

A CASE OF CERVICOFACIAL ACTINOMYCOSIS

S. Racheva, Zh. Dimitrova, Stoian Pavlov, Zl. Lolev
*Department of Dermatology and Venerology,
Medical University – Varna, Bulgaria*

RESUME:

The Actinomycosis in humans is rarely observed in Europe. It is presented as a chronic, suppurative, granulomatous infection, originally caused by *Actinomyces israelii* (found as normal microflora in the mouth cavity) and dependent on the associative bacterial flora.

The most frequent form of Actinomycosis in humans is the Cervicofacial, followed by the thoracic, abdominal etc.

The slow development of the infection, the thick tumorlike nodules which it forms and the abscesses and fistulas usually hinder the diagnosis of the Actinomycosis and lead to other initial diagnoses – tumors, phlegmons, etc.

This is a case of Cervicofacial Actinomycosis in a 59-year-old patient, with whom a tumoral nodule in the facial area tends to grow slowly. Originally the nodule was diagnosed as a planocellular papillomatous on the right side of the gums and the hard palate, affecting the skin of the zygomatic area secondarily with formation of fistulas.

Key words: Actinomycosis, cervicofacial

INTRODUCTION:

The Actinomycosis in humans is rarely observed in Europe. Separate cases are described in literature. It is presented as a slowly evolving chronic, suppurative, granulomatous infection. It is caused by a bacterium of the *Actinomyces* class, supporting in the mouth cavity and dependent in its clinical expressions on the associative bacterial flora.

Diagnosing the Actinomycosis is usually hard for the clinical worker, as it involves a wide differential diagnosis and is achieved after a number of misdiagnoses. The case described herein is no exception.

DESCRIPTION OF THE CASE:

The patient is a 57-year-old man who in 2003 for a period of 4-5 months had a painful tumor-like nodule in the right zygomatic area. Originally, a papillomatous formation appeared, followed by swelling, thickening, and reddening of the right cheekbone, where a month later three fistulas appeared filled with abundant purulent exudation.

A 'Fistula' operation on the right side of the nose thirty years before was reported, as well as serious loosening of the teeth on the right upper jaw seven years before.

In the surgery ward, the three biopsies performed revealed a histological picture of an inverting planocellular papilloma. The computer tomography of the perinasal cavities visualizes a widespread soft tissue lesion of inverting nature, a destruction of the right branch of the maxilla and infiltration of the soft facial tissues. The patient was hospitalized in the Clinic of Dermatology, a year after the initial symptoms and six months after the biopsies.

DERMATOLOGICAL STATUS:

The changes are localized in the right infraorbital and zygomatic area as a considerable edema and infiltrate of 'wooden' thickness, and the skin covering it is of livid erythematous color. At this background there are three fistula openings from which purulent yellowish exudation is discharged spontaneously and upon pressure. The fistula openings are covered with yellow-green crusts, which upon removal reveal vegetation and exudative surfaces. The lower eyelid is highly erythematous and edematous. (Fig. 1).



Figure 1.

In the mouth cavity, in the right gum area – a tumorous formation with papillomatous surface and numerous whitish granulous changes; the formation covers

part of the palate and the vestibulum of the mouth cavity



(Fig. 2).

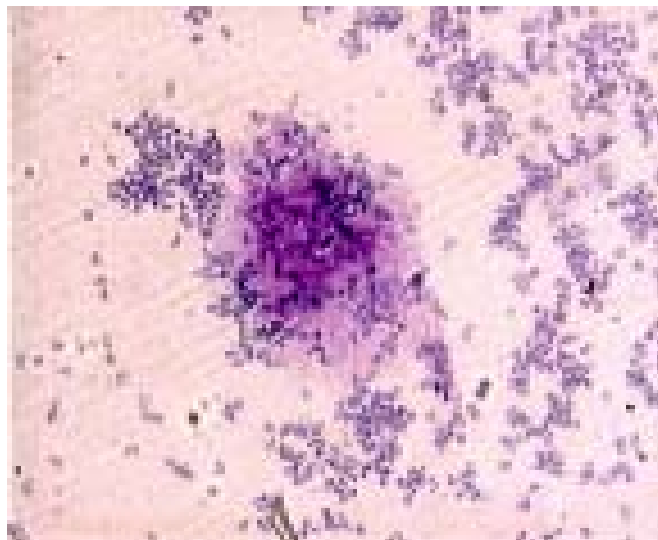
Figure 2.

