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ORIGINAL ARTICLE

The Use of Minimally Invasive Surgical Techniques in Pediatric Patients with Partial Anomalous Pulmonary Venous Return

Parsiyel Anormal Pulmoner Venöz Dönüş Anomalili Pediatrik Hastalarda Minimal İnvaziv Cerrahi Tekniklerin Kullanımı

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ABSTRACT

Aim: The use of minimally invasive procedures in low-risk congenital heart surgeries has increased recently. Compared to traditional median sternotomy, minimally invasive techniques offer better cosmetic results and provide more satisfaction to both the patient and their parents. Partial anomalous pulmonary venous return (PAPVR) can be safely repaired using these techniques.
Material and Methods: The perioperative data of right-sided PAPVR patients who were operated on with minimally invasive approaches in our clinic between March 2019 and January 2023 were reviewed retrospectively. The perioperative data of the patients' including type of surgery, cardiopulmonary bypass duration, cross-clamp duration, postoperative cardiac rhythm, total operation duration, total drainage, total intensive care unit duration and total hospital stay duration were obtained. The results were compared with the data in the current literature.
Results: During the study period, 14 patients underwent surgical repair. Five (36%) of them were female and nine (64%) were male. Patients underwent right infra-axillary vertical thoracotomy (RIAVT) and right anterolateral thoracotomy (RALT) had mean ages of 48±26.6 and 42±18.2 months, respectively. High venosum type atrial septal defect (ASD) was detected in 12 (85.7%) of the patients. The most frequently used surgical treatment was the double patch technique which was used in 12 patients (85.7%). This was followed by single patch repair in one patient (7.1 %). The Warden procedure was used in one patient (7.1%) who was operated on with the RIAVT technique. Patients' cross-clamp, cardiopulmonary bypass (CPB), and total operation durations were comparable to those reported in the medical literature.
Conclusion: RIAVT and RALT are two of the frequently used minimally invasive surgical techniques in congenital heart surgery. After gaining sufficient experience, both of these methods can be safely applied to PAPVR repair.

Keywords: Anomalous pulmonary venous return, pediatrics, minimally invasive surgical procedure, thoracotomy

ÖZ

Amaç: Son zamanlarda, düşük riskli doğumsal kalp cerrahisinde minimal invaziv prosedürlerin kullanımı artmaktadır. Geleneksel median sternotomiye kıyasla minimal invaziv teknikler daha iyi kozmetik sonuçlar sunmakta ve hem hastaya hem de ebeveynlerine daha fazla memnuniyet sağlamaktadır. Pediatrik hastalarda , parsiyel anormal pulmoner venöz dönüş anomalisi (PAPVD) , bu teknikler kullanılarak güvenle onarılabilmektedir.
Gereç ve Yöntem: Mart 2019 ile Ocak 2023 tarihleri arasında kliniğimizde minimal invaziv yaklaşımlar kullanılarak ameliyat edilen sağ taraflı PAPVD hastalarının perioperatif verileri retrospektif olarak incelendi. Hastaların perioperatif verileri çerçevesinde , uygulanan cerrahi tipleri, kardiopulmoner bypass süreleri, kross klemp süreleri, postoperatif kardiyak ritimleri , toplam operasyon süreleri, toplam drenajları, toplam yoğun bakım süreleri , toplam hastanede kalış süreleri ve mortalite verileri toplandı. Sonuçlar mevcut literatürdeki verilerle karşılaştırıldı.
Bulgular: Çalışma döneminde 14 hasta opere edildi . Bunların beşi (%36) kadın ve dokuzu (%64) erkekti. Sağ infraaksiller vertikal torakotomi(SIAVT) ve sağ anterolateral torakotomi(SALT) yapılan hastaların yaş ortalamaları sırasıyla 48±26,6 ve 42±18,2 aydı. Hastaların 12'sinde (%85,7) yüksek venöz tip atriyal septal defekt tespit edildi. En sık kullanılan cerrahi tedavi çift yama tekniği olup, bu yöntem 12 hastada (%85,7) kullanıldı. Bunu, bir hastada (%7,1) tek yama onarımı izledi. SIAVT tekniği ile ameliyat edilen bir hastada (%7,1) Warden prosedürü uygulandı. Hastaların kross klemp, kardiopulmoner bypass ve toplam operasyon süreleri, tıbbi literatürde bildirilenlerle benzer olarak izlendi.
Sonuç: SIAVT ve SALT , konjenital kalp cerrahisinde sıkça kullanılan minimal invaziv cerrahi tekniklerdir. Yeterli deneyim kazandıktan sonra, bu yöntemlerin her ikisi de PAPVD onarımında güvenle uygulanabilir.

