Case Report

A Case of Multiple Aneurysms

Mototaka Murakami, M.D.,* Tetsuo Yokoyama, B.S.,**
Akira Itakura, B.S.,** Eiji Murakami, M.D.,**
Seiichi Murakami, M.D.,*** Kenji Shiotani, M.D.,***
Itaru Yamakawa, B.S.,*** and Fujitsugu Matsubara, M.D.****

Multiple aneurysms of the major branches of the aorta are rare, although the incidence of multiple aneurysmal formation of the aorta ranged from 5 to 20 per cent of all the aortic aneurysms. In 1958, Held reported a sixth case of multiple aneurysms in large arteries, naming it "Wurzelknollenkrankheit der großen Arterien."

This paper describes briefly a case of multiple aneurysms involving the right and left subclavian arteries, the right renal artery and the abdominal aorta, associated with hypertension and non-functioning kidney of the involved side.

Case Report

T.N., a 45-year-old man was admitted to the First Department of Surgery of this Hospital because of swelling in the right supraclavicular fossa, on November 8, 1962. One year before entry the patient was found to have hypertension and proteinuria for the first time. In October, 1962 he noticed of a swelling in the right supraclavicular fossa. Although he had complained of infrequent dull abdominal pain for the last 3 years before entry, he did not recall having had hematuria, nausea or vomiting. Family history and past history were non-contributory. Physical examination disclosed two masses, one in the right and one in the left supraclavicular fossae. A murmur over the abdominal aorta was also discovered. Blood pressure was 170/120 mm.Hg and equal in the both arms, as were the brachial pulses. Diagnosis of aneurysms of the right and the left subclavian arteries was made. On November 18, aneurysm of the right subclavian artery was extirpated. On December 10, 1962 he was referred to the Second Department of Internal Medicine.

On admission, physical examination revealed a poorly nourished, chronically

* , ** From the Second Department of Internal Medicine, School of Medicine, Kanazawa University, Kanazawa.
* Professor of Internal Medicine.
*** From the First Department of Surgery, School of Medicine, Kanazawa University, Kanazawa.
**** From the Central Clinical Laboratory, University Hospital, Kanazawa University, Kanazawa.
ill man, but in no acute distress. Blood pressure was 204/114 mm.Hg in the left arm and was unobtainable in the right arm. Pulse was regular with rate of 64 per min. Brachial pulses were difficult to feel in the right arm. The pupils reacted to light and accommodation. There was no periorbital edema. The scar of the previous operation was present on the right upper anterior chest. A pulsatile, non-tender, elastic, soft mass of about 2 by 5 cm. with a harsh systolic murmur was noted in the left supraclavicular fossa. The lungs were clear to percussion and auscultation. The heart was not enlarged, sounds were normal and no murmurs were detected. The liver and the kidneys were not palpable. No abdominal masses were palpable. There were harsh systolic and diastolic murmurs around the umbilicus, heard best at the area 2 fingerbreadths to the left of the umbilicus and a systolic thrill was palpated at the same area. Reflexes were normal. The upward movements of the right upper limb were limited due to previous operation. No pretibial or pedal edema was present.

Laboratory Data: The urine was straw colored, with an acid reaction, trace of albumin, no sugar and 4-5 white blood cells per low power field. Examination of the blood revealed the hemoglobin of 58 per cent, the red cell count of 4,700,000 and the white cell count of 4,300, with 68 per cent neutrophils, 5 per cent band forms, 2 per cent basophils and 23 per cent lymphocytes. The platelets were 64,000 per cubic mm. The prothrombin time was 20.1 sec. The serologic test for syphilis was negative in the blood. The nonprotein nitrogen was 19 mg., the total protein 7.8 Gm. (the albumin 35.3 per cent, the α1-globulin 6.8 per cent, the α2-globulin 9.7 per cent, the β-globulin 16.1 per cent and the γ-globulin 32.1 per cent), the cholesterol 195 mg. and the β-lipoprotein 457 mg. per 100 ml. The sodium was 140 mEq., the potassium 4.7 mEq., the calcium 4.4 mEq. and the chloride 99 mEq. per L. The inorganic phosphorus was 3.5 mg. per 100 ml. Erythrocyte sedimentation rate was 23 mm. in 1 hour and 57 mm. in 2 hours (Westergren). C-reactive protein was positive, 1+. The thymol turbidity test was 4.7 units, and the zinc sulfate test was 17.6 units. The daily urine volume was between 600 and 2,300
Fig. 3. Retrograde pyelogram.

