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■ Letter

An Unusual Case of Fulminant Type 1 Diabetes Developed during Puerperium

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Dear sir,

Fulminant type 1 diabetes is a newly identified subtype of type 1 diabetes, characterized by pancreatic islet cell destruction, rapid hyperglycemia, and ketoacidosis. During the onset, the hemoglobin A1c (HbA1c) levels are normal or near-normal. Diabetic ketoacidosis (DKA) and complete β-cell destruction occur within a short period of time. The disease is more likely to develop during the third trimester of pregnancy.[1,2] The disease has not been fully characterized. It accounts for 15%–20% of Japanese patients with type 1 diabetes accompanied by ketosis or ketoacidosis at onset.[3] We report a rare case of fulminant type 1 diabetes that developed during puerperium.

A 20-year-old woman presented in the emergency room 1 week postpartum ($G_1P_1L_0$) with several episodes of vomiting, labored breathing, and deterioration of consciousness. On examination, she was severely dehydrated, had cold extremities, and exhibited acidotic breathing. Investigation revealed severe DKA.

She underwent a cesarean section for gestational hypertension and uncontrolled blood sugar, which were detected on admission with labor pain. Her blood sugar and HbA1c levels were 463 mg/dL and 6.3%, respectively. She had no history of gestational diabetes, and the antenatal screening results were unremarkable. Her infant developed severe hypoxic-ischemic encephalopathy, which resulted in multiple organ dysfunction syndrome and eventually, death. Upon initiating insulin therapy, her blood glucose level normalized.

During the postpartum HbA1C report, her doctor decided to stop insulin, and check her fasting glucose and C-peptide levels after 1 week without insulin. However, she presented to the emergency department with severe DKA after 2 days. She had a normal body mass index (23.1 kg/m^2) and unremarkable personal and family history. The laboratory findings on admission are shown in Table 1. Autoantibodies to glutamic acid decarboxylase and islet cells were negative, and the fasting C peptide was <0.005 ng/mL. She was treated for DKA. High doses of insulin were administered (>80 U/d) to attain and maintain euglycemia. The patient recovered completely. She continued receiving insulin under regular follow-up. Consent for publication of this case report was obtained from the patient.

The rapid onset and progression to severe DKA in the absence of autoantibodies and C-peptide that initially occurs during labor, despite good antenatal follow-up, favors the diagnosis of fulminant type 1 DM. The normal serum amylase level may be due to the time delay in the estimation (1 week after onset). Based on these findings, viral infection and the

Table 1. Laboratory findings at admission

Variable	Value
Blood: total count	37,400 cells/cmm
Differential count	P78 L16 M6
Casual plasma glucose	673 mg/dL
Urine acetone	++++
Urine sugar	++
Arterial blood gas analysis	pH: 6.94; $PaCo_2$: 9.5 mm Hg; PaO_2: 133 mm Hg; HCo_3^2: 1.9 mmol/L
Liver function test	Normal
Renal function test	Normal
Serum electrolytes	Na: 126 mEq/L; K: 4.55 mEq/L; Cl: 92 mEq/L
Serum amaylase	Normal
C-reactive protein	3.2 mg/dL

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subsequent immune reaction in genetically susceptible individuals cause beta-cell destruction and fulminant type 1 diabetes.[4)]

CONFLICT OF INTEREST

No potential conflict of interest relevant to this article was reported.

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