Acute Sheehan syndrome following massive postpartum hemorrhage due to vulvar hematoma

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Acute Sheehan syndrome appearing within 6 weeks postpartum is a rare form of hypopituitarism caused by postpartum hemorrhage. A 37-year-old Japanese woman experienced a vulvar hematoma after spontaneous labor at 40 weeks gestation, leading to massive postpartum hemorrhage (estimated total blood loss of 3,000 mL). Despite successful cesarean delivery and hematoma drainage, she presented 28 days postpartum with anorexia, fatigue, and hyponatremia after initial recovery. MRI revealed a swollen pituitary gland with subacute hemorrhage, confirming the diagnosis. Hormonal replacement therapy with levothyroxine, hydrocortisone, estrogen, and progesterone was initiated. This is the first reported case of acute Sheehan syndrome following a vulvar hematoma, a condition typically not considered a risk factor for this syndrome. The case highlights the importance of considering rare complications like acute Sheehan syndrome in patients with massive postpartum hemorrhage, even when the pathogenesis is clinically common, such as vulvar hematomas.

Key words: acute Sheehan syndrome, hypopituitarism, postpartum hemorrhage, vulvar hematoma

The patient was a 37-year-old Japanese woman (gravida 1, para 0). She had no significant medical or family history and no problems during pregnancy after conceiving naturally. Spontaneous onset of labor occurred at 40 0/7 weeks of gestation, and vulvar hematoma on the left side with severe pain acutely developed following the frequent pushing during delivery in the third trimester (Figure 1A). She was urgently transported by ambulance from a midwifery clinic 36 km away to our hospital, which took 30 minutes by ambulance, for treatment. Cesarean section was performed for pain management and prolonged labor, delivering a healthy 2,981-g female infant. Subsequently, the hematoma was surgically drained (Figure 1B); we could not identify bleeding vessels after evacuating the hematoma, which was followed by hemostasis and suturing with interrupted stitches using absorbable 2-0 Vycril rapid (Ethicon Japan, Tokyo, Japan). After suturing the vulvovaginal lacerations, a Penrose drain was inserted to prevent hematoma reformation. The estimated total blood loss was approximately 3,000 mL, including 1,000 mL blood with amniotic fluid during cesarean section and 2,000 mL blood in the vulvar hematoma.

On the first day after delivery, the patient's general condition, blood

FIGURE 1
(A) The left vulva is greatly enlarged due to a hematoma. (B) The left vulva shrinks after surgical hematoma drainage

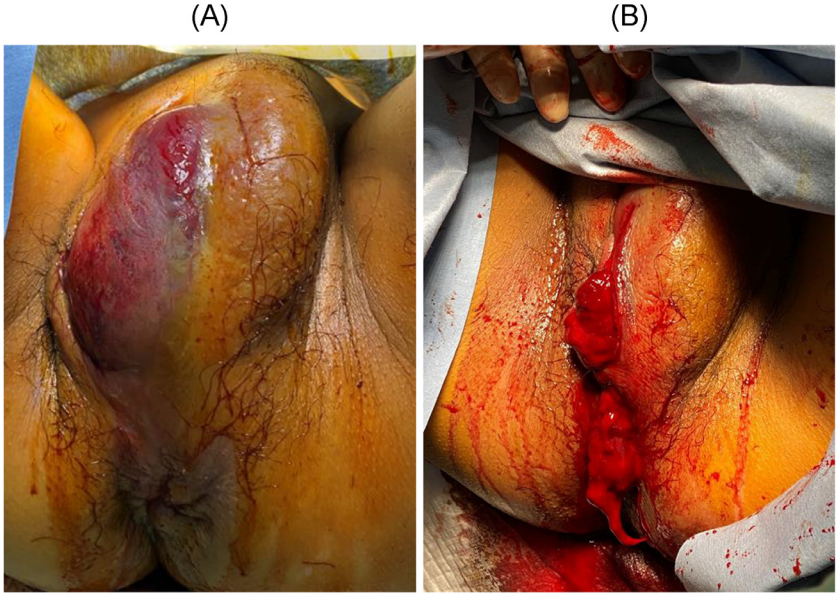

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TABLE
Postpartum hormonal profile

