Case Report

Sheehan's Syndrome — A Case Report

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Anterior pituitary infarction occurring in the postpartum period and the ensuing hypopituitarism is called Sheehan's syndrome. Postpartum hemorrhage is an important precipitating factor for Sheehan's syndrome. Here we describe a primigravida with severe postpartum hemorrhage who remained drowsy and failed to produce breast milk in the puerperium. Her MRI T2 & FLAIR revealed hyperintense and enlarged anterior pituitary gland consistent with anterior pituitary infarction. Her cortisol and prolactin levels were also low. After replacing corticosteroids her clinical condition improved. Failure to lactate, failure of return of menses in a puerperal woman along with typical MRI findings and low anterior pituitary hormone levels suggests Sheehan's syndrome. Assessing hormonal deficiencies and replacing them forms the cornerstone of management.

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Key words **: Sheehan's syndrome, Anterior pituitary, Postpartum hemorrhage.**

In 1937 Harold L Sheehan, a Pathologist at Glasgow Royal Maternity Hospital performed autopsy of women who died in late pregnancy. He found that nearly 12 out of 76 had destruction of anterior pituitary whose common clinical feature was hemorrhagic shock. He compiled his experience in a monograph titled 'postpartum hypopituitarism' which was published in 1982. He was also a man of modesty who had aversion to the term Sheehan's syndrome and preferred postpartum pituitary necrosis. His exemplary work on postpartum hypopituitarism has stood the test of time. Anterior pituitary infarct due to puerperal hemorrhage is called as Sheehan's syndrome[1]. Six percent of all hypopituitarism is attributed to Sheehan's syndrome. In India 3 percent of women above 20 years develop postpartum pituitary necrosis[2]. The clinical presentation of this entity can be acute (less common) or chronic. Here, we report a case of an acute presentation of Sheehan's syndrome in a young female who had massive postpartum hemorhage (PPH).

Case Report

A 25 year old primigravida with twin gestation of 34 weeks delivered dead born male babies via labour naturalis. She had abruption of placenta, leading to massive postpartum hemorrhage. Subsequently she developed features of HELLP syndrome and disseminated intravascular coagulation. Her clinical course was further complicated by acute pulmonary edema and acute kidney injury. She was intubated and ventilated soon after delivery and required inotrope support to maintain hemodynamics. She was transfused 10 units of blood and blood products including platelets and fresh frozen plasma (FFP). Initially she was conscious and oriented. General examination revealed severe pallor and anasarca. Her temperature was 102.2°F, pulse 118/min, and a supine blood pressure of 130/80 mm Hg. Systemic examination revealed normal heart sounds with bibasal crackles and a soft abdomen. There was trickling of blood with few clots per vaginally. On laboratory evaluation She had low hemoglobin (Hb -7 g/dl), raised urea levels (blood urea 110 mg/dl), liver enzyme (ALT 118 U/L), uric acid (18 mg/dl) and Lactate dehydrogenase (4300 U/L). Her electrolytes were within the normal range. She had prolonged prothrombin time (19 seconds) hence an elevated international normalized ratio (1.42).

On day 2 of post partum,her blood clots were evacuated under general anesthesia with ultrasound guidance. At the end of procedure she sustained sudden cardiac arrest but was resuscitated successfully. Around 3 litres of blood clots were removed and blood products were replaced along with uterotonic infusion. Post operatively, she continued to bleed per vaginally for which uterine massage, FFP transfusion and prostaglandin infusion were administered. After a few hours her bleeding stopped.

On day 6 of post partum, she still remained drowsy and somnolent. She was not able to move her limbs and her speech was very slow. She also had lactation failure. Computed tomography(CT)of brain revealed enlarged pituitary gland with prominent stalk (Fig 1). Magnetic resonance imaging (MRI) T2/FLAIR of brain showed hyperintense lesion in an enlarged pituitary gland (anterior portion) suggestive of anterior pituitary infarct. Hormonal

Editor's Comment :

- **Sheehan's syndrome should be suspected in a postpartum woman with massive uterine bleed following childbirth, hypotension, agalactorrhea, and failure of return of menses.**
- **Careful assessment and replacement of hormones is necessary to avoid life threatening consequences.**

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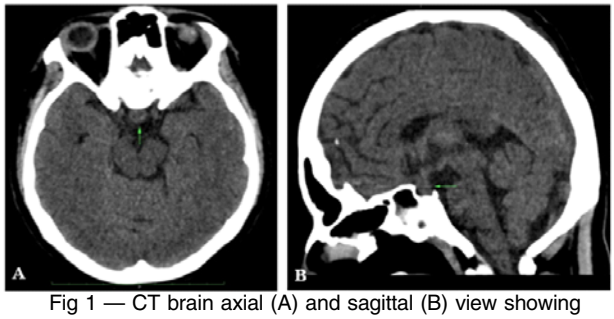

Fig 1 — CT brain axial (A) and sagittal (B) view showing enlarged pituitary with prominent stalk

assays including thyroid stimulating hormone (TSH), cortisol and prolactin were performed. Low prolactin (7.23 ng/ml) and cortisol levels (1.8 mcg/dl at 6 am) were noted. In the background of PPH, her reduced alertness, failure of lactation, low cortisol and prolactin levels along with MRI which was consistent with anterior pituitary infarction made us to diagnose Sheehan's syndrome. Corticosteroid was replaced (hydrocortisone 50 mg per day intravenous) along with hematinics and physiotherapy. After 48 hours of hormonal replacement her sensorium improved and was discharged on 14th day successfully.

