



Case Report

Focal epilepsy and psychosis symptoms: A case report and review of the literature

Fares Jaballah^{*}, Islem Romdhane, Jasser Nasri, Mohamed Ferhi, Nawras Bellazrag, Yosra Saidi, Jihene Mannai

Department of Psychiatry, Ibn El Jazzar University Hospital of Kairouan, Sousse University, Tunisia

ARTICLE INFO

Keywords:

Case report
Comorbidity
Diagnosis
Focal epilepsy
Psychosis
Schizophreniform

ABSTRACT

Background: Epilepsy is still a real mental health problem; although most epilepsies are curable, their psychopathological consequences are often significant and complex to manage. In this framework, the association of epilepsy with psychotic disorders has long been known.

Case presentation: To discuss the links between epilepsy and psychosis, we report the observation of a 52-year-old man, treated for complex focal epilepsy, admitted to a psychiatric department for auditory and visual hallucinations and a behavioural disorder not improved by antiepileptic treatment alone or an antipsychotic alone.

Discussion: Psychotic symptoms in epilepsy can be part of intercritical, post-critical or alternative psychoses. In our patient's case, the psychotic symptoms were post-critical and alternative. It was probably an associated schizophreniform disorder. Emotional indifference and activity restriction are rarely encountered in this setting, while rapid mood fluctuations are frequent. Delusional themes are often mystical, fueled by auditory and unusual visual hallucinations. Negative disorders are rare.

Conclusion: Epileptic psychoses have not been identified as nosographic entities in the psychiatric classification systems (DSM-V and ICD-10), which poses a problem in recognizing these disorders. Therefore, a collaboration between psychiatrists and neurologists is necessary to understand this complex comorbidity better, avoid diagnostic errors, and optimize management.

1. Introduction

Epilepsy is still a real mental health problem; although most epilepsies are curable, their psychopathological consequences are often significant and complex to manage [1]. The association of epilepsy with psychotic disorders has long been known [2,3]. These disorders have been individualized under per-critical psychosis, post-critical psychosis and alternative psychosis. The question then arises as to the relationship between these two types of disorders: is it a chance association, or is there a specific relationship between the two and what are the therapeutic implications?

Based on the analysis of an observation highlighting the clinical and therapeutic issues of the epilepsy-psychotic disorder comorbidity, a bibliographic review was conducted to study the relationship between these two entities.

This case report has been reported in line with the SCARE Criteria 2020, Agha RA, Franchi T, Sohrabi C, Mathew G, for the SCARE Group.

The SCARE 2020 Guideline: Updating Consensus Surgical CAse REport (SCARE) Guidelines, International Journal of Surgery 2020; 84:226–230 [27].

2. Case presentation

The patient is 52 years old, with a family history of generalized tonic-clonic epilepsy in a maternal cousin, without a family history of follow-up in a psychiatric setting, was referred to the psychiatric setting by an emergency physician for abnormal perceptions, sensory (auditory and visual) with hetero-aggression towards his family of sudden onset for five days. Mr. has known an episodic evolution for 22 years. He is not improved by the antiepileptic treatment with a total of 3 hospitalizations simulating brief psychotic episodes within the framework of a bad therapeutic observance and resolution lower than one month, secondary to symptoms described by the family as "focal epileptic seizures" which last 2–3 days before the onset of psychotic symptoms. He was a day

^{*} Corresponding author. Mohamed Rchid Ridha, Ksar hallal, 5070, Tunisia.
E-mail address: jaballah.fares@yahoo.fr (F. Jaballah).



Fig. 1. Brain MRI, frontal section.

