

CASE REPORT

Cervicofacial actinomycosis in a middle aged Saudi man

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ABSTRACT

Actinomycosis is a chronic rare disease, caused by Actinomyces species.

It has a broad spectrum of clinical presentations mimicking malignancy, tuberculosis and nocardiosis. Therefore the physician should have a high index of suspicion.

The most common presentation of actinomycosis is the cervicofacial form.

Here we describe the first case of lumpy jaw syndrome to be reported in Saudi Arabia.

INTRODUCTION

Actinomycosis is a rare chronic bacterial disease with cervicofacial actinomycosis being the most frequent clinical form, and the most common clinical manifestation is “lumpy jaw syndrome”, which is associated with odontogenic infection, representing approximately 60% of all reported cases.^{1,2,3,4} Other clinical presentations usually involve the thoracic, abdominopelvic regions, and the CNS.²

Cervicofacial actinomycosis is a relatively rare condition worldwide. There is no predilection for age, race, season, or occupation.⁵

It is considered an endogenous infection that requires a breach in the mucous membrane to become pathogenic.^{1,2,3}

It is important to have a high index of suspicion of this entity as it can mimic malignancy, tuberculosis, or nocardiosis.^{1,2,3} Physicians should also be aware of the diversity of presentations of actinomycosis.

To our knowledge, this is the first case of lumpy jaw syndrome to be reported in Saudi Arabia.

CASE

48 year old Saudi gentleman, who has no known medical problems, presented with a painless mass over his right jaw line for 1 month. The lesion has been increasing in size. He had history of a dental procedure 2 months prior to the development of this mass, and was given a course of prophylactic antibiotics. There is no history of trauma, fever, weight loss or loss of appetite.

On clinical examination, the patient had a 4 by 4 cm indurated non tender erythematous brown round tumor over the right mandible, which was attached to underlying structures, with no overlying skin changes. There were no draining sinuses or discharge (Fig. 1).

There was no cervical lymphadenopathy. On oral examination, there were no draining sinuses

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Fig. 1 Single subcutaneous indurated erythematous tumor over right mandible.



Fig. 2 Upon biopsy, there was white granules and purulent discharge.

and patient had acceptable oral hygiene. When we biopsied the mass with punch biopsy there was purulent discharge with white granules (Fig. 2). A 4 mm punch biopsy with crushed granules was sent to histopathology, and for bacterial culture.

RESULTS

Histopathology showed: deep dermal and subcutaneous inflammation (hematoxylin and eosin, original magnification x 40) (Fig. 3)

Culture showed: no growth, most likely because patient was on frequent courses of antibiotics.

GMS stain: shows multiple filamentous bacteria (GMS, original magnification x 40) (Fig. 4)

Which supports the diagnosis of actinomycosis. Patient was given 1 gram of amoxicillin/ clavulanate twice daily for 1 month, and follow up with his dentist.

Upon follow up, the patient had excellent improvement.

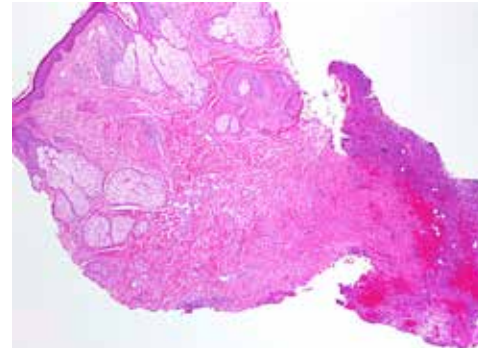


Fig. 3 Deep dermal and subcutaneous inflammation.(hematoxylin and eosin, original magnification x 40).



Fig. 4 GMS stain: shows multiple filamentous bacteria(GMS, original magnification x 40).

DISCUSSION

Actinomyces spp. are filamentous branching anaerobic Gram-positive bacilli which are normal commensal inhabitants of the oral and buccal cavities, gastrointestinal and urogenital tract. Actinomycosis is caused by the Actinomyces genus, with Actinomyces israelii being the most prevalent species isolated in human infections, reaching almost 70% of cases, and is found in most clinical forms of actinomycosis.^{1,2,3}

Other species have also been described, such as *A. gerencseriae*, *A. meyeri*, *A. odontolyticus*, *A. naeslundii*, *Actinomycesgeorgiae*, *Actinomycespyogenes*, or *A. viscosus*.⁶

Cervicofacial actinomycosis typically presents as a slowly enlarging painless subcutaneous indurated mass, which may evolve into multiple draining abscesses with sinus tracts opening on the skin or oral cavity, with a thick yellow exudate with characteristic sulfur granules.^{1,2,3}

