TUBERCULOUS AORTIC-OESOPHAGEAL FISTULA
REPORT OF A CASE

By Lim Cheng Hong,
(Medical Unit II, General Hospital, S'pore)
and K. Sugai,
(Pathology Department, General Hospital, S'pore)

It is well-recognised that tuberculosis can affect blood vessels, and an endarteritis is a common finding in the walls of tuberculous cavities in the lungs. However, larger vessels like the aorta are less commonly affected, and in spite of the high incidence of the disease in this region, there have been no reports of this complication in the local literature. It may be that these lesions have not been particularly looked for, or opportunities for necropsy in these cases have not arisen. It is, therfore, considered worthwhile to record the following case of miliary tuberculosis, who died after a massive haematemesis from a rupture of the aorta into the oesophagus.

CASE REPORT

A 28 year old Chinese housewife, T.K.S., was admitted to General Hospital, Singapore on 7th April 1964. She had been well until one month before admission when she developed a cough, productive of whitish sputum. The cough was worse at night. There was no associated fever, chills or chest pain. She was treated by a General Practitioner with cough mixtures, and improved after two weeks. An X-ray chest taken at S.A.T.A. was reported as being normal.

Two weeks before admission, she developed a cough again, also productive of whitish sputum. This time she had a fever associated with chills, rigors and sweats. The fever was quite high, and intermittent in character. It was worse in the mornings and nights. Other symptoms she complained of were anorexia, lethargy and generalised weakness. There was no haemoptysis. Bowel movements and micturition were normal. She had amenorrhoea for the preceding two months.

PHYSICAL EXAMINATION

On examination, patient was pale, thin and had a temperature of 101°F. There were no enlarged lymph nodes. The head and neck, respiratory system, cardiovascular system and entral nervous system were clinically normal. B.P. was 130/80 and all the peripheral pulses were equal and normal. In the abdomen, the liver was enlarged, 3 fingers breadth below the costal margin, soft with a smooth edge. The spleen was just palpable. A diagnosis of P.U.O. was made.

INVESTIGATIONS

Investigations done showed a haemoglobin of 86%, total white cell count of 7,200/cu. mm. with a normal differential count. Blood films for malarial parasites were negative, and blood culture grew B. alkaligenes. Blood for Widal and Weil-Felix, Paul Bunnell Test and agglutination for B. abortus and B. melitensis were all negative. The erythrocyte sedimentation rate was 12 mm. in the 1st hour (Westergren). The sputum was repeatedly examined for acid fast bacilli, but without success. Urine examination showed a trace of albumin and on microscopy, there were occasional R.B.C. and W.B.C. The Mantoux Test 1:1000 was negative but 1:100 was strongly positive. X-ray chest showed normal cardiac size and configuration. There was a suggestion of mottled shadowing fairly evenly distributed throughout both lungs.

PROGRESS

While in the ward, the patient continued to have a swinging temperature between 99°F and 104°F, associated with chills and occasionally rigors.

In view of the clinical picture, hepatosplenomegaly and radiological findings, she was treated as a case of miliary tuberculosis with Streptomycin and Isoniazid. She seemed to respond to treatment, the fever being reduced to lower levels by the 4th day of treatment.

Fig. 1. Tuberculous aortitis. Thoracic aorta shows perforation.

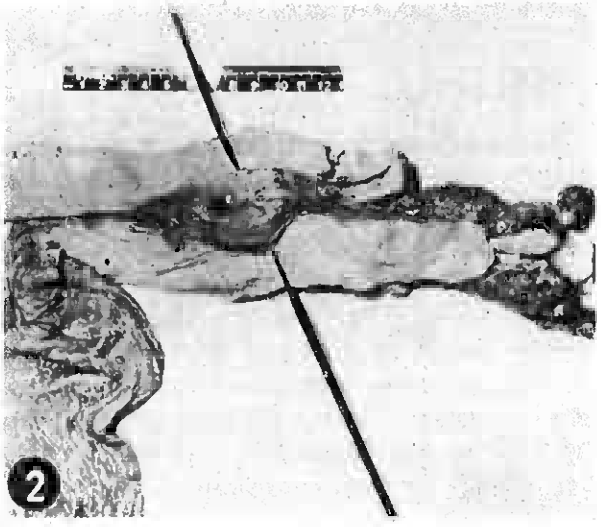

Fig. 2. Tuberculous aortitis. Perforation of the aorta covered by a thick granulation tissue in the mediastinum.

