Gender-related differences in octogenarians with congenital coronary artery fistula: a report of two cases and a review

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Published online: 29 September 2011
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Abstract

Aim To highlight gender-related differences in octogenarians with a congenital coronary artery fistula (CAF).

Materials and methods We present two elderly female patients with a congenital fistula, a septuagenarian and a nonagenarian, and review the world literature between 1954–2010.

Results The septuagenarian patient presented with easy fatigability and the nonagenarian patient with acute myocardial infarction contralaterally to the fistula. Coronary angiography (CAG) demonstrated a coronary-pulmonary artery fistula (CPF). The nonagenarian patient underwent percutaneous coronary intervention of the right coronary artery. CAG revealed a CPF associated with a huge multiple aneurysmal formation. Data from 57 mainly symptomatic patients with a mean age of 75.3 years (range 70–87 years) were collected. The cohort was subdivided

into female (mean age 84.3 years) and male (mean age 75.2 years) subgroups and compared with each other. Multi-origin (bilateral and multilateral) was prevalent in females, 40% versus 12% in males. Aneurysmal formation was found in females and males in 40% and 18%, respectively. Ethnicity was 65% Caucasian and 35% Asian. Multi-origin fistulas were prevalent in the Asian (45%) compared with the Caucasian (11%) subset.

Conclusions A septuagenarian and a nonagenarian female patient with congenital CAF are presented. On reviewing the literature, important differences were found between elderly females and males with congenital CAF.

Keywords Congenital anomaly · Coronary angiography · Coronary artery fistula · Elderly population · Surgical ligation · Percutaneous therapeutic embolization

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Introduction

Most congenital coronary artery fistulas (CAFs) are identified incidentally during conventional coronary angiography (CAG). Large congenital CAFs are infrequent anomalies and may be symptomatic and detected in the early life. Small congenital CAFs are frequent coronary artery anomalies which usually remain asymptomatic. Young patients <30 years of age are usually asymptomatic [1]. CAFs may remain silent for years to give rise to symptoms of complications starting to occur after the third decade of life; they may also be incidentally found during diagnostic CAG. The most common silent fistulas are coronary-pulmonary artery fistulas [2, 3], as was the case of the incidentally found fistula in 1 of the 2 patients presented here.

CAFs may be isolated or associated with other congenital anomalies [4–6]. Currently, CAFs and their anatomical morphology are increasingly being detected, non-invasively and invasively, due to the widespread application and availability of computed tomography scanning [7], multi-detector computed tomography [8], cardiovascular magnetic resonance [9–11], transthoracic echocardiography [12], transoesophageal echocardiography (TEE) [13] and CAG [12, 14, 15]. The incidence is estimated at 0.1–0.4% of the adult coronary angiographic population [16, 17]. However, an abnormal shadow on the chest X-ray may bring CAF under the attention of practitioners [9, 11, 18, 19]. Several non-invasive investigation techniques have been used for the functional assessment of CAF, such as myocardial perfusion scanning [20, 21], contrast TEE [13] and dobutamine stress echocardiography [22, 23].

Gradual dilatation of the fistulous vessels, spanning over many years, may be observed with increasing age and this may explain the late onset of symptoms and the delayed development of congestive heart failure and atrial fibrillation in the elderly subjects [24–28].

In the elderly, CAF can be managed with conservative medical management [29–34], with a percutaneous therapeutic embolisation approach [21, 35–38] or with surgical closure techniques [1, 6, 28, 39–43].

Materials and methods

Case reports

Case 1: A septuagenarian female patient was evaluated for complaints of a few months of reduced exercise capacity and rapid fatigability; she sometimes had an oppressive feeling in the chest. Selective CAG showed normally originating epicardial right (RCA) and left (LCA) coronary arteries. A coronary artery-pulmonary artery fistula was visible (Fig. 1) between the proximal segment of the left anterior descending coronary artery (LAD) and the pulmonary artery (PA). She was treated medically. She remains free of symptoms.

