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CASE REPORT
Congenital heart disease

Foetoscopy-assisted balloon valvuloplasty in a human foetus with disadvantageous intrauterine position: a case report

Thomas Kohl [1]*, Ibrahim Akin[2], Juliane Frommberger[3], Nadja Riehle[1], and Dietmar Schranz[1]

[1]German Center for Fetal Surgery & Minimally-Invasive Therapy (DZFT), Mannheim University Hospital (UMM), Theodor-Kutzer-Ufer 1-3, 68167 Mannheim, Germany; [2]Department of Cardiology, Mannheim University Hospital (UMM), Theodor-Kutzer-Ufer 1-3, 68167 Mannheim, Germany; and [3]Department of Anesthesiology, Mannheim University Hospital (UMM), Theodor-Kutzer-Ufer 1-3, 68167 Mannheim, Germany

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Background Some foetuses scheduled for balloon valvuloplasty present with unfavourable lies that render a successful procedure unlikely or impossible. In these situations, foetal posturing previously has been achieved by maternal laparotomy. As a less invasive means, we demonstrate the feasibility of a minimally invasive foetoscopic approach.

Case summary Percutaneous ultrasound-guided foetal balloon valvuloplasty for severe aortic valve stenosis was attempted in a human foetus at 29 + 4 weeks of gestation under general maternofoetal anaesthesia. Unfortunately, prior to the procedure, the foetus had been observed on several occasions remaining in a dorsoanterior cephalic position. Therefore, the left ventricle could not be accessed by the conventional percutaneous ultrasound-guided approach. In order to achieve the desired foetal lie, foetoscopic assistance was employed: using a standardized foetoscopic setup, a foetoscope and two graspers, the foetus was rotated in dorsoposterior position. After this manoeuver, successful balloon valvuloplasty was achieved. Mother and foetus tolerated the procedure well and complications were not observed.

Discussion Foetoscopy-assisted foetal posturing offers itself as an alternative to maternal laparotomy in foetuses presenting with a persisting disadvantageous position at the time of balloon valvuloplasty. Due to the increased risks of preterm rupture of membranes and earlier delivery posed by the foetoscopic approach, this technique may preferably be used in more mature foetuses when foetal posturing cannot be achieved by other means.

Keywords Case report • Foetus • Aortic stenosis • Pulmonary atresia • Valvuloplasty • Intervention • Foetoscopy • Foetal surgery

Learning points

- Foetal valvuloplasty is performed in human foetuses to preserve left or right ventricles from the detrimental effects of severe semilunar valve obstructions.
- Foetoscopy-assisted foetal posturing offers itself as an alternative to maternal laparotomy in foetuses presenting with a persisting disadvantageous position at the time of foetal valvuloplasty.

* Corresponding author. Tel: +49 175 597 1213, Fax +49 621 383 5797, Email thomas.kohl@umm.de
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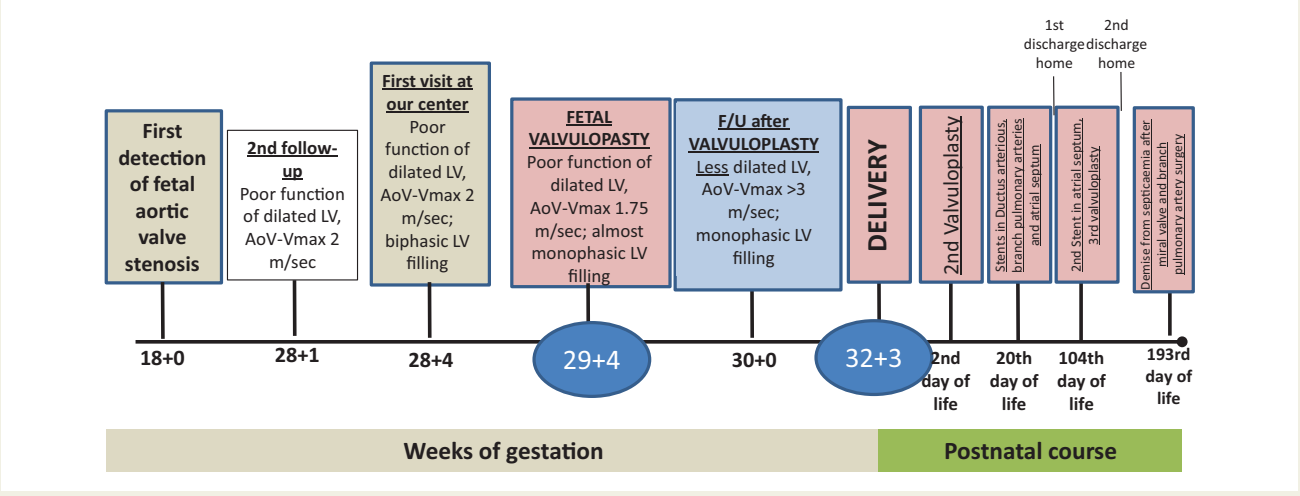
Introduction