	Normal range	Day 28 postpartum	Day 139 postpartum	Day 332 postpartum
ACTH (pg/mL)	0–60	4.2	1.5	1.8
Cortisol (μg/d)	4.3–20	<0.2	18.8	6.6
TSH (μIU/mL)	0.35–4.80	0.37	0.6	0.3
Free T3 (pg/mL)	2.30–4.00	2.56	2.91	2.78
Free T4 (ng/dL)	0.90–1.70	0.46	0.4	1.09
PRL (ng/mL)	3.50–30.0	11.6	16.6	13
GH (ng/mL)	0.1–2.7	1.45	1.0	0.9
Somatomedin-C (ng/mL)	109–265	53	144	137
FSH (mIU/ml)	0.4–11	NA	5.4	4.56
LH (mIU/mL)	0.08–7.3	NA	3.0	3.14

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pressure, and urine output were normal, although her hemoglobin and fibrinogen levels were 7.1 g/dL and 216 mg/dL, respectively. The patient received a 4,000-mL transfusion during intra- and post-operation comprising 2,000 mL of red blood cells and 1200 mL of fresh frozen plasma each. On the second day after delivery, her hemoglobin and fibrinogen levels were 8.9 g/dL and 338 mg/dL, respectively. Therefore, we didn't add transfusion. On the other hand, the kidney function profile revealed blood urea nitrogen of 12 mg/dl, serum creatinine of 1.36 mg/dl, and uric acid of 5.6 mg/dl, and infusion was continued with 2000 ml/day because kidney function was slightly decreased not including abnormal urinalysis. On the fifth day after delivery, a Penrose drain was removed, and there was no reconstitution of the hematoma on the seventh day after delivery. On day 13 after delivery, the patient was prescribed sodium ferrous citrate 100 mg/day (Ferromia®, Eisai, Tokyo; containing 100 mg of iron in 1.2 g of powder) because her hemoglobin level was 9.7 g/dL and discharged from the hospital since the vulvovaginal wound was clear without bleeding and her kidney functional profile improved to a normal level.

On day 28 after delivery, the patient visited the hospital because of anorexia and fatigue, and laboratory data showed low sodium (Na^+, 116 mEq/L) and chloride (Cl^-, 84 mEq/L) levels. We immediately administered a NaCl infusion to compensate for the Na^+ level by 10 mEq/day. Sheehan syndrome was suspected, and other tests were performed, including pituitary gland magnetic resonance imaging (MRI). Table lists the patient's hormonal levels. T1-weighted MRI showed a swollen pituitary gland with subacute hemorrhage (Figure 2A). The MRI results did not indicate lymphocytic hypophysitis as a differential disease; thus, the most probable diagnosis was acute Sheehan syndrome, which we confirmed.

Initially, the adrenal insufficiency was treated via intravenous administration of hydrocortisone (20 mg), followed by oral administration of levothyroxine (50 μg/day) and hydrocortisone (20 mg/day), which improved their condition (Figure 3). At 105 days postpartum, repeat MRI demonstrated an atrophic pituitary gland (Figure 2B) with no clinical findings of neurologic compromise. Furthermore, the patient had pituitary

FIGURE 2
(A) Sagittal T1-weighted magnetic resonance imaging shows a swollen pituitary gland with subacute hemorrhage on day 28 postpartum (white arrow) and (B) an atrophic pituitary gland on day 105 postpartum (white arrow)

