



Clinical Characteristics and Surgical Outcomes of Coronary Artery Aneurysm: A Case Series

Zahra Azizi¹, Bahador Baharestani², Maziar Gholampour Dehaki², Alireza Yaghoubi², Mohammad Hassan Ghafarinejad², Ali Sadeghpour Tabaei² and Alireza Alizadeh Ghavidel^{3,*}

¹Faculty of Health, York University, Toronto, Canada

²Department of Cardiac Surgery, Rajaie Cardiovascular Medical and Research Center, Iran University of Medical Sciences, Tehran, Iran

³Heart Valve Disease Research Center, Rajaie Cardiovascular Medical and Research Center, Iran University of Medical Sciences, Tehran, Iran

*Corresponding author: Heart Valve Disease Research Center, Rajaie Cardiovascular Medical and Research Center, Iran University of Medical Sciences, Tehran, Iran. Email: aaghavidel@rhc.ac.ir

Received 2019 January 23; Revised 2019 February 10; Accepted 2019 February 13.

Abstract

Background: Coronary arteries aneurysm (CAA) is one the rare and life-threatening abnormalities of the cardiovascular system.

Objectives: Our study aims to present clinical characteristics and surgical results of patients with aneurysm of coronary artery in Iranian population.

Methods: This is a retrospective case series of eight patients with aneurysmal coronary artery, who underwent surgical repair in an Iranian tertiary referral centre from 2004 - 2014 and were followed up till 2017 (mean follow-up: 8.6 ± 3.8 years). Patients' demographics, clinical characteristics, past medical histories and surgical data were recorded retrospectively. All data were analyzed by SPSS software (IBM Corp. IBM SPSS Statistics for Windows, version 22.0. Armonk, NY: IBM Corp.).

Results: Overall eight patients with CAA, consisting of 6 (75%) male and mean age of 35.21 ± 23.76 (0.75 - 67) years, were analyzed. Dyspnea and chest pain were the most frequent symptoms among patients (3 (37.5%)). Moreover, hypertension and coronary artery disease (CAD) were the most common comorbidities amongst our subjects. Angiography of patients showed concomitant CAD in 6 (75%) patients including four (50%) three-vessel disease and two (25%) two-vessel disease. Left anterior descending (LAD) was the most prevalent aneurysmal coronary artery 4 (50%). Six (75%) patients underwent coronary artery bypass graft (CABG) at the same time due to coronary artery disease. One patient had post operative bleeding and underwent re-operation and 2 patients had pericardial effusion that didn't require surgical intervention. There was no perioperative MI, respiratory problems, acute kidney insufficiency (AKI), infections and in-hospital or late mortalities in patients after surgery.

Conclusions: Majority of patients diagnosed and treated for CAA, had concomitant coronary artery disease at the time of testing and underwent CABG. There were no peri and post-operative mortality, and all patients showed satisfying outcome. In addition to the benefits of early diagnosis and intervention in prognosis of patients, larger studies are warranted to identify the possible risk factors of this condition to identify those at risk in order to reduce the consequent complications.

Keywords: Coronary Arteries Aneurysm, Clinical Characteristics, Coronary Artery Disease, Surgical Outcome

1. Background

Coronary artery aneurysms (CAA) are abnormal dilations along the coronary arteries, known as one of the most life-threatening diseases of the cardiovascular system (1-9). While the etiology of this disorder is not clear, it has been associated with concomitant atherosclerosis, history of percutaneous coronary intervention (PCI), drug abuse, autoimmune and infectious disorders (3, 5, 7). History of Kawasaki's disease has also been known to be correlated with the occurrence of CAA and ischemic myocardial infarctions during adulthood secondary to compromised circulation in coronary arteries (4, 7). Surgical interven-

tion is the treatment of choice in patients with CAA refractory to medical therapies (7).

