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CASE REPORT

Sodium-glucose co-transporter-2 inhibitor-associated euglycemic diabetic ketoacidosis that prompted the diagnosis of fulminant type-1 diabetes: A case report

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Author contributions: Yasuma T, Tanaka S, and Nishihama K were responsible for clinical treatment and clinical follow-up; Yasuma T prepared the first draft of the manuscript; Eguchi KC, Inoue CC, Maki KC, Okano Y, and Uchida A contributed with resources and acquisition of data; Suzuki T, D’Alessandro-Gabazza CN, Yano Y, and Gabazza EC contributed to the interpretation of the data and made an intellectual contribution to the manuscript’s preparation.

Informed consent statement: Written informed consent was obtained from the patient to publish clinical details and images.

Abstract

BACKGROUND

Fulminant type 1 diabetes mellitus (FT1DM) is a subtype of type 1 diabetes mellitus characterized by an abrupt onset and a rapid and complete functional loss of islet β cells. It is a very rare disease generally associated with ketoacidosis and the absence of circulating pancreatic islet-related autoantibodies. Diabetic ketoacidosis with normal blood glucose levels has been reported during sodium-glucose co-transporter 2 (SGLT2) inhibitor therapy.

CASE SUMMARY

The patient was a 43-year-old woman that consulted a medical practitioner for malaise, thirst, and vomiting. Blood analysis showed high blood glucose levels (428 mg/dL), a mild increase of hemoglobin A1c (6.6%), and increased ketone bodies in urine. The patient was diagnosed with type 2 diabetes mellitus. The patient was initially treated with insulin, which was subsequently changed to an
INTRODUCTION

Fulminant type 1 diabetes mellitus (FT1DM) is a rare subtype of type 1 diabetes mellitus. Abrupt onset of the disease with a rapid and complete loss of islet β cell function is a characteristic finding of FT1DM. Although this metabolic disorder’s clinical manifestations are nonspecific, it can lead to diabetic ketoacidosis and increase death risk if not promptly diagnosed and treated with insulin. Unlike patients with acute-onset type 1 diabetes mellitus, patients with FT1DM are negative for circulating autoantibodies. Therapy with sodium-glucose co-transporter-2 (SGLT2) inhibitor may trigger diabetic ketoacidosis with normal blood glucose levels. Here, we report a case of diabetic ketoacidosis with almost normal blood glucose levels following therapy with SGLT2 inhibitor that prompted the diagnosis of FT1DM.
CASE PRESENTATION

Chief complaints
The patient was a 43-year-old woman that consulted the Mie University Hospital because of fatigue and vomiting.

History of present illness
The patient first consulted a medical practitioner because of sudden malaise, thirst, and vomiting in April 2019. A laboratory analysis disclosed increased blood glucose levels (428 mg/dL), a mild increase of hemoglobin A1c (6.6%), and increased ketone bodies in urine. She received insulin therapy for four days. Because the serum anti-glutamic acid decarboxylase antibody was negative, she was diagnosed with type 2 diabetes mellitus. The treatment was then switched from insulin therapy to oral medication with metformin 500 mg/d, empagliflozin 10 mg/d, and vildagliptin 100 mg/d. The patient's general condition improved, and she was discharged two days after switching to oral treatment when her one-point blood glucose level decreased to 203 mg/dL.

Two days after discharge from the medical practitioner's clinic, she consulted Mie University Hospital's outpatient department complaining of fatigue and vomiting. The clinical findings on examination were as follows: Height 159.3 cm; body weight 58.6 kg, body mass index 23.0 kg/m², blood pressure 128/83 mmHg, heart rate 107 beats/min, body temperature 37.4 °C, and peripheral oxygen saturation (SpO2; normal level > 95%) at room air 98 %.

History of past illness
She had no medical history of any disease.

Physical examination
The patient's physical examination showed notable dryness of the oral cavity.

