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Case Report

Recurrent hypoglycemia, a rare case of insulin autoimmune syndrome in a young African American male

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ABSTRACT

Insulin autoimmune syndrome (IAS) is an uncommon cause of spontaneous hypoglycemia from hyperinsulinemia due to autoantibodies against endogenous insulin (Jian-Ping Chu, 2016). These individuals have no prior exposure to exogenous insulin. We report a case of a 35-year-old African American male, who presented to Vaughn Regional Medical Center in Selma, AL, after he was found to have seizures from hypoglycemia, with a blood sugar of 63 on presentation. He was never diagnosed with diabetes in the past, nor did he have a history of seizure disorder. He continued to be hypoglycemic during the initial period of his hospital stay. His fasting insulin level was 27 mIU/l (normal is less than 25, with presence of insulin autoantibodies (IAA), and a negative workup otherwise. This led us to include IAS as one of our differentials for his hypoglycemia.

1. Introduction

Insulin autoimmune syndrome (IAS), also called Hirata's disease, as it was first reported by Hirata et. Al in 1970 [1], is a rare disease that causes spontaneous hypoglycemia. The characteristic features of IAS are repeated hypoglycemia with hyperinsulinemia, high serum level of IAA, and no history of exposure to exogenous insulin [2]. While many cases have been reported in east Asian countries, there have been very few reported cases amongst the Western World. Out of the reported 23 cases from the United States (US), only two reported cases were from the African American population [3].

2. Case report

A 35-year-old African American male, weighing 131 kg, BMI of 42, was brought to the emergency room after he was found unresponsive in the library, with tremors/twitches in his face and eyes on April 6, 2018. His subsequent presentation was consistent with resultant Todd's Paralysis L > R. He was admitted to the ICU for new onset seizure disorder and blood work was obtained. The initial blood work in the ER showed a blood glucose of 63. He had no documented history of diabetes mellitus and was not on any insulin. His other medical problems were significant for hypertension, asthma, migraine headaches, anxiety, schizoaffective disorder-depression type, and morbid obesity. He was prescribed Depakote, Invega, Zyprexa, and Remeron for his psychiatric problems, but had not taken those medications for at least a month prior to presentation. His social history was positive for occasional consumption of beer, and his family history was positive for diabetes. He had been admitted to an inpatient psychiatric facility in December 2017 for suicidal and homicidal ideations, and his medications were adjusted after that.

This patient continued to be hypoglycemic during this admission with the lowest glucose level measured at 29. No infectious etiology was found. He was started on D10% drip at 175 cc/hr and maintained on this for 5 days to prevent recurrent hypoglycemia. His blood glucoses were monitored and were in the 100s range the entire time while on the D10 drip. His new onset seizure was attributed to hypoglycemia. Once he tolerated oral feeds, his glucose improved. His liver, kidney, thyroid, and pancreatic function, were all normal. His Urine Drug Screen was positive for tricyclic antidepressants. HIV was negative. CT head and MRI brain were negative for any significant findings, negative for CVA. EEG was normal. Diabetes specific labs showed he had an HbA1c of 6.3%, insulin level at 27, C-peptide was 4.4 (NL is > 2) indicating endogenous insulin production; proinsulin level 8.8, b-hydroxybutyrate 0.3, Islet cell ab neg, GAD ab <5.0. ANA titer was negative. Serum cortisol level was 13 (WNL). Insulin antibody positive with elevated levels at 6.8.

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The patient was treated with suggested oral diet with small frequent ADA compliant meals when he was more awake and alert. Neurology was consulted at time of admission for seizures, and tele-psychiatry was consulted for his psychiatric issues. The patient's medications were continued. He continued to improve and feel better and was discharged after 11 days of admission and monitoring, with instructions to follow up with a psychiatrist, neurologist (with continued Depakote) and a free clinic for uninsured people.

3. Discussion

IAS is characterized by a) recurrent hypoglycemia; b) hyperinsulinemia; c) elevated insulin autoantibodies (IAA) and d) no prior exposure to exogenous insulin. It is the third leading cause of hypoglycemia in Japan after insulinoma and extra pancreatic neoplasm. Although majority of the cases were reported from Asian countries, IAS has been recognized in the western world as well [4].

IAS affects males and females equally, and it is seen more frequently in patients more than 40 years old. Although it is not common in children, there have been few reported cases worldwide.

The affected individual may present with various symptoms like confusion, altered mental status, or be in a state of coma on presentation. Our patient had new onset seizure due to hypoglycemia. IAS should be one of the differentials in all patients presenting with hypoglycemia to avoid expensive tests and surgical interventions.

Exposure to drugs like methimazole, sulfonylurea, proton pump inhibitors, antibiotics like penicillin can trigger IAS [5]. Some antipsychotic medications have a side effect of hypoglycemia, but our patient was not taking his medications at the time. Viral infections, autoimmune diseases, or hematological disorders have been shown to trigger IAS. A nutritional supplement used to treat diabetic neuropathy, alpha-lipoic acid, has also been reported to cause IAS [6]. Association between IAS and HLA haplotypes are strong [5]. Although the exact mechanism is unknown, the presumed mechanism is that the body attacks its own insulin, which is released in response to a meal. The attacked insulin dissociates in an uncoordinated manner causing a further increase in insulin release, and resultant hypoglycemia [3]. There is no real treatment besides diet control and small frequent low carbohydrate meals to avoid hypoglycemia. Use of glucocorticoids have shown reduction in hypoglycemic episodes and improved lab results with negative insulin autoantibodies [6].

In the case presented, the patient did not have autoimmune disease. He was on oral medications for psychiatric disorder which have not been reported to cause IAS. He was not treated with glucocorticoids. Adjusting his meals to small frequent diabetic diet resolved the hypoglycemic episodes.

4. Conclusion

IAS should be high amongst the differential in patients being evaluated for spontaneous unexplained hypoglycemia in non-diabetic patients. While it may not be prevalent in the western world, it still mandates the workup when insulin levels are high. This will prevent from unnecessary diagnostic procedures and surgical interventions.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.dsx.2019.05.009.

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