

# RIGHT VENTRICULAR OUTFLOW TRACT PSEUDO-ANEURYSM AFTER RECONSTRUCTION WITH SMALL INTESTINAL SUBMUCOSAL (CORMATRIX) PATCH – A WORD OF CAUTION

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## Abstract

*Tetralogy of Fallot is very prevalent with correction techniques well standardized. Whenever infundibular incisions are needed, patch reconstruction seems mandatory. Recently, the small intestinal submucosal (CorMatrix, MAC's Medical Group,) patch was introduced, with optimal results in pre-clinical studies. However, clinical results do not match its pre-clinical promises, particularly when used in right ventricular outflow tract and pulmonary artery reconstructions. We describe a case of Tetralogy of Fallot for which small intestinal submucosal (CorMatrix) patch was used as a trans-annular patch, with development of a massive pseudo-aneurysm.*

## INTRODUCTION

Tetralogy of Fallot (TOF) is very prevalent with correction techniques that are well standardized - all producing predictable results with an immediate mortality risk below 2%.<sup>1</sup> Whenever infundibular incisions are needed, patch reconstruction seems mandatory and options vary between synthetic and biological materials.<sup>2</sup> Recently, small intestinal submucosal (CorMatrix) patch was introduced and claimed having perfect stability and low incidence of retraction and calcification, given its decellularization process.<sup>3</sup> We describe a case of TOF for which small intestinal submucosal (CorMatrix) patch was used as a trans-annular patch, with apparent immediate good results but soon developing a massive pseudo-aneurysm and severe right ventricle out-flow tract obstruction (RVOTO).

## MATERIAL AND METHODS

A 5 years old patient with TOF and hypoplastic intrapulmonary pulmonary arteries was initially submitted to a right modified Blalock Taussig shunt in 2013. Five years later, he underwent total correction, consisting of trans-atrial ventricular septal defect closure with a Dacron

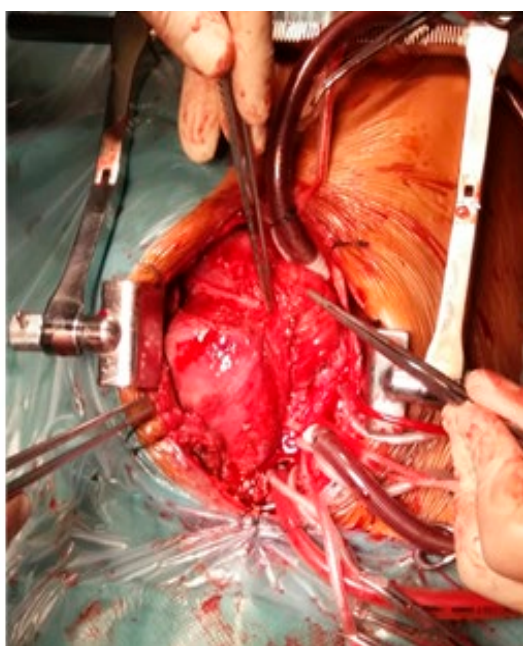
patch and RVOTO relieve by muscular section and extended infundibular, main pulmonary trunk and bifurcation enlargement, using a trans-annular single small intestinal submucosal (CorMatrix) patch. Taking into consideration the hypoplastic pulmonary arteries, we created a 3mm atrial septal defect. Weaning of bypass was uneventful with direct systolic right ventricle pressure measurement of 38 mm Hg. We collected blood samples at the pulmonary artery, superior vena cava and aorta and performed gasometric analyze. There was no evidence of intra-cardiac shunt, despite the presence of an iatrogenic atrial septal defect. Immediate postoperative period was uneventful. An echocardiogram at discharge revealed no residual pressure gradient across RVOT, but a residual gradient of 70 mmHg was present at the level of pulmonary artery (PA) origin, bilaterally. There was mild pulmonary valve regurgitation. Taking into consideration that the patient was clinically asymptomatic, with pulse oximetry values of 99-100%, he was discharged home, with indication of echocardiographic reevaluation one month later. The echocardiogram revealed systemic right ventricle pressure and stenosis at the origin of right PA (5,7 mm) and left PA (3 mm). Pulmonary regurgitation was now free. There were no signs of infection. An angio-CT scan evidenced a giant RVOT pseudo-aneurysm (66 x 46 mm) compressing pulmonary

