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Cervicothoracic Actinomycosis Presenting As Superior Venacava Syndrome: A Case Report

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Abstract
Actinomycosis is a slowly progressive infection caused by anaerobic or microaerophilic bacteria, primarily of the genus Actinomyces that colonize the mouth, colon, and vagina. Mucosal disruption might lead to infection nearly at any site within the body. The clinical presentations of actinomycosis are myriad. Thoracic actinomycosis is a very rare condition contributing 15% of actinomycosis cases. Here we present a rare case of cervicothoracic actinomycosis presenting as superior venacava syndrome. Actinomycosis is uncommon, and most physician's personal experience with its clinical presentations is limited. Actinomycosis remains a diagnostic challenge, even for a skilled clinician as it mimics tuberculosis and malignancy. Actinomycosis has been called the most misdiagnosed disease, and it has been said that no disease is so often missed by experienced diagnosticians.
Keywords: *Actinomycosis, Superior Venacava Syndrome.*

Case History

This is a case of 22 years old housewife hailing from Wayanad presented with multiple swellings around the neck for 2 years duration, swelling of face and dyspnea on exertion for 2 months. Initially it was small painless raised lesion in the right submandibular region with gradual enlargement, pus filling, and breaking down to discharge serous and occasionally yellowish powdery material. Multiple similar lesions were seen in the surrounding area-neck, upper chest wall, breast and axillae which heals spontaneously with scarring. And now she came with increase in the severity of the lesion. No similar lesion in the abdomen, legs and genital region. Her swelling started in the face followed by neck with distension of veins over the neck, abdomen and chest. It was associated with occasional cough, palpitation, dyspnea on exertion and dysphagia. There was associated loss of weight and appetite. She also gives history of low grade fever since 1 week. No history of swelling of legs, abdominal distension. No history of decreased urine output, frothy urine, hematuria. No history of chest pain,

hemoptysis. She is a chronic betel nut chewer. Bowel and bladder habits were normal. She takes mixed diet. She is a mother of 3 children with no complications during pregnancy. She is having regular menstrual cycles. No history of IUCD use. There was no significant family history.

Examination

She was conscious, oriented with temperature of 100°F.Her pulse rate was 110 bpm which was regular with low volume, Blood pressure was100/70 mm of hg and respiratory rate of 32/ min. She had pallor with bilateral axillary lymph nodes which were soft and non-tender. Facial puffiness was present. There was no cyanosis, clubbing, breast was asymmetrical and thyroid was not appreciable. Glossitis was present. Multiple confluent, tender nodules of varying size which were fixed to underlying subcutaneous tissue distributed over neck, upper back, chest, left breast, and right infra mammary region, left axilla with surface of skin showing multiple sinuses draining yellowish discharge was present[figure1, 2].There were hyperpigmented scars with regular and irregular borders. She also had facial oedema with distended veins around neck, thorax and abdomen. There was nicotine staining of teeth with very poor oral hygiene. Trachea could not be appreciated. Chest Expansion was1 cm. Tubular breathing was present in left infraclavicular and inframammary area. There was coarse crepitations present in the right infraclavicular area, right mammary area and right infra axillary area. There was no murmurs or additional heart sounds. Distended veins were present over the anterior abdominal wall with flow above downwards [figure3]. Liver was palpable 3cm below right costal margin and it was nontender with smooth consistency and well defined borders. Traube's space resonant. Nervous system was within normal limits.

Summary

22 years old betel nut chewer with poor oral hygiene having nodular lesions with discharging sinus and scars, remitting and relapsing last 2 years now came with increase in severity of lesion with yellowish discharge and features of superior venacaval syndrome. Possibilities of infectious etiologies like Actionomycosis, Nocardiosis, Botryomycosis, Tuberculosis and Eumycetoma were considered as there was combined dermatological and respiratory manifestations. Malignancies like Cutaneous T cell Lymphoma were also considered.

