

## Clinical outcomes of primary arterial switch operation in treatment of Taussig-Bing anomaly

*Taussig-Bing anomalisinin tedavisinde primer arteriyel switch ameliyatının klinik sonuçları*

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### ABSTRACT

**Background:** This study aims to present the outcomes of primary single-stage arterial switch and ventricular septal defect closure operation in patients with Taussig-Bing anomaly.

**Methods:** Between November 2004 and November 2015, a total of 41 patients (33 males, 8 females; median age 38 days; range 4 to 1,239 days) who underwent primary arterial switch procedure with the diagnosis of Taussig-Bing anomaly were retrospectively analyzed using the hospital database. Demographic data, echocardiograms, cardiac catheterization reports and operative notes were reviewed.

**Results:** In-hospital mortality occurred in six patients (14.3%). Risk factors associated with in-hospital mortality were cardiopulmonary bypass time ( $p=0.047$ ; Exp (B)=1.0222) and aortic arch obstruction ( $p=0.023$ ; RR=NA). The mean follow-up was 3.5 (range 0.1-10.9) years. A total of 27 (77%) of 35 survivors were followed. Late mortality was seen in three patients (11.1%). Freedom from reoperation was 96% at one year and 84% at five and 10 years. The only factor associated with late mortality was the coronary pattern 1LAD-2R, Cx ( $p=0.025$ ; RR=16; %95 CI= 2-128).

**Conclusion:** Primary arterial switch operation is safe and effective in the repair of Taussig-Bing anomaly. Despite an increased risk in patients with aortic arch obstruction, single-stage approach seems to be preferable.

**Keywords:** Arterial switch operation; congenital heart disease; double-outlet right ventricle; Taussig-Bing anomaly; transposition of great vessels.

### ÖZ

**Amaç:** Bu çalışmada Taussig-Bing anomalili hastalarda primer tek aşamalı arteriyel switch ve ventriküler septal defekt kapatılması ameliyatının sonuçları sunuldu.

**Çalışma planı:** Hastane veri tabanı kullanılarak Kasım 2004 - Kasım 2015 tarihleri arasında Taussig-Bing anomalisi tanısı ile primer arteriyel switch ameliyatı yapılan toplam 41 hasta (33 erkek, 8 kız; medyan yaş 38 gün; dağılım 4-1.239 gün) retrospektif olarak incelendi. Demografik veriler, ekokardiyografi ve kardiyak kateterizasyon raporları ve ameliyat notları incelendi.

**Bulgular:** Altı hastada (%14.3) hastane mortalitesi izlendi. Hastane mortalitesi ile ilişkili risk faktörleri kardiyopulmoner baypas süresi ( $p=0.047$ ; Exp (B)=1.0222) ve arkus aort obstrüksiyonu ( $p=0.023$ ; RR=NA) idi. Ortalama takip süresi 3.5 (dağılım 0.1-10.9) yıl idi. Sağkalan 35 hastanın toplam 27'si (%77) takip edildi. Üç hastada (%11.1) geç mortalite izlendi. Yeniden ameliyat olmama oranı birinci yılda %96 ve beş ve 10. yıllarda %84 idi. Geç mortalite ile ilişkili tek faktör 1LAD-2R, Cx koroner arter paterni idi ( $p=0.025$ ; RR=16; %95 CI= 2-128).

**Sonuç:** Taussig-Bing anomalisinin tedavisinde primer arteriyel switch ameliyatı güvenli ve etkilidir. Arkus aort obstrüksiyonu olan hastalarda risk artışına karşın, tek aşamalı yaklaşım tercih edilebilir görünmektedir.

**Anahtar sözcükler:** Arteriyel switch ameliyatı; doğumsal kalp hastalığı; çift çıkımlı sağ ventrikül; Taussig-Bing anomalisi; büyük arterlerin transpozisyonu.



