Case Report
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The Successful Replacement of Aortic Valve and Ascending Aorta in Patients with Type A Aortic Dissection in the Postpartum Period. The Analysis of Literature and Demonstration of Own Observations

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ABSTRACT Aortic dissection is a rupture of the inner layer of the aorta with subsequent penetration of blood into the degeneratively altered middle layer with the formation of false lumen and true lumen. Pregnancy is one of the risk factors for the development of aortic dissection. The incidence of aortic dissection during pregnancy is only 0.0004% of cases.

AIM OF THE STUDY To analyze national and foreign literature, as well as share own clinical observations in the diagnosis and treatment of patients with aortic dissection in the postpartum period.

MATERIAL AND METHODS Two patients after successful childbirth, in the late postpartum period, were admitted with a diagnosis of aortic dissection type A according to Stanford.

CT angiography confirmed the presence of Stanford type A aortic dissection. After additional examination, surgical treatment was performed to replace the aortic valve and ascending aorta under artificial circulation, with a satisfactory clinical result.

CONCLUSION The diagnosis of aortic dissection should be considered in all pregnant women with chest pain, as this condition often goes undiagnosed.

The pregnancy period is one of the risk factors for the development of aortic dissection with a high mortality rate. The likelihood of developing aortic dissection in women at risk peaks in the third trimester and the first 12 weeks after delivery.

The risk group should include women with confirmed syndromic and non-syndromic genetic diseases, bicuspid aortic valve, coarctation of the aorta, or at least one major criterion indicating the presence of aortopathy (ectopia lentis, aortic aneurysm, habitus, genetic testing).

If Marfan syndrome is present, surgical intervention should be considered if the maximum aortic diameter is more than 4.5 cm before pregnancy. In women with Marfan syndrome and aortic dissection in the family history, as well as in the presence of more aggressive genetic diseases (Loeys-Dietz syndrome, Ehlers-Danlos syndrome), it is possible to consider preventive surgical treatment for an aortic diameter of 4.0 cm or more.

The delivery in high-risk patients is recommended to be performed in a hospital that has a cardiac surgery service and an "aortic" team.

Keywords: aortic dissection, pregnancy, aortic valve and ascending aorta replacement
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AD - aortic dissection
BCS - brachiocephalic vessels
CT - computed tomography
ECG - electrocardiography
EchoCG - echocardiography

INTRODUCTION

Aortic dissection (AD) is a rupture of the inner layer of the aorta with subsequent penetration of blood into the degeneratively changed middle layer with the formation of two lumens (true and false) [1].

The causes of AD are diseases associated with changes in the vessel wall: syndromic and non-syndromic genetic diseases (Marfan syndrome, Ellers-Danlos, Loeys-Dietz, Shereshevsky-Turner, etc.), degenerative acquired lesions of the aorta, bicuspid aortic valve, iatrogenic factors [2].

Aortic dissection during pregnancy is rare, occurring in only 0.0004% of cases [3]. During pregnancy, a number of hormonal changes occur in a woman's body: an imbalance occurs between collagen and elastin due to an increase in the concentration of estrogen, the structure of elastic fibers in the medial layer of the aorta changes and the morphology of the vessel changes due to an increase in the concentration of progesterone [4].

Hemodynamic changes occur due to increased activity of the sympathetic system and activation of the renin-angiotensin-aldosterone system, resulting in an increase in heart rate and cardiac output. The volume of circulating blood almost doubles due to the addition of the placental circulation. The above-mentioned changes in the structure of the aortic wall and hemodynamic changes are most characteristic of the third trimester of pregnancy and the first 12 weeks after delivery and play a fundamental role in the pathogenesis of AD [5, 6].

Aortic dissection during pregnancy and the early postpartum period is a life-threatening condition associated with a high risk of maternal and fetal mortality, most often associated with a genetic predisposition. Analysis by *FF Immer et al.,* showed that 50% of patients with aortic dissection during pregnancy or the early postpartum period have a genetic syndromic disease (mainly Marfan syndrome) [7].

The tactics of managing pregnant and postpartum women with aortic dissection remains a complex multidisciplinary task and is associated with solving a number of problems, including routing, timing and sequence of surgical intervention and delivery.

We report two successful clinical observations of patients whose pregnancy and postpartum period were complicated with acute aortic dissection type A according to Stanford.

