Management of Ruptured Arteriovenous Malformation During Pregnancy- Case Report.

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Case report

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Abstract

Background: Over the years care of pregnant woman has improved all over the world. However, still a significant percentage of the causes of pregnant mortality are subarachnoid haemorrhages due to rupture of arteriovenous malformation (AVM). According to available data, the percentage is as high as 5-12%. Neurosurgical operations in pregnant women require the cooperation of doctors of many specializations and are associated with close supervision of the patient due to the increased risk of complications. There are no guidelines regarding brain hemorrhages in pregnancy. This is the reason why case reports in this field are valuable as source of medical knowledge.

Case presentation: A 28-year-old patient was admitted in a severe condition, GCS: 3, to the ICU due to massive intracerebral bleeding with a puncture into the ventricular system from the AVM in left frontal lobe. The family informed about the possibility of patient's pregnancy, TVUS was performed and the sixth week fetus was visualized in the uterus. After gynaecological and neurosurgical consultation the successful surgery was made.

The patient was hospitalized for 5 months at the ICU. The stay was complicated by pneumonia and an urinary tract infection that were successfully cured. The patient was under supervision of gynaecologists, the pregnancy was developing properly, tests were carried out as recommended. The patient underwent psychotherapy, rehabilitation and music therapy.

At 35th week of gestation elective caesarean section was performed. A female infant of 2400 g was born, on the Apgar scale in 1st, 3rd, 5th minutes, 6,10,10 points respectively. Labour was complicated by mothers seizures and respiratory distress. After three days the patient's condition improved. Currently patient is rehabilitated as an outpatient.

Conclusions: The above case emphasizes the need for cooperation between many specialties, physiotherapists and midwives, which enabled effective therapy of a pregnant woman and saving her life in such a critical situation. In addition interdisciplinary treatment ended positively for both mother and child.

Background

Neurological conditions are one of the main causes of mothers' deaths.[6] Common during pregnancy are subarachnoid hemorrhage and intracerebral hemorrhages, which are mainly caused by ruptured aneurysms and arteriovenous malformations (AVM). Additional aneurysm and AVM rupture inducing factors during pregnancy are i.e. hemodynamic or hormonal changes. It is possible that hormonal changes alter the organization and composition of the arterial and venous intima.[10] Probably it can be associated with reticulum fibers fragmentation, decrease in acid mucopolysaccharides and loss of the normal corrugation of elastic fibers. These changes are presumably caused by increased levels of estrogen, progesterone, human chorionic gonadotropin (β -hCG) and relaxin and occur during physiological pregnancy.[2, 3] The risk of AVM rupture depends on gestational age, higher risk is observed

during second and third trimester than during first trimester and postpartum [8] The highest risk of AVM rupture is observed during second trimester when the highest cardiac output is observed.[11] According to study the risk of rupture of AVM is 8.1% during pregnancy.[4, 7] Among all pregnancies the risk of aneurysm rupture is in the range of 0.001-0.01%. [2] Maternal fatality rate is 83%, whereas for intracerebral hemorrhages it is not well established in corresponding literature. Intracerebral hemorrhage is an interdisciplinary problem that require cooperation of many specialists, physiotherapists, psychologists and all medical staff. This case report shows possible outcome of neurosurgical treatment of vascular disorders during pregnancy. It also outlines challenges of intensive therapy during pregnancy.

Case Presentation

A 28 years old woman, not diagnosed with hypertension had not presented any alarming signs until she lost consciousness and had episode of emesis at her workplace. She was transported to the nearest hospital where computer tomography (CT) and neurological consultations were ordered. CT scan revealed intraventricular bleeding and arteriovenous malformation in left frontal lobe. [Fig. 1.]. Patient presented critical condition: GCS: 3; anisocoria: left pupil> right pupil. After diagnostic tests patient was admitted to intensive care unit (ICU). Patient was given IV pharmacological neuroprotection (thiopental 100 mg/h, xylocaine 100 mg/h, 20% mannitol 3x 100 ml). Respiratory therapy: SIMV-VC, FiO2 0,4, SpO2 98-100%, cardiologically stable: BP 90-100/60-70 mmHg, HR 90-100/ min, CVP 0-5 mmHg, diuresis preserved, body temperature: 36,2-38,9 C degrees. Patient was disqualified from neurosurgical treatment.

