

Cervicofacial actinomycosis - report of two cases

V. Bochev, I. Angelova and N. Tsankov

S U M M A R Y

Human actinomycosis is a bacterial infection caused primarily by *Actinomyces israelii*, a resident of the oral cavity. The cervicofacial region is most commonly affected; thoracic and abdominal forms of the disease have also been distinguished. Actinomycosis is sometimes difficult to diagnose and it should be borne in mind in the differential diagnosis of numerous infectious and non-infectious diseases. We describe two patients with cervicofacial actinomycosis which on initial examination were diagnosed as "submandibular phlegmona".

K E Y W O R D S

**actinomyco-
sis, cervico-
facial, sub-
mandibular
phlegmona,
differential
diagnosis**

Introduction

Actinomycosis is an infectious disease with a worldwide distribution caused by anaerobic and microaerophilic bacteria residing in the oral cavity. The disease was known in cattle as early as the beginning of the 19th century and even though similar conditions were observed in humans these were not recognized as actinomycosis (1). Actinomycosis is often difficult to diagnose as it can mimic numerous infectious and non-infectious diseases. It is nowadays uncommon in Europe and the possibility that we may face a patient with actinomycosis is therefore underestimated. Recently we observed two cases of cervicofacial actinomycosis diagnosed initially as phlegmona.

Case report 1

A 28-year-old woman complaining of painful swelling in the left submandibular region came for a consultation to the Department of maxillo-facial surgery in October 1999. The swelling had appeared 10 days prior to the admission and Tetracycline 4 x 250 mg/d had been administered with unsatisfactory therapeutic result. As the symptoms persisted, the patient was admitted to the department because of a purulent infection of the left submandibular region.

The examination upon the admission revealed a soft tissue swelling in the left submandibular region. The overlying skin was reddened, edematous and tender on palpation. Figure 1. Masticatory spasm (degree II)

was present.

The X-ray examination revealed a semi-retained inferior left wisdom-tooth. An intraoral surgical incision of the left temporomandibular fold was performed, the discharge was purulent with several sulfur granules. The biopsy specimen of the same area was consistent with actinomycosis. Figure 2.

The semi-retained tooth was extracted. The patient was treated with Penicillin 4 x 2 000 000 E for 30 days with good result.

Case report 2

A 28-year-old man was seen at the Department of maxillo-facial surgery because of a progressively growing swelling in the left submandibular area.

Physical examination revealed an indurated painful nodule on the lateral aspect of the left cheek. The opening of the mouth was considerably restricted. The patient was admitted to the department with the clinical diagnosis of a phlegmona of the left submandibular region.

The X-ray evaluation revealed the presence of bilateral inferior semi-retained molars. Figure 3.

Surgical incisions in the left submandibular region were performed with "granules" present in the purulent discharge. The biopsy specimen revealed the presence of *Actinomyces israelii*. The patient was successfully treated with Penicillin 4 x 2 000000 E for 30 days.

Discussion

Human actinomycosis is caused primarily by *Actinomyces israelii*, an anaerobic gram-positive, branched, filamentous bacterium, residing as commensal in periodontal pockets and gingival crevices, in carious teeth, dental plaques, tonsillar crypts or in periodontium. It is the most significant, but not the sole causative agent of the disease. The penetration is facilitated by dental extraction and trauma. The pathogens may enter the respiratory tract by inhalation or the intestines by ingestion. Spread by contiguity is another pattern of propagation.

Numerous clinical manifestations of the disease have been described. The cervicofacial region, the chest and the abdomen are most commonly affected, other possible sites of infection are the extremities, lacrimal glands, kidneys, genital organs, bones and the central nervous system. Actinomycosis is mostly found in young adults, women are less frequently affected than men.

The disease has not been subjected to systematic epidemiological studies, the reported incidence of the different forms varies significantly. Cervicofacial actinomycosis accounts for approximately 50% to 60% of



Figure 1. Case 1. Reddened and swollen skin with a small erosion in the center, left submandibular region.

all cases (3), thoracic between 13% and 15% (4,5), while the incidence of the abdominal form is estimated to be between 2.8% (6) and 63% (5).

