CASE REPORT

Anomalous left anterior descending artery arising from the pulmonary trunk: a rare cause of angina

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SUMMARY

A 60-year-old man with a medical history of hypertension and dyslipidaemia presented to our rapid access chest pain clinic with a 2-month history of chest pain on exertion. An exercise stress test was arranged, which showed electrical evidence of inducible ischaemia. Subsequently, a coronary angiogram revealed an anomalous left anterior descending artery arising from the main pulmonary artery that received grade 3 collaterals from a large right coronary artery arising from the aorta. The circumflex arises from the right coronary sinus with a retroaortic course and provided collaterals to the anomalous left anterior descending artery. The patient was managed medically with the recommended pharmacological measures for stable angina and responded well with complete resolution of his symptoms, and he is currently under regular follow-up in the cardiology outpatient department.

BACKGROUND

Coronary artery anomalies (CAAs) are a rare but an important entity in the field of cardiovascular medicine. It is very important to recognise CAA as they lead to myocardial ischaemia and sudden cardiac death.^{1 2} Pulmonary artery (PA) origin of the left anterior descending (LAD) artery is an extremely rare anomaly with an estimated frequency of 0.0008%.³

Figure 1 Aortogram showing absence of a left coronary artery origin from the left coronary sinus (arrow).

CASE PRESENTATION

We report a case of a 60-year-old man with a medical history of dyslipidaemia and hypertension. His general practitioner referred him to our rapid access chest pain clinic with a 2-month history of left-sided chest pain radiating across the chest on moderate to severe exertion. He was an ex-smoker and had no family history of any heart disease. Clinical examination was unremarkable apart from an elevated blood pressure at 160/90 mm Hg.

INVESTIGATIONS

An ECG was performed that showed sinus rhythm with left ventricular hypertrophy, but there were no changes suggestive of ischaemia. We proceeded to an exercise stress test, where the patient reached stage 3 of the standard Bruce protocol without any symptoms. However, there were electrical changes consistent with myocardial ischaemia. We then performed an elective diagnostic cardiac catheterisation, which suggested the presence of an anomalous LAD artery arising from the main PA as there was no visible left coronary artery origin on an aortogram (figure 1). The large right coronary artery (RCA) provided grade 3 retrograde collaterals to the anomalous LAD (online supplementary video 1, figure 2). The circumflex artery appeared to arise from the right coronary sinus and also provided collaterals to the anomalous LAD (figure 3). A subsequent CT coronary angiogram (CTCA) confirmed the anomalous origin of the LAD from the main PA (figures 4, 5 and 6) and demonstrated the origin of the circumflex from the right coronary sinus with a retroaortic course (figure 7). Transthoracic echocardiography showed normal left ventricular function with left ventricular hypertrophy.

TREATMENT

The patient was managed medically with aspirin, statin, beta blocker and optimisation of his blood pressure medications.

OUTCOME AND FOLLOW-UP

The patient's symptoms were resolved, his blood pressure was optimally controlled and his exercise tolerance was greatly improved. He is currently under regular follow-up with the cardiology outpatient department. Because he was rendered asymptomatic, we decided to continue managing him medically and to consider surgery if symptoms recur.

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Figure 2 Coronary angiogram showing a large right coronary artery that provides collaterals (arrow) to the left anterior descending artery that arises from the pulmonary artery.

DISCUSSION

Anomalous pulmonary origin of a coronary artery is a rare but serious congenital cardiac anomaly. Mortality from this anomaly can be as high as 85% if untreated, and a majority of such cases die during childhood.⁴ Clinical manifestations are usually in the form of signs and symptoms of cardiac failure or sudden cardiac death in some patients. Patients usually present with the

Figure 3 Coronary angiogram showing anomalous left circumflex arising from the right coronary cusp (white arrow), with collaterals filling retrograde into the anomalous left anterior descending artery and pulmonary artery (blue arrow).

