J. Fetal Med. (December 2022) 9:121–124
https://doi.org/10.1007/s40556-022-00348-4

CASE REPORTS

Prenatal Diagnosis of Ductus Arteriosus Aneurysm: A Case Report and Literature Review

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Received: 27 October 2021 / Accepted: 12 July 2022 / Published online: 7 November 2022

Abstract The ductus arteriosus (DA) is a vascular shunt between the main pulmonary artery and the proximal descending aorta, allowing the oxygenated blood from the placenta to bypass the fetal pulmonary circulation and supply oxygen-rich blood to the systemic circulation. Anomalies within the ductus arteriosus can compromise fetal circulation. Ductus arteriosus aneurysm (DAA) is a localized saccular or tubular dilation of the DA and is considered a rare lesion. However, many reports in the literature describe DAA in the infancy period, with limited reports describing the lesion prenatally. We diagnosed a case of ductus arteriosus aneurysm with mild hypoplasia of the transverse arch prenatally at a 37-week gestational age. Both resolved spontaneously in the third week of life without complication.

Keywords Ductus arteriosus aneurysm · Prenatal · Fetal echocardiography

Case Report

A 38-year-old G8P3 woman was referred for fetal echocardiography at 37-weeks gestation with a history of seizures and on antiepileptic medication. She previously had three miscarriages, one due to blighted ovum and others for unknown reasons and she also had one termination of pregnancy for gestational trisomy 18 at 19 weeks of gestation age. Her current fetus had a normal chromosome complement on amniocentesis. She was a mother of three healthy children. There was no family history of connective tissue disease or genetic disorders, and there is no history of diabetes mellitus or systemic hypertension during pregnancy. Fetal echocardiography showed a normal four-chamber view, left ventricle outflow tract view, and bicaval view. The sagittal view showed a large ductus arteriosus aneurysm measuring15mm x 9 mm connecting to the main pulmonary artery proximal to the left pulmonary artery origin with mild arch hypoplasia at the ductus arteriousus insertion point. Color flow showed a turbulent 'swirling' appearance within the aneurysm with normal antegrade flow into the descending aorta. Three vessel view, the most crucial view, was obtained by sliding the transducer cranially from the four-chamber plane toward the upper fetal mediastinum. It demonstrated round cross-sections of the main pulmonary artery, the ascending aorta, the superior vena cava, and the abnormally large saccular vessel representing DAA. Color Doppler of three vessels' view showed swirling flow within the ductus aneurysm (Fig. 1). No thrombus in the aneurysm, arrhythmia, or other complications were noticed on fetal echocardiography. Spontaneous vaginal delivery and postnatal echo were recommended. The pregnancy continued on an uneventful course. After delivery, the baby was stable and asymptomatic with a normal Apgar score. A primary postnatal echocardiogram on the first day of life showed a large tortuous patent ductus arteriosus measuring 3 mm with a significant aneurysm measuring10mm x10mm at the pulmonary artery insertion. However, there was no arch obstruction or pulmonary artery compression (Fig. 2). Another follow-up echocardiogram in the third week of life

