

Non-typhoid *Salmonella* mycotic aneurysm of the aortic arch

Stella PY Wong 黃佩茵
Thomas KK Lai 賴國強
WL Ng 伍煥良
WK Luk 陸衛光

Salmonella mycotic aneurysm is a rare but potentially fatal condition. Mortality is high without timely intervention. The clinical presentation is protean and early diagnosis requires a high degree of clinical alertness. Prompt surgical intervention and prolonged antimicrobial therapy are keys to successful treatment. We report an 81-year-old man with an atypical presentation of *Salmonella* mycotic aneurysm in the aortic arch. The case highlights the need to evaluate all patients over 50 years with non-typhoid *Salmonella* bacteraemia for possible endovascular infections. Contrast-enhanced computed tomography is useful for making an early diagnosis of this disease.

Case report

An 81-year-old man with a history of laparoscopic cholecystectomy and diabetes mellitus was admitted thrice to Tseung Kwan O Hospital in August 2003 for fever. During the first two episodes he also complained of prostatism and dysuria and a tentative diagnosis of urinary tract infection was made. Routine blood tests were unremarkable except for the following abnormal liver function tests: alkaline phosphatase 218 U/L (reference range, 40-129 U/L), and alanine aminotransferase 99 U/L (reference level, <41 U/L). Repeated urinary cultures were negative and he was discharged after resolution of the fever with antibiotic therapy.

During his third admission he presented with fever, vague abdominal pain, cough, and sputum for 1 week. His laboratory results were essentially normal except for persistently abnormal liver function tests and an erythrocyte sedimentation rate (ESR) raised to 95 mm/h (reference range, 2-10 mm/h) and C-reactive protein (CRP) level of 80.6 mg/L (reference level, <3.0 mg/L). A chest radiograph (CXR) done on admission was normal. He was not jaundiced and had no abdominal tenderness or hepatomegaly. An ultrasound of the abdomen showed increased periportal echogenicity without ductal dilatation and a small left basal pleural effusion. The septic work-up was repeated and the patient was treated empirically with cefoperazone/sulbactam.

On day 5, his blood cultures grew group D *Salmonella* sensitive to cefotaxime, ceftriaxone, and levofloxacin. His fever subsided on day 10 after a change of therapy to ciprofloxacin but the patient suddenly developed mild haemoptysis on day 11. A repeated CXR showed a new ill-defined shadow in the medial aspect of the left upper lung field adjacent to the aortic knuckle (Fig 1a). A bronchoscopic examination was declined by the patient but a contrast-

Key words

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Tseung Kwan O Hospital, Tseung Kwan O, Hong Kong:
Department of Medicine
SPY Wong, MRCP
Department of Pathology
WK Luk, FRCPA, FHKAM (Pathology)
Department of Radiology
TKK Lai, FRCP, FHKAM (Radiology)
Department of Medicine and Geriatrics,
United Christian Hospital, Kwun Tong,
Hong Kong
WL Ng, FRCP (Glasg), FHKAM (Medicine)