Figure 4 Multislice coronary CT showing the anomalous origin of the left anterior descending artery from the main pulmonary artery.

symptoms in the first year of life.⁵ Survivors to adulthood can be asymptomatic or manifest symptoms of myocardial ischaemia, and in some patients, mitral regurgitation, cardiomyopathy or arrhythmias can be a presenting feature.⁵

An increasing number of reports of adult patients with anomalous left coronary artery arising from pulmonary artery (ALCAPA) has been noted. This may be due to the development of collaterals from an enlarged RCA that supply the left myocardium. ALCAPA is extremely rare, but isolated anomalous left anterior descending coronary artery arising from pulmonary artery (ALADAPA) is even much rarer.⁶ Although considered to have a more benign course in comparison with ALCAPA, ALADAPA is a potential cause of cardiac failure, myocardial ischaemia and even sudden cardiac death in adults.⁷ Our patient remained asymptomatic until the age of 60 due to the presence of extensive collaterals from the RCA and left circumflex. We believe his symptoms appeared late in life probably due to atherosclerosis in the collateral vessels supplying his anomalous LAD. He also had

Figure 5 Multislice coronary CT showing the anomalous origin of the left anterior descending artery from the main pulmonary artery (arrow).

Figure 6 CT coronary angiogram 3D reconstruction showing the anomalous origin of the left anterior descending artery from the main pulmonary artery.

another coronary anomaly, that is, his left circumflex arose from the right coronary cusp. The latter was first described in 1933, and usually in this anomaly, the left circumflex courses posterior to the aorta and supplies the lateral wall of the left ventricle. This anomaly is relatively common with an estimated frequency of 0.32%–0.67% and is generally considered benign when there is no evidence of coronary atherosclerosis.¹⁸

Ismail *et al* reported a similar case of a 63-year-old man with ALADAPA who was treated successfully with surgical revascularisation.⁹ Another case of a 9-year-old boy who died suddenly

Figure 7 Multislice coronary CT showing the anomalous origin of the circumflex artery from the right coronary cusp.

after climbing numerous stairs was found to have ALADAPA after postmortem examination.¹⁰ The American Association for Thoracic Surgery published a review in which they reported 10 patients in their adulthood who were treated surgically for an anomalous coronary artery from the PA between 1960 and 2011. Only one patient had ALADAPA while ALCAPA was present in seven patients, and the remainder had anomalous RCA arising from the PA.¹¹

In the past, invasive coronary angiography and postmortem examination were the diagnostic tools for this anomaly. Recently, CTCA and cardiac MRI have become the preferred non-invasive modalities to visualise coronary anatomy and diagnose anomalies such as ALADAPA.¹² Pulsed-wave echo-Doppler can show a predominant systolic flow of the left coronary ostium and is regarded a characteristic for ALCAPA anomaly.¹³

The mainstay of treatment of this anomaly is considered to be surgical reimplantation of the anomalous coronary artery onto the aorta. Left internal mammary artery ligation to the proximal part of the anomalous artery is also considered an acceptable alternative but should be reserved for cases where transfer onto the aorta is not feasible.^{11 14} Medical management has been reported as an option in the absence of disabling symptoms and the presence of extensive collaterals to the anomalous vessel.¹⁵

In conclusion, we report an extremely rare case of ALADAPA associated with an anomalous circumflex arising from the right coronary cusp. Invasive angiography and CTCA are very useful in identifying these anomalies. Surgery is the mainstay of treatment and usually results in a significant improvement in symptoms and clinical course.

Learning points

- ▶ Anomalous left anterior descending coronary artery arising from pulmonary artery (ALADAPA) is an extremely rare but important cause of myocardial ischaemia in adults.
- ▶ Diagnosis of ALADAPA can be easily made during invasive cardiac catheterisation or CT coronary angiogram.
- ▶ Patients with this anomaly can remain asymptomatic until the development of angina in adulthood.
- ▶ Some patients can present with more than one coronary artery anomaly.

Contributors All authors made a significant contribution to this case report. AI conducted the design of the work, data collection and drafting the article. SPP organised the images and participated in preparation of the manuscript. PK conducted critical revision of the article and final approval of the version to be published.

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