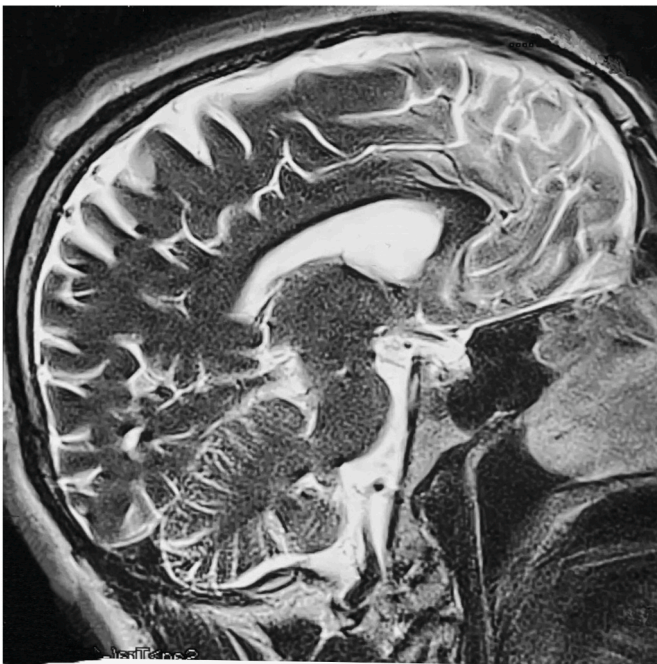


Fig. 2. Brain MRI, sagittal section.

labourer, married and living with his wife and five children. His wife described him as a calm, reticent person with a tendency to social isolation. The onset of the morbid disorders of the concerned person was at 22 years old, marked by the appearance of nocturnal crises made of sudden awakenings accompanied by dystonia at the level of the left lower and upper limb objectified in our service with video surveillance (video 1-2), sometimes secondarily generalized with the notion of

urinary leakage and most critical amnesia. These seizures were of brief duration, repeated in a stereotyped way and followed by partial amnesia of the facts. Initially, these nocturnal events were well tolerated by the patient's family and attributed to a transient sleep disorder due to stress. Secondly, a few days after the clinical aggravation and the number of attacks (2–3 attacks per week), there was an appearance of visual perceptions only at night, in the form of zoopsies, which evolved in a paroxysmal way, preceded by multiple somatic complaints, especially headaches, nausea and a sensation of epigastralgia. Since then, the patient has become very anxious; he complains of instinctual disorders: chopped sleep and memory and concentration disorders. A feeling of derealization and anticipatory anxiety invaded these nights, which distracted him most of the time and affected his socio-professional functioning. A brain magnetic resonance imaging (MRI) showed a slight dilatation of the Virchow Robin lenticulostriate type 3 spaces with a punctiform T2 hyper signal of the peduncles (see Figs. 1 and 2). A right frontal subcortical millimetric T2 hyper signal was retained by dilating the peripheral Virchow Robin spaces. On electroencephalogram (EEG): a low-voltage and short trace that could be correlated to the drug used in the hospital. The diagnosis of focal epilepsy could not explain the whole clinical picture; the hypothesis of a comorbid psychotic disorder, in this case, a schizophreniform psychosis, was likely. This justified his being put on an antipsychotic. After discussion and collaboration with our colleagues in the department of neurology, the patient was then put on carbamazepine at a dose of 600 mg/day (he had an intolerance to sodium valproate in a previous hospitalization confirmed biologically by hepatic cytolysis) in combination with amisulpride 400mg/day and a short half-life benzodiazepine: only one dose/night: diazepam 5mg.

Supplementary video related to this article can be found at <https://doi.org/10.1016/j.amsu.2022.104862>

The evolution under treatment was marked by a clear decrease in the frequency of nocturnal attacks. However, the auditory perceptions persisted for a few days after the initiation of treatment and the decrease of the nocturnal crises. The picture was enriched by other elements, including mental automatism, delirium with themes of influence and mystic-religious and terrifying visual hallucinations during the first two days of treatment. For the disorders described above, the patient was hospitalized in our department for 25 days and discharged after the disappearance of the psychotic disorders and epileptic seizures. The evolution was quite good, with attenuation of the abnormal perceptions and distancing of the delusional convictions. After the resolution of the psychotic symptoms, the patient was essentially cared by the neurology department, with a cessation of antipsychotic treatment and spaced visits, each three months.

3. Discussion

In this patient, the epileptic seizures, which had occurred at the age of 22 years, reappeared at the age of 41 years, 42 years and 50 years, respectively, requiring three hospitalizations in the context of poor compliance with his antiepileptic treatment with the notion of a free interval between seizures. The clinical presentation was psychomotor agitation, heteroaggression and insomnia. The nocturnal seizures of paroxysmal awakenings and dystonia correspond a priori to frontal/temporal nocturnal epilepsy. In this type of epilepsy, often unrecognized by the patient and his family, the seizures occur exclusively during sleep and sometimes pose a problem of differential diagnosis with paroxysmal motor manifestations related to a sleep disorder [4]. Whereas the auditory and neurovegetative symptoms with the experiential phenomena of the beginning, evolving in a paroxysmal way, point to temporal epilepsy.

