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Diabetic Striatopathy Complicated by Fatal Laminar Cortical Necrosis: A Case Report

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Case Report

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Abstract

Background: Diabetic striatopathy (DS), or non-ketotic hyperglycemia-induced hemichorea-hemiballism, is a rare movement disorder that occasionally serves as the initial presentation of previously undiagnosed diabetes. While its course is generally favorable, it can exceptionally be complicated by laminar cortical necrosis (LCN), a formidable and most often fatal condition.

Case Presentation: A 60-year-old male with a history of poorly controlled type 2 diabetes was admitted for acute hemichorea-hemiballism associated with severe non-ketotic hyperglycemia of 11 g/L (1100 mg/dL). Brain MRI confirmed diabetic striatopathy involving the basal ganglia. Despite initial improvement under haloperidol and glycemic correction, the patient presented two weeks later with sudden neurological deterioration characterized by behavioral disturbances and hallucinations. Follow-up MRI revealed laminar cortical necrosis, leading to the patient's death within 48 hours.

Discussion and Conclusion : This case illustrates an extremely rare and severe clinical association between diabetic striatopathy and laminar cortical necrosis. It highlights that while magnetic resonance imaging (MRI) is essential for early diagnosis and monitoring, the onset of this cortical complication turns a typically benign metabolic syndrome into a fatal outcome, emphasizing the need for future multicenter studies to better identify its risk factors.

Introduction

Diabetic striatopathy (DS), or non-ketotic hyperglycemia-induced hemichorea-hemiballism, is a rare movement disorder occurring in the context of hyperglycemia without ketosis. It frequently serves as the initial presentation of previously undiagnosed diabetes in 17% of cases [1, 2]. In 88.1% of cases, it manifests as hemichorea-hemiballism (HC-HB) [2]. Neuroimaging most commonly reveals isolated putaminal involvement or combined involvement of the putamen and caudate nucleus [2].

The exact pathophysiological mechanisms of these striatal abnormalities during non-ketotic hyperglycemia remain controversial [3, 4]. The two most strongly supported theories rely on tissue ischemia induced by hyperviscosity and/or acute metabolic disturbances, alterations in neurotransmitters such as gamma-aminobutyric acid (GABA), and blood-brain barrier dysfunction [1, 5, 6, 7].

Although this metabolic and functional syndrome usually follows a favorable course with glycemic correction, it can occasionally take an unexpected and formidable clinical turn. Among these rare evolutionary pitfalls is the occurrence of laminar cortical necrosis (LCN), an exceptional complication with a most often fatal prognosis. The medical literature reports only rare cases associating these two entities. Because of this rarity, there are few large-scale studies on its incidence, demographic characteristics, symptom correlation, and the localization of striatal abnormalities [1, 2, 8].

Herein, we report the case of a patient presenting with severe non-ketotic hyperglycemia revealed by a hemichorea-hemiballism syndrome, whose evolution was fatal due to the onset of laminar cortical necrosis.

Case Presentation

A 60-year-old right-handed man, with a medical history of type 2 diabetes mellitus treated with insulin and poor compliance, was admitted to the emergency department for the acute onset of involuntary, uncontrollable, and random abnormal movements affecting the right hemibody. These movements were abrupt, violent, and large-amplitude, with a proximal component and a tendency to worsen with emotional distress.

Initial laboratory workup revealed severe hyperglycemia of 11 g/L (1100 mg/dL) without ketosis, associated with a glycated hemoglobin (HbA1c) level of 12%. Fluid and electrolyte balance, as well as renal and hepatic functions, were unremarkable. Retroviral and syphilitic serologies were negative, and there was no clinical or biological evidence suggesting an infectious process.

Initial brain magnetic resonance imaging (MRI) revealed hyperintensity on FLAIR (Fig. 1A) and diffusion-weighted imaging (DWI) (Fig. 1B) sequences in the left head of the caudate nucleus and the left insular region, associated with a corresponding low apparent diffusion coefficient (ADC) signal (Fig. 1C). Gradient-echo sequence showed no evidence of hemorrhage or calcification (Fig. 1D).

