CLINICAL CASES

Dent. Med. Probl. 2014, **51**, 4, 553–557
ISSN 1644-387X

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Primary Oral Myiasis – Rare Case Report

Pierwotna muszyca jamy ustnej – opis rzadkiego przypadku

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A – research concept and design; **B** – collection and/or assembly of data; **C** – data analysis and interpretation; **D** – writing the article; **E** – critical revision of the article; **F** – final approval of article

Abstract

Oral myiasis is a rare disease caused by dipterous *larvae* that feed on the host's dead or living tissues, liquid body substances or ingested food. It is mostly reported in developing countries and in the tropics. Incidence of oral myiasis is comparatively lesser than that of cutaneous *myiasis* as oral tissues are not permanently exposed to the external environment. Cases of oral myiasis have been reported to occur following dental extraction, nosocomial infection, in drug addicts, visits to tropical countries, in psychiatric patients and conditions that are likely to cause prolonged mouth opening, like mouth breathing during sleep, senility, alcoholism and mental retardation. The flies are attracted to the fetid odor due to neglected oral hygiene or fermenting food debris. Persistent mouth opening facilitates the deposition of the eggs by the adult fly, with India's subtropical climate conducive to their breeding. Early removal of the maggots through various means is usually curative.

Herein, a case of oral myiasis in the maxillary anterior region of a 60-year-old mentally challenged female is being reported. The clinical findings, etiology and the importance of oral health in medically compromised people are also discussed (**Dent. Med. Probl. 2014, 51, 4, 553–557**).

Key words: myiasis, maggots, oral cavity.

Streszczenie

Muszyca jamy ustnej jest rzadką chorobą wywoływaną przez wędrujące larwy muchówek, które żywią się martwymi lub żywymi tkankami gospodarza, jego płynami biologicznymi lub trawionym pokarmem. Najczęściej była opisywana w krajach rozwijających się lub tropikalnych. Przypadki muszycy jamy ustnej są rzadsze niż skóry, ponieważ tkanki jamy ustnej nie są eksponowane w sposób ciągły na środowisko zewnętrzne. Opisywano je po usunięciu zębów, w zakażeniach szpitalnych, u alkoholików i narkomanów, po pobycie w krajach tropikalnych, u pacjentów leczonych psychiatrycznie mających często otwarte usta, u śpiących z otwartymi ustami, w wieku podeszłym, alkoholizmie i u osób upośledzonych umysłowo. Owady są przyciągane przez halitozę wywołaną zaniechaniem higieny jamy ustnej i fermentacją resztek pokarmowych. Trwale otwarcie ust ułatwia złożenie jaj przez dojrzałe latające muchówki żyjące głownie w Indiach i klimacie subtropikalnym. Szybkie usunięcie larw w różny sposób zawsze działa leczniczo.

W pracy przedstawiono przypadek muszycy w przedniej części szczęki u 60-letniej kobiety z zaburzeniami psychicznym. Opisano objawy kliniczne, etiologię i znaczenie zdrowia jamy ustnej u pacjentów obciążonych chorobami układowymi (**Dent. Med. Probl. 2014, 51, 4, 553–557**).

Słowa kluczowe: muszyca, larwy, jama ustna.

Myiasis is derived from the Greek word "*myia*", meaning fly and "*asis*", meaning disease. Myiasis is caused by dipterous *larvae* that feed on the host's dead or living tissues, liquid body substances or ingested food [1]. Myiasis was first described by F. W. Hope in 1840. Oral myiasis is a rare pathology in humans. The first case of oral myiasis was reported by Laurence in 1909. Incidence of oral myiasis is comparatively lesser than that of cutaneous myiasis as oral tissues are not permanently exposed to the external environment. Cases of oral myiasis have been reported to occur following dental extraction, nosocomial infection, in drug addicts, visits to tropical countries, in

psychiatric patients and conditions that are likely to cause prolonged mouth opening, like mouth breathing during sleep, senility, alcoholism and mental retardation [2]. The flies are attracted to the fetid odor due to neglected oral hygiene or fermenting food debris. Persistent mouth opening facilitates the deposition of the eggs by the adult fly, with India's subtropical climate conducive to their breeding [3].