Case 2: A nonagenarian woman was admitted for percutaneous primary coronary intervention (PCI) due to acute inferior wall myocardial infarction. Successful PCI was performed of the totally occluded RCA with placement of a bare metal stent with resulting good flow and resolution of symptoms. Emergent CAG demonstrated a normal origin of the RCA and LCA with huge dilatation of the left main stem. A fistula was visible between the anterolateral branch of the LCA and the PA associated with excessive aneurysmal (51×48 and 52.5×48 mm) formation (Fig. 2). Medical treatment was initiated. She is still doing well.

Data of the two patients are summarised in Table 1.

Fig. 1 Coronary-pulmonary artery fistula (CPF) in a septuagenarian female patient: Originating from the proximal segment of the left anterior descending (LAD) artery and terminating into the pulmonary artery (PA). Origin and termination had a single opening and the fistulous vessel showed a tortuous pathway

Literature review

We performed a literature search of the Medline database using the PubMed search interface to identify reports dealing with congenital CAFs in octogenarians. The search

Fig. 2 In a nonagenarian female, from the proximal portion of the anterolateral branch of the left coronary artery, a huge fistula originated and terminated into the PA associated with two large saccular aneurysms measuring 52.5×48 mm and 51×48 mm (arrow)

Table 1 Data of two elderly patients with congenital coronary artery fistulas

Case/age/gender	Comorbidity	Clinical presentation	Cardiac murmur	CAG/CAF	Management
1–73 F	Hypertension COPD	Reduced exercise capacity, rapid fatigability, chest pain	SM	LAD-PA	CMM Metoprolol 50 mg/day Nifedipine 60 mg/day Seretide 250/50 µg b.i.d
2–94 F	Hypertension PAF	AMI, contralateral to the shunt	SM	AL-PA Aneurysmal formation CAD	CMM/PCI of RCA Metoprolol 150 mg/day Acenocoumarol Lisinopril 20 mg/day Clopidogrel 75 mg/day Spironolactone 50 mg/day

F female, COPD chronic obstructive pulmonary disease, PAF paroxysmal atrial fibrillation, AMI acute myocardial infarction, SM systolic murmur, CAG coronary angiography, CAF coronary artery fistula, LAD left anterior descending coronary artery, AL anterolateral coronary artery, PA pulmonary artery, CAD coronary artery disease, CMM conservative medical management, PCI primary coronary intervention, RCA right coronary artery.

included English and non-English literature between 1954 and 2010. This search resulted in 52 published reports. The following criteria were stipulated in order to obtain a homogenous cohort of study: patients were ≥ 70 years of age, clinical presentation and confirmation of diagnosis and treatment modality were stated in the paper. Patients with acquired traumatic iatrogenic or accidental CAF were excluded. Subjects with congenital coronary artery-ventricular multiple microfistulas were not included.

Results

Among the reported cases in the English and non-English literature between 1954 and 2010, we found a total of 57 symptomatic patients, except 2 (who were asymptomatic), with 70 fistulas. Forty (70%) were females and 17 (30%) males. The mean age was 75.3 years (range 70–87 years). According to gender, 2 subsets were formed and compared with each other Table 2.

Gender and clinical presentation

There were 70% females (mean age 84.3, range 70–87 years) and 30% males (mean age 75.2, range 70–85 years). Females differed significantly from males in that the CAFs were reported twice as often as in males.

The clinical presentation contained a single symptom in 35 patients and was composed of multiple symptoms in 16, giving a total of 70 symptoms; these data were not reported in 4 of the patients. Left-sided congestive heart failure (CHF) was reported 13 times and right-sided CHF once, while dyspnoea was present 18 times. Chest pain occurred 12 times, angina pectoris 6 times and acute coronary syndrome once. Furthermore, recurrent and chronic pericardial effusion,

haemopericardium and tamponade were each reported once and an abnormal shadow on chest radiograph brought 4 patients under medical attention. Palpitations, presyncope and syncope were the presenting symptom 6 times, fatigue 4 times and infective endocarditis in 1 patient.

Supraventricular arrhythmias were reported 14 times: atrial fibrillation in 13 cases and supraventricular tachycardia in 1 case.

Associated, congenital and acquired, cardiac anomalies were 4 disorders seen in 2 patients and non-cardiac disorders in 8 of the patients. The concomitant congenital lesions comprised atrial septal defects twice, bicuspid aortic valve once and pulmonary valvular stenosis once. Insignificant and significant valvular heart disease was reported in 7 and in 2 of the patients, respectively.