Correspondence to: Dr WK Luk
E-mail: lukwk@ha.org.hk

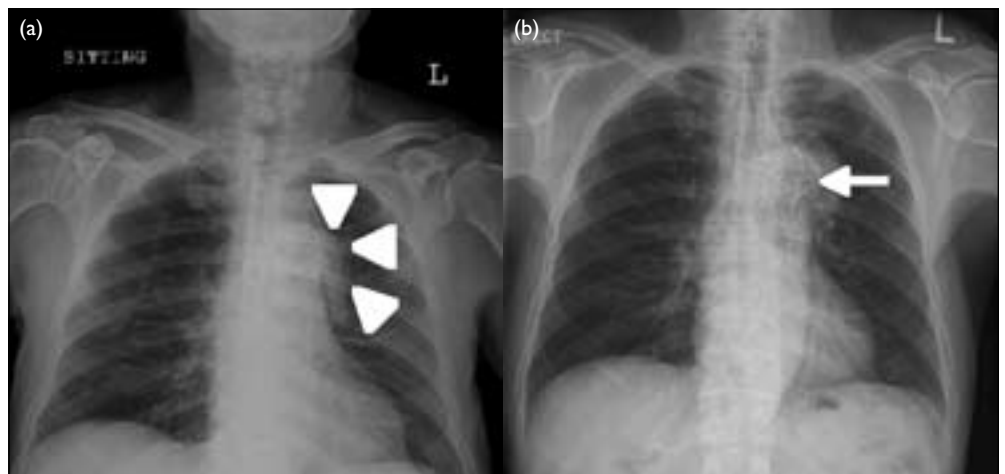


FIG 1. (a) A chest radiograph on day 11 showed an abnormal opacity around the aortic knuckle (arrowheads). (b) Endovascular graft in situ at the aortic arch region (arrows)

enhanced computed tomography (CT) scan of the thorax performed on day 18 showed a 3.5 x 1.9 cm distal aortic arch saccular aneurysm with adjacent haemorrhage (Fig 2a, b). His haemoptysis became more profuse and the patient was promptly transferred to the surgical unit. Endovascular stenting of the aortic arch aneurysm was performed on day 19 and the patient had an uneventful recovery (Fig 1b). His ESR decreased and the liver function tests returned to normal. Unfortunately, the patient suffered an ischaemic stroke 6 months after his initial presentation. A CT scan done at that time showed a patent endovascular graft with no evidence of leakage or thrombosis (Fig 2c, d). The operative risks and the patient's preferences were taken into consideration and a decision was made to put the patient on life-long oral levofloxacin instead of proceeding to further grafting surgery.

Discussion

Non-typhoid *Salmonella* are food-borne pathogens that commonly cause a mild, self-limiting gastroenteritis. A transient bacteraemia may occur in 2 to 8% of these patients.¹ The species appears to have a predilection for invading damaged endothelium in the heart and arterial walls¹ leading to a spectrum of cardiovascular infections including mediastinitis, pericarditis, endocarditis, aortitis, mycotic aneurysms, and infection of cardiac devices.¹⁻⁴ In one large Taiwanese series, the common serogroups responsible for endovascular infections were group C (47%), group D (32%), and group B *Salmonella* (21%).⁵ Advanced age and diabetes mellitus, both of which were likely to contribute to significant atherosclerosis, as evidenced by the occurrence of ischaemic stroke, were risk factors predisposing our patient for *Salmonella* endovascular infection.

The reported incidence of endovascular infections in patients with non-typhoid *Salmonella* bacteraemia ranges from 9 to 10% in western countries^{1,4} to 16.2% in a recent Taiwanese series.² The most frequent sites for infective aortitis include the infrarenal abdominal aorta, followed by the thoracic and suprarenal abdominal aorta.^{3,5} Mycotic aneurysms of the aortic arch are reportedly rare.⁶ Manifestations of endovascular infections can be protean and non-specific. Common presentations include fever, unremitting sepsis, breakthrough and relapsing bacteraemia. Localised symptoms may relate to pressure effects from aneurysms, contagious extension of infection, or impending aneurysmal rupture.^{3,7} Rare presentations include psoas and pelvic abscesses, vertebral osteomyelitis, endobronchial masses,⁸ haemoptysis,⁹ and gastro-intestinal bleeding due to aortobronchial or aortoduodenal fistula formation.³ Failure to consider these unusual complications of non-typhoid *Salmonella* bacteraemia can lead to catastrophic consequences. In a report by Dransfield and Johnson,⁸ the possibility that a mycotic aneurysm may present