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Taussig-Bing anomaly was first described by Taussig and Bing in 1949.<sup>[1]</sup> Although various alternative repair strategies have been developed through the years,<sup>[2-5]</sup> the closure of the ventricular septal defects (VSDs) tunneling the left ventricular outflow tract to the pulmonary valve followed by arterial switch operation was accepted as the method of choice, particularly in the past decade.<sup>[6-10]</sup> One of the concerns regarding the management has been the high coincidence of Taussig-Bing anomaly with aortic arch obstruction which was spotted early in the surgical experience.<sup>[11]</sup> The recent literature has addressed into the debate on whether the arch obstruction can be managed on the same stage as the Taussig-Bing repair or whether a staged approach is more suitable in selected cases.<sup>[12-14]</sup> Of note, favorable results with a single-stage approach have been described in the recent cohorts.<sup>[8,9]</sup> However, with the focus shifting to the follow-up of patients undergoing single-stage primary arterial switch operation and VSD closure with or without arch reconstruction, late mortality and re-interventions have become other subjects of concern. Despite the increasing number of reports about the early primary repair of Taussig-Bing anomaly, controversial results and risk factors for re-intervention and late mortality calls for additional data from large cohorts.

In the present study, we aimed to define basic parameters such as mortality, early, and late complications, re-intervention rates and to analyze risk factors for mortality, complications, and re-intervention on a cohort of Taussig-Bing anomaly patients treated with primary single-stage arterial switch operation.

## PATIENTS AND METHODS

Using the hospital database, a total of 41 patients (33 males, 8 females; median age 38 days; range 4 to 1,239 days) who underwent primary arterial switch procedure with the diagnosis of Taussig-Bing anomaly between November 2004 and November 2015 were retrospectively analyzed. The diagnosis of Taussig-Bing anomaly was based on the double-outlet right ventricle (DORV) and subpulmonary VSD according to a recent and common definition of the disease.<sup>[9]</sup> Preoperative echocardiograms and operative notes were reviewed to confirm the diagnosis. The patient charts and computer database were retrospectively reviewed. The patients with missing follow-up data within the past six months were contacted by phone interviews. In-hospital mortality was defined as death before discharge.

The study protocol was approved by the Ethics Committee of Dr. Siyami Ersek Thoracic and

Cardiovascular Surgery Training and Research Hospital. The study was conducted in accordance with the principles of the Declaration of Helsinki.

## Statistical analysis

Statistical analysis was performed using the IBM-SPSS Statistics version 21.0 software (IBM Corp., Armonk, NY, USA). Categorical variables were expressed in frequency (percentage), and continuous variables were expressed in median (min-max) for skewed data or mean  $\pm$  standard deviation (SD) for values with normal distribution. Survival and freedom from reintervention were estimated using the Kaplan-Meier method. The association between the categorical variables and mortality or reintervention were analyzed using the chi-square or Fisher's exact tests based on the number of patients. For continuous variables, the Mann-Whitney U test was used. Relative risk (RR) and 95% confidence interval (CI) were calculated for the associations between categorical variables. A *p* value of  $<0.05$  was considered statistically significant.

## RESULTS

During the study period, a total of 42 patients were operated with the diagnosis of Taussig-Bing anomaly. However, one patient who underwent the Senning procedure and ventricular septal defect closure was excluded from the analysis. The remaining patients who underwent primary arterial switch procedure were included in the study.

The median bodyweight at the time of operation was 3.7 (range 2.5 to 8.5) kg. None of the patients received palliative intervention or surgery before the operation or had a preoperative infection. Three patients had apical VSDs in addition to subpulmonary VSDs of the Taussig-Bing anomaly. Aortic arch obstruction developed in 21 patients (51%): 18 patients had a hypoplastic aortic arch, two patients had the coarctation of aorta, and one patient had an interrupted aortic arch. Table 1 shows preoperative anatomic characteristics of the patients.

