

Actinomycosis of the tongue – a literature review

Promienica języka – przegląd piśmiennictwa

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Słowa kluczowe: promienica języka, promienica u dziecka, promienica twarzowo-szyjna.

Abstract

Actinomycosis is a specific chronic inflammation caused by anaerobic or microaerophilic gram positive bacteria from the Actinomyces family. They form a natural part of the bacterial flora found in the mouth, digestive and respiratory tract. The bacteria mentioned above can cause actinomycosis after previous damage to the natural anatomical barrier, which is skin or mucous membrane. This inflammation occurs especially in states of reduced immunity. Actinomycosis occurs in three clinical forms: cervico-facial, pulmonary or pulmono-thoracic and abdomino-pelvic. Actinomycosis of the tongue is a rare form of infection. To date, incidental occurrence of actinomycosis at this location has been reported in the literature. In the available literature we have found 21 cases of actinomycosis described so far. The authors present their own effective diagnostics and treatment of actinomycosis and review the literature on this topic.

Streszczenie

Promienica jest swoistym, przewlekłym zapaleniem wywołanym przez beztlenowe lub mikroaerofilowe bakterie Gram-dodatnie z rodziny *Actinomyces*. Stanowią one naturalną część flory bakteryjnej występującej w jamie ustnej, przewodzie pokarmowym i oddechowym. Wspomniane wyżej bakterie mogą wywołać aktinomykozę po wcześniejszym uszkodzeniu naturalnej bariery anatomicznej, jaką jest skóra bądź błona śluzowa. Ten stan zapalny pojawia się szczególnie u pacjentów z obniżoną odpornością. Aktinomykoza występuje w trzech postaciach klinicznych: szyjno-twarzowej, płucnej lub płucno-piersiowej i brzuszno-miednicznej. Promienica języka jest rzadką postacią infekcji. Do tej pory odnotowano rzadkie występowanie promienicy w tej lokalizacji. W dostępnym piśmiennictwie znaleźliśmy 21 przypadków promienicy języka. Autorzy przedstawiają własną, skuteczną diagnostykę oraz leczenie promienicy języka u dziecka oraz dokonują przeglądu literatury na ten temat.

Actinomycosis is a specific chronic inflammation caused by anaerobic or microaerophilic Gram-positive bacteria from the actinomyces family. Bacteria from the actinomyces species are usually used for pathogenicity. They form a natural part of the bacterial flora found in the mouth, and digestive and respiratory tract. The bacteria mentioned above can cause actinomycosis after previous damage to the natural anatomical barrier, i.e. skin or mucous membrane. This inflammation occurs especially in states of reduced immunity. Of the main species (*A. israeli*, *naeslundii*, *viscosus*, *odontolyticus*, *meyeri*, and *pyogenes*), *Actinomyces israeli* is the most common pathogen causing radiant infection. Actinomycosis occurs in three clinical forms: cervico-facial, pulmonary or pulmono-thoracic, and abdomino-pelvic. The most frequently type is (in as much as about

50% of cases) the first type [1]. Cervico-facial actinomycosis is the most common actinomycosis, responsible for 50–70% of cases of disease. It usually develops as a complication of oral procedures or in people who do not comply with oral hygiene. The cervico-facial actinomycosis is most often located in the submandibular, parotido-maseteric region and neck. Oral involvement is considered rare. Hence, the location in the tongue region file may mimic another pathological process including neoplastic. In the initial stage of the disease, painless swelling of the soft tissues of the mandible is usually used, treatments are available that take into account the use of shuffling. The skin in the fistula area is usually red or bruised. As the disease progresses, the affected skin hardens. The evolution of the clinical picture from the infiltrative to purulent

