Case Reports

A Case of the Superior Vena Caval Syndrome Presumed to Result from the Postoperative Disorder of Bilateral Clavicular Osteomyelitis

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THE superior vena caval syndrome has been extensively studied since the first description by William Hunter in 1757.¹⁾ Recently it has become to be diagnosed more accurately and reports appear increasingly because of the progressive improvements in the diagnostic technics. This report is a case of the superior vena caval syndrome that is assumed to result from the postoperative disorder of bilateral clavicular osteomyelitis.

CASE REPORT

A 59-year-old man, a draper, was admitted to the First Department of Internal Medicine, Kanazawa University Hospital on March 1, 1962, complaining of venous dilatation of thoracic and abdominal walls, emaciation, general malaise, and palpitation and dyspnea on exertion. In 1935, the patient noticed of a slight pain on movement of his bilateral shoulders which, however, were not swollen. He had been suffering from the same episodes every winter and every rainy season until June, 1946, when he noticed of severe pains never experienced by that time and the movements of his bilateral upper extremities were remarkably restricted. As the pain did not disappear completely in spite of a spa treatment for a week, he consulted a physician by whom the diagnosis of neuralgia was made. Although the pain was gradually relieved by two months treatment, details of which are unknown at present. He thereafter, began to suffer from frequent attacks of severe pains in the bilateral

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shoulders. Slight pains in the chest, waist and extremities were also complained of. Fever was never noticed. In September, 1951, he visited the First Department of Surgery, Kanazawa University Hospital because of a swelling in the right clavicular area associated with a severe spontaneous pain where he was told for the first time that the cause of shoulder pains he had been suffering from for a long time was chronic osteomyelitis of the clavicle, for which an operation was performed on October, 1951. Twenty days after the first operation the second one was done because there occurred a swelling and an acute pain at the left clavicular area, too. After these operations he noticed by himself that his posture became gradually kyphotic and the rotation or tilting of the neck was increasingly limited because of pains. In addition, he began to complain of rachialgia and lumbago in June, 1953, when the kyphosis became remarkable in degree and the movement of the neck was severely limited.

Fig. 1. Venous dilatation and tortuosity a) on the anterior chest and abdominal walls, and b) on the right and c) on the left aspect of the chest

Since December, 1961, he noticed the dilatation of superficial veins of thoracic and abdominal walls and a gradual decrease of his body weight. In January, 1962, he caught a cold, and thereafter he felt general malaise, and palpitation and dyspnea on exertion.

Family history was non-contributory.

On admission, physical examination revealed a small, poorly nourished man who seemed to be chronically ill. Temperature was 36.7°C, pulse 80 per min. and regular, respirations 20 per min. and blood pressure 110/78 mm.Hg. As shown in Fig. 1a), b) and c), marked distension and tortuosity of veins of thoracic and abdominal walls were observed. The direction of the blood flow in these vessels was downward on the whole. The pupils were circular, equal in size and reacted promptly to light. Fundoscopic examination demonstrated grade I finding of Keith-Wagener classification. The lips were not cyanotic. No lymphadenopathy was noted. The cardiac dullness was normal, heart sounds were of fairly intensity and no murmurs were audible in any areas. The lung was clear to percussion and to auscultation. The abdomen was flat and no ascites was elicited. The liver, spleen and kidneys were not palpable. The vertebral column showed a remarkable kyphosis centered at T-VII.

