

SURGICAL CORRECTION OF A PULMONARY ARTERY BRANCH ORIGINATING ABNORMALLY FROM AORTA

1Arben Baboci, 1Edvin Prifti, 2Massimo Bonacchi, 1Altin Veshti, 1Numilla Kuneshka, 2Vittorio Vanini

1. Division of Cardiac Surgery, University Hospital Center of Tirana, Albania

2. Division of Cardiac Surgery, Policlinico Careggi, Florence, Italy

Implantation of the anomalous PA to the main PA trunk was performed by: I) direct anastomosis in 3 patients with AOLPA; II) interposition of a synthetic graft in one patient with AOLPA; III) employing an autologous pericardial patch in 2 patients with AORPA; IV) using an aortic flap in 4 patients with AORPA. The mean follow-up time was 37.7 months.

Results: Three patients died postoperatively due to progressive heart failure unresponsive to inotropic support. Early postoperative pulmonary hypertension crisis was identified in another patient. Within 2 years after surgery, the residual gradient across the anastomotic site was significantly lower in patients undergoing correction employing adjunctive autologous tissues, 9.5 ± 4.6 mmHg versus 21 ± 7.2 mmHg ($p=0.045$) in patients undergoing direct anastomosis or interpositioning of a synthetic graft. Similarly, the Tc-99m scintigraphy demonstrated a significantly lower lung perfusion in patients undergoing AOPA implantation without employing autologous tissues for increasing the AOPA length 57 ± 5.6 (%) versus 72 ± 4.5 (%) ($p=0.011$).

Conclusion. The AOPA from the aorta is a rare but important entity, necessitating a scrupulous preoperative and intraoperative evaluation. The techniques employing autologous tissues for enlarging and lengthening the AOPA seem to be associated with better results in terms of postoperative restenosis.

Key words: Anomalous origin of the pulmonary artery, pulmonary scintigraphy, restenosis

Introduction

Anomalous origin of one pulmonary artery (AOPA) from the aorta is a rare congenital cardiac malformation that should be distinguished from other heart defects associated with an anomalous blood supply to the lungs such as patent ductus arteriosus, major collaterals between the systemic and pulmonary circulation and truncus arteriosus (1). This type of cardiac malformation was

described firstly by Fraentzel in 1868 (2). Since then, almost 250 cases have been reported in the literature with a high mortality among patients not surgically treated (1,3-5). The AOPA from the aorta is frequently associated with other cardiac malformations (6-9) and rarely is presented as an isolated anomaly (10-12). In 1961 Armer et al (13) reported the first successful anatomic repair of anomalous origin of the right pulmonary artery (AORPA) using a polyester fiber graft. Different techniques have been successfully employed to reimplant the AOPA to the main PA trunk (MPA) such as direct implantation (11,14-15), end-to-end anastomosis with a synthetic graft (3,13), homograft patch (3-4, 14), and "aortic-ring" flap (9,12). However, postoperative restenosis across the anastomotic site is frequently observed. We are reporting our experience with the surgical repair of the AOPA including an overall literature review.

Patients and methods

Between January 1995 and March 20013, 10 consecutive patients presenting AOPA in normally connected heart underwent surgical correction. Patients with common arterial trunk with non-confluent pulmonary arteries were excluded from the study.

-Patients characteristics: The preoperative patients' characteristics are given in Table 1. Three patients presented isolated AOPA. Six patients presented AORPA and 4 AOLPA. In one case, the origin of the AOLPA was from the findings and surgeon's preference.

Technique I. Consisted in direct implantation of the AOPA to the MPA. This technique was employed in patient 3 and 6 presenting AOLPA (Figure 2A&B)(Table 1). The AOLPA was identified and encircled with a sylastic vascular loop. Then it was carefully mobilized for a long tract, including the first part of the lobar branches, and temporarily occluded with a tourniquet. The AOPA was detached from the aorta and directly anastomosed to the MPA with a continuous 6-0 Prolene suture.

The anterior half of the suture line was completed with interrupted sutures to allow tissue growth. The aortotomy was sutured primarily or with an autologous pericardial patch.

