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Case report/ Kazuistyka

Central diabetes insipidus in neonate born at 24 weeks of pregnancy – Case report and review of literature

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ABSTRACT

Background: Central diabetes insipidus is a condition associated with the dysfunction of hypothalamic-pituitary axis and, consequently impaired synthesis and secretion of vasopressin. This condition is rarely found in preterm neonates which is why we decided to report it. Also, there are no guidelines on how to treat this condition in neonates, so it is important to share our experience and provide information on the therapies we used therapies and their results. **Case presentation:** In our paper we describe a case of a male infant born at 24 weeks of gestation, who suffered an intraventricular hemorrhage of grade IV severity. The hemorrhage caused damage to the posterior pituitary lobe which in turn resulted in central diabetes insipidus. Oral desmopressin therapy was administered. **Conclusion:** In our case we achieved positive clinical results – normalization of diuresis and natremia. It shows that oral desmopressin can be taken into consideration for treatment of diabetes insipidus in preterm neonates. This case report can be useful for both neonatologists and endocrinologists. It is also important to compare our case with similar cases to expand our knowledge about this rare disease in neonates and to find possible therapies.

Background

Total and extracellular water accounts for a high percentage of the newborn's body weight. It is very important that water-electrolyte balance be properly maintained, especially in preterm infants. Processing of liquids in the body is influenced by many factors: fluid delivery, gestational age, ambient temperature and humidity, body weight, mechanical ventilation, as well as others. Other regulating factors are those of a hormonal nature. The primary hormone with direct impact on diuresis is vasopressin. Disorders related to this hormone may result either from inadequate secretion of vasopressin (central diabetes insipidus, CDI) or from impaired responsiveness of renal tubules to this hormone (nephrogenic diabetes insipidus). CDI, a condition rarely present in newborn infants, is caused by deficient secretion of vasopressin by the posterior lobe of the pituitary gland, causing an impairment of urine concentrating ability. In terms of clinical symptoms, it manifests itself by polyuria,

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low urine osmolality and hypernatremia. The development of CDI may be associated with a variety of disease classification units – it may be idiopathic, caused by genetic predispositions, or may follow a trauma or tumor [1].

In the patient, whose case is described in this paper, CDI was caused by an injury to the pars nervosa of the pituitary gland, resulting from grade IV intraventricular hemorrhage (according to Papille grading criteria). Oral desmopressin (a synthetic analog of vasopressin) therapy was administered and a positive clinical result was obtained. There are no published guidelines on how to treat this condition in neonates, so it is important to share experience our and provide information on therapies we used and their results.

Case presentation

A male neonate, born to a G5, P2 mother was delivered vaginally in breech position at 24 weeks of gestational age, with 790 g body weight at birth. The infant's Apgar score was 1, 1, 1 and 3 at 1, 3, 5 and 10 min of life, respectively. Umbilical cord pH was 7.33 (−3.2 mEq/l) and 7.0 (−9.7 mEq/l). The baby was intubated in the labor ward and artificially ventilated. External cardiac massage was performed, as well as 4 doses of adrenalin. Following resuscitation, the patient was admitted to NICU (Neonatal Intensive Care Unit) of the Neonatology Clinic in Poznan. Because of respiratory distress SIMV (Synchronized intermittent mandatory ventilation) mechanical ventilation was administered. Based on an X-ray, symptoms of grade III respiratory distress syndrome as well as inflammation of the lungs were diagnosed. In his first hour of life the patient received surfactant (Survanta by AbbVie; dosage 4 ml/kg bw); empirical therapy using broad-spectrum antibiotics was also applied: ampicillin at 50 mg/kg/every 12 h and gentamicin at 5 mg/kg every 48 h, and this line of therapy continued until day 5 of postnatal life. Afterwards a targeted therapy was used (meropenem at 20 mg/kg/every 8 h and vancomycin at 10 mg/kg/every 18 h) because of a positive colony of methicillin-resistant Staphylococcus haemolyticus (MRS) in bronchial content up to 20 day of postnatal life. At day 27 of postnatal life, a course of steroid therapy was administered for prevention of bronchopulmonary dysplasia. Conventional ventilation was provided until day 45 of life. Afterwards, noninvasive ventilation support (NIV) was used and, finally, ended at day 101 of life. The postnatal period was also complicated by a ruptured spleen and bleeding into the peritoneal cavity (managed conservatively due to the very serious condition of the patient).

