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# CUTANEOUS MYIASIS IN PATIENTS WITH PRE-EXISTING DERMATOSES ENCOUNTERED OVER A SHORT SPAN – AN INTERESTING CASE SERIES

Dermatology

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## ABSTRACT

**BACKGROUND:** Myiasis is the infestation of tissues of living vertebrates by the larvae of *Dipterous flies*. Clinically, myiasis can be divided into obligatory and facultative. Depending on the body parts involved, Myiasis can be Ophthalmic, Nasopharyngeal, Intestinal or cutaneous. Cutaneous myiasis is of three forms: Migratory,Furuncular and Wound myiasis. The most common agent of wound myiasis in world and India is *Chrysomia bezziana*.**CASE HISTORY:** We encountered 5 cases in pre-existing dermatosis in a short period of one month. Five patients were encountered during 1 month period (December 2017) with male to female ratio3:2.Out of 5 patients,2 patients with Bullous disease, one patient with Harlequin ichthyosis, one patient with Hypertrophic lichen planus and one patient with Plica polonica with pediculosis. The infestation occurred in ulcerative wounds and the larvae were deeply inserted in living tissue. They were treated with turpentine oil occlusion and surgical removal. The maggots were sent for speciation to the Department of Microbiology and Parasitology and it was finally detected to be the larvae of *Chrysomia bezziana*.**CONCLUSION:** Cutaneous myiasis is a rare tropical disease. Early detection and maintaining good hygiene will prevent complications. All the above reported cases belong to obligatory wound myiasis with one rare case of Harlequin ichthyosis with wound myiasis which was not encountered in previous studies. We have reported this case series for its rarity of occurrence in a short period probably influenced by seasonal change(winter) which has not yet reported before.

## KEYWORDS

Cutaneous Myiasis,*Chrysomia bezziana*,Bullous disease, Harlequin ichthyosis, Hypertrophic lichen planus,Plica polonica with pediculosis

## INTRODUCTION

Myiasis is the infestation of tissues of living vertebrates by the larvae of Dipterous flies. **"Myia"** means fly, **"asis"** means disease. It is commonly seen in domestic and wild animals but occurs rarely in humans also. Clinically, myiasis can be primary or secondary. Primary myiasis is caused by biophagus larvae (feed on living tissue) whereas, secondary myiasis is due to necrobiophagus (feeds on dead tissue).[1]Ecologically, myiasis can be obligatory or facultative. Obligatory parasites require living tissue for larval development whereas, facultative parasites usually develop on carrion or vegetable matter, but may occasionally develop on living tissue.[2]

Depending upon the body parts involved, anatomically myiasis can be classified into ophthalmic, nasopharyngeal, intestinal, urogenital or cutaneous. Cutaneous myiasis is more common in unhygienic people with poor sanitation. The most common agents of wound myiasis in world are *Chrysomya bezziana, Cochliomyiahominivorax and Wohlfahrtiamagnifica*. In India, *C. bezziana* is the commonest. A study done in Guangdong, China indicated that *C. bezziana* myiasis appears to be largely neglected as a serious medical condition, with human cases only reported in 16 countries despite this fly species being recorded in 44 countries worldwide.[3]

This disease appears to have been under-recognized as a serious medical condition. Though cutaneous myiasis is rare, we encountered 5 cases in a pre-existing dermatosis in a short span of one month during December 2017 in Dermatology department of Tirunelveli Medical college hospital in South Tamilnadu, India.

### Case history

### Case 1

A 13years old female child residing in Ambai town presented toOPD with painless bloody discharge from scalp for 2 days. The patient had Plica polonica, i.e. matting of scalp hair of 8 months duration associated with pediculosis. On shaving the hair, multiple punched out ulcers with foul smelling discharge over the vertexand occipital scalp with live maggots creeping out were seen**(Fig 1a).** Also, multiple crusted erosions and excoriations over arms, forearms and nape of neck were present. She was diagnosed to have wound myiasis with numerous larvae. Pus culture and sensitivity reported to be coagulase negative staphylococcus which was managed with corresponding antibiotics. CT brain was taken to rule out deeper tissue involvement and it was found normal. Maggots were extracted and sent for speciation. Finally, the patient was discharged after complete healing of the ulcers**(Fig1b).**

**Figure 1**: **Plica polonica(tonsured) with wound myiasis in a 13 year old girl**. (a) Multiple punched out ulcers with creeping maggots (b)Healing ulcers after treatment

