Scrofuloderma from the acromioclavicular joint presenting as a chronic ulcer in an immunocompetent host

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ABSTRACT

A 53-year-old woman presented with a chronic, non-healing painless ulcer over her right clavicular area of a two-month duration. Skin biopsy, microbiological and radiological investigations confirmed the diagnosis of scrofuloderma arising from an underlying tuberculous infection of her acromioclavicular joint. She was treated successfully with anti-tuberculosis therapy with complete healing of the ulcer. Awareness of this uncommon presentation of osteoarticular tuberculosis may assist in earlier diagnosis.

Keywords: acromioclavicular joint, articular tuberculosis, cutaneous tuberculosis, scrofuloderma, tuberculosis.

Singapore Med J 2007; 48(9):e243–e245

INTRODUCTION

There has been a rise in the incidence of tuberculosis in recent years. This increase appears to be due to several factors including development of resistance to the most commonly-used chemotherapeutic substances, human immunodeficiency virus (HIV) infection, and increased travel and immigration. In spite of this, cutaneous tuberculosis remains an uncommon infection. Osteoarticular tuberculosis, especially with involvement of sites other than the spine or weight-bearing joints, is also rare. Scrofuloderma, which refers to the involvement of the skin overlying a contiguous tuberculosis focus, may uncommonly arise from an infected bone or joint. We report an unusual case of scrofuloderma arising from an underlying infection of the acromioclavicular joint in an immunocompetent female adult.

CASE REPORT

A 53-year-old woman presented with a two-month history of a chronic painless non-healing ulcer over her right clavicular area associated with purulent discharge.

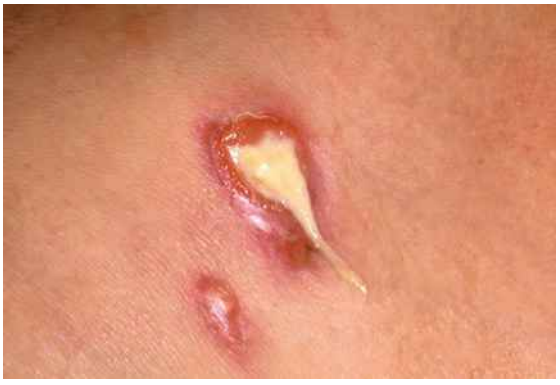

Fig. 1 Photographs shows a suppurative ulcer with a sloughy, purulent base, surrounding erythema and induration.

The lesion started as a papule that subsequently ulcerated with copious purulent discharge. There was no preceding history of trauma. Systemic review was unremarkable, with no recent weight loss or unexplained pyrexia. She was from a neighbouring developing country and had been working in Singapore for the past 14 years. Physical examination showed a 1 cm punched-out, suppurative ulcer with a sloughy purulent base, with surrounding erythema and induration (Fig. 1). Her right shoulder's range of movement was also limited by the pain. There were no palpable lymph nodes and systemic examination did not reveal any abnormalities.

Routine laboratory investigations were normal, except for a raised erythrocyte sedimentation rate of 77 mm/hour. An initial pyogenic culture yielded the growth of *Enterococcus* species and a course of systemic antibiotics was prescribed. Failure to respond to therapy prompted further investigations. A punch biopsy specimen obtained from the edge of the ulcer showed the presence of caseating granulomas (Fig. 2). Ziehl-Neelson (ZN) stain and periodic acid-schiff stain of the specimen were negative. Mantoux test was positive with erythema and induration of 15 mm. Radiograph of the right shoulder joint revealed dislocation of the acromioclavicular joint with erosions at the lateral end of the clavicle and multiple small bone debris within the joint, consistent

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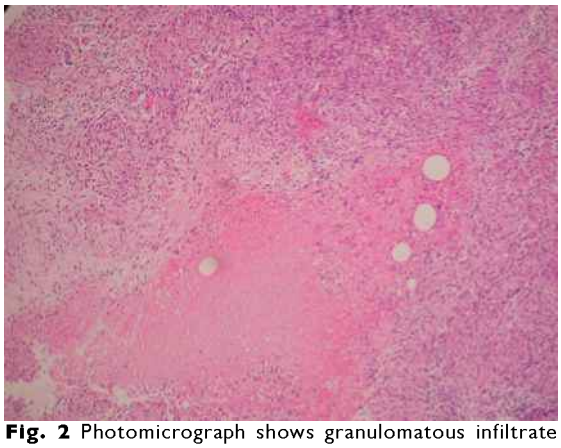

Fig. 2 Photomicrograph shows granulomatous infiltrate of epithelioid cells, lymphocytes and multinucleated giant cells surrounding a caseating necrotic core (Haematoxylin & eosin, ×100).

Fig. 3 Right shoulder joint radiograph shows dislocation of the acromioclavicular joint with erosions at the lateral end of the clavicle and multiple small bone debris within the joint.

Fig. 4 Follow-up photograph shows the healed ulcers with scarring over both supraclavicular regions.

with infective arthritis and osteomyelitis (Fig. 3). Chest radiograph did not show any pulmonary disease. Mycobacterial culture of the tissue specimen was positive for fully sensitive *Mycobacterium tuberculosis,* but the detection of *M. tuberculosis* DNA by polymerase chain reaction (PCR) turned out negative. Fungal cultures were also negative. HIV serology was non-reactive.