## Surgical procedure

The operations were performed or supervised by four different surgeons, while the main approach remained homogenous. Operative details are demonstrated in Table 2. Arterial switch operation was performed under moderately hypothermic cardiopulmonary bypass. Ventricular septal defects were approached mainly through the neo-aortic valve; however, additional approaches were applied as dictated by the malformation. The VSD closure was performed using the continuous technique. The trap-door technique

**Table 1. Anatomic characteristics (n=41)**

Characteristic	n	%
<b>Aortopulmonary artery relationship</b>		
Anteroposterior	33	81
Side-by-side	8	19
<b>Coronary configuration</b>		
1LAD, Cx-2R	28	68
2R, LAD, Cx	6	15
1LAD-2R, Cx	4	10
1LAD, R-2Cx	2	5
1R-2R, LAD, Cx	1	2
<b>Intramural course of left coronary artery</b>		
Aortic arch obstruction	4	10
None	20	49
Hypoplastic aortic arch	18	44
Intrerrupted aortic arch	1	2
Coarctation of aorta	2	5
<b>Other cardiac malformations</b>		
Patent foramen ovale	15	37
Atrial septal defect	9	22
Apical ventricular septal defect	2	5
Branch pulmonary artery stenosis	2	5
Right ventricular outflow tract stenosis	2	5
Mitral straddling	1	2

LAD: Left anterior descending coronary artery; Cx: Circumflex coronary artery; R: Right coronary artery.

was routinely employed for coronary button transfer. Coronary distribution is demonstrated in Table 1. In side-by-side position, five patients (63%) and in

anteroposterior configuration 23 patients (70%) had 1LAD, Cx-2R configuration of coronary arteries. In three patients with 2R, LAD, and Cx coronary artery pattern (Table 1), coronary reallocation technique was used.<sup>[15]</sup> Decision of whether to perform LeCompte maneuver was dictated by the anatomy; however, mostly it was not performed in patients with side-by-side relationship of aorta and pulmonary artery. Pulmonary artery reconstruction was augmented using the autogenous pericardium.

Additional aortic arch pathologies were treated simultaneously during the procedure. In two patients with isolated aortic coarctation, an extended end-to-end anastomosis was performed. The patient with aortic interruption was repaired with partial anastomosis of the distal arch and descending aorta, and the anastomosis was partially augmented with patchplasty. In the remaining patients with arch hypoplasia, the technique used was anatomy-dependent; however, it was mostly either some form of extended end-to-end anastomosis, patchplasty or a combination of these techniques based on the surgeon's preference (Table 2). An end-to-end anastomosis was performed between descending aorta and distal aortic arch. In patients repaired with patchplasty, the descending aorta was transected and the ductal tissue was entirely resected. In case that discrete coarctation accompanied the arch hypoplasia, the segment was removed and partial anastomosis between the distal arch and descending

**Table 2 Operative data (n=41)**

Characteristic	n	%	Mean±SD
<b>Operative times</b>			
Bypass time			213±59
Cross-clamp time			162±39
Antegrade cerebral perfusion	16	39	30±10
Total circulatory arrest	6	15	26±13
<b>Ventricular septal defect closed through</b>			
Right atriotomy	19	46	
Right ventriculotomy	2	5	
Neo-aorta	15	37	
Combined	5	12	
<b>Ventricular septal defect patch</b>			
Pericardium	23	56	
Polytetrafluoroethylene	17	41	
Dacron	1	2	
LeCompte maneuver	34	81	
Neopulmonary patch fresh pericardium	7	17	
<b>Arch repair procedure</b>			
Patchplasty	13	31	
Extended end-to-end anastomosis	8	20	

SD: Standard deviation.

aorta was performed, and the proximal aortic arch was augmented using the autologous pericardium. A gluteraldehyde-treated autologous pericardial patch was used as the patch material.