Technique II. A synthetic graft was employed in the patient 2. A standard postero-lateral thoracotomy was performed. The AOLPA was identified just beneath the ductus arteriosus. The MPA was identified easily after opening the pericardium. The AOLPA was clamped at its origin. Then the descending aorta was clamped just above and beneath the AOLPA, which was detached at its origin. A 6mm Goretex prothesis was anastomosed end-to-end to the AOLPA and then, end-to-side to the MPA.

Technique III. In patient 5, presenting AORPA from the contro postero-lateral wall of the ascending aorta and in patient 1, the distance between the AORPA and the MPA was small enough, to permit a direct posterior anastomosis between them, and later an autologous pericardial patch was employed to enlarge the anterior aspect of the anastomosis.

Technique IV. In all other patients with the AORPA originating from the right postero-lateral aspect of the ascending aorta, we prolonged the length of the AORPA using the aortic ring.

IVA (Single aortic flap technique). In patient 4, 7 and 9 (Table 1), (Figure 3A, 3B, 3C) the ascending aorta, MPA and its branches were mobilized. Then ascending aorta was transected, just above and beneath the origin of AORPA. The resulting aortic ring was fashioned into an elongated tube for the pulmonary artery and the far end was connected to the MPA side-to-Fend (10). End-to-end anastomosis of the aorta in front of the AORPA was performed (Figure 4).

IVB. (Double flap technique) In patient 8, the AORPA originated from the right antero-lateral aspect of the ascending aorta (Figure 3D and 3E). We employed a modified technique as previously described (16). The AORPA is carefully mobilized for a long tract, including the first part of the lobar branches, and temporarily closed with a tourniquet. The ascending aorta is transected obliquely, just above and beneath the AORPA, providing a symmetric large aortic ring almost 1.5 times more than the RPA diameter (Figure 5A). Then, an anterior vertical incision of MPA, at the origin of the LPA, is performed extending superiorly to the half of LPA circumference and inferiorly to the MPA. Two transversal incisions, creating a right angle with the first longitudinal incision, nearly half MPA circumference, are performed, creating a

posterior pulmonary flap (Figure 5A). Under direct vision, after measuring the length of the created pulmonary flap, the aortic ring is cut transversely, living a small aortic flap posteriorly, and an anterior aortic flap is created. The ascending aorta is retracted posteriorly and an end-to-end anastomosis is performed (Figure 5B). The pulmonary flap is sutured to the small posterior aortic flap, anterior to the ascending aorta. The anterior aortic flap is laid above the pulmonary flap and further to MPA and sutured (Figure 5C).

Results

The in-hospital mortality was 30% (three patients). Postoperatively, patient 1 (Table 1) developed low cardiac output, acute renal failure and pulmonary hypertension crisis and died in the 5th postoperative day due to progressive heart failure unresponsive to inotropic support. Patient 9 and 10 developed severe low cardiac output and died during the early postoperative period. Early postoperative pulmonary hypertension crisis was identified in patient 7 that was managed by intravenous prostacyclin. The same patient necessitated mechanical ventilation for 11 days. Chylopericardium was identified in patient 6 (Table 1), which was treated with pleural draining and parenteral nutrition during the first two postoperative weeks.

Within 1 year after surgery all patients were alived and underwent cardiac catheterisation. The mean residual gradient for all patients was 14.4 ± 8.2 mmHg. The residual gradient across the anastomotic site was significantly higher in patients undergoing correction according technique I or II, 21 ± 5.8 mmHg, than the AOPA implantation according technique III or IV 9.5 ± 4 mmHg ($p=0.026$). Similarly, the Tc-99m scintigraphy demonstrated a significantly lower lung perfusion (the lung perfused from the respective AOPA compared with the contralateral lung) in patients undergoing AOPA implantation according to technique I and II 57 ± 5.6 (%) versus patients undergoing technique III or IV 72 ± 4.5 (%) ($p=0.011$).

