

Failure of Balloon Dilatation in Mid-cavity Obstruction of the Systemic Venous Atrium After the Mustard Operation

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SUMMARY. Mid-cavity obstruction of the systemic venous atrium developed after the Mustard operation in a child with transposition of the great arteries. Balloon dilatation (BD) was performed twice, to a maximum theoretical transverse diameter of 18 mm. Each time obstruction was initially relieved, but recurred within months. The usefulness of balloon dilatation therapy requires long-term follow-up. Results from currently reported experience do not suggest a major therapeutic role for this procedure in children.

KEY WORDS: Mid-cavity obstruction — Systemic venous atrium — Balloon dilatation — Interatrial baffle

Case Report

The patient underwent 2 cardiac catheterizations and a Mustard procedure at other institutions. The first catheterization performed at 1 day of age revealed dextro-transposition of the great arteries, intact ventricular septum and mesocardia; a balloon atrial septostomy was performed with good clinical result. Pre-operative cardiac catheterization at 14 months of age demonstrated low left ventricular pressure, equalization of pressure across the atrial septum, and an aortic saturation of 82%. Mustard operation was performed at 20 months of age (4/6/79) using pericardium to construct the interatrial baffle. During closure of the sternotomy, a cardiac arrest occurred requiring external cardiac compression. Prior to hospital discharge, a right-to-left atrial shunt was indicated by radionuclide studies.

At the first catheterization at Children's Hospital of San Diego, a large right-to-left shunt was confirmed: the aortic saturation was 82% and the ratio of pulmonary-to-systemic flow (Qp/Qs) was 0.5. The interatrial baffle was disrupted at its anterosuperior edge. Although a narrow area was apparent

angiographically within the systemic venous atrium, no significant gradient was detected across this region; pressure in the superior vena cava was normal (mean = 5 mm Hg). Other pressures were also normal for a patient after venous switch operation: left ventricle = 24/6, main pulmonary artery = 20/10, right ventricle = 90/7, and ascending aorta = 90/60 mm Hg.

A revision of the interatrial baffle was performed at age 3 years (10/9/80) using dacron to close the atrial septal defect. The pericardium previously used to create the interatrial baffle was noted to be thick. Subsequently, the patient began to develop cyanosis and edema of the head as well as bulging of the neck veins, most noticeable during exercise. At repeat catheterization (5/15/81), there was no residual interatrial communication, pressure was elevated (24 mm Hg) in the superior vena cava, and a discrete area of mid-cavity obstruction was seen in the systemic venous atrium (Fig. 1-A, B).

After obtaining informed consent, a BD was performed on 7/7/81 (Fig. 1-C, D) and the initial result was considered satisfactory (Table 1; Fig. 1-E, F). Relief of superior vena cava syndrome was noted clinically. Three months after the dilatation procedure, symptoms recurred. The patient's hemodynamic, as well as angiographic, condition (Table 1 and Fig. 2-G, H) had returned completely to predilatation status. A 2nd BD was performed (10/14/81; Fig. 2-I, J) again with initially good effect: pressure gradient was reduced, flow increased, and the obstruction was less severe angiographically (Fig. 2-K, L). At most recent evaluation, 7 months after the 2nd dilatation procedure,

