

PRENATAL DIAGNOSIS OF COARCTATION AND INTERRUPTION OF THE AORTIC ARCH BY 2B AND 3-DIMENSIONAL ECHOCARDIOGRAPHY

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ABSTRACT — **OBJECTIVE.** Coarctation and interruption of the aortic arch is occupied the 4th place among all the congenital heart defects. Prenatal diagnosis of it is very important for the planning of the delivery and the The purpose of this study was to evaluate the diagnostic possibilities of the sighting of the fetus echocardiography in the two-mode imaging and a three-dimensional reconstruction for prenatal detection of aortic arch abnormalities.

MATERIALS AND METHODS. The prospective study was based on the analysis of prenatal ultrasound echocardiography datasets in a two-dimensional mode and 3D-reconstruction of 2 fetus groups, were examined between 20 and 36 weeks of gestation . 50 "healthy" fruit (I group) were included in the first (control) group. The second group consisted of 15 fetuses with coarctation and interruption of the aortic arch (II group).

Volume data sets of all fetuses, examining by 2D US, were acquired by 3D and cine 4D using spatiotemporal image correlation (STIC) software. Various additional rendering tools were applied. Color and power Doppler were added to the volumes acquired. A retrospective offline analysis was performe. Neonatal echocardiography and pathological examination were performed to verify the prenatal diagnosis.

RESULTS. In the analysis of 3D - «cardiac volume» in 37 patients (74%) of groupe I, aortic arch in longitudinal section was successfully visualized to exclude the pathology. 2 group of fetuses (aortic arch abnormalities, n=15): In 7 (46%) cases abnormality of the heart and great vessel was demonstrated by 2D and 3/4D ultrasound volume did not add to the information in the 2D loop. In 2 cases (16%) 3/4D ultrasound had added value in achieving or enhancing diagnosis in 2/6 of diagnosed cases and a definitive diagnosis was made only after 3D examination . This was 1 coarctation of aorta and 1 interrupted aortic arch, Type A. In 5 cases (33%) we had fals-positive results and 2D and 3/4D ultrasound examination were equivalent in it.

CONCLUSION. The datasets from 15 patients suspected of having prenatal coarctation and interruption of the aortic arch demonstrates the complexity of Interpretation echographic data

in forming the final diagnosis, prognosis, and the choice of further tactics. The main method of prenatal diagnosis disease of the aortic arch main method, in our view, is the two-dimensional imaging mode while 3/4D-modes are optional, but very usefull in several cases. However, no single module is sufficiently accurate for the diagnosis of aortic arch abnormalities, each case requires different and appropriate module of visualization. 3/4D addition enhances precision of diagnosis by providing planes and data that «flesh out» the 2D ultrasound examination.

KEYWORDS — prenatal diagnosis, fetal echocardiography, coarctation of aorta, interruption of the aortic arch

TOPICALITY

Coarctation and interruption of the aortic arch is occupied 4th place among all the congenital heart defects. Among the critical CHD incidence of coarctation of the aorta is 10% and interruption of the aortic arch — 1% (1, 15).

A high percentage of critical forms of coarctation of the aorta due to the need for prenatal detection of these forms of cardiac defect. Being a typical duct-dependent pathology, in which livelihoods newborn maintained until the closing of the ductus arteriosus, critical forms of pathology of the aortic arch require emergency specialized care after birth. Less severe forms of aortic coarctation, in which a moderate narrowing of the aortic isthmus that is light on the severity of clinical symptoms after birth. An estimated 60% to 80% of newborns with isolated coarctation of the aorta are sent

home as "healthy" children (1). This result is in a late detection of defects in this group of children, with the deteriorating results of delayed surgery (1, 15).

Prenatal diagnosis of pathology of the aortic arch is an important task of prenatal medicine, the solution of which will provide timely specialized care after birth, reduce infant and child mortality rates.

However, the detection of prenatal pathology of the aortic arch is associated with objective difficulties and the percentage of diagnosis of this disease is less than 43%.

Prenatal diagnosis of malformations of the aortic arch is the subject of a small number of studies (4,8,15), it confirms the diagnostic complexity of this anomaly. In this regard, there is a need for new diagnostic techniques that improve accuracy and results of diagnostic search for examination of the fetal heart.

