

A Case of Recalcitrant Actinomycosis Unresponsive to Antibiotic Therapy

Dear Editor,

Actinomycosis is a chronic granulomatous disease caused by gram-positive, anaerobic *Actinomyces* bacteria.¹ These commensal microbes can cause opportunistic infection following mucosal breach. Classically, histology shows granulomatous inflammation with abscesses or sinuses containing sulphur granules, bearing clusters of gram-positive, non-acid-fast bacteria with club-shaped ends within dilated infundibulocystic cavities.² Treatment involves long-term antibiotics, sometimes with adjunctive surgery.

We introduce an 86-year-old retired bus driver who presented with a 3 x 4 cm discoloured right thigh induration (Fig. 1), stable for more than 30 years without treatment. During the past year, it had occasionally expressed purulent, foul-smelling discharge. He denied prior trauma, soil exposure and insect bites.

Gram stain showed many gram-positive cocci in clusters and few gram-negative bacilli. Cultures yielded methicillin-resistant and sensitive *Staphylococcus aureus* (MRSA and MSSA). An x-ray excluded bony involvement. A skin biopsy was done, showing pseudoepitheliomatous hyperplasia with dilated infundibulocystic cavities containing keratinaceous debris and large clumps of gram-positive bacteria, in association with chronic dermal inflammation (Fig. 1). Together with the clinical presentation, actinomycosis was considered the most appropriate diagnosis.

The patient was given repeat, prolonged courses of amoxicillin-clavulanate, ciprofloxacin and cephalexin. Despite bouts of transient improvement, the lesion continued to enlarge to 11 x 10 cm, with visible sulphur granules 2 years later. Repeat cultures showed MSSA, *Escherichia coli* and *Pseudomonas*. Oral clindamycin and trimethoprim-sulfamethoxazole were given.

Five years after first presentation, the lesion was 40 x 15 cm, from greater trochanter to knee (Fig. 1). Repeat biopsies showed similar histology, but no organisms. Itraconazole was given empirically for occult fungal infection or eumycetoma, without clinical improvement. Cultures again yielded *Actinomyces*, *Enterococcus* and *Pseudomonas*. The patient was restarted on ciprofloxacin, amoxicillin-clavulanate and trimethoprim-sulfamethoxazole.

Actinomycosis is rare and can mimic malignancy and other infections.³ Moreover, the skin is an unusual site and this



Fig. 1. Slide A shows the patient's first presentation with a 3 x 4 cm hyperpigmented, non-tender induration on the right posterior thigh. In slide B, histology from skin biopsy shows pseudoepitheliomatous hyperplasia with dilated infundibulocystic cavities containing keratinaceous debris, large clumps of gram-positive bacteria and occasional neutrophilic foci. There was a mixed dermal infiltrate of lymphocytes, histiocytes, plasma cells and neutrophils. (Hematoxylin and Eosin stain, magnification x 25). Slide C shows the lesion 5 years later, showing extension with sinus tracts and sulphur granules.

patient lacked risk factors, including diabetes and trauma.⁴ Co-infection with "companion bacteria" (e.g. *Staphylococci*, *Streptococci*) is common in actinomycosis⁵ and may render diagnosis challenging, as isolation of *Actinomyces* is commonly unsuccessful.¹ Less likely differentials include botryomycosis and mycetoma, both also chronic granulomatous infections with sulphur granules. Mycetoma infections are subdivided into eumycetoma, caused by fungal organisms, and actinomycetoma. Actinomycetoma is caused by aerobic, sometimes weakly acid-fast gram-positive filamentous bacteria (e.g. *Nocardia*), in contrast to the *Actinomyces* species implicated in actinomycosis. Another differential is pyoderma vegetans, although histology here showed no suprabasal clefting or acantholysis.

While *Actinomyces* rarely develop penicillin resistance, coverage for co-pathogens is essential.¹ Interestingly, despite multiple prolonged antibiotic courses, this patient's lesion remained progressive. He declined amikacin and spectinomycin, as well as surgical management. Going forward, he will likely require continued long-term antibiotics.

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