INTRODUCTION
Since the advent of surgery for tetralogy of Fallot (TOF) nearly seven decades ago,[1] primary repair of TOF has been one of the most commonly performed procedures with acceptable early and late results. A successful repair entails effective relief of right ventricular outflow tract (RVOT) obstruction. This is achieved in most cases by a right ventriculotomy followed by patch augmentation of the RVOT. Taking into consideration various angiographic, surgical, and autopsy series of patients with TOF, anomalous coronary arteries (ACA) have been reported up to 14%.[2] Repair of TOF with ACAs has been fraught with the lurking dangers of myocardial ischemia and sudden cardiac death.[3] The main reasons behind intraoperative nonvisibility of ACAs, is due to myocardial bridging,[4] overlying epicardial fat,[5,6] or by pericardial adhesions due to previous palliative surgery.[7,8] Notwithstanding these, an effective knowledge of the possible coronary patterns and all the possible surgical options are essential for optimal surgical outcomes following repair of TOF. The presence of a significant coronary artery or its branch crossing the RVOT necessitates a change in the surgical strategy. This article is an attempt to review this surgical challenge [Figure 1].

EMBRYOLOGY
The development of coronary arteries during embryogenesis is a complex interplay of several factors and is a less explored field of cardiac embryology. Variation in the origin, number, and pattern of the major coronary arteries is far greater than the variations seen in valves, myocardium, and/or great vessels. Initially, the developing heart is covered by the rapid migration of simple epithelial cells originating from the proepicardial organ (PEO) that completely envelopes the heart.[9] Few of these epicardial cells lose contact with
the surface epithelium and develop into freely migratory mesenchyme and move into the developing subepicardial connective tissue space. This is an example of the epithelial-to-mesenchymal transition. This mesenchyme then migrates through the entire myocardium and gives rise to the coronary vasculature.[10] These initial phases of coronary vasculogenesis occur in the absence of blood flow, and the caliber of proximal and distal arteries is governed by forces other than blood flow. It is noteworthy that the entire coronary vasculature develops without blood flow within them [Figure 2].

During the latter half of embryogenesis, the developing coronary arterial system establishes connection with the ascending aorta after aortopulmonary rotation. There was an initial controversy of whether the coronary arteries grow “from” or “into” the ascending aorta. However, it is now accepted that the coronary arteries grow and penetrate into the aorta through local apoptotic mechanisms.[11,12] The entry of coronary arteries into the aorta occurs after the aortopulmonary rotation, and therefore, when the aortic position is final, the coronary arteries enter the closest aortic sinus.[13,14] The aortic annulus in TOF rotates in a clockwise manner during embryologic septation of the great arteries, which relocates the left coronary artery more posteriorly and the right coronary artery (RCA) more anteriorly. This influence of aortopulmonary rotation on anomalous coronary patterns in TOF has been evaluated using angiographic studies.[15]

ANOMALOUS CORONARY ARTERIES IN TETRALOGY OF FALLOT

Almost 14% of patients with TOF have coronary anomalies, and among them, 2%–9% have an ACA crossing the RVOT.[2,16] This factor greatly influences the surgical approach to the lesion. These include a large conal artery, left anterior descending artery (LAD) arising from the right sinus-of-Valsalva, LAD arising from the proximal RCA, or dual LAD [Figure 3]. In an autopsy study of >100 hearts with TOF, Meng et al.[17] further subclassified the latter into type 1 (coronary artery crossing proximal to the pulmonary annulus) and type 2 (coronary artery crossing at some distance from the pulmonary annulus). Dual LAD coronary arteries can be found in 1.8% of patients—whereby not all accessory LAD from RCA would actually cross the RVOT to reach the anterior interventricular groove, but all LAD from the right sinus of Valsalva would do so.[18] Distinguishing between a large conal (cristal artery of Spitzer-described by Meng et al.) branch of the RCA (6.4% of patients) versus dual LADs is decided by whether the branch reaches the anterior interventricular groove or not. This means that a large conal artery from RCA can be considered a normal variant if it is not reaching the anterior interventricular groove. An abnormal coronary artery, the size of which is similar to or larger than RCA and whose course goes near the apex of the heart, is called as a large conal branch. Moreover, a large conal branch with a typical morphology of LAD reaching the apex is defined as an “accessory LAD.”