The overall diagnosis was complex frontotemporal epilepsy since there was a loss of contact and total amnesia during the post-critical episode. This diagnosis was retained based on various anamnestic and clinical arguments. However, apart from these stereotyped epileptic seizures, the subsequent enrichment of the symptomatology by sensory

hallucinations (auditory and visual) concomitant with the seizures and recognized as pathological by our patient, the delusions with mystical-religious themes and influence and the mental automatism, without disturbance of consciousness, evoked a psychosis associated with epilepsy. The challenge is to recognize this comorbidity despite the polymorphism of this frontotemporal partial epilepsy and the atypicality of the psychotic disorders developed by our patient during her illness [5]. Indeed, the visual hallucinations of zoopsia type and the delirium presented constitute one of the originalities of our observation and are not usual in focal seizures and should draw the attention of neuropsychiatrists toward an associated psychotic disorder. The amnesia of behavioural disorder and hetero aggressiveness, another originality of the observation, is rare in epileptic psychosis, contrary to the mystical delirium. These clinical data refer to a schizophreniform disorder; however, can we speak of a schizophreniform disorder in the epileptic subject and what are, in this case, its specificities?

The literature suggests that epileptic patients have a risk of developing a psychosis 6 to 12 times higher than the general population [6,7], and this risk is higher in the case of temporal epilepsy [8]. Does the question remain as to what type of epileptic psychosis is involved in our case? An attempt to classify epileptic psychoses, taking into account the duration of the psychotic episode and the chronology of its appearance with the epileptic seizures, as well as the level of consciousness [9], distinguishes three types [3]:

- Episodic psychoses where consciousness is often altered (acute psychosis, post-critical psychosis).
- Alternative psychoses, where the level of consciousness is variable.
- Permanent or chronic psychoses where consciousness is preserved (schizophreniform inter-critical psychoses).

1. Critical and peri-critical psychoses

During some partial epileptic seizures, psychotic-like manifestations such as dreaming or hallucinations may occur. Moreover, the patient we have presented also experienced critical disorders at the beginning of the evolution of his epilepsy, made of auditory and visual hallucinations of the zoopsia type. These manifestations are easily linked to epilepsy because of their short duration, stereotyped character and especially their different experience by the patients [3]. Post-critical confusion does not usually pose a diagnostic problem, given its brevity and obvious link to epileptic Seizure. In contrast, status epilepticus is prolonged in time and could thus wrongly suggest a psychiatric condition. However, the confusion is characterized by maladaptive defence reactions, oppositionism, agitation and anxiety.

The spontaneously favourable evolution, within a few hours, and the history of epilepsy will allow to re-establish the diagnosis a posteriori. The EEG is always the main diagnostic tool since it is constantly abnormal: it shows either spike activity testifying to the critical nature of the episode or slow anomalies of variable distribution and amplitude pointing to a prolonged post-critical state [10]. Critical or peri-critical psychoses are usually brief and limited to the Seizure's duration or status epilepticus. Nevertheless, their clinical presentation in the form of authentic confusional states within the framework of the Seizure renders the qualification "psychotic" of these episodes as a kind of semantic slip highly questionable [1]. Post-critical psychosis is an entity of fairly recent individualization in the literature [11]. A precise chronology characterizes it: a seizure or a burst of seizures, usual post-critical confusion, return to normal consciousness, an interval of complete lucidity, and then the sudden appearance of a brief psychotic disorder evolving in full consciousness or with discreetly altered consciousness [12]. It appears in patients with refractory focal epilepsy, evolving for at least ten years [10]. It often consists of complex focal temporal seizures and secondarily generalized seizures [11,13] with frequently inter-critical bitemporal abnormalities on the EEG [14]. The psychotic disorder occurs after a series of closely spaced seizures, typically in the form of a burst of seizures.

