

Case report**Postpartum Hemorrhage Complicated by Pan-hypopituitarism: Sheehan's Syndrome; Case Report, 2022****Berhe Tesfai¹, Okbu Frezgi¹, Khalid Hussien²**¹MD, Obstetrics and Gynecology Resident, Orotta College of Medicine and Health Science, Post-Graduate, Ministry of Health, Asmara, Eritrea²MD, Obstetrician and Gynecologist, Associate Professor, Orotta college of Medicine and Health Science, Post-Graduate, Ministry of Health, Asmara, Eritrea**Abstract.**

Background: Sheehan's syndrome is hypopituitarism due to postpartum ischemic necrosis of the pituitary gland. Due to its potentially life-threatening and very rare complications; it still remains important to pregnant women, clinicians, and public health services around the world.

Case Report: 38 years parity three Eritrean mother from Ghone subzone of Gash Barka, Eritrea; had history of postpartum hemorrhage and blood transfusion before 10 years (2011) on her last delivery. She presented with history of failed lactation, amenorrhea and infertility of 10 years. She had also constipation, lethargy and generalized body weakness with her current complaint. Hormone analysis revealed pan-hypopituitarism of most anterior pituitary hormones and Magnetic Resonance Imaging of the brain revealed empty sella. She was started on Levothyroxine and fertility supplements based on her fertility desire, and on subsequent follow-ups, her menses was resumed and constipation had subsided and she was attached to endocrinology department for further follow-up and management.

Conclusion: The history of failed lactation, amenorrhea and infertility following postpartum hemorrhage, hormone analysis and imaging findings were consistent with a rare complication of Sheehan's syndrome. This case was reported for its rarity. Physicians should aware about this complication and should have high index of suspicion for cases with common manifestation of uncommon syndromes.

Key words: Pan hypopituitarism, Sheehan's syndrome, postpartum hemorrhage, Eritrea

Abbreviations

MRI: Magnetic resonance imaging

CBC: complete blood count

TSH: thyroid-stimulating hormone

LH: luteinizing hormone

FSH: follicle-stimulating hormone

Introduction

Sheehan's syndrome is hypopituitarism due to postpartum ischemic necrosis of the pituitary gland and was first described in 1937 by Sheehan [1]. It is rare complication which occurs in 1 out of every 100,000 births globally and is the most common cause of hypopituitarism in low- or middle-income countries [2, 3]. The prevalence is much higher in developing countries, as high as 3.1% in India where more than half of the affected individuals had home deliveries [4].

The underlying process leading to Sheehan's syndrome is the infarction of the physiologically enlarged anterior pituitary lobe and secondary to the compression of the blood vessels supplying the gland by the enlarged gland itself or due to grossly decreased blood supply during intrapartum or postpartum events [5].

The diagnosis of Sheehan's syndrome is determined by the patient's history and physical examination, and confirmed by laboratory tests [6]. Hemorrhagic shock during pregnancy is a key leading point in diagnosis and failure to lactate is often a common initial complaint in patients with Sheehan syndrome [6]. Many of them also report amenorrhea after delivery [7]. The pathophysiology of Sheehan's syndrome has been classically attributed to a transient hypo perfusion that provokes infarction, necrosis, and consequent dysfunction in a physiologically enlarged pituitary gland (due to pregnancy) [8, 9].

Due to its delayed diagnosis, clinical presentation, and potentially life-threatening complications, Sheehan's syndrome still remains important to pregnant women, clinicians, and public health services around the world [10]. Moreover, not every woman who suffers postpartum hemorrhage develops Sheehan's syndrome and when they do, it mani-

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Received: November 18, 2022**Published:** December 14, 2022

Citation: *Berhe Tesfai. Postpartum Hemorrhage Complicated by Pan-hypopituitarism: Sheehan's Syndrome; Case Report, 2022. Int Clin Img and Med Rew. 2022; 3(3): 1126.

feats within a wide spectrum of time, suggesting that there are other factors that influence its appearance. [11–13].

Case Report

A 38 years Gravid 3 Parity 3 Eritrean mother from a subzone called Ghone, Gash Barka Zone, visited Orotta National Maternity Referral Hospital on 27/01/2022. She had history of postpartum hemorrhage and was transfused blood before 10 years during delivery of her third child. At that time, she had history of uterine atony with massive vaginal bleeding, headache, generalized body weakness and altered level of consciousness. Her vaginal bleeding was stopped after she was managed with misoprostol, oxytocin, and transfused with three units of blood in her nearby hospital. Her previous pregnancies were unremarkable and have no history of similar problem.

