Case Report

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Management of oral myiasis: A rare entity

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Abstract

Myiasis is one of the foremost daunting parasitic infestations. Although pandemic, it is more often found in tropical and subtropical countries where poor hygiene, poor housing infrastructure, warm humid climate and proximity to domestic animals prevail. Mostly it occurs secondary to serious medical conditions such as cerebral palsy, cancerous lesions. Although many treatment protocols are available, the standard treatment focuses on antibiotic therapy concomitantly with mechanical removal of larvae and necrotic tissues. This is a case of intraoral myiasis in a twenty-year old female patient with cerebral palsy that was managed using the suffocation approach and mechanical debridement.

Key words: Cerebral palsy; Maggots infestation; Maggot; Myiasis.

INTRODUCTION

Myiasis is the infestation of living tissues caused by the larvae of fly species within the arthropod order Diptera. The word myiasis is derived from the Greek word "myia," which means fly[1].

It was first described by Laurence and F. W. Hope in 1909. Zumpt coined the term 'myiasis' in the year 1940 for the condition in which the dipterous larvae invade the human or other vertebrate animals and feed on host's dead or living tissue, liquid body substances, and ingested food for certain period of time[2]. The various species which cause myiasis include C. hominivorax, Oestrus ovis, Wohlfahrtia magnifica, Chrysomya bezziana, Hypoderma bovis, Cordylobia anthropophaga, Hypoderma tarandi, Calliphora vicina, Musca nebulo, Musca domesticus and Lucilia sericatab[3-5]. The various sites affected include head and neck region, skin, anus, and vagina[1]. The standard treatment focuses on antibiotic therapy concomitantly with mechanical removal of larvae and necrotic tissues[6]. Surgical debridement of the wound and extraction of larvae are most commonly done under local anaesthesia or general anaesthesia[1]. The suffocation approach forces aerobic larvae to surface in search of air where they can be removed with the aid of forceps or tweezers[7]. Due to the underlying systemic conditions and the prevailing condition of the patient, treatment under general anaesthesia was thought to be a risk factor for the patient. Hence, we tried the suffocation technique with turpentine oil.

CASE REPORT

A developmentally disabled bedridden 20-year-old female patient was brought to the department of Paediatric and Preventive Dentistry with the complaint of peeking of worms from the oral cavity for two days. The patient's mother informed that the patient was apparently asymptomatic two days back, later she noticed two worms coming out from the oral cavity.

Complete history revealed that patient was diagnosed with cerebral palsy since birth, followed by severe mental retardation. The developmental milestones were delayed

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and she developed spastic quadriplegia (cerebral palsy involving all the four limbs), kyphoscoliosis along with impairment of speech. Familial history revealed that there is no history of any consanguineous marriage. The patient had two siblings, who were healthy with no diagnosed disorders. The patient was completely dependent on her mother for her day-to-day activities including hygiene and nutrition.

General examination revealed severe wasting of all the forelimbs and pigmentation at the joints along with kyphoscoliosis. Extraoral examination revealed swelling in the infraorbital region on the left side, extending from lower border of eye to the ala of the nose and in the premaxillary region extending in between the nasolabial folds. The region over the swelling was normal, with no rise in temperature and no evidence of discolouration. Due to speech impairment, tenderness on palpation was not elicited by the patient. Intraoral examination revealed a full set of permanent teeth except the third molars, poor oral hygiene, class I molar relation with increased overjet and mouth breathing habit. A traumatic wound was noticed in the anterior maxillary region in the labial gingival tissue region extending from the right lateral incisor to the left canine. Gingival inflammation and recession were seen in the maxillary anterior teeth region along with peeling of the gingival tissue. Few maggots were seen peeking from inside of traumatic wound. Gentle lifting of the mucosa revealed multiple white coloured wiggly worms. Based upon all these features the patient was diagnosed by primary and accidental type of myiasis (Kettle's classification - 1984, Patton – 1922)[8].

