OCCULT FILARIAISIS: A STUDY OF TWO CASES, WITH REVIEW OF LITERATURE
Superna Ganguly¹, Sadhana Bagde², Rashmi Gupta³, Amit Tiwari⁴

ABSTRACT: Occult Filariasis is a term used to designate lesions in which microfilariae are not found in peripheral blood, although they may be seen in tissues. These lesions present differently from those with classical disease and resemble many non-filarial lesions from which they are difficult to distinguish clinically. This is a report of two such cases where diagnosis of filariasis was entertained only after cytological examination.

KEYWORDS: Filariasis, Wuchereria Bancrofti, Brugia Malayi.

INTRODUCTION: Lymphatic filariasis is a major public health problem in India with 99.4% cases being caused by Wuchereria bancrofti. Occult Filariasis is a term used to designate lesions in which microfilariae are not found in peripheral blood, although they may be seen in tissues. These lesions present differently from those with classical disease and resemble many non-filarial lesions from which they are difficult to distinguish clinically.

CASE REPORT: Clinical Presentation: Case No. 1: 21yr old male presented with swelling in left axilla since 15 days, and fever with cough since 1 month. On examination, a single, firm, axillary swelling was observed, measuring 4cm x 3cm x 1.8cm. No other lymph node was enlarged. A clinical diagnosis of tubercular lymphadenitis was entertained.

Case No. 2: 34yr old female presented with a painless swelling in right arm since 15 days. On examination, a single, non-tender, ill-defined, soft swelling was observed in the medial aspect of right arm, measuring about 2cm in diameter. There was no history of similar swelling(s) anywhere else in the body. A clinical diagnosis of Lipomatosis was made, aided by radiological findings of diffuse proliferation of fat in medial and post compartments of the arm.

FINE NEEDLE ASPIRATION: Multiple aspirations from representative lesions were obtained in both cases using 22G disposable needle and 10ml disposable syringe. In the 1st Case, 2 ml of whitish fluid aspirated, whereas in the 2nd Case, aspirate consisted of blood mixed material that did not give an oily or fatty appearance. Fixed and air dried smears were stained by Hematoxylin & Eosin, PAP and Giemsa stains respectively.

CYTOPATHOLOGICAL IMPRESSION: Case No. 1: Smears were hypercellular, consisting of a mixed inflammatory infiltrate comprising of neutrophils, eosinophils and macrophages seen against a lymphocytic background. Multiple fragments of adult filarial worm as well as...
microfilariae of Wuchereria bancrofti were seen in the smears. Focal areas showed granulomatous response.

Case No. 2: Smears showed numerous degenerated, coiled and organized microfilariae of Wuchereria bancrofti, embryonated eggs and fragments of adult worms surrounded by fragmented granulomas and a mixed inflammatory infiltrate comprising acute and chronic inflammatory cells including eosinophils. Background showed necrotic debris. Aspirates from adjoining sites showed normal, mature adipocytes, but no evidence of adipose tissue lesion.

ZEIHL-NEILSEN STAIN: In both cases, due to presence of granulomatous lesions, ZN Stain was done to rule out AFB. Both cases were negative for AFB.

PERIPHERAL SMEAR: Peripheral eosinophilia (9% in Case I and 12% in Case II) was noted in both cases. No microfilariae were seen in the peripheral smears of both the patients (both thick and thin smears examined).

HISTOPATHOLOGY: In both the cases, once the infectious and non-neoplastic nature of the lesions was explained, the patients refused to undergo biopsy.

DISCUSSION: Eight filarial parasites infect humans, of which four, i.e., Wuchereria bancrofti, Brugia malayi, Onchocerca volvulus and Loa loa are responsible for the most serious filarial infections. Wuchereria bancrofti, the most widely distributed filarial parasite of humans, affects an estimated 110 million people. It was first reported in India by Lewis in 1872. Lymphatic Filariasis is a major public health problem in India with 99.4% cases being caused by Wuchereria bancrofti whereas Brugia malayi is responsible for 0.6% of the problem, being uncommon except for a few areas in eastern India. In both our cases, the causative organism was W. bancrofti, as evidenced by detection of sheathed microfilariae with pointed tail-tip devoid of nuclei.

Bancroftian Filariasis is a vector–transmitted parasitic infection caused by the nematode Wuchereria bancrofti with man as the only known definitive host. Natural vectors for W. bancrofti infection are Culex quinquefasciatus (fatigans) mosquitoes in urban settings and anopheline or aedean mosquitoes in rural areas.

