

First Record of Indigenous Furuncular Myiasis due to *Cordylobia anthropophaga* In Unusual Ecologic Niche, Makkah, Saudi Arabia

ABSTRACT

Background: *Cordylobia anthropophag*, one of the known furuncular myiasis-associated fly, has long been limited to the Afrotropical region. In Saudi Arabia, Southwest region, part of the Afrtropical region, has been considered the sole endemic region of *C. anthropophag* outside Africa.

Aim: the aim of current study was to investigate suspected cases of myiasis recorded among 7 different children belonging to 3 different households in Makkah province, Midwest of Saudi Arabia.

Methodology: The cases were referred to the Maternity and Children Hospital on 3 different occasions extended from September 2014 to February 2016. All cases were subjected to history taking and clinical examination. Suspected maggots were recovered from associated lesions and were subjected to thorough parasitological and taxonomic examination.

Results: Clinical examination revealed suspected lesions of cutaneous myiasis in different parts of children bodies. History taking revealed no past history of wounds or travelling to Southwest region or tropical sub-Saharan Africa. Based on shape, size, cuticle spine pattern and the posterior spiracles, suspected maggots were identified as 3rd instars of *C. anthropophag*.

Conclusion: To the best of our knowledge the current study is the first record of indigenously acquired cases of *C. anthropophag*-induced furuncular myiasis in Midwest region of Saudi Arabia. This interesting finding might denote a remarkable change in the geographic epidemiology of *C. anthropophag* with the first known incidence of autochthonous transmission pattern outside its conventional Afrotropical endemic region. Indeed, further investigations are required to confirm this spectacular record.

Keywords: *Cordylobia anthropophaga*, indigenous furuncular myiasis, Makkah and Saudi Arabia.

1. INTRODUCTION

Myiasis is defined as the infestation of human and animal tissues by the larvae of dipteran flies. In human, myiasis is usually classified according to the clinical presentation into cutaneous myiasis, traumatic myiasis, and myiasis of body cavities [1]. While the term traumatic myiasis is used when maggots of dipteran flies infest open wounds, cutaneous myiasis always refers to penetration of intact skin by dipteran maggots [1-3].

Furuncular myiasis (FM) is one of the common forms of cutaneous myiasis in which a boil-like lesion, with one or more maggots, developed in affected skin. FM could be developed into what is called migratory myiasis when maggots migrate aimlessly through burrows in the skin and produce numerous furuncular lesions [4].

Cordylobia anthropophaga (tumbu fly) and *Dermatobia hominis* (human botfly) are the most commonly associated dipteran flies with furuncular myiasis. However, these 2 types appears to have distinct geographical preferences, where *C. anthropophaga* is the common cause of FM and reportedly endemic in tropical sub-Saharan Africa, while *Dermatobia hominis* is endemic in Central and South America [5,6]. Tumbu fly-induced FM not only differs than that of human botfly in term of geographical location, but also in the mode of transmission. In human botfly, eggs are carried to the host by a blood-sucking insect, where the hatched larvae invade exposed skin of the trunks, head and limbs. On the other hand, eggs of the tumbu fly, are however deposited on the soil or wet and soiled clothes hung outside for

drying where the hatched larvae invade unexposed skin of the buttocks, trunk and limbs in contact with the wet clothes [3,6].

Diagnosis of furuncular myiasis depends mainly on the epidemiological history of the case such as recent travel to endemic areas [7]. Clinical manifestations of FM are not specific and are often confused with other conditions such as actinomycosis, leishmaniasis, tuberculosis induced furuncular lesions, insect bite, cellulitis, and sebaceous cyst. However, diagnosis of FM is usually confirmed by the extraction and morphological identification of the larvae from the lesion [4,5].