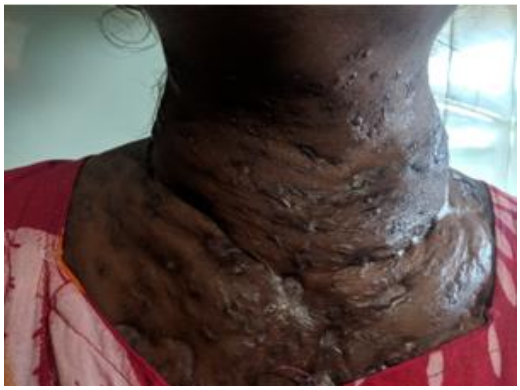


Figure 1: Multiple confluent, tender nodules of varying size which were fixed to underlying subcutaneous tissue

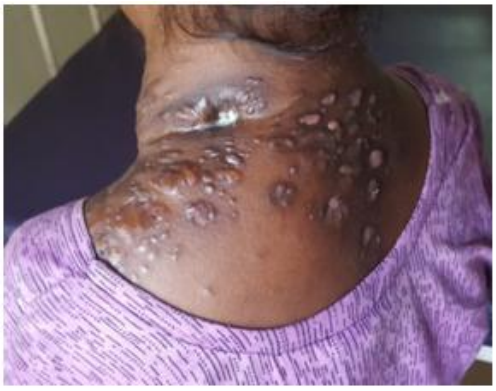


Figure 2 showing multiple sinuses draining yellowish discharge

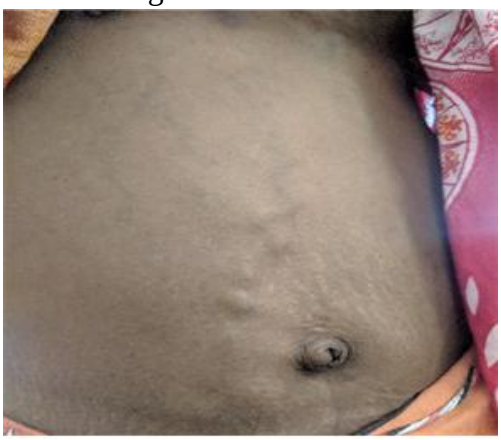


Figure 3 showing dilated veins over abdominal walls with flow above downwards

Test	Value	Test	Value
Haemoglobin	7.6	Total Bilirubin/Conjugated Bilirubin	0.5/0.2
Total Count	13300	Total Protein/Albumin	6.2/2.4
Differential Count	L14N82M4	Alanine Amino Transferase/ Aspartate AminoTransferase /ALP	32/12/212
Platelet Count Mean corpuscular volume(MCV)	3.93 74	Urine Microscopy	ALB-TRACE Pus 1-2
Random Blood sugar	112	PT/INR	16.3/1.2
Serum Electrolytes	139/3.5	ESR	124
C-Reactive Protein	184	HIV/HBsAg/Anti HCV	Negative

Investigations

Her routine blood investigations revealed anemia with low MCV and peripheral smear suggestive of iron deficiency anaemia. She had neutrophilic leukocytosis with ESR of 124. Chest X-ray done showed homogenous infiltrates in the bilateral upper lobe with mediastinal widening and lesion extending into multiple tissueplanes[figure4].Pus draining from the lesion site was cultured which revealed growth of methicillin resistant Staphylococcus aureus and was started on antibiotics according to the drug sensitivity. Mantoux test was negative and sputum cultures were sterile and negative for acid fast bacilli. USG venous Doppler study revealed bilateral internal jugular vein (IJV) thrombus with normal flow in the carotid artery. FNAC of axillary lymph nodes were suggestive of reactive hyperplasia. We proceeded with CECT thorax which showed bilateral IJV thrombus, brachiocephalic veins were not visualized with superior vena cava (SVC) contrast opacifications and heterogeneously enhancing soft tissue density in cervical space, infected collection/oedema in carotid space. There was also features of mediastinitis and aorto-pulmonary nodes were present [figure 5]. Skin biopsy taken from the lesion site showed ulcers with granulation tissue and foreign body giant cell reaction[figure 6]. Epidermis was hyperplastic. Dense inflammatory infiltrate composed of neutrophils and eosinophils forming micro abscess around darkly basophilic actinomycotic colonies was seen which was positive for Gomorimethenamine silver stain [figure 7]. Fungal culture showed yellow wrinkled colonies, Gram stain showed beaded branched Gram-positive filamentous rods. Staining for AFB was negative. Hence a diagnosis of cervicothoracic actinomycosis complicated by superior venacava syndrome co infected by MRSA species was made. She was started on injection Vancomycin 1g IV bd, injection Crystalline Penicillin 4million units IV Q4H.She was anticoagulated for thrombosis in the jugular veins and she was supplemented with iron orally and other supportive measures. Patient improved with antibiotics, she became afebrile and her lesions started healing with no discharge from sinuses and she was discharged with advice of antibiotics for 6 months.