Cardiac evaluation and Doppler ultrasound of the supra-aortic trunks were normal. Given these radiological findings in the context of severe hyperglycemia and poorly controlled diabetes, a metabolic etiology (diabetic striatopathy) was established.

Initial clinical course was marked by a progressive regression of involuntary movements within 48 to 72 hours under haloperidol, combined with the gradual correction of hyperglycemia. However, two weeks later, the patient presented with acute neurological deterioration characterized by behavioral disturbances, agitation, oneiric confusion, and visual hallucinations (the patient reported seeing animals and experiencing a sensation of free fall). Performed urgently in response to this clinical worsening, the follow-up brain MRI revealed spontaneous gyrate hyperintensity of the left cortical ribbon on diffusion-weighted imaging sequences (Fig. 2E) and of the insular region on the FLAIR sequence (Fig. 2F). The electroencephalogram (EEG) showed global and diffuse slowing of the background activity, without epileptiform paroxysms (Fig. 3).

The subsequent clinical course was rapidly fatal, with death occurring in a context of neurological failure 48 hours later

Discussion

1. Clinical and Epidemiological Features of Diabetic Striatopathy

Diabetic striatopathy is a rare disorder that can occur in patients presenting with hyperglycemia without ketosis. It occurs in 96.6% of cases in patients with known type 2 diabetes and serves as the initial presentation of diabetes in 17% of cases, with mean blood glucose and glycated hemoglobin concentrations of 414 mg/dL and 13.1%, respectively [2, 6]. In its typical presentation, hemichorea-hemiballism is the revealing clinical sign in 88.1% of cases [5, 6], with MRI showing a unilateral hyperintense lesion affecting the basal ganglia, which may disappear after the correction of hyperglycemia [1, 9].

Its estimated prevalence is approximately 1 per 100,000 diabetic patients, with a predominance in elderly Asian women with long-standing diabetes [1, 2, 7]. Although our patient is male and of sub-Saharan origin, the clinical picture serves as a typical illustration of diabetic striatopathy, suspected in the presence of hemichorea-hemiballism-type abnormal movements in the context of glycemic imbalance [6].

In our observation, the usual metabolic mechanisms do not entirely explain the persistence of abnormal movements after the correction of hyperglycemia, a particularity that could be a consequence of the occurrence of laminar cortical necrosis.

2. Clinical and Radiological Diagnosis

Hemichorea-hemiballism can also occur in the setting of acute ischemic or hemorrhagic stroke affecting the contralateral basal ganglia, particularly the putamen [5, 6, 10]. However, in our case, the diagnosis of diabetic striatopathy was established based on clinical and radiological arguments, notably the acute onset of right hemichorea-hemiballism in the context of non-ketotic hyperglycemia, with brain MRI showing hyperintensity of the left caudate nucleus head.

Another particularity of our observation was the secondary occurrence of laminar cortical necrosis (LCN), a rare clinicoradiological entity. Brain MRI is the only modality capable of detecting it on T1-weighted sequences [1, 4, 10] and monitoring its evolution on FLAIR and diffusion-weighted sequences [11, 12]. LCN can also be observed following prolonged arterial hypotension, hypoglycemia or hyperglycemia, epileptic disorders, or cardiorespiratory arrest. The patient was neither hypotensive nor hypoglycemic, and the EEG revealed no epileptiform abnormalities suggestive of status epilepticus (Fig. 3).

Follow-up MRI revealed spontaneous gyrate hyperintensity of the left cortical ribbon, visible on diffusion-weighted sequences (Fig. 2E) and FLAIR (Fig. 2F), without systematic distribution within a vascular territory, which formally ruled out a stroke. Spontaneous gyrate hyperintensity of the cortical ribbon on MRI constitutes the typical radiological signature of laminar cortical necrosis in the subacute phase.

