



Case Report

# Human Cervico-Facial Actinomycosis: Actinomyces in Chronic Granulomatous Disease, Epidemiological and Clinical Comments

Silvia Tortorici\*

Department of Oral- Surgery University of Palermo, Italy

University of Palermo Via del Vespro N 129 CAP 90127, Palermo, Italy

\*Corresponding author: Silvia Tortorici, Department of Oral- Surgery University of Palermo, Italy

Citation: Tortorici S (2022) Human Cervico-Facial Actinomycosis: Actinomyces in Chronic Granulomatous Disease, Epidemiological and Clinical Comments. Ann Case Report 7: 963. DOI: 10.29011/2574-7754.100963

Received: 20 September 2022, Accepted: 24 September 2022, Published: 27 September 2022

## Abstract

In a retrospective investigation performed in the department of Surgical Odontostomatology of Palermo University Polyclinic from 2012 to 2021, the Author examined 10 cases of crevice-facial actinomycosis, taking into consideration age range, gender distribution, predisposing factors and symptoms. In concordance with reports in literature, she found that the disease was per mandibular in 65%; she also report the diagnostic methods and therapeutic approaches used in the study.

**Keywords:** Skin fistula; Actinomycosis; Differential diagnosis

## Introduction

Actinomycosis is a chronic infectious disease, caused by an anaerobic Gram-plus germ belonging to the Antinomies family; It is a rare disease occurring in three different clinical forms -crevice-facial, thoracic and abdominal. The crevice-facial site is the most common of the three types, and is found in about 50% of the cases; it is also known as lumpy jaw and involves both the soft and hard tissue of the head and neck region: the scalp, the paranasal sinuses, the palate, the parotid gland, the tear glands, the cheeks, the lower jaw, the tongue, the larynx and the lower pharynx. Antinomies provokes a chronic productive and colliquative inflammatory reaction known as “actinomycotic granuloma”. The aim of this study was to define the diagnostic allorhythmia of crevice-facial actinomycosis by drawing attention to its differential features compared with other diseases showing similar ulcerative and infiltrating clinical aspects.

## Materials and Methods

A retrospective investigation conducted in the department of Surgical Odontostomatology of Palermo University Polyclinic

from 2012 to 2021, led to the diagnosis of 10 cases of crevice-facial actinomycosis in patients ranging from 16 to 71 years of age (mean 45.67 years) and made up of 5 men (50 %) and 5 women (50 %). The patients presenting fistulas were affected by enema, swelling of the soft tissues with the formation of skin abscesses and general symptoms such as fever and weight loss. The following predisposition factors were identified from the patients’ medical history:

- 2 patients had periodontal pockets
- 3 patients had previously undergone tooth extraction
- 5 patients had deep dental decay

In no case was there lymph node involvement. All the patients underwent an X-ray examination and anatomy-pathological analysis involving desquamate or fine-needle aspiration cytology, either by means of the F.N.A.B. method or by exudate sampling.

## Discussion

Human actinomycosis, a chronic, granulomatous infectious disease, has been recognized for a long time [1], and its causative agent, originally named Streptothrix Israeli (currently Antinomies Israeli), was described in 1896 by Kruse [2]. It was not until

1951 that another Antinomies species, *Antinomies naeslundii*, was implicated in actinomycotic lesions in humans [3], while *Antinomies odontolyticus* and *Antinomies viscous* (first named as *Odontomyces viscous*) were described in 1958 and 1969, respectively [4-6]. It is well established that actinomycosis is an endogenous infection. The causative *Antinomies* species reside on mucosal surfaces and gain access to deeper tissues via trauma, surgical procedures, or foreign bodies, which disrupt the mucosal barrier. Inside the tissue, these bacteria form masses consisting of aggregates of branching, filamentous bacilli [7-9]. Actinomycosis is defined as a hard mass-type lesion with a specific histopathological structure. There are a large number of case reports of actinomycosis in the literature, but in most cases, diagnosis has been based solely on clinical and histopathological findings. In the majority of early reports, microbiological confirmation of diagnosis was lacking. Even when microbiological assessment was included, culture was typically the only method used. If, however, antimicrobial treatment had been started before sample collection, the results of culture may be falsely negative. The increasing introduction of molecular bacterial detection and identification methods is helping to overcome such problems. A large number of *Antinomies* species have been described since the description of *A. Israeli*, *A. naeslundii*, *A. odontolyticus*, and *A. viscous*. In addition, reassignments within some species, such as *A. naeslundii* and *A. viscous*, have occurred [10]. However, only some human-associated *Antinomies* species, including *A. Israeli*, *Antinomies gerencseriae*, and *Antinomies graevenitzii*, may be involved in classical actinomycosis [11,12]. A wide range of *Antinomies* species are being increasingly associated with infections at many body sites [11,13,14]. *Antinomies Meyer*, *Antinomies naoi*, and *Antinomies turicensis* are emerging as important causes of such infections. Although actinomycosis is relatively rare, at least in Western populations [8], recently reported observations implicating *A. Meyer* in brain abscesses [15] and *Actinobaculum scholia* (currently *Actinotignum scholia*) in urosepsis [16] and the introduction of advanced microbiological techniques, which can identify even very fastidious organisms, have resulted in an increased awareness of *Antinomies* and other Gram-positive, non-spore-forming bacilli in clinical microbiology.

