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LETTER TO EDITOR

Central nervous system vasculitis: a rare manifestation of leptospirosis presenting as bilateral cortical blindness

Sir,

Leptospirosis is the most widespread zoonosis in the warm and humid region caused by spirochetal infection.[1] It is an occupational disease with clinical manifestation varies from subclinical, self-limited infection to severe and potentially fatal disease.[2] The most frequent neurologic manifestation in leptospirosis is aseptic meningitis. Other manifestations are encephalitis, cerebellitis, myelitis, intracranial bleed, and movement disorders.[3] Here, we report a case of central nervous system (CNS) vasculitis in severe leptospirosis.

A 28-year-old female presented with a history of fever and diarrhea. After 5 days of her illness she developed dyspnea, orthopnea and bilateral leg swelling and progressive decrease in urine output. She received three sessions of hemodialysis for her persistent anuria and deranged renal function before referral to our center. On examination her blood pressure was 92/60 mmHg PR - 100/min, Jugular venous pressure was raised and was icteric with subconjunctival hemorrhage. She had bilateral basal crepitations, has S3 gallop and tender hepatomegaly. CNS examination was normal at presentation. She had global hypokinesia with 20% ejection fraction in two-dimensional echocardiography. Dark ground microscopy was showing motile spirochetes in the urine sample and positive serum immunoglobulin M [Table 1]. Based on these findings, diagnosis of leptospirosis with severe myocarditis, ischemic transaminitis, and acute renal failure was made. On the 15th day of admission, the patient's urine output starts improving. She had about 5-6 L of urine daily on 18-19th day, however on 20th day she had two episodes of generalized tonic clonic seizures along with sudden painless loss of vision in both the eyes. Magnetic resonance imaging brain was showing bilateral occipital and left thalamic infarct. She was started on low dose steroid. Her vision started improving on the 23rd day. She was discharged on 28th day with complete recovery of her vision, serum creatinine of 1.8 mg/dl and repeat two-dimensional echocardiography was showing 54% ejection fraction.

Rare manifestation like CNS vasculitis has been reported, which can be due to direct endothelial damage by leptospira or late due to immune mediated mechanism.[4,5] Microscopically, perivascular round cell infiltration of small and medium sized blood vessels along with a patchy demyelination are the prominent features in vasculitic lesion. It can present as stroke with hemiparesis

Table 1: Laboratory investigation

Investigation	7th day of her illness (at admission)	15th day of her illness	20th day of her illness	28th day of her illness (at discharge)	35th day
Hemoglobin (g/dl)	9.3	9.1	9.4	11.8	11.6
TLC	8600	10,600	9600	7300	6500
DLC (N/L)	75/18	70/19	74/15	70/20	72/22
Platelet	1,88,000	2,12,000	1,60,000	2,50,000	1,90,000
Blood urea (mg/dl)	173	101	80	51	30
Serum creatinine (mg/dl)	5.3	5.7	3.48	1.8	1.3
Serum bilirubin total (mg/dl)	1.7	0.9	0.9	0.8	0.8
Direct (mg/dl)	1.1	0.5	0.6	0.5	0.5
Indirect (mg/dl)	0.6	0.4	0.3	0.3	0.3
SGOT (U/L)	289	46.50	44	40	35
SGPT (U/L)	794	5.32	24	9.1	12.0
PTI/INR	1.2	1.0	1.0	1.1	1.0
Total protein (g/dl)	6.5	6.4	7.0	7.0	6.8
Albumin/globulin (g/dl)	3.6/2.9	3.5/2.9	3.56/3.45	4.0/3.0	3.8/3.0
Complement C3 (mg/dl) Normal (88-165)	72			70	92
Complement C4 (mg/dl) Normal (14-44)	16.2			19	21
ANA	Negative				
dsDNA	Negative				
P-ANCA	Negative				
C-ANCA	Negative				
Leptospira serology					
IgG (U/ml)	18.0 (positive)	19.0 (positive)			
<10-negative					
10–15-inderminate					
>15-positive					
IgM (U/ml)	29.0 (positive)	26.0 (positive)			
<15-negative					
15–20-inderminate					
>20-positive					
Scrub typhus IgM	Negative				
Malaria antigen	Not detected				
Widal serology	Negative				

TLC=Total lymphocyte count, DLC=Differential leucocyte count, SGOT=Serum glutamic oxaloacetic transaminase, SGPT=Serum glutamic pyruvic transaminase, PTI=Protein thiolation index, INR=International normalized ratio, ANA=Anti-nuclear antibodies, dsDNA=Double stranded Deoxyribonucleic acid, P-ANCA=Perinuclear anti-neutrophil cytoplasmic antibodies, C-ANCA=Cytoplasmic anti-neutrophil cytoplasmic antibodies

or monoparesis. In our case it manifested as bilateral cortical blindness.

Ours is a unique case in the sense that the patient presented with CNS manifestation in the convalescent phase, recovering from severe leptospirosis and simultaneously producing 5–6 L of urine output daily. This situation can prone the patients for intravascular dehydration and create a fertile ground for immune complex deposition or thrombosis in small vessels. Whether the pathogenesis is vacuities or thrombosis is difficult to predict without biopsy. However, indirect evidence in the form clinical presentation of stroke with ischemic infarct, presentation in the convalescent phase and response to steroid indicate immune complex mediated vacuities. As our patient showed clinical improvement with steroid, biopsy was deferred. The clinician should be aware of the rare manifestation of leptospirosis, which can present even in a convalescent phase, especially in patients with multiorgan involvement.

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