Case Report

A Rare Co-existent Case of Splenic Microfilariasis and Pancreatic Solid Pseudopapillary Epithelial Neoplasm - A Double Jeopardy!

Abstract

Filariasis is a major public health concern in tropical and subtropical countries like India with *Wuchereria bancrofti* accounting for 90% of lymphatic filariasis. Rarely observed are extra lymphatic manifestations caused by interaction of immune system with microfilaria and their diffusible products. Among various organs involved, splenic involvement is a rare extra lymphatic manifestation of filariasis and can masquerade clinicoradiologically as metastasis when associated with a known malignancy or as a primary malignancy like lymphoma. Hereby, we present an unusual case of coincidence of splenic filariasis with pancreatic solid pseudopapillary epithelial neoplasm in a 20-year-old woman associated with peripheral blood eosinophilia.

Keywords: *Eosinophilia, filariasis, granuloma, solid pseudopapillary epithelial neoplasm*

Sangeeta Kini, Rima N Kamat, Sneha Janjal, Heena M Desai

Department of Pathology, BYL Nair Ch. Hospital and TNMC, Mumbai, Maharashtra, India

Introduction

Filariasis, commonly seen in the tropical and subtropical regions, is caused by *Wuchereria bancrofti*, *Brugia malayi,* and *Brugia timori*. These organisms affect lymphatics and also involve extra lymphatic tissue; tropical pulmonary eosinophilia being one of the prototypes.[1] The extralymphatic filarial clinical manifestations are heterogeneous causing diagnostic dilemma, clinic-radiologically as well on histopathology especially when associated with malignancies.[2] Till date there is no evidence of association of splenic microfilariasis with a neoplasm.

We present an interesting case of coincidental occurrence in a young female of pancreatic solid pseudopapillary epithelial neoplasm (SPEN) with splenic granulomatous microfilariasis which masqueraded as metastasis in a known primary neoplasm.

Case Report

A 20-year-old female presented with a complaint of pain in abdomen for 1–2 months not associated with nausea or vomiting.

Blood investigations showed mild increase in white blood cell count (16,400/cmm) and eosinophilia (polymorphs 56%, lymphocytes 18%, and eosinophils 26%) with absolute eosinophil count of 4,264/cmm. Erythrocyte sedimentation rate was raised (130 mm at the end of 1 h).

Radiological investigations (ultrasonography abdomen) revealed a well-defined heterogeneous mass measuring 9.5 cm × 9 cm × 2 cm arising from the tail of pancreas with internal and peripheral vascularity highly suggesting SPEN. Tumor markers done such as AFP, CEA, and CA 19.9 were within normal limits. The patient was taken for exploratory laparotomy and pancreatic mass excised. Intraoperatively, spleen was found to be enlarged with multiple tiny nodules on capsular surface. A suspicion of metastasis was raised and splenectomy was performed.

Gross of pancreatic mass showed a well-encapsulated mass measuring 10 cm × 9 cm × 7 cm with normal pancreatic tissue at one end. Cut surface is solid cystic with yellow white and brown spongy areas [Figure 1]. Spleen weighed 118 g with capsular and cut surfaces showed many tiny whitish nodules of 2–5 mm size largely distributed in subcapsular region.

Histopathological examination of the pancreatic mass showed features of SPEN of pancreas. Features of malignancy like nuclear atypia and invasion (vascular,

How to cite this article: Kini S, Kamat RN, Janjal S, Desai HM. A rare co-existent case of splenic microfilariasis and pancreatic solid pseudopapillary epithelial neoplasm - A double Jeopardy! Int J App Basic Med Res 2024;14:131-3.

Submitted: 20-Oct-2023
Revised: 19-Feb-2024
Accepted: 18-Mar-2024
Published: 24-May-2024

Address for correspondence:
Dr. Sangeeta Kini,
A-802, Keshan Kunj3,
Sec 14, Plot 19, Sanpada,
Navi Mumbai - 400 705,
Maharashtra, India.
E-mail: sangukini@yahoo.co.in

Access this article online

Website: https://journals.lww.com/IJAB

DOI: 10.4103/ijabmr.ijabmr_464_23

Quick Response Code:

perineural, and capsular) were not identified, however, foci of dense infiltration of eosinophils were seen [Figure 2]. Immunohistochemical profile showed positivity for B Catenin, CD10, loss of E Cadherin, and negativity for Chromogranin further reinforcing the diagnosis and excluding the differentials. Histopathology of nodules in spleen revealed multiple granulomas composed of histiocytes, inflammatory cells, dense infiltration of eosinophils forming eosinophilic microabscesses, and occasional multinucleate giant cells [Figure 3]. A diligent search identified microfilariae in one of the fibrohistiocytic granuloma. Peripheral blood smear (PBS) examination was recommended in view of marked tissue and blood eosinophilia. Microfilaria with features of *W. bancrofti* was detected on PBS and buffy coat preparation [Figure 3]. The presence of microfilaria in blood appropriately justified blood eosinophilia and intense granulomatous reaction in spleen. Postoperatively, the patient was started on antifilarial treatment and recovery was uneventful.

