

# Headache after Delivery, a Misleading Presentation of Sheehan's Syndrome: A Case Report

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## Abstract

Post dural puncture is one of the most common causes of postpartum headache. Other causes are benign primary headache disorders (migraine, tension-type headache) and secondary headache disorders such as stroke, ischaemia or pituitary apoplexy and venous sinus thrombosis. Headache in the new mother after delivery, mandates a differential diagnosis because it is not appropriate to focus only on the most common diseases, such as post dural puncture headache. In this report we discuss a case of postpartum headache with delayed cause diagnosis in complicated labour and delivery.

**Keywords:** Sheehan's syndrome; Post delivery headache; Dural puncture; Delivery; Oxytocin

## Introduction

Post dural puncture is one of the most common causes of postpartum headache. Other causes are benign primary headache disorders (migraine, tension-type headache) and secondary headache disorders such as stroke, ischaemia or pituitary apoplexy and venous sinus thrombosis [1]. In this report we discuss a case of postpartum headache with delayed cause diagnosis in complicated labour and delivery.

## Case Report

We report the case of a 35-y-old female, admitted to our ICU 5 days after delivery for agitation, headache, nausea, vomiting, anisocoria, photophobia, polyuria and hypertension.

Because of failure of progression in the birth canal, after repeated and unsuccessful attempts of spinal anaesthesia, delivery was done under general anaesthesia. Before Caesarean delivery, the haemoglobin was 12.4 mg/dL, and coagulation pattern and platelets count were normal. Delivery was affected by inadequate uterine contraction, which required a supplemental dose of sulprostone (0.1 mg IV), in addition to our institutional dose (20 IU) of oxytocin. For concomitant blood loss, hypotension and vaginal dripping, vasopressors were used and, at the end of surgery, a Bakri balloon was inserted. The patient had been awakened from general anaesthesia and monitored in the recovery room. Two hours after Caesarean the patient underwent a laparotomic surgery because of the persistence of haemorrhage (Table 1). A cervix uterine laceration was observed and a local suture was applied. She received 1 packed red blood cells (PRBC). At the end of surgery, she was transferred to the recovery room, where she received 2 more PRBC, 600 ml of fresh frozen plasma and 2000 IU of antithrombin. Oxytocin infusion was started. After haemodynamic stabilisation the patient was transferred to a gynaecology ward with a restored coagulation pattern (Table 1) and the vaginal packing in place. The next day the packing was removed, no blood loss was observed, laboratory and haemodynamic parameters were stables, urine output was normal without pharmacological stimulation.

On the 3<sup>rd</sup> post-operative day the patient developed headache, nausea and vomiting without cervical rigidity or photophobia. The anaesthesiologist in charge rejected the hypothesis of post dural puncture headache despite a consistent number of attempts of spinal anaesthesia being performed. Furthermore, a severe hyponatraemia (115 mmol/l) was found. The neurologist consulted and revealed just a cognitive decay, probably caused by hyponatraemia; no other

pathological signs were found. Natraemia remained low despite the infusion of normal saline in the gynaecology ward. No lactation onset was observed and the patient was unable to breast-feed the child.

On the 5<sup>th</sup> day after delivery she developed an alteration of mental status, photophobia and oliguria and she was transferred from the department of gynaecology to the intensive care unit.

At the ICU admission the neurological evaluation showed a mental slowing probably related to metabolic disorders. The lab test showed: hyponatraemia, hypokalaemia, hypomagnesaemia, hypochloraemia, metabolic acidosis with compensatory respiratory alkalosis. Pituitary apoplexy was considered and a CT scan was performed. The tomography showed cerebral oedema and no ischemia or cerebral mass was found. Hypertonic saline and parenteral nutrition were started, hypokalaemia and hypomagnesaemia were corrected. Hypertension was detected and alpha-methyl dopa was started. The following day an MRI showed an enlargement of adenohypophysis without haemorrhage and no compression of the optic chiasm; neurohypophysis was normal. No cerebral oedema was confirmed. The cardiac transthoracic echography showed a normal cardiac function and contractility. During the ICU stay, the lab tests showed a reduction of cortisol, hypoprolactinaemia (abnormally low, given the recent birth) and reduction of gonadotropins. These values were compatible with Sheehan's syndrome (SS) with secondary adrenal insufficiency and associated syndrome of inappropriate antidiuretic hormone secretion (SIADH). Therapy with hydrocortisone and levothyroxine was started leading to a normalisation of all the metabolic values.

