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CASE REPORT

ORAL MYIASIS CONVERTING TO ORAL SQUAMOUS CELL CARCINOMA

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ABSTRACT: INTRODUCTION: Oral Myiasis, a condition of infestation of the body by fly larvae (maggots) is a rare pathology in humans. It is associated with poor oral hygiene, alcoholism, senility, suppurating lesions, severe halitosis. It is seen frequently in tropical countries and hot climatic regions. The reported cases in literature of oral Myiasis associated with oral cancer are few. The treatment is a mechanical removal of the maggots but a systemic treatment with Ivermectin, a semi-synthetic macrolide antibiotic, has been used successfully for treatment for oral myiasis. We present a case of 55yr old male alcoholic patient with oral myiasis with extensive proliferative growth of oral cavity. Our patient was managed with manual debridement and administration of systemic ivermectin along with antibiotic coverage. Incisional biopsy of the proliferative lesion showed well differentiated squamous cell carcinoma. Thus our patient showed presence of oral myiasis in association with oral squamous cell carcinoma.
KEYWORDS: Oral Myiasis, Maggots, Oral squamous.

INTRODUCTION: SUMMARY: Tissues of the oral cavity can be invaded by parasitic larvae of house flies, this condition is called as oral Myiasis. Myiasis comes from the Greek term "myia" meaning fly and was first introduced by Hope in 1840.[1] It was defined by Zumpt as the infestation of live human and vertebrate animals with dipterous larvae, which feed on living or dead host tissue, liquid body substance or ingested food for a certain period of time.[2] The larval stages of dipterous flies, are usually known as maggots.[3] Foremost decription of Oral Myiasis was given by Laurence in 1909.[4]

More than 80 different species have been reported to cause this condition in human beings. Clinically myiasis can be classified as (i) primary myiasis – larvae that feed on living tissue by biphagous larvae. (ii) second art myiasis- larvae that feed on dead tissue caused by necrobiphagous flies. Based on anatomic site can be classified as (i) cutaneous myiasis (ii) myiasis of external orifices and (iii) myiasis of internal organs. Depending on the involved tissue as (i) accidental myiasis (larvae ingested along with food), (ii) semi specific (larvae laid on necrotic tissue in wound), (iii) obligatory myiasis (larvae affecting the undamaged skin).[5]

The most common anatomic sites for myiais are the nose, eye, lung, ear, anus, vagina and more rarely, the mouth.[6] The different parts of the body such as cutaneous, urogenital, ophthalmic, nasopharyngeal, intestinal, and the oral cavity may be infested by the larvae. Oral Myiasis is more commonly ascribed to predisposing anatomical, medical conditions where oral cavity is being exposed to the external environment for a prolonged time, mouth breathing, anterior open bite, incompetent lips, cerebral palsy, following tooth extractions, neglected mandibular fracture, patients undergoing mechanical ventilation, and certain local pathological conditions such as cancrum oris, and oral malignancies.[7]

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Higher incidence is reported in tropical, subtropical regions of Africa, America, and South East Asia, where warm, humid climate prevail almost throughout the year; commonly seen among people where sanitation, personal hygiene is often ignored. The infestations can strike organs or tissues that are accessible to egg-laying and development of the larvae, which feed on living or necrotic tissue and body fluids, and necrotic lesions that provide an ideal substrate.[8] The flies lay over 500 eggs directly on the diseased tissue. These eggs hatch and the larvae get their nourishment from the soft tissue. The common symptoms associated are painful growth with ulceration and itching due to crawling movement of the larvae.

The oral cavity is rarely affected by this infestation and is associated with poor oral hygiene, alcoholism, senility, suppurating lesions, severe halitosis and other conditions. Some time larvae come out crawling out from nose or oral cavity. They cause extensive damage to soft tissue, death may occur due to meningitis. The larvae in the mouth are observed as worms/mugs/maggots.[9]

We present a rare case of oral myiasis with association with squamous cell carcinoma.

CASE: A 55 yrs old male patient visited the department with a chief complaint of mass in the mouth with foul odour since a month. The patient also complained of difficulty in breathing and in speech. Patient was alcoholic since 25 yrs.

Extra oral examination showed a necrotic and proliferating extensive mass protruding out from the mouth with an ulcer present on the chin area (Fig. 1, 2). A fetid odour was also present with the same. Regional lymphadenopathy was noticed which was hard and non-tender on palpation.

Intraoral examination of the patient was not possible as the proliferating lesion had occupied the entire oral cavity which had also caused depression of the tongue which could be seen on examination.

The ulcer on the chin showed presence of maggots. On the basis of history and clinical findings a diagnosis of Oral myiasis was made.

Cotton gauze impregnated with turpentine liniment was placed over the ulcerative lesion for approximately 10 min. Mechanical removal of larvae was done (Fig. 3) using turpentine oil along with surgical debridement of necrotic tissue. About 80-90 larvae were removed (Fig. 4).

The patient was prescribed inj. ivermectin and antibiotics and was recalled after 3 days for surgical removal of the proliferative lesion.

