CASE HISTORY REPORT

ABSTRACT

Myiasis is a condition caused by the invasion of tissues by larvae of Diptera flies. This phenomenon is well documented in the skin especially among animals and people in tropical and subtropical areas. The condition causes extensive tissue destruction as the larvae, at least for a certain period, feed on the host's dead or living tissue, liquid body substances, or ingested food. Mouth breathing during sleep, poor oral hygiene, alcoholism, senility, mental disability, cerebral palsy, and hemiplegia may facilitate the development of myiasis. We present a case report of oral myiasis in a 22-year-old male with cerebral palsy and severe mental retardation treated successfully by manual removal of the larvae by topical application of turpentine oil and oral systemic therapy with ivermectin.

KEY WORDS: oral myiasis, child, management, mental retardation, cerebral palsy, ivermectin

Oral myiasis: a case report

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Introduction

The term myiasis is derived from the Latin word "muia" which means fly and "iasis" means disease. The term was coined by Hope in 1840.[1] Laurence first described oral myiasis in 1909.[2] Myiasis has been defined by Zumpt (1965) as the infestation of live human and vertebrate animals with dipterous larvae, which, at least for a certain period, feed on the host's dead or living tissue, liquid body substances, or ingested food.[3] The incidence is higher in tropical and subtropical zones of Africa and the America. The flies responsible for the condition prefer a warm and humid environment; therefore myiasis is restricted to the summer months in temperate zones while it can be seen all year round in the tropics.[4]

Various classifications have been suggested in the literature. Clinically, myiasis can be classified as primary and secondary.[5] *Primary myiasis* is caused by larvae which feed on live tissue, which is commonly seen in cattle, and is rare in humans. *Secondary myiasis* is caused by flies that feed on dead tissue. This is more common and seen in patients with necrotic cavity lesions. Myiasis can be classified, depending on the condition of the involved tissue,[6] into *accidental myiasis*, when larvae ingested along with food produce infection, *semi-specific myiasis* where the larvae are laid on necrotic tissue in wounds, and *obligatory myiasis* in which larvae affect undamaged skin. Based on the anatomic sites, affected myiasis is subdivided into cutaneous myiasis, myiasis of external orifices (aural, ocular, nasal, oral, vaginal, and anal), and myiasis of internal organs (intestinal and urinary).[7]

This paper presents a case of oral myiasis in a 22-year-old male with cerebral palsy and mental retardation.

Case report

A developmentally disabled 22-year-old male was brought to the Department of Pediatric Dentistry with a chief complaint of worms in the mouth for 3 days. Complete history revealed that patient was diagnosed with cerebral palsy, severe mental retardation, quadriplegia, and kyphoscoliosis. He also suffered from seizure disorder, which was controlled with medication (carbamazepine, 100 mg, twice/day.). He was also diagnosed with coloboma of the right eye 3 years prior. Familial history revealed a consanguineous marriage. He had two siblings, who were healthy with no diagnosed disorders.

Patient, who resided in a rural setting on the outskirts of Mumbai, India along with his family, was completely dependent on his mother for his day-to-day cleanings and feedings. Mother gave a history of cleaning his oral cavity with only a wet cloth for the past 22 years. It was reported that the mother first noticed "worms" in his mouth 3 days back and

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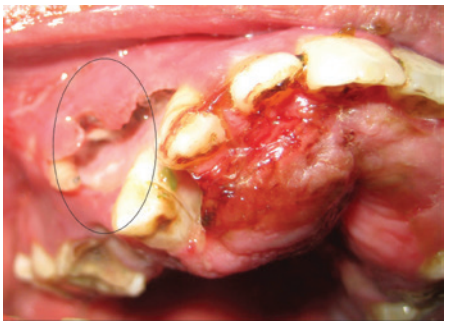

Figure 1. Ulcerative lesion on the buccal gingiva seen with a protruding larva.

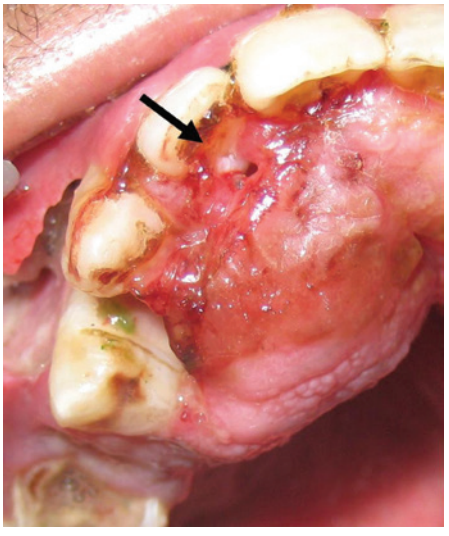

Figure 2. Palatal swelling seen with a protruding larva.

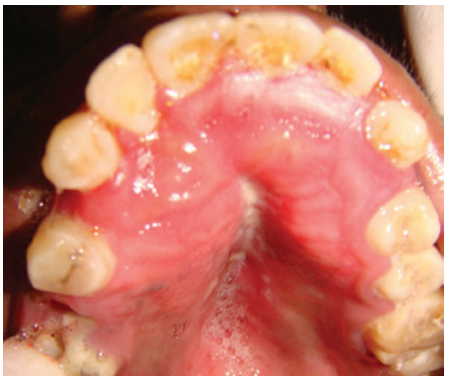

Figure 3. Satisfactory healing 1 month postoperatively.

Figure 4. Two of the larvae removed from the lesion.