Table nr. 1. Preoperative demographic and clinical characteristics, early and late postoperative outcome

Patient	1	2	3	4	5	6	7	8	9	10
Variables										
<u>Demographic data</u>										
Age(days)	8	53	37	18	11	34	248	105	75	20
Sex	F	m	m	m	f	m	m	f	m	m
Weight(kg)	2.8	4.6	4.1	3.3	3.4	3.1	7.4	5.8	3.7	3
Syndromes		Waarden bourg					Di Georg e			
<u>Clinical data</u>										
Pulmonary Hypertension	IV	I	III	II	II	II	III	II	II	IV
Congestive heart failure	+	-	+	-	+	-	+	-	-	-
Pulmonary artery branch	R	L	L	R	R	L	R	R	L	R
Anomalous origin	AsAo	Thorac Ao	AsAo	AsAo	AsAo- left	AsAo	AsAo	AsAo	As Ao	As Ao
Anomal. Ass	DIV, PVS	DIV	Fallot	DIV	-	-	DIA	DIA	-	-
Rp/RS	0.45:1	0.4:1	-	-	-	0.38- 1	0.43-1	-	-	-
<u>Surgery data</u>										
Surgical procedure	RAP	Graft	DR	RAR	RAP	DR	RAR	RAR	RA R	DR
Associated surgical procedure	DIV- c,PVct	-	DIV- c	DIV- c,TP	DIV-c	-	-	DIA- c	DI Ac	-
Employment of CPB	+	-	+	+	+	+	+	+	+	+
<u>Postoperative data</u>										
Chylopericardium	-	-	-	-	-	+	-	-	-	-
Bleeding	+	-	-	+	-	-	+	-	+	-
Low cardiac output	+	-	+	-	-	-	+	-	+	+
Peritoneal dialysis	+	-	+	-	-	-	-	-	+	-
Hospital stay	5	23	18	12	28	32	22	31	1	3
Outcome	Died	Survived	Survi ved	Survi ved	Surviv ed	Survi ved	Surviv ed	Survi ved	Die d	Die d
<u>Follow-up</u>										
Follow-up interval(months)	68	61	41	33	27	24	10			
Residual gradient(mmHg)	-	29	19	15	8	15	11	4		
Status		-	OK- reop						OK	

LEGEND: LA-Left atrium; LV-Left ventricle; Ass-Associated; AsAo-Ascending aorta; m-males; f-females; L-left; R-right; DR-Direct implantation; CPB-Cardiopulmonary bypass; RAR-Implantation using the aortic ring; PDA-Persistent ductus arteriosus; DIV-Interventricular septal defect; DIV-c -Closure of the interventricular septal defect; DIA-Interatrial septal defect; DIA-c -Closure of the interatrial septal defect; PVS-Pulmonary valve stenosis; reop-reoperation; PVct-Pulmonary Commissurotomy; TP-Transannular patch enlargement of the right ventricular outflow tract

At follow-up all patients were alive. Patient 2 (Table 1) underwent reoperation at 28 months after the first surgical procedure: the employed graft presented significant stenosis in both anastomotic sites. He underwent graft's replacement employing

an 8-mm Goretex graft combined with left pulmonary arterioplasty. All survivors underwent serial echocardiographic examinations during follow-up, demonstrating a progressive regression of the ventricular hypertrophy and dimensions.

Discussion

AOPA from the aorta remains a field of investigation in cardiac surgery due to the low incidence of this congenital malformation (4,15,17). The largest reported series includes only 16 patients with this anomaly undergoing surgery over a 36 years' period (4).

Little is known about the embryogenesis and pathogenesis of this malformation, although an association has been suggested with the CATCH 22 syndrome complex (6-7, 18-19), including DiGeorge syndrome (19). Different authors have reported the important role of the neural crest cells in the development of the third and fourth pharyngeal pouch derivatives as well as the aortic arches and the trunco-conal part of the heart (20). It is plausible that deletions interesting the chromosomal band 22q11 may cause some degree of derangement in the neural crest. Similar defects have recently been described in association with the fetal Valproate syndrome (21), another condition linked to disorders of the neural crest cells migration. In our series, one patient presented the Waardenburg syndrome, which is associated with the neural crest-derived melanocyte deficiency. Aru et al (22) included this anomaly as an aortic arch anomaly complex hypothesizing that a failure of media fusion of the AOPA with the MPA results in persistence of the aortic sac from which the AOPA originates. In the absence of the left sixth arch, the AOPA may not find connection to the MPA and consequently the aortic sac connection persists.

