Cutaneous granular bacteriosis, a rarely diagnosed infection of the head and the neck

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doi:10.1111/j.1365-2230.2008.03036.x

Summary

A young man presented with a large multilobulated and mutilating tumour of the scalp, which had been relapsing for years. Histological examination of a biopsy from the lesion revealed chronic inflammation with granulation tissue and presence of granules with eosinophilic periphery, which was positive for Gram, Grocott and periodic-acid–Schiff stains. A large excision was performed. Cultures grew *Staphylococcus aureus*. The patient was treated with penicillin G, but 4 weeks after the start of treatment, new small nodules appeared over the same area. All these new nodules disappeared within 2 weeks the addition of clindamycin and cotrimoxazole. This triple antibiotic treatment was carried on for 18 months, and the patient remained disease-free after a follow-up of 4 years. Although the lesions were clinically and histologically suggestive of actinomycosis, culture revealed that they were caused by a completely different organism. We suggest grouping such lesions under a single term ''granular bacteriosis'' and combining surgery with broad-spectrum antibiotics covering *Actinomyces* species and botryomycosis-causing organisms (mainly *Staphylococcus*).

Chronic suppurative lesions of the skin showing grains on histopathological examination are a rare disorder characterized by resistance to systemic antibiotics and high recurrence rates. There is confusion regarding the aetiology and terminology, which needs to be clarified.[1,2]

Report

A 17-year-old boy presented to the dermatology department with a large mutilating tumour of the scalp. When he was 12 years of age, a small asymptomatic nodule of 10 mm had developed over the temporal area, which was surgically removed. Two months later, he had the first recurrence, which also removed. At the age of 14 years, the tumour again recurred and grew very rapidly. Surgical excision was performed, followed by a skin graft. At this time, the pathology report noted acute and chronic inflammation with granulation tissue. Postoperative treatment with rifampicin and clarithromycin was started. Six months later, while the patient was still being treated with antibiotics, the tumour recurred for the third time.

On physical examination at our clinic, a large multilobulated tumour was seen, covering the temporal, retroauricular and periauricular areas. It was smooth on palpation, discharging pus and exudates on gentle pressure (Fig. 1).

Samples were taken for both aerobic and anaerobic cultures. Cultures grown from samples of pus were identified as *Pseudomonas aeruginos*a and methicillin-sensitive *Staphylococcus aureus*. Histological examination of a biopsy from the lesion found chronic inflammation with granulation tissue, granules with eosinophilic periphery, and small bunching microorganisms (Fig. 2). The granules were positive for Grocott silver, Gram and periodic-acid–Schiff stains (Fig. 3), but

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Conflict of interest: none declared.

Accepted for publication 1 July 2008

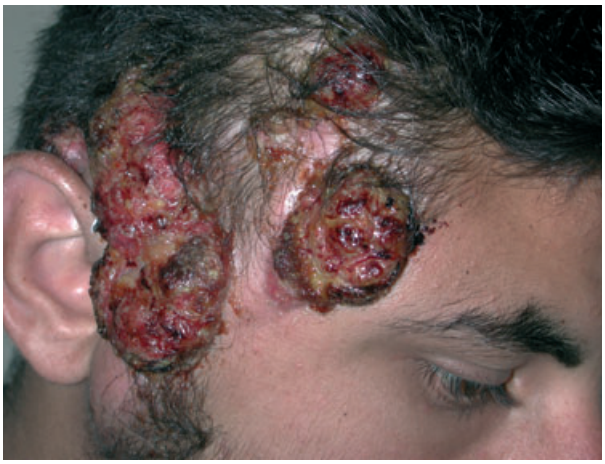

Figure 1 Large multilobulated tumour covering the retroauricular, periauricular and temporal areas.

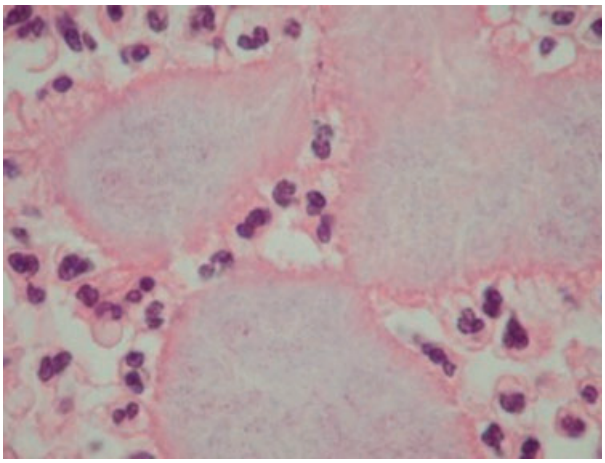

Figure 2 Chronic inflammation with granulation tissue and presence of granules with eosinophilic periphery, and small bunching microorganisms (haematoxylin and eosin; original magnification × 400).

