patient became seizure free and was able to decrease his antiseizure medications (CBZ from 600mg bid to none, and LTG from 150mg bid to 75mg bid) over four months.

One month after resection the patient developed some psychiatric features including difficulty accepting his seizure free status, obsessions of contamination, hyperactive and manic episodes, decreased behavioural flexibility, and significant ethanol abuse. His neuropsychological reassessment showed no change, and his EEG showed theta slowing over the right temporal region but no epileptiform activity. His psychiatric features initially resolved after increasing lamotrigine to 125mg twice a day and treatment with risperidone for one month. His alcohol abuse also resolved after spending some time in a rehabilitation center.

He restarted working full time four months post resection surgery. Unfortunately, after a few months the patient started to abuse ethanol again with significant negative impact on his performance at home and work well as one generalized tonic clonic seizure due to ethanol withdrawal. Later, he developed depression and aggression towards his family. He was referred to a psychiatrist and treatment with venlafaxine at 75mg per day was started for depression and dextroamphetamine 5mg one to two times per day was also started which improved his cognitive slowing. One year after the temporal lobectomy he was referred again to an ethanol-dependence rehabilitation center. He remains seizure free after two years of follow-up with intermittent burst of alcohol ingestion.

This patient developed depression, decreased behavioural flexibility, and significant ethanol abuse after right temporal lobectomy. As temporal resection and discontinuation of CBZ occurred around the same time, it is difficult to identify one of these factors as an independent cause for ethanol abuse. Studies in animal models have shown that ethanol reward appears to depend on an interaction with the GABAA receptor, dopamine, and opioid peptides in the reward pathway that includes the limbic system[4].

Therefore, mesial temporal and limbic resections presumably affect ethanol abuse in humans; however, it is not known if the unilateral resection such as in our patient will have a significant clinical effect. Our patient had familial predisposition to ethanol abuse, which could have remained dormant due to treatment with carbamazepine for many years. After epilepsy surgery, CBZ was discontinued as he became seizure free and therefore his alcohol dependence may have been revealed. Previous case series and randomized open-labeled clinical trials have shown that CBZ and oxcarpazepine likely decrease alcohol dependence and recurrence of abuse after a period of abstinence[5].

This is the first case reporting the onset of alcoholism after a right temporal resection. As we mentioned in our discussion this patient had other risk factors to develop alcoholism including the family history of alcoholism and the withdrawal of CBZ. Overall we believed that the onset of alcoholism was multifactorial, although the mesial and neocortical structures of the right temporal region could be involved in the pathophysiology of alcoholism. In conclusion based in our case it is important to consider strong family history of ethanol dependence as a possible relative contraindication for temporal lobectomy or discontinuation of CBZ post operatively. Further assessment of this issue in a large number of patients is required to confirm the association.

Farzad Moien-Afshari, José F. Téllez-Zenteno
Royal University Hospital, University of Saskatchewan, Saskatoon, Saskatchewan, Canada

References

1. Wiebe S, Blume WT, Girvin JP, Eliasziw M. A randomized, controlled trial of surgery for temporal-lobe epilepsy. N Engl J Med. 2001; 345(5):311-18.
2. Wilson SJ, Saling MM, Lawrence J, Bladin PF. Outcome of temporal lobectomy: expectations and the prediction of perceived success. Epilepsy Res. 1999; 36(1):1-14.
3. Butman J, Allegri RF, Thomson A, et al. Behavioral flexibility impairment with negative feedback in refractory temporal lobe epileptic patients with unilateral amygdala and hippocampal resection. Actas Esp Psiquiatr. 2007; 35(1):8-14.
4. Koob GF. Drugs of abuse: anatomy, pharmacology and function of reward pathways. Trends Pharmacol Sci. 1992; 13(5):177-84.
5. Ait-Daoud N, Malcolm RJ, Jr., Johnson BA. An overview of medications for the treatment of alcohol withdrawal and alcohol dependence with an emphasis on the use of older and newer anticonvulsants. Addict Behav. 2006; 31:1628-49.

