

# Granulomatous Mastitis – A Rare Presentation of Atypical Mycobacterial Infection

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## ABSTRACT

We report a rare case of granulomatous mastitis (GM) caused by nontuberculous mycobacteria (NTM). The causative microbe was rapidly growing atypical mycobacteria species – *Mycobacterium chelonae* and *abscessus* complex. The patient presented with tender breast swelling and underwent trucut biopsy after 2 weeks of failed treatment with antibiotics. The histopathology showed GM and acid-fast bacilli. Based on the pathology report, antitubercular treatment was started without any further delay, while waiting for the culture report. However, the culture grew atypical mycobacteria (NTM) resulting in change of the drugs. Identification of the causative organism plays a major role as the patient has to receive appropriate treatment.

**Key words:** Granulomatous mastitis, *Mycobacterium chelonae* and *abscessus* complex, nontuberculous mycobacterial mastitis

## INTRODUCTION

Nontuberculous mycobacteria (NTM) are referred to as atypical mycobacteria, environmental mycobacteria, and mycobacteria other than tuberculosis (MOTT).<sup>[1]</sup> Rapidly growing mycobacteria are established pathogens and one of the four species of atypical mycobacteria.<sup>[2]</sup> They are ubiquitous in the environment found in soil, dust, and water, able to infect both immunocompetent and immune-compromised persons.<sup>[3,4]</sup> *M. chelonae* and *Mycobacterium abscessus* group are rapidly growing mycobacteria that can cause pulmonary, skin, and soft tissue infections. *M. chelonae* and *M. abscessus* group are a very rare cause of mastitis with very few reported cases.<sup>[5]</sup> We report a case of granulomatous mastitis (GM) in a healthy female of reproductive age group from a non-endemic region (Oman) for mycobacterial infections. This is probably the first case report of atypical mycobacterial infection presenting as GM from this region.

## CASE REPORT

A 33-year-old lady presented with breast swelling, fever, and pain for 1 week. She was treated with a course of antibiotics without any benefit before attending our hospital from a local health center. The fever had no particular pattern, no cough, or weight loss. Local examination showed diffuse enlargement of the breast with tenderness, redness, and warmth. Peau d'orange, discharging sinus, or nipple discharge were negative. No palpable lymph nodes in the axilla. Her history was nothing contributory other than tubectomy and one C-section. The patient was again prescribed a course of antibiotics, referred to ultrasonographic (USG) examination, and to follow-up after a week. USG showed diffuse inflammation of the breast with streaks of fluid without obvious collection and few benign lymph nodes in the axilla. As there was no improvement noticed by the patient and the surgeon, a trucut biopsy of the breast and punch biopsy from the inflamed skin were done. Histopathological examination showed breast tissue with granulomas, mixed inflammation with foci of

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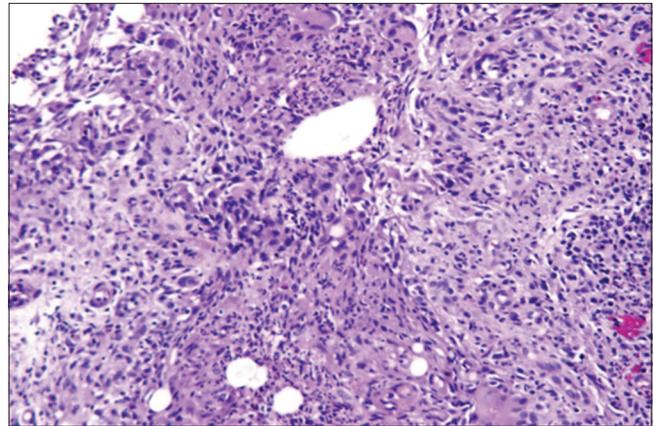
necrosis, and few cystic spaces lined by neutrophils [Figure 1]. Ziehl–Neelsen and Gram’s staining were done, which showed acid-fast bacillus (AFB) and Gram-positive bacilli, respectively [Figures 2 and 3]. A diagnosis of granulomatous inflammation with mixed infection suggestive of tubercular mastitis and cystic neutrophilic GM (CNGM) was rendered. The culture was suggested to confirm tubercular mastitis and to rule out atypical mycobacteria.

Antitubercular treatment (ATT) was started based on the pathology report while awaiting culture result. The culture showed growth of rapidly growing atypical mycobacteria. Specific species identified were *M. chelonae* and *M. abscessus* complex. ATT was discontinued immediately (which she received for 9 days) and switched over to combination therapy consisting of clarithromycin and amikacin. By now, the patient’s condition deteriorated with increased breast size, tenderness, and developed discharging sinuses at the biopsy sites. Repeat USG showed few tiny pockets of communicating fluid collections in the breast parenchyma with thickened skin and normal nipple. Her biochemical investigations showed a raised C-reactive protein. The erythrocyte sedimentation rate was not done. The patient requested a referral to a different hospital and discontinued at our center.

