# Orificial Tuberculosis: Presenting as a Refractory Perianal Ulcer

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*Sir,*

Orificial tuberculosis (OTB) is a very rare form of true cutaneous tuberculosis, which is clinically characterized by the appearance of ulcerative lesions affecting the oral, genital or anal mucosa and adjacent skin as a result of infectious spread from the primary focus in the lung or gastrointestinal tract (1). Yates & Ormerod (2) reported that among 47 cases of cutaneous tuberculosis in the Blackburn district, UK, during a 15-year period, only one case had OTB. We report here a 55-year-old Japanese patient who appeared healthy but had OTB and pulmonary tuberculosis.

## CASE REPORT

A 55-year-old Japanese man presented with a 1-year history of painful perianal ulceration. His past medical history and family history were unremarkable. He had developed a subcutaneous perianal nodule with a malodorous discharge about 1 year earlier which had been surgically treated under diagnosis of perirectal abscess, although histological examination was not taken at this time. Subsequently, the lesion became ulcerated and gradually enlarged despite conventional therapeutic treatment for 1 year.

Clinical examination showed a painful superficial ulceration about 5 cm in diameter with a cheesy and malodorous discharge. The lesion was reddish coloured, tender, haemorrhagic and partly necrotic. The edge was slightly elevated. The lesion extended from the perianal region to mainly the right buttock, but slightly extended to the left buttock as well (Fig. 1). There were no other cutaneous eruptions nor regional lymphadenopathy. He denied any constitutional symptoms such as weight loss, night sweat, fever, cough or abnormal bowel habit.

A skin biopsy taken from the ulcerated area showed an epithelioid granulomatous inflammatory infiltrate with central caseous necrosis, surrounded by Langhans'-type giant cells (Fig. 2A). Acid-fast bacilli were seen in the granuloma and necrotic area by Ziehl-Nielsen stain (Fig. 2B) and were rapidly confirmed by polymerase chain reaction (PCR) as *Mycobacterium tuberculosis*. Cultures from the biopsy tissue and the ulcer exudates also grew *M. tuberculosis*.

The chest X-ray showed bilateral patchy infiltrates affecting the upper and middle field (Fig. 3). The results of PCR and culture of sputum were positive for *M. tuberculosis*, but those from urine and stool were negative. Colonoscopy only demonstrated a polyp in the anal canal. Laboratory results demonstrated normal complete blood count, elevated C-reactive protein of 5.1 mg/dl (normal 0.0–0.4 mg/dl), while other biochemistry data were normal. Hepatitis virus, syphilis and HIV serologies were non-reactive. The Mantoux test was positive.

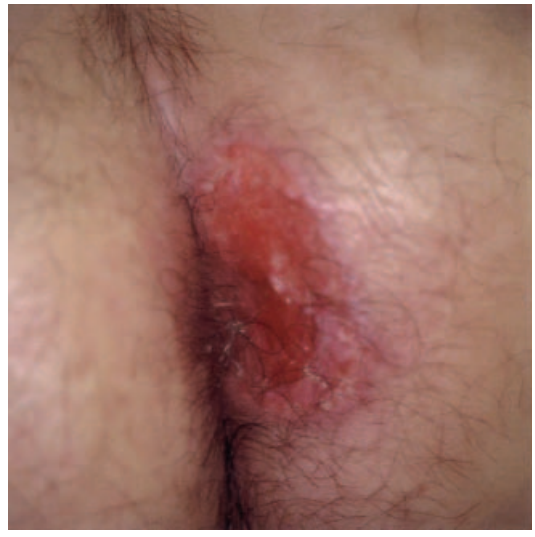

*Fig. 1.* Perianal ulcer with cheesy and malodorous discharge.

The patient was hospitalized with a diagnosis of OTB and pulmonary tuberculosis. He was treated with isoniazid 400 mg daily, rifampicin 450 mg daily, pyrazinamide 1200 mg daily and ethambutol 750 mg daily. The ulcer promptly improved but the treatment was discontinued due to liver dysfunction 2 weeks later.

