Pregnancy complicated by idiopathic central diabetes insipidus and oligohydramnios

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Ethics Committee Approval

This study was approved by the Ethics Committee of Iwase General Hospital (#191103).

ABSTRACT

Maternal diabetes insipidus does not usually adversely affect the course of pregnancy. We present a rare case of central diabetes insipidus diagnosed at 31 weeks of gestation with fetal oligohydramnios successfully treated with intranasal desmopressin. To date, only three cases of diabetes insipidus with oligohydramnios have been reported.

KEY CLINICAL MESSAGE

This is the fourth case reported worldwide of central diabetes insipidus (cDI) with oligohydramnios. In general, cDI does not adversely affect pregnancy, it can present with oligohydramnios, but this situation can be improved by treatment of cDI.

INTRODUCTION

Central diabetes insipidus is a rare endocrine disease with predominant symptoms of polydipsia and polyuria. It has been shown that central diabetes insipidus does not affect the course of pregnancy; however, a few cases complicated by oligohydramnios have been reported. Herein, we report a case of central diabetes insipidus with oligohydramnios, the fourth of such case ever reported.

CASE PRESENTATION

Case history and examination

The patient was a 30-year-old primigravida who was 168 cm tall and weighed 50 kg, without a significant medical history. She was not taking any medication, and there was no significant family history. There were no high-risk social factors.

She was managed at a nearby maternity clinic from the start of her pregnancy. The pregnancy progressed without any particular problems; however, at 31 weeks and 4 days of gestation, an amniotic fluid index (AFI) of 0 was identified during a prenatal check-up. The biophysical

profile of the fetus was favorable except for oligohydramnios. The patient was hospitalized at the clinic and hydrated with 2,000 mL/day intravenously to increase her amniotic fluid volume.

Additionally, the medical staff noted that she drank large amounts of water (4,000–6,000 mL/day) during hospitalization and had experienced severe thirst before her pregnancy. This symptom is indicative of an endocrine disorder; hence, she was referred to our institution and hospitalized at 32 weeks of gestation.

Investigations

We identified polydipsia and hypotonic polyuria using urine specific gravity (1.001), urine osmolarity (51 mOsm/kgH_20), and serum osmolarity (281 mOsm/kgH_20). The estimated fetal weight and amniotic fluid volume were 2,107 g (+1.39 standard deviation) and AFI 4, respectively. Findings from a plain head magnetic resonance imaging scan were consistent with central diabetes insipidus, whereas findings were unclear regarding the high signal of the posterior pituitary gland in a T1-weighted image. We carefully conducted a water deprivation test at 32 weeks and 4 days of pregnancy while monitoring the fetus and mother; this led to excessive thirst within 2 hours and rapid increase of serum Na level to 149 mEq/L, with no increase in urine osmolarity. The test was then immediately discontinued.

Differential diagnosis and treatment

This case is novel in that the diagnosis of diabetes insipidus was made on the basis of oligohydramnios. Oligohydramnios is usually associated with fetal diseases such as fetal circulatory failure and renal disease when preterm prelabor rupture of membranes is excluded, and with maternal causes such as hypertensive disorders during pregnancy and the use of certain medications. Amniotic fluid volume is a crucial factor in assessing the wellbeing of the fetus; if severe oligohydramnios is observed, as in this case, we need to consider whether early delivery of the fetus is warranted. In this case, careful observation

determined that immediate delivery of the fetus was not necessary, and maternal hydration, which is known to be effective against isolated oligohydramnios, was instituted.[1] Hospitalization for this purpose led to the diagnosis of diabetes insipidus in the patient. In cases of suspected diabetes insipidus, it is critical to differentiate between psychogenic polydipsia, central diabetes insipidus, renal diabetes insipidus, and transient diabetes insipidus of pregnancy.

Results were consistent with idiopathic central diabetes insipidus; thus, we started transnasal administration of desmopressin. We noted reduced water intake and urinary volume after the initiation of desmopressin, along with increased amniotic fluid volume (AFI 12). The patient was discharged at 33 weeks and 6 days of pregnancy, and the subsequent amniotic fluid volume was well maintained (AFI 9-16). She had a spontaneous rupture of the membrane at 38 weeks and 5 days of gestation and had vaginal delivery of a baby boy (3,434 g) . The infant had an umbilical cord blood gas of pH 7.380 and an Apgar score of 8 points at 1 minute and 10 points at 5 minutes. The infant remained in good condition.

Outcome and follow-up

A magnetic resonance imaging scan of the patient's head taken during her 1-month postpartum check-up showed similar results to that taken during pregnancy; thus, it was suggested that central diabetes insipidus may have existed even before the pregnancy. At the time of this article's publication, the patient had continued to receive desmopressin treatment at a local institution for about 8 months.

DISCUSSION

Although cases of gestational diabetes insipidus with oligohydramnios, such as the present case, are rare, they have been recorded in the literature, and three cases have demonstrated improvement in amniotic fluid volume with desmopressin treatment.[2-4] Table 1 shows the

treatment course for each case. The mechanism as to how diabetes insipidus reduces amniotic fluid volume remains unknown. However, in a report on the relationship between changes in maternal and fetal plasma osmolality, it was described that the increase in maternal plasma osmolality was accompanied by an increase in fetal plasma osmolality. This suggests that fetal urine production may be affected by changes in plasma osmolality.[5,6] This potentially play a role in suppressing fetal urine production. In terms of water movement through fetal membranes, it has been reported that membrane permeability increases with gestational age and that membrane water flow can be changed by both hydrostatic and osmotic forces[7]. Although there have been studies on the increased permeability of amniotic fluid to the maternal side due to overexpression of aquaporin 1 and 3, there have been no studies specific to diabetes insipidus, and further studies are needed[8,9]. Although the majority of previous studies suggest that diabetes insipidus does not adversely affect the course of pregnancy, this report increases the number of cases of oligohydramnios associated with diabetes insipidus, and should contribute to future studies on pregnancies with this condition [10-12].
Oligohydramnios may be used to diagnose central diabetes insipidus, as was shown in the present case. Moreover, we demonstrated that oligohydramnios with central diabetes insipidus may be treated by the administration of desmopressin.

AUTHORSHIP

Karin Imaizumi: Writing the paper

Shun Yasuda: Corresponding author, Idea or design of the study

Naoya Toba, Takayuki Okazaki Toma Fukuda, Tsuyoshi Murata, Aya Kanno,Hyo Kyozuka, Makiho Ishibashi, Fumihiro Ito: Critical revision of the paper

Akiko Yamaguchi, Keiya Fujimori: Approval of the final draft

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CONFLICT OF INTEREST STATEMENT

The authors report no conflicts of interest.

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TABLE 1. Cases of diabetes insipidus that presented with oligohydramnios

Author	Year	Age	Parity	Medical history	Symptoms	Cause	Delivery weeks/ days	Delivery	**Clinical course**
Hanson RS et al.[2]	1997	14	0	None	Uterine contraction, genital bleeding, edema	Pituitary dysplasia	38w	Transvaginal	Good
Park JW et al.[3]	2017	30	0	None	Anorexia	Idiopathic central DI†	37w	Caesarean section	Good
Choi HS et al.[4]	2018	37	0	None	Thirstiness, polyuria	Rathke's cleft cyst	37w 4d	Caesarean section	Good
Imaizumi K et al.		30	0	None	Thirstiness, polyuria	Idiopathic central DI†	38w 5d	Transvaginal	Good

† diabetes insipidus