

Actinomycosis in Differential Diagnosis of Cervicofacial Mass: A Case Report

Servikofasiyal Kitlenin Ayırıcı Tanısında Aktinomikozis: Olgu Sunumu

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Summary

Actinomycosis is a rare soft tissue infection caused by Gram-positive, anaerobic bacteria. It is rarely seen in childhood. However, most of the cases are misdiagnosed. Thus, the true prevalence cannot be estimated. The major clinical forms of actinomycosis are; cervicofacial, thoracic, abdominal and pelvic forms. The most frequent clinical form is cervicofacial actinomycosis. Diagnosis of disease and differential diagnosis from numerous infectious and noninfectious diseases are very difficult. Cervicofacial actinomycosis is not commonly thought of in the first step of the differential diagnosis of cervicofacial mass. We presented a 9-year old boy diagnosed with cervicofacial actinomycosis. (*J Pediatr Inf* 2009; 3: 28-30)

Key words: Cervicofascial mass, actinomycosis, child

Özet

Aktinomikozis, gram pozitif anaerobik bakterinin neden olduğu, ender görülen bir yumuşak doku enfeksiyonudur. Bu enfeksiyon hastalığı çocukluk çağında oldukça ender görülür. Ancak, olguların çoğunluğunun yanlış tanı alması nedeni ile gerçek prevalansı bilinmemektedir. Aktinomikoz enfeksiyonunun başlıca klinik formları, servikofasiyal, torasik, abdominal ve pelvik aktinomikozistir. Servikofasiyal aktinomikozis hastalığının en sık görülen klinik formudur. Gerek hastalığın tanısı, gerekse çeşitli enfeksiyon ya da enfeksiyon dışı hastalıklardan ayırıcı tanısı oldukça güçtür. Özellikle servikofasiyal aktinomikozis, servikofasiyal kitlelerin ayırıcı tanısında düşünülmemektedir. Bu yazıda, servikofasiyal aktinomikozis tanılı 9 yaşındaki erkek olgu sunuldu.

(*Çocuk Enf Derg* 2009; 3: 28-30)

Anahtar kelimeler: Servikofasiyal kitle, aktinomikozis, çocuk

Introduction

Actinomycosis is an uncommon suppurative and granulomatous chronic infectious disease. It is caused by Gram-positive, pleomorphic non-spore-forming, non-acid-fast anaerobic or microaerophilic bacilli of the genus *Actinomyces*. In humans, actinomyces are often normally found in the oral cavity, the gastrointestinal tract and the female genital tract (1). Numerous clinical manifestations have been described. The most frequent clinical form is cervicofacial actinomycosis (2).

This disease is infrequent in children. However, the diagnosis of actinomycosis is usually under-recognized, so the real prevalence must be higher. In additionally, diagnosis of disease and dif-

fertial diagnosis from numerous infectious and noninfectious diseases are very difficult. Therefore actinomycosis should be included in the differential diagnosis of a soft tissue mass of the cervicofacial area.

Case Report

A 9-year-old boy, presented with a six months history of an enlarging left submandibular mass. He denied fever, chills, weight loss and other constitutional symptoms. The patient had been evaluated due to cervicofacial mass in the plastic and reconstructive surgery clinic. Physical examination had revealed a very hard, fluctuant and painless left submandibular mass measuring

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3x3.5 cm in diameters. There had been no skin drainage at his initial visit. The lesion had been excised due to differential diagnosis of cervicofacial mass, especially malignant neoplasm, under general anesthesia.

Histological examination showed actinomycotic granules located in the abscesses formation with suppuration background. Granules were characterized with round-oval masses with basophilic appearance and a finely eosinophilic border (Fig. 1). Histochemically, actinomycete filaments were colored deep bluish-purple with tissue Gram stain and deep Brown with Gomori Methenamine silver stain (Fig. 2).

