

Cervicofacial actinomycosis following jaw pathology – Case Reports

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Abstract

Cervicofacial actinomycosis is a specific, primarily chronic inflammatory process caused by Gram-positive anaerobic, microaerophilic *Actinomyces* bacteria. The infection usually results from the skin or mucosal lesions. The following paper reports two cases of Cervicofacial actinomycosis in patients admitted in 2014 to Maxillofacial Surgery Department in Lublin, Poland. Diagnosis of culture-positive actinomycosis was based on histopathology findings and the presence of actinomyces colonies. Cervicofacial actinomycosis requires surgical treatment, oral cavity sanitation, and in the case of presenting actinomycosis abscesses formation, combined surgical incision and drainage. Long-term penicillin antibiotic therapy is necessary.

Key words

cervicofacial actinomycosis, actinomyces abscess, surgical treatment

INTRODUCTION

Cervicofacial actinomycosis is a specific, primarily chronic inflammatory process caused by Gram-positive anaerobic *Actinomyces* bacteria. The pathogenic *Actinomyces* species do not exist free in nature but are commensals and normal inhabitants of the oropharynx and the gastrointestinal and female genital tracts [1, 2, 3]. *Actinomyces* are soil saprophytes which lies dormant in the oral cavity (in the gingival pockets, carious defects or periapical lesions and palatine tonsils niches) and may become activated under certain circumstances. Actinomycetaceae encompasses a group of commensals, the most common is *A. israelii*, followed by *A. naeslundii*, *A. viscosus*, *A. meyeri*, *A. odontolyticus*, *Arachnia propionica* and *Bifido-bacterium dentium* [3, 4].

The development of cervicofacial actinomycosis results from impaired immunity accompanied by micro- or macro-traumas or persisting non-specific inflammation. Actinomycosis is classified according to the site of infection into [1, 3, 4]:

- orocervicofacial actinomycosis (33–65% of cases);
- abdominopelvic actinomycosis (15–20% of cases);
- thoracic actinomycosis (10–15% of cases).

Orocervicofacial actinomycosis occurs mainly in middle-aged patients, more frequently in men than women. It rarely occurs in children and adolescents [1, 3]. The most common sites of presentations include: perimandibular region, cervical, parotid-masseter or buccal. Orocericofacial actinomycosis develops in the majority of cases, and patients who suffer from the condition usually have poor oral hygiene and/or have undergone some invasive dental procedures or facial traumas [1, 2, 3, 5]. Actinomycosis is often misdiagnosed due to the fact that its clinical presenting features may be similar to various other conditions, including malignant neoplasms [3, 6]. Typical clinical manifestation is characterised by

subcutaneous tissue infiltration which becomes firm over time, and draining sinus tracts formation discharging pus containing actinomyces colonies, so-called 'sulphur granules' [2, 5]. The manifestations are characteristic but not pathognomonic to actinomycosis [4].

The diagnosis of actinomycosis is made on the basis of bacteriological and histopathological examination. The presence of actinomycetes in the purulent material is in the form of the 'sulfur granules' that appear as round or oval eosinophilic masses with basophilic terminal 'clubs' on haematoxylin-eosin stained sections. Sulfur granules are not specific for actinomycosis because they represent a localized immunologic response to an antigen-antibody precipitation occurring in infectious (e.g. fungi, parasites and bacteria) or other non-infectious processes (e.g. around inert material, in hypereosinophilic syndrome and allergic conjunctival granuloma) [3, 6, 7]. Direct isolation of actinomyces from a clinical specimen or from sulfur granules is necessary for a definitive diagnosis. However, the failure rate for isolation via anaerobic culture is high owing to previous antibiotic treatment, inadequate methodology, or overgrowth of the slow growing actinomyces by bystander organisms, such as *Bacteroides*, *Fusobacterium*, or *Aggregatibacter* [6, 8].