### Mortality

In-hospital mortality occurred in six patients (14.3%). Four patients were unable to be weaned off cardiopulmonary bypass, and two patients died in the early postoperative period; one due to unexplained cardiac arrest on postoperative Day 4 and the other due to sepsis on postoperative Day 18. All non-survivors had additional aortic arch obstruction addressed on the same procedure. Of four patients who were unable to be weaned off cardiopulmonary bypass, one had additional apical and midmuscular VSD which was unable to be closed during the repair, which was thought to be a potential reason for low cardiac output state postoperatively. In the other three patients, one had 2R, LAD, Cx pattern and the other three had 1LAD, Cx-2R coronary artery pattern. In these patients, there were signs of coronary malperfusion on direct observation. In each of these patients, a second period of cardioplegic arrest was instituted and coronary osteal repositioning was performed. In one of the patients, additionally LeCompte was taken down. The risk factors associated with in-hospital mortality were cardiopulmonary bypass time ( $p=0.047$ ; Exp (B)=1.0222) and aortic arch obstruction ( $p=0.023$ ; RR=NA).

### Early postoperative period

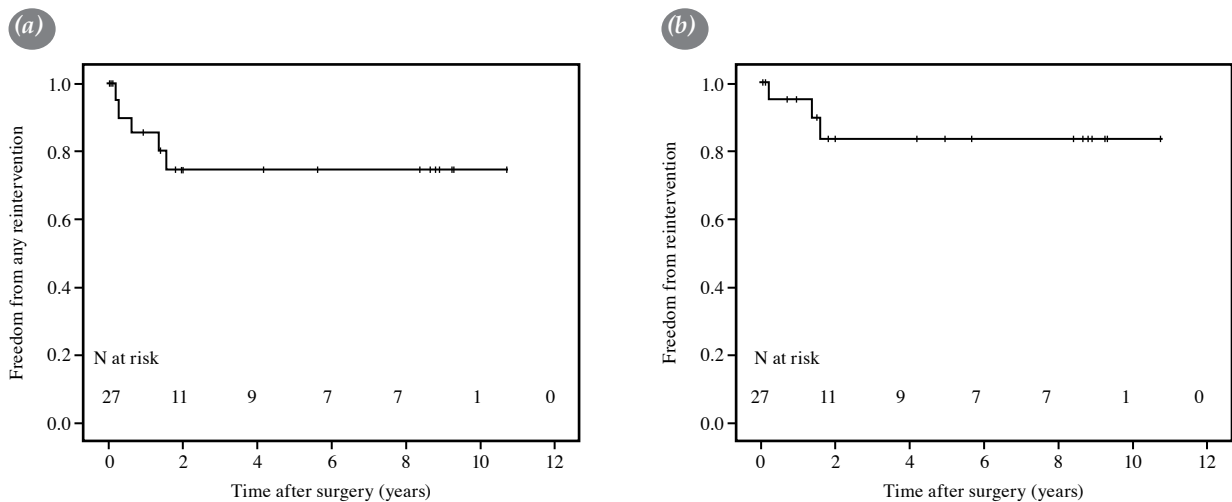
The patients were ventilated for a median duration of four (range 1 to 55) days with a median length of stay in the intensive care unit (ICU) of nine

(range 2 to 100) days. Median postoperative hospital stay of the hospital survivors was 15 (range 7 to 109) days. Of 37 intraoperative survivors, 13 patients (35%) had delayed sternal closure.

### Follow-up

Of 35 survivors, 27 were followed with a mean follow-up of 3.5 (range 0.1-10.9) years. Late mortality was seen in three patients (11.1%). Two patients died two years postoperatively at home due to unknown reasons. Another patient died nine years postoperatively due to ventricular fibrillation.

