

## Atypical cases of filariasis from a non-endemic area

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**Summary** Filariasis can present in many different ways and pose significant dilemma to the clinician. We report four atypical cases of filariasis which presented as abdominal mass, cervical lymph node enlargement, fever in pregnancy and nosocomial febrile illness respectively. All the four cases were treated successfully with oral antifilarial agents. It is essential to be aware of such atypical presentations of filariasis so that prompt therapy can be initiated.

**Keywords:** *Wuchereria bancrofti*, pregnancy, lymphadenopathy

### 1. Introduction

Filariasis is a neglected tropical disease which poses a significant burden in endemic areas. An estimated 68 million people are infected worldwide, with 40 million being symptomatic (1,2). India shares a heavy burden of the disease and despite having a successful national program, it is not rare to find people from endemic areas presenting with swollen limbs and scrotal swelling even today (3). The social, psychological and economic implications of this disease are enormous. Unfortunately, the disease is often missed in the early stages and when it manifests atypically. It can present in many different ways including fever, lymphangitis, cough or wheeze and limb or scrotal swelling. Since it may be incidentally detected in the blood or tissue aspirate, it is imperative for the laboratory technicians and clinicians to be aware of such findings. Timely diagnosis and appropriate management can significantly decrease the morbidity associated with the disease. We present four atypical cases of filariasis which created significant diagnostic dilemmas but were successfully managed.

### 2. Case Report

#### 2.1. Case 1: Filariasis mimicking a malignant abdominal tumor

A forty-eight-year old female, resident of Uttar Pradesh, presented with a lump in the right lumbar area, which developed over a year. Her blood investigations were normal. Magnetic Resonance Imaging (MRI) of the abdomen revealed a large midline, complex cystic mass of size 8.5 cm in the greater pelvis showing fluid-debris level as well as non-dependent membranous structures within (Figure 1A). The ovaries were seen separately from the mass. There was another complex cystic mass of size 5 cm in the right pararenal space of retroperitoneum, inferior to the right kidney (Figure 1B). Some of the loculi showed T2-hypointense debris. In addition, conglomerate tubular channels were seen along the bilateral external and internal iliac vessels, which raised the suspicion of dilated lymphatics (Figure 1C). The magnetic resonance lymphangiogram subsequently performed confirmed the lymphatic nature of these tubular channels. The cystic lesions in the pelvis and retroperitoneum also showed partial opacification with gadolinium, suggesting lymphatic communication and hence the origin of these cysts. Imaging differentials of multifocal macrocystic lymphatic malformation as well as of

