Case Report

Palate Tuberculosis with Paradoxical Lymphadenitis

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Abstract

Oral cavity involvement in tuberculosis (TB), particularly palatine, is extremely rare and mostly described in case reports. Management of these cases usually responds to classic antitubercular therapy. Some serious complications such as paradoxical reactions (PRs) may however occur, making it more challenging for physicians to treat and to manage. We present a case of a 30-year-old female patient with a history of juvenile idiopathic arthritis and systemic lupus erythematosus who presented a bifocal form of TB involving the palate and the cervical lymph nodes. Follow-up after 2 months of proper antitubercular treatment revealed a PR of the lymph nodes contrasting with a favorable outcome of the oral lesions. It seems useful to raise all clinicians' awareness to suspect TB when they deal with chronic drug-resistant oral erosions and to keep in mind the diagnosis of PR when there is a worsening of one lesion and a favorable outcome of another.

Keywords: Oral ulcers, palatine tuberculosis, paradoxical reaction

Introduction

Oral cavity involvement in tuberculosis (TB) is rare, occurring in 0.2%–1.5% of extrapulmonary cases.[1] The most common sites are the tongue followed by the gingiva.[2] Palate involvement is exceptional. Diagnosis essentially relies on histological findings. Although it is believed to be a curable benign disease, management of TB can be challenging, particularly when complications such as paradoxical reactions (PR) occur.[3] We hereby present a case with a palatine and cervical lymph node TB complicated with a PR of the lymph nodes.

Case Report

A 30-year-old female Caucasian housewife from a rural area with a history of juvenile idiopathic arthritis has been followed in our department since 2008 for systemic lupus erythematosus (SLE) in its cutaneous (acute and discoid lesions) and immunologic form. She has been on hydroxychloroquine since the diagnosis of SLE. No family or personal history of TB was recorded. In December 2017, the patient was admitted for gingivostomatitis occurring after a tooth extraction that was resistant to usual treatment and antibiotics. On admission, the general state was not altered and she had no fever. She weighed 45 kg (body mass index 14.36 kg/m²). Mucocutaneous examination found poor oral hygiene. She had a large palatine inflammation with multiple superficial infracentimetric nonnecrotic ulcerations [Figure 1a]. Examination also found three firm inflammatory right cervical lymphadenopathies in the submandibular and deep anterior region measuring 1.5–2 cm. In laboratory tests, white blood cell count was 8400/mm^3 (neutrophil polynuclear level count: 5600/mm^3 and lymphocyte count: 2000/mm^3). Hemoglobin level was 11.2 g/dL, mean corpuscular volume was 89.2 μ^3, and platelet count was at 295,000/mm^3. Ferritin level was at 77 ng/mL, C-reactive protein at 20 mg/L, and albumin at 35 g/L. Creatinine level and hepatic enzymes were within the normal range. Tuberculin intradermal skin test was positive with a 15 mm induration. Serology for HIV was negative. The patient had no clinical or biological sign of SLE flare.

Cervical ultrasonography found bilateral hypoechogenic cervical lymphadenomegalies (right IIB and IIA chains: 18 mm × 26 mm) and (the left IB chains: 20 mm × 7 mm). Facial computed tomography (CT)-scan found an irregular osteolytic lesion of the maxillary bone next to the 14th tooth

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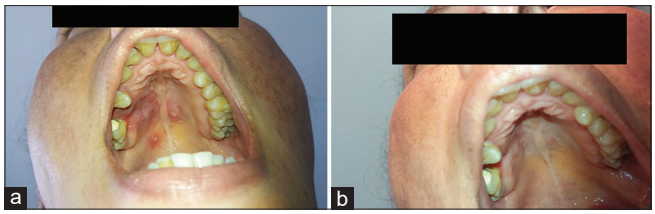

Figure 1: (a) Before treatment: Large palatine inflammation with multiple superficial infra-centimetric nonnecrotic ulcerations. (b) After treatment: good evolution

space that was compatible with an infectious etiology. Chest radiography and abdominal ultrasonography were normal. A biopsy of one ulcerated palatine lesion showed an inflammatory granulomatous tissue containing gigantic and epithelioid cells with caseous necrosis. This aspect was in favor of caseofollicular necrosis. Thus, the diagnosis of palatine and lymph node TB was made considering the high specificity of caseum in TB and the epidemiologic context. National antitubercular scheme therapy was prescribed with isoniazid 100 mg/day, rifampicin: 600 mg/day, ethambutol: 1200 mg/day, and pyrazinamide 1000 mg/day for 2 months followed by bitherapy (Isoniazid and rifampicin) with good adherence and without side effects.

