TWO-DIMENSIONAL AND DOPPLER ECHOCARDIOGRAPHIC FINDINGS IN A CASE OF SUBAORTIC "FIBROUS SAC"

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We described a 37-year-old man with a subaortic "fibrous sac", admitted for congestive heart failure. On 2-dimensional echocardiography a saccular structure was seen to extend from the left coronary cusp of the aortic valve to the outflow tract of the left ventricle. By color Doppler imaging, a grade 3 aortic regurgitation was recognized. Aortic regurgitant flow was recorded from the left coronary cusp to the saccular lesion. When congestive heart failure became exacerbated, the repeat examination showed the regurgitant flow passing through the perforated bottom of this lesion and reaching the left ventricular cavity. On microscopic examination of the excised valve, capillary proliferation and inflammatory changes were recognized near the annular region of the left coronary cusp. The edge of the valve leaflet and the other 2 cusps were intact. It is likely that our patient had a mycotic aneurysm near the aortic ring. We speculate that aortic regurgitation followed inflammation. It dilated the left ventricular cavity and contributed to congestive heart failure. Inflammation also weakened the tissue near the annulus, causing it to protrude into the subaortic region thus forming a small aneurysm. It may have grown to become a large saccular structure under high aortic pressure. That is, it became a "giant" endocardial pocket with inflammatory process. Finally, the rupture of this sac caused a massive aortic regurgitation, exacerbating congestive heart failure.

INFECTIONOUS endocarditis often causes aortic ring abscess. The abscess spreads to the adjacent sinus of Valsalva, extending to the left atrium or the right ventricular cavity or even into the myocardium. But there have been no reports of inflammation of the aortic ring causing the formation of a saccular structure protruding to the left ventricular cavity without the prominent destruction

of aortic root. We report here on a man who had a fibrous sac extending to the left ventricular cavity who had a macroscopically normal aortic root and sinus of Valsalva.

A thirty-seven year old man felt acute precordial pressure with cold sweats and nocturnal dyspnea on April 23, 1988. Cardiomegaly and heart murmur were noted in a hospital. He was diagnosed as having congestive heart failure. After treatment with furosemide and digoxin, he was transferred to the Osaka City University Hospital for further study on June 7. He had no history

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Fig.1. Two-dimensional echocardiographic findings. An abnormal structure protrudes into the left ventricle during diastole (white arrow) and disappears during systole (left ventricular end-diastolic dimension, 72 mm and end-systolic dimension, 53 mm).

of rheumatic heart disease.

On admission, the blood pressure was 120/48 mmHg and the pulse rate was 110 beats/min. Heart murmurs were heard as follows: a grade 3/6 holosystolic murmur at the apex, a grade 2/6 systolic ejection murmur and a grade 3/6 early diastolic murmur in the third and fourth intercostal space at the left sternal border, respectively. Crepitant rales were audible in both lungs. He did not have any manifestations of collagen disease or Marfan syndrome.

On laboratory evaluation, leucocytosis and an elevated C-reactive protein were recognized. The serum test for syphilis was negative. He had a few transitional events of low grade fever during the hospitalization, but blood cultures were negative. An electrocardiogram showed sinus tachycardia, I° AV block and left ventricular hypertrophy. Chest x-ray showed pulmonary congestion and the cardio-thoracic ratio was 65%. Two-dimensional echocardiography showed a markedly dilated left ventricle, with a saccular structure extending from the left

coronary cusp of the aortic valve to the out-flow tract of the left ventricle (Fig. 1). It bulged into the left ventricle in diastole but disappeared in systole. It bordered on the interventricular septum and on the base of the anterior mitral leaflet (Fig. 4, left). The other cusps were intact. On Doppler echocardiography, mitral, tricuspid and aortic regurgitations were detected. Aortic regurgitant flow was recorded in 2 locations: one at the commissure between the right and non-coronary cusps and the other from the left coronary cusp to the saccular lesion (Fig. 2, upper). Angiographically, aortic regurgitation was determined to be of grade 3.

While waiting for aortic valve replacement, congestive heart failure was exacerbated. On repeat Doppler echocardiography, the bottom of the saccular lesion was found to be perforated with regurgitant flow passing through the perforated portion and reaching the left ventricular cavity (Fig. 2, lower). During the operation on August 1, macroscopically, the sinuses of Valsalva were normal. Of the excised 3 cusps, the left

Fig.2. Color Doppler flow imaging and schematic description. Aortic regurgitant flow is detected in the structure (left upper). After perforation of the saccular structure, the regurgitant flow passes through the hole (white arrow) and reaches the left ventricular cavity (left lower). AV=aortic valve, LA=left atrium, LV=left ventricle.

coronary cusp was smaller than the others (Fig. 3) and continuous with the saccular structure. This was 5.5 cm by 4.0 cm in size with a hole at the bottom. The right and non-coronary cusps were almost intact and without vegetation. Histologically, near the annular region capillary proliferation and mild inflammatory cell infiltration were recognized (Fig. 5). The edge of the valve leaflet was intact. The wall of the saccular

lesion was fibrous and partially elastic. The saccular structure bordered on the endocardium of the interventricular septum through the loose connective tissue.