A suffocation technique with turpentine oil was used for removal of worms[9-10]. A gauge pack was prepared by adding a few drops of turpentine oil to it. The gauge was packed in the anterior maxillary vestibular region and kept in place for 10 to 15 minutes, after which the larvae were removed with the help of tweezers. No local anaesthesia was administered during the procedure. There was no evidence of bleeding from the traumatic wound as the larvae caused the necrosis of the underlying tissues.

The treatment was performed in two sittings and in each sitting approximately 7-8 larvae were removed with the help of tweezers. The second sitting was performed after three days as the approximate life cycle of the larvae is 50–60 hours[11]. The larvae were cylindrical in shape and creamy white in colour with tapered heads and high motility. The larvae were approximately 1.2 cm in length and in the head region a pair of dark hooks were evident. These larvae were preserved in 10% formalin and sent to the department of pathology for identification, where they were identified as larvae of Musca domestica (common housefly).

Systemic antibiotics, syrup Augmentin - amoxicillin/clavulanate potassium (288mg/5ml) thrice daily, syrup Flagyl - Metronidazole (400 mg/5 ml) twice daily, Pan 40 - Pantoprazole twice daily were prescribed for five days, along with Quadrajel ointment (chlorhexidine gluconate1% W/W, lignocaine 2% W/W, metronidazole 1% W/W) for local application.

The parents were educated and motivated regarding the benefits of good personnel hygiene and nursing care. The closure of the traumatic wound was appreciated three days after using the turpentine oil. The patient was kept under review for one month and no recurrence was detected.

Figure 1: General examination.

Figure 2: Frontal view.

Figure 4: Removal of larvae.

Figure 3: Intraoral photograph.

Figure 5: Mechanical debridement.

Figure 6: Larvae removed from wound.

DISCUSSION

Myiasis is usually among the five most common dermatologic conditions, accounting for 7.3% to 11% of cases[12]. The prevalence of the disease is high in tropical and subtropical regions such as India, Tunisia, Brazil, and Australia[3,13]. Oral myiasis is a rare pathology in humans compared to other forms of myiasis, as the tissues of the oral cavity are not permanently exposed to the external environment[14-16]. Males are commonly affected than females[17].

Aetiology: At least 86 different species of Diptera can infect man with larvae that invade skin and body cavities[14,18]. Myiasis is commonly caused by dipteran clade Calyptratae which consists of four families: Calliphoridae, Sarcophagidae, Oestridae and Muscoidea[19].

Classification: Myiasis has been classified depending on various factors such as:

- location on the host body
- type of host parasite relationship
- type of tissues fed by larvae (clinical classification)
- condition of tissue involved

Based on location on the host body[20]:

- Dermal, subdermal, nasopharyngeal, internal organs and urogenital.

Based on the type of host parasite relationship[21]:

- Obligatory, facultative, or pseudomyiasis.

Clinical classification depending upon the type of tissues fed by larvae[22]:

- Primary myiasis is caused by biophagous larvae feeding on living tissue and is rare in humans.
- Secondary myiasis caused by necrobiophagous flies feeding on dead tissue in a necrotic cavity or lesion.

Depending upon the condition of tissue involvement[14]:

- Accidental (larvae ingested along with the food),
- Semi-specific (larvae laid on necrotic tissue in wounds), and
- Obligatory (larvae affecting the undamaged skin).

Clinical signs: Head and neck region, skin, anus and vagina are the sites most commonly affected. In the head and neck region, ears, eyes, oral cavity, nose, paranasal sinuses, lymph nodes, mastoid region and tracheostomy wounds are mostly affected[1,13,23,24]. The anterior part of the oral cavity is more commonly affected than the posterior because it is easily accessible to flies[25-27] and the sites most commonly involved are the anterior segments of the maxillary and mandibular jaws and the palate[4,14].

The various risk factors include poor oral hygiene, advanced age, low socio-economic status, medical comorbidities such as a history of craniomaxillofacial trauma and malignant tumors,[28] cerebral palsy,[21] neglected fractures,[29] poor oral hygiene,[30-31] mouth breathing, anterior open bite,[32-33] cancrum oris,[28] patient undergoing radiotherapy,[34] person living in close proximity to animals[18] and debilitated patient with neglect of nursing[35].

