Original Research Paper

Volume - 13 | Issue - 04 | April - 2023 | PRINT ISSN No. 2249 - 555X | DOI : 10.36106/ijar

General Surgery

AN INTERESTING CASE OF HYPONATREMIA – SHEEHAN SYNDROME

Dr. Jaya Lalwani

Dr. Gunasekaran N.

ABSTRACT Sheehan syndrome (postpartum hypopituitarism) results hypopituitarism because of necrosis of the pituitary gland due to hemorrhagic shock in pregnancy. This disease is a rare complication with different clinical presentations, occurs years after the complication hence there is a significant delay in the diagnosis. In our study, we report a case of 46 years old female patient presented with illness of vomiting since one month and unexplained fatigue for the past 10 years. She also had amenorrhea. She was diagnosed to be hypothyroid two years back and on medication (thyroxine 50 mcg). On clinical examination her blood pressure was 90/60mm of Hg. Laboratory investigations revealed hyponatremia. Complete clinical examination, hormonal studies, MRI brain showed hypopituitarism due to Sheehan's syndrome. The patient improved clinically after hydrocortisone injection and her sodium level is normal. She is still under medication. Though this is a rare complication, the patient detailed past medical history, clinical examination and laboratory tests helps the clinicians for an early diagnosis of this syndrome.

KEYWORDS : postpartum hypopituitarism, hyponatremia, Sheehan syndrome.

INTRODUCTION:

Sheehan's syndrome is also known as postpartum hypopituitarism, postpartum pan hypopituitarism,postpartum panhypopituitary syndrome, and postpartum pituitary necrosis. Sheehan's syndrome was described by H.L.Sheehan in 1937. It is characteristically caused by ischemic necrosis of pituitary gland which occurs due to spasm of its arterioles at the time of severe hemorrhage or shock (usually postpartum) complicating child birth (1). Pituitary gland is an endrocrine gland that is suspended from the brain by infundibulum (pituitary stalk). The vascularisation of pituitary gland is by superior hypophyseal artery (supplies infundibulum,pars tuberalis, and inferior hypophyseal artery (supplies posterior lobe). Pars distalis has unique circulation to receive hormones from the hypothalamus and posterior pituitary gland but it is vulnerable to hypotension during postpartum period. It is rare in developed countries due to advanced techniques in obstetric care. Sheehan's syndrome is common in developing countries and the prevalence was estimated throughout the world is 1 in 1,00,000 women who had given birth at home (2). The main cause for the formation of Sheehan's syndrome is the pregnancy. In pregnancy, there is physiologically enlargement of pituitary gland usually preceeded by postpartum hemorrhage which leads to arterial spasm in smaller vessels and results in infarction and necrosis of this gland (3). Restricted blood supply to the pituitary gland following untreated severe hypotension associated with postpartum hemorrhage is the main cause for the development of Sheehan's syndrome. The pathogenesis of Sheehan's syndrome is still remains uncertain. Not every patient with the history of massive postpartum hemorrhage nor does every massive postpartum hemorrhage lead to Sheehan's syndrome. The important risk factor in the pathogenesis of Sheehan's syndrome are vasospasm,thrombosis or vascular compression of hypophyseal arteries,history of previous birth associated with postpartum, number of births, smaller size of sella turcica, autoimmune component, and genetic predisposition (4).Sheehan's syndrome may be acute or chronic. The acute Sheehan's syndrome includes no lactation, hypotension and hypoglycemia. The chronic symptoms includes no lactation, amenorrhea, fatigue, anemia , hypothyroidism and hypoadrenal (5). Depending on the degree of tissue destruction, the hypopituitarism occurs immediately or after a delay of several years (6-8).

Hyponatremia is also not common in acute form of Sheehan's syndrome. In addition, in patients with Sheehan's syndrome the spontaneous pregnancy is very rare (9). Sheehan's syndrome with initial hyponatremia and hypoglycemia is rarely noted in literature (10). Early diagnosis and proper treatments are very important to decrease the morbidity and mortality. Hence, in our study we are presenting an interesting case of Sheehan's syndrome with hyponatremia.