On the 8<sup>th</sup> post-operative day the patient was retransferred to the gynaecology ward. The cerebral MRI was repeated a few days later and confirmed the adenohypophysis apoplexy, with normal neurohypophysis. On the 10<sup>th</sup> post-operative day she was discharged to home with hormone replacement therapy and made a complete recovery.

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TIME	Before delivery	1 h PD	2 h PD	4 h PD	9 h PD	11 h PD	15 h PD	24 h PD
Hb (g*dL <sup>-1</sup> )	12.4	9.8	8.2	8.8	7.5	8.6	8.6	7.8
PLT (1000*dL <sup>-1</sup> )	232	196		159	151	154	156	165
aPTT ratio	0.78	0.67		0.9	0.8	0.83	0.84	
INR	0.87	0.97		1.15	1.02	1.04	1.01	
Fibrinogen (mg*dL <sup>-1</sup> )	296	251		155	223	247	319	
AT (% of activity)	75	58		33	58	115	95	
Na (mmol*L <sup>-1</sup> )			132	140	135	135	136	134
D-dimer				2507		1214		
Lactate (mmol*L <sup>-1</sup> )			2.11		2.09			

Note: PD: post delivery; Hb= Hemoglobin; PLT: platelets; AT:Antitrombin; Na: plasma sodium.

Table 1: Table shown laboratory test performed during the hospital stay, after delivery.

## Discussion

Headache affects 4 in 10 women are within one week after delivery [2].

The most common cause is tension (39%) followed by pre-eclampsia or eclampsia (24%), post dural puncture headache (16%), migraines (11%), and haemorrhage, thrombosis or vasculopathy (10%) [3].

The percentage of postpartum headache due to SS and the incidence of SS in developed countries is not clear; the prevalence of hypopituitarism in the adult Caucasian population in Spain is 29-45.5/100,000 without sex differences and 6-10% of those cases (male and female) are due to Sheehan's syndrome [4].

Sheehan's syndrome is a rare complication of pregnancy, consisting of the necrosis of the pituitary gland. Usually it occurs after excessive blood loss or extremely low blood pressure during or after childbirth. These factors can be particularly damaging to the pituitary gland, which enlarges during pregnancy, destroying hormone producing tissue so that the gland can't function normally. The presence of disseminated intravascular coagulation (i.e. in amniotic fluid embolism or HELLP syndrome) also appears to be a co-factor in its development. The anterior pituitary is supplied by a low-pressure portal venous system [5]. These vulnerabilities, when affected by major haemorrhage or hypotension during the peripartum period, can result in ischaemia of the anterior pituitary regions leading to necrosis. The posterior pituitary region is usually not affected because it has a direct arterial supply.

The necrosis of the pituitary gland can lead to panhypopituitarism or to only selective

pituitary deficiencies depending on the extent of the ischaemic damage. Headache is not the most common presentation of SS but mental disturbances are frequent, including psychosis. Failure to lactate and breast involution are the earliest signs in the completed form because of prolactin deficiency. At a later time symptoms of hypothyroidism and hypoadrenalism occur and the persisting of amenorrhoea becomes evident. Pituitary failure is usually suggested by laboratory tests revealing low free thyroxine, oestradiol and cortisol levels, as well as with low or inappropriately normal thyroid-stimulating hormone, follicle-stimulating hormone, luteinising hormone, and ACTH (adrenocorticotrophic hormone) levels.

The symptoms revert when SS is treated with hormone replacement, which may include glucocorticoids, levothyroxine, pulsatile GnRH (gonadotropin-releasing hormone) or exogenous gonadotropins for ovulation induction, growth hormone [6,7].

Hypopituitarism can lead to hyponatraemia but this is rarely the acute presentation of SS. There are several hypotheses on the mechanisms which cause hyponatraemia in SS. Both hypothyroidism

and glucocorticoid deficiency can cause decreased free water clearance and therefore hyponatremia. Another possible mechanism is the inappropriate secretion of the antidiuretic hormone that occurs in cases of adrenocorticotropin deficiency [8,9]. Hyponatraemia may be euvolemic, hypovolemic or hypervolemic and the treatment depends on the type of hyponatraemia. Hyponatraemia related to SS is usually euvolemic. Acute hyponatraemia induces cerebral oedema that leads to neurological symptoms such as seizures, impaired mental status or coma and death [10,11].

