



Orofacial Myiasis Associated With Squamous Cell Carcinoma: A Case Report

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ABSTRACT

Myiasis is a parasitic infestation in vertebrate animals caused by dipterous fly larvae that feed on living or necrotic tissue of their host. It is considered a rare pathology that is related to local, systemic and environmental predisposing factors. Few cases of oral myiasis related to squamous cell carcinoma (SCC) have been reported in the literature. Thus, it can be said that the frequency of this binomial is relatively low or underreported. Objective and case report: The present work aims to report the case of a 68-year-old patient, with psychiatric disorders, in a situation of abandonment, affected by orofacial myiasis associated with SCC. Furthermore, we discuss the diagnosis and treatment considered most appropriate. Discussion:

The involvement of myiasis, in advanced stages, highlights a tissue destruction that can lead to severe functional sequelae, as in the reported case, which showed destruction of the lower and middle third of the right hemiface, leading to exposure of the oral cavity and compromising chewing and phonation, in addition to aesthetics. The association of myiasis with SCC increases the severity of both conditions by accelerating the process of destruction by larvae and increasing the severity of the carcinoma, a fact that was observed in the case reported. Treatment, even because of the rarity of the involvement, becomes challenging in these cases, since there are no well-established protocols in the literature, leading to the need for an accurate and individual assessment of the case to establish an appropriate therapy. Conclusion: It is of utmost importance for health professionals to investigate any possible neoplastic lesion associated with myiasis. Due to the importance of this binomial, it should become part of the treatment protocol to perform biopsy of the lesion and, consequently, anatomopathological analysis.

Keywords: Myiasis, Squamous Cell Carcinoma, Face, Mouth Neoplasms.

1 INTRODUCTION

Myiasis is a parasitic infestation in vertebrate animals caused by dipterous fly larvae that feed on the living or necrotic tissues of their host^{1,2,3}. This affection in humans, in general, is frequently found in tropical and subtropical climatic regions of the planet^{1,2,4,5,6}; therefore, India and Brazil are the most affected countries^{2,4}.

The disease is capable of infesting different regions of the human body, such as the skin, paranasal sinuses, ophthalmic, nasopharyngeal, otologic, intestinal, oral, urogenital and cerebral cavities, among others¹⁻⁷. Oral myiasis, first described by Laurence in 1909, is considered a rare pathology^{2,6}, because its incidence is comparatively lower than its cutaneous form, since oral tissues are not permanently exposed to the external environment⁸. The risk factors that can be observed in this condition are poor oral hygiene, absence of lip seal, mouth breathing, oral and maxillofacial trauma, suppurative lesions, malignant neoplasms, senility, drug use, alcoholism, low socioeconomic level, neurological deficiencies, street

situation, immunosuppression and delay in seeking medical care¹⁻⁷. Of these, the malignant neoplasm seems to be the most challenging factor, and there is no consensus as to the initial treatment of the myiasis-malignant neoplasm binomial.⁹

Squamous Cell Carcinoma (SCC), also known as epidermoid carcinoma or squamous cell carcinoma of the mouth, is a malignant neoplasm that originates from the lining epithelium, considered the most common head and neck neoplasm¹⁰. However, few cases of oral myiasis associated with SCC have been reported in the literature. Thus, it can be said that the frequency of this binomial is relatively low or underreported.¹

Given the above, this paper aims to report the case of a female patient, 68 years old, in a situation of abandonment, affected by orofacial myiasis associated with squamous cell carcinoma. Furthermore, we sought to discuss the diagnosis and treatment considered most appropriate for the case. This study was followed according to the SCARE Criteria.¹¹

2 CASE REPORT

Female patient, 68 years old, Caucasian, university graduate, homeless, extremely vulnerable, missing from her family since 2016, referred from another health unit to the emergency service of Oral and Maxillofacial Surgery and Traumatology (MBCT) of the State Hospital of Aparecida de Goiânia Caio Louzada in June 2021, due to the presence of myiasis and foul odor in the right hemiface region. According to information gathered from the accompanying health professional, his health history included a diagnosis of psychiatric disorders. A previous endodontic treatment that was not completed was also mentioned.

