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CASE REPORT

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Aneurysmal coronary arteriovenous fistula closing with covered stent deployment: A case report and review of literature

Samad Ghaffari, Fariborz Akbarzadeh, Leili Pourafkari

Cardiovascular Research Department of Tabriz University of Medical Sciences, Tabriz, Iran

Abstract

Coronary arteriovenous fistula (CAVF) is a rarely encountered congenital coronary anomaly, in which coronary artery blood flow bypasses the myocardial capillary network and usually drains to a heart chamber or great vessel. It is usually asymptomatic. However, the risk of symptoms and complications increases with age. High output heart failure, pulmonary hypertension, myocardial ischemia and infective endocarditis may complicate the course of this anomaly. The appearance, and even rupture, of a saccular aneurysm is one of the even rarer complications of CAVF. Here we describe a 57 year-old patient with incidental finding of an aneurysmal CAVF which was treated successfully using covered stent. (Cardiol J 2011; 18, 5: 556–559)

Key words: coronary fistula, coronary angiography, covered stent

Case report

A 57 year-old diabetic male with atypical chest pain and a mediastinal mass above heart contour in his chest roentgenogram (Fig. 1) was referred to our clinic for further evaluation. An exercise tolerance test was inconclusive, so we decided to proceed with multi-slice computed tomographic (MSCT) coronary angiography which showed a calcified mass over the heart with a stalk originating from a rudimentary diagonal branch of left anterior descending artery (LAD), and another (rather smaller) mass distal to it (Figs. 2, 3). Coronary angiography was performed. A calcified ovoid mass was present over the heart at fluoroscopy which was confirmed to be an aneurysmal coronary arteriovenous fistula originating from diagonal branch (Fig. 4); other parts of coronary arteries were angiographically normal. The largest diameter of mass was about 4 cm, and because of the narrow stalk and large volume of the sacs the drainage site of the fistula was poorly visualized (Fig. 5) but seemed to

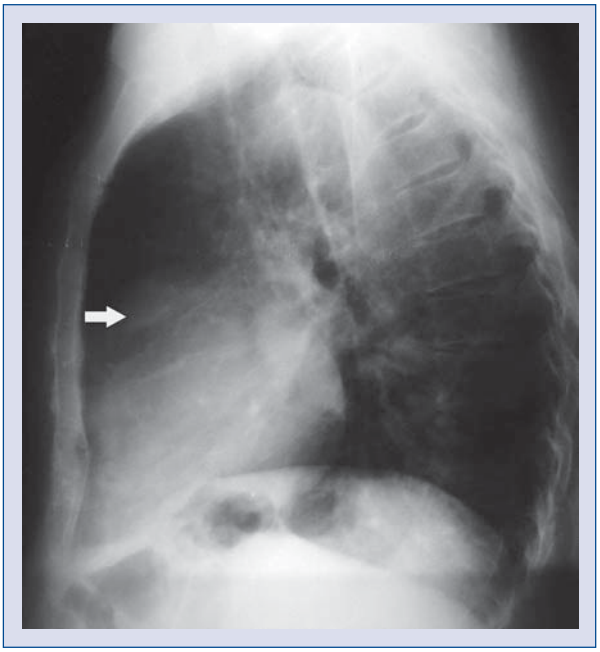

Figure 1. Lateral chest roentgenogram showing the mass above heart contour.

Address for correspondence: Fariborz Akbarzadeh, MD, Associate Professor in Cardiology, Cardiovascular Research Department of Tabriz University of Medical Sciences, Daneshgah st., Postal Code: 5166615573, Tabriz, Iran, tel: +98 411 3357770, fax: +98 411 3344021, e-mail: f_akbarzadeh@yahoo.com

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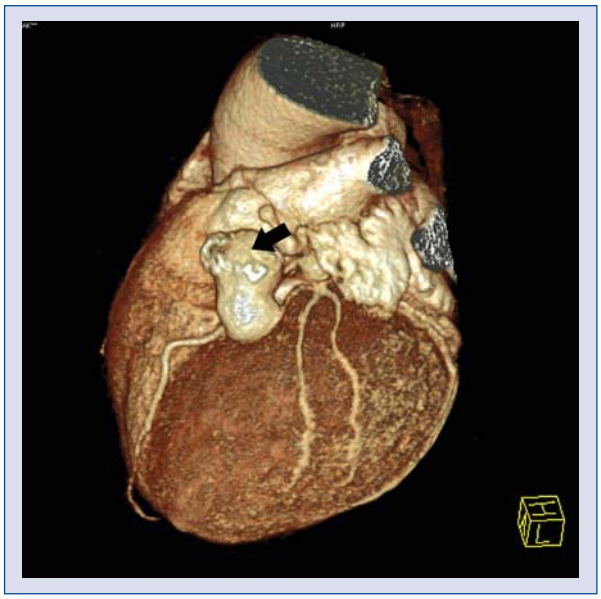

Figure 2. Volume rendered multi-slice computed tomographic image showing the calcified mass over heart contour.