We reviewed the age, the associated malformations and the surgical procedure undertaken through the literature during the last 30 years, in patients with anomalous origin of one pulmonary artery from the aorta undergoing repair (Table 2). There were identified 96 reports in patients with anomalous origin of one pulmonary artery from the aorta undergoing surgery. Almost 60% of patients were under 6 months of age at operation. Major associated heart defects were identified in 30% patients. The overall mortality was 16%.

The AOPA may be isolated or associated with other congenital heart defects. Most of the reported cases presented some major associated cardiovascular defect such as tetralogy of Fallot, interventricular septal defect, patent ductus arteriosus (4, 7). Surprisingly, we did not observe in any case the association with aorto-pulmonary window, interrupted aortic arch or aortic isthmal hypoplasia (18). The right aortic arch has been reported in almost 50-75% of patients with AOLPA (4,6,8).

Different cases have been reported with isolated malformation (10-12), although its incidence is very limited. In our series, there were 3 cases with isolated malformation. Two of them presented AOLPA and one AORPA.

According to some previous studies, the echocardiography is sufficient in recognising the AOPA (6,11). Complete diagnosis employing echocardiography is demonstrated by the presence of two concordant ventricular outflow tracts, absence of the usual MPA bifurcation pattern and the right or LPA arising directly from the aorta with the MPA continuing with the contralateral pulmonary branch. We believe that this is not always easily demonstrated by echocardiography alone (11). The third patient in our series, after undergoing echocardiographic examination in another institution was referred with the diagnosis of transposition of the great arteries and patent ductus arteriosus. Cardiac catheterisation showed AORPA and patent ductus arteriosus arising from the LPA. Cardiac catheterisation remains the gold standard diagnostic procedure in such cases and should be indicated preoperatively in all patients.

Most commonly the surgical correction has been achieved within the first month of age (4,15), although, successful repair has been reported in adults (10-11,17,23). Fontana et al (5) found a 30% one-year survival in a series of 23 cases with AOPA not surgically treated. Successful correction of this anomaly in the first days of life, even in prematures, has been reported (14). Early repair is preferred to avoid pulmonary hypertension and irreversible pulmonary vascular disease. Serious pulmonary vascular disease has been observed in patients with AOPA as early as the third month of life (24). Several mechanisms have been postulated for the development of pulmonary vascular disease: high pulmonary blood flow, circulating vasoconstrictor substances, neurogenic crossover from the unprotected lung, and left ventricular failure. In our series, the patient presenting AOLPA from the descending aorta necessitated catheterisation control due to presence of respiratory symptoms. Moderate restenosis across the anastomotic site was associated with ipsilateral lung hypoperfusion at ^{99m}Tc pulmonary scintigraphy. The pulmonary biopsy (at reoperation) demonstrated a Heath-Edwards grade-II pulmonary vascular disease of the respective lung.

The origin site of the AOPA is different. Some authors (15) advocate the existence of two forms: a proximal one - the AOPA originates from the ascending aorta close to the valvar plane, and a

distal one – the AOPA arises via a patent ductus arteriosus (1). The third form of presentation is the origin site close to the innominate artery, or in extreme cases the AORPA may originate from the innominate artery itself (25). In 8 cases in this series, the AOPA originated from the respective proximal posterolateral part of the ascending aorta. Usually, the origin is from the respective posterolateral aspect of the ascending aorta (4). Nevertheless, a more antero-lateral origin of the AORPA from the ascending aorta was detected in one patient, while an anomalous origin of the AORPA from the contro postero-lateral wall of the ascending aorta was diagnosed in another case. In another patient, the AOLPA originated from the descending aorta. It is postulated that the ductus arteriosus and the AOLPA develop from the left sixth aortic arch (23). In our case we did not find the ductus arteriosus remnant indicating a complete absence of the left sixth aortic arch in this patient.