The last decade has been steadily increasing number of publications on the use of 3/4-dimensional scanning modes (5, 6, 15) as a means of improvement of prenatal diagnosis of heart diseases, including diseases of the aortic arch.

The purpose of this study was to evaluate the diagnostic possibilities of the sighting of the fetus echocardiography in the two-mode imaging and a three-dimensional reconstruction for prenatal detection of coarctation and interruption of the aortic arch, the study of the evolution of fetal defect with the analysis and definition of outcomes, postnatal prognosis and determining the causes of diagnostic errors and their possible solutions.

MATERIALS AND METHODS

The prospective study was based on the analysis of prenatal ultrasound echocardiography datasets in a two-dimensional mode and 3D-reconstruction of 2 fetus groups. 50 "healthy" fetus (I group) were included in the first (control) group. The second group consisted of 15 fetuses with coarctation and interruption of the aortic arch (II group).

The study was conducted in the perinatal cardiology center based Russia, Moscow, Bakoulev SCCVS, Perinatal Medical Cardiocenter from 2011 to 2012.

Cases of isolated coarctation of the aorta were included in the study or the combination of "small" heart anomalies:

- with hypoplasia and stenosis of the aortic valve – 2 cases;
- with bicuspid aortic valve – 2 cases;
- with ventricular septal defect (VSD) – 9 cases;
- defective aortic-pulmonary septum – 1 case.

These observations were excluded from this study:

- the combination of this disease with combined CHD because the changes of hemodynamics

and morphometry of fetal heart in these cases are determined, first of all, a complex abnormality of the fetal heart, and do not reflect his true changes associated with obstruction in the aortic arch;

- with a positive parents decision to finish the pregnancy (post-mortem study of abortions were not performed).

The second group of pregnant women's age ranged from 24 to 39 years (Table 1, 2, 3) and averaged 29.8 years. The percentage of "age" of patients (over 35 years) was extremely low (1 case) — 6% of the total surveyed.

The reason for the patients to come in our department were: 1) suspicion of CHD in the fetus (II group), 2) a family history (in most cases – the birth of children with CHD from previous pregnancies) or private desire to be tested in the cardiocenter (I group).

All patients in group I were examined twice – in the 20–27 and 30–34 weeks gestation. 5 patients in Group II were examined twice like the group I and 10 patients were examined once.

In all cases the diagnosis is established by the fetal echocardiography, including a study of the 2D-scan mode using pulsed wave, color and power Doppler mapping and 3D-multiplanar mode scan. The study was conducted on the unit Voluson 730 Pro (GE Medical System) with convex and 3/4D transducer 3.5–5 MHz in the software Fetal Cardio mode by transabdominal access.

The protocol of the fetal heart examine, developed and introduced into clinical practice in Bakoulev SCCVS, Perinatal Medical Cardiocenter, which is based on the segmental approach to Van Praag in modifying Becker.

The following key indicators were studied to assess morphology and hemodynamics and comparative analysis: diameters of ascending part, the proximal and distal part of the arc, the diameter of the isthmus, the diameter of the descending aorta, the diameter of the ductus arteriosus and evaluation ratio of isthmus diameter to the diameter of ductus arteriosus. These parameters were carried out in the gray scale in the longitudinal projection of the aortic arch and in transversal-section — V-sight of connection ductal arch and the aorta.

The nature of blood flow in the aortic arch was assessed by color Doppler mapping and power Doppler mode.

Additional parameters, indirectly characterized by the pathology of the aortic arch, were used in the study:

- Ratio of left and right end-diastolic ventricular dimensions (the presence of ventricular disproportion),

- diameters of the great vessels, and their ratio (the presence of arterial disproportion),
- thickness of ventricular myocardium (presence left ventricular hypertrophy),
- diameter of atrioventricular and semilunar valves, their ratio of the left and right heart ,
- the dimension of the right atrium.