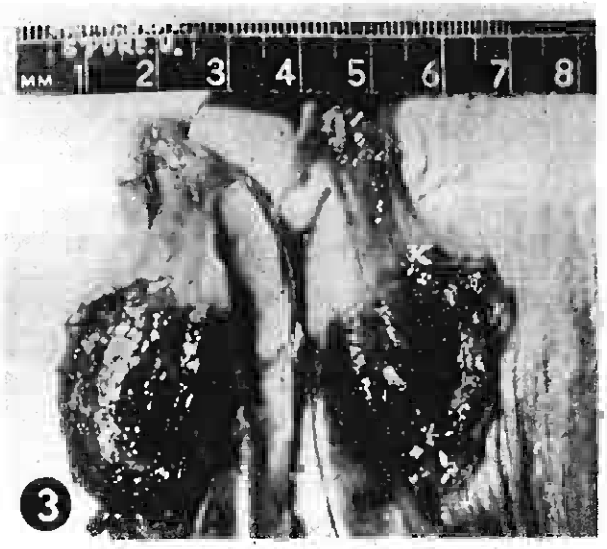

Fig. 3. Tuberculous aortitis. Oesophago-aortic fistula in granulation tissue.

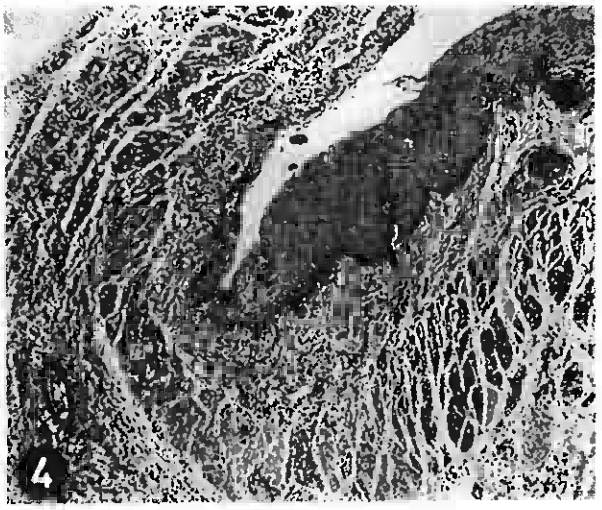

Fig. 4. Photomicrograph illustrating granulation tissue with epithelioid and giant cell tubercles at the edge of the perforation of the oesophagus.

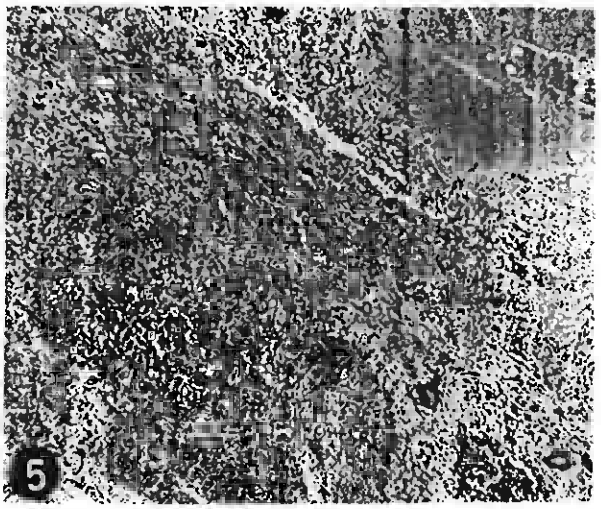

Fig. 5. Photomicrograph illustrating tubercles and caseous foci in the adventitia of the aorta near the perforation. Lower portion of the picture is that of a tuberculous lymphadenitis which is continuous to the adventitia. (H-E, X 45).

