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Gingival Myiasis on Oral Squamous Cell Carcinoma in Turkey: A Case Report

Oral Squamöz Hücreli Karsinom Zemininde Gelişen Miyaz Olgusu

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ABSTRACT

Myiasis is a disease caused by tissue invasion of diptera larvae and eggs. Oral myiasis is mostly related to old age, poor oral hygiene, suppurative lesions, anatomical disorders and cancer cases. Oral squamous cell carcinoma (OSCC) is an important risk factor for myiasis. This report presents the case of an 82-year-old woman who presented with gingival myiasis developing on the background of OSSC. The patient was diagnosed with OSSC in the hospital. Myiasis larvae were identified and sent to the National Parasitology Reference Laboratory for identification. Thus, development of myiasis on OSCC background was shown in Turkey for the first time. Myiasis larvae have been identified as the 3rd phase of the larvae *Sarcophaga* sp. development (Diptera: Sarcophagidae). As a result, myiasis cases are sporadic in Turkey, and it can be avoided by controlling fly population and by paying attention to hygiene. Controlling myiasis is an important public health problem and should be considered in a single health concept, as it causes health problems in both humans and animals. The findings of this case will draw attention to the importance of dealing with myiasis factors, which is a public health problem.

Keywords: Larvae, myiasis, oral squamous cell carcinoma, *Sarcophaga* sp.

ÖZ

Miyaz, diptera larva ve yumurtalarının doku istilasından kaynaklanan bir hastalıktır. Oral miyaz nadir görülen, yaşlılık, zayıf ağız hijyeni, süpüratif lezyonlar ve kanser olguları ilgili bir durumdur. Oral skuamöz hücreli karsinom (OSCC) miyazda önemli bir risk faktörüdür. Bu raporda, 82 yaşında bir kadın hastada OSCC zemininde gelişen miyaz sunulmuştur. Hasta, ağız içinde ovoid bir lezyon ile diş hekimine müracaat etmiş, sonra hastanede OSSC tanısı konmuş ve bu zeminde miyaz larvaları tespit edilmiştir. Larvalar Ulusal Parazitoloji Referans Laboratuvarı'na gönderilmiştir. Laboratuvar çalışmaları ile tespit edilen larvaların üçüncü dönem *Sarcophaga* sp. (Diptera: Sarcophagidae) larvaları olduğu tanımlanmıştır. Böylece, Türkiye'de ilk defa OSCC zemininde gelişen miyaz olgusu gösterilmiştir. Sonuç olarak, Türkiye'de miyaz sporadiktir ve sinek popülasyonunu kontrol ederek ve hijyene dikkat edilerek önlenebilir. Miyaz ile mücadele bir halk sağlığı sorunudur. İnsanlarda ve hayvanlarda karşılaşıldığından tek sağlık kavramı içinde değerlendirilmelidir. Bu olgu sunumu ile, bir halk sağlığı problemi olan miyaz etkenleri ile mücadeleye önem verilmesinin üzerinde durulması amaçlanmıştır.

Anahtar Kelimeler: Larva, miyaz, oral skuamöz hücreli karsinom, *Sarcophaga* sp.

INTRODUCTION

Myiasis is a parasitic infestation of human/animal tissues and cavities with dipterous larvae and eggs. The term myiasis is derived from the Greek words ''myia,'' which means fly and ''iasis,'' which means disease. This term was first proposed by Hope in 1840 (1) and oral myiasis was first defined by Laurence in 1909 (2,3). The clinical menifastations of myiasis depend on the fly species, the invasion level and stage of the larvae and the infestation area. Flies that cause myiasis belong to the class of Insecta, order Diptera, and are classified into seven different families (calliphoridae,

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sarcophagidae, oestridae, hypodermatidae, gasterophylidae, glossinidae and muscidae), which can infest the skin and body cavities. Myiasis is divided into three groups; i- obligatory (the larvae can affect undamaged skin and require a living host for their development), ii- facultative (free living larvae capable to initiate myiasis to destroy nectoric tissue), iii- accidental (free living larvae which cause myiasis when the larvae are injested accidentally). Myiasis can be also classified according to the anatomical area of infestation; i- cutaneous myiasis, ii- myiasis of external orifices, iii- visceral myiasis. In addition, it can be divided clinically into; i- primary and ii- secondary myiasis. Myiasis most often occurs in the skin wounds, nose, eyes, sinuses, ears, and vagina. Occasionally, it can be observed seen in the mouth (2,4,5).

Sarcophaga sp. is a member of the Sarcophagidae family and a known group of flies which cause myiasis in Europe and Turkey. They are called flesh flies, and their adults are twice the size of house flies. These flies generally leave their eggs or larvae in the nasal cavity and cause myiasis on living tisues. In this study, we present a case of *Sarcophaga* sp. myiasis seen in the oral cavity.

