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Case Report

Condition that Simulate the Patent Ductus Arteriosus that Closed by a New Novel

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Introduction

The diagnosis of aorto-atrial fistula is a challenging matter. The cause is due to rare cases that can be diagnosed until-now. The high incidence of suspicion is mandatory to complete the diagnosis. The presence of abnormal facial features is one of the guide for diagnosis of this anomaly. There are two classification of aorto-atrial fistula, the first one is related to the opening of the fistula and the other is related to the ending point of fistula.

In relation to the opening point, there are three type according to the origin from one of the three cusps of the aorta. However, the other classification is related to the drainage point to the atrium either to the right or to the left one.

Totally there are 12 types of aorto-atrial fistula and the clear diagnosis is done by using contrast computed tomography and well delineation done by the cardiac characterization. Once diagnosis by cardiac catheterization done. The closure is the solution to prevent more inter-cardiac shunt flow. The closure is important to avoid complications as thrombus formation and fistula rupture.

Our Case

A female girl, full term single vaginal delivery. Stable vital signs. All laboratory investigations were normal. The patient shows facial signs of Di gorge syndrome, laboratory genetic study was sent.

The cardiac examination shows a mummer at the right parasternal area at the level of fourth intercostal space. The mummer is not common to be heard in this area. The echocardiography is done. But the diagnosis not matches with the site of the audible mummer.

Echocardiography show initily diagnosis as large PDA (Figure 1A). At that time no need to start any medication. Follow up at the age of 1 week, the patient developed complete symptom and signs of heart failure. Anti-failure medication were started and follow up after 2 weeks shows no improvement. Echocardiography follow up done and shows abnormal S shape vessel that is not seen before (Figure 1 B). This is explained due to high right atrium pressure post-delivery that prevent shunting to be clear unless the pulmonary pressure decrease. The diagnosis is supported by the absence of contentious flow of PDA and the pulsitil wave of the artery was seen (Figure 1 C).

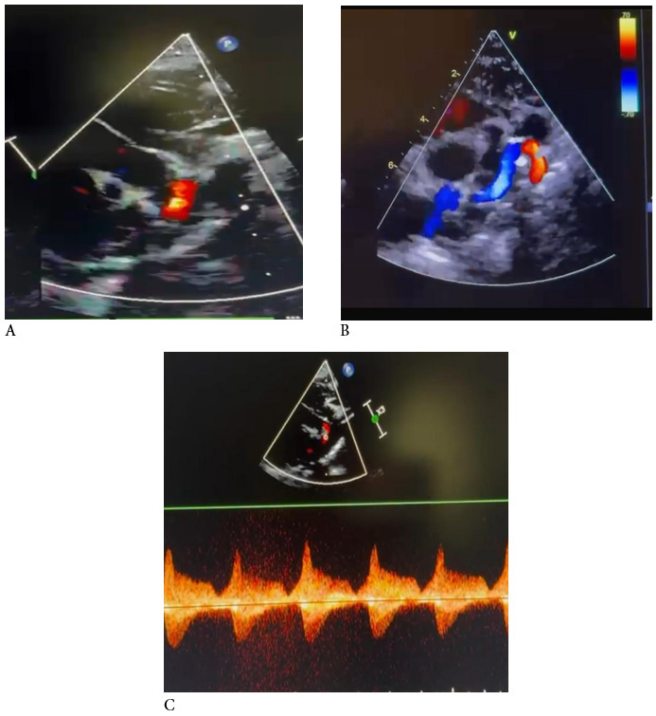


Figure 1A: Echocardiography at the level of the PDA window shows the flow like PDA that give false result in which the lateral angulation to the prope give the next image with a characteristic abnormal S shape vessel can be diagnosed (B) and the confirmation done by absence of the contentious murmur that commonly seen in PDA (C).

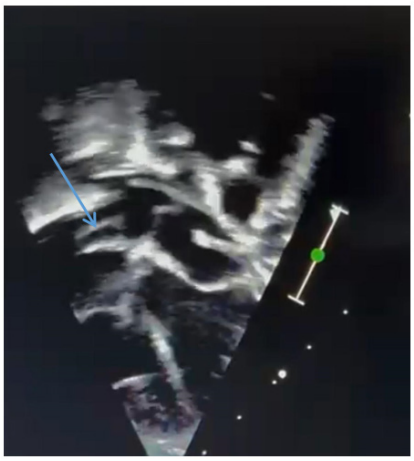


Figure 2: The mouth of the fistula is seen open in the right atrium (arrow). that take origin from the aortic right cusp.

