

Lung Shadow in Neonate: An Unusual and Treatable Cause of Cyanosis

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Abstract

Persistence of lung shadow is usually diagnosed and treated as congenital pneumonia. Sometimes clinical status cannot be co-related by the disease. We came across a similar situation where a neonate presented with lung shadow and mild cyanosis since birth due to an uncommon cause and diagnosed as pulmonary arteriovenous malformation. He was treated by percutaneous closure of arteriovenous malformation.

Keywords: Lung shadow; Pulmonary AV fistula; Device closure

Case Report

A 13 days old baby, 2nd of twins, Full term small for gestational age, weighing 1.5 kg, having history of poor feeding since day 5 of life, was referred to us with suspected congenital heart disease. Baby had mild cyanosis (SpO₂ in room air = 84%) with systemic examination findings being unremarkable. Chest radiograph showed rounded opacity in right lower lung field (Figure 1). Echocardiography done with Phillips iE-33 with broad band transducer at our centre revealed small muscular ventricular septal defect (VSD), small patent ductus arteriosus (PDA) with left to right shunt. In view of desaturation and unexplainable cyanosis, contrast echocardiography was done which revealed filling of left atrium and ventricle after 3 beats suggestive of pulmonary arteriovenous malformation (PAVM). Computed Tomography (CT) angiography scan was done to see detail anatomy of the lesion and it confirmed single large pulmonary arteriovenous malformation of lower lobe of the right lung. In view of stable clinical condition with stable oxygen saturation he was advised close follow up.

Baby was brought to us at 4 months of age with severe respiratory distress and cyanosis came. His oxygen saturation in the room air was 55 to 60%. Repeat Echocardiography showed spontaneous closure of VSD and PDA, normal ventricular function and dimension. Therefore it was decided to close the PAVM percutaneously.

Cardiac catheterization and device closure of PAVM was done under general anaesthesia with proper antibiotic cover. On right pulmonary artery angiogram we observed almost immediate filling of left atrium (Figures 2a and 2b). There were multiple AV malformations



Figure 1: Chest X-ray showing rounded opacity in right lower lung (encircled).



Figure 2a: Angiogram showing RPA and filling of LA in the same frame through PAVM.

involving right middle and lower lobe with three major feeding vessels measuring 6 mm, 5 mm and 4 mm which were closed by Amplatzer vascular plug II 12 mm, Amplatzer vascular plug II 10 mm and Amplatzer vascular plug IV 8 mm respectively (Figures 2c, 2d and 3a, 3b). Saturation improved drastically from 71% (on 100% FiO₂) to 100% (Po₂=248 on 60% FiO₂) immediately after the procedure. Most important precaution to be taken during the procedure is to avoid thromboembolism as one is working in pulmonary vein. Procedure was performed uneventfully. Baby was extubated in cardiac catheterization lab only after completion of procedure and got discharged on next day of procedure.

Discussion

Central cyanosis in newborns and infants is usually due to cardiovascular and respiratory causes. High index of suspicion is what

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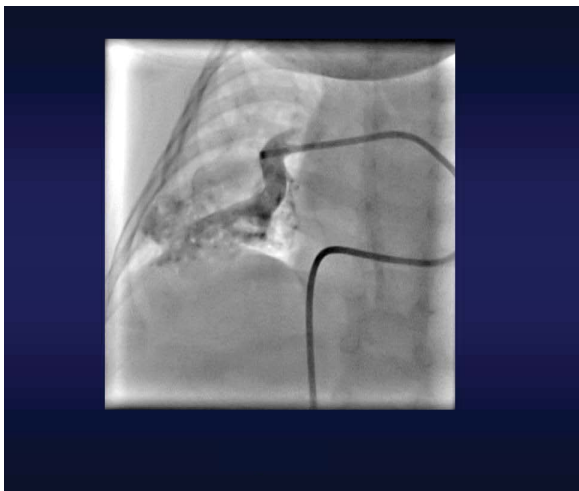


Figure 2b: RPA angiogram showing large feeding channel to PAVM.