Fig. 4. Transfemoral aortogram showing an aneurysm of the right renal artery and multiple aneurysmal dilatation of the abdominal aorta.

Fig. 5. Superimposed renograms obtained over right and left renal regions, following the injection of 0.5 μc./Kg. I\(^{131}\).Hippuran. The right renogram showed a diminution in segment A and absence of segment B.

ml. the average being 1,500 ml. The maximum concentration and dilution of the urine were 1.025 and 1.006, respectively. The phenolsulphophthalein test showed 13.5 per cent excretion of injected dye in 15 min. and 49.5 per cent total in 2 hours. The creatinine clearance was 55.9 ml. per min. Antibody to rabbit anti-human

* Normal value, negative.
kidney serum was positive in a titre of 1:2 on one occasion and 1:4 on another oc-
casion.* Funduscopic examination revealed thin sclerotic arteries and small hemor-
rhages. The chest X-ray film was normal except for the slight widening of the
aortic arch. Transfemoral aortogram (Fig. 1) before extirpation of the aneurysm
of the right subclavian artery failed to outline the both subclavian arteries.

Fig. 6. Renal scintillation scanning, 3 hours after the intravenous
injection of 50 μc./Kg. Hg²⁰⁹-Neohydrin.

Fig. 7. Records of vascular sounds. Upper left, at apex. Lower left,
over the aneurysm of the left subclavian artery. Upper right, at the area 2
fingerbreadths to the left of the umbilicus. Lower right, at the area 2 finger-
breadths to the right of the umbilicus.
The thoracic aorta, the innominate, and the both common carotid arteries were within normal limits. Excretory urograms (Fig. 2) failed to show the right pelvis clearly, but the upper urinary tract of the left side was well visualized on the 7 min. exposure. Cystoscopy disclosed no flows of urine or injected indigocarmine from the right ureter. Retrograde pyelographic study (Fig. 3) revealed a slightly dilated right collecting system. The size of the right kidney measured on X-ray film was 5.6 by 8.5 cm. and that of the left was 7.5 by 15 cm. Transfemoral aortograms (Fig. 4) demonstrated aneurysm of the right renal artery and multiple aneurysmal dilatation of the abdominal aorta, extending down to the level of the renal arteries. The radioisotope renogram from the right renal area was typical of non-

Fig. 8. Extirpated aneurysm of the right subclavian artery.

Fig. 9. Wall of artery without thrombus, showing intimal fibrosis, thickening and vacuolation (Hematoxylin-eosin stain).

Fig. 10. Wall of the aneurysm near the thrombus. Note severe fibrosis and thickening of intima and thinning of irregularly fibrosed media (van Gieson's stain).
functioning or absent kidney (Fig. 5). The left renogram was within normal limits. Scintigram (Fig. 6) failed to scan the right kidney, while it suggested the hypertrophied left kidney. Electrocardiograms revealed inverted U waves in leads V_4 and V_5. In leads II, III, aV_L and aV_R the downstrokes of the QRS complex were notched. Vectorcardiograms were unremarkable. Records of vascular sounds (Fig. 7) revealed normal cardiac sounds and systolic murmur over the aneurysm of the left subclavian artery. Around the umbilicus there were late systolic and diastolic murmurs, heard best at the area 2 fingerbreadths to the left of the umbilicus.

Findings of the Previous Operation: On November 18, 1962 the right subclavian artery was entered through a claviculotomy. In the artery, a saccular aneurysm of about 5 cm. in diameter was found, 5 cm. from the origin of the right common carotid artery (Fig. 8). Moderate adhesion to the adjacent right subclavian vein was present. Aneurysm was extirpated. It contained red-grey material adherent to the vessel wall. Arterial reconstruction with teflon graft was attempted without

**Fig. 11.** Aneurysm, filled with thrombus. Difficult to identify each layer of aneurysmal wall due to fibrosis (Hematoxylin-eosin stain).

