—Short Communication—

Diabetes Insipidus Presenting with Oligohydramnios and Polyuria During Pregnancy

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We report a case of subclinical central diabetes insipidus (DI), due to Rathke's cleft cysts, that was initially misdiagnosed as transient DI of pregnancy because it presented in the third trimester of pregnancy. A 37-year-old primigravida visited the Department of Obstetrics in the 30th week of gestation due to polyuria. She was admitted due to oligohydramnios; the amniotic fluid index was 3.24. A vasopressin challenge test was performed and her urine osmolality increased by >100% from baseline after the administration of desmopressin. Because central DI or transient DI of pregnancy was suspected, we prescribed her a desmopressin nasal spray. She gave birth to a relatively healthy baby at 37 weeks and 4 days of gestation. Several months after delivery, discontinuation of desmopressin resulted in recurrence of her polyuria. Magnetic resonance imaging of her brain revealed Rathke's cleft cysts, and finally central DI was diagnosed. (J Nippon Med Sch 2018; 85: 191–193)

Key words: diabetes insipidus, pregnancy, Rathke's cleft cyst

Introduction

Diabetes insipidus (DI) is an uncommon complication in pregnancy with an incidence of approximately 1 in 30,000 pregnancies[1]. DI most classically presents with polyuria and polydipsia. It occurs due to loss of the kidneys'ability to concentrate urine and the resulting aquaresis. It is very important to diagnose and treat DI quickly because it can cause changes in maternal volume status and osmotic homeostasis as well as affecting the health of the fetus. Here, we present a rare case of subclinical central DI due to Rathke's cleft cysts that presented as polyuria and oligohydramnios in the third trimester of pregnancy.

Case Report

A 37-year-old woman presented to the Department of Obstetrics with polyuria in the 30th week of her first pregnancy. She was admitted to the Department of Obstetrics due to oligohydramnios; the amniotic fluid index measured at admission using the 4-quadrant technique was 3.24, which was significantly lower than the reference range (8–18). She did not have any underlying disease or history of medication use and reported no abnormal findings in previous antenatal visits in another obstetric hospital. She had complained of increased thirst a few weeks earlier, and her daily urine volume measured in the ward was approximately 5–7 L. On the day of consultation, her laboratory data were as follows: serum sodium, 146 mEq/L; serum osmolality, 336 mOsm/kg; urine osmolality, 126 mOsm/kg; blood urea nitrogen, 8.1 mg/dL; serum creatinine, 0.76 mg/dL.

During the vasopressin challenge test, her urine osmolality increased by >100% from baseline 30 minutes after the administration of desmopressin, and the effect persisted for up to 2 hours. Based on this, our diagnosis was central DI or transient DI of pregnancy. Since she refused to undergo immediate brain magnetic resonance imaging (MRI), we decided to continue cautiously with desmopressin nasal spray. Initially, 10 μg of desmopressin was sprayed twice a day, and desmopressin use was adjusted based on urine volume and serum sodium level. Finally, with 10 μg of desmopressin 4 times a day, her daily urine

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Table 1 Results of the vasopressin challenge test

	1st test (gestational age 30+3 weeks)		2nd test (postpartum 4 months)	
	Serum osmolality (mOsm/kg)	Urine osmolality (mOsm/kg)	Serum osmolality (mOsm/kg)	Urine osmolality (mOsm/kg)
Baseline	304	130	295	110
30 minutes	303	360	290	197
60 minutes	300	325	301	282
90 minutes	295	413	293	357
120 minutes	293	367	300	373

