



Case report

Orofacial tuberculosis: A diagnostic challenge

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ABSTRACT

Tuberculosis is typically a pulmonary chronic infectious disease with a high prevalence in developing countries which carries a substantial rate of mortality. Extrapulmonary disease may occur, mainly second to the endogenous spread of the pathogen from the primary site. Oral or mandibular involvement represents a minority among the reported cases. A 12-year-old female patient with a diffuse left-sided facial swelling and dull pain that gradually developed over 8 months, presented to us with misdiagnosis and poor management. Examination was found to have a firm swelling involving the buccal region, and left posterior mandible with intact overlying skin and mucosa, and palpable cervical lymphadenopathy. Imaging showed a heterogeneous osteolytic lesion of left ramus extending to the surrounding soft tissue. The diagnosis with oro-facial tuberculosis was established by histopathological study and confirmed by the Mantoux test and polymerase chain reaction. Although it occurs rarely, oro-facial tuberculosis has detrimental local and systemic effects, and devoid of characteristic clinical and radiographic features, poses a diagnostic challenge.

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Introduction

Tuberculosis (TB) is a chronic granulomatous disease, potentially involving multiple body systems, caused by *Mycobacterium tuberculosis* (MT) [1]. TB has been second only to HIV/AIDS as the greatest killer worldwide, especially in developing countries, where its prevalence is high [2]. About 10.4 million new cases were diagnosed in 2015 imposing a worldwide health issue [3]. Typically, the infection occurs when the bacilli transmitted to the pulmonary alveoli through the airborne route via close contact with infected persons causing pulmonary TB [1]. Extrapulmonary presentation of TB can arise mainly second to the endogenous spread of the microorganisms from the primary site, through hematogenous or lymphatic routes and seed into other organs, such as lymph nodes, meninges, kidneys, bone, and skin [2,4,5]. However, the disease may exist through direct inoculation of the infecting pathogens in the susceptible sites, without evidence of systemic involvement [6,7]. Oro-facial TB with jaw involvement is considered a rare finding, comprising about 3% of all reported cases, and posing a diagnostic challenge to the clinician [8]. The majority are usually secondary to endogenous spread of systemic infection [2,5,9].

We report a case of: oro-facial TB involving the angle, the ramus of the mandible and the surrounding masticatory muscles in a 12-year-old female patient.

Case presentation

A 12-year-old female patient came with her parent to the morning consultation clinic of our institute suffering from a diffuse swelling of the cheek and lower jaw on the left side with dull pain and discomfort aggravated by swallowing. The symptoms developed and gradually worsened over 8 months and was associated with weight loss and generalized weakness that had been subjectively attributed to the decreased nutrition and appetite. Initially, the patient suffered from a painful intraoral swelling involved the left side of the lower jaw. Management was sought at a local dental clinic, where extraction of a carious deciduous tooth in the area was performed. Later as the symptoms progressed, she was referred to the maxillofacial surgery unit of a local hospital. Two biopsies were taken separately from the region, only yielding inconclusive findings of inflammatory cells and fibrosis.

The patient was referred to our institute for further investigations and management. On examination, there was a slightly tender firm left-sided swelling involving the cheek, not crossing the lower border of the mandible with intact overlying skin (Fig. 1). The patient had normal mouth opening and showed no neurological deficit. Palpable mobile lymph nodes were evident at ipsilateral submandibular and deep cervical levels with no

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Fig. 1. Swelling involves the left cheek and the lower jaw with intact overlying skin.

tenderness. Intra-orally, the swelling involved the ipsilateral buccal region and extended medially to the posterior lower alveolus, retromolar trigone and the ascending ramus of the mandible, with erythematous overlying mucosa (Fig. 2). Computed tomography (CT) scan, revealed an expansile lesion involving the left masticatory region, extending from the condylar process to the angle of the mandible, with bicortical erosion, and osteoblastic activity (Fig. 3). The CT findings were reported as a potentially malignant tumor. Routine laboratory blood tests were unremarkable. Incisional biopsy under local anesthesia was obtained from



Fig. 2. Intra oral swelling and expansion of the involved tissue with fair oral hygiene and intact mucosa. Evidence of mucosal scar from a previous intervention over the external oblique buccal ridge.

the lesion lateral to the ramus of the mandible. The histopathological study showed a dense chronically inflamed fibrous tissue with evidence of ill-defined granulomas with scattered multinucleated Langhans giant cells. The Ziehl-Neelsen (ZN) stain for acid-fast bacilli (AFB) detection yielded negative results in both the lesion specimen and the pharyngeal expectorate. The chest x-ray of the patient revealed bilateral enlarged hilar lymph nodes. Further investigations by Mantoux test produced 25 mm of induration at 48 h, and a PCR for MTB of blood was positive. Quadruple anti-tubercular therapy was then started at a specialized center of respiratory diseases. At the latest follow-up, one year after the diagnosis, the swelling nearly disappeared with complete resolution for the accompanying symptoms.