CASE REPORT

An 82-year-old female patient visited a dentist with a complaint of a tongue wound. The patient was directed to the hospital by dentist due to the complicated apperance of the wound, and there was indications for a poor oral hygiene. A moderately sized lesion was observed in the patient's mouth. On physical examination, an ovoid lesion of approximately 4 cm was detected in the right half of the sublingual region that crosses the midline and not descends to the floor of the mouth. Induration around the lesion and fixed lymph nodes of 1 cm in the left upper jugular, 1 cm in the middle jugular, and approximately 3 cm in the right upper jugular were detected by palpation. A biopsy of the lesion showed that the patient was suffering from a well-differentiated squamous cell carcinoma. After debridement of the lesion, a large number of larvae were detected within the lesion. Two larvae removed alive from the lesion were sent to the parasitology laboratory for examination, where they were identified as the third instar larvae of *Sarcophaga* sp. using standart taxonomic keys (6,7).

For this identification, microscopic preparations were prepared by dissection of the cephalopharyngeal skeleton and anterior and posterior stigmas of the larvae. The shape, number and order of larval segments, peritemes and perithem buttons were evaluated morphologically. It was determined that the larvae were 10-12 mm in size and consisted of 12 segments, which were seen as transverse spiral bumps (Figure 1A). The back end of the *Sarcophaga* sp. larvae had a pit-like structure that enclosed the posterior stigmas. The posterior stigmas consisted of three long slits, while the middle one was straight and the others were curved (6,7) (Figure 1B, C).

It was observed that the anterior stigmas extending from the lateral surfaces of the second segment were small and had a fan-like structure consisting of 13 branches, while the cephalopharyngeal skeleton was typical to *Sarcophaga* sp. (6,7) (Figure 2A, B).

In the literature, local application of various substances such as ivermectin, turpentine oil, mineral oil, ether, chloroform, ethyl chloride, mercury chloride, creosote, saline, phenol, calomel, olive oil, iodoform are recommended for the control of larvae. In addition, it is necessary to apply treatments that prevent the development of secondary bacterial infection in the skin

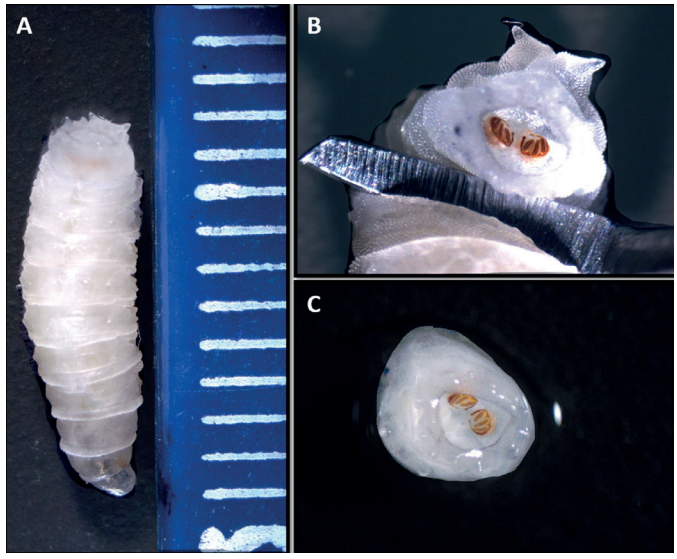


Figure 1. A) *Sarcophaga* sp. 3rd stage larvae, B, C) Posterior stigmas (posterior spiralce)

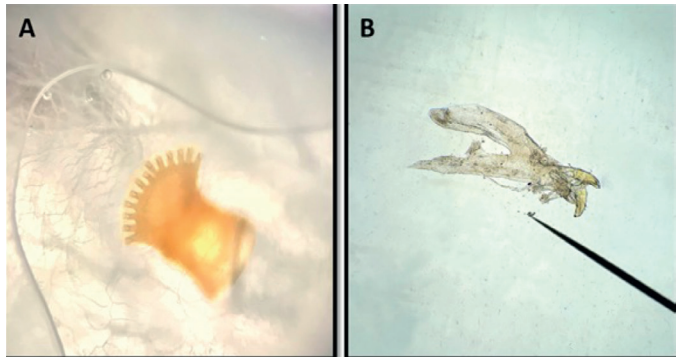


Figure 2. A) Anterior stigmas (anterior spiracles), B) Cephalopharyngeal skeleton

surrounding the lesion where myasis agents are located. In the present case, the larvae were physically removed from the wound once a day until they were completely cleaned, and antibiotic treatment was initiated in terms of infection while the patient was being treated for cancer. However, the patient's condition deteriorated soon and she died thereafter.

