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INTERMEDIATE

CASE REPORT: CLINICAL CASE

Coronary Artery Aneurysm After Drug-Eluting Stent Implantation Causing Coronary-Bronchial Fistula

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ABSTRACT

Coronary artery aneurysm (CAA) after drug-eluting stent implantation is rare, with a reported incidence of 0.3% to 6.0%. Most of these aneurysms are asymptomatic. Hemoptysis as a presentation of CAA is very rare. The patient in our case had CAA after zotarolimus-eluting stent implantation and presented with hemoptysis resulting from a leaking coronary-bronchial fistula. (**Level of Difficulty: Intermediate.**) (J Am Coll Cardiol Case Rep 2020;2:1692–7) © 2020 The Authors. Published by Elsevier on behalf of the American College of Cardiology Foundation. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/).

HISTORY OF PRESENTATION

A 42-year old man presented to the Base Hospital Delhi Cantt, New Delhi, with a 3-day history of cough, hemoptysis, pain in the left lower chest, and generalized weakness, 8 weeks after he underwent left main (LM) coronary artery bifurcation percutaneous coronary intervention (PCI). On admission, the patient's vital signs were as follows: pulse rate, 84 beats/min; blood pressure, 134/88 mm Hg; and respiratory rate, 16 breaths/min. The patient was afebrile on presentation. The general and systemic examinations were unremarkable.

LEARNING OBJECTIVES

- Consider leaking CAA as a differential diagnosis in patients with hemoptysis after DES implantation, so that proper diagnostic and management steps can be taken.
- In patients with CAA as a differential diagnosis, early CT coronary angiography must be planned to identify this serious complication in a timely manner.
- Plan early coronary angiography followed by definitive management in case of a CT angiography result indicative of leaking CAA, so that critical lifesaving time is not lost.

PAST MEDICAL HISTORY

Eight weeks before the current presentation, the patient initially presented to the hospital with anginal chest pain of 10 days' duration. The electrocardiogram showed ST-segment elevation, T-wave inversion, and poor progression of the R-wave in the anterior chest leads. A 2-dimensional echocardiogram showed left anterior descending (LAD) coronary artery territory hypokinesia with preserved thickness and a 30% ejection fraction. Coronary angiography

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The authors attest they are in compliance with human studies committees and animal welfare regulations of the authors' institutions and Food and Drug Administration guidelines, including patient consent where appropriate. For more information, visit the *JACC: Case Reports* author instructions page.

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showed a dominant, normal right coronary artery, LM distal plaque, LAD osteoproximal plaque followed by thrombotic cutoff, and a nondominant left circumflex (LCX) artery having osteoproximal 60% to 70% stenosis (**Figures 1A and 1B**, Video 1). He was scheduled to undergo PCI on the LAD through the right radial route. PCI was performed using a 6-F extra back-up guiding catheter. However, the LM artery was dissected with compromised flow to both the LAD and the LCX. The patient became hypotensive, with ongoing angina. Bailout LM bifurcation PCI was performed with the TAP (T and Protrusion) by using Resolute Onyx stents (Zotarolimus-Eluting Coronary Stent System, Medtronic, Minneapolis, Minnesota), 4 × 22 mm from LM to proximal LAD, 3 × 22 mm from proximal to mid-LAD, 2.75 × 22 mm in mid-LAD, 2.75 × 18-mm in osteoproximal LCX, and 2.5 × 18 from LCX to the obtuse marginal major overlapping previous stent. All stents were deployed at 12 atm. Final kissing balloon inflation was done with a 3.5 × 15 mm noncompliant (NC) balloon in the LM to LAD and a 3 × 15 mm NC balloon in the LM to LCX at 10 atm, and the proximal optimization technique in the LM was performed with a 4.5 × 8 mm NC balloon at 20 atm. The patient became angina free and normotensive, with Thrombolysis In Myocardial Infarction (TIMI) flow grade 3 in the left coronary system (**Figures 2A and 2B**, Video 2). The patient was discharged after 4 days on dual antiplatelet therapy consisting of Ecosprin (a proprietary formulation containing acetyl salicylic acid) and clopidogrel, and he was asymptomatic until his presenting symptoms developed.

DIFFERENTIAL DIAGNOSIS

The differential diagnosis included lung parenchymal infection (bacterial or tubercular), a noninfective lung parenchymal lesion such as a tumor, pulmonary embolism, and coronary artery aneurysm (CAA).

