Aorto-cavitary Fistula: A Complication of Infective Endocarditis
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ABSTRACT

Aorto-cavitary fistulae are rare complications of infective endocarditis. The diagnosis, in the absence of concomitant aortic valve disease, replacement of aortic valve with homograft or prosthetic valve, periannular abscess and negative blood culture, requires a high index of suspicion and has important prognostic and management significance. The sensitivity of the Modified Duke Criteria is challenged in this case report with a documented right sinus of valsalva fistula to the right ventricle seen on transthoracic echocardiography.

Keywords: Aorto-cavitary, Duke criteria, fistula, infective endocarditis

INTRODUCTION

Aorto-cavitary fistulae were initially described in necropsy specimens (1). A rare complication of infectious endocarditis with Staphylococi aureus being documented as the most common cause has been reported on both sides of the Atlantic in necropsy and retrospective studies (1–4). Aorto-cavitary fistulae have been documented to occur from all three aortic valve sinuses to all four cardiac chambers with no preponderance from any specific aortic sinus to a specific cavity, with predominantly left to right shunts (1–5).

The incidence of infective endocarditis has remained the same despite the advent of antibiotics with increasing efficacy and still requires a high index of suspicion (6–9). Infective endocarditis involving the adjacent structure to the aortic root with extension into the aortic valve and subsequent formation of periannular abscess and erosion of the sinus of valsalva has been the theory proposed by some as the method of development of the fistulous communication between the aorta and the cardiac chambers (1–7). There is a greater likelihood of a ventricular septal defect occurring at the same time (2). The infecting organism in decreasing frequency noted in both necropsy patients and retrospective studies has been staphylococcus, streptococcus, pneumococcus, enterococcus, and other bacterium and fungal infections. Negative blood cultures do not make infective endocarditis less likely, as prophylactic antibiotics given consistently prior to cardiac surgery or for any other cause before considering a diagnosis of infective endocarditis, can cause such a result after the 1st, 2nd and 3rd week at 36%, 79% and 100%, respectively (2–9).

The Modified Duke Criteria was used to determine the diagnosis of infective endocarditis (10). Two-dimensional transthoracic echocardiograms with colour Doppler were used to confirm discrete masses as vegetations and direction of flow of the aorto-cavitary fistula, respectively.
The aorto-cavitary fistula was identified as an abnormal communication between the aorta and the right ventricle with high velocity, turbulent and continuous flow. Interventricular (VSD) flow and aortic regurgitation were measured using published, internationally accepted standard. The recommended standard for antibiotic prophylaxis, therapy and sensitivity of organisms identified was used for antimicrobial therapy (7–9, 11). The data recorded in the patient’s medical records were used to follow the patient’s progress and development of complications.

CASE REPORT
A 16-year old female, who is known to have tetralogy of Fallot, was symptomatic with recurrent hypercyanotic episodes associated with syncope, chest pain and breathlessness on mild exertion (New York Heart Association [NYHA] III–IV) immediately prior to corrective surgery. Repeated phlebotomy was required for polycythaemia. Significant findings on examination were plethoric mucous membranes, central cyanosis, marked clubbing with intermittent episodes of cardiopulmonary distress secondary to hypercyanotic spells. Her weight was 45 kg and height 117 cm. Oxygen saturation on air was 51% and saturation on 15 litres of oxygen was 76%, while the respiratory rate in between episodes of respiratory distress was 20/minute; the pulse rate was 100/minute, and blood pressure was 92/55 mm Hg.

Cardiovascular examination revealed a parasternal heave, single 2nd heart sound and a long ejection systolic murmur 3/6 at the upper left sternal edge. There were no signs of right or left heart failure. She was only on propranolol. Haematology results prior to surgery showed: HB 18.6 g/dL, PCV 56%. Electrocardiogram showed right axis deviation and right ventricular hypertrophy. Chest X-ray showed normal cardiothoracic ratio with oligoemic lung fields.

Transthoracic echocardiographic findings were consistent with a diagnosis of tetralogy of Fallot with right ventricular outlet obstruction at infundibular, valvar and supravalvar level and a gradient of 84 mmHg across a dysplastic pulmonary valve. Relatively small, but confluent pulmonary arteries were seen. Perimembranous ventricular septal defect (VSD) with right to left shunt was present and there was normal origin and course of both coronary arteries.

Cardiac catheterization confirmed the transthoracic echocardiographic findings. Right ventricular pressures were systemic with good biventricular function. Single VSD, normal coronary arteries and confluent pulmonary arteries, confirming good anatomy for biventricular repair, was noted. In order to prevent hypercyanotic spells, prophylactic propranolol was used and no attempt was made to cross the right ventricular outflow tract.