Although *C. anthropophaga* itself has long been limited to tropical and subtropical sub-Saharan Africa [8], cases of tumbu fly-induced FM, are not uncommon in regions outside Africa. However, such cases are usually recorded in travelers returning from endemic areas [9,10]. In Saudi Arabia, human cutaneous myiasis in general, and *C. anthropophaga* infestation in particular are not very common. However, indigenous cases of tumbu fly-related FM were repeatedly reported in Saudi Arabia during the periods of 80s and early 90s of the last century in the province of Asir [11,12] and more recently in the province of Al-baha [13]. Both provinces are located at the South Western region of Saudi Arabia, a region with an ecology of great resemblances to that of subtropical Africa with a rainy, humid climate and surrounding forests environment [13], which make it a potential endemic focus of indigenous FM outside Africa. The current study described seven different cases of human cutaneous FM caused by maggots of *Cordylobia anthropophaga* in Makkah region, Midwest/Saudi Arabia. To the best of our knowledge these cases that were reported among never travelled children represent the first record of indigenous furuncular myiasis outside the conventional ecologic niche of *Cordylobia anthropophaga*.

2. Methodology

2.1. History and clinical examination

All cases were subjected to thorough clinical examination of suspected lesions to record site and shape of the lesions. In addition, all cases were subjected to complete history taking that included medical history, past lesions or trauma as well as travel history. Blood samples were collected from suspected cases for complete blood count.

2.2. Taxonomic identification of extracted maggots

Suspected larvae were collected by gentle squeezing of the lesions and grasping of the maggots painlessly using strile forceps. Cephaloskeleton and posterior spiracles was dissected and examined microscopically after the standard procedures of fixation, dehydration and clearing. The extracted larvae were identified based on the size, body shape, Cephaloskeleton, spine pattern and posterior spiracles morphology as previously described [8].

3. Results

3.1. Clinical presentation and history of cases

Clinical examination of suspected cases revealed that suspected lesions had initially started as a painful itchy swelling in various regions of the patient body including face, shoulder, abdomen, back and limbs. No medical history of systemic malaise, trauma, bites or chronic disease as diabetes or history of wound in affected areas was recorded. History of previous travel to any of the known endemic areas was not evident. Upon clinical examination, there was a cutaneous boil-like lesion, associated with cellulitis spreading over their face, abdomen, back and the limb. Hematological parameters were within normal range and there was neither bleeding nor pus discharge from the crater, but there was what seemed like creamy contents at the bottom layer of the boil. Suspected maggots were obtained after squeezing -with or without the help of small surgical incisions- of the lesions. After removal of the maggots, the left 1 × 1 cm with a depth of about 2 cm sinus-shape wound was dressed local anti-parasitic and antibiotic. Patients were placed on ampicillin/ cloxacillin capsules, tetanus toxoid injection, analgesics and daily

86 wound dressing with EUSOL. One week post treatment, patients were seen in the clinic to ensure that all
 87 the furuncle had healed.
 88 Seven cases belonging to 3 different households were presented on 3 distinctive incidences with a history
 89 of boil-like swellings and skin abscess. In the First incidence, an 11 years-old male child was referred to
 90 the Maternity and Children Hospital, Makkah province, Saudi Arabia at 3rd of September, 2014 with 2
 91 suspected lesions on the face and abdomen (Figure 1A,B). In the second incidence, three siblings aged 2
 92 month male, 5 years female and 16 years female children were referred to the Maternity and Children
 93 Hospital, Makkah province, Saudi Arabia at 23rd of October, 2014 with a single suspected lesion on the
 94 abdomen and/or limb (Figure 1 C,D). In the third incidence, three siblings aged 5 years old female, 7
 95 years old male and 15 years old male children were referred to the Maternity and Children Hospital,
 96 Makkah province, Saudi Arabia at 8th of February, 2016 with multiple lesions. The 7 years old male child
 97 was presented with 4 lesions on the back (Figure 1E). The 5-years old female child and the 15 years old
 98 male child were presented each with a single lesion at the limb (Figure 1F).

