

Cite this article as: Kulyabin YY, Voitov AV, Nichay NR, Soynov IA, Zubritskiy AV, Bogachev-Prokophiev AV. Single-stage off-pump repair of coarctation of the aorta and ventricular septal defects in children. *Interact CardioVasc Thorac Surg* 2022; doi:10.1093/icvts/ivac186.

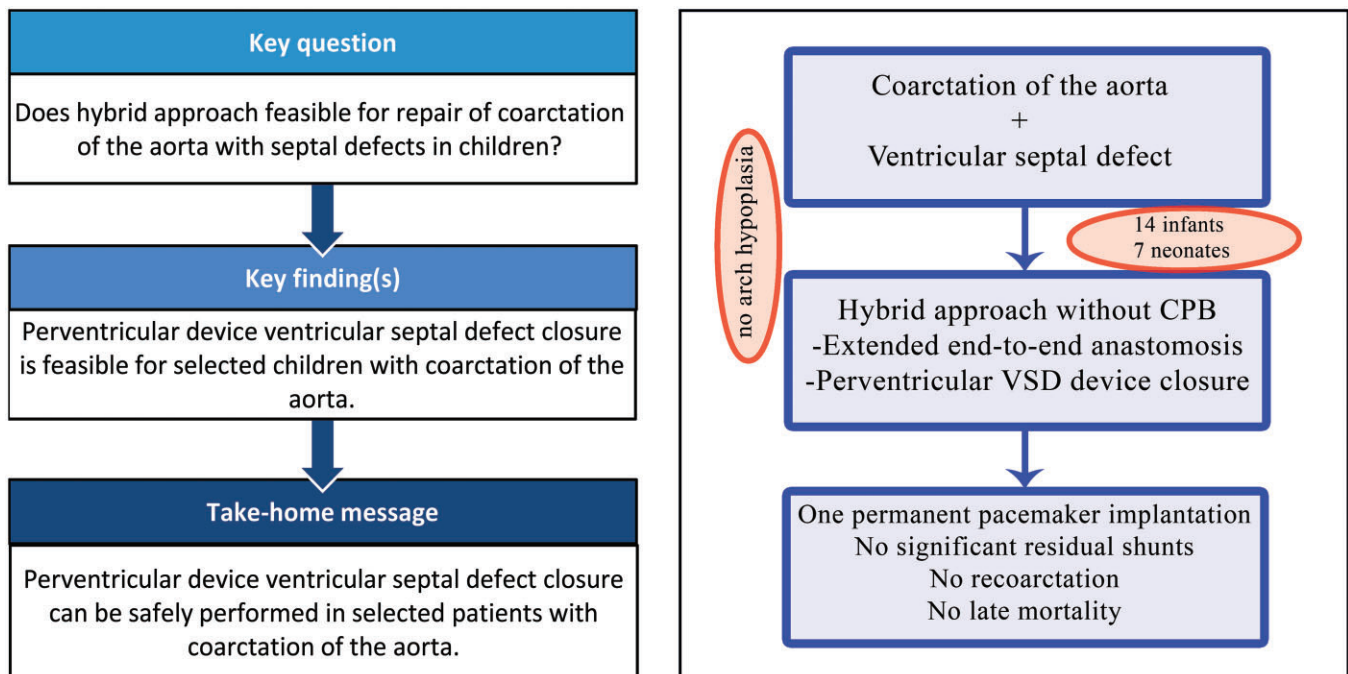
Single-stage off-pump repair of coarctation of the aorta and ventricular septal defects in children

Yuriy Y. Kulyabin *, Alexey V. Voitov, Nataliya R. Nichay, Ilya A. Soynov, Alexey V. Zubritskiy and Alexander V. Bogachev-Prokophiev

Department of Pediatric Cardiac Surgery, E. Meshalkin National Medical Research Center, Novosibirsk, Russian Federation

* Corresponding author. 630055, Rechnunovskaya str., 15, Novosibirsk, Russian Federation. Tel.: +7(953)-888-9890; fax: +7(383)-332-2437; e-mail: y.y.coolyabin@gmail.com (Y. Kulyabin).

