Oral Myiasis: A Rare Case Report and Literature Review

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Abstract
Myiasis is a rare disease caused by infestation of tissue by larvae of flies. Oral myiasis is still “rare” and “unique” owing to the fact that oral cavity rarely provides the necessary habitat for a larval lifecycle. Herein, we present a case of extensive gingival myiasis in a 12-year-old mentally retarded, epileptic child as well as a literature review.

Key words: Myiasis; Mouth; Epilepsy; Ivermectin

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INTRODUCTION
Although the term myiasis was coined by F.W. Hope in 1840, it was first described by Laurance in 1909 [1]. Myiasis has been defined as a pathological condition in which there is an infestation of living mammals with dipterous larvae, which, for at least a certain period, feed on living or dead tissue in the host and develop as parasites [2]. In humans, the most commonly affected sites are the nose, eyes, skin wounds, sinuses, lungs, ears, gut, gall bladder, vagina, nasal cavities and rarely the mouth [3].

The occurrence of myiasis in the oral cavity is unusual as oral tissues are rarely exposed to the external environment [2]. Globally, a higher incidence of oral myiasis (OM) is observed in tropical and subtropical regions of Africa, America and South East Asia due to favorable climatic conditions [4]. It is classified as primary myiasis (caused by biophagous larvae also called obligatory myiasis) or secondary myiasis (caused by necrobiophagous larvae also called facultative myiasis) [1]. Oral factors like incompetent lips, poor oral hygiene, halitosis, anterior open bite, nocturnal mouth breathing, extraction wounds, facial trauma, ulcer-like lesions and oral carcinoma are predisposing factors for the occurrence of OM [3]. Oral myiasis caused by the Chrysomya bezziana (Cb) species is very rare in humans [5], which makes this case more unique.

CASE REPORT
A 12-year old boy was reported with a chief complaint of swelling in the maxillary anterior region especially in the gingiva. Presence of worms was reported in his mouth, noticed by his parents for two days. He reported a maxillofacial trauma a week ago, leading to avulsion of teeth #11 and #21.
The patient’s medical history revealed that he was epileptic (not under any antiepileptic drug) and mentally retarded since birth; consequently, he was unable to do his daily routine and required the help of his parents for daily activities. Intraoral examination revealed live larvae in the socket of tooth #11 and lingual sulcus of teeth #12 and #13 along with a swelling in the right premaxillary region and larvae showing a wriggling movement. There was an empty socket in place of teeth #11 and #21 and Ellis class I fracture in teeth #12, #13 and #22. Gingiva was red in color, soft and edematous in consistency and generalized bleeding on probing was observed. In addition, laceration of upper labial mucosa, incompetent lips, poor oral hygiene, halitosis and mouth breathing habit were also noticed (Fig 1). Radiographic examination could not be conducted due to the lack of patient cooperation. A cotton swab impregnated with turpentine was then placed at the opening of the socket for 5 minutes. Dozens of larvae rushed out from the infected area, measuring one inch in length, which were then manually removed one by one with the help of clinical forceps (Fig. 2) and sent for entomological examination on meat media.

The patient was then placed on oral ivermectin 6 mg once daily for three days and oral amoxicillin 250 mg three times daily for seven days. The procedure of manual removal of larvae was repeated once daily for three consecutive days. A total of 33 live larvae were removed from the wound. On the fourth day the oral cavity was free of larvae. The wound was debrided, together with adequate general care provided for the patient. Healing was uneventful. The patient’s parents were advised to maintain proper oral hygiene for their child and advised to rinse his mouth twice daily with 0.2% chlorhexidine gluconate for 15 days under supervision. The entomologist report was suggestive of Cb. On recall visit, oral prophylaxis was followed by topical application of 1.23% acidulated phosphate fluoride gel, restoration for teeth #12, #13, #22 and replacement of the missing teeth.

**DISCUSSION**

Literature review revealed that there are a number of species, which can cause OM. Multiple larvae infestation has been commonly reported [1-14]. Male predominance has been noted (because of their outdoor activities and negligence of oral hygiene) [2]. It may be proposed that this leads to halitosis leading to attraction of flies as seen in almost all cases. However, in some cases it may be due to serious medical conditions, which are debilitating.
These predisposing factors made our patient a prime target for the disease. The anterior part of the oral cavity including both jaws and the palate is commonly affected by OM suggesting direct inoculation of tissues [10] as described in the majority of cases but involvement of the posterior [8] areas of both jaws is rare. Clinically, OM may present as oral mucosal swelling [7], gingival inflammation [3,5,12], laceration [2,3,11] (as in our case), ulceration [1,6,11,13], periodontal disease [7,10], non-healing extraction wound [8], jaw bone fracture [2] and secondary infestation to cancrum oris [15]. Myiasis in human beings is caused by many species of larvae belonging to the order Diptera [5]. The Cb, (screw-worm fly), belongs to the genera Calliphoridae, whose larvae develop only in living tissues, and human cases of Cb infestations are rare [5,6,9]. The female adult Cb fly lays around 150–200 eggs on exposed wounds of mucous membranes of the nose, mouth and ear. The eggs hatch after 24 hours and the larvae thrive on living tissues for 5–7 days. They then wriggle out of the wound and fall to the ground to pupate.

The peculiarity of this maggot infestation is its ability to cause tissue invasion even without pre-existing necrosis [5]. The traditional management consists of manual removal of maggots. If there are multiple larvae in advanced stages of development and tissue destruction, local application of several substances will help remove all the larvae [1]. If mechanical removal is impossible, placement of a variety of substances like petroleum, nail polish, animal fat, bees wax, paraffin, hair gel and mineral oil will deprive the oxygen and help in resolution of infested wounds either by killing the larvae or by forcing them superficially where they can be removed [13]. Systemic prescription of oral ivermectin 6mg once daily for three days [13,16,17] (which is a semi-synthetic macrolide antibiotic derived from Streptomyces avermitilis) often results in recovery of patients suffering from OM. It acts by blocking the nerve impulses on nerve endings through the release of gamma amino butyric acid, which leads to paralysis and subsequent death of the parasite. It is also safe for human use. Flushing of gingival wounds with topical application of nitrofurazone has shown promising results with no further intervention [18].

CONCLUSION
Oral myiasis can be prevented by controlling fly population and maintaining personal and oral hygiene. Special attention is required for medically, physically and mentally compromised patients. Early intervention can help avert complications.

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