### Case 2

A 44years oldmale coming from suburban Tirunelveliwith chief complaints of tense fluid filled blisters over trunk and upper limb of 1month duration. The patient mistook it as a chickenpox and didn't take bath for a month. He presented to our OPD with painful irregular ulcer of 2x2x1 cm over inner aspect of left lower arm with maggots in it and also few purulent crusted and ulcerated plaques with numerous maggots over the lower third of left forearm**(Fig2a,b&c).** Swelling due to cellulitis and fasciitis of the entire forearm with compartment syndrome of left hand was present and was relieved by fasciotomy. The maggots were extruded manually as well as surgically after turpentine oil soaks applications. The patient was diagnosed as bullous pemphigoid with clinical and histopathological examinations and was treated with systemic steroids and antibiotics. Daily cleaning and dressing were done for the ulcers. After a month, all lesions healed well and he was discharged.**(Fig.2d)**

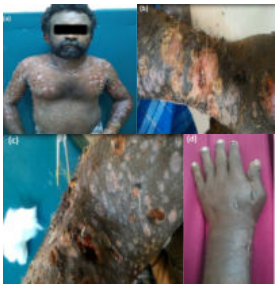


**Figure 2: Bullous pemphigoid in 44 year male** (a) Multiple tense fluid filled blisters

(b) multiple ulcers with slough and maggots over left forearm (c) An oval ulcer with maggots extruding from left elbow (d) Healed fasciotomy scars after treatment of fasciitis and compartment syndrome

**Case 3**

A 9years old male child from urban slum ofTirunelveli who is a survivor of Harlequinichthyosisborn to 3rd degree consanguineous parents presented withpainless swelling over right forearm for 1week duration**(Fig 3a).** On examination, upon removing the thick flake of scales, an oval shaped ulcer of size 4x2.5x0.5cm with multiple maggots protruding outwith head stuck inside was seen in the extensor aspect of the right upperforearm just below the elbow crease giving an appearance of "**cut pomegranate**"**(Fig 3b).**Similarly, single oval ulcers of size 1x1cmwith maggots inside it were seen over the volar aspect of same forearm (Fig 3c). Manual extrusion of maggots with turpentine oil soaks was done**. (Fig 3 a to c)**

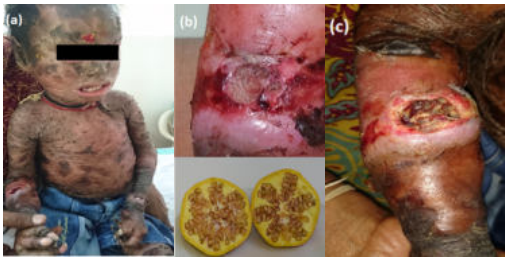


**Figure 3: A 9 year old boy, a survivor of Harlequin ichthyosis** (a) Face and torso show polygonal plates of adherent scales (b) Ulcer over right elbow with closely studded whitish maggots mimicking **"cut pomegranate appearance"** (c) A large ulcer over volar aspect of right forearm filled with classical old world screw worms

**Case 4**

A 20 years old male, resident of Shenkottai town presented with an ulcerative growth over dorsum of left foot for 20 days. H/o foul smelling discharge and visualisation of two worms from the growth were present. Patient had multiple itchy skin lesions presenting over the left leg since the age of 9 years. Personal hygiene was good. Examination revealed well defined fleshy vegetative growth of size 6x4 cm over anteromedial aspect of left foot justabove the ankle coveredwith necrotic slough, serosanguinous discharge and few creeping maggots from one end of the growth. Multiple small hyper pigmented keratotic plaques in a linear fashion were also present over the left lower shin. We diagnosedit as hypertrophic lichen planusand biopsy had proven to be consistent with clinical diagnosis. Maggots were extruded manually and patient was treated with antibiotics followed by intralesional steroid.

**Case 5**

A 45 years old female patient from a rural area, a known case of Pemphigus foliaceous, on irregular treatment with systemic steroids had presented to our OPD with acute exacerbation for 10 days. On the day of admission, patient had adherent crusted plaques with purulent foul-smelling discharge from the frontal and temporal scalp. Shehad not taken bath for 10 days preceding this episode. After four hours, patient developed left sided facial swelling. After cleaning and thorough examination, we noticed two linearpainfululcers of size 4x0.5x0.5cm and 2x0.5x0.5cmrunning anteroposteriorly over left temporal scalp margin behind the ear with 3 live maggots seen in total**(Fig 4a).**Manual extrusion of maggots done with the help of turpentine oil soaks**(Fig 4b).**