To the Editor

A Case of Bilateral Homonymous Hemianopsia with Macular Sparing

A 55-year-old woman presented in December 2008 with a complete left homonymous congruent hemianopsia. Multiple territories stroke was diagnosed, including the right occipital lobe. At this time, a superior left lobe tumor of lung was suspected and out-of-hospital investigated. She was discharged with ASA and atorvastatin. In January 2009, she presented with complete right homonymous hemianopsia but was still able to see with her central field of vision. The Goldmann's test showed a sparing of central vision of about 5 to 10 degrees on both sides, with the exception of an inferior right homonymous quadranoptic macular defect. (Figure A) Once again, multiple territories stroke was diagnosed, including one in her left occipital lobe.

The pneumology team diagnosed a lung adenocarcinoma (3b stage). The complete cardiovascular investigation was normal. An exhaustive hematologic testing has shown anticardiolipin antibodies (IgM) increased at 14.6 UPL (N<9.0). ASA has been stopped and she left hospital with dalteparin. At the end of February, she demonstrated improved vision in the right temporal crescent by Goldmann's visual field test.

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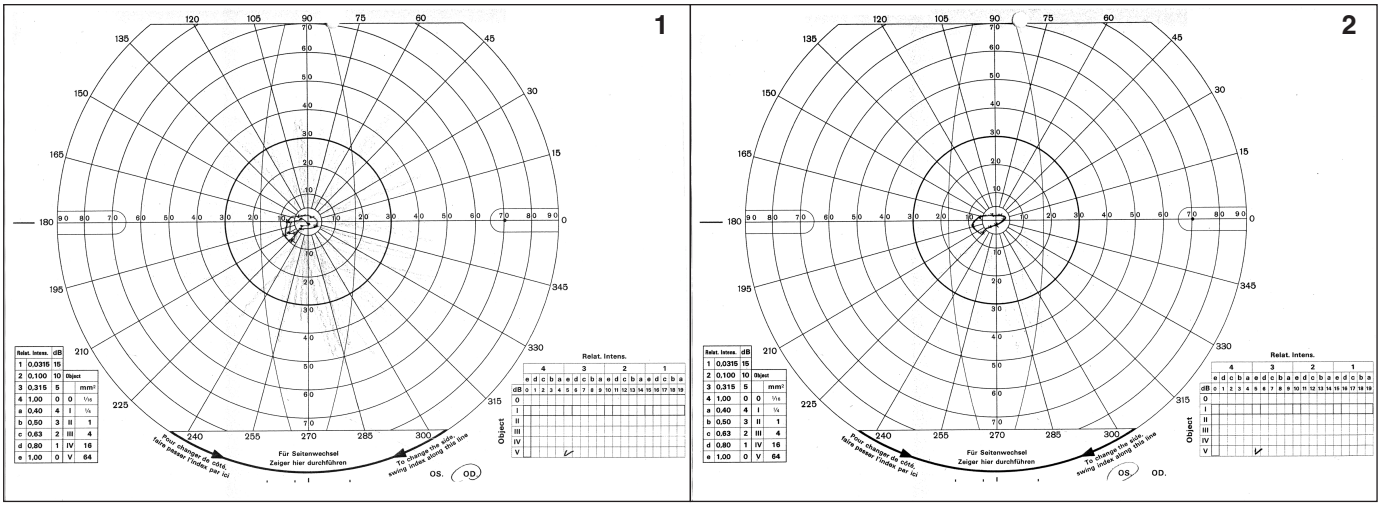

Figure A: *Goldmann's tests of 01-2009 showing macular sparing. Middle of circle shows the intact visual field.*

In December 2009, a left middle cerebral artery stroke was diagnosed while she was taking delteparin. (Figure B) She died in January 2010 from a pericardiac and peritoneal carcinomatosis.

