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REVIEW

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Mondor's disease of the breast in Asia: a forgotten diagnosis for the front-line clinicians—a case report and literature review

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Abstract

Background Mondor's disease is a rare, however, benign and self-limiting condition which can occur in the breast. Diagnosis of Mondor's disease of the breast is not frequently made as this may be due the lack of awareness on this condition. Although majority of the documented cases were described to be idiopathic, it is wise to properly investigate for other causes such as hypercoagulability or an underlying malignancy. Only nine available case reports pertaining Mondor's disease of the breast in Asia since 2011 were identified and reviewed.

Main body of abstract We present a case of a 47-year-old, Malaysian, Malay, female who presented with a right breast swelling which was referred to the Radiology Department to rule out breast abscess. However, further clinical examination and radiological assessment proved it to be superficial thrombophlebitis of the lateral thoracic vein along the upper outer quadrant of her right breast. This condition was attributed to hypercoagulable state secondary to her newly diagnosed diabetes mellitus. After strict diabetic control, short course of antibiotics and symptomatic treatment, complete resolution of signs and symptoms were noted during her follow-up 6 weeks later.

Short conclusion Mondor's disease of the breast is believed to be more common than reported as many patients and even clinicians may not pay close attention to this complaint due to the mild symptoms of pain and its self-limiting nature. Raising awareness of the disease through this case review will broaden the scope of differential diagnosis for front-line clinicians when approaching patients presenting with a breast lump.

Keywords Mondor's disease, Superficial thrombophlebitis, Breast, Diabetes mellitus, Asia

Background

Mondor's disease is a condition which is defined by superficial vein thrombophlebitis that can occur in any part of the body such as breast, upper arm, abdomen, groin and penis [1–5]. It is a rare condition, with breasts being the commonest location for this seemingly infrequent disease occurrence. The incidence rate for Mondor's disease of the breast have been reported to be less than 0.8% [6].

The rarity of this disease may be attributed to the lack of awareness regarding this condition by healthcare professionals, hence causing it to be underdiagnosed. A total of nine cases of Mondor's disease involving the breast have been gathered in Asia since 2011, with only one case reported in Malaysia. It is undeniable that awareness regarding this condition is low as it is also underreported.

Below we discuss on a case of a middle-age, Malaysian, female, who presented with a right breast swelling that was mildly tender on palpation with overlying skin erythema. She was also diagnosed with diabetes mellitus during presentation. The patient was initially treated for breast abscess by the front-line clinician in

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the Emergency Department and subsequently referred to the Radiology Department for further sonographic assessment.

Main text

Case presentation

A 47-year-old, Malaysian, Malay, female, with no known past medical or surgical history presented to the Emergency Department with complaints of right breast swelling associated with itchiness, soreness and fever for three days' duration. Otherwise, she denied any history of trauma, insect bites or recent surgical procedures. She denied family history of haematological disorders or autoimmune conditions.

Upon assessment, the patient was alert, conscious and not septic looking. She recorded normotensive blood pressure with normal heart rate. However, there was documented low-grade fever of 37.7 °C with high random blood glucose levels recording 17.6 mmol/L. Physical examination by the front-line clinicians in the Emergency Department noted a mildly tender, palpable breast lump at the upper outer quadrant of right breast. Mild overlying skin erythema was noted. No regional lymphadenopathy was documented.

Biochemical panel revealed increased inflammatory markers, with white blood cell count of 12.0×10^3/uL and C-reactive protein of 159.4 mg/L. Further investigations were sent to investigate patient's high blood glucose status. Urine ketone was negative with normal venous blood gas reading, hence ruling out possibility of diabetic ketoacidosis. Other laboratory results showed high fasting blood glucose of 11.0 mmol/L and HbA1C of 9.0%, which confirmed the diagnoses of diabetes mellitus. Referral was made to the Radiology Department for further ultrasound assessment to rule out the possibility of breast abscess.

