CASE REPORT

Breast filariasis or inflammatory breast carcinoma? Reaching a diagnosis

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SUMMARY

Lymphatic filariasis is a neglected tropical disease caused by the parasite *Wuchereria bancrofti*. Involvement of the breast is relatively uncommon and may affect only 10% of individuals with lymphatic filariasis. We present a case of an elderly woman with breast pathology. The condition clinically appeared as a malignancy. Routine diagnostic tests including tissue histopathological studies failed to clinch a diagnosis. However, a serological test helped in identifying the pathology, which turned out to be a manifestation of breast filariasis, and aided in instituting appropriate therapy for this condition.

BACKGROUND

Lymphatic filariasis, also known as elephantiasis, is considered a neglected tropical disease. It is caused due to infection caused by *Wuchereria bancrofti* (most commonly), a parasite transmitted into the blood stream by the bite of a female culex mosquito and subsequently passes into the lymphatic system. The manifestations resulting from involvement and obstruction of the lymphatic system vary from acute dermatolymphangioadenitis to lymphoedema and elephantiasis. Lymphoedema of various parts of the body, such as the limbs, genitalia and breasts, results in grotesque deformities, which are a source of pain and embarrassment, besides being a social and financial burden to the affected individual. According to the 2015 WHO factsheet, over 120 million people are infected, and about 40 million disfigured and incapacitated, by lymphatic filariasis.¹ Inflammatory breast cancer is a rare but rapidly proliferating breast malignancy presenting as an erythematous breast without a palpable breast mass, similar to filariasis of the breast. Considering the uniformly poor prognosis of the malignancy, as well its different strategy of management, it is necessary to reach an early accurate diagnosis in such a situation. We present a case where lymphatic filariasis of the breast mimicked an inflammatory carcinoma and confounded the management.

an ill-defined lump in the centre and a few enlarged axillary lymph nodes on the left, in the anterior axillary fold. Considering the age of the patient and clinical findings, a provisional diagnosis of inflammatory breast carcinoma was made.

INVESTIGATIONS

A mammography revealed thick and coarse trabecular markings as well as diffuse skin thickening of the left breast (figure 2A). The mammogram of the right breast was found to be normal. An ultrasoundogram of the left breast revealed diffuse thickening of the skin and periareolar region, and increased hypoechogenicity of retroareolar breast parenchyma with evidence of diffuse subcutaneous oedema. An enlarged axillary lymph node was also detectable in the ultrasonogram. So the suspicion of inflammatory breast carcinoma was further strengthened. However, a fine-needle aspiration cytology (FNAC) study from the left breast did not reveal any significant findings while the FNAC of the left axillary lymph node revealed reactive hyperplasia of the lymphoid cells. The patient was then subjected to an incisional biopsy of the left breast over the lesion. Histopathology study revealed inter-anastomosing slit-like spaces lined by spindle cells and ropy collagen in the intervening spaces (figure 2B). In addition, several scattered stellate-shaped cells, patchy mild lymphoplasmacytic infiltrates and foci of vascular proliferation were recorded. A pathological diagnosis of pseudoangiomatous stromal hyperplasia of the left breast was made. In addition, immunohistochemistry results for estrogen receptor (ER), progesterone receptor (PR), CD34 and CD31 were negative. As the investigations failed to confirm a diagnosis of malignancy, an alternative diagnosis was considered. Since the patient came from a filarial endemic zone, an Og4C3 test for filarial antigen in a serum sample was ordered. A positive value of 8192 antigen units (AU) (reference value: up to 128 AU

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CASE PRESENTATION

A 73-year-old woman presented to the surgical outpatient department, with a slow growing swelling of the left breast for the past 3 months and pain with redness over the breast for the last 10 days. Inspection revealed redness over the left breast with a peau d'orange appearance of the skin (figure 1). There was, however, no ulceration over the breast and no discharge from the nipple. Palpation revealed mild tenderness over the breast,

Figure 1 (A and B) Left breast showing redness and a peau d'orange appearance.

Figure 2 (A) Mammogram of the left breast revealing thick and coarse trabecular markings as well as diffuse skin thickening with normal right breast. (B) High-power microscopic study of the left breast tissue reveals slit-like spaces lined by spindle cells and ropy collagen in the intervening spaces, with several scattered stellate-shaped cells and patchy mild lymphoplasmacytic infiltrate.

is considered normal) confirmed a diagnosis of lymphatic filariasis of the left breast. A final diagnosis of acute filarial dermatolymphangitis of the left breast, with ipsilateral axillary adenopathy, was made.

DIFFERENTIAL DIAGNOSIS

Owing to the advanced age of the patient, alongside the ill-defined slow growing breast swelling with evidence of inflammation on the surface, a differential diagnosis of breast cellulitis, inflammatory breast carcinoma, breast cellulitis and filariasis of the breast was considered. Breast cellulitis could be easily ruled out as there was no history of fever or undue tenderness on breast examination. However, a diagnostic differentiation between malignancy and a benign filariasis posed a challenge. While the biopsy for evidence of malignancy proved negative, a positive ELISA for filarial antigen helped in clinching the diagnosis.