Anahtar Kelimeler: Anormal pulmoner venöz dönüş, pediatri, minimal invaziv cerrahi prosedür, torakotomi

Introduction

It is estimated that 1% of all congenital heart defects (CHD) involve a partial anomalous pulmonary venous connection (1). It is a congenital anomaly characterized by one or more pulmonary veins, but not all, draining directly or indirectly into the right atrium instead of the left atrium. Indirect ways of pulmonary venous drainage might be tributaries of the superior vena cava (SVC), inferior vena cava (IVC) , coronary sinus or left innominate vein (2). There are more than 20 anatomical variations of PAPVR. However , abnormal drainage of the pulmonary veins originating from the right lung is the most prevalent form (3). An additional

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secundum atrial septal defect (ASD) is observed in approximately 80% of patients (4). In approximately 50% of these, ASD is of the sinus venosus type (5).

The application of minimally invasive techniques in congenital cardiac surgery are gradually increasing in the pediatric population. Due to their safe application and cosmetic benefits, these techniques are widely accepted as the "new standard" by many centers, particularly in patients with low-risk CHD. The most significant drawbacks of these techniques are that achieving a clear surgical exposure is more difficult than the conventional sternotomy and that a considerably long learning period is required for safe surgery (6). Currently, the combination of the right-sided PAPVR and ASD can be repaired safely by using a variety of minimally invasive techniques (6-8).

In this study, we hypothesized that minimally invasive surgical techniques can be safely used by experienced surgeons in pediatric patients with PAPVR. In order to investigate the safety and reproducibility of minimally invasive techniques in the surgical repair of the right-sided PAPVR and ASD in pediatric patients, we analyzed the data on perioperative morbidity and mortality, comparing them with the data in the current literature.

Material and Methods

This retrospective study was conducted between March 2019 to January 2023, by reviewing the medical records of all pediatric patients diagnosed with right-sided PAPVR concomitant with ASD and operated using minimally invasive surgical techniques. Excluded from the study were the patients with Scimitar syndrome or other concomitant complex cardiac anomalies as well as patients with conduction abnormalities before surgery. Indications for surgery include presence of an ASD with at least two pulmonary veins draining to right atrium, SVC or its tributaries resulting in dilatation of right sided cardiac structures or significant left-to-right shunt leading to Qp/Qs ratio of greater than 1.5:1 on an invasive cardiac catheterization study. In selected cases where the pulmonary vein connections could not be clearly observed by echocardiography, computed tomography imaging was utilized (9).

The patients were operated on after the localization of all pulmonary veins to the right atrium or SVC was determined. Since 2020, in accordance with our clinical policy we have started to use minimally invasive surgical techniques in low risk cardiac pathologies. RIAVT has become the most preferred technique in our clinic. In addition, we also frequently employ RALT. The surgical method that would be used was left to the discretion of the senior cardiac surgeons performing the operation. The perioperative data of the patients' including cardiopulmonary bypass time, cross-clamp time, postoperative cardiac rhythm, total operation time, total drainage, total intensive care unit time and total hospital stay time were obtained.

Statistical Analysis

Categorical measurements were summarized as numbers and percentages, and numerical measurements were summarized as mean and standard deviation. All statistical analyses were performed using IBM SPSS (Statistical Package for the Social Sciences) version 25.