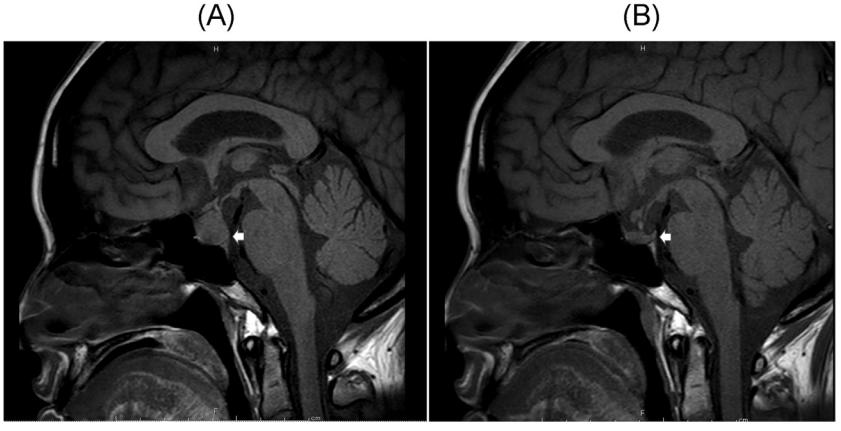

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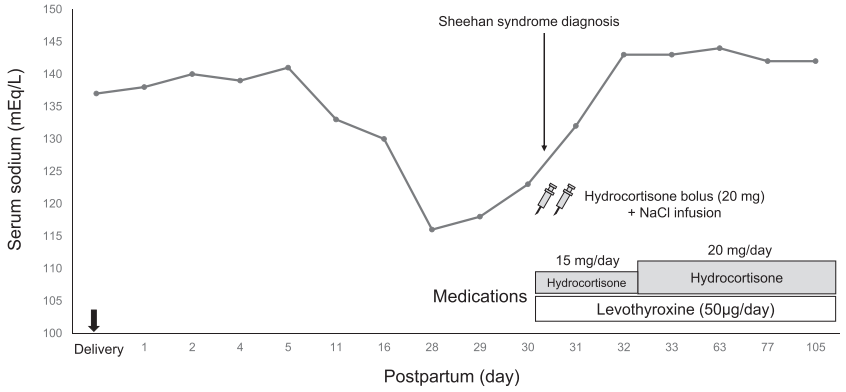
FIGURE 3
Temporal change in serum sodium following intervention with hormone therapy and NaCl infusion



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amenorrhea and required hormonal replacement therapy with estrogen and progesterone, which must be continued until the perimenopausal age.

Sheehan syndrome, an endocrine disorder characterized by hypopituitarism, is caused by pituitary gland collapse following massive postpartum hemorrhage due to uterine atony, amniotic fluid embolism, retained placenta, uterine rupture, and disseminated intravascular coagulation.[1] Some reports indicate that the duration between the occurrence of symptoms and postpartum hemorrhage in Sheehan syndrome could be several years.[1,2] On the other hand, Sheehan syndrome appearing within 6 weeks postpartum is considered an acute and rare pathogenetic syndrome. These patients experience adrenal insufficiency, diabetes insipidus, hypothyroidism, and panhypopituitarism following delivery.[3] Our patient presented to the hospital immediately after discharge with complaints of anorexia and fatigue and was subsequently diagnosed with hyponatremia. Eventually, they were diagnosed with acute Sheehan syndrome with central hypothyroidism, secondary hypoadrenalism, central enuresis, growth hormone deficiency, and pituitary hypogonadism. Matsuzaki et al.[3] recently reviewed the literature on acute Sheehan syndrome, reporting that uterine bleeding, such as uterine atony or retained placenta, is a strong causative factor for acute Sheehan syndrome. However, vulvar hematoma is not a risk factor for Sheehan syndrome following postpartum hemorrhage.

Postpartum hematomas, especially those that form in the upper vaginal wall or within the retroperitoneal space, can cause more bleeding than expected, or "hidden bleeding," and can induce maternal hemorrhagic shock.[4] In this case, vulvar hematomas localize in the space of the urogenital diaphragm, and the levator ani muscle is susceptible due to wounds at the bleeding point in the descending branch of the uterine artery; therefore, it can be recognized as a fluctuant and intensely painful swelling (on palpation) protruding from the vulva.[5] However, to the best of our knowledge, no reports have shown maternal massive hemorrhage from vulvar hematomas derived from vulvar hematoma and resulting in Sheehan syndrome. Therefore, this is the first report of a postpartum hematoma in the vulvar area leading to Sheehan syndrome.

Although vulvovaginal hematomas sufficiently predict Sheehan syndrome development in rare conditions with massive hemorrhage, such as placenta previa or accreta, they are frequently observed in clinical practice and represent a common pitfall for obstetricians in diagnosing Sheehan syndrome. ■

CRediT authorship contribution statement

Wataru Saito: Data curation, Conceptualization. **Kuniaki Ota:** Writing – original draft, Conceptualization. **Toshifumi Takahashi:** Writing – review & editing. **Mika Sugihara:** Methodology, Investigation. **Takehiko Matsuyama:** Visualization, Supervision. **Yoshiaki Ota:** Supervision, Resources, Project administration. **Koichiro Shimoya:** Writing – review & editing, Supervision. ■

TWEETABLE STATEMENT

Vulvovaginal hematomas are a pitfall for obstetricians in diagnosing Sheehan syndrome.

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