Case Report

# Dengue Fever with An Unusual Radiological Manifestation : Mini - Boomerang Sign.

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**ABSTRACT**

Here we present a serologically and virologically diagnosed case of dengue fever with excruciating headache along with obtundation. MRI was ordered to see for the type of CNS involvement which revealed a hyper-intense lesion at the splenium of corpus callosum, reported as acute non-hemorrhagic infarct. Non congruent neurological and hematological features despite of a dreadful lesion on MRI, created a diagnostic dilemma. Extensive literature search brought forth case reports with similar self-limiting, benign lesions of various etiologies with varied pathogenic mechanisms. Transient affection of splenium of corpus callosum, is called as reversible splenial lesion syndrome (RESLES), which on MRI, morphologically highlights splenium to appear like a boomerang. This hyperintense lesion could be complete or partial to be called as "Boomerang Sign" or "Mini- Boomerang Sign" respectively. This case report emphasizes to refrain from unnecessary investigations considering the non-sinister nature of such lesions.

**Key words :** Dengue fever, Splenium of corpus callosum, Boomerang sign, Reversible splenial lesion syndrome.

## Introduction :

Dengue virus (DENV) is single stranded RNA virus of family Flaviviridae, responsible for dengue and severe dengue[1] in more than 100 countries in the WHO regions with Asia representing 70 % of the global burden[2]. Dengue encephalopathy, though commonly seen and reported, encephalitis is lesser known and published. Few literatures have reported dengue encephalitis with typical MRI changes of viral encephalitis and associated neurological features[3]. Isolated transient hyperintense lesion of splenium of corpus callosum have been reported in literature and is called as reversible splenial lesions syndrome[4] (RESLES), which on MRI, morphologically highlights splenium to appear as a "Boomerang". This boomerang shaped hyperintense lesion could be complete or partial to be called as Boomerang sign or Mini-Boomerang sign. Here we present a case of Dengue fever with transient and partial splenial hyperintense lesion without any congruent hematological and neurological features, emphasizing it's non sinister nature and urge to refrain from unnecessary investigation.

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## Case Report :

Patient was 26 years old male, engineering student with no pre-morbidities. There was no significant family, personal, social, medical and surgical history. Patient presented with complaints of fever with rigors, myalgia, excruciating retro bulbar headache and extreme prostrations.

***General examination :*** Temp-102$^0$F, heart rate - 100/min, respiratory rate - 24/min and blood pressure - 120/80 mmHg. There was no pallor, icterus, cyanosis or lymphadenopathy.

***Systemic examination :*** Respiratory system - chest clear, air entry equal both sides, no adventitial sounds, CVS - S1, S2 were normal, no murmurs, Per Abdomen - No guarding, rigidity and tenderness were appreciated, liver and spleen were not palpable and the bowel sounds were normal. CNS - Patient was conscious, responding to commands but at the same time seemed confused with GCS of 14 / 15 (E4V4M6), pupils were bilaterally equal and normally reacting to light with bilateral planters being flexor and a normal fundoscopy.

***Routine investigations :*** CBC, LFT, KFT, Malarial Antigen, Dengue profile, urine - routine/ microscopy were sent. Patient was started on PPIs,

Antipyretic, Artisunate, Clindamycin and Ceftriaxone. The above course was started as empirical therapy keeping in mind probability of bacterial meningitis and cerebral malaria as our patient had fever with rigors and febrile confusion, suggesting either bacterial or parasitic involvement of CNS on primary evaluation.

Preliminary blood reports revealed thrombocytopenia to the extent 60,000/mm$^3$ and NS1antigen, IgM, IgG were positive for dengue. CSF study, though was planned, was withheld in view of thrombocytopenia and absence of brain scan, hitherto.

Further with the above said treatment, patient had lesser peak and frequency of fever, platelets reached to a nadir of 55,000/mm$^3$ and started to rise there after, amidst all this patient had paroxysm of excruciating headaches associated with nausea, vomiting, photophobia and obtundation for which brain scan was sorted, as probability of intracranial hemorrhage was considered in view of thrombocytopenia, even typical viral pattern of CNS involvement as seen in any other viral encephalitis was anticipated.