Each fetus heart was taken in a three-dimensional reconstruction ("heart volume") in the 4-chamber view. 3D «volume» multiplanar mode imaging was used to obtain transversal-sectional and longitudinal sections of the aortic arch and ductal arch. The criterion of successful visualization of the aortic arch was to obtain continuous images of the ascending aorta move in an arc, the isthmus and the proximal descending aorta. Criterion for successful imaging of ductal arch was to obtain continuous images of the outflow of the right ventricle, pulmonary valve and arteria, ductus arteriosus and proximal descending aorta.

Information on each patient is entered into the database, made in Excel. Standard Excel functions were also used to calculate the values of arithmetic studied variables (M) and standard deviation (\pm sd). The bulk of the statistical data is processed using the software package STATISTICA company StatSoft, Inc., (USA), version 6.0.

RESULTS

All 50 patients of the control group were examined in the first two months of life and confirmed the absence of disease of the heart and great vessels was confirmed.

Examined parameters in the control group were like indicators of the fetal heart at different stages of pregnancy, previously calculated in Bakoulev SCCVS, Perinatal Medical Cardiocenter on a representative sample of 2,000 women with normal pregnancy and no fetal pathology (link to the book).

One of the new morphometric criteria, which was not used before, was the ratio of isthmus diameter to the diameter of ductus arteriosus. In 44 patients of the control group (88%), the ratio was more than 0.8, that is, the diameter of the isthmus was greater or almost equal to the diameter of the ductus arteriosus. In 6 fetuses (12%), this ratio in the range 0,76–0,8 mainly by increasing the diameter of the ductus arteriosus.

In the analysis of 3D-«cardiac volume» in 37 patients (74%) of groupe I, aortic arch in longitudinal section was successfully visualized to exclude the pathology. In 7 patients (14%), visualization of the aortic arch was successfully for trace its continuity, but it could not exclude the narrowing of the isthmus. In 6 patients (12%) of the control group, it was not possible to visualize the

aortic arch by the three-dimensional reconstruction.

Groupe II consisted of 3 subgroups (A, B, C). A sub-group consisted of patients without CHD (Table 1), subgroup B — patients with coarctation of the aorta, which needed only dynamic control after birth without surgery (Table 2), subgroup C — patients with coarctation of the aorta, requiring surgery and interruption of the aortic arch (Table 3).

Subgroup A. 5 infants (33%) of the 15 patients of the group II had clinical and echocardiographic examination in the neonatal period to exclude the presence of the disease. All five «healthy» newborns had prenatal predictors of fetal aortic coarctation appeared after 30 gestation week:

- ventricular disproportion: in 4 cases (80%) — the ratio $RV / LV > 1.6$;
- arterial disproportion: in 5 cases (100%) — ratio of the diameter of the aorta to the diameter of the pulmonary artery < 0.8 ;
- the ratio of isthmus diameter to the diameter of ductus arteriosus in 3 cases (60%) — 0,66–0,7 and in 2 (40%) cases — more than 0.7;
- ventricular septal defect in 3 cases (60%) had small size (up to 2 mm) and 2 (40%) patients in this subgroup interventricular septum was intact;
- 1 patient (20%) — had tricuspidal regurgitation 2+;
- in 2 (40%) patients — pericarditis.

Subgroup B. 2 out of 15 patients of the study group (13%) had a pressure gradient on the isthmus near 20 mmHg in neonatal period and needed dynamic control but no surgery.

Manifestation ultrasound indicate pathological changes in these two cases were in the third trimester of pregnancy. In both cases:

- absence of ventricular disproportion
- intact interventricular septum,
- arterial disproportion,
- the ratio of isthmus diameter to the diameter of ductus arteriosus — 0.6–0.7.

In one case, the image of the aortic arch In a three-dimensional reconstruction was received successfully and showed normal heart, in the second case it was highly suspicious for the presence of tubular coarctation.

Subgroup C. 8 cases out of 15 (53%) were from the subgroup C.

2 cases of 8 (25%) with postnatal diagnosis — interruption of the aortic arch (Fig. 3, 4). Both cases displayed prenatal ultrasound signs of manifestation of obstructive lesions of the aortic arch earlier — at 23 and 24 weeks of pregnancy:

- No (in both cases) ventricular disproportion,
- ventricular septal defect - 5.4 and 6.3 mm;
- the presence in one case of hypoplasia and steno-

Table 1.

