SELECTED REPORTS

Aneurysm of the Left Sinus of Valsalva Draining into the Right Atrium*

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We report the case of an arteriovenous fistula connecting the aorta to the right atrium, due to a ruptured aneurysm of the left sinus of Valsalva. Clinical, echocardiographic, and angiographic aspects of this unusual lesion are discussed and correlated. Emphasis is put on the diagnostic value of the echocardiogram; a characteristic dense echo parallel to the posterior aortic wall is seen, along with signs of right ventricular volume overload. Combined with the case history and auscultatory finding (a continuous murmur on the right sternal edge), this may lead to the correct diagnosis by noninvasive means.

Aneurysms and fistulas of the sinuses of Valsalva are rare congenital lesions. This is particularly true for those originating from the left sinus of Valsalva. Although patients with this lesion are usually asymptomatic in childhood and adolescence, rupture may cause heart failure, sometimes of acute onset. Cardiac surgery nowadays allows successful repair of this lesion. An early diagnosis is therefore important, especially since clinical symptoms are not always conclusive.

Recently, the echocardiographic signs of aneurysms of both the right and left sinuses of Valsalva have been described. Since symptoms, clinical signs, and echocardiographic and angiographic findings differ markedly according to the site of the aneurysm and its hemodynamic consequences, we would like to report the features of a fistula connecting the left sinus of Valsalva to the right atrium.

CASE REPORT

The patient, a 16½-year-old Ethiopian boy, was first found to have a cardiac murmur at the age of 14 years. Two years later, he was hospitalized in Addis Ababa, Ethiopia, because of acute dyspnea and nonspecific pain in the chest. A diagnosis of patent ductus arteriosus complicated by subacute bacterial endocarditis was made, and the patient was transferred to the University Hospital of Geneva, Switzerland.

On admission, clinical examination revealed a boy in good health, with blood pressure of 130/70 mm Hg, a regular pulse rate of 60 beats per minute, and no signs of cardiac failure. A grade-3/6 continuous "machinery-type" murmur was audible maximally at the right sternal border and radiated towards the left sternal border and both clavicles.

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Figure 1. Echocardiogram demonstrating aneurysm (AN) of left sinus of Valsalva as echo parallel to and behind posterior aortic wall. RVOT, right ventricular outflow tract; AOV, aortic valve; AO, aorta; and LA, left atrium.

The electrocardiogram showed sinus rhythm, an axis of +30°, first-degree ativoventricular block, and left ventricular hypertrophy. The chest x-ray film revealed cardiomegaly and increased pulmonary vascular markings.

The echocardiogram showed increased right and left ventricular diameters and slight dilatation of the pulmonary artery and of the aorta; abnormal septal movement of type B was shown on the sweep. There was slight diastolic vibration of the left side of the septum and of the mitral valvular leaflets, especially the anterior leaflet. A constant band 12 mm in diameter was visible parallel to the posterior aortic wall, appearing at the transition from the anterior mitral valvular leaflet to the posterior aortic wall (Fig 1). Within the band, lines of echoes were seen moving parallel to the echoes of the aortic valve.

Cardiac catheterization revealed pressures at the upper limit of normal in the right side of the heart. The end-diastolic pressure of the left ventricle was raised after injection of contrast medium (116/20 mm Hg). Oximetric studies revealed a left-to-right shunt of 2.5 L/min and a ratio of pulmonary-to-systemic flow of 1.9. The aortic angiogram showed moderate aortic regurgitation. From the left sinus of Valsalva, which was dilated, just below the orifice of the left coronary artery, a fistulous conduit arose, which was 10 to 12 mm in diameter and drained into the right atrium (Fig 2 and 3). The conduit skirted the left atrium directly behind and slightly below the aortic cusps. The selective coronary arteriograms were normal.

At cardiac surgery a dilated left sinus of Valsalva was found, with a normal left coronary orifice. To the right and slightly below this orifice, another opening gave origin to a conduit 8 to 10 mm in diameter, through which a Nélaton's probe could be advanced into the right atrium. The fistula ran along the upper border of the left atrium. It was sectioned and sutured as close as possible to the sinus of Valsalva, as well as at its right atrial extremity.

Histologic examination of the resected segment, which had an external diameter of 13 mm with a wall that was 2 mm thick, showed disorganized smooth muscular fibers and a few elastic fibers separated by an accumulation of acid mucopolysaccharides. Intimal thickness was also increased.

The postoperative course was uneventful. Echocardio-
still not completely understood. A congenital weakness at the junction of the aortic media and the anulus fibrosum certainly contributes to the formation of an aneurysm.¹

In several review articles and case reports,¹,²,³,⁵,⁶ a predominance of male patients was found. Most frequently, the right aortic sinus is involved, and ruptures occur into the cavities of the right side of the heart. The left sinus of Valsalva is infrequently the site of formation of an aneurysm. One would expect such an aneurysm to rupture into an adjacent structure, such as the pulmonary artery, the left atrium, or the left ventricle, and not into the right atrium. An aneurysm of the left sinus of Valsalva draining into the right atrium is an extremely rare lesion, and some authors⁷ have doubted its existence altogether.

