REVIEW ARTICLE

Biventricular repair in double outlet right ventricle with non committed VSD: impossible to possible

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Abstract

Double outlet right ventricle is relatively an uncommon cyanotic congenital heart disease. The most simple meaning of DORV is origin of both aorta and pulmonary artery is from right ventricle. But, in reality, DORV is full of complexities. Its clinical spectrum spans from VSD like presentation to TOF to TGA. Biventricular repair is treatment of choice for DORV committed VSD with or without pulmonary stenosis with excellent results but surgical treatment of DORV non-committed VSD remained controversial. Because of the technical challenges, the common surgical strategy for DORV non-committed VSD is single ventricle pathway. Now, with understanding of the anatomy and physiology of this subset, gradually these difficulties in surgical repair are being overcome. This review article will enlighten the various methods of biventricular repair of DORV non-committed VSD with and without pulmonary stenosis and condensing the indications for univentricular repair to a very limited space.

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Introduction

Double outlet right ventricle (DORV) constitutes 1-3% of all congenital heart diseases and its incidence varies between 3-9 per 100,000 live births [1,2]. Pathologic description of this entity dates back to as early as 1703 but it was Withham who popularized the term "DORV" [3].

Normally, the aorta takes origin from left ventricle and pulmonary arterial trunk from right ventricle but double outlet right ventricle is a type of ventriculo-arterial connection in which both great arteries takes origin either entirely or predominantly from morphologic right ventricle. Presence of a ventricular septal defect (VSD) is the utmost requirement for left ventricle to deliver its output to systemic circulation.

Classification provided by Lev and Bharati [4] guides about relation of VSD with great arteries which helps in surgical correction of DORV. First successful repair by Kirklin and Castaneda in 1977 [5,6] opened the door for surgical management of various forms of DORV. Many techniques have been described with excellent results to tunnel the left ventricular flow to aorta in DORV committed VSD with and without pulmonary stenosis (PS). Biventricular repair of DORV non-committed VSD remained controversial because of poor early and late results.

Since, VSD is located in the inlet portion of right ventricle , construction of an unobstructed tunnel from LV to aorta is extremely difficult and not without severe complications therefore, Fontan pathway is used as an alternative method to relieve the symptoms. Single ventricle pathway, in long terms, is associated with great morbidity and complications of Fontan circulation ultimately require heart or heart and lung transplant. As we all know biventricular repair is always superior than univentricular repair, options were explored regarding 2 ventricular repair for DORV non-committed VSD. In DORV non-committed VSD, this VSD is the only outlet for LV, so providing a circulation in series is the technically most difficult challenge for the surgeons.

Now, a good number of techniques are available which will provide a series circulation without obstructing any outflow tract and ventricular chamber along with excellent early and late results.

Anatomic Consideration of DORV Non-committed VSD (fig 1)

It accounts for 10% of DORV [7]. Both great vessels arise entirely from right ventricle (RV) (200%) [8-10]. According to Van Praagh, in DORV where the distance between the superior edge of VSD and any great vessel annulus is greater than aortic valve diameter is labeled as non-committed VSD [11-13]. This VSD is usually in contact with tricuspid valve and located below the posterior limb of trabeculoseptomarginalis (TSM). VSD is restrictive in approximately 2/3rd of cases and has natural tendency to close with time [10,14].

The great vessels relationship varies from aorta right and posterior to pulmonary artery (PA) which is normal relation to D- malposed, L-Malposed, anteroposterior, and side-by-side relation. Fibro-aortic discontinuity and 2 well developed infundibulum are constantly present. The conal septum stands entirely in RV and it can be shifted towards posterior limb of TSM during development causing subaortic obstruction. This subset may present with and without PS. PS is present in nearly 1/3rd cases and results from shifting of conal septum towards anterior limb of TSM. Coronary arrangement is “Inverted Coronary anatomy”.

Non- committed VSD is divided into 2 subgroups –

A. Truly non-committed VSD [15]: Here, VSD is situated in inlet septum, apical septum, or as a part of atrioventricular septal defect or conoventricular (perimembranous) VSD distant to great arteries because of length of subarterial conus.
B. Indirectly non-committed VSD [13]: VSD is not directly non-committed type and can opens into outlet portion of RV but because of the presence of extensive ventriculoinfundibular fold (VIF), long outlet septum, extreme dextro position of aorta, or aberrant chordae tendinae in outlet of RV disturbs the direct relationship of VSD to semilunar orifice. Stellin and Coworkers extended the meaning to include structures obstructing the space between VSD and orifice of great vessels, for example – leaflets of straddling valve.

