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Coexistence of tuberculous chancre on a chronic nonhealing wound and bilateral submandibular scrofuloderma

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Abstract

Cutaneous tuberculosis is caused by *Mycobacterium tuberculosis* with varied clinical features depending on the dissemination route and host immune status. Scrofuloderma is a type of cutaneous tuberculosis that often occurs in locations where there is an infected lymph node or bone underneath, whereas tuberculous chancre often appears in sites that are prone to trauma. Although several cases have been reported, the coexistence of more than one type of cutaneous tuberculosis is very rare. We report a 21-year-old immunocompetent male with a chronic nonhealing lesion on the left tibia followed by bilateral purulent ulcers on the submandibular area. Acid-fast bacilli examination was positive for *M. tuberculosis*, and the patient was diagnosed with tuberculous chancre and scrofuloderma, respectively. This case showed the importance of high clinical suspicion of cutaneous tuberculosis and the possibility of the coexistence of more than one type of cutaneous tuberculosis in order to avoid misdiagnosis and delay in treatment.

Keywords: coexistence, scrofuloderma, tuberculous chancre, chronic nonhealing wound

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Introduction

Tuberculosis is an infection caused by *Mycobacterium tuberculosis* and, although it most commonly affects the lung, it may infect any organ, including the skin. Out of 14% of extrapulmonary tuberculosis cases, only 1 to 2% are of cutaneous origin (1). Cutaneous tuberculosis can be further classified based on clinical presentation, inoculation route, and host immune status (2, 3).

Scrofuloderma is a type of cutaneous tuberculosis resulting from direct mycobacterial spread from organs under the skin such as the lymph nodes, especially the cervical lymph node (tuberculous lymphadenitis) (4). Scrofuloderma in the submandibular area is often associated with pulmonary tuberculosis. Tuberculous chancre, on the other hand, is caused by the entry of *M. tuberculosis* through the use of non-sterile syringes, tattooing, mouth-to-mouth resuscitation, tooth extraction measures, or surgical wounds (1). However, despite the various clinical presentations of cutaneous tuberculosis, it usually occurs as a single entity, and the coexistence of more than one type of cutaneous tuberculosis in the same subject is very rare.

We report a rare case of tuberculous chancre masquerading as a chronic nonhealing lesion followed by the development of bilateral submandibular scrofuloderma.

Case report

A 21-year-old man presented with a chief complaint of non-healing postoperative wounds on the left tibia following a closed tibial fracture procedure 5 months earlier. In addition, 4 months after the surgery, two lumps appeared on the left and right submandibular areas that developed into nodules and ulcers. No skin lesion was reportedly observed before surgery. There was no fever or pain, and prolonged cough > 2 weeks was denied. The patient had received multiple courses of various antibiotic therapy after surgery to no avail. The patient had no history of tuberculosis treatment, but his mother and relatives living in the same house had completed 6 months of therapy for pulmonary tuberculosis. The patient had no remarkable medical history. He had been vaccinated with the Bacillus Calmette–Guérin (BCG) vaccine.

Physical examination revealed several large nodules overlying bilateral cervical lymph nodes with ulceration, caseous secretion, and a livid edge in the right submandibular area (Fig. 1). On the left tibia an ulcer with livid border and caseous secretion on the edge of the surgical wound with an edematous and erythematous surrounding was observed (Fig. 2). There was no inguinal lymph node enlargement.

Radiograph examination of the chest and tibia was within normal limits and showed no signs of lung tuberculosis or osteomyelitis, respectively. Ziehl–Neelsen staining of the lesion on the submandibular area was positive for acid-fast bacilli. GeneXpert MTB/RIF was positive and HIV serology test was negative. Histopathological examination of the lesion on the left tibia showed pseudoepitheliomatous epidermal hyperplasia and epithelioid granuloma with Langhans giant cells and lymphocyte infiltration (Fig. 3).