Received 4 June 2022; accepted 26 June 2022



Abstract

OBJECTIVES: The appropriate approach for surgical repair of coarctation of the aorta with a ventricular septal defect (VSD) remains controversial. This study evaluated the outcomes of primary repair of VSDs with periventricular device closure without cardiopulmonary bypass through a left thoracotomy in patients without arch hypoplasia.

METHODS: We selected 21 patients aged <1 year, including 7 neonates, who underwent repair of coarctation of the aorta with periventricular device closure of a VSD.

RESULTS: The median occluder size was 6 (range, 5–8) mm. The median mechanical ventilation time was 14 (range, 2–68) h, and the median duration of hospital stay was 11 (range, 7–16) days. No reoperations were required to correct VSD shunting, and the median residual shunt size was 1 (range, 1–2) mm. The median follow-up period was 13 (range, 4–31) months. No late deaths were reported, and no

Meeting presentation: The abstract was submitted for the 33rd EACTS annual meeting in Lisbon.

haemodynamically significant pressure gradient at the anastomotic site was observed. The median distal aortic arch z-score was 0.39 (range, -0.1 to 0.9). Only 1 patient had a permanent pacemaker implanted towards the end of the follow-up period.

CONCLUSIONS: Periventricular device closure can be used safely for closure of VSD in children with coarctation of the aorta without a hypoplastic aortic arch, even in neonates, to reduce the risk of prolonged cardiopulmonary bypass. This hybrid approach can be performed with a low incidence of rhythm disturbances and residual shunting. However, a meticulous assessment of the VSD anatomy is essential to avoid any unfavourable events.

Keywords: Ventricular septal defect • Coarctation of the aorta • Hybrid surgery

LIST OF ABBREVIATIONS:

AV	Atrioventricular
CoAo	Coarctation of the aorta
CPB	Cardiopulmonary bypass
PVDC	Periventricular device closure
VSD	Ventricular septal defect

INTRODUCTION

Coarctation of the aorta (CoAo) is frequently associated with septal defects in neonates and infants. However, the appropriate approach for its surgical repair remains controversial. Most centres prefer a single-stage repair under cardiopulmonary bypass (CPB) with simultaneous closure of the septal defects [1, 2]. Aortic arch reconstruction under CPB with hypothermia is necessary in patients with aortic arch hypoplasia, but small patients are vulnerable to the effects of prolonged CPB time and myocardial ischaemia. Therefore, various perfusion methods are used to minimize CPB-induced complications during repair [3–5]. Repair through a left thoracotomy with or without palliative pulmonary artery banding can be performed in some patients with CoAo without arch hypoplasia. A hybrid approach with periventricular device closure (PVDC) of the ventricular septal defect (VSD) could also be an option for some patients to avoid CPB for primary repair.

We present our experience with the hybrid approach of aortic arch reconstruction with PVDC of VSD for the treatment of complex CoAo.

PATIENTS AND METHODS

Ethical statement

The ethics committee of the E. Meshalkin National Medical Research Center approved this study. Informed consent was obtained from patients' parents or guardians.

Participants

We retrospectively evaluated the medical records of >200 patients with obstructive aortic arch lesions who underwent surgery at the Novosibirsk National Medical Research Center from January 2014 to December 2019. A total of 21 patients (12 boys and 9 girls; 7 neonates) were included in the study according to the inclusion criteria (Fig. 1). We included patients aged <1 year with CoAo without severe aortic arch hypoplasia with perimembranous or muscular VSD sized <7 mm that was suitable for device closure. Patients with severe aortic arch hypoplasia

(transverse arch z-score < -2.0) or interrupted aortic arch, large VSDs (>7 mm), inlet-type VSDs or concomitant intracardiac lesions (large atrial septal defects, multiple VSDs, subvalvular aortic stenosis and aortic valve stenosis) were excluded from the analysis.

