**Spinal Stroke in a Pregnant Female with an Endodermal Cyst of the Cervical Spinal Cord (a Case Report and Literature Review)**

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**Objective.** The study purpose was to present a clinical case of spinal stroke in a pregnant female, which was caused by an endodermal cyst of the cervical spinal cord, and to analyze treatment tactics.

**Results.** A 20-week pregnant female presented with acute transverse spinal cord injury at the C3—C5 level. CT revealed an extramedullary space-occupying lesion in the ventrolateral position, with compression of the spinal cord at this level. The patient was transferred to the Neurosurgical Institute on the 5th day after disease onset. The patient underwent surgery on the 7th day after disease onset. Doctors of various specialties participated in preparation for surgery. During surgery, total resection of the space-occupying lesion and spinal cord decompression were performed. An obstetrician-gynecologist conducted intraoperative fetal monitoring by ultrasound. The histological diagnosis was an endodermal cyst. There was no improvement of neurological symptoms in the early postoperative period. After stabilization of the condition, the patient was discharged for follow-up care at the place of residence. According to the follow-up report, the patient died due to multiple organ failure. The child was alive, in serious condition, under mechanical ventilation.

**Conclusion.** In the case of spinal stroke, the decision on treatment tactics should be made no later than 12 hours after its onset; otherwise, the outcome is usually unfavorable, and a neurological deficit is irreversible. The decision about continuing pregnancy should be made individually in each case, and an approach to the choice of appropriate treatment tactics should be multi-disciplinary.

**Keywords:** spinal stroke, extramedullary tumor, spinal cord tumor, endodermal cyst, pregnancy.

Spinal stroke, especially at the cervical level, poses a serious danger to the patient’s life and health. In our practice, we were faced with a similar situation where the patient was a female with incomplete pregnancy. This article discusses the experience in treatment of the patient and associated problems.

**Clinical case**

A 24-year-old female patient M. was transferred from a regional hospital at the place of residence to the Burdenko Neurosurgical Institute (BNI). The patient was at 20 weeks gestation.

According to a medical history, the patient suffered from neck pain since the end of August 2015 and received pain relievers. In the evening of September 05, 2015, she noted numbness and growing weakness in the upper and lower extremities. In the morning, she could not move the extremities. The patient was transported by ambulance to a regional hospital; after examination by a neurologist and neurosurgeon, she was immediately transferred to the Critical Care Department. The patient’s condition at admission was evaluated as serious. The patient was seriously conscious and complained of neck pain, especially during head movements. Breathing was spontaneous and adequate. Bilateral anesthesia of all kinds of sensitivity, starting at the C3—C5 level of the spinal cord, and flaccid tetraplegia were present. Reflexes in the upper and lower extremities were absent. There was acute urinary retention.

Within 1 day of hospitalization, the patient’s condition began to deteriorate rapidly; respiratory failure worsened, which required intubation of the trachea and ventilation. On the second day of stay at the hospital, after stabilization of the vital functions, the patient underwent spiral computed tomography (SCT) of the cervical spine, which revealed a space-occupying lesion in the vertebral canal at the C3—C5 level, which was located extramedullary and markedly compressed the spinal cord (Fig. 1). Blood test indicators were without significant abnormalities, except for hypokalemia (3.0 mmol/L).

Ultrasound of the uterine cavity confirmed a normally developing pregnancy at 20 weeks. There were no obstetric indications for abortion.

On the 3rd day after the development of focal symptoms, the patient was transferred to the BNI for surgical treatment. The patient’s condition at admission was serious: she was in the supine position in a passive pose. Ventilation was performed through an orotracheal tube. The patient followed simple instructions (she answered simple questions by nodding of the head, articulated, and closed the eyes on command) but quickly exhausted; there were no movements in all the extremities; there was a loss of all types of sensation, starting at the C5—C6 level of the spinal cord and below. The patient had hyperthermia of up to 38.7 °C and arterial hypotension that required intravenous infusion of vasopressors (norepinephrine), and worsening respiratory failure (acceptable SpO2 figures were reached only at FiO2=0.8). X-ray of the lungs revealed bilateral infiltrative shadows in the lower lobes and perihilar portions. The patient underwent therapeutic fibrobronchoscopy...
accompanied by removal of a small amount of viscous purulent sputum. Given the episodes of desaturation and desynchronization with the respirator, elevation of the CRP level, fever, pulmonary radiographic findings, nature of the underlying disease, and expected need for prolonged ventilation, a puncture tracheostomy was performed on the second day of stay at the Critical Care Department of the BNI. On ventilation with positive end expiratory pressure of 10 cm and FiO2=0.6 as well as norepinephrine infusion, the patient’s condition was stabilized, after which (on the 5th day of stay at the BNI), she underwent magnetic resonance imaging (MRI) of the cervical spinal cord (Fig. 2) and brain. MRI revealed an extramedullary space-occupying lesion at the C3—C5 level, which was located in the vertebral canal on the right anteriorly, compressing the spinal cord and displacing it to the left posteriorly. The lesion had a high T2 signal (Fig. 2a, c), differing from that of free CSF, and relatively clear smooth contours. T1-weighted MRI revealed signs of parietal hemorrhage in the lesion (Fig. 2b). The spinal cord at the pathology level and I or II vertebra below it, as well as the medulla oblongata, had signs of vasogenic edema. Series of T1 and T2-weighted axial MRI scans of the brain revealed no pathological changes.