Result of MRI brain startled us, as it revealed a hyperintense lesion at the splenium of corpus callosum, in DWI images, reported as acute non-hemorrhagic infarct, ***Figure 1***

On account of radiological finding of non-hemorrhagic infarct, entire hematological work-up for thrombocytopenia, including CBC, PT, PTT, Lupus anticoagulant, Anti-cardiolipin antibodies and proteins C and S, was done, and came out to be normal.

Even clinically there were no neurological features of hemispheric disconnection such as apraxia of left hand, pseudo neglect, alien left hand, agraphia, alexia and visual apraxias, pertaining to the site of CNS involvement on MRI.

More so, MRI features of infarct were localized to Diffusion-weighted images (DWI) and did not have concomitant changes in T1 / T2 and Flair images.

A neurological opinion was asked for, and taking into consideration the coagulation, neurological and radiological aspects of the lesion seen in MRI, it was though to be a non sinister lesion of lesser pathological gravity.

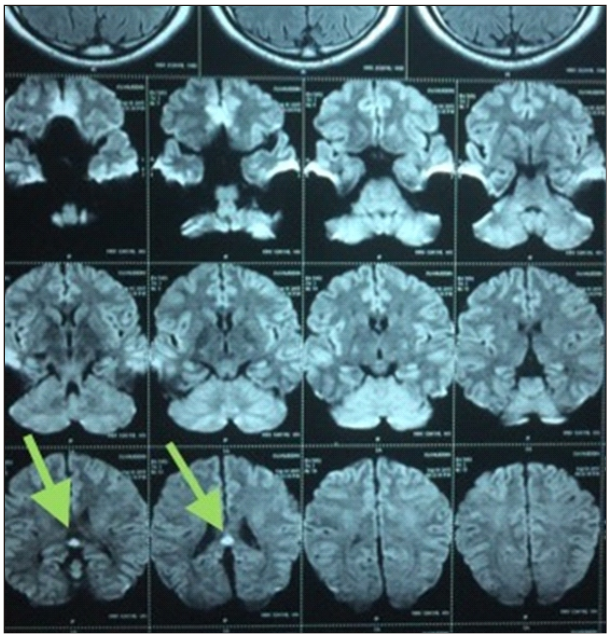

***Figure 1 :*** **DW images on MRI showing hyperintense lesions at splenium of corpus callosum, reported as acute non-hemorrhagic infarct.**

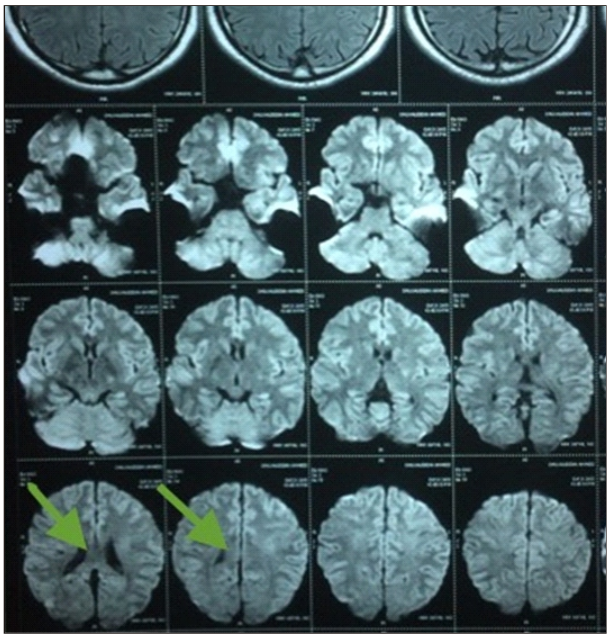

***Figure 2 :*** **Follow-up MRI at six weeks showing complete resolution of splenial hyperintensity, previously seen in DW images.**

Neurologist labelled this headache as stress related vascular headache and advised addition of Tab. Flunarizine to the ongoing treatment. Patient was fairly comfortable, without any headache and with platelet count above 100,000/mm$^3$ on day 7.