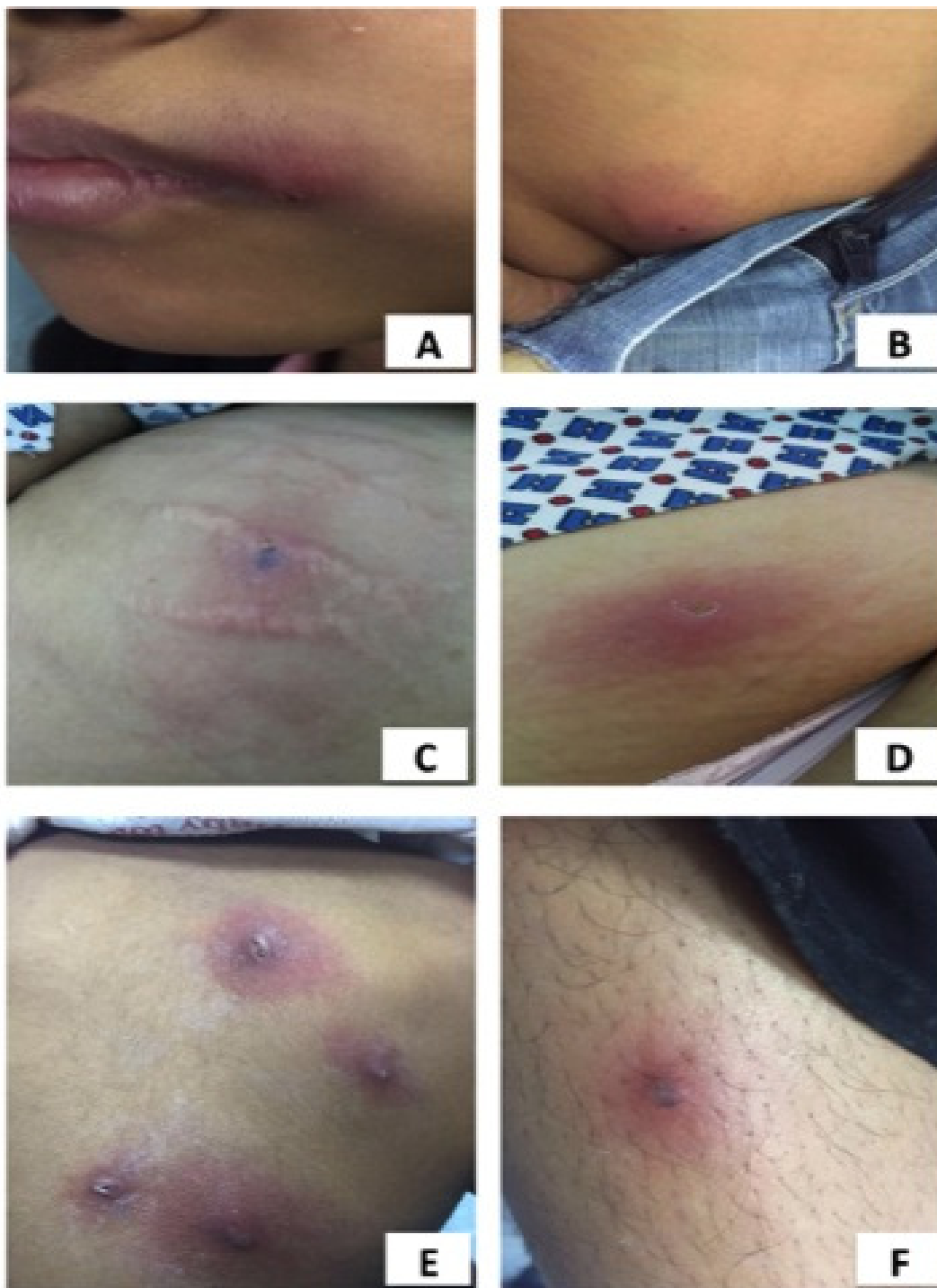


Figure 1. Representative gross appearance of cutaneous boil-like lesions associated with cellulitis in different body locations of referred cases. (A) Cutaneous boil-like lesion in 11-years old child face; (B) Cutaneous boil-like lesion in 11-years old child abdomen; (C) Cutaneous boil-like lesion in 5-years old child abdomen; (D) Cutaneous boil-like lesion in 15-years old child limb; (E) 4 cutaneous boil-like lesions in 7-years old child back and (F) Cutaneous boil-like lesion in 30-years old patient's limb.

3.2. Morphology and identification of extracted maggots

Extracted larvae were identified as typical third-instars of *C. anthropophaga* based on the size, body shape, Cephaloskeleton, spine pattern and posterior spiracles morphology. Briefly, the maggots were oval in shape with a pointed anterior and a blunt posterior end, creamy white in color and with a size range of 7-12 mm (Figure 2A). The oral area contained a pair of toothed, spade-like hooks (Figure 2B). The body consisted of twelve clearly marked segments with transverse black bands covered with backwardly directed black spines Spade-like hooks (Figure 2C). Posterior spiracles were not widely separated and lacked a chitinous rim with a closed peritreme and three slightly sinuous slits (Figure 2D).

