

## Cervical actinomycosis: A rare diagnosis in cervical lymphadenopathy

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### Abstract

Actinomycosis is a rare infectious disease caused by an anerobic gram-positive non-spore forming bacteria that resulting in chronic granulomatous inflammation with formation of multiple abscesses. This infection can manifest as a swelling in the cervicofacial region. It is very sensitive to penicillin but often requires a prolonged course of antibiotics. We present the case of a patient with a 2x2cm neck lump who was treated with complete surgical excision and a short two-week course of antibiotics. Histopathologically, there were several foci of abscess, only one revealing actinomycosis with it's characteristic sulphur granule appearance. Cervicofacial actinomycosis still remains a rare disease that can lead to fatal outcomes. Although difficult to diagnosed, it can be successfully treated surgically with a short two-week course of antibiotics compared to the traditional six-week course.

### Keywords

rare neck lump; cervical lymphadenopathy; neck infection; actinomycosis

### Introduction

Actinomycosis is a rare disease caused by anaerobic, gram-positive non-spore forming branching filamentous bacteria [1-3]. Due to it's anaerobic nature, the bacteria is difficult to culture [1]. Actinomycosis is very sensitive to penicillin but often needs a prolonged course of antibiotics of about 4-6 weeks [4]. The incidence is approximately 1 per 40,000 and can present as a chronic progressive swelling or as a rapidly enlarging mass. Previously, it was known to be a fatal disease however with the widespread use of antibiotics, improved dental hygiene and water fluoridation, the incidence have been decreasing [1,5].

### Case Presentation

A 31-year-old Indian man presented to the Otolaryngology-Head & Neck clinic with a one-month history of a painless right neck swelling, gradually increasing in size. There was no history of fever, oral cavity infection or dental trauma. The patient was otherwise healthy without significant past medical history.

Physical examination of the neck showed a firm non-tender swelling at level III, measuring approximately 2.0cm. There were no signs of cutaneous fistulae. There were no abnormal findings on endoscopy of the post nasal space and larynx.

Hematological investigations were unremarkable revealing only a slightly elevated white cell count ( $11.9 \times 10^3/\mu\text{L}$ ). Inflammatory markers were normal and mantoux test was negative. Fine needle aspiration was inconclusive. A Computed Tomography (CT) scan of the neck revealed a well-defined vividly enhancing lesion in the right neck laterally displacing the sternocleidomastoid. (Figure 1) It measured approximately 2.0cm in diameter and extended from the angle of the mandible inferiorly into the cervical region. It appeared separate from the overlying muscles.

Based on the CT findings, he promptly underwent an excisional biopsy of the mass under general anesthesia. Intraoperatively, the mass was found to be firm and fibrotically adherent to surrounding structures from the upper border of the angle of the mandible to the lower border of the cricoid cartilage. Postoperative recovery was uneventful. Histological examination showed an enlarged lymph node with several foci of dense collection of neutrophils forming abscesses surrounded by variable amount of foam cells, lymphocytes, plasma cells and fibrosis (Figure 2a). One of the abscesses revealed colonies of actinomycosis surrounded by neutrophils forming sulphur granule appearances (Figure 2b). The adjacent muscle and fatty tissue showed focal inflammation without evidence of malignancy.

The patient was placed on intravenous ampicillin/sulbactam (Unasyn) for three days as an inpatient on the ward and then oral Unasyn for another ten days as an outpatient. There were no complications or evidence of recurrence after three months follow-up.

## Discussion

In 1938, Cope classified actinomycosis infection into three distinct clinical forms: cervicofacial, pulmothoracic and abdominopelvic with the cervicofacial form being the most common constituting 42% of all presentations [6]. The sites most commonly involved in this form include the submandibular space, cheek, parotid gland, teeth, tongue, nasal cavity, gingival, oral space, hypopharynx, larynx and cervical lymph nodes [7,8]. Actinomycetes are constituents of normal oral flora and are only able to cause an infection if there is a mucosal breach [3]. The disease has been reported to affect people in their third to sixth decade in life [7].

### Clinical signs and symptoms

Actinomycosis was previously described as “lumpy jaw” disease due it's common appearance in the submandibular region. The chronic form can typically present itself as an insidious swelling over weeks, months or years with suppurative features. The most common systemic manifestations are a low-grade fever; chills, lethargy and some weight loss however patients are usually asymptomatic. The bacteria spreads by burrowing through tissue planes and rarely has lymphatic or hematogenous spread [1,9].

### Diagnosis

CT scans are useful to evaluate the extent of disease such as its infiltration to muscle and degree of bony destruction. Ultrasonography is useful to identify the multiple small loculations or abscess

characteristic of actinomycosis. Galium scintigraphy or MRI are useful to differentiate between chronic inflammation and tumour by assessing the lesions vascularity [1]. The definitive diagnosis however is based upon histological findings.

## Histology

Actinomycosis has the propensity to develop into an abscess and drain superficially through a sinus in one-third of patients [5]. They can form fibrous thickening of capsule nodulation with fibrous bands, multiple abscesses with central loculation and scattered neutrophil laden macrophages [10]. Our patient did not have a full blown abscess however he did have several foci of abscesses that allowed us to culture the bacteria demonstrating the characteristic purulent crystalized granules with yellow “sulfur-like” appearance in one abscess colony [6]. The microbiological diagnosis is often challenging because of its anaerobic nature which makes it particularly sensitive to oxygen. Therefore the bacteria has to be promptly cultured in an anaerobic medium with a prolonged incubation time of up to three weeks [6].