Surgical technique

Aortic arch hypoplasia was assessed using the z-score (Peterson's calculator) [6]. CoAo without arch hypoplasia or with only distal arch hypoplasia (distal arch z-score < -2.0; transverse aortic arch z-score > -2.0) was considered for extended end-to-end anastomosis without CPB.

CoAo repair was performed through a posterolateral thoracotomy in the third intercostal space. The isthmus, descending aorta and aortic arch with brachiocephalic vessels were dissected. The ductus arteriosus was ligated, and the aorta was closed with Satinsky clamps. Next, the isthmus with ductal tissue was resected, and an extended end-to-end anastomosis was performed.

When single-stage repair with PVDC was considered, a ministernotomy was performed in most patients (18 patients); in 3 patients, PVDC was performed through a left lateral thoracotomy, extending it anteriorly to visualize the RV wall (the RV wall was lifted with the forceps to find the proper puncture site). The right ventricle was punctured opposite the VSD under transoesophageal echocardiography monitoring. A guidewire was introduced into the left ventricle through the VSD, and a MemoPart VSD occlusion device (Lepu Medical Technology, Shanghai Shape Memory Allow Co., Ltd., Shanghai, China) of appropriate size (VSD size + 1 mm) was placed in a stepwise manner.

Statistical methods

The Shapiro–Wilk W-test was performed to assess the normality of continuous variables before further analysis. Continuous data are presented as medians and ranges. Categorical data are presented as rates and frequencies. Statistical analyses were performed using Stata 13 (StataCorp LP, College Station, TX, USA).

RESULTS

The baseline characteristics are presented in Table 1. No in-hospital deaths were reported. The median occluder size was 6 (5–8) mm. Seven patients in the cohort were neonates.

One patient presented with temporary atrioventricular (AV) conduction disorder (second-degree AV block, Mobitz type I), which was resolved on the third postoperative day after glucocorticosteroid and dobutamine drug therapy in the intensive care unit. No other significant rhythm disturbances were noted.