The laboratory studies revealed 67 per cent hemoglobin, 443×10^4 red blood cells per cubic millimeter, color index 0.76 and 7,900 white blood cells per cubic millimeter with normal differential count. The sedimentation rate was 130 mm. per Hr. and 142 mm. per 2 hrs. The serological test for syphilis was negative in the blood. Urinalysis showed a + test for albumin; the sediment contained 2 to 3 red blood cells, 5 to 6 white blood cells per high-power field, one flat epithelial cell per 5 or 6 high-power fields, one small round cell per 7 or 8 high-power fields and one granular cast per 10 high-power fields. Sugar, urobilinogen, urobilin and bilirubin were all negative. The stool was negative for occult blood, no parasites or ova were seen. The icterus index was 6, the cobalt reaction $R_{3(4)}$, the cadmium reaction R_{10} , the Takata reaction (-) at 3 Hrs. and ($\pm$) at 24 Hrs., the CCF test 1 (+) at 24 Hrs. and 3 (+) at 48 Hrs.; the bromsulfalein test showed 6 per cent retention of the dye at 45 min. The total serum protein was 7.5 Gm. per 100 ml., with albumin-globulin ratio of 0.35; the serum protein fractions showed 26.1 per cent albumin, 7.8 per cent α_1 -globulin, 9.9 per cent α_2 -globulin, 11.6 per cent β -globulin and 44.6 per cent γ -globulin. The serum alkaline phosphatase activity was 2.7 units (Bessey-Lowry), total serum cholesterol 158 mg. per 100 ml. The serum sodium was 137 mEq., potassium 4.0 mEq., calcium 4.9 mEq. and chloride 96 mEq. per liter. The CRP was 4+, RA-test negative and ASL-O 100 Todd units. The venous pressure of the upper extremities was 105 mm. of water in the right and 95 mm. of water in the left side, and that of the lower extremities was 45 mm. of water in the right and 30 mm. of water in the left side; when the lower part of the chest was bound with a rubberband, a rise in pressure equivalent to 25 mm. of water was observed. The circulation time measured with decholin was 23 sec. and the time measured with ether was 10 sec.

The roentgenogram of the chest indicated prominence of aortic arch and deformity, irregular trabeculation, hardening and dissolution of bilateral clavicles (Fig. 2). The roentgenogram of the spinal column showed the findings compatible

Fig. 2. Roentgenogram of the chest showing prominence of aortic arch and deformity, irregular tracheation, hardening and dissolution of bilateral clavicles.

Fig. 3. Roentgenogram of the spinal column indicated the finding of spondylitis ankylopoetica totalis.

Fig. 4. Phlebogram in the right side; most of the contrast medium flowed from the venous plexus-like anastomosis developed between the right jugular vein and the right subclavian vein, through the bilateral brachiocephalic veins, into the superior vena cava.

Fig. 5. Phlebogram in the left side; a part of the contrast medium flowed from the anastomosis through the left brachiocephalic vein into the superior vena cava. The most upper part of the film was overshadowed by cracks due to technical error.

with spondylitis ankylopoetica totalis (Fig. 3). X-ray examination of the esophagus and stomach did not show any abnormalities. The tracheo-bronchogram was interpreted almost normal.

The phlebogram in the right side revealed that most of the contrast medium flowed from the right jugular vein into the superior vena cava through the venous plexus-like anastomosis developed between the right jugular vein and

the right subclavian vein and through the bilateral brachiocephalic veins and a part flowed into the thoracic subcutaneous veins (Fig. 4). The catheter inserted into the right superficial jugular vein passed readily to the superior vena cava through the right brachiocephalic vein. The phlebogram in the left side, being overshadowed by cracks of the film due to technical error, revealed that most of the contrast medium flowed from the left jugular vein into the left external

Fig. 6. Schematic representation of both-sided phlebograms on the same paper.

- a: right brachiocephalic vein.
- b: left brachiocephalic vein.
- c: superior vena cava.

thoracic vein and a part flowed from the anastomosis, although not so well developed as in the right side, into the superior vena cava through the left brachiocephalic vein, situation of which was displaced to the right side (Fig. 5). The catheter inserted into the left superficial jugular vein could not enter into the left brachiocephalic vein. Fig. 6 is a schematic representation of the both-sided phlebograms on the same paper which shows clearly (1) the difference of degree in development of the venous plexus-like anastomosis between the jugular vein and the subclavian vein, (2) the relationship between the anastomosis and the brachiocephalic veins, and (3) the relation of the brachiocephalic veins to the superior vena cava.