Discussion

Curiously, even if it is well known that occipital stroke can cause a hemianopsia with macular sparing, there is only one 1951 report in the literature of bilateral occipital stroke with macular sparing.[1]

Macular sparing is a debated subject since many years in neurology. Many hypotheses exist to explain the phenomenon and one of them is the bilateral representation of macular vision in occipital cortex.[2] This concept is vanishing because there is no anatomical evidence to prove it. Our case, with bilateral lesions, is against this theory since the second lesion should have cancelled the initial macular sparing cause by the first stroke.

Another hypothesis is simply an incomplete damage to the striate cortex, where the macular vision is largely represented in the occipital pole and operculum.[2] In fact, 50 - 60% of the posterior striate cortex represents ten degrees of central vision.[3] This hypothesis could probably explain in part or by itself the phenomenon, since the redundancy of macular representation in occipital lobe may confer a certain function resistance to lesions.

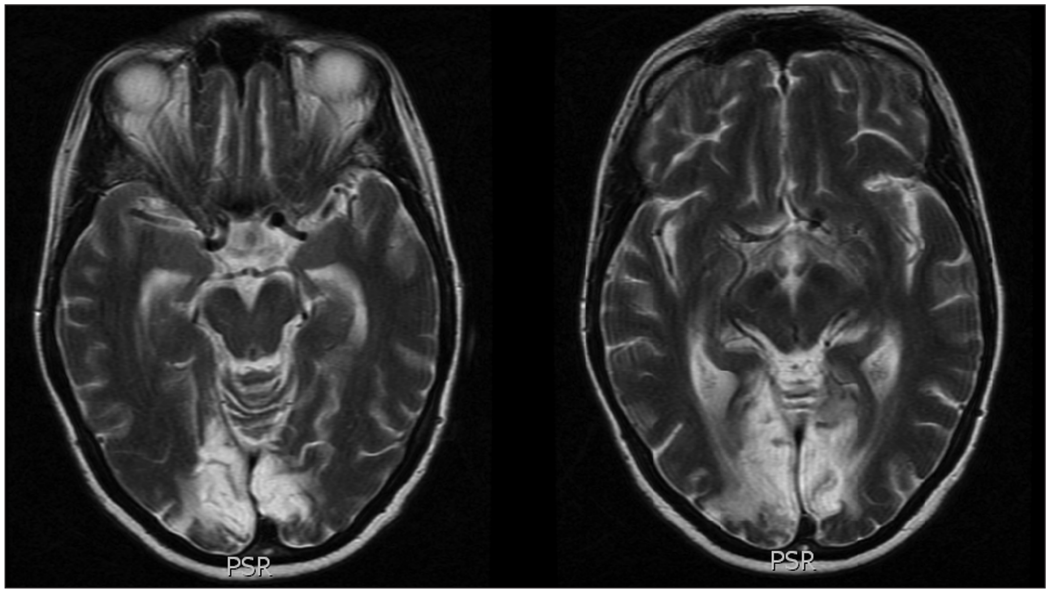

Figure B: *MRI of 12-2009 showing increased T2 signal in both occipital lobes.*

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Otherwise, the most frequent cause of hemianopsia is vascular events, followed by trauma and brain tumor.[4] Consequently, a third hypothesis, the most accepted one, to explain macular sparing is a double vascularization of the posterior striate cortex. Posterior cerebral artery supplied occipital lobe by four branches (posterior temporal, anterior temporal, parieto-occipital and calcarine). Calcarine artery supplies the whole striate cortex in fifty percent of people.[2] The middle cerebral artery also supplies occipital lobe with the posterior temporal artery and the artery of the angular gyrus.[5] Another vascular explanation for macular sparing is bifurcation of posterior temporal artery before the calcarine artery. Occipital cortex could be supplied by the posterior temporal artery even if there is a thrombosis in calcarine artery. This explanation may be incomplete because posterior temporal artery supplies occipital cortex in only fifty percent of cases, but as we know, macular sparing is far of being a universal phenomenon in ischemic stroke.[2]

Many others hypotheses are written in literature, but to our opinion, the more relevant are listed above.

On the other hand, anticardiolipin antibodies are a cause of arterial and venous thrombosis. It is also known that anticardiolipin antibodies are frequently elevated in association with malignancy. In our case, we suppose that a hypercoagulable state is the cause of recurrent strokes in this patient.