CASE REPORT:

A 46 years old female patient presented with illness of vomiting since one month and unexplained fatigue for the past 10 years. These symptoms started after the delivery of her second child birth in home after which she developed excessive vaginal bleeding and was treated locally and survived. After her delivery, She had amenorrhea and failed to breast feed. Due to family issues, she did not undergo any gynecological evaluation. She had a history of dizziness,cold intolerance,diarrheoa,vomiting and abdominal pain repeatedly and was treated by nearby clinic symptomatically. These symptoms worsened for the past one month. Routine investigations were done previously in the other hospital where she was diagnosed to be hypothyroid for 2 years and was on Tablet .Thyroxine 50mcg. On clinical examination her blood pressure was 90/60mmHg. Systemic examination was unremarkable.

Laboratory investigations revealed hyponatremia (sodium:127mEq/L). Serum cortisol level was 11.55 mg/dl. Serum FSH and LH levels were subnormal. MRI brain revealed empty sella.

She was started on injection hydrocortisone and she improved clinically and her sodium levels came back to normal. She is currently on oral hydrocortisone 10 mg TDS and Tablet .Thyroxine 50mcg.

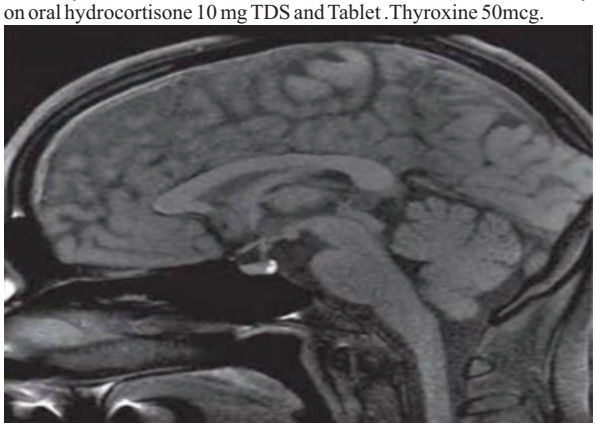

Fig 1: MRI image showing diminished pituitary gland.

DISCUSSION:

Sheehan's syndrome accounts for 0.5% of all known cases of hypopituitarism in females. The pathogenesis behind Sheehan's syndrome is due to decreased vascularisation of the physiologically enlarged pars anterior or due to hyperplasia of prolactin secreting cells because of increased oestrogen secretion or by the spasm in arterioles or vascular compression of supplying hypophyseal vessels due to enlarged gland during postpartum events.The average time between the previous obstetric event and the diagnosis of Sheehan's syndrome was 6 to 13 years was reported in 124 pateints (11).

Hyponatremia is the common electrolyte imbalance, seen in 33–69% of cases. Hyponatremia is an earlier symptom of acute Sheehan's

syndrome which accounts for 60% of the reported cases (12-14). Acute hyponatremia is life-threatening because this causes cerebral edema. There are several pathophysiological mechanisms for the formation of hyponatremia in this Sheehan's syndrome. Hypothyroid and hypoadrenal causes hyponatremia due to increased ADH (15, 16). In our study, we reported a case of hyponatremia in Sheehan's syndrome may be due to hypothyroidism. Gopal et al, recently reported the hematological abnormalities in 65 patients with Sheehan's syndrome in that 80 % of the patients were with anaemia (17). In our case we also noted the patient presented with anaemia. Abere Genetu et al reported that in female patients the first clinical presentation to diagnosis was her lack of lactation and amenorrhea (18). Our patient also presented with the symptom of amenorrhea. Khan SA et al diagnosed Sheehan's syndrome based on the features of hormone deficiency, a suggestive obstetric history and decreased levels of basal hormones (free T3, T4, TSH, FSH, LH, estrogen, prolactin, cortisol, and insulin like growth factor)(19). In our case, the laboratory investigations revealed hyponatremia. Serum cortisol level was 11.55 mg/dl. Serum FSH and LH levels were subnormal. Fleckman AM and Otsuka F in their studies they found that the patient MRI brain showed empty sella syndrome which was compatible with Sheehan's syndrome (20,21). In our patient, MRI revealed an empty sella. Increased awareness of this condition among the peoples will helps in earlier diagnosis that lowers morbidity and mortality.

CONCLUSION:

Clinicians should know that hyponatremia might be an initial manifestation of this disease. High index of suspicion for Sheehan's syndrome needed a patient detailed past medical history, clinical examination and laboratory tests for an early diagnosis. Sheehan's syndrome is still a common problem in our country, especially in rural areas. Early diagnosis and proper managements are very important to decrease morbidity and mortality of this syndrome.

