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Cover Page Footnote

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CASE REPORT

Primary Oral Myiasis of Anterior Maxilla in An Intellectually Disabled Female – A Case Report

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ABSTRACT

"Myiasis" is a term used to represent invasion caused by fly larvae that affect organs and tissues of human beings and feeds on the host tissue. It is a rare and non-specific pathology that varies according to the species of fly larvae and the area of the body involved. Various factors like extraction wounds, uncontrolled diabetes, necrotic tissues, alcohol addiction, poor oral hygiene, immunocompromised conditions and people with special care needs like debilitated or mentally challenged individuals predispose to develop myiasis. We hereby report a case of oral myiasis of the maxillary anterior region of the palate and labial mucosa, in a 16-year-old female patient with special care needs who for her daily activities is dependent. The diagnosis was confirmed by the presence of pulsating larvae in the lesion and was managed by conservative suffocation therapy involving the turpentine oil topical application followed by the mechanical removal of larvae along with surgical debridement and post-operative antibiotics.

Key words: antibiotic therapy, larva, maxilla, myiasis, palate, parasitic diseases, turpentine

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INTRODUCTION

The Reverend Frederick William Hope coined the name myiasis in 1840, assuming it was derived from the Latin words 'muia,' meaning fly, and 'iasis,' meaning disease. Laurence was the first to describe oral myiasis in 1909. It was later explained as a dipterous larvae infestation on live human animals and vertebral animals that nourishes the animal host's dead or alive tissue, liquid body contents, or swallows the food in ill people for at least a specific period.¹

Myiasis has been reported in warm, humid environments such as the tropical or subtropical regions of Southeast Asia, America, and Africa. Since the oral cavity rarely supplies the essential habitat for the larval lifecycle, oral myiasis is a rare condition. As a result, the incidence of cutaneous myiasis is higher in sites other than the oral cavity, such as the nose, eyes, ear, vagina, skin, and nasopharynx.²

Incompetent lips, poor oral hygiene, anterior open bite, nocturnal mouth breathing, extraction wounds,

facial trauma, and ulcer-like lesions are all risk factors for the development of oral myiasis.³ Due to its ease of access, the anterior part of the maxilla is the most commonly affected site, where substantial tissue loss can be detected, beginning with tiny ulcers holding living larvae. The presence of live larvae is a defining factor in the diagnosis of myiasis.⁴

Oral myiasis is a rare but dreadful and debilitating infection that affects patients with special care requirements due to their inability to express their concerns to their parents or physicians. Therefore, understanding the nature of this disease, its origin, treatment, and preventative measures are critical for oral health care practitioners. Therefore, the current paper presents a case of primary oral myiasis of the maxillary anterior region of the palate and labial mucosa in a 16-year-old female patient with special healthcare needs.

CASE REPORT

A 16-year-old female patient, accompanied by her parents, presented to the outpatient department of Oral Medicine and Radiology at a private institute with a primary complaint of swelling, pain, and discomfort in the upper front teeth region of the jaw for 2-3 days. The patient's caregiver noted gingival swelling and growth with live larvae in the maxillary anterior region, as well as bleeding that caused pain in the patient with no history of associated malodor. The patient had a habit of mouth breathing, and further history revealed that the patient had an intellectual disability; therefore she was dependent on her caregiver for daily activities. Due to weak or poor motor control, the patient had altered salivary flow that resulted in the drooling of saliva and involuntary hand and arm control was the root cause for her inability to brush her teeth. These conditions predisposed her to have a poor oral hygiene that lead to periodontal diseases and other dental problems.

Extraoral examination revealed diffuse swelling of size 2.5 x 2 cm that was soft and non-fluctuant near the left anterior cheek region with no involvement of lymph nodes. Palpation of the swelling revealed no tenderness. In addition, the patient had incompetent lips with increased labial fullness (Figure 1). Intra orally, erythematous, and edematous gingiva were noted from the 12-26 region both labially and palatally, along with bleeding on probing. Friable gingiva was noted to result in exposure of the underlying bone periosteum, forming a dome-shaped "warble" seen solely on the palatal aspect. Fistulas of the labial vestibule on either side of the frenum were noted, along with many live pulsating larvae that tried to ooze out of the fistula which were several, white in color, had a conical shape morphology with a pointed anterior end and a black posterior end.

