Author's response to reviews

Title: An uncommon case of random fire-setting behavior associated with Todd paralysis

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Author's response to reviews: see over
Dear Dr. Lon S Schneider

MS: 4523864586157599
An uncommon case of random fire-setting behavior associated with Todd paralysis. Masayuki Kanehisa, Katsuhiko Morinaga, Yoshihiro Maruyama, Taiga Ninomiya, Yoshinobu Ishitobi, Yoshihiro Tanaka, Jusen Tsuru, Hiroaki Hanada and Jotaro Akiyoshi

We are most grateful to you and to the reviewers for the helpful comments on the original version of our manuscript. We have taken all these comments into account and now submit, herewith, a revised version of our paper.

We have addressed all the comments by Reviewers 1 & 2, as indicated below, and we hope that the explanations and revisions of our work are satisfactory.

Reviewer 1
This article describes brain perfusion SPECT images in a young man with a sudden onset of a bizarre and random fire-setting behavior. Although this case report seems to be very interesting, there are several following issues;

1. Background) Unusual abbreviation of PP (postictal paresis) had better not be used.

We removed PP as an abbreviation for postictal paresis.

2. Neuroimaging data, 2.4) Fig.1 did not show MRI findings.

We have added this information to Fig. 1.

3. Neuroimaging data, 2.4) Ref. 8 is not related to eZIS.
We deleted the reference:

4. Neuroimaging data, 2.4) What tracer was used for brain perfusion SPECT? Authors should clarify.

We added the following information: “…..using an ethyl-cysteinate-dimer labeled with technetium- 99m (99mTc-ECD) (FUJIFILM RI Pharma Co., Ltd. Tokyo) as a radiotracer…..”.

5. Neuroimaging data, 2.4) Original SPECT images in ictal and interictal phase should be demonstrated.

We now include original SPECT images for ictal and interictal phases (Figure 3).

We added the following information to the figure legends: “Original Tc-99m ECD-SPECT images; (A) ictal SPECT; (B) interictal SPECT.”

6. Neuroimaging data, 2.4) Details of previously established normal database at author’s institute were not described. Please cite previous reference.


7. Neuroimaging data, 2.4) eZIS results are shown in Fig.2 not in Fig.1.

We changed the phrase “eZIS revealed hyperperfusion in frontal cortex (Fig. 1).” to “SISCOM revealed hyperperfusion in the frontal cortex (Fig. 3, 4, 5).”

8. Neuroimaging data, 2.4) Details of the way to acquire ictal perfusion SPECT in this patient have to be described. When was the tracer injected after the onset of ictus?
We injected the tracer 130 seconds after the onset of ictus.

We added the sentence “We injected the tracer 130 seconds after the onset of ictus”.

9. Neuroimaging data, 2.4) Authors described that eZIS revealed hyperperfusion in frontal cortex. However authors described hypoperfusion in frontal cortex in this patient. Authors may be mixed up in the evaluation of eZIS results. Again authors should demonstrate original SPECT images along with eZIS results.

We deleted the sentences “However, lesions in the hypoperfused regions within the frontal cortex and basal ganglia might result in a disconnection and deactivation of the frontal cortex. Consistent with this concept are more recent observations linking cognitive inflexibility with prefrontal depletion of serotonin (Clarke et al., 2004). Moreover, and in agreement with our findings, neuropsychological deficits (including memory impairment) have been associated with hypoperfusion within the frontal cortex and basal ganglia (Chukwudelunzu et al., 2001; Tatemichi et al., 1992)”.

We have included original SPECT images during ictal and interictal phases (Figure 3).

10. Fig.2, Fig.3 eZIS results) Lower and upper limit of Z-score had better being set to 2 to 6 with extent threshold of 300 pixels in Fig.2. Similarly Lower and upper limit of Z-score had better being set to 1 or 2 to 6 in Fig.3.

We set the lower and upper Z-score limit and 2 to 6 (respectively) with an extent threshold of 300 pixels in Fig. 4 and Fig. 5.


We have now used SISCOM instead of eZIS software.

We added the reference:

12. Conclusion) Authors had better refer to SPECT findings.

We added the following sentence in the conclusion: “In conclusion, SISCOM from ictal and interictal Tc-99m ECD-SPECT showed hyperfusion and ictal minus interictal Tc-99m ECD-SPECT showed specific hyperfusion in the frontal cortex”.