The cervicofacial variant is characterized by the appearance of solid sub- or supramandibular nodules or swellings, the overlying skin becoming purple to violet

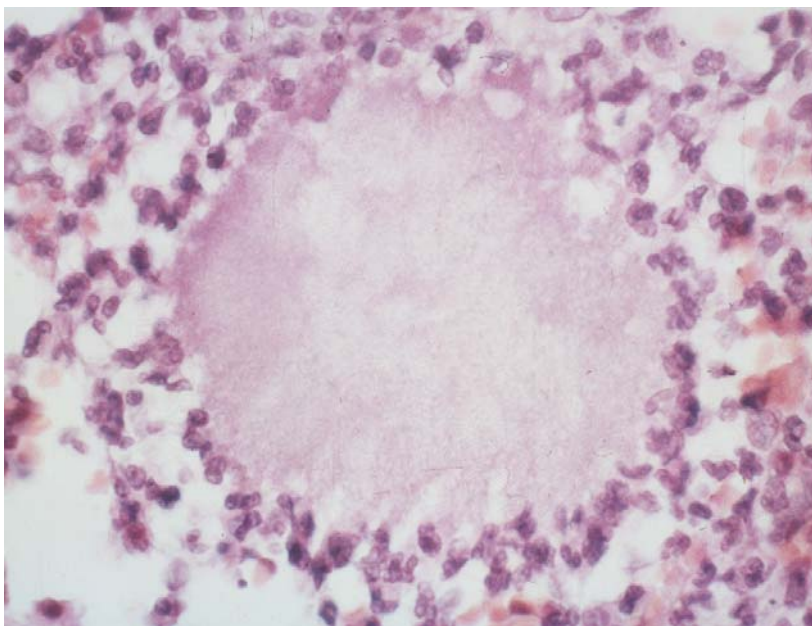


Figure 2. Case 1. A sulfur granule, histopathology: a homogenous center and clubs at the periphery

and warm. A retrospective study of 317 patients treated at the Department of dental surgery of the University of Cologne has shown that the mandibular region was the primary site of cervicofacial actinomycosis in 53.6%, the cheek in 16.4%, the chin in 13.3%, the submaxillary ramus and angle in 10.7%, the upper jaw in 5.7% and the mandibular joint in 0.3%. Periostitis and posttraumatic osteomyelitis were present in 11.7% (7). The onset was unnoticed rather than acute, and careful history taking often revealed preceding dental extractions and other therapeutic procedures thus suggesting the origin of the infection. Subsequently fistulae or ulcerations arise with a seropurulent discharge containing minute yellowish granules. Masticatory spasm may be present. In contrast, the general state of health is seldom affected.

The diagnosis of the cervicofacial form on clinical examination alone is difficult at the onset, unless the physician is aware of it, but becomes easy once the granules are discharged. However, a number of inflamma-



Figure 3. Case 2. Semi retained lower molars bilaterally

tory, infectious and noninfectious diseases should be considered in differential diagnosis. Abscesses and fistulae due to pyogenic bacteria are usually sudden in onset and accompanied by fever, lymphadenopathy, alteration in the general health and leucocytosis.

Subcutaneous tuberculosis with fistulae or scrofuloderma are believed to be rare nowadays, as is tertiary syphilis. Benign or malignant tumors of the cervicofacial region are less likely to be confused with actinomycosis. As a rule, we suggest that any soft tissue swelling on the face and neck should be investigated for actinomycosis.

Abdominal actinomycosis affects predominantly the ileocecal region, but any segment of the intestines can be the primary site of the disease. The symptoms are not disease-specific: a colicky abdominal pain with irradiation to the anus, urinary bladder and to the thigh. Physi-

cal examination reveals a poorly demarcated indurated mass in the abdomen that may later propagate to the retroperitoneal space or drain through the abdominal skin forming numerous fistulae.

The presenting symptoms of pulmonary actinomycosis are also non-specific: fever, cough, thoracic pain, dyspnea and as a consequence - general impairment of health. The sputum is mucopurulent or even sanguineous. With the appearance of fistulae, the disease has spread to the mediastinum, the pericardium, and finally to the skin of the chest. X-ray examination is mandatory in such cases to estimate the degree of organ involvement.

Unusual presentations of the disease include osteitis of the mandible (8), maxillotemporal osteoarthritis (9), renal actinomycosis (10), actinomycosis of the central nervous system (11,12), and perianal actinomycosis (13).

The biopsy specimen of an actinomycetic infection shows a central neutrophilic lobulated abscess that contains a number of granules surrounded by granulation tissue. Actinomycetic sulfur granules appear in HE stained sections as basophilic structures with elongated eosinophilic clubs that radiate from the periphery (14). The granules contain gram-positive, non-acid fast bacteria initially misclassified as fungi. Actinomycosis can be differentiated from nocardiosis in which granules consist of acid fast branched bacilli, and botryomycosis containing non-filamentous cocci (bacilli) respectively (15).

