We experienced a case with a huge aortic valve aneurysm, and its etiology was strongly suspected as due to infectious endocarditis from the pathological findings, blood cultures, and clinical course.
history was unremarkable except for a tonsillectomy due to tonsillitis at the age of 60 years. His elder brother suffered from diabetes mellitus, but his other family history was unremarkable.

**Present Illness**: On May 28, 1999, the patient suddenly experienced dyspnea on exertion, and he visited a near-by hospital. On chest X ray examination, the Cardio-thoracic Ratio (CTR) was 53%, but a right pleural effusion was noted. He was hospitalized with the diagnosis of congestive heart failure. After being hospitalized, trans-thoracic echocardiography was performed, and mitral regurgitation, aortic regurgitation, and aortic valve doming out into the left ventricular chamber were noted. He was transferred to our hospital for a detailed examination and treatment.

**Physical Findings on Admission**: The patient's height was 167 cm, weight 67 kg, body temperature 36.0 degrees Centigrade, blood pressure 140/60 mmHg, and he had a regular heart rhythm at 60 beats per minute. Neither anemia nor icterus or conjunctiva was found. No crackles were audible over the lung field. A Levine II/VI systolic murmur auscultated from the second intercostal space on the right sternal border and a Levine III/VI diastolic murmur at the fourth intercostal space on the left sternal border was audible.

**Laboratory Findings on Admission**: The blood tests on admission revealed a slightly high value for an inflammatory reaction (Table 1).

**Chest X Ray Findings**: The CTR ratio was 51.5%, and the vascular shadow in the lung field was increased (Fig. 1).

**ECG Findings**: The heart rhythm was normal sinus rhythm, but a slight ST segment depression in leads II, III, aVR, V5, and V6 was noted (Fig. 1).

**Trans-Thoracic Echocardiography**: The aortic diameter was 42 mm, left atrial diameter 38 mm, and left ventricular end-diastolic diameter 52 mm, revealing a slight enlargement and dilatation of the aorta and left ventricle. The left ventricular ejection fraction was 61%. Moderate

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Table 1. Laboratory Data

<table>
<thead>
<tr>
<th>Blood Cell Count</th>
<th>Biochemical Test</th>
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<tbody>
<tr>
<td>WBC 7500/μl</td>
<td>TP 7.2 g/dl</td>
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<tr>
<td>RBC 4.52×10^12/μl</td>
<td>GOT 15 IU/L</td>
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<tr>
<td>Hb 131 g/dl</td>
<td>GPT 14 IU/L</td>
</tr>
<tr>
<td>Plt 26×10^9/μl</td>
<td>LDH 2.14 IU/L</td>
</tr>
<tr>
<td>Cr 1.1 mg/dl</td>
<td>CK 2.34 mg/dl</td>
</tr>
<tr>
<td>BUN 18 mg/dl</td>
<td>BUN 2.0 mg/dl</td>
</tr>
<tr>
<td>K 4.2 mEq/L</td>
<td>Na 144 mEq/L</td>
</tr>
<tr>
<td>Cl 110.8 mEq/L</td>
<td>C-reactive protein&lt; 40</td>
</tr>
</tbody>
</table>

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![Fig. 1](image1.png) Chest radiography on admission demonstrating a Cardio-thoracic ratio of 51.5% and pulmonary congestion. The electrocardiogram demonstrated sinus rhythm with ST depression in leads II, III, aVF, V5, and V6.

![Fig. 2](image2.png) Transthoracic echocardiography in the parasternal long-axis view demonstrating an aortic valve aneurysm on the left coronary cusp. LCC: left coronary cusp.
mitral valve regurgitation and aortic valve regurgitation (Decay slope 4.2 m/s²) accompanying the left coronary cusp prolapsing into the left ventricular chamber with bulging along the long axis view of left ventricle was noted (Fig. 2).

Trans-Esophageal Echocardiography: Moderate mitral valve regurgitation and slight aortic regurgitation was noted. On the aortic valve, the left coronary cusp leaflet exhibited bulging and prolapsing, and the non-coronary cusp leaflet also exhibited prolapsing into the left ventricle (Fig. 3).

