



A Rare Case of 66-Year-Old Woman with Right Lumpy Jaw Syndrome and Pathological Fracture of the Mandible

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Abstract

Lumpy jaw is an extremely rare disease with few cases reported in the literature. This report presents the case of a lumpy jaw with mandibular pathological fracture associated with actinomycosis osteomyelitis, due to a dental infection. The case was managed surgically. An overview of the entity including literature review of the clinical presentation, diagnosis and emerging treatment criteria was discussed. Clinicians should be aware of the possibility of actinomycosis arising in the setting of dental infection, and the importance of bone biopsy and cultures in arriving at a definitive and timely diagnosis.

Introduction

Lumpy jaw syndrome is well known by veterinarians. It is frequently found in dogs, swine and horses. The clinical picture of this disease consists of a large swelling and/or abscess that grows in the face and neck regions of the infected animal, following an odontogenic infection. In humans, this syndrome is particularly associated with poor oral hygiene, and mandibular osteomyelitis, periodontal disease and periapical dental abscess [1-5].

Lumpy jaw syndrome is mainly due to anaerobic polymicrobial infection; including *Actinomyces* spp (filamentous Gram-positive bacilli belonging to the human commensal flora) [1-5]. Lumpy jaw syndrome is the most common clinical form of cervico-facial actinomycosis and can be associated with skin fistula.

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Case Presentation

A 66-year-old female was referred to the Department of Oral Maxillofacial Surgery because of a large hard lump occupying the posterior lower right jaw "lumpy jaw". The swelling/lump was severe which left the patients in severe deformity and difficulty in eating and swallowing. The lower right second molar was extracted 1 year prior to referral and the patient could not recall ever experiencing pain. However, on clinical examination, the lower right body of the bony mandible was fully exposed and the tooth socket seemed like a freshly extracted one. A CT scan of the area revealed ill-defined bony changes with osteolytic and osteosclerotic areas associated with a pathological fracture of the right body of the lower jaw (Figure 1). The patient underwent surgical intervention with a preliminary clinical and radiological diagnosis of actinomycosis. Decortication of the lower jaw and thorough debridement of necrotic tissue was carried out. Histopathological evaluation was carried out for the curetted necrotic tissue. The patient responded well to surgery and high doses of post operative, intravenous and then oral amoxicillin for 6 months (Figure 1).

Culture of the tissue was positive for *Actinomyces* and Gram-positive cocci were observed on cultures of the involved tissue. On histopathological examination, trabeculae of necrotic woven bone enclosing *Actinomyces* granules with bone marrow and a number of partially resorbed bony sequestra, nonspecific inflammatory cell infiltrates, vascular proliferations, and granulation tissue were seen. Within the granulation tissue were granules surrounded by polymorphonuclear leukocytes. The periphery of the *Actinomyces* granules showed radiating, basophilic filaments and eosinophilic, club-shaped ends (Figure 2).

Discussion

Signs and symptoms

Cervicofacial actinomycosis usually involves tissues surrounding the upper and/or lower jaw. Approximately 50% of cases affect the body of the mandible itself, cheek (15%), chin (15%), and ascending ramus (10%) [1-5]. Typically, the disease presents as a slowly progressive painless

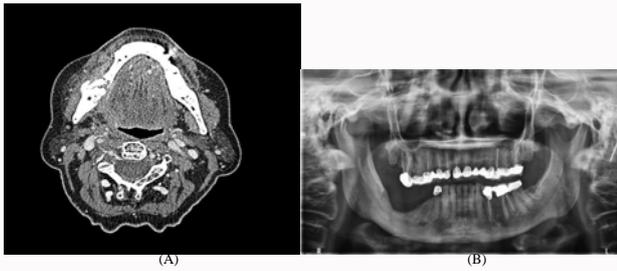


Figure 1: A) CT scan image revealed ill-defined bony changes with osteolytic and osteosclerotic areas associated with a pathological fracture of the right body of the lower jaw (red arrow). B) Post operative panoramic X-ray shows full recovery of the patient with optimum bone healing of the fracture of the lower right jaw.