2. Objectives

Although, there is a number of literature reviews on this subject, environmental and genetic characteristics of this disease can be unique in different regions, therefore in this study we sought to report a descriptive analysis about clinical characteristics and surgical outcome of patients with CAA, who had undergone surgical intervention in a tertiary referral center in Iran.

3. Methods

This is a retrospective case series of 8 patients with CAA who underwent surgical repair between 2004 to 2014 in a tertiary referral center in Iran and were followed up till 2017 (mean follow-up: 8.6 ± 3.8 years).

After obtaining informed consent, patients' clinical characteristics, past medical histories and surgical data were recorded retrospectively. Moreover, pre and post-operative echocardiographic, X-ray and angiographic data were also gathered for each patient.

This study was approved by Ethics Committee of Iran University of Medical Sciences.

Surgical repair was performed under general anesthesia with median sternotomy approach using cardiopulmonary bypass for all patients. Two techniques were used for repair of aneurysmal section of the coronary arteries based on the location and size of the aneurysm and also surgeons' preference. For small aneurysms suture ligation of the proximal and distal parts of aneurysm and its related small branches using 5-0 polypropylene sutures (PROLENE® Polypropylene Suture|Ethicon) with or without pledgets. For the large ones, after un-roofing the aneurysm by a longitudinal sharp incision and removing the organized clots and debris the proximal, distal ends and also small branches (including septal branches) were ligated by the same techniques. Concomitant surgeries including coronary artery bypass graft (CABG) were performed according to patients' pathologies (Table 1).

Descriptive statistics were performed and reported as mean \pm standard deviation (median) for continuous variables and frequency (percentage) for categorical variables. P value under 0.05 was considered as statistically significant. All data were analyzed by SPSS software (version 23, IBM Corp).

4. Results

Total of 8 patients, 6 (75%) male with the mean age of 35.21 ± 23.76 years (0.75 - 67) were analyzed.

Overall eight patients with CAA, consisting of 6 (75%) male and mean age of 35.21 ± 23.76 (0.75 - 67) years, were analyzed.

Dyspnea and chest pain were the most frequent symptoms (3 (37.5%)). One patient (12.5%) reported palpitation, and 1 (12.5%) was symptom free. Physical examination showed palpable thrill in one (12.5%) patient, and abnormal lung sound in another one (12.5%). Moreover, patients past medical history showed, hypertension in 2 (25%) patients, coronary syndrome in 2 (25%), Ischemic heart disease in 3 (37.5%), rheumatoid heart disease in 1 (12.5%), pre-

vious steroid use in 1 (12.5%), and family history of CAA in one (12.5%) patient.

Pre-operative echocardiography showed valve disease in 5 (62.5%) patients. The most common valve disease was pulmonary insufficiency (PI, 3 (37.5%)) and mitral regurgitation (MR, 3 (37.5%)), however only one patient had severe conditions and required mitral valve replacement. Only one patient (12.5%) had pre-operative pericardial effusion.

Pre-operative angiography of patients showed concomitant coronary artery disease in 6 (75%) patients amongst whom, 4 (50%) had three-vessel disease and 2 (25%) had two-vessel disease. Left anterior descending (LAD) was the most prevalent aneurysmal coronary artery (4 (50%)). In addition, one patient (12.5%) had concomitant coronary artery fistula.

The result of CXR was normal in all patients, except in one patient (12.5%) that showed increased CT ratio and pulmonary vascular malformations (PVM).

Amongst eight patients who underwent surgical repair, 6 (75%) patients underwent concomitant CABG due to Coronary artery disease (Table 1).

Pre and post-operative hemoglobin were 12.9 ± 2.3 (9 - 17) g/dL and 12.05 ± 1.9 (9.6 - 14.3) g/dL respectively, while, pre and post-operative creatinine were 0.9 ± 0.34 (0.3 - 1.3) g/dL and 1.2 ± 0.1 (1.1 - 1.3) g/dL, respectively.