**Fig. 12.** Elastic fibers revealing fragmentation, tortuosity and partial thickening of elastic fibers (left), fragmentation and scarcity of elastic fibers (middle), and scattering of few elastic fibers (right) (Weigert’s elastic tissue stain).
success, because of fragility of arterial wall, and both ends of the artery were ligated.

Histologic Findings of the Extirpated Aneurysm: The wall of the aneurysm showed marked arteriosclerotic changes, with relatively recent mural thrombus. The media was thin and partly disappeared, so that identification of each layer was difficult. There was marked proliferation of capillaries and hyalinized connective tissue fibers in the intima and in the adventitia. Minimal accumulation of sudanophilic material was present in the intima. Elastic fibers were coarsely thickened, tortuously fragmented and partly disappeared (Fig. 9-12).

**DISCUSSION**

Aneurysm of the major branches of the aorta is relatively infrequent. According to White\(^5\)) the chief sites of aneurysms other than in the aorta are the popliteal, femoral, carotid, subclavian, innominate, axillary and iliac arteries, and the chief sites of visceral aneurysms are the splenic and hepatic arteries. Aneurysms may be single or multiple. Brindley and Stembridge,\(^4\) collecting 369 necropsy cases of aortic aneurysms and reviewing the literature, stated that the incidence of multiple aneurysms of the aorta was from 5 to 20 per cent. Multiple aneurysms other than in the aorta are extremely rare. In 1958, Held\(^1\) reported a sixth case of multiple aneurysms in large arteries, naming it "Wurzelknollenkrankheit der großen Arterien." Our case had at least 4 aneurysms, i.e. in the right and left subclavian arteries, the right renal artery and the abdominal aorta. We did not ascertain whether there was any other aneurysm elsewhere. Careful auscultation disclosed no murmurs other than those already mentioned, however, in view of multiplicity of involvement of the case, a possibility exists that he might have another aneurysm and in the course of time it would be discovered.

The cause of our case is obscure. Serologic test for syphilis was negative and there were no evidences for syphilis clinically. In a few reported cases of multiple aneurysms, Lepow et al.\(^2\) reported a case caused by elastic tissue defect, but in our case the result of elastic tissue stain did not coincide with it. Held's case\(^1\) had history of severe serum anaphylactic shock, which the author took significant. No such a history could be obtained from our case. Although the tissue specimen showed marked arteriosclerotic changes, conclusion could not be drawn from the limited material. Arteriosclerosis might be a secondary change formed on the wall of preexisting aneurysm. Also we could not exclude arteritis or congenital weakness in vessel wall as suggested by Abrahams and Cockshott,\(^3\) for which there was no basis to support in our case.

Another point of interest is that the patient had hypertension. Six cases collected by Held\(^1\) including his own case, all were hypertensive. Although, Held made no mention about causal relationship between hypertension and
aneurysms, it is not unreasonable to assume that hypertension had caused multiple aneurysms. Recently renovascular hypertension has attracted attention as curable hypertension. Among many causes of renovascular hypertension, renal artery aneurysm was not infrequent. In the case reported here, an aneurysm was found in the right renal artery, with resultant renal functional impairment. As the patient had had no history suggestive of renal infarction and pressure gradient of the right renal artery was not measured, we could not conclude whether hypertension was cause or result of aneurysms. However, all available data were highly indicative of renovascular hypertension, caused by thrombotic narrowing of the right renal artery aneurysm. We intended to perform him arterial reconstructive surgery or at least nephrectomy. Unfortunately, however, experience at previous operation that the arterial wall was too fragile to manipulate made us not operate him on.

Summary

(1) A case of multiple aneurysms involving the right and left subclavian arteries, the right renal artery and the abdominal aorta, is presented.

(2) The pathogenesis of these lesions was obscure, although the aneurysm of the right subclavian artery showed severe arteriosclerotic changes.

(3) He had hypertension and functional impairment of the right kidney. Renal artery aneurysm as a cause of renovascular hypertension was briefly discussed.

References