Different surgical techniques have been employed. The direct anastomosis of the AOPA to the MPA is most frequently used in the previously described series (4, 11, 14-15). End-to-end anastomosis with a synthetic graft (3, 15), interposition of a homograft patch (3-4, 15), aortic flap (9, 12), or interposition of autologous pericardial patch, for increasing the AOPA length, have been successfully employed in specific cases, when direct implantation of the AOPA was not possible. In our experience, we employed four techniques, depending on the anatomic characteristics of the malformation. In our hands, the direct implantation was associated with more anastomotic suture tension predisposing to higher risk of dehiscence. Therefore we preferred this approach, which could easily be performed without extracorporeal circulation, when the

distance between the AOPA and the MPA was minimal.

Mortality in our series was 30%. We believe that a learning curve can explain the improvement in clinical outcome, however the presence of associated heart defects and preoperative heart failure should be considered as predictors for poor postoperative results.

Occurrence of stenosis at the anastomotic site has been a major problem in some reported series remaining the main late complication (4). The gradient across the anastomotic site accurately demonstrates the stenosis. The residual gradient seems to be higher and the respective lung less perfused when repair was accomplished according to the direct reimplantation technique or synthetic graft interposition. These findings, demonstrate that direct reimplantation and synthetic graft interposition fail to gradually accommodate larger volumes with patient's somatic growth, due to growth failure of the anastomotic site. The other techniques, (consisting in interposition of autologous tissue for increasing the length and coaptation between the AOPA and the MPA) offer better results in term of late restenosis.

Conclusion

We may conclude that the AOPA from the aorta is a rare but important entity, necessitating a scrupulous preoperative and intraoperative evaluation. The techniques employing autologous tissues for enlarging and lengthening the AOPA seem to be associated with better results in terms of postoperative restenosis. Patients presenting this anomaly may undergo surgical correction with acceptable outcome.

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Figure nr.1. Preoperative angiography: A. (patient 2) The anomalous left pulmonary artery originating from the descending aorta. B. (patient 5) The anomalous right pulmonary artery originating from the contralateral wall of the ascending aorta.

LEGEND: AOLPA-Anomalous Origin Left Pulmonary Artery; AO-Aorta; TH-AO – Thoracic Aorta; AORPA-Anomalous origin Right Pulmonary Artery.

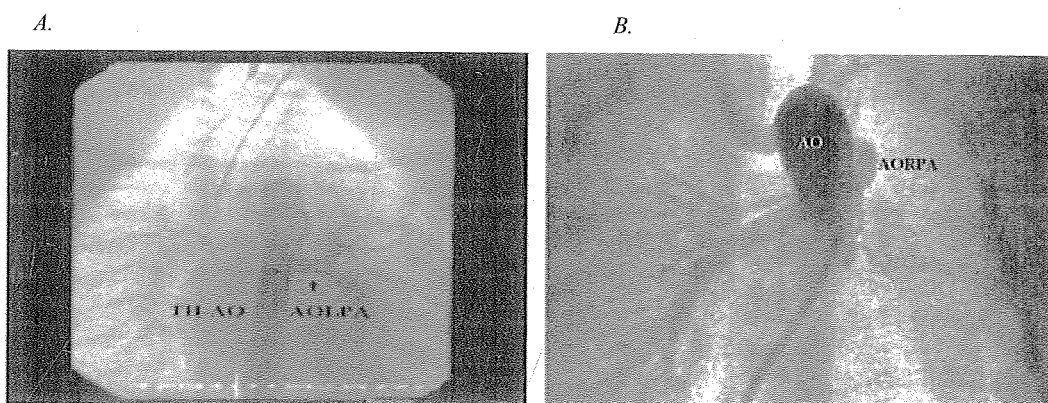


Figure nr. 2. A. Anomalous origin of the left pulmonary artery from the ascending aorta in patient 6. B. Echocardiographic view demonstrating the anomalous origin of the left pulmonary artery in patient 3.