DISCUSSION

Myiasis is a parasitic invasion of tissues by living larvae, usually seen in tropical and subtropical countries, and often caused by poor hygiene (8). Myiasis particularly affects the elderly humans and develops also secondary to poor alcoholism, and suppurative lesions (9). After the female flies lay their eggs on the damaged tissues, the larvae can settle in the deep tissues. Cases of myiasis in the mouth and nasal cavity can cause dangerous complications which can result even with death. Larval infestation causes progressive destruction and cavitation and ultimately adheres tightly, forming a fibrous capsule that causes difficulties in dissection during surgery (10). The severity of tissue destruction depends on the number of larvae that settle in the host tissue.

Although oral myiasis is mostly seen in tropical countries, it can also occur in temperate climates when oral hygiene is inadecvate and in immuno-surpressed patients. It is known that patients with suppurative lesions, severe bad breath, facial trauma,

Table 1. Oral myiasis cases which were reported from Turkey

No	Author, publication year	Case year	City	Gender	Age	Localization	Species	Systemic diagnosis	Treatment
1	Ciftçioğlu et al.(12)	1996	Ankara	M	80	Orotracheal myiasis	*Wohlfahrtia magnifica* (Diptera: *Sarcophagidae*)	Intensive care patient, poor general condition	The patient was lost
2	Erol et al. (13)	2000	Diyarbakır	W	4	Myiasis in the upper lip lesion	*Calliphora* spp.	Healthy but poor oral hygiene	The patient could not be followed after mechanical debridement
3	Gursel et al. (14)	2002	Konya	M	26	Oral cavity (Gingival myiasis)	Diptera: *Calliphoridae*	Healthy but poor oral hygiene	Mechanical debridement, patient recovered in six months after periodontal flap surgery
4	Yazar et al. (15)	2002	Kayseri	M	15	Oral cavity	*Sarcophaga* sp.	*M. tuberculosis* was detected in the CSF taken after meningeal symptoms	Mechanical debridement, the patient died on the 30th day
5	Büyükkurt et al. (16)	2008	Erzurum	M	4	Oral cavity (Gingival myiasis)	*Wohlfahrtia magnifica* (Diptera: *Sarcophagidae*)	Unavailable	After mechanical debridement, recovered in 3 months
6	Balcioglu et al. (17)	2008	Manisa	W	65	Subungual myiasis	*Calliphora* spp.	Chronic psychosis	Mechanical debridement and recover
7	Arslan et al. (19)	2013	Van	-	2	Oral cavity (Gingival myiasis)	-	Unavailable	Mechanical debridement and recover
8	Çetin Özdemir et al. (20)	2013	Gaziantep	M	43	Gingival groove region (gingival myiasis)	*Wohlfahrtia magnifica* (Diptera: *Sarcophagidae*)	Unavailable	Mechanical debridement and recover
9	Taş Cengiz et al. (21)	2019	Van	M	63	Oral cavity (Gingival myiasis)	Diptera: *Calliphoridae*	Intracerebral hemorrhage, respiratory failure and hypertension	Mechanical debridement, the patient died on the 6th day

persistent mouth opening, thumb sucking habits and advanced periodontal diseases, lesions in tooth extraction areas, fungal infections of the buccal mucosa, oral carcinomas and epilepsy are susceptible to oral myiasis (11).

In Turkey, nine oral myiasis cases were reported (12-22) (Table 1), showing that kind of myiasis was mainly observed in the elderly and younger ages.

Cases of myiasis developing in different types of carcinoma have been reported worldwide (22-32). The most common malignant lesion in the head and neck region is squamous cell carcinoma (SCC) (33). Oral SCC (OSCC) is a possible risk factor for myiasis for humans or other mammals. In oral myiasis cases, larvae can cause itching and irritation, as well as cause serious and sometimes life-threatening bleeding by destroying vital tissues. With this article a myiasis case is reported in developing OSCC ground for the first time in Turkey. Myiasis occurring on OSCC background were reported five times from India and four from Brazil (24,25).

Prevention is based on the fight against adult flies. Limiting myiasis is an important issue for public health. Poor health conditions and insufficient sanitation are important risk factors for myiasis, especially in underdeveloped countries with low socioeconomic status. It is very important for people living in endemic areas to not leave their wounds open and avoid sleeping outdoors. Physicians should consider clinical examinations in terms of myiasis during the months of May-October, when the fly population is intense throughout our country. In addition, it is known that myiasis can be significantly reduced by informing the people in the regions dealing with animal husbandry, training the personnel who are interested in wound care in health units or hospitals, and combating the factors.

INFORMATION

This study was presented as a poster at 38th International Turkısh Microbiology Congress (04-08 November 2018 Antalya, Turkey).

* Ethics

Informed Consent: Since the patient was lost when myiasis was identified, the patient's approval could not be obtained.

Peer-review: Externally and internally peer-reviewed.

* * Authorship Contributions

Concept: B.Y., Design: B.Y., C.B., Data Collection or Processing: B.Y., C.B., N.K., Analysis or Interpretation: B.Y., C.B., N.K., S.K., Literature Search: B.Y., Writing: B.Y.

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