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Outcomes of Surgical and Transcatheter Closure of Congenital Coronary Artery Fistulas: Results from a National Audit Database

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Abstract

Congenital coronary arterial venous fistulae are very rare anomalies. We analyzed data from a national procedure database and identified 79 (0.09%) catheter and surgical closures of coronary fistulae out of total of 88061 procedures performed over a 12 year period. Thirty four patients (43%) underwent surgical closure while 45 (57%) patients had catheter closure. Sixty six patients (83.5%) had complete follow-up. There were no early deaths, and mortality at 10 years was 4.8%. Freedom from fistula reintervention was 91.2% for the trans catheter group *vs*. 100% in the surgical group (p=0.11). We found no differences in outcomes between patients with isolated fistulas and those associated with other congenital defects. Surgical or catheter treatment of coronary fistulae is attainable with no early mortality and good medium term outcomes. However, reintervention rates appear to be higher in trans catheter patients.

Keywords: Coronary arteries abnormalities; Arteriovenous fistulas; Cardiac surgical procedures; Catheter ablation

Abbreviations

CAF: Congenital Coronary Artery Fistulas; NICOR: National Institute for Cardiovascular Outcomes Research; SD: Standard Deviation

Introduction

Congenital Coronary Artery Fistulas (CAF) is very rare termination anomalies. The reported incidence in patients undergoing cardiac catheterization ranges from 0.13-0.6% [1]. Without treatment, patients may develop heart failure secondary to a left to right shunt, myocardial ischemiaby "coronary steal", bacterial endocarditis due to turbulent flow and, rarely, aneurysmal rupture [2]. Current literature results following CAF repair originate from small series and reviews [1-5]. Surgery offers excellent outcomes [1,4,5], however transcatheter closure is emerging as a less invasive alternative. The present study reports 79 consecutive arteriovenous CAF patients treated with transcatheter or surgical closure with an emphasis on reinterventions and survival outcomes.

Material and Methods

The dataset

The National Institute for Cardiovascular Outcomes Research (NICOR) collects data on cardiac procedures from all the UK heart units with the aim of reporting outcomes following surgical and transcatheter procedures (available at https://nicor4.nicor.org.uk/). We identified 79 patients who underwent a coronary fistula procedure between April 2000 and March 2013 from 20 centers.

Statistical analysis

Frequencies are given as absolute numbers and percentages, continuous values as mean (SD) or median (inter-quartile range). Comparisons of proportions were done with the Fisher Exact test. Estimates of long term survival and freedom from reintervention were done with the Kaplan Meier method using mortality (all cause) and reoperations (coronary) as failure events and comparisons of survival and reintervention between groups were evaluated with the log-rank test.

Results

Table 1 shows the demographic, clinical, procedural and follow-up data of the surgical and catheter groups. Out of a total of 88061 patients undergoing catheter and surgical procedures, coronary fistulas repairs were performed in only 0.09% of cases. Sixty six patients (83.5%) had complete follow-up. Figure 1 illustrates the freedom from coronary reintervention at 10 years, by procedure type. There were no early deaths, and mortality at 10 years was 4.8%. No coronary reinterventions were observed in the surgical group, while

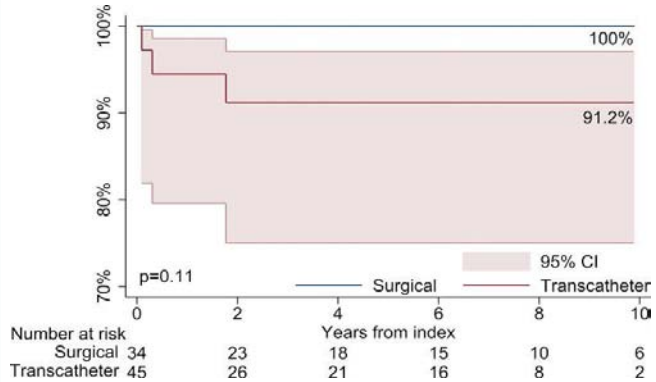

Figure 1: Freedom from coronary reintervention, by procedure type (surgical or transcatheter), Kaplan Meier estimated survivor function.

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Table 1: Demographic, clinical, procedure and follow-up data in patients undergoing coronary fistula repair, by repair type.