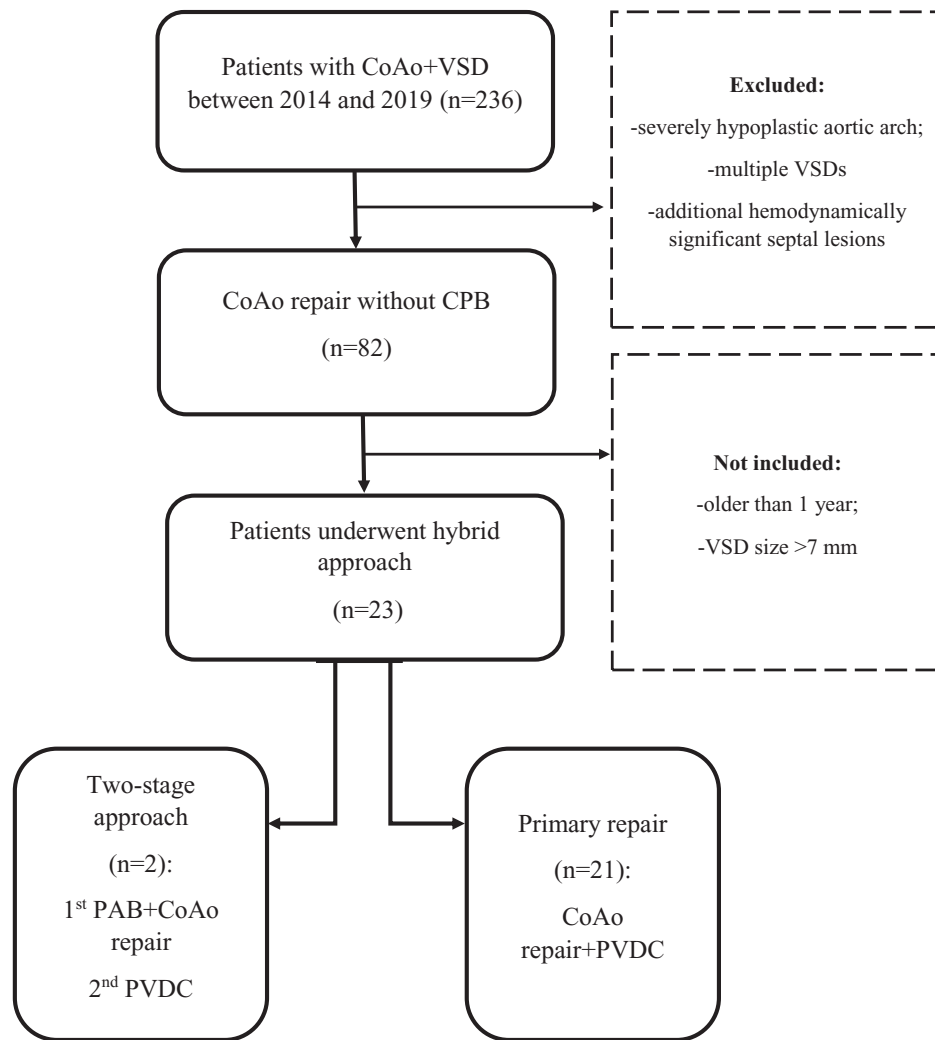


Figure 1: Flow chart of the selection of patients with coarctation of the aorta for retrospective analysis. CoAo: coarctation of the aorta; CPB: cardiopulmonary bypass; PAB: pulmonary artery banding; PVDC: periventricular device closure; VSD: ventricular septal defect.

Table 1: Baseline characteristics

Parameter	n = 21
Gender	
Female	9 (42.8%)
Male	12 (57.1%)
Age, days	71.1 (20-320)
Weight, kg	4.8 (3.4-11.2)
Neonates, n (%)	7 (33.3)
Z-score of aortic isthmus	-4.9 (-5.79--4.75)
Z-score, distal aortic arch	-1.66 (-1.81--1.26)
Device size, mm	6 (5-8)
Device type, n	
I	4
II	10
IV	7

Table 2: Postoperative data

Parameter	N = 21
Early deaths, %	0
Duration of mechanical ventilation, h	14 (2-68)
Hospital stay, days	11 (7-14)
Pressure gradient, mmHg	8.3 (6-12)
Follow-up, months	13 (4-31)
Recoarctation rate, %	0
Residual VSD rate, n	4 (19)
Residual VSD shunt, mm	1 (1-2)
Rhythm disturbance, n	1 (4.3)
Z-score distal arch	0.39 (-0.1 to 0.9)
Survival, %	100

VSD: ventricular septal defect.

The median mechanical ventilation time was 14 (2-68) h. The postoperative results are presented in Table 2. The median follow-up period was 13 (4-31) months. No late deaths were reported (Table 2). No haemodynamically significant pressure gradient was observed at the anastomotic site, and the median distal aortic arch z-score was 0.39 (-0.1-0.9). No reoperations

were required to correct VSD shunting, and the median residual shunt size was 1 (1-2) mm.

Only 1 patient presented with a rhythm disturbance 1 year postoperatively. He had a temporary second-degree AV block in the early postoperative period, which was reversed; a permanent pacemaker implant was recommended.