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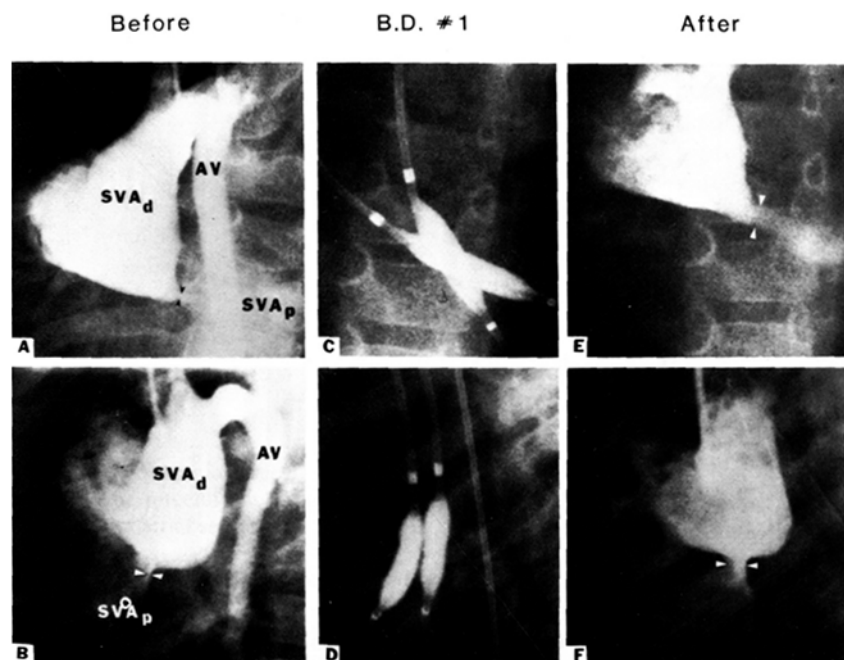


Fig. 1. Angiograms before and immediately after first BD of mid-cavity obstruction in the systemic venous atrium. Each pair of panels includes an anteroposterior frame above a lateral frame. A & B Before 1st BD, severe obstruction is seen between the distal systemic venous atrium (SVA_d) near the superior vena cava, and the proximal venous atrium (SVA_p) near the mitral valve. AV = azygous vein. C & D First BD procedure using 2 balloons with an aggregate transverse diameter of 12 mm. E & F Immediately after BD #1, when the mean gradient was decreased from 16 to 8 mm, enlargement of the narrowed area is seen. (Compare with A & B).

Table 1. Effects of Balloon Dilatation (BD)

Date	Condition	Mean Pressure (mm Hg)		
		SVA distal	SVA proximal	Gradient
7/7/81	Pre-BD	20	4	16
	Post-BD	15	7	8
10/14/81	Pre-BD	20	3.5	16.5
	Post-BD	8	6	2
4/15/82	—	18	4	14

the patient is asymptomatic but has returned, angiographically and hemodynamically (Fig. 3-M, N; Table 1), to his predilatation status.

Materials and Methods

Pressures at catheterization were recorded through fluid-filled systems after zero reference was determined by radiographic method [16]. Gradients in the systemic venous atrium were determined without a catheter across the obstruction. Before dilatation therapy, an angiogram in the superior vena cava confirmed that a tiny opening persisted between the 2 compartments in the systemic venous atrium (Fig. 1-A, B). Balloon dilatation was performed with Medi-Tech BD catheters (Stanco) introduced through the right and left basilic veins; the balloons were 6 mm wide when inflated and 20 mm long and the catheters were 6/20. The obstruction was first dilated with 6 rapid pulses of inflation/deflation at 60 to 65 PSI delivered by a Gruntzig balloon inflation pressure regulator (USCI). At first inflation, a marked indentation was noted on the balloon, but this was only minimally present at the 6th inflation. A 2nd 6/20 catheter was passed across the obstruction with both balloons deflated. Via a "Y"-

connector, both catheters were connected to the Gruntzig pressure regulator and both balloons were inflated/deflated rapidly 5 times at a pressure of 60 to 65 PSI. Again, an indentation was seen at commencement of bi-balloon dilatation which was essentially abolished at the final inflation (Fig. 1-C, D). The maximum theoretical transverse diameter achieved by the two 6 mm wide balloons was 12 mm; however, the actual transverse dimension was probably less than 12 mm because the obstruction deformed the balloon(s) to some degree.

At the 2nd BD, the same general procedure was used. Two balloon dilatation catheters were introduced (6/20 and 12/40) percutaneously via the right internal jugular vein and the right femoral vein through No. 9 French sheaths. Rapid inflation/deflation was done for eight pulses, using the 6/20 catheter from above. The 12/40 catheter was then advanced from below. The 40 mm length of the catheter complicated the therapy since in most positions, when inflated, the long catheter occluded both the obstruction and the inferior vena cava, thereby preventing all systemic venous return. After 4 brief trial inflations, a catheter position was found that allowed maintenance of systemic arterial pressure. Six bi-balloon inflation/deflation cycles were used at 60 to 65 PSI, the longest inflation period being 90 seconds. Even after the series of dilatations, indentation is noted (Fig. 2-I, J) in both balloons, indicating a transverse diameter less than the theoretical maximum of 18 mm (6 + 12). With both catheters remaining inflated, the 6/20 catheter was withdrawn into the distal systemic venous atrium.