The onset is abrupt, after a free interval of 24 hours to 1 week after the last Seizure. The psychiatric picture is characterized by an acute delirium associated in variable proportions: mood disorders, often in the foreground, auditory hallucinations, perceptual disorders, delusions of religious themes or grandeur, and delusions of persecution [13]. The critical EEG is normal, and the intercritical EEG shows the frequency of independent bitemporal foci [1]. Affected patients have a more frequent psychiatric history when compared to matched non-psychotic epileptics [12]. The course is self-limiting after a maximum of one week [14], with amnesia of the episode and the possibility of subsequent recurrences of identical episodes.

In some severe cases, psychotropic treatment is essential to manage behavioural problems.

2. Alternative psychosis:

Contrary to post-critical psychosis, which follows a period of activity of the epileptic disease, alternative psychosis appears when the seizures disappear, suggesting a swinging phenomenon [3]. The first description of this phenomenon was made by Landolt, who, as early as 1953, noted a correlation between the evolution of the psychotic process and the changes in the EEG: the focus of paroxysmal anomalies that were active before and after the psychotic episode disappeared during this episode [15]. He called this normalization of the EEG "forced normalization of the EEG"; a concept characterized by the fact that, with the onset of the psychotic state, the EEG improves or even becomes completely normal compared to the previous tracings. This state, which may last several weeks, can be triggered by the initiation of antiepileptic treatment and interrupted by seismotherapy.

According to a review of the literature, Tellenbach [16] associated this concept of "forced normalization", which is only electroencephalographic, with the term "alternative/alternating psychosis" to insist on the disappearance of seizures during a psychotic episode, whatever the EEG tracing. This type of psychosis is rare. Generally, the episodes are brief, but sometimes they can last for several weeks. There is often a common prodromal phase with anxiety, hypochondriacal complaints and insomnia. Then, there is delirium in the state phase with an essentially imaginative mechanism and a slightly altered consciousness. Hysterical, depressive, manic, dysphoric symptoms and hypochondriacal concerns may complete the clinical picture. Alternative psychosis can be encountered in both focal and generalized epilepsies, but the simultaneous presence of tonic-clonic and nonconvulsive seizures accompanied by altered consciousness (either absences or complex partial seizures) seems necessary [17].

3. Schizophreniform intercritical psychoses:

Chronic inter-critical psychoses have an evocative clinical presentation in epileptic patients, such as schizophrenia [18]. This entity gave rise to the term schizocomial psychosis [19] to describe this type of psychosis, whereas, in the Anglo-Saxon literature, the reported cases have been referred to as "schizophrenia-like psychoses of epilepsy" [20–22]. Clinically, different forms can be observed. A paranoid form [3] can be observed, where patients with sensitive or suspicious personality traits will develop auditory hallucinations with an often interpretative theme. However, one cannot speak of paranoid delusions because of a lack of systematization. A paranoid form can also be observed [3], which is considered a true schizophrenic psychosis of epilepsy.

On the semiological level, some nuances have been noted [12]: in epileptic psychoses, an affective indifference and a restriction of activities are rarely encountered, whereas rapid fluctuations of mood are frequent. Delusional themes are often mystical, fueled by auditory and unusual visual hallucinations. Deficit disorders are rare. Epilepsy begins before age ten and lasts about 14 years between the onset of epilepsy and psychosis [12]. There is no premorbid schizoid personality or family

history of schizophrenia. The existence of temporal epilepsy is considered the main risk factor, independently of the intrinsic severity of epilepsy measured by seizure frequency. Finally, the evolution of epileptic psychoses seems less deficient than endogenous schizophrenia [23].

We can thus bring the case of our patient under study closer to this nosographic entity: schizophreniform psychosis. Indeed, after years of evolution of his frontotemporal epilepsy, he presented a paranoid delusion with mystical and influential themes and a hallucinatory mechanism (auditory, visual and cenesthetic hallucinations).

The absence of a family history of schizophrenia, alteration of the course of his thought, and important repercussions on his social and school functioning corroborate this diagnostic hypothesis. It remains to be noted that the dilation of the Virchow Robin spaces objectified on MRI, and according to a recent review of the literature [24–26], does not have a clinical impact, nor does it support the origin of epilepsy.

4. Conclusion

The clinical observation presented and the review of the literature illustrate the variety of psychotic disorders that can be associated with epilepsy during its course and their specificities. This comorbidity implies the existence of a causal relationship between epilepsy and psychosis, the explanation of which remains hypothetical. Epileptic psychoses, although they present clinical and evolutionary specificities, have not been identified as nosographic entities in the psychiatric classification systems (DSM-V and ICD-10), which poses a problem in recognizing these disorders.