Discussion

The jawbone is rarely affected by the tubercular infection, and the mandible is more susceptible than the maxilla [10]. Various routes have been proposed for the access of the infecting bacilli to the jaws; through dental extraction sockets, break of the mucosal lining, direct spread from adjacent tissue or via a spread of systemic disease through the hematogenous and/or lymphatic systems [11].

Clinically, this form of TB may manifest in variable pictures. It can cause superficial or central bone destruction with a variable soft tissue extent, usually accompanied by subperiosteal abscess collection that gives the lumpy jaw appearance, with or without intra- or extra- oral draining sinuses [2]. Cervical tubercular lymphadenopathy is a usual distinctive feature in these cases. It may occur as the sole presentation or may accompany other oro-facial hard and soft tissue TB, typically producing a non-tender, discrete firm, or fluctuant masses with or without draining sinuses [8]. The CT scan showed here bicortical osteolytic changes of the left ramus with an expansile heterogeneously enhanced lesion, representing the spread of the tuberculous tissue accompanied by foci of abscess collection and osteoblastic activity to the attached musculature. The imaging characteristics of oro-facial TB may mimic a neoplastic lesion [2]. Accordingly, a tentative assumption of malignancy was made initially in the absence of further investigations.

In most typical cases, the diagnosis of pulmonary TB is mainly made by the clinical and radiographic findings and confirmed by the demonstration of AFB in the sputum [12]. However, in extrapulmonary disease, the clinical and radiographic signs could be more nonspecific. The diagnosis generally needs a histological examination of tissue and demonstration of the infective organisms in the specimen [13]. In the present case, the patient had suffered from progressive symptoms for about 8 months without curative management. The diagnosis was initially delayed due to the assumption of odontogenic infection that was treated by dental extraction and multiple doses of antimicrobials. With the absence of typical and suggestive clinical signs and symptoms and inconclusive histopathological study of two biopsies, the patient was referred to our institute. The histologic picture of TB characterized by the presence of caseating and non-caseating granuloma with multinucleated Langhans giant cells [13]. AFB can be demonstrated in the specimen using ZN stain; however, that does not necessarily yield positive results [2]. In this case, the Mantoux test had produced a TB-indicative delayed hypersensitivity reaction in the forearm skin, and the PCR probe tested positive for the bacilli DNA in the patient's blood. The enlargement of the hilar lymph nodes that revealed by the patient's chest imaging, implied a primary hilar disease from previous exposure to TB, a premise that has been well endorsed by Bernardo [14] and Pagel et al. [15]. The dissemination of the disease to the cervical lymph nodes and then to the other oro-facial region from the hilar



Fig. 3. CT scan imaging. A) 3D reconstruction demonstrates the cortical osteolytic changes of the ramus. B) Coronal section shows the bicortical bone changes of the left ramus and the surrounding heterogeneous lesion of the related musculature. C) The extent of the lesion in axial view.

disease has been reported and explained through the lympho-hematogenous spread [16]. Dissemination to the posterior mandible and the related musculature, in the absence of direct AFB inoculation, can be due to the rich blood supply of the area [17]. Accordingly, Andrade attributed the distinctive cortical erosion of the ramus mostly seen in those cases to the preferential involvement of the attached muscles by the cervical disease due to their rich blood supply [18]. Efficient medical treatment of TB may necessitate 6–9 months of chemotherapy, in which, isoniazid with other frequent drug combinations of rifampin, pyrazinamide, and ethambutol constitute the cornerstone of the regimen [19].

Conclusion

Clinicians should always keep in mind the differential diagnosis of TB in the management of chronic cases that are refractory to routine therapy. Although the disease may exhibit ambiguous presentation and associated with high morbidity, the targeted investigations and the efficient collaboration between the medical personnel, prompt toward the correct diagnosis and curative treatment.

Author contribution

Sabah Abdulaziz Issa: Data curation, formal analysis, Methodology, Supervision, Validation, and Visualization.

Hussein Ali Abdulnabi: Conceptualization, Data curation, Formal analysis, Project administration, resources, Software, Writing- original draft & -review & editing.

Mustafa Emaduldeen Jameel: Data curation, Formal analysis, Investigation, Resources, Writing - original draft.

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Ethical approval

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Informed consent

Informed consent was obtained from the patient to publish the case.

Declaration of Competing Interest

None.

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