Based on the clinical features and supporting examination, a diagnosis of tuberculous chancre of the left tibia and bilateral submandibular scrofuloderma were established. The patient was treated with category 1 tuberculosis treatment consisting of a 2-month intensive phase and 4-month continuation phase. The intensive phase regimen comprised of rifampicin 600 mg/day, ethambutol 100 mg/day, isoniazid 450 mg/day, and pyrazinamide 1,000 mg/day. This was followed by a continuation phase that comprised of rifampicin 600 mg and isoniazid 450 mg given three times weekly. Both lesions showed significant clinical improvement after 2 weeks and healed completely after a month of therapy (Fig. 4).

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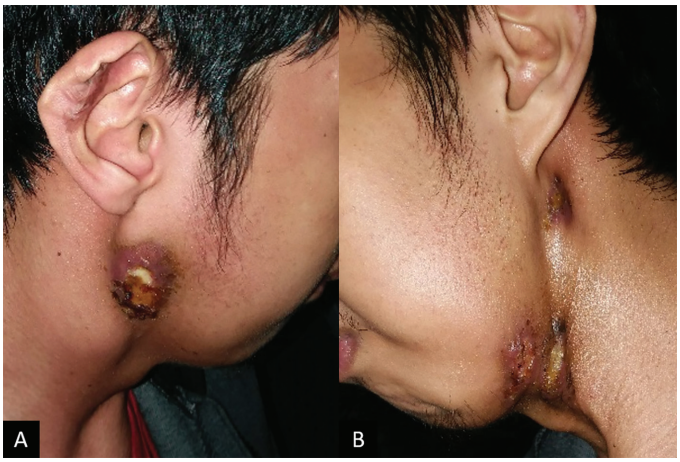

Figure 1 | Nodules on the right (A) and left (B) submandibular area.

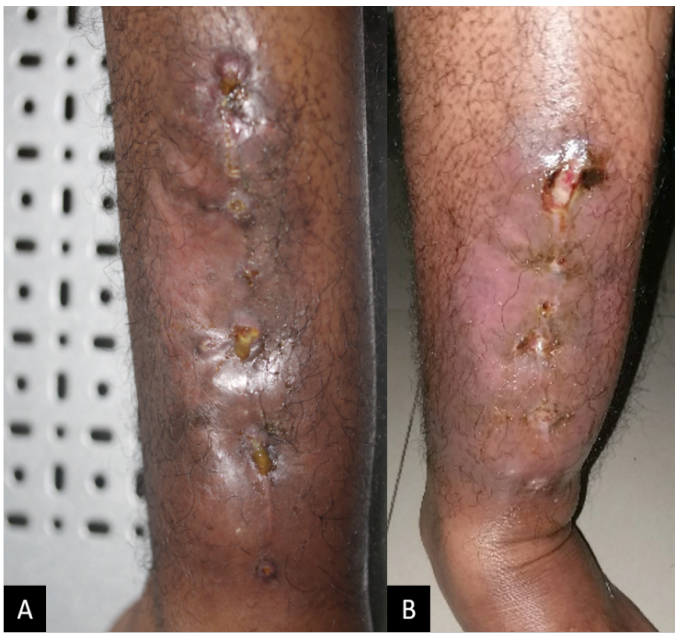

Figure 2 | Chronic non-healing ulcer on the left tibia (A), which was purulent (B).