REFERENCES:

1. Sheehan HI. Postpartum necrosis of the anterior pituitary. Journal of Pathology and Bacteriology. 1937;45:189-214.
2. Matsuwaki T et al. Evaluation of obstetrical factors related to Sheehan Syndrome. J Obstet Gynecol 2013:22;279-283.
3. Diri H et al. Sheehan Syndrome; new insights into a old disease. Endocrine 2016; 51: 22-31.
4. C. Shivaprasad, (2011). "Sheehan's syndrome: newer advances," Indian Journal of Endocrinology and Metabolism, vol. 15, supplement 3, pp. S203–S207.
5. Gei-Guardia O, Soto-Herrera E, Gei-Brealey A and Chen-Ku CH. Sheehan's syndrome in Costa Rica: Clinical experience with 60 cases. Endocr Pract. 2011;17(3):337-44.
6. B. A. Laway, S. A. Mir, M. I. Bashir, J. R. Bhat, J. Samoon, and A. H. Zargar, (2011). "Prevalence of hematological abnormalities in patients with Sheehan's syndrome: response to replacement of glucocorticoids and thyroxine," Pituitary, vol. 14, no. 1, pp. 39–43.
7. D. Gokalp, A. Tuzcu, M. Bahceci et al., (2011). "Analysis of thrombophilic genetic mutations in patients with Sheehan's syndrome: is thrombophilia responsible for the pathogenesis of Sheehan's syndrome?" Pituitary, vol. 14, no. 2, pp. 168–173.
8. M. Sert, T. Tetiker, S. Kirim, and M. Kocak, (2003). "Clinical report of 28 patients with Sheehan's syndrome," Endocrine Journal, vol. 50, no. 3, pp. 297–301.
9. C. Putterman, Y. Almog, Y. Caraco, D. J. Gross, and E. Ben-Chetrit, "Inappropriate secretion of antidiuretic hormone in Sheehan's syndrome: a rare cause of postpartum hyponatremia," *American Journal of Obstetrics and Gynecology*, vol. 165, pp. 1330–1333, 1991.
10. Singhania P, Singh S, Banerjee R, Singhania B, Banerjee I, Maitra S. Hyponatremia - A rare and emergency presentation of Sheehan's syndrome. Pak J Med Sci. 2010;26(3):713-5.
11. D. Gokalp, G. Alpagat, A. Tuzcu et al., "Four decades without diagnosis: Sheehan's syndrome, a retrospective analysis," Gynecological Endocrinology, vol. 32, pp. 904–907, 2016.
12. Y. Y. Huang, M. K. Ting, B. R. Hsu, and J. S. Tsai, "Demonstration of reserved anterior pituitary function among patients with amenorrhea after postpartum hemorrhage," Gynecological Endocrinology, vol. 14, pp. 99–104, 2000.
13. C. Ratarasarn, R. Rajatanavin, and T. Himathongkam, "Salient clinical features of Sheehan's syndrome," Journal of the Medical Association of Thailand = Chotmaihet thangphaet, vol. 72, pp. 41–47, 1989.
14. P. C. Pham, P. A. Pham, and P. T. Pham, "Sodium and water disturbances in patients with Sheehan's syndrome," American Journal of Kidney Diseases The Official Journal of the National Kidney Foundation, vol. 38, p. E14, 2001.
15. F. Kelestimur, "Sheehan's syndrome," *Pituitary*, vol. 6, pp. 181–188, 2003.
16. R. W. Schrier, "Body water homeostasis: clinical disorders of urinary dilution and concentration," Journal of the American Society of Nephrology Journal of the American Society of Nephrology, vol. 17, pp. 1820–1832, 2006.
17. Gokalp D, Alpagat G, Tuzcu A, Bahceci M, Tuzcu S, Yakut F, et al. Four decades without diagnosis: Sheehan's syndrome, a retrospective analysis. Gynecol Endocrinol. 2016;32(11):904–7.
18. Abere Genetu et al .A 45-year-old female patient with Sheehan's syndrome presenting with imminent adrenal crisis: a case report.Journal of medical case reports.15;229;2021.
19. Khan SA et al. Sheehan's syndrome: a case report. Int J Reprod Contracept Obstet Gynecol. 2016 Sept;5(9):3221-3222.
20. Fleckman AM, Schubart UK, Danziger A, et al. Empty sella of normal size in Sheehan's syndrome. Am J Med 1983;75:585–91.
21. Otsuka F, Kageyama J, Ogura T, et al. Sheehan's syndrome of more than 30 years' duration: an endocrine and MRI study of 6 cases. Endocr J 1998;45:451–58.