主動脈弓的非傷寒沙門氏桿菌感染性動脈瘤

沙門氏桿菌感染性動脈瘤是一種罕見但可致命的疾病，患者沒有得到及時治療會有高死亡率。此症臨床病徵多變，醫護人員需具備極高的警覺性，才可在發病初期確診此症。及時施行手術並延長抗生素治療是成功醫治這疾病的關鍵所在。本報告記述一位81歲男性患者，在主動脈弓裏發現非典型沙門氏桿菌感染性動脈瘤。本病例揭示，50歲以上患非傷寒沙門氏桿菌血症病者，都需要評估是否有血管腔內感染。造影加強的電腦斷層照相術有助在疾病早期診斷此症。

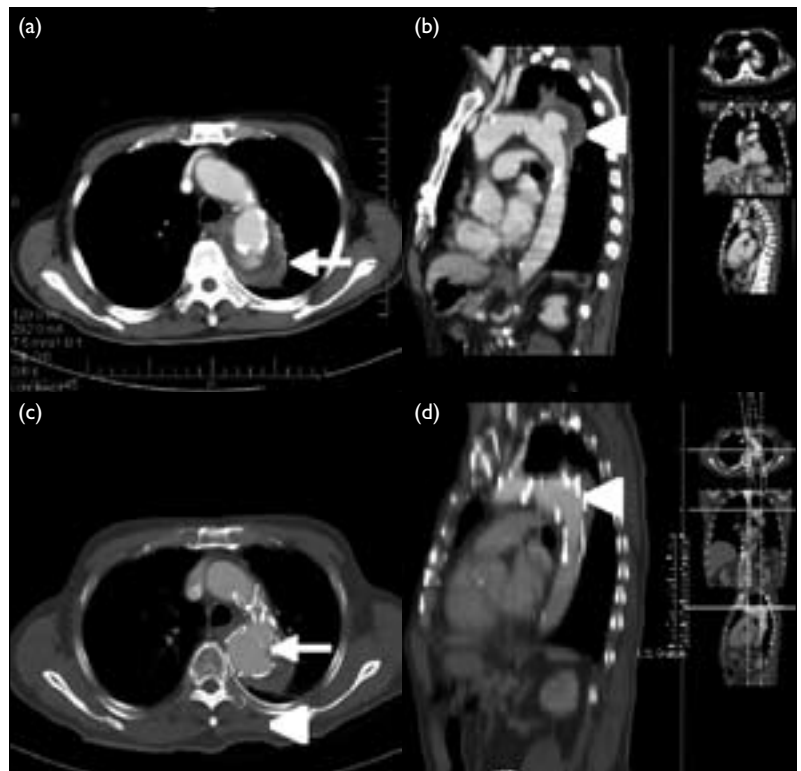


FIG 2. Contrast-enhanced computed tomographic scans showing (a) focal aneurysmal dilatation at the level of aortic arch surrounded by poorly enhancing periaortic soft tissue density (arrow), (b) oblique sagittal reconstruction image with thoracic aorta in profile, demonstrating the position of the aneurysm (arrowhead) in relation to the aorta. (c) Follow-up scan 6 months after operation in widened window width setting showing the hyperdense stent graft in situ (arrow) at the aortic arch with no evidence of contrast leakage. (d) Oblique sagittal reconstruction of image in (c) demonstrating the exclusion of the aneurysm from the aortic lumen by the stent graft (arrowhead)

as an endobronchial mass was not suspected and the lesion was biopsied, resulting in a massive and fatal haemorrhage.

In the past, angiography with arterial catheterization was the gold standard for diagnosing infected aneurysms. With the advent of fast multi-detector spiral scanners, CT scanning has become the preferred initial investigation in patients with suspected arterial aneurysms.^{7,10} Computed tomographic features suggestive of infective aneurysms include periaortic soft tissue density with