phase with the formation of abscesses and fistulas and skin involvement is characteristic of actinomycosis. In contrast, the nodular form progresses imitates tumour growth, which makes it difficult to recognise the actinomycosis. In addition, unlike other inflammatory processes, actinomycosis does not cause severe general symptoms indicative of an inflammatory process. We rarely observe fever leukocytosis or an increase in laboratory symptoms of inflammation. An increase in lymph nodes is usually not observed. Thoracic actinomycosis accounts for 15–20% of cases of the disease. Usually infection occurs as a result of aspiration into the respiratory tree of contents from the mouth and throat containing actinomycetes. People with epilepsy and alcoholics are predisposed. Occasionally, oesophageal perforation leads to infection, or inflammation is transmitted through the abdomen or neck. This form of actinomycosis usually occurs in the form of an inflammatory lung infiltrate, which, if not treated, can affect the pleura, pericardium, and even the chest wall. Symptoms are coughing, dry or wet, shortness of breath, chest pain, and fever. The abdomines and pelvis actinomycosis constitute 10–20% of cases of the disease. It usually develops in people who have undergone bowel surgery, especially because of diseases that occur when the bowel is broken, including post appendix or diverticulum perforation or after foreign body aspiration. The disease usually proceeds as a slowly growing inflammatory tumour, often located in the left iliac region with fever, change in bowel habits, abdominal pain, nausea, and vomiting. The starting point of the abdominal actinomycosis are usually the intrauterine contraceptive devices. Diagnosis of actinomycosis is based on clinical picture, additional tests, direct examination of fistula secretions to visualise bacteria, culture of the secretions in anaerobic conditions, and granularity examination to determine the characteristics of actinomycetes in the

microscopic image. Imaging studies and histopathological examination of the tissues involved are also helpful. Treatment of actinomycosis involves the use of antibiotics, usually for a period of 6 months to one year. In some cases, surgical treatment is necessary: incision and drainage of abscesses or fistula removal [2]. The authors present a very rare case of actinomycosis in a 11-year-old boy.

Child was admitted to the ward due to non-healing thickening of the tongue on the left side for about three months. A painless lesion was found: a small epithelial defect in the central part without purulent filtration, and without limited mobility of the tongue. The diameter of the lesion was 2 × 1 cm, and blood laboratory tests did not reveal significant deviations from normal. Blood test marker for inflammation of C-reactive protein (CRP) < 1 mg/l, OB 9 mm/h, white blood cells (WBC) 5.1 thousand/ μ l. Ultrasound of neck lymph nodes revealed no enlarged lymph nodes. The lesion had been treated with several antibiotics by paediatric doctors without significant improvement. A swab test taken on an outpatient basis showed no signs of bacterial or fungal growth. Due to the very unclear clinical picture suggesting tumour growth, excision biopsy was decided. The procedure was performed under general anaesthesia. The postoperative course was without complications. On day 7 the sutures were removed, and the wound healed correctly. Results of a histopathological examination showed a fragment of the tongue, partly covered with flat multilayer epithelium, with the presence of sub-epithelial inflammatory infiltrate with numerous *Actinomyces* colonies (GMS +). Due to the histopathological results obtained, penicillin was orally administered at a dose of 1.5 million IU every eight hours for a period of 6 weeks. A child without complaints without signs of the inflammatory process within the tongue (Figures 1 A, 2).

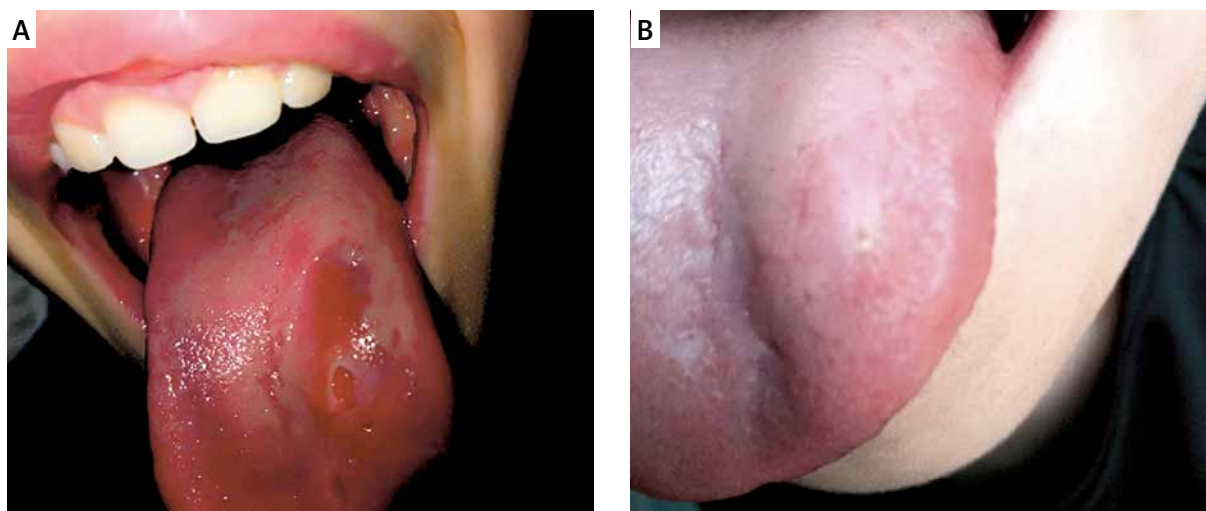


Figure 1. Tongue actinomycosis: **A** – before treatment, **B** – after treatment