Discussion

Sheehan's syndrome or postpartum pituitary necrosis results in anterior pituitary hormonal deficiency due to infarction of anterior pituitary. Postpartum hemorrhage, autoimmunity, coagulation abnormalities and small sellaturcica are some of the risk factors for developing postpartum pituitary necrosis. Our patient had postpartum hemorrhage which could have caused transient hypoperfusion of anterior pituitary resulting in infarction and necrosis in a physiologically enlarged gland. The production of growth hormone and prolactin is affected the most. In severe necrosis TSH and ACTH secretion is also affected. Diabetes insipidus is rare and can occur if the stalk is damaged[2]. The clinical spectrum varies from immediate postpartum circulatory collapse to mild central hypothyroidism manifesting many years later after the inciting event. Postpartum hypotension and hypoglycemia are the clinical clues in acute presentation. Failure to lactate is a feature of subacute presentation[3]. Secondary hypothyroidism and secondary adrenal insufficiency occurred in all 28 patients of Sheehan's syndrome studied by Sert et al whereas Banzal et al reported in 97 % and 90 % of his patients (n=30) respectively[4,5]. The secretion of prolactin and cortisol were affected in our patient as expected. Laboratory abnormalities usually reveal a normocytic anemia, hyponatremia and hyperlipidemia. Hypernatremia can occur if stalk is damaged. MRI findings include T1 hypointense and T2 hyperintense enlarged anterior pituitary which is consistent with infarction as noted in our patient. Irregular enhancement occurs with Gadolinium contrast. Sequential MRI will show atrophied pituitary resulting in empty sella. Lymphocytic hypophysistis is the most relevant differential diagnosis as it also occurs commonly in peripartum period. It's a nonneoplastic inflammatory condition in which lymphocytes infiltrate pituitary gland resulting in enlargement and impaired production of hormones. Uniform enhancement with gadolinium contrast

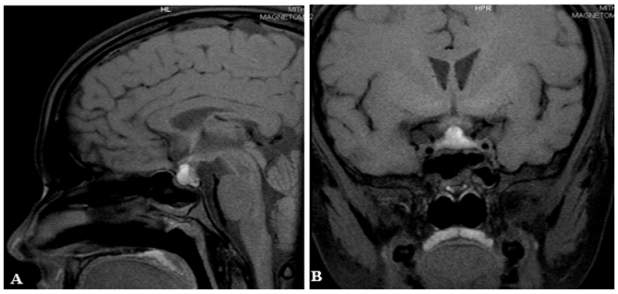

Fig 2 — MRI brain- FLAIR sagittal (A) and coronal view (B) showing enlarged and hyperintense anterior pituitary suggestive of anterior pituitary infarct

and cystic appearance in MRI will help in differentiating this entity from Sheehan's syndrome. Pituitary tumors usually exhibit uniform enhancement but can cause diagnostic confusion particularly if necrosis occurs[6].

Management of postpartum hemorrhage aggressively can prevent hypoperfusion of the pituitary. Cord clamping, controlled cord traction and usage of uterotonics are some of the techniques to avoid PPH[2]. Assessment of hormonal deficiency guides further management. Thyroxine supplement should be done only after corticosteroid replacement as thyroxine enhances the metabolism of steroids which may result in adrenal insufficiency. Gonadal steroids can be replaced in pre-menopausal women to maintain bone density and to improve quality of life. The benefit of growth hormone replacement is uncertain[1].

Conclusion

Sheehan's syndrome should be suspected in a postpartum woman with massive uterine bleed following childbirth, hypotension, agalactorrhea, and failure of return of menses. Diagnosis can be delayed as the timing of presentation and symptom range varies widely. MRI can help in diagnosing anterior pituitary infarction.Careful assessment and replacement of hormones is necessary to avoid life threatening consequences and also to improve the quality of life.

References

1 Tessnow AH, Wilson JD — The Changing Face of Sheehan's Syndrome. The American Journal of the Medical Sciences [Internet]. Elsevier BV; 2010 Nov; **340(5):** 402-6.

2 Karaca Z, Laway B, Dokmetas H, *et al* — Sheehan syndrome. *Nat Rev Dis Primers* **2:** 16092 (2016).

3 Matsuzaki S, Endo M, Ueda Y, Mimura K, Kakigano A, Egawa-Takata T, *et al* — A case of acute Sheehan'ssyndrome and literature review: a rare but life-threatening complication of postpartum hemorrhage. BMC Pregnancy and Childbirth [Internet]. Springer Science and Business Media LLC; 2017 Jun 14; **17(1):**

4 Sert, Murat & Tetiker, Tamer & Kirim, Sinan & Kocak, Mikail — Clinical Report of 28 Patients with Sheehan's Syndrome. Endocrine journal. 50. 297-301(2003).10.1507/endocrj.50.297.

5 Banzal S, Ayoola EA — Sheehan'ssyndrome in Saudi Arabia. *Int J Gynaecol Obstet* 1999; **66:** 181-2.

6 Kaplun J, Fratila C, Ferenczi A, Yang WC, Lantos G, Fleckman AM, *et al* — Sequential Pituitary MR Imaging in Sheehan Syndrome: Report of 2 Cases. American Journal of Neuroradiology [Internet]. *American Society of Neuroradiology (ASNR)* 2008 Feb 22; **29(5):** 941-3.