Chest X-Ray

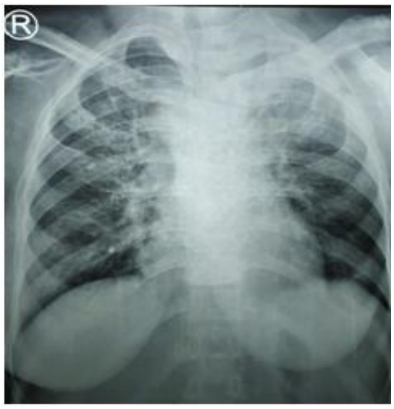
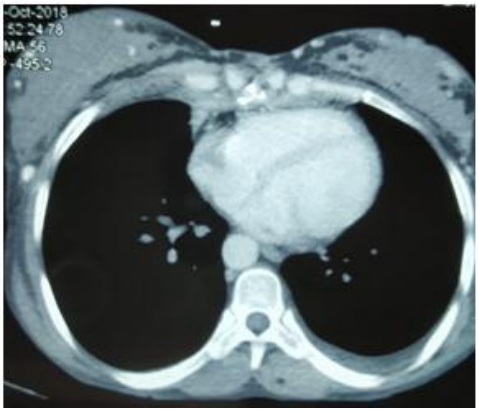


Figure 4: Chest X-ray showing pulmonary infiltrates and mediastinal widening

CECT Thorax



Skin Biopsy

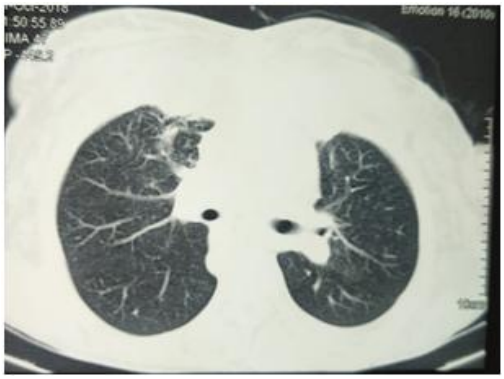


Figure 5: CECT Thorax showing lesion extending into multiple level of tissue planes[blue arrow] with mediastinal extension[black arrow]

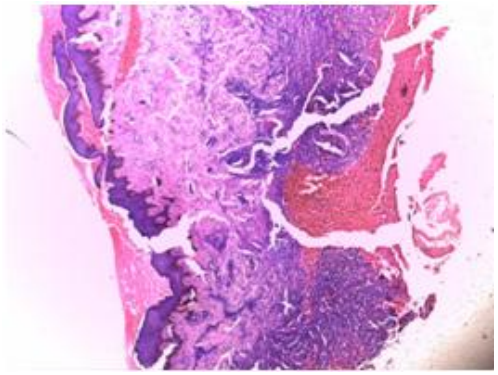


Figure 6: Ulcers with granulation tissue and foreign body giant cell reaction [blue arrow]