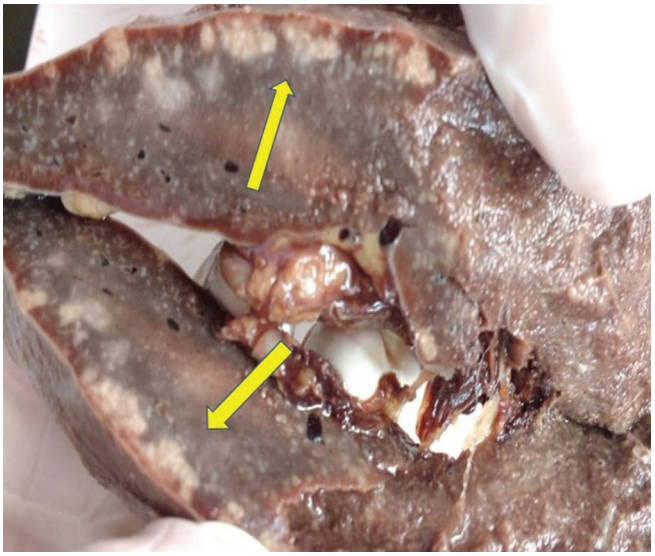

Figure 1: Gross of spleen with capsular and cut surfaces showing whitish nodules of 2–5 mm in size (arrows)

Discussion

Filariasis is a common public health problem in the southeast Asia including Indian subcontinent. The filarial parasites are known to cause dilatation of lymphatic channels and lymphangiectasia. The extralymphatic filariasis is heterogeneous as largely it is either due to microfilaria *per se* or tissue reaction caused due to its diffusible products.[3] The microfilariae are disseminated to various organs through blood and produce immune mediated reaction at these sites. In the lungs, they produce tropical pulmonary eosinophilia. Unlike other sites, spleen is an unusual site for bancroftian or Wuchereria filariasis.[4]

In *W. bancrofti*, a granulomatous response composed of macrophages, foreign-body giant cells, eosinophils, and lymphocytes is seen which is either toward deposition of hyaline substance (Splendore–Hoeppli phenomenon) which have the remains of the microfilariae or occasionally presence of microfilaria *per se* as seen in the present case in spleen. This reaction is the result of host–parasite interaction, mostly hypersensitivity to microfilarial antigen instead of direct damage from the parasite. Literature search highlights such rare incidental splenic microfilarial granulomatous lesions.[5]

Association of filarial parasite with malignancy has been described where the adult form of the filaria lodge in lymph vessels and due to lymphatic blockage in neoplasms they appear in tissue fluid or on surface material, but its role in tumorigenesis is not explained. The coexistence of microfilaria and malignancy may be coincidental. Sane and Patel reported a case where adult filarial worm was identified in cystic teratoma of ovary.[6] Gupta *et al*. reported five cases of microfilaria as an incidental finding in cavernous hemangiomas, transitional cell carcinoma, non-Hodgkin's lymphoma, follicular carcinoma of thyroid, and germ cell tumor of testis.[7] Mohan *et al*. reported a case

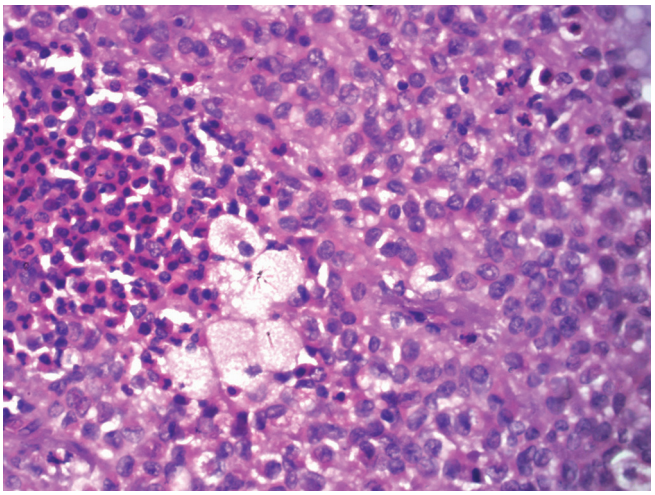

Figure 2: Photomicrograph showing microscopic appearance of pancreatic SPEN with tissue eosinophilia (H and E, ×400)