The principal pathologic changes in lymphatic filariasis result from inflammatory damage to the lymphatics, which are caused by adult worms and not by microfilariae. Although scrotal lymphatics and inguinal lymph nodes are preferred locations for adult worms, the lymphatic system of the lower limbs, retroperitoneal tissues, spermatic cord, epididymis and mammary glands are also involved. There are also reports of worms found in aspirates of the axillary lymph node, as seen by us in Case I. Some other unusual sites are breast, thyroid, maxillary antrum, eyeball, cervical scrapes, bronchial aspirates, bone marrow and body fluids. Sometimes it might be an incidental finding with other inflammatory or neoplastic lesions. In Case II, the lesion presented as a ill-defined soft tissue swelling in the arm, clinically and ultrasonographically misdiagnosed as a subcutaneous lipoma. Filariasis of superficial soft tissue of the upper extremity, though uncommon, has been reported.
The adult filarial worms reside in the lymphatic system from where the gravid female releases large number of microfilaria which may pass through the thoracic duct and pulmonary capillaries into the peripheral blood and occasionally microfilaraemia may be found in the infected patient. However; many cases of tissue filariasis present without microfilaraemia. In these cases, fragments of adult filarial worm, mainly the gravid female with double uterus is usually seen in the tissue aspirates. Sometimes sheathed microfilariae, coiled microfilariae, oval ova, embryonated eggs or non-gravid female worm may also be found. The diagnosis is further corroborated by associated findings like predominance of eosinophils, or granulomas, or both in the same aspirate. Adherence of inflammatory cells and macrophages to microfilariae is also seen. These are in concurrence to our findings as fragments of adult filarial worm as well as large numbers of sheathed microfilariae of W. bancrofti were seen by us in both cases. In Case II, we also saw embryonated eggs of adult gravid female. An abundance of mixed neutrophilic and eosinophilic inflammatory infiltrate, a granulomatous response to the parasites, as well as adherence of the inflammatory cells to the microfilariae was observed in both the cases. A negative ZN Stain ruled out Tubercular Granulomas.

**The clinical presentations of Lymphatic (Bancroftian) Filariasis are:**

i. Asymptomatic (subclinical) microfilaremia

ii. Hydrocele

iii. Acute adenolymphangitis

iv. Chronic lymphatic disease

v. Occult or Cryptic Filariasis

The term Occult Filariasis is commonly used to designate filarial infections in which microfilariae are not found in the peripheral blood although they may be seen in tissues. The clinical features of occult filariasis are different from those of the classical disease, and resemble many non-filarial diseases from which they are difficult to distinguish. Some of these “occult” clinical manifestations are tropical pulmonary eosinophilia, glomerulopathies, filarial arthritis, nerve palsies, endomyocardial fibrosis and filarial granulomas in the breast.

- Tropical Pulmonary Eosinophilia (TPE): First described by Frimodt-Moller and Barton in 1940, TPE is most common in the Indian Subcontinent even though Bancroftian Filariasis is prevalent all over the world. It manifests with cough, fever, breathlessness, malaise and abdominal pain. Investigations show peripheral eosinophilia, raised ESR and diffuse miliary lesions or increased bronchovascular markings on X-ray. Microfilariae are not seen in peripheral blood, but may be found in lung, liver and lymph nodes. Antifilarial IgE antibody titres may be raised.

- Filarial Arthritis: This form of arthritis usually affects the knee joints and is fairly common in endemic areas. It is usually monoarticular, but two-joint involvement also occurs. Filarial arthritis exclusively involves the large joints and involvement of small joints rules out the diagnosis. The patients have normal to moderately elevated eosinophil counts and ESR, but ASO titers are normal. Microfilariae are not seen in peripheral smears. X-ray of
involved joints show only soft tissue swelling and no bony abnormality. The pathogenesis of the disease is obscure.

- **Glomerulopathies:** Acute glomerulonephritis associated with lymphatic filariasis has been reported by Chugh et al and Chaturvedi et al. Renal biopsies of the patients showed diffuse mesangial proloferative glomerulonephritis with C₃ deposition on the basement membrane. Demonstration of filarial antigens and specific filarial antibodies in the immune complexes deposited in the glomeruli provide definitive evidence of filarial etiology.

- **Endomyocardial fibrosis (EMF):** Endomyocardial fibrosis is a rare disease with a geographical distribution in the areas endemic for filariasis. The occurrence of eosinophilia and EMF with Loeffler's Syndrome and finding of filarial antibodies in majority of patients with EMF supports the theory that EMF may be filarial in origin.

- **Filarial Granulomas in the Breast:** This manifestation is particularly prevalent in India and Sri Lanka where W. bancrofti is the predominant species. It has not been reported for areas endemic for Brugian filariasis. Filarial granulomas of breast present as firm-to-hard breast lumps which may be grossly confused with malignant tumors. Microscopy shows an eosinophilic granulomatous response to filarial parasites (both adult worms and microfilariae) in various stages of degeneration.

