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Clinical Case Report

A case of *Brucella* aortitis associated with development of thoracic aortic aneurysm and aortobronchial fistula

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ABSTRACT

Objectives: The purpose of this case report is to document the occurrence of granulomatous aortitis complicated by formation of a saccular aneurysm and aortobronchial fistula due to *Brucella* infection.
Methods: A 65-year-old man with a history of feral swine hunting presented with hemoptysis and was found to have a saccular thoracic aortic aneurysm and associated aortobronchial fistula. The aneurysm underwent operative repair with closure of the aortobronchial fistula.
Results: Histopathological examination of the aneurysm wall revealed evidence of granulomatous aortitis. Cultures of the blood and aortic wall tissue were positive for *Brucella suis*.
Conclusions: Although rare, *Brucella* infection should be considered in the differential diagnosis of aortic aneurysm with granulomatous aortitis.

1. Case presentation

The patient is a 65-year-old man with a medical history of hypertension and hyperlipidemia. The patient originally presented to an outside hospital with massive hemoptysis and was found to have an aortobronchial fistula with an associated saccular descending thoracic aortic aneurysm that was treated by a thoracic endovascular aortic repair (TEVAR) procedure. He recovered uneventfully with resolution of the hemoptysis. An infectious etiology of the fistula was not suspected; therefore, no cultures were collected and no antimicrobial therapy was given.

Twenty months later, the patient then presented to a regional hospital with recurrent hemoptysis and was treated with antibiotics for a presumed pneumonia. After evaluation with bronchoscopy, he was found to have a clot in the left lower lobe of the lung, with high suspicion for aortobronchial fistula as the result of the prior TEVAR procedure. He was transferred for repair of the aortobronchial fistula.

The patient underwent resection and graft replacement of the descending thoracic aortic aneurysm using a Dacron tube graft with extensive resection and debridement of the aortic wall and thrombus. An aneurysm of 5 cm in size extending from the thoracic level of T4 to T8 was found with erosion into the superior segment of the left lower lobe of the lung (Image 1A). The superior left lower lobe of the lung was surgically resected due to chronic abscess and purulence. A biopsy of the aortic wall along with blood and sputum was sent for culture (Image 2A). A pedicled omental flap was created and used to completely wrap the newly replaced Dacron graft also providing a vascularized barrier between the Dacron graft and the remaining lung parenchyma (Image 1B). The patient awoke neurologically intact, extubated, and had an uneventful convalescence.

Histology of the aortic wall tissue revealed severe granulomatous aortitis, with extensive inflammation of the intima, lymphohistiocytic infiltrates with multinucleated giant cells, foci of necrosis, and fibrous thickening of the adventitia and vasa vasorum (Image 2B and C). Gram, GMS, PAS, Fite, Warthin–Starry, and AFB stains were negative. The left lower lobe lung tissue sections showed extensive hemorrhage and organizing pneumonia with acute neutrophilic inflammation, necrosis, and chronic lymphocytic inflammation (Image 2D), likely caused by the chronic fistula.

After 3 days of incubation, the blood culture was positive, and Gram stain revealed gram-negative coccobacilli (Image 3A). Culture from the blood and the aortic wall grew a tiny gram-negative bacilli that was oxidase and urease positive, which was suspicious for *Brucella*. Isolates were sent to the City of Houston Laboratory and were confirmed by polymerase chain reaction to be *Brucella suis*. The patient was discharged home with 6 weeks of IV 100 mg doxycycline.

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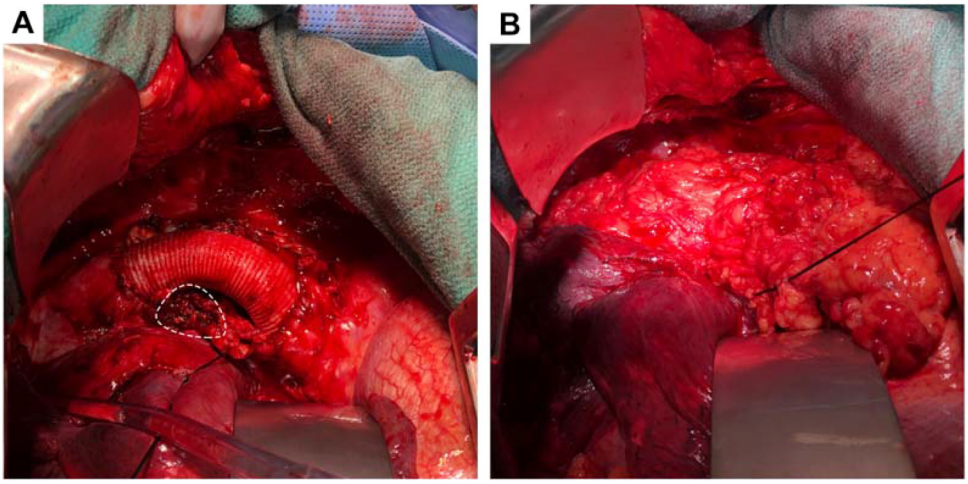