Cervicofacial actinomycosis usually involves tissues surrounding the upper (maxillary expansion of the jaw) or lower mandible, including the mandible itself in approximately 50% of cases, cheek (15%), chin (15%), and submaxillary ramus and angle (10%). More rarely, the mandibular joint could be involved.⁵ Acute suppurative forms with rapid abscess formation are less common and are usually febrile and painful.⁵

It is important to have a high index of suspicion of this entity as it mimics malignancy, tuberculosis, or nocardiosis.^{1,2,3}

Because *Actinomyces* species usually do not spread lymphatically as a result of the size of the organism, actinomycosis is rarely associated with regional lymphadenopathy.^{7,8,9} Thus, in distinguishing actinomycosis from a malignancy, the diagnosis of actinomycosis should be considered if there is a mass with a few reactive lymph nodes or no lymph nodes involved.^{7,8,9}

Furthermore, it can disseminate to other organs such as lung, gastrointestinal tract and brain.⁵

The thoracic form of the disease accounts for 15%-30% of the cases of actinomycosis, which presents as bronchopulmonary infection, pleural, chest wall, or mediastinal involvement.¹⁰

Pulmonary actinomycosis probably results from aspiration of oropharyngeal or gastrointestinal secretions into the respiratory tract.¹¹ It also may extend to the mediastinum and chest wall across anatomic barriers, such as fissures and pleura.³

In central nervous system infection, there is usually hematogenous spread from a distant primary site such as the lung, oral cavity or pelvis.¹² Although, direct extension from face, jaw or paranasal sinuses through connective tissue planes or base of skull foramina can occur.¹³

Therefore, physicians should also be aware of the diversity of presentations of actinomycosis. Actinomycotic infection should be considered in the following clinical situations: (a) a series of manifestations including chronicity, extensive propagation across tissue planes, and firm to hard mass-like features, which are frequently confused with malignant disease; (b) drainage of an abscess by a sinus tract, which may spontaneously close and reform elsewhere; and (c) temporary improvement after a short course of antibiotic treatment, followed by frequent relapses.³

There are a number of factors predisposing to actinomycosis, including poor oral hygiene (such as dental caries, infections, and periodontal problems) and breaching of oral mucosa (dental extraction, gingival trauma, local tissue damage by neoplasia or irradiation, surgery), diabetes mellitus, male, immunosuppression, malnutrition, alcoholism.^{1,2,3}

Although it is not considered an opportunistic infection, it is more likely to occur in immunosuppressed individuals, such as patients with HIV.

The gold standard for diagnosing cervicofacial actinomycosis is histological examination and bacterial culture of affected tissue. Histologic examination is more sensitive.⁵ Because there is a high rate of failure in culturing the bacteria, mostly due to prior antibiotic treatment, inhibition of growth by contaminant or concomitant microorganisms, suboptimal culture conditions or short term incubation.¹¹ Gram staining can show Gram-positive filamentous branching bacteria at the periphery of the granule that is highly suggestive of actinomycosis. Immunofluorescence techniques have poor sensitivity but are highly specific.⁵ Treatment of cervicofacial actinomycosis may include surgical intervention in cases of large abscesses, bone or maxillary sinus involvement, or excessive fibrous tissue.² Treatment and prevention of dental caries and other dental problems is essential.⁵

Although there are no randomized controlled trials evaluating antibiotic regimens for cervicofacial actinomycosis, the treatment of choice is oral amoxicillin, since most isolates are susceptible to beta-lactams. Antibiotics are usually administered for a long time reaching up to 12 months, depending on tissues involved and patient response.⁵ For cervicofacial infection, penicillin G (50-75 mg/kg/day intravenously in four daily divided doses) for 4 to 6 weeks may be followed by oral penicillin V (30-60 mg/kg/day administered in four divided doses) for 2 to 12 months.²

The course of antibiotics can be shortened to as short as 4-6 weeks, in uncomplicated cases, or if there was adequate surgical intervention, no bone involvement, and rapid satisfactory patient response.^{14,15} Mild cervicofacial actinomycosis

may be managed with just 2 months of oral penicillin V or doxycycline 2 to 5 mg/kg per day administered in two equal portions at 12 hour intervals without surgical intervention.⁴

Surgical therapy is often indicated for curettage of bone, resection of necrotic tissue, excision of sinus tracts, and drainage of soft tissue abscesses. Although surgery facilitates recovery, it is usually not curative by itself.⁴

In conclusion, Actinomycosis can present in a number of different pictures, so the physician should be aware of the diversity of presentations, and have a high index of suspicion. Once diagnosis is made patient should be treated with an adequate course of antibiotics.

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