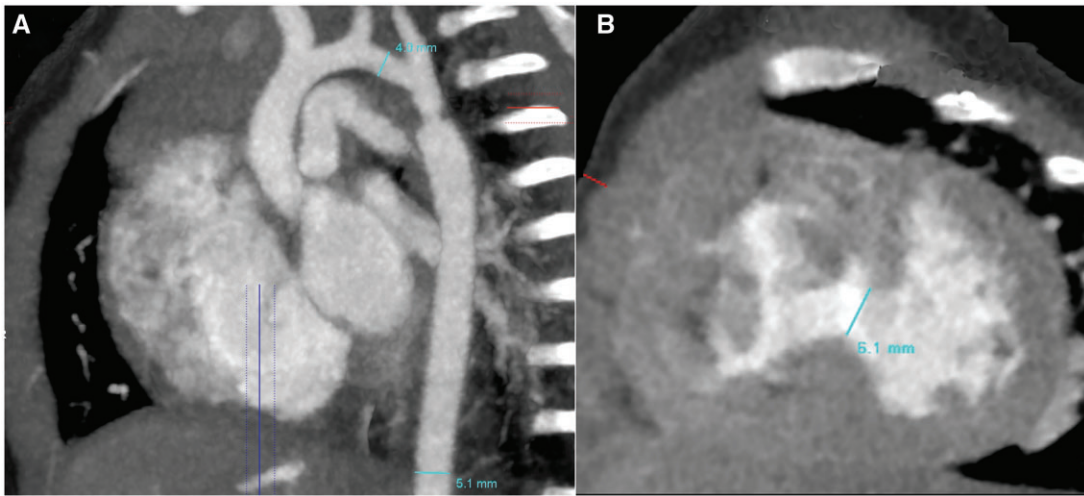


Figure 2: Patient with (A) coarctation of the aorta and (B) muscular ventricular septal defect.

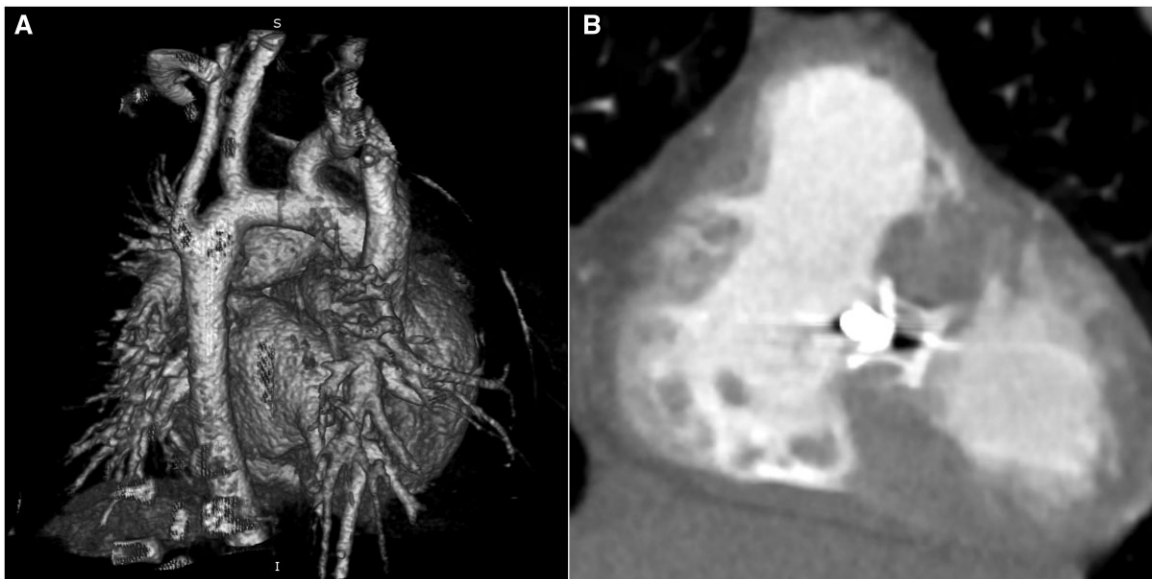


Figure 3: Result of the hybrid approach. Computed tomography imaging showing (A) the aortic arch after extended end-to-end anastomosis and (B) showing the device in the ventricular septum.