Case report 1

A 36-year-old patient was transferred to the N.V. Sklifosovsky Research Institute for Emergency Medicine from a medical institution in Moscow with a diagnosis of "Acute aortic dissection type *A* according to *Stanford* " on the 12th day after spontaneous birth.

From the anamnesis: it is known that since childhood she was seen by an ophthalmologist with a diagnosis of ectopia of the lens. Also the patient's high height—194 cm, was noteworthy. During planning and during the pregnancy itself, there was no concern for the presence of genetic disorder, and therefore echocardiographic examination (EchoCG) and computed tomography (CT) were not performed. According to the patient, the pregnancy was threatening to be terminated in the second trimester. A month before giving birth, she noted the appearance of a sharp pain syndrome in the chest, accompanied by hypotension. When contacting a therapist, according to an electrocardiographic study (ECG), no pathology was detected and no further examination was carried out.

According to medical documentation, 12 days ago, the first spontaneous birth at 39 weeks, a live, full-term baby was born. She was discharged from the hospital on the 4th day. On the 9th day, she noticed severe swelling of the lower extremities and began to worry about shortness of breath during physical activity. On the 12th day there was a repeated episode of acute pain in the chest with a sharp increase in shortness of breath. The ambulance team hospitalized her at the Moscow City Clinical Hospital. Examination using CT angiography revealed *Stanford* type *A aortic dissection*. The patient was urgently transferred to the N.V. Sklifosovsky Research Institute for Emergency Medicine for surgical treatment. To confirm the diagnosis and determine surgical tactics, CT agiography with ECG synchronization was performed.

EchoCG: a hyperechoic structure was located in the lumen of the ascending aorta and aortic arch. The diameter of the aortic root at the sinus of Valsalva is 56 mm, the ascending aorta is 81 mm, the aortic arch is 35 mm. Aortic valve: tricuspid, thickened cusps. Violation of central cooptation of the leaflets due to prolapse of the right coronary cusp into the cavity of the left ventricle. Severe regurgitation. Echo signs of hydropericardium.

CT angiography with ECG synchronization: contents up to 11 mm wide are determined in the pericardial cavity. Thoracic aorta: diameter at the root level — 85 mm, ascending section — 61 mm, arch — 30 mm, descending section — 38 mm. The structure in the lumen of the aorta is heterogeneous due to the presence of double-circuitry from the level of the ascending aorta, which extends throughout the arch, descending and abdominal aorta. The false lumen occupies 50% of the true lumen, there is no significant compression of the true lumen, the visceral branches extend from the true lumen (Fig. 1).

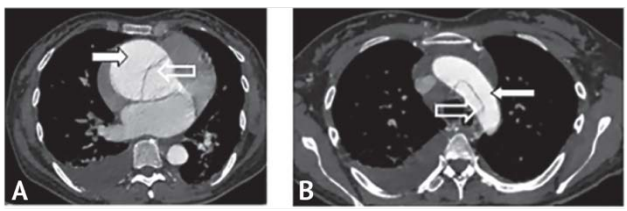

Fig. 1. CT angiography with electrocardiography synchronization. A - ascending aorta; B - aortic arch

Note: white arrow — false lumen of the aorta, contour arrow — true lumen of the aorta

Considering the futility of conservative therapy and the direct threat to life, emergency surgical treatment was performed: the replacement of the aortic valve and ascending aorta with a valve-containing conduit with reimplantation of the coronary artery ostia using the *Kouchoukos technique* under conditions of artificial circulation and circulatory arrest.

Progress of the operation. The design of the heart-lung machine: right subclavian artery – right atrium. Myocardial protection by pharmacocold cardioplegia (Custodiol 3,000 ml) into the coronary sinus. Moderate hypothermia.

The pericardium was moderately tense. After opening the pericardium under pressure, 200 ml of fresh blood was released. The aorta was pear-shaped with a maximum diameter of up to 8.0 cm (Fig. 2), the wall was purple-bluish in color, the aortic arch was 3.0 cm.

