CASE REPORT

Two cases of high-output heart failure as initial presentation of iliac arteriovenous fistula

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SUMMARY

We present two cases of females in their 40s presenting with biventricular heart failure being the consequence of a large arteriovenous fistula. Both patients had undergone abdominal surgery several years prior to the heart failure event with the initial finding of moderate pulmonary hypertension and high-output heart failure. CT revealed a large arteriovenous fistula between the common iliac artery and vein which subsequently was closed percutaneously.

BACKGROUND

Heart failure is a common disease, with an increased mortality and morbidity despite appropriate treatment. Identification of the causes potentially leading to heart failure is thus important. Arteriovenous malformations may lead to a significant shunting and consequently heart failure. It is thus important to consider if heart failure could be the cause of arteriovenous shunting. Although it is a rare cause, corrective measures may lead to dramatic improvement.

CASE PRESENTATION

Case 1

A 46-year-old obese (body mass index 37 kg/m²) woman, smoker for nearly 30 years, was referred to the outpatient clinic of a local hospital with shortness of breath and a new systolic cardiac murmur detected prior to an elective cervical disc prolapse surgery. The patient experienced mild palpitations in addition to increasing dyspnoea during the last 6 months and was found to be in New York Heart Association (NYHA) functional class 2. The patient presented with a normal clinical examination except for a cardiac murmur, described as a grade 2 holosystolic murmur above the aortic and pulmonic ostium with no diastolic components.

Case 2

A 48-year-old obese (body mass index 36 kg/m²) woman was referred to the outpatient clinic of a local hospital with shortness of breath and a new systolic cardiac murmur detected prior to an elective spinal stenosis surgery. The patient experienced chest pain in addition to increasing dyspnoea during the last 12 months and was found to be in NYHA functional class 2. The patient presented with a normal clinical examination except for moderate oedema of the lower extremities and a cardiac murmur, described as a grade 3 holosystolic

murmur above the aortic and pulmonic ostium with no diastolic components.

INVESTIGATIONS

Case 1

An ECG was performed which was normal, and the initial transthoracic echocardiographic (TTE) examination demonstrated normal left ventricular (LV) and right ventricular (RV) systolic function and a mild tricuspid regurgitation and estimated RV systolic pressure of 70 mm Hg, suggestive of severe pulmonary hypertension. As a consequence, the patient was further examined with a lung function test which demonstrated moderately reduced restrictive lung function with a forced expiratory volume in 1 s (FEV₁) of 1.8 L (65%), Tiffeneau Index 82%, carbon monoxide transfer factor (TL_{CO}) 105%, total lung capacity 74% and a saturation of 99%, thought to be related to the patient's obesity and with no possible explanation for the suspected pulmonary hypertension.

The patient was re-evaluated, and a new TTE examination was performed (figure 1) which demonstrated a mildly dilated LV (LV end-diastolic diameter 59 mm) with normal systolic and a grade 2 diastolic function (LV ejection fraction 50%–55%, E/A 1.8 deceleration time 170 ms e'_{sep} 9 cm/s E/e'_{sep} 14, left atrial volume index 130 mL) and increased stroke volume (112 mL) and cardiac output (9.5 L). The RV was moderately/severely dilated (RV end-diastolic diameter 48 mm), with normal systolic function (tricuspid annular plane systolic excursion (TAPSE) 28 mm) and a stroke volume of 108 mL. A moderate tricuspid regurgitation was demonstrated with a systolic gradient of 52 mm Hg with increased right atrial pressure (vena cava 31 mm and no collapse) suggestive of pulmonary hypertension although pulmonary acceleration time (218 ms) and non-invasively assessed pulmonary resistance¹ (1.6 Woods Units) were in the normal range. The TTE examination was thus most suggestive of biventricular volume overload, and the patient was re-evaluated to identify a potential extracardiac shunt. A murmur was noted in the left inguinal region, and an arteriovenous fistula was suspected. This suspicion was confirmed by a subsequent CT angiography (figure 2) which demonstrated a large arteriovenous fistula between the left common iliac artery and vein, thought to be the consequence of a hysterectomy performed in 2010.

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Figure 1 Transthoracic echocardiographic evaluation at the time of diagnosis demonstrating (A) dilated left ventricle of 59 mm, (B) increased right ventricular stroke volume with a normal acceleration time, (C) pulmonary hypertension with a tricuspid valve regurgitation gradient of 52 mm Hg and (D) dilated vena cava suggesting a high central venous pressure.