DISCUSSION

Surgical repair of VSD in infants is routinely performed under CPB with a low complication rate and few deaths. Although this method has been partially replaced by transcatheter device closure to improve clinical outcomes, the endovascular approach is limited by the vessel size and the anatomical features of the VSD [7]. PVDC, described by Amin *et al.* in 1998, is an increasingly used hybrid method that has no restrictions, unlike the percutaneous transcatheter method [8]. However, the risk of valve distortion or heart block is higher in small patients. Thus, the results of device closure are controversial. Our recent randomized study showed the suitability of this method in patients of any age [9]. Although few patients had transitory rhythm disturbances during the study, some of them returned with complete AV block and had a pacemaker implanted permanently. Some authors have mentioned that, in case of early complete AV block due to

mechanical injury during manipulation of the delivery system or during device implant, conversion to open surgery should be considered [7, 10]. The cause of late-onset complete AV block remains unknown, but it probably occurs as a result of chronic inflammation and fibrosis due to the device, which is more likely to occur in infants with large defects.

The simultaneous closure of VSD and atrial septal defects or patent ductus arteriosus has been reported [11, 12]. Our experience has shown that this method is safe and effective in treating congenital septal defects that do not require surgical repair under CPB. Indications for PVDC should not be overestimated because implanting a large device may lead to more severe complications than those of the conventional approach, such as aortic regurgitation, tricuspid regurgitation and rhythm disturbances. We used a cut-off VSD size ≤ 7 mm for PVDC in children. However, the VSD size should be correlated with the patient's body surface area.

CoAo repair in small patients often requires aortic reconstruction under CPB with deep hypothermia or selective cerebral perfusion for brain protection. However, this approach poses additional risks to the patient, such as renal injury and neurological events, that could compromise the postoperative recovery [5, 13, 14]. In some patients, CoAo without arch hypoplasia can be repaired through a left thoracotomy; however, the presence of VSD usually necessitates primary repair under CPB. We selected patients with perimembranous or large muscular VSD that required closure for CoAo and VSD repair using the hybrid approach. This method helps to avoid repair under CPB and a prolonged stay in the intensive care unit. However, the hybrid approach has some limitations that are related mainly to the patient's body mass and VSD size. We did not include in this study the patients with VSDs with well-defined margins that are too large for PVDC, in whom we considered placing a band on the pulmonary trunk to allow the patient's growth until the subsequent assessment. As one could see, only a few patients from the whole CoAo + VSD cohort underwent repair using PVDC. This method could be especially helpful for the closure of muscular VSDs in the mid-portion of the septum (Figs. 2, 3) with a low risk of residual shunting.

LIMITATIONS

The limitations of our analysis include its retrospective nature, the small number of cases reviewed and the short follow-up period (median was approximately 1 year). Although the intermediate-term follow-up results seem to be satisfactory, it is necessary to assess long-term rhythm disturbance rates. This study did not include a comparison group, and further studies with a control group are required to assess the effectiveness of PVDC.

CONCLUSIONS

PVDC can be used safely for closure of VSD in children with CoAo without a hypoplastic aortic arch, even in neonates, to reduce the risk of prolonged CPB. This hybrid approach can be performed with a low incidence of rhythm disturbances and residual shunting; however, a meticulous assessment of the VSD anatomy is essential to avoid any unfavourable events. Further studies are required to define the best candidates for this approach.

FUNDING

This study did not receive any specific financial support from funding agencies in the public, commercial or not-for-profit sectors.

Conflict of interests: none declared.

Data availability

The data underlying this article are available in the article.

AUTHOR CONTRIBUTIONS

Yuriy Kulyabin: conceptualization; investigation; methodology; writing—original draft; writing—review & editing. **Alexey Voitov:** conceptualization; data curation; methodology; visualization. **Nataliya Nichay:** data curation; investigation; methodology; writing—review & editing. **Ilya Soynov:** data curation; formal analysis; methodology. **Alexey Zubritskiy:** data curation; formal analysis; investigation; visualization. **Alexander Bogachev-Prokophiev:** conceptualization; project administration; writing—review & editing.