Surgical Technique

Central cannulation was performed in all patients. In patients who underwent RALT and RIAVT, 4-5 cm incisions were made in the fourth intercostal space to gain access to the thorax (Fig.1a). After retracting the lung from the surgical field with a moist sponge, the pericardium was opened 2 centimeters anterior to the phrenic nerve in the superior inferior direction. Holding sutures were placed at frequent intervals. Removing the slack of the previous holding suture after each one is a crucial manipulation for improving surgical exposure and bringing cardiac structures closer together. The atrial appendage was then retracted in an inferior direction with another suspending suture. This procedure provides exceptional exposure of the aortic root (Fig.1b). After exposing the entire aortic root and ascending aorta, the pericardium over the SVC was released without damaging the phrenic nerve by sharp dissection up to the junction with the left innominate vein. All anomalous pulmonary veins draining into SVC or cavoatrial junction and azygos vein were exposed (Fig.1b). After aortic cannulation, selective cannulations of the SVC and IVC were performed. SVC cannulation was performed at the junction point of right and left innominate veins with either a right-angle metal-tipped cannula or a small diameter plastic straight cannula (Fig.1c). Subsequently, cardiopulmonary bypass was initiated and continued under mild hypothermia (34˚C). The heart was then arrested with del-Nido cardioplegia solution after achieving targeted temperature.

Three different repair techniques were used in patients following cardiac arrest; double-patch repair, single patch repair and Warden procedure. The preferred surgical method was chosen after careful consideration of several aspects. These include; (1) distance between the entrance point of pulmonary venous connection to SVC and cavoatrial junction, (2) the presence of persistent left SVC (PLSVC) and the diameter of right SVC and (3) the localization of the ASD in the interatrial septum. In cases with wide SVC and short distance (<1 cm) between the pulmonary venous connection and the cavoatrial junction, the single patch technique was used. If the patient had a well-developed superior limbus combined with a small SVC or if the pulmonary venous connection to the SVC was more than 1 cm from the cavoatrial junction, the double patch or Warden procedure was preferred. All restrictive ASDs was expanded towards the direction of the fossa ovalis. If the patient's PLSVC or ASD was not of the high venosum type, we preferred the double patch technique or Warden procedure rather than the single patch technique.

Care was taken to ensure that the patch was sewn generously to avoid stenosis in the baffle, and the double patch or Warden procedure was favoured in every case where SVC stenosis was suspected to develop after the interior patch was sutured (Fig.2).

Results

During the study period, 14 patients underwent surgical repair. Five (36%) of them were female and 9 (64%) were male. The mean ages of the patients who underwent RIAVT and RALT 48±26.6 and 42±18.2 month, respectively. The mean weight of the RIAVT and RALT patients were 14.6 kg and 24 kg, respectively.

In the majority of cases, 12 patients (85.7%) had both the right upper and right middle lobe pulmonary veins draining into right-sided heart structures. ASD of the high venosum type was identified in 12 patients (85.7%). On 12 patients (85.7%), the double patch technique was employed most frequently in surgical treatment. This was then followed by a single patch repair in one (7.1%) patient. The Warden procedure was performed on only one patient (7.1%) underwent RIAVT surgery. In both minimally invasive techniques, the cross-clamp time, CPB time and total operation time were comparable. Nevertheless, due to the complexity of the surgical procedures involved in the double patch and Warden procedures, the duration of these operations was slightly longer than those of the single patch technique. Table 1 lists the demographic, anatomical and surgical details of the patients.

The postoperative intensive care morbidity variables of the patients, such as the need for mechanical ventilation, the total length of stay in the intensive care unit, and the total amount of drainage were similar to each other, and no significant difference was observed between the RIAVT and RALT techniques. However, CPB, cross-clamp and total operation time was slightly longer in the RALT group. This was attributed to the fact that fewer patients were operated on and less experience was gained in the group with less RALT.

During the surgical procedure, none of the patients required inotropic support. All patients were extubated within the first six hours postoperatively. No mortality was observed in any patient. In addition, no complications that would prolong their hospital stay were observed in any of the patients.