Additional coronary arterial anomalies include a single RCA (0.3% of patients) and a single left coronary artery (0.2% of patients), both of which can cross the RVOT. In another series,[17] a single left coronary artery was far more common than a single RCA. It was also shown that when the RCA arises from a single left coronary artery and passes anterior to the aorta, it was liable to cross the RVOT. In the same series, the variant of RCA from the left sinus of Valsalva was unlikely to
cross the RVOT as it would mostly pass behind the aorta, whereas RCA from LAD though less common was likely to cross the RVOT. There can be a right ventricular branch arising from the LAD and crossing the RVOT, narrow distance between LAD and RCA and coronary fistulas with connection to bronchial arteries. It is also noteworthy that in patients of TOF with abnormal coronary artery patterns, the left coronary artery is dominant in 28% of patients as demonstrated in a study by Baraona et al., which involved detailed literature search of patients with conotruncal anomalies. Coronary artery anomalies are more frequent in patients with more aortic overriding though the difference has not been proven to be statistically significant. The high take-off of a coronary artery may be present in TOF and can be associated with an intramural course at its origin. This may give rise to difficulties during aortic cross-clamping at operation, as well as produce problems during selective arteriography. Intramural course and acute angulation at the origin of a coronary artery have been implicated in sudden death during exercise. Furthermore, there may be atresia of the proximal aspects of coronary vessels in patients with TOF.

**PREOPERATIVE EVALUATION**

Echocardiographic delineation has found an increasing role just as in the evaluation of transposition of the great arteries and anomalous origin of the left coronary artery from the main pulmonary artery (ALCAPA). Initial reports from Santoro et al. and Saraçlar et al. have documented the efficacy of the preoperative coronary-echocardiogram in these patients. In protocols established by Need et al., the coronary arteries were interrogated mainly from the parasternal short- and long-axis views. From the standard parasternal short-axis view, the left main coronary artery origin is traced from the left sinus of Valsalva and clockwise rotation of the transducer with a slight apical angulation can clearly trace the course of LAD through the anterior interventricular groove along with the depiction of the origin of the left circumflex coronary. The RCA is depicted with high-up parasternal short-axis view for detailing the size and extent of the conal branch and to rule out any anomalous origin of the LAD from the RCA [Figure 4a and b]. From the parasternal long-axis view, the left main, LAD and circumflex coronaries are imaged between the aortic root and
the main pulmonary artery (MPA). From these views, the infundibular free wall shows coronary coursing its epicardial surface. Current practice is to have further imaging if the echocardiographic evaluation is equivocal.[26] Technological advances like the introduction of dual-source scanners have improved the capabilities of multi-slice computed tomography [Figure 4c and d]. The temporal resolution of dual-source multi-slice computed tomography (DSCT) is now 83 ms, allowing coronary artery evaluation in very young infants with high heart rates. The radiation dose can also be kept low when a prospective, sequential, electrocardiographically triggered acquisition protocol is obtained.

In many centers, it is still a common practice for all patients with TOF to undergo routine preoperative cardiac catheterization for delineation of coronary anatomy despite limitations, technical difficulties of selective coronary angiography and potential complications. In pure oblique projections, the aortogram often fails to depict the origins of the coronary arteries or anatomic relation to the RVOT.[27] In infants, the imaging requires modification of angling and view of normal aortogram itself to obtain the desired information on the coronary anatomy in TOF. Due to anterior angulation of the ascending aorta in TOF, getting an end-on view of the aorta is facilitated by a maximum caudal tilt with wedge support placed underneath the child and left oblique angulation of up to 30° which would facilitate bringing the RVOT anteriorly and to the left of the aortic valve and any vessel across this area would then be visible.[28]

The 2008 American College of Cardiology/American Hospital Association guidelines recommends that when TOF is associated with ACA, the anatomy of the coronary anomaly should be determined before any intervention on the RVOT is undertaken.[29]

MANAGING A PATIENT WITH TETRALOGY OF FALLOT AND ANOMALOUS CORONARIES

Systemic to pulmonary artery shunts

A systemic-to-pulmonary shunt for TOF remains a relatively less popular option as primary repair in infancy has been favored by most surgeons worldwide, though this issue is debatable.[30] The palliative shunt is a surgical option for a symptomatic patient who is below the age and/or weight cutoff for primary repair as an institutional/surgeon policy.[30]