Physical examination of the breast by a radiology clinician noted cord-like swelling over the right upper outer quadrant. Ultrasound revealed a non-compressible, hypoechoic, beaded tubular structure along the right upper outer quadrant as shown in Fig. 1. Intraluminal echogenicity was visible with the absence of colour Doppler flow within. There was minimal surrounding soft tissue hyperechogenicity and peripheral vascularity depicted, in keeping with local inflammatory changes. Otherwise, no concerning mass was identified in the right breast. Mammography performed for completion of assessment demonstrated superficial tubular density along the right upper outer quadrant which corresponds to the ultrasound findings (Fig. 2).

Overall history, physical examination, biochemical profile and radiological findings were suggestive of Mondor's disease of the right breast, likely secondary to uncontrolled diabetes. She was also treated for right breast mastitis. Patient was admitted for intravenous antibiotics and tight diabetic control. Her fever subsided quickly with resolution of overlying right breast skin erythema. Thereafter, she was discharged with oral antibiotics and anti-inflammatory drugs for five days' duration. Aside from that, she was prescribed with an oral hypoglycaemic agent and insulin injection regime to ensure well-controlled blood glucose levels. During outpatient clinic follow-up, patient was well with no active complaints. Her blood glucose level was also well controlled. Ultrasound scan for reassessment after six weeks post-treatment revealed resolved thrombophlebitis of the superficial right lateral thoracic vein of her right breast as demonstrated in Fig. 3.

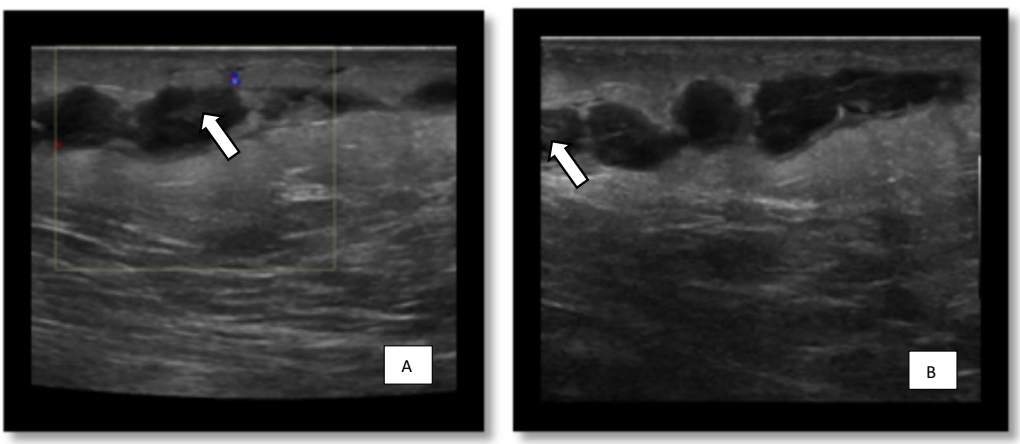

Fig. 1 **A, B** Ultrasound scan of the concerned palpable swelling in the right upper outer quadrant which revealed a beaded appearance of the superficial vein (right lateral thoracic vein) with intraluminal echogenicity within (arrow). The absence of internal colour Doppler flow was demonstrated