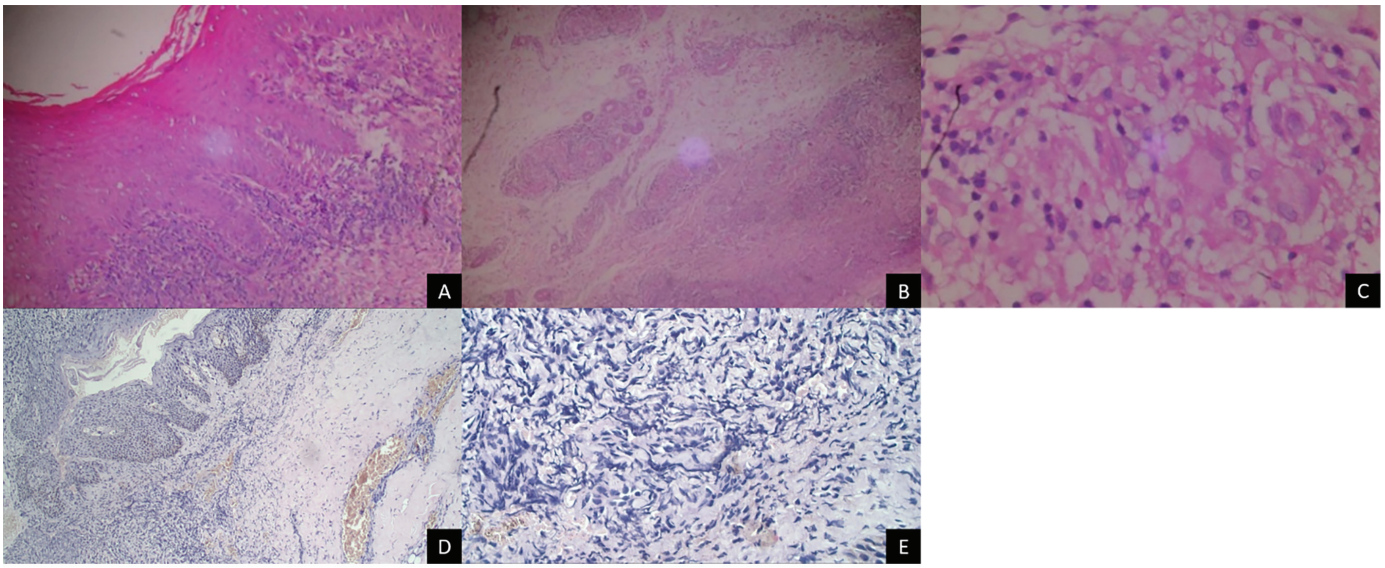

Figure 3 | Histopathological examination of the lesion on the left tibia showed (A) pseudoepitheliomatous epidermal hyperplasia (4×) and (B and C) epithelioid granuloma with Langhans giant cells with lymphocyte infiltration (40× and 100×, respectively). Histopathological examination of the lesion on the neck showed (D) psoriasiform epidermal hyperplasia (4×) and (E) epithelioid histiocytes with lymphocyte infiltration (100×).

