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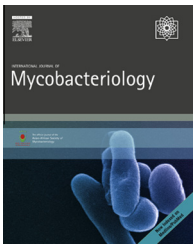

Case Report

Primary oral tuberculosis in a patient with lepromatous leprosy: Diagnostic dilemma

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ABSTRACT

Pulmonary tuberculosis (TB) is the most common form of TB. Primary infection can also affect the pharynx, cervical lymph node, intestine, or oral mucosa. Historically, the observed incidence of concomitant infection with leprosy and TB is high. However, reports of concomitant infection in modern literature remain scarce. Most cases reported in the literature had borderline/lepromatous leprosy and pulmonary tuberculosis. Extrapulmonary tuberculosis is reported in only 3.2% of leprosy cases. To the best of our knowledge, this is the first case report of primary oral tuberculosis of the tongue in a patient with lepromatous leprosy with Type 2 lepra reaction. The patient was referred to Directly Observed Treatment, Short-Course clinic and started on Category I treatment. She received oral prednisolone for lepra reaction, which was subsequently tapered and stopped, however, she continued to receive other antileprotic drugs (thalidomide and clofazimine). The patient's general condition improved and she is on regular follow up.

Case report

A 45-year-old female came to the outpatient department with complaints of low-grade fever for 2 weeks, erythema and edema of the whole body, and painless ulcers in the tip of the tongue. She had a history of leprosy, which was treated and cured 15 years ago. In the dermatology outpatient department, she was evaluated for leprosy. Slit-skin smear was taken from different sites such as the nose, eyebrows, earlobes, and the results indicated 4+ grading for all the aforementioned samples. Based on this result, the patient was diagnosed as a case of lepromatous leprosy with Type 2 reaction and we decided to treat her with multidrug therapy.

To examine the pathology of the ulcer of the tongue, sample tissue from the tip of the tongue was collected and sent for various examinations such as KOH mount and fungal culture, Giemsa stain for histoplasmosis, bacterial culture and acid-fast staining, and histopathological examination. Surprisingly, Ziehl–Neelsen staining showed acid-fast bacilli, which were beaded and not uniformly stained, a finding characteristic of tubercle bacilli. Histopathological examination of the sample showed a granulomatous reaction with central caseous necrosis and epithelioid and giant cells suggestive of tuberculosis (Fig. 1).

Nested polymerase chain reaction was performed after extracting the DNA from the sample tissue. The extracted

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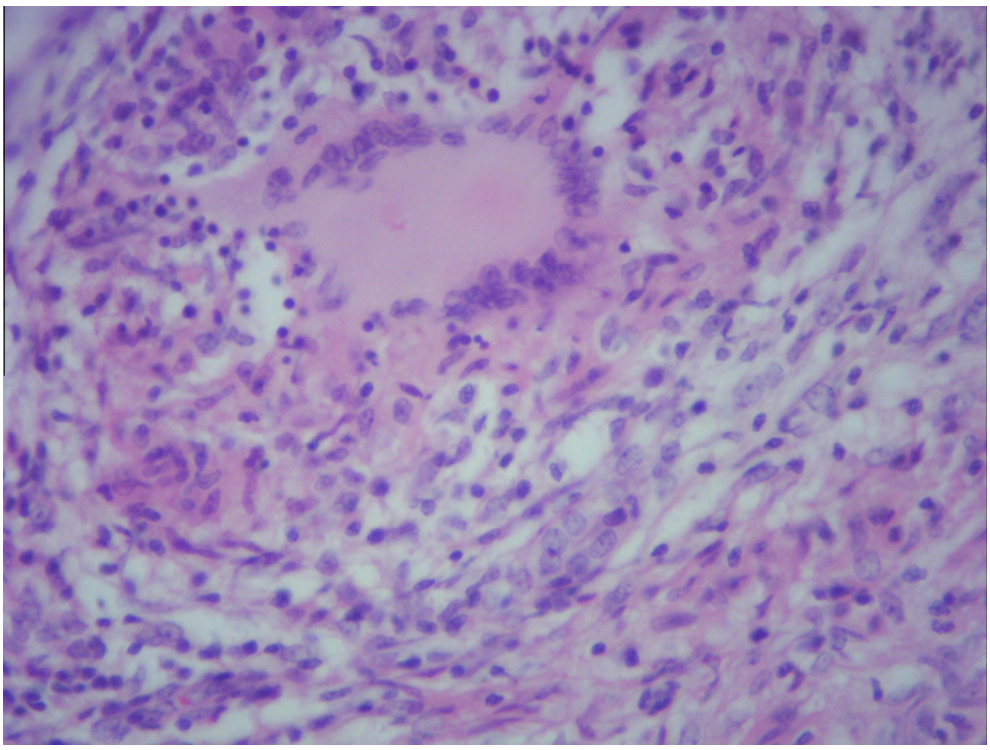

