

## Case Report

# *Bordetella hinzii*: An Unexpected Pathogen in Native Valve Endocarditis

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### ABSTRACT

*Bordetella hinzii*'s route of transmission to human hosts and its pathogenicity remain unclear. Only a few cases have established this species as an opportunistic zoonotic disease. We introduce the first reported case of native aortic valve endocarditis presenting with fulminant aortic valve insufficiency that responded to conventional medical and surgical treatment. The patient did not have predisposing factors to this unusual infection. This case may provide a better understanding of the disease process, transmission, and pathogenicity of *Bordetella hinzii*.

### RÉSUMÉ

La voie de transmission de *Bordetella hinzii* à l'hôte humain et la pathogénicité de cette bactérie demeurent incertaines. Seuls quelques cas de zoonose considérés comme étant attribuables à cette bactérie ont été documentés. Nous vous faisons part du premier cas signalé d'endocardite de la valve aortique naturelle s'étant manifestée par une insuffisance aortique fulminante répondant aux traitements médicaux