Department of Neurology, St. John's Medical College Hospital, Bengaluru, Karnataka, India
E-mail: chakkuthom@hotmail.com

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Charles Bonnet syndrome in a case of cerebral venous thrombosis with fMRI-EEG correlation

Sir,

Charles Bonnet syndrome (CBS) is characterised by complex visual hallucinations with preserved insight in a patient with impaired vision or organic brain pathology. Occipital cortical and other cerebral resections, stroke, multiple sclerosis and temporal arteritis are the organic brain diseases associated with CBS. CBS in cerebral venous thrombosis (CVT) is not yet documented in English literature.

A 27-year-old labourer presented with 4 days history of headache, recurrent vomiting and gradual onset of painless bilateral vision loss. He complained of seeing transient images of vivid human figures like men in uniform, lady combing her hair, lady standing with a shawl, a crying baby, landscapes, buildings and also animals like cat. They lasted only for few seconds to minutes and occurred infrequently almost daily. Patient was aware these were non-existent. They occurred more often in dim light and in evenings. The complex visual hallucinations (VH) disappeared on looking at them and when someone talked to him. There was no past history of psychiatric illness. He had no perception of light in left eye and visual acuity was 3/60 in the right eye. Fundus revealed florid papilledema with haemorrhage and macular exudates. Perimetry revealed enlarged blind spot with concentric constriction of visual fields. There were no other neurological deficits. Computed tomography (CT) brain showed hyper dense superior sagittal sinuses with empty delta sign. [Figure 1a and b]. Magnetic resonance imaging (MRI) showed filling defect in superior sagittal sinus and right transverse sinus and loss of flow void in superior sagittal sinus in T2. MR-venogram revealed thrombosis of superior sagittal sinus, right transverse and straight sinus and few cortical veins [Figure 2b]. Repeat imaging (10 days after onset) showed additional appearance of parenchymal bleed in right inferior frontal gyrus, left temporal region and multiple micro bleeds in susceptibility weighted images. [Figure 3a-c]. Resting functional MR-images (fMRI) were acquired using a 3T scanner (Skyra, Siemens, Erlangen, Germany). For design specification and model of the fMRI the patient was asked to press the start response button at the onset of hallucination and press the end response button at the end of hallucination so as to record the

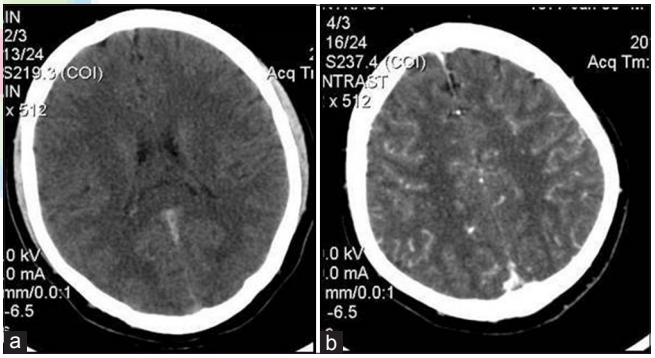

Figure 1: (a and b) Hyper dense superior sagittal sinus in plain and empty delta sign on contrast scan respectively

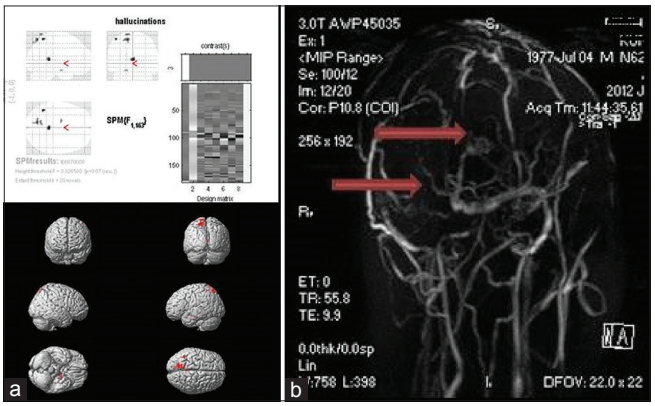

Figure 2: (a) FMRI showing activation of bilateral (more on dominant side) parietal and temporal regions and (b) MR Venography showing non visualisation of superior sagittal sinus and right transverse sinus

