

“Filarial Dance Sign” with Cytological Detection of Microfilaria: Our Experience over a Two-Year Period

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Abstract

Introduction: Filariasis is a major health problem in tropical countries including India. Adult worms of *Wuchereria bancrofti* reside in the lymphatics and cause progressive lymphatic vascular dilation and various pathologic lesions in organs such as lower limbs, spermatic cord, epididymis, testis, retroperitoneum, and rarely in the female breast. Adult worms can be demonstrated on real-time ultrasound (USG) by their characteristic wriggling movements known as “filarial dance sign” (FDS). Demonstration of microfilariae in fine needle aspiration cytology (FNAC) smears of the swellings with FDS play an important role in the prompt diagnosis of the disease.

Purpose: The current study aims to assess the role of FNAC in demonstration of microfilaria in the cases which showed “FDS” on real-time high-resolution ultrasonography.

Materials and Methods: A total number of 13 cases comprising breast swelling, scrotal swelling, and inguinal swelling showing FDS on real-time high-resolution ultrasonography were subjected to USG guided FNAC for demonstration of microfilaria.

Results: Of 13 cases, five cases were of breast swelling, six cases were of scrotal swelling, and two cases were of inguinal swelling. Filarial dance was demonstrated in ultrasonography in all the cases. On FNAC, adult gravid female worm with eggs and microfilaria were seen in one case of breast swelling and one case of scrotal swelling; microfilariae were detected in two cases of breast swelling, five cases of scrotal swelling and both the cases of inguinal swelling.

Conclusion: High-resolution ultrasonography is a useful technique for diagnosing not only inguino-scrotal filariasis but also filariasis of the breast. USG guided FNAC in these patients can lead to prompt diagnosis of the disease and initiation of treatment.

Key words: Breast, Filariasis, Fine needle aspiration cytology, Microfilaria, Ultrasonography, *Wuchereria bancrofti*

INTRODUCTION

Lymphatic filariasis affects more than 120 million people worldwide, and WHO has identified filariasis as the second leading cause of permanent and long-term disability after leprosy.^{1,2}

Lymphatic filariasis in humans is commonly caused by human parasitic nematode *Wuchereria bancrofti* along with

Brugia malayi and *Brugia timori*.¹ Filariasis of skin and subcutaneous tissue is caused by *Onchocerca volvulus* and *Loa loa*.

In endemic areas, filariasis causes a spectrum of diseases including (1) asymptomatic microfilaremia, (2) recurrent lymphadenitis, (3) chronic lymphadenitis with swelling of dependent limb or scrotum (elephantiasis), and (4) tropical pulmonary eosinophilia.³

Adult worms of *W. bancrofti* reside in the lymphatics and cause progressive lymphatic vascular dilation and various pathologic lesions in organs such as lower limbs, spermatic cord, epididymis, testis, retroperitoneum, and rarely in the female breast.^{4,5} A majority of infected individuals in filarial endemic communities are asymptomatic.

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Filariasis of the breast is a rare cause of breast lump.² It is a cause of concern as the patients with breast filariasis presenting with lump are often clinically suspected to be malignant. Genital filariasis in India presents commonly as secondary vaginal hydroceles. Adult filarial worms can be demonstrated in the dilated lymphatics on real-time high-resolution ultrasound (HRUS), by their characteristic wriggling movements known as “filarial dance sign” (FDS).^{6,7} FDS is a diagnostic sign of inguino-scrotal filarial infection and breast filariasis.^{6,8}

Conventional mode of diagnosis of filariasis is by demonstration of microfilaria in peripheral blood smear. Despite high incidence, it is infrequent to find microfilariae in fine needle aspiration cytology (FNAC) smears and body fluids. There have been reports of incidental detection of microfilariae in FNAC smears and body fluids.^{5,9} The current study aims to assess the role of ultrasound (USG) guided FNAC in demonstration of microfilaria in the cases which showed “FDS” on real-time ultrasonography.

MATERIALS AND METHODS

The study was conducted at the Department of Pathology in collaboration with the Department of Radiology at Rama Medical College, Hospital and Research Centre, Mandhana, over a period of 2-years from September 2013 to August 2015.

The study group comprised 13 patients in whom the FDS was seen on real-time HRUS with use of 10 MHz linear probe. Of thirteen cases, five cases were of breast swelling, six cases of scrotal swelling, and two cases were of inguinal swelling. Both the cases of inguinal swellings were male patients.