The patient repeatedly consulted an obstetrician-gynecologist and underwent daily ultrasound of the uterine cavity, which revealed signs of threatened miscarriage. There were no obstetric indications for abortion. The patient was offered surgery for removal of the space-occupying lesion at the C3—C5 level with preserving pregnancy; the patient and her mother gave a written consent.

On the 7th day of stay at the BNI, the patient underwent surgery: microsurgical resection of the extramedullary space-occupying lesion at the C3—C5 level using ultrasound monitoring of the fetus. Anesthesia included intravenous propofol (infusion under control of anesthesia depth) + intravenous bolus of fentanyl and rocuronium during surgery. Arterial pressure was monitored directly through a catheter placed in the left radial artery. Continuous intravenous infusion of norepinephrine was performed throughout surgery due to the tendency to arterial hypotension.

The patient was placed in the left lateral position that was considered as the most physiological one, given the gestational age and the need for intraoperative ultrasound monitoring of the fetus. The patient’s head was fixed with a Mayfield clamp; the patient underwent a laminectomy at the C3—C5 level. The dural sac was stretched, not pulsed, and prolapsed into the wound. After opening of the dura mater, about 3 mL of fluid discharge released into the wound, which was collected for testing. The spinal cord was pale and ischemic. After excision of arachnoid adhesions, a pale-gray space-occupying lesion located anterolaterally on the right was found; it grossly compressed and displaced the spinal cord. After identifying poles of the lesion, its capsule was opened. The space-occupying lesion contained viscous yellowish liquid. The space-occupying lesion and its capsule were resected en bloc (Fig. 3).

An urgent biopsy revealed that the resected lesion had an endodermal origin.

Surgery was performed in the presence of an obstetrician-gynecologist, with continuous ultrasound monitoring of the fetal condition (Fig. 4).
The duration of surgery was 1.5 h. The total time of anesthesia, including positioning of the patient, was about 4 h. The amount of intraoperative blood loss was less than 150 mL.

After surgery, the patient was transferred to the Critical Care Department where she continued receiving intensive therapy. No changes in the neurological status occurred.

Postoperatively, the patient’s condition remained unstable: she was conscious; episodes of fever, up to 39 °C, persisted, without any effect of administration of antipyretics; artificial ventilation through the tracheostomy was continued. There was marked production of viscous sputum, requiring sanitation bronchoscopy; the tendency to arterial hypotension persisted, which required continuation of intravenous infusion of norepinephrine; salt-wasting syndrome typical of patients with a high level of spinal cord injury developed [1].

A control MRI study of the cervical spinal cord confirmed complete resection of the space-occupying lesion (Fig. 5).

A histological examination of the lesion capsule revealed the presence of cell-free masses with hemorrhages as well as fibrous tissue with cylindrical epithelium, the morphological structure of which was typical of an endodermal cyst (Fig. 6).

A histological examination of the discharge collected after opening of the dura mater revealed necrotized tissue with hemorrhages (cerebral detritus).

On intensive therapy, the patient’s general condition gradually stabilized: fever, hypoalbuminemia, and anemia regressed. There was no improvement in the neurologic status. According to an examination by an obstetrician-gynecologist, the fetus continued to develop normally; there were no indications for abortion.

Given expected prolonged subsequent treatment and rehabilitation and upon agreement with relatives, the patient was transferred to the Critical Care Department of a regional hospital at the place of residence.