Mean post-operative ICU stay was 7 ± 8.94 (3 - 27) days with a median of 3 days, while mean hospital stay was 14 ± 2.13 (11 - 18) days with a median of 14 days. Mean CPB time was 31.05 (41 - 134) minutes.

Only one (12.5%) patient had post-operative bleeding and required reoperation. One patient (12.5%) required inotrope use and 3 (37.5%) had blood transfusion. Two patients (25%) had pericardial effusion post surgery, however none of them required surgical intervention. There was no perioperative MI, respiratory problems, acute kidney insufficiency (AKI), infections, in-hospital or late mortality in patients after surgery during our mean follow up time of 8.6 ± 3.8 years.

5. Discussion

The result of our study showed dyspnea and chest pain as the most common symptoms among patients. Hypertension and coronary artery disease were the most prevalent comorbidities among patients. The majority of our patients underwent simultaneous CABG with their aneurysmal repair. Only one patient had post operative bleeding that required re-operation. Two patients had pericardial effusion, however none of them required surgical intervention. There was no perioperative MI, respiratory problems, AKI, infections, in-hospital or late mortalities in patients af-

Table 1. Individual Characteristics, Angiographic, and Surgical Findings of Patients with CAA

Patients	Sex	Age, y	Pre-Op EF	Pre-OP NYHA	OP Date	Follow-Up, y	Comorbidities	Aneurysmal Coronary Artery	Concomitant Surgery	Type of Graft and Target Vessels
Patient 1	Male	47	40	2	2004	13	IHD	RCA	CABG (3 VD, 5 graft)	SVG to diagonal, LIMA to LAD, SVG to RV branch, SVG to PDA, SVG to PL
Patient 2	Female	0.75	74	-	2005	12	ASD, SDS, coronary fistula	LAD	Coronary fistula repair, PFO closing, aneurysmorrhaphy	-
Patient 3	Male	45	35	-	2006	11	HTN, DLP, IHD, MI, MR	LCX	CABG (3VD, 3 graft)	LIMA to LAD, SVG to RCA, SVG to OM
Patient 4	Male	67	30	1	2007	10	HTN	LAD	CABG (3VD, 3 graft), Endarterectomy	LIMA to LAD, SVG to OM, SVG to RCA
Patient 5	Male	16	50	-	2008	9	IHD, MI, PI	LCX	CABG (2SVD, 2 graft)	LIMA to OM1, SVG to OM2
Patient 6	Male	20	55	2	2009	8	Double chamber RV, VSD, PS, AS, AI	RCA	AVR, PS repair, VSD closure, aneurysmorrhaphy	-
Patient 7	Female	63	40	2	2014	3	IHD, PI, TR, MR, PE	LAD	CABG (3VD, 3 graft), MVR	LIMA to LAD, SVG to OM, SVG to diagonal
Patient 8	Male	23	55	-	2014	3	PI, AI, MR	LAD	CABG (2VD, 2 graft)	LIMA to LAD, SVG to diagonal

Abbreviations: AI, aortic insufficiency; AS, aortic stenosis; AVR, aortic valve replacement; CABG, coronary artery bypass graft; LAD, left anterior descending; LCX, left circumflex artery; LIMA, left internal mammary artery; MVR, mitral valve replacement; OM, obtuse marginal; OP, operation; PDA, posterior descending artery; PFO, patent foramen ovale; PI, pulmonary insufficiency; PL, posterolateral; PS, pulmonary stenosis; PS repair, pulmonary stenosis repair; RCA, right coronary artery; RV, right ventricle; SVG, saphenous vein grafts; TR, tricuspid Regurgitation; VD, vessel disease; VSD, ventricular septal defect.

ter surgery. All patients showed satisfying outcome after surgery.