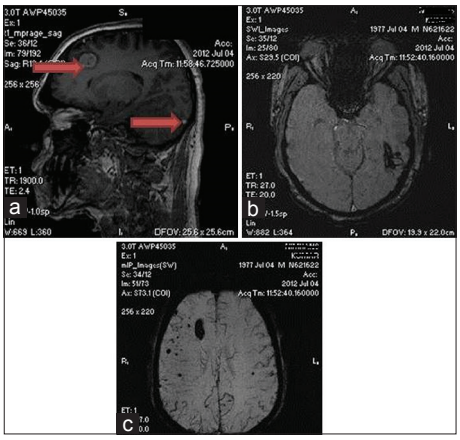

Figure 3: (a) T1 Sagittal view showing frontal sub acute bleed with loss of flow void in right transverse sinus. (b and c) susceptibility weighted images showing right frontal, left temporal bleeds and micro bleeds with spared parietal lobe

onset and duration of hallucination. 3 sets of visual hallucination in fMRI scan of 9 minutes were recorded. Analysis was performed using statistical parametric mapping (SPM8; Welcome Department of Cognitive Neurology, London). EEG data were recorded using a 32-channel MR compatible EEG system (Brain Products, Gilching, Germany). Functional MRI acquired during hallucination revealed activation of bilateral parietal and temporal cortex. [Figure 2a]. Electroencephalography during rest and visual hallucinations was normal.

He also had prothrombotic risk factors like alcoholism, low B12 levels, hyper homocysteinemia and polycythaemia. Patient was started on low dose subcutaneous heparin 5000 U 6 hourly for 7 days overlapped later with acenocoumarol. Visual acuity was monitored regularly and repeat MR-venogram after 2 weeks showed resolution of parenchymal bleed. His follow up perimeter revealed improvement in the visual field. Acuity gradually improved over next 2 weeks to 6/12 in right eye and 6/24 in left eye. As his vision started improving, his hallucinations disappeared over the next 2 weeks period.

CVT presents with a wide spectrum of neurologic features.[1] Acute vision loss with papilledema was described in a patient with severe occipital lobe oedema secondary to venous congestion, pseudo tumour cerebri and cortical venous thrombosis.[2] Intracranial venous sinus thrombosis leading to intracranial hypertension is uncommon.[3] Severe visual loss due to CVT rarely occurs (2% to 4%). On studying 624 cases, Ferro *et al*.[4] found that persistent visual loss (visual field defects in 6 patients and decreased visual acuity in 21 patients) was more frequent in patients diagnosed as CVT with raised ICT with a delay of more than 4 days from onset of symptoms. Purvin *et al*.[1] described reduced visual acuity in ten patients of cerebral venous thrombosis with raised ICT.

Several pathologic conditions like focal epilepsy, migraine, diffuse lewy body disease, brainstem or thalamic lesions can cause complex hallucinations. Mechanisms hypothesised are epileptic hallucinations due to irritative process on the cortical centres, visual pathway lesions causing defective visual input and defective visual processing and brainstem lesions affecting various neurotransmitters.[5] Central to the genesis of complex visual forms in man is the visual association (extra striate) cortex.[5,6]

Manford *et al*.[5] described two cases of visual hallucinations in abnormal visual field in posterior cerebral infarct involving the occipital cortex. He proposed the "deafferentation" theory and "perceptual release" theory. Ffytch *et al*.[7] and Santhouse *et al*.[8] linked certain regions of fMRI signal activation to specific hallucinatory experiences. There was no occipital cortex lesion in our patient and epileptic activity was ruled out by EEG. Vision loss was due to raised ICT with Cerebral venous thrombosis. Hence we hypothesise, that the VH and CBS seen in our patient was probably due to venous congestion and sensory deprivation of visual cortex with subsequent cortical release of extrastriate cortex as evident by FMRI –EEG studies. Management of CBS is supportive and few drugs like sodium valproate, gabapentin, olanzapine and carbamazepine can alleviate symptoms. The CBS in our patient resolved with management of CVT.

