

## CLINICAL VIGNETTE

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# *Bordetella holmesii* Bacteremia and Endocarditis in a Patient with a History of Splenectomy

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### **Introduction**

Asplenia may be due to either functional or anatomical purposes, such as splenectomy associated with trauma or splenectomy indicated for a variety of hematologic diseases.<sup>1</sup> Regardless of etiology, patients with asplenia are at an increased risk for severe bacterial infections, including systemic infections caused by Gram-negative organisms.<sup>1,2</sup> The following case describes a patient with a history of childhood Hodgkin's lymphoma and history of splenectomy, who presented with symptoms of new-onset congestive heart failure. He was ultimately found to have *Bordetella holmesii* bacteremia complicated by infective endocarditis requiring surgical intervention.

### **Case Presentation**

A 49-year-old man presented with a 2-week history of shortness of breath, dyspnea on exertion, and bilateral lower extremity swelling. He had never experienced these symptoms previously. The patient also complained of a constant, non-productive cough for 3 days. He denied any fevers, chills, hemoptysis, chest pain, or palpitations. He denied any recent travel or sick contacts. The remainder of his review of symptoms was unremarkable. His past medical history was significant for a congenital heart murmur, obstructive sleep apnea, and childhood Hodgkin's lymphoma, treated with chemotherapy, radiation, and splenectomy at age 10. He denied any current or prior smoking or alcohol abuse, and his family history was noncontributory.

On initial presentation, his vitals were notable for a temperature of 37.2°C, blood pressure 97/53, heart rate 104, respiratory rate 16, and oxygen saturation of 95% on room air. Physical examination was significant for 2/6 early systolic murmur loudest in the right upper sternal border, rales bilaterally, and 1+ bilateral lower extremity edema to the distal thighs. Laboratory evaluation was notable for a white blood cell 17.8 x 10<sup>9</sup>/L, basic natriuretic peptide 1618 pg/mL, troponin T 0.02 ng/mL, and D-dimer 1.60 mcg/mL. The remaining routine chemistry and blood cell counts were normal. Chest X-ray showed bilateral interstitial markings concerning for interstitial pneumonia. Given the elevated D-dimer, a CT Pulmonary Angiogram was done which did not reveal any pulmonary emboli but did show findings consistent with pulmonary edema.

Because of the leukocytosis and chest x-ray showing possible interstitial pneumonia, blood cultures were drawn on admission and intravenous ceftriaxone and azithromycin were initiated to cover for community-acquired pneumonia. The patient was admitted to the hospitalist service for further management.

A transthoracic echocardiogram revealed an ejection fraction of 40-45%, global hypokinesis, moderate aortic stenosis with an aortic valve area of 1cm<sup>2</sup> with mild aortic regurgitation, and moderate mitral regurgitation. No valvular vegetations were appreciated. Cardiology consultation was requested, and intravenous diuresis was initiated given clinical, laboratory, and radiographic findings of acute decompensated systolic heart failure. After several days of diuresis, the patient reported improvement in his prior shortness of breath and lower extremity edema. Eventually, he was transitioned to carvedilol, lisinopril, and low-dose oral furosemide. However, lisinopril and furosemide had to be held due to episodes of hypotension, which improved once these medications were discontinued. Cardiology believed that the patient would likely need an elective aortic valve replacement in the future.

On hospital day 3, the initial blood cultures grew gram-negative bacilli. At this time, Infectious Diseases consultation was requested. Preliminary cultures grew *Moraxella* species, possibly from a pulmonary source given the initial chest X-ray findings. A CT Abdomen/Pelvis was done to rule out an intra-abdominal source, which was unremarkable. There was a delay in sensitivity reporting for the *Moraxella* cultures, and per the Microbiology lab, it would take an additional two weeks for the sensitivities to finalize. Given the patient's significant improvement from admission, Infectious Diseases believed the patient could be transitioned to oral levofloxacin to complete a two-week course of antibiotics. The patient was soon discharged home with appropriate outpatient Infectious Diseases and Cardiology follow-up.

