

CASE REPORT

PRIMARY ACTINOMYCOSIS OF BREAST – A CASE REPORT

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ABSTRACT: Primary actinomycosis of breast is a rare disease. Here we report one such case in a 25 year old lactating mother from West Bengal with a breast lump and multiple discharging sinuses, clinically normal otherwise with no remarkable clinical history. Histopathology from breast lump suggested granulomatous mastitis. Microbiological investigations isolated *Actinomyces israelii* and *Staphylococcus aureus*. A diagnosis of primary actinomycosis of breast was made and clinical response was followed up with overall improvement in patient's condition.

INTRODUCTION: Rare cases, though rare, do occur and thus deserve reporting. Primary actinomycosis of breast is one such clinical entity of which only about 32 cases⁽¹⁴⁾ have been reported worldwide since its first description by Ammentorp in 1893⁽⁷⁾. Actinomycosis of breast is primary when portal of entry is through nipple in contrast to secondary where thoraco-pulmonary actinomycosis invades breast tissue after extending through ribs and thoracic muscles⁽¹⁴⁾. Such a case poses challenge to the clinicians, pathologists and microbiologists. Without proper diagnosis, the disease progresses and leaves disappointment not only to the care-givers but also traumatizes patient with disfigurement and unnecessary drug effects.

Here we report one such case from Calcutta School of Tropical Medicine, West Bengal, India.

CASE REPORT: A 25 year Muslim lactating mother, having a 3 year old son, presented with right-sided breast lump for about 5 months with development of multiple sinuses. There was tender erythematous breast swelling with multiple sinuses mainly in the periareolar region. Sinuses discharged thick, sticky pus with no sulfur-like granule. No history suggestive of tuberculosis, leprosy, other granulomatous diseases or any other specific disease could be obtained nor any history of obvious local trauma. Clinically also no suggestive signs regarding the etiology could be elicited but revealed right axillary lymphadenopathy. Clinicians had tried 2 months of empirical Anti-Tubercular drugs with apparent deterioration followed by its discontinuation. Excision biopsy from the breast lump revealed absence of neoplastic cells and acid-fast bacilli but presence of infiltration by histiocytes and chronic inflammatory cells along with non-caseating granulomas and neutrophilic microabscesses i.e. suggestive of granulomatous mastitis. Her Chest skiagram was normal and blood reports showed: hemoglobin 12.7g/dl, leukocyte count 9500/cu.mm. (Neutrophil 68%, Lymphocyte 26%, Eosinophil 4%, Monocyte 2%), ESR 90 mm 1st hr., Platelet-3.6 lac/cu.mm., normal liver function test, non-diabetic and negative for HIV-1 & 2.

Microbiological procedures were aimed at isolating possible pathogens including fungal etiology. Pus was collected from depth of the sinuses. Direct smears had plenty of pus cells obscuring any obvious microorganism in view and acid-fast staining by Ziehl-Neelsen method and modified Ziehl-Neelsen method (using 1% sulfuric acid) were negative (see Fig.1). A 10% potassium hydroxide preparation of the pus was negative for fungal element but was put to fungal culture in

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Sabouraud's dextrose agar medium at 25 °C for 4 weeks. Bacteriological culture was done at 37 °C both aerobically and under partial anaerobic condition using Fortner's method (see Fig.2). Overnight incubation yielded growth of *Staphylococcus aureus*. Culture on 5% sheep blood agar media by Fortner's method was examined daily without disturbing the partial anaerobic condition which after 10 days yielded growth of *Actinomyces israelii*. Fungal culture was negative.

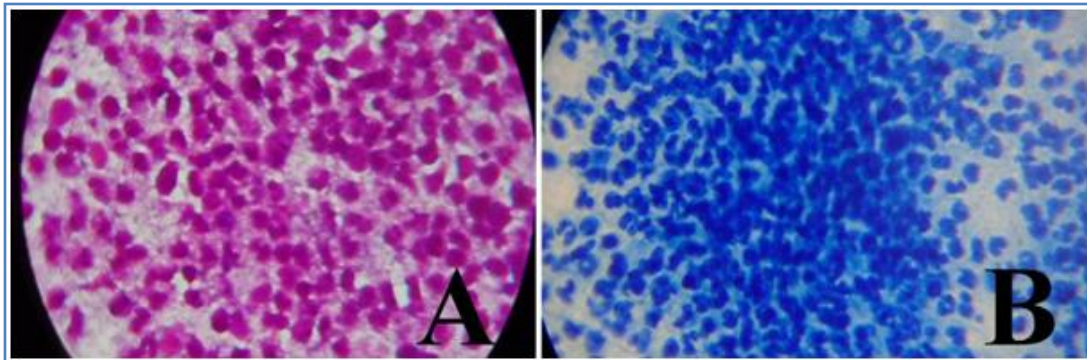


Fig. 1: Direct smears showing plenty of pus cells. (A) Gram stain & (B) Acid-fast stain.