At the request of the family transferred to the higher reference hospital, where lifesaving ventricular drainage was performed. Patients family reported that she might be pregnant, transvaginal ultrasonography, revealed the inherence of six-weeks pregnancy. Level of β -hCG was 30 049 mIU/ml. A magnetic resonance imaging (MRI) of the head was made, and revealed: blood in ventricular system, lateral ventricles enlarged. In the left frontal lobe there was exposed 32x27x33 mm AVM in the form of hemangioma with vast draining vessel emptying into superior sagittal sinus. In maximum intensity projection there was observed 3mm aneurysm of anterior connective artery. Digital Subtraction Angiography (DSA) was made; patient received 80 ml of contrast. Pelvic protection with individual dosimeter was applied to patient [Table 1.].

Table 1
Digital Subtraction Angiography individual dosimeter measurements.

Cumulative fluoroscopy time:	3:12 m:ss
Cumulative Air Kerma:	108 mGy
Total number of acquired images:	148
Total number of acquired exposure images:	148

Interdisciplinary team of doctors made decision to perform neurosurgery after patient had been stabilized in eighth week of pregnancy. Goals of neurosurgery were to clip an aneurysm of anterior connective artery, remove intracerebral hematoma and AVM. It was made from frontopterional approach. Surgery was difficult, because malformation was highly vascularized, vessels were very small and fragile. AVM had following main vessels: two coming off left frontal artery, three big coming off right middle cerebral artery and the biggest emptying into superior sagittal sinus. The size of AVM was about 32x27x33 mm [Fig. 2.]. The surgery was complicated by abundant, recurring hemorrhage that was controlled by Surgicel. Whole operation lasted for over fifteen hours, blood loss was estimated for 1500 ml. Therapy with 20% mannitol and nimodipine was implemented during the operation. Patient also received infusion of 2g MgSO₄. After surgery level of β -hCG decreased slightly from 84 361,90 mIU/ml to 74 246,90 mIU/ml due to blood transfusion.

After surgery patient was unconscious, ventilated mechanically, cardiologically stable. Patient stayed under IV anaglosedation for four days. On the fourth day of hospitalization she got fever and was diagnosed with pneumonia caused by *Klebsiella oxytoca* and *Pseudomonas aeruginosa*, treated with antibiotics effectively. State of patient was getting improved, however two weeks from operation hydrocephalus was diagnosed. [Fig. 3.].

Patient was operated again. Before neurosurgery patient received progesterone per vaginal (PV) in case of blood loss, which could result in β -hCG decrease leading to miscarriage. Goals of neurosurgery were to remove intraventricular hematoma and implant medium pressure ventriculoperitoneal shunt to the frontal horn of right lateral ventricle. Patient under general anesthesia, skin was incised on the length of 3 cm within postoperative scar in the right frontal area. To the frontal lobe of right lateral ventricle an endoscope was entered. Significant amount of organizing clots and yellowish membranes were observed, the ventricle was rinsed and after few minutes satisfactory visibility was achieved. The ventricular septum was perforated. Left lateral ventricle was entire filled with clots. Lateral ventricles, third ventricle and intraventricular foramina were cleaned. In typical way the ventricular drain was implanted to frontal horn of right lateral ventricle and peritoneal drain to peritoneum. The ventriculoperitoneal shunt was implanted.

Patient's condition was improving, she regain her consciousness. Patient started gradually opening eyes spontaneously and obey commands. Few days of improvement were followed by few days of regression when patient presented stupor. Attempts to extubate were made, patient periodically was breathing efficiently and periodically with help of continuous positive airway pressure (CPAP). After two months of stay at hospital tracheotomy was performed, patient was breathing spontaneously and coughing effectively. During intensive care patient was diagnosed twice with pneumonia, once with urinary tract infection, many times with asymptomatic bacteriuria, every was treated in accordance with the antibiogram. Level of CRP was increased during total time of hospitalization until delivery. Additionally patient presented fever which was treated with physical methods and paracetamol.

Patient was fed by feeding tube for three months of hospitalization, after that percutaneous endoscopic gastrostomy feeding was applied with oral administration of water and Nutridrinks. During intensive care patient was rehabilitating effectively, she attended to psychotherapy, music and speech therapy.

Two and half months from surgery seizures occurred. Patient, 57 kg weight, was given valproic acid 1g IV per day. Level of valproic acid after two months was below lower therapeutic window value (50-100µg/ml), so the dose was increased to 1,5g IV per day.