Destruction of the alveolar process and the maxilla on the right.

Three enlarged peripheral lymph nodes are found – one submandibular, two – behind the right sternocleidomastoideus. Size – about 0.5 cm in diameter, slightly painful upon palpation, soft, elastic and loose as far as the surrounding structures are concerned.

The patient was afebrile; no affected internal organs were found. The paraclinical tests were within norms, except for an accelerated ESR.

The tests for lues were negative. Intradermal tuberculin test - 15 mm. Two fistula material cultures in a medium for aerobes and anaerobes /lab. ¹1083- 25.05.04 / – *Staphylococcus aureus* was isolated. Gram staining – abundant dross. Two cytosmears of fistular exudate / ¹1563 – 14.05.04/ - purulent exudation with complexes of pseudox-



anthomous cells, keratinocytes and actinomycetous dross. (Fig.3).

Figure 3.

A 13-month long treatment was applied: 60 days - crystal Penicillin / 12 million Å daily/, followed by 20 days – Rifampicin (200 mg daily), together with Íспен tablets(2x 10000 mg). During the following months the therapy continued with once weekly application of Retarpen (2400000 E) and intermittent two-week courses with Rifampicin.

After a one-year treatment the edema, erythema and infiltrate on the skin decreased considerably, as well as the fistular exudation. The tumor-like formation in the mouth cavity diminished considerably in size, lost its papillomatous structure, and no whitish grains on the surface were ob-



served. (Fig.4)

Figure 4.

DISCUSSION:

The opinion that there is exceptionally low frequency of Actinomycosis in the European countries is dubious, but there is a lack of sufficient statistics for the infection. It is often diagnosed after several misdiagnoses, and resembling a tumor or an abscess it remains undiscovered. Therefore anaerobic flora has to be sought persistently.(18).

The Actinomycosis can be caused in humans by *Actinomyces Israelii* - a grampositive, anaerobic, filamentously branching-out, no-spores-forming bacteria, which is part of the saprophytic microflora in human mouth cavity. It is found in the tonsillar crypts, the tooth plaque, periodontium and the periodontal sacks. Tooth manipulations or facial traumas can provoke the malady as a slowly developing, granulomatous, suppurative infection. Predominant is the opinion that with Actinomycosis endogenous infection is developed upon penetration of this anaerobe / in tooth manipulations or traumas/ into the deep tissues. Of great importance for the development of the malady is the associative microflora

- other anaerobic and aerobic bacterial agents which cause the development of abscesses, fistulas and purulent exudation in the area of the original granulomatous infiltrate. The spread of the process overcomes the anatomic borders of the organ structures and the skin is affected secondarily. Susceptible to the infection are HIV positive patients (11).

Various localizations of the infection are described. Undoubtedly, the most frequent is the cervicofacial form - 50-60% (2, 17, 21), followed by the thoracic - 13-15% (6, 15) and abdominal - 20% (15). Described are also renal Actinomycosis (19), Actinomycosis of the central nervous system (10, 14, 22), and perianal (7).

The cervicofacial actinomycosis is usually preceded by a tooth extraction accident or trauma. The formation of a nodular formation with 'wooden' thickness of the infiltrate follows and with the secondary affection, the skin becomes purple to livid erythematous and warm. The process can be presented clinically as maxillotemporal osteoarthritis (8), as osteitis of the mandibula (20), periosteitis (21) or posttraumatic osteomyelitis (21).

Of the cervicofacial forms of the actinomycosis the most frequent is the one localized in the submandibular triangle and the mandibular area - 53% (2, 21). But there are descriptions of other locations when various facial structures, external auditory canal (5), larynx (16) are affected.

The case described herein is of interest because of the comparatively rarity of this disease in the dermatological practice and the long route of the patient to his final diagnosis.

The slow clinical development, the localization of the process and the morphological peculiarities (thick infiltrate, fistulas with seropurulent exudation) led to Actinomycosis.

The histology of biopsy material did not contribute to the confirmation of the diagnosis, which corresponds to the data in literature. The histological picture is specific (4), but dross could be scarce or lacking (3). The microscopic smear test of fistular exudation dross is found in not more than 40% of the cases, and the culture test is positive in less than 50% (1). Usually the diagnosis is based either on the morphology of the bacterium with the dross or on a direct find (in a smear of exudation, culture) of dross (1). Fine needle biopsy is also recommended (9). With the described case dross was found in the purulent exudation smear test (twice), and also in the anaerobic culture.