In the early postoperative period, a heart rhythm other than normal sinus rhythm was observed in two patients. Since the heart rates of these patients were within the acceptable range, no additional intervention was performed, and they were discharged with close monitoring. In the first month of follow-up, it was observed that one patient with low atrial rhythm treated with the double patch technique returned to normal sinus rhythm. The rhythm of the remaining patient did not change in the first month of follow-up and close monitoring continued. The variables in postoperative intensive care unit and early-term following discharge are shown in Table 2.

Table 1. Demographic and Surgical Data of the Patients

	Minimal Invasive Surgery(N=14)	
Route of Surgery	RIAVT(N=10)	RALT (N=4)
Age (month)	48±26.6	42±18.2
Weight (kg)	14.6±3.51	24±2.64
Male Sex, n (%)	6(60%)	3 (75%)
Right pulmonary veins		
RUL	1(10%)	1(25%)
RUL+RML	9(90%)	3(75%)
RUL+RML+RLL	0(0%)	0(0%)
High venosum ASD, n	9(90%)	3(75%)
Surgical Technique		
Double Patch ,n	9 (90%)	3(75%)
Single Patch ,n	0(0%)	1(25%)
Warden, n(%)	1(10%)	0(0%)
Cross clamp time(min)	35.6±4.9	49±17.7
CPB time(min)	61.6±7.8	75.6±24.5
Operation Duration (h)	4.55±0.62	4.82±0.76

RIAVT; right infra axillary vertical thoracotomy, RALT; right anterolateral thoracotomy, RUL; right upper lobe, RML; right middle lobe, RLL; right lower lobe, ASD; atrial septal defect, CPB; cardiopulmonary bypass

Table 2. The variables in postoperative intensive care unit and early-term following discharge

	Minimal Invasive Surgery(N=14)	
Surgical Approach	RIAVT(N=10)	RALT (N=4)
Mechanic Ventilator Time (h)	6.4±2.2	5.8±3.3
ICU stay time (d)	2.2±0.2	2.4±0.6
Total Drainage (ml)	171±37.5	185±34.1
Total Hospital Stay (d)	4.1±0.6	4.2±0.4
Early Postoperative rhythm		
Normal sinus rhythm	8	4
Lower atrial rhythm	2*	0
Junctional rhythm	0	0
Rhythm at the postoperative 1st month		
Normal sinus rhythm	9	4
Lower atrial rhythm	1*	0
Junctional rhythm	0	0

RIAVT; right infra axillary vertical thoracotomy, RALT; right anterolateral thoracotomy, RUL; right upper lobe ,RML; right middle lobe, RLL; right lower lobe ICU ; intensive care unit ,* Double Patch repair was used

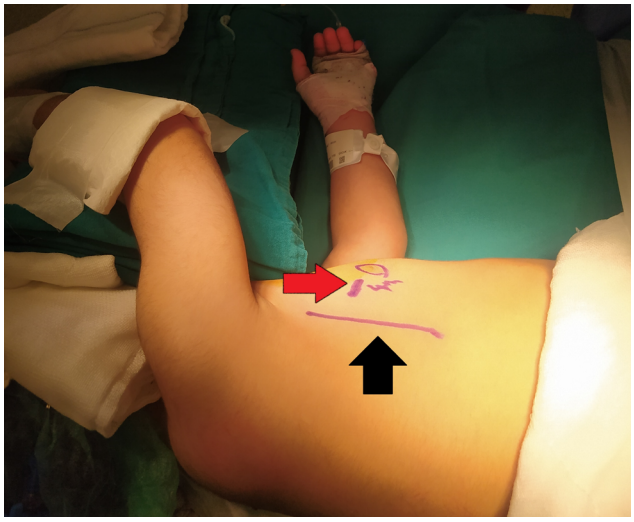

Fig 1a: Preoperative marking of the patient : the red arrow indicates the 4th intercostal space , the black arrow shows the vertical skin incision to be performed.