Laboratory examinations
Table 1 showed the results of the laboratory analysis performed at Mie University Hospital. Arterial blood gases demonstrated metabolic acidosis (pH, 7.18; pCO₂, 18 mmHg; HCO₃, 6.6 mEq/L; base excess, -19.3 mmol/L; A-gap, 27.6 mmol/L), with normal level of lactic acid (1.0 mmol/L). Fasting blood glucose (184 mg/dL) was mildly elevated with increased glycated hemoglobin/hemoglobin A1c (HbA1c) (7.3%). Urinalysis showed high levels of glucose (2000 mg/dL) and ketone bodies (3+). The plasma levels of 3-hydroxybutyric acid (6585 μmol/L) were markedly high. The renal function markers (blood urea nitrogen 9.1 mg/dL; serum creatinine 0.40 mg/dL) were within the normal range. These findings were compatible with the diagnosis of diabetic ketoacidosis with mild hyperglycemia.

Further diagnostic work-up
Additional laboratory analysis showed reduced urinary C-peptide level (8.6 μg/d) and decreased fasting serum C-peptide level (0.3 ng/mL). However, islet-related autoantibodies, including anti-glutamic acid decarboxylase antibody, insulin autoantibody, islet cell antibody, insulinoma-associated protein-2 antibody, and zinc transporter-8 autoantibody, were undetectable. An abdominal computed tomography showed a normal pancreas, and the blood level of pancreas exocrine enzymes, including amylase and lipase, were within the normal range.

FINAL DIAGNOSIS
We made the diagnosis of FT1DM based on the laboratory findings at Mie University Hospital and the severe hyperglycemia (≥ 16.0 mmol/L), and a mild increase in HbA1c (< 8.7%) observed at the medical practitioner's clinic.

TREATMENT
On the first day of hospitalization at Mie University Hospital, besides suspending oral antidiabetic agents, the patient received intravenous fluid infusion (1500 mL),
including 20 g of dextrose and 2 units of regular insulin. After this treatment, her metabolic acidosis substantially (pH, 7.32; pCO₂, 25.9 mmHg; HCO₃⁻, 12.9 mmol/L) improved. Because the patient had reduced blood potassium levels (2.8 mEq/L), 10 mEq/d potassium was administered together with 2000 mL/d of intravenous fluid infusion, including 60 g dextrose and 6 units of regular insulin. Blood glucose level fluctuated between 156 mg/dL and 195 mg/dL during the first two days after hospitalization. On the third day of hospitalization, oral food intake and multiple daily injections of insulin were initiated together with a gradual reduction in fluid administration. Urinary ketone bodies became negative on day 7.

### OUTCOME AND FOLLOW-UP

The patient was discharged on day 14 with the therapeutic indication of subcutaneous injection of insulin glargine (18 U) before sleep and subcutaneous injection of insulin aspart (12U-6U-10U) before each meal.