Embryogenesis of DORV [16]

DORV is type of conotruncal malformation and results from halts in development of normal conotruncus in various stages. Normally, two conus, namely subaortic and subpulmonary conus are present beneath the respective great arteries during development of human heart. At this stage the entire truncus overlies the bulbar cavity which is connected to primitive ventricle through bulboventricular foramen. With the gradual absorption of subaortic conus, the aorta attains position over the LV,right and posterior to PA along with aortomitral continuity. The

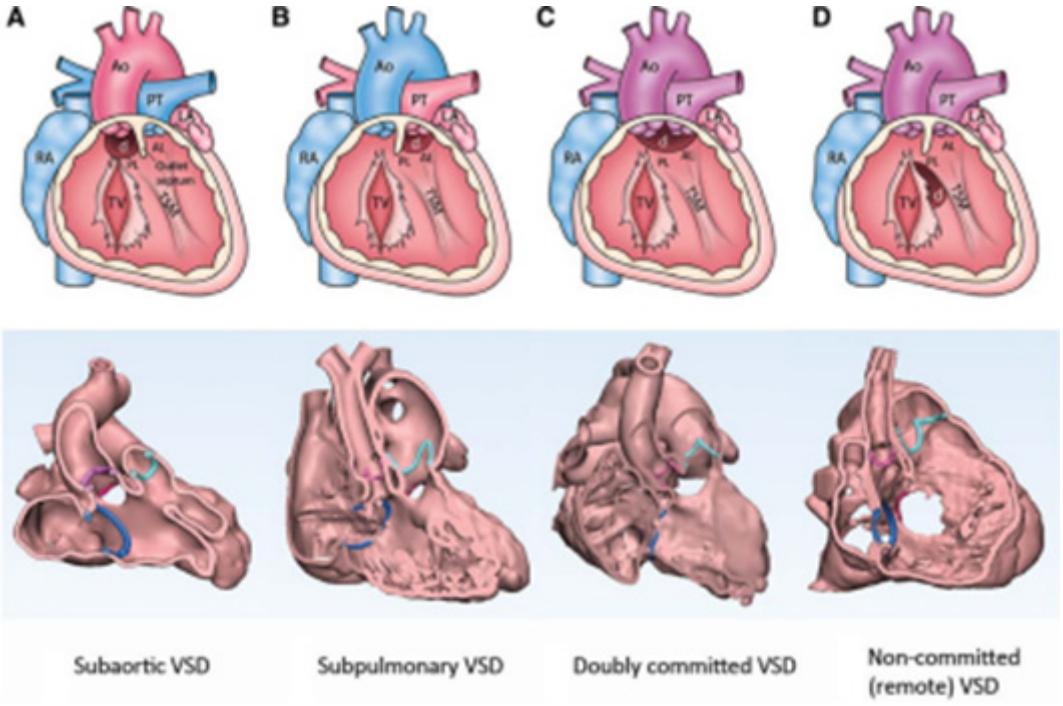

Fig 1: Lev et al's[9] classification of ventricular septal defects (VSDs) in double outlet right ventricle. The subaortic (A), subpulmonary (B), and doubly committed (C) VSDs classically involve the outlet part of the septum and therefore cradled between the anterior and posterior limbs of the trabecula septomarginalis (TSM). The classic noncommitted or remote VSD (D) involves the inlet part of the ventricular sep tum behind and below the posterior limb of the TSM. AL indicates anterior limb; Ao, aorta; d, ventricular septal defect; LA, left atrium; PL, posterior limb; PT, pulmonary trunk; RA, right atrium; and TV, tricuspid valve (reference no. 29)

subpulmonary conus persists causing discontinuity between tricuspid valve and pulmonary valve.

Anderson and his colleague studied TOF, DORV, and TGA with VSD and concluded that development of various spectrum of DORV is explained by departure from the normal development of portions of bulboventricular loop. Conal malrotation, changes in the position of anterior portion of muscular interventricular septum, and differential conal malabsorption form the basis of their hypothesis. Counter clockwise rotation of conal septum along with varying degree of nonabsorption of subaortic conus result into varying degree of aortomitral discontinuity and overriding of aorta into RV. The degree of conal septum rotation and site of insertion of the septal part of conal septum determines the specific types of DORV. Persistence of both conuses allow the aorta to continue with the RV with various degree of overriding of both ventricular outflow tract and persistence of bulboventricular foramen serves as the outlet for left ventricle.

So, in DORV non-committed VSD, persistence of both conus separates the bulboventricular foramen away from both the semilunar valve and its flow is directed to right ventricle than to any of the great artery. Deviation of conal septum towards subpulmonary or subaortic region results in stenosis of respective outflow tracts.