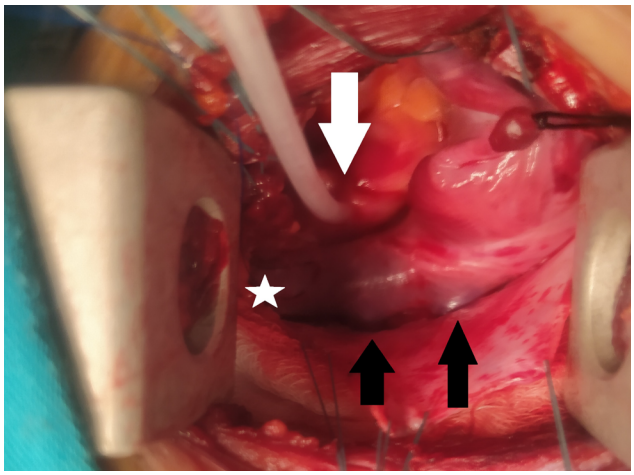

Fig 1b: The view of he surgical field from the primary surgeon's side: White asteriks indicates the SVC , black arrows indicate anomalous pulmonary veins draining to SVC and atriocaval jounction and white arrow shows aortic purse string sutures.

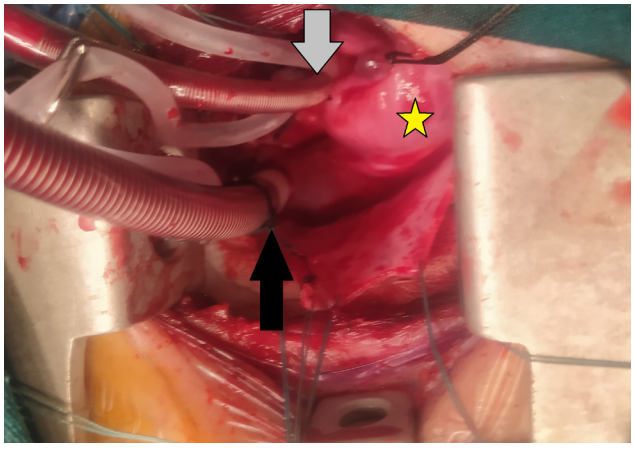

Fig 1c: Central cannulation of the patient: Black and grey arrow indicate SVC and aortic cannulations, respectively.Yellow asteriks marks the right atrium..

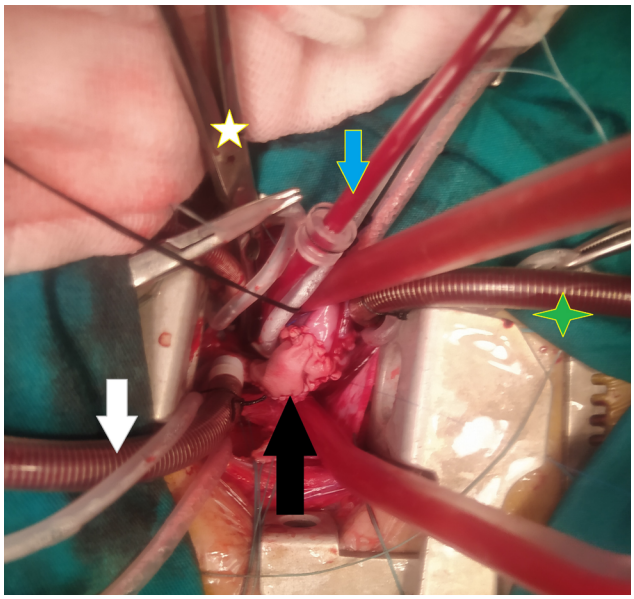

Fig 2: Surgical field of the patient during cardioplegic arrest : White , blue and black arrows indicate SVC cannula, cardioplegia cannula and pericardial patch , respectively. White arrow marks aortic cross clamp and green arrow indicates IVC cannula.

