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Case Report

Diabetic Hemichorea at the Euglycemic State:

A Case Report and an Earlier Unique Imaging before Hemichorea Symptoms Appeared

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Abstract

Hemichorea-hemiballism (HCHB) is an infrequent but specific complication of diabetes mellitus. It usually occurs in the Asian elderly female who experienced high blood glucose. However, it needs to be cautious to diagnose because it can be also found in the male/female whose blood glucose is under good control. Here, we reported a man, who went through this situation where his serum glucose was normal (1-4). Although its exact pathogenesis is unknown, many articles took for the fluctuation of serum glucose (5). In addition, we observed that his head computed tomography (CT) scan presented relevant changes before his symptoms occurred. Hence, our findings suggest that HCBC may potentially be prediagnosed through patients' computed tomography.

Key words: Hemichorea, Diabetes mellitus, Hyperglycemia, Involuntary movement, Normal glucose level

Introduction

Diabetic Hemichorea is an unusual disease which always happens when the blood glucose is under high level. Many cases have reported that the

involuntary movement of unilateral limbs are the common and characteristic symptoms of Hemichorea-hemiballism. According to the mainstream opinions, the abnormal movement was caused by the high glucose level (5-6). However, these days, there appeared some special

cases with hemichorea while the patients' blood glucose was under normal level. This phenomenon has raised attentions from many physicians' especially neurologists. Herein, we report a rare case about diabetic hemichorea at normal blood glucose. A patient was admitted twice by our hospital. At the first time, he experienced an acute stroke with high plasma glucose, and at the second time, he suffered from an unwilling movement with his right extremities with normal plasma glucose. At the same time, we also present some special findings with his head CT which could be a possible evidence to prediagnose the Diabetic Hemichorea.

Case report

A 55-years-old Asian man who had been diagnosed type 2 diabetes for 2 decades, experienced an involuntary movement of his right upper and lower extremities for 6 days. His bad glucose level had damaged his retinas and kidneys. In addition, he also had hypertension and smoking history for 30 years. Before this admission, he had been admitted to hospital due to an acute ischemic stroke and awful glucose levels (plasma glucose 404.1 mg/dL, HBA1C 19.4%) 3 weeks ago. However, his plasma glucose level had been controlled well when he was discharged from hospital. At this time, his blood tests showed plasma glucose 111.96 mg/dL, and HBA1C 12%. Neurophysical examinations didn't show any other positive physical signs except unconscious dyskinesia of his right limbs. The head CT revealed a hyperdensity in the left putamen, and the T1 weighted imaging (T1WI) also revealed left putamen high signal density while low signal density in T2WI. Interestingly, hyperdensity in the left putamen was discovered through retrospective study of the images of his head CT.

