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Case Report

A rare case of actinomycosis in routine fnac of the face in a tertiary care center in Central India

Puja Singh[1,*], **Meena Singrol**[2], **Amar Gangwani**[1]

[1]*Dept. of Pathology, Bundelkhand Government Medical College, Sagar, Madhya Pradesh, India*
[2]*Atal Bihari Vajpai Government Medical College, Vidisha, Madhya Pradesh, India*

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ABSTRACT

Actinomycosis is a chronic suppurative bacterial infection. It is caused by gram-positive, non-acid fast, filamentous, anaerobic to microaerophilic bacilli. Actinomycosis on the root of the nose is a rare entity. We report a case of 38 years old male who presented with swelling over the root of the nose since birth. Mucoid material was aspirated. Microscopic examination reveals a filamentous structured colony and crystal against the granular proteinaceous background. Features suggest benign cystic lesion and secondary actinomycosis infection, negative with an acid-fast bacilli stain consistent with Actinomyces species. As stated in most of the literature, mainly males are affected by this condition. Very little information is available on the use of fine-needle aspiration cytology (FNAC) to diagnose these lesions. We are reporting a case of an actinomycotic lesion that was reliably diagnosed with FNAC. So, we emphasized here that FNAC is aninexpensive, rapid, and simple outpatient procedure that helps in the rapid diagnosis of Actinomycosis.

1. Introduction

A persistent, purulent bacterial illness known as actinomycosis is brought on by gram-positive, non-acid fast, filamentous, anaerobic to microaerophilic bacilli.[1] The name actinomycosis means “ray fungus”. Hence organism’s filamentous appearance may resemble fungi.[2] Actinomyces Israeli is a causative agent of actinomycosis. Actinomyces Israelii, the current name for the causal agent Streptothrix Israeli, was first identified as the cause of human actinomycosis by Kruse in 1896.[3]

Actinomycosis is characterized by abscess formation, tissue fibrosis, and draining sinuses. The most common age group is between the age of 20 to 60 years with a peak incidence of 40 to 50 years. The incidence of the disease is greater in males than in females. The male-to-female ratio is 3:1.[4]

Actinomycosis normally lives as commensals in the human oral cavity, respiratory and digestive tracts.[5] It becomes invasive by breaking the integrity of the mucus membrane and the presence of devitalized tissue to invade deeper body structure and cause disease through a mucosal lesion, they gain access to the subcutaneous tissue.[6] It normally colonizes the human mouth, digestive tract, and genital tracts. Usual clinical presentations arepelvic actinomycosis (in women with an intrauterine device), cervicofacial actinomycosis (following dental foci of infection), and pulmonary actinomycosis (in smokers with poor dental hygiene).[7]

Actinomycosis very often imitates malignancy, tuberculosis, or Nocardiosis, it spreads continuously and progressively and formscold abscesses.[8] Both Actinomyces and Nocardia cause similar clinical syndromes involving the bone, lung, soft tissue, joint, and central nervous system. Hence, they are often called “the great masqueraders”. Leading to delayed diagnosis of actinomycosis and

* *Corresponding author*.
E-mail address: dr.pujasingh@gmail.com (P. Singh).

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nocardiosis. Both infections are characterized by the development of abscesses and a protracted course of illness. Patients usually arrive with fistulous tracts and draining sinuses, and extended courses of parenteral and oral treatments are needed to cure these infections (e.g., months to years).[2]

Actinomyces species were once believed to be fungi due to their branching filaments. However, now they are classified as higher prokaryotypic bacteria. In this way, they are closely related to Nocardia species. The Actinomyces are pleomorphic, non-acid fast, gram-positive, and delicately filamentous. Nocardia species are morphologically similar to actinomyces on gram staining and also clinically resemble actinomycosis. Some Nocardia species are somewhat acid-fast, and all species thrive aerobically.[9] Pathological investigation and culture are the bedrock of diagnosis. Necrosis with yellowish sulfur granules and filamentous gram-positive pathogens, that resemble fungi, are common microscopic findings.[8]

2. Case Presentation

A 38 years old male presented with only solitary swelling over the midline between both the eye for 3 years. On examination single swelling was seen, measuring 1cm x 1cm, firm, mobile and non-tender. Mucoid material is aspirated. Swelling is reduced in size after aspiration. Clinically, we suspected an epidermoid cyst which on microscopy showed the presence of actinomycosis.

On microscopic examination, using PAP and Geimsa stains the smear's revealed a colony of filamentous structures and crystals against the granular proteinaceous background. The features suggested benign cystic lesion with secondary actinomycosis infection as represented in Figures 1 and 2, 10x, and 40x respectively showing the characteristic features of actinomycosis. Differential diagnoses included Nocardia. Nocardia can be differentiated from Actinomyces by performing AFB staining, as Nocardia exhibits a varying degree of acid fastness due to the mycolic acid content of the cell wall. Actinomycosis is a specific granulomatous chronic infection, therefore results of routine blood tests areusually normal, leukocyte values are high, as consistent in our case as the patient had a marked neutrophil leucocytosis and the rest of the blood parameters were within normal limits.