Therefore, at the end of our work, to be able to help in the management of such cases in the future, a collaboration between psychiatrists and neurologists becomes necessary for a precise description of the psychotic disorder, on the one hand, and of the epilepsy, on the other hand, in order to better understand this complex comorbidity, to avoid diagnostic errors and to optimize the management and especially to ensure a good follow-up for the patients.

Further research on this subject seems to be necessary to better understand this comorbidity and to allow an adequate and appropriate comparison with other researches.

Data availability statement

All data generated or analyzed during this study are included in this article. Further enquiries can be directed to the corresponding author.

Provenance and peer review

Not commissioned, externally peer reviewed.

Ethical approval

I APPROVE that there was necessary to get an ethical approval.

Patient consent: the patient has given consent for possible publication of this case report.

Sources of funding

I CONFIRM that there isn't any sources of funding for my research.

Author contribution

OUR STUDY was a team-work. All authors contributed on data collection, data interpretation and writing the article.

Trail number registry

1. Name of the registry:
2. Unique Identifying number or registration ID:

3. Hyperlink to your specific registration (must be publicly accessible and will be checked):

Guarantor

I CONFIRM FULL RESPONSIBILITY.

Consent

I CONFIRM.

Declaration of competing interest

I CONFIRM that there isn't any conflicts of interest.