LEGEND: AOLPA-Anomalous Origin Left Pulmonary Artery; AO-Aorta; TH-AO – Thoracic Aorta

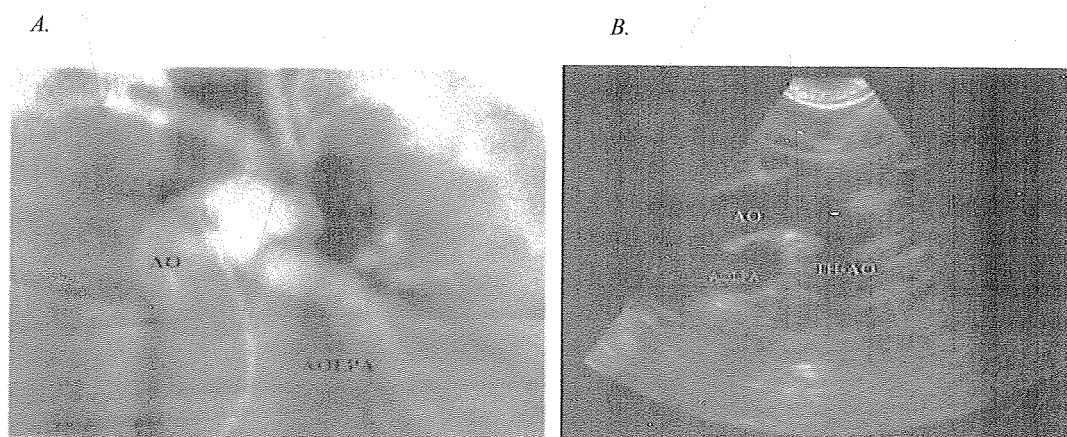
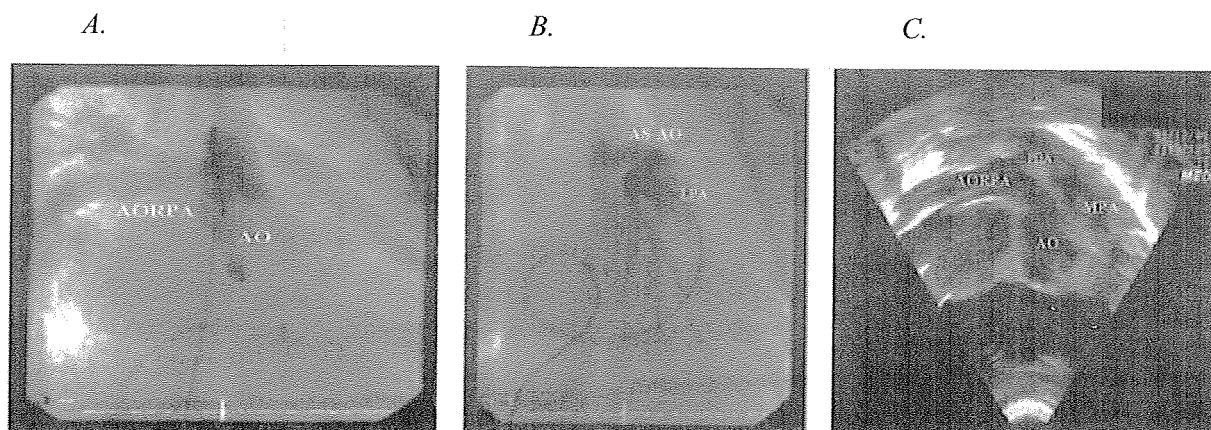
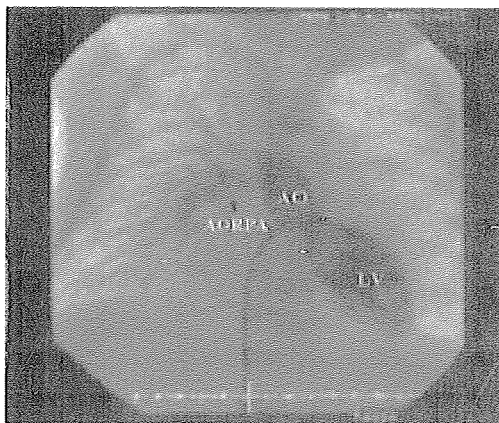