CAA is an extremely rare disease amongst the population who undergo cardiovascular surgical intervention with prevalence rate ranging from 1.5% - 6.5% (4, 5, 8, 10). CAA is mostly asymptomatic, however, in symptomatic cases, the most common clinical manifestations are similar to coronary artery disease (CAD) (8, 11). Dyspnea and chest pain were the most prevalent symptoms among our patients. In some cases presence of murmur has been reported (11). Only one patient in our study revealed palpable thrill.

CAD has been known as the most notable comorbidity in most cases of CAA (1, 2, 11). This has been supported by a recent review by Boyer et al. (5) describing CAD as the most common etiology of CAA. This is very similar to our findings showing concomitant CAD in the majority of patients who underwent CABG during aneurysmal repair procedure. Additionally, studies have discussed the association of CAA and hypertension (11). This hypothesis can be notable since Hypertension was one of the prevalent comorbidities beside CAD among our patients.

As shown in most studies, CAA has a wide range of signs and symptoms and a single symptom can not be diagnosed based on one symptom. Inokuchi et al. (4) reported strawberry tongue an atypical presentation of CCA secondary to Kawasaki disease. Therefore, diagnosis of this disease can be quite challenging in patients without CAD, since angiographic testing is rarely done on patients without CAD symptoms.

There is no consensus on the best approach for treatment of CAA, and the treatment options mostly depends on patients underlying condition and clinical presenta-

tion (11).

A prospective corono-angiography registry of patients with CAA, showed 3.1% mortality peri-admission and 20.7% mortality within median follow-up time of 52 months (27 - 84). While the most frequent procedure was percutaneous revascularization, surgical intervention was performed in 12.9% of the patients. Significant bleeding was reported in 10.1%, arterial thromboembolic events in 1.8%, and stroke in 3.9%. Death rate was 0% in simple percutaneous transluminal coronary angioplasty (PTCA), 35% in conventional stent, 7.8% in drug-eluting stent and 21.2% in surgical method, while, The MACE (death and/or un-stable angina and/or myocardial infarction and/or heart failure and/or stroke) was reported in 60%, 50%, 37.3% and 19.2% of the reported procedures, respectively (12). Our result demonstrated no perioperative MI, respiratory problems, AKI, infections, in-hospital or late mortalities after surgery.

5.1. Limitations

The main limitation of our study was the retrospective design and small sample size of the patients.

5.2. Conclusions

Aneurysm of the coronary artery remains of the most life-threatening abnormalities of the vascular system. Although the prevalence of such disease is low, its diagnosis is extremely difficult without CAD symptoms. Majority of patients diagnosed and treated for CAA, also manifested CAD at the time of testing. While several studies have discussed different pathophysiology of CAA, the exact mechanism of this disease is still unclear. In addition to the benefits of early diagnosis in prognosis of patients,

larger studies are warranted to identify the possible risk factors of this condition to identify those at risk in order to reduce the consequent complications. Majority of patients diagnosed and treated for CAA in our study had concomitant coronary artery disease at the time of testing and underwent CABG. There was no perioperative MI, respiratory problems, AKI, infections, in-hospital or late mortality in patients after surgery. All patients showed satisfying outcome after surgery.

Footnotes

Authors' Contribution: Study concept and design: All authors; acquisition of data: Zahra Azizi; analysis and interpretation of data: Zahra Azizi; drafting of the manuscript: All Authors; critical revision of the manuscript for important intellectual content: All authors; statistical analysis: Zahra Azizi; administrative, technical, and material support: Zahra Azizi, Alireza Alizadeh Ghavidel; study supervision: Zahra Azizi, Alireza Alizadeh Ghavidel.

Ethical Approval: This study was approved by Ethics Committee of Iran University of Medical Sciences (code: 94014, date: 9/12/94).

Conflict of Interests: None declared.

Financial Disclosure: The authors have no disclosures.

Funding/Support: None.

Patient Consent: After obtaining informed consent, patients' clinical characteristics, past medical histories and surgical data were recorded retrospectively.