Case 2

An ECG was performed which was normal, and the initial TTE examination demonstrated normal LV and RV systolic function, mild dilatation of the RV and estimated RV systolic pressure of 75 mm Hg with severe dilation of the inferior vena cava suggestive of severe pulmonary hypertension. As a consequence, the patient was further examined with a lung function test that demonstrated reduced restrictive lung function with an FEV₁ of 2.8 L (99%), forced vital capacity (FVC) of 3.3 L (100%), Tiffeneau Index 85%. As a consequence of symptoms and echo findings, pulmonary embolism was suspected, and a thoracic CT scan and a pulmonary ventilation-perfusion scintigraphy were performed; both examinations were interpreted as normal although the CT scan corroborated the echo findings demonstrating a severe dilated inferior vena cava and mild dilatation of the RV. Pulmonary function was further examined with a TICO 121%, total lung capacity 100% and a saturation of 95% with no possible explanation for the suspected pulmonary hypertension.

The patient was referred to our centre for a right-heart catheterisation that confirmed moderate pulmonary hypertension with a pulmonary pressure of 67/27 mm Hg (mean pulmonary artery pressure (PAP) 42 mm Hg) with no reversibility on nitrous oxide inhalation. Central venous pressure (10 mm Hg), pulmonary capillary wedge pressure (14 mm Hg) and pulmonary resistance (1.9 Woods Units) were normal. The cardiac output was severely elevated at 14.5 L/min. Arterial blood gasses were analysed

for oxygen saturation from the aorta (96%), pulmonary artery (86%), right ventricle (87%), right atrium (84%), inferior vena cava (88%) and superior vena cava (70%), and as a consequence a left-right shunt was suspected. The patient was re-evaluated with a TTE with agitated saline infusion with no signs of a cardiac right-left shunting. The previous CT scan was re-examined, and an arteriovenous fistula was suspected, thought to be the consequence of a previous cholecystectomy performed in 2006. The patient underwent angiography that demonstrated a large shunt between the right common iliac artery and vein.

TREATMENT

Case 1

The patient was referred to the department of interventional radiology, and an endovascular closure of the fistula was thought to be the most appropriate option. The fistula was treated under local anaesthesia with the use of two Perclose ProGlide (Abbott) devices. The stent graft with diameter 16×16×93 Medtronic was deployed across the fistula and control angiography demonstrated a well-positioned stent graft with complete exclusion of the fistula (figures 3 and 4). Haemostasis was obtained without complications. Due to the magnitude of the shunt, it was decided to perform haemodynamic monitoring during the procedure. Before the procedure, the systemic pressure (217/88, mean arterial pressure 126 mm Hg), pulmonary pressure (63/39 mm Hg,

Figure 2 CT angiography demonstrating a large arteriovenous fistula between the left common iliac artery and vein.

Figure 3 Angiography demonstrating a well-positioned stent graft with complete exclusion of the fistula.

Figure 4 CT angiography demonstrating normalisation of the diameter of the iliac vessels and no sign of persistence of the fistula.

mean pulmonary artery pressure 48 mm Hg) and central venous pressure (37 mm Hg) were highly elevated. In addition, cardiac output (14.2 L) and mixed venous oxygen saturation (SVO₂) (84%) estimated by the thermodilution method were found elevated. Immediately after the procedure, central venous pressure (18 mm Hg) and cardiac output (8.0 L) dramatically decreased although pulmonary artery (69/31 mm Hg, mean pulmonary artery pressure 46 mm Hg) and systemic pressures remained elevated (192/94 mm Hg, mean artery pressure 127 mm Hg).

Case 2

The fistula was treated under local anaesthesia with the use of two preclosure ProGlide (Abbott) devices. A Medtronic stent graft with a diameter of 16×13×82 mm was deployed, and control angiography showed a successfully closed fistula.

OUTCOME AND FOLLOW-UP

Case 1

The patient underwent a clinical examination and a TTE examination the day after and 3 months after the procedure including CT control which showed normalised diameter of the iliac vessels and no sign of persistence of the fistula (figure 4). She experienced immediate symptom improvement and 3 months after the procedure, the patient was completely asymptomatic. This improvement was paralleled by changes in LV and RV geometry and haemodynamics. LV size (LV end-diastolic diameter 61 mm; 54 mm) and RV size (RV end-diastolic diameter 44 mm; 30 mm) showed no immediate changes, but normalised after 3 months while LV (79 mL; 60 mL) and RV (78 mL; 65 mL) stroke volume were immediately reduced after the procedure and further normalised after 3 months. Although LV ejection fraction remained in the normal range (55%), diastolic grade remained abnormal although improved from grade 2 (E/A 1.6, deceleration time 180 ms, e'_{sep} 3.6 cm/s, E/e' 34) the day after procedure, to grade 1 (E/A 0.8, deceleration time 215 ms, e'_{sep} 4.4 cm/s, E/e' 16) after 3 months. This improvement in diastolic function was the consequence of a reduction in LV preload demonstrated by