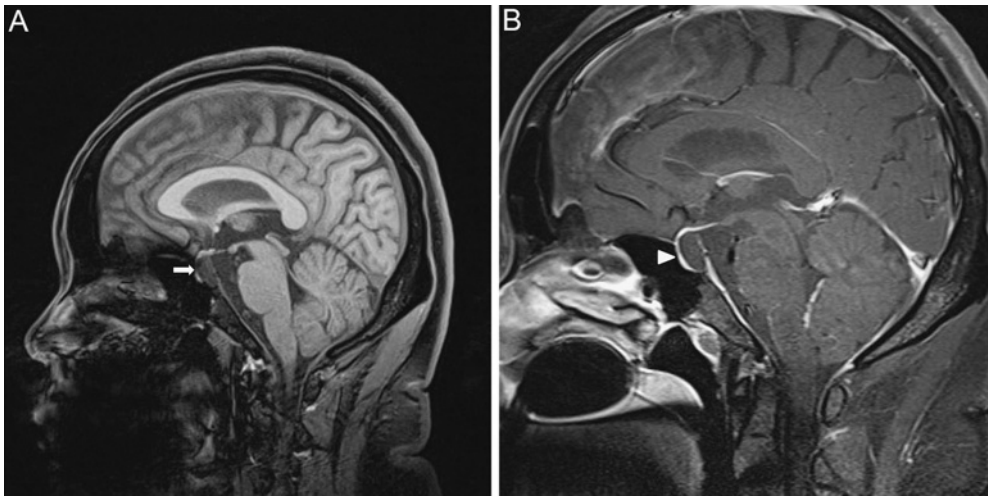

Fig. 1 (A) A sagittal T1-weighted magnetic resonance (MR) image showing a 1.2×0.9×1.4 cm sized well-defined low signal intensity sellar lesion in the posterior aspect of the pituitary gland (white arrow). (B) A gadolinum-enhanced T1-weighted image showing no contrast in the cyst and a thin enhancing rim of surrounding compressed pituitary tissue (arrow head). Rathke's cleft cyst.

volume was maintained at approximately 1−2 L without hyponatremia until delivery. The amniotic fluid index measured just before delivery was 9.1, which was considerably better than that seen at the time of admission. She gave birth via cesarean section to a 2,680 g baby with an Apgar score of 10 points at 37 weeks and 4 days of gestation.

Her urine volume and serum sodium level were monitored after delivery, desmopressin was gradually decreased, and its use was discontinued 5 weeks after delivery. However, the patient complained of increased urinary frequency at 4 months postpartum, due to which we performed the vasopressin challenge test once again. The test results suggested central DI (**Table 1**), and an MRI of the brain was performed to determine the cause. A 1.2×0.9×1.4 cm sized Rathke's cleft cyst was detected on the MRI. Based on this, she was diagnosed with central DI due to the Rathke's cleft cyst and was advised to continue using the desmopressin spray. At present, she has no health-related problems.

Discussion

Central DI in pregnancy is a relatively uncommon complication of pregnancy[1]. The diagnosis of central DI may be known prior to pregnancy; however, in rare cases, pregnancy can unmask subclinical central DI. Placental vasopressinase is a cysteine aminopeptidase which inactivates vasopressin, oxytocin, and other small peptides[2]. Vasopressinase is produced from the 7th gestational week by trophoblasts, increasing 1,000-fold between the 7th and 40th weeks and reaching maximal levels in the third trimester as the placental volume increases[2]. Increased clearance of endogenous vasopressin by excessive vasopressinase activity in placental trophoblasts during pregnancy further decreases the already low vasopressin levels as the gestation progresses[3]. DI of pregnancy is treated using desmopressin, a synthetic form of vasopressin, which is known to be more resistant to vasopressinase than endogenous vasopressin. If the patient's oligohydramnios had been untreated, it could have had a serious adverse effect on the fetus' health. Thus, desmo-

pressin was administered, which successfully maintained the pregnancy and enabled a near-term delivery with minimal complications.

Rathke's cleft cysts, remnants of the embryonic Rathke's pouch, are usually asymptomatic. DI with symptomatic Rathke's cleft cysts have been found in 0–19% of patients[4]. We could find only two previous cases of DI caused by a Rathke's cleft cyst that presented during pregnancy[5,6]. To the best of our knowledge, this is a unique case of subclinical central DI due to a Rathke's cleft cyst that presented as polyuria and oligohydramnios during pregnancy.

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Conflict of Interest: None declared

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