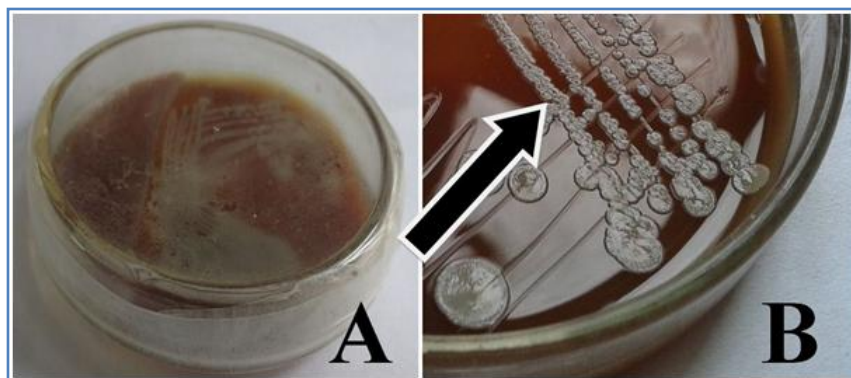


Fig. 2: (A) Fortner's method of culture & (B) Close-up view. Arrow points to the small, irregular, rough *Actinomyces* colonies.

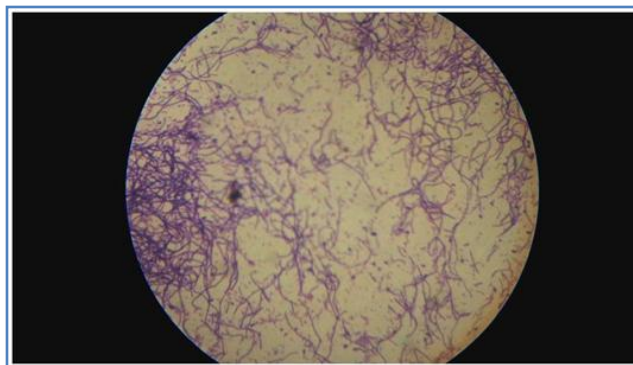


Fig. 3: Gram-positive, extremely fine, filamentous forms of *Actinomyces israelii* (1000X)

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Case was therefore diagnosed to be a case of primary actinomycosis of breast with *Staphylococcus aureus* as co-pathogen.

Identification of *Staphylococcus aureus* was done by Gram stain, catalase positivity, presence of clumping factor, coagulase and golden yellow pigment, and biochemical tests. Antibiogram on Mueller-Hinton agar media using disc-diffusion method showed its sensitivity to Linezolid, Vancomycin, Ceftriaxone, Amikacin, Cefoxitin and Cotrimoxazole; and resistance to Amoxicillin-Clavulanic acid, Azithromycin and Piperacillin-Tazobactam.

Actinomyces israelii was identified from findings of Gram-positive extremely fine filamentous forms (see Fig.3), non-pigmented and non-hemolytic irregular rough colony morphology (see Fig.2), Catalase negativity, positive nitrate reduction test, negative urea hydrolysis test, negative Voges-Proskauer reaction and by extended biochemical tests to determine different sugar fermentation (see Table). Fermentation of glucose, maltose, sucrose, mannitol, arabinose, sorbitol were found to be positive. For biochemical tests standard methods were followed as described by Slack & Gerencser⁽¹³⁾, Schoefield & Schaal⁽¹¹⁾ and were interpreted accordingly^(11,13,15).

Test	Result	Comment
Catalase	Negative	Excludes <i>Nocardia</i> , <i>Nocardiosis</i> , <i>Streptomyces</i> , <i>Mycobacterium fortuitum</i> , <i>Bifidobacterium dentium</i> , <i>Actinomyces neuii</i> , <i>Actinomyces viscosus</i>
Nitrate reduction	Positive	Excludes <i>Actinomyces turicensis</i> , <i>Actinomyces radingae</i> , <i>Actinomyces europaeus</i>
Colony pigment/ color	Grayish-white Non-pigmented	Excludes <i>Actinomyces odontolyticus</i>
Colony texture	Rough	Excludes <i>Actinomyces naeslundii</i> (geno.1), <i>Actinomyces gerencseriae</i>
Urea hydrolysis test	Negative	Excludes <i>Actinomyces naeslundii</i> , <i>Actinomyces viscosus</i>
Voges-Proskauer test	Negative	Suggest <i>Actinomyces israelii</i>
Fermentation of glucose, maltose, sucrose	Positive	
Fermentation of arabinose, sorbitol	Positive	

Table: Phenotypic tests with results obtained arranged in order of shortlisting the probable species of *Actinomyces* isolated by culture.