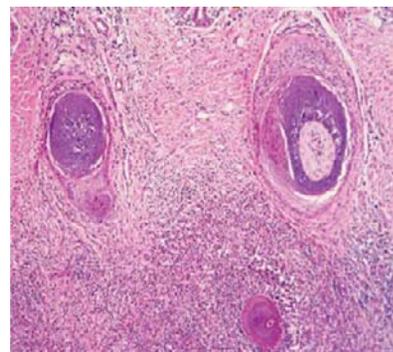


Figure 1. Histology showing actinomycosis sulfur granules with surrounding inflammation.
Source: Graham Library of Digital Images, Department of Dermatology, Wake Forest University, Winston-Salem (NC), USA

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The current study presents two cases of patients admitted to Maxillofacial Surgery Department at the Medical University in Lublin, Poland, who developed actinomycosis due to inflammatory lesions. In these case reports the authors used dental nomenclature of teeth numeration which consists of two numbers. The first number describes the quadrant of the oral cavity: 1 – upper right, 2 – upper left, 3 – lower left, 4 – lower right. The second number describes which tooth referred to, from 1 – 8, e.g. tooth 17 is first upper right molar and 48 is the third lower right molar.

CASE REPORT 1

A 41-year-old female patient was admitted to hospital due to inflammatory cheek and left parotid-masseter infiltration (Fig. 2, 3). The patient reported pain in the infiltrated region and had been feverish for a week. Antibiotic therapy was implemented and a panoramic radiograph ordered. The RTG showed a left mandibular primordial cyst with a retained tooth (38), as well as three other cysts, one cyst in the maxilla – in the region of teeth 21–23, and two cysts in the right mandibular region – region of teeth 43–47) (Fig. 4). Furthermore, poor oral hygiene – dental defects, numerous carious teeth and gangrenous roots, were found. Extraoral incision and drainage of the left cheek abscess were performed under local anaesthesia Lignocainum 2% cum Noradrenalino 0.00125% Polfa. A computed tomography scan was carried out to determine the precise location of the three cysts visible in the panoramic radiography (Fig. 5, 6). In the course of hospital treatment, an active purulent sinus tract appeared at the top of the inflammatory lesion in the region of the left parotid-masseter. Material for bacteriological and histopathological examination was collected. Mandible and maxilla cysts were removed and 17 teeth extracted under general anaesthesia. Histopathological examination confirmed actinomycosis; serologic investigations were not performed. The patient



Figure 2. Inflammatory infiltration in the left parotid-masseter in a 41-year-old female patient



Figure 3. Inflammatory infiltration in the left cheek of a 41-year-old female patient

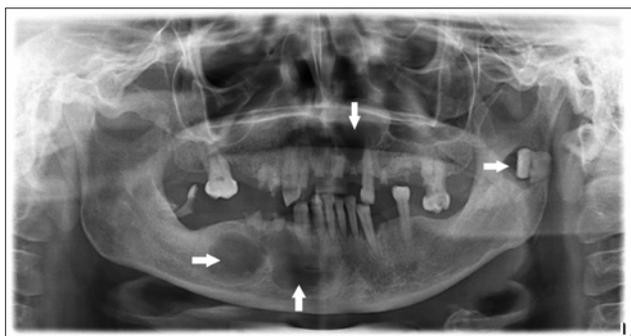


Figure 4. Panoramic radiography of a 41-year-old female patient which shows a left mandibula primordial cyst with retained tooth 38, cyst in the maxilla (region of teeth 21–23), and two cysts in the right mandibula (region of teeth 43–47)



Figure 5. Axial CT scan presenting a cyst in the maxilla in the region of teeth 21–23

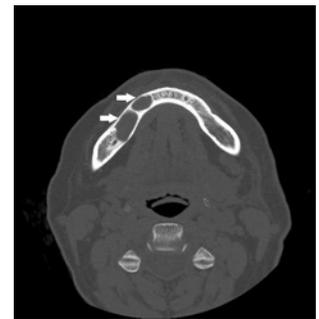


Figure 6. Axial CT scan presenting two cysts in the mandibula

was treated with amoxicillin-clavulanic acid for 3 months which produced complete clearance of the orocervicofacial actinomycosis. After the hospital treatment, the patient reported for regular health checks which did not reveal any complications.

CASE REPORT 2

A 19-year-old male patient was admitted to hospital due to twice occurring inflammations of the right mandibula angle region. The first inflammatory episode occurred two months earlier and dental extraction of tooth 48 was performed. After approximately 6 weeks recurrent inflammation of the same region, a purulent skin sinus tract appeared. Panoramic radiograph revealed an osteolytic lesion in the region of the right maxilla angle (Fig. 7). Intraoral incision of the abscess and drainage were performed, intravenous antibiotic treatment with Taromentin and Metronidazole was administered. Computed tomography (CT scan) revealed osseous restructure with periosteum detachment in the region of the right maxilla angle (Fig. 8). The patient was classified to revision of the above-mentioned region under general anaesthesia. Sequestrotomy of the right maxilla angle was performed and some material for bacteriological and histopathological examination was collected. Local and general condition improved significantly during the following days of hospitalization. Histopathological examination confirmed actinomycosis. The patient was discharged in good condition, and continuation of antibiotic therapy with Ospen (Phenoxymethylpenicillinum) 1.5 mln i.u. 3 × daily for 3 months was recommended. The patient reported for control health checks which confirmed the efficacy of the treatment.