References

- [1] N. Adachi, et al., Psychoses and epilepsy: are interictal and postictal psychoses distinct clinical entities? *Epilepsia* 43 (12) (déc. 2002) 1574–1582, <https://doi.org/10.1046/j.1528-1157.2002.22402.x>.
- [2] E. Lu, N. Pyatka, C. J. Burant, et M. Sajatovic, « Systematic literature review of psychiatric comorbidities in adults with epilepsy », *J. Clin. Neurol.* Seoul Korea, vol. 17, n° 2, p. 176–186, avr. 2021, doi: 10.3988/jcn.2021.17.2.176.
- [3] B. de Toffol, Les psychoses postictales, *Rev. Neurol.* (Paris) 165 (10) (oct. 2009) 769–773, <https://doi.org/10.1016/j.neurol.2009.07.007>.
- [4] V. Stal, Nuits mouvementées et bruyantes : comment différencier parasomnies et crises d'épilepsie frontale nocturnes ? Intérêt de l'enregistrement vidéo-EEG-polysomnographique, *Médecine Sommeil* 6 (2) (2009) 65–73, <https://doi.org/10.1016/j.msom.2009.05.001>, avr.
- [5] Interventions for psychotic symptoms occurring with epilepsy (consulté le 21 juillet 2022), https://www.cochrane.org/CD006118/EPILEPSY_interventions-psychotic-symptoms-occurring-epilepsy.
- [6] R. Torta, et al.R. Keller, Behavioral, psychotic, and anxiety disorders in epilepsy: etiology, clinical features, and therapeutic implications, *Epilepsia* 40 (s10) (1999) s2–s20, <https://doi.org/10.1111/j.1528-1157.1999.tb00883.x>.
- [7] P. Sachdev, Schizophrenia-like psychosis and epilepsy: the status of the association, *Am. J. Psychiatr.* 155 (3) (mars 1998) 325–336, <https://doi.org/10.1176/ajp.155.3.325>.
- [8] L. Altshuler, R. Rausch, S. Delrahim, J. Kay, P. Crandall, Temporal lobe epilepsy, temporal lobectomy, and major depression, et, *J. Neuropsychiatry Clin. Neurosci.* 11 (4) (nov. 1999) 436–443, <https://doi.org/10.1176/jnp.11.4.436>.
- [9] N. Adachi, et al., Psychoses and epilepsy: are interictal and postictal psychoses distinct clinical entities? *Epilepsia* 43 (12) (2002) 1574–1582, <https://doi.org/10.1046/j.1528-1157.2002.22402.x>.
- [10] B. De Toffol, Les troubles psychiatriques dans les épilepsies partielles pharmacorésistantes, *Epilepsies* 17 (2005) 17–23.
- [11] S.J. Logsdail, et al.B.K. Toone, Post-ictal psychoses: a clinical and phenomenological description, *Br. J. Psychiatry* 152 (2) (févr. 1988) 246–252, <https://doi.org/10.1192/bjp.152.2.246>.
- [12] B. de Toffol, P. Corcia, J. Praline, K. Mondon, L'ère de la prise en charge globale et de la qualité de vie Impact et enjeux des comorbidités psychiatriques dans les épilepsies, et, *Epilepsies* 19 (4) (oct. 2007) 231–241.
- [13] K. Kanemoto, J. Kawasaki, I. Kawai, Postictal psychosis: a comparison with acute interictal and chronic psychoses, et, *Epilepsia* 37 (6) (1996) 551–556, <https://doi.org/10.1111/j.1528-1157.1996.tb00608.x>.
- [14] O. Devinsky, et al., Postictal psychosis: a case control series of 20 patients and 150 controls, *Epilepsy Res.* 20 (3) (1995) 247–253, [https://doi.org/10.1016/0920-1211\(94\)00085-B](https://doi.org/10.1016/0920-1211(94)00085-B), mars.
- [15] L. Hans, « Serial electroencephalographic investigations during psychosis episodes in epileptic patients and during schizophrenic attacks, in: *Forced Norm. Altern. Psychoses Epilepsy Wrightson Biomed, Publ. Ltd, 1998, pp. 25–48. First published in 1958 (re-edited).*
- [16] H. Tellenbach, Epilepsy as a convulsive disorder and as a psychosis. On alternative psychoses of paranoid nature in "forced normalization"(Landolt) of the electroencephalogram of epileptics, *Nervenarzt* 36 (1965) 190–202.
- [17] P. Wolf, et al.M.R. Trimble, Biological antagonism and epileptic psychosis, *Br. J. Psychiatry* 146 (3) (1985) 272–276.
- [18] B.K. Toone, E. Garralda, M. Ron, The psychoses of epilepsy and the functional psychoses: a clinical and phenomenological comparison, et, *Br. J. Psychiatry* 141 (3) (sept. 1982) 256–261, <https://doi.org/10.1192/bjp.141.3.256>.
- [19] H. Ey, « *Traite des hallucinations: I-II* », 1973.
- [20] M.M. Perez, et al.M.R. Trimble, Epileptic psychosis—diagnostic comparison with process schizophrenia, *Br. J. Psychiatry* 137 (3) (sept. 1980) 245–249, <https://doi.org/10.1192/bjp.137.3.245>.
- [21] The psychoses of epilepsy | journal of neurology, neurosurgery & psychiatry (consulté le 5 avril 2022), <https://jnnp.bmj.com/content/69/1/1.short>.
- [22] S. Benzahra, « SCHIZOPHRENIE ET EPILEPSIE : LIENS CLINIQUES ET PSYCHOPATHOLOGIQUES », 2021. Consulté le: 12 avril 2022. [En ligne]. Disponible sur: <http://ao.um5.ac.ma/xmlui/handle/123456789/19071>.

- [23] S. Qureshi, J. Siddiqui, F. Ayari, Interictal psychosis- a case of psychosis of epilepsy, *et, Psychiatry Behav. Sci.* 10 (3) (2020) 161, <https://doi.org/10.5455/PBS.20200117070405>.
- [24] S. Gronier, et al., Dilatations géantes des espaces de Virchow-Robin symptomatiques, *Rev. Neurol. (Paris)* 169 (11) (nov. 2013) 898–902, <https://doi.org/10.1016/j.neurol.2013.05.003>.
- [25] R.E. Feldman, et al., Quantification of perivascular spaces at 7T: a potential MRI biomarker for epilepsy, *Seizure* 54 (janv. 2018) 11–18, <https://doi.org/10.1016/j.seizure.2017.11.004>.
- [26] C. Liu, et al., Quantification of visible Virchow-Robin spaces for detecting the functional status of the glymphatic system in children with newly diagnosed idiopathic generalized epilepsy, *Seizure* 78 (mai 2020) 12–17, <https://doi.org/10.1016/j.seizure.2020.02.015>.
- [27] R.A. Agha, T. Franchi, C. Sohrabi, G. Mathew, for the SCARE Group, The SCARE 2020 guideline: updating Consensus surgical CAse REport (SCARE) Guidelines, *Int. J. Surg.* 84 (2020) 226–230.