Case Report

A rare case of Actinomycosis of the pyriform sinus mimicking malignancy

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Abstract

Actinomyces is a commensal in the oral cavity, digestive tract and genital tract. However invasive Actinomycosis occurs when the host immunity is breached due to local or systemic immunocompromised states. The most common presentation is a slowly growing mass with cutaneous draining sinuses. However the presentation is varied. There are cases of actinomycosis mimicking as tuberculosis and malignancy. The diagnosis is very important as the management varies drastically depending on the diagnosis. This case report highlights the unique presentation of actinomycosis in clinical and radiological diagnosis. Hence histopathological diagnosis is always mandatory in the diagnosis of actinomycosis.

Keywords: Actinomycosis, Pyriform fossa, Head and neck malignancy, Differential diagnosis of malignancy

Introduction

Actinomycosis is a chronic suppurative bacterial infection caused by Actinomycesis raelii [1]. It is a gram positive, filamentous, branching and micro-aerophilic bacterium. It is usually a commensal in the oral cavity especially the tonsilar crypts, digestive and genital tract. Invasive Actinomycosis occurs when the mucocutaneous barrier is injured [2]. The common predisposing local factors are poor oral hygiene and tobacco usage. The systemic causes include diabetes, malignancies and other immuno compromised states. Based on the site of occurrence, actinomycosis is clinically divided into three types as cervico facial (55%) which is the most common type followed by abdominopelvic (20%) and pulmonothoracic (15%)[3]. Actinomycosis usually presents as a slowly growing, painless mass with surrounding induration. In long standing cases, there may be abscess formation with cutaneous draining sinuses.

Case Report

A 55 years old male patient presented to the outpatient department with complaints of pain in the right side of the throat for 6 months. He had difficulty in swallowing and odynophagia. He had three episodes of haemoptysis which was self-limiting for 3 months. He was a chronic smoker, tobacco chewer and alcoholic for more than 20 years. Systemic examination revealed uncontrolled diabetes and hypertension on irregular treatment. The chest x-ray, CT chest and the routine blood investigations were normal. On local examination, his oral hygiene was poor. Fibre optic laryngoscopic evaluation revealed an ulceroproliferative lesion in the right pyriform fossa with edema of the adjoining mucosa (Picture1). Computed tomography of the neck reveals moderately enhancing soft tissue density thickening of right pyriform sinus, aryepiglottic fold extending upto the cricoid cartilage. No obvious erosion of cartilage is noted (Picture2). There is no cervical lymphadenopathy. A provisional diagnosis of malignancy was made based on the clinical and radiological findings. However, in order to confirm the diagnosis, it was decided to biopsy the lesion. Direct

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laryngoscopy and biopsy was performed under general anaesthesia. The histopathology surprisingly revealed the characteristic basophilic sulphur granules as irregular clusters surrounded by eosinophilic clubs (Picture3). The surrounding stroma revealsan intense neutrophilic andlymphoplasmacyticinflammatory infiltrate. Periodic acid Schiff'sstain demonstrates negative staining of the filaments within the granules (Picture4). The patient was treated with amoxicillin (500mg) and clavulanic acid (125 mg) thrice daily for 3 months and complete recovery was noted as evidenced by the follow up laryngoscopy.

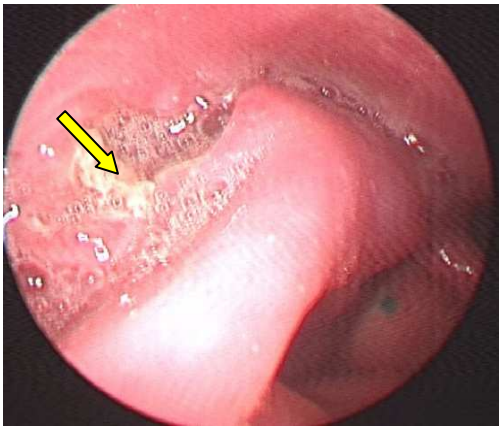


Fig 1: Flexible laryngoscopy showing ulcer proliferative lesion in right pyriform sinus with surrounding mucosal edema

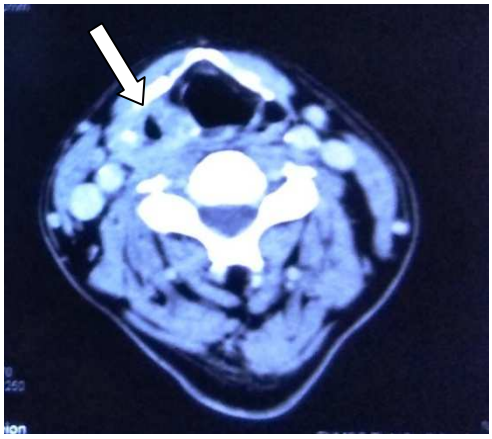


Fig 2: Axial plane contrast enhanced computed tomography of neck shows moderately enhancing soft tissue density thickening of right aryepiglottic fold and pyriform sinus