INVESTIGATIONS

The patient's hematologic and biochemical parameters, as well as his chest radiograph, were normal. A complete blood count showed no leukocytosis, and blood culture showed no growth of organisms. A 2-dimensional echocardiogram showed LAD territory hypokinesia and a 45% ejection fraction. Contrast-enhanced computed tomography (CT) of the chest showed a proximal LCX aneurysm compressing the atrioventricular (AV) junction, a left atrial (LA) and left ventricular (LV) lateral wall intramural hematoma (**Figures 3A to 3C**), and adjacent lung parenchymal consolidation.

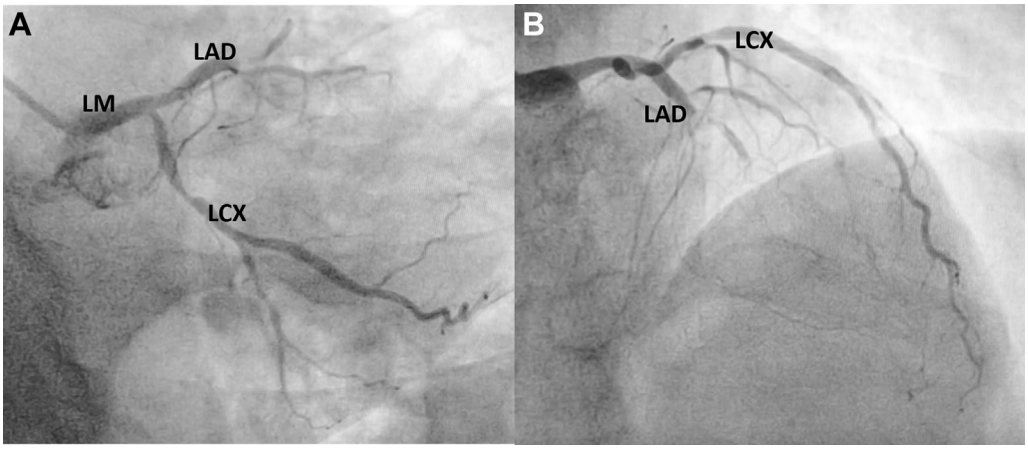
MANAGEMENT

The patient was started on intravenous antibiotics, which resulted in resolution of hemoptysis. Coronary angiography was planned; however, before the procedure, the patient had an episode of massive

ABBREVIATIONS AND ACRONYMS

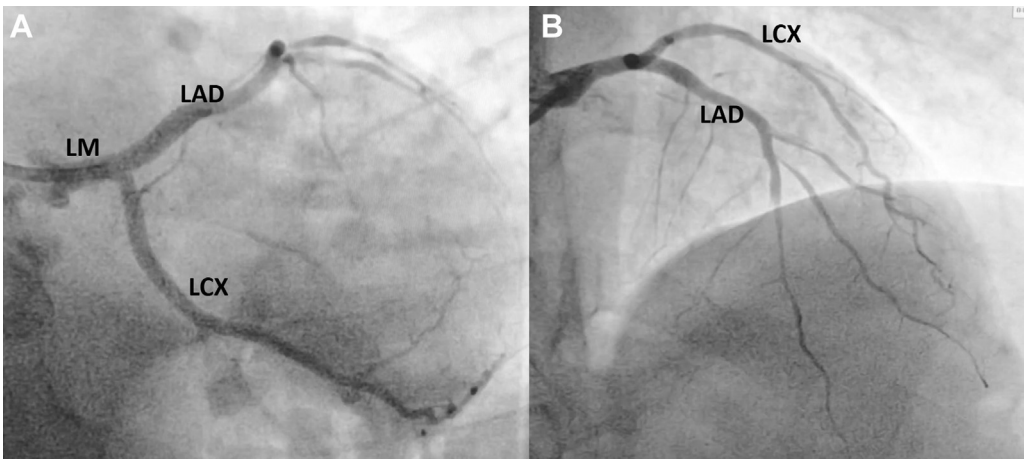
BMS = bare metal stent(s)
CAA = coronary artery aneurysm
CT = computed tomography
DES = drug-eluting stent(s)
ECG = electrocardiogram
LA = left atrial
LAD = left anterior descending
LCX = left circumflex
LM = left main (coronary artery)
LV = left ventricular
NC = noncompliant
PCI = percutaneous coronary intervention

FIGURE 1 Coronary Angiography at Initial Presentation



(A and B) Coronary angiography showing diseased left main (LM), left anterior descending (LAD) and left circumflex (LCX) arteries. There is thrombotic cutoff at the mid-left anterior descending artery and 60% to 70% stenosis in the osteoproximal left circumflex artery.