3. Treatment and Evolution

3.1. Treatment

The management of diabetic striatopathy is based primarily on hospitalization and the correction of hyperglycemia [4]. Involuntary movements often improve within weeks following glycemic control combined with antidopaminergic medications [1, 7]. Our patient was started on haloperidol concomitantly with the correction of hyperglycemia.

3.2. Evolution

In the absence of complications, DS generally has a good prognosis. Recurrence remains possible in 18% of cases, even after the normalization of blood glucose levels [1, 7]. In our case, the clinical evolution was initially favorable under treatment, marked by a regression in the frequency and amplitude of abnormal movements. However, two weeks later, the patient presented with clinicoradiological deterioration characterized by the onset of a confusional state with hallucinations, the identification of laminar cortical necrosis on the follow-up brain MRI, and subsequent death occurring 48 hours later.

4. Study Limitations

Our work has certain limitations that should be highlighted. The absence of more invasive complementary examinations or a post-mortem neuropathological study precluded an exhaustive exploration of the intimate cellular and ischemic cascade that led to the occurrence of laminar cortical necrosis [11, 13]. Finally, the rarity of the association between these two entities (diabetic striatopathy and laminar cortical necrosis) limits the comparison of our case with large series in the literature, highlighting the need for future multicenter studies to better identify the risk factors for this fatal complication.

Conclusion

Although hemichorea-hemiballism is a classic and well-documented complication of non-ketotic hyperglycemia, it remains underdiagnosed. It can serve as the initial presentation of previously undiagnosed diabetes or occur during major glycemic imbalance. Laminar cortical necrosis represents a rare, formidable, and particularly severe secondary complication. In this context, brain MRI proves essential for the early detection of these lesions and their evolutionary monitoring.

Abbreviations

ADC
Apparent Diffusion Coefficient
DS
Diabetic Striatopathy
DWI

Diffusion-Weighted Imaging
EEG
Electroencephalogram
FLAIR
Fluid-Attenuated Inversion Recovery
HbA1c
Glycated Hemoglobin
HC-HB
Hemichorea-Hemiballism
LCN
Laminar Cortical Necrosis

Declarations

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Authors' Contributions

General contribution : All authors contributed to this work. **Conceptualization :** M.Albakaye, Y. Maiga, S.H. Diallo, N. Kissani. **Topic identification and manuscript writing:** M. Albakaye, Y. Maiga, N. Kissani. **Supervision :** Y. Maiga, N. Kissani, S.H. Diallo, T. Coulibaly, G. Landouré. **Iconography:** A. Coulibaly, S. Diallo, M. Dao, Z. Traoré. **Final manuscript version correction/review:** Y. Maiga, N. Kissani, S. Diallo, M. Albakaye, S. Boubacar, D. Ben, G. Landouré, T. Coulibaly.

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Availability of data and materials

Not applicable.

Declarations

Ethical approval and consent to participate

Ethical approval for this study was not needed as per institutional ethics policy, as this study is a case report of a single patient and did not include protected health information, data analysis, or testing of a hypothesis. Written informed consent from the patient attendant for the study and the publication was obtained.

Consent for publication

Written informed consent for the publication of this case report and any accompanying images was obtained from the patient's legal proxy.

Indeed, at the time of evaluation, the patient presented with severe acute neuropsychiatric symptoms, including confusion, agitation, hallucinations, and severe hyperreactivity to physical or verbal stimuli (which triggered abnormal movements and a distressing sensation of falling). Due to this altered mental status, the patient was clinically assessed as lacking the capacity to fully comprehend the nature of the publication, weigh its risks and benefits, and provide informed consent. Consequently, to protect the patient's rights and best interests, written informed consent for publication was legally and ethically obtained from the accompanying family member and legal proxy.

Competing interests

No conflicts of interest are related to this article.