With the patient in the supine position, all swellings were scanned with high-resolution ultrasonography. Doppler imaging was used to delineate lymphatic vessels/spaces for differentiation from blood vessels. Both real-time video and still images were stored on hard drive with recording of microfilarial movements. All 13 patients were subjected to USG guided fine-needle aspiration, after taking consent. FNAC was done by 22-23 G needle attached to 10 ml syringe. The needles were placed in the areas which had positive findings for the FDS. Alcohol-fixed smears were stained with Hematoxylin and Eosin stain and air-dried smears were stained with Leishman stain. Routine blood counts and peripheral smears for microfilaria were obtained from all patients.

RESULTS

Of 13 cases with “FDS,” five cases (38.5%) were of breast swelling, six cases (46.1%) were of scrotal swelling, and two cases (15.4%) were of inguinal swelling. Clinical details and

age distribution of patients were variables (Table 1). Of five cases of breast swelling, in three cases left breast and in two cases right breast was involved. The patients with breast swelling (four out of five) were predominantly in the fourth decade and were diagnosed clinically as fibrocystic disease. One patient with breast swelling was 55 years old, and clinically a suspicion of malignancy was raised. Of six cases of scrotal swelling, two involved the left side, and four involved the right side. Two patients also had a hydrocele. Both the inguinal swellings were on the left side. Peripheral blood smear and cytologic findings are discussed in Table 2.

High-resolution ultrasonography of breast swellings showed well-defined cystic lesions in all the cases. In two cases, multiple cystic lesions were seen. All the cysts were well-defined and thin walled. The largest cyst measured 2.5 cm × 1 cm, and the smallest cyst measured 1 cm × 0.8 cm. In all five cases, the cyst cavity showed linear echogenic structure. The elongated worm showed slow, repetitive dancing movements on real-time ultrasonography. No calcification was obvious in the cyst. A diagnosis of breast filariasis with live adult worm was suggested. On USG guided FNAC, adult gravid female worm with eggs and coiled microfilariae were seen in one case (Figure 1) of breast swelling. In two cases, cytology smears revealed microfilaria along with few inflammatory cells (Figure 2). Two cases (15.4%) did not show either microfilaria or adult worm in cytology smears.

Table 1: Clinical data of all the cases included in present study

Clinical data	Breast swelling	Scrotal swelling	Inguinal swelling
Number of cases	5	6	2
Clinical findings			
Age range (mean) year	32-55 (mean 40)	25-37 (mean 31.2)	15-26 (mean 20.5)
Swelling	5	6	2
Pain	5	4	-
Fever	2	4	1
Erythema	1	2	-

Table 2: PBS and cytologic findings of all the cases included in present study

PBS and cytology details	Breast swelling	Scrotal swelling	Inguinal swelling
Peripheral smear findings			
Eosinophilia	2	3	1
Microfilaremia	-	1	-
Cytologic findings			
Hemorrhagic aspirate	2	1	1
Fluid aspirate	1	2	1
Adult worm in aspirate	1	1	-
Microfilariae in aspirate	2	5	2
Polymorphs	2	2	1
Eosinophils	1	3	-

PBS: Peripheral blood smear

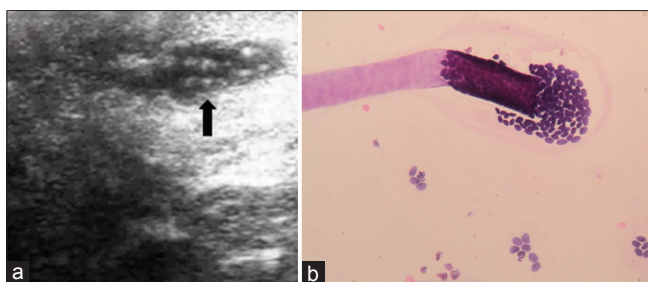


Figure 1: (a) B-mode high-resolution ultrasound of left breast showing well defined lobulated cystic lesion in upper outer quadrant showing internal actively mobile linear echogenic structures (filarial dance sign), (b) cytology smear from breast aspirate showing adult filarial worm with eggs (H and E x100)