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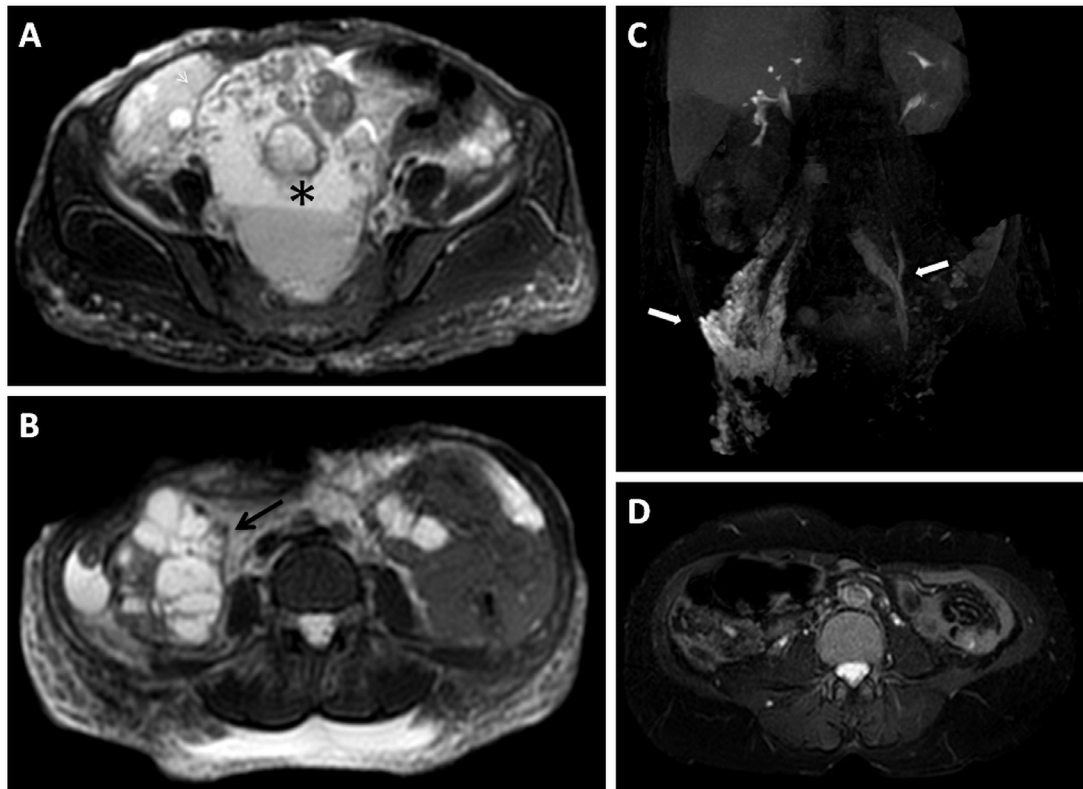
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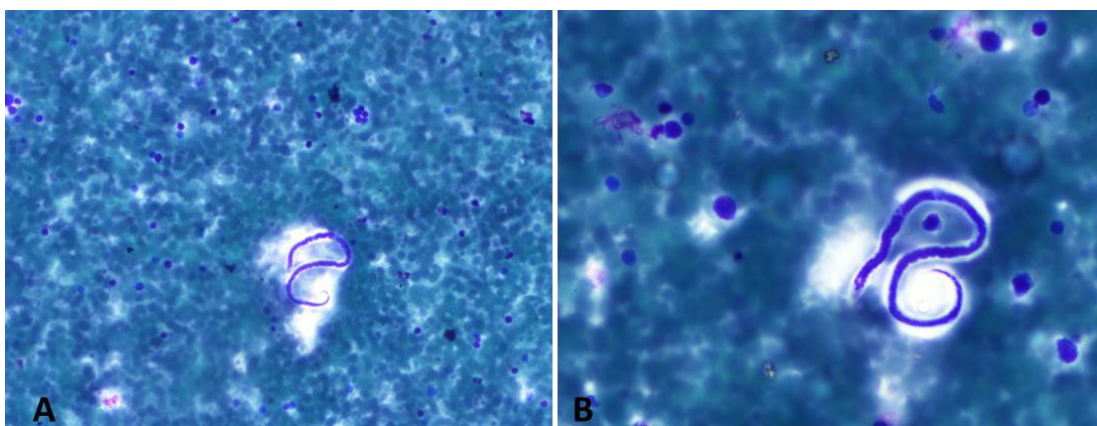
cystic mesothelial and germ cell neoplasms were kept. Since imaging could not rule out malignancy, fine needle aspiration cytology (FNAC) of the pelvic mass was done, which revealed microfilaria (Figure 2). She was treated with oral diethylcarbazine (DEC) for twenty-one days, subsequent to which the abdominal lump gradually decreased in size. The follow-up MRI performed at one year showed complete resolution of the pelvic and retroperitoneal lumps as well as the lymphatic dilatation (Figure 1D).

## 2.2. Case 2: *Microfilariae* in the lymph node

A eleven-year-old female, resident of Delhi, presented with on and off fever with chills for six months. On physical examination, she was found to have an enlarged right cervical lymph node measuring 4 × 2 cm. Her routine blood investigations were normal. Mantoux test was negative. Cytopathology of the lymph node aspirate revealed reactive changes with presence of microfilariae. She was treated with DEC and



**Figure 1.** Fat suppressed T2-weighted MRI showing (A) large complex pelvic cystic mass (asterisk) with fluid-debris level as well as numerous T2-hypointense membranous contents, and (B) a complex multiseptate cyst (arrow) in the retroperitoneum on the right side. Some of the loculi show differential T2-hypointensity, consistent with debris. (C) Coronal maximum intensity projection images of MR lymphangiogram showing dilated lymphatics (open arrows) along the bilateral external and common iliac vessels. (D) Representative post-treatment T2-weighted MRI at the level of the second image showing complete resolution after one year of therapy.