## Differential diagnosis

Lymphadenopathy is a common clinical finding in neoplastic and inflammatory conditions. Infectious disease that should be considered in the differential include cat scratch disease, lymphogranuloma venerum, infectious mononucleosis, syphilis and HIV infection due to the similar histological appearance which involves an outer zone of granulation tissue and a central zone of abscess surrounding actinomycosis colonies [10]. A very close differential diagnosis is nocardiosis which contains similar looking granules which are acid fast branched bacilli. They respond to sulfadiazines as opposed to actinomycosis which respond to penicillin [1,6,8].

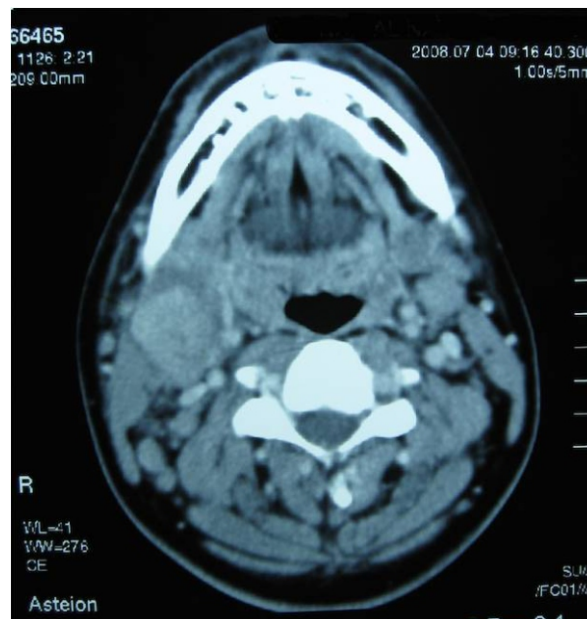
## Treatment

Actinomycosis is very sensitive to penicillin and hence the treatment of choice for this disease with alternatives being clindamycin, erythromycin or tetracycline. [1,8]. There is no standard treatment, however the literature recommends at least a course of at least 4-6 weeks of antibiotics [4]. This could even be prolonged up to 12 months to obtain adequate penetration through the sclerotic tissue that forms around the bacteria. [1]. In our case above, the total length of antibiotics given was less than two weeks. Perhaps a complete surgical excision of the mass prevented the need for a longer course of antibiotics. According to Nagler et al, patient's with actinomycosis do not respond well to antibiotics before degranulation and curettage of the lesion due to compartmentalization of the organism within the granulation tissue and the sulphur granules [2].

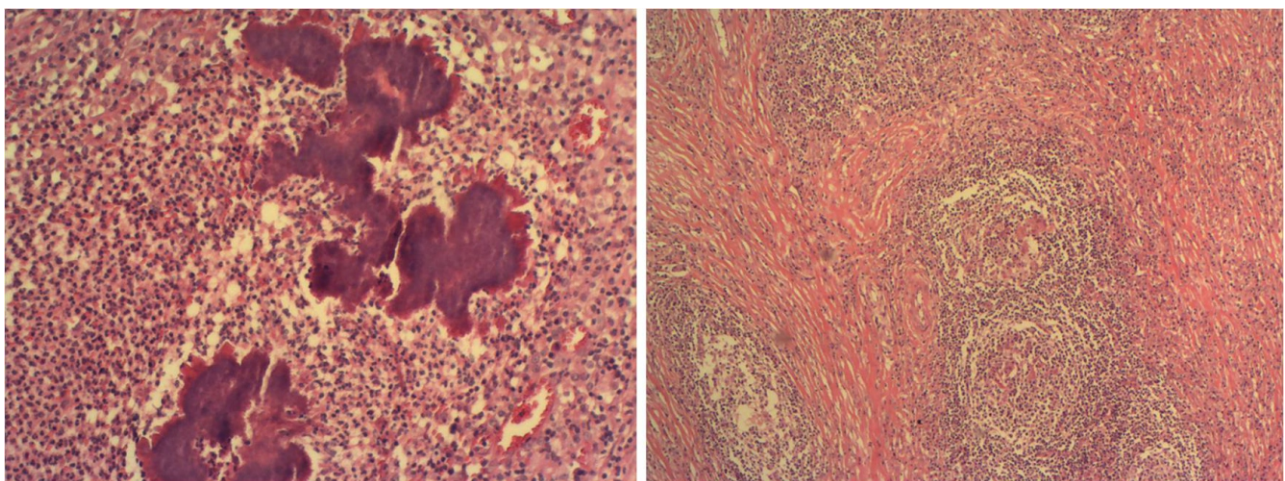
## Conclusion

Cervicofacial actinomycosis still remains a rare disease that can lead to fatal outcomes. Although it is difficult to diagnosed, it should be considered in our differentials because it may be treated adequately and promptly with a simple surgical excision and a short course of antibiotics.

## Figures



**Figure 1:** Axial CT scan of the neck showing a well defined enhancing lesion in the right neck displacing the sternocleidomastoid muscle laterally.



**Figure 2:** A. Enlarged lymph node with several foci of dense collection of neutrophils forming abscesses surrounded by variable amount of foam cells, lymphocytes, plasma cells and fibrosis. B. In one of the abscesses there were colonies of actinomycosis surrounded by neutrophils surrounding normal lymph node architecture.

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