On physical examination, she was in a regular general state, disoriented in time and space, pale, dehydrated, hemodynamically stable, spontaneous breathing without effort with episodes of coughing and dysentery. The orofacial examination revealed a vegetative and ulcerated swelling located on the right side of the face, extending from the middle third of the face to the submandibular trigone region, with approximately 10 cm in its largest diameter, transfixated to the oral cavity, with raised edges, irregular, hardened to palpation, infiltrated, necrotizing aspect, infected by larvae, with limited mouth opening, foul odor and algesia to manipulation (Figures 1 and 2).



Figure 1: Initial clinical aspect of the lesion, in lateral view, showing tissue necrosis and presence of larvae.



Figure 2: Clinical aspect of the lesion, in 3/4 view, showing raised, irregular and hardened borders on palpation.

Initial laboratory tests showed high levels of C-reactive protein (98.6 mg/L), leukocytosis (15,330 mm³); erythropenia with critical RBC results (1.77 mm³); anemia (3.4 mm³); and low hematocrit levels (12.3 mm³); which made hemotransfusion with 3 bags necessary. The test for COVID-19 was positive, requiring isolation for 14 days, and no significant complications were observed. The tomographic study without iodine contrast injection showed discontinuity solutions in soft tissue regions in the right hemiface, without bone alterations.

Treatment was initially performed with surgical procedure under local anesthesia with lidocaine and vasoconstrictor, mechanical removal of the larvae, debridement of devitalized tissues, and irrigation with ether solution asphyxiating substance (Figure 3 and 4).



Figure 3: Extensive facial lesion with presence of multiple larvae and lesion transfixing to the oral cavity. Procedure performed under ether solution irrigation.



Figure 4: Immediate clinical appearance after complete removal of the larvae and tissue debridement.

The deeper larvae were removed with the patient resting in bed in the subsequent days, associated with the use of ether and daily local dressing with activated charcoal. The drug therapy used involved the administration of ivermectin 6mg of 12/12 hours for 2 days, associated with systemic intravenous antibiotics, namely ceftriaxone 1g of 12/12 hours for 7 days and clindamycin 600mg of 06/06 hours for 7 days (Figure 5, 6 and 7).



Figure 5: First postoperative week, revealing the beginning of granulation tissue at the margins of the defect and drying of the central part of the wound.



Figure 6: Second postoperative week, showing advanced healing process.



Figure 7: Clinical appearance after 1 month of follow-up.

Due to the aspect of the tissue lesion and the possibility of association of myiasis with malignant lesions, the CTBMF team performed an incisional biopsy of the region, which is considered an important step in the treatment protocol. The specimen was sent for pathological analysis, which revealed tissue compatible with SCC (figure 8).