Granules may be scarce or even absent (16), that is why culture and subsequent identification are necessary for the diagnosis. However *Actinomyces* are difficult to grow even in enriched media and the diagnosis is confirmed by culture in less than 50% (17). In such cases the diagnosis is based on the morphology of granules and bacteria or on direct examination of granules.

Penicillin in high doses and under hospital surveillance is the treatment of choice. It should be given on long-term basis (30 days on the average). If penicillin can not be used, erythromycin 2.0 gms/24 hours, tetracycline 2.0-3.0 gms/24 hours or lincomycin 2.0 gms/24 hours should be administered (18). Surgical intervention is a valuable treatment modality, especially in cases of thoracic and abdominal involvement. Complete recovery is expected in 90% of patients with cervicofacial actinomycosis (19).

Conclusion

At present actinomycosis is estimated a rare disease in Europe. Difficulties arise from the fact that it can mimic numerous common conditions in human pathology. Therefore we consider that any soft tissue tumoral formation on the face and neck should be investigated for actinomycosis.

REFERENCES

1. Grigoriu D, Delacrétaz J, Borelli D. Actinomycosis: Medical mycology, Editiones Roche, Basle, Switzerland, 1987, 425-32
2. Cirillo-Hyland V, Herzberg A, Jaworsky C. Cervicofacial actinomycosis resembling a ruptured cyst. *J Am Acad Dermatol*, 29, 1993, 308-11
3. Pordy RC. Lumpy jaw due to *Actinomyces meyerii*: Report of the first case and review of the literature. *Mt Sinai J Med* 1988;55:190-3
4. Cope VZ.:Actinomycosis. University Press, London, Oxford 1938.
5. Mousseau A, Mousseau-Brodu CM.: L'actinomycose abdominale, *J Chir*, Paris 1973;106: 565.
6. Almeida F, 1939, quoted in: Negroni P: Micosis profundas. Vol I: Los micetomas. El Ateneo, Buenos Aires, 1954.
7. Schaaal KP, Beaman BL. Clinical significance of actinomycetes. In: Goodfellow M, Mordarski M, Williams ST, eds. The biology of actinomycetes. New York: Academic Press, 1983:389-424.
8. Sartory A, Roger R, Meyer J. :Deux cas d'actinomycose inactivee sans grains microscopiques. *Bull. Soc Méd CVII* 16, 1932 and 3rd suppl. 125, 1933.
9. Grigoriu D, Maillard FG, Goumaz FC: L'actinomycose. *Rev Méd Sues Romande* 1969; 90: 325-36.
10. Rosenblum PS. Renal actinomycosis. A case report. *Urol and Cutan Rev* 1949; 53: 329
11. Intile JA, Richest JH. Cervicofacial actinomycosis complicated by meningitis. *J Am Med Ass* 1962; 181: 724-6.
12. Martin WJ, Nichols DR, Wellman et al. Disseminated actinomycosis treated with tetracycline. *Arch Intern Med* 1956; 97: 252
13. Gayraud A, Grosieux-Dauger C, Durlach A. et al. Actinomycose cutanéé périanale et fessiere, *Ann Dermatol Venereol* 2000; 127: 393-6
14. Burden P. Actinomycosis. *J Infect* 1989; 19: 95-9.
15. Wee SH, Chang SN, Shim JY et al. A case of primary cutaneous actinomycosis. *J Dermatol (Tokyo)*, 2000;27:651-654
16. Brown JR. Human actinomycosis, a study of 181 subjects. *Hum Pathol* 1973; 4: 319-330.
17. Ashton N, Cook C. Allergic granulomatous nodules of the eyelid and conjunctiva. *Am J Ophthalmol* 1979; 87: 1-28
18. Lerner PI. The lumpy jaw, cervicofacial actinomycosis. *Infect Dis Clin North Am* 1988; 2: 203-19
19. Katz BJ, Kalter DC, Bruce S. Subcutaneous nodules in a man diagnosed as having tuberculosis. *Arch Dermatol* 1988; 124: 121-2, 124-5.

A U T H O R S '
A D D R E S S E S

Valo Bochev MD, Department of Maxillo-facial Surgery, University of Medicine, Sofia, Bulgaria, St. Georgi Sofiiski Blvd., 1431 Sofia
Irena Angelova MD, Department of Dermatology, University of Medicine, Sofia, Bulgaria, St. Georgi Sofiiski Blvd., 1431 Sofia
Nikolai Tsankov MD, PhD, same address
e-mail: elfyda@mail.bg