

# Actinomycosis of the Cheek

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## SUMMARY

**Introduction** Actinomycosis is an uncommon chronic granulomatous infection first described by Bollinger in 1877. The infection is caused by actinomyces species and it is characterized by slow contiguous spread and suppurative inflammation, formation of multiple abscesses and sinus tracts with possible drainage of "sulfur granules".

**Case Outline** We report an unusual case of actinomycosis of the cheek that occurred 6 years after buccal odontogenic abscess. A 56-year-old male was referred to the Department of Oral Surgery because of a painless swelling of the left cheek, which initiated three weeks prior to the referral. The diagnosis of actinomycosis was confirmed by histopathologic examination. In accordance with the diagnosis oral penicillin was prescribed for four months with complete resolution.

**Conclusion** This case of actinomycosis is presented as a rarity. For proper diagnosis, careful examination and a high degree of clinical suspicion are necessary.

**Keywords:** actinomycosis; buccal; granulomatous infection

## INTRODUCTION

Actinomycosis is a rare chronic granulomatous infection caused by actinomyces species; it is characterized by a slow contiguous spread and suppurative inflammation, formation of multiple abscesses and sinus tracts with possible drainage of "sulfur granules" [1]. Actinomyces, first described by Bollinger in 1877, are gram-positive, non-acid-fast, anaerobic or facultative anaerobic bacteria which are very difficult to cultivate [2]. This commensal bacteria usually inhabits the human oral cavity, respiratory and digestive tracts, and becomes invasive when it gains entry to the submucosal or subcutaneous tissue through cutaneous or mucosal lesions [3]. Infection is more common in rural areas than in cities. Antecedent conditions that can predispose infection are: maxillofacial trauma, tooth extraction, periodontal pockets, poor dental hygiene, immunodeficiency or conditions such as osteonecrosis after radiotherapy or after orthognathic surgery. However, there have been a few cases where the focus of infection could not be specified. The various sites of infection were described: the scalp, the forehead, the nose, the paranasal sinuses, the palate, the parotid gland, the temporal bone, the lacrimal glands, the minor salivary glands, the cheeks, the lower jaw, the tongue, the lip, the larynx and the lower pharynx [4-11]. Although uncommon, and at present estimated a rare disease in Europe [12], actinomycosis is an important clinical entity because of its difficult diagnosis due to non-specific imaging findings, symptoms that can mimic other diseases and because of long-lasting treat-

ment. Although cervicofacial actinomycosis occurs infrequently, it should be included in the differential diagnosis when images show a soft-tissue mass with inflammatory changes and an infiltrative nature in the cervicofacial area. A CT scan and MRI are not sufficient for distinguishing actinomycosis from malignant tumoral masses [13]. Actinomycosis is now uncommon in Europe and the possibility that we may face a patient with actinomycosis is therefore underestimated [12].

We report an unusual case of actinomycosis of the cheek that occurred 6 years after buccal odontogenic abscess, intraoral incision and extraction of tooth 36.

## CASE REPORT

A 56-year-old male was referred to the Department of Oral Surgery because of a painless swelling in the left cheek, which he had noticed three weeks prior to the referral. Empirical antibiotic therapy prescribed by a primary care physician did not lead to the decrease in size of the swelling. The patient who dwells in an urban area, without any pets in his household, firmly denied any contact with animals or visiting rural areas in the last 6 years, he also denied any recent odontogenic symptoms or maxillofacial trauma prior to the appearance of the swelling. However, the patient did mention having a buccal abscess 6 years earlier because of a decayed tooth 36, which was then extracted. An intraoral incision was also performed. After that painful experience the patient took good care of his remaining teeth