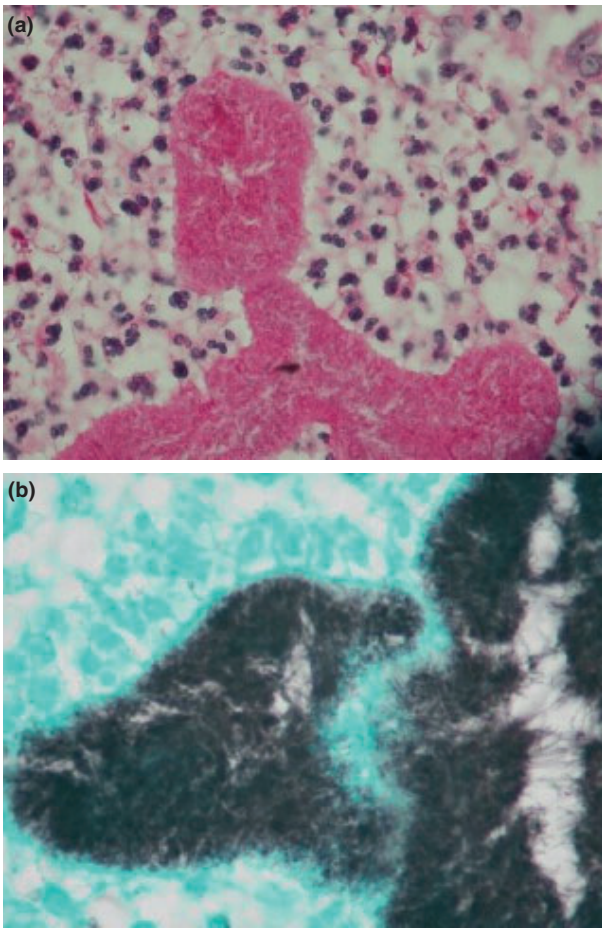

Figure 3 Granules were positive for (a) periodic acid–Schiff (original magnification × 400) and (b) Grocott silver (original magnification × 400).

negative for Ziehl–Neelsen stain. These pathological findings were consistent with actinomycosis. Results of serological testing for human immunodeficiency virus and a Mantoux test for tuberculosis were negative. A cerebral computed tomography scan showed an extension to the superficial parotid gland.

A large excision with superficial parotidectomy was performed, and completed with a skin graft. The pathology report confirmed actinomycosis. Bacteriological cultures grew *S. aureus*.

Intravenous penicillin G 24 MU/day was started. Four weeks later, new small nodules resembling the initial lesion appeared over the graft and on the resection margins. Histopathological examination showed the same features as before, and repeated cultures for fungus, aerobes and anaerobes were negative. Clindamycin 600 mg/8 h and trimethoprim–sulfamethoxazole (800 and 160 mg, respectively, every 8 h) were added to the previous treatment. All the new nodules disappeared after only 2 weeks, but the triple antibiotic treatment was carried on for 18 months, and the patient remained disease-free after a follow-up of 4 years.

Chronic and recurrent infections of the skin showing grains on histopathology have conflicting terminology in the medical literature. The word 'mycetoma' has been used to designate such lesions, with the most widely used classification dividing mycetomas into eumycetomas caused by true fungi, and actinomycetomas produced by actininomycetes, mainly of the genera Nocardia, Streptomyces and Actinomadura.[3]

This terminology cannot apply to other infections with similar clinical findings, such as actinomycosis and botryomycosis.

Actinomycosis is caused by *Actinomyces israelii* or other species of *Actinomyces* or related bacteria, most commonly affecting the cervicofacial site. The branching bacteria causing actinomycosis are nonacid-fast anaerobic or microaerophilic bacteria, whereas the agents of actinomycetoma are always aerobic and sometimes weakly acid-fast. Culture is negative in >50% of cases.[4–6]

Botriomycosis is a very complex entity with multiple causative organisms. *S. aureus* was the first to be isolated,[7] followed by *Escherichia coli*, *Bacteroides* spp., *Proteus* spp., *Propionibacterium acnes*, *Corynebacterium* spp., and *Neisseria* spp., most of them being low virulence strains.[8,9] *S. aureus* is still the major causal agent (40% of cases) followed by *P. aeruginosa* (20%).[1] The inflammatory reaction is characterized by the presence of granules with (as in actinomycotic mycetomas) an eosinophilic periphery corresponding to the immune response of the host.[10] However, unlike granules of actinomycosis, botryomycosis granules do not stain with Grocott–Gomori silver stain.[1]

This case was clinically suggestive of 'mycetoma', more accurately of 'granular bacteriosis', and had recurred after three previous surgical interventions. Once a fungal aetiology was excluded, the differential diagnosis included actinomycosis and botryomycosis. Although we had one positive culture for *Pseudomonas* (probably a contamination) and repeated positive cultures for *S. aureus*, we decided to treat it as if it was actinomycosis rather than botryomycosis for two main reasons. Firstly, the granules on pathology were very suggestive of actinomycosis, showing small surrounding filaments, and were positive for both Grocott silver and Gram stains. Secondly, the patient had relapsed in spite of long-term antibiotic treatment (rifampicin and clarithromycin) known to be effective against *S. aureus*. After another clinical and histopathological relapse, we added clindamycin and trimethoprim–sulfamethoxazole in order to cover actinomycosis, nocardiosis, and *S. aureus* infection, after which complete clearance occurred rapidly, and the patient remained disease-free after 4 years of follow-up.

This case raises an important issue: knowing that *Actinomyces* cultures are very difficult to obtain, how should we treat lesions that are clinically and histologically suggestive of actinomycosis, but which have negative cultures or even cultures that are positive for totally different bacteria? Should they be treated as actinomycosis, botryomycosis or both? We believe it is preferable to bring these lesions together under a single term 'granular bacteriosis'. Hence, it would be defensible to combine surgery with broad-spectrum antibiotics covering *Actinomyces species as* well as botryomycosis-causing organisms (mainly *S. aureus*). The difficulty of reaching a precise bacteriological diagnosis is partly the cause of the confusion that exists in the literature, but clear definitions should be considered of lesser importance if treatment is successful.

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