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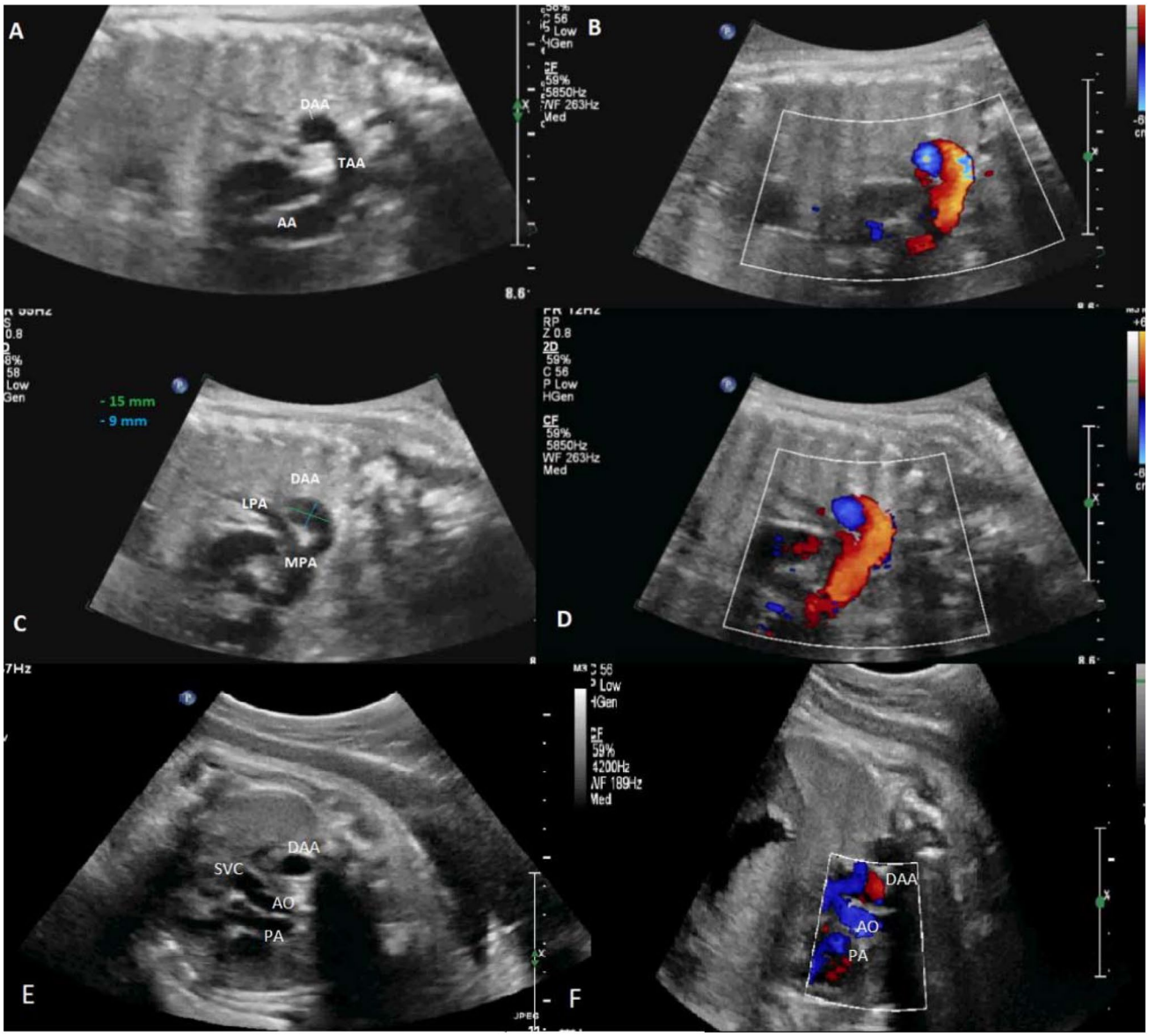

Fig. 1 Fetal echocardiography: **a** The 2D sagittal view shows a large ductus arteriosus aneurysm with mild hypoplastic transverse aortic arch. **b** The Color Doppler sagittal view showing swirling flow in ductus arteriosus aneurysm. **c** 2D view showed a large ductus arteriosus aneurysm joining the main pulmonary artery. **d** Color Doppler view showing swirling flow in ductus arteriosus aneurysm. **e** 2D view shows an abnormal three-vessel view in the presence of a large vessel in relation to the aorta and pulmonary artery, which represented DAA. **f** Color Doppler three-vessel view showing swirling flow in DAA. DAA: ductus arteriosus aneurysm, TAA: transverse aortic arch. MPA: main pulmonary artery. LPA: left pulmonary artery. SVC: superior vena cave. AO: aorta, PA: pulmonary artery

showed complete resolution of the DAA with spontaneous patent ductal arteriosus closure.