Figure 2c: Angiogram showing occlusion of this feeding channel through AVP. Two more channels seen opacified after the occlusion of first-one.

is required if we have to reach an uncommon diagnosis like pulmonary AV malformation. Basic investigation like chest X-Ray is important to provide a clue to the diagnosis. Our case was misdiagnosed as Transposition of great vessel in a peripheral centre. Misdiagnosis in Pediatric echocardiography is well reported [1] especially if it is not done by a trained Pediatric echocardiographer. Once the diagnosis was made, at that stage we decided to keep baby under close follow up as he was clinically stable and was maintaining relatively good oxygen saturation.

Pulmonary AV

Pulmonary arteriovenous malformations are abnormal direct communications between pulmonary arteries and pulmonary veins bypassing the capillary circulation with an aneurysmal dilatation [2-4]. Effectively it causes intrapulmonary right to left shunting leading to cyanosis. It could be congenital or acquired [2-4]. Hereditary hemorrhagic telangiectasia or hereditary generalized angiomas are frequent causes of congenital PAVMs. AV fistulae in the lungs can be

single or multiple and known to progress. Symptomatology depends upon the extent of the disease. In the current scenario, recommendations suggest that all PAVMs should be treated with embolisation therapy and that surgery is to be reserved for individuals with PAVMs that are not amenable to embolisation or have other contraindications to embolisation, such as an allergy to contrast material [5]. Surgical options like ligation of fistula, Segmental resection or lobectomy are more invasive and more morbid. Taylor et al. [6] pioneered the technique of therapeutic embolization of pulmonary artery. Recently Amplatzer Vascular Plug (AVP) has become preferred choice of device to embolize PAVM [7-10]. Indications for closure of pulmonary AVMs are hypoxemia, neurological symptoms (transient ischemic attacks, migraine, stroke, brain abscess), hemorrhage, progressive enlargement of the lesion, and presence of a 3 mm or larger feeding vessel [2-4].

When our case started showing signs of severe hypoxaemia, we took him for transcatheter closure of PAVMs. As per CT report we



Figure 3a: Post procedure fluoroscopy showing three vascular plugs in the right lower lung.

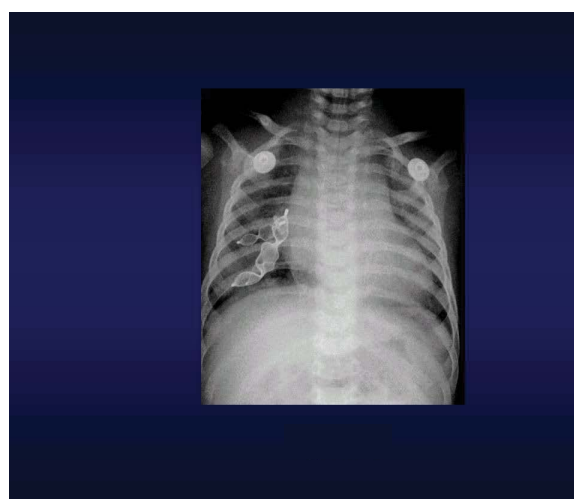


Figure 3b: Post procedure chest X-ray showing three vascular plugs in the right lower lung.

were expecting one feeding vessel to the malformation. On contrast injection of RPA, initially we saw one major feeding vessel but as we closed it, another channel was seen filling the malformation which was also closed (by another vascular plug). Again on repeat injection one more feeding channel was noticed that also needed closure. As soon as we closed third channel, oxygen saturation increased to 100%. Final RPA angiogram showed complete closure of the fistula. Tricky situation in our case was opening up of new feeding channels one after the other which often occurs as reported in the past [11]. When channels are small and multiple, it is difficult to occlude all of them. Additionally there are chances that segmental branches of pulmonary artery are blocked if multiple devices are deployed leading to risk of lung infarction. Fortunately our patient was not too much cyanosed in the newborn period at weight of 1.5 kg; else, the risk of complications could have been more.

Conclusion

Few lessons from our case are important for the clinicians (1) Unusual causes of cyanosis in newborn and infants should always be kept in the mind. (2) Even chest X-Ray can point towards noncardiac cause of cyanosis. (3) Cyanotic infant must be referred for evaluation at Pediatric cardiac centre. (4) Detailed echocardiography and CT pulmonary angiography both are usually required to reach the anatomical details of the PAVM. (5) A very safe nonsurgical intervention exists for the rare cyanotic disease like PAVM in infants.

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