Figure 5 – Transthoracic echocardiographic evaluation after occlusion of the fistula demonstrating (A) reduction in left ventricle diameter to 55 mm, (B) normalisation of right ventricular stroke volume, (C) normalisation of pulmonary systolic pressure with a tricuspid valve regurgitation gradient of 22 mm Hg and (D) normalisation of vena cava suggesting a normalisation of central venous pressure.

a dramatic decrease in vena cava diameter (17 mm; and 13 mm) and reduction in LA volume index (137 mL; 48 mL). In addition, tricuspid gradient decreased and even normalised after 3 months (34 mm Hg; 22 mm Hg) (figure 5).

Case 2

The patients was examined 6 months after the procedure and demonstrated no signs of heart failure, normalisation of pulmonary blood pressure (34/10 mm Hg) and complete resolution of symptoms.

DISCUSSION

The haemodynamic consequence of extracardiac arteriovenous fistulas is the shunting of oxygenated blood from the arterial system to the venous system, depriving oxygen extraction from the peripheral tissue and leading to limb hypoperfusion. In order to maintain an adequate peripheral perfusion, the heart will adapt by dilating and increasing cardiac output and eventually leading to a condition of biventricular volume overload. The increased flow across the heart valves may lead to systolic murmurs, and transvalvular flow velocities may increase solely as the consequence of increased flow rather than to obstruction. Particularly, the increased gradients through the pulmonary valve may lead to increased RV pressures mimicking pulmonary hypertension due to pulmonary disease. In our case, the patient presented solely with shortness of breath, and no symptoms of limb ischaemia, and consequently, a pulmonary cause was suspected initially. The occurrence of high tricuspid gradients in the setting of increased RV stroke volume and normal acceleration time in the pulmonary artery flow should raise the suspicion of a shunt with volume load of the RV. This can further be examined by estimating the pulmonary artery vascular resistance which can be estimated and non-invasively approximated by echocardiography ($10 \times V_{TR} / VTI_{RVOT}$).¹ When this condition

additionally is accompanied by increased LV stroke volume, an extracardiac shunt should be suspected. Although in the first case one could expect the arteriovenous fistula to be a complication to the previous coronary angiography, arteriovenous fistulas is a rare complication occurring in 0.1%–0.4% depending on the use of a vascular closing device,² and most arteriovenous fistulas would include the femoral artery rather than the iliac artery. Another possibility could be that the fistula could be the consequence of prior abdominal surgery. A minor vascular injury of the iliac artery during surgery may lead to a small communication between the artery and vein that eventually may lead to a large arteriovenous fistula with time. In a larger registry with nearly 80 000 women undergoing hysterectomy, 0.3% experienced circulatory complications although the paper did not account for numbers experiencing arteriovenous fistula.³ However, the association has been described in a previous case report with a woman presenting with a pelvic arteriovenous fistula after hysterectomy.⁴

Learning points

- ▶ In a case of heart failure and pulmonary hypertension with no obvious explanation, arteriovenous fistula should be considered.
- ▶ This is particularly true when biventricular heart failure with high cardiac output is present. In addition to echocardiography, right-sided heart catheterisation may help clearing the cause of high-output heart failure and may raise the suspicion of a non-cardiac shunt.
- ▶ Arteriovenous fistula is rare but may occur in patients with history of surgical and/or interventional procedures.

Contributors All four authors have contributed significantly to this paper and fulfil the criteria for being coauthor. All authors have read and approved the final manuscript. JSD has contributed in the entire manuscript, in the conception and design of the paper and has performed all echocardiographic examinations. CA has contributed in the revision of the entire manuscript, has performed the right-heart catheterisation during the procedures and has helped interpreting data. SD has contributed in the revision of the entire manuscript, has provided CT scans before and after the procedure, has performed the closure of the fistulas and has equally helped interpreting data. JEM has contributed in the revision of the entire manuscript, has performed the diagnostic right-sided heart catheterisation and has provided insightful comments about the interpretation of all acquired data.

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