After the above mentioned management, she gained her consciousness and attempts to breastfeed her neonate as usual of her previous neonates. Unfortunately, lactation was failed and neonate was started on formula feeding. On the following years, she had visited different hospitals for amenorrhea and infertility for about 10 years. She had also constipation, lethargy and generalized body weakness associated with these complaints. She denied any history of head trauma, surgery, radiation exposure, diabetes mellitus and hypertension.

Physical examination was unremarkable on her visit to Orotta National Maternity Referral hospital on 27/01/2022. During her current visit, she was investigated with complete blood count (CBC), hormone analysis, thyroid function test and magnetic resonance imaging (MRI) of brain. The CBC revealed hemoglobin 10.2g/dl, hematocrit of 30.5%, and platelet of 195.65x103/μL. The hormone analysis indicated low level of most anterior pituitary hormones (Table: 1) and the MRI of the brain revealed empty sella (Image: 1). Based on her findings, she was counseled about her condition and started on Levothyroxine, combined oral contraceptive pills and other fertility supplements based on her fertility desire. On subsequent clinical evaluation, constipation had subsided and her menses had resumed and was referred to endocrinology clinic for further follow-up and management.

Table 1: Hormone analysis results, 28/01/2022

Variables	Results	Range of reference
Estradiol	3.41 pg/mL	15-350
FSH	2.91 mIU/mL	26.7–133.4, postmenopausal
LH	1.72 mIU/mL	5.16- 61.99, postmenopausal
Progesterone	<0.05 ng/mL	1.8-24
Prolactin	2.67 ng/ml	5.18-26.5
Testosterone	<2.5 ng/dl	8 - 60
T uptake	1.41 TBI	0.8-1.3
TSH	2.75 μIU/ml	0.27-4.2
T4	2.16 μg/dl	5.13-14.06

TSH: thyroid-stimulating hormone, LH: luteinizing hormone, FSH: follicle-stimulating hormone



Figure 1: Brain magnetic resonance imaging shows an empty sella on different views.

Discussion

One of the most clinical manifestations of Sheehan’s syndrome is failed lactation following postpartum hemorrhage, which this patient had similar to other studies [6, 14, 15]. Failure to lactate is often a common initial complaint in patients with Sheehan syndrome [6]. Even though this could be the first manifestation in most cases with Sheehan syndrome, some cases present with delayed failed lactation which complicated the diagnosis and outcome.

This case revealed amenorrhea associated with her other clinical findings. Many cases also report amenorrhea after delivery [7]. The most common clinical presentation was failure to resume menstruation [15]. This is mainly caused by the pituitary insult and its impaired function following the postpartum hemorrhage. This further complicated her with infertility and became challenging for management.

The hormone analysis displayed pan-hypopituitarism and symptoms consistent with Sheehan’s syndrome. This was similar to other studies that hypothyroidism, hypoprolactinemia and low levels of FSH and LH of the anterior pituitary gland [16]. The growth hormone and cortisol level were not determined (unavailable), but may spectacle further function of the pituitary gland.

The brain MRI of this patient exhibited empty sella which was consistent with most studies that six of seven patients had partially or

completely empty sella on MRI [16]. This imaging finding was steady with the history, clinical presentation and laboratory results of the case, resulted due to the pituitary ischemia following the postpartum hemorrhage.

Even though the presentation of the syndrome was acute; the diagnosis was delayed due to many constrains. This patient had history of failed lactation and amenorrhea following postpartum hemorrhage, which shows acute presentation of Sheehan's syndrome. This was similar to a case report from Ethiopia [14]. The duration of diagnosis for this case was late, about 10 years similar to other studies [10], [15]. The diagnosis can be delayed in some patients if they have partial and delayed clinical presentation and if physicians had no high index of suspicion about the complex presentation of the disease.

Conclusion

The history of postpartum hemorrhage, failed lactation, infertility and amenorrhea, the laboratory and imaging findings were consistent with Sheehan's syndrome. The presentation was acute but the duration of diagnosis was late in this case similar to most studies. This case is reported due to its rare presentation. Health professionals should aware about these unusual complications as their delayed diagnosis may result in devastating complications.

Declarations

Acknowledgments: Authors acknowledge the patient for using her data

Funding: Not applicable

Availability of additional data: Additional data are available from the corresponding author upon request.

Competing of interest: Authors didn't have any conflict of interest to disclose

Ethical approval and consent to participate: Written informed consent was obtained from the patient to present in the case report

Author's contribution: All authors have contributed on data analysis, interpretation and writing

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