3. Discussion

Actinomycosis is a chronic, rare, and slowly progressive granulomatous disease. Actinomycetaceae family filamentous Gram-positive anaerobic bacteria are the culprits. (genus Actinomyces).[10] The organisms may resemble fungi due to their filamentous appearance.[2]

Cervicofacial swellings are the most common manifestation of actinomycosis accounting for 50- 70% the

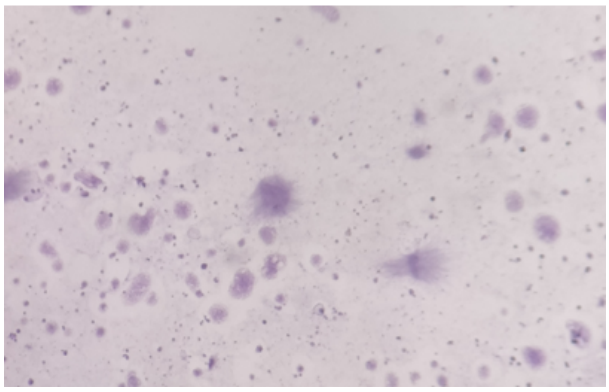

Fig. 1: Filamentous structure of actinomycosis (10x) shows presence of filamentous structured organisms in a clear protinaceous background devoid of any inflammatory infiltrate.

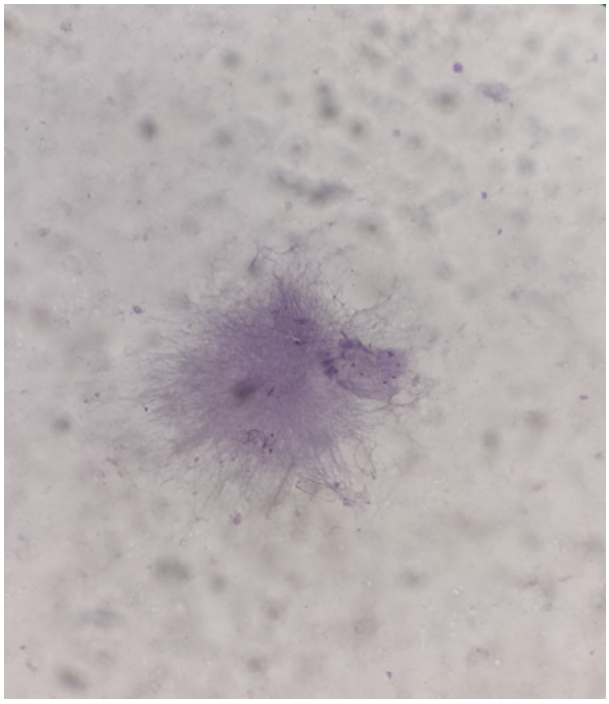

Fig. 2: Filamentous structure of actinomycosis (40x) high power view shows the clear filamentous projections of the actinomycosis. The clear absence of hyphae and septate is noted along with clear background.

of reported cases.[11] The incidence of the disease is greater in males along with the mean age group of 40 to 50 years.[4] This is in line with our case as the site is the root of the nose of a 38-year-old male.

Actinomycosis is often characterized by abscess formation, tissue fibrosis, and draining sinuses.[4] However, in our case, single swelling of size 1.0 x 1.0 cm was present with no draining sinuses and abscess formation.

Actinomyces are difficult to isolate as more than 70% of cultures are negative.[12] In our case, there were no obvious

clinical features suggestive of actinomycosis in any other body part. Clinically it was suspected as an epidermoid cyst. The presence of actinomycosis was an incidental finding.

FNAC is a good tool to diagnose actinomycosis. It is a safe, rapid, and relatively inexpensive outpatient procedure for rapid diagnosis.[13,14] In our case, Actinomyces was distinguished from its fungal mimickers, Nocardia, with the help of an acid-fast stain, which is positive in Nocardia.

Actinomycosisis often misdiagnosed because it can mimic other conditions such as malignancy and tuberculosis. A high level of clinical suspicion is needed to diagnose and cure actinomycosis. The microscopic presence of sulphur granules (yellow granules) and Splendore–Hoeppli phenomenon (antigen antibody complex) are usually associated with actinomycosis but were absent in the present case.[15,16] It is especially crucial to make early diagnoses in patients with indolent, unresolving, or relapsing chronic inflammatory disease. Hence, special efforts must be made to look for this microorganism. Material aspirated, in actinomycosis,is found to be an inflammatory exudate and rich in neutrophils along with a few lymphocytes, histiocytes, and plasma cells.[16] In contrast, in our case, the material aspirated had mucoid consistency and was deficient in inflammatory cells on microscopy. The cytological image of actinomycosis appears as a filamentous structure or clumps of the fibrillary organism.

4. Conclusions

The diagnosis of actinomycosis is often delayed due to a lack of clinical suspicion. Actinomycosis often mimics neoplasia both clinically and radiologically. Obtaining the specimen and performing a successful culture is also difficult. So the cytological diagnosis of actinomycosis is a good tool to diagnose and should always be borne in mind when doing FNAC of the face region. It is minimally invasive and quick. Thus avoiding unnecessary or extensive surgery. In our case, it was clinically suspected as an epidermal cyst which turned out as actinomycosis upon microscopy. Actinomycosis at the root of the nose is a rare entity. Aspirated material is usually exudative in nature and rich in acute inflammatory cells. In contrast in our case report, aspirated material was mucoid and deficient in inflammatory cells. We must also emphasize that FNAC is the technique of choice for actinomycosis diagnosis.

5. Source of Funding

None.

6. Conflict of Interest

None.

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Author biography

Puja Singh, Associate Professor https://orcid.org/0000-0002-6182-5597

Meena Singrol, Senior Resident

Amar Gangwani, Professor

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