Advantages

In patients with a coronary artery crossing RVOT, performing a shunt can be justified to “buy time” and delay placement of an RV-PA conduit to minimize the number and probability of re-operations for conduit failure.[31]

Right ventricular outflow tract reconstruction in tetralogy of Fallot with coronary anomaly

The first-ever mention of dealing with a cohort of TOF with ACA patients comes from the initial work by Senning[32] In this series, out of 27 patients, 4 patients had an anomalous LAD from RCA, and one patient had a single coronary artery. In the initial years of such on-table surprises, things were grim. Though coronaries were being transected (Kirklin et al.[4] Friedman et al.,[6] Gadboys et al.[33]), a growing concern among surgeons developed, which gradually led to the conclusion that a treacherous coronary vessel sitting over the precious infundibular area is a murky substrate for unsuspecting mayhem. The fact that there definitely was a transition point of dawning of this realization comes from the evidence in a series by Berry and McGoon from Mayo Clinic, Rochester[37] of eight deaths (30%) of the 27 TOF patients with ACA. The mortality rate was 100% before 1961 and 17% thereafter, and most deaths were due to myocardial infarction caused by injury to the ACA. In the same series, relief of pulmonary stenosis was performed through a ventriculotomy caudal to the anomalous artery or, if necessary, caudal and cephalad to the artery. If the stenosis was still unrelieved or if the ratio of right ventricular-to-left ventricular peak pressure was >0.9, placing a conduit from the caudal ventriculotomy to the MPA followed. A similar strategy of careful placing of ventricular incision and relief of pulmonary stenosis followed by conduit placement, if needed, was common practice in several surgical series till 1970s.[8]

The first-ever comparative study came from the Mayo Clinic in 1987, when Humes et al.[34] reported details of management of 20 ACA in TOF out of 416 patients who underwent complete repair of TOF between 1973 and 1984. Twelve of these patients had RVOT patch reconstruction, and eight had placement of a conduit from the right ventricle (RV) to the pulmonary artery (PA). The outcome of this study was almost equivocal in terms of superiority of either approach, most probably because of the small sample size and follow-up. However, it was concluded that appropriateness of specific procedure depends on the exact location and degree of tortuosity of the anomalous artery and the level and severity of RVOT obstruction.

Trans-atrial approach with or without pulmonary arteriotomy

The trans-atrial approach was first described by Hudspeth et al.[35] in 1963, who described its successful use in 10 consecutive patients. Though he described its usefulness in avoiding a ventriculotomy, its utility in cases of coronary crossing RVOT was also recognized.

Technique

The approach consists of the closure of the ventricular septal defect and maximum possible relief of RVOT.
obstruction through the right atrium. A pulmonary arteriotomy may be added to either inspect the pulmonary valve, to complete an inadequately performed pulmonary valvotomy (as judged by an inability to pass an adequately sized Hegar’s dilator) or to slightly extend the incision onto the right ventricular free wall. Only that much of the RV wall is incised as it is necessary to relieve obstruction. Subsequently, this approach was successfully used and reported by several authors.

**Outcomes**

The largest series was reported by Karl et al.\[^{36}\] from Melbourne in 1992 who described this approach in 366 patients with 0.5% mortality and actuarial freedom from reoperation for any reason of 95% at 5-year and 10-year follow-up.

In a series of 611 consecutive patients with TOF, Brizard et al.\[^{37}\] from the same institute reported 36 patients of TOF with ACA. The infundibular obstruction was addressed from the right atrium and the pulmonary valve annulus. The pulmonary valve and annulus were incised whenever felt to be of inadequate diameter. When the anomalous vessel would complicate infundibular stenosis with the threat of coronary injury, the incision was made parallel to the vessel and toward the interventricular septum in case of LAD from RCA or toward the right atroventricular groove in case of an anomalous RCA from the LAD or the LCA [Figure 5a and b]. Thirty-four patients out of 36 had a trans-atrial-transpulmonary repair with avoidance of a conduit; 25 of these needed a limited transannular patch. They also described a slight modification of the approach by extending the right ventriculotomy (whenever necessary) parallel to the anomalous coronary, which avoided the need of a conduit in all but 2 patients.\[^{37}\] The mean RVOT gradient was 19 mm Hg at follow-up, similar to that seen in patients with normal coronary arteries (15 mm Hg in their own series).