## Symptoms

The incubation period ranges from two months to a year. In the crevice-facial form, a hard, painless swelling with unclear margins forms in the soft tissues; in time, this becomes fluctuating and small, multiple, communicating abscesses surrounded by granulation tissue are formed the purulent material contains yellowish sulphurous granules. The infectious process spreads to the surrounding tissues such as the pharynx, the tongue, the saliva glands, the jaw bones, with resulting otitis, or the skin, forming fistulas in various points; sometimes, but not often, the infection may enter the blood stream and cause a general form

which may be fatal. When the skin surface is involved, there is a painless pubescence, with fluctuating zones alternating with hard, wooden areas; there may be one or more syringed openings. If no surgical action is taken, the fistulas tend to heal and new openings may form. This process gives rise to retraction of the scar tissue, deformation of the skin and aesthetic damage. The clinical variants of the crevice-facial form are:

- chronic otitis
- osteocytes lesion with granulation tissue which presents as polycystic at X-ray examination [8]
- on the tongue (3% of cases), with the formation of a hard nodule attached to the muscular layer [9]
- at the pterygomaxillary site with lockjaw
- as a par apical or paradental abscess [ 10,11]

The regional lymph nodes are rarely involved and if so, without pain [12].

## Anatomy-pathological aspect

The main element is an “actinomycotic granuloma”, made up of a network of threads and lavaform formations; the peripheral tissue is granulomatous and full of plasma cells, fibroblasts, giant-cells and blood vessels; there is also an infiltrate of polymorph nucleates. An actinomycotic granuloma may develop into a purulent form with several fistulas in the case of multiple granulomas or into a productive form with connectivization tendencies.

## Diagnosis

The osteocutaneous fistula simulates an odontogenic abscess, especially when the clinical picture has been altered by an improper use of antibiotics; furthermore, the presence of sulphurous granules visible to the naked eye, should suggest the presence of an actinomycosis. Diagnosis is based not only on the symptoms, but also on the identification of *Antinomies Israeli* in the sierra-purulent sputum or in the biopsy specimens. Fine-needle aspiration is a useful sampling method in order to avoid contamination with a specific amicrobic flora [13]. Silver meth enamine shows up the threads bunched up in balls or tufts [14]. Another useful diagnostic method is an *Antinomies* culture on glioglycolate and on agar-glucose [15,16]. This diagnostic work-up is essential for a differential diagnosis compared with other ulcerative diseases or infiltrative masses of the jaw [17]. A CT scan and MRI are not sufficient for distinguishing actinomycosis from malignant humoral masses [18,19].

## Learning points

- Human actinomycosis is an a specific granulomatous chronic bacterial infection caused by *Antinomies Israeli* a Gram-positive, non-acid-fast, anaerobic bacteria and a normal commensal flora of the oral cavity that cannot penetrate healthy tissue and requires mucosal breakdown as a prerequisite for infection.

- Surgical excision of the mass followed by histopathological staining and special staining with modified Gram, Gomora meth enamine silver and periodic acid-Schiff of the tissue remains the only resolute approach to make a definitive diagnosis.
- A very close differential diagnosis of actinomycosis is nocardiosis but the granules in the latter consist of acid-fast branched bacilli. Botryomycosis may also have similar presentation but the granules in this case contain non-filamentous cocci.
- To conclude, any soft tissue mass or swelling on the cervicofacial area should be investigated for cervicofacial actinomycosis.

A biopsy should be performed when a small alveolar carcinoma lesion is found together with massive infiltration of the surrounding tissues; in such cases the presence of actinomycosis may complicate the situation [20]. It is also essential to perform a histological examination in cases where a parotid carcinoma includes a hard swelling which simulates an actinomycotic in durative mass and which may develop into a skin fistula in such cases the regional lymph nodes are not always involved [21]. Diagnostic doubts are also necessary in cases of oral tuberculosis and of ulcerated syphilitic gummi.

## Treatment

A therapy with penicillin for at least 3 months is the mainstay of the treatment of actinomycosis. Alternatively, second line drugs like erythromycin, tetracycline may also be used in patients who are allergic to penicillin. Cephalosporin's may also be used in the acute phase along with steroids to eliminate the residual inflammatory reaction [22].

