A buccal mucosa ulcer as the first sign of tuberculosis

Paulo S. S. Pina, Celso A. Lemos, Suzana C. O. M. de Sousa

Department of Stomatology, Faculty of Dentistry, Universidade de São Paulo (USP), Sao Paulo, SP, Brazil

INTRODUCTION

Tuberculosis (TB) is a bacterial infectious disease characterized by granulomatous lesions caused by Mycobacterium tuberculosis. TB bacillus was discovered by Robert Koch in 1882, and it has been a global health problem for centuries. Although its prevalence reduced decades ago, most of the estimated number of cases in 2018 occurred in the South-East Asia Region (44%); smaller proportions of cases occurred in the region of the Americas (2.9%).

Although lungs are affected primarily, extrapulmonary lesions can also occur as they may spread to other sites through self-inoculation via infected sputum, blood or lymphatic system. Oral TB lesions are rare and, probably because of that, they have largely become a forgotten diagnosis in the mouth. In general, oral manifestations of TB are rare and may be found in patients with isolated oral mucosa lesions, we must consider the possibility of oral manifestation of systemic diseases, even without typical clinical signs and symptoms.

Keywords: Buccal mucosa, granulomatous chronic disease, tuberculosis

Address for correspondence: Dr. Paulo S. S. Pina, Department of Stomatology, Faculty of Dentistry, Universidade de São Paulo, Av. Prof. Lineu Prestes, 2227 – Butantã, São Paulo, SP, Brazil.
E-mail: ps.souzapina@usp.br
Submitted: 18-Dec-2021, Revised: 11-Feb-2022, Accepted: 26-Feb-2022, Published: 17-Oct-2022

How to cite this article: Pina PS, Lemos CA, de Sousa SC. A buccal mucosa ulcer as the first sign of tuberculosis. J Oral Maxillofac Pathol 2022;26:399-403.

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Access this article online

Quick Response Code:
Website: www.jomfp.in
DOI: 10.4103/jomfp.jomfp_443_21

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CASE REPORT

A 61-year-old black male patient presented to the Stomatology Clinic of a dental school reporting an ulcerated lesion on the buccal mucosa. The patient stated to be a smoker, and he did not had any underlying diseases. Extra-oral examination showed facial asymmetry caused by swelling of the right side of the face. In the intra-oral examination, an ulcerated, red lesion with yellowish areas, measuring approximately 4 cm was noted extending from the labial commissure to the right buccal mucosa [Figure 1]. According to the patient, the injury was symptomatic and had been present for 4 months.

Clinical findings were suggestive of infectious or neoplastic disease. Under local anesthesia, an incisional biopsy was performed. Histopathological findings showed fragments of mucosa lined by stratified, squamous, epithelia. In the lamina propria, numerous granulomatous formations, with several Langhans-like multinucleated giant cells and numerous blood vessels were noted [Figure 2]. Also, it was observed that the granulomatous arrangement extended to involve the underlying muscle [Figure 3]. PAS staining revealed Candida pseudo-hyphae on the surface of the epithelium, and the Ziehl–Neelsen histochemical reaction was negative. The final anatomopathological diagnosis was as a chronic granulomatous inflammatory process, and despite the negativity to Ziehl–Neelsen, further detailed investigations for tuberculosis were suggested.

Chest radiography depicted hollow macronodular lesions suggestive of an inflammatory-infectious/granulomatous process of bronchogenic dissemination [Figure 4]. Sputum alcohol-acid–resistant bacilli (BAAR) testing was positive. After 3 months of initiating drug therapy for TB, there was a total regression of the oral lesions, in addition to a recovery of 10% of body weight after finishing the treatment regimen.

DISCUSSION

The World Health Organization (WHO) states that around 10 million people fall ill with TB each year, worldwide. Although the disease can affect anyone, most people who develop it (about 90%) are adults with a male: female ratio of 2:1. Globally, almost 2 billion people are infected and are, therefore, at risk of developing the disease. In 2018, Brazil showed a markedly lower incidence rate per capita of TB, however, it is still among the 30 high tuberculosis burden countries, accounting for 87% of all estimated incident cases on earth. [3]

Mycobacterium tuberculosis infection of the human host often affects the lungs, and it is known as pulmonary TB. However, it can also manifest in other sites of the body, and then it is referred to as extrapulmonary.[7] The extrapulmonary affection can vary between 5% and 50%,[6,8] and regarding the head and neck, the prevalence ranges from 10% to 35%.[8] Tuberculosis oral lesions have an unusual occurrence, with the incidence being less than 0.5%–1% amongst all the TB patients.[9] The most common affected area in the mouth is the tongue, followed by the gums, buccal mucosa and lips.[6]