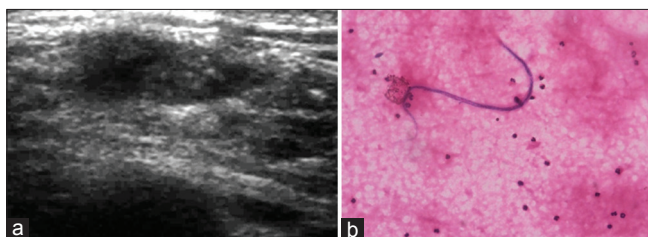


Figure 2: (a) High-resolution ultrasound of right breast showing well defined lobulated cystic lesion in upper medial quadrant with filarial dance sign, (b) cytology smears from breast shows microfilaria in a background of inflammatory cells (H and E x400)

B-mode HRUS of scrotal swellings showed small, well-defined cystic lesions in the subcapsular region of testicular parenchyma and adjacent to epididymis with no flow on color Doppler study. Multiple, echogenic linear structures with a persistent rigorous wriggling motion were observed within these. Free fluid was observed in the scrotal sac in two cases. USG guided FNAC revealed adult worm with coiled microfilaria in one case (Figure 3). Rest five cases showed microfilariae in cytology smears. Peripheral blood smear revealed microfilaremia in one case of scrotal swelling.

HRUS of inguinal swellings showed dilated anechoic channels (lymphatics) with no flow on color Doppler study. Multiple actively motile elongated tubular structures were seen within these channels. USG guided FNAC revealed microfilariae in both the cases (Figure 4). In all the cases, the microfilariae showed the presence of hyaline sheath and well-separated nuclei with absence at the cephalic end and tail tip.

All the patients were followed-up after 3 weeks of oral therapy with DEC. Four patients (2 scrotal, 1 breast, and 1 inguinal swelling) were lost to follow-up. Rest all cases showed decrease in pain and swelling. On repeat ultrasonography, 3 patients of breast swelling, 2 patients of scrotal swelling, and 1 patient of inguinal swelling demonstrated no linear echogenic structures or movements.

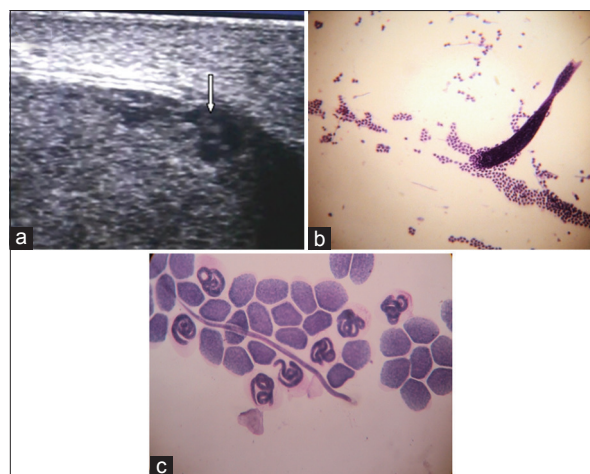


Figure 3: (a) B-mode high resolution ultrasound of left testes showing well-defined cystic lesion in subcapsular region with internal undulating tubular echogenic structures (filarial dance sign), (b) cytology smear from testicular aspirate shows adult female worm with eggs (H and E x100), (c) cytology smear showing numerous microfilarial eggs, coiled microfilaria and an uncoiled microfilaria (H and E x1000)

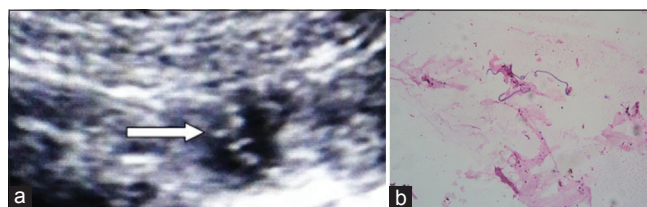


Figure 4: (a) Ultrasound of inguinal region showing well defined lobulated cystic lesion in left inguinal region with filarial dance sign, (b) cytology smear from inguinal swelling shows multiple microfilariae in a background of inflammatory cells

One patient of breast and scrotal swelling showed linear echogenic structures, but they did not have the movements.