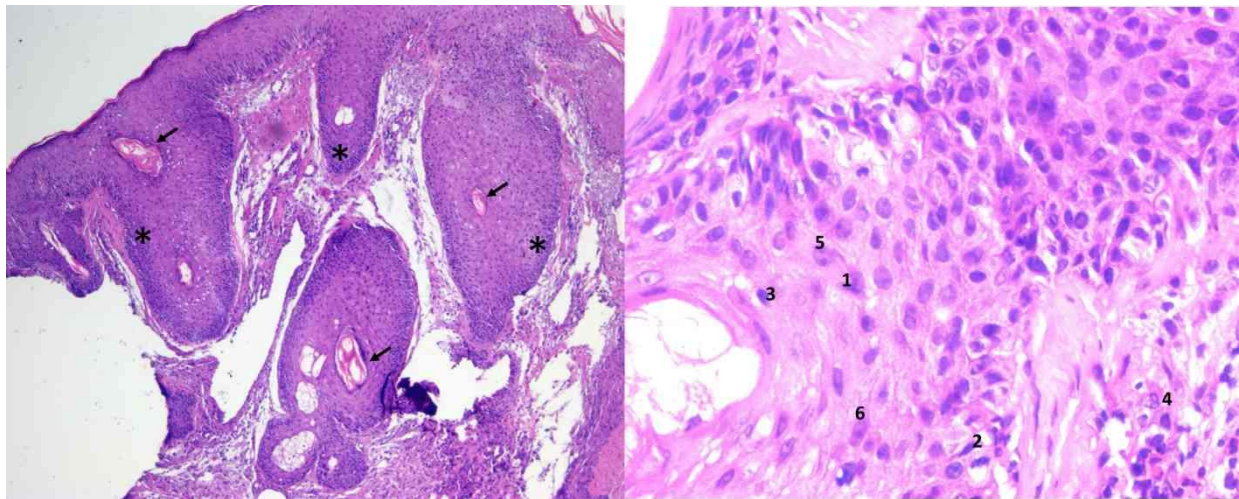


Figura 8– Fotomicroscopia em menor (A) e maior (B) aumento coradas em hematoxilina e eosina, aumento original $\times 5$ (A) $\times 40$ (B). Os cortes microscópicos revelam a presença de tecido epitelial neoplásico bem diferenciado que invade o tecido conjuntivo (Asterisco). Nota-se a presença de formação de pérolas córneas em A (seta preta). Em B nota-se a presença de figuras de mitoses atípicas (1), pleomorfismo nuclear (2), hiper cromatismo (3), binucleação (4), nucléolos múltiplos e proeminentes (5) e alteração da relação núcleo citoplasma (6). Achados compatíveis com carcinoma de células escamosas bem diferenciado (Grau I).

After the resolution of the myiasis infection and the diagnosis of malignant neoplasm, the patient was discharged from the CTBMF and referred to Araújo Jorge Cancer Hospital for treatment of the pathology and facial reconstruction with the Head and Neck Surgery team. However, due to the terminal stage of the disease, the patient died within approximately 3 months, and palliative treatment was instated during this period.

3 DISCUSSION

The term myiasis is derived from the Greek "myia", meaning fly, and is used to designate the invasion of living human or other mammalian tissues by dipterous larvae. These feed on living or necrotic tissue of the host and body substances such as liquid or ingested food^{3,12}. Thus, they can be classified as primary (obligate) or secondary (facultative). Primary myiasis originates from biophagous larvae that feed on living tissue and are extremely uncommon in humans. However, secondary myiasis, which is the most common form in humans, arises from necrophagous flies that feed on necrotic tissues^{1,2,3}, as described in this case.

The life cycle of the fly begins with an egg, followed by the larval stage, pupa, and finally evolving into the adult stage. The insects are usually attracted to pre-existing wounds or neoplasms. A female fly can lay about 100 to 500 eggs in these lesions, which hatch after about 15 hours and release larvae that feed on the adjacent tissues. Foul-smelling lesions are formed rapidly, attracting other flies and, consequently, more eggs. These, on average, emerge from the host after 4-7 days as pre-pupae, causing extensive tissue destruction and forming spaces called "pockets", which [then](#) migrate from the wound into the environment and pupate after a week (intermediate stage between larva and fly). As adults, they metamorphose into flies and can infect new hosts, closing the cycle that usually lasts 24 days^{4,6,7}. Therefore, it is imperative to

start treatment of the disease early, because the longer the waiting time, the greater the tissue destruction and, consequently, the functional damage.

Much has been discussed about the importance of predisposing factors for myiasis infection, which may be local, systemic and environmental. Local factors include poor oral hygiene, suppurative lesions, trauma, absence of lip seal, mouth breathing, anterior open bite, digital sucking habit, halitosis and periodontal disease. Systemic factors are: cerebral palsy, epilepsy, poor general hygiene, senility, drug and/or alcohol use, neurological deficiencies, mental disorders, diabetes, and immunosuppression. Finally, environmental factors include low socioeconomic level, street situation and hot and humid climate^{1,2,3,4,5,6,7}, conditions that are compatible with the reported case.