In the present case the fistula ran along the left atrium and behind the aorta; the normal left coronary artery originated adjacent to it. Several hypotheses can be advanced for the origin of this abnormality. First, it could represent an accessory left coronary artery communicating with the right atrium. Secondly, and less likely, the structure could be a false aneurysm resulting from an epicardial hematoma which ruptured secondarily into the right atrium.² Histologic findings do not suggest either of these hypotheses.

In our opinion the fistula is most likely an aneurysm of the left sinus of Valsalva, penetrating into the transverse sinus of the pericardium, approaching and finally rupturing into the right atrium when the boy was 14 years old. The histologic findings were compatible with this interpretation. The diagnosis may be suspected by the onset of symptoms late in childhood and by the clinical findings; a continuous murmur at the right sternal border is not likely to be due to a ductus, since isolated right patent ductus arteriosus is extremely rare.⁸ In the differential diagnosis a coronary arteriovenous fistula was excluded by selective coronary angiograms.

The echocardiogram was conclusive in this case. The presence of an echo parallel to the posterior margin of the aortic root which moves in the same direction and is accompanied by dilatation of the aortic root was first described by Nanda et al¹ as findings thought to be diagnostic of aortic root dissection; however, in a 16-year-old boy with no evidence of Marfan’s syndrome⁹ or congenital syphilis, dissection of the aorta is very unlikely, and an aneurysm of the sinus of Valsalva is much more probable. Very recently, Alter et al¹⁰ have reported a similar echocardiographic finding due to a nonruptured aneurysm of the left sinus of Valsalva.

Angiograms delineated the aneurysm with its origin slightly inferior to the left coronary arterial orifice and running behind the aortic root, which corresponds exactly with the echocardiographic picture showing a dense echo parallel to the aortic root. The presence of echoes moving within this structure is difficult to interpret. They could reflect late systolic filling of the aneurysm and diastolic emptying, as suggested by Wong et al¹¹ in their case report. Increased right ventricular diameter and paradoxic septal movement, indicative of right

FIGURE 2. Diagram of aortogram in right oblique position demonstrating aneurysm (An) of left sinus of Valsalva draining into right atrium (RA). Ao, Aorta; RV, right ventricle; and LV, left ventricle.

grams and angiograms two months after surgery showed that marked dilatation of the left sinus of Valsalva persisted and that the aortic regurgitation was unchanged.

FIGURE 3. Corresponding aortogram in right oblique position. Aneurysm of left sinus of Valsalva has its origin below left coronary artery and drains into right atrium.
ventricular volume overload, suggested ruptures into the right atrium or right ventricle.

Finally, the fluttering of the anterior mitral valvular leaflet and of the left ventricular side of the septum noted during diastole on the echocardiogram indicated aortic insufficiency. Cineangiograms demonstrated that the aortic insufficiency was due to traction of the valvular leaflet by the dilated and enlarged left sinus of Valsalva.

In conclusion, the echocardiographic findings of a vascular structure behind the aorta associated with signs of aortic regurgitation and signs of right ventricular volume overload, suggested a tentative diagnosis of aneurysm of the left sinus of Valsalva with rupture into the right atrium or right ventricle. This diagnosis was subsequently confirmed by cardiac catheterization and at surgery.

REFERENCES

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Gastric Rupture Complicating Post-Extubation Laryngeal Edema and the Use of a Manual Resuscitation Bag

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This report describes a patient who developed gastric rupture as a result of post-extubation laryngeal edema and the use of a manual resuscitation bag. The association between oxygen administration and gastric rupture is reviewed.

Gastric distension resulting from ventilatory assistance or resuscitative efforts is a common occurrence. Gastric rupture is, however, quite rare, and no reports of this complication associated with either upper airway obstruction and/or the use of manual resuscitation bags could be located in the literature.

CASE REPORT

An 82-year-old white woman with stigmata of severe rheumatoid arthritis and myxedema was admitted to Henry Ford Hospital because of right lower lobe pneumonia. Though initially stable, a respiratory arrest occurred 12 hours after admission because of retained secretions. The patient was intubated and ventilated mechanically for the next three days while chest physiotherapy and suctioning were performed.

On the fifth hospital day she was extubated, but ten minutes later developed marked stridor that did not respond to aerosolized racemic epinephrine. Reintubation was difficult because of the patient's agitation, and she was supported with supplemental oxygen delivered by face mask and a Puritan manual resuscitation (PMR) bag during the procedure. Marked gastric distension developed that did not respond to nasogastric suctioning, and a chest radiograph (Fig 1) revealed a large pneumoperitoneum without pneumomediastinum.

An exploratory laparotomy revealed a 5 cm laceration of the lesser curvature of the gastric wall, which was oversewn. The patient tolerated the procedure well and was uneventfully extubated 48 hours later. Her pulmonary status gradually improved during the remainder of her hospitalization.

Figure 1. Chest radiograph after reintubation. A large pneumoperitoneum is evident.