DISCUSSION

Filarial infection is endemic in the tropics including India, part of Sri Lanka, Africa, and the Far East. There are eight species of filarial parasite, out of which only three, *W. bancrofti*, *Brugia malayi*, and *B. timori* are known to cause lymphatic filariasis.¹⁰ 90% of all filariasis cases in the world are caused by *W. bancrofti*, followed by *B. malayi* and *B. timori*.¹¹

Man is the definitive host, and the mosquito is the intermediate host for bancroftian and brugian filariasis. *Culex quinquefasciatus* transmit *W. bancrofti* and *Mansonia* mosquitoes transmit Brugian filariasis. The microfilaria of *W. bancrofti* and *B. malayi* display nocturnal periodicity.

Mosquitoes deposit infective larvae on the skin and transmit the infection. The larvae develop into adult worms over a

period of 6 months-2 years in the lymphatics. Adult worm produces microfilaria, which circulate in the blood stream.¹²

The majority of infected individuals in endemic areas remain asymptomatic throughout their life. Symptomatic patients may show an acute phase of disease characterized by fever, muscle pain, lymphangitis, lymphadenitis, and lymphedema. The chronic stage may be characterized by lymphadenopathy, lymphedema, hydrocele, and elephantiasis.

The lower extremities and genitalia are the most common sites of involvement, followed by the upper extremities. However, the organisms may be found in any organ of the body causing a mass lesion in the absence of the classic signs of filariasis.¹³

Breast filariasis is a rare cause of breast lump. It is the host inflammatory immune reaction, which is responsible for the wide spectrum of symptoms and signs associated with the filarial affliction of breast.¹⁴ Intact worms produce minimal tissue reaction while degenerating parasites provoke inflammatory cell infiltration- mainly eosinophils and occasional development of epithelioid granulomas.¹⁴

Before the description of the FDS on ultrasonography, there were no methods available to detect adult filarial worms *in vivo*. By localizing the adult worms on ultrasonography, the response to therapy can be assessed. Amaral *et al.* first reported FDS in 1994.¹⁵ They described the movements of live adult filarial worms in the lymphatic vessels as “peculiar, random-appearing movements of objects inside a vessel-like structure with persistent, random, twirling movements. This sign is unique and specific for filarial adult worms.

Chaubal *et al.* reported a series of eight cases of scrotal FDS.⁹ In two patients, they aspirated the dilated lymphatic channels and demonstrated microfilariae in the aspirates. In the present study, 13 patients with scrotal, breast, and inguinal swellings demonstrated positive FDS. All the patients were subjected to USG guided FNAC, and in 11 patients, filariasis was confirmed by cytology smears. Earlier, there were single case reports for filarial dance in breast mass;⁸ however, the present study shows five cases of breast swelling with FDS.

Despite the high incidence of this parasite in the endemic zone, detection of adult gravid filarial worm or eggs in FNAC is extremely rare. Kapila and Verma⁵ and Azad *et al.*¹⁶ reported the presence of adult filarial worms in soft tissue swellings. Chakrabarthi *et al.*¹¹ reported adult worm in breast lump aspirate. In our study on USG guided FNAC, adult worm were demonstrated in cytology

smears in one case of breast swelling, and one case of scrotal swelling. Demonstration of microfilaria in FNAC smears is also an uncommon finding. In the present study, microfilariae were seen in cytology aspirates of nine cases. This finding confirms that USG guided FNAC in patients with positive FDS is an excellent tool for demonstration of microfilaria or adult worm. FDS on HRUS correlates with active release of microfilariae by the worms and hence indicates active infection. In two cases of breast swelling with FDS, microfilaria could not be demonstrated in cytology smears.

The response of adult filarial worms to diethylcarbamazine is demonstrated by many authors.¹⁷ The response is quite variable in different patients, and even in the same patient, different adult worms may show varying response. Ultrasonography, being the only modality that can show the adult worms, is very useful in the follow-up period to document the response of worms to the drug. Complete absence of worm movements on follow-up examination was taken as a positive response. In our study, 8 (61.5%) of 13 patients had a positive response to the drug, which correlates with results from previous studies.

CONCLUSION

High-resolution ultrasonography is a valuable technique for diagnosing filarial infection in symptomatic patients. This study emphasizes on the importance of the classic and diagnostic USG finding of FDS. USG guided FNAC in the patients presenting with FDS helps in detecting microfilaria or adult worm and thus plays a significant role in recognition of the disease and institution of specific treatment. The appearance of motile filarial worms on ultrasonography correlates with active release of microfilariae into the lymphatic vessels and hence indicates active infection.

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