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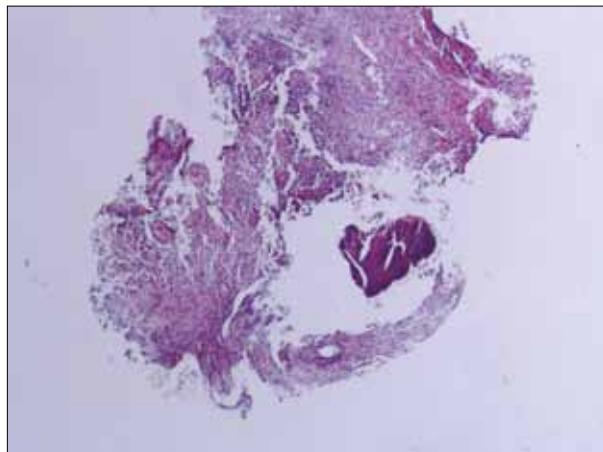
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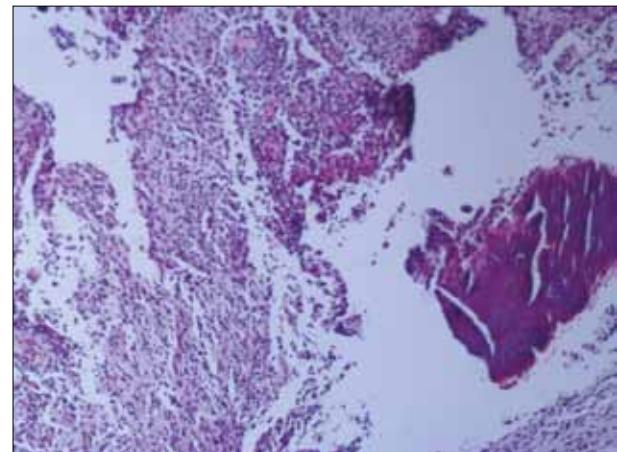
**Figure 1.** Left cheek with slightly erythematous overlaying skin



**Figure 2.** Intraoral finding, partially edentulous arches with no abnormality



**Figure 3.** Biopsy specimen composed of granulation tissue surrounding an abscess cavity with "drusa" formation of actinomycete colonies (Hematoxylin and eosin,  $\times 40$ )



**Figure 4.** Biopsy specimen composed of granulation tissue surrounding an abscess cavity with "drusa" formation of actinomycete colonies (Hematoxylin and eosin,  $\times 100$ )

and visited his primary dentist on a regular basis. The patient had no record of serious disease in his medical history and no sign of immunodeficiency. The possibility of inoculation of the bacteria with eventual biting of the buccal mucosa was excluded since the patient did not have molars in the upper and lower left jaws, which were extracted earlier except tooth 36. The patient did not use partial dentures. According to that, the only possible moment of inoculation of actinomycetes was 6 years earlier, at the time when tooth 36 was extracted and incision was performed because of an odontogenic abscess in the buccal region. Clinical examination revealed a facial asymmetry, with swelling of the left cheek measuring cca  $2 \times 2$  cm with a slightly erythematous overlaying skin (Figure 1). Upon palpation the mass was round, indurated and movable. No significant cervical lymphadenopathy was noted. Intraoral examination showed partially edentulous arches with no abnormality discovered, with well maintained oral hygiene (Figure 2). Routine panoramic radiograph was performed with a normal radiological finding. The patient was referred to the Cytology Department for cytological examination. Cytological finding showed subacute inflammation and was non-specific. Thus it was decided to perform an incisional biopsy under local anesthesia. The patient gave

informed consent for incision, which revealed granulation tissue with discharge of yellowish "sulfur granules".

Histopathologic examination revealed a specimen composed of granulation tissue surrounding an abscess cavity with amorphous blue masses of actinomycete colonies in a characteristic "drusa" formation.

The diagnosis of actinomycosis was confirmed by histopathologic examination showing chronic granulomatous inflammation with the presence of multiple granules of actinomyces surrounded by polymorphonuclear neutrophils (Figures 3 and 4). According to the diagnosis oral penicillin was prescribed for four months with complete resolution.

## DISCUSSION

Actinomyces species is divided in several subgroups. Actinomycosis in humans is mainly caused by *Actinomyces israelii*. There are at least 5 types of actinomycosis based on the site of involvement. Cervicofacial actinomycosis is most frequent and accounts for around 60% of all cases; it occurs in the oral cavity and in the region of the head and neck. *Actinomyces* species is commensal in the oral

cavity as well as in the overall gastrointestinal tract, and it is believed that caries, periodontal disease, mucosal injuries, poor hygiene and immunodeficiency can lead to the development of actinomycosis. Pathogens can enter the respiratory tract by inhalation and cause thoracic actinomycosis which mainly involves the lungs and mediastinum. Abdominal actinomycosis most frequently occurs after surgery, pelvic and generalized types of actinomycosis are also described in the literature [14, 15, 16].