Echocardiography

Apart from establishing the diagnosis of DORV, the important points in DORV non-committed VSD to be checked on echocardiography to guide type of surgical

technique to be used are as follows –

1. Segmental situs
2. Atrioventricular concordance
3. Origin of both great arteries from morphologic RV along with their degree of overriding of both outflow tracts.
4. Relationship of both semilunar valves to each other
5. Adequacy of both ventricles.
6. VSD – size, location, its relationship to the origin of aorta and PA and tricuspid valve. Distance between superior margin of VSD and orifice of both semilunar valve. Presence of additional VSD.
7. Distance between tricuspid and pulmonary valve.
8. Outflow obstruction in right and left ventricle.
9. Anatomy and size of tricuspid and mitral valve
10. Systemic and pulmonary venous connection anomalies.
11. Coronary artery anatomy and relation to right ventricular outflow tract
12. To see the feasibility of intraventricular tunnel repair –
    A. Can blood from LV be directed to any great vessel in an unobstructed way
    B. Is Tricuspid to pulmonary valve distance adequate (it should be equal to or more than aortic orifice diameter)
    C. Is resection of infundibular septum required
    D. Is enlargement of VSD required
    E. Are there tricuspid valve attatchments to the infundibular septum or straddling which will require to be tackled during tunnel formation.
13. Associations –
    ASD
    PDA
    Coarctation of aorta
    Interrupted aortic arch
    Atrioventricular septal defect
    Heterotaxy/isomerism
    Straddling and overriding, stenosis or atresia of atrioventricular valves
    Pulmonary hypertension

Surgical Management

An adequate space is required in right ventricle to accommodate intraventricular tunnel without causing obstruction to right ventricular outflow. Therefore, definite procedure is deferred for the age after 3 months and when weight of the baby is approximately 6 - 8 kg.

Pulmonary overcirculation may require PA Banding or decreased pulmonary blood flow may require BT shunt formation to buy some time for definitive repair.

Goals of surgery of two ventricular repair -

1. To separate systemic and pulmonary circulation
2. To establish an unobstructed LV to aortic continuity
3. To establish adequate RV to PA continuity
4. To repair associated lesions

Indications for univentricular repair –

1. Associated Apical VSD or more than 2 multiple VSD
2. Heterotaxy
3. Underdevelopment/hypoplasia of LV/ RV
4. Hypoplasia or atresia of tricuspid or mitral valve
5. Straddling of Atrioventricular valve (type C)

Surgical Techniques

A. To route DORV non-committed VSD to aorta –
   1. Long single patch intraventricular tunnel repair [5,6,17,18]
   2. Multiple patch intraventricular tunnel repair [19]
   3. Tubular graft intraventricular tunnel repair [20,21]
   4. Kawashima repair [22]
   5. Aortic root translocation (Bex Nikaidoh and Modified Nikaidoh procedure)

B. To route DORV non-committed VSD to Pulmonary artery –
   1. Intraventricular tunnel to PA with atrial switch operation [23]
   2. Intraventricular tunnel to PA with arterial switch operation [8]
   3. Damus-Kaye-Stansel procedure [24]
   4. Pulmonary root translocation [25]

C. DORV non-committed VSD with PS –
   1. Tunnel from LV to aorta with RVOT patch repair/ REV/Rastelli/ RV to PA conduit
   2. Tunnel from LV to aorta with Nikaidoh procedure/modified Nikaidoh operation
   3. Tunnel from LV to PA with arterial switch operation [8]
   4. Tunnel from LV to PA with pulmonary root translocation [25]
   5. Tunnel from LV to PA with double root translocation [26]
   6. Tunnel from LV to aorta with Half Turn Truncal Switch procedure [27]

D. LV apex and descending thoracic aorta valved conduit with VSD closure and RVOT reconstruction (McGoon technique) [28].

Technical points to be considered when routing the VSD to aorta –

- Aggressive resection of subaortic conus
- Selectively enlarging the VSD anteriorly and superiorly, reducing the angle of intracardiac tunnel
- Tricuspid chordae reimplantation to the baffle
- Tricuspid anteroseptal commissure reattachment to the baffle
- Tricuspid anteroseptal commissure folding