DISCUSSION

The superior vena caval syndrome results from the obstruction of the superior vena cava⁽¹⁾⁻¹²⁾ or bilateral brachiocephalic vein,^(13),14) or from arteriovenous communication between the ascending aorta and the superior vena cava.^(6),15),16)

The causes of superior vena caval syndrome are numerous as shown

in Table I, among which the thoracic tumor is the most important and its percentage tends to increase more and more in recent years while cases due to aortic aneurysm has decreased remarkably owing to the development of anti-luetic treatment.

Mediastinitis (traction or compression by scar tissue) is another important cause of the superior vena caval syndrome. Recently cases

Table I. Causes of Superior Vena Caval Obstruction

Authors	No. of cases	Thoracic tumors No. (%)	Aortic aneurysms No. (%)	Mediastinitis No. (%)	Thrombosis No. (%)	The others No. (%)
Fischer (1904)	252	126 (50.0)	91 (36.0)	15 (6.0)	14 (5.5)	Tuberculous phlebitis 4 (1.5) Actinomycosis 1 (0.5) Obliteration of pericardium 1 (0.5)
Hussey et al. (1946)	35	13 (37.1)	12 (34.3)			Leukemia 2 (5.7) Unverified 8 (22.9)
McIntire et al. (1949)	250	94 (37.6)	60 (24.0)	62 (24.8)	23 (9.2)	Etiology not stated 11 (4.4)
Hinshaw (1949)	125	48 (38.4)	29 (23.2)	16 (12.8)	17 (13.6)	Acute leukemia 3 (2.4) Pericardial constriction 2 (1.6) Pneumothorax 1 (0.8) Mitral stenosis 1 (0.8) Undetermined 8 (6.4)
Klassen et al. (1951)	12	7 (58.5)	1 (8.3)	1 (8.3)	1 (8.3)	Hodgkin's disease 1 (8.3) Constriction of superior vena cava by pericardium 1 (8.3)
Roswit et al. (1953)	51	47 (91.9)	4 (8.1)			
Schechter (1954)	22	20 (90.8)	1 (4.6)			Mediastinal and cervical lymphnode involvement with active tuberculosis 1 (4.6)
Calkins (1956)	21	11 (52.4)	2 (9.5)			Thyroid 2 (9.5) Undiagnosed 6 (28.6)
Bruckner (1958)	23	19 (82.6)	1 (4.4)	1 (4.4)	2 (8.6)	

from chronic luetic and tuberculous mediastinitis have decreased owing to the development of anti-luetic and anti-tuberculous treatment and on the contrary idiopathic chronic fibrous mediastinitis shows a tendency to increase gradually. Although the etiology of idiopathic chronic fibrous mediastinitis is of course undetermined, some antecedent etiological factors such as upper respiratory infection,^{13), 17), 18)} trauma,^{4), 18)} bronchopneumonia^{1), 4)} and others^{1), 17)} can be often detected.

In our case, a significant formation of collateral circulation was observed as the dilatation and tortuosity of veins of thoracic and abdominal walls and was clearly ascertained in phlebogram. As a result of remarkable development of collateral circulation, the venous pressure was found to be only slightly elevated and no delay in circulation time nor edema in the upper body were observed. Of venous blood coming from the right jugular vein, a part flowed into the right external thoracic vein, while most of venous blood coming from the left jugular vein flowed into the left external thoracic vein. Venous blood of each side was considered to flow into the inferior vena cava through the thoracic subcutaneous vein, abdominal subcutaneous vein, superficial epigastric vein and femoral vein.

Collateral circulation of this case might arise gradually owing to the formation of the venous plexus-like anastomosis, the displacement of the left brachiocephalic vein to the right side and the stenosis of the bilateral brachiocephalic veins, as shown in Fig. 4, 5 and 6.

Our case, although the possibility is still remained in the congenital abnormality of the brachiocephalic vein, may principally result from the postoperative disorder of bilateral clavicular osteomyelitis. Spondylitis ankylopoetica totalis and the persistence of bilateral active chronic clavicular osteomyelitis may play a supplementary role in the formation of the superior vena caval syndrome.

SUMMARY

A case of the superior vena caval syndrome which presumably resulted from the postoperative disorder of bilateral clavicular osteomyelitis is described.

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