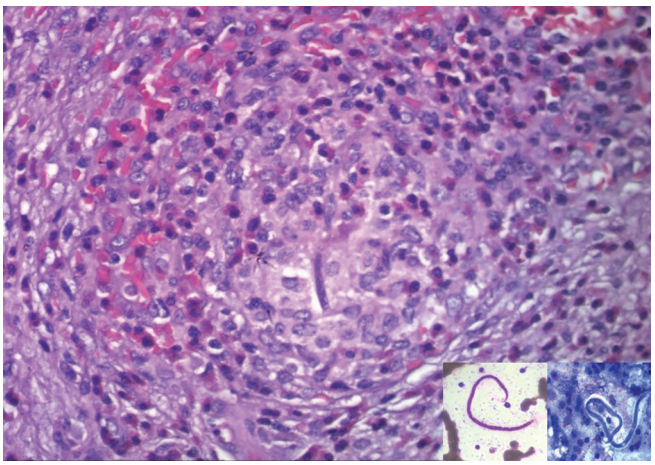

Figure 3: Spleen showing intense granulomatous reaction and eosinophilia surrounding the microfilaria (H and E, ×400\). Inset- Microfilaria on peripheral smear and buffy coat (Wright's stain ×1000)

of infiltrating ductal carcinoma of breast with coexistent microfilaria.[8] Sometimes, microfilariasis may present as a retroperitoneal mass.[9]

With respect to the above-mentioned reports where there is coincidental demonstration of microfilaria within the neoplasm on histology or in malignant fluid on cytology, the present case did not identify microfilaria within pancreatic SPEN but there is coexistence of microfilaria in spleen which masqueraded as splenic metastasis intraoperatively and prompting splenectomy.[10]

The extreme tissue eosinophilia observed both within the pancreatic neoplasm and splenic granuloma prompted to search for parasite in the present case which further was confirmed on PBS and buffy coat preparation. Microfilariasis may not be associated always with peripheral blood or tissue eosinophilia.[11]

To the best of our knowledge and with thorough literature search, this is first case of Pancreatic SPEN coexisting with splenic granulomatous microfilariasis.

Conclusion

We highlight through this case report that awareness is essential of coexistence of microfilariae in neoplastic conditions while dealing with associated extralymphatic chronic granulomatous lesions with or without tissue/peripheral blood eosinophilia thereby prompting right clinical management.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

References

1. Dreyer G, Dreyer P, Piessens WF. Extralymphatic disease due to bancroftian filariasis. Braz J Med Biol Res 1999;32:1467-72.
2. Agarwal PK, Srivastava AN, Agarwal N. Microfilariae in association with neoplasms. Acta Cytol 1990;26:488-90.
3. Cook GC. Discovery and clinical importance of the filariases. Infect Dis Clin North Am 2004;18:219-30.
4. Andola SK, Anita AM, Yevoor K, Patil R. Extralymphatic filarial disease in spleen (occult filiariasis) – A report of 2 cases. RGUHS J Med Sci 2012;2:61-3.
5. Sahu G, Verma Y, Jajoo S, Shrivastava P. Filariasis with squamous cell carcinoma: A hidden surprise. Int J Appl Basic Med Res 2020;10:59-61.
6. Sane SY, Patel CV. A filarial worm in the wall of a cystic teratoma of the ovary – (A case report). J Postgrad Med 1989;35:217-8.
7. Gupta S, Sodhani P, Jain S, Kumar N. Microfilariae in association with neoplastic lesions: Report of five cases. Cytopathology 2001;12:120-6.
8. Mohan N, Agrawal R, Kumar P. Breast ductal carcinoma with coexistent microfilaria: Diagnosed on cytology. Trop Parasitol 2018;8:103-5.
9. Giri A, Kundu AK, Chakraborty M, Das S. Microfilarial worms in retroperitoneal mass: A case report. Indian J Urol 2000;17:57-8.
10. Burchard GD, Reimold-Jehle U, Bürkle V, Kretschmer H, Vierbuchen M, Racz P, *et al.* Splenectomy for suspected malignant lymphoma in two patients with loiasis. Clin Infect Dis 1996;23:979-82.
11. Garg N, More S, Sharma M, Kotru M. Microfilaria: Not always associated with eosinophilia. Trop Doct 2019;49:210-2.