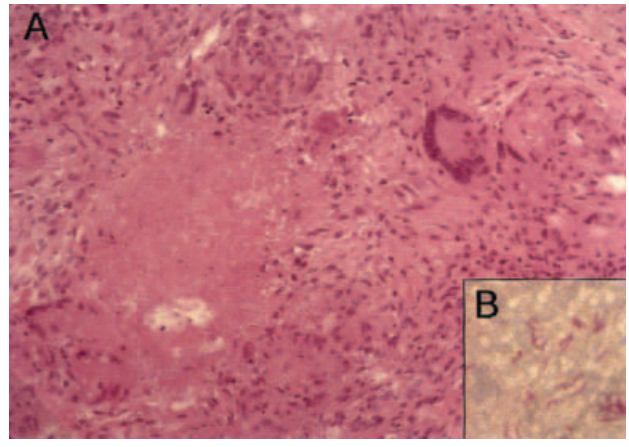


*Fig. 2.* (A) Skin biopsy showing an epithelioid granulomatous inflammatory infiltrate with central caseous necrosis, surrounded by Langhans'-type giant cells (haematoxylin-eosin stain ×100). (B) Numerous acid-fast bacilli (Ziehl-Nielsen stain ×200).



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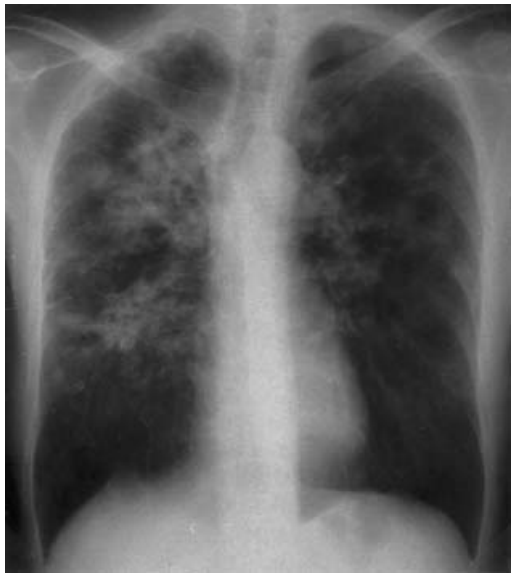

*Fig. 3.* Chest X-ray showing bilateral patchy infiltrates affecting the upper and middle field.

*In vitro* testing later showed *M. tuberculosis* to be susceptible to all antituberculosis drugs except for ethambutol. Over the next month, the ulcer enlarged again to the same size. A new, modified regimen with streptomycin 1000 mg every other day, isoniazid 400 mg daily and rifampicin 150 mg daily was administered. The ulcer completely healed after 2 months of therapy. Sputum culture became negative. The treatment was discontinued after a total of 9 months and there has not been any recurrence during 6 months of follow-up.

## DISCUSSION

Tuberculosis of the skin is classified into (i) exogenous infection and endogenous spread; (ii) conditions caused by BCG vaccination; and (iii) tuberculids (3). OTB is a very rare form of endogenous spread of *M. tuberculosis* characterized by rapidly ulcerating brownish papules in the oropharynx or gastroanal tract (4). The course of the disease might be acute and due to the miliary spread of bacilli; untreated OTB frequently leads to death within months. OTB arises as a result of auto-inoculation of the acid-fast bacilli from endogenous sources such as the lungs, gut or urinary tract. Other possible routes of spread are haematogenous, lymphatic or direct extension (5). The most widely accepted hypothesis in the case of perianal tuberculosis is that the skin lesions arise as a consequence of auto-inoculation from swallowed bacilli-containing sputum into defects in the perianal mucosa, usually after trauma (1, 3). This mechanism might be involved in our case. To our knowledge, only 11 cases of perianal ulceration as OTB have been reported in the English literature so far. Concomitant pulmonary tuberculosis was present in nine of these cases (1, 2, 5–9).

A diagnosis can be made by histopathological examination and culture from skin biopsy. PCR is also an effective diagnostic technique, as it allows a more rapid diagnosis of rare or atypical manifestations of tuberculosis infection. Cultures should always be done to confirm PCR results and determine drug resistance (4).

OTB is usually a symptom of advanced pulmonary, intestinal or, rarely, genitourinary tuberculosis with a most unfavourable prognosis (3). Our case, however, is very rare in that the patient was apparently healthy and free from systemic symptoms. Although cutaneous tuberculosis is currently rare, it is important to include OTB in the differential diagnosis of recalcitrant skin ulcer and perform skin biopsy and culture to avoid any undue delay in the diagnosis and treatment of the condition.

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