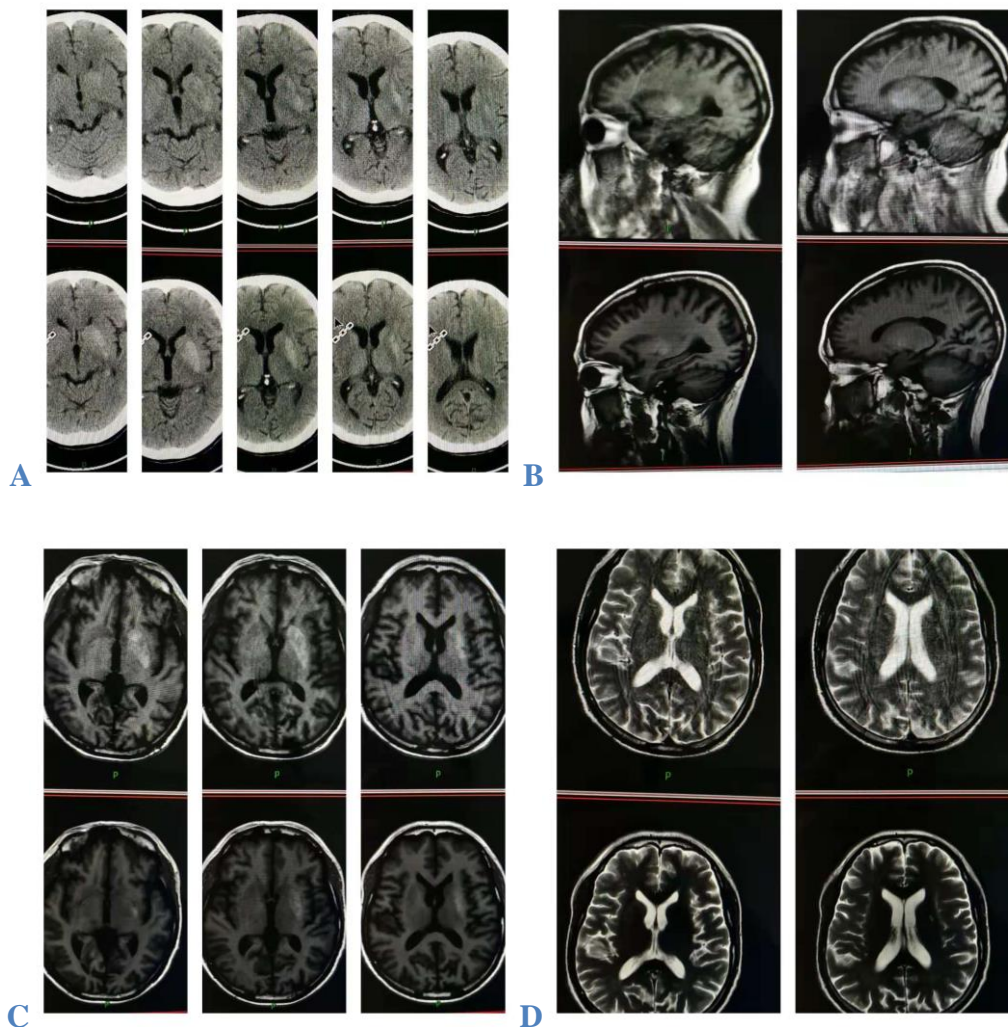
Discussion

In the article, we presented a middle-aged male with hemichorea-hemiballism (HCHB) whose plasma glucose was under good control through formal treatment. By coincidence, he has got an acute ischemia stroke in the left putamen 3 weeks ago, but his head CT scan showed a hyperdensity in the left putamen. The precise pathogenesis remains unclear; thus, our conjectures was raised by combining to certain similar case.

The mechanism in many articles maybe attribute to ischemia infarction (7). As we all know, hyperglycemia makes our blood sticky and syrupy, and slow down the blood flow rate (7) which will cause ischemia, especially the striatum. Yutaka Suzuki et al. compared the focal ischemia of cerebral between patients who developed cerebral infarction and hyperglycemia using the 99m Tc-ECD Patlak Plot method (ECD-SPECT) (7). They found that ischemia and the change of relative blood perfusion which were caused by hyperglycemia were like acute ischemia stroke. Therefore, they presumed that the cause of hemichorea was related to ischemia.

By the same token, we presume that the great and rapid change of blood glucose level would cause local ischemia resulting in contralateral body uncontrolled movements. Firstly, hyperglycemia will damage vascular endothelial cells, and the fluctuation of blood sugar will give rise to much worse damage. Secondly, the fluctuation of blood sugar will generate many superoxide free radicals, which will increase oxidative stress and endothelial cells apoptosis. All these damages to endothelial cells finally broke the integrity of

blood vascular and resulted in ischemia.



Figures 1. The contrast of image changes between this admission and the last admission in every picture.
 A). The hyperdense in the putamen was weakened and faded than 3 weeks ago in the head CT scan
 B). The high signal in the putamen was stronger and larger than 3 weeks ago in the T1WI. (sagittal)
 C). The high signal in the putamen was stronger and larger than 3 weeks ago in the T1WI. (coronal)
 D). No obvious high/low signal was found in the T2WI. (The upper is present and the lower was 3 weeks ago)

Meanwhile, ketone bodies had been considered to play an important role in delayed onset hemichorea (4). Hsiao-Shan Cho et al. reported that acetoacetatecan, which was a kind of ketone bodies, can be used as a GABA substitute to temporarily compensate for the hyperglycemia-induced GABA depletion (4). If our body was under a hyperglycemia situation, each cell can not take well advantage of insulin to lower the high glucose level. Perhaps, the

very fast change of glucose level can not discharge the insulin resistance immediately, so the main energy of brain cells were still ketone bodies. However, it was still just a hypothesis that need more evidence to prove its facticity.

Furthermore, we found a phenomenon where the hyperdense of computed tomography was likely earlier than the absence of hemichorea, under sustained hyperglycemia. There was a similar case reported by Nobuhito Nakajima in

2014 (8). As to the pathogenesis of the occurrence of putaminal abnormalities on CT and T1WI, the best hypotheses involves gemistocytes, calcification and petechial haemorrhage (9-10).

Conclusion

Our case presented in this article indicated the possibility of the appearance of hemichorea under good control of glucose level (11-12). Besides, we found that the changes in CT image maybe earlier than the appearance of hemichorea, which provided a new way for clinicians to identify and predict this infrequent disease, to adopt some intervening measures to prevent it happening.

Declarations

1. *Consent to publication*

We declare that all authors agreed to publish the manuscript at this journal based on the signed Copyright Transfer Agreement and followed publication ethics.

2. *Ethical approval and consent to participants*

Not applicable.

3. *Disclosure of conflict of interests*

We declare that no conflict of interest exists.

4. *Funding*

None

5. *Availability of data and material*

We declare that the data supporting the results reported in the article are available in the published article.

6. *Authors' Contributions*

Authors contributed to this paper with the design (JD), literature search (JD), drafting (JD), revision (DKG and XHH), editing (JD) and final approval (DKG and XHH).

7. *Acknowledgement*

None

8. *Authors' biography*

None

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