Discussion

The use of minimally invasive surgical methods in congenital heart anomalies continues to increase. Contrary to the disadvantages of traditional median sternotomy, minimally invasive techniques provide superior cosmetic outcomes and greater patient and parent satisfaction. In many cases of CHD, minimally invasive surgical procedures have become the standard of care in recent years as a result of advancing surgical techniques and growing experience. Among these techniques, right vertical or horizontal infra-axillary thoracotomy, ministernotomy and RALT are the techniques used most commonly. In many high-volume centers around the world, low-risk CHD have substantially been repaired using these techniques, and the conventional median sternotomy has gradually been used less frequently. The most common congenital heart anomaly in which these techniques are used is secundum ASD. In addition, many other anomalies such as, PAPVR, ventricular septal defect, partial or complete atrioventricular septal defect, double chamber right ventricle and tetralogy of fallot can also be repaired using these methods. However, in more complex congenital heart anomalies, conventional sternotomy is still recommended because minimally invasive techniques cannot provide sufficient exposure to allow comfortable access to the cardiac structures to perform successful surgical repair.

RALT and RIAVT have been used safely in the surgical repair of right-sided PAPVR and ASD. Unlike the standard secundum ASD repair, selective cannulation of the superior vena cava is one of the most challenging parts of the surgical procedure. It is troublesome to insert metal-tipped right-angle cannulas into the SVC, especially in cases where the anomalous

pulmonary vein has connected to the SVC at superior segments. In these cases, the use of smaller, straight cannulas may provide simplicity in surgical technique. Alternatively, the thorax may be reentered through the 3rd intercostal space for more comfortable SVC and aortic cannulation. Thus, the surgical procedure can be completed using both intercostal spaces. However, in certain cases where the thorax has been entered through the fourth intercostal space, IVC cannulation and snaring may still be challenging. Especially for inferior type sinus venosus secundum ASDs with weak inferior rims, it is crucial to cannulate the IVC as distally as possible. For this reason, we cannulate the IVC with CPB support in cases where exposure is insufficient and do not hesitate to cleave the lower rib to gain enough exposure instead of performing peripheral cannulation if necessary. As a result of our experience with post-discharge patient follow-ups, we did not observe any cosmetic or functional abnormalities in patients whose lower ribs were severed and repaired at the conclusion of the surgical procedure.

There are many studies that report PAPVR repair could be conducted with minimally invasive approaches. Zubritskiy et al.(8) reported that they successfully performed the Warden procedure using the right infra axillary thoracotomy approach in 21 pediatric patients with an average weight of 25 kg, and diagnosed with PAPVR and ASD. In that study population, central cannulation was performed in all patients and surgical repair was performed with ventricular fibrillation under CPB support. No morbidity or mortality was observed in the patients in the postoperative period and all patients were discharged with a normal sinus rhythm. Similarly, Rao et al.(7) reported that they performed the surgical treatment of 14 pediatric patients with PAPVR and sinus venosus type ASD via a modified RIAVT. The average weight of the patients was 21 kg. Peripheral cannulation was performed using the internal jugular vein and femoral vein in patients instead of central venous cannulation. They stated that with this method, the venous cannulas that disturb clear exposure of the surgical field can be avoided and the skin incision can be kept shorter (7). In the study of Amirghofran et al. (10), in which minimally invasive methods and conventional sternotomies were compared in PAPVR and ASD repair in pediatric and adult patients, it was reported that surgical repair of patients could be successfully performed with an anterior mini thoracotomy method. The mean age of 25 pediatric patients who underwent minimally invasive approaches was 4.99 years. It was stated that peripheral cannulation was avoided and central venous cannulation was applied to these patients whenever possible. In addition, high SVC cannulation was not used selectively in pediatric patients, instead, a malleable cannula was sent through the right atrial appendage to the upper segments of the SVC to provide drainage. In all patients, double patch repair was performed and SVC stenosis was only detected in a 3.5-year-old patient. This patient was reoperated to relieve the SVC stenosis. The authors concluded that the anterolateral mini thoracotomy method can be safely applied to pediatric patients (10).