In a case series by Kalra et al.,\[^{38}\] a routine trans atrial approach was used and if required, transpulmonary approach was used where the pulmonary annulus needed to be addressed. One concern that they raised was that under-resection of the hypertrophied anterior wall of the RV to prevent “button holing” of the anterior wall of the RV and injury to the ACA was associated with higher gradients across the RVOT. The reason for this concern was perhaps due to their finding that all their patients except two had an RVOT gradient ≤ 30 mm Hg at late evaluation.

**Van Son technique**

The first alternative technique of RVOT reconstruction in patients with TOF and ACA devised by van Son\[^{39}\] in San Francisco drew its inspiration from the work of Barbero-Marcial et al.\[^{40}\] in truncus arteriosus repaired without the use of an extracardiac conduit.

**Technique**

A longitudinal incision was made in the RVOT extending distal to the LAD and bringing out an inverted u-shaped PA flap backward all the way over the anomalous coronary (a second bridge over the artery) to reach the edge of the ventriculotomy. This was then followed by a hood of oval-shaped glutaraldehyde-treated pericardial patch or pulmonary homograft patch that was sutured circumferentially sutured to the edge of the ventriculotomy, both edges of the PA flap, and the edge of the pulmonary arteriotomy [Figure 6a and b].

**Advantage**

The main advantage was the growth potential of the surgically created outflow from a flap of viable autologous vascular tissue.

**Outcomes**

Like transannular patches, this technique has inevitable pulmonary insufficiency. However, during follow-up ranging from 3 to 7 years, Dandolu et al.\[^{41}\] found that the only reintervention required was for muscular obstruction proximal to the right ventricular incision.

**Double outflow technique**

Dandolu et al.\[^{41}\] described an interesting approach called the “double outflow technique” in patients of TOF with anomalous coronaries.

**Technique**

In this technique, the native RVOT flow is untouched. A ventriculotomy is placed below the anomalous coronary through which infundibular resection is performed. A flap of the anterior PA wall is turned down and sutured through which infundibular resection is performed. A ventriculotomy is placed below the anomalous coronary through which infundibular resection is performed. A flap of the anterior PA wall is turned down and sutured through which infundibular resection is performed.
autologous vascular tissue. The authors described the procedure in 4 patients out of which 1 patient required reintervention for recurrent RVOT obstruction. In another unique two separate patch' technique, one patch for the pulmonary annulus and the other patch for the infundibular portion of the RVOT below the coronary artery was placed strategically in the available space with good results. A simplified modification of the double-barrel technique was suggested by Shivaprakasha.

**Flaps**

**Technique**

Bonchek described an interesting technique in 1976, where they mobilized an anomalously crossing LAD and patched the RVOT with a Teflon patch in an 11-year-old girl. This technique can be used if the aberrant coronary artery is unusually tortuous and without RVOT branches. This allows its mobilization along with surrounding epicardium and underlying myocardium and the placement of a slightly undersized transannular patch to obviate any stretching and compression of the coronary artery.

**Limitations**

This technique, therefore, is not practical in all situations but remains one of the possible surgical options.

**Innovations**

**Translocation methods**

Tchervenkov et al. translocated the entire small MPA without the addition of a Lecompte maneuver. This keeps the right PA behind an enlarged and anteriorly displaced aorta. Hence, this technique was particularly useful in patients with an aberrant coronary artery near the pulmonary valve annulus. The MPA is divided just distal to the annulus with the proximal end being oversewn. The distal end is then translocated onto the right ventriculotomy site keeping the aberrant coronary artery behind, thus reconstituting the design of the artery above a displaced bridge.

**Limitations**

There are concerns regarding the possibility of right PA compression by the ascending aorta in a non-Lecompte design.

**O’Blenes method**

In a unique case of anomalous origin of LAD from RCA with the left pulmonary artery (LPA) arising from the ascending aorta in TOF with absent pulmonary valve cusps, the primary repair was done with the use of Lecompte maneuver by O’Blenes et al.

