Actinomycosis in Differential Diagnosis of Cervicofacial Mass: A Case Report

Servikofasiyal Kitlenin Ayıncı 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: Cervicofacial mass, actinomycosis, child

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 underestimated, so the real prevalence must be higher. In additionally, diagnosis of disease and differential 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...
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 examination 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 histopathological 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-
le regions of infection are the extremities, skin, brain, lac-
rimal 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 swel-
ling over the lower border of the mandibule, 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 extrac-
tion, oral and maxillofacial surgery, maxillofacial trauma,
dental pulp exposure and endodontic treatment, periapi-
cal infection or granuloma, periodontal disease, local
anesthesia, tooth eruption, poor hygiene and tonsillec-
tomy are important risk factors cervicofacial acti-
nomycosis (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 acti-
nomycetic 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 sur-
gical excision of lesion due to differential diagnosis of
cervicofacial mass was diagnosed cervicofacial actinomyc-
osis by histopathological 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, erythromy-
cin, clindamycin, doxycycline or tetracycline for a period
of 6-12 months (12). Selvin et al. described cases of esop-
hageal and of cervicofacial actinomycosis treated suc-
cessfully 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 antibio-
tic 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 child-
ren. Due to the opportunistic characteristics of the acti-
nomycotic infection, early differential diagnosis of acti-
nomycosis 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 acti-
nomycosis is still a difficult diagnosis. In conclusion, any
soft tissue mass or swelling on the cervicofacial area sho-
uld be investigated for cervicofacial actinomycosis.

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