Of 27 survivors, five patients (18.5%) required surgical or transcatheter reintervention. These included coarctation balloon angioplasty and pulmonary artery balloon angioplasty in two patients (7.4%), right ventricular outflow tract reconstruction in two patients (7.4%), and aortic repair for regurgitation in one patient (3.7%). Of two patients with right ventricular outflow tract reoperations, none of them had preoperative subaortic stenosis. One of these reoperations was for stenosis on valvular level which required transannular patch augmentation. In the other patient, supravalvular pulmonary stenosis at the anastomotic level was repaired with patch augmentation. The patients who underwent pulmonary balloon angioplasty also had stenosis at the supra-annular level. Neither of these patients had preoperative subaortic stenosis. Reintervention was required at a median duration of 0.6 (range 0.2 to 1.55) years postoperatively. Freedom from catheter or surgical intervention was 87% at one year and 76% at five and 10 years. Freedom from reoperations was 96% at one year and 84% at five and 10 years (Figure 1).



**Figure 1.** (a) Freedom from any catheter or surgical reintervention over time in years. (b) Freedom from surgical reintervention over time in years.

The only factor associated with late mortality was the coronary pattern 1LAD-2R, Cx ( $p=0.025$ ; RR=16; %95 CI= 2-128). None of the factors reached the level of significance in predicting reintervention. In the most recent follow-up echocardiogram, one patient had right ventricular dysfunction (2.8%), two patients had left ventricular dysfunction (5.6%), nine patients had grade two or three aortic insufficiency (25%), five patients had mild aortic stenosis (13.9%), three patients (8.3%) had neo-aortic root dilatation, six patients had mild-to-moderate left ventricular outflow tract obstruction (16.7%), 12 patients had mild pulmonary stenosis (33.3%), and four patients had aortic coarctation (11.1%).

## DISCUSSION

To the best of our knowledge, this is one of the largest cohorts of single-stage primary arterial switch operation for Taussig-Bing anomaly in a relatively short time period. Our results have the variability of being the product of four different surgeons and using modifications in surgical technique regarding the patch material, approach for closure of VSDs, and arch repair technique. However, these modifications were unable to be demonstrated as the risk factors for early or late mortality and morbidity.

Primary arterial switch operation, as opposed to primary palliation, has been established particularly in the past decade as the method of choice in the treatment of Taussig-Bing anomaly.<sup>16,71</sup> Primary repair has the advantage of avoiding multiple surgeries and also, with primary palliation, patients are exposed to a longer period of cyanosis and have the risk of developing heart failure, pulmonary vascular disease, and aortic insufficiency.<sup>181</sup> In cases with complex anatomy, prematurity, and preoperative complications Schwarz et al.<sup>171</sup> proposed that a two-stage approach can still be preferable. However, there is still a limited of data about the time of surgery and patients requiring primary palliation.

Some types of aortic arch obstruction accompany the Taussig-Bing anomaly with a rate of up to 66%.<sup>171</sup> Recent series equivocally advocate the repair of the arch pathology on the same stage during the arterial switch operation.<sup>16,7,101</sup> The technique of aortic arch repair vary among different clinics and include end-to-side repair,<sup>171</sup> integrating neo-aortic stump into the arch anastomosis or aortic relocation,<sup>1101</sup> and routine patch augmentation.<sup>161</sup> Our cohort is heterogeneous regarding the technique of the arch repair; mainly extended end-to-end anastomosis and patch augmentation were the techniques applied based on the surgeons' preference.

Our recoarctation rate was 7.4% and any correlation with the technique used was unable to be demonstrated. Further large-scale, randomized studies are required to obtain long-term effects of the arch repair methods used.

In addition, subaortic right ventricular tract obstruction was reported in rates up to 75% in patients with Taussig-Bing anomaly,<sup>171</sup> although others reported lower rates around 15%.<sup>16,81</sup> In our study, the rate of preoperative subaortic stenosis was coincidentally lower (5%). Therefore, none of the patients required right ventricular outflow tract reconstruction at the initial repair.