Image 1. (A) Intraoperative image of aortic aneurysm repair showing site of aortobronchial fistula (dotted line). (B) A pedicled omental flap wraps the newly replaced Dacron graft to provide a vascularized barrier between the Dacron graft and the remaining lung parenchyma.

On follow-up questioning, the patient reported being an active feral swine hunter, engaging in both field dressing and consumption of the animals.

2. Discussion

The differential diagnosis for this pathological case was based on the inflammatory pattern observed in the histology of the aorta. Of the four major inflammatory patterns [1], this case most closely aligned with a granulomatous/giant cell pattern, consisting of clusters of epithelioid macrophages with or without giant cells or granulomas. Noninfectious causes of granulomatous/giant cell pattern include giant cell aortitis, Takayasu's aortitis, and granulomatosis with polyangiitis. Common infectious causes include syphilis, tuberculosis, and fungus, although brucellosis is a rare presentation. Positive blood cultures for *Brucella* prompted further questioning of the patient, revealing his proclivity for hunting, field dressing, and consuming wild boar.

Mycotic aneurysm results from infection of the arterial wall. Four different routes of invasion by microorganisms of the arterial or aortic wall have been identified. These are (1) implantation of bacteria on a

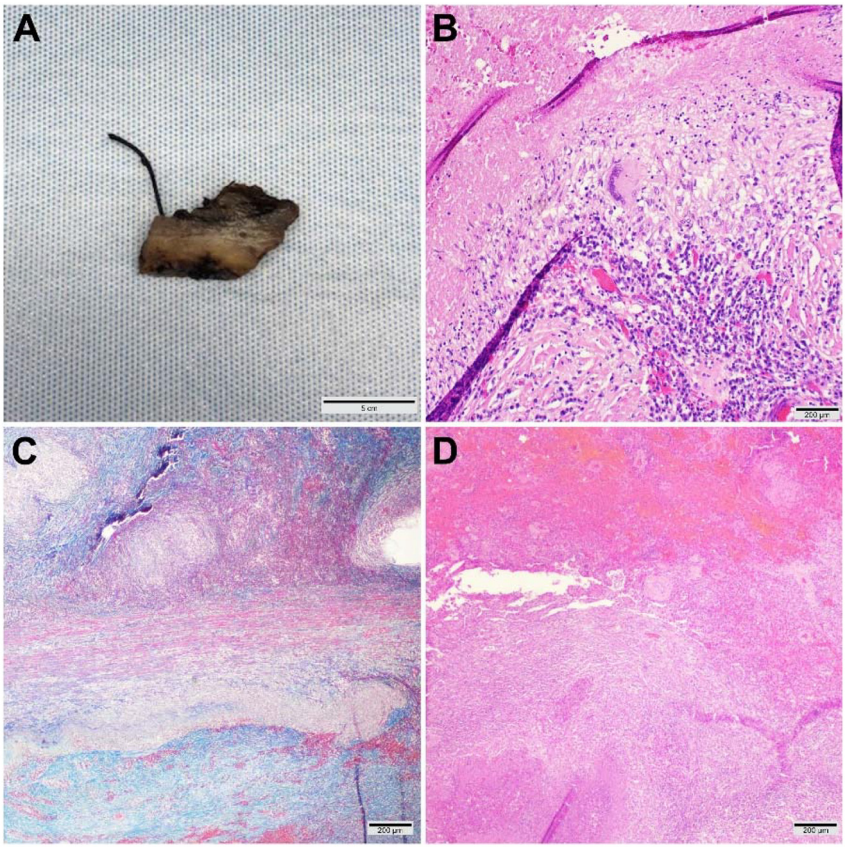

Image 2. (A) Gross image of the aortic wall. (B) Histology of aortic wall showing granulomatous inflammation. (C) Trichrome stain of aortic wall fibrous thickening of the adventitia and vasa vasorum. (D) Histology of the lung showing extensive hemorrhage and organizing pneumonia with acute neutrophilic inflammation, necrosis, and chronic lymphocytic inflammation.