Discussion

The mechanism of ductus aneurysmal formation remains unclear. However, the possible causes of DA aneurysm include inadequate fibronectin secreted by the subendothelial proliferation of smooth muscle cells and abnormal elastin, collagen deposition, congenital weakening of the ductal wall, abnormal intimal cushion formation, and defective elastin [1]. Helen Taussig suggested that delayed closure of the aortic end in the ductus arteriosus results in exposure of the ductal wall to systemic pressure, which may cause a DA aneurysm [2]. DA aneurysms have been associated with chromosomal abnormalities such as trisomy 21 and 13, or syndromes such

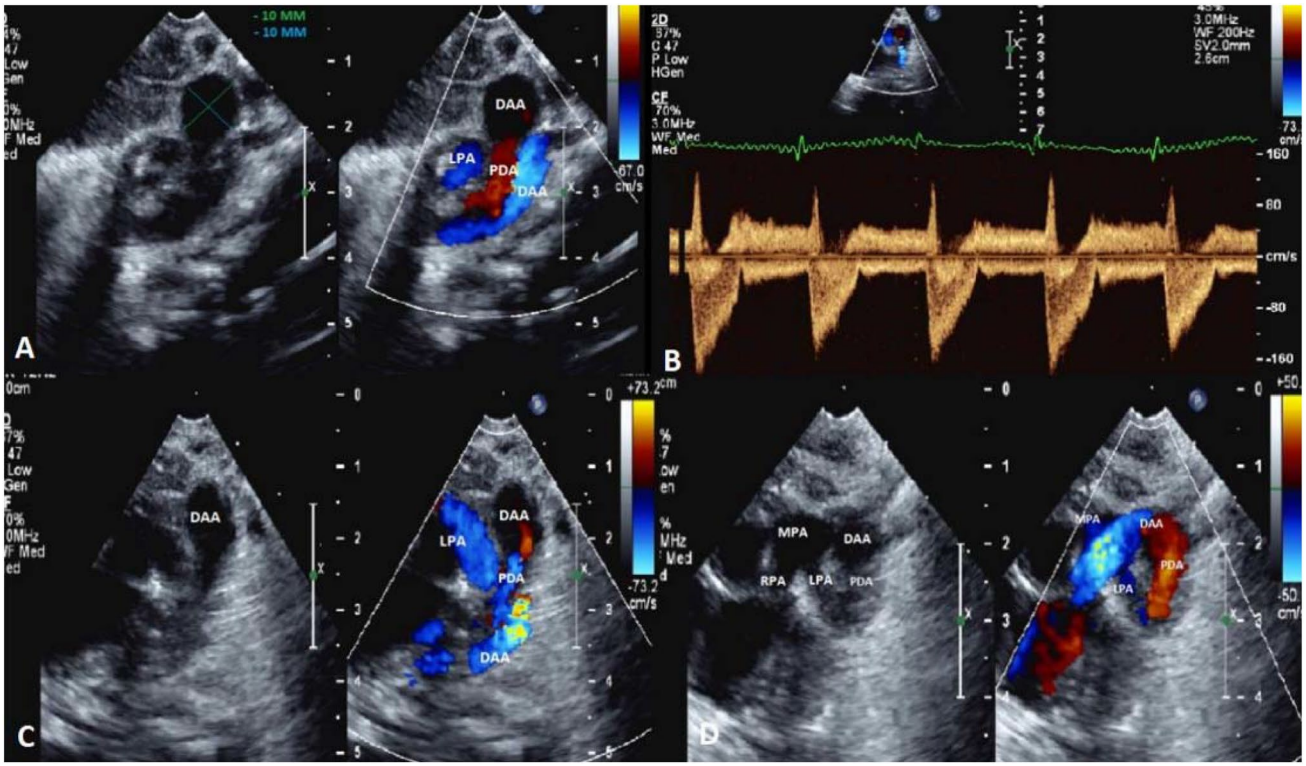

Fig. 2 Transthoracic echocardiography: **a** Ductal view showing patent ductus arteriosus and large saccular ductus arteriosus aneurysm in 2D and color Doppler. **b** Continuous-wave (CW) Doppler interrogation of ductus arteriosus showed bidirectional flow due to swirling flow in DAA. **c** Modified ductal view 2D and color Doppler showed swirling flow in ductus arteriosus aneurysm. **d** Modified ductal view 2D and color Doppler showed the flow of ductus arteriosus and ductus arteriosus aneurysm to the main pulmonary artery. DAA: ductus arteriosus aneurysm, PDA: patent ductus arteriosus, DAO: Descending aorta. MPA: main pulmonary artery. LPA: Left pulmonary artery, RPA right pulmonary artery

as Smith-Lemli-Opitz syndrome, and have been described in connective tissue diseases such as Marfan and Ehlers-Danlos syndrome, as well as Larsen syndrome. [3, 4]. Fetal echocardiography is the first modality in the diagnosis of DAAs. However, there are no specific diagnostic criteria for defining a neonatal DAA. Some authors consider the ductus arteriosus to be dilated if it is greater than 95% of the DA's average width or saccular dilatation with a maximal internal diameter equal to or larger than the adjacent transverse arch or descending aorta [1]. The absolute size of DAAs in the reported series ranged from 6.5 to 24 mm in maximal diameter, with some authors considering a DAA diameter greater than 10 mm as the criteria for a large DAA [3]. Others classified DAAs as small (< or = 7 mm.) or large (> or = 8 mm.) based on their diameter. The smaller ones commonly close spontaneously in 70% of the cases, while the larger ones are associated with more complications [5]. Most ductus arteriosus aneurysms resolve spontaneously. However, serious complications include thromboembolism, compression of adjacent structures, left pulmonary artery stenosis, and