R. Subasree, Girish B. Kulkarni, Veerendra Kumar M., Rose Dawn Bharath[1], Sandhya M[1]., Ravi Yadav, Sailesh Modi, Nitish Kumar, Rajanikant Panda[1]

Departments of Neurology, and [1]Neuroimaging and Interventional Radiology, National Institute of Mental Health and Neurosciences, Bangalore, Karnataka, India
E-mail: subasree.ramakrishnan@gmail.com

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Levetiracetam for tardive dystonia: A case report

Sir,

Tardive syndrome (TS) is a group of hyperkinetic or hypokinetic movement disorders and sensory symptoms sharing the same pathophysiological basis. This neurological disorder most frequently occurs as the result of long-term or high-dose use of antipsychotic drugs. The etiological theories and treatment strategies have been elucidated recently.[1,2] Of late, levetiracetam is gaining importance as a novel therapeutic agent for TS.[3-8] This report presents a case of trifluoperazine-induced tardive dystonia who partially responded to adjunctive treatment with levetiracetam.

A 46-year-old lady presented to the psychiatry outpatient services with abnormal, spontaneous, repetitive, nonrhythmic, and dystonic movements involving the cranial and cervical region. Patient had blepharospasm, grimacing, puckering, pouting and clenching, and neck muscle dystonia. The movements decreased on resting her head to a hard surface, increased with anxiety and disappeared in sleep. She had been prescribed escitalopram 20 mg with clonazepam 1 mg for panic disorder by a private practitioner, along with trifluoperazine 10 mg possibly for agitation. There was no history of any other psychiatric/medical/surgical illness in the past. On examination, her vitals were stable. Other than the abnormal movements, she had no other neurological findings. Her Abnormal Involuntary Movements Scale (AIMS) score was 22. As her symptoms started after neuroleptic exposure, a diagnosis of tardive dystonia with panic disorder was considered. Blood biochemistry, complete blood picture, ECG, EEG, and brain imaging were essentially normal. Mental state examination revealed anxiety/depressive features with speech difficulty. She was followed-up in outpatient department for the next 1 year, during which she was tried on multitude of drugs—tetrabenazine (100 mg), trihexyphenidyl (8 mg), clonazepam (2 mg), valproate (1,000 mg), pregabalin (600 mg), tizanidine (0.6 mg), baclofen (60 mg), and clozapine (75 mg) for adequate duration at tolerable doses in various combinations. She showed minimal improvement with clozapine (AIMS score - 21), but dose could not be titrated above 75 mg due to tachycardia and new onset 't-wave' inversion because of which it was subsequently discontinued. While she was on tetrabenazine (100 mg) and clonazepam (1.5 mg), she was prescribed levetiracetam, dose of which was gradually increased to 1,000 mg in divided doses. Her AIMS score decreased to 14 following 1 month of prescription. She had never experienced such significant improvement. Mirtazapine (15 mg) was also added for her anxiety and depressive symptoms.

In previously reported cases, levetiracetam was administered as monotherapy.[3-8] Our patient was already on tetrabenazine and clonazepam, but showed improvement only after the addition of levetiracetam. It can be argued that probably the improvement could be a late therapeutic effect of the other drugs, but considering they were tried at adequate (tolerable) dosages for adequate time it seems less likely. Levetiracetam acts on synaptic vesicle protein 2 (SV2); thereby inhibiting the release of various neurotransmitters and modulating glutamatergic, GABAergic, dopaminergic, and cholinergic systems.[7,9,10] Levetiracetam seems to be effective with respect to all the proposed pathophysiological theories of TS.[1] And as such, it has been found to be useful in almost all kinds of hyperkinetic disorders.[9]

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Ajish Mangot, Satyakant Trivedi, Ravindra Kurrey, Vaibhav Dubey

Department of Psychiatry, People's College of Medical Sciences and Research Centre, Bhanpur, Bhopal, Madhya Pradesh, India
E-mail: dr.ajish@outlook.com

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