

Case Report

Reversible cardiomyopathy as a rare presentation of sheehan's syndrome-case report and review of literature

Mohmmad Hayat Bhat, Farhana Bagdadi*, Asma Rafi, Parvaiz Ahmad Shah

Department of Medicine, Government Medical College Srinagar, Jammu and Kashmir, India

Received: 26 July 2017

Accepted: 19 September 2017

*Correspondence:

Dr. Farhana Bagdadi,

E-mail: farhanasirajmir@gmail.com

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ABSTRACT

Sheehan's syndrome is a clinical condition characterised by post-partum panhypopituitarism caused by necrosis of the pituitary gland. The hypervascularity of the pituitary gland during pregnancy makes it vulnerable to arterial pressure changes and prone to haemorrhage in the post-partum period. The cardinal features are lethargy, secondary amenorrhea and lactational failure. The diagnosis in immediate post-partum period is difficult and require a high degree of suspicion. Cardiac involvement in sheehan's syndrome is known but rare.

We hereby present a case of post-partum cardiomyopathy with relatively poor response to anti failure treatment. Patient however responded to hormone replacement including glucocorticoids and levothyroxine, after proper confirmation of the hypopituitary state. Cardiac dysfunction markedly reversed with the institution of replacement therapy.

Keywords: Hypopituitarism, Reversible dilated cardiomyopathy, Sheehan's

INTRODUCTION

Post-partum panhypopituitarism due to necrosis of the pituitary gland resulting from severe post-partum hemorrhage qualifies for the typical Sheehan's syndrome.¹ A high index of suspicion is needed to diagnose Sheehan's syndrome in the immediate post-partum period while dealing with a case of peripartum cardiomyopathy.² Leading clues to diagnosis and initiation of appropriate hormonal therapy go a long way in reversing the clinical condition.

In this report, we present a case of dilated cardiomyopathy in a 30-year-old female who had history of pregnancy induced hypertension in the last trimester, followed by postpartum hemorrhage. She presented as acute decompensated heart failure and responded sub optimally to anti failure treatment. She was given appropriate hormone replacement after hypopituitarism

was confirmed both biochemically and radiologically. Patient had a dramatic response as replacement treatment was initiated. This clearly proved to be the cornerstone in her management with prompt reversal of her cardiac dysfunction.

CASE REPORT

A 30-year-old female para 3 with history of normal vaginal deliveries, had her last normal vaginal delivery 10 days back at home which got complicated by post-partum hemorrhage that required hospital admission with transfusion of three units of packed red cells. There was a history of pregnancy induced hypertension in the last trimester. After getting discharged from the hospital she gradually developed easy fatigability, shortness of breath, orthopnea and swelling of both feet. Clinical examination revealed patient was confused with pulse rate of 84/min (regular), respiratory rate of 24 breaths/min and BP

90/60mmHg. Cardiovascular examination revealed a leftward shifted diffuse apex beat with an S3 gallop.

Her complete blood counts revealed bicytopenia (HB-7.2gm/dl, TLC-1790/cumm, N28.8%; L55%; E 7%; PLT-1.69lacs). Bone marrow aspiration and biopsy showed erythroid hyperplasia with predominant normoblastic maturation. Biochemistry revealed low sugars (blood sugar 60mg/dl) and azotemia (creatinine 2.0mg/dl). Chest X-ray showed cardiomegaly and ECG revealed diffuse T wave inversions in the chest leads. There was echocardiographic evidence of dilated cardiac chambers with moderate global hypokinesia and moderate LV systolic dysfunction with EF-40-45%. In view of significant peripartum history (post-partum hemorrhage, decompensated heart failure) and low blood sugars a clinical suspicion of Sheehan's syndrome emerged. It was confirmed biochemically (FSH-0.10IU/ml; LH-0.02IU/ml; Cortisol-0.0IU/ml; Prolactin-3.7IU/ml, TSH 8.0IU/ml, Free T4) and CEMRI evidence of partially empty sella with normally located PPBS on contrast enhanced MRI of the sellar/parasellar region Figure 1.

