Myiasis from *Sarcophaga* spp. in a patient with cutaneous lymphoma

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SUMMARY

Human autochthonous myiasis is uncommonly reported in Europe. This report describes a case of myiasis of a wound caused by *Sarcophaga* spp. Suffering from cutaneous lymphoma, the patient showed, at the level of his scalp lesions, the presence of larvae that were removed during curettage surgery; they were subsequently identified as belonging to the genus *Sarcophaga*. Preservation of these larvae in 10% formalin did not allow identification at the species level using molecular methods.

Keywords: myiasis, sarcophaga, cytotoxic epidermotrophic lymphoma.

INTRODUCTION

The term "myiasis" was proposed for the first time by Hope (1840) and describes a parasitic infestation caused by the larvae of flies (Diptera, Brachiceri, Ciclorrafa) through the invasion of vital or necrotic tissues [1]. The disease occurs widely all over the world. However it is subject to seasonal variations, the prevalence of which is related to latitude and the life cycle of the various species of flies [2]. The majority of cases occurs in tropical and sub-tropical countries, because flies prefer a hot-humid environment. Consequently, in temperate zones, myiasis occurs mainly in the summer months, while it is found throughout the year in tropical regions [3]. Poor hygiene and low socio-economic status, metabolic disorders (diabetes), cutaneous neoplasms, and psoriasis, often favor the establishment of cutaneous myiasis [4-6]. Larvae frequently responsible for myiasis in both domestic animals and humans belong to the Calliphoridae and Sarcophagidae family and to *Rhinoestrus* spp., *Gasterophilus* spp., *Hypoderma* spp., *Chrysoma* spp. [7]. According to an ecological classification based on the type of contact with the host, the following can be distinguished:

1) specific myiasis, in which the insect must necessarily perform a phase of larval development in the tissues, even intact, of a host;
2) occasional (optional) or semi-specific myiasis in which a normally free-living larva can live part of its life cycle in a host, a behavior favored by the presence of accidental skin lesions and,
3) myiasis in which the eggs or larvae are accidentally swallowed with food [8-10].

Agents responsible for specific myiasis, such as members of the *Oestridae* family, usually have long periods of relationship with the host, routinely parasitize the mjoik pathogenicity; on the contrary, agents responsible for occasional myiasis such as *Sarcophaga* and *Calliphora* have brief relationships with the host, parasitize superficial tissues such as the skin, and have a high degree of pathogenicity [11]. Clinically, in humans, the most frequent sites of infestation are the skin and only rarely other districts such as eye, ear,

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nose, urogenital or intestinal tract, and exceptionally the CNS [12]. Cutaneous myiasis can, in turn, include furuncular, migratory (creeping) and wound myiasis, depending on the type of infesting larvae [13]. In the literature, various dermatological conditions associated with cutaneous myiasis have been described, especially ulcerative lesions which can be complicated by this infestation [10-14]. In the present clinical case, we describe the diagnosis and treatment of a cutaneous myiasis of necrotic tissue caused by *Sarcophaga* spp. in a patient with a cutaneous lymphoma (CD8 +).

CASE REPORT

The patient was a Caucasian male, aged 34, resident in eastern Sicily, Italy, who came to the observation of the Dermatology Unit of the Hospital Vittorio Emanuele in Catania, Sicily, Italy, for the first time in August 2017. The patient, residing in an urban area with poor sanitary conditions, reported that he had been treated in previous years with pharmacological therapy, though it was not specific, for a cutaneous pathology diagnosed as psoriasis, obtaining modest clinical benefit. No other relevant pathology was reported. At the time of admission, laboratory tests were performed that documented: Ab anti-HIV negative, VDRL and TPHA negative. At the clinical inspection, the patient presented lesions in various parts on the skin: crusts and squamous crusts detected on a green-yellow skin on an erythematous background, with a diameter ranging from a few millimeters to 10 cm, with a widespread localization, but primarily attributable to the face, trunk, limbs and scalp. In the scalp, the lesions were also purulent. Under them, we identified the presence of numerous cavities within which - in a purulent environment - we could see the presence of 5 whitish larvae, about 15 mm in size, actively mobile (Figure 1). The lesions were cleaned with saline solution, chlorhexidine 0.5% was applied and, finally, the necrotic tissue and larvae were removed. A biopsy was also performed which subsequently allowed the diagnosis of Cytotoxic Epidermal Lymphoma, classified in the CD8 + Lymphoma group (Figure 2). Therefore, chemotherapeutic therapy was undertaken using prednisolone with the addition of cyclophoshamide, doxorubicin, etoposide, vincristine (CHOEP pro-

Figure 1 - Purulent lesion with larvae in the scalp.

Figure 2 - Patient affected by Cytotoxic Epidermal Lymphoma.

Figure 3 - Larva of Sarcophaga.

Figure 4 - Particular of the peritrema.