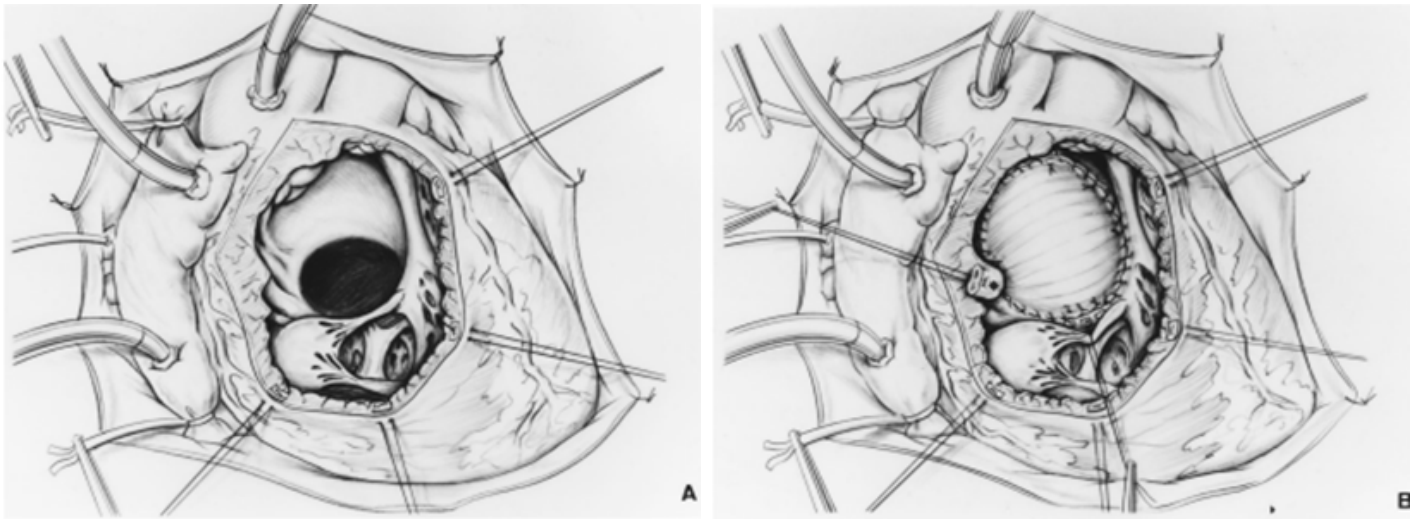

Fig 2: Intraoperative transventricular view of a heart having DORV with non-committed conoventricular VSD. (A) The VSD is considerably distant from both arterial valves. (B*) baffle construction almost always requires parietal band section. The insertion of the baf¯e patch includes both interrupted pledget supported and continuous prolene sutures (reference no. 17)

Intraventricular Baffle Repair

Tunnel repair technique with a patch placed in a way that directing the LV blood through the VSD to aorta (figure 2).
If there is Pulmonary stenosis component is present then infundibular resection, pulmonary valvotomy, infundibular patch repair, and transannular patch placement is done as per the requirement of situation.

Multiple Patch Technique

In this technique, the distance between tricuspid and pulmonary valve is not considered as contraindication for intraventricular routing.

VSD is tunneled to aorta. This repair is suitable for side by side, d – malposed, l-malposed , and anteroposterior great artery relationship.

VSD is enlarged in all cases even if it is nonrestrictive. 3 patches of bovine pericardium are used for tunnel formation (figure 3).