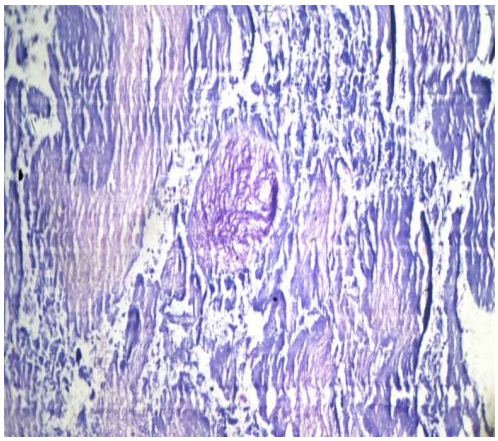
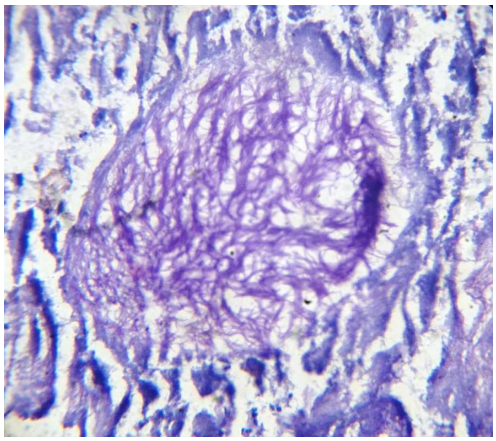


Fig 3: Hematoxylin and Eosin staining of actinomycotic colonies in histopathology section



Picture-4: Periodic acid schiff's staining of actinomycosis

Discussion

Actinomycosis can involve any part of the body. Many cases of actinomycosis involving the breast [4], pharynx [5], larynx [6], bronchi [7], pancreas [8], urachal remnants [9] and extremities [10] are reported in the literature. The clinical presentation is varied and can mimic malignancy as in our case or even tuberculosis. Many cases of actinomycosis misinterpreted as malignancy are reported in the literature [5,11-13]. The differential diagnosis of malignancy ranges from congenital cysts, inflammation, and infection to benign tumours. In this antibiotic era, infections mimicking malignancy are rare. One such rare occurrence is actinomycosis involving the pyriform fossa presenting as an ulceroproliferative mass. Radiological investigations such us computerised tomography or magnetic resonance imaging are essential to diagnose the underlying pathology. However, the radiological investigations can be misleading as in our case and hence a biopsy is always the golden standard in making the accurate diagnosis.

Our patient presented with recurrent episodes of haemoptysis. Haemoptysis is one of the most common clinical presentations of pulmonary actinomycosis as reported by a study [14]. Volpi et al [5] in their study reported a similar case of actinomycosis involving the

pyriform fossa. Their patient also presented with haemoptysis. Hence it may be concluded that haemoptysis is one of the presenting symptoms of thoraco pulmonary actinomycosis. The cases of thoraco pulmonary actinomycosis do not present with regional lymphadenopathy as in our case supported by other studies [2,5,7].

Hence a diagnosis of non-neoplastic pathology could be suspected in cases clinically presenting as malignancy without cervical lymphadenopathy [2]. Our patient had all the risk factors for an invasive actinomycosis with compromised local and systemic immunity. The histopathological diagnosis of actinomycosis is done by routine hematoxylin and eosin stain. In histopathological sections, irregular tangled masses of actinomycotic granules also called as sulphur granules.

The sulphur granules reveal delicate filaments which are colonies of gram positive bacteria. Periodic acid Schiff staining reveal the negative staining of the bacteria.

Diagnosis is very important as treatment varies based on the etiology. Early diagnosis is vital as invasive Actinomycosis involving the pyriform sinus can form an abscess, disseminate locally into the larynx and lung or spread hematogenously.

The treatment is essentially a course of antibiotic therapy and follow up. The drug of choice for actinomycosis is beta lactam antibiotics [15]. Surgical intervention is needed in long standing cases with complications such as abscess, sinus tract or fistula formation.

Conclusion

Actinomycosis is one of the rare differential diagnosis to be considered in head and neck malignancies. The most common presentation of actinomycosis involving the pyriform fossa is haemoptysis in addition to pain and difficulty in swallowing.

The clinical and radiological findings can be misleading and hence biopsy is mandatory for definitive diagnosis.

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