

Actinomycosis of submandibular gland: an unusual presentation

Actinomicosi cervico facciale: una presentazione insolita

L. BUBBICO, M. CARATOZZOLO¹, F. NARDI², G. RUOPPOLO³, A. GRECO³, M. VENDITTI⁴
ENT Specialist, Department of Biomedical Sciences, Italian Institute of Social Medicine, Rome; ¹ 4th Department of Surgery, University "La Sapienza", Rome; ² Cytopathology Unit, University "La Sapienza", Rome; ³ 2nd ENT Department, University "La Sapienza", Rome; ⁴ Department of Infectious Diseases, University "La Sapienza", Rome, Italy

Key words

Salivary glands diseases • Actinomycosis • Case report

Parole chiave

Malattie delle ghiandole salivari • Actinomicosi • Caso clinico

Summary

An unusual presentation of oro-facial actinomycosis, mimicking the clinical appearance of a malignant lesion is reported. The patient, a 74-year-old female, presented with a right submandibular mass, which slowly grew in size over a period of about 2 months, and a modest dysphagia. A painless cervical mass was palpable over the submandibular region. The rhinopharyngo-laryngeal region, explored by flexible fiberoptic examination, was normal. At ultrasonography, a 2x2 cm infiltrating dyshomogeneous mass, involving the right submandibular gland, was visible. No connection with adjacent organs was found. There was no associated cervical lymphadenopathy. Ultrasound-guided fine-needle aspiration cytology, performed on lesion, revealed no evidence of malignancy. The presence of characteristic colonies of actinomyces infection was found. The patient was treated initially with tetracycline chloridrate 100 mg: 1 tablet every 12 hours for 7 weeks, but a repeat ultrasonography showed no resolution. A further fine-needle aspiration cytology showed no actinomyces infection in the specimen. The patient was treated with methylprednisolone, 20 mg every 24 hours, for 5 days. After steroid treatment, the patient has been well and, upon repeat ultrasonography, total resolution of the submandibular lesion was confirmed. In conclusion, the clinical presentation of cervicofacial actinomycosis is variable and may mimic a malignant lesion or chronic granulomatous infections. Diagnostic and therapeutic findings are discussed.

Riassunto

Segnaliamo il caso di una donna di 74 anni, che si è presentata alla nostra osservazione con una massa sottomandibolare destra, non dolente, accompagnata da una modesta disfagia. L'esame rino-faringo-laringoscopico eseguito mediante fibroscopio flessibile, non ha rivelato lesioni nella regione esaminata. All'esame ultrasonografico era visibile una massa disomogenea di 2 cm di diametro che coinvolgeva la ghiandola salivare sottomandibolare di destra. Nessun segno di coinvolgimento degli organi adiacenti è stato trovato. Non erano presenti linfadenopatie latero-cervicali. L'esame citologico, eseguito mediante ago-aspirazione, non ha rivelato segni di malignità, ma la presenza delle caratteristiche colonie di actinomyces. La paziente è stata trattata inizialmente con tetraciclina cloridrato 100 mg ogni 12 ore per 7 settimane, ma la ripetizione dell'esame ultrasonografico non ha mostrato nessun miglioramento. L'esame citologico di controllo dimostrava l'assenza di infezione da actinomyces. La paziente è stata trattata con prednisolone 20 mg/die per 5 giorni. Dopo il trattamento la paziente ha ottenuto la risoluzione totale dell'infezione sottomandibolare, confermata dall'esame ultrasonografico di controllo. A 2 anni di distanza non si sono riscontrate recidive. L'esordio clinico dell'actinomicosi cervico facciale è subdolo e può simulare lesioni maligne o infezioni granulomatose croniche. Vengono discussi l'approccio diagnostico e la condotta terapeutica.

Introduction

Actinomycosis infection is a sporadic chronic infection caused by anaerobic Gram positive bacteria with filamentous appearance, erroneously classified in the past as part of the mycetes families. The infection is rare and usually presents in 3 distinct localizations: cervico-facial, pulmonary, abdomino-pelvic; it may also involve the female genital organs, in the event of intra-uterine device use^{1,2}.

The cervico-facial form is the most common, involving between 50-70% of the cases reported in the liter-

ature. The incidence of the disease is not completely known: during the Seventies, in the Cleveland, United States area, an incidence/year of 1 case in 300,000 was found³. There is a slight male prevalence in young adults 3:1^{2,4}. There is no racial predisposition or geographic factors. Of the various actinomyces species causing actinomycosis infection in the human (*A. israeli*, *A. bovis*, *A. naeslundii*, *A. viscosus*, *A. odontolyticus*), *A. israeli* is that most frequently isolated, it takes the name from the first discoverer of the disease (J. Israeli in 1878)⁵. Actinomyces is a commensal and normal inhabitant of the human oral cavi-

ty and gastro-enteric tract. The infection in humans occurs in the presence of dental manipulation, dental extractions, poor oral hygiene or maxillo-facial trauma. A tear in the oral mucosa is the port of entry of infection.