Cardiothoracic surgical correction following standard procedure and use of prophylactic antibiotics was successful with saturation of 100% postoperatively. She was extubated within 24 hours. She subsequently had a prolonged admission requiring re-intubation on day four, because of chest infection associated with tachycardia. Her temperature remained normal and saturations were maintained above 96%. Repeat transthoracic echocardiograms confirmed clinical signs of a good repair with no vegetation or dehiscence of VSD patch. Repeated blood cultures and other cultures were negative. There were persistent lung signs, with normal neurological, renal and cardiac function. There were no signs of cardiac failure.

On day 18 post surgery, multiple resistant acinetobacter was noted in the sputum with a spike in temperature and an increase in her white blood cell count which started to decrease with appropriate changes in antibiotics within 48 hours.

On day 32, she was returned to the ward post-extubation. Her saturation was noted to be 77% on air and 100% saturation in three litres of oxygen. A cardiac murmur was noted. Repeat transthoracic echocardiogram showed right ventricular to aortic shunt with predominant right to left shunt indicating suprasystemic right ventricular pressures (Figs. 1 and 2). Moderate aortic regurgitation was noted (Fig. 2). There was dehiscence of the upper and lower end of the VSD patch. There was a mobile vegetation on the right ventricular aspect of the VSD patch. Ventricular function was preserved and still normal. There were no perianular abscesses.

Surgical re-intervention to correct defects noted on the echocardiogram was done on day 60 post surgery, when she was considered stable, but she developed uncontrolled, intractable congestive cardiac failure. She had poor cardiac output despite maximum inotropic support and died two days post re-intervention.
The significant clinical findings associated with annular lesions and has more extensive peri-valvular tissue native aortic valves complicated predominantly with peri-valvular abscesses by two-dimensional echocardiography. J Am Coll Cardiol 2006; 49: 1254–60.

The sensitivity of the Modified Duke Criteria was challenged in this patient. At no point during the patient’s postoperative period did she satisfy the original Duke or Modified Duke Criteria for the diagnosis of infective endocarditis. On day 32 post-operatively when the vegetation was noted, there was one major criterion only. Using the Modified Jones criteria, she needed one minor criterion as well for the diagnosis of infective endocarditis to be “possible” (6–9). The use of prophylactic antibiotics affected the likelihood of having a positive blood culture where it has been shown that three weeks post use of antibiotics, negative blood cultures occur at 100%. The difficulty with diagnosis of infective endocarditis with the Modified Jones Criteria has been substantiated with case reports by Li et al (10).

The original Duke Criteria in 1994 was initially designed to help with clinical research, to encourage and facilitate prospective comparison of features of patients with infective endocarditis. This criteria has, however, been used worldwide to make or substantiate the diagnosis of infective endocarditis, making its use to do same, difficult. The Modified Duke Criteria has added specificity and sensitivity, which correlates to approximately 80% by some authors (7–10). Careful and accurate clinical assessment with a high index of suspicion is still required for the diagnosis of infective endocarditis, the incidence of which has not decreased over the past few decades. Use of clinical judgement and modern technology since the advent of transthoracic endocardiogram (TTE), which the Duke Criteria relied on, is important. The tool that should be used with much greater sensitivity and specificity, even with a negative TTE, is the transoesophageal echocardiogram.

The aorto-cavitary fistulae have been documented in native aortic valves complicated predominantly with perianular lesions and has more extensive peri-valvar tissue destruction. The significant clinical findings associated with the fistulous development are moderate to severe heart failure, renal failure and an increasing age, which in some studies constitute the major risk factors for death (2–6). Heart failure rates increase with the greater likelihood of development of third degree heart block as the Triangle of Koch where the atrioventricular node is located adjacent to the aortic valve root. In the case of the index patient, recurrence of the VSD was secondary to the dehiscence of the VSD patch following infection and extensive necrosis of the surrounding cardiac tissue (7–20). Other potential complications include paradoxical embolization, cerebral micro-bleeds, cerebral infarction and renal failure (2–8, 12–20).

Suprasystolic pressures secondary to an unresolved lung infection led to a right to left shunt occurring across the aorto-right ventricular fistulae as seen in Fig. 2.

Aorto-cavitary fistulae are rare complications of infective endocarditis and are associated with high morbidity and mortality with or without reintervention and require a high index of suspicion for diagnosis (1–20).


