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## INTRODUCTION

The more common occurring type of pulmonary venous return anomaly is a partial anomalous connection of the right pulmonary veins (PARPVC) to the superior vena cava (SVC). It is encountered in approximately 10% of patients with atrial septal defects (ASDs). In most cases, interatrial communications present as sinus venosus defects (1). The typical surgical method for repairing this anomaly is forming a baffle, which redirects blood flow from the pulmonary veins (PV) ostia through the ASD to the left atrium (LA), with or without relevant enlargement of the upper cavoatrial junction. This method is

## Long-term results of two different techniques in patients with supracardiac partial anomalous pulmonary venous connection via right thoracotomy approach. A prospective study

### Abstract

**Aim:** This study compared outcomes following the double-patch and Warden procedures for correcting partial anomalous connection of the right pulmonary veins to the superior vena cava via right thoracotomy approach.

**Material and Methods:** Hundred patients, aged <18 years old, and with partial anomalous connection of the right pulmonary veins to the superior vena cava were randomly selected into double-patch method (n=0) and Warden procedure (n=50) groups. The median follow-up was 31.5 (range, 10-45) months. The primary endpoint was sinus node dysfunction at the mid-term follow-up period.

**Results:** No early or late mortality occurred. In the early postoperative period, sinus node dysfunction was observed in 32.4% and 7% of cases after double-patch technique and the Warden procedure, respectively (risk ratio, 6.15; 92% confidence interval, 1.45-26.35; p=0.02). At follow-up, sinus node dysfunction remained in two (5%) patients after double-patch correction. All patients had normal sinus rhythm after the Warden procedure. No early or late pacemaker implantation occurred in either group. No patients had significant pulmonary veins or superior vena cava stenosis.

**Conclusion:** The double-patch technique and Warden procedure both showed best mid-term consequences without mortality. The Warden procedure was linked with less sinus node dysfunction in the early postoperative period than the double-patch technique. There was no significant various differences in sinus node dysfunction at the mid-term follow-up.

**Keywords:** Anomalous pulmonary venous connection, Right thoracotomy approach, Warden procedure, Double-patch method, Sinus node dysfunction, Pulmonary vein or superior vena cava obstruction

said to be associated with rhythm disturbances (accounting for up to 70% of sinus node dysfunction (SND) early after surgery and 25.2% of those at mid-term follow-up), including SVC and PV stenosis in the early and late postoperative periods (2). Another surgical technique, which was popularized by Warden et al., involves baffling the SVC ostium to the LA through the ASD, with dividing and reimplanting the cranial SVC stump into the right atrial appendage (RAA) (Warden procedure, cavoatrial anastomosis) (3). From this point of view, this procedure can prevent sinus node dysfunction, reduce arrhythmogenic effects, is associated with SND absence at follow-up, and can reduce the risk of SVC and PV stenosis (4). This study compared outcomes

of correcting PARPVC to the SVC with the double-patch (DP) method and Warden procedure (WP).

## MATERIAL AND METHODS

Between October 2018 and March 2021, 100 patients with PARPVC to the SVC were enrolled in this prospective. The study complied with the Declaration of Helsinki, was approved by the local ethics committee, and was conducted according to the protocol and using minimal invasive surgical techniques. Written informed consent was signed by all patients (their parents) before participation. Surgery was indicated for symptomatic and asymptomatic patients in accordance with current recommendations: Qp:Qs ratio >1.5, right ventricle dilatation, and increased pulmonary artery pressure.

Enrolled patients were randomly assigned into two groups using block randomization and a computer-based random numbers table. Surgical repair was performed using the DP technique in the first group, and WP was used in the second group. The groups were comparable with respect to baseline characteristics (Table 1).

The primary endpoint was sinus node dysfunction at the mid-term follow-up period. Secondary endpoints were: superior vena cava stenosis, right pulmonary vein stenosis, duration of cardiopulmonary bypass, aortic cross-clamping, and induced ventricular fibrillation.

**Table 1. Patients' baseline characteristics**

	Double patch method (n=50)	Warden procedure (n=50)
Female, n (%)	30 (60%)	25 (45%)
Age, months	45 (35-88)	55 (42-95)
Weight, kg	17.8 (14.0-21.5)	22.1 (20.5-31.5)
Qp/Q s	2.1(1.8-2.3)	1.9(1.5-2.7)
Pulmonary artery pressure	40(35.5-42)	40(31.5-48)

### Surgical Technique

For the surgical approach, right minimal invasive thoracotomy approach, used in all patients from both groups. The choice of approach was based on the surgeon's preference.

After the surgical approach was accomplished, aorto - bicaval cannulation was performed (with high SVC cannulation). Normothermic bypass was initiated. Induced ventricular fibrillation was used during repair through right mid-axillary thoracotomy.

**Double-Patch Method** - For this type of repair, a right atriotomy incision was continued through the cavoatrial junction (on its lateral surface) to the SVC. A diepoxy treated bovine pericardial patch was used for PV baffling and cavoatrial junction plasty, which was sutured with 5/0-6/0 polypropylene

### Warden Procedure

The SVC ostium was redirected to the LA using a diepoxy- treated bovine pericardial patch with emphasis on patch fixation on its upper margin (non-transmural suturing was used in this area). After cardiac rhythm restoration, the SVC was transected above the upper PV ostium in an oblique fashion, and the caudal stump was oversewn with running suture. For cavoatrial anastomosis, 5/0-6/0 polydioxanone sutures were used.