The aortic wall in the proximal part was dissected by 2/3 of its diameter. At the level of the sinotubular junction, along the anterior surface of the aorta, an intimal defect measuring 3.0×1.0 cm was visualized. The dissection extends to the right coronary and non-coronary sinuses with dissection of the ostium of the right coronary artery (*NERI type B*) [8]. The aortic valve is tricuspid, the cusps were thickened, degeneratively changed, cooptation of the cusps is impaired due to dilatation of the sinotubular junction, prolapse of the right coronary cusp into the cavity of the left ventricle was observed due to detachment of the commissure between the right and non-coronary cusps. It was decided to replace the aortic root and aortic valve.

The aortic valve and aortic sinuses were excised with further formation of the ostia of the coronary arteries on platforms (1 cm^2).

At a core temperature of 28°C, the clamp was removed from the aorta, and circulatory arrest was initiated with selective antegrade unilateral perfusion of the brain through the right subclavian artery. The aortic arch was dissected by 2/3 of its diameter. No secondary fenestrations were found in the aortic arch. The ascending aorta was completely excised at the level of the brachiocephalic vessels (BCV) and part of the lesser curvature. A linear vascular prosthesis was used to replace the ascending aorta using the "half-arch" technique.

The circulatory arrest was terminated, artificial circulation was restored in full through the additional branch of the prosthesis, and warming of the patient began.

Prosthetics of the aortic root and aortic valve was performed using a valve-containing conduit (with a mechanical aortic valve prosthesis) with reimplantation of the coronary artery ostia using the *Kouchoukos technique*. Interprosthetic anastomosis. (Fig. 3)

The duration of artificial circulation was 190 minutes. The duration of aortic clamping was 71 minutes. Circulatory arrest lasted 17 minutes.

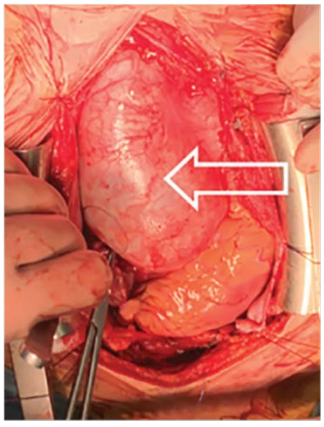

Fig. 2. Dissection of the ascending aorta

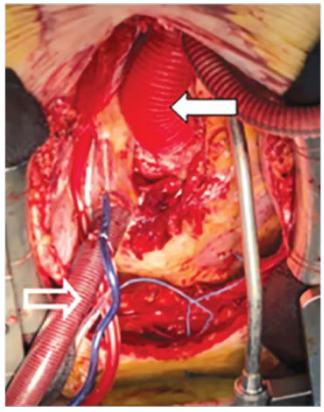

Fig. 3. The final version of the operation of replacing the aortic root and aortic valve with a valve-containing conduit.
Note: white arrow — valve-containing conduit, contour arrow — right atrial venous cannula

Tracheal extubation after 11 hours. Vasopressors were discontinued on the 2nd day. The patient was transferred to the clinical department on the 2nd day. According to echocardiography, the function of the mechanical prosthesis in the position of the aortic valve was satisfactory. The repeated CT angiography of the aorta revealed thrombosis of the false lumen at the level of the aortic arch and the initial section of the descending thoracic aorta, and residual dissection of the distal sections of the descending thoracic and abdominal aorta. On the 9th day the patient was discharged from the hospital in satisfactory condition. According to the results of a morphological study of the aortic wall, the duration of the dissection was more than 30 days (subacute stage).

Case report 2

The 35-year-old patient was transferred to the N.V. Sklifosovsky Research Institute for Emergency Medicine from a medical institution in Moscow with a diagnosis of "Acute aortic dissection type *A* according to *Stanford*" on the 4th day after the second childbirth by cesarean section.

From the anamnesis it is known that from the first year of life she was observed by a cardiologist for a genetic syndromic disease (Marfan syndrome, confirmed by a genetic test). The patient has a complicated family history; her grandfather and father with confirmed Marfan syndrome died of aortic dissection at the age of 27 years. When planning and carrying her first pregnancy, the patient was under close supervision of a cardiologist, with periodic echocardiography examinations. The first pregnancy proceeded without complications. The delivery was performed by caesarean section. The second pregnancy was terminated in the first trimester. Further, the patient underwent annual echocardiography studies; the diameter of the aortic root before the third pregnancy was 5 cm; CT angiography of the aorta was not performed.

