Primary Cutaneous Actinomycosis of the Femorogluteal Region: Two Case Reports

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Actinomycosis is a rare, chronic infection characterized by induration, multiple abscesses and draining sinuses (1). Cervicofacial actinomycosis is the most common form, which is usually caused by deep oral mucosal injury. The major pathogen is *Actinomyces israelii*, a commensal of the oropharyngeal flora (2). Actinomycosis of the lower part of the body, especially the primary cutaneous form, is very unusual (3–5). *A. turicensis* is a recently identified distinct Actinomycetes species (6) found particularly in infections of the lower part of the body, especially in the anorectal and urogenital regions (3, 7).

CASE REPORTS

Case 1. A 47-year-old man presented with extensively spreading, superficial ulcerations, erythematous indurated plaques and nodules in the femorogluteal, perianal and genital regions for 3 years (Fig. 1A). Routine laboratory examination revealed elevated C-reactive protein levels (1.81 mg/dl; normal < 0.5 mg/dl), elevated neutrophil counts (77%; normal 50–70%) and lymphopaenia (13.3%; normal 25–40%) with a total lymphocyte count of 1,091/µl. HIV and syphilis serologies were non-reactive. Bacteriological culture of a biopsy demonstrated *A. turicensis*, *Prevotella intermedia* and *Eubacterium limosum*. *Staphylococcus aureus*, *Citrobacter koseri*, *pseudomonas* species and streptococci (group A) could be co-cultured from a bacterial swab. Histological examination revealed inflammatory infiltrate composed of neutrophils showing focal epidermotropism, lymphocytes, histiocytes, plentiful plasma cells, and focal granulomatous inflammation with multinucleated giant cells. In the subcutaneous interlobular septa, fistulas and fibrosis were detected. No sulphur granules were found. Anti-bacille Calmette-Guérin antibody immunostain (BCG), periodic acid-Schiff (PAS), and Giemsa stains were negative. Computed tomography (CT) showed no relevant abnormalities. Intravenous treatment with benzylpenicillin (24 million units/day) was started in combination with oral trimethoprim+sulphamethoxazole (320+1,600 mg/day) for 6 weeks, followed by amoxicillin (2 g/day) orally for 3 months. Five months later, a decrease in the number and extent of ulcerations, induration and nodules was detected, and the exudate disappeared (Fig. 1B).

Case 2. A 69-year-old man without comorbidities had a one-year history of indurations, ulcerations and nodules in the right femorogluteal region, with fistulas, foetid secretion, and enlarged regional lymph nodes (Fig. 2A).

Routine laboratory blood examinations and urinalysis were normal, except for highly elevated C-reactive protein levels (15.2 mg/dl; normal < 0.50 mg/dl) and lymphopaenia (13.0%; normal 25.0–40.0%) with a total lymphocyte count of 1,170/µl. Microbiological cultures of the exudate showed the presence of *A. turicensis* accompanied by secondary flora (*Prevotella bivia*, *Streptococcus pyogenes*, *S. aureus*, *Enterococcus faecalis*, intestinal flora). In addition, microbiological culture of a biopsy showed *Bacteroides* species. Histological findings comprised chronic fistulating, scarring dermatitis, and were almost identical to the findings in case 1, including the absence of sulphur granules. Staining with PAS, BCG, tissue Gram, and Grocott-Gömöri methenamine-silver nitrate were negative. A CT scan showed distinct cutaneous and subcutaneous inflammatory infiltration without visceral involvement (Fig. 2B). Therapy was started with benzylpenicillin 30 million units/day intravenously for 29 days. As no substantial clinical improvement occurred, intravenous clindamycin was added (1,800 mg/day for 17 days). Treatment was continued with intramuscular depot injections of benzathine penicillin (2.4 million units) once a week. A follow-up 4 months later showed only a slight improvement, with reduction of discharge and foetor, and significantly softer findings on palpation.

Although skin reconstructive surgery was proposed in both cases, the patients were lost to further follow-up.