### Statistical Method

Sample size calculation was conducted with the G\*Power 3.1 application (<http://gpower.hhu.de>) and was based on an 18.1% expected difference (5). Categorical variables were presented as numbers (%). Continuous variables were presented as median (Q1,Q3) unless otherwise specified. The Mann-Whitney U test and Fisher's exact test were used for inter-group comparisons. For comparison of one quantitative characteristic among three or more independent groups, the Kruskal-Wallis test was used. In case of statistically significant between – group differences, pairwise comparisons using the Mann – Whitney U test with Bonferroni's correction was utilised (6).

## RESULTS

The median cardiopulmonary bypass (CPB) and ventricular fibrillation (VF) time were significantly lower in the WP group than in the DP group (p=0.005 and p<0.0003, respectively).

Sinus venosus defect was present in the majority of patients (65% in the DP and 72.5% in the WP group). The interatrial septum was intact in two patients from the WP group. Atrial septal defect enlargement was performed in more than half of patients in both groups.

Kruskal-Wallis variance analysis showed statistically significantly different CPB and VF duration between sub- groups, divided by approach (p<0.002).

No mortality occurred in the early postoperative period. The median intensive care unit stay was 1 day in both groups. The median mechanical ventilation time in the DP and WP groups was 8 (3,9) hours and 4 (2,5) hours, respectively (p=0.12). Sinus node dysfunction was observed in 32.5% of cases after DP correction and in 7% after WP (risk ratio, 6.50; 90% confidence interval, 1.32-25.27; p=0.02). Temporary atrial pacing was required more often in the DP group (22.5% in the DP and 3.5% in the WP group (p=0.03)).

There was no PV or SVC stenosis in the DP group (p=0.25). Turbulent blood flow in the SVC was observed in two patients after Warden procedure with peak gradients

of 9 mmHg and 11mmHg, respectively. However, no superior vena cava syndrome (SVCS) was observed in these patients. The surgical approach used during the WP did not affect SVC stenosis development (p=0.15).

At discharge, SND signs according to 24-hour Holter ECG

were revealed in 8 (15%) patients from the DP and one (3.5%) patient from the WP group, but this difference did not reach statistical significance ( $p=0.45$ ). Rhythm disturbances were not clinically relevant. No patients underwent permanent pacemaker implantation.

All patients were discharged in good clinical condition. The median hospital stay was 12 (8,15) and 12 (7,18) days in the DP and WP groups, respectively ( $p=0.91$ ), and its duration was not associated with surgical approach ( $p=0.42$ ).

## DISCUSSION

This study was conducted in a prospective, randomized manner. It assessed early and mid-term outcomes of the Warden procedure compared to those following surgery with the DP technique. It is believed that this is the first study with a prospective design dedicated to comparing outcomes after different procedures for repair of PARPVC to the SVC (7). The most significant complications after repair of PARPVC to the SVC are sinus node dysfunction, and systemic and pulmonary venous obstruction. Any surgical technique could pose a risk for these events, which are caused by the specific anatomy in the region of this anomaly (8).

Trauma to the sinus node or its blood supply elements can cause serious rhythm disturbances, which can require permanent pacemaker implantation. Up to 6% of patients with partial anomalous pulmonary venous connection have been reported to undergo pacemaker implantation at late follow-up (125 T 99 months) (9). Hypothetically, procedures, excluding cavoatrial incision, such as Warden procedure or transcaval technique could minimise the risk of arrhythmias, which has been supported by some retrospective studies (10). In recent studies dedicated to the Warden procedure, the occurrence of rhythm disturbances in the early postoperative period was 0%-6%. In cases of double-patch correction, it occurs in up to 55% of patients early after surgery, with a reduction to 30.5% at discharge (11). In the current study, SND occurred less often and no statistically significant difference was found between the DP and WP groups at discharge. Sinus node dysfunction is likely caused by peculiarities specific to how surgical techniques are performed at different centers. The risk of SVC obstruction during any type of procedure dictates the necessity of caudal SVC enlargement. The literature reports variability in the SVC stenosis rate after different types of surgery (3%-5.2%) (12). Iyer et al. compared single-patch and double-patch methods and revealed that the single-patch technique carried a higher risk of SVC stenosis. The results for the Warden procedure and its modifications have demonstrated a low rate of SVC obstruction, despite being a more complicated technique, which, in some cases, has necessitated enlargement of the anastomosis with a patch (13). All authors emphasize the importance of wide cavoatrial anastomosis and careful resection of all intra-atrial trabeculations, which can cause future stenosis. Two patients in the current study had mild SVC stenosis (WP group), and there were no clinical manifestations of SVCS.

Pulmonary venous obstruction is relatively uncommon after PARPVC to SVC correction. Patients develop PV obstruction after the Warden procedure in 1.6% of cases and after the DP technique in 0.5% of cases, which can require re-operation (14). Among the current study participants, no significant SVC or PV stenosis was observed at follow-up, but further evaluation is necessary to draw definitive conclusions regarding these complications.

## CONCLUSION

Right mid-axillary thoracotomy surgical approach for PARPVC to the SVC correction, has previously been shown to be safe. Both the DP method and WP can be efficiently performed through RMT. Ventricular fibrillation, used instead of cardioplegia in this approach, did not show any influence on intraoperative and early postoperative courses.

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**Data Availability Statement:** The data that support the findings of this study are available from the corresponding author upon reasonable request.

**Ethics committee approval:** The study complied with the Declaration of Helsinki, was approved by the Scientific Surgery Center named after M. Topchubasov and was conducted according to the protocol and using minimal invasive surgical techniques.

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