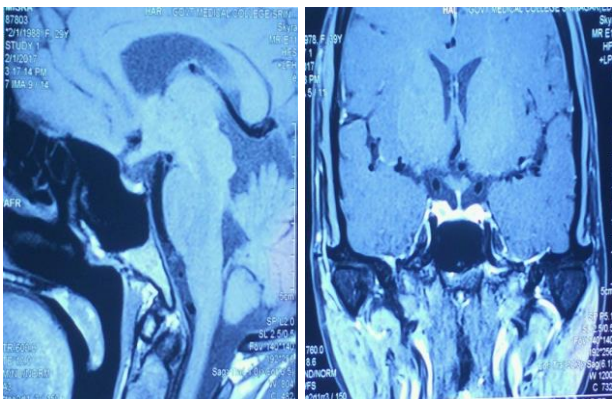


Figure 1: Post contrast MRI pituitary showing partial empty sella with thin rim of pituitary tissue in place.

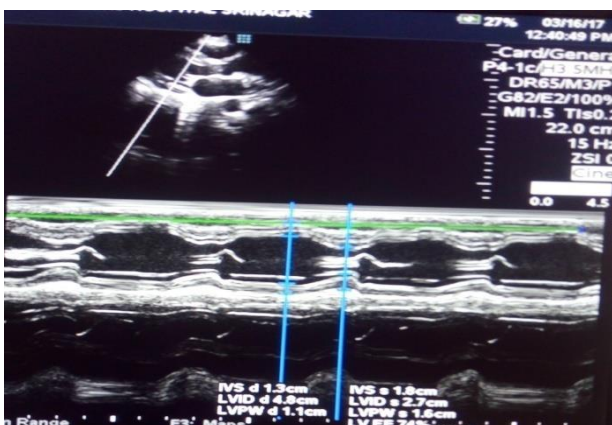


Figure 2: Echocardiography showing normal chamber size and ejection fraction.

Patient was diagnosed as a case of Sheehan's syndrome with dilated cardiomyopathy. Patient received anti-failure

medication (oxygen, diuretics, ACE inhibitors) initially with relatively lesser response. Once appropriate hormonal replacement for hypopituitarism was done, patient had near normal recovery. Serial monitoring of her blood counts and biochemistry showed significant improvement and got normalized. Echo cardiography after three months revealed normal LV systolic functions with EF=70% and complete resolution of hypokinesia. Figure 2.

Anti-failure treatment was withdrawn on follow-up in view of improved cardiac abnormalities. She is doing well on follow up and is on levothyroxine and prednisolone only.

DISCUSSION

Our patient was diagnosed as Sheehan's syndrome in immediate post-partum period with echo cardiographic evidence of dilated cardio myopathy which is a very rare presentation and difficult to diagnose. Patient was initially thought to have peri partum cardiomyopathy but a proper history, clinical accumen, biochemical evidence of hypopituitarism and imaging helped us to reach a proper diagnosis. She had history of pregnancy induced hypertension and had post-partum hemorrhage in immediate post-partum period which required blood transfusion. Pregnancy, pregnancy induced hypertension and postpartum hemorrhage are all risk factors for pituitary apoplexy. Although pituitary apoplexy is an endocrinological emergency caused by acute hemorrhage or acute infarction in the pituitary gland where patient can present with acute headache, vomiting, cranial nerve palsies. However, our patient had sub clinical presentation/apoplexy which was diagnosed 2weeks after delivery.

She gradually improved with correction of hypocortisol and hypothyroid state and cardiac abnormalities completely reversed once she was euthyroid and eucortisolemic. Although various types of cardiac abnormalities have been described in patients with sheehan's syndrome like pericardical effusion, dilated/hypertrophic cardiomyopathy which reverse with replacement therapy.^{3,4} There are only few case reports of Sheehan's syndrome with reversible cardiomyopathy in the literature and presentation and diagnosis in immediate post-partum period is rarest acute manifestation.⁵⁻⁹ Although GH deficiency leads to sub clinical left ventricular dysfunction and is thought to be the cause for the dilated cardiomyopathy in such patients. But our patient received only thyroxine replacement and steroids and anti-failure measures and showed marked improvement on follow up. So, role of growth hormone deficiency as a cause of cardiomyopathy needs to be seen and explained.