rim enhancement due to periaortic inflammation and haematoma. Other signs include an eccentric, thickened aortic wall without calcification; periarterial collections or adjacent osteomyelitis, and gas in the aneurysmal sac.³ Angiography is now mainly reserved for cases where interventional procedures like endovascular stent insertion are being planned, as in our case. Changes in plain radiographs can often be subtle but can still provide a major clue. The sudden appearance of new periaortic infiltrates on our patient's CXR along with *Salmonella* bacteraemia and fresh haemoptysis led to a prompt diagnosis and early surgical intervention. This patient's mycotic aneurysm could have been diagnosed earlier if patients older than 50 years with non-typhoid *Salmonella* bacteraemia are actively evaluated for possible endovascular infections.¹ This is especially true if the patient has underlying risk factors such as pre-existing atherosclerotic diseases (peripheral vascular disease, previous ischaemic strokes, and coronary artery disease) as well as the traditional cardiovascular risk factors including diabetes mellitus and hypertension.^{2,3}

Soravia-Dunand et al⁷ reviewed 140 cases of aortitis due to *Salmonella* and reported that increased survival was associated with the early use of bactericidal antibiotics and early surgical intervention. In their review, the mortality rate was 40% for patients who received combined surgical and medical treatment while 96% of those patients who received medical therapy alone died.⁷ Different antimicrobial regimens with varying success rates have been described, including combinations of ampicillin or cefotaxime with gentamycin, ceftriaxone alone or fluoroquinolones.³ A case series found that among 34 *Salmonella* isolates from people with infective aortic aneurysms, 65% were ampicillin resistant and 41% were resistant to ciprofloxacin but none was resistant to cefotaxime.⁵ Therefore, third-generation cephalosporins are recommended as the initial drugs of choice for *Salmonella*-induced aneurysms.

Early surgical intervention is the cornerstone of treatment for endovascular infections. Conventional surgical treatment of mycotic aneurysms consists of wide debridement of the infected tissues, extra-anatomic revascularisation or in-situ graft replacement with resection of the infected aneurysm and insertion of an end-to-end tube graft.^{3,4,11} More recently, endovascular repair is emerging as an alternative therapy for patients with high operative risks prohibiting open surgery as in our patient. Endovascular stent graft repair obviates

the need for thoracotomy, aortic cross-clamping and extracorporeal circulation and is associated with reduced blood loss and possibly decreased perioperative morbidity and mortality.¹² Ting et al¹³ and Kotzampassakis et al¹⁴ have reported encouraging short- and medium-term success. Endovascular procedures do not allow surgical debridement and removal of the infected tissues so delayed septic and haemorrhagic complications may occur. Concerns about the durability of endoscopic stent grafts have also been raised.¹⁵ We suggest an endograft may be considered an effective temporary measure while preparing the patient for a more definitive elective intervention. More long-term follow-up data are needed to help define the role and durability of this surgical approach. Prolonged antibiotic therapy and long-term follow-up are mandatory.

There is no consensus regarding the ideal duration of systemic antimicrobial therapy. As a general rule, postoperative antimicrobial therapy for 6 to 8 weeks seems adequate for preventing a relapse.³ Lifelong therapy is suggested in some cases.^{3,7,12} Biochemical parameters of inflammation (white cell count, CRP, and ESR) may help with disease monitoring and to guide the duration of antibiotic treatment. Lifelong suppressive antibiotic therapy was offered to our patient because of the unacceptable operative risks he faced and the concern that late recurrent infections of in-situ prosthetic material may occur.

Conclusion

We have reported a rare case where a man with an aortic arch mycotic aneurysm due to *Salmonella* presented with recurrent fever and haemoptysis. The importance of prompt recognition and combined medical and surgical treatment is illustrated by this case. Patients presenting with non-typhoid *Salmonella* bacteraemia associated with fever or unremitting sepsis, unexplained symptoms of back, abdominal or chest pain, or haemoptysis should be expeditiously and extensively evaluated for possible infective aortitis, especially if they are older than 50 years and have risk factors for atherosclerosis or reduced immunity. A targeted CT scan should be performed urgently. If a mycotic aneurysm is found, surgical resection with in-situ bypass grafting should follow as soon as possible. Endovascular repair is an effective alternative in selected patients. Adequate preoperative and postoperative antimicrobial therapy is imperative.

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