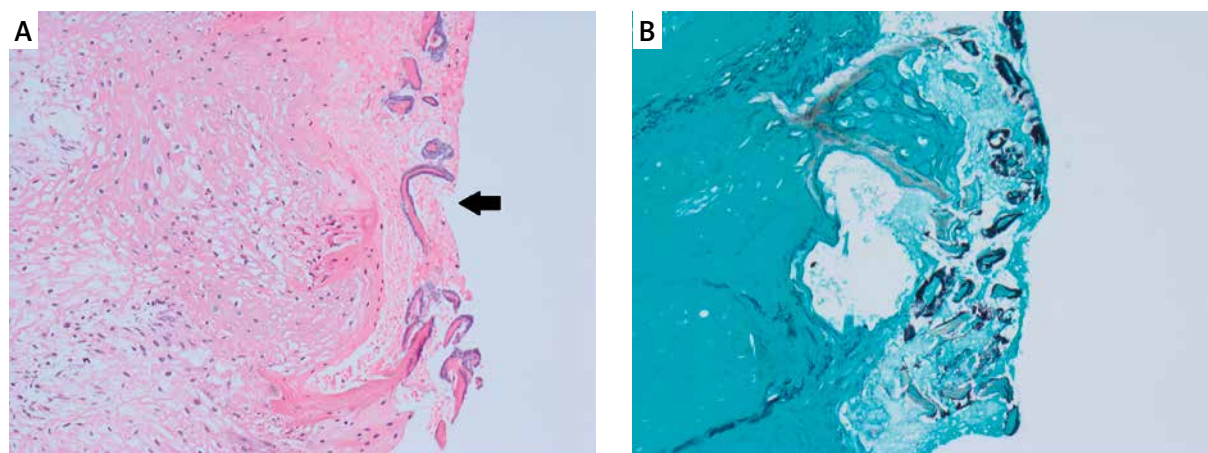


Figure 2. Confirmation of actinomycosis in microscopic examination. **A** – Haematoxylin and eosin staining. Magnification 100×. **B** – Staining GMS. Magnification 100×

Actinomycosis of the tongue is a rare form of infection. To date, incidental occurrence of actinomycosis at this location has been reported in the literature (Table 1 [3–23]). Actinomycosis of the tongue usually occurs on its lateral surface because it is the area most susceptible to injury. The uniqueness of the case presented by us also results from the very young age of the patient. Actinomycosis is more common in middle-aged men and is often associated with earlier discontinuation of the skin or mucosa membrane or co-occurrence of other inflammatory diseases. As a possible factor inducing the occurrence of actinomycosis in children, an earlier injury in the form of cutting or biting is suggested [2]. Although in the presented case there is no identification of the cause of inflammation, in our opinion it is important for the diagnostic process and subsequent treatment to indicate possible factors predisposing to development of actinomycosis in children. These include mechanical damage to the mucosa, poor oral hygiene, tooth replacement, parafunctions associated with inserting objects into the mouth, and recurrent acute inflammation of the mouth, palatine tonsils, and throat and associated immunosuppression. As a result of infection and disruption of the mucous membrane, actinomyces penetrate into the tissues of the tongue. Slow growth and no discomfort cause the lesion to grow. The histological structure of the tongue and its location favour the development of the tuberous form of actinomycosis. The rarity of actinomycosis within the tongue, its nodular form imitating tumour growth, and the absence of other systemic and local signs of inflammation make it difficult to recognise and treat. Due to the atypical picture, antibiotic therapy is usually included, which, when used for too short a time, causes diagnostic difficulties and finally confirms or excludes initial diagnosis. Collected swabs in 50–76% [2] are sterile, as in our case. Usually, surgical intervention and collection of material for histopatholog-