References

1. Daoud AS, Pankin D, Tulgan H, Florentin RA. Aneurysms of the coronary artery. Report of ten cases and review of literature. *Am J Cardiol.* 1963;**11**:228–37. doi: [10.1016/0002-9149\(63\)90064-X](#). [PubMed: [14025069](#)].
2. Befeler B, Aranda MJ, Embi A, Mullin FL, El-Sherif N, Lazzara R. Coronary artery aneurysms: Study of the etiology, clinical course and effect on left ventricular function and prognosis. *Am J Med.* 1977;**62**(4):597–607. doi: [10.1016/0002-9343\(77\)90423-5](#). [PubMed: [300567](#)].
3. Swaye PS, Fisher LD, Litwin P, Vignola PA, Judkins MP, Kemp HG, et al. Aneurysmal coronary artery disease. *Circulation.* 1983;**67**(1):134–8. doi: [10.1161/01.CIR.67.1.134](#). [PubMed: [6847792](#)].
4. Inokuchi R, Kurata H, Harada M, Aoki Y, Matsubara T, Doi K, et al. Coronary artery aneurysms after adult-onset Kawasaki disease. *Circulation.* 2013;**127**(15):1636–7. doi: [10.1161/CIRCULATIONAHA.112.130153](#). [PubMed: [23588962](#)].
5. Boyer N, Gupta R, Schevchuck A, Hindnavis V, Maliske S, Sheldon M, et al. Coronary artery aneurysms in acute coronary syndrome: Case series, review, and proposed management strategy. *J Invasive Cardiol.* 2014;**26**(6):283–90. [PubMed: [24907086](#)].
6. Heizer J, Petersen TC, Flemmer MC. Multiple coronary aneurysms in a young adult with acquired immunodeficiency syndrome. *Oxf Med Case Reports.* 2016;**2016**(5):109–12. doi: [10.1093/omcr/omw036](#). [PubMed: [27168940](#)]. [PubMed Central: [PMC4860520](#)].
7. Jahollari A, Sarac A, Tafaj O, Ozal E. Surgical management of a giant right coronary artery aneurysm. *Heart Surg Forum*; 2014.
8. Crawley PD, Mahlow WJ, Huntsinger DR, Afiniwala S, Wortham DC. Giant coronary artery aneurysms: Review and update. *Tex Heart Inst J.* 2014;**41**(6):603–8. doi: [10.14503/THIJ-13-3896](#). [PubMed: [25593524](#)]. [PubMed Central: [PMC4251331](#)].
9. Misra A, Majumder B, Tandel V, Ahmed A. Giant aneurysm of right coronary artery: A case report and review of literature. *Nigeria J Cardiol.* 2016;**13**(2):144. doi: [10.4103/0189-7969.187732](#).
10. Dolapoglu A, Ott DA. Giant right coronary artery aneurysm associated with a fistula draining into the superior vena cava. *Tex Heart Inst J.* 2016;**43**(4):360–2. doi: [10.14503/THIJ-15-5450](#). [PubMed: [27547153](#)]. [PubMed Central: [PMC4979401](#)].
11. Abou Sherif S, Ozden Tok O, Taskoylu O, Goktekin O, Kilic ID. Coronary artery aneurysms: A review of the epidemiology, pathophysiology, diagnosis, and treatment. *Front Cardiovasc Med.* 2017;**4**:24. doi: [10.3389/fcvm.2017.00024](#). [PubMed: [28529940](#)]. [PubMed Central: [PMC5418231](#)].
12. Nunez-Gil IJ, Terol B, Feltes G, Nombela-Franco L, Salinas P, Escaned J, et al. Coronary aneurysms in the acute patient: Incidence, characterization and long-term management results. *Cardiovasc Revasc Med.* 2018;**19**(5 Pt B):589–96. doi: [10.1016/j.carrev.2017.12.003](#). [PubMed: [29276176](#)].