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Case Report

Large retroperitoneal filarial lymphangiectasia

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Introduction

Human lymphatic filariasis is caused by infections with *Wuchereria bancrofti*, *Brugia malayi*, or *Brugia timori*. Currently there are about 31 million microfilaraemics, 23 million cases of symptomatic filariasis, and about 473 million individuals potentially at risk of infection in India.¹ The most common manifestation of the disease is hydrocele followed by lower limb lymphedema and elephantiasis.¹ In India and the neighboring countries, both hydrocele and lower limb lymphedema are common. Other forms of the disease such as tropical pulmonary eosinophilia and chyluria occur less frequently. While the laboratory diagnosis of filariasis has conventionally been made by demonstrating microfilariae in peripheral blood smear, there have been reports where diagnosis was made by finding microfilariae in material obtained by fine needle aspiration of various organs including thyroid, soft tissue, bone marrow, epididymis, lungs, breast, gastric brushing, hydrocele fluid and retroperitoneal

cyst.^{2–5} We report a rare case of filariasis presenting as hematuria without chyluria that on evaluation revealed a large retroperitoneal mass extending from mediastinum to pelvis.

Case report

A twenty-one year old patient, an professional athlete, was admitted with sudden onset, painless, intermittent gross hematuria after exercise. There was no history of loin pain, dysuria, increased frequency of urination, whitish urine, swelling of feet or facial puffiness. He never had any constitutional symptoms like fever, anorexia or weight loss. There was no history of bleeding manifestations like epistaxis, purpura or hemoptysis. There was no significant past history. His mother had lymphedema of one lower limb. Clinically he was averagely built and nourished. There was no lymphadenopathy or pedal edema. There was no evidence of any inguino-scrotal swelling. Both spermatic cords appeared to be mildly thickened. Investigations revealed Hb 14 gm%, total leukocyte count of 6600/cumm, polymorphs 70%, Lymphocytes 21%, Eosinophils 8% and absolute eosinophil count of 820/cumm. Urine examination showed numerous red blood cells but no casts, dysmorphic red blood cells, white blood cells or malignant cells. 24 h urinary protein was within normal limits. Urine culture was sterile. Cystoscopy showed bladder walls to be congested but there was no mass or bloody efflux from any ureteric orifice and a diagnosis of cystitis was entertained. Ultrasonography (USG) of abdomen showed an abdominal mass in retroperitoneal area with ill defined margins and cystic and solid areas within. Both the kidneys were normal but both proximal ureters were moderately dilated. Intravenous urography (IVU) was suggestive of external compression of right renal pelvis, bilateral dilation

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Fig. 1 – CT scan picture of the large retro peritoneal, diffuse, mass lesion extending between the two renal hila, encasing the aorta, IVC and the renal arteries.

of proximal third of both the ureters and extrinsic compression of the bladder.

Contrast Enhanced Computerized Tomography (CECT) showed a large retro peritoneal, diffuse, non-enhancing low density mass extending superiorly from just below the diaphragm to the pelvis inferiorly and laterally to the renal hila, encasing aorta, IVC, renal artery and proximal iliac vessels [Fig. 1]. The mass was encasing segments of both ureters causing ureteral dilatation but there was no significant hydronephrosis. The pancreas was displaced anteriorly by the mass. Urinary bladder appeared elongated with evidence of extrinsic compression bilaterally. MRI of the chest and abdomen confirmed a fluid filled, non-enhancing retroperitoneal mass as before, hypointense in T1 and hyperintense in T2, no fatty, hemorrhagic or calcific component and anteriorly displaced pancreas. The great vessels and branches were encased without luminal compromise. In view of the

Fig. 2 – Repeat CT after a course of antifilarial drugs showing a significant reduction in the size of the lesion.

presentation and findings on imaging a diagnosis of haeman-gioma/lymphangiectasia was made.

To confirm the diagnosis USG guided FNAC of abdominal mass was performed which showed blood with increased eosinophils and plenty of live microfilarial worms. On high-resolution USG of the scrotum, thickened spermatic cord was seen. Patient was started on Tab Diethylcarbamazine 100 mg TDS for 21 days. Patient responded well and became asymptomatic. Hematuria ceased. Patient has been on regular follow up for last three years. The retroperitoneal mass has reduced significantly in size but the patient remains symptomatic with hematuria on and off [Fig. 2].

Discussion

Human lymphatic filariasis has many nonconforming presentations and the worm has been recovered from several atypical tissues.²⁻⁵ The adult worms live in the lymphatics throughout the body and may cause extensive lymphangiectasia by obstructing lymph flow. Filariasis is the most common cause of acquired lymphedema in the world. Our patient did not have any hydrocele or lower limb lymphedema.

Filariasis causing retroperitoneal masses is rare even in regions where filariasis is endemic.⁶ The exact pathogenesis of the lesion remains speculative. Obstructed lymphatic vessels, rupture of lymphatics causing extravasation of chyle and the presence of ectopic lymphatic tissue have been propounded as the possible etiologies.⁷ A few cases have been reported in literature and that too mostly from the Indian subcontinent. Mehta et al⁸ reported a filarial abscess in their study of retroperitoneal tumorous lesions. Gupta et al⁹ diagnosed an infected retroperitoneal cyst to be of filarial origin by aspiration cytology and treated it surgically. Kapoor⁶ reported a cystic lesion in the retroperitoneum extending from the sub hepatic region to the pelvis and causing compression of the inferior vena cava. FNAC revealed live microfilariae of *Wuchereria bancrofti* on microscopic examination. Ahn et al published the CT and MRI findings of a dilated thoracic duct and diffuse retroperitoneal lymphangiectasis due to Bancroftian filariasis.¹⁰ Patients with retroperitoneal filarial lymphangiectasis usually present with abdominal pain and constitutional symptoms like fever.^{7,8} None of the patients in the various case reports published to date of filarial retroperitoneal masses have had hematuria alone as their presenting symptom. Our case has probably had the most extensive retroperitoneal lesion reported so far and presented only with hematuria due to extrinsic bladder compression. The treatment remains a course of antifilarial drugs and most of these lesions have significant regression with resolution of the ureteric dilatation on drug therapy alone. The role of surgery appears to be very limited in the absence of secondary infection.

Conclusion

We have described a case of filariasis with extensive retroperitoneal mass lesion. Lymphatic filariasis presenting

as an isolated retroperitoneal mass without any peripheral sign of filariasis is a rare occurrence.

Conflicts of interest

The authors have none to declare.

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