Clinically, myiasis is classified as: (a) primary myiasis – when *larvae* feed on living tissue and (b) secondary myiasis – when *larvae* feed on dead tissue. Depending upon the condition of the involved tissues it is of 2 types: (a) accidental myiasis – when *larvae* get ingested along with food (b) semi-specific myiasis – when the larvae are laid on necrotic tissue of the wound c) obligatory – require living tissue for larvae development (d) facultative – require necrotic tissue for flies to lay eggs and incubate them. Based on anatomic site, it can be classified as: (a) cutaneous myiasis, (b) myiasis of external orifices an (c) myiasis of internal organs [2].

Primary myiasis, also called obligatory myiasis, is caused by biophagous *larvae* (which feed on living tissues). Secondary myiasis (facultative myiasis) is caused by necrobiophagus *larvae* (which feed on dead tissues). The most common anatomical sites for myiasis are the nose, eyes, skin wounds, sinuses, ears, lungs, gut, gall bladder, vagina, nasal cavities, and rarely, the mouth. However, specific type of flies can penetrate the healthy skin and produce myiasis [4]. Here authors report an uncommon case of primary oral myiasis.

Case Report

A 60-year-old mentally challenged female patient was presented to our hospital with swelling of the right middle one-third of the face and the upper lip since the previous day. A general physical examination revealed a compromised state, as the patient was accompanied by the attender. The history of patient revealed that she was unable to walk, dumb and deaf but was perfectly healthy 5 years ago and no details were available regarding her medical condition. She was not on any medication and has never been hospitalized before. The patient belonged to a low socio-economic status with poor built and was poorly nourished, uncooperative, and not well oriented to time, place and person. The patient's attender gave a history of tooth avulsion one day back and severe, continuous pain since then. Extraoral examination revealed facial asymmetry with swelling of the upper lip and periorbital edema on the right side

Fig. 1. Swelling of upper lip and periorbital edema

Ryc. 1. Obrzęk górnej wargi i oczodołów

(Fig. 1), TMJ was clinically normal with palpable and non-tender submandibular lymph nodes.

Intraoral examination revealed a necrotic area in the maxillary anterior region involving the vestibular sulcus and labial mucosa extending from the right canine to the left canine, measuring 6 cm × 4 cm with numerous maggots (Fig. 2). Border was irregular and the surrounding area

Fig. 2. Necrotic area in the maxillary anterior with numerous maggots

Ryc. 2. Martwica przedniej części szczęki z licznymi larwami

was erythematous with no bleeding or pus discharge evident from the necrotic area. The associated tooth, right upper central incisor exhibited significant mobility and the left upper central incisor was missing. Otherwise, there was poor oral hygiene with generalized periodontitis and root stumps with respect to 16, 36, 46. Clinical diagnosis was made as oral myiasis with classical presentation of maggots. The patient was subjected to radiographic examination with patient's attender's consent and occlusal radiograph revealed diffuse rarefaction of alveolar bone in the periapical region of maxillary central incisors suggesting superficial erosion of cortices. Well-defined lamina dura of 21 was appreciable suggestive of recent tooth exfoliation. The floor of the nasal cavity and maxillary sinus were unaltered. External root resorption was evident in 11, 12, 22 and well defined radiopacity in crown of 11 suggestive of ornamental filling (Fig. 3). Other hematological investigations were advised which were in normal limits.

The immediate treatment included the manual removal of all maggots with a tweezer. Around 50–60 maggots were removed (Fig. 4). The area was then washed with saline, followed by irrigation with betadine (Fig. 5). Broad spectrum antibiotics amoxycillin with clavaualnic acid and ibuprofen with paracetamol (i.v. route) were prescribed for 5 days along with Ivermectin (semisynthetic macrolide antibiotic isolated from *Streptomyces avermitlis*) 6 mg single dose was administered for 3 consecutive days. The same procedure was followed for 3 days and the patient responded well (Fig. 6). The upper right central incisor was extracted as it had grade III mobility (Fig. 7). The patient's attenders were educated about personal and environmental hygiene. Patient showed drastic improvement and was followed for a week's time and after which the patients other oral problems were attended.