The third pregnancy proceeded without complications; according to echocardiography, the diameter of the ascending aorta was 4.6 cm. Despite the presence of risk factors and the diameter of the ascending aorta, surgical intervention was not suggested.

According to medical documentation, she was discharged from the hospital on the 3rd day after the second delivery by cesarean section at 39 weeks. On the 4th day, she noted severe acute pain in the chest against the background of an increase in blood pressure to 180/90 mm Hg. She was admitted to the medical institution in Moscow. During the examination, CT angiography with ECG synchronization revealed aortic dissection of type *A* according to *Stanford* (Fig. 4). The patient was urgently transferred to the Research Institute of Emergency Medicine for surgical treatment.

CT angiography with ECG synchronization: in the pericardial cavity there is content up to 10 mm wide. Thoracic aorta: diameter at the root level — 55 mm, ascending aorta — 48 mm, aneurysm of the ascending aorta with intimal detachment and the formation of true and false lumens (Fig. 4).

EchoCG: the diameter of the aortic root at the level of the sinuses of Valsalva — 50 mm, the ascending aorta — 47 mm. In the projection of the sinuses of Valsalva, the ascending section and the aortic arch, a mobile hyperechoic structure (intima) was located. Severe aortic valve regurgitation.

Ultrasound examination of the pelvic organs: condition after cesarean section (4th day). No additional structures were identified in the uterine cavity.

Considering the futility of conservative therapy and a direct threat to life, emergency surgical treatment was performed: replacement of the aortic valve and ascending aorta with a valve-containing conduit with reimplantation of the coronary artery ostia using the *Kouchoukos technique* under conditions of artificial circulation and circulatory arrest.

Progress of the operation

The design of the heart-lung machine: right subclavian artery — both vena cava. Myocardial protection by pharmacocold cardioplegia (Custodiol 3,000 ml) into the coronary sinus. Moderate hypothermia.

At a core temperature of 28°C, the clamp was removed from the aorta, and circulatory arrest was initiated with selective antegrade unilateral perfusion of the brain through the right subclavian artery. The aortic arch was dissected by 2/3 of its diameter; no secondary fenestrations were found in the aortic arch. The ascending aorta at the level of the BCS and part of the lesser curvature were completely excised. A linear vascular prosthesis was used to replace the ascending aorta using the "half-arch" technique.

The circulatory arrest was terminated, artificial circulation was restored in full through the additional branch of the prosthesis, and warming of the patient began.

Prosthetics of the aortic root and aortic valve was performed using a valve-containing conduit (with a mechanical aortic valve prosthesis) with reimplantation of the coronary artery ostia using the *Kouchoukos technique*. Interprosthetic anastomosis (Fig. 5).

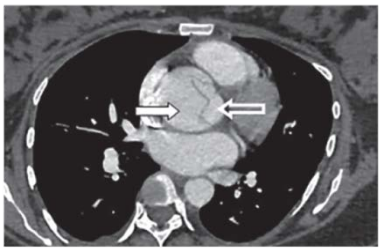

Fig. 4. CT angiography with electrocardiography. Ascending aorta. Note: contour arrow – true lumen of the aorta, white arrow – false lumen of the aorta

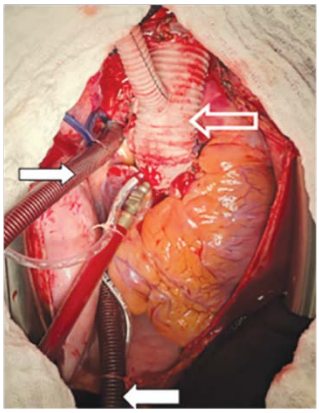

Fig. 5. The final version of the operation of replacing the aortic root and aortic valve with a valve-containing conduit. Note: contour arrow – valve-containing conduit, white arrows – venous cannulas of the superior and inferior vena cava

The duration of artificial circulation was 168 minutes. The duration of aortic clamping was 126 minutes. Circulatory arrest lasted 28 minutes.

Tracheal extubation after 10 hours. Vasopressors were discontinued after 9 hours. The patient was transferred to the clinical department on the 3rd day. According to echocardiography, the function of the mechanical prosthesis in the position of the aortic valve was satisfactory. According to the control multislice CT scan of the aorta with contrast enhancement, thrombosis of the false lumen was revealed along its entire length. On the 10th day the patient was discharged from the hospital in satisfactory condition.