Further information on the fate of the patient and the child was obtained from the doctors of appropriate clinics by phone. The patient’s condition remained relatively stable within a month after surgery; then, an infectious process in the lungs worsened. This fact and the need for antibacterial therapy with high doses of potentially fetotoxic antibiotics, as well as significant intrauterine fetal growth retardation, served as the basis for the decision to terminate the pregnancy. Preterm labor surgery (cesarean section) was performed at 29 weeks gestation, and a premature baby (boy) with a body weight of 780 g, with an Apgar score of 6, was born. The child was intubated and transferred to a specialized perinatal center for artificial ventilation. According to the available information, the child’s condition remained severe by the time of manuscript preparation; the child had several intracranial hemorrhages and was on artificial ventilation. The mother’s condition after cesarean section rapidly deteriorated. In the setting of bilateral pneumonia, sepsis, and progressive multiple organ failure, she died.

Discussion

Spinal pathology is the second most common disease after traumatic brain injury among neurosurgical diseases in Russia. Degenerative spine diseases are the most common spinal pathologies. Primary tumors of the spinal cord are relatively rare and account for only 10—15% of all CNS tumors, or 1—2 cases per 100,000 people per year [2]. Manifestations of spinal cord tumors during pregnancy refer to casuistic cases. For example, A. Moles et al. in their recent work on symptomatic vertebral hemangiomas [3] (ones of the most common spinal tumors) in pregnant females note that only 27 similar cases have been reported in the literature since 1948. Spinal cord tumors in pregnant females are even rarer. Our patient had an extremely rare pathology — an endodermal cyst.
An endodermal cyst is a cystic lesion resulting from endogenous dysgenesis; it is often combined with other developmental anomalies [4]. The endodermal cyst wall consists of columnar epithelium with cylindrical, cuboidal, or goblet cells that usually produce mucin. Inside, the endodermal cyst is lined with the ciliated epithelium. Depending on the location, there are intraspinal (intradural extramedullary) cysts, which are most common, and intracranial cysts that are usually located in the posterior cranial fossa, anteriorly to the brainstem. Intraspinal cysts often occur in the lower cervical and upper thoracic spine. The cysts typically have an intradural extramedullary (mainly anterior) location [5]. Endodermal cysts usually have an isointense
or hypointense signal on T1-weighted MR images and a hyperintense signal on T2-weighted MR images and are not contrast-enhanced. The lack of tracer uptake in the cyst wall distinguishes endodermal cysts from other cystic lesions, such as cystic schwannomas, cystic meningiomas, cystic ependymomas, and cysticercosis [6]. Differential diagnosis with arachnoid, epidermal, and dermoid cysts should be performed. Despite the benign nature, endodermal cysts can recur, therefore the main surgical treatment option is total cyst resection [5, 6].

The symptoms of spinal cord space-occupying lesions usually develop slowly and can be represented by pain as well as motor and sensation disorders [7]. An acute onset occurs extremely rare and usually is a consequence of concomitant spinal stroke, as it obviously occurred in the presented case. This is confirmed by the rapid development of a neurological deficit and the presence of hemorrhages in histological specimens. It is important that the rate of deterioration and the severity of clinical symptoms should be considered and profoundly influence the management of these patients.

Examination and, if necessary, surgical intervention should be performed as soon as possible, no later than the “golden” day, and preferably earlier [8—10]. In the presented case, surgery should be performed immediately, even at the place of residence, to provide at least decompression of the spinal cord. Transfer to the BNI and stabilization of the patient’s general condition took some time. The delay in surgery resulted in irreversible changes in the tissues of the spinal cord, as evidenced by the morphological picture of the fluid discharge released after dissection of the dura mater, represented by a necrotic tissue with hemorrhages.

In general, the management of pregnant females with spinal pathology, especially in cases of severe neurological symptoms, is extremely complex, and information on this topic is quite limited [11—14]. Usually, one or two clinical cases with almost full-term pregnancy or in the postpartum period are reported. In this regard, the most interesting is a paper by I. Han et al. [11], which summarizes the experience of managing 10 pregnant patients with different spinal pathologies at one clinic for 13 years, and an article by K. Vijay et al. [14], which describes 10 patients with vertebral hemangiomas. Summarizing the data provided by the authors of these two papers, we believe that two points are of crucial importance: 1) neurological status of the pregnant female and its dynamics and 2) gestational age of the fetus. In the case of life-threatening neurological symptoms, full-term or almost full-term pregnancy (32 weeks or more) almost uniquely necessitates the following approach: cesarean section and subsequent surgical treatment of the mother. In the case of life-threatening neurological

Fig. 4. Intraoperative ultrasound examination of the fetus.