**Technique**

After separating LPA from ascending aorta, the MPA is transected and brought anterior to the aorta by the Lecompte maneuver. This is followed by the final connection to a carefully placed right ventriculotomy incision, just proximal to the anomalous LAD, thus avoiding the use of any conduit.

**Sub coronary suturing**

**Technique**

Tchervenkov et al. described a unique technique of sub coronary suturing where a continuous U-shaped suture line starts from the pericardial patch, going under the coronary artery, and coming out all the way lateral to it into the epicardium. Then the suture is brought back from epicardium to endocardium under the ACA and coming through the pericardial patch, thus maintaining the coronary artery on an island of myocardium.

**Advantages**

This technique is especially useful if aberrant coronary artery travels not only below the pulmonary annulus but also is close to LAD, leaving very little space, in between, for placing a patch.
Managing Conal arteries

Surgical management of TOF with ACA is further made difficult by a well-developed conal artery, which is associated with a small but normally positioned LAD. These conal arteries cross anomalously over the RVOT. In such a case reported by Asano et al.,\textsuperscript{[16]} with a small pulmonary annulus, the conal artery was transected intraoperatively to generate a favorable postoperative right ventricular pressure to left ventricular pressure ratio but only to land up with extracorporeal membrane oxygenation to rescue the patient from critical left ventricular failure.

Conduits

The use of RV-PA conduit is a possible surgical option. The origin of the conduit is from a ventriculotomy placed below the ACA. However, with the growth of the patient, conduit-patient mismatch and conduit replacement is inevitable. With the availability of a variety of techniques described above, in the current era, the conduit should be rarely needed.\textsuperscript{[47]}

INJURY TO CORONARY ARTERIES

Instances have been reported where an anomalously crossing coronary artery was accidentally divided while performing a ventriculotomy. This situation is not unlikely in patients where such a coronary is intramyocardial. Perfusion to the distal part of a divided coronary has been successfully achieved using either a left internal mammary artery to coronary anastomosis\textsuperscript{[48]} or using a saphenous vein graft.\textsuperscript{[49]} Cooley et al.\textsuperscript{[48]} reported successful internal thoracic artery grafting to an anomalous LAD in a 16-month-old patient. Postinjury, after ligation of the proximal end of the divided anomalous LAD, its distal cut end was probed to confirm that the artery was the blood supply to the anterolateral surface of the left ventricle.
and ventricular septum. The left internal mammary artery, which was 1.0 mm in diameter, was used for an end-to-side anastomosis to the coronary artery using 7-0 monofilament polypropylene suture with good result. In a patient reported by Shaffer et al.,[49] after a ventriculotomy incision was performed in the hypoplastic infundibulum, the cut ends of a large coronary artery were visible in the myocardial wall. After probing, the artery was identified as an aberrant LAD from the RCA which might have been intramycocardial. Blood flow to the coronary artery was however restored using a saphenous vein graft that was anastomosed end-to-side to the coronary artery.

In another case report by di Carlo et al.,[50] inadvertent transection of an anomalous LAD coming from the RCA was managed by an aorta-coronary bypass graft with a 4 mm polytetrafluoroethylene conduit. Early re-catheterization proved patency of the polytetrafluoroethylene graft. However, a myocardial scintiscan performed 3 months after the operation demonstrated reduced perfusion of the areas supplied by the left coronary system.

In a case report by Bhutani et al.,[51] during the extension of the pulmonary arteriography across the annulus to the infundibulum to repair the hypoplastic annulus, the LAD was divided accidentally. The left IMA was grafted to the LAD distal to the site of transection. Later digital subtraction angiogram in right anterior oblique view demonstrated patent left IMA and patent right ventricular outflow patch with distal pulmonary arteries in RV angiogram in the same sitting.

In a case report by Luo et al.,[52] in which the patient with TOF had an anomalous origin of the RCA from the left coronary, the transected proximal RCA was mobilized and was reimplanted into a 4 mm opening into the ascending aorta. The authors suggested that using this technique, conduits can be avoided, and at the same time, the growth potential of the translocated native coronary artery can be maintained.

**CONCLUSION**

A firm preoperative diagnosis of ACA in TOF guides the surgeon to optimal techniques to manage these patients. From a plethora of available techniques, the one that works best in a particular situation needs to be chosen depending on the surgeon preference. Repair using a conduit should be the last resort.

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There are no conflicts of interest.

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