References

1. Oh SH, Lee KY, Im JH, Lee MS. Chorea associated with non-ketotic hyperglycemia and hyperintensity basal ganglia lesion on T1-weighted brain MRI study: a meta-analysis of 53 cases including four present cases. J Neurol Sci. 2002 Aug 15;200(1-2):57-62. DOI: 10.1016/s0022-510x(02)00133-8. PMID: 12127677.
2. Chua CB, Sun CK, Hsu CW, Tai YC, Liang CY, Tsai IT. "Diabetic striatopathy": clinical presentations, controversy, pathogenesis, treatments, and outcomes. Sci Rep. 2020 Jan 31;10(1):1594.DOI: 10.1038/s41598-020-58555-w. PMID: 32005905; PMCID: PMC6994507.
3. **.**Battisti, C., Forte, F., Rubenni, E. *et al.* Two cases of hemichorea-hemiballism with nonketotic hyperglycemia: a new point of view. *Neurol Sci* **30**, 179–183 (2009). https://doi.org/10.1007/s10072-009-0039-5
4. **.** Lin JJ., Lin GY., Shih C., Shen WC. Presentation of striatal hyperintensity on T1-weighted MRI in patients with hemiballism-hemichorea caused by non-ketotic hyperglycemia: Report of seven new cases and a review of literature. *J Neurol* **248**, 750–755 (2001).https://doi.org/10.1007/s004150170089
5. . Sayantan Ray., Sukharanjan Howlader., Sumit Chakraborty., Partha Pratim Chakraborty., Sujoy Ghosh. Hemichorea-Hemiballism as the First Presentation of Type 2 Diabetes. Clin Diabetes. 2015 Apr;33(2):87-9. DOI: 10.2337/diaclin.33.2.87. PMID : 25897191 ; PMCID : PMC4398017
6. . Hammami N., Nagi S. , Chaabouni M. , Boudabous S. , Drissi C. , Sebai R. , Ben Hamouda M. Hémiballisme secondaire à une hyperglycémie sans cétose. Journal of Neuroradiology (2011)38, 308-312. DOI: 10.1016/j.neurad.2011.02.001.Epub 2011 Apr 12. PMID : 21489632.
7. **.** Dixit A. Nonketotic hyperglycemia hemichorea and hemiballismus: a case report. J Med Case Rep. 2024 Mar 12;18(1):154. doi: 10.1186/s13256-023-04332-y. PMID: 38468351; PMCID: PMC10929169.

8. Cosentino C , Torres L , Nuñez Y , Suarez R , Velez M , Flores M.Hemichorea/Hemiballism Associated with Hyperglycemia: Report of 20 Cases. *Tremor Other Hyperkinet Mov.* **6**,402 (2016). https://doi.org/10.7916/d8dn454p
9. Lai PH, Tien RD, Chang MH, Teng MM, Yang CF, Pan HB, Chen C, Lirng JF, Kong KW. Chorea-ballismus with nonketotic hyperglycemia in primary diabetes mellitus. AJNR Am J Neuroradiol. 1996 Jun-Jul;17(6):1057-64. PMID: 8791916 ; PMCID: PMC8338595
10. Abe Y, Yamamoto T, Soeda T, Kumagai T, Tanno Y, Kubo J, Ishihara T, Katayama S. Diabetic striatal disease: clinical presentation, neuroimaging, and pathology. Intern Med. 2009;48(13):1135-41. doi: https://doi.org/10.2169/internalmedicine.48.1996.
11. Tung, CS., Guo, YC., Lai, CL. ,Liou LM. Irreversible striatal neuroimaging abnormalities secondary to prolonged, uncontrolled diabetes mellitus in the setting of progressive focal neurological symptoms. *Neurol Sci* **31**, 57–60 (2010). https://doi.org/10.1007/s10072-009-0127-6
12. Sawada, H.; Udaka, F.; Seriu, N.; Shindou, K.; Kameyama, M.; Tsujimura, M. MRI demonstration of cortical laminar necrosis and delayed white matter injury in anoxic encephalopathy. *Neuroradiology* **1990**, *32*, 319–321, https://doi.org/10.1007/bf00593053.
13. Ohara, S., Nakagawa, S., Tabata, K. and Hashimoto, T. (2001), Hemiballism with hyperglycemia and striatal T1-MRI hyperintensity: An autopsy report. Mov. Disord., 16: 521-525. https://doi.org/10.1002/mds.1110