The patient was referred to our Department of Pediatric Infectious Disease. The patient denied any history of facial trauma or recent dental extraction. In the physical examination, postoperative scar tissue measuring 2cm the in left lower border of the mandible was observed. The oral examination and the remainder of the head and neck ex-

amination were normal. The patient was afebrile. He had no cervical and other lymphadenopathy. Organomegaly was not observed. His teeth were vital, and there were no clinical signs of dental infection. Chest x-ray and PPD measurement were normal. The laboratory evaluation was entirely normal. None residual or additional mass due to actinomycosis were detected by computed tomography (CT) scan of head, neck and paranasal sinus tract. Abdominal ultrasonographic evaluation that performed due to evaluation of abdominal actinomycosis was normal.

The patient with underwent surgical excision of lesion to distinguish the other possible diagnosis. In conclusion, he was diagnosed as cervicofacial actinomycosis by clinical signs and histopatological examination. The abdominal and thoracic types of disease were not detected. Amoxicillin treatment was administered for 12 weeks.

Discussion

Actinomycosis is an infectious disease with a worldwide distribution caused by gram-positive, pleomorphic non-spore-forming, non-acid-fast anaerobic or microaerophilic bacilli of the genus *Actinomyces*. The disease was known in cattle as early as the beginning of the 19th century (3). At present lowering of the incidence is related to widespread use of antibiotics, because *Actinomyces* is sensitive to many antibiotics. *Actinomyces* are in the normal flora of the gastrointestinal tract and mouth. These microorganisms are usually non-virulent in nature, however an interruption of the protective mucosal barrier, and alteration of the resident microbial flora play a crucial role in infection (4). It is mostly found in adults, women are less frequently affected than men. Infection is very rare in infants and children. When found in children, it rarely spreads beyond superficial cervicofacial lesions (2).

Actinomycosis is often difficult to diagnose as it can mimic numerous infectious such as tuberculosis or a fungal infection and noninfectious diseases such as malignant neoplasm of cervicofacial area (5). A neoplasm may also result in an enhancing solid mass. Cervical infection usually presents with matted cervical lymphadenopathy. The absence of lymphadenopathy may be beneficial evidence in differentiating cervicofacial actinomycosis from a malignancy. The nodal characteristics and lymph node features may be helpful in differentiating cervicofacial actinomycosis from other infectious and noninfectious disease in cervicofacial area. Besides the organism causing actinomycosis usually does not spread by way of the lymphatic system due to the size of the bacterium. Thus, the regional lymphadenopathy is uncommon (6). In our case, regional lymphadenopathy was not detected and the malignant neoplasm had excluded by histopathological investigation.

Numerous clinical manifestations of the disease have been defined. Cervicofacial, thoracic and the abdominal parts of the body are most commonly affected, other liab-



Figure 1. Actinomycotic granule in an abscess. HE x 100

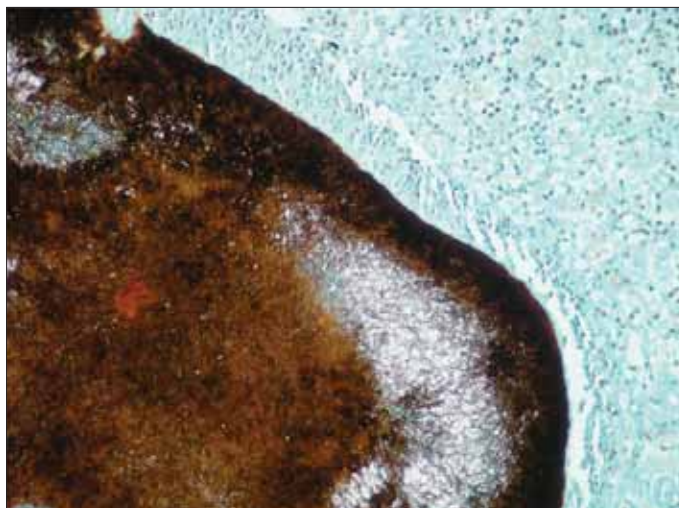


Figure 2. Delicate filaments are visible especially in the right part of the granule. Gomori Methenamine Silver stain x 2000