TREATMENT

Therapy was decided based on standard treatment regimen for filariasis. The patient was started on a regimen of diethylcarbamazinecitrate at a dosage of 6 mg/kg/day for 4 weeks. A course of antibiotic (ofloxacin tablets, 400 mg two times a daily for 7 days) was added into the regimen. The patient was advised follow-up visits at 1, 3, 6 and 12 weeks. At the end of the 3-week clinical assessment, a reduction in the redness and peau d'orange appearance was noted. At the end of 12 weeks, the patient was relieved of the heaviness and pain in the left breast while the peau d'orange appearance, redness and left axillary lymphadenopathy disappeared completely (figure 3).

Figure 3 (A and B) Left breast after therapy showing reduction of redness and disappearance of the peau d'orange appearance.

OUTCOME AND FOLLOW-UP

With the start of antifilarial therapy, the patient rapidly improved symptomatically. Three weeks after therapy, the inflammatory redness and peau d'orange appearance decreased considerably. Six weeks post-therapy, the patient was completely free of symptoms; however, a minor discrepancy of breast size (left bigger than right) persisted—the patient was, however, not bothered by this.

DISCUSSION

Lymphatic filariasis is a neglected tropical disease caused most commonly by the parasite *W. bancrofti* and transmitted by the female culex mosquito. Involvement of the breast in lymphatic filariasis is relatively uncommon.² Few studies have demonstrated the role of FNAC and biopsy in confirming the presence of this parasite. Ultrasonogram of the breast and mammography have been reported to detect the parasite as well.³ Inflammatory carcinoma of the breast is a rare, highly malignant and fatal form of breast cancer. Clinically, it is characterised by rapid onset of symptoms including erythema, peau d'orange appearance of skin and breast oedema with presence of axillary lymphadenopathy. Involvement and obstruction of the dermal lymphatics with tumour emboli results in the typical clinical picture described above.

Tissue diagnosis reveals dermal and stromal tumour emboli, a finding that is considered a hallmark of inflammatory breast cancer (IBC).⁴

In spite of the very similar clinical picture of both these conditions, few literature reports of one condition mimicking the other can be found.⁵ However, early diagnosis in those cases could be reached by demonstration of filarial parasite in the

breast tissue or nipple secretions.^{2 5} Our report, nonetheless, demonstrates the possibility of non-demonstration of the parasite in tissue studies. In such a situation, an Og4C3 filarial antigen assay could help in confirming the filarial pathology. Quantitative filarial antigen levels can be detected by the commercial kit, Og4C3 ELISA (TropBio, Townsville, Australia). This technique helps in detection of circulating filarial antigen (CFA) in serum and plasma, as well as in other body fluids. Studies have satisfactorily demonstrated the high sensitivity of the Og4C3 ELISA assay in positive individuals.^{3 6} Many studies have shown the specificity of Og4C3 testing to be around 95%. Demonstration of high titres of CFA using the Og4C3 ELISA assay, as seen in this patient, is suggestive of active *W. bancrofti* infection.^{6 7} The interpretation of tests may change with regard to the patient's epidemiology, depending on whether the patient belongs to an endemic or non-endemic zone. False-positive results may be seen with low titres below 128 AU and when the patient is suffering from nematode infestation. To the best of our knowledge, ours is the first reported case of filariasis of the breast mimicking an inflammatory carcinoma, where the diagnosis was reached by demonstrating CFA using the Og4C3 ELISA assay. The Og4C3 ELISA assay test can also be used as a cure marker. The serum values of CFA gradually decrease after 3–6 months of treatment. The test can be repeated every 6 months to monitor the prognosis of the disease. In this patient, however, symptomatic relief and clinical resolution of the inflammatory findings helped determine the cure. Considering the ease of conducting this test and convenience of performing it on blood collected any time of the day, as well as the high sensitivity, the Og4C3 ELISA assay can be considered the main initial investigative tool to ascertain filariasis of the breast.

Patient's perspective

(Translated from Odia Language)

I was initially very worried when I developed swelling and redness of my left breast. When I shared my symptoms with my relatives, most of them suspected it to be breast cancer. I consulted few doctors and they conducted multiple tests on me. This further increased my fears. I felt really afraid. The doctors then suggested this test for filaria. My fears were decreased when they said that my breast problem was not cancer but due to filaria. I have heard about filaria in my village. Some people have thick swollen legs due to which they cannot walk properly. I did not know it could affect my breast also. The doctors advised me on the medicines to take for filaria, which I took regularly. Three months later, I was relieved that the redness and swelling had come down. I now feel better.

Learning points

- ▶ Lymphatic filariasis of the breast is a condition that may lead to grotesque deformity of the breast when left untreated.
- ▶ Inflammatory carcinoma of the breast is a malignant condition that sometimes mimics a benign lymphatic filariasis of the breast.
- ▶ Histopathological study of the breast tissue helps in differentiating the two conditions.
- ▶ Occasionally, when tissue diagnosis fails to identify breast filariasis, an ELISA test for circulating filarial antigen, the Og4C3 test, will help in diagnosis.
- ▶ Institution of appropriate therapy for early breast filariasis helps in alleviating symptoms as well as provides emotional relief to the patient.

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Contributors All the authors significantly contributed to the preparation of manuscript. DKP provided the clinical data and images, and conceived the case study. DPM and MMM reviewed the literature and drafted the manuscript. All the authors approved the final manuscript.

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