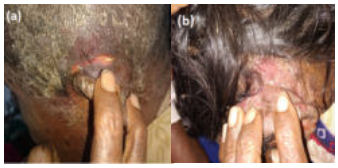


**Figure 4: A 45 yearfemale with pemphigus foliaceous** (a) Linear ulcers behind left ear with maggots before treatment (b) after treatment

The maggots were either manually or surgically extruded and sent for speciation to Microbiology Department, who with the help of the Department of Parasitology, Veterinary college and Research Institute,TANUVAS, Tirunelveli certified it as larvae of *Chrysomya bezziana* in all the cases. **(Table 1)**

**Table 1: Summary of cases of Wound myiasis**

| S. NO. | Age / Sex | Primary dermatoses | Sites of myiasis | Complications | Season/ month | Species of larva |
|---|---|---|---|---|---|---|
| 1. | 13/ F | Plica polonica | Parietal scalp | Scalp and facial cellulitis | Winter & Rainy/ December | *Chrysomia bezziana* |
| 2. | 44/ M | Bullous pemphigoid | Left lower arm & forearm | Cellulitis, fasciitis, compartment syndrome | Winter & Rainy/ December | *Chrysomia bezziana* |
| 3. | 9/ M | Harlequin ichthyosis | Left upper forearm | Localised cellulitis | Winter & Rainy/ December | *Chrysomia bezziana* |
| 4. | 20/ M | Hypertrophic lichen planus | Left foot | - | Winter & Rainy/ December | *Chrysomia bezziana* |
| 5. | 45/ F | Pemphigus foliaceous | Left temporal scalp | Facial cellulitis | Winter & Rainy/ December | *Chrysomia bezziana* |

## DISCUSSION

### Epidemiology

Worldwide, variable number of cases of myiasis have been reported till now involving varying anatomical locations like cutaneous, oral, nasal, ophthalmic, aural, urogenital and enteric myiasis. Historically cutaneous myiasis have been reported from leprosy ulcers, necrotic lesions of adenocarcinoma, squamous cell carcinoma, basal cell carcinoma, filarial lymphoedema, diabetic foot etc.[4]Different geographical areas have different types and species of flies producing myiasis across the world.

Myiasis has a widespread incidence all over the world especially in tropical countries like India which fulfill all the favourable conditions for the abundant growth of myiasis-causing flies. *C.bezziana* is the most common species encountered in cutaneous traumatic or wound myiasis in India as well as the world [5–7]. *Oestrus ovis* is the commonest species isolated fromophthalmomyiasis[8] and *Musca domestica* has been isolated from oral myiasis.[9]Rarely *C.bezziana* also causes oral, ophthalmic, rectal and urogenital myiasis.

*C. bezziana* is also called as old-world screw worm. It belongs to the order *Diptera,*family *Calliphyridae* and suborder *Cyclorrhpha.*[10]They can be identified based on the characteristics of a mature larva and patterns of anterior and posterior spiracles. These larvae are photophobic and penetrates deep into the tissues by sharp mouth, hooks and anchoring intersegmental spines.Ecological classification takes into account the level of parasitism of the host, agent and specific life cycle**(Table 2).**[10]

**Table 2: Ecological classification for myiasis**[10]

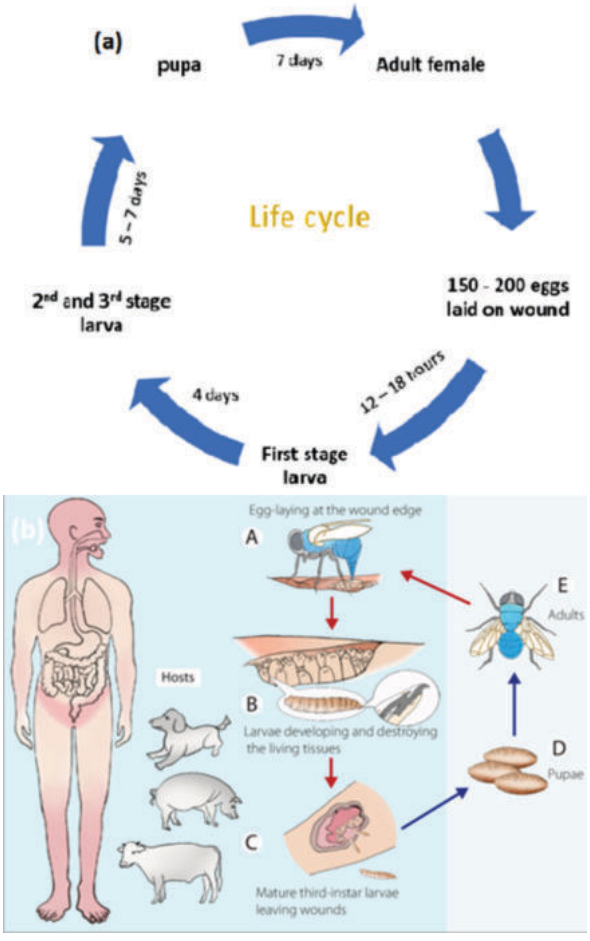
| Classification | Description |
|---|---|
| 1. Specific/ obligatory | Parasite dependent on host for part of its life cycle |
| 2. Semispecific/ facultative | |
| (i) Primary | Free living and may initiate myiasis |
| (ii) Secondary | Free living and unable to initiate myiasis; may be involved once the animal is infested by other species |
| (iii) Tertiary | Free living and unable to initiate myiasis; may be involved when host are near death |