First patch – it is semilunar shaped patch, secured to the margin of VSD with

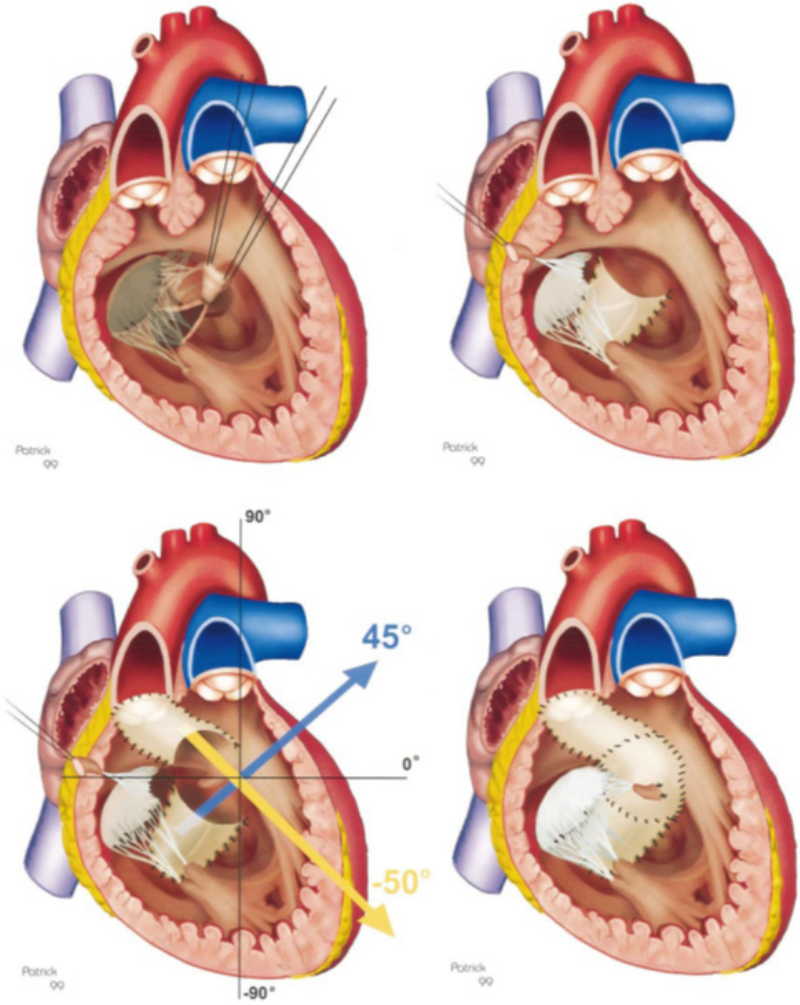

Fig 3: Reference no. 19

interrupted mattress sutures with pledgets. Long axis of this patch form 45-degree angle to an imaginary line drawn through the center of aortic annulus. As the tricuspid valve mainly septal cusp overhung the defect and therefore was interposed between VSD and aortic orifice, its papillary muscle was detached from the septum and secured with interrupted stitches for later reinsertion at the tunnel patches. If it is not feasible to reinsert the papillary muscle, then their resection and that of a portion of the corresponding septal cusp was performed followed by bicuspidization of the tricuspid valve.

Second patch - Through the transverse or oblique right ventriculotomy excise the obstructive subaortic conus and often the portion of infundibular septum. Second patch is more distal patch and roughly oval in shape.it is secured around the aortic orifice and subaortic conus with long axis of patch perpendicular to the long axis of the first patch.

Third patch – it is the intermediate patch and trapezoid in configuration. It is sutured to the septum, septal portion of tricuspid valve, and both patches.

PS , if present, is relieved by pulmonary valvotomy or infundibular resection with or without transannular bovine pericardium or valved extracardiac conduit.

Tubular Graft Intraventricular Tunnel Repair

This technique is useful for DORV non-committed VSD with or without PS and with d-malposed and l-malposed great artery relationship. Patient should be more than 2 years.

Operative steps (figure 4)

It is done through right atriotomy and right ventriculotomy.

Measure the shortest and longest distance between VSD and aortic annulus. Resect the hypertrophic muscular bands in the free wall of bilateral conus. VSD is enlarged leftward, anteriorly, and superiorly through trabeculoseptomarginals. VSD diameter should be 3mm larger than aortic annulus in cardioplegic heart. A PTFE conduit (figure 5) of 16 mm size (< 5 year age) or 19 mm (>5 year age) is prepared by trimming the both ends with appropriate inclines so that shortest length of conduit is made equal to the distance between superior edge of VSD and

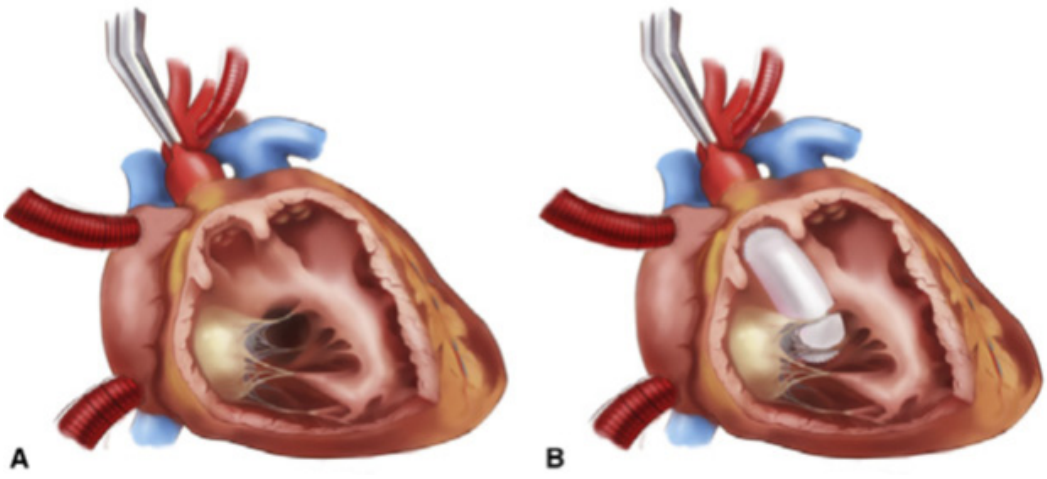

Fig 4: The intraventricular conduit is constructed using a vascular graft to connect the VSD to the aorta (reference no. 21)