Several diseases can cause an abnormality of the subaortic region. Congenital subaortic stenosis can be classified into the discrete or muscular type, but we could not discern a discrete membranous lesion or muscular ridge! During the operation, there was no

Fig.3. Macroscopic appearance of the resected aortic leaflets (left) and left coronary cusp (right). Right and non-coronary cusps appear normal. The left coronary cusp is abnormally enlarged but the proper leaflet (black arrow) has shrunk and is continuous with the perforated saccular lesion (white arrow).
R=right coronary cusp, N=non-coronary cusp, L=left coronary cusp.

Fig.4. Schematic description of the saccular structure. Left: the shaded area depicts the lesion. The proper leaflet is smaller than normal. The dotted line indicates the proper ring. Right: the saccular structure protrudes into the left ventricle (arrow).
LCC=left coronary cusp, NCC=non-coronary cusp, RCC=right coronary cusp, MV=mitral valve IVS=inter-ventricular septum.

evidence of rupture of the sinus of Valsalva? In this patient, the mitral leaflets were normal and the saccular structure had no chorda-like tissue characteristic of an accessory mitral valve³. Cases of aortic ring abscess have been reported using 2-dimensional or Doppler echocardiography. Scanlan et al⁴ reported an aortic ring abscess as

an echolucent or echo-free space by 2-dimensional echocardiography. Pollak et al⁵ reviewed 11 published cases in addition to their 4 cases. Thirteen of totally 15 cases had aortic ring abscess, including 5 prosthetic aortic valves. Saner et al⁶ reported 9 additional cases. All had underlying disease of a prosthetic or congenital bicuspid aortic valve. The reports differentiated between an aortic root abscess and a mycotic aneurysm. The former was defined as purulent material contained within a fibrous capsule and did not communicate with the lumen. The latter was defined as an aneurysm with its origin in the aortic annulus, sinuses or wall caused by an infectious break in the internal wall forming a blind sac connected with the aortic lumen. The progression of these infectious processes into the subannular structures may lead to disruption of the membranous septum or the aortic-mitral intervalvular fibrosa.

We could not detect any cavity which was seen in an aortic ring abscess as an echolucent or echo-free space extending into the left ventricular myocardium or left atrium. As revealed by surgery, unlike an aortic ring abscess, the sac did not contain purulent fluid and communicated with the aortic lu-

Fig.5. Histology of the left coronary cusp and schematic description.

The dotted line indicates the proper ring. A: the edge of the valve leaflet (left upper), B: the tissue near the annulus (right upper) and C: a portion of the saccular structure (left lower) (Elastica von Gieson stain, $\times 50$). D: near the annular region, capillary proliferation and mild mononuclear cell infiltration were seen (right lower, Hematoxylin-Eosin stain, $\times 100$).

LCC=left coronary cusp.

men. Recently, Morimoto et al⁷ reported a similar patient who had a cystic lesion of the aortic valve, that is, an aortic valve aneurysm. The lesion in our patient is different from an aortic valve aneurysm because the wall of the saccular structure involved the annulus and the inflammation appeared in the valve annulus and not the valve leaflet (Fig. 5). Therefore, it is more likely that our patient had a mycotic aneurysm near the aortic ring than an aortic ring abscess or a valve aneurysm. But microscopically, most of the wall of the saccular structure had collagen and elastic fibers, and not infiltration of inflammatory cells. The growth of the sac may be caused by not only inflammation, but also an aortic regurgitant jet. The regurgitant jet sometimes causes endocardial thickening or an endocardial pocket⁸. Originally, it is cusp-like in size without inflammation. But the lesion may become a "giant" endocardial pocket if the inflammatory process adds to it. We speculate that aortic regurgitation followed infectious endocarditis. It dilated the left ventricular cavity and contributed to congestive heart failure. In-

flammation also spread and weakened the adjacent tissue between the annulus and the bottom of the sinus of Valsalva, causing it to protrude into the subaortic region like a small aneurysm (Fig. 4, right). Under high aortic pressure, it may have grown to become a large saccular structure. Finally, the exacerbation of congestive heart failure may have been caused by rupture of this sac.

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