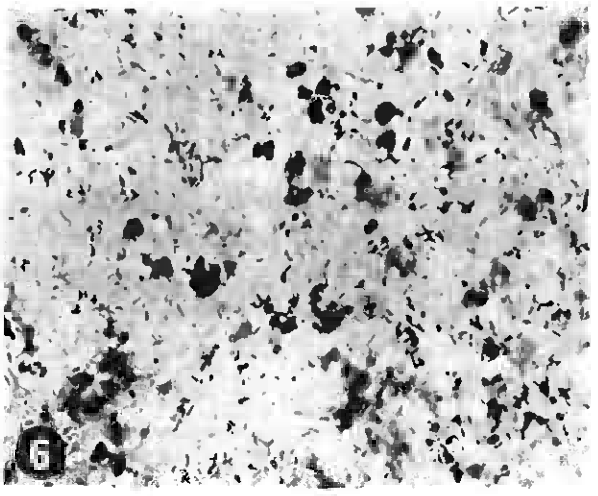

Fig. 6. Photomicrograph illustrating strongly positive acid-fast bacilli at the margin of the perforation of aorta. (Z-N. X 500).

However, a few days later, the temperature began to rise again. One morning, about two weeks after admission, she complained of retrosternal pain on swallowing. No obvious cause for this new symptom was found. The same evening, patient was sitting in a chair when she suddenly slumped forward, and appeared to lose consciousness. Fresh blood gushed out from her mouth and nose. She also vomited some blood and food material, became cyanosed, gasping for air. She was carried to her bed, but died almost instantaneously.

At Autopsy (A810/64): The body was that of a thin Chinese female young adult. There was marked pallor of the skin. A small amount of blood oozed out of the mouth and nostrils.

Skeletal and Nervous Systems: n.a.d.

Cardio-Vascular System: Heart n.a.d. Aorta showed a perforation (Fig. 1) 0.5 cm. in diameter in right anterior aspect of thoracic portion at the point 2 cm. below a closed ductus arteriosus. The perforation was externally covered by a thick granulation tissue (Fig. 2) 8 × 4 × 3.5 cm., probing of which revealed a fistula 3.5 cm. long, 0.6 cm. in diameter. Through this fistula the aorta communicated with the oesophagus. The granulation tissue was haemorrhagic and densely adherent to the oesophagus, showing caseous lymph nodes embedded. No particular foreign bodies were found in this granulation tissue.

Respiratory System: Both lungs were pale pink, well aerated, but revealed widespread miliary tubercles throughout the cut surfaces.

Digestive System: Oesophagus showed a perforated ulcer (Fig. 3) which was 3.5cm. long and 1.5 cm. wide at the point 10 cm. below an interarytenoid fold. The floor of the ulcer was composed of a haemorrhagic granulation tissue in the center of which there was a perforation communicating with above said fistula. The stomach contained about one litre of fluid and clotted blood. Liver showed pale parenchyma with miliary tubercles. Gall bladder and bile tract n.a.d.

Urinary System: Kidneys pale in colour, showing on cut surfaces some tubercles. Ureters and urinary bladder n.a.d.

Endocrine System: Thyroid showed a few tubercles. Adrenals n.a.d.

Haematopoietic System: Spleen pale red and soft, with miliary tubercles. Peribronchial and peritracheal lymph nodes were enlarged and caseous.

Reproductive System: Uterus, bilateral ovaries and tubes n.a.d.

Microscopically: Sections through the granulation tissue revealed a specific granulation tissue containing epithelioid and giant cell tubercles and areas of caseation necrosis. Caseation and tubercles together with haemorrhages were also noted at the margin of perforation on both aortic and oesophageal sides. (Fig. 4).

