excreted in the feces [23]. Most cases respond to treatment and regular cystoscopy controls at 3 and 6 months are recommended [24]. On the other hand, some reports also show recovery without treatment, most likely achieved by an immune response capable of destroying the eggs and larvae [25] (Figure 2).

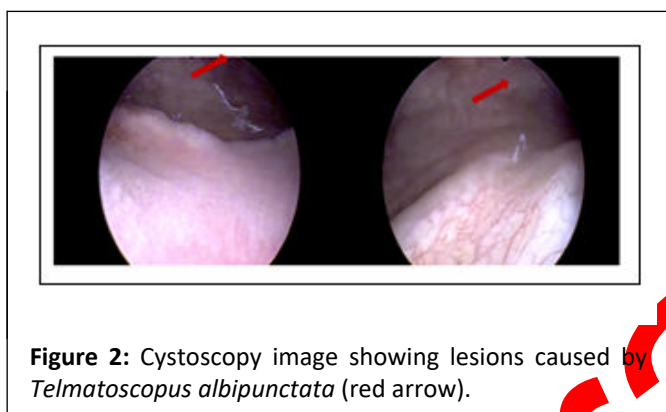


Figure 2: Cystoscopy image showing lesions caused by *Telmatoscopus albipunctata* (red arrow).

Conclusion

In Mexico, there are few documented cases with cutaneous predominance and those affecting the urogenital tract are limited to genital skin, such as the scrotum. As it occurs mainly in rural areas, its presentation is unusual and there are no characteristic data, only documentation of the larvae. This is the fifth reported case of *Clogmia albipunctata* affecting the urinary tract worldwide. This is the first case reported in Mexico.

Acknowledgments

The authors would like to acknowledge the support and guidance of Dr. José Antonio Carmona Suazo, Professor of the Neurocritical assistance course.

Funding

Christian Arias-Reyes received a doctoral scholarship from the Fonds de Recherche de Québec-Santé (FRQ_S). Jorge Soliz is funded by the Canadian Institutes of Health Research (CIHR).

Conflicts of Interest

None declared.

Ethical Approval

Not required.

References

- Chan JCM, Lee JSW, Dai DLK, Woo J (2005) Unusual cases of human myiasis due to old world screwworm fly acquired indoors in Hong Kong. *Trans R Soc Trop Med Hyg* 99: 914-918.
- González MM, Comte MG, Monárdez PJ, Díaz de Valdés LM, Matamala CI (2009) Accidental genital myiasis by *Eristalis tenax*. *Rev Chilena Infectol* 26: 270-272.
- Makarov DV, Bagga H, Gonzalgo ML (2006) Genitourinary myiasis (maggot infestation). *Urology* 68: 889.
- Soler Cruz MD (2000) Myiasis studies at Spain over the last 100 years. *Ars Pharmaceutica* 41: 19-26.
- Tarazona S, Sot R (1989) Myiasis incidence for patients on clinical review. *Kasmera* 17: 31-39.
- Kokcam I, Saki CE (2005) A case of cutaneous myiasis caused by *Wohlfahrtia magnifica*. *J Dermatol* 32: 459-463.
- Park P, Lodhia KR, Eden SV, Lewandrowski KU, McGillicuddy JE (2005) Pin-site myiasis: A rare complication of halo orthosis. *Spinal Cord* 43: 684-686.
- Nevil EM, Basson PA, Schroomraad JH, Swanepoel KA (1969) A case of nasal myiasis caused by the larvae of *Telmatoscopus albipunctatus* (Williston) 1893 (*Diptera: Psychodidae*). *S Afr Med J* 43: 512-514.
- Mullen GR, Lance D (2009) *Medical and veterinarian entomology*. California: Elsevier Academic Press, USA.
- Faulde M, Spiesberger M (2013) Role of the moth fly *Clogmia albipunctata* (*Diptera: Psychodinae*) as a mechanical vector of bacterial pathogens in German hospitals. *J Hosp Infect* 83: 51-60.
- Mohammed N, Smith KGV (1976) Nasopharyngeal myiasis in man caused by larve of *Clogmia (=Telmetoscopus) albipunctatus* Williston (*Psychodidae, Dipt.*). *Trans R Soc Trop Med Hyg* 70: 91.
- Smith KGV, Thomas V (1979) The intestinal myiasis in humans caused by *Clogmia (=Telmatoscopus) albipunctata* larvae. Williston (*Psy-Chodidae, Diptera*). *Trans R Soc Trop Med Hyg* 73: 349-50.
- Kamimura K, Arakawa RA (1986) Case report on urinary myiasis due to the moth fly, *Telmatoscopus albipunctata*. *Med Vet Entomol* 37: 161-2.
- Siddig A, Al Jowary S, Al Lizzi M, Hopkins J, Hall MJR, et al. (2005) Seasonality of old world screwworm myiasis in the Mesopotamia valley in Iraq. *Med Vet Entomol* 19: 140-150.
- Akhoundi M, Ranorohasimanana NM, Brun S, Lacroix CK, Lzri A (2022) *Clogmia albipunctata* (*Nematocera; Psychodidae*) as the Etiologic agent of Myiasis: True or False? *Diagnostics (Basel)* 12: 21-29.
- El-Dib NA, El Wahab WMA, Hamdy DA, Ali MI (2017) Case report of human urinary myiasis caused by *Clogmia albipunctata* (*Diptera: Psychodidae*) with morphological description of larva and pupa. *J Arthropod Borne Dis* 11: 533-538.
- Jeremy-Depatureaux A, Rouleau D, Thivierge K, Cecan A, Beaudin VL, et al. (2019) Urinary myiasis: Not your typical urinary tract infection. *J Travel Med* 26: taz081.

18. Shimpi R, Patel D, Raval K (2018) Human urinary myiasis by *psychoda albipennis*: A case report and review of literature. *Urol Case Rep* 21: 122-123.
19. Garcia-Zapata MTA, De Souza Júnior ES, Fernandes FF, Santos SFO (2005) Human pseudomyiasis caused by *Eristalis tenax* (Linnaeus) (Diptera: Syrphidae) in Goiás. *Rev Soc Bras Med Trop* 38:185-187.
20. Sherman RA, Hall MJ, Thomas S (2000) Medicinal maggots: An ancient remedy for some contemporary afflictions. *Annu Rev Entomol* 45:55-81.
21. López C (2006) Myiasis. *Dermatology Journal* 50: 94-104.
22. El-Badry AA, Salem HK, El-Aziz YAE (2014) Urinary myiasis by *Clogmia albipunctata* larvae. *J Vector Borne* 51:247–249.
23. González Canga A, Sahagún Prieto AM, Díez Liébana MJ, Martínez NF, Vega MS, et al. (2008) The pharmacokinetics and interactions of Ivermectin in humans-A mini-review. *AAPS J* 10: 42-46.
24. Delgado Guerrero F, Reyes Vela C, Torres Aguilar J, Arellano Cuadros R, Hernandez Leon O (2015) Scrotal myiasis: Case report and bibliography review, *Urology. Mexican Academy Bulletin* 30: 41-42.
25. McCoy OO, Rabley A, Prasad MM, Rovner ES (2016) A case of uncomplicated urinary myiasis in a healthy female. *BMJ Case Reports* 2016: bcr2016214783.

Retracted