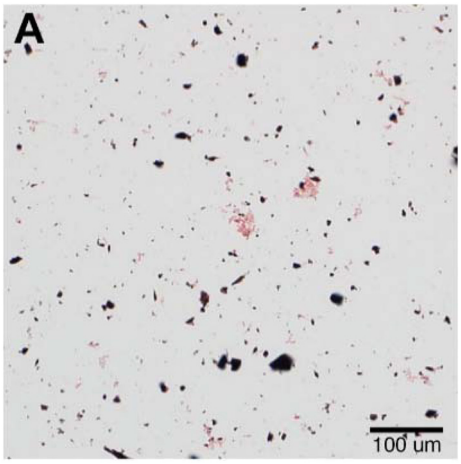

Image 3. Bacterial culture slide image showing gram negative coccobacilli.

normal or abnormal intimal surface, (2) embolization by bacteria into the vessel wall via the vasa vasorum, (3) direct invasion of the vessel from a contiguous extravascular site of infection, and (4) traumatic inoculation of infected material into the vessel wall [2]. An apparently normal aorta, an atherosclerotic plaque, and a preexisting aneurysm may be infected by any one of these four mechanisms. Infection of the arterial wall may occur by seeding during bacteremia or by septic emboli to the vasa vasorum with infective endocarditis. Circulating microorganisms can colonize (1) a nonaneurysmal aorta, with or without atherosclerotic plaques (infective aortitis); (2) a preexisting aortic aneurysm; or (3) a posttraumatic false aneurysm [2,3].

In retrospect, the patient likely developed a mycotic aneurysm resulting from the *Brucella* aortitis, leading to its invasion of the adjacent lung parenchyma and causing initial massive hemoptysis. Appropriately, the patient was initially treated using TEVAR to address the immediate hemoptysis, to become stabilized, and to allow recovery. It has been our view, however, that this approach for aortobronchial fistulas resulting from mycotic thoracic aneurysms is only a temporizing measure or a "bridge" to a more definitive procedure, which was what this patient ultimately underwent.

Infection of the aorta by *Brucella* is a relatively rare event; a systemic review of the literature found only 44 cases of *Brucella* aortitis [3]. However, due to the difficulty of diagnosis of brucellosis, the true number of cases may be underreported. The majority of cases (43%) were reported in Mediterranean countries, with 11% of cases in North America. The ascending and abdominal aorta were the most common sites of localization, and the most common presentations of brucellosis were mycotic aneurysm followed by aortic root abscess and pseudo-aneurysm, with a high percentage of complications including spine involvement, rupture, and fistulization [3].

Brucellosis results in a chronic granulomatous infection caused by intracellular bacteria. Brucellosis is endemic mainly in California and Texas, which make up over 50% of all U.S. cases [4,5]. Mechanisms of human infection include direct contact of mucosal membranes or through lacerations in the skin, consumption of infected meat or milk products, and inhalation of the aerosolized bacteria [6]. Infection by the *Brucella* organism involves invasion into the mucosa, where it is ingested by phagocytes, most are eliminated by phagolysosomes, some will survive and replicate in the endoplasmic reticulum, inducing a host response from natural killer cells and T lymphocytes [7,8]. There are 10 species in the *Brucella* genus, with the most common pathogenic species to humans consisting of *B. mellitensis*, *B. abortus*, and *B. suis*. *B. suis* is believed to be a sexually transmitted disease among feral swine, as it has been isolated from the sexual organs of boars; there is minimal evidence of clinical illness or effect on viable pregnancies [9]. Human infection by *B. suis* is relatively rare compared to other species. Recently reported cases in the literature have all been associated with contact during feral swine hunting [10–12].

Brucella can be difficult to diagnose as it is a slow-growing organism in culture, although molecular testing techniques such as multilocus sequence typing, multiple-locus variable number tandem repeat analysis, and whole-genome sequencing are increasingly being utilized [4]. Due to the patient's history of field dressing wild boar in a geographical area with endemic *Brucella* in feral swine, we hypothesize that he was exposed to *Brucella* hematogenously during field dressing, which colonized the aorta to form the mycotic aneurysm, leading to the formation of the thoracic aortic aneurysm and aortobronchial fistula, causing the patient to present with hemoptysis. Although relatively rare, *Brucella* infection should be considered in the differential diagnosis of aortic aneurysm with granulomatous aortitis.

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