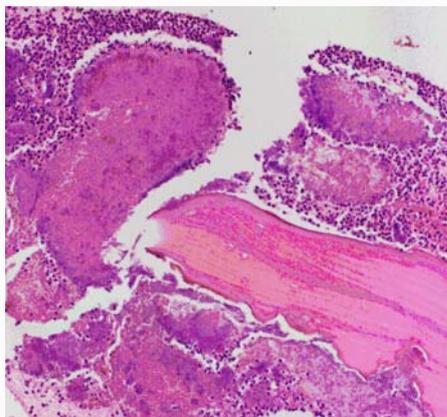


Figure 2: Photomicrograph sections showing multiple lamellar bony trabeculae and few soft tissue fragments including fibrous tissue, skeletal muscle fibers and inflammatory cell infiltration. Several foci show devitalized bony bits (sequestrum). Also seen within the inter-trabecular spaces and lying separately are variable sized colonies of long filamentous organisms with sunray appearance. Many colonies show surrounding Splendore-Hoeppli phenomenon. (H&E stain x20).

indurated swelling, evolving into multiple abscesses with draining sinus tracts on the skin surface or oral mucosa. Associated with a thick yellow exudate with characteristic sulfur granules [2]. At severe cases, pain and trismus can occur, linked with masticatory muscles infiltration. Acute suppurative forms with rapid abscess formations are less common and are usually febrile and painful. Regional adenopathy is rare. Bone involvement is observed in approximately 10% of cases [6].

Although most cases are of odontogenic origin and concern the perimandibular regions, many other locations of primary infections have been described, including the tongue, sinuses, middle ear, larynx, lachrymal pathways, and thyroid gland [1-6].

Diagnosis

Imaging findings are usually non diagnostic. Oral panoramic radiograph is fundamental to diagnose odontogenic abscesses, which will require dental extraction. MRI and CT scan not only may demonstrate the involvement of skin and soft tissues, but are also useful to examine bone involvement [1,2,7-10].

Diagnosis can be difficult, and especially differentiating between neoplastic conditions, malignant hemopathy, and other cervicofacial infections such as nocardiosis or mycobacterial infections. The gold standard for diagnosing actinomycosis of the lower jaw (lumpy jaw)

is histological examination and bacterial culture of an abscess or of a suspected bone, if actinomycosis is suspected. It is recommended to incubate bone samples for two weeks for microbiological cultures. Bacteria frequently reduce their growth capabilities in chronic osteomyelitis [1,7-9]. A false negative result of the cultures is common before surgery due to the frequent prescription of oral antimicrobials especially in patients with odontogenic cervicofacial actinomycosis.

Taking, for instance, the bacterial diagnosis for patients with prosthetic joint infection, the sensitivity decreased from 76.9% to 47.8% to 41.2% for bone sample culture as the antimicrobial-free interval before surgery decreased from greater than 14 days to 4 to 14 days, to 0 to 3 days, respectively [9]. As a result, in patients with chronic mandibular osteomyelitis suspected to have cervicofacial actinomycosis, cessation of antimicrobials at least 14 days prior to surgery is mandatory to facilitate the growth of *Actinomyces* spp. in cultures. In typical cases, i.e. especially in patients with lumpy jaw syndrome, *Actinomyces* spp. have always been targeted by antimicrobial therapy, regardless of the results of microbiological cultures or the result of pathology [1,7-10].

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Current and emerging treatment options

Surgical management can be required for drainage of voluminous abscesses, marsupialization of chronic sinus tracts, excision of recalcitrant fibrotic lesion, and/or debridement of necrotic bone tissue in case of osteomyelitis [1,7,9]. Treatment of dental caries and/or apical abscesses is essential, often necessitating dental avulsions.

There is a lack of randomized controlled trials to evaluate antibiotic regimens for cervicofacial actinomycosis. Most isolates are susceptible to beta-lactams, and the treatment of choice is a prolonged course of oral amoxicillin. As the penetration of beta-lactams in bone is low (10% to 20% of the administered dose) [10], intravenous high doses of amoxicillin (up to 200 mg/kg/day) or penicillin G (up to 24 MIU/day) has to be used initially in severe cases. Acceptable alternatives include clindamycin, macrolides (erythromycin, clarithromycin, or azithromycin), and doxycycline, which has a better bone penetration [1-5,7-10]. The adjunction of a companion drug such as metronidazole or a beta-lactamase inhibitor is controversial, but may help in these frequent polymicrobial infections [1,10]. The traditional prolonged course of up to 6 to 12 months of treatment can likely be shortened if an optimal surgical resection of infected tissues has been performed, in the absence of bone involvement, and if a satisfactory patient response to treatment is rapidly observed. Indeed, several observations have reported satisfactory cure rates with 4 to 6 week antimicrobial therapy [1-5,7-10].

In summary, clinicians should be aware of the possibility of actinomycosis arising in the setting of dental infection, and the importance of bone biopsy and cultures in arriving at a definitive diagnosis in order to plan a timely and efficient management of this rare disease entity.

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