Reviewer 2

Reviewer's report:

1. Minor issues not for publication:

Re Discussion: The phrase “guilty by reason of sanity” better reads as: he was found guilty, having been found to have no mental illness or defect. Further they might just briefly mention that their diagnosis had implied that their patient lacked the capacity to form mens rea [21] which “negated criminal responsibility”.

(The verdict “guilty” does not necessarily negate their diagnosis.)

We changed the sentence “The case was closed with a verdict of guilty by reason of legal sanity” to “He was found guilty, having been found to have no mental illness or defect….”.

2. Major Compulsory Revisions:

a) The Conclusion would gain if the authors would detail their suggested “disconnection of frontal lobe structures (which specifically? “as a possible pathogenic mechanism”.

We added the conclusion “The present case suggests a disconnection of frontal lobe structures as a possible pathogenic mechanism.”

b) It would be important to mention epilepsy and to specify what type of epilepsy, the authors seem to mean a nonconvulsive behavioral type.

We added the sentence “We diagnosed this patient as suffering from complex partial
seizures”.

c) further, it would help to elaborate on their finding that Todd paralysis had been pre-existing in their case, since their Discussion suggests “an association between an isolated, first-time arson offense and Todd’s paralysis after a seizure”. (Did the patient have other seizures too without offensive behavior, possibly with other bizarre non-criminal behavior?

We added the sentence “Here we describe a case of a first-time arsonist who suffered Todd’s paralysis prior to the onset of a bizarre and random fire-setting behavior. The patient did not appear to have seizure without offensive behavior.”

3a. Further, Re. Discussion (see also lc) the authors might add an explanation what suggested to them the reference “temporal and frontal lobe dysfunction in violent offenders …more pronounced in the dominant hemisphere [16]”. So far they had mentioned “hypoperfused regions within the frontal cortex and basal ganglia”. It might also help to specify prefrontal cortex vs. motor cortex within this context.

We changed the sentence “Neuropsychological and neuropsychiatric findings suggest temporal and frontal lobe dysfunctions in violent offenders and these dysfunctions appear to be more pronounced in the dominant hemisphere (Volavka et al., 1992)” to “Neuropsychological and neuropsychiatric findings suggest frontal lobe dysfunction in violent offenders and these dysfunctions appear to be more pronounced in the dominant hemisphere (Volavka et al., 1992).”

We changed the sentence “eZIS revealed hypoperfusion in cingulate cortex, basal ganglia and hyperperfusion in frontal cortex” to “SISCOM revealed hyperperfusion in the frontal cortex”.

b) Also re “Disconnection of the frontal lobe….. “ Here the authors list impairments of prefrontal lobe functions alone. It would help if they could here specify which ones of those functions are “disconnected “.

We changed the sentence “Here we argue that, in our case, disconnection of the frontal lobe resulted in impairments of cognition, primarily executive functions involving planning and judgment, as well as partial loss of memory and impulse control
functions” to “Here we argue an impairment of the prefrontal cortex in our case. The disconnection of the frontal lobe resulted in impairments in cognition, primarily executive functions involving planning and judgment, as well as partial loss of memory and impulse control functions.”

c) “We recommended continuing prophylactic treatment with antiepileptic medication to help prevent further hypoxic-ischemic insults.”. It would help if the authors could elaborate on their implication that suggests to the reviewer that a previous hypoxic-ischemic insult had occurred which had somehow been association with seizures. (Actually the authors stated previously that antiepileptic medication had made the patient symptom free, which would speak for a diagnosis of seizures).

We added the reference:

4. Medical History 2.2. Mental status examination
a) It would much help if the authors would not only briefly cite AXIS I – V of DSM IV-TR but could add a more details. For example, what was the patient’s general motoric behavior, speech and affect? How dif he experience his acts and feel after the fire setting? What did fire mean to him, any memories of fires, etc?

We added the sentences: “His general motor behavior did not show a gait disturbance. His speech was smooth and his affect was euthymic. He was indifferent to his actions. After the fire setting he felt foggy.”

b) Further, it would be helpful to briefly differentiate the use of the term “impulsive”, given that this term applies not only to the common use of it as contained in the diagnostic category of impulsivity disorders” which are associated with strong emotions and typically exist virtually life long behavior. By contrast, the authors appear to imply “impulsive behavior” in the sense of non-intentional sudden behavior without emotional involvement(!) as is typical of any kind of seizure, which the authors apparently mean.

