Reversible Cardiomyopathy Accompanied by Secondary Adrenal Insufficiency

Yukinori Ikegami, MD, PhD; Tadashi Fukuda, MD; Rie Jo, MD; Yukihiro Momiyama, MD, PhD

Case Report

A 41-year-old woman was admitted to our hospital because of worsening dyspnea for 2 weeks. She experienced fatigue and shortness of breath on physical activity (5 metabolic equivalents) for 8 years after delivery, and she considered this normal. She also had meningitis 3 months before presentation that resolved in 2 weeks. There was no evidence of ischemia or infection, and she had no history of alcohol abuse or illegal drug use. Twelve-lead electrocardiography showed sinus rhythm with low voltage, chest radiography showed pulmonary congestion and a dilated cardiac silhouette (Figure 1A), echocardiography showed a dilated left ventricle (Figure 1B) with reduced ejection fraction of 25%, and cardiac magnetic resonance imaging demonstrated low ejection fraction without specific findings of inflammation, fibrosis, or deposition. Diuretic injection improved dyspnea in 2 days. Low-dose β-blocker administration was started for idiopathic cardiomyopathy with reduced ejection fraction; however, the β-blocker was discontinued because it caused hypotension. Two days after admission, blood tests indicated a low cortisol level (0.1 μg/dL; normal range, 10.4–26.4 μg/dL). The thyroid-stimulating hormone level was high (Table); however, the main endocrine pathological condition was believed to be adrenal insufficiency. Cortisol was administered for adrenal insufficiency, and her hypotension improved in 3 days and fatigue in 2 weeks. In 2 months, her physical activity level increased to that present before delivery and the thyroid-stimulating hormone level returned to normal. Considering the long-term fatigue after delivery and the results of endocrine function tests (Table) and brain magnetic resonance imaging that revealed an empty sella (Figure 2), she was diagnosed with secondary adrenal insufficiency caused by idiopathic cardiomyopathy. Because cardiomyopathies accompanied by secondary adrenal insufficiency caused by Sheehan syndrome have been reported to be reversible,1–3 we continued oral cortisol therapy alone, and her cardiac function gradually improved. Complete recovery of ejection fraction was noted 2 months after the start of oral cortisol therapy (Figure 1C and 1D). In addition, her B-type natriuretic peptide level improved from 1399.6 pg/mL at admission to 26.7 pg/mL (normal range, <18.5 pg/mL) 2 months after treatment. She has been on oral glucocorticoid therapy alone for 1.5 years, and follow-up with echocardiography and magnetic resonance imaging has indicated normal sustained cardiac function.

Discussion

Few cases of reversible cardiomyopathy accompanied by Sheehan syndrome have been reported.1–3 These cases have the common characteristics of a late diagnosis of Sheehan syndrome after delivery (>2 years) and recovery of cardiac function after hormone replacement therapy, including glucocorticoid therapy. One of the previous cases was initially diagnosed with peripartum cardiomyopathy and showed no improvement of cardiac function with standard medications for heart failure until hormone replacement therapy was initiated.3 Another case was administered hormone replacement therapy without standard medication for heart failure because of adrenal crisis.1 Sheehan syndrome is characterized by hypopituitarism caused by ischemic necrosis from blood loss and hypovolemic shock during and after childbirth, resulting in multisystem endocrine insufficiency. Thus, the previous cases were treated with multiple courses of hormone replacement therapy. In the present case, the patient had no complications during delivery, massive bleeding, or hypovolemic shock, except for fever after delivery that persisted for a night and resolved without medication. Considering these findings, her history of fatigue and monophyletic endocrine insufficiency and the positive response to cortisol replacement therapy, hypophysitis rather than Sheehan syndrome was more likely the cause of her secondary adrenal insufficiency, with exacerbation of adrenal insufficiency after meningitis. To our knowledge, no case of hypophysitis-induced secondary adrenal insufficiency along with heart failure successfully treated with cortisol replacement therapy alone has been reported in the literature. The findings of this case are consistent with those of previous cases1–3 in terms of long-term fatigue that was considered normal after delivery. Therefore, adrenal insufficiency may remain undiagnosed after delivery, suggesting the possible existence of women with undiagnosed endocrine insufficiency, who may have heart failure in the future. In conclusion, we re-emphasize the importance of the evaluation of the underlying disease with idiopathic cardiomyopathy that might be curable.
Disclosures

None.

References


Key Words: adrenal insufficiency ■ cardiomyopathy ■ echocardiography ■ magnetic resonance imaging ■ women

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<td>Triliodothyronine-T3, pg/mL</td>
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Figure 1. A. Chest radiography showing pulmonary congestion and a dilated cardiac silhouette. B. Echocardiography showing a dilated left ventricle (left ventricular end-diastolic diameter, 50 mm) and an ejection fraction of 25%. C. Chest radiography performed 2 months after admission showing no pulmonary congestion and a normal cardiac silhouette. D. Echocardiography performed 2 months after admission showing improvement in the size of the left ventricle (left ventricular end-diastolic diameter, 42 mm) and an ejection fraction of 60%.

Figure 2. Brain magnetic resonance imaging showing an empty sella (red arrow).
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