Figures

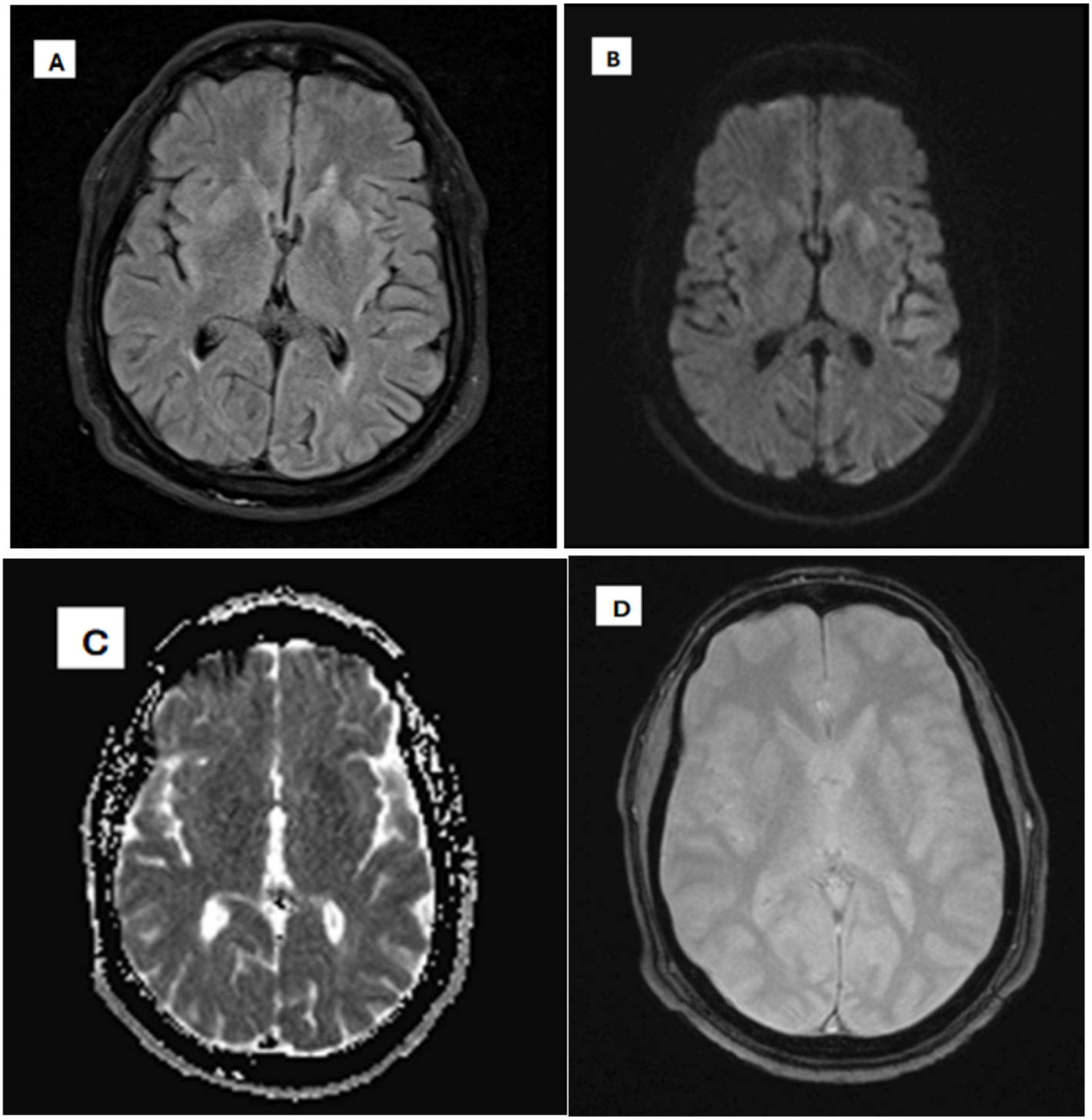


Figure 1

Initial brain MRI showing hyperintensity on FLAIR (Figure 1A) and diffusion-weighted imaging (Figure 1B) sequences in the left head of the caudate nucleus and the left insular region, associated with a corresponding low ADC signal (Figure 1C). The gradient-echo