**Figure 2.** Fine needle aspiration smear from abdominal lump showing microfilaria in a background of blood and inflammatory cells (A). A higher power view of the microfilaria surrounded by a faintly staining sheath (B).

doxycycline, subsequent to which the fever resolved and did not recur at four months of follow. However, the lymph node swelling was persistent.

### 2.3. Case 3: Detection of microfilariae in the screening peripheral smear of a pregnant patient with fever

A 24-year-old pregnant female, resident of Delhi, presented in the second trimester of her pregnancy with fever, chills and joint pain lasting three days. The physical examination was unremarkable. Her routine peripheral blood smear showed microfilariae. Oral albendazole was initiated and the patient became afebrile on the second day of treatment. She remained asymptomatic at six months of follow-up and no microfilariae were seen in follow-up smear.

### 2.4. Case 4: From the postoperative ward for fever

A 55-year-old hypertensive male from Delhi presented acutely with altered sensorium and hemiparesis. Unenhanced computed tomography (CT) of the brain showed acute bleed in the left basal ganglion with midline shift and intraventricular extension. He underwent craniotomy with hematoma evacuation and frontal external ventricular drain (EVD) insertion. On the seventh postoperative day, he developed high-grade fever with chills. Physical examination revealed no focus of infection. He was started on broad-spectrum antibiotics, however there was no improvement in the fever. His blood counts, liver and kidney function tests, blood and urine cultures, cerebrospinal fluid

examination, serum procalcitonin levels and chest radiograph were normal. Blood mount showed motile microfilariae (Figure 3), which were identified as *Wuchereria bancrofti* on giemsa staining (Figure 4). He was started on oral DEC and became afebrile on the second day of treatment.

### 3. Discussion

Filariasis is a major public health problem in the tropical world with both lymphatic and extralymphatic manifestations. A retrospective analysis of the records of our outpatient clinics and infectious disease consultations from 2017 revealed 20 patients of proven filariasis. Five patients showed atypical presentations, with the findings of one among them (massive splenomegaly with bone marrow aspirate showing microfilariae) having been already published (4).

Microfilariae have been detected incidentally in unsuspected swellings in many reports from across the world (5-8). In such cases, microfilariae may either be the direct etiologic agent for the swelling or may have gotten entrapped in an already existent swelling. Diagnosing extralymphatic filariasis remains a clinical challenge as it is often unsuspected and as it mimics non-filarial diseases. Our series describes four cases of filariasis which presented at unusual and rare sites, but were promptly diagnosed and managed.

The first case showed complex cystic masses in the pelvis and retroperitoneum, which usually raise the suspicion of macrocystic lymphatic malformation or malignancy. Since the ovaries were separately visualized, the possible differentials included primary cystic mesothelial and germ cell neoplasms. Because of the unusual morphology, filariasis was not kept as a differential until the cytology revealed otherwise. Timely diagnosis and prompt institution of antihelminthic therapy enabled complete recovery

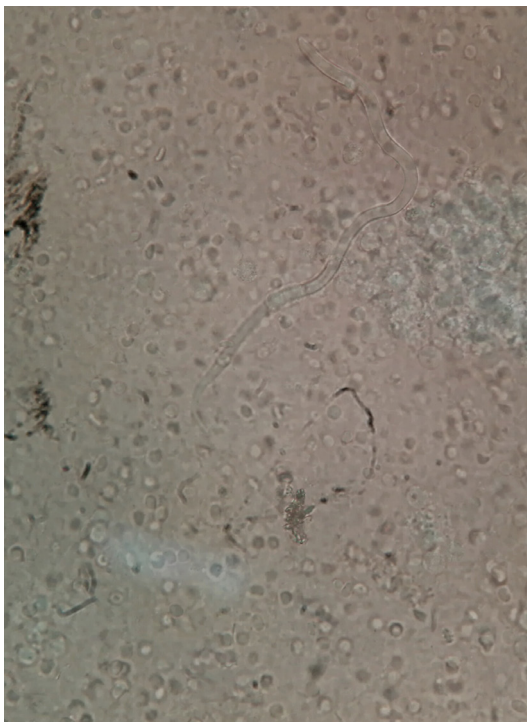


Figure 3. Wet mount of blood showing microfilaria.