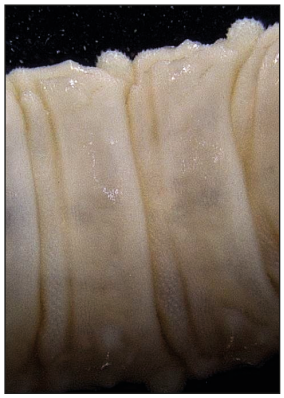

Figure 5 - Small thorns on the larval body.

tocol). The therapy was suspended after the administration of only two cycles due to the worsening of the clinical conditions and replaced by administration of brentuximab i.v. Despite these therapies, the patient went to exitus after a short time, due to the neoplastic condition.

Identification of larvae The morphological identification of the larvae was performed microscopically, using a Leica MZ 12 stereoscopic microscope, with a micrometric eyepiece and a glossy one connected to the camera. The images of the detected structures were obtained through a Leica MZ 205A stereoscopic microscope (equipped with the Leica Application Suite v. 4.2.0). The morphological characteristics allowed us to establish the correct identification to the genus *Sarcophaga* (Meigen, 1826), at the third stage of development. The larva of *Sarcophaga* is cylindrical (Figure 3), of a whitish-yellow color, larger than that of Calliphoridae and unlike the latter, it presents the posterior spiracles inside a cavity. The three spiracular fissures, indicative of the third stage of development, were not parallel to each other. The peritrema is light and does not close to form the posterior region (Figure 4). The body has small thorns (Figure 5), free of chitin and therefore not very evident. In this case it was not possible to identify species with molecular methods due to the conservation of larvae in 10% formalin solution [10].

DISCUSSION

In countries with high levels of hygiene, myiasis is not frequent and the few cases found are therefore of considerable clinical-epidemiological relevance [7] .

According to a review conducted by Amandeep and Zorawar Singh, globally from 1914 to 2015, the cases of myiasis mainly refer to the species belonging to the families Calliphoridae (12 species) and Sarcophagidae (9 species) followed by Oestridae (5 species), Muscidae (4 species), Psychodidae (2 species), Syrphidae (1 species), Phoridae (1 species), Drosophilidae (1 species), Dryomyzidae (1 species), and Gasterophilidae (1 species) [15].

In their review, Zammarchi et al. described 703 cases of autochthonous human myiasis in Italy in the period 1957-2013, of which 98% were caused by *Oestrus ovis*, *Linnaeus* and only 2% caused by other species [16]. Already in the mid-1950s, 414 Italian cases had been reported and at least one case of myiasis from *O. ovis*, 77% of which were diagnosed in shepherds [17]. The case of myiasis from *Sarcophaga* spp. described here therefore falls within the aforementioned 2% of Italian case studies.

In the last forty years, in the world, only thirty cases of human myiasis related to the *Sarcophaga* genus have been described, and in Italy, among the latest reports, there was one case of myiasis in 2003 and another of cutaneous myiasis in 2008, in a polytraumatized patient [15-18]. *Sarcophaga* is a genus of diptera that includes about 2,500 species with worldwide diffusion, agents of numerous types of myiasis depending on the parasitic anatomical district. From a clinical point of view, larval activity is oriented to necrotic tissues, exudates excretions and stagnant secretions in anatomical cavities, generally in

association with more or less severe infectious processes [19]. The morphological identification of the genus takes place without particular difficulty, however, within the genus, the species are difficult to recognize, both in the adult and in the larval stages [20]. The ovoviviparous female has the characteristic of not laying eggs on the tissues, but of giving birth directly to larvae at the first stage (L1). The L1 larvae are able to feed autonomously immediately after their release on the host [19]. The larval development continues in the thickness of the dermis through two other evolutionary stages, L2 and L3, distinguishable from each other morphologically, before entering the so-called post-feeding phase in which the larvae will no longer feed on necrotic tissues, but will move away from the host and will complete their metamorphosis that will change them into an adult insect [21]. In almost all hosts, including humans, the infestation on healthy skin occurs in very young subjects, because the hooks of the larval buccal apparatus are generally not strong enough to penetrate the skin of adults. This suggests that in our case, being an occasional myiasis caused by Sarcophaga, the patient's necrotic lesions, caused by the underlying neoplastic pathology, constituted a predisposing condition for the onset of the infestation [22].

The purulent appearance of the larval host lesions certainly suggests the presence of an underlying bacterial infection. The association between myiasis and bacterial infection has already been reported in the literature. The presence of suppurative lesions attracts the adult female [19, 23]. Several bacterial species have been found in lesions hosting larvae of diptera, many of which belong to the *Enterobacteriaceae* family [23]. In our case, the clinical appearance of the lesions suggested a possible coexistence of myiasis and bacterial infection (presence of greenish-yellow pus), however, no bacteriological investigation was performed on the lesions. Therefore, our case shows that it is important to be aware of myiasis, and the possible correlation with infections, to ensure proper wound management, using appropriate treatment methods [24].

Conflict of interest

The authors declare that there are no conflicts of interest.

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