REFERENCES

- [1] Tchervenkov CI, Tahta SA, Jutras L, Béland MJ. Single-stage repair of aortic arch obstruction and associated intracardiac defects with pulmonary homograft patch aortoplasty. *J Thorac Cardiovasc Surg* 1998;116: 897–904.
- [2] Cho SH, Cho YH, Jun TG, Yang JH, Park PW, Huh J *et al.* Outcome of single-stage repair of coarctation with ventricular septal defect. *J Card Surg* 2011;26:420–4.
- [3] Kreuzer M, Sames-Dolzer E, Schausberger L, Tulzer A, Ratschiller T, Haizinger B *et al.* Double-arterial cannulation: a strategy for whole body perfusion during aortic arch reconstruction. *Interact CardioVasc Thorac Surg* 2018;27:742–8.
- [4] Hammel JM, Deptula JJ, Karamlou T, Wedemeyer E, Abdullah I, Duncan KF. Newborn aortic arch reconstruction with descending aortic cannulation improves postoperative renal function. *Ann Thorac Surg* 2013;96: 1721–6. discussion 1726.
- [5] Kulyabin YY, Bogachev-Prokophiev AV, Soynov IA, Omelchenko AY, Zubritskiy AV, Gorbatykh YN. Clinical assessment of perfusion techniques during surgical repair of coarctation of aorta with aortic arch hypoplasia in neonates: a pilot prospective randomized study. *Semin Thorac Cardiovasc Surg* 2020;32:860–71.
- [6] Pettersen MD, Du W, Skeens ME, Humes RA. Regression equations for calculation of z scores of cardiac structures in a large cohort of healthy infants, children, and adolescents: an echocardiographic study. *J Am Soc Echocardiogr* 2008;Aug21:922–34.
- [7] Yang L, Tai BC, Khin LW, Quek SC. A systematic review on the efficacy and safety of transcatheter device closure of ventricular septal defects (VSD). *J Interv Cardiol* 2014;27:260–72.
- [8] Amin Z, Berry JM, Foker JE, Rocchini AP, Bass JL. Intraoperative closure of muscular ventricular septal defect in a canine model and application of the technique in a baby. *J Thorac Cardiovasc Surg* 1998;115:1374–6.
- [9] Voitov A, Omelchenko A, Gorbatykh Y, Zaitsev G, Arkhipov A, Soynov I *et al.* Outcomes of periventricular off-pump versus conventional closure of ventricular septal defects: a prospective randomized study. *Eur J Cardiothorac Surg* 2017;51:980–6.
- [10] Huang X-S, Luo Z-R, Chen Q, Yu L-S, Cao H, Chen L-W *et al.* A comparative study of periventricular and percutaneous device closure treatments for isolated ventricular septal defect: a Chinese single-institution experience. *Braz J Cardiovasc Surg* 2019;34:344–51.
- [11] Voitov A, Omelchenko A, Gorbatykh Y, Bogachev-Prokophiev A, Karaskov A. Combined periventricular septal defect closure and patent ductus arteriosus ligation via the lower ministernotomy approach. *Interact CardioVasc Thorac Surg* 2018;26:323–4.
- [12] Ling Y, Wang Y, Fan Q, Qian Y. Combined periventricular closure of ventricular septal defect and atrial septal defect via lower ministernotomy. *J Cardiothorac Surg* 2018;13:125.
- [13] Algra S, Jansen N, van der Tweel I, Schouten A, Groenendaal F, Toet M *et al.* Neurological injury after neonatal cardiac surgery: a randomized, controlled trial of 2 perfusion techniques. *Circulation* 2014;129:224–33.
- [14] Kulyabin YY, Gorbatykh YN, Soynov IA, Zubritskiy AV, Voitov AV, Bogachev-Prokophiev AV. Bogachev-Prokophiev AV Selective Antegrade Cerebral Perfusion With or Without Additional Lower Body Perfusion During Aortic Arch Reconstruction in Infants. *World J Pediatr Congenit Heart Surg* 2020;11:49–55.