The incisional biopsy was done for the proliferative lesion and histopathologically, the section showed, stratified squamous epithelium with proliferation. The connective tissue was invaded by tumor, showing malignant epithelial cells with large hyperchromatic pleomorphic nucleus and a moderate amount of cytoplasm. Keratin pearl formation was also noted at places. The stroma showed dense inflammatory cells predominantly lymphocytes and plasma cells. Overall features were of well-differentiated squamous cell carcinoma (WDSCC).(Fig. 5)

DISCUSSION: Oral Myiasis occurs in vertebrate animals, main parasites being flies of order of diptera (maggots) which feed on the host's dead or living tissue. It is a rare condition which occurs in human beings. The life cycle of a fly begins with the egg stage followed by the larva,

the pupa and finally the adult fly. Sood et al described that the larva can be divided into three stages depending upon the size and life span. During the first and second stage, the larva has segmental hooks which are directed backward. These hooks help the larva to anchor itself to the surrounding tissue. The presence of these hooks made removal of the larva from its host difficult. The larval stage lasts from six to eight days in which period they are parasitic to human beings. The larvae are photophobic and therefore they hide themselves deep into the tissues and also to secure a suitable niche to develop into pupa. The host tissue is destroyed due to the release of toxins by the larvae. The surrounding bacteria decompose the tissue due to the release of proteolytic enzymes and the larvae feed on this rotten tissue. The infected tissue frequently releases foul smelling discharge. The interaction of toxin or enzyme released by the larvae-bacteria can also cause bony erosion.[10,11]

The developmental transition via the larval stage requires moisture, necrotic tissue and suitable temperature in an intermediate host. The number of larvae which are developing depends on the number of viable eggs deposited in the tissue. Oral myiasis which is a parasitic entity is mainly seen in tropical countries like India due to warm and humid environment that aids their breeding.[12]

In literature males have been found to be more commonly affected than females due to their more outdoor activities and negligience towards oral hygiene, which is in agreement with the finding of the present case. The lesions are usually located in the anterior part of the oral cavity affecting the anterior segments of both jaws and the palate which suggests direct inoculation of the tissues. Rarely, posterior portions of the oral cavity are involved due to ingestion of infected material.[13]

Low socioeconomic status, immuno compromised state, neglected oral hygiene, debilitated and unhygienic living conditions and alcoholism were the main contributing factors responsible for myiasis in the present case. It was predicted that the flies were attracted to the bad mouth odour due to negligence of oral hygiene or fermenting food debris and also due to the proliferating lesion from the oral cavity. Persistent mouth opening also facilitates in the deposition of the eggs by the adult fly. The association of Myiasis with oral malignancy can be explained as the fungating and necrotic wounds are common among cancer patients in India because many patients have advanced neglected tumors because of low socio-economic status and lack of knowledge.[14] A rare case of nasal myiasis with palatal complication has been reported in a 15 year old.[15] In the present case the patient was an alcoholic and belonged to low socioeconomic status with poor sanitary condition.

Extensive oral Myiasis infestation associated with squamous cell carcinoma in the head and neck region are reported in the literature, but are very few.[16,17,18,19,20,21] Our case also showed an association with well differentiated squamous cell carcinoma.

Treatment and Management: The treatment and management of a patient with myiasis includes mechanical removal of larvae along with use of turpentine oil which aids in blocking their respiratory sinuses which forces them to emerge on the surface resulting in an increase in their accessibility. Ether, iodoform and choloroform are the other such agents which can also be used.[13]

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Nutritional support of the patient was maintained in addition to multi vitamin tablets were prescribed to the patient. Secondary bacterial infection in surrounding skin with antibiotics was essential in our line of treatment. Generally the antibiotic regimen used may include ampicillin, amoxicillin or metronidazole.[22]

Systemic ivermectin has been tried in many patients with successful results. Ivermectin is a semi- synthetic macrolide antibiotic, obtained from Streptomyces avermitilis, which is the only antibiotic which has been found to be effective against maggots. Ivermectin can be administered via oral or intravenous route. Mechanism of action of ivermectin is through blocking nerve impulses on the nerve endings of the larvae through the release of gamma aminobutyric acid (GABA) and linking to the receptors, which causes palsy and death.[23,24]

Malignancy should be managed surgically followed by reconstruction.

In the present case, elimination of the maggots was achieved with turpentine oil with manual debridement and the patients were administered systemic antibiotics. With administration of systemic ivermectin. Incisional biopsy was done which was diagnosed as moderately differentiated squamous cell carcinoma. Surgical resection with pectoralis major myocutaneous flap with reconstruction was planned, however, patient withdrew from treatment and was lost to follow up. After six months relatives reported that patient succumbed to the disease.

Take Home Message:

- Educate and sensitizing the general population for maintaining good personal and oral hygiene, especially the fly population.
- Special care needs to be taken in medically compromised, dependent patients as they are unable to maintain their basic oral hygiene.
- Early diagnosis can prevent deeper penetration facilitating diminution of morbidity and mortality.

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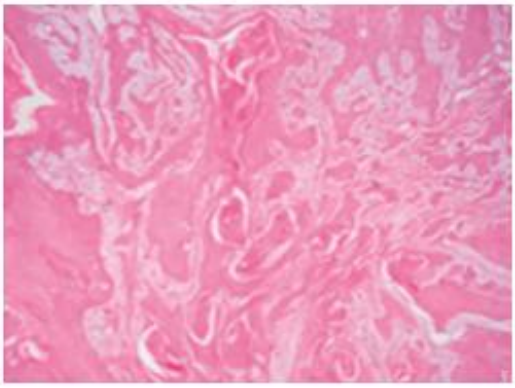

Figure 1

Figure 2

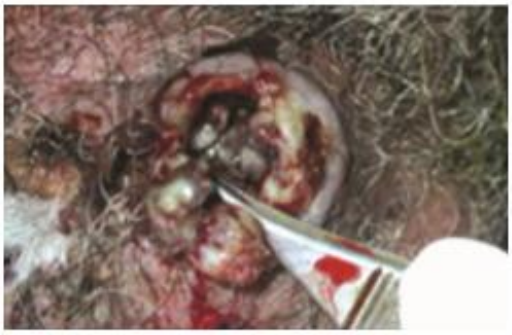

Figure 3

Figure 4

Figure 5

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