sequence showed no evidence of hemorrhage or calcification (Figure 1D).

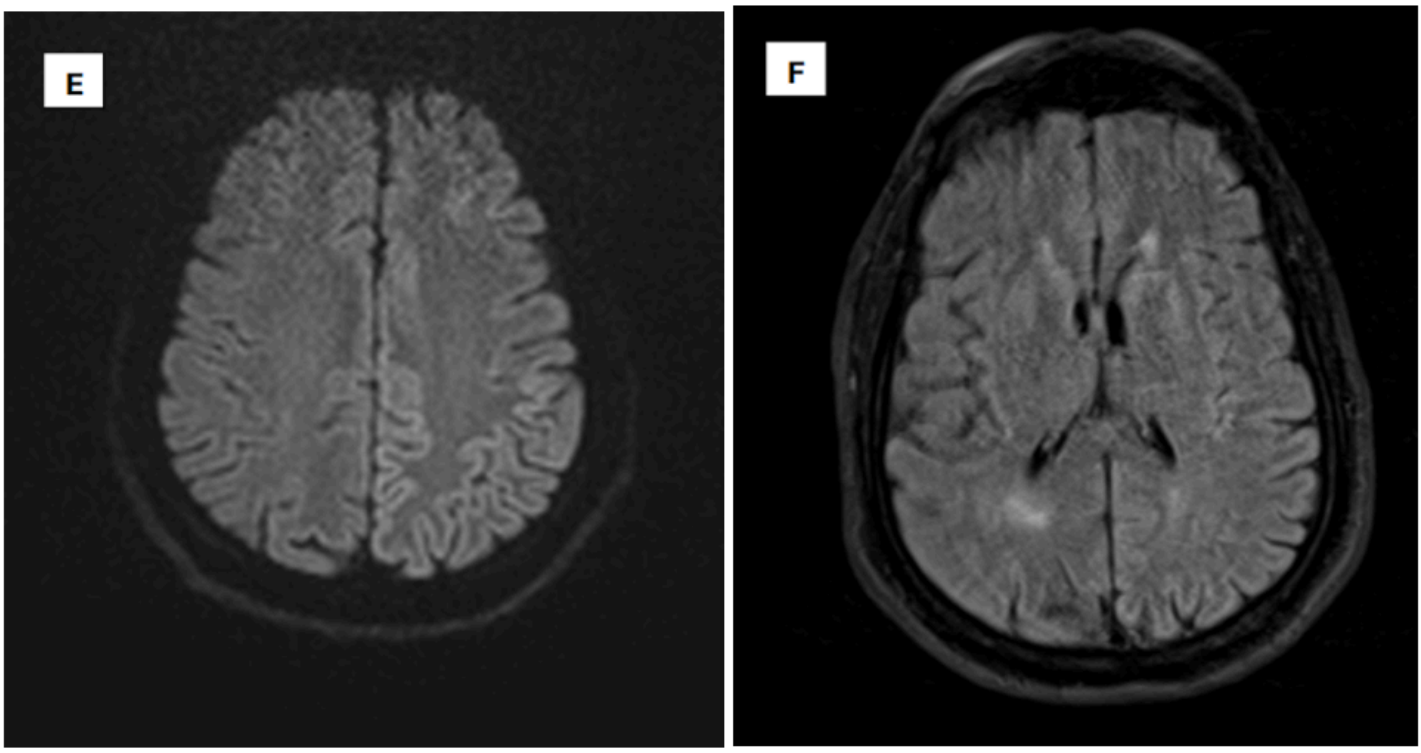


Figure 2

Follow-up brain MRI revealed spontaneous gyrate hyperintensity of the left cortical ribbon on diffusion-weighted imaging sequences (Figure 2E) and of the insular region on the FLAIR sequence (Figure 2F).