A diagnosis of scrofuloderma with underlying tuberculous infection of her right acromioclavicular joint was made and the patient was commenced on rifampicin, isoniazid, ethambutol and pyrazinamide. One week after initiation of treatment, she reported clear improvement of symptoms with decreased pain and an increase in the right shoulder's range of movement. Four months after initiation of anti-tuberculosis treatment, she developed left supraclavicular non-tender lymphadenopathy that subsequently broke down, forming an ulcer. A possibility of non-compliance to treatment was suspected and the patient was placed on directly observed therapy with improvement of the ulcer. Rifampicin, isoniazid, pyrazinamide and ethambutol were given for a total of five months, followed by seven months of dual therapy with rifampicin and isoniazid, with eventual complete healing of both ulcers (Fig. 4).

DISCUSSION

Cutaneous tuberculosis remains a rare infection, with an incidence of 3.5% reported among patients with organ tuberculosis.[1] Although scrofuloderma is one of the commonest forms of cutaneous tuberculosis reported in some series,[2] most cases resulted from an infected lymph gland, and less commonly as a result of an infected joint or bone. There have also been reports of scrofuloderma following surgical drainage of the joint[3] and infection of the lacrimal system.[4] This case highlights scrofuloderma arising from underlying acromioclavicular joint involvement which is exceedingly rare. Tuberculous osteomyelitis and arthritis are also uncommon, especially with involvement of sites other than the spine or weight-bearing joints, accounting for 10% of extrapulmonary tuberculosis. It runs a long and insidious course, and unusual presentation with chronic discharging sinus or non-healing ulcer is possible.[5] A long preceding history may be present for scrofuloderma, with chronic infection of a 35-year duration previously reported.[6] As seen in this case, concomitant infection with aerobes or anaerobes frequently occur, preventing the early diagnosis of tuberculosis if not suspected. Presence of purulent discharge or features suggestive of a carbuncle can cause further confusion and misdiagnosis as a bacterial abscess.[7] Other diagnoses that need to be considered include deep mycotic infections, melioidosis, atypical mycobacteriosis, tertiary syphilis and cutaneous malignancies.

Although a positive culture remains the gold standard for diagnosis of tuberculosis, PCR may

actually have a higher sensitivity than culture. A further advantage for PCR is the possibility for early diagnosis and institution of treatment in these patients. In a prospective study by Negi et al comparing the sensitivity of PCR test with conventional ZN-stained microscopy, conventional Lowenstein-Jensen (LJ) culture and radiometric BACTEC system, a significant difference was seen in the sensitivities for different tests at 74.4% for PCR test, 33.79% for ZN smear examination, 48.9% for LJ culture and 55.8% for BACTEC culture.[8] In pulmonary tuberculosis, where the uses of PCR tests are most widely evaluated, sensitivities of 40%–70% and specificity of about 95% have been reported for smear-negative (paucibacillary) disease. A previous study at our centre found an overall sensitivity of 73% of the PCR assay for patients with paucibacillary cutaneous tuberculosis, when only one out of the 14 cases, where tissue cultures were performed, yielded a positive culture result.[9] The authors concluded that when paucibacillary tuberculosis is suspected, a negative PCR test does not rule out the diagnosis of cutaneous tuberculosis, but a positive test result can enhance the clinician's confidence in instituting anti-tuberculous treatment. Interestingly, DNA of *M. tuberculosis* was not detected in our patient, though mycobacterial culture was positive. A plausible explanation for the negative PCR result in our patient may be due to sampling error. In another study, it was reported that only one out of three patients with scrofuloderma had a positive PCR result. In this study, the patient with a positive PCR result had disseminated tuberculosis with positive smear and culture for *M. tuberculosis* as well.[10] As scrofuloderma is usually a paucibacillary condition, a negative PCR result is not unexpected. In addition, any discharge should also be sent for mycobacterial smear and culture.

A six-month regimen including four drugs in the initial two months (rifampicin, isoniazid, pyrazinamide plus ethambutol or streptomycin), followed by rifampicin and isoniazid in the four-month continuation phase is highly effective in patients with fully sensitive organisms. This standard six-month regimen is now recommended by the British and American Thoracic Societies. For osteoarticular tuberculosis, the American Thoracic Society recommends six- to nine-month duration of therapy for patients with drug sensitive disease.[11] In addition, a high index of suspicion of non-compliance to treatment is necessary in patients with relapse or new areas of involvement mid-therapy, as highlighted in this case. Prolonged therapy should also be considered for patients slow to respond to otherwise adequate treatment and in the presence of dissemination. Though not necessary for cure, surgery may play a supportive role in draining of abscesses, debridement, fusion or replacement of joints that are significantly damaged. Tissue or fluid obtained during surgery can also be sent for diagnostic confirmatory studies.

A chronic, non-healing ulcer is an uncommon presentation of osteoarticular tuberculosis. An increased awareness of the re-emergence of cutaneous tuberculosis will allow for the prompt diagnosis and management of this increasingly common skin disorder.

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