Cardiac Catheterization Examination: The left ventriculography demonstrated Sellers II mitral regurgitation. Aortography demonstrated Sellers III aortic regurgitation, and also the left coronary cusp leaflet was bulging and prolapsing and the non-coronary cusp leaflet was prolapsing into the left ventricle (Fig. 4).

Clinical Course after Admission: The patient was afebrile and his blood tests revealed a normal white blood cell count (7500 /mm³) and relatively low CRP value (3.4 mg/ml). Blood cultures were also negative, thus his condition was simply diagnosed as congestive heart failure due to combined valvular heart disease. Digoxin and flosemide were administrated, resulting in gradual improvement in his congestive status. However, he later developed anterior chest pain while he was still in the hospital, which was accompanied by ST-T segment depression on his ECG during the attack. Although there was a slight possibility that this was secondary due to the aortic regurgitation, an infusion of nitroglycerine effectively improved the symptoms and ECG changes. These problems seemed to be easily controlled by oral medical treatment, and he was discharged on June 17th. From July 5th, he developed nocturnal dyspnea in the supine position, thus he was re-admitted to our hospital. The echocardiogram demonstrated no remarkable change in the end-diastolic diameter of the left ventricle (55.5 mm) or the left ventricular ejection fraction (76%), but his blood cultures were positive for Staphylococcus epidermidis, thus he was diagnosed with worsened congestive heart failure due to combined infectious endocarditis. Treatment was started with diuretics (flosemide), cardiotonic agents (mirurinon), and antibiotics (imipenem), but his congestive status did not improve. The inflammatory laboratory findings were unchanged after his admission (WBC 5800 /mm³, CRP 2.1 mg/ml), and the thickening of the mitral leaflet was detected to have worsened with the echocardiography, thus his infectious endocarditis was
refractory to this treatment. The patient appeared to be intolerable to this medical treatment, and surgical intervention was chosen.

**Surgical Treatment and Findings** : The chest was opened with the median sternotomy approach, and fibrous adhesions were found around the aorta, pulmonary artery, and superior vena cava. Blood was drawn from a venous cannula inserted into the superior and inferior vena cavae and fed and supplied to the ascending aorta via an aortic cannula for extracorporeal circulation. When the aorta was opened with an incision, a tear in the surrounding tissue of the aortic valve 2 mm beneath the valve was found. This tear was 22 mm long, reaching to the posterior surface of the mitral leaflet, surrounded by the lateral wall of the left ventricle and its papillary muscle connected to the mitral leaflet. On the center portion of the left coronary cusp leaflet, there were two valvular aneurysmal formations. The larger aneurysm reached to a size of 20 X 23 mm, but no perforation was found. The leaflets of the right coronary cusp and non-coronary cusp were thickened. The mitral leaflet was thickened and the tendon of the anterior commissure was shortened, and this anterior leaf was removed and replaced with a CarboMedics Valve (fai 23 mm) using nine 2-0 ethibond threads. After the aortic valve replacement was finished, the patient's circulation was resumed from the extracorporeal circulation without any problems.

**Pathological Findings** : The left coronary cusp leaflet that was removed had an aneurysmal formation in its central portion, and it was bulging out into the left ventricular side (Fig. 6). The pathohistological study revealed infiltration of inflammatory cells, most of which were neutrophils infiltrating the tissue, and this was also accompanied by mucoid degeneration (H-E stain).

**Clinical Course and Treatment after the Surgical Operation** : The tissue that was removed during the operation was tested for bacterial studies, but no bacillus was detected and the culture was negative. However, the blood cultures had already resulted in positive findings for *Staphylococcus epidermidis*, as mentioned, thus antibiotic therapy was continued, though the combination was altered to use piperacillin, clindamycin, and imipenem, after the operation. The inflammatory findings and congestive status of the failing heart gradually improved, and the chest X ray examination was found to have improved in regard to the CTR before he was discharged on September 30th.
DISCUSSION

Valvular aneurysms are a group of diseases of various etiologies with the same structural disorders, which are especially easily found due to the progress of echocardiographic diagnostic technologies. Aneurysmal changes are likely to form on mitral or aortic valves, but cases with them solely on the aortic valve are extremely rare. As far as we could review, there have been only nine such aneurysmal cases, and there have only been three aortic valve cases in Japan.