### Table 1 Laboratory data

<table>
<thead>
<tr>
<th>Blood cell count</th>
<th>Normal range</th>
<th>Units</th>
<th>Biochemical examination</th>
<th>Normal range</th>
<th>Units</th>
<th>Arterial blood gas analysis (room air)</th>
<th>Normal range</th>
<th>Units</th>
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<tbody>
<tr>
<td>White blood cell</td>
<td>3300-8600</td>
<td>/μL</td>
<td>Total protein</td>
<td>6.6-8.1</td>
<td>g/dL</td>
<td>pH</td>
<td>7.35-7.45</td>
<td>mmHg</td>
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<tr>
<td>Red blood cell</td>
<td>4.5</td>
<td>4.5</td>
<td>Albumin</td>
<td>8.0-20.0</td>
<td>mg/dL</td>
<td>pCO₂</td>
<td>115.2</td>
<td>&gt; 80  mmol/L</td>
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<tr>
<td>Hemoglobin</td>
<td>35.1-44.1</td>
<td>%</td>
<td>Creatinine</td>
<td>0.4-0.79</td>
<td>mg/dL</td>
<td>HCO₃⁻</td>
<td>6.6</td>
<td>22.0-26.0 mmol/L</td>
</tr>
<tr>
<td>Platelet</td>
<td>15.8-34.8</td>
<td>× 10⁴</td>
<td>Uric acid</td>
<td>5</td>
<td>mg/dL</td>
<td>Base excess</td>
<td>-19.3</td>
<td>-2.0-+2.0 mmol/L</td>
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<td>Urinalysis</td>
<td>1.015</td>
<td></td>
<td>Na</td>
<td>131</td>
<td>mg/l</td>
<td>Lactic acid</td>
<td>1.0</td>
<td>0.9-1.7 mmol/L</td>
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<td>Specific gravity</td>
<td>1.005-1.030</td>
<td></td>
<td>Cl</td>
<td>102</td>
<td>mg/l</td>
<td>Blood ketone bodies</td>
<td>3</td>
<td></td>
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<tr>
<td>pH</td>
<td>5.5</td>
<td>4.5-7.5</td>
<td>Ca</td>
<td>8.7</td>
<td>mg/dL</td>
<td>Blood ketone bodies</td>
<td>8.8-10.1</td>
<td>mg/dL</td>
</tr>
<tr>
<td>Glucose</td>
<td>2000</td>
<td>mg/dL</td>
<td>P</td>
<td>2.6</td>
<td>mg/dL</td>
<td>Acetoacetate</td>
<td>1784</td>
<td>&lt; 55  μmol/L</td>
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<tr>
<td>Protein</td>
<td>100</td>
<td>mg/dL</td>
<td>AST</td>
<td>20</td>
<td>U/L</td>
<td>3-Hydroxybutyrate</td>
<td>6585</td>
<td>&lt; 85  μmol/L</td>
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<tr>
<td>Ketone body</td>
<td>5 (±)</td>
<td>U/L</td>
<td>ALT</td>
<td>6</td>
<td>U/L</td>
<td>Blood ketone bodies</td>
<td>43</td>
<td>U/L</td>
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<tr>
<td>Blood</td>
<td>6 (±)</td>
<td>U/L</td>
<td>LDH</td>
<td>124-222</td>
<td>U/L</td>
<td>GAD autoantibody</td>
<td>3</td>
<td>U/mL</td>
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<td>Diabetic parameters</td>
<td>ALP</td>
<td>178</td>
<td>γ-GTP</td>
<td>9-32</td>
<td>U/L</td>
<td>IA-2 autoantibody</td>
<td>4</td>
<td>U/mL</td>
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<tr>
<td>HbA1c</td>
<td>7.3</td>
<td>%</td>
<td>T-Bil</td>
<td>0.5</td>
<td>mg/dL</td>
<td>Insulin autoantibody</td>
<td>0.4</td>
<td>&lt; 0.4 U/mL</td>
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<tr>
<td>Glycoalbumin</td>
<td>23.2</td>
<td>%</td>
<td>CRP</td>
<td>0.12</td>
<td>mg/dL</td>
<td>Islet cell antibody</td>
<td>&lt; 0.14</td>
<td>U/mL</td>
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<td>Blood glucose</td>
<td>184</td>
<td>mg/dL</td>
<td>Total-cholesterol</td>
<td>242</td>
<td>mg/dL</td>
<td>ZnT8 autoantibody</td>
<td>&lt; 10.0</td>
<td>&lt; 15.0 U/mL</td>
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<td>Serum C-peptide</td>
<td>0.3</td>
<td>ng/mL</td>
<td>Triglyceride</td>
<td>230</td>
<td>mg/dL</td>
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<td>Urine C-peptide</td>
<td>8.6</td>
<td>pg/d</td>
<td>Amylase</td>
<td>35</td>
<td>U/L</td>
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AST: Aspartate aminotransferase; ALT: Alanine aminotransferase; ALP: Alkaline phosphatase; T-Bil: Total bilirubin; CRP: C-reactive protein; GAD: Glutamic acid decarboxylase; IA-2: Insulinoma-associated protein-2; BUN: Blood urea nitrogen; LDH: Lactate dehydrogenase; HbA1c: Glycated hemoglobin/hemoglobin A1c.
Here, we report a case of F1DM with diabetic ketoacidosis. This case report underscores the importance of considering the diagnosis of F1DM in all patients with
severe hyperglycemia even in the absence of circulating autoantibodies and the possibility of euglycemic diabetic ketoacidosis in patients treated with SGLT2 inhibitors. SGLT2 inhibitors are frequently used as antidiabetic drugs for their cardiovascular[20][21] and renal benefits[22][23]. Recently, Japan approved the use of dapagliflozin and ipragliflozin in combination with insulin for type 1 diabetes, and in May 2020, the United States Food and Drug Administration approved the indication of dapagliflozin for heart failure. The major availability of SGLT2 inhibitors predicts an increase in the number of medical indications of SGLT2 inhibitors in the coming future. Therefore, major awareness of the potential adverse events of SGLT2 inhibitors is fundamental to avoid misdiagnosis in daily clinical practice.

REFERENCES