Similar to our study, early mortality was reported in the recent studies as 2.2 to 20%.<sup>17,8,10,12,14,16,171</sup> Staged repair and higher weight, and coronary abnormalities were reported to be main risk factors for mortality.<sup>113,171</sup> Soszyn et al.<sup>181</sup> attributed their decreased mortality to complete resection of sub-neo-aortic obstruction and overall improvement of perioperative care. Although our cohort showed an increased risk in patients with an arch pathology, in the recent cohorts, it has been shown that additional arch pathology can be addressed without an increased risk of mortality or reoperations.<sup>16,91</sup> Therefore, single-stage approach still seems to be preferable in patients with accompanying arch obstruction.

Furthermore, with an increased number of patients during follow-up after arterial switch operation for Taussig-Bing anomaly, late mortality has become a concern. Despite a cohort with no late mortality,<sup>181</sup> others have reported late mortality mostly with an unexplained etiology.<sup>16,7,91</sup> In our patient cohort, sudden nature of the late mortality primarily indicates arrhythmia, and an underlying coronary problem cannot be ruled out. One of the largest series about arterial switch operation for the transposition of great arteries which includes angiographic studies in the perioperative period and follow-up suggested that 89% of postoperative coronary events were observed within the first three months.<sup>1181</sup> It can be proposed that performing coronary angiograms in selected patients before discharge can improve late results. Vergnat et al.<sup>161</sup> also suggested routine angiography postoperatively for Yacoub type C coronary pattern and all patients with positive electrocardiography findings. On the contrary, in the present study, Yacoub type B or, in Leiden terminology, 1LAD-2R, Cx coronary pattern was associated with late mortality. This is contradictory to Vergnat et al.'s<sup>161</sup> study, since they did not find such an association, although 10% of their population consisted of patients with type B (1LAD-2R, Cx) coronary

morphology. The variability in results among different clinics can be the effect of even minute differences in the surgical technique applied. However, it seems that with increasing data, at least some of the late mortality can be linked to abnormal coronary patterns. This, in turn, raises the question whether these patients would require additional diagnostic measures during follow-up. Currently, the use of positron emission tomography and myocardial perfusion scans have been investigated in the follow-up of arterial switch patients;<sup>[19,20]</sup> however, guidelines for active surveillance of these patients have not been developed, yet.

In addition, it has been reported that 20 to 40% of patients require some types of reintervention.<sup>[7-9,13,17]</sup> Reintervention was observed mostly within the first one or two years, as in this cohort.<sup>[9,12,21]</sup> Right ventricular outflow tract obstruction was observed up to 9 to 10 years after arterial switch operation for transposition of the great arteries.<sup>[22,23]</sup> The number of patients under follow-up up to 9 to 10 years after arterial switch operation for Taussig-Bing anomaly is limited both in this cohort and in the literature; therefore, whether a higher number of reintervention is to be expected in the years to come still remains unknown. Consistent with our results, right ventricular outflow tract obstruction has been the most common reason for reintervention.<sup>[9,12]</sup> Although LeCompte maneuver, inadequate mobilization of pulmonary arteries, and mismatch in size of great arteries have been accused previously,<sup>[12]</sup> we were unable to find any risk factors in this cohort.

On the other hand, this study has the limitations of a retrospective design. Data were limited with follow-up available to only 77% of hospital survivors for a relatively short follow-up period. Additionally, some of the important questions in Taussig-Bing anomaly repair including risk factors associated with late mortality are still difficult to answer with the limited population of this study.

In conclusion, primary arterial switch operation is safe and effective in the repair of Taussig-Bing anomaly. Despite an increased risk in patients with aortic arch obstruction, single-stage approach still seems to be preferable. Once the basic principles of the operation are met regarding coronary transfer, ventricular septal defect closure, and arch repair, specific modifications in surgical technique do not seem to have any functional significance. However, the effect of coronary pattern on late mortality should be further investigated, as this can improve the follow-up routines to detect the probable causes of unexpected death events.

#### Declaration of conflicting interests

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