Based on the presence of live maggots (Figure 2) and the patient's principal complaint, history, and clinical examination, the present case was tentatively classified as primary oral myiasis of the maxillary anterior region of the palate and labial mucosa. Informed consent was obtained from the patients' parents to continue with further care and management.

Radiological examination was performed to determine the extent of the underlying bone injury. Intra-oral periapical radiographs (IOPAR) of teeth 11-13 and 21-23 teeth revealed radiolucency in the periapical regions of 11, 12, 21, and 22; horizontal bone loss of 2-3 mm apical to the CEJ; and altered trabecular pattern (hazy radiolucency) suggestive of localised periodontitis in teeth 11-13 and 21-23 teeth. Figure 3 shows a maxillary anterior occlusal radiograph with characteristics similar to those reported by IOPAR. An orthopantomogram was not considered because of the poor motor control.

Given the high morbidity, conservative care with suffocating therapy with turpentine oil followed by surgical debridement was planned. The afflicted spots were thoroughly investigated under aseptic conditions. Cotton pellets soaked in turpentine oil were kept in cavitations for 5 - 10 min before the maggots were harvested using cotton tweezers in an exploratory technique after the cotton was removed. The treatment was performed several times in a single visit, combined with intermittent, profuse normal saline irrigation for three consecutive days separated by 48 hours. Exploration of the wound revealed a plethora of burrows and tunnels, several of which were inhabited by live larvae. When feeding on surrounding tissues. the larvae had tunneled into the deep soft tissues, detaching the gingiva and mucoperiosteum from the bone (Figure 4).

During the first visit, approximately 24 live maggots were removed (Figure 4). After debridement, the wound was irrigated with 3 percent hydrogen peroxide diluted in normal saline. Susp. Amoxicillin 250mg + clavulanic acid 125mg, Susp. Metronidazole 200mg, and Ibuprofen 100mg + paracetamol 162.5 mg were prescribed as 10 ml b.i.d. for 7 days.

The same process was performed at the second visit, resulting in the removal of approximately 22 live maggots, followed by surgical curettage, debridement, irrigation, and post-operative instructions (Figure 5). The method resulted in a lack of maggots on the third visit; therefore, only surgical debridement with povidone-iodine mixed in saline irrigation was performed (Figure 6). Three sessions of suffocation therapy resulted in a total of 46 live maggots. Hence turpentine oil was enough to remove the live maggots, and further management was supplemented by the use of antibiotics. The patients' caretakers were instructed to continue the drugs for three days and to return for review after three, seven, and fifteen days. During the follow-up appointments, healing was deemed satisfactory.

Caretakers play a crucial role in maintaining the oral hygiene of patients with intellectual disability and hence they should be trained adequately in both prophylactic measures (adaptation of food according to masticatory ability of the patient, prevention of aspiration of food and other objects, prevention of localized source of infection i.e. calculus deposits, proper brushing and oral hygiene maintenance) as well as therapeutic measures (identifying cause of localized pain or discomfort to the patient, bringing patient to dentist for regular follow ups). They should be well equipped in coping strategies as these patients are generally not well cooperative. So strategies such as supervision of the brushing, performing oral hygiene during the bath, seeking the help of the qualified dentist,



Figure 1. Facial profile at Day 1

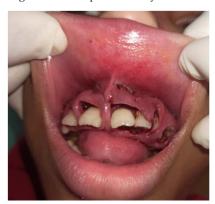


Figure 2. Day 1 – Pre-operative intraoral view







Figure 3. Intraoral periapical radiographs of 11-13 and 21-23 and maxillary anterior occlusal radiograph reveals horizontal bone loss and ill defined radiolucency at the periapical region of 11, 12, 21 and 22.

utilizing physical restraints e.t.c are employed to deliver oral hygiene care to patients with special health care needs. Caretakers must identify the obstacles to dental health, overcome informational obstacles, control the infection, overcome physical and behavioral obstacles and provide unique oral health care meeting the specific need of the patient.