The treatment of Actinomycosis is efficient with high doses of Penicillin injections for a long time (for at least a month), combined with Erythromycin, Rifampin, lincomycin, per os (12). Some authors recommend therapy with quinolones - ciprofloxacin (13).

In our case the 13-month-long treatment with a penicillin product in combination with Rifampin (suitable for treating the associative bacterial flora) showed good results.

CONCLUSIONS:

The case is interesting not only because of its being rarely diagnosed, but also because of its long history, length and hard-to-establish diagnosis.

The meeting with this rare infection requires it to be considered by various specialists as Actinomycosis in patients with soft tissue tumor-like formations with fistulas on the face and neck.

The meeting with this rare infection requires it to be considered by various specialists as Actinomycosis in patients with soft tissue tumor-like formations with fistulas on the face and neck.

sis of the larynx, Ear Nose Throat J, 1992, 71, 356-358.

17. Pordey RC, Lumpy Jaw due to Actinomyces meyeri: Report of the first case and review of the literature, Mt Sinai J Med, 1988, 55, 190-3.

18. Richtsmeier WJ, Johns ME, Actinomycosis of the head and neck, Clin Lab Sci, 1979, 11(2), 175-202.

19. Rosenblum PS, Renal actinomycosis. A case report, Urol and Cutan Rev, 1949, 53, 329.

20. Sartory A, Roger R, Meyer J, Deux cas d'actinomycose inactive sans grains microscopiques, Bull Soc Med, CVII 16, 1932 and 3rd suppl. 125, 1933.

21. Schaal KP, Beaman BL, Clinical significance of actinomyces. In: Goodfellow M, Mordarski M, Williams ST, The biology of actinomycetes, New York, Academic Press, 1983, 389-434.

22. Smego RA, Actinomycosis of the central nervous system, Rev Infect Dis, 1987, 9, 855-865.

BIBLIOGRAPHY:

1. Ashton N, Cook C, Allergic granulomatous nodules of the eyelid and conjunctiva, Am J Ophthalmol, 1979, 87, 1-28.

2. Bochev V, Angelov I, Tsancov N, Cervicofacial actinomycosis - report of two cases, Acta Dermatoven, APA, 2003, 12, 3 105-108.

3. Brown JR, Human actinomycosis a study of 181 subjects, Hum Pathol, 1973, 4, 319-330.

4. Burden P, Actinomycosis, J Infect, 1989, 19, 95-9.

5. Chang CJ, Lalwani AK, Lanser MJ, Actinomycosis of the external auditory canal, Otolaryngol Head Neck Surg, 1993, 108, 73-75.

6. Cope VZ, Actinomycosis, University Press, London, Oxford 1938.

7. Gayraud A, Grosieux-Dauger C, Durlach A, Actinomycose cutanee perianale et fessiere, Ann Dermatol Venereol, 2000, 127, 393-6.

8. Grigoriu D, Maillard FG, Goumaz FC, L'actinomycose, Rev Med Sues Romande, 1969, 90 325-36.

9. Hong IS, Mezgebe HM, Gaiter TE,

Loffon J, Actinomycosis of the neck, diagnosis by fine needle aspiration biopsy, J Nafl Med Assoc, 1993, 85, 145-146.

10. Intile JA, Richest JH, Cervicofacial actinomycosis complicated by meningitis, J Am Med Ass, 1962, 181, 724-6.

11. Kingdom TT, Tami TA, Actinomycosis of the nasal septum in a patient infected with the human immunodeficiency virus, Otolaryngol Hed Neck Surg, 1994, 111, 130-133.

12. Lerner PI, The lumpyjaw, cervicofacial actinomycosis, Infect Dis Clin North Am, 1988, 2, 203-19.

13. Macfarlane DJ, Tucker LG, Kemp RJ, Treatment of recalcitrant actinomycosis with ciprofloxacin, J Infect, 1993, 27, 177-180.

14. Martin WJ, Nichols DR, Wellman, Disseminated actinomycosis treated with tetracycline, Arch Intern Med, 1956, 97, 252.

15. Mousseau A, Mousseau-Brodu CM, L'actinomycose abdominale, J Chir, Paris, 1973, 106, 565.

16. Nelson EG, Tubor AG, Actinomyco-