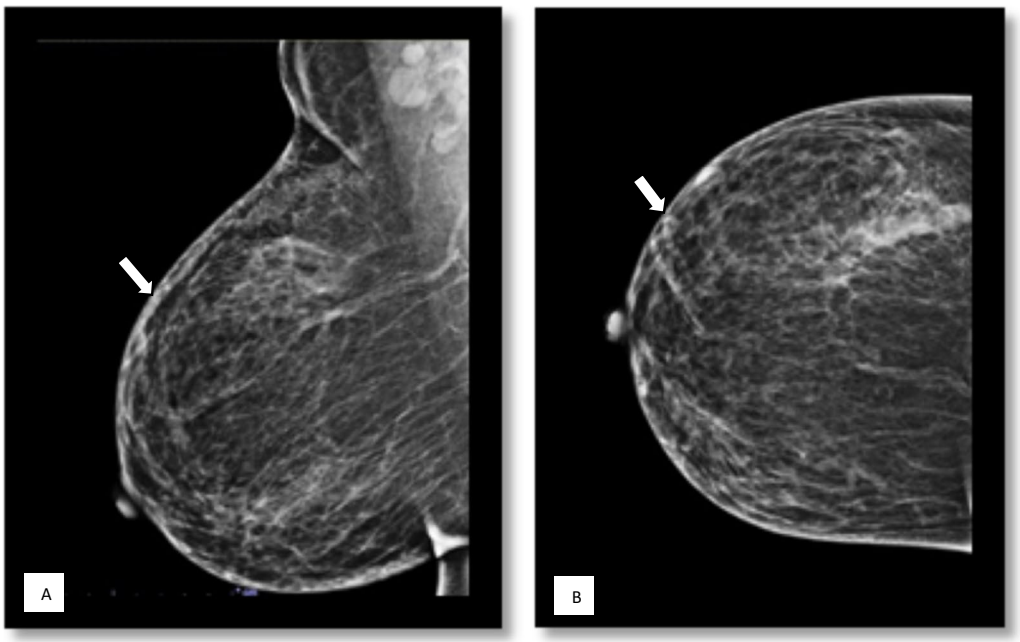

Fig. 2 Right mammogram [**A** Mediolateral oblique, **B** Craniocaudal] demonstrates superficial tubular density in the upper outer quadrant (arrow) which corresponds to the patient's sonographic and clinical findings. No associated concerning calcification or distortion identified

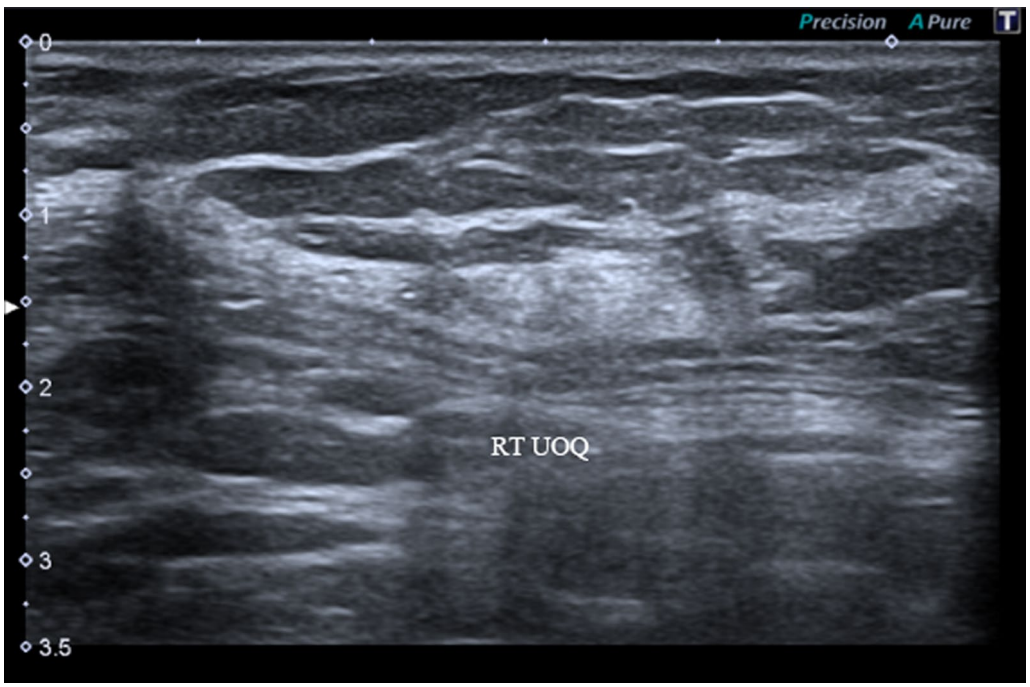

Fig. 3 Repeat ultrasound of the upper outer quadrant of right breast which confirmed the absence of previously seen beaded appearance of the superficial lateral thoracic vein. This confirmed resolution of the superficial thrombophlebitis of her right breast