Gender and cardiac murmur

Thirty-five cardiac murmurs were reported in 33 patients (58%). The murmur was continuous in 20 (57%), systolic in 13 (37%) and diastolic in 2 (6%) of the patients. Four patients had no audible murmur (7%); these data were not recorded in 20 (35%).

Cardiac murmur was reported in 27/40 female patients (68%) but in 6/17 (35%) males. It was continuous in 17 females and in 3 males, systolic in 10 females and 3 males, and diastolic in 1 each. Only 1 female and 3 male patients had no audible murmur; these data were not recorded in 12 females and 8 males.

Gender and fistula characteristics

Unilateral fistulas were detected in 44, bilateral in 11 and multilateral in 2 of the patients. The origin ($n=70$) of the fistulas was the LCA in 63% and the RCA participated in

Table 2 Gender differences in elderly patients ($n=57$) with congenital coronary artery fistulas

Age rang in years (mean)	Females $n=40$ (70%) 70-87 (84.3)	Males $n=17$ (30%) 70-85 (75.3)
Clinical presentation		
Single symptom	23/40 (57%)	12/17 (71%)
Multiple symptoms	13/40 (33%)	3/17 (18%)
Not reported	4/40 (10%)	2/17 (11%)
Congestive heart failure and dyspnoea	24×	8×
Chest pain [#]	10×	8×
Abnormal chest x-ray	3×	1×
Pericardial effusion	3×	0
Cardiac murmur		
Patients with murmur	27 (68%)	6 (35%)
No murmur	1	3
Not reported	12	8
Mean pulmonary/systemic flow ratio	$n=19$	$n=6$
Qp:Qs	1.9 : 1.0	1.7 : 1.0
Fistula characteristics		
Unilateral	29 (72%)	15 (88%)
Bilateral	9 (23%)	2 (12%)
Multilateral	2 (5%)	0
Multi-origin	11/40 (40%)	2/17 (12%)
Origin ($n=70$)		
Left coronary artery	34/51 (67%)	10/19 (53%)
Right coronary artery	17/51 (33%)	9/19 (47%)
Termination		
Pulmonary artery	24/40 (60%)	9/17 (53%)
Coronary sinus	10/40 (25%)	4/17 (24%)
Aneurysmal formation	16/40 (40%)	3/17 (18%)
Associated congenital cardiac lesions	4×	0
Associated acquired cardiac disorders		
Coronary artery disease	4	5
Supraventricular arrhythmia	8	5
Associated non-cardiac conditions	3	1
Management		
Surgical ligation	21 (53%)	6 (35%)
Percutaneous therapeutic embolisation	4 (10%) (Failed $n=2$)	3 (18%) (Failed $n=1$)
Conservative medical management	13 (32%)	5 (29%)
Not reported	2 (5%)	3 (18%)

[#]=chest pain (angina pectoris and atypical chest pain)

37% of the fistulas. Origin from the left main stem was reported in 4, from the LAD in 28 and from the circumflex coronary artery (Cx) in 14 of the fistulas. The termination ($n=57$) was into the PA in 33 (58%), ending in the coronary sinus and right atrium in 14 (25%), drainage to the right ventricular outflow tract in 2, right ventricle in 2, the right superior vena cava in 2, and terminating in 1 of each of the following: left persistent superior vena cava, left ventricle, left atrium and pulmonary vein.

Associated aneurysmal formation was depicted in 19 out of 57 patients (33%). Significant obstructive coronary artery

disease was present in 8 patients and minor narrowing or a normal coronary arterial tree in 49 of the subjects.

Multiple origin (bilateral and multilateral) was prevalent in females (40%) versus 12% in males. Bilateral CAFs are common in females: 23% versus 12% in men.

In nearly half of the patients ($n=25$), the step-up oxygen was documented. The calculated mean Qp:Qs was 2:1. Mean pulmonary to systemic flow ratio (Qp:Qs) was 2.3:1 in females and 1.7:1 in males.

Bilateral fistulas formed 19% (11/57). Of these, combined fistulas from the LAD+RCA were predomi-

nant (73%): from Cx+RCA this was 18% and from LM+RCA 9%.