National database procedures (over 12 year period)	Overall n=88061	Surgery n=51666	Catheter procedures n=36395	
Coronary fistula procedures (n,%)	Overall n=79 (0.09)	Surgical repair n=34(0.06)	Catheter repair n=45 (0.12)	p value 0.006
Demographics				
Age, y Median (IQR)	11.9 (3.1-54.0)	12.6 (3.0-52.7)	7.8 (3.6-52.7)	0.8
Age group (n,%)				
Neonate	1 (1.3)	0 (0)	1 (2.2)	0.6
Infant	9 (11.4)	6 (17.6)	3 (6.6)	0.1
Child	31 (39.2)	12 (35.3)	19 (42.2)	0.3
Adult	38 (48.1)	16 (47.1)	22 (46.9)	0.6
Gender (n,%)				
Male	39 (49.4)	16 (47.1)	23 (51.1)	0.5
Female	40 (50.6)	18 (52.9)	22 (48.9)	0.5
Associated pathology (n, %)*	25 (31.3)	19 (55.9)	6 (13.3)	<0.001
Procedural details				
Other concomitant procedures†		20 (58.8)		
Bypass duration, min (median, IQR)		117.5 (75-150)		
Cross clamp duration, min (median, IQR)		61.5 (44.5-100.5)		
Procedure duration, min (median, IQR)			106.4 (66.4-165)	
Follow-up				
Hospitalization, days (median, IQR)	4 (1-7)	7 (6-13)	1 (1-3)	<0.001
30 day mortality	0	0	0	
Follow-up period, years (median, IQR)	4.39 (0.9-7.7)	4.84 (1.0-9.6)	4.39 (0.6-7.1)	0.3
Long term outcomes‡				
10 year survival (%, CI)	95.2 [85.3;98.4]	93.0 [74.6;98.2]	97.1 [80.9;99.6]	0.4
10 year freedom from coronary reintervention (%, CI)	95.1 [85.5;98.4]	100	91.2 [75.0;97.1]	0.11
Coronary reinterventions	3	0	3	
Surgical ligation	0	0	1	
Re-do catheter procedures	0	0	2	

*Tetralogy of Fallot (n=4), Pulmonary atresia and VSD (n=4), Aortic valve stenosis (n=1), Supravalvular aortic stenosis (n=1), ALCAPA repair and MR (n=1), ALCAPA repair and RVOT stenosis (n=1), Ascending aortic aneurysm repair and LVOT obstruction (n=1), Left ventricular isomerism and aortic atresia and AVSD (n=1), Mitral and tricuspid pathology (unknown) (n=1), Mitral atresia (n=1), MR and ASD (n=1), VSD (n=1), Pulmonary valve stenosis (n=1), Pulmonary atresia (n=1), Transposition of great arteries, Tricuspid atresia and ASD (n=1), Tricuspid valve endocarditis (n=2), Tricuspid Valve regurgitation (n=1), Superior vena cava persisting to coronary sinus (n=1).

†CABG (n=5), CABG and AVR (n=1), RVOT obstruction repair (n=3), Supravalvular aortic stenosis (n=1), ASD closure (n=1), Pulmonary valve replacement (homograft and arterioplasty (n=1), Pulmonary and tricuspid valve replacement (n=1), mitral valve repair and ASD closure (n=1), Damus-Kaye-Stansel operation and total cavopulmonary connection (n=1), Mitral valve replacement (n=1), Mitral and tricuspid valve procedure (unkonwn) (n=1), tricuspid procedure (unknown (n=1), Pulmonary atresia repair and VSD repair (n=1),VSD closure (n=1).

‡ From Kaplan Meier estimate

ALCAPA: Anomalous Left Coronary from the Pulmonary Artery; ASD: Atrial Septal Defect; AVSD: Atrioventricular Septal Defect; CABG: Coronary Artery By-pass Grafting; MR: Mitral Regurgitation; LVOTO: Left Ventricular Outflow Tract; RVOT: Right Ventricular Outflow Tract; VSD: Ventricular Septal Defect.

a total of 3 patients from the transcatheter group had either a repeat transluminal occlusion (n=2) or a surgical ligation. Out of these, one patient required three occlusion procedures in three years. The freedom from fistula reintervention was 91.2% for the transcatheter group *vs.* 100% in the surgical group (p=0.11). No differences in outcomes were found between patients with isolated fistulas and those with associated abnormalities.

Discussion

Our study demonstrates that surgical closure can be achieved with no operative mortality and excellent long term outcomes. Liberthson et al. [2], in a meta-analysis of 174 surgical CAF repairs, found a mortality of up to 4%. However, more recent series mirror our results reporting 100% long term survival and closure rates in patients treated with surgery [1,4,5]. This led others to consider surgery a

standard against which catheter techniques should be compared [4].

More than half our patients had transcatheter closure with no procedural or long term mortality and significantly lower hospital stay. There were only 3 patients who required a fistula reintervention, all of them in the transcatheter group, which shows a trend towards higher reintervention rates for this group. In 33 patients treated with transcatheter embolisation, Armsby et al. reported no deaths or long term morbidity [3]. The median follow-up time was only 2.8 years and there were no reinterventions reported. However, the authors found residual fistulas in 5 cases (19%).

Transcatheter embolisation is not possible or effective in all cases [4]. Patients with associated cardiac defects that require correction or complex distal coronary anatomy will generally be listed for surgery. This possibly explains why in our study the surgical patients had significantly more associated cardiac defects.

The main limitation of this study is the lack of preoperative imaging to define the anatomy and number of fistulas and incomplete follow-up data. Hence we could not draw conclusions regarding the indication and selection criteria for surgery or catheter closure. What we know is that all decisions were reached in a multidisciplinary setting. The general approach is that in situations of clinical equipoise the less invasive treatment is preferred. Also, the relatively small group with few events only permitted a limited statistical analysis. Nevertheless, this retrospective cohort offers an overview of contemporary practice and outcomes as all patients who underwent treatment for this condition were included in the data extract.

Conclusion

CAF repair via transcatheter or surgical approach is attainable with no early mortality and good long-term results. Although the subgroups are not matched, reintervention rates appear to be higher in transcatheter patients.

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