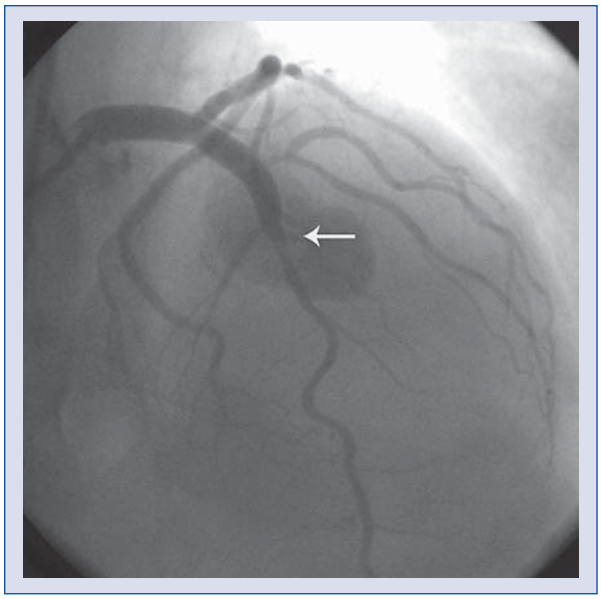

Figure 4. Coronary angiography image showing the mass filling via small rudimentary diagonal branch.

Figure 3. Multi-slice computed tomography angiography image showing the calcified mass with narrow stalk above left anterior descending artery.

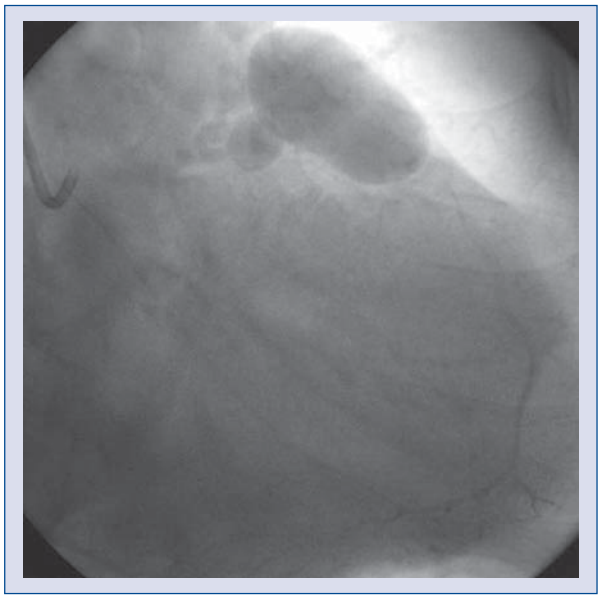

Figure 5. Image of aneurysmal sacs at the end of a forceful coronary injection showing the second small mass and the termination of fistula connection near the pulmonary artery.

end in pulmonary artery. A 3.5/19 covered stent (Jomed) was deployed in the LAD (covering the origin of the diagonal artery) and was redilated with a 4/10 balloon to ensure complete sealing. Control coronary angiography six months later showed patent stent with complete sealing of the fistula connection (Fig. 6).

Discussion

Congenital coronary arteriovenous fistula (CAVF) is a rare anomaly through which coronary blood flow is usually shunted into a cardiac chamber, great vessel, or other structures, bypassing the myocardial capillary network [1]. The first case of CAVF was reported by Krause in 1865 [2]. It is usually a congenital coronary anomaly, but rarely it is acquired secondary to chest trauma or iatroge-

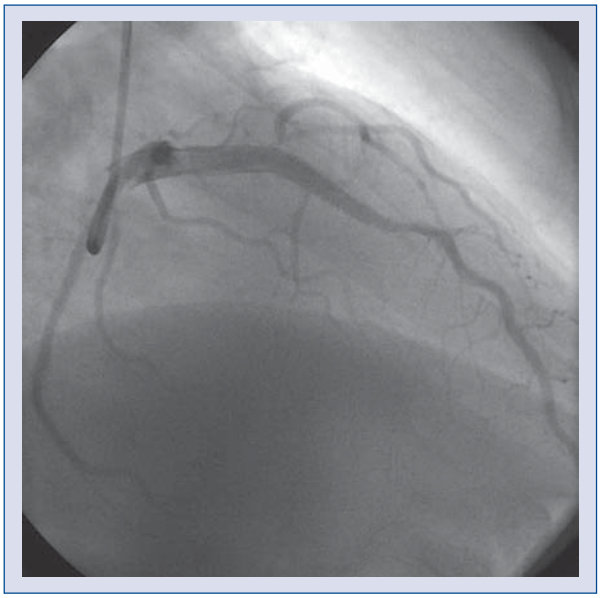

Figure 6. Control angiography image obtained six months after the index procedure showing the patent covered stent and complete sealing of the aneurysmal sac.

nic secondary to percutaneous interventions [3–5]. Coronary fistulas to pulmonary artery have been reported in Takayasu's arteritis [6]. Congenital CAVF is reported in about 0.2% of routine coronary angiograms [7, 8]. The right coronary artery (or its branches) is the commonest site of origin of CAVFs (constituting 55% of cases) and the second commonest site is the left coronary artery (about 35% of cases) [9]. Low pressure heart chambers or great vessels are common drainage sites of these fistulas. Fistulous drainage occurs into the right ventricle in 41%, right atrium in 26%, pulmonary artery in 17%, left ventricle in 3%, coronary sinus in 7% and superior vena cava in 1% [10]. Thus, a left to right shunt is present in about 90% of cases. Fistulous dilatation is usually uniform, but aneurysm formation is reported in around one in five cases [8].