le regions of infection are the extremities, skin, brain, lacrimal glands, kidneys, genital organs and bones. The most frequent clinical form is cervicofacial actinomycosis (55%) (7). Presentation of cervicofacial form is variable. It can develop in tongue, larynx, hypopharynx, lacrimal gland, mandible, scalp, paranasal sinus, palate, parotid gland. If the infection extends to the facial and maxillary bones, periostitis or osteomyelitis may develop. Cervicofacial actinomycosis presents in two distinct pattern; "lumpy jam", which is a slowly enlarging, fluctuant painless swelling over the lower border of the mandible, or a greatly spread infections that involves the submandibular area. Both forms spread very slowly. (8). Our patient showed the lumpy jam pattern. The other possible infection regions of cervicofacial form and osteomyelitis or periostitis were excluded in our patient by CT scan of head, neck and paranasal sinus tract. Indeed, the other forms of disease were excluded by clinic and radiological examination.

Mucous contusion or abrasion, sinusitis, tooth extraction, oral and maxillofacial surgery, maxillofacial trauma, dental pulp exposure and endodontic treatment, periapical infection or granuloma, periodontal disease, local anesthesia, tooth eruption, poor hygiene and tonsillectomy are important risk factors of cervicofacial actinomycosis (2). In our case, these risk factors were excluded and he evaluated as idiopathic.

Actinomycosis is diagnosed by examining the exudate and infected tissue. Gram staining reveals gram positive long-branching filaments. The biopsy specimen of an actinomycetic infection shows a central neutrophilic lobulated abscess that contains a number of granules surrounded by granulation tissue. (6,9). However, Actinomyces are difficult to grow even in enriched media and the diagnosis is confirmed by culture in less than 50% (3). In such cases, the diagnosis is based on the morphology of granules and bacteria or on direct examination of granules (3). When the often ambiguous clinical presentation and the possibility of neoplasia are taken into consideration, actinomycosis is seen to be a pathologic condition. Actinomycosis is the initial diagnosis in fewer than 10% of documented cases (2). Therefore, incisional biopsy is often undertaken to determine a diagnosis (6). Our patient with underwent surgical excision of lesion due to differential diagnosis of cervicofacial mass was diagnosed cervicofacial actinomycosis by histopatological examination.

This infectious disease was a relatively frequent and often fatal disease before the widespread availability of antibiotics (10). Harvey et al. reported a 62% mortality rate prior to the antibiotic era (11). Conventional therapy for actinomycosis is high-dose intravenous penicillin G for 4-6

weeks, followed by oral penicillin, amoxicillin, erythromycin, clindamycin, doxycycline or tetracycline for a period of 6-12 months (12). Selvin et al. described cases of esophageal and of cervicofacial actinomycosis treated successfully with short-term antibiotic therapy. Indeed, they pointed out that cervicofacial actinomycosis is especially responsive to brief courses of antibiotic treatment. (4). Surgical intervention may be necessary adjunct to antibiotic treatment. Complete recovery is expected in 90% of patients with cervicofacial actinomycosis. In our case, the amoxicillin treatment was started for 12 weeks after the excision of mass.

Actinomycosis is a very rare infectious disease in children. Due to the opportunistic characteristics of the actinomycotic infection, early differential diagnosis of actinomycosis is very important. Cervicofacial actinomycosis can present a diagnostic difficult situation for the clinician due to its capability to mimicked as other infectious and neoplastic disease. In the other word, cervicofacial actinomycosis is still a difficult diagnosis. In conclusion, any soft tissue mass or swelling on the cervicofacial area should be investigated for cervicofacial actinomycosis.

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