Ultrasound-guided foetal valvuloplasties have been performed since the late 1980s in foetuses with severe aortic valve obstruction in order to safe their left ventricles for a postnatal biventricular circulation.[1–5] Whereas the procedure is in the truest sense of the word *straightforward* when the foetus presents in a favourable dorsoposterior position, some foetuses present repeatedly with unfavourable positions that render the procedure impossible. In order to overcome this obstacle, maternal laparotomy has been used to facilitate moving the foetus into a position suitable for intervention.[6] Our case report describes the use of foetoscopic assistance as a less invasive means for improving foetal position.

Timeline



Case presentation

A 31-year-old pregnant woman—with no family history of congenital heart disease—had been referred to our centre because of foetal aortic valve stenosis, first detected at 18 + 0 weeks of gestation. Maternal transabdominal foetal ultrasound imaging at 28 + 4 weeks of gestation now revealed severe foetal aortic valve stenosis with a poorly contracting dilated left ventricle, severe mitral valve regurgitation, and local endocardial fibroelastosis. Severe mitral regurgitation most likely results from papillary muscle ischaemia. In foetuses with severe aortic valve stenosis, the mitral valve apparatus is most often the first area affected by the development of endocardial fibroelastosis. The mitral valve inflow was still biphasic and the regurgitant velocity exceeded 4 m. The maximum systolic flow velocity across the thickened aortic valve was 2 m/s. The flow across the oval foramen was left to right and inside the aortic arch retrogradely.

In order to salvage the function of the left ventricle, percutaneous ultrasound-guided foetal balloon valvuloplasty was attempted at 29 + 4 weeks of gestation under general maternofoetal anaesthesia. At that time, the function of the left ventricle had deteriorated further as the mitral valve flow integral exhibited now mostly an a-wave filling. As a technical obstacle to the procedure, the foetus had been consistently observed lying in a dorsoanterior cephalic position. Therefore, the left ventricle pointed posteriorly and could not be accessed by the conventional percutaneous ultrasound-guided approach.

In order to achieve the desired foetal position with the least maternal procedural trauma, foetoscopic assistance was employed: three 11 Fr catheter sheaths were percutaneously placed along the left flank of the foetus into the amniotic cavity (*Figure 1*). In order to improve foetal visualization, the amniotic cavity was then insufflated with carbon dioxide. Following these manoeuvers, using a foetoscope and two graspers introduced through the trocars, the foetus was rotated in dorsoposterior position and the upper extremities postured along the foetal sides (*Figure 2*).

After successful foetoscopic foetal posturing (*Figure 3*), the insufflation gas was removed and an 18 gauge needle percutaneously advanced into the foetal left ventricle and placed underneath the obstructed aortic valve by maternal transabdominal foetal echocardiographic guidance (*Figure 4*). Successful foetal balloon valvuloplasty was then achieved via the needle shaft employing a 3.5 mm—coronary angioplasty catheter (*Figure 2*). Mother and foetus tolerated the procedure well and complications were not observed. At the end of the procedure, all interventional materials were removed and the maternal skin incisions closed with single stitches. In the days after foetal balloon valvuloplasty marked improvement of flow across the aortic valve and an increase in flow velocity from 1.75 m/s prior to the intervention to more than 3 m/s after the intervention were documented (*Figure 5*).