## References

1. Israel J (1878) Neue Beobachtungen auf dem Gebiete der Mykosen des Menschen. Arch Pathol Anat 74: 15-53.
2. Kruse W (1896) Systematik der Streptothricheen und Bakterien, In Flugge C. (ed), Die Mikroorganismen, 3rd ed, 2: 48-96.
3. Thompson L, Lovestedt SA (1951) An Actinomyces-like organism obtained from the human mouth. Proc Staff Meet Mayo Clin 26: 169-175.
4. Batty I (1958) Actinomyces odontolyticus, a new species of actinomycete regularly isolated from deep carious dentine. J Pathol Bacteriol 75: 455-459.
5. Howell A Jr, Jordan HV, Georg LK, Pine L (1965) Odontomyces viscosus, gen. nov., spec. nov., a filamentous microorganism isolated from periodontal plaque in hamsters. Sabouraudia 4: 65-68.
6. Georg LK, Pine L, Gerencser MA. (1969) Actinomyces viscosus, comb. nov., a catalase positive, facultative member of the genus Actinomyces. Int J Syst Bacteriol 19: 291-293.
7. Smego RA, Foglia G. 1998. Actinomycosis. Clin Infect Dis 26: 1255-1261.
8. Wong VK, Turmezei TD, Weston VC (2011) Actinomycosis. BMJ 343: d6099.
9. Russo TA (2009) Agents of actinomycosis. In Mandell GL, Bennett JE, Dolin R (ed), Principles and practice in infectious diseases, 7th ed Elsevier, Philadelphia, PA 2: 2864-2873.
10. Henssge U, Do T, Radford DR, Gilbert SC, Clark D, et al (2009) Emended description of Actinomyces naeslundii and descriptions of Actinomyces oris sp. nov. and Actinomyces johnsonii sp. nov., previously identified as Actinomyces naeslundii genospecies 1, 2 and WVA 963. Int J Syst Evol Microbiol 59: 509-516.
11. Hall V (2008) Actinomyces gathering evidence of human colonization and infection. Anaerobe 14: 1-7.
12. Pulverer G, Schutt-Gerowitt H, Schaal KP (2003) Human cervicofacial actinomycoses: microbiological data for 1997 cases. Clin Infect Dis 37: 490-497.
13. Schaal KP, Lee HJ (1992) Actinomycete infections in humans a review. Gene 115: 201-211.
14. Wade WG, Könönen E (2011) Propionibacterium, Lactobacillus, Actinomyces, and other non-spore-forming anaerobic gram-positive rods, p 817–833. In Versalovic J, Carroll KC, Funke G, Jorgensen JH, Landry ML, Warnock DW (ed), Manual of clinical microbiology, 10th ed ASM Press, Washington, DC.
15. Kommedal Ø, Wilhelmsen MT, Skrede S, Meisal R, Jakovljevic A, Gaustad P, Hermansen NO, Vik-Mo E, Solheim O, Ambur OH, Sæbø Ø, Høstmælingen CT, Helland C (2014) Massive parallel sequencing provides new perspectives on bacterial brain abscesses. J Clin Microbiol 52: 1990-1997.
16. Cattoir V (2012) Actinobaculum schaalii: review of an emerging uropathogen. J Infect 64: 260-267.
17. Johnson JL, Moore LV, Kaneko B, Moore WE (1990) Actinomyces georgiae sp. nov., Actinomyces gerencseriae sp. nov., designation of two genospecies of Actinomyces naeslundii, and inclusion of A. naeslundii serotypes II and III and Actinomyces viscosus serotype II in A. naeslundii genospecies 2. Int J Syst Bacteriol 40: 273-286.
18. Lawson PA, Falsen E, Akervall E, Vandamme P, Collins MD (1997) Characterization of some Actinomyces-like isolates from human clinical specimens: reclassification of Actinomyces suis (Soltys and Spratling) as Actinobaculum suis comb. nov. and description of Actinobaculum schaalii sp. nov. Int J Syst Bacteriol 47: 899-903.
19. Collins MD, Pascual C (2000) Reclassification of Actinomyces humiferus (Gledhill and Casida) as Cellulomonas humilata nom. corrig., comb. nov. Int J Syst Evol Microbiol 50: 661-663.
20. Yassin AF, Hupfer H, Siering C, Schumann P (2011) Comparative chemotaxonomic and phylogenetic studies on the genus Arcanobacterium Collins et al. 1982 emend. Lehen et al. 2006: proposal for Trueperella gen. nov. and emended description of the genus Arcanobacterium. Int J Syst Evol Microbiol 61: 1265-1274.
21. Greub G, Raoult D (2002) "Actinobaculum massiliae," a new species causing chronic urinary tract infection. J Clin Microbiol 40: 3938-3941.
22. Lancella A, Abbate G, Foscolo AM, Dosedegani R (2008) two unusual presentations of cervicofacial actinomycosis and review of the literature. Acta Otorhinolaryngol Ital 28: 89-93.