Two days after discharge, the patient presented to his Cardiology appointment and reported recurrence of his shortness of breath and dyspnea on exertion. At that time, his Cardiologist re-initiated low-dose furosemide with plan for outpatient right-and-left cardiac catheterization as well as transesophageal echocardiogram within the following two days. The day after

this Cardiology appointment, the patient could no longer tolerate his symptoms and presented once again to the emergency department for further evaluation. His vitals on this second presentation were as follows: temperature 36.6°C, heart rate 104, blood pressure 105/61, respiratory rate 18, and oxygen saturation of 95% on room air. Chest X-ray re-demonstrated pulmonary edema. His laboratory evaluation was notable for a white blood cell count  $9.18 \times 10^9/L$ , BNP 343 pg/mL and troponin I 0.29 ng/mL, with repeat 0.72 ng/mL. Electrocardiogram was negative for any dynamic ST changes. The patient was admitted to the coronary care unit and subsequently underwent cardiac catheterization showing nonobstructive left coronary disease. A transesophageal echocardiogram was also performed which revealed a left ventricular ejection fraction of 45%, a mobile mass on the aortic valve leaflet of the left coronary cusp, severe aortic regurgitation, and an aneurysm of the sinus of Valsalva. Given concern for native valve endocarditis, both Cardiothoracic Surgery and Infectious Diseases consultations were requested. Ceftriaxone was initiated, and the patient was taken to the operating room for aortic valve replacement, repair of the sinus of Valsalva aneurysm, mitral valve valvuloplasty, and repair of a patent foramen ovale. He tolerated these surgical procedures well. The Infectious Diseases consultants contacted the Microbiology Lab to inquire about the blood cultures from his initial hospitalization in order to obtain final sensitivities. Eventually, it was revealed that the original microorganism was incorrectly identified as *Moraxella* species, and the correct organism was *Bordetella holmesii*. It was pan-sensitive to multiple antibiotics, and the patient was transitioned to a 6-week course of intravenous cefepime. Eventually, the patient was transferred to a post-acute rehabilitation facility for ongoing rehabilitation and completion of antibiotic treatment.

## Discussion

*Bordetella holmesii* (*B. holmesii*) is a rare Gram-negative coccobacillus that was first isolated in 1983, and first described in the literature in 1995.<sup>2,3</sup> This organism is often associated with invasive infections such as bacteremia, meningitis, septic arthritis, and endocarditis in primarily immunocompromised patients. In recent years, it has also been associated with invasive infections in patients with a history of functional or anatomical asplenia.<sup>3</sup> Since its identification in 1995, there have been fewer than 10 case reports describing *B. holmesii* endocarditis in asplenic patients.

In a literature review of 30 identified cases of *B. holmesii* bacteremia in the United States, 22 of these patients were asplenic.<sup>3</sup> Most of these patients suffered from sickle cell anemia, but three patients had undergone splenectomy for lymphoma treatment or staging, such as in our patient. The majority of the asplenic patients presented initially with a non-specific febrile syndrome, and 20 cases required hospitalization for further evaluation and treatment. Eleven cases were given a primary diagnosis of *B. holmesii* bacteremia. The remaining cases included two patients with endocarditis, one who had a history of Hodgkin's lymphoma and eventually required sur-

gery for aortic valve replacement. There were no deaths amongst this cohort, and all patients were treated with intravenous antibiotics, including third-generation cephalosporins, fluoroquinolones, and carbapenems.

The case presented in this report describes a patient who shared similar features to this literature review. He had several risk factors for invasive *B. holmesii* infection, including a history of childhood Hodgkin's lymphoma and associated splenectomy for staging purposes. His clinical course was complicated by both bacteremia and infective endocarditis, treated with both surgical valve replacement and antibiotics. There are several issues to highlight in this case. The patient's initial blood cultures took almost four weeks for isolation of the correct organism. Another reported case of *B. holmesii* endocarditis in a post-splenectomy patient, also took four weeks for the correct organism to be isolated.<sup>2</sup> This has an impact on timing and selection of appropriate therapy. Another interesting point in our case is that the initial transthoracic echocardiogram did not reveal any vegetations. His new-onset heart failure symptoms were thought to be due to advanced aortic stenosis versus delayed sequela of his prior chemotherapy and radiation treatments. Because the patient's white blood cell count had improved, he was never febrile, and his presenting symptoms had improved with treatment, the initial suspicion for infective endocarditis was rather low. If *B. holmesii* had been isolated in the initial blood cultures much sooner, perhaps a transesophageal echocardiogram would have been performed on his initial hospitalization given his history of asplenia.

## Conclusion

Patients with a history of asplenia are at an increased risk for serious invasive bacterial infections. Though the most commonly associated culprits are encapsulated organisms, it is important to also consider other pathogens that may cause systemic disease. Early clinical suspicion for these rare pathogens may lead to prompt treatment and reduced mortality. Patients with asplenia should also be properly educated on precautions to prevent serious infections, including appropriate immunizations.

## REFERENCES

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