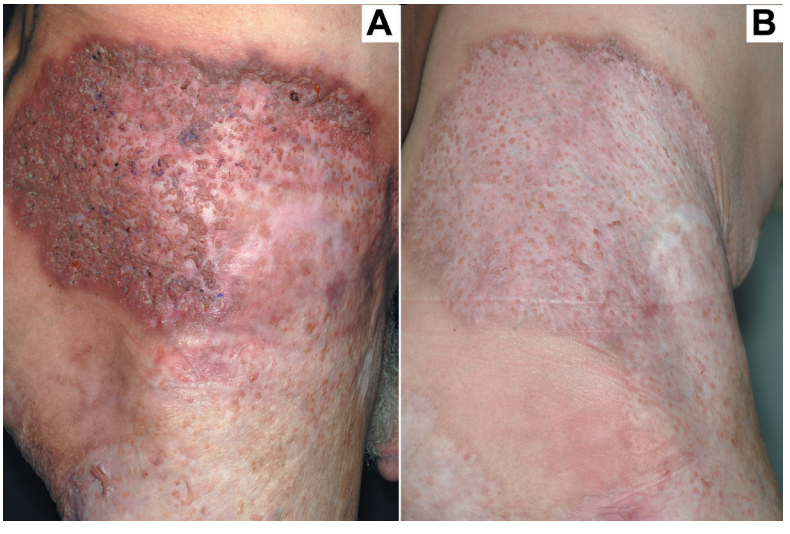

Fig. 1. Case 1: superficial ulcers, erythematous indurated plaques and exudation in the femorogluteal region (A) before and (B) 5 months after combined antibiotic therapy.

DISCUSSION

Both of the cases described above demonstrate that diagnosis and treatment of actinomycosis is challenging. Correct diagnosis results from specific clinical, microbiological, and histopathological findings (2, 4). *In vivo*, some of the Actinomycetes conglomerate into colonies of 1–2 mm in size, so called "sulphur granules" leading to the granular character of the pus. These particles are almost pathognomonic, but their absence does not exclude the diagnosis of actinomycosis (8–10). As many specimens contain only a few granules, their detection may be difficult (8). The histological evidence for a chronic, fistulating, fibrotic and subcutaneous granulation

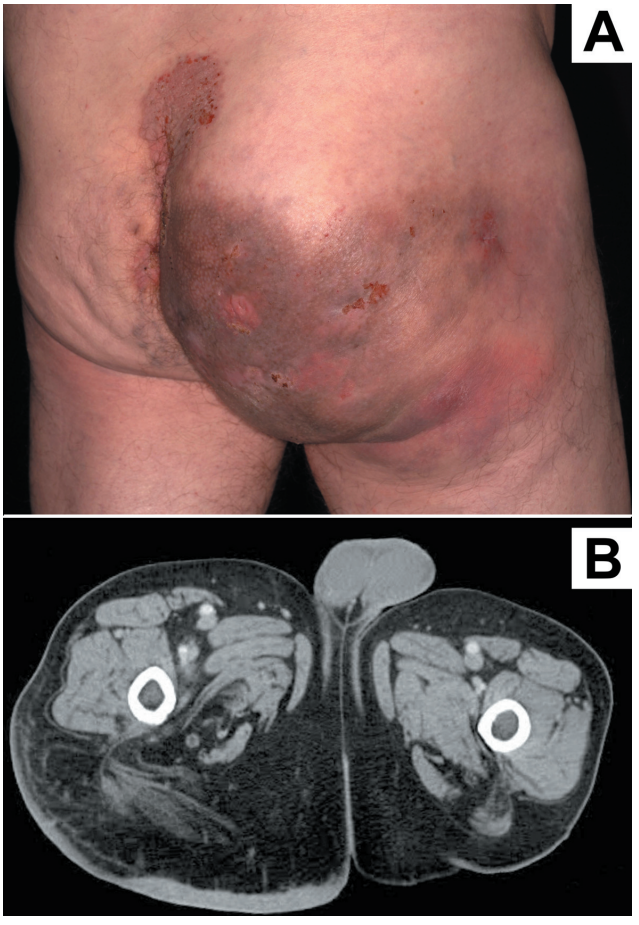

Fig. 2. Case 2: extensive indurated plaques, draining sinuses and ulcers in (A) the right femorogluteal region and (B) computed tomography (CT) scan showing involvement of the subcutis.

tissue supports the diagnosis if microbiological analysis reveals actinomyces. Sulphur granules could not be found in either of our patients, and indeed, they can be absent in actinomycosis caused by all Actinomyces spp., including *A. turicensis* (10). The lipophilic growth behaviour of *A. turicensis* could explain its increased association with primary cutaneous, sebum-rich areas (7, 10). We speculate that, in our two cases, in which the origin of infection remained unknown, exogenous cutaneous trauma might have enabled inoculation of bacteria, as described previously (11).

Actinomycosis is generally a mixed infection. Adequate antibiotic treatment, therefore, must also include the spectrum of the secondary flora (9). Several authors recommend a combination therapy including amino penicillins and beta-lactam inhibitors as first-line treatment (9, 12). Depending on the synergistic accompanying flora, combination with aminoglycosides or lincosamides could be essential because of their good tissue penetration (4, 9). Nevertheless, the two cases described here demonstrate that, despite adequate, high-dosed antibiotic therapy, a chronic refractory course can develop if actinomycosis is diagnosed too late.

The authors declare no conflicts of interest.

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