Most patients with CAVFs are asymptomatic [11]. The symptoms of CAVF vary depending on the patient's age, the site of origin of the fistula, size, the presence of any associated aneurysm and the drainage site of the fistula. CAVFs tend to grow over time [12]. Large left to right shunt of these fistulas may cause pulmonary hypertension or high output heart failure and myocardial ischemia [13, 14]. Myocardial ischemia/infarction can occur from reduced coronary blood flow distal to the fistula and has been seen in patients with coronary fistulas without evidence of coronary atherosclerosis [13–15]. Occasionally, incidental finding of a continuous murmur, characteristically heard over the left sternal border and at the apex, leads to the diagnosis of CAVF [9]. Aneurysmal CAVFs may have a compressive effect on neighboring cardiovascular structures. Thrombosis inside this aneurysm may lead to acute ischemic events. Rupture of this aneurysm is rare and usually occurs in large giant aneurysms [7]. It is a catastrophic event and may present with chest pain, pericardial effusion, cardiac tamponade or even sudden cardiac death and could mimic the presentation of acute aortic dissection [7, 11, 16, 17]. The frequency of this complication, as with the frequency of previously described symptoms, increases over time [16].

Two-dimensional echocardiography is valuable in the diagnosis of fistula, but transesophageal echocardiography is complementary. However, echocardiography is operator-dependent and limited by the availability of a good acoustic window, and it may not identify the complete course of the fistula [18].

Selective coronary angiography is the best way to image a coronary artery aneurysm and fistula [13]. But it is invasive, and the entire course of the vessel may be difficult to define due to its complicated three-dimensional geometry, shown in two dimensions fluoroscopically [19]. MSCT may be better than catheter angiography because of its ability to show the fistula separately from the adjacent cardiovascular structures, along with any aneurysm or obstruction along its course [18]. Utsunomiya et al. [19] proposed that MSCT imaging should be considered the technique of choice for both diagnosis and follow-up examination of a coronary anomaly.

The natural history of coronary artery fistulas varies, and spontaneous closure secondary to spontaneous thrombosis, although uncommon, has been reported [20, 21]. The management is controversial and recommendations are based on anecdotal cases or small retrospective series [9]. Prophylactic precautions of subacute bacterial endocarditis are recommended, as bacterial endocarditis is a recognized complication [9]. Antiplatelet therapy is recommended, especially in patients with distal coronary artery fistulas and abnormally dilated coronary arteries [9]. It is widely accepted that fistula closure should be considered in symptomatic patients with heart failure, myocardial ischemia or high shunt ratios to prevent further complications, but it remains controversial in asymptomatic patients [8, 13, 22]. Some observations of progressive enlargement during the follow-up period are reported [23, 24]. Ito et al. [16] reported a case of CAVF with significant enlargement and rupture of aneu-

rysm in a short time course (three years). Therefore some authors recommend fistula closure be considered in CAVFs associated with a large aneurysm so as to prevent later complications [11]. Considering the rather benign natural history of small CAVFs, a conservative approach may be favored in such cases. If these small fistulas are associated with aneurysmal dilatation, a strategy of close observation and early surgical or interventional closure in those with progressive dilatation during follow-up might be considered [16]. Percutaneous closure is another alternative to surgery with good reported success [14, 22, 25]. Catheter closure can be performed with a variety of techniques, including detachable balloons, stainless steel coils, controlled-release coils, controlled-release patent ductus arteriosus coils, patent ductus arteriosus plugs, regular and covered stents, and various chemicals [14, 22, 25]. Most fistulas treated with catheter intervention were occluded with coils [9]. Results from the transcatheter and surgical literature show that both approaches have similar early effectiveness, morbidity, and mortality [22]. There are few reports of CAVF closure using covered stents [14, 26]. Although technically it is a relatively easy procedure, it should be emphasized that post-dilatation is generally necessary after deployment of covered stents to ensure complete sealing of the fistula and to reduce the possibility of restenosis.

Conclusions

Our case represents an incidental finding of a CAVF in an adult patient with associated aneurysm measuring more than 4 cm in its major diameter. It emphasizes the role of MSCT in diagnosing and delineating the origin and course of fistula. Although surgery or coil embolization are potential alternatives, given our experience with covered stents we decided to proceed with the latter option. Control angiography six months after the initial procedure showed no evidence of restenosis in our patient. After 15 months follow-up, the patient remains well and symptom-free.

Acknowledgements

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