Fig. 5. Postoperative T1- and T2-weighted MR images of the cervical spinal cord in the sagittal (a) and axial (b) projections; there is no residual cyst; the area of vasogenic edema and ischemia at the cervical level is decreased in size.
symptoms and early pregnancy, the authors of both works suggest abortion because of the risk of fetal exposure to X-rays and potentially fetotoxic drugs during diagnosis and surgery in the first trimester of pregnancy. We consider this recommendation as not quite reasonable, especially given the recommendations of the American Society of Radiologists, which contain the only limitations related to the use of gadolinium as an MRI tracer and a moderate risk of newborn hypothyroidism associated with the use of iodine-containing contrast agents [15].

Parturition in the case of incomplete pregnancy is an approach that always requires serious argumentation. A complication in premature newborns, such as intracranial hemorrhage, is a well-known phenomenon with the rate of occurrence, according to various authors [16], ranging from 35 to 90%. The cause for this high rate of the severe complication is now known: a highly vascularized embryonic matrix is a substrate of the developing brain. It is located in the subependymal space of the lateral ventricles and undergoes a reverse development at the 32―34th week of pregnancy. Capillaries of this embryonic matrix are immature and fragile and lack autoregulation mechanisms, which sets the ground for hemorrhage in the case of even a slight increase in cerebral blood flow [16]. In our opinion, the only solution in our situation was an attempt to preserve pregnancy by all means. Obviously, our colleagues had solid indications for operative delivery in the case of incomplete pregnancy. However, the consequences of prematurity in the form of typical complications affected the further fate of the baby.

Conclusion

The presented clinical case is a rare coincidence of unfavorable circumstances: the presence of an extramedullary space-occupying lesion in the patient, lesion-induced spinal stroke, and incomplete pregnancy. All these factors influenced the unsatisfactory outcome of the disease. An endodermal cyst is a rare cystic lesion of the intradural space, which we encountered for the first time in our practice. Spinal stroke caused by an anterior endodermal cyst at the cervical spinal cord level in a pregnant female had not been reported in the literature before, so we tried to present in detail this case and the features of microsurgery, anesthesia, and critical care. Regarding the management of the patient, we believe that the decision of surgery should have been made within the first 12 h after the disease onset. The outcome of later interventions is unfavorable in most cases, and the neurological deficit is irreversible. Treatment of patients with combined pathology requires participation of various specialists. In our case, the fetus condition was controlled by obstetrician-gynecologists. Unfortunately, prolongation of pregnancy up to 37―38 weeks was excluded in the presented situation. Was it possible to avoid this treatment outcome? In our view, under these circumstances, unfortunately not.

There is no conflict of interest.
The article describes a rare clinical case of intensive therapy and surgical treatment of a 24-year-old patient who was admitted to the Neurosurgical Institute at 20 weeks gestation with spinal stroke associated with a space-occupying lesion of the cervical spinal cord — an endodermal cyst.

Undoubtedly, the presented clinical case is of great practical importance and real interest. Pregnancy combined with spinal pathology is rarely encountered by the neurosurgeon and neurointensivist in their practical work and is always associated with considerable difficulties in choosing the treatment strategy and tactics. In the present case, the authors encountered a combination of unusual and, unfortunately, extremely unfavorable factors that occurred simultaneously in the patient. These factors included a very rapid development of the clinical and radiographic picture of the tumor in the form of cervical stroke, a rare histological type of the space-occupying lesion, and delayed radical surgery (because the patient had no neurological symptoms. Unfortunately, surgical care, at least decomposition of the spinal canal, was not provided in the first hours after admission of the patient to a local hospital. Since the patient was admitted to the Neurocritical Care Department, which included reversal of life-threatening conditions, such as salt-losing syndrome and hyperthermia. The perioperative period was complicated by not quite ordinary phenomena, the genesis of which as well as initial severity of the patient’s condition are not clear because of limited world experience. Preserving pregnancy in this situation is also debatable.

Undoubtedly, only joint efforts of doctors of different specialties in a well-equipped hospital are able to solve the problems of treatment of spinal cord space-occupying lesions in the pregnant patient with severe and rapidly worsening neurological symptoms. Unfortunately, surgical care, at least decompression of the spinal canal, was not provided in the first hours after admission of the patient to a local hospital. Despite the fatal outcome of the disease for the mother and dubious prospects for the newborn, this clinical case demonstrates the clinical case of intensive therapy and surgical treatment of a 24-year-old patient who was admitted to the Neurosurgical Institute at 20 weeks gestation with spinal stroke associated with a space-occupying lesion of the cervical spinal cord — an endodermal cyst.

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