In PAPVR repair, the superiority of single patch, double patch and Warden procedures to each other in terms of the development of sinus node dysfunction, SVC stenosis or pulmonary venous baffle stenosis in the long term has not been fully clarified. Each technique has its own advantages and disadvantages. Single patch repair is a practical and reproducible method in patients with large SVCs and where the pulmonary venous connection is close to the SVC-right atrial junction. However, it is not recommended in cases where the SVC is small and the pulmonary venous connection is away from the SVC-right atrial junction due to the development of potential stenosis in SVC or baffle. In these patients, double patch repair or the Warden procedure is preferred. Although SVC stenosis can usually be avoided with double patch repair, it may still develop in cases where the pulmonary venous connection is very high in SVC or the left SVC is accompanied and the right SVC is smaller than the normal size (8,11). In such cases, the Warden procedure may be a more viable option. Although the long-term survival results of both surgical techniques are excellent, there are publications showing that the Warden procedure gives better results than the double patch technique in terms of sinus node dysfunction (12, 13). Despite the fact that the Warden procedure appears to be a safe and effective surgical technique, it is not without its drawbacks. Particularly in cases where the right atrial appendage is underdeveloped and the SVC caudal segment is short, end to end anastomosis of the SVC to the right atrial appendix cannot be performed and pericardial augmentation or PTFE tube graft interposition may be required to reduce anastomotic tension. In addition, the pectinate muscles in the appendix must be radically resected to prevent stenosis at SVC- right appendix anastomosis (3). Patients undergoing this surgery should be monitored for stenosis in this region during the follow-up period (2, 3, 14).

Double patch repair has been our first choice in the vast majority of PAPVR cases in our clinic (2). This was because all of our patients had a well-developed SVC and we were confident that the patch on the SVC lateral wall would greatly relieve SVC flow. Although it is thought to cause sinus node dysfunction in the long term (11), we did not encounter such a complication in the patients we operated on with this method in our clinic. None of our patients needed a permanent pacemaker. There are many scientific publications showing that the Warden procedure is a superior surgical method that offers very good results in terms of postoperative mortality and morbidity in PAPVC patients (12, 15-18). In one of our patients, we preferred to use the Warden procedure because the pulmonary vein-SVC connection was located very superiorly. There were three reasons why we did not prefer the double patch in this patient. The first reason was that the diameter of the SVC was not wide enough and we suspected that the baffle to be created might critically narrow the total diameter of the SVC. The second

reason was that a very long and wide buff had to be created to divert the pulmonary vein to the left atrium. The third reason was that internal baffle creation was a technically difficult procedure due to the high insertion of the pulmonary vein to the SVC and the presence of the venous cannula close to the surgical area.

The most important limitation of this study is that the small number of cases in the RIAVT and RALT groups made it impossible to compare these two minimally invasive techniques with each other in terms of early and long-term results. In addition, the long-term echocardiographic follow-up of the patients has not been completed yet. After obtaining these data, the reliability of these two techniques used in terms of the development of stenosis in the venous structures where patches are used and the development of cardiac rhythm problems can be clarified.

Conclusion

In conclusion, minimally invasive surgical techniques have better cosmetic results than conventional sternotomy incisions. All surgical techniques that are used in right-sided PAPVR repair with conventional sternotomy methods can also be applied easily and safely with minimally invasive methods. In the following years, minimally invasive approaches may become standard practices in right-sided PAPVR repair.

Ethics Committee Approval: The study was evaluated by the Ankara City Hospital Ethics committee and received ethical approval on 01.02.2023 with the number E2-23-3285. Every procedure was carried out in compliance with the moral guidelines outlined in the 1964 Helsinki Declaration and its subsequent revisions.

Informed Consent: Informed consent was obtained from each subject.

Peer-review: Internally peer-reviewed.

Authorship Contributions: MY wrote the primary draft of the article. BST took part in collecting the data of the article. ANE took a primary role in the preparation of the ethics committee file of the article. YŞÖ and HAG worked on the editing of the article. AA reviewed the final version of the article. The authors have read and approved the final manuscript.

Conflict of Interest: The authors disclosed no conflicts of interest.

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