**Figure 1**

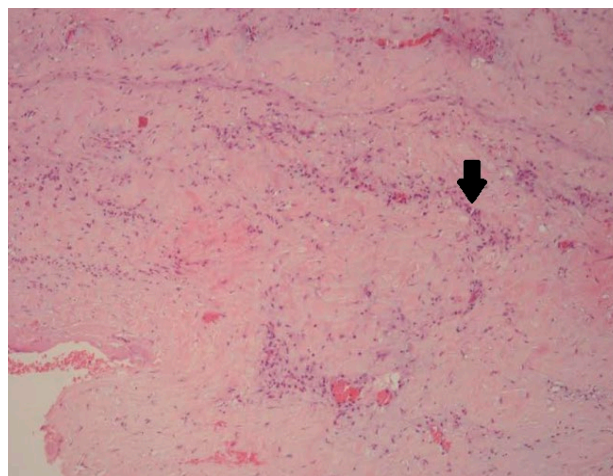
Angio-TC showing pseudo-aneurysm dimension and space relationship.

bifurcation and producing bilateral branch stenosis (Figure 1). Patient was scheduled for urgent reoperation. RVOT area was dissected, revealing a massive pseudo-aneurysm (Figure 2), the wall consisting of patch remains and organized clots. Visually, the patch seemed to have been disintegrated. There was no evidence of rupture of the suture lines. There was massive inflammation and adhesions, the

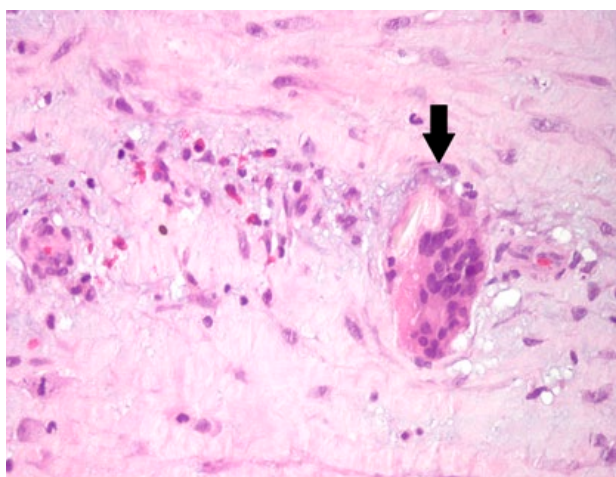
PA bifurcation was compressed and grossly distorted by the pseudo-aneurysm. The pseudo-aneurysm was resected and RVOT continuity was re-established using a Contegra 16 graft interposed between the slit open bifurcation and the ventricular incision. A patch of pericardium was used to augment the origin of both PA's. Post-operative period was uneventful. A fragment of the pseudo-aneurysm wall was sent for pathology, showing small fragments of residual patch material, engulfed in fibrosis, hemorrhage and mononuclear cell infiltrate consisting on eosinophils and giant multinuclear cells in a foreign body reaction (Figure 3 and 4).

**Figure 2**

Intra-operative appearance of the pseudo-aneurysm.

**Figure 3**

Histological exam of pseudo-aneurysm wall with a small amplification with H&E showing marked eosinophilic infiltration (arrow).

**Figure 4**

*Histological exam of pseudo-aneurysm wall with a great amplification with H&E showing a giant multinucleated cell in a foreign body reaction (arrow).*

## RESULTS

We describe a case of TOF and hypoplastic arteries, corrected by a trans annular patch using small intestinal submucosal (CorMatrix) patch. One month later, the patch material was disintegrated leaving place to a massive and fast-growing pseudo-aneurysm, compressing PA bifurcation. There was no infection by clinical and laboratory methods and no evidence of rupture of the suture lines.

## DISCUSSION

Small intestinal submucosal (CorMatrix) patch was firstly used in 2010 and received FDA approval as a pericardial substitute and for intra cardiac corrections. This is a de-cellularized patch having the needed protein structure to ensure constructive remodelling. The reabsorption process starts early but extends till 3-6 months, in pre-clinical studies. Supposedly the host tissue would replace the patch material between 3 and 6 months. Previous reports showed conflicting results regarding the use of this type of patch for RVOT and or pulmonary reconstruction, namely in the paediatric group, with a very high rate of reoperations. Among these, there are two cases reporting TOF repair, both needing reoperation, at 2 months and 13 months,

and showing similar pathology findings, as for our case.<sup>4</sup> The only multicentric study evolving over 100 cases with small intestinal submucosal (CorMatrix) patch also revealed a high incidence of reoperation, particularly on the right side of the heart.<sup>5</sup> Clinical results regarding the use of small intestinal submucosal (CorMatrix) patch do not match its pre-clinical promises, particularly for use in the RVOT and pulmonary artery reconstructions.<sup>6</sup>

In our case, the absence of signs or symptoms of infection and the integrity of the suture lines suggest that the pseudo-aneurysm resulted of the characteristics of the patch material, in the context of residual right ventricular hypertension.

## CONCLUSION

We understand it is a single case report. However, in view to literature evidence and our present case, small intestinal submucosal (CorMatrix) patch should be used carefully for right heart reconstructions.

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