Patient was discharge the same day on PPI, MVI, Flunarizine and nutritional supplements, with an advice to inform in case of any further fever, headache or neurological deterioration. Follow up at 6 weeks with MRI brain, showed complete resolution of the splenial lesion, ***Figure 2***

**Discussion :**

Corpus callosum is the largest commissural white matter bundle in the brain containing 200-500 million inter hemispheric fibers[5], splenium exceptionally receives its blood supply from the vertebrobasilar system, while rest of the corpus callosum is supplied by the carotid system. Transient signal abnormality involving solely the splenium of corpus callosum on MRI is not frequently encountered in clinical practice, tempting the treating physician to subject the patient to a vast array of diagnostic and therapeutic interventions. Occurrence of this abnormality was first described by Chason et al. as a transient post-ictal focal edema denoting trans hemispheric propagation of seizure through the corpus callosum[6]. Since then various etiological factors have been associated with the transient hyper intensity of the splenium, ***Table 1.*** Various hypothesis have been put forth to explain the transient splenial changes; 1) Breakdown of blood-brain barrier, producing transient focal edema, in seizures[6]. 2) Reversible demyelination due to toxicity of anti-epileptic drugs (AED)[7]. 3) Sudden cessation of long term AED leads to alteration of arginine-vasopressin (AVP) system, resulting in hydric imbalance and cytotoxic edema[8]. 4) Viral antigens and receptors on the antibodies induced by the antigens have specific affinities for receptors on splenial axons, leading to raised inflammatory cytokines, IL-6, causing inflammation of splenium[9]. In our case, on premise of absent systemic metabolic derangements, encephalopathy was ruled out. Considering encephalitis MRI was advised, which showed a hyperintense lesion at the splenium of corpus callosum, reported as acute non-hemorrhagic infarct. Extensive hemophilia work-up was done which came to be normal, even there were no neurological signs of hemispheric disconnection such as apraxia of left hand, pseudo neglect, alien left hand, agraphia, alexia and visual apraxias[10]. In presence of a lesion at splenium with non-congruent

***Table 1*** **- Clinical conditions associated with transient splenial hyperintensity**

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| **Epilepsy** |
| Seizures |
| AED overdose |
| Abrupt drug withdrawal |
| **Infections** |
| Encephalitis |
| Salmonella |
| Malaria |
| Rota virus infection |
| **Demyelinating** |
| ADEM |
| SLE |
| **Metabolic** |
| Hypo / hypernatremia |
| **Hypoglycemia** |
| Renal failure |
| **Vascular** |
| Cerebrovascular disease |
| Post cardiac arrest |
| Hypertensive encephalopathy |
| Pre-eclampsia |
| Posterior reversible encephalopathy syndrome |
| Migraine with aura |
| **Miscellaneous** |
| Malnutrition-Vitamin B12 deficiency |
| Drug toxicity-cyclosporine, fluorouracil, metronidazole |
| High altitude cerebral edema |
| Trauma-axonal injury |

ADEM = Acute disseminated encephalomyelitis,
AED = Antiepileptic drugs,
SLE = Systemic lupus erythematosus

hematological and neurological features led to a diagnostic dilemma and we sorted to search the literate for similar such findings in the past. Literature showed similar findings as in our case, to mention a couple are, isolated transient splenial involvement in a case of refractory epilepsy, here the splenial involvement was complete giving a "Boomerang sign"[10]. Another example is partial involvement of splenium in a case of hemicrania continua, this partial splenial involvement is called as "Mini- Boomerang sign"[10], both the lesions were transient and showed subsequent resolution at six and three months respectively, similar to what was observed in our case.

## Conclusion :

Isolated transient hyperintense splenial lesions without congruent hematological and neurological features are usually benign and non-sinister in nature and should refrain the treating physician from unnecessary investigations.

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