The patient was regularly consulted by gynecologist. During the first trimester, β -hCG was measured daily and consultations were held several times a week. Transvaginal and transabdominal ultrasonography were performed. Pregnancy, FHR and CRL were evaluated, the fetus was developing properly. Ordered virological tests were negative. Supplementation was initiated in accordance to pregnancy guidelines.

At the 25 PW cervix shortened to 2cm. Patient was given progesterone PV and she was transferred to department of pregnancy pathology in other hospital. Foetal vital signs were monitored using ultrasound, CTG. Patient was given steroid therapy with dexamethasone (6mg) in four doses. Throughout the period of hospitalization, there was no uterine contractions noted. Elective caesarean section (C-section) was performed in 35PW. A 2400 g baby was delivered with Apgar score of 6, 10 and 10 at 1st, 3rd and 5th minute. C-section was complicated by respiratory distress and seizures and patient needed to be under anesthesiologist's supervision. Further hospitalization was complicated by three episodes of seizures. The level of valproic acid was less than therapeutic and patient received 1,5 g of valproic acid p.o. per day. Patient with daughter were discharged from the hospital nine days after C-section.

Patient after treatment stays at home under neurosurgical supervision. Suffers paraparesis more pronounced on left side. Lovett scale for left lower limb 2 points, left upper limb 2/3 points and 3/4 for right upper and 2/3 lower limb. Patient communicates with several words, blinking of eyelid and charts. Patient stays conscious and aware.

Discussion And Conclusions

Almost every medicine and radiation dose that is ordered, needs to be taken under consideration. Safe dose of radiation during pregnancy is 50 mGy.[9] Patient got many medicines from FDA category C, but all of them were indispensable. Blood pressure was regulated with norepinephrine, in order to avoid fetal hypoxia. Mannitol 20% or nimodipine were used in imminent danger of brain swelling or secondary ischemic stroke. Patient during DSA had individual radiation detector and it showed after examination 108 mGy. It was impossible to resign from DSA before such a big risk-based surgery. The risk of miscarriage was rather big during surgery and hospitalization, but saving mothers life was a supreme value. Numerous consultations, that were made to keep patient in stable state and keep pregnancy. Obstetrics consultations were made more than once per week during first trimester.

Most intracranial aneurysm and AVMs are asymptomatic, so its detection is hard. Among risk factors of aneurysm or AVM rupture there is hypertension and that factor can be safely treated. Hypertensive

disorders of pregnancy affect up to 10% pregnancies.[5] Effective treatment of hypertension allows to avert from hypertension complications during pregnancy that leads to fetal and maternal morbidity and mortality. Widely used and proved to be safe during hypertension in pregnancy are: methyldopa, labetalol, extended-release nifedipine and hydralazine.[1]

There are no guidelines regarding brain hemorrhages in pregnancy. That is the reason, why therapeutic process in that case depended on doctors commitment and courage to perform therapy that is very rare and complicated. The case shows that it could be made safe even in such critical condition.

Multidisciplinary work and cooperation between doctors, physical therapists, psychologists, nutritionists, microbiologists and other medical staff is a basis for successful treatment of patients.

Declarations

Ethics approval and consent to participate: This case report has been processed positively by The Bioethics Committee of the Nicolaus Copernicus University functioning at Collegium Medicum in Bydgoszcz. Permission number: KB 24/2019. Written informed consent for participation was obtained from the patient.

Consent for publication: Written informed consent for publication was obtained from the patient.

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Authors' contributions: All authors made substantial contributions to conception and design, or acquisition of data, or analysis and interpretation of data. M.W.S. provided the conception, supervised all the writing process, revised it critically for important intellectual content and gave final approval of the version to be submitted; M.G. provided the conception and design; S.K. supplied the design of study and acquisition of data and drafting the manuscript; P.B. was responsible for analysis and interpretation of data. All authors read and approved the final manuscript.

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Figures

Figure 1

Preoperative non contrast CT scan, revealing intracerebral hemorrhage with ventricular extension. A, B, C: Axial view; D: Coronal view.

Figure 2

Preoperative DSA, showing AVM, its vascularity coming off: A, B: left frontal artery; C, D: right middle cerebral artery and emptying into sagittal sinus. Additional middle connecting artery aneurysm.

Figure 3

Postoperative MRI showing complication – hydrocephalus (before shunt implantation). A, B, C: axial T1; D, E, F: axial T2 flair; G, H: axial T2