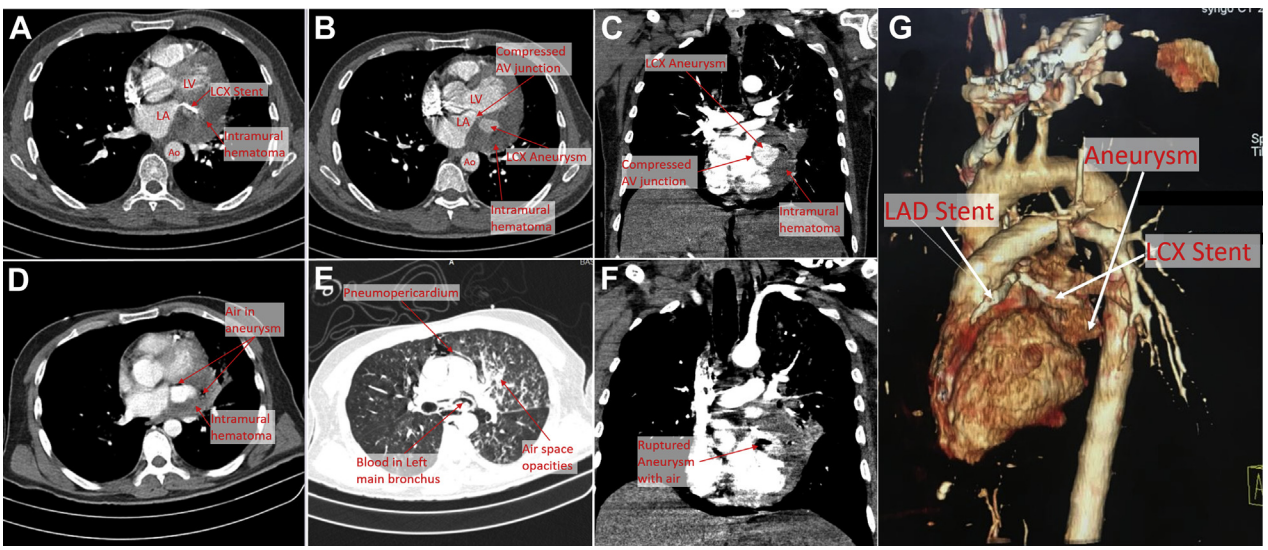
FIGURE 2 Coronary Angiography



(A and B) Coronary angiography after left main (LM) bifurcation angioplasty. Abbreviations as in Figure 1.

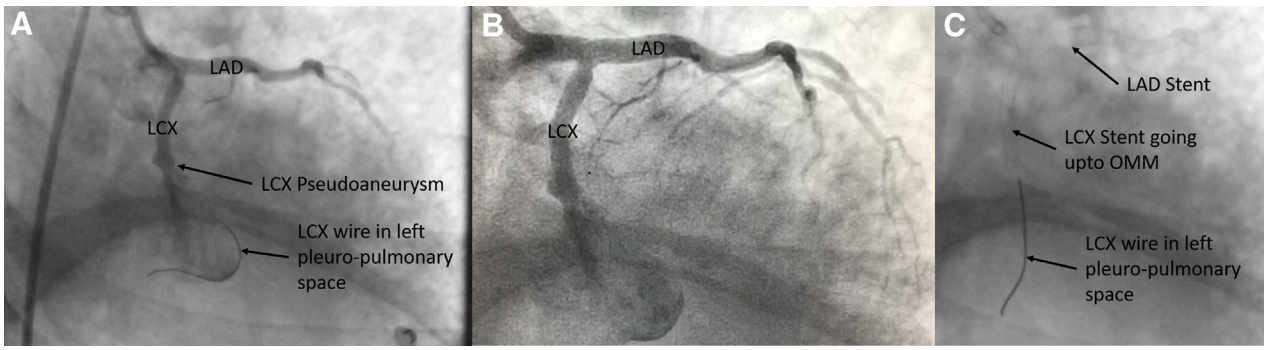
hemoptysis and cardiopulmonary arrest. He was revived after 10 min of cardiopulmonary resuscitation. Repeat contrast-enhanced CT of the chest showed air in the ruptured LCX aneurysm and a LA-LV lateral wall intramural hematoma, pneumopericardium, blood in left main bronchus, and air space opacities (Figures 3D to 3F). He was taken for emergency coronary angiography, which showed a

