Cystic Neutrophilic Granulomatous Mastitis with Corynebacterium and Pseudomonas species: A comparison

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Introduction

Cystic neutrophilic granulomatous mastitis (CNGM) is an uncommon subtype of granulomatous mastitis (GM) affecting young to middle-aged parous women. Correct diagnosis is often delayed due to its rarity and wide differential diagnoses including invasive carcinoma, tuberculosis, fungal infection, sarcoidosis and foreign-body type reaction. Misclassification as idiopathic lobular GM is common and likely due to lack of awareness about this relatively new entity, as well as poor visualization and difficulty growing of the fastidious and elusive Corynebacterium species; a key feature of this unique disease. Although there is no universally accepted diagnostic criteria, granulomatous inflammation with epithelioid histiocytes and neutrophil rimmed lipid vacuoles containing gram-positive rod shaped bacteria, are strongly suggestive of CNGM. However, other infectious causes exist and should be considered in the absence of Corynebacterium. Here we present two cases of this rare entity with Corynebacterium and Pseudomonas species, including a brief review of the literature.

Methods

Case 1 is 42-year-old woman with a past medical history of left breast mastitis and abscess treated with incision and drainage (I&D) six months prior, presented with fever and worsening right breast pain and swelling without improvement on oral antibiotics treatment without improvement. Ultrasound revealed suspected abscess with I&D, excisional biopsy, and cultures sent to pathology for evaluation. Gram stain was performed.

Case 2 is a 33-year-old ESRD on hemodialysis female with a 4-month history of right breast pain, erythema, and swelling that became worse in the prior month. Imaging reported multiple right breast abscesses and I&D with biopsy, and culture was done. Gram stain was performed.

Results

On H&E, Case 1 and Case 2 both revealed a histopathological pattern distinctive of CNGM. This includes granulomatous inflammation with multinucleated giant cells (Figure 1), as well as formation of clear lipid-filled cyst-like spaces or vacuoles surrounded by a rim of neutrophils (Figure 2).

Gram stain performed on Case 1 showed rare, gram-positive rod shaped organisms within the lipid filled vacuoles (Figure 3a and 3b.) suggestive of Corynebacterium species.

Gram stain performed on Case 2 was negative for microorganisms (Figure 3.) but culture identified Pseudomonas species.

Figures

Case 1

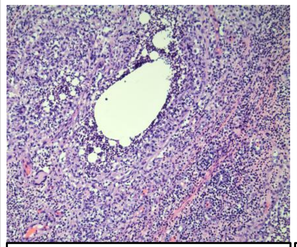

Figure 1. 40x H&E Granulomatous inflammation with multinucleated giant cells and lipid vacuoles.

Case 2

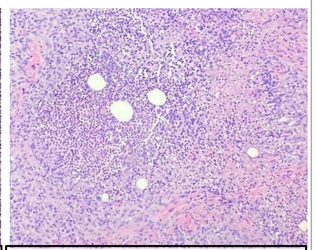

Figure 1. 20x H&E Granulomatous inflammation with multinucleated giant cells and lipid vacuoles.

Results (Continued)

Case 1

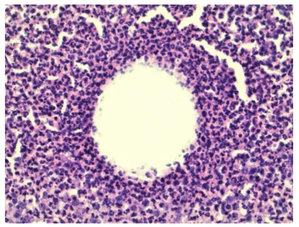

Figure 2. 40x H&E Lipid vacuoles rimmed by neutrophils.

Case 2

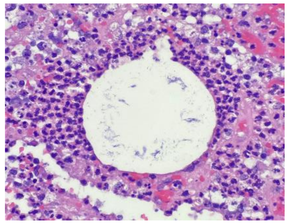

Figure 2. 40x H&E Lipid vacuoles rimmed by neutrophils.

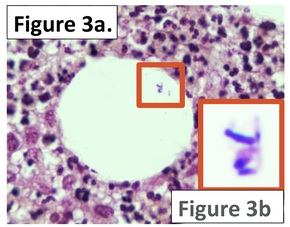

Figure 3. GRAM stain at 60x (3a.) and 100x oil immersion (3b.) showing rare gram positive rod-shaped organisms.

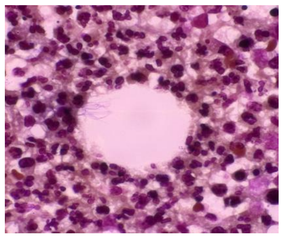

Figure 3. GRAM stain at 60x showing negative microorganisms.

Conclusions

CNGM is a rare but increasingly recognized cause of mastitis, often associated with Corynebacterium ssp. infection. It has a distinct histopathological pattern with Gram-positive Corynebacterium spp. within the vacuoles. Corynebacterium are a component of normal skin flora and it has been postulated that the it may gain entry to breast tissue via lactiferous ducts during lactation and following breast trauma, including nipple piercing and breast core biopsy. It can be very difficult to identify, with prolonged antibiotic therapy required. Therefore, clinical suspicion of CNGM with Corynebacterium is essential to ensure the use of appropriate medium for evaluation.

However, other infectious causes exist and should be considered in the absence of Corynebacterium spp.. Pseudomonas has been identified as the most common bacterial cause of granulomatous mastitis in general. Therefore, proper investigation of other infectious agents with special stains and cultures should be performed to ensure proper medical management.

Diagnosis of CNGM often missed or delayed due to its rarity and many potential mimickers. Radiology findings are usually not reported, but if available, includes abscess, edema on ultrasound, and masses/asymmetry on mammogram. A high index of suspicion among pathologists and clinicians managing patients with breast disease is essential for accurate diagnosis and appropriate management of CNGM.

References & Acknowledgements

To view the reference list, please scan the QR code below.