Discussion

Balloon dilatation of obstructive lesions has been shown to be beneficial in some forms of coronary artery disease [2, 4, 5, 14], in peripheral arterial obstruction [6, 7, 10, 15], and in 1 patient with pulmonary veno-occlusive disease [12]. Recently,

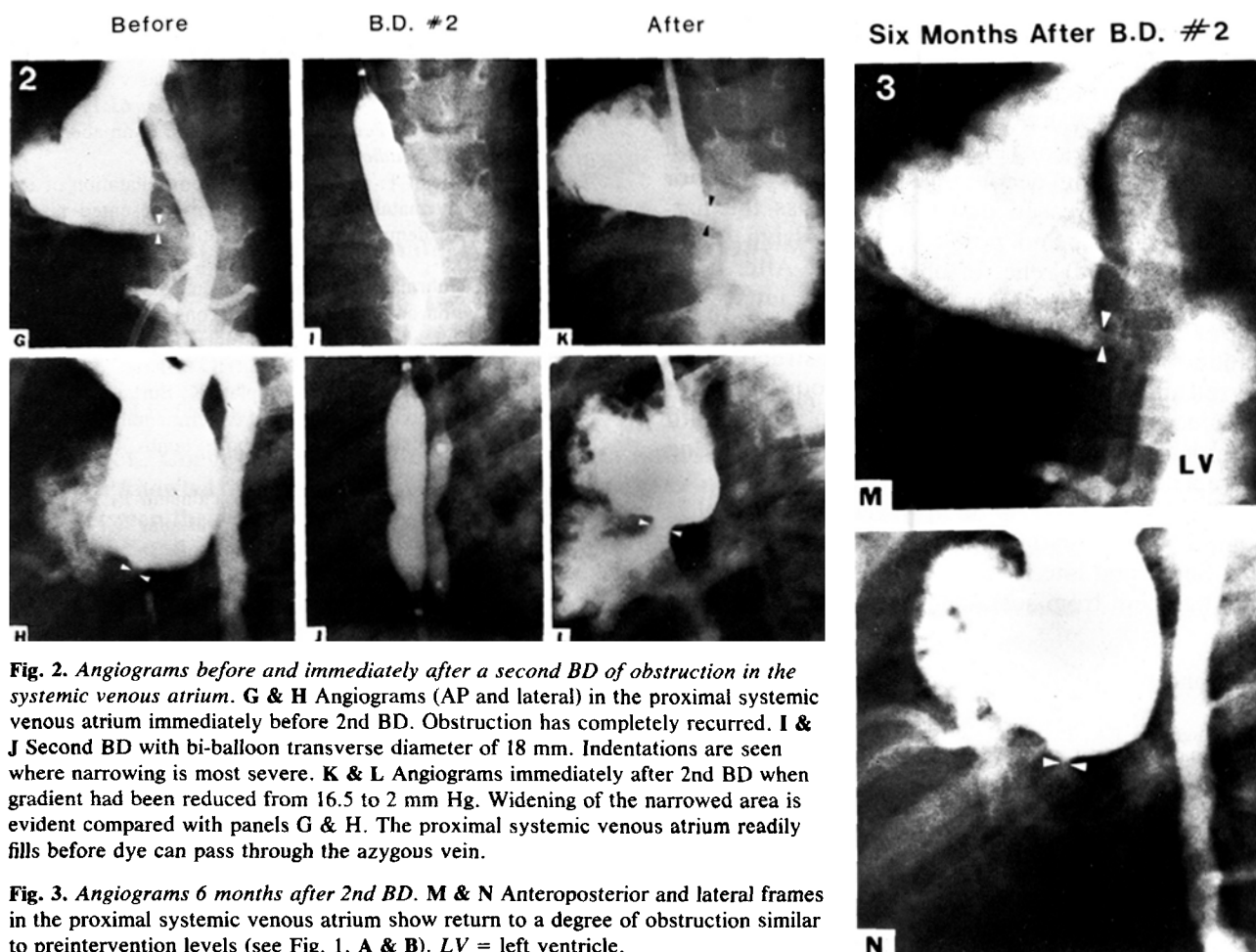


Fig. 2. Angiograms before and immediately after a second BD of obstruction in the systemic venous atrium. **G & H** Angiograms (AP and lateral) in the proximal systemic venous atrium immediately before 2nd BD. Obstruction has completely recurred. **I & J** Second BD with bi-balloon transverse diameter of 18 mm. Indentations are seen where narrowing is most severe. **K & L** Angiograms immediately after 2nd BD when gradient had been reduced from 16.5 to 2 mm Hg. Widening of the narrowed area is evident compared with panels **G & H**. The proximal systemic venous atrium readily fills before dye can pass through the azygous vein.

Fig. 3. Angiograms 6 months after 2nd BD. **M & N** Anteroposterior and lateral frames in the proximal systemic venous atrium show return to a degree of obstruction similar to preintervention levels (see Fig. 1, **A & B**). **LV** = left ventricle.

there has been considerable interest in this mode of therapy for children with obstructive cardiovascular defects, either congenital or postsurgical.

Balloon dilatation has been studied in experimental models of congenital heart disease, specifically coarctation of the aorta [7] and branch pulmonary artery stenosis [1]; relief of obstruction was found after balloon dilatation. However, both animal studies and postmortem human heart data [3] indicate splitting of the vascular intima and media on pathologic inspection. It is unknown if this is a deleterious side effect [7] or the mechanism of improvement [1].

Driscoll et al [3] reported results of BD therapy in infants with pulmonary vein stenosis; relief of obstruction was shown hemodynamically, with a decrease in gradient after balloon dilatation. However, the stenoses uniformly recurred.

Lock et al [8] used BD therapy in 4 children with congenital obstructions: 2 infants with pulmonary vein stenosis, 1 infant with coarctation of the aorta and atrioventricular canal defect, and 1 teenager with multiple narrowings in the brachiocephalic