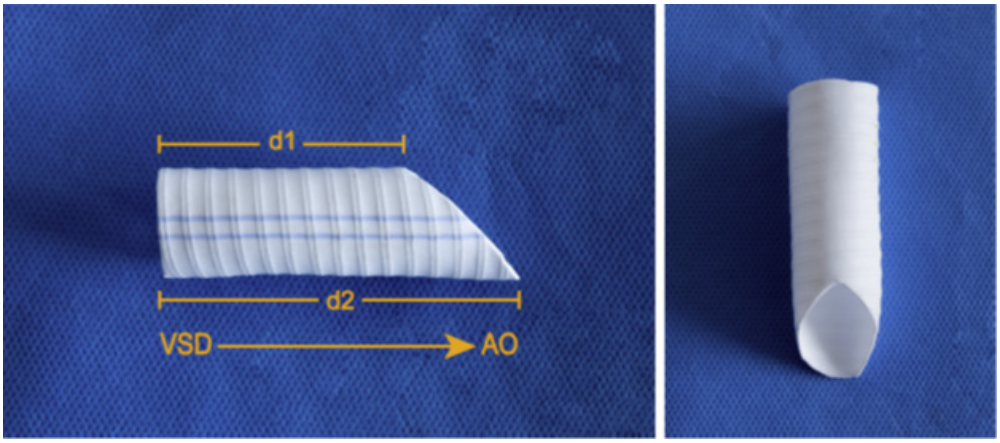

Fig 5. Schematic diagram of intraventricular conduit. The shortest length is equal to the distance between the superior edge of the VSD and the inferior edge of the aortic annulus (d1). The longest length of the conduit is equal to the distance between the inferior edge of the VSD and the superior edge of the aortic annulus (d2). VSD, Ventricular septal defect; AO, aorta (reference no. 21)

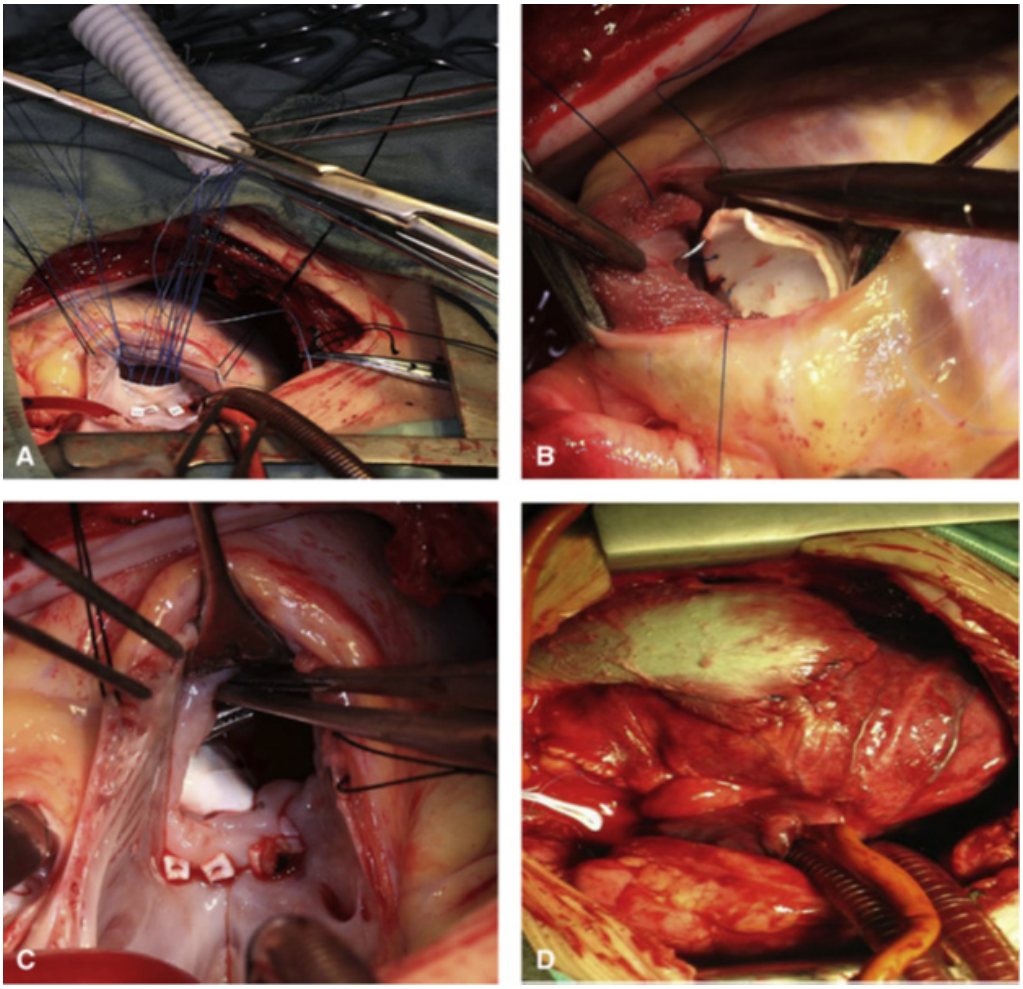

Fig 6: A, Polytetrafluoroethylene (Gore-Tex; WL Gore & Associates, Flagstaff, Ariz) vascular prosthesis was used for the patients. The defect end was anastomosed with interrupted suture. B, the aortic end was anastomosed with continuous suture. C, After the establishment of the conduit, the tricuspid valves and chordae were carefully checked. D, Valved conduit (bovine jugular vein conduit) was used to reconstruct RVOT for pulmonary atresia (reference no. 21)

inferior edge of aortic annulus and largest length to the distance between inferior edge of VSD and superior edge of aortic annulus. Conduit is anastomosed to VSD through the tricuspid valve in interrupted manner and to the aortic annulus through the right ventriculotomy in continuous suture manner (figure 6).