Patient N ^o	Age	Gestation age	Ventricular disproportion	Arterial disproportion	Ao/duct.	VSD (mm)	Prenatal diagnosis	Postnatal diagnosis
1	31	36	++	+	0.60	1.5	AC	Normal
2	30	33	-	+	0.77	1.9	AC	Normal
3	28	32	++	+	0.75	1.4	AC	Normal
4	28	34	++	+	0.90	-	AC	Normal
5	32	32	++	+	0.85	-	AC	Normal

Ao/duct — Istmal to Ductal ratio, VSD — ventricular septal defect, AC — aortic coarctation, ++ — strong feature, + — moderate feature, - — no feature

Table 2.

Patient N ^o	Age	Gestation age	Ventricular disproportion	Arterial disproportion	Ao/duct.	VSD (mm)	Prenatal diagnosis	Postnatal diagnosis	Outcome
6	33	35	-	+	0.9	-	AC	AC	Need for surveillance
7	29	36	+	+	0.8	-	AC	AC	Need for surveillance

Ao/duct — Istmal to Ductal ratio, VSD — ventricular septal defect, AC — aortic coarctation, ++ — strong feature, + — moderate feature, - — no feature

Table 3.

Patient N ^o	Women's age	Gestation age	Ventricular disproportion	Arterial disproportion	Ao/duct.	VSD (mm)	Prenatal diagnosis	Postnatal diagnosis	Outcome	
8	25	23	-	++	Ductal-4	6.3	Interrupted aortic arch	Interrupted aortic arch Type B	Surgery	Microdiletion p22 q 11
9	29	24	-	++	0.3	5.4	AC	Interrupted aortic arch Type A	Intranatal death	
10	39	24	-	+	0.4	5	AC	AC	Surgery death	
		30	-	-	Ductal-5	5.3	Interrupted aortic arch?			
11	30	23	-	-	0.7	3.1	AC	AC	Surgery	
		33	+	+	0.66	3.2				
12	27	24	-	+	0.71	2.6	AC	AC	Surgery	
		32	+	+	0.64	2.8				
13	27	25	-	+	0.7	3	AC	AC	Surgery	
		32	+	+	0.7	3.1				
14	24	28	++	+	0.5	-	AC	AC	Death at the age of 3 days	Multiple malformation
		32	++	+	0.49	-				
15	33	31	++	-	0.57	3	AC	AC	Death at the age of 2 days	Multiple malformation

Ao/duct — Istmal to Ductal ratio, VSD — ventricular septal defect, AC — aortic coarctation, ++ — strong feature, + — moderate feature, - — no feature

sis of the aortic valve;
 - arterial disproportion: $\text{diametrAo/D LA} = 0.5$ and 0.38;

- the diameter of the ductus arteriosus 4 and 4.5 mm (22 and 23 ned.gestatsii);
 - inability to visualize the aortic arch;

- in one case — fetal hypotrophy and impaired fetoplacental blood flow.

In both cases dynamic echocardiographic control was not made: in one case — geographical distance and in the second case — fetal death due at 28 weeks gestation (at the autopsy — interruption of the aortic arch type C). In one case, after the birth, Computed Tomography-angiography (Fig. 4) — interruption of the aortic arch type B, with karyotypic analysis — deletions of chromosome p22q11. Newborn was operated successfully at the age of two days. In one of two cases, in 3D-image reconstruction, the interruption of the aortic arch can clearly be seen (Fig. 3).

6 of 8 cases (75%) with verified postnatal diagnosis — coarctation of the aorta, which needed in surgical correction - characterized by a variety of degree of obstruction and concomitant extracardiac disease. 2 of 6 (33%) of cases of coarctation of the aorta had the presence of multiple fetal malformations: in 1 case — kidney aplasia and dysplasia of the right hand and fetal hypotrophy, in the case of 2-facial cleft in combination with polyhydramnios. In these cases, the suspicion of disease of the cardiovascular system in fetuses arised at 27 and 30 weeks of pregnancy.