fatal spontaneous rupture or dissection [6, 7]. Lund et al. reviewed sixty-five cases and found a complication rate of 31% in infants younger than two months, while it increased significantly to 66% in children between 2 months and 15 years. Therefore, the author suggested urgent surgical intervention for DAA patients older than two months [4]. Dyamenahalli et al. previously reported nine patients with a prenatal diagnosis of DAA. Although all patients were asymptomatic, three were complicated by thrombus formation within the aneurysm [3]. Sheridan et al. reported a fetus with DAA, which was detected early in pregnancy at a 22-week-gestational age and was complicated by massive thrombosis in the pulmonary artery and fetal hydrops that ended in fetal death [8]. Similar to our case, DAA was diagnosed antenatally. However, the fetus in our report was asymptomatic and was delivered in a stable condition without complications. Ganesan et al. reported a fetus with DAA measuring 10 mm that was identified at 34 weeks gestational age. The pregnancy was uneventful, and the infant was asymptomatic after birth. The DAA had spontaneously resolved without complications around the third week after delivery

[9]. Similar to our report, the DAA was diagnosed prenatally in the third trimester, and the fetus was asymptomatic with an uncomplicated pregnancy. Most of the patients with DAA are asymptomatic and need only observation. However, surgical resection of the ductal aneurysm has been proposed if the DAA persists beyond the neonatal stage or DAA with thrombus formation extending into adjacent vessels or DAA with significant compression of adjacent structures [3]. Careful monitoring of fetuses with DAA and early detection of complications requires interdisciplinary care, including collaboration with fetal and pediatric cardiologists and perinatal specialists.

Conclusions

Three vessel view imaging in Fetal Echocardiography plays a pertinent role in the identification of DAA and should be advocated especially in the third trimester. Though most DAA aneurysms resolve spontaneously, complications may occur in the prenatal and neonatal periods. Hence a postnatal cardiac evaluation by a pediatric cardiologist is recommended in fetuses with DAA.

Acknowledgements None.

Source of Funding None.

Declarations

Conflict of interest None.

Ethical approval The study was approved by King Faisal Specialist Hospital and Research Center with Abdullah International Medical Research Center (KFSH-RC).

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