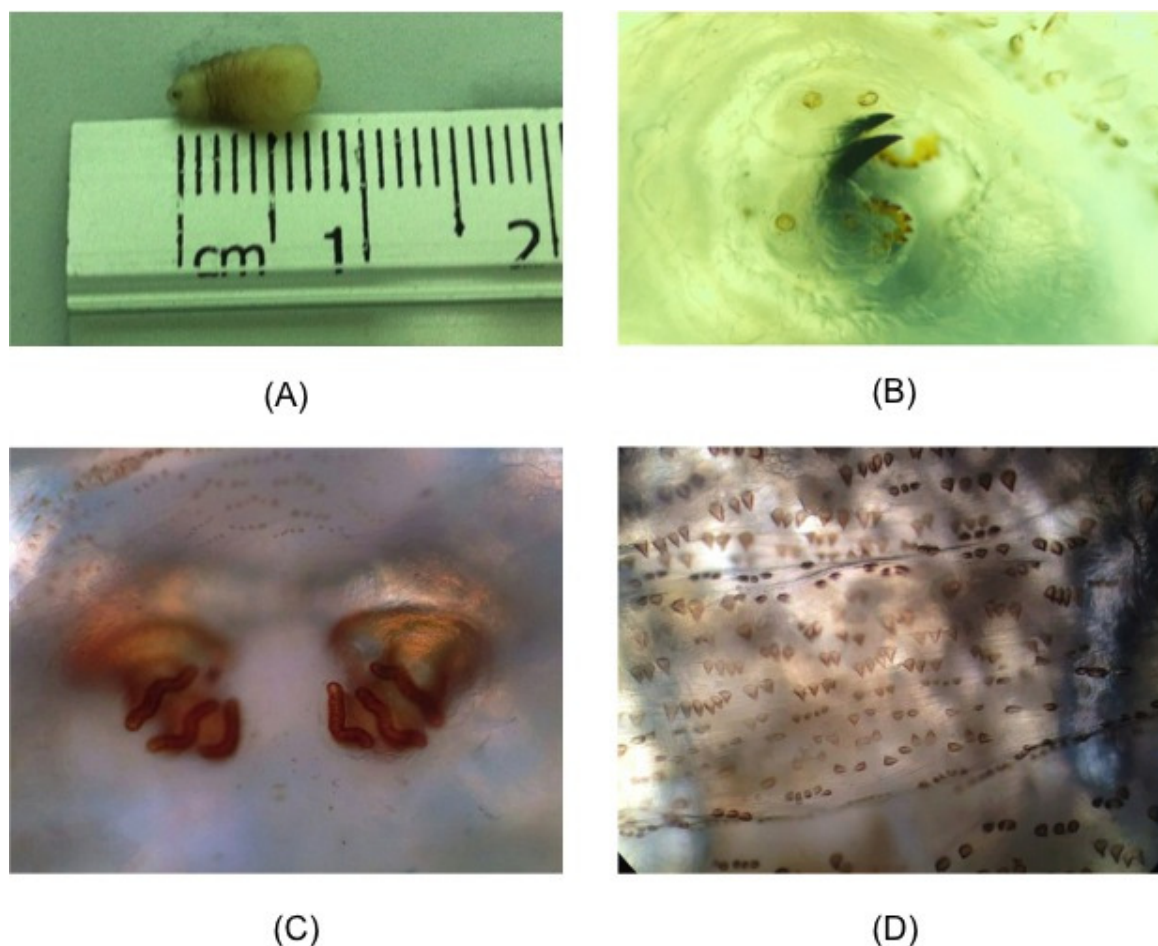


Figure 2: Morphological features of extracted maggots. (A) Gross appearance of one of the maggots (Third instars larva) of *Cordylobia anthropophaga* extracted from skin lesions of referred cases. (B) Anterior end of extracted maggot showing oral area of larva of *Cordylobia anthropophaga* with a pair of tooth (Lateral view). (C) Posterior spiracles of extracted maggots showing characteristic pattern of 3rd. instar larva of *Cordylobia anthropophaga* with weak chitinous rim and closed peritreme with three slightly sinuous slits. (D) Spade-like hooks and transverse black bands on the abdomen.