In both cases, it was noted:

- the presence of ventricular disproportion $RV/LV=1.5$ and 1.6,
- the ratio of isthmus diameter to the diameter of ductus arteriosus 0.5 and 0.7,
- in one case ventricular septal defect — 6mm and arterial disproportion — 0.7 and diffuse pericarditis,
- in the second case — the central muscular defects — 3 mm and defect aorto-pulmonary septum — 5 mm and the absence of arterial disproportion. Both infants had low Apgar score — 4–5 and 4–6 scores, and died fa the age of 2 and 3 days. The diagnosis was verified at autopsy.

In 1 of 6 cases aortic coarctation (16%) the manifestation of obstructive process of the aortic arch was at 23 weeks' gestation:

- the presence of arterial disproportion (0.6),
- a low the ratio of isthmus diameter to the diameter of ductus arteriosus — 0.4,
- multiple ventricular septal defects with a maximum size apical localization — 5 mm
- no ventricular disproportion.

So, prenatal diagnosis at 23 weeks gestation — tubular hypoplasia of the aortic arch, critical coarctation.

Dynamic examination at 32 weeks showed the ultrasound image changed: no ventricular and arterial imbalances, multiple ventricular septal defects wiyh

max — 5,4 mm, the diameter of the ductus arteriosus — 6 mm, the area of the isthmus was not visualized in 2D mode and 3D-multiplanar scanning. Prenatal diagnosis — interruption of the aortic arch.

After birth — critical aortic coarctation with tubular hypoplasia were diagnosed in CT-angiography. The child was operated on the 5th day of life and died on the 10th day after the surgical.

In 3 of 6 cases of aortic coarctation (50%) infants were successfully operated in one week of life and feel good now (Fig. 2). In these cases, prenatal ultrasound diagnostic identified:

- 1 case — ventricular disproportion — 1.4;
- in 2 cases — arterial disproportion — 0.7 and 0.72;
- in all cases, the ratio of isthmus diameter to the diameter of ductus arteriosus — 0.6–0.7;
- in 1 case — the bicuspid aortic valve; in one case — aortic kinking of the proximal descending aorta;
- in 2 cases — ventricular septal defect — 2.6 and 3.4 mm.

Ultrasound manifestations of this disease occurred in 1 case in 31 weeks, and in two cases — at 23 and 25 weeks of gestation. In 3D-mode in one of three cases the image of aortic arch was visualized and showed aortic coarctation, but can not determine its degree (Table 1).

DISCUSSION

The small size of the group of patient give the results of this study should be considered preliminary, to assist in determining the main directions of scientific research in the broader project.

The datasets from 15 patients suspected of having prenatal coarctation and interruption of the aortic arch demonstrates the complexity of Interpretation echographic data in forming the final diagnosis, prognosis, and the choice of further tactics.

The reason for the high percentage of false-positive results (33%, which is consistent with the data of foreign literature) is still not definite. However, analysis of the gestational age of pathological process manifestation and subsequent outcomes, reveals the following pattern. Patients with suspected coarctation or interruption of the aortic arch, identified before 25 weeks of pregnancy, did not have there were no false positive results. Pathological process manifestation in the 3rd trimester of pregnancy is accompanied by a high rate of false-positive results (50%). It is known that the aortic arch coarctation refers to progressive disease and may manifest not only in the early neonatal period at time the process of closing the ductus arteriosus, but



Fig. 1. Patient 11. Gestational age — 23 weeks. Aortic coarctation. 3D-reconstruction, multiplanar mode



Fig. 2. Patient 11. Age — 3 days. Aortic coarctation. Computed Tomography — angiography, multiplanar mode