The adventitia of the thoracic aorta near the perforation contained tubercles and caseous foci, and was in continuity with tuberculous lymph nodes (Fig. 5).

Acid-fast organisms were strongly positive at the margins of the perforation in aorta (Fig. 6) and oesophagus, within granulation tissue and lymph nodes.

Miliary tuberculosis in lungs, liver, kidney, spleen and thyroid confirmed.

Anatomcal Diagnosis: Tuberculous aortitis of the thoracic aorta with perforation into oesophagus and resulting in massive haemorrhage. Tuberculous lymphadenitis of peribronchial, peritracheal and mediastinal lymph nodes. Miliary tuberculosis of the lungs, liver, kidney, spleen and thyroid.

DISCUSSION

There have been many reports and reviews of tuberculosis affecting the aorta in the literature 1-14. The diagnosis in most of these cases was made either at autopsy or at operation, and there does not seem to be any way of making a definite clinical diagnosis ante-mortem. However, the development of an aortic aneurysm in a patient with extensive tuberculosis should lead one to recognise the possiblity of tuberculous aetiology.

Wetteland and Scott (1956) reported a case of tuberculous aortic aneurysm which had ruptured into the respiratory tract, the lesion having spread from softening of hilar lymph nodes of a primary pulmonary tuberculous complex. They observed from their review of the literature that rupture usually followed the formation of a false aneurysm, and this occurred mostly into a cavity or hollow viscus, for example, peritoneal cavity, oesophagus, stomach, duodenum, jejunum, pericardium and trachea.

Meehan et al (1957) reviewed 19 cases of T.B. aortitis, and all the cases excepting one, had aneurysmal formation. Their own case had a dissecting aneurysm of the abdominal aorta, the dissection occurring in the adventitia instead of the usual location in the media.

Stiefel J.W. (1958) noted that of the 21 cases of tuberculous aortic aneurysm reported by Gellerstedt and Swafenburg in 1933, 12 cases ruptured. Four cases of aortic perforation did not have aneurysmal formation, as in the present case reported.

Volini et al (1962) reviewed 99 cases reported in the literature and also three cases of their own. They also observed the high incidence of rupture with fatal haemorrhage in cases with aneurysmal formation.

It would then appear that cases of tuberculosis affecting the aorta with aneurysm carry a grave prognosis if left untreated. There have been a few reports of surgical repair in these cases. In 1956, Rob and Eastcott reported the first case of successful resection of an aortic aneurysm of tuberculous aetiology, and treatment with a prosthesis. There have been other reports since including de Prophetis et al (1959), Kline and Durant (1961), Penn I (1962), Yeoh, C.B. et al (1963).

This is an uncommon case of miliary tuberculosis complicated by perforation of the aorta into the oesophagus, and the formation of an aortic-oesophageal fistula. There is a high incidence of miliary tuberculous infection in the cases of tuberculosis of the aorta reported in the literature. This is easily understood when one considers the widespread involvement of lymph glands, bone and the bacteraemia in miliary tuberculosis. At necropsy in this patient, there was extensive miliary infection, and practically every organ in the body was studded with tubercles. It would appear that the infection in this case spread from a mediastinal lymph node which had eroded into the aorta and the oesophagus. The significance of the retrosternal pain on swallowing was not recognised, and in retrospect would appear to have been caused by the oesophageal ulceration and impending rupture. The possibility of a foreign body causing an aortic-oesophageal fistula in this case was considered, but there was no evidence of this and histology confirmed the tuberculous aetiology.

SUMMARY

A case of miliary tuberculosis complicated by an aortitis and rupture of the aorta into the oesophagus is reported. A brief review of the literature on the subject is made.

The difficulty of making a clinical diagnosis is stressed. The mode of infection in this case appeared to be an extension of the tuberculous process from a mediastinal lymph node into the aorta and the oesophagus. Necropsy showed that this resulted in the formation of an aortic oesophageal fistula and fatal haemorrhage.

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