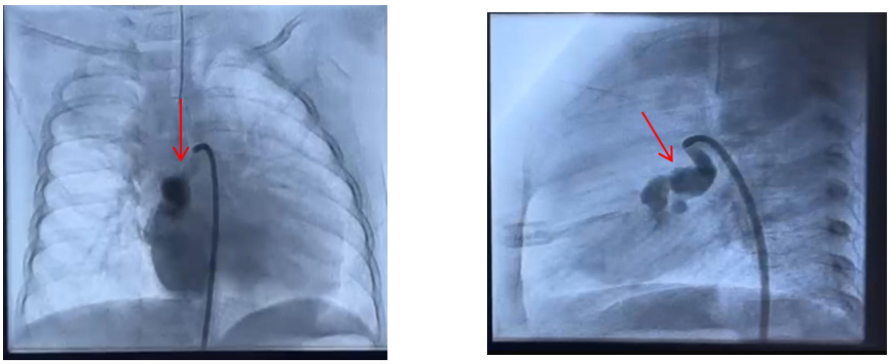
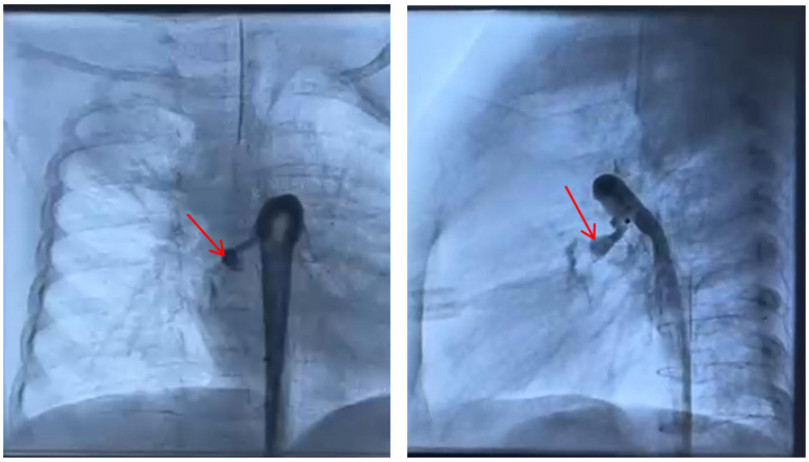


Figure 3A: Shows an anterio posterior view of the aortography at the origin of fistula. The dye pass through the vessele and drain to the right atrium (Arrow). B: Lateral view of the aortography in which the S shape fistula is clearly seen (Arrow).



A B

Figure 4A: Shows an anterio posterior view of the aortography shows no more dye pass in the fistula. The device is well fixed in the mouth of the fistula (Arrow). B: Lateral view of the aortography there is absence of flow through the previous S shape vessel S shape fistula is clearly seen (Arrow).

Follow up echocardiography show (Figure 2) the mouth of the fistula is seen opening in the right atrium. The right atrium was dilated and there is turbulence flow extend to the right atrium.

For clear diagnosis cardiac caheterization was done and aortography on both anterioposterior and lateral views were done in both pre device closure (Figure 3) and post device closure (Figure 4).

Method

Trancathere closure done. The use of a 16 mm Amplatzer vascular plug II (AGA Medical). Through a femoral approach, a 4 Fr venous and a 4 Fr arterial sheath were used. A 3 Fr Concierge Amplatz Left 2 guiding catheter was positioned in the aorta at the fistula entrance and a 0.035 in Terumo hydrophilic guide wire was advanced along the fistula, until the right atrium was reached. The wire was then snared and an arteriovenous loop was created. A 7 Fr Amplatzer Delivery System was used to deploy the device through the atrial end.

Discussion

The presence of abnormal connection between the aorta and the atrium is accidentally discovered. The patient may be asymptomatic or suffering from unexplained heart failure.

Any patient shows signs of Di George syndrome should have a detailed echocardiography and looking for this anomaly.

The pharyngeal arch arteries are important in the development of aorta and neck branches. Any abnormal connection seen in relation to the aorta is highly occurred due to abnormal genetic code for the development of aotro-atrial connection. The fistula is due to resorption of normal developing structure with down regulation of the function of mesenchymal cells.

The cause is due to changes of the progenitors arise from second heart field (SHF). These progenitors responsible for regulation of the development of endothelial cells which line the (PPAs). In cases of decreased numbers of SHF- derived vascular progenitors as in case of absence of VEGFR2, an angiogenetic response take place from the nearby veins and venous-derived cells migrate into pharyngeal arches, proliferate, incorporate into the PAAs, acquire arterial identity, and allow for morphogenesis of new vascular network ended by vascular connection [1-5].

Therefore, there are two genetic code that result in aorta- atrial connection. These are Tbx1 and VEGFR2 (vascular endothelial growth factor receptor 2) are responsible for the development and the number of SHF-derived endothelial cells (ECs).

The endothelium of the pharyngeal arch arteries (PAAs) is formed from the mesodermal core of the arches, whereas the vascular smooth muscle is predominantly cardiac neural crest [6-9].

There are two forms, either an aorto-atrial tunnel or a aorto-atrial fistula. The aorto-atrial tunnel is due to overregulation of mesenchymal cells [9]. This result in a true new vessel formation that connect between the aorta and the atrium. The fistula is due to resorption of normal developing structure with down regulation of the function of mesenchymal cells.

Conclusion

Any abnormal murmur detected in site away from the normally position of hearing should be examined carefully for this abnormal vessel. The closure of the aorto-atrial fistula is mandatory to prevent further complication.

The patient with Di Gorge syndrome may have abnormal genetic code that need further evaluation as abnormal vessels can be developed.

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