In our case, a semi-compliant balloon with an inflated diameter of 3.5 mm was used that fit through the shaft on an 18 gauge needle. As

Figure 1 Top—External setup during foetoscopy-assisted posturing of the foetus in the carbon dioxide insufflated amniotic cavity via three 11 Fr trocars that were placed along the left uterine wall. Bottom—External setup during the exact moment of the inflation of the valvuloplasty catheter across the aortic valve.

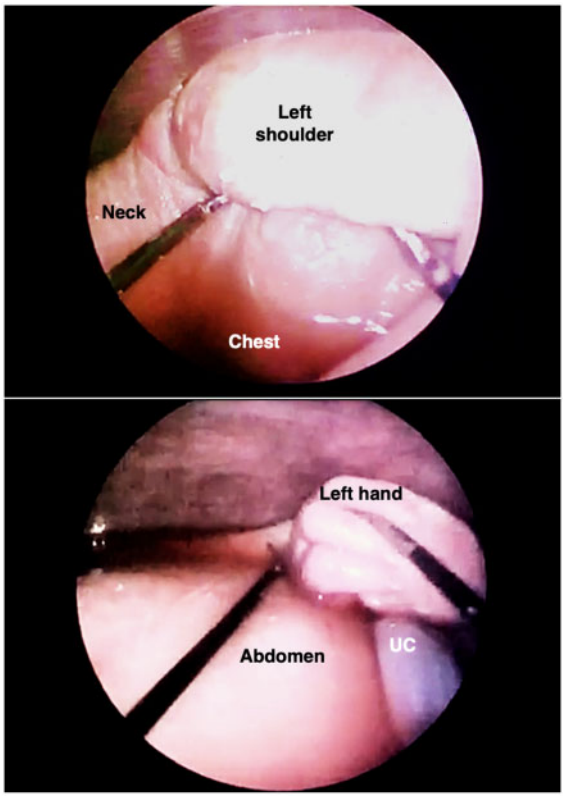


Figure 2 Top—Following partial carbon dioxide insufflation of the amniotic cavity in order to optimize visualization of intra-amniotic contents, the foetus is being rotated 180 degrees from its dorsoanterior position. Bottom—After having achieved the desired dorsoposterior lie, the left arm is also moved aside in order to achieve unhindered access into the foetal left ventricle.

a result, the inflated diameter of the balloon in this 29 + 4 weeks old foetus was smaller than the aortic annulus (4.5 mm—z = +0.1). Whereas decompression and improved function of the left ventricle were observed for about 2 weeks, with further foetal growth, the fixed stenosis became more and more effective again. This was the main reason, why the foetus already was delivered in the third week after prenatal intervention and scheduled for postnatal re-valvuloplasty on the second day of life with a 5 mm balloon. At the time of resubmission of this report, the infant has undergone stent insertions in the ductus arteriosus and atrial septum, as well as insertion of flow occluders into both pulmonary branches. By these interventions, the foetal circulation can be maintained and blood flow into the pulmonary arteries be reduced. In addition, two more aortic valvuloplasties were carried out. Using these measures, left ventricular function improved over time. Unfortunately, the baby died at 6 months of age from *Escherichia coli* septicaemia most likely from a long-standing central venous line after another cardiac surgery addressing its mitral valve regurgitation and branch pulmonary artery obstruction.

The percutaneous ultrasound-guided and foetoscopy-assisted procedure had been performed following parental informed consent and in accordance with the ethical standards for human experimentation established by the Declaration of Helsinki.

