



## Case illustrated

## Primary tuberculosis cutis orificialis; a different face of the same coin

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## ABSTRACT

Primary tuberculosis cutis orificialis (TCO) is a rare form of extrapulmonary TB. The lack of respiratory symptoms and similarity of the presentations to other oral lesions can pose a diagnostic dilemma. Hence, delaying treatment and potentially devastating consequences

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A 51-year-old female was referred to our service because of a three-month history of a painful and non-healing oral ulcer on the right buccal mucosa associated with a weight loss of 4 kg. Her history is significant for well-controlled seronegative rheumatoid arthritis (RA) with methotrexate and golimumab for the two years. She denied fever, painful joints, or cough. Examination of her mouth was limited by pain when she fully opened her mouth, but a single right buccal ulcer opposite to the retromolar trigone was seen (Fig. 1A). It was tender with ragged margins (around 2.1 cm). No other findings or lymphadenopathy were observed. The differential diagnoses included squamous cell carcinoma, infectious etiologies, methotrexate related, or RA related. Laboratory tests were within normal limits except for a CRP level of 40 mg/L (0–5). Anti-CCP, HIV, and syphilis serology were negative. Punch biopsy showed necrotizing granulomatous inflammation with scattered acid-fast bacilli (Fig. 2A and B). The fungal culture was negative. However, MTB (GeneXpert MTB/RIF) was positive. Subsequently, a fully sensitive MTB strain was isolated from the culture. CXR demonstrated no pulmonary involvement, and MRI of the jaw excluded osteomyelitis. She was started on standard 6 months TB therapy with pain resolution and complete healing of the ulcer (Fig. 1B). Golimumab was stopped. She had no recurrence for six months during follow-up.

Primary TCO occurs when the oral mucosa is affected without pulmonary involvement [1]. It represents 0.01–5% of all TB cases [1]. Children and adolescents are affected more than adults, while

tongue and palate are the most common sites for infection [2]. Various morphological lesions exist including ulcers, nodules, plaques, and fissures [3]. It can cause osteomyelitis in the adjacent bone [1]. Typically, primary TCO is a single and painless lesion. The differential diagnoses are squamous cell carcinoma, chronic traumatic ulcer, medications, syphilis, and other granulomatosis diseases [3]. Notably, golimumab at least doubles the risk for TB activation [4].

The first step to clinch the diagnosis is to consider TCO while evaluating any chronic oral lesions [1]. Fundamentally, TB MTB PCR can secure the diagnosis promptly pending the histopathological diagnosis to rule out other potential causes [1]. The mainstay of management is 6 months of TB therapy [3].

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## Author contributions

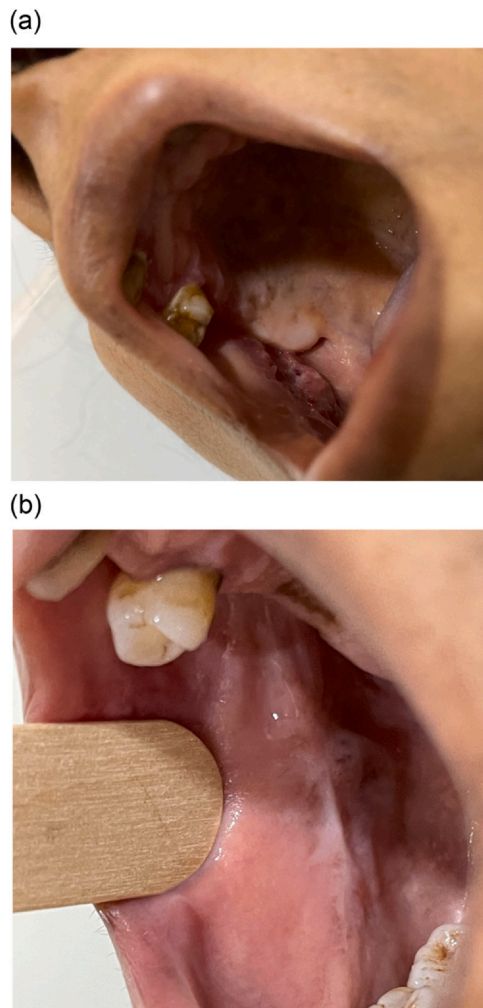
**GA:** Clinical management, data acquisition and manuscript writing. **WG:** Clinical management, contribute to data acquisition, manuscript preparation and final proof reading.

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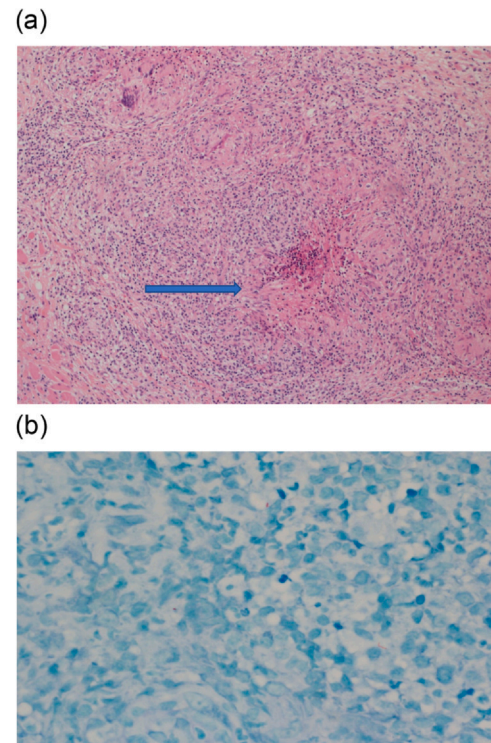
**Fig. 1.** A: Single right buccal ulcer with ragged margins. B: Right buccal ulcer demonstrating significant improvement after six months of the TB therapy.

#### Conflict of Interest

the authors declare that they have no competing interests.

#### Compliance with Ethical Standards

Ethics approval and permission was obtained to publish the case reports from the institutional review board which is in line with international standards,



**Fig. 2.** A: Punch biopsy showed necrotizing granulomatous inflammation consistent with TB (arrowed). B: Punch biopsy with Ziehl-Neelsen stain showed scattered acid-fast bacilli.

#### References

- [1] Kakisi OK, Kechagia AS, Kakisis IK, Rafailidis PI, Falagas ME. Tuberculosis of the oral cavity: a systematic review. *Eur J Oral Sci* 2010;118:103–9. <https://doi.org/10.1111/j.1600-0722.2010.00725.x>
- [2] Mignogna MD, Muzio LL, Favia G, Ruoppo E, Sammartino G, Zarrelli C, et al. Oral tuberculosis: a clinical evaluation of 42 cases. *Oral Dis* 2000;6:25–30. <https://doi.org/10.1111/j.1601-0825.2000.tb00317.x>
- [3] Krawiecka E, Szponar E. Review paper Tuberculosis of the oral cavity: an uncommon but still a live issue. *Adv Dermatol Allergol* 2015;4:302–6. <https://doi.org/10.5114/pdia.2014.43284>
- [4] Baddley JW, Cantini F, Goletti D, Gómez-Reino JJ, Mylonakis E, San-Juan R, et al. ESCMID study group for infections in compromised hosts (ESGICH) consensus document on the safety of targeted and biological therapies: an infectious diseases perspective (soluble immune effector molecules [I]: anti-tumor necrosis factor- $\alpha$  agents). *Clin Microbiol Infect* 2018;24(Suppl 2):10. <https://doi.org/10.1016/j.cmi.2017.12.025>