For d-malposed aorta position, conduit is placed below the conal papillary muscle and for the l –malposed aorta position the conduit is placed over the conal papillary muscle.

RVOT obstruction is treated with infundibular or transannular patch or RV to PA conduit in case of severe PS or pulmonary atresia.

For complete atrioventricular septal defect patients, atrioventricular valve plasty is done and ostium priumum ASD is repaired with the patch.

Advantages

- Simple technique
- It avoids complicated manipulation between conal papillary muscle and chordate of tricuspid valve, therefore, prevents impairing of tricuspid and pulmonary valves.
- It is suitable for patients with PS
- It does not require translocation of coronary arteries
- It may protect LV function by reducing the intraventricular septal injury and avoiding the significant akinetic area and flow turbulence in LVOT as seen in baffle tunnel repair.

DAMUS-KAYE-STANSEL Procedure [24]

When DORV non-committed VSD is associated with significant subaortic stenosis then VSD is routed to pulmonary artery, division of main Pulmonary artery, connection of aorta to proximal puylmonary artery, and restoring the RV to PA continuity with placement of valved extracardiac conduit between distal pulmonary artery and RVOT.

Aortic Root Translocation (BEX – NIKAIDOH Operation)

Indications

- Remote VSD
- Presence of abnormal chordae of the mitral valve which are attached onto the conal septum, some of them can be sacrificed which are not essential while preserving the essential chordate
- Major abnormal insertion of the tricuspid valve opposite onto the conal septum
- Anteroposterior relationship of the great arteries
- For side-by-side relationship, the coronary button harvesting and reimplantation is required.

Distance between the aorta and PA should be equal or more than 5 mm.

Contraindications

- Complex coronary artery anatomy, major coronary crossing RVOT precluding the harvesting of aortic root from RV.
- Presence of posterior looping coronary artery which is at risk during suturing the aortic root to the pulmonary annulus.

Operative steps

Aortic root is harvested from the RV after extensive mobilization of coronaries.