Figure 7. Panoramic radiograph showing osteolytic lesion in the region of the right maxilla angle in a 19-year-old male patient.



Figure 8. Axial CT scan indicating osseous restructure with periosteum detachment.

DISCUSSION

Actinomyces were originally classified as fungi. These bacteria may be present in the oral cavity of healthy people and development of the disease requires tissue integrity disruption [2]. Actinomycosis can have various clinical presenting features. In the presented study it manifested in Case 1 as inflammatory skin, subcutaneous tissue infiltration with a purulent maxilla, and in Case 2 as mandibular cysts and mandible inflammation to form the bone sequestrum. The disease took the form of a hard brownish mass with purulent fistulas, especially in the mandibular angle area.

Kolm describes a woody cheek infiltration with three red-bluish nodules discharging purulent material. A skin biopsy was performed which showed a deep inflammatory infiltrate and epithelial lamellas, which suggested the diagnosis of a ruptured epidermoid cyst [6]. Oro-cervicofacial actinomycosis is not a common disease process, and develops within specific oxidoreductive parameters that characterise a particular ecosystem. Absolute identification is performed by anaerobic culture, biochemical tests and gas chromatography. Unfortunately, the microorganism is delicate and sensitive to oxygen. *Actinomyces* is identified in culture in fewer than 50% [7, 8, 9, 10]. It usually results from trauma, surgery to the oral mucosa or dental manipulations. It can also develop in an erupted third molar. Some authors note that many cases of the disease are seen in patients who have an unfavourable socio-economic background with poor oral hygiene [11].

Radiographic examination, both basic and advanced, does not allow a specific diagnosis due to the lack of pathognomonic signs and symptoms. However, it is useful to determine the exact location and extent of the pathologic process [3, 8]. Radiographic examination also determines the type and scope of surgical treatment. In Case 1 in the presented study, the maxilla cysts were enucleated and maxilla sequestrotomy was performed.

The basic treatment of actinomycosis is by antibiotic therapy, with penicillin and its derivatives as the drugs of choice [2, 5, 6, 8, 9, 10]. A possible scheme for the antibiotic treatment of oro-cervicofacial actinomycosis may consist of amoxicillin plus clavulanic acid, or possibly ampicillin plus sulbactam. The initial dose is 3 × 2 g amoxicillin plus 3 × 0.2 g clavulanic acid per day for 1 week, and 3 × 1 g amoxicillin plus 3 × 0.1 g clavulanic acid per day for another week.

Chronic cervicofacial infections may require up to 4 weeks of treatment [9]. In the case of allergy to penicillin, clindamycin, erythromycin, clarithromycin and metronidazole are used [3, 9]. The use of fluoroquinolones and tetracyclines, however, is not advised [3, 9, 10].

The duration of the antibiotic therapy depends on the progression of the disease and the patient's response to the treatment [1, 8, 11]. The antibiotic regimen should initially be administered intravenously until clinical improvement is demonstrated, followed by oral administration over 2–12 months [1, 6]. According to Wong, the traditional recommendation of 6–12 months may not be necessary in all patients, because oro-cervicofacial disease has been cured after short courses of 2–6 weeks of antibiotic therapy (oral and intravenous), combined with surgical drainage [3].

Surgical management is indicated in the case of tissue necrosis, sinus tract formation, and in patients with abscesses which require incision and drainage, as well as in cicatrix removal [3, 6].

Prognosis for oro-cervicofacial actinomycosis is good and complete recovery is achieved in 90% of patients [1].

CONCLUSIONS

Actinomycosis is a specific, primarily chronic inflammatory process which is difficult to diagnose in its early stages due to non-typical clinical presenting features. Final diagnosis is made on the basis of histopathological examination. Oro-cervicofacial actinomycosis requires surgical treatment, oral cavity sanitation, in the case of radiating abscesses formation, surgical incision and drainage. It is important to complete treatment with long-term antibiotic therapy. Prognosis is favourable in the majority of cases.

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