The maggots cause infestation to the humans either by direct inoculation into wounds or through the ingestion of infected materials like meat. Flies which act as the intermediate host, lay approximately 500 eggs in the tissues. The larvae hatch in about eight to ten hours, after which they invade into the surrounding tissues and cause inflammation and discomfort to the patient due to the decomposition of the tissue caused by the proteolytic enzymes released by the surrounding bacteria[36-37].

Larvae form a fibrous capsule and firmly adhere to it and this poses a difficulty in dissection during surgical procedure. As the larvae are photophobic; they tend to hide deep into the tissues for a suitable niche to develop into pupa[38].

The burrowing of the larvae causes the separation of the mucoperiosteum from the bone creating a patent opening with induration of the marginal tissues and it appears as a dome shaped "warble". Infestation is mostly seen subcutaneously and it produces a furunculated or boil-like lesion, also called as "berne". Larvae position themselves with their heads down to expose their posterior spiracles to the air, which makes their respiration possible. The backward segmental hooks are useful for the anchoring of the larvae to the surrounding tissue[14,39].

Treatment: Although a variety of treatments oppose the lack of a protocol, the standard treatment focuses on antibiotic therapy concomitantly with mechanical removal of larvae and necrotic tissues[6]. Surgical debridement of the wound and extraction of larvae are most commonly done under local anaesthesia or general anaesthesia[1]. The suffocation approach forces aerobic larvae to surface in search of air where they can be removed with the aid of forceps or tweezers[7].

Various substances like mineral oil, ether, oil of turpentine, [9-10,14,29] chloroform,[40] mercuric chloride, ethyl chloride, creosote, phenol, saline, calomel, gentian violet, white head varnish, olive oil, petroleum jelly, heavy oil, beeswax, nail polish, adhesive tape, butter, chewing gum, native tobacco leaf and iodoform can be used for the suffocation approach[7,39,41-42]. Broad spectrum antibiotics such as ampicillin, amoxicillin or metronidazole and topical use of nitrofurazone and ivermectin can be advocated along with multivitamin tablets for nutritional support[22].

Ivermectin is administered as a single dose of 150-200 µg/kg body weight. It is rapidly absorbed and reaches high blood concentrations within a relatively short

period of time due to which the larvae are quickly expelled from the wound. Ribeiro et al. suggests the oral administration of ivermectin at a dose of up to 300 µg/kg [that is, the patients weighing 40 to 60 kg: two tablets (12 mg) and patients weighing 60 to 90 kg: three tablets (18 mg)] for three consecutive days, does not cause any intercurrence and eliminates all larvae in cavitary myiasis. Shinohara et al. and Gealh et al. recommend the use of ivermectin without debridement for patients with cutaneous myiasis[22,33,43].

Secondary infection of the wound by bacteria is uncommon because of the bacteriostatic activity in the gut of larvae, which prevent the undesirable overgrowth of pyogenic bacteria[44]. In cases of secondary infection, systemic antibiotic therapy alone can be beneficial[40,45].

Preventive strategies: Control measures of the fly population have emerged as an alternative strategy to prevent infestation. Basic sanitation and eradication of foci of adult fly proliferation are the first steps in the combat of this disease. Limiting myiasis directly relates to curbing the fly population by efficient waste disposal supplemented by spraying with an insecticide, providing the patient with a physical barrier and good personnel and nursing care[14,46].

Complications: The sequelae of myiasis include blindness, hearing loss, massive tissue destruction, salivary gland involvement, in addition to the development of an oroantral communication and considerable bone and soft tissue loss and even death may also occur.

Even though myiasis may be self-limiting and non-fatal in some cases, the patient and relatives report with huge psychological distress. Few larvae can destroy vital tissues, inducing serious or even life-threatening haemorrhage[43].

Thus, establishment of the diagnosis and immediate treatment are necessary to interrupt progression of the disease and to prevent more devastating sequelae.

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