Actinomycosis is an endogenous infection, there is no person to person transmission.

The disease is characterized by an abscess-like formation surrounded by a granulomatous inflammatory reaction.

In the cervico-facial presentation, the infection develops silently, in a subacute form, as a solid mass, slowly increasing in size, without pain. It rarely involves the regional lymph nodes.

Case report

In March 2001, a 74-year-old female presented with a right, painless, submandibular mass, which slowly increased in size over a period of about 2 months, with a modest dysphagia. A neck mass, approximately 2x2 cm, located in the submandibular region, was palpable. The cervical lymph nodes were not palpable. Her social history was significant for cigarette smoking (20 cigarettes/day for approximately 50 years).

At ultrasonography (US), a 2x2 cm infiltrating dyshomogeneous mass involving the right submandibular gland was detected. There was no associated cervical lymphadenopathy. Figure 1 shows the US features.

The rhino-pharyngo-laryngeal region, explored by flexible fiberoptic examination, was normal.

Paranasal sinuses, thorax and oesophagus, studied by standard radiology, were normal.

The cytological study performed on specimens collected from the lesion, by US-guided fine-needle aspiration, showed the presence of micro-organisms with a filamentous appearance, PAS positive, compatible morphologically with actinomyces colonies. A diagnosis of actinomycosis of the right submandibular gland was made.

The patient, in consideration of the advanced age, has been treated with minocycline chloridrate (Minocin 100), 1 tablet every 12 hours, periodically monitoring hepatic and renal parameters.

After 50 days of antibiotic treatment, US examination showed no decrease in the size of the mass.

Seven days after suspension of the treatment, a second cytological study of the lesion showed no actinomycosis infection in the specimen.

Since the clinical features remained unchanged, the patient was treated with a cycle of methylprednisolone (Urbason 20 mg/day for 5 days). On 2nd day of treatment, the cervical mass gradually decreased, until total resolution occurred on the 5th day, confirmed by US findings.

At 2-year follow-up, no recurrence was found.



Fig. 1. Ultrasonographic features.

Discussion

Submandibular salivary gland actinomycosis, in a cervicofacial localization, is an extremely rare, but not exceptional, event.

Early diagnosis of actinomycosis is rare since it is usually difficult to diagnose on clinical features alone.

In the presence of a neck mass, imaging studies are usually carried out as a first choice diagnostic technique.

Several imaging modalities are used in making the differential diagnosis in a cervico-facial mass, e.g. computed tomography (CT) scan, nuclear magnetic resonance (NMR), US, which, albeit, are generally non specific. These techniques provide only quantitative information (limits and borders of the lesion, homogeneity and density of the content, localization, invasion of surrounding organs, etc.).

Frequently, definitive diagnosis is only made with a cytological study and is based on the identification and/or isolation of the organism, and presence of sulphur granules. According to Weese et al.⁴, the characteristic sulphur granules in the specimen are present in only about 35-55% of cases. The diagnosis, in this case, appears definitive⁶⁻¹⁷.

Isolation of the organism and its identification, instead, takes from 2 to 3 weeks. Cultures should be placed immediately in anaerobic conditions and incubated for approximately 48 hours.

When the cervico-facial infection is circumscribed, of small size and is not associated with draining fistulas, the medical approach is preferred to the surgical approach.

Actinomyces are usually susceptible to several antibiotics: penicillins (Penicillin G), chloramphenicol, tetracycline, erythromycin, clindamycin, imipenem, streptomycin and cephalosporins.

The current recommended therapy for all clinical forms of actinomycosis infection, is high dose penicillin over a prolonged period (benzatin-penicillin G) at a

dosage of 10-20 mil. U/day iv for 4-6 weeks, continuing with oral penicillin treatment, even after total resolution of symptoms.

In our case, we preferred to use the minocycline chlorhydrate (Minocin 100) ⁶, for oral administration, since it is better tolerated and more manageable (home use). In our case, 7 weeks of antibiotic treatment with tetracycline were necessary, to remove actinomycetes infection from the tissue.

The steroid used has been effective in removing the residual inflammatory granulomatous reaction and in hastening recovery. In actinomycosis infection, the swelling formation is characterized by a mixed suppurative and granulomatous inflammatory reaction, surrounded by polymorphonuclear neutrophils. The granulomatous reaction remains in the organ long after disappearance of the pathogenic agent from the tissue, decreasing slowly.