| | |
|---|---|
| 3. Accidental/ pseudomyiasis | Freeliving larva and not able to complete its life cycle; causes pathological reaction when accidentally in contact with the host |

**Lifecycle**

The adult female lays 150 to 200 eggs on the wound which hatches out to first stage larva within 12 to18 hours. Second and third stage larvae develop in another 5 days and this larvae feed in thesub dermal cavity. Mature larvae drop to the ground and pupate in the environment within a week. Larvae tend to leave their host during the night and early morning, probably to avoid desiccation. The pupal stage is temperature dependent with warm weather favouring growth which lasts for 1 week to 2 months.[11]After approximately onemonth, the adults emerge to mate and repeat the cycle**(Fig 5a &5b).**[12]

**Figure 5** (a) Life cycle of *Chrysomya bezziana* (b) Schematic depiction of *C.bezziana* life cycle [adopted from Spradbery (2002). Zoonotic myiasis caused by *C.bezziana* involving hosts including warm-blooded animals and human beings]




**Pathophysiology**

The wounds with alkaline discharge and necrosis are attractive to flies.[13] Larvae of these species produce lysozymes that destroy normal tissues**.** A characteristic odour is produced by infested wounds that attracts gravid flies to lay additional batches of eggs. Temperature appeared to exert influence with relative humidity and rainfall positively influencing the occurrence of myiasis. Many conditions like diabetes and people with sequelae of leprosy, alcoholism with neuropathy, visual defects, mental, neurologic, or psychiatric disturbances, drug addicts, ulcers of the legs due to deep vein thrombosis, as well as those with hyperkeratotic lesions of the skin, have a greater tendency to suffer infestations, due to their lack of sensitivity and they have higher probability of having such lesions.[14] These individuals who do not receive special attention for wound dressing, loss of motility and consciousness, poor social conditions, poor overall hygiene, advanced age and injuries resulting from the continuous exposure of skin and mucosa to secretions all seem to favour the occurrence of the myiasis.

**Clinical features**

Cutaneous myiasis is of three forms: migratory,furuncular and wound myiasis. Wound myiasis or traumatic myiasis, mostly caused by *Cochliomyiahominivorax*, *Chrysomya bezziana,* and *Wohlfahrti amagnifica* occur when larvae infect neglected wounds which are necrotic, haemorrhagic or pus filled. The patient's usual complaints are offoul smelling purulent discharge with pain, pruritus and delayed wound healing with crawling of larva. The lesions may be accompanied by lymphangitis and regional lymphadenopathy. Secondary bacterial infection is the most common complication.[2]More severe cases may be accompanied by fever, chills, pain, bleeding from the infested site, cellulitis, compartment syndrome, osteomyelitis, neutrophil leucocytosis, and hyper eosinophilia.

Furuncular myiasis is commonly caused by *Dermatobia hominis* and *Cordylobiaanthropophaga*. It occurs after penetration of larvae into healthy skin. The typical furuncular lesion is a papule or nodule with central punctum that exudes serosanguinous discharge. The symptoms include extreme pain, pruritus and crawling sensations due to movement of larvae. The other clinical variants include vesicular, bullous, ulcerative, pustular, erosive and ecchymotic types.[10]

Migratory myiasis or creeping myiasis occurs when larvae migrate in the skin. They burrow inside the skin, producing the migratory pattern of the lesions. The depth of the tunnel and migration speed of 1- 30 cm /day are the factors responsible for clinical symptoms. The two most common agents of human migratory myiasis are *Gasterophilus intestinalis* and *Hypoderma spp.* The burrowing *G.intestinalis* larva is located in the lower layers of the epidermis; the Hypoderma larva burrows subcutaneously and forms a less distinct, erythematous, linear lesion and reported to cause ascites, pneumopericardium, hemopericardium and intracerebral myiasis.[15] They can invade the nervous system, eye, and ear, with possible blindness, paralysis, or death.