Figure 4. Peripheral blood smear (Giemsa stain) showing microfilaria of *Wuchereria bancrofti*.



without surgery. Only four reports of filariasis presenting as abdominal masses or cysts are available in the current literature, all of which are from the Indian subcontinent (9-12). Except for one, all showed large unilocular retroperitoneal cysts which were resected. Giri *et al.* (12) reported a complex solid-cystic retroperitoneal mass which, similar to our case, resolved completely on oral DEC therapy. Although the exact mechanism of the origin of abdominal cysts in filariasis is not known, possible hypothesis includes lymphatic obstruction resulting in rupture and extravasation of chyle as well as the involvement of ectopic lymphatic tissue (9). Although rare, keeping a high index of suspicion can result in timely diagnosis, which avoids surgery and significantly changes the course of the disease.

Many case series and reports have shown incidental detection of microfilaria in lymph node aspirates (6-8). Presence of microfilariae in lymph nodes may only be an incidental finding with no proven causal association. Cases of microfilariae in aspirates from malignant lymph nodes have also been reported, although the association with malignancy is controversial (13-17). It is important to be aware of this observation and so that the samples are carefully screened. Timely diagnosis can prevent future morbidity and complications.

There is no definitive evidence as to whether pregnancy, surgery or other forms of stress can activate the dormant worm and release more microfilariae into blood. No cases of filariasis presenting as nosocomial fever has been described in the literature. However, two among the four cases in our series presented for the first time in periods of stress (pregnancy and postoperative period respectively). Whether these presentations were incidental or had any possible relation to stress remains speculative. Another noteworthy finding in our case series was that none of the patients had eosinophilia. This highlights the fact that eosinophil counts may be normal in filariasis and the diagnosis should not be ruled out for the mere absence of eosinophilia.

There have been several reports of filariasis from non-endemic areas such as Delhi. Two among the four patients belonged to Delhi and had no history of travel to other endemic areas. Literature suggests that transmission of filariasis requires prolonged stay in endemic areas and hence is uncommon in travelers.

Detection of microfilaria or adult worm in the laboratory samples should prompt the physician to initiate treatment so as to avoid any long-term consequences of the disease. DEC is highly effective in eliminating microfilaria, but has only modest activity against the adult worm. Many recent studies support the synergistic effect of combining this drug with doxycycline. Doxycycline acts by targeting *Wolbachia*, which engages in endosymbiotic relationship with the filarial parasite and is important in maintaining the reproductive capacity of the worm (18-20). Albendazole

and ivermectin have been used in combination as alternatives. Ivermectin and doxycycline should be avoided in pregnancy. In our series, all the patients responded well to treatment with oral anti-filarial drugs (DEC or albendazole). All patients tolerated the drug well and did not report any side effects or intolerance. All patients showed improvement in symptoms at follow up.

As we envision a world free of filariasis, the World Health Organization (WHO) has launched the Global Programme to Eliminate Lymphatic Filariasis (GPELF) by 2020. WHO recommends preventive chemotherapy with annual mass drug administration (MDA) of albendazole together with ivermectin or with DEC. Enhanced public health strategies, disease awareness, prompt diagnosis and treatment can help us in paving the path towards elimination.

In conclusion, these cases highlight the unusual and diagnostically challenging presentations of filariasis. It underscores the fact that timely diagnosis and prompt management can prevent severe complications and long-term sequelae. Each patient who gets diagnosed and treated moves us a step closer towards the vision of filariasis-free world.

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