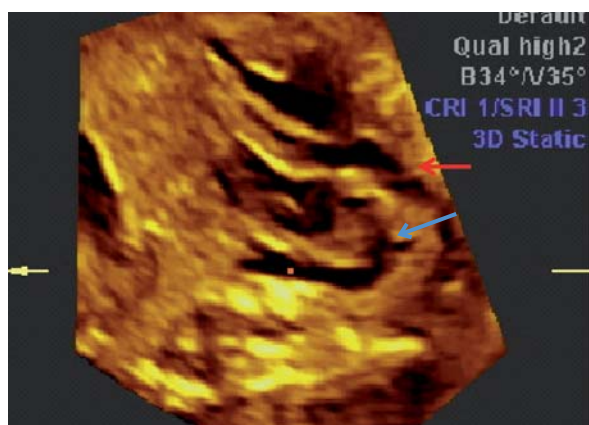


Fig. 3. Patient 8. Gestational age — 23 weeks. Interruption of the aortic arch (red arrow). 3D-reconstruction, multiplanar mode. Blue arrow — ductus arteriosus



Fig. 4 Patient 8. Age — 2 days. Computed Tomography — angiography, 3D-reconstruction. Interruption of the aortic arch, Type B (red arrow). Blue arrow — ductus

in infancy, childhood and even adolescence. The first ultrasound signs of disease process prenatal appearance is possible in the second and in the third trimester. However, the ventricular disproportion may be effect of functional disorders in transient hypoxia, abnormality function fetoplacental complex, anemia in pregnancy in the 3rd trimester of pregnancy. In this case, ventricular and arterial imbalances and physiological isthmus hypoplasia of the aortic arch in the fetus like a coarctation.

The emergence these echographic fetal heart changes during the second trimester of pregnancy is correlated with the absence of false-positive results and the maximum expression of anatomical changes of the aortic arch (aortic arch interruption and tubular hypoplasia with critical coarctation). Echographic pattern in these cases: the absence of ventricular disproportion, large ventricular septal defect (more than 5 mm), with the most pronounced in the whole study group arterial disproportion and the lowest the ratio of isthmus diam-

eter to the diameter of ductus arteriosus. Unfortunately, the echographic study in case of interruption of the aortic arch was not done. The dynamic control of fetal heart with a tubular hypoplasia and critical coarctation at 32 weeks of pregnancy showed lost of coarctation echographic signs (ventricular and arterial disproportion). It may be effect of the development of collateral circulation and adequate hemodynamic functioning right-left shunts (ductus arteriosus, ventricular septal defect and patent foramen ovale). Perhaps, this defect evolution, changing described ultrasound picture, is one of the causes of false-negative results of studies.

Group of fetuses with isolated coarctation of the aorta, which requires dynamic control after birth and requiring surgery, were similar ultrasound picture of the poor echographic manifestations. Group had a unexpressed (or absence) of ventricular disproportion

and arterial disproportion, ratio of the diameter of the isthmus to the diameter of the ductus arteriosus close near to normal, the presence of small ventricular septal defect (up to 3.5 mm), or their absence. However, important is early appearance of these signs (the 2nd trimester) in the case of coarctation requiring surgical treatment and late manifestation of the process (in the 3rd trimester) cases requiring medical supervision only. Poor echographic manifestations of defect in this group are causes the difficulty of diagnosis and prediction of disease progression.

The complexity of interpreting the data in case of a combination of aortic coarctation with multiple malformations of the fetus is to change the influence of echocardiographic picture abnormal intracardiac anatomy and the violation of the fetoplacental circulation with the occurrence of hypoxia. Echographic pattern generally reflects the effect on the anatomy of the heart combination of different factors.

The main method of prenatal diagnosis disease of the aortic arch main method, in our view, is the two-dimensional imaging mode while 3/4D-modes are optional, but very usefull in several cases. One of the obvious drawbacks of the method is worse resolution three-dimensional mode to 2D-mode. Significant loss of image quality is observed in the construction of virtual planes that do not coincide with the plane in the direction of the initial scan (4-chamber section of the heart), whereas in the transvers-sections of the image quality does not change significantly. This explains the tendency of foreign researchers to analyze the anatomy of the heart in transverse planes.

Nevertheless, the most obvious advantages of the method of volume reconstruction should be noted: 1. The possibility of obtaining images of virtual planes, hard to the study of the fetus in two-dimensional mode, but having a certain diagnostic value. 2. The possibility of studying the fetal heart in the mode off-line. 3. Transferability pick up "heart volume" in the form of digital data for remote consulting physicians for primary diagnosis and dynamic monitoring patient.

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