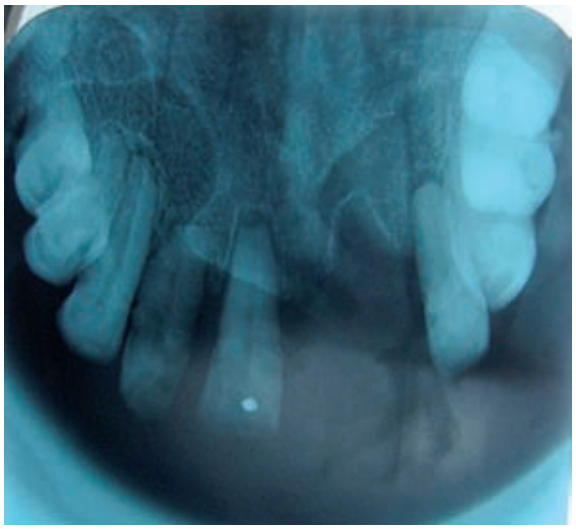

Fig. 3. Circle: ornamental filling: up broad arrow: intact lamina dura; down broad arrow: rarefaction of cortical bone; narrow down arrows: external root resorption

Ryc. 3. Zdjęcie radiologiczne przedniej części szczęki

Fig. 4. Removed maggots

Ryc. 4. Usunięte larwy

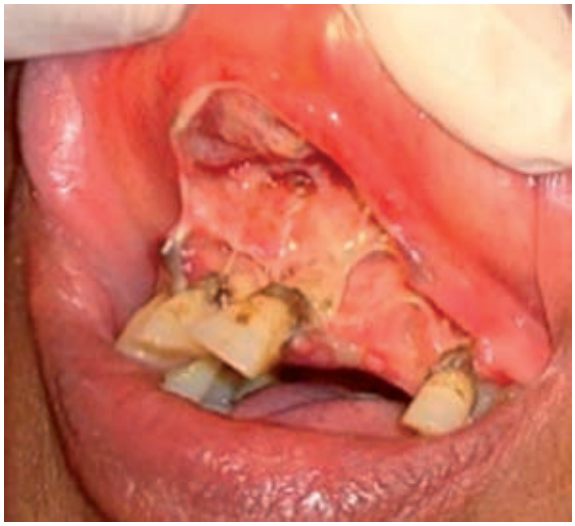

Fig. 5. After removal of maggots (first day)

Ryc. 5. Po usunięciu larw (pierwszy dzień)

Discussion

Myiasis is defined as an infestation of live human and vertebrate animals with dipterous *larvae* that feed on the host's dead or living tissue, liquid body substances or ingested food. They infest several parts of the body as in cutaneous, ophthalmic, oral, urogenital, nasopharyngeal and intestinal myiasis [5, 6].

Low socioeconomic status, immunocompromised state, debilitated and unhygienic living conditions are the main contributing factors respon-

Fig. 6. Marked reduction in swelling 2 days after treatment

Ryc. 6. Zmniejszenie obrzęku 2 dni po zabiegu

Fig. 7. Extraction of upper right central

Ryc. 7. Usunięcie górnego siekacza przyśrodkowego

sible for myiasis. The risk factors for oral myiasis include suppurative lesions, facial trauma, mouth-breathers, extraction wounds, fungating carcinomas, diabetes and peripheral vascular diseases, cerebral palsy and hemiplegia and others conditions. As in the present case, the patient was from a low socioeconomic status, with poor oral hygiene and mentally retarded [1].

During the lifecycle of the parasite, the developmental transition *via* the larval stage requires an intermediate host. The prevailing oral hygiene status provided the suitable substrate and temperature for the *larvae*. Eggs hatch within 24 h and the stage of *larvae* lasts for 2 weeks during which they are parasitic to human beings. The *larvae* release toxins to destroy the host tissue. Proteolytic enzymes released by the surrounding bacteria decompose the tissue on which the *larvae* feed. They are photophobic and tend to hide deep into the tissues for a suitable niche to develop into pupa. The present case also showed the *larvae* burrowed deep inside the wound in upper labial vestibule [2, 3].

The treatment consists of topical application of turpentine oil, mineral oil, chloroform, ethyl chloride or mercuric chloride followed by the manual removal of the larvae and surgical debridement. Recently, a systemic treatment with Ivermectin has been used for the treatment of oral myiasis. The cases of oral myiasis with no existing medical illnesses recover completely on removal of larvae [7, 8].

It is usually people with mental or physical disability, who are affected because of the difficulties in maintaining good oral hygiene due to poor manual dexterity and negligence by parents/guardians. Therefore, it becomes necessary for these patients to be exposed to a dental examination at regular intervals to prevent such diseases which if left unnoticed may have fatal complications.

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Conflict of interest: None declared

Received: 24.06.2014
Revised: 23.07.2014
Accepted: 8.08.2014

Praca wpłynęła do Redakcji: 24.06.2014 r.
Po recenzji: 23.07.2014 r.
Zaakceptowano do druku: 8.08.2014 r.