4. Discussion

Cordylobia anthropophaga-induced furuncular myiasis has long been endemic in tropical sub-Saharan Africa.¹⁴ Although FM due to *C. anthropophaga* used to be restricted to the Afro-tropical regions with an autochthonous pattern of transmission, several reports have recently recorded multiple cases of FM in

several countries as UK [15], USA [16], Spain [17], and China [7], outside the conventional endemic regions of the disease. Although most cases were reported among travellers returning from endemic areas [9,10,18], sporadically reported cases were also recorded among un-travellers in Spain and Portugal suggesting indigenously acquired cases in these non-endemic areas as well [19].

Despite of the little available information on epidemiology of myiasis in Saudi Arabia, few reports had already recorded furuncular myiasis among Saudi patients during the last decade. Interestingly, most incidences of FM were recorded as being indigenous cases that were restricted to Asir Province, a region located southwest of Saudi Arabia [11,12]. The fact that ecological features of Asir region is quite similar to that of the tropical sub-Saharan Africa, have suggested it as the sole Saudi endemic focus outside Africa. More recently, a case of furuncular myiasis proved to be induced by *C. anthropophaga* where recorded in Al-Baha province, which is 300-Km north to Asir, yet sharing the same Afrotropic ecological features of Asir region [13,20,21].

In the current study, 7 cases of FM, belonging to 3 different households were recorded among local residents of Makkah region over a period extended from September, 2014 to February, 2016. All cases were recorded among children with age range from 2 Month up to 16-years where the high risk is probably due to relatively thin skin and lower immunity with the lack of exposure. This finding is in agreement with previous studies, which showed increased infective rate in infants and children as well as pregnant and nursing mothers with physiological impaired immunity [18]. Furuncular myiasis is usually presented as a nodular swelling that develops into a boil-like tender lesion with the symptoms of pruritus, pain, and movement sensation commonly reported [1,22]. Lesions tend to affect unexposed body regions as limbs, back, abdomen, buttocks and shoulders and to lesser extend affecting face and other exposed skin regions [4,23,24]. The central pore of the lesions might exude serous or purulent fluid and usually have one or more maggots that may protrude from the pore with its respiratory spiracle bearing posterior segment [1]. In the current study, the site and number of suspected lesions as well as the clinical presentation of these lesions were suggestive of *C. anthropophaga* type of FM. This was confirmed by the easy extraction and taxonomic studies of collected maggots. However, the fact that none of the currently recorded cases were previously travelled to any of the known endemic areas of *C. anthropophaga* neither in Tropical Africa nor in southwest region of Saudi Arabia suggests indigenous acquisition of the disease. With the exclusion of the southwestern region, Saudi Arabia is considered one of the driest countries in the world. In fact Asir and Al-Baha provinces, as parts of the southwestern region, is considered by many authors part of the Afrotropical zone [25]. On the other hand, Makkah region is far from sharing the same ecological features of the Afrotropic zone with a more desert and water deficit conditions [26]. These facts make Makkah environment far from being conventional permissive ecological environment for *C. anthropophaga*. However the un-travelling history of the presented cases to any of the known *C. anthropophaga* endemic areas raise a very interesting question about the possibility of autochthonous pattern of transmission in this non-endemic region. A question that might denote a dramatic change in the geographical distribution of such myiasis causing flies, which indeed needs further investigations to document the inhabitation and successful life cycle completion of *C. anthropophaga* in Makkah region.

5. CONCLUSION

To the best of our knowledge, the current report is the first to record cases of indigenously acquired FM in Makkah region, Midwest of Saudi Arabia. The recording of indigenously acquired cases of FM in a dry desert climate region among never travelled patients to any of the known endemic places of *C. anthropophaga* might represent the first cases of autochthonous transmission pattern outside its conventional endemic niche. Indeed, the current report represents an interesting finding that clearly shows that calliphorine species is not restricted to Afrotropical regions, yet further epidemiological and ecological investigations are required to gain more insights on this interesting finding.

Consent

All participated persons has given, either via themselves or via their parents or legal guardians, a written consent that they acknowledge the inclusion of their data anonymously in the study and that they cannot be identified via the paper.

Ethical Approval

Not applicable

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