Discussions

Mondor's disease was first introduced in 1939 by Henri Mondor, who described this condition as thrombophlebitis involving superficial veins of the anterior chest wall. Following that, Mondor's disease have also been documented to involve other areas, such as abdomen, penis, groin and upper limb [1–5]. In this case review, we will be focusing on Mondor's disease of the breast. This condition has been reported to mostly involve the thoracoepigastric, superficial epigastric and lateral thoracic veins [6]. In our patient, right lateral thoracic vein was affected.

Mondor's disease can be further subdivided as primary and secondary Mondor's disease. Primary Mondor's disease of the breast is described as superficial thrombophlebitis of unknown cause, recording about 50–60% of all cases of breast Mondor's disease [6]. Six out of nine cases reviewed in Asia since 2011 demonstrated no specific cause to explain the presentation of superficial thrombophlebitis of the breasts in their patients and, hence, concluded as primary Mondor's disease [1, 7–11].

On the other hand, secondary Mondor's disease of the breast have been reported to be attributed to direct trauma, iatrogenic post-surgical procedures, local inflammation or a hypercoagulable state [8]. The definite pathogenesis of Mondor's disease is not well documented. However, it has been hypothesised that trauma, blood stasis, inflammation, hypercoagulability and underlying cancer may be precursors for the formation of thrombosis within the affected superficial vein. Subsequent adjacent fibroblastic proliferation of subcutaneous tissue was believed to cause the thrombosed vein to adhere closely to the overlying skin, leading to skin retraction and hence a characteristic palpable cord-like swelling [6].

Two case reports in Saudi Arabia and Oman presented on hypercoagulopathy being the cause for superficial thrombophlebitis of the breast in the reported patients. The similarity between these two literatures was that the female patients consumed oral contraceptive pills for approximately two-week duration prior to presentation. Upon discontinuation of the oral contraceptives, complete resolution of the condition was observed during follow-up, four to six weeks later [12, 13].

In our patient, blood investigations confirmed that she had diabetes mellitus, which was taken as a risk factor predisposing her to a hypercoagulable state, hence leading to the occurrence of superficial thrombophlebitis of the right breast.

Chronically high level of blood glucose is known to cause many detrimental effects. Hyperglycaemia is believed to cause activation of coagulation system, leading to a hypercoagulable state, hence increasing risk of developing venous thrombosis. This condition is further aggravated by concurrent infection or inflammation. A case–control study on risk of venous thrombosis in diabetic patients by Hermanides et al. described that superficial thrombophlebitis were seen in 12% of patients while a majority of patients (82%) had deep venous thrombosis and a minority (6%) had calf vein thrombosis. This shows that in any case of superficial thrombophlebitis, it is wise for clinicians to consider diabetes mellitus as a contributing factor and not miss this diagnosis as well [14].

Patients classically present to the Emergency Department with complain of painless or mildly painful cord-like structure within the superficial layers of the breast, corresponding to the affected vessel. It is often associated with erythema and oedema. All the nine cases of Mondor's disease of the breast gathered in Asia since 2011, described similar complaint of a cord-like superficial swelling over the breast. In our patient, she initially presented with a complain of mildly painful right breast lump. On further history taking, patient described this lump as a palpable, long, cord-like structure in her right upper outer breast.

Mondor's disease of the breast can typically be diagnosed by thorough history taking and clinical examination with confirmation by ultrasonography as well as supplementary mammography.