In developing countries like India, myiasis is a sign of neglected wound care.[4] And endorsing to this statement, we encountered all 5 patients with poor personal hygiene, low socio-economic status and neglected wound care. This might be one of the reasons for emergence of cutaneous myiasis in such patients. Public unawareness and their false spiritual beliefs lead to improper self-care and finally end up in hospitals at critical stages. For these reasons and to prevent or treat complications we must admit all the patients and manage them as in our cases.

We have reported here cutaneous myiasis in various dermatoses like Plica polonica, bullous pemphigoid, harlequin ichthyosis, hypertrophic lichen planus and pemphigus foliaceous. All the five patients were encountered during themonth of December (winter and rainy season) with male to female ratio of 3:2. Two cases lived in suburban areas whereas 3 cases hailed from rural areas. Literatures have shown various types of myiasis in various dermatoses like seborrheic keratoses, bullous disorders, cutaneous malignancies, etc but none have reported cutaneous myiasis in harlequin ichthyosis, plica polonica and hypertrophic lichen planus.[5,7,16]It is also rare and unique that we had encountered the cases altogether during the month of December. High index of suspicion and close examination of wounds and hyperkeratotic skin conditions are necessary to keep a vigil on cutaneous myiasis which is apparently the need of the hour to detect the cases early and prevent complications.

**Diagnosis**

Diagnosis is made easily by clinical inspection of wound. In above cases on direct examination, the larvae were creamy white in colour, with cuticular spines, and varied in size due to different stages of presentation, from 5 to 15 mm. They had strong robust mouth hooks, with four to six papillae on the anterior spiracles, incomplete posterior spiracular peri-treme and pigmented dorsal tracheal trunks in the terminal twelfth larval segment. Based upon these findings they were confirmed to be larvae of *Chrysomya bezziana***(Figure 6a,b,c).**

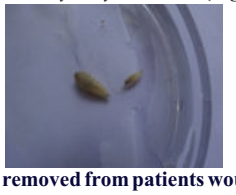


**Figure 6 a - Maggots removed from patients woun**

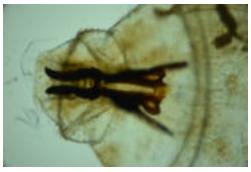


**Figure 6 b - *C.bezziana* L3 Anterior end (40X)**

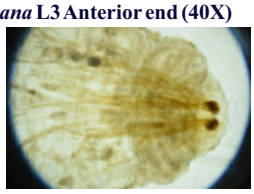


**Figure- 6 c - C.bezziana L3 Posterior end(40X)**

In difficult cases, dermoscopy is useful in showing yellow structure with black barb like spines.[17] Ultrasound has been used to confirm furuncular myiasis; when it fails, colour doppler sonography was able to visualize the continuous movement of internal fluids of larva and was helpful to remove it.[18] After removal, larvae should be preserved in a solution of 70% ethanol. This will preserve the larval length, colour and morphology which can be seen with the aid of a microscope. Formalin should not be used because it causes excessive hardening of larval tissue and makes it difficult to process.

### Treatment

Sanitation, personal hygiene, wound care and extermination of flies are the modalities of prevention and control. Some occlusive agents used for removal are turpentine oil, liquid paraffin, petrolatum, nail polish, animal fat , hair gel, mineral oil, bacon , and bee wax.[16] Topical application of these substances blocks respiratory exchange and either kills the larva directly or induces it to migrate upward, where it can be removed manually with toothed forceps. Surgical methods for extracting larvae include injecting1% lignocaine(anesthetizing the larva through injection of 1% lidocaine hydrochloride around the pore to prevent it from anchoring its spines), placing a cruciate incision adjacent to the central pore, and removing the larva, usually with forceps.[19] Antibiotics are implicated to prevent secondary infection and larvicidal agents like oral Ivermectin given as a single dose of 150–200 mcg/kg of body weight activates the release of gamma amino butyric acid, which induces the death of the larvae. [9]

## CONCLUSION

Cutaneous myiasisseems to be a common but under reported tropical disease. Early detection of cases and maintaining good hygiene is important to prevent complications. All the cases in our series belonged to obligatory wound myiasis.Wound myiasis in caseswith Harlequin ichthyosis, Plica polonica and hypertrophic lichen planus has not beenreported inliterature before. We present this case series for its rarity of occurrence within a short period, probably influenced by seasonal change(winter and rainy season) when the domestic flies favour the indoor atmosphere, which has not yet been reported.

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