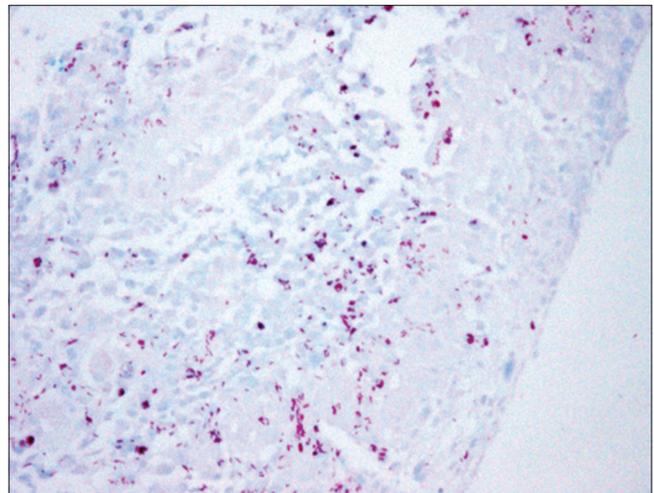
With the help of our infectious department team, we got to know that the patient has shown a near complete recovery. Now, more than 3 months since diagnosis, most of the sinuses are healed and the breast swelling has reduced. She received a third drug linezolid in addition to the above mentioned two drugs from the referral hospital.

## DISCUSSION

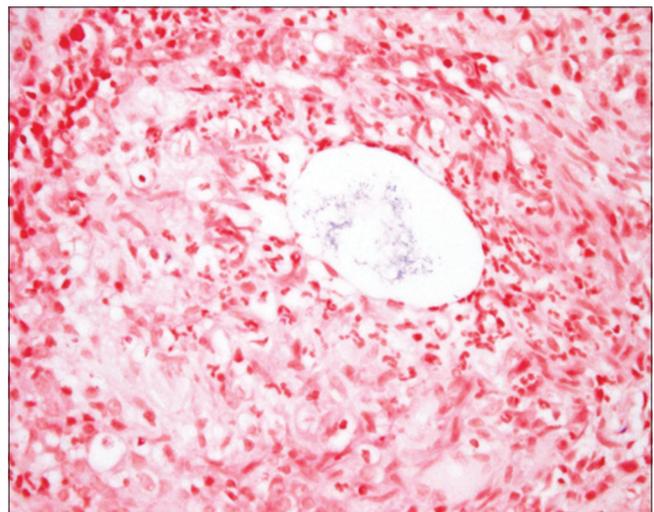
Atypical mycobacteria are ubiquitous in the environment both in soil and aqueous sources. They can remain nonpathogenic inhabiting the body surfaces.<sup>[3]</sup> Various infections caused by them are pneumonia, lung abscess, lymphadenitis, skin, and soft tissue (post-traumatic or post-surgical), gastrointestinal, meningeal, bone and joint, disseminated, and intravenous catheter infections.<sup>[2]</sup> *M. abscessus* and *M. chelonae* both cause skin and soft tissue infections usually post-traumatic or post-injection abscesses.<sup>[3,4]</sup> A localized cutaneous form following trauma and disseminated forms in immunocompetent persons can be seen with *M. abscessus* and *chelonae*.<sup>[2]</sup> Diagnosis is by simple AFB staining (ZN), culture, and genome detection by polymerase chain reaction (PCR).<sup>[1]</sup> Isolation of the *Bacilli* in a shorter time period in the Lowenstein–Jensen culture medium is a clue to suspect NTM.<sup>[5]</sup> More than one culture have to be done as *M. abscessus* and *chelonae* survive harsh decontamination procedures and are able to contaminate the growth media.<sup>[3]</sup> As our patient had no history of surgical procedures (injections or aspiration), most probably, post-traumatic cutaneous



**Figure 1:** Breast tissue shows granulomas with giant cells, cystic spaces lined by neutrophils and inflammation (hematoxylin and eosin stain, ×40)



**Figure 2:** Zeihl–Neelsen stain showing many rod-shaped acid-fast bacillus (×20)



**Figure 3:** Gram’s stain showing Gram-positive bacilli in the cystic space (×40)

infection can be suggested. ZN staining was positive for AFB aiding in diagnosis. Culture grew rapidly growing atypical mycobacteria. PCR was not done. The treatment is with antimicrobials and surgical drainage.<sup>[3]</sup> Combination therapy consisting of 2–3 drugs usually clarithromycin combined with linezolid, imipenem, amikacin, ceftazidime, or fluoroquinolones is reported to be effective against rapid growers for a period of 3–6 months.<sup>[5]</sup> No definite-time period is mentioned and the treatment may last for more than 6 months.<sup>[6]</sup> Our patient has completed 3 months of combination antimicrobial treatment without surgical intervention. Surgical procedures could result in diffuse and ugly scarring. The biopsy showed cystic spaces lined by neutrophils with Gram-positive bacilli in the cystic spaces. A granulomatous response with Gram-positive bacilli in cystic spaces is known to be associated with corynebacterium infection.<sup>[7]</sup> This needs to be culture proven and the condition is referred to as CNGM. Corynebacterium culture is difficult as they are highly fastidious microbes. However, corynebacterial infections are treated with lipophilic antimicrobials. Clarithromycin is a lipophilic drug and was one of the multidrugs included in our patient's drug profile. Hence, although not proven by culture, the treatment of CNGM has also been taken care.

## CONCLUSION

Our case highlights that not all cases of lobular GM are self-limiting idiopathic disease; instead, it could be a rare case of atypical mycobacterial infection. Atypical mycobacteria need to be ruled out in spontaneous breast infections with no response to the usual antimicrobials in healthy females. Appropriate diagnosis and treatment are very essential as this is a treatable condition with complete recovery.

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