DISCUSSION

Parasitosis is a parasitic infection or infestation in which a person is infected or infested with parasites. Infestations were first documented in 1840, with open wounds and dead tissue serving as the principal sites of infection as well as the involvement of body cavities



Figure 4. Day 1 postoperative with removed maggots.



Figure 5. Day 2 postoperative with removed maggots



Figure 6. One week post operative

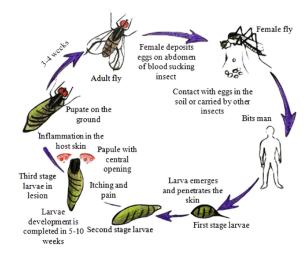


Figure 7. Lifecycle of lava causing oral myiasis

such as the ear, nose, and oral cavity.² Oral myiasis is a secondary, unpredictable condition caused by the invasion of such parasites by live fly larvae. Shira documented the first instance of oral myiasis in 1943, and Lim conducted a literature analysis of this illness

in 1974.⁵ The most prevalent causative species of oral myiasis are divided into four families: Calliphoridae, Sacrophagide, Oestridae, and Muscoidea.⁶ The most prevalent larval species that cause myiasis in India belong to the Muscidae family, particularly M. *domesticus*, also known as the housefly. The life cycle of the larvae from its initial stage of contact with the host tissue until its complete development is depicted in the Figure 7.⁷

As larvae have a strong preference for warm and humid temperatures, tropical conditions promote their growth; therefore, these conditions are restricted to tropical and subtropical locales.8 People who are unwell, mentally challenged, or elderly are more likely to develop myiasis because they rely on their families for everyday duties. As a result, the caregivers must be aware of how to detect, avoid, and manage sickness. Poor oral hygiene, as well as other predisposing conditions such as poor nutrition, decreased body resistance, alcoholism, neurological disorders, oral and maxillofacial trauma, mouth breathing, incompetent lips, or inability to close the lips due to malocclusion, are just a few of the factors that attract flies for larval deposition.⁹⁻¹¹ Maggots are conical in appearance, with a pointed anterior end that resembles a hook, and a blunt black posterior end that is sometimes misidentified as the eyes of the larvae. However, the black posterior end is the part through which the larvae breathe as they feed on the deep tissues, and is known as the spiracle, the larvae's respiratory apparatus.

Due to her neurological state, the patient was fully dependent on her caregiver in this case, and this dependence, combined with poor dental hygiene, mouth breathing, and lip incompetence contributing to a lack of lip seal, resulted in infestation of larvae in the oral cavity. Because she was unable to speak or articulate the symptoms of infestation due to aphasia, the caregiver observed the indication of abnormality in this case, which was pulsating live larvae in the oral cavity, a hallmark clinical sign of myiasis. ^{12,13} The inability of the patient to and comply limits the professional's ability to intervene. ¹⁴ Therefore, regular examination and evaluation of the oral cavity are needed as preventive measures to avoid the occurrence of such conditions or other oral pathologies in the future.

Oral myiasis is classified into two categories based on the sequence in which it occurs: primary, in which the infection is confined to the initial site, such as in cases of palatal or maxillary sinus involvement; and secondary, in which the infection infiltrates from the primary site and lodges elsewhere. Primary myiasis occurs when larvae feed on living tissues, whereas secondary myiasis occurs when larvae feed on dead tissues. A classification system was developed based on the presence of larvae and their long-term survivability, with two basic categories: anatomical and ecological. The larvae are anatomically classified as

migratory, furuncular, bloodsucking, sanguinivorous, cutaneous myiasis, cavitary myiasis, and wound myiasis. There are three types of myiasis: 1) pseudo or accidental (ingestion of larvae with food), 2) obligatory or specific (affecting intact skin), and 3) facultative or semi-specific.¹⁶

Current literature reveals that oral myiasis is broadly classified into^{17,18}

- 1. Larvae were found to be active outside the body.
- 2. Larvae develop below the uninterrupted skin where they hide.
- 3. Larvae live in cavities in the urinary and gastrointestinal tracts.
- Young larvae or eggs are deposited within wounds or normal cavities inside the body.