In our patient headache was the presentation of SS. Psychomotor agitation and photophobia arose two days later as signs of hyponatraemia and the CT scan confirmed the cerebral oedema. Both headache and hyponatraemia are not usual in the early presentation of SS.

Lack of breastfeeding, associated with clinical symptoms, led clinicians to suppose acute pituitary damage.

The development of cerebral oedema had been aggravated by the use of uterotonic. In this patient, oxytocin and sulprostone were given at high dose for the treatment of severe atonic-post-partum haemorrhage; they may have contributed to the development of acute hypotonic hyponatraemia [12,13].

Hypotension, bradycardia and other possible side effects of sulprostone therapy were transient in our patient and were quickly corrected with fluid infusion and vasopressors. Sulprostone doesn't interfere with the pituitary gland. In this case, furosemide had complicated an already precarious metabolic status, worsening hyponatraemia and inducing a polyuria. In Sheehan's syndrome, usually, potassium is normal [14]. Adrenal aldosterone production is not dependent on the pituitary gland. The transient hypokalaemia observed in our patient could be related to diuresis induced by furosemide.

Headache in the new mother after delivery mandates a complete differential diagnosis and it is not appropriate to focus only on the most common diseases.

It is mandatory not to minimise the problem, since this can lead to not considering life-threatening diseases and may lead to a delay on life-saving therapies.

## References

1. Klein AM, Loder E (2010) Postpartum headache. *Int J Obstet Anesth* 19: 422-430.
2. Goldszmidt E, Kern R, Chaput A, Macarthur A (2005) The incidence and etiology of postpartum headaches: a prospective cohort study. *Can J Anaesth* 52: 971-977.
3. Stella CL, Jodicke CD, How HY, Harkness UF, Sibai BM (2007) Postpartum headache: is your work-up complete? *Am J Obstet Gynecol* 196: 318.e1-7.

4. Regal M, Paramo C, Sierra SM, Garcia-Mayor RV (2001) Prevalence and incidence of hypopituitarism in an adult Caucasian population in northwestern Spain. *Clin Endocrinol* 55: 735-740.
5. <http://www.ncbi.nlm.nih.gov/bookshelf/br.fcgi?book=endocrin&part=A1257> under heading Sheehan's syndrome.
6. DiZerega G, Kletzky OA, Mishell DR (1978) Diagnosis of Sheehan's syndrome using sequential pituitary stimulation tests. *Am J Obstet Gynecol* 132: 348-353.
7. Ozbey N, Inanc S, Aral F (1994) Clinical and laboratory evaluation of 40 patients with Sheehan's syndrome. *Isr J Med Sci* 30: 826-829.
8. Boulanger E, Pagniez D, Roueff S (1999) Sheehan syndrome presenting as early post-partum hyponatremia. *Nephrol Dial Transplant* 14: 2714-2725.
9. Putterman C, Almog Y, Caraco Y, Gross DJ, Ben-Chetrit E (1991) Inappropriate secretion of antidiuretic hormone in Sheehan's syndrome: a rare cause of postpartum hyponatremia. *Am J Obstet Gynecol* 165: 1330-1333.
10. Manisha Sahay, Rakesh Sahay (2014) Hyponatremia: A practical approach. *Indian J Endocrinol Metab* 18: 760-771.
11. Verbalis JG, Goldsmith SR, Greenberg A, Korzelius C, Schrier RW, et al. (2013) Diagnosis, evaluation and treatment of hyponatremia: Expert panel recommendations. *Am J Med* 126(Suppl 10): S1-42.
12. Spasovski G, Vanholder R, Allolio B, Annane D, Ball S, et al. (2014) Clinical practice guideline on diagnosis and treatment of hyponatraemia. *Eur J Endocrinol* 170: G1-47.
13. Schmitz T, Tararbit K, Dupont C, Rudigoz RC, Bouvier-Colle MH, et al. (2011) Prostaglandin E2 analogue sulprostone for treatment of atonic postpartum hemorrhage. *Obstet Gynecol* 118: 257-265.
14. Schrage S, Sabo L (2001) Sheehan syndrome: a rare complication of postpartum hemorrhage. *J Am Board Fam Pract* 14: 389-391.