On ultrasound imaging, typically there will be evidence of a long, hypoechoic or anechoic, tubular, thrombosed vessel, sometimes beaded in appearance, seen on the subcutaneous layers of the breast. In our patient as well as described in the other case reviews, a tubular hypoechoic structure with the absence of venous flow on colour Doppler study can be identified, as demonstrate in Fig. 1 [6, 8]. Occasionally, it may be associated with surrounding soft tissue inflammatory changes. Further assessment by mammography may often be futile as it frequently reveals normal findings in a large proportion of patients. Sometimes, superficial tubular structure can be visualised as shown in Fig. 2, which was observed in our case [6].

Radiological imaging is not only important to confirm the diagnosis of Mondor's disease, but also crucial in identifying other possible causes for it, such as an underlying malignancy. Wong et al. described a case whereby diagnosis of Mondor's disease was made purely based on clinical history and examination. No complementary ultrasound or mammography was done for the patient to confirm the diagnosis of Mondor's disease of the breast. During subsequent follow-up five months later, routine mammogram was ordered which unfortunately demonstrated the presence of a highly suspicious breast mass. This mass which was later biopsy proven to be invasive ductal carcinoma with metastasis to the ipsilateral lymph nodes [15]. Thus, it is imperative for clinicians to perform thorough clinical examination and early breast imaging for all patients presenting with breast complaints to avoid

delayed diagnosis and mismanagement which may lead to devastating complications if missed.

All studies in Asia since 2011 confirmed that Mondor's disease is generally a self-limiting condition which resolves spontaneously within four to six weeks. Often, only symptomatic treatment with anti-inflammatory analgesics are prescribed [8]. In a retrospective unblinded study by Shirah et al., analysis on the effectiveness of oral diclofenac sodium versus topical patch of diclofenac sodium was done on 172 women with Mondor's disease from 2001 to 2010. This study revealed that diclofenac sodium patch was more effective in pain relieve and fast regression of Mondor's disease of breast as compared to oral agents [16].

With regard to this case report, the patient was treated with anti-inflammatory drugs, analgesics and antibiotics as well as insulin injection and oral hypoglycaemic agents. She showed complete resolution of signs and symptoms after six weeks during follow-up.

Conclusions

In conclusion, even though Mondor's disease of the breast has been described in literature for more than 100 years, this condition is still known to be a rare entity. A plausible reason for this could be due to this condition being underdiagnosed and, hence, underreported because of the lack of awareness on it.

Mondor's disease of the breast is not commonly considered as a differential diagnosis for patients presenting with breast complaints. Majority of patients presenting to the Emergency Department with focal breast swelling alongside with non-specific inflammatory symptoms are commonly investigated and treated for mastitis or breast abscess. As in this case report, Mondor's disease of the breast was not a diagnosis under consideration of the primary team, however, was only identified by the radiology clinician upon further history taking, clinical examination and ultrasonography. Additionally, upon diagnosis of Mondor's disease of the breast, front-line clinicians must never immediately dismiss it as an idiopathic presentation because it is important to rule out possible causes such as hypercoagulable state as described in our case. Another important and distressing condition which all clinicians would never want to overlook is an underlying breast malignancy.

Developing awareness on this condition, its causes, risk factors and radiological findings as detailed in this manuscript would prove beneficial for clinicians to avoid delay in diagnosis and mismanagement as well as provide reassurance to patients.

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PV was the main author in writing the manuscript and compiling information. FTP helped in writing the manuscript and grammar correction. All authors have read and approved the final manuscript.

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Declarations

Ethics approval and consent to participate
Patient gave informed consent to use their data in case write up. No ethics approval is needed from the Hospital Queen Elizabeth II, Sabah, Malaysia, for a case write up.

Consent for publication
Written informed consent was obtained from the patient for publication of this case report and accompanying images.

Competing interests
The authors declare that they have no competing interests.

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