At 2 months of life hypernatremia was observed (maximum level: 167 mmol/l at day 38) with concomitant polyuria (8 ml/kg/h; plasma osmolality: 324 mOsm/l, urine osmolality: 149 mOsm/l). Renal function parameters were normal (creatinine: 0.95 mg/dl, urea 37.1 mg/dl), as well as levels of cortisol, aldosterone and plasma renin activity. No decreases in Na^+ levels were observed when supply of liquids was increased to 200 ml/kg of body weight and supply of Na^+ was reduced. Based on the results of additional tests, CDI was diagnosed. At 60 day of life oral desmopressin therapy was introduced at a dose of 4.2 μg/kg per day in two doses, which normalized diuresis and results of laboratory tests (Na 138 mmol/l, plasma osmolality 280 mOsm/l, urine osmolality 418 mOsm/l). At 151 days of life the infant was discharged from the hospital and was referred as an outpatient to the Clinic of Pediatric Endocrinology at Poznań University of Medical Sciences.

Cranial ultrasound scan revealed hemorrhage into lateral ventricles of the brain: with grade III severity on the right side and grade II severity on the left according to Papille criteria. Subsequent examination revealed a malacic cavity connecting to the lumen of the right lateral ventricle (status following grade IV hemorrhage). At 144 days of postnatal life a cranial MRI (Magnetic Resonance Imaging) technique was performed with used TSE (Turbo Spin-Echo) sequence with T1- and T2-weighted images, FLAIR (Fluid Attenuated Inversion Recovery) sequence, DWI (Diffusion-Weighted Imaging), SWI (Susceptibility Weighted Imaging) technique with contrast. The results indicated dilatation of the ventricles, deepening of the posterior horns and rounding of the anterior horns. Hemorrhage to the lateral ventricles, the fourth ventricle, choroid plexuses, periventricular white matter, and the right hemisphere of cerebellum were found with malacic and scarring lesions. Hypoplasia of the cerebellum and brain stem was also present. The ~4 mm pituitary gland was correctly positioned, with no signal in the posterior pituitary lobe visible in T1-weighted images, which demonstrated a lack of neurohypophysis. Post contrast, intensive enhancement of hypothalamus and pituitary stalk was found. The pituitary stalk, shifted to the right side, showed no focal changes in the suprasellar region (Fig. 1).

Discussion

Etiology and risk factors for CDI in neonates

Diabetes insipidus is caused by impaired response to vasopressin or inadequate secretion of this hormone. Both nephrogenic and central DI can have transient character. NDI is due to genetic defect of vasopressin receptor or to immaturity of renal tubules which declines along with the growth of a neonate. CDI can be a result of irreversible failure of pituitary gland or transient disruption of its function. Congenital malformations are permanent, but CDI caused by some drugs, infections, inflammations or increased pressure of surrounding structures is frequently transient. CDI is a condition which rarely manifests itself in newborn infants. It is definitely more prevalent in children and adults. Publications on this disease unit in premature infants are therefore rather scarce. Retrospective analysis of patients hospitalized at endocrinology units in the UK between 1992 and 2011 confirmed 19 cases of CDI. The causes of CDI identified in the analysis were varied: septo-optic dysplasia, chromosomal abnormalities, microcephaly, Ohtahara syndrome, MPHD (multiple pituitary hormone deficiency), CNS (Central Nervous System) tumor growth and isolated idiopathic diabetes insipidus. In the analyzed group an increase in the incidence of CDI among preterm neonates was not found, relative to term infants. It was demonstrated, however, that in neonates born before week

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