From the above-mentioned classification, it is clear that the present case is related to the fourth variant of the classification system.

Oral myiasis can be treated using several methods. The most common method is mechanical removal or surgical debridement of the larvae under local anesthesia supplemented with appropriate antiseptics, antibiotics, or anthelmintic therapy. To eject maggots, chemicals such as turpentine oil, negasunt, ethyl chloride, ether, mercuric chloride, creosote, saline, iodoform, chloroform, clove oil, calomel, phenol mix, petroleum jelly, olive oil, gentian violet, camphor, and sodium hypochlorite were initially utilized. 19,20 Aerobic larvae asphyxiate and migrate to the shallow region of the cavitations, which assists in the evacuation of these larvae with minimal injury to the tissues and larvae.²¹ Surgical debridement was performed under local anesthesia, with access to the afflicted location acquired through a sulcular incision and flap reflection. After exposure, larvae were removed using hemostatic or clinical forceps and sutured.

In this case, turpentine oil was used to remove larvae from the affected location. It causes reversible tissue necrosis, which leads to epithelial hyperplasia, hyperkeratosis, and ulceration, all of which regress once the stimulus is removed.¹⁷ Turpentine oil is one such asphyxiating agent that acts as a topical irritant, blocking the larvae's respiratory sinuses, thereby making the larvae forcibly to come out in search of oxygen. This helps in the removal of the larvaes with the help of forceps or tweezers. Usage of chemicals in suffocating or asphyxiating the larvae and removing them is also known as suffocation therapy. Also care should be taken to prevent the laceration or disruption of larvae as any remnants of larvae can lead to undesirable inflammation and further infection which inturn is difficult to manage. Ivermectin is a semi-synthetic macrolide antibiotic that produces gamma-aminobutyric acid, which attaches to larvas' receptor and causes the organism to die.²² It is an anti-parasitic drug isolated from *Streptomyces* avermitillis that can be administered in 6 mg doses once a day or routinely for 3-5 days.²³ Ampicillin, augmentin,²⁴ polymyxin B,⁸ and cefazolin are examples of broad-spectrum antibiotics that can be administered alone or in conjunction with metronidazole.¹⁰ Ivermectin use can cause dizziness, fever, migraines, muscular and joint pain, and cutaneous eruptions.¹⁶

Oral hygiene is an important aspect for individuals with intellectual and developmental disabilities, as they depend on their caregivers to provide it. Hence, knowledge of various normal and abnormal common oral conditions is necessary for caregivers to improve the quality of life of such patients. Proper training of caregivers can help them identify the oral conditions that require interventions at an early stage, which can help them to reduce or prevent common oral diseases such as gingival or periodontal conditions, thereby improving the overall health care of a vulnerable population. In addition, patient behavior can also be a concern, as it creates an additional barrier for assessing dental care because it either makes parents hesitant to bring their child to the dentist or makes it difficult for the dentist to manage them to carry out any dental treatment accordingly. Therefore, a collaborative approach between dentists and caregivers is required to benefit patients with intellectual or developmental disabilities.

CONCLUSION

As the cliché goes, "prevention is better than cure," and this is especially true in the case of oral myiasis, where greater personal care and quality of life combined with an early and accurate diagnosis can block the growth of the disease. The general population should be aware of the importance of oral health through health campaigns. Patients who are physically or intellectually challenged require special attention when it comes to oral hygiene, and it is our moral obligation as dentists to ensure that these patients have access to all available therapies for successful management of this pathological condition. In order to prevent re-infestation after treatment, patient caregivers should be counseled regarding the maintenance of proper oral hygiene.

CONFLICT OF INTEREST

None declared.

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We would like to thank Dr. S M Kotrashetti, Professor, Department of Oral and Maxillofacial Surgery for guiding the suffocation therapy and Dr. Ram Surath Kumar for his contribution in pictorial representation of life cycle of larvae. We also thank Dr. Nagarajan V, Associate Professor and Head, P.G. Department and Research Centre in English, for his significant contribution in language editing service.

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