Drainage into the PA was described in 60% of the female versus 53% of male subjects and into the right atrium/coronary sinus this was 25% versus 24% in males.

Mild to moderate pulmonary hypertension was described in 3 reports [12, 28, 44]. Myocardial infarction (MI) was documented in 3 octogenarian subjects; 2 males and 1 female (all had an Asian ethnic origin). In 2, MI was ipsilateral in the absence of coronary artery disease (CAD) and in 1 it was contralateral to the fistulous connection with significant CAD.

Gender and management

Different treatment modalities were performed; in 27 (47%) patients surgical ligation was conducted, in 7 (12%) percutaneous therapeutic embolisation (PTE) attempts to obliterate the fistula were undertaken, in 18 (32%) patients a conservative medical management (CMM) was chosen and 1 was reported post-mortem but was treated medically during life. Therapeutic strategy was not reported in 5 (9%) subjects. Two female patients refused surgical ligation. In 3 patients percutaneous therapeutic embolisation failed to obliterate the fistula.

Surgical ligation was the frequently applied treatment strategy of choice in both female and male genders (53% vs 35%) followed by CMM (33% vs 29%) and PTE (10% vs 18%).

Octogenarians and ethnicity

The ethnic origin was Caucasian in 37 patients (65%, 24 women and 13 men) and Asian in 20 (35%, 16 women and 4 men). Of the Asian subset, 65% developed an aneurysm. In the Caucasian population this was 16%.

Of the 19 patients (16 females and 3 males) (mean age 76.6, range 70–87 years) with aneurysmal formation, 68% had an Asian origin and 32% a Caucasian origin. Aneurysmal formation was more common in females with an Asian ethnic background (75%). Among the Asian ethnic group, the bilateral and multilateral fistulas were 45% compared with 11% in the Caucasian subset.

Statistical analysis

All data are presented as means, percentages, or range.

Discussion

Data describing a large cohort of octogenarians with congenital CAF are still lacking.

Congenital CAFs are often detected during infancy and early adulthood because of developing symptoms, complications, or presence of a continuous cardiac murmur. As has been demonstrated by our presented cases, it is quite remarkable that congenital CAFs are still diagnosed in the 7th and 8th [45] or 9th decade of life [46–48]. CAFs that remain undetected or not obliterated in childhood have been reported to cause symptoms and complications in adulthood because of chronic volume overload [49]. Furthermore, Liberthson et al. found that 63% of patients older than 20 years of age had either symptoms or complications of CAF [49].

On the other hand, small-sized congenital CAFs have been reported to be associated with longevity with a benign natural history [50]. Several case studies and reports, as was the case in our septuagenarian and nonagenarian patient, have elucidated the long-term survival in this category of patients with a limited-size CAF [16].

It has been found that approximately 75% of CAFs detected incidentally are small and clinically irrelevant [2]. Although it has been reported that small, asymptomatic CAFs arise much more commonly from the left coronary artery (LCA) (87%) [3], in contrast, in this review of the elderly symptomatic population we found that the origin of the CAF was the LCA in 66%, which was associated with moderate-size L-R shunt (Qp:Qs 2:1). The mean pulmonary-systemic flow ratio was larger in females 2.3:1 compared with males 1.7:1.

The current elderly cohort from the literature review contained subjects mainly described from case reports; the CAF terminated into the PA in 58% and coronary sinus/right atrium in 25%. Sapin et al. recognised that fistulas described in case reports are more frequently associated with a continuous murmur and more likely to develop symptoms and complications requiring surgical intervention [51]. Indeed we found that 47% of the population under review was surgically treated and in 12% percutaneous therapeutic embolisation was attempted. Fifty-three percent and 35% of females and males, respectively, were surgically treated. Furthermore, fistulas detected incidentally by CAG are more often multiple, originating from the left coronary artery and draining into the PA, as was found in our presented cases, or the left ventricle [51–54] or terminating into the pulmonary vein or right ventricular outflow tract [55]. In this elderly population, bilateral and multilateral fistulas were present in 23% of the patients.