FIGURE 3 Computed Tomography Angiography



Computed tomography angiography of the thorax and mediastinal window showing **(A)** a left circumflex (LCX) artery stent in the axial view with an adjacent aneurysm and a large left atrial (LA)-left ventricular (LV) lateral wall intramural hematoma compressing the left atrioventricular (AV) junction in the **(B)** axial and **(C)** coronal views. Computed tomography angiography on the day of massive hemoptysis shows the **(D)** presence of air within the aneurysm and **(F)** a left atrial–left ventricular lateral wall intramural hematoma. **(E)** Lung window showing pneumopericardium, a soft tissue density in the left main bronchus suggestive of blood and air space opacities resembling alveolar hemorrhage. These findings were suggestive of ruptured left circumflex aneurysm and its communication with the left main bronchus and left lung. **(G)** A 3-dimensional reconstruction image showing a left circumflex aneurysm as well as a left circumflex stent. Ao = aorta; LAD = left anterior descending artery.

FIGURE 4 Coronary Angiography After Rupture of LCX Aneurysm



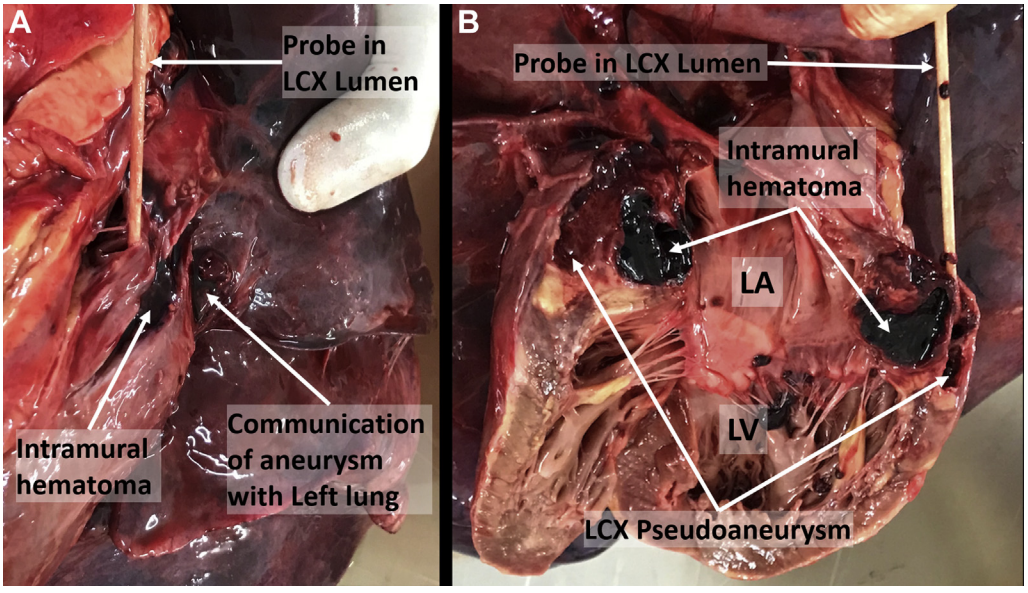
(A and B) Coronary angiography showing a leaking left circumflex (LCX) aneurysm. **(C)** Wire could not be negotiated into the distal LCX artery and instead went into the left pleuropulmonary space. LAD = left anterior descending artery.