ical examination results in a diagnosis and, accordingly, proper treatment. Based on the presented case and literature analysis, it should be stated that the dominant form of actinomycosis within the tongue is a nodular form with oedema, enlargement of the tongue in the form of limited nodular hyperplasia or generalised macroglossia. The clinical diagnosis of actinomycosis may pose difficulties due to the often nonspecific symptoms, similarity to other diseases, and lack of awareness due to its rare occurrence. Therefore, actinomycosis should be differentiated from tuberculosis, mycobacterial atypical infection, and above all tumour growth, fungal infection, subcutaneous tissue inflammation, or nocardiosis. In our opinion, the final diagnosis should always be based on histopathological examination. In the fight against the disease, the treatment of choice is the use of high doses of penicillin for up to several months. Incorrect, overly short-duration pharmacotherapy can cause the disease to recur. In turn, clindamycin is recommended for patients allergic to penicillin; other possibilities include the use of erythromycin, tetracycline, streptomycin, lincomycin, or doxycycline. In the case of abscess formation, surgical treatment is also necessary, consisting of the incision and drainage of oozing lesions. Actinomycosis, even in people with normal immunity, requires intensive and long-term (up to 12 months) antibiotic treatment (penicillin), sometimes in combination with surgical management. Based on the presented case and analysis of the literature, the recommended treatment method for the actinomycosis of the tongue in the form of a tumour is intensive antibiotic therapy and surgical treatment consisting of resection. A more severe course and disseminated form of *Actinomyces* infection is observed in the case of reduced immunity: in persons after organ transplantation, chronically treated with corticosteroids, patients with lymphoproliferative hyperplasia, or infected with HIV. Disseminated infection

Table 1. Studies reporting cases of actinomycosis of the tongue

Studies	Age	Sex	Symptoms	Treatment
Our case	11	Male	Mass	Excision and penicillin
D'Amore <i>et al.</i> 2020 [3]	52	Male	Swelling	Clarithromycin
Ahmed <i>et al.</i> 2018 [4]	60	Female	Swelling	Amoxicillin
Sadeghi <i>et al.</i> 2019 [5]	66	Male	Macroglossia	Penicillin
Aneja <i>et al.</i> 2017 [6]	14	Male	Mass	Excision and amoxicillin-clavulanic acid
Jat <i>et al.</i> 2017 [7]	44	Female	Mass	Excision and doxycycline
Rocha <i>et al.</i> 2017 [8]	44	Female	Necrotic tissue	Local debridement
Escoda <i>et al.</i> 2013 [9]			Two ulcerated lesions	Clindamycin
Kurtaran <i>et al.</i> 2011 [10]	54	Female	Mass	Excision and combination of amoxicillin-clavulanic acid and metronidazole
Habibi <i>et al.</i> 2008 [11]	54	Female	Mass	Excision and intravenous penicillin
Enoz 2007 [12]	39	Female	Nodular lesion	
Atespare <i>et al.</i> 2006 [13]	52	Female	Mass	Excision and amoxicillin-clavulanic acid
Alamillos-Granados <i>et al.</i> 2000 [14]	74	Female	Painless indurated ulcer	Minocycline
Vasquez <i>et al.</i> 1997 [15]	31	Man	Nodules	Amoxicillin
Gerbino <i>et al.</i> 1998 [16]	No data	No data	Nodules	Penicillin
Ficarra <i>et al.</i> 1993 [17]	57	Female	Mass	Penicillin
Morris <i>et al.</i> 1992 [18]	50	Male	Nodular lesion	No data
Isalska <i>et al.</i> 1991 [19]	64	Female	Swelling	Amoxycillin
Brignall and Gilhooly 1989 [20]	55	Male	Swelling	Penicillin
Kuepper and Harrigan 1979 [21]	35	Male	Mass	Penicillin
Uhler and Dolan 1972 [22]	43	Male	Mass	Excision and penicillin
Sodagar and Kohout 1972 [23]	47	Female	Mass	Excision

may include the lungs, skin, central nervous system, and muscular system.

In our opinion, due to the rare occurrence of actinomycosis of the tongue, uncharacteristic clinical picture, often lack of actinomycosis growth in bacteriological smears, earlier empirical inclusion of antibiotic in our opinion, excision biopsy is the basic source that can determine the diagnosis of actinomycosis and for most all exclude the diagnosis of tumour growth.

Conflict of interest

The authors declare no conflict of interest.

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