We changed the term “impulsive” to “impulsive behavior”.
b) It would be helpful if the authors could explain under AXIS III why they mention “history of Todd’s paralysis, which raises the possibility of a delirious state as a possible cause for the observed fire setting behavior [4].” This Ref. [4] re. hyperglycaemic delirium. Had the latter occurred in patient’s history? or do the authors mean this reference as a background differential diagnosis?

We cited the Ref. [4] as a background differential diagnosis.

c) The authors mention “observed fire-setting behavior”. This implies that there had been witnesses observing the fires-setting. It would be of great interest if there had been reports by such witnesses.

We did not mean to imply that somebody witnessed the fire-setting. This sentence meant that the patients reported the fire-setting.

We deleted the phrase “observed fire-setting behavior”.

5. To Case Report 2.5 Neurological examination

a) It would help if the authors could clarify whether the patient had a seizure during a neurological examination

The patient did not have a seizure during a neurological examination.

We changed the sentence “Bilateral postictal paresis of the bilateral legs (motor deficits without sensory deficits) lasted approximately 5 minutes” to “Bilateral postictal paresis of the bilateral legs (motor deficits without sensory deficits) lasted approximately 5 minutes without seizure.”

b) to 2.6 “Other diagnostic procedures” (obviously not during the fire-setting)

The authors might specify whether they assigned any significance to the “ictal discharge of repetitive spikes localized in the frontal, central and parietal regions Fig 2A)”.

In regard to the parietal region it might enhance their presentation within the context of their report that the patient had at times disturbance of consciousness)

We added the sentence “The patient had at times a disturbance of consciousness. Recent literature has emphasized the role of the parietal lobe in consciousness (Custers and Aarts, 2010; Haggard, 2008)”.

We added the references:


6. Background
a) “Here we describe a case of…first-time arsonist who suffered Todd’s paralysis prior to the onset of a bizarre and random fire-setting behavior”. It would be helpful if the authors might be more specific in their expression, e.g. by stating that Dodd’s paralysis had been pre-existing in their case. - Not also 2c) reDiscussion, where the authors suggested “an association between an isolated first-time arson offence and Todd’s paralysis…after a seizure”. This needs clarification, probably within the context of the pre-existing Todd paralysis. Such a clarification also pertains to Summary.

We changed the sentence “In contrast, our case suggests an association between an isolated, first-time arson offence and Todd’s paralysis” to “Here we describe a case of a first-time arsonist who suffered Todd’s paralysis prior to the onset of a bizarre and random fire-setting behavior.”

b) To their list of previous studies of fire-setting cases the authors might chose to add the proposed partial epilepsy of ‘limbic psychotic trigger reaction” (LPTR). LPTR is two-fold consistent with established experimental knowledge: LPTR symptomatology is analogous to that elicited in primates by the neuro-
physiological mechanism of seizure kindling. Further, LPTR is also “intriguingly similar” to seizures evoked by direct electrical stimulation of the mesotemperobasal brain region in pre-surgery patients (Wieser HG, 1983; 1993). LPTR’s so far 24 published cases include four fire-setters whose acts had been unplanned, non-intentional, and remembered afterwards (Pontius AA & Wieser HG. Can memories kindle nonconvulsive behavioral seizures in ‘limbic psychotic trigger’ reaction’? Epilepsy & Behavior 2004, 5:775-783; Pontius AA Motiveless fire-setting implication partial seizure kindling by reviving memories of fires in ‘limbic psychotic trigger reaction’. Perceptual & Motor Skills 1999, 88::970-982).

We added the sentences “Finally, we would also like to mention the phenomenon of ‘limbic psychotic trigger reaction’ (LPTR). LPTR symptomatology is analogous to that elicited in primates by the neuro-physiological mechanism of seizure kindling. Further, LPTR is also “intriguingly similar” to seizures evoked by direct electrical stimulation of the mesotemperobasal brain region in pre-surgery patients (Wieser, 1983; Wieser et al., 1993). LPTR’s 24 published cases include four fire-setters whose acts had been unplanned, non-intentional, and remembered afterwards (Pontius, 1999; Pontius & Wieser, 2004.).”

We added the references:


7. Summary
The same advice pertains to summary as that mentioned under Background 6a
and under Conclusion 2c).

We added the following sentences to the summary “We describe a case of a first-time arsonist who suffered Todd’s paralysis prior to the onset of a bizarre and random fire-setting behavior. The patient did not appear to have seizure without offensive behavior”.

Sincerely,

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Thank you for your attention to our paper.
Sincerely yours,
Jotaro Akiyoshi, MD, PhD