In both our cases patients presented with misleading clinical symptoms due to which a clinical diagnosis of Classical Filariasis was not entertained in either case. The diagnoses were further confounded by absence of microfilaremia in both cases. However, moderately high eosinophil counts were encountered in both cases, which, after correlating with the FNAC findings, were confirmed to be a manifestation of Tropical Pulmonary Eosinophilia. Thus, by correlating the absence of diagnostic clinical features and peripheral microfilaremia with the manifestation of Tropical Pulmonary Eosinophilia, a final diagnosis of Occult Filariasis was made in both the cases.

**Filariasis is diagnosed by two methods**

- **Direct Method:** By visualization of adult worm in tissues, or of microfilariae in blood or body fluids. (FNAC, Biopsy, X-Ray, Doppler USG)

- **Indirect Method:**
  1) Peripheral blood eosinophilia.
  2) Elevated Serum Ig E Conc.
  3) Antifilarial serum antibodies.
  4) ELISA
  5) Rapid format immunochromatographic card test.
  6) PCR.

In both the cases reported here, diagnosis was clinched by direct visualization of the parasites on FNAC and demonstration of peripheral blood eosinophilia in the absence of microfilaremia. While biopsy was refused by the respective patients in both cases, in the 2nd Case, USG was not helpful diagnostically.
CONCLUSION: Our report of these 2 cases of Occult Filariasis is concurrent with prior literature on similar presentations. It reminds the tropical pathologist to maintain a high index of suspicion for any subcutaneous swelling, as occult filariasis needs to be considered in the differential diagnosis of all such swellings. Even though many sophisticated investigative modalities are available for diagnosis of occult filariasis, FNAC; being a cheap, simple and easy procedure has an edge over the others. Especially in endemic areas, FNAC has a special role to play where expensive investigations are out of the reach of many a poor patient.
BIBLIOGRAPHY:
1. Harrison's Principles of Internal Medicine. 18th Edition. Editors: Longo, Fauci, Kasper et al. Publisher: McGraw Hill; Pgs. 1746-51.
2. Chaturvedi P, Gawdi A, Dey S. Occult Filarial Infections. The Natl. Med J India. 1990 Jan-Feb; 3(1): 7-9.
3. National vector borne disease control programme, Directorate general of health services, ministry of health & family welfare. http://nvbdcp.gov.in/filariasis.html
4. Khare P, Kala P, Jha A, Chauhan N, Chand P. Incidental Diagnosis of Filariasis in Superficial Location by FNAC: A Retrospective Study of 10 Years. J Clin Diagn Res. 2014 Dec; 8(12):FC05-8. doi: 10.7860/JCDR/2014/9906.5266. Epub 2014 Dec 5.
5. Adhish Basu, Sarath Chandra sistla, surendra kumar verma, S Jagdish. Lymphovaryx in the axilla- an unusual presentation of filariasis. Filaria Journal 2006, 5:9.
6. Dey P, Radhika S jain A. Microfilaria of weechereria bancrofti in a lymph node aspirate. A case report. Acta cytol. 1993 sep-oct;35(5):745-6.
7. Microfilariae in lymph node aspirates. Sah SP, Rani S, Mahto R. Acta Cytol. 2002 Jan-Feb; 46(1):73-5.
8. Varghese R, Raghuveer CV, Pai MR, Bansal R. Microfilaria in cytologic smears; a report of 6 cases. Acta Cytol. 1996 Mar-Apr; 40(2): 299-301.
9. Yenkeshwar PN, Kumbhalkar DT, Bobhate SK. Microfilaria in FNAC: a report of 22 cases. Indian J Pathol Microbiol. 2006 July; 49(3):365-9.
10. Srikanth S. Microfilaria in forearm swelling aspirate: An unusual finding. J Mahatma Gandhi Inst Med Sci 2014; 19:59-61.
11. Garbyal RS, Arundhati, Bohra A, Bhujun KH. An unusual presentation of filariosis: a case report. Acta Cytol. 2008 Mar-Apr; 52(2):204-06.
12. Tandon N, Bansal C, Sharma R, Irfan S. Role of fine needle aspiration cytology in diagnosing filarial arm cysts. BMJ Case Rep. 2013; 2013: bcr2013009677. doi: 10.1136/bcr-2013-009677. Published online 2013 May 17.
13. Fourth report of a WHO expert committee on lymphatic filariasis. Filariasi. WHO Tech Rep Ser 1984; 702:1-112.
14. Textbook of Medical Parasitology. 2nd Revised edition. Author: Pranabeshwar Chakraborty. Publisher: New Central Book Agency; Pgs.196-198.