It is important to emphasize that after inoculation of actinomycetes or inadequate first treatment, actinomycosis can reoccur years after the first inoculation [17, 18]. However, the reason why actinomycosis may manifest itself years after inoculation still remains unknown [19].

Actinomycosis often mimics other diseases because of non-specific radiological findings and non-specific symptoms that involve indurated swelling characterized by a slow progress and with the formation of multiple sinus tracts that lack response on the empirical application of antibiotics. It is important to mention that the appearance of "sulfur granules", although a helpful sign, is not pathognomonic for actinomycosis since they can also be present in nocardiosis [20].

Lymphatogenous spread of actinomycetes is rare because of the size of bacteria; cervical lymphadenopathy eventually develops in the late stages of the disease, which can be helpful in the differential diagnosis from malignant neoplasms. Because of the above given reasons and due to a difficult cultivation of bacteria, the diagnosis of actinomycosis still

remains difficult. The disease is therefore often misdiagnosed as tumorous, granulomatous disease or fungal infection [13, 21]. In obtaining the definitive diagnosis, incisional biopsy and histopathological analysis are necessary. Typical microscopical findings for actinomycosis indicate two zones; an outer zone of granulation and a central zone with multiple granules representing colonies of actinomycetes [22]. Because of diagnostic difficulties, actinomycosis is also known in the literature as a great mimicker, or when referring to the cervicofacial actinomycosis as a great masquerader of head and neck disease [23, 24]. Treatment of actinomycosis consists of surgery, primarily of incision and drainage, and of antibiotic therapy. Inadequate short term antibiotic therapy can result in the relapse of infection after apparent cure. To prevent the chance of relapse of the disease, prolonged antibiotic therapy is necessary [25]. Penicillin is the antibiotic of choice, but for patients who are allergic to penicillin valid alternatives include administration of tetracycline, clindamycin, erythromycin and cephalosporin antibiotics [26].

In conclusion, we must emphasize that the prognosis of the disease is good after an early diagnosis and adequate treatment. The presented case of actinomycosis of the cheek 6 years after tooth extraction is presented as a rarity, which clinicians need to take into account in the differential diagnostics of tumors and similar lesions in the soft tissue of the oral cavity and the region of the head and neck. Because of difficult diagnosis a high dose of clinical suspicion is needed.