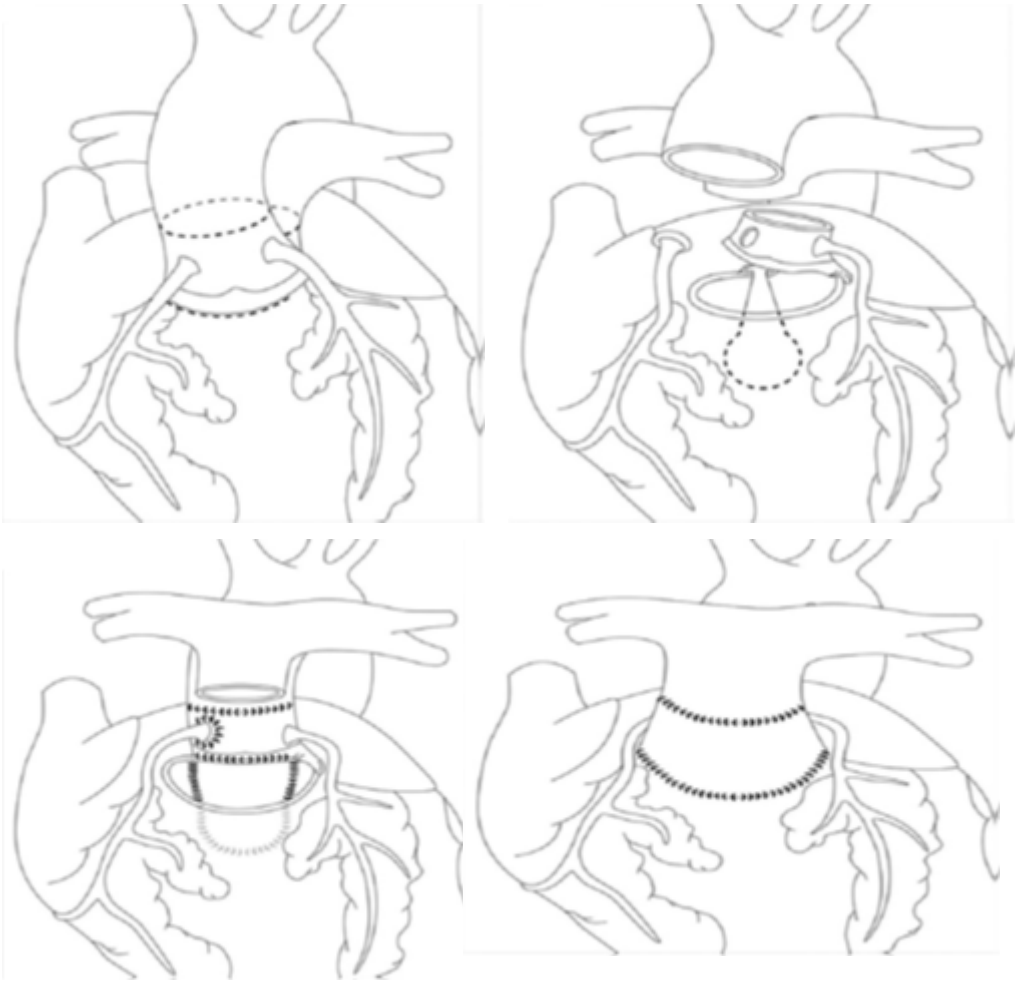

Fig 7: Operative procedure. (A) Transverse incision of the aorta below the aortic valve and aortic root harvest. (B) Aortic root pivot rotation and the Lecompte maneuver. (C) Left ventricular outflow tract reconstruction with VSD extension and patch closure. (D) MPA reconstruction with a pulmonary homograft. VSD, ventricular septal defect; MPA, main pulmonary trunk (reference no. 30)

The main PA is transected near its annulus. The aortic root is translocated into the pulmonary annulus. The conal septum is divided and VSD is closed with a patch routing the LV to aorta. RVOT is reconstructed by external RV to PA conduit or by adding LeCompte maneuver along with RV to PA patch reconstruction.

Modified NIKAIDOH Procedure

This operation is suitable for DORV non-committed VSD with pulmonary atresia.

Operative steps (figure 7)

After oblique right atriotomy and detailed analysis of the anatomy, the aorta is transected above the sinotubular junction and pulmonary artery is transected just above the annulus. The right coronary button is harvested with mobilization of its proximal part. Right ventriculotomy is done through the transverse incision below the aortic annulus and extended circumferentially around the aortic valve except immediately below the left coronary artery maintaining a muscle rim of 3-5mm. As such, the left coronary artery is left intact. Interventricular septum and the pulmonary annulus are transected just below the pulmonary valve annulus to

extend the VSD and to create the space for aortic root. Now, rotate the aortic root clockwise and anastomosed to the rim of pulmonary annulus and transected edge of infundibular septum (Pivot Rotation). The new plane of VSD (from the crest of original VSD to the free edge of aortic root circumference) is closed with bovine pericardium. After LeCompte maneuver, distal aorta is anasomosed to the proximal aorta. Right coronary button is implanted on aorta. RVOT reconstruction is done with pulmonary homograft conduit.

Pulmonary Root Translocation [25]

Indications

- DORV – VSD pulmonary RVOT obstruction and malposed great arteries (VSD committed to PA)
- DORV subpulmonary VSD with malposed great arteries with complex coronary anatomy, not suitable for arterial switch operation
- DORV-AVSD with malposed great arteries and pulmonary stenosis

Advantages

- It keeps the aorta untouched with its original anterior position without coronary artery manipulation.
- LeCompte maneuver is not required therefore pulmonary trunk is not lying close to the sternum hence no risk of bleeding during a reintervention
- Partial resection of conal septum favors construction of an obstruction free LVOT. Therefore, low incidence of LVOT obstruction which is an important problem of Rastelli operation.

Double Root Translocation [26]

It aims at retrieving the normal geometry of the left and right ventricular outflow tract and maximally preserving the function and growth potential of aortic and pulmonary valves. It can be done in presence of atrioventricular discordance. Presence of bilateral conus, pressure gradient of more than 35 mmHg across the RVOT, and diameter of pulmonary annulus at least one third of aortic valve, and distance between pulmonary and mitral valve of more than 10mm are important requirement for double root tranlocation.

Indications

- TGA VSD PS with subpulmonary VSD or VSD distant from aorta with anteroposterior relationship of the great arteries.
- DORV non-committed VSD with PS

Contraindications

- Excessive hypoplasia of the pulmonary valve.
- Complex coronary artery pattern e.g. origin of left main coronary or only the left anterior descending artery or left circumflex coronary artery from the sinus 2.
- Severe associations e.g. AVSD, Atrioventricular valve straddling, Swiss Cheese VSD.

Abbreviations

DORV	-	double outlet right ventricle
VSD	-	ventricular septal defect
TOF	-	tetrology of Fallot

TGA	-	transposition of great arteries
PS	-	pulmonary stenosis
LV	-	left ventricle
RV	-	right ventricle
TSM	-	trabeculoseptomarginalis
PA	-	pulmonary artery
ASD	-	atrial septal defect
PDA	-	patent ductus arteriosus
LVOT	-	left ventricular outflow tract
RVOT	-	right ventricular outflow tract
BT Shunt	-	Blalock Taussig Shunt

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