In conclusion, we think this case illustrate one more proof that, at least in our case, macular sparing is more likely explained by a vasculature hypothesis or by a large representation of macular vision in striate cortex rather than by a bilateral occipital macular representation.

Anne-Marie Dufresne, Martin Savard
Quebec, QC, Canada

References

1. Steiner A. Homonymous hemianopia, bilateral with macular sparing, of vascular origin. Am J Ophtalmol. 1951;34(8):1170-3.
2. Leff A. A historical review of the representation of the visual field in primary visual cortex with special reference to the neural mechanisms underlying macular sparing. Brain Lang. 2004;88: 268-78.
3. McFadzean R, Brosnahan D, Hadley D, Mutlukan E. Representation of the visual field in the occipital striate cortex. Br J Ophthalmol. 1994;78:185-90.
4. Zhang X, Kedar S, Lynn MJ, Newman NJ, Biousse V. Homonymous hemianopias: Clinical–anatomic correlations in 904 cases. Neurology. 2006;66:906-10.
5. Tanaka R, Miyasaka Y, Yada K, Mukuno K. Bilateral homonymous hemianopsia due to temporal herniation, with sparing of central vision: Case report. Neurosurgery. 1992;31(4):787-91.

To the Editor

Paraneoplastic Encephalomyelitis, Stiff Person Syndrome and Breast Carcinoma

Paraneoplastic syndromes (PNS) are the rarest non-metastatic neurological complications of cancers in which no specific etiology can be identified. Among PNS, stiff person syndrome is characterized by "rock-hard" rigidity and painful spasms in distal limbs, most frequently legs, and abnormal foot and/or hand posture. General posture is rigid; ambulation is difficult and may result in falls. Paraneoplastic syndromes are postulated to be autoimmune diseases. Stiff person syndrome associated to cancer display high antibody titers directed against neuronal proteins[1,2].

We present the case of a woman first diagnosed with stiff person syndrome, which led to the diagnosis of an invasive ductal carcinoma of the breast. Paraneoplastic antibodies titers previously reported in literature were all negative.

Case Report

A 30-year-old woman presented to the hospital in October 2006 for involuntary muscle contractions of the legs, making walking difficult with occasional falls. Spasms were painful, waking her in the night, and it was possible to provoke them by applying pressure to the lumbar region. Legs and arms were stiff, legs being much worse. Spasms were treated with benzodiazepine with very minimal improvement.

The investigation included the following (in chronological order): cerebral magnetic resonance imaging (MRI), cervical/thoracic/dorsal MRI, electromyogram, electroencephalogram, lumbar/sacral MRI and mammography. Mammography revealed the presence of a mass and asymmetric density in her left breast. Percutaneous biopsy revealed an invasive ductal carcinoma, grade I/III, estrogen receptor-positive, progesterone receptor-positive and HER2-negative. Positron emission tomography revealed one metastasis in left internal mammary node chain and another one in left axillary region. There was no other metastasis.

Cerebral and spinal magnetic resonance imagery showed neither lesion nor metastasis. Lumbar puncture showed slightly elevated white cells count ($6x10^6$/L), but normal proteinorachia. The following anti-neuronal autoantibodies were performed and were all negative: anti-Hu, anti-Ri, anti-Yo (anti Purkinjee cells antibodies (ab)), anti-glutamic acid decarboxylase (GAD) ab, anti-amphiphysin ab, anti-CRMP-5 ab, anti-striated muscle ab, anti-P/Q-Type calcium channels ab, anti-N-Type calcium channels ab, acetylcholine receptor binding ab and anti-acetylcholine receptor ganglionic neuronal ab.

She received intravenous immunoglobulin (IVIG) treatment in November 2006 with no effect. Lumpectomy and axillary dissection were performed on December 11th, 2006. Pathology reports a T_2 $N_{1/18}$ M_0 invasive ductal carcinoma. Neurological symptoms were not improved following surgery. Gabapentin and clonazepam were added to treatment, improving spasms control

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