Figure nr. 3. Preoperative examinations' data in patient 7 - A. Anomalous origin of the right pulmonary artery from the ascending aorta. B. The origin of the left pulmonary artery from the main pulmonary artery trunk and the visualization of the ascending aorta through the patent ductus arteriosus. C. Echocardiographic examination showing the anomalous origin of the right pulmonary and the presence of ductus arteriosus; and in patient 8 - D. Anomalous origin of the right pulmonary artery from the right lateral aspect of the ascending aorta. E. The origin of the left pulmonary artery from the main pulmonary artery trunk. LEGEND: AOLPA-Anomalous Origin Left Pulmonary Artery; LPA-Left Pulmonary Artery; RPA-Right Pulmonary Artery; MPA- Main Pulmonary Artery; AO-Aorta; AS-AO – Ascending Aorta; TH-AO – Thoracic Aorta; AORPA-Anomalous origin Right Pulmonary Artery; PDA-Patent Ductus Arteriosus; LV-Left Ventricle; RV-Right Ventricle



D.



E.

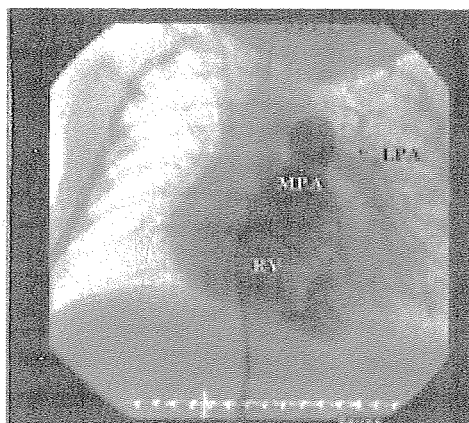


Figure nr. 4. SINGLE AORTIC FLAP TECHNIQUE. The reimplantation of the right pulmonary artery to the main pulmonary trunk by using an aortic ring

LEGEND: LPA-Left Pulmonary Artery; MPA-Main Pulmonary Artery; AO-Aorta, RPA-Right Pulmonary Artery; AV-Aortic Valve; PV- Pulmonary Valve

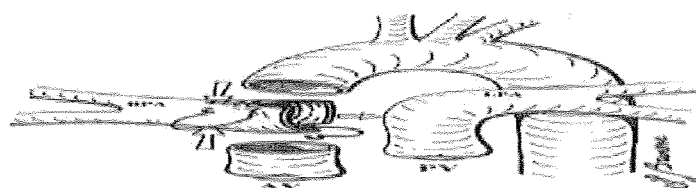
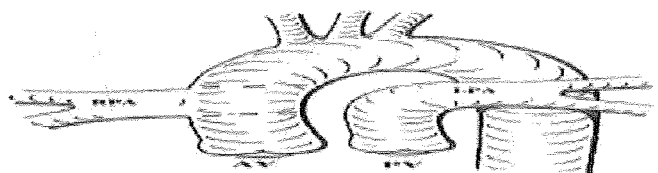
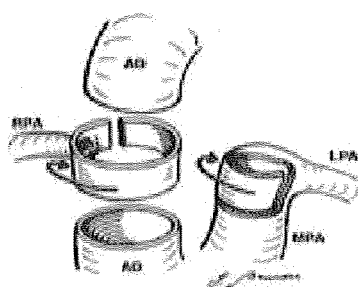


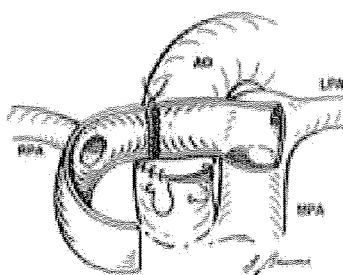
Figure nr. 5. DOUBLE FLAP TECHNIQUE. A) Aortic and pulmonary flap preparation. B) Anterior positioning of the aortic and pulmonary flaps and posterior anastomosis. C) Newly created communication between anomalous right pulmonary artery and main pulmonary artery.

LEGEND: LPA-Left Pulmonary Artery; MPA-Main Pulmonary Artery; AO-Aorta, RPA-Right Pulmonary Artery

A.



B.



C.