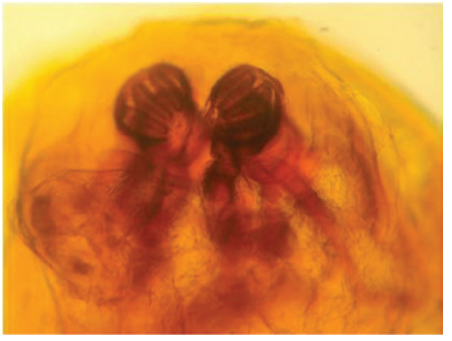

Figure 5. Microscopic examination of the larva.

sought consultation from a physician who then referred him to the department of pediatric dentistry clinics. It was reported that nine worms were removed from the patient's oral cavity in those 3 days at his residence using Nilgiri oil.

On examination, the patient was afebrile. He weighed only 16 kg. General physical examination revealed severe emaciation and kyphoscoliosis. Oral examination revealed incompetent lips, anterior open bite, and very poor oral hygiene with the presence of supra- and sub-gingival calculus. An ulcerative lesion was present on the buccal aspect of the gingiva extending from the mesial aspect of the maxillary right canine to the mesial aspect of the maxillary right first molar. Live larvae were seen protruding from this lesion (Figure 1). Also a soft, fluctuant, and erythematous swelling was observed extending from the palatal aspect of the maxillary right first premolar to the palatal aspect of the maxillary left first premolar. It had an orifice through which larvae were seen protruding out (Figure 2).

Patient was immediately hospitalized and intravenous antimicrobials including cefexime (750 mg, twice/day) and metronidazole (150 mg, thrice/day) were started to control secondary infection. A total of 13 maggots were removed manually by placing cotton pellets impregnated with turpentine oil at the orifice of the buccal and palatal lesions for approximately 10 minutes. Following this, the patient was given a single dose of an oral suspension containing 3 mg of ivermectin and 400 mg of albendazole. In following days no more maggots were found. After 5 days the patient was found to be in satisfactory health and was discharged.

Follow-up examination 1 month later revealed a decrease in the palatal swelling. The buccal lesions had healed satisfactorily (Figure 3). Mother reported absence of maggots in the healing wound.

Parasitology

The larvae that were removed measured 13 mm in length on an average, were whitish in color and segmented (Figure 4). The larvae were sent to the veterinary laboratory for parasitology. The specimens were treated overnight in 10% KOH, dehydrated in ascending grades of alcohol, cleared in clove oil and mounted on glass slides with Canada Balsam, and examined under the microscope. On the basis of microscopic examination of the cephalopharyngeal apparatus and the posterior peritremi, the larva was identified as Chrysomia species (Figure 5).

Discussion

Prevalence of oral myiasis in tropical countries may be more than that reported in the literature. Mouth breathing during sleep, poor oral hygiene, alcoholism, senility, mental disability, cerebral palsy, and hemiplegia may facilitate the development of myiasis.[8] Local factors such as halitosis can attract the flies. Tropical conditions are more

favorable for multiplication of the flies responsible for oral myiasis.

In this patient, poor oral hygiene, halitosis, and poor lip seal along with unhygienic living conditions may have exposed him to oral myiasis. Such cases have been reported in the literature.[5,9,10,12]

Flies causing myiasis belong to the family of Diptera and its seven different species (Calliphoridae, Sarcophagidae, Oestridae, Hypodermatidae, Gasterophylidae, Glossinidae, and Muscidae).[11] In the presence of favorable conditions, the female fly deposits its eggs. After hatching, the larvae develop in the warm, moist environment, burrow into oral tissues, obtain nutrition, and grow larger. This causes progressive tissue destruction and cavitation. The larval stage lasts for 6 to 8 days during which they are parasitic to human beings. The subsequent host tissue reaction produces a fibrous capsule to which the larvae adhere. The larvae have backward directed segmental hooks with which they anchor themselves to the surrounding tissue. The larvae also move out of the tissues through small orifices.[5] They are photophobic and tend to hide deep into the tissues for a suitable niche to develop into pupa. The diagnosis of myiasis at an early stage can prevent involvement of deeper tissues. The condition can be completely benign and asymptomatic, result in mild to acute pain, or in extreme cases cause death of the patient.[12]

Prevention of human myiasis involves control of fly populations and general cleanliness such as reducing decomposition and covering wounds. Proper oral hygiene is essential to ensure against oral myiasis.

The traditional management for myiasis involves mechanical removal of larvae. When there are multiple larvae, local application of several substances including iodoform, ethyl chloride, mercuric chloride, creosote, saline or turpentine oil or systemic butazolidine and thiobendazole have also been used to ensure the complete removal of larvae. Recently, systemic ivermectin has been used successfully in severe cases.[4]

Pharmacology of ivermectin

Ivermectin is a systemic antihelmenthic that was introduced for professional use in the 1980s. It is derived from a group of natural substance, avermectin, which is obtained from actinomycetes. The medicine was initially used in humans as a prophylactic in the treatment of filariasis. It has also been used in the treatment of scabies. For the treatment of myiasis, usually a single oral dose of ivermectin, 0.2 mg/kg body weight, has been recommended. It is contraindicated in children below 3 years of age or less than 15 kg body weight.[13] It is quickly absorbed and gives a high blood concentration in a relatively short period of time. Mechanism of action 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, causing palsy and death. Acetylcholine, which is the main peripheral neurotransmitter in mammals, is not affected by ivermectin, maintaining a security margin when it is used.

It is usually well tolerated by patients. The adverse effects of ivermectin include dermal eruptions, fever, dizziness, migraines, muscular pains, lymphangitis, and pain in joints.[14]

In our patient we chose to use systemic ivermectin in addition to mechanical removal of larvae to ensure complete control. Patient showed satisfactory healing of the oral lesion after this treatment.

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