Besides being uncommon, orofacial TB presents clinically in a variety of aspects. Thus, it can be misdiagnosed, especially whether the diagnosis of TB is unknown.[10] In the case reported, the microorganisms could not be
visualized using Ziehl–Neelsen histochemical reaction although the morphology strongly suggested a TB infection. Moreover, interestingly, despite the radiographic examination of the patient's chest being highly suggestive of TB, a sputum smear microscopy was not positive at the first examination, only at the second. As presented by Kakisi et al.[11] (2010) in a systematic review, negative sputum reactions can occur as they showed in 12/73 of their samples. They have also shown that there is a scarcity of mycobacteria, especially in oral lesions, probably due to the cleansing effect of saliva, the scarcity of lymphoid

| Author/year                | Country | Gender | Age | Site of Involvement | Baseline disease | Form of TB                |
|----------------------------|---------|--------|-----|---------------------|------------------|---------------------------|
| Present case               | Brazil  | Male   | 61  | Buccal mucosa       | None             | Secondary                 |
| Razem et al. (2021)        | Marocco | Female | 36  | Tongue              | None             | Primary                   |
| Kechaou et al. (2021)      | Tunisia | Female | 30  | Palate              | Arthritis and Systemic Lupus | Primary                   |
| Sachdeva et al. (2020)     | India   | Female | 12  | Lip                 | None             | Primary                   |
| Hamid et al. (2020)        | India   | Female | 37  | Gingiva             | None             | Primary                   |
| Hamid et al. (2020)        | India   | Female | 45  | Palate              | None             | Primary                   |
| Hamid et al. (2020)        | India   | Male   | 49  | Tongue              | None             | Primary                   |
| Hamid et al. (2020)        | India   | Male   | 23  | Gingiva             | None             | Primary                   |
| Fragozo et al. (2020)      | Portugal| Male   | 44  | Tongue              | Not informed     | Not informed              |
| Kim et al. (2019)          | South Korea | Male | 57  | Tongue              | None             | Secondary                 |
| Esteves et al. (2019)      | Brazil  | Female | 61  | The floor of the mouth and gingiva | Rheumatoid arthritis and osteoporosis | Primary                   |
| Esteves et al. (2019)      | Brazil  | Female | 67  | Lower lip           | Pulmonary emphysema | Secondary                 |
| Nico et al. (2018)         | Brazil  | Male   | 22  | Lip                 | Not informed     | Not informed              |
| Nico et al. (2018)         | Brazil  | Male   | 39  | Lip                 | Not informed     | Not informed              |
| Nico et al. (2018)         | Brazil  | Male   | 42  | Tongue              | Psoriatic arthrits | Not informed              |
| Vineetha et al. (2018)     | India   | Female | 65  | Gingiva             | Not informed     | Secondary                 |
| Trawinski et al. (2018)    | Germany | Female | 29  | Buccal mucosa       | Crohn’s disease  | Secondary                 |
| Zhang et al. (2017)        | China   | Male   | 52  | Gingiva and palate  | Not informed     | Not informed              |
| Bhuiya et al. (2017)       | India   | Male   | 29  | Upper lip and palate | Immunocompetent   | Secondary                 |
| Parajuli R; Maharjan S (2017) | Nepal | Male   | 78  | Tongue              | Hypertension     | Not informed              |
| Bos et al. (2017)          | Croatia | Male   | 68  | Tongue              | None             | Secondary                 |
| De Souza et al. (2016)     | Brazil  | Male   | 61  | Upper lip and buccal mucosa | Diabetes         | Secondary                 |
| Gupta et al. (2014)        | India   | Male   | 24  | Lip                 | None             | Primary                   |
| Nagaraj et al. (2013)      | India   | Male   | 43  | Tongue              | Not informed     | Secondary                 |
| Gill et al. (2010)         | India   | Female | 32  | Gingiva             | Not informed     | Primary                   |
| Kumar et al. (2010)        | India   | Male   | 38  | Tongue              | None             | Primary                   |
| Ebenezer et al. (2006)     | India   | Male   | 40  | Buccal mucosa       | None             | Primary                   |
| Ebenezer et al. (2006)     | India   | Female | 7   | Gingiva             | Not informed     | Not informed              |
| Sezer et al. (2004)        | Turkey  | Male   | 46  | Buccal and alveolar mucosa extending to the oropharynx | None | Secondary                 |

Table 1 shows demographic and clinical data of previous reported cases of oral tuberculosis manifestation.
tissue in the tongue, and the antagonist oral commensals microorganisms. Also, material collected from lesions in sites such as the soft palate and lips are more likely to be negative for mycobacterium. According to McKee et al. (2012), occasionally, the diagnosis of tuberculosis is sometimes a hard task, and the identification of the bacillus and consequent definition of the diagnosis must be confirmed by other tests, such as sputum culture or yet by a therapeutic trial of anti-tuberculosis drugs\textsuperscript{[12]}

It is well elucidated that immunosuppressed patients with certain comorbidities (such as diabetes, alcoholism, neoplasms, human immunodeficiency virus (HIV) and those on long-term steroid therapy\textsuperscript{[13]} Tobacco habits, oral trauma and poor oral hygiene are also considered risk factors\textsuperscript{[13]} In the literature, the majority of cases of TB oral manifestation are mostly seen in elderly patients, whereas primary infection appears to be limited to younger individuals [Table 1].\textsuperscript{[6,14-32]} The patient of our study only declared to be a smoker, not reporting any systemic comorbidities or use of corticosteroids. As our patient did not state any baseline disease, fever or use of medications, added to the fact that he was an elderly man and a smoker, the clinician suspected that the lesion could be a carcinoma. Kakisi et al.\textsuperscript{[11]} (2010) noted that most of the patients (94%) were unaware of their TB infection. Therefore, they recommend an immediate investigation for TB in those cases where ulcers are typically showing necrosis, irregular borders and unresponsiveness to antibiotic treatment or to corticosteroids. According to Bansal et al.\textsuperscript{[10]} (2015), TB may present clinically as a single or multiple, superficial or deep, painful or painless ulcers with an irregular border, which tends to increase slowly in size.

CONCLUSION

Thus, in patients with isolated oral mucosa lesion, dentists must be aware of the possibility of an oral manifestation of a systemic disease.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

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