The patient was provided in-patient care with inj. Ampicillin (1g) QDS and tab. Doxycycline (100mg) BD which showed significant improvement with closing of sinus openings and stoppage of discharge but only one recurrence of discharge after few weeks. It was examined microbiologically and no microorganism could be isolated. The patient was discharged with tab. Clarithromycin (500mg) BD, tab. Ciprofloxacin (750mg) BD and tab. Cotrimoxazole-DS BD and was followed up at 2 monthly intervals. No further discharge was noted and healing of sinus openings ensued within few months.

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DISCUSSION: Actinomycosis is caused by gram-positive anaerobic filamentous bacteria that are part of the normal oral flora. Usually, it represents a chronic infection characterized by pus, fibrosis and fistulas that drain sulfur-like granules. The main clinical forms of actinomycosis are cervicofacial, thoracic, abdominal, and, in women, pelvic. Dissemination to other organs may occur by spatial contiguity.

Actinomycosis of breast is rare and is usually secondary to thoracopulmonary actinomycosis⁽¹⁴⁾. Primary actinomycosis of the breast starts at the nipple; most of the abscesses are retropapillary⁽⁷⁾. Possible causes of this condition observed by Cope and quoted by Lloyd-Davies include trauma, lactation and kissing⁽⁷⁾. Local mucosal injury causes actinomycotic infection of the lactiferous ducts. Most cases of actinomycosis of the breast have involved premenopausal women like our case⁽¹⁴⁾. The differential diagnosis should include inflammatory carcinoma, chronic suppurative mastitis, tuberculosis, syphilis, and chronic osteomyelitis of the ribs⁽¹⁴⁾. Treatment consists of the prolonged administration of penicillin and, if necessary, surgery. Till date, about 32 cases of primary actinomycosis of breast have been reported with *Actinomyces israelii* being the commonest isolate. Other species of *Actinomyces* that have been isolated from such rare cases include *A. viscosus*, *A. europaeus*, *A. turicensis* and *A. radingae*⁽¹⁴⁾.

Our case lack history of obvious local trauma and other related systemic diseases. Absence of suggestive symptoms and signs along with a normal chest skiagram excluded pulmonary involvement.

In our case, the possible mode of infection is through lactation. *Actinomyces* sp. colonize human oral cavity in early childhood. Sarkonen et al showed that prevalence rates of total Actinomycetes flora increases from 31% to 97% within first 2 years of life⁽¹⁰⁾.

Our case also demonstrated that a catalase-negative *Actinomyces* can cause granulomatous inflammation which may seem in contrary to usual observation of association of granulomatous inflammation with catalase-positive microorganisms. But actually it may not be, since we were able to isolate a catalase-positive co-pathogen *Staphylococcus aureus*. Studies have demonstrated presence of companion bacteria in actinomycotic lesions which act as co-pathogens and lowers oxygen tension at tissue level for *Actinomyces* sp. to survive and multiply^(4,8). *Staphylococcus* sp. is one of these companions along with *Actinobacillus* sp., *Fusobacterium* sp., *Bacteroides*, *Hae mophilus* sp., *Eikenella* sp. Etc^(1,4,5,6).

Culturing *Actinomyces* from primary actinomycosis breast has been possible in 50% of the cases⁽¹⁴⁾. We were successful in culturing *Actinomyces israelii*. Fortner's method⁽³⁾ of partial anaerobic culture for *Actinomyces*, where inoculation of clinical material is done along with a heavy inoculum of *Serratia marscecens* on blood agar in a glass petridish sealed with plasticine or paraffin-film, was found to be an easy, low-cost and effective method. It also allowed frequent inspection without disturbing the low oxygen environment of the system.

CONCLUSION: Breast abscess with multiple sinuses should raise a possibility of actinomycotic involvement apart from other causes. It needs proper judgement, skill and most importantly, patience, since culture isolation is not so prompt and easy. The microbiologist must be cautious in this regard as proper management depends on the report. The mind must know what the eyes are seeing!

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