Within 10 weeks of treatment, the palatine lesions completely disappeared [Figure 1b], whereas the size of the cervical lymphadenopathies was significantly increasing [Figure 2]. Cervical, thoracoabdominal and pelvic CT-scan showed multiple confluent cervical adenomegalies (right IIB chains whose biggest measured 22 mm × 24 mm). There was also one adenomegaly with a necrotic center (right III chains: 20 mm × 25 mm) that was repressing the right internal jugular vein without causing thrombosis. Calcified lymph nodes of the right hilum were also observed. Biopsy of the submandibular lymph node found caseous necrosis and no signs of associated lymphoma. Bacterial samples using polymerase chain reaction assays isolated a complex *Mycobacterium tuberculosis* sensitive to both isoniazid and rifampicin. The patient was completely adherent to treatment and no adverse events were reported. Paradoxical reaction (PR) was therefore diagnosed. We decided to take up a triple agent antitubercular therapy by re-adding ethambutol. The lymph node biopsy was complicated with fistulization surgically treated with favorable outcomes. The patient received a total of 30 months of treatment with restoration of a normal general state, disappearance of palatine lesions, and stabilization of lymph nodes.

Discussion

Our case highlights a bifocal localization of TB involving palate and cervical lymph nodes. Multiple risk factors of oral TB are described in the literature such as poor dental hygiene, smoking, or trauma.[1] Our patient had a tooth extraction before the palatine inflammation and a poor dental hygiene. Furthermore, she is considered at risk of immunosuppression even if her SLE was quiescent.

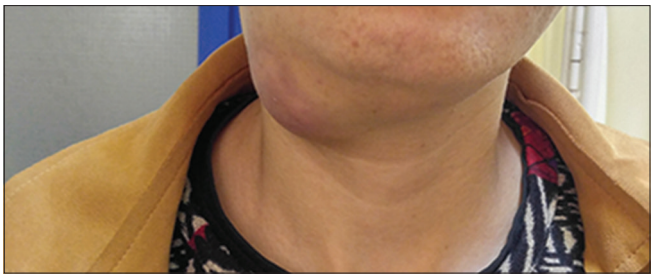

Figure 2: Paradoxical reaction with increasing of the size of submandibular lymph nodes

Positive diagnosis of PR was retained after excluding a drug resistance, a poor adherence to treatment, and an associated lymphoma.[3,4] The eccentricity of PR in our case comes from the contrast between the good evolution of oral cavity lesions and the worsening of lymphadenitis.

When referring to current literature, none of the oral cavity TB cases was complicated with a PR.[1-3,5-8] Yet, in some cases, when TB involves more than one site including the lymph nodes, it is likely that PR causes a worsening of lymphadenitis with a better outcome of the other localizations as described in our patient.[5,7] It may be related to the frequency of lymph node involvement in TB compared to other sites.

Multiple risk factors of RP have been described: High bacterial load, lower age, male gender, local tenderness, anemia, hypoalbuminemia, low lymphocyte count at baseline, size of lymph node ≥3 cm, and associated extra-lymph node TB.[6-8] The delay of occurrence of PR in our observation was two months, which was concordant to the delays described in the literature.[3,9]

For our patient, the lymph node biopsy was not only performed to exclude an associated lymphoma but also to avoid the risk of compression considering the CT-scan findings.

Treatment of PR is still debatable. Anti-tubercular treatment should not be suspended.[3,5,7,10] In some cases, a quadritherapy had to be taken up again.[7,10] In our patient, we decided to take up a third antitubercular agent, ethambutol. In most cases, antitubercular treatment should be extended to even 30 months, depending on the PR localization and the evolution.[3,7] Corticosteroids can be used.[1] Drainage aspiration can be an interesting alternative to surgery but beware of the risk of fistulization and recurrence.[9]

Declaration of patient consent

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

Research quality and ethics statement

The authors followed applicable EQUATOR Network (http://www.equator-network.org/) guidelines, notably the CARE guideline, during the conduct of this report.

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Conflicts of interest
There are no conflicts of interest.

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