Discussion

Our case shows that in foetuses with unfavourable positions, foetoscopy-assisted foetal posturing can be employed in order to improve foetal position and enable balloon valvuloplasty for severe semilunar valve obstruction. The minimally invasive approach offers itself as a less invasive alternative to maternal laparotomy. This more invasive approach has previously been used by other teams in order to enable interventions in foetuses presenting with disadvantageous positions at the time of intervention.[6]

Apart from optimizing foetal position for percutaneous ultrasound-guided foetal balloon valvuloplasties, foetoscopic assistance has been employed by our group for foetal transoesophageal echocardiography during intervention.[7] Furthermore, the three-trocar technique combined with insufflation will be a prerequisite for minimally invasive foetoscopic pacemaker insertion.[8]

The minimally invasive techniques for foetal cardiac intervention were initially developed in foetal sheep models and—combined with a safe anaesthesia protocol for human foetoscopic surgery—clinically used for foetal surgery for spina bifida and for haemodynamically compromised foetuses with congenital high airway obstruction

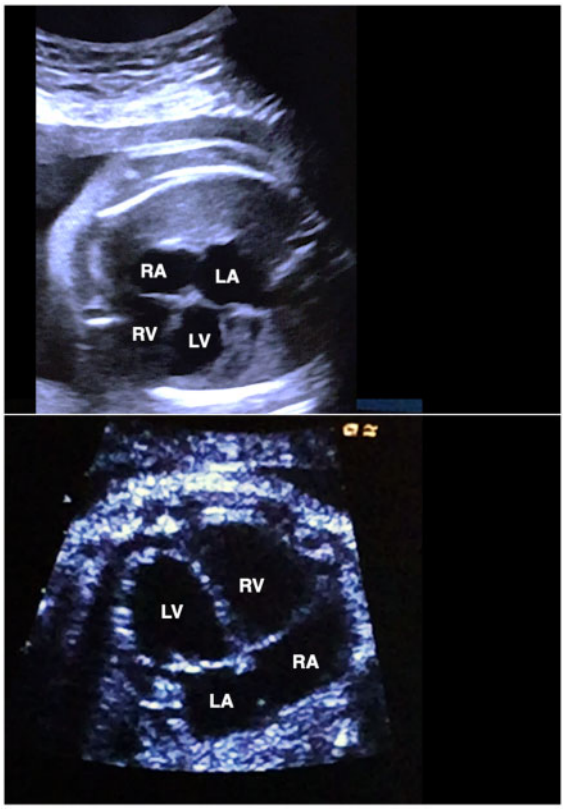


Figure 3 Top—This image demonstrates the unfavourable lie of the foetus before posturing. With the apex of the left ventricle pointing to the posterior uterine wall, percutaneous puncture of the left ventricle is not possible. Bottom—This image demonstrates the favourable lie of the foetus following successful foetoscopy-assisted posturing. After evacuation of the insufflation gas, percutaneous transuterine access into the foetal left ventricle can now easily be achieved. LA, left atrium; LV, left ventricle; RA, right atrium; RV, right ventricle.

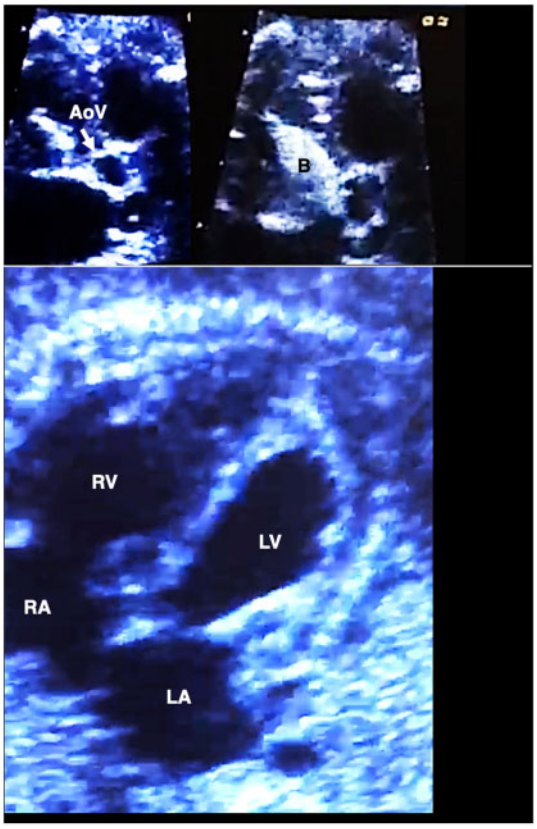


Figure 4 Top left—The needle shaft (left to the arrow) can be seen underneath the aortic valve (arrow). Top right—Ultrasound image during inflation of the 8 mm long balloon with a diameter of 3.5 mm. Bottom—This image demonstrates the decompressed, now smaller left ventricle following successful balloon valvuloplasty of the aortic valve. AoV, aortic valve; LA, left atrium; LV, left ventricle; RA, right atrium; RV, right ventricle.

syndrome.[9–13] From these previous experiences, we assumed that the approach would most likely be technically successful but also be safe and well tolerated by both mother and her foetus with a cardiac malformation.