According to the results of a morphological study of the aortic wall, the duration of the dissection was less than 2 days ("acute" stage).

DISCUSSION

In the literature, most often there are isolated clinical observations of pregnancies complicated by acute aortic dissection. One of the largest studies is the work of *A.C. Braverman et al.* according to the International Registry of Acute Aortic Dissection (*IADD*), where pregnancy-related type *A aortic dissection accounts for 0.13% of all cases (13 cases over 21 years) [9].*

The pregnancy period is one of the risk factors for the development of AD with a high mortality rate. The likelihood of developing AD in women at risk peaks in the third trimester and the first 12 weeks after delivery [2, 10–12].

The risk group should include women with confirmed syndromic and non-syndromic genetic diseases, bicuspid aortic valve, coarctation of the aorta, or at least one major criterion indicating the presence of aortopathy (ectopia lentis, aortic aneurysm, *habitus,* genetic testing).

The principles of medical support for pregnant women with genetic diseases are actively discussed by leading experts in these fields, but clinical guidelines have a low degree of recommendations and level of evidence [6]. If Marfan syndrome is present, surgical intervention should be considered if the maximum aortic diameter is more than 4.5 cm before planning pregnancy. In women with Marfan syndrome and aortic dissection in the family history, as well as in the presence of more aggressive genetic diseases (Loeys-Dietz syndrome, Ellers-Danlos syndrome), it is possible to consider preventive surgical treatment for an aortic diameter of 4.0 cm or more [2].

In cases where, at the stage of preconceptional preparation, aortic dilatation is detected in a woman, which does not require surgical intervention at the time of the study, it is recommended to carry out dynamic monitoring of the aortic diameter, through echocardiography, at least once during each trimester of pregnancy, before discharge from the hospital and in the first 12 weeks after delivery.

If aortic dissection is verified during pregnancy, at a period of more than 28 weeks, it is necessary to consider the option of performing simultaneous surgical intervention. The first stage is delivery by cesarean section, the second is aortic surgery.

Women at risk must be treated with extremely high clinical vigilance in order to prevent the development of serious complications associated with aortic pathology. A multidisciplinary approach to pregnancy management is required, with mandatory monitoring of aortic diameter throughout pregnancy and in the early postpartum period.

The presented clinical examples show successful cardiac surgical treatment of AD as a pregnancy complication. However, based on the morphological study, it can be assumed that in the first observation the patient experienced an acute period of aortic dissection type *A* according to *Stanford* and successfully underwent spontaneous childbirth, being in the subacute period without verification of the diagnosis with a very high risk of developing aorta-associated complications. Despite the patient having signs of connective tissue dysplasia (ectopia lentis, specific *habitus*), the diagnosis of a syndromic genetic disease was not established. Chest pain with hypotension was not taken into account by primary care specialists.

In the second clinical example, due to the established diagnosis of a genetic disease (Marfan syndrome), the patient was dynamically observed by a cardiologist from childhood. The first pregnancy proceeded without complications and successfully ended in delivery by cesarean section; the patient's second pregnancy was terminated at 9 weeks. Before planning the third pregnancy, the diameter of the ascending aorta according to echocardiography was 4.6 cm, which in the presence of Marfan syndrome is a direct indication for surgical intervention [2]. Despite all the known risk factors, aortic diameter when planning before pregnancy and multidisciplinary monitoring during pregnancy, surgical intervention was not offered at any stage, and therefore the development of aortic dissection could not be avoided.

As a conservative therapy, the European Society of Cardiology recommends preventive therapy with beta blockers for aortopathies throughout pregnancy in women with Marfan syndrome and non-syndromic genetic diseases [6], which was not done in our cases.

CONCLUSION

The diagnosis of aortic wall dissection should be considered in all pregnant women with chest pain, as this condition often goes undiagnosed.

Considering the increase in the frequency of pathologies of the cardiovascular system in pregnant women and in the postpartum period (aortic dissection, damage to the heart valves, etc.), requiring emergency surgical intervention, it is recommended to perform an echocardiographic study as one of the preconception preparation measures throughout pregnancy and early postpartum period in risk groups.

It is recommended that delivery in high-risk patients be carried out in a hospital that has a cardiac surgery service and an "aortic" team on staff.

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