Transthoracic echocardiography is definitely a powerful tool for the diagnosis of aortic valve aneurysms. The characteristic findings are that a sack-like form is demonstrated beneath the aortic valve facing towards the left ventricle during the diastolic phase, this sack-like form disappears or shrinks during the systolic phase, and the sack-like form is demonstrated as a ring-like form in the short axis view. The same findings were also demonstrated in the present case, and this patient was diagnosed as having an aortic valve aneurysm.

The etiology of valvular aneurysms is mostly known to be due to infectious endocarditis, and other causes have been reported to be congenital, rheumatic fever, aortitis syndrome, Bechet syndrome, Marfan syndrome and so on. Morimoto 1) and Iemura 2) proposed a mechanism for aneurysm formation which was initiated by the erosive changes or abscess formation due to infection, and the aortic diastolic pressure affect on such a lesion resulted in the dilation and formation of the aneurysm. Safhir et al. 3) classified aneurysms into true and pseudo-aneurysms from their histological characteristics as to whether or not they had endocardium over the surface of the aneurysm. The former (True) is supposed to have endocardium, because it gradually dilates at the weak valvular site which was left during the antibiotic treatment. The latter (Pseudo), on the other hand, is supposed to form with rapid dilatation, thus the endocardium is damaged in this process. Four years prior to the recent incidence, the present case had a past history of tonsillitis, which was treated with antibiotics. Since it was refractory to medical treatment, surgical therapy was also undergone for that also. From the consideration of this long and related clinical history, the aneurysm was presumed to be a true aneurysm, and was confirmed by the histopathological study.

In the case of mitral valve aneurysms, Edwards 4) and Reid 5) pointed out the importance of their anatomical relationship to their continuity, and suggested that they are formed by a direct hit from the blood jet flow due to aortic regurgitation towards the mitral leaflet, which may create a weak site resulting in a bulge formation with the pressure load. Like this example, the mitral valve is likely to be affected by various pressures and/or blood stream loads. Thus the mitral valve may have a greater chance to form a valvular aneurysm than the aortic valve. With the aortic valve, the pressure load may be the only related factor, and this fact may explain the reason why they are so rare with the aortic valve. The case presented here had inflammatory cell infiltration revealed by the histopathological study, and the etiology was presumed to be endocarditis. Further, the reason for having a huge aneurysm was due to its massive destructive effect on valve structure.

Reference

感染性心内膜炎により著明な大動脈弁膜症を呈した 1 例

抄 録

僧帽弁膜症の症例は散見されるが大動脈弁膜症は稀である。我々は感染性心内膜炎に伴ない巨大な大動脈弁膜症を形成した症例を経験したので報告する。

症例は 65 才男性。3 ヶ月前から胸部絞扼感出現。近医にて心不全の診断にて入院。経胸壁心エコー上、大動脈弁閉鎖不全、僧帽弁閉鎖不全、大動脈弁の左室内突出を指摘され当科に転院。入院時、拡張期逆流性雑音を聴取し、炎症所見を認めた。経胸壁心エコー、経食道エコー上左冠尖の瘤状膨瘤を認め大動脈弁膜症が疑われた。抗生剤、利尿剤による治療を開始したが、心不全の進行と僧帽弁前尖の肥厚を認めめたため、2 術置換術を施行。手術所見では左冠尖に 20 〜 23 mm の弁膜症を認めた。切除組織の細菌培養は陰性であったが、血液培養にて表皮ブドウ球菌が検出されていることと、病理所見上高度な弁破壊を伴う炎症性細胞浸潤を伴うことなどから、その発形成に感染性心内膜炎が関与していると考えた。

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