It is a good rule, when the lesion fails to decrease in size after appropriate antibiotic therapy, to repeat the cytological examination, before programming other treatment procedures.

The clinical appearance of cervico-facial actinomycosis is variable and may mimic malignant lesions or other chronic diseases, such as chronic granulomatous infections, and inflammatory disease.

Conclusions

Although cervico-facial actinomycosis occurs infrequently, it should be included in the differential diagnosis of: neoplasm (non-Hodgkin lymphomas), of chronic and granulomatous suppurative lesions of the cervical region (nocardiosis, tuberculosis, etc.).

Actinomycetes is a potential microbial contaminant of head and neck surgery and may complicate a major surgical oncologic head and neck procedure ⁷.

When the actinomycosis infection is circumscribed, is diagnosed early and is not associated with draining fistulas, medical treatment is preferred to surgery. The surgical approach is indicated for excision or drainage of empyemas, or when the outcome of medical treatment is not satisfactory.

Use of tetracycline, for easy management (home setting), is advised in patients allergic to penicillin and in the elderly.

Use of a steroid as adjunctive therapy, after disappearance of the pathogenic agent, has been shown to be effective to speeding up recovery.

The prognosis of actinomycosis of the submandibular gland, when circumscribed, without drainage fistulas and diagnosed early, is usually excellent.

References

- Vera Alvarez J, Marigil-Gomez M, Abscal-Agorreta M. *Fine needle aspiration cytology of cervicofacial actinomycosis*. Acta Cytol 1993;37:109-11.
- Schaal KP, Lee HJ. *Actinomycete infections in humans – a review*. Gene 1992;115:201-11.
- Das DK, Gulati A, Bhatt NC, Mandal AK, Khan VA, Bhambhani S. *Fine needle aspiration cytology of oral and pharyngeal lesions. A study of 45 cases*. Acta Cytol 1993;37:333-42.
- Weese WC, Smith IM. *A study of 57 cases of actinomycosis over a 36- year period*. Arch Intern Med 1975;135:1562-8.
- Sa'do B, Yoshiura K, Yuasa K, Arijji Y, Kanda S, Oka M, Katsuki T. *Multimodality imaging of cervicofacial actinomycosis*. Surg Oral Med Oral Pathol 1993;76:772-82.
- Stenhouse D, Mc Donald DG, Mc Farlane TW. *Cervico-facial and intra-oral actinomycosis; a 5-year retrospective study*. Br J Oral Surg 1975;13:172-82.
- Bennhoff DF. *Actinomycosis: diagnostic and therapeutic considerations and a review of 32 cases*. Laryngoscope 1984;94:1198-217.
- Israeli J. *Neue Beobachtungen auf dem Gebiete der Mykosen des Menschen*. Virchows Archiv Pathol Anat 1878;74:15-53
- Smego RA, Foglia G. *Actinomycosis. State of the art clinical article*. Clin Infect Dis 1998;26:1255-61.
- Sah SP, Mishra A, Rani S, Ramachandran VG. *Cervicofacial actinomycosis: diagnosis by fine needle aspiration cytology*. Acta Cytol 2001;45:665-7.
- Zitsch 3rd RP, Bothwell M. *Actinomycosis: a potential complication of head and neck surgery*. Am J Otolaryngol 1999;20:260-2.
- Fradis M, Zisman D, Podoshin L, Wellisch G. *Actinomycosis of the face and neck*. Arch Otolaryngol 1976;102:87-9.
- Hong IS, Mezgebe HM, Gaiter TE, Lofton J. *Actinomycosis of the neck: diagnosis by fine needle aspiration biopsy*. J Natl Med Assoc 1993;85:145-6.
- Kwartler JA, Limaye A. *Cervicofacial actinomycosis: pathologic quiz case*. Arch Otolaryngol Head Neck Surg 1989;115:524-6.
- Smego RA. *Actinomycosis of the central nervous system*. Rev Infect Dis 1987;9:855-65.
- Rice DH. *Chronic inflammatory disorders of the salivary glands*. Otolaryngol Clin North Am 1999;32:813-8.
- Guiral H, Risco J, Mayayo E. *Submaxillary sialolithiasis due to Actinomyces israelii. A case report*. An Otorrinolaringol Ibero Am. 1998;25:247-53.

■ Received June 6, 2003.
Accepted July 7, 2003.

■ Address for correspondence: Dott. Luciano Bubbico, Via Nemorense 93, 00199 Roma, Italy.
Fax: +39 06 86206602.