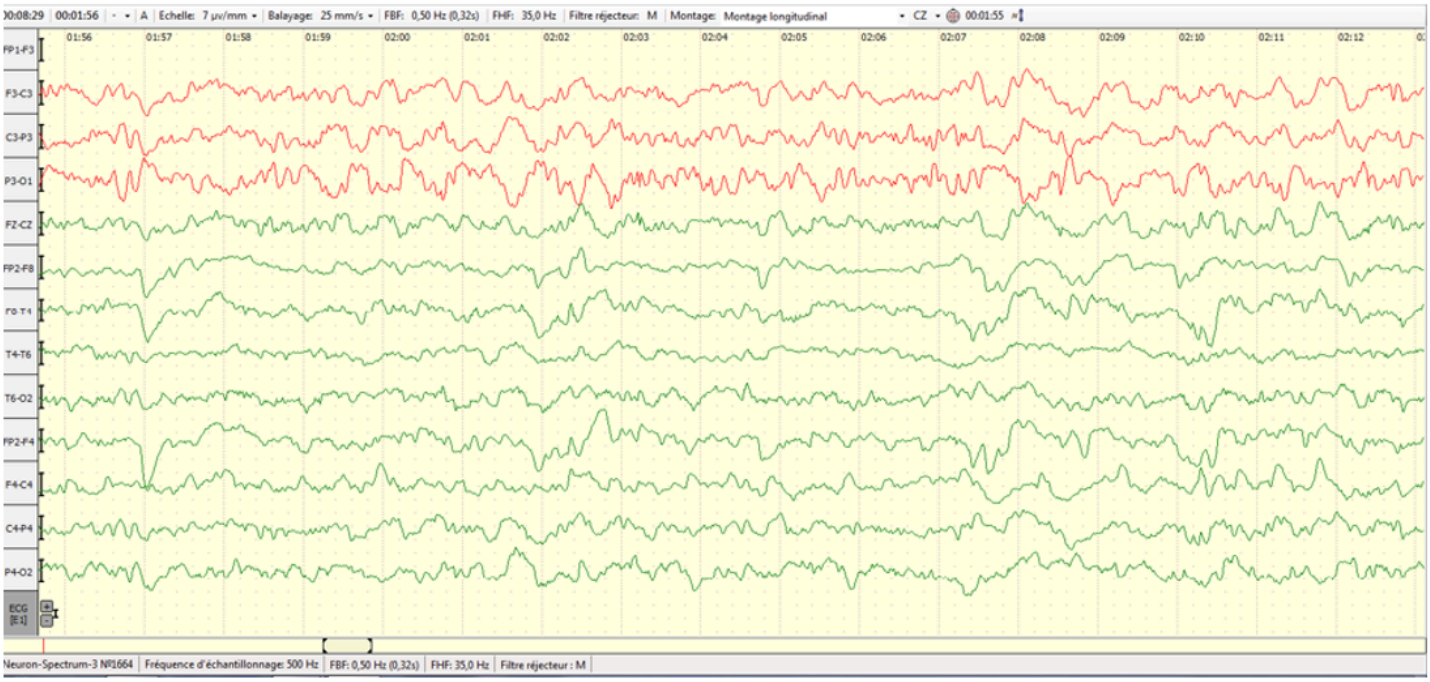


Figure 3

EEG showed global and diffuse slowing of the background activity, without epileptiform paroxysms