Fig. 1 – Histopathological examination showing central caseous necrosis and epithelioid and giant cells suggestive of tuberculosis.

DNA was amplified using two sets of primers coding for IS6110 (insertion sequence), which is specific for *Mycobacterium tuberculosis*. After amplification, gel electrophoresis was carried out, which showed a band corresponding to IS6110 (219 bp; Fig. 2). Based on this result, oral tuberculosis of the tongue was diagnosed. To identify the primary focus of infection, we carried out further examinations. Chest X-ray showed clear lung fields without features of tuberculosis. Computed tomography of the thorax and abdomen did not reveal any primary focus. Venereal Disease Research Laboratory and human immunodeficiency virus (HIV) tests were not reactive. A provisional diagnosis of primary oral tuberculosis of the tongue in lepromatous leprosy was made and we initiated treatment for both leprosy and tuberculosis. The patient was advised to stop taking dapsone and instead received oral prednisolone, thalidomide, and clofazimine. The patient was referred to the Directly Observed Treatment, Short-Course clinic and started on Category I treatment. The oral prednisolone dose was subsequently tapered and stopped, however, she continued to receive the other antileprotic drugs. The patient's general condition improved and she was on regular follow up. We obtained Institutional Ethical Committee approval and informed consent from the patient for reporting this case.

Discussion

Historically, the observed incidence of concomitant infection with leprosy and tuberculosis (TB) is high. However, reports of concomitant infection in modern literature remain scarce. Estimation of the annual new case detection rate (ANCDR) in India, where both TB (ANCDR 181/100,000 in 2011) [1] and leprosy (ANCDR 10–35/100,000 in 2011) [2] remain endemic, suggests that only 0.019 cases of concomitant infection/100,000 population would be detected.

It is hypothesized that reduced cell-mediated immunity plays a role in reactivation of latent TB or superinfection with TB in multibacillary patients. Trindade et al. [3] recently investigated the cell-mediated responses of two patients who were diagnosed with borderline leprosy and TB [3]. However, they were unable to find any aberrant response of the interferon-gamma/interleukin-12/23 axis on immunological evaluation. Therefore, further studies are needed to support this hypothesis.

Although there are many well-known risk factors for TB, including HIV infection, diabetes mellitus, transplant patients on immunosuppression, and birth and travel in the developing world, the use of steroids and development of TB are controversial. One weakness in the argument for steroids increasing the risk of developing TB is that a large number of leprosy patients, especially those with multibacillary leprosy, go on to develop lepra reactions, which require steroid treatment. This means that there is a high rate of steroid prescriptions in leprosy cases. With an estimated ANCDR for co-infection of 0.019/100,000 patients (in India), the incidence of patients started on steroids is approximately 66 times greater than the estimated incidence of co-infection with both diseases [4]. Therefore, longitudinal work with active screening of patients before commencing treatment is necessary to identify any temporal relationship between steroids and TB development.