Case Report

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Chronic odontogenic osteomyelitis and facial actinomycosis of six-month duration

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Introduction: An uncommon association of chronic odontogenic osteomyelitis and an abscess in the right mandibular region with facial actinomycosis of 6 months duration after several short-term therapy failures is described.

Case presentation: The patient was a 55-year-old man with chronic inflammation of the right parotid area of the face. *Actinomyces israelii*, *Fusobacterium nucleatum* and *Parvimonas micra* were isolated from specimens of the subtemporal abscess. Therapy started as clarithromycin and was followed by amoxicillin/clavulanate because of the resistance patterns of co-infecting *F. nucleatum*.

Conclusion: In this case of facial actinomycosis, a successful therapeutic outcome involved a prolonged therapy as well as the detection of drug-resistant co-existing anaerobic bacteria.

Keywords: *Actinomyces*; actinomycosis; *Fusobacterium*; osteomyelitis; susceptibility.

Introduction

Actinomycosis is a rare chronic infection. If untreated, the disease can lead to a high (>60 %) mortality rate (Falchero *et al.*, 1997). Bacteriological diagnosis is difficult because slow-growing *Actinomyces israelii* is masked by faster-growing co-infecting bacteria in clinical specimens and the characteristic sulfur granules may be single/absent in parts of specimens (Wong *et al.*, 2011). This study revealed a case of odontogenic osteomyelitis of the right mandibular region in association with facial actinomycosis involving *A. israelii* and two anaerobic species.

Case report

A 55-year-old man was admitted to the Maxillo-Facial Surgery Hospital and hospitalized for 11 days. The onset of symptoms had started 2 months previously, 2 days after extraction of tooth 47 (right mandibular second molar) because of an exacerbated periodontal abscess. For this reason, the patient had taken consecutively doxycycline, tetracycline/oleandomycin, ampicillin and ciprofloxacin, each for 5–10 days, without clinical improvement. At the time of admission, purulent infection in the right mandibular region and an abscess in the right subtemporal region were observed. The patient had enhanced dental calculus. Orthopantomography showed osteolysis (10 × 10 mm) near teeth 47 and 48 with two sequestrums. Histological examination revealed chronic fibrosing osteomyelitis with sequestrums. The sedimentation rate was at

28 mm h⁻¹. Incisions and open drainage of the affected tissues and focus of osteomyelitis were performed. The patient was treated with lincomycin (600 mg every 8 h) for 8 days and, after clinical improvement, he was discharged.

Two months later, because of a relapse, specimens from the parotid space were taken for anaerobic culture (Fig. 1). The skin surface was cleaned with iodophor for 1 min and incisions were made. Curettings of the infected tissue were placed into Stuart transport medium (BBL) and processed within 2 h. The specimens were inoculated onto Schaedler blood agar (BBL) with vitamin K/haemin, and incubated

Fig. 1. Clinical observation of the patient with facial actinomycosis after incisions.

anaerobically at 37 °C for 14 days. Isolates were identified according to Wadsworth Anaerobic Bacteriology Manual (Jousimies-Somer *et al.*, 2002), as well as by API Rapid ID 32A (bioMérieux). Susceptibility to penicillin, clarithromycin, metronidazole, amoxicillin/clavulanate and ampicillin/sulbactam was determined by an agar dilution method (Jousimies-Somer *et al.*, 2002).

Single sulfur granules were seen in direct Gram-stained smears. Both specimens grew *A. israelii*, *Fusobacterium nucleatum* and *Parvimonas micra*. *A. israelii* grew after 11 days incubation. An 8-day treatment with oral clarithromycin (500 mg every 12 h) was started together with surgical incisions of the affected tissues. The third specimen, taken 12 days after the first one, showed no bacterial growth. However, smears revealed single, Gram-positive filamentous *Actinomyces*-like rods, and thus the treatment was prolonged by oral amoxicillin/clavulanate (625 mg every 12 h) for an additional 2 weeks. Gradual clinical improvement and healing were achieved. Nine months and 3 years later, the follow-up examinations confirmed that the patient was fully recovered.

Discussion

The male sex, age of the patient, tooth extraction and severe dental calculus were predisposing factors for actinomycosis (Wong *et al.*, 2011; Badre *et al.*, 2013). The *P. micra* strain was susceptible to all antibacterial agents. The penicillin resistance of *F. nucleatum* (MIC 2 mg l⁻¹) according to European Committee on Antimicrobial Susceptibility Testing (EUCAST) breakpoints and the elevated clarithromycin MICs against *F. nucleatum* (MIC 2 mg l⁻¹ against the first isolate and 4 mg l⁻¹ against the second isolate) and *A. israelii* (MIC 2 mg l⁻¹) could be due to the previous antibiotic treatments. As the treatment should initially cover both *Actinomyces* spp. and the other co-infecting bacteria at the site of infection (Reichenbach *et al.*, 2009), the therapy started with clarithromycin because of penicillin resistance of the co-existing anaerobic bacteria. Because of possible side effects of a prolonged clindamycin therapy, macrolides are an alternative to penicillin for treating actinomycosis (Badre *et al.*, 2013). β -Lactamase-positive *F. nucleatum* isolates have been reported (Jousimies-Somer *et al.*, 2002; Mosca *et al.*, 2007). *F. nucleatum* resistance to penicillin and clarithromycin (2 mg l⁻¹) motivated a treatment prolongation with amoxicillin/clavulanate to prevent a relapse of

infection. All isolates were susceptible to amoxicillin/clavulanate and ampicillin/sulbactam.

Actinomycosis requires longer therapy compared with that for most other infections because of the species-specific antibiotic tolerance of *A. israelii*, and the induration and avascularity of infected areas (Barnard *et al.*, 1996; Reichenbach *et al.*, 2009). In some cases of orofacial actinomycosis, shorter (2–6 weeks instead of the classical 6–12 months) therapy combined with surgical drainage has been reported to be successful (Wong *et al.*, 2011). In the present case, the treatment lasted over 3 weeks and was curative.

In conclusion, actinomycosis is a rare but serious chronic infection. Clinical and bacteriological diagnosis and therapy of actinomycosis are specific and difficult. The patient was treated inappropriately with successive courses of antimicrobial agents, including ciprofloxacin, which has no activity against most anaerobes. The clinical progress of the disease strongly emphasizes the need for early surgery, earlier anaerobic cultures and more prolonged antimicrobial therapy for severe odontogenic infections. In this case of facial actinomycosis, the successful therapeutic outcome required a prolonged therapy as well as the detection of drug-resistant co-existing anaerobic bacteria.

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