## REFERENCES

- Nakagawa Y, Asada K, Doi Y. Oral maxillofacial actinomycosis – clinical and bacteriological studies of 13 cases. *Jpn J Oral Maxillofac Surg.* 1985; 31:110-5.
- Bollinger O. Ueber eine neue Pilzkrankheit beim Rinde. *Zentralblatt Medizinische Wissenschaft.* 1877; 15:481-90.
- Volante M, Contucci AM, Fantoni M, Ricci R, Galli J. Cervicofacial actinomycosis: still a difficult differential diagnosis. *Acta Otorhinolaryngol Ital.* 2005; 25(2):116-9.
- Belmont MJ, Behar PM, Wax MK. Atypical presentations of actinomycosis. *Head Neck.* 1999; 21(3):264-8.
- Tamura T, Ryoke K, Kidani K, Takubo K, Nakabayashi M, Amekawa S. A case of actinomycosis of the minor salivary gland in the buccal region. *Oral Sci Int.* 2008; 5(2):131-4.
- Tortorici S, Burruano F, Buzzanca ML, Difalco P, Cabibi D, Maresi E. Cervico-facial actinomycosis: epidemiological and clinical comments. *Am J Infect Dis.* 2008; 4(3):204-8.
- Bartell H, Sonabend ML, Hsu S. Actinomycosis presenting as a large facial mass. *Dermatol Online J.* 2006; 12(2):20.
- Che Y, Tanioka M, Matsumura Y, Kore-Eda S, Miyachi Y. Primary cutaneous actinomycosis on the nose. *Eur J Dermatol.* 2007; 17(2):167-8.
- Cucuroccia B, Gubinelli E, Fazio M, Girolomoni G. Primary cutaneous actinomycosis of the forehead. *J Eur Acad Dermatol Venerol.* 2003; 17(3):331-3.
- Schwartz HC, Wilson MC. Cervicofacial actinomycosis following orthognathic surgery: report of 2 cases. *J Oral Maxillofac Surg.* 2001; 59(4):447-9.
- Sanchez-Estella J, Bordel-Gomez MT, Cardenoso-Alvarez E, Gabarito-Solovera E. Actinomycosis of the lip: an exceptional site. *Actas Dermosifiliogr.* 2009; 100(9):817-32.
- Bochev V, Angelova I, Tsankov N. Cervicofacial actinomycosis – report of two cases. *Acta Dermatovenerol Alp Panonica Adriat.* 2003; 12(3):105-8.
- Park JK, Lee HK, Ha HK, Choi HY, Choi CG. Cervicofacial actinomycosis: CT and MR imaging findings in seven patients. *AJNR Am J Neuroradiol.* 2003; 24(3):331-5.
- Marr CR, Sebelik ME, Hodges JM. Actinomycosis presenting as a temporal mass. *Otolaryngol Head Neck Surg.* 2005; 132(1):163-4.
- Palonto F, Preti G, Vione N, Cavalot AL. Actinomycosis of the masseter muscle: report of a case and review of the literature. *J Craniofac Surg.* 2003; 14(6):915-8.
- Milojković M, Mrcela M, Rubin M, Pajtler M. The case of asymptomatic primary actinomycosis of the greater omentum in the patient with intrauterine contraceptive device. *Coll Antropol.* 2007; 31(4):1169-71.
- Habibi A, Salehinejad J, Saghafi S, Mellati E, Habibi M. Actinomycosis of the tongue. *Arch Iranian Med.* 2008; 11(5):566-8.
- Tambay R, Côté J, Bourgault AM, Villeneuve JP. An unusual case of hepatic abscess. *Can J Gastroenterol.* 2001; 15(9):615-7.
- Meade RH. Primary hepatic actinomycosis. *Gastroenterology.* 1980; 78(2):355-9.
- Graybill JR, Silverman BD. Sulfur granules. Second thoughts. *Arch Intern Med.* 1969; 123(4):430-2.
- Pant R, Marshall TL, Crosher RF. Facial actinomycosis mimicking a desmoid tumour: case report. *Brit J Oral Maxillofac Surg.* 2008; 46(5):391-3.
- Brown JR. Human actinomycosis. A study of 181 subjects. *Hum Pathol.* 1973; 4(3):319-30.
- Ngow HA, Wan Khairina WM. Cutaneous actinomycosis: the great mimicker. *J Clin Pathol.* 2009; 62(8):766.
- Rankow RM, Abraham DM. Actinomycosis: masquerader in the head and neck. *Ann Otol Rhinol Laryngol.* 1978; 87(2):230-7.
- Peterson LJ, Ellin IE, Hupp JR, Tucker MR. *Contemporary Oral and Maxillofacial Surgery.* St. Louis: Mosby; 1998.
- Martin MV. Antibiotic treatment of cervicofacial actinomycosis for patients allergic to penicillin: a clinical and in vitro study. *Br J Oral Maxillofac Surg.* 1985; 23(6):428-34.

## Актиномикоза образа

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### КРАТАК САДРЖАЈ

**Увод** Актиномикоза је ретка хронична грануломатозна болест коју је први пут описао Болингер 1877. године. Болест је узрокована актиномицетама и одликује се спорим напретком, гнојном упалом, стварањем мултиплних апсцеса и фистула с могућим лучењем „сумпорних гранула“.

**Приказ болесника** Приказујемо необичан случај актиномикозе образа шест година након букалног одонтогеног апсцеса. Мушкарац стар 56 година упућен је у Завод за оралну хирургију због безболног отока у подручју левог образа, ко-

ја је започела три недеље пре упућивања у завод. Дијагноза актиномикозе потврђена је патохистолошким налазом. У складу са дијагнозом преписан је пеницилин током четири месеца, након чега је дошло до излечења.

**Закључак** Овај случај актиномикозе је ретка појава. За правилну дијагнозу потребни су пажљив преглед болесника и висок степен клиничке сумње.

**Кључне речи:** актиномикоза; букально; грануломатозна инфекција

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