The most common complication is the development of amniotic fluid leakage, occurring at a mean of 29 weeks of gestation.[14] Despite this problem, a mean age at delivery of ~33 weeks of gestation is reached with maternal bedrest and infection prophylaxis. Given the high quality of neonatal cardiac intensive care management, interventions, and surgery, this age usually suffices to take care of a premature infant with still impaired ventricular function.

In conclusion, foetoscopy-assisted foetal posturing offers itself as a less invasive alternative to maternal laparotomy in foetuses with persisting disadvantageous positions at the time of balloon valvuloplasty.

Lead author biography

Prof. Dr med. Thomas Kohl leads the German Center for Fetal Surgery & Minimally-Invasive Surgery (DZFT) at Mannheim University. He is one of the pioneers in the field of foetal cardiac intervention. His most important contribution is the non-invasive treatment of hypoplastic cardiovascular foetal structures by chronic intermittent hyperoxygenation.

Supplementary material

Supplementary material is available at *European Heart Journal - Case Reports* online.

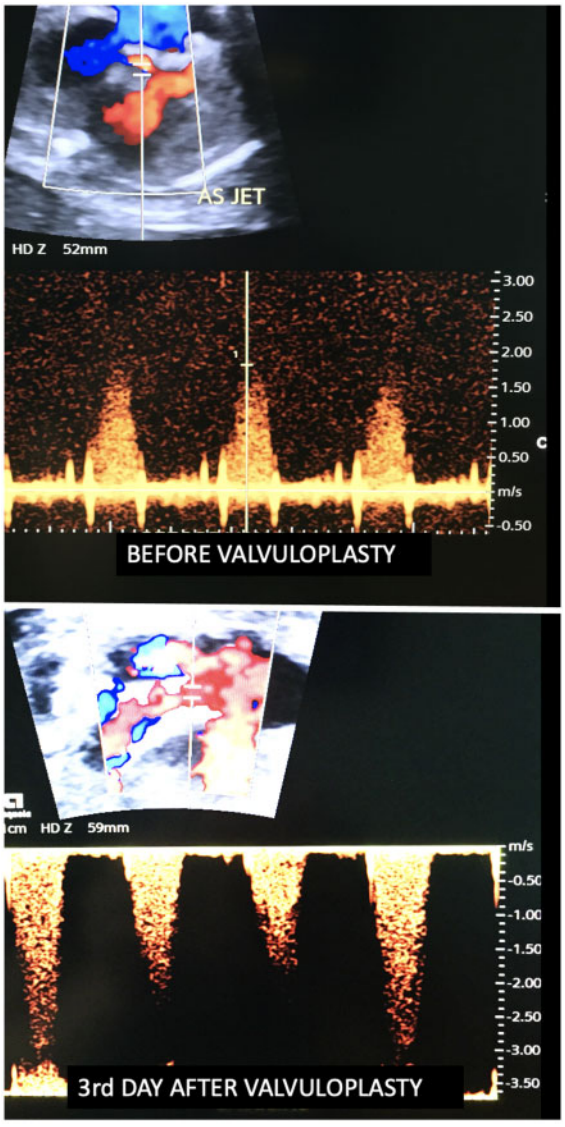


Figure 5 Top—Doppler interrogation before and—bottom—on the third day after foetoscopy-assisted foetal balloon valvuloplasty shows an increase of the flow velocity across the aortic valve from ~1.75 m/s before to more than 3 m/s after the intervention.

Slide sets: A fully edited slide set detailing this case and suitable for local presentation is available online as Supplementary data.

Consent: The authors confirm that written consent for submission and publication of this case report including images and associated text has been obtained from the patient in line with COPE guidance.

Conflict of interest: None declared.

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