Aneurysmal formation has been reported in unilateral [42, 56] and bilateral [18, 19, 52] fistulas. Rupture of congenital aneurysmal fistulas occurred more often in female patients, especially when there is a saccular type of aneurysm originating from the LAD [57]. It has been described earlier that female subjects are prone to develop rupture of an associated aneurysm, haemopericardium and

cardiac tamponade [57, 58]. Aneurysmal formation (33%) complicated the fistulas of the reviewed elderly subjects. Thirteen out of 20 patients (65%) with an Asian ethnicity developed an aneurysm compared with 16% of the Caucasian subset.

Collins reported that dyspnoea was present in 36% of patients [21] and Sapin et al. [51] noted that the most frequent clinical presentations were symptoms of dyspnoea or fatigue (25%), congestive heart failure (20%), and angina pectoris (20%) [51] and that the pulmonary hypertension associated with CAF terminating into the right heart side is usually mild to moderate [12, 28, 44]. In our current review, congestive heart failure and dyspnoea were responsible for 46% of clinical presentation.

Sapin et al. reported that the typical continuous cardiac murmur of a moderate or large CAF was present in 40% of patients, isolated systolic (20%) and diastolic (3%) murmurs were heard, but 10% had no audible cardiac murmur and it was not reported in a quarter of the elderly patients [51]. Our findings are in accordance with those of Sapin et al., with an exception that the rate of reported continuous murmur was higher among the reviewed patients; 35 cardiac murmurs were reported in 33 (58%) of the reviewed patients. It had a continuous character in 57%, was systolic in 37% and in 6% a diastolic component was heard. Four patients (7%) had no audible heart murmur at all and in 20 (35%) the murmur was not recorded. Systolic murmur alone was reported by several authors [59–61]. Cardiac murmurs are twice as frequent in women (68%) compared with men (35%).

Approximately 10–30% of patients with congenital CAF also have concomitant congenital cardiovascular disorder including tetralogy of Fallot, patent ductus arteriosus and atrial or ventricular septal defect [62, 63]. In the current review, 4% of the patients had other congenital anomalies. Coronary artery disease combined with CAF was diagnosed in 40% of patients in the series of Cebi et al. [6] but in the present review, we found associated coronary artery disease (CAD) in only 14% of the whole cohort. Our octogenarian patient had no CAD and she was treated medically. Conservative medical management was applied in 32% of the reviewed cohort.

Myocardial infarction (MI) was documented only in 3 octogenarian subjects with an Asian ethnic origin. In 2, MI was isplateral and in 1 it was contra-lateral to the fistulous connection. In our Caucasian nonagenarian patient, MI was located contra-laterally to the shunt with total occlusion of the RCA.

The present paper discusses the difference between the two genders as well as differences in the clinical presentation and treatment strategies of elderly reviewed patients with congenital CAF. To the best of our knowledge, our presented nonagenarian patient is the oldest ever reported

female with congenital CAF (between the anterolateral branch of the LCA and the PA) associated with multiple aneurysmal formation. This case emphasises the longevity of unrecognised and untreated patients with congenital CAF even when complicated with aneurysms.

Conclusion

This paper shows that octogenarian females differ significantly from males in that CAF is reported twice as much in females (70% vs 30%) than males. Bilateral CAF (23% vs 12%) is prevalent in females and termination into the PA is frequent in both genders (60% vs 53%). Associated aneurysmal formation was commonly found in females (40% vs 18%) and surgical ligation was the frequent treatment strategy of choice in both genders (53% vs 35%). Moreover, we found that aneurysmal formation was significantly higher in the Asian ethnic subgroup (65%) compared with the Caucasian subset (16%).

Limitations of the study

This review aimed to determine gender-related differences in octogenarians with congenital CAF comparing several variables. Longitudinal and clinical prospective studies examining the effect and nature of aneurysmal CAF in the elderly are lacking and thus required for determination of the fate of these disorders and their impact on longevity.

In the current paper, the majority of the reviewed patients were collected from case reports. Case reports with specific issues are easily published and other similar cases are not reported. Only interesting case reports with specific issues are likely to be published. This may be a source of publication bias. Therefore, the data must be interpreted with some caution.

Acknowledgement The authors appreciate the assistance of librarians of Hospital Group Twente, Mrs. A. Geerdink and Mr. D. Maas, during the preparation of the manuscript.

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