Parikh et al has reported a 37-year-old female who concomitantly presented with lymphocytic hypophysitis, pan hypopituitarism, post-partum cardiomyopathy and

pnemonitis. Patient received steroids, levothyroxine, ACE inhibitors, beta blockers and showed marked improvement on follow up for 2years and was maintained on prednisolone and levothyroxine. The woman was found to be growth hormone deficient but was not replaced.^{10,11} So the role of steroids and levothyroxine is important for such changes to take place. There has also been some evidence that Sheehan's syndrome has an auto immune basis and auto immune etiology has been suggested for cardiomyopathy.¹⁰⁻¹² Wane et al has reported cardiomyopathy secondary to hypocalcemia and hypocortisolemia as a part of autoimmune polyglandular syndrome type 1 which was reverted to normal after appropriate correction.¹²

Our patient had some atypical biochemical finding like bicytopenia and azotemia which subsequently reversed in addition to cardiomyopathy. Although acute renal failure as a presentation of sheehan's syndrome has been reported.¹³ We believe that patient developed hypotension during hospital course which could be the possible explanation of acute renal insult and had bicytopenia at presentation which later on improved but could not be explained in acute sheehan's. We believe that autoimmunity and acute hormonal deficiencies both plays a role in these hematological and cardiac abnormalities and need to be studied further.

Funding: No funding sources

Conflict of interest: None declared

Ethical approval: Not required

REFERENCES

1. Kaufman MS, Ganti L, Holmes J, Schachel P. First Aid for the Obstetrics and Gynecology Clerkship. McGraw Hill Professional; 2010.
2. Schragar S, Sabo L. Sheehan syndrome: a rare complication of postpartum hemorrhage. J Americ Board of Family Practice. 2001;14(5):389-91.
3. Sheehan HL. The incidence of postpartum hypopituitarism. Am J Obstet G. 1954;68(1):202-23.
4. Zargar AH, Masoodi SR, Laway BA, Shah NA, Salahuddin M, Siddiqi MA, Kour S. Clinical spectrum of Sheehan's syndrome. Annals of Saudi medicine. 1996;16(3):338-41.
5. Laway BA, Alai MS, Gojwari T, Ganie MA, Zargar AH. Sheehan's syndrome with reversible dilated cardiomyopathy. Ann Saudi Med. 2010;30(4):321-4.
6. Vasanti N, Asha H, Paul T, Sunithi M. Sheehans syndrome with reversible cardiomyopathy. A case reports. 2014;3(5):44.
7. Wang SY, Hsu SR, Su SL, Tu ST. Sheehan's syndrome presenting with early postpartum congestive heart failure. Journal of the Chinese Medical Association. 2005;68(8):386-91.
8. Bao S, Fisher S. Repairing a "broken heart" with hormone replacement therapy: case report of cardiogenic shock due to undiagnosed pituitary insufficiency. Endocrine Practice. 2011;18(2):e26-31.
9. Parikh A, Ezzat S. Complete anterior pituitary failure and postpartum cardiomyopathy. Endocrine practice. 2006;12(3):284-7.
10. Goswami R, Kochupillai N, Crock PA, Jaleel A, Gupta N. Pituitary autoimmunity in patients with Sheehan's syndrome. Journal Clinic Endocrinol Metabol. 2002;87(9):4137-41.
11. Sundstrom JB, Fett JD, Carraway RD, Ansari AA. Is peripartum cardiomyopathy an organ-specific autoimmune disease? Autoimmunity reviews. 2002;1(1):73-7.
12. Wani AI, Farooqui KJ, Bashir MI, Lone AA, Masoodi SR. Autoimmune polyglandular syndrome type 1 with reversible dilated cardiomyopathy: complete recovery after correction of hypocalcemia and hypocortisolemia. J Peadiatr Endocrinol Metab. 2013;26(3-4):373-6.
13. Bhat MA, Laway BA, Allaqaband FA, Kotwal SK, Wani IA, Banday KA. Acute renal Failure: A rare presentation of Sheehan's syndrome. Indian J Endo and Metab. 2012;16(2):306-9.

Cite this article as: Bhat MH, Bagdadi F, Rafi A, Shah PA. Reversible cardiomyopathy as a rare presentation of sheehan's syndrome-case report and review of literature. Int J Adv Med 2017;4:1713-5.