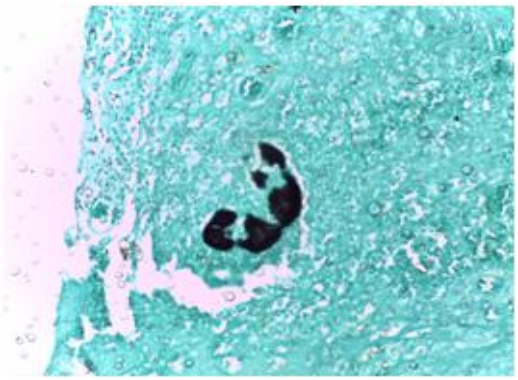


Figure 7: Darkly basophilic actinomycotic colonies was seen which was positive for Gomorimethenamine silver stain [blue arrow]

Discussion

Actinomycosis is an infrequent invasive bacterial disease that has been recognized for over a century[(1)]. *Actinomyces* spp. most common being Actinomyces israelii are filamentous Gram-positive bacilli, mainly belonging to the human commensal flora of the oropharynx, gastrointestinal tract, and urogenital tract[2–5]. Actinomycosis is endemic, occurring worldwide. It has no predilection for age, race, season, or occupation, and although not considered an opportunistic infection, it has been described in patients with HIV, leukemia, and in patients with other causes of immunodeficiency [6]. However, no underlying disease or immunosuppression is found in most patients. These infections usually involve the cervicofacial, thoracic and abdominopelvic regions, and the CNS, and no person-to-person transmission has yet been documented. Thoracic actinomycosis is frequently confused with lung cancer, lymph node blocks, or mediastinal tumors. The thoracic variety of the disease accounts for approximately 15% of the cases. Thoracic pain is considered the first symptom in many cases, but the examination and inspection of available material stained by direct microscopy is crucial. Thoracic actinomycosis is attributed to the aspiration of oropharyngeal secretions. The usual mechanism of the thoracic variety is by infection of the lung. Very few cases of thoracic actinomycosis with superior vena cava obstruction has been described in the literature. This infection is considered to extend across fissures and through the pleura. Mediastinal actinomycosis is provoked by direct local extension from the neck. Air space consolidation with adjacent pleural thickening are radiologic findings suggestive of pulmonary actinomycosis. CT findings overlap with those of other inflammatory and neoplastic conditions, including soft tissue mass, with various degrees of infiltration and abscess formation[7]. There are no radiological signs that are indicative of thoracic actinomycosis, and chest X-ray findings can mimic a wide variety of diseases, including

pulmonary infiltrate (suggestive of mild pneumonia) and micronodular infiltrate accompanied by pulmonary cavitation or large masses (suggestive of neoplasia), pleural effusion being common. In advanced cases, a CT scan of the chest can reveal involvement of the chest wall, mediastinal involvement, and pleural involvement. An image of diffuse involvement that crosses anatomical boundaries is highly suggestive of pulmonary actinomycosis. The differential diagnosis of pulmonary actinomycosis includes recurrent pneumonia, pulmonary infarction, lung cancer, Wegener's granulomatosis, Nocardiosis, pulmonary sequestration, and bronchogenic cyst[8]. It is difficult to make a diagnosis of pulmonary actinomycosis for the following reasons. First, Actinomyces are difficult to culture because cultures require brain/heart-enriched agar and the organisms grow best at a temperature of 37℃ in an atmosphere of 6-10% ambient carbon dioxide. Moreover, cultures of Actinomyces should be observed for up to 21 days to allow for adequate growth. Second, the histological diagnosis requires the presence of actinomycetes filaments or sulphur granules in purulent matter from infected tissue. The main principle of treatment is the use of high-dose intravenous penicillin for a long duration, and that generally 18-24 million units of penicillin per day are given for 2-6 weeks followed by oral penicillin for 6-12 months. In the present case, we initially administered Intravenous penicillin 24 million units per day for 4 weeks which is normally effective in actinomycosis as surgical intervention would cause increased risk of morbidity and mortality. The patient's lesions were healing over time and there was no discharge from lesion. The scars and disfigurement are remaining. We decided to follow the classical treatment in the literature, and she was discharged on oral penicillin along with oral anticoagulation therapy for jugular vein thrombosis.

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