Some cases are secondary to medical or anatomical conditions, such as neglected fractures, malignant neoplasms and delay in seeking medical care^{1,2,3,4,5,6,7}. Therefore, corroborating the literature, the report presented evidenced clinical conditions of poor oral hygiene, suppurative lesion, senility, mental disorder, street situation, low socioeconomic level, geographic location such as subtropical Brazil, medical condition secondary to malignant neoplasm, and delay in seeking medical care, which were the determining factors for the development of orofacial myiasis.

Individuals with head and neck cancer have an increased risk of developing myiasis⁷. According to the authors Girardi and Scrofernecker - 2017, individuals with this condition usually present neglected and advanced tumors, with areas of necrotic tissue exposure, becoming an attractive region for egg deposition⁷. This situation is compatible with the reported case and corroborates other authors, since there are well-reported predisposing factors, such as the local, systemic and environmental ones cited above.

The association between oral myiasis and malignant neoplasms was analyzed in a literature review by Passos and Arruda in 2020. The study states that the most affected individuals are from rural areas, of low socioeconomic level, predominantly male (81.8%), with a mean age of 57.1 years, who suffered mainly from squamous cell carcinoma⁴, conditions that were partially corroborated in this study.

SCC constitutes about 90% of oral cancers, causing significant morbidity and mortality. This entity results in various morbidities such as pain, bleeding on provocation, loss of function, facial disfigurement, extraoral fungi and tissue necrosis². This is evidenced by this case, in which the larvae in question fed on necrotic tissues, stemming from the malignant condition of the SCC.

The association of myiasis with SCC increases the severity of both conditions by accelerating the process of destruction by the larvae and increasing the severity of the carcinoma, a fact that was observed in the reported case⁷. The treatment, even because of the rarity of the involvement, becomes challenging in these cases, since there are no well-established protocols in the literature, leading to the need for an accurate and individual assessment of the case to establish an appropriate therapy.

The collection of information through a good anamnesis is extremely important for a correct therapeutic indication. This is something already established in health services and is part of the basic actions in patient care¹³. However, in the case presented here, since the patient was unassisted by friends

or family, the anamnesis could not be initially performed with the necessary quality, since the patient had psychiatric disorders and no one was with her. For this reason, and because there are studies correlating the myiasis-CCE binomial, also to elucidate possible local causal factors in addition to the systemic and environmental ones, the incisional biopsy was chosen.

There is no specific treatment model for the management of myiasis. A combination of local and systemic treatment is recommended for cases of oral myiasis associated with OHC². The local treatment modality is preferably mechanical removal associated with the application of topical smothering agent, with the aim of forcing the larvae to move out of the host tissue, followed by debridement of the wound in question. Systemically, ivermectin has been used in the treatment of oral myiasis, since this drug activates the release of γ -aminobutyric acid, which induces the death of the larvae and their spontaneous elimination². The use of systemic antibiotic can also be administered if there are signs of secondary bacterial infection². In the present case, surgical treatment of mechanical removal of larvae, topical use of a smothering substance, daily dressing of activated charcoal associated with the use of ivermectin and systemic antibiotic therapy were shown to be effective in curing the myiasis, although the patient's final outcome was death, a result of the terminal stage of CEC.

4 FINAL CONSIDERATIONS

Considering the work performed here and the literature referred to, it could be observed that individuals affected by head and neck cancer belong to a risk group for developing myiasis on their wounds, a condition that may accelerate the progression of the disease, sometimes lethal. Undoubtedly, preventive approaches in patients, when already diagnosed with malignant neoplasia, include basic health care, access to primary health care, and control of the fly population. These are primary care to prevent the disease. Once affected, a proper treatment protocol should be followed, as shown in our work.

Therefore, it is known that the patient will not always have a previous diagnosis of malignant neoplasia. Thus, it is of utmost importance that the health professional pay attention to the investigation of any possibility of neoplastic lesion associated with myiasis. Due to the importance of this binomial, it should become part of the treatment protocol to perform a biopsy on the lesion and, consequently, an anatomopathological analysis.

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