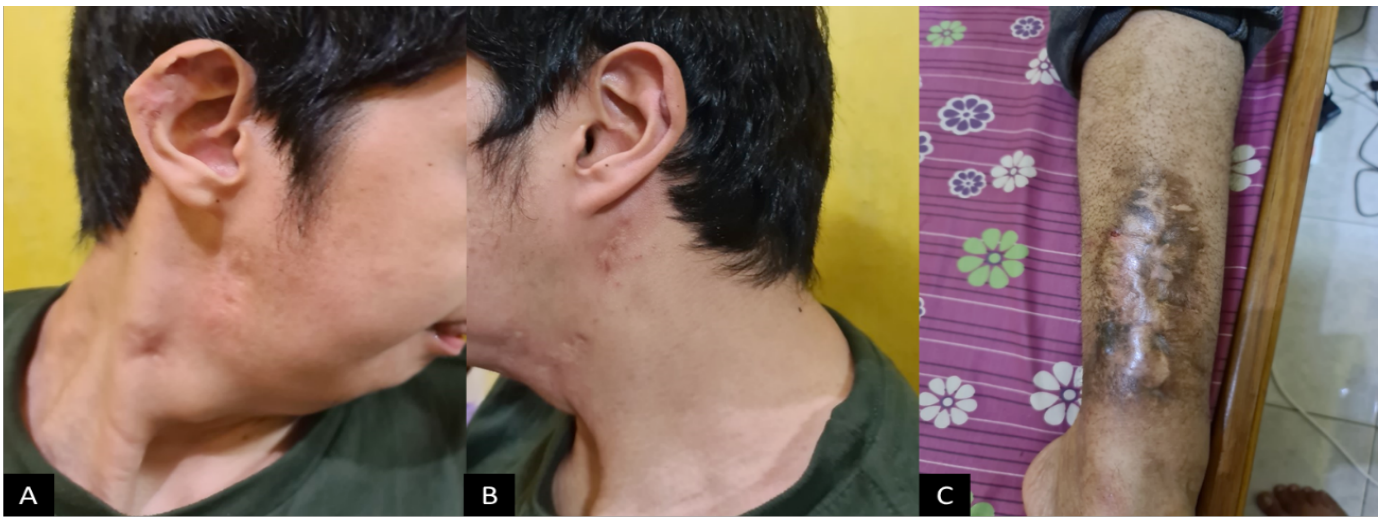

Figure 4 | Significant improvement of all lesions on the submandibular area (A and B) and left tibia (C).

Discussion

Inoculation of *M. tuberculosis* into the skin can occur exogenously through syringes, tattooing, dental extraction surgery, injury, or a surgical wound, and is known as a primary infection or tuberculous chancre (4). Clinically, it presents as brownish-red papules or a plaque with an incubation period of 2 to 4 weeks that softens and forms an ulcer with a firm, undermining border within a few weeks. Within 3 to 8 weeks after the primary lesion arises, regional lymphadenopathy, appearing as a cold abscess, with ulcers and sinuses with caseous fluid may develop. The coexistence of primary lesions and lymphadenopathy is known as primary complexes (3). Owing to its multibacillary nature, bacilli dissemination may occur through the lymphatic flow and eventually reach distant lymph nodes (1). Detection of *M. tuberculosis* in circulation is related to the dissemination to multiple lymph nodes in patients with lymphadenitis tuberculosis (5). The patient in this case had a nonhealing wound for more than 4 months with a clinical picture of the livid-colored ulcer that was consistent with tuberculous chancre. Exogenous bacilli inoculation might have occurred during the initial trauma resulting in the fracture, during an intraoperative procedure, or during wound healing. In this case, we suspected that the inoculation occurred postoperatively because the family members residing in the same house as the patient were being treated for pulmonary tuberculosis. One hypothesis is that the inoculation occurred 5 months earlier, post-surgery, when the treated family members with lung tuberculosis were still contagious.

The development of nodules in the left and right submandibular areas indicated a lymphatic spread from the lesion on the tibia. Although the primary focus of scrofuloderma originated from the lung in the majority of cases, the normal findings on chest X-ray and acid-fast bacilli sputum examinations excluded the possibility of pulmonary tuberculosis as the primary focus of the infection and confirmed the diagnosis of tuberculous chancre and resultant bilateral scrofuloderma, respectively. It is interesting to observe that the scrofuloderma was located distant from the tuberculous chancre and that there was no regional lymph node (inguinal lymph node) enlargement. A similar phenomenon has been noted in previous reports (6–8). In addition to the distant location, scrofuloderma was found in both sides and not only on the same side as the tuberculous chancre.

The coexistence of more than one type of cutaneous tuberculosis is rare. Tuberculosis verrucosa cutis has been reported along with scrofuloderma (6, 9) and lupus vulgaris (8, 10), and a recent study reported the coexistence of tuberculous gumma and tuberculosis verrucosa cutis (11). However, to our knowledge, this study is the first to report the coexistence of tuberculous chancre and scrofuloderma. In addition, our case was interesting because the emergence of scrofuloderma served as a central clinical clue in revealing the diagnosis of tuberculosis chancre that masqueraded as a chronic nonhealing wound following a tibial fracture operation. This further signified the importance of high clinical suspicion of cutaneous tuberculosis due to its varied clinical manifestations.

Conclusions

The coexistence of tuberculous chancre and bilateral submandibular scrofuloderma is rare. Physicians should be vigilant of this possibility to avoid misdiagnosis and delay treatment.

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