ruptured fusiform aneurysm in the proximal LCX artery that was bleeding into the left pleuropulmonary space (**Figures 4A and 4B**, Video 3). The LM artery was hooked with a 7-F extra back-up catheter, both LAD and LCX were wired, and a plan was made to place a covered stent across the bleeding aneurysm. However, because of high flow from the ruptured site, the LCX wire went into the pleuropulmonary space (**Figure 4C**, Video 4) and could not be negotiated to the distal LCX. Because of continuous bleeding, the patient was hemodynamically unstable, and a 4 × 19 mm covered stent (Graftmaster, Abbott Vascular, Santa Clara, California) was placed from the LM to the LAD across e LCX, thereby controlling the LCX leak. The patient's hemodynamic status improved after the intervention. Thereafter, the patient developed generalized tonic-clonic seizures secondary to hypoxic-ischemic encephalopathy, bleeding gastric ulcers, ventilator-associated pneumonia, and right lung collapse, and he was treated with intravenous antiepileptic agents, endoscopic hemoclips, upgraded antibiotics, and bronchoscopy, respectively. The patient remained critically ill, with ongoing hypoxic-ischemic encephalopathy, a poor Glasgow Coma Scale score, ventilator-associated pneumonia, and sepsis. Five days post-aneurysm rupture, the patient had a sudden cardiac arrest and died of his illness. Autopsy revealed a 1.5 × 1.5 cm pseudoaneurysm around the proximal LCX stent with an adjacent 5.2 × 5.8 cm intramural hematoma in the LA and LV lateral wall compressing the left AV junction (**Figures 5A and 5B**). The ruptured aneurysm was also seen to be communicating with left main bronchus and left lung parenchyma through a fistula between the wall of the left atrium and the lower lobe of the left lung. The LCX artery showed overlapping in situ metallic stents in its lumen with aneurysmal dilation. The histopathology report found sections from the LA and LV walls showing intramural hematoma. Sections of the LA and LV walls had features of old myocardial infarction with granulation tissue and fibrosis, with splaying of cardiac myocytes. Sections from the LCX artery showed a dilated lumen with an atheroma within the wall of the coronary artery. There was disruption of the tunica intima and underlying media with adherent thrombus suggestive of a pseudoaneurysm. The absence of significant inflammation of the vessel wall ruled out the presence of a mycotic aneurysm.

DISCUSSION

CAAs after PCI are rare (incidence, 0.3% to 6.0%), and most of these lesions are pseudoaneurysms (1–3). CAA after PCI has been reported with bare-metal stents (BMS), as well as with drug-eluting stents (DES) (2). Mechanisms of CAA after PCI include coronary dissection and medial injury resulting from the use of oversized balloons or stents, high-pressure balloon inflation, atherectomy, and laser angioplasty (1–3). CAA is seen more commonly with DES as compared with BMS (2). DESs elute antiproliferative drugs to prevent restenosis; however, this may also delay coronary healing. In addition to this mechanism, medial inflammation secondary to a hypersensitivity reaction to the drug, polymer, or stent platform may also predispose patients to CAA formation after DES implantation (4–6). This complication is more frequently seen after bailout or complex coronary procedures. In this case, a pseudoaneurysm

FIGURE 5 Autopsy Specimen



(A and B) Autopsy specimen showing a ruptured left circumflex (LCX) pseudoaneurysm with an adjacent left atrial (LA)–left ventricular (LV) lateral wall intramural hematoma.

developed across the LCX stent after bailout LM bifurcation PCI with a zotarolimus-eluting stent. In addition, the possibility of extension of the LM dissection to the LCX, as well as balloon dilatation or coronary wire-induced medial injury and intimal dissection, could not be ruled out. These mechanisms, in conjunction with the antiproliferative action of the DES, could explain the formation of the pseudoaneurysm. Although most CAAs are asymptomatic, presentation secondary to ischemia caused by aneurysmal thrombosis or distal embolization can occur (7,8), and rupture of CAAs, albeit rare, may lead to ischemia and life-threatening tamponade (7,9,10). Hemoptysis as a presentation of CAA, as seen in this case, is rarely reported. The aneurysm across the LCX stent gradually eroded the pericardium, left pleural membrane, and left lung parenchyma to communicate with airways and leading to hemoptysis. Rupture of the CAA led to massive hemoptysis, hemopneumopericardium, pneumomediastinum, left hemopneumothorax, and air space hemorrhage. Management of CAAs depends on presentation, rate of expansion, size, and evidence of infectivity. Asymptomatic, small CAAs can be managed medically with dual antiplatelet agents (8). Percutaneous interventions for symptomatic aneurysms include covered stent implantation (as done in this case), as well as balloon or stent-assisted aneurysmal coil embolization (8). Giant CAAs (>20 mm), CAAs involving the LM artery, and infective aneurysms require surgical excision and coronary artery bypass (2,8). This report was approved by the institutional ethical committee of Base Hospital Delhi Cantt, New Delhi.

CONCLUSIONS

Although CAAs are relatively rare complications of PCI, they can have disastrous complications. The presence of atypical presentations such as hemoptysis in a post-PCI patient should not preclude this diagnosis, especially in patients who underwent complicated or bailout PCI. Hence, in such patients, CAAs should be kept in the differential diagnosis. Early CT angiography must be planned because the presence of an aneurysm warrants urgent coronary angiography and definitive management.

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KEY WORDS complication, coronary angiography, percutaneous coronary intervention

APPENDIX For supplemental videos, please see the online version of this paper.