vessels. The 3 infants had no significant improvement and all died. Three children with left pulmonary artery stenosis after repair of tetralogy of Fallot experienced moderate improvement after BD therapy. Lock concluded that this approach was of little benefit in pulmonary vein stenosis but can be "moderately successful" in pulmonary artery stenosis.

In a recent case report, Rocchini et al [13] described a 15-month-old child with total anomalous pulmonary venous drainage who developed severe postoperative obstruction at the intraatrial baffle; reoperation did not relieve the pulmonary venous hypertension. Percutaneous transluminal angioplasty was performed (maximum transverse diameter = 6 mm) decreasing the gradient from 30 to 16 mm Hg. Fourteen hours after the procedure, the baby died and autopsy examination revealed endothelial desquamation and disruption of the vena cava interna at the site of balloon dilatation. The authors concluded that the death of their patient "should not deter attempts at angioplasty in other patients with superior vena cava obstruction."

In our patient, we believed that the postoperative obstruction was in part due to scarring and adhesions of the (resected) atrial septum and the interatrial baffle. It was hoped that a fibrotic element in the mid-cavity lesion (Fig. 1-A, B) would be broken by BD, allowing decompression of the superior vena cava, increased direct flow across the narrowed segment, and possibly, progressive (or at least maintained) relief of obstruction. After completion of each dilatation, a significant immediate improvement was confirmed, similar to Rocchini's patient [13]. However, twice the obstruction recurred and it seems likely that the balloon dilatation stretched the narrowed area but (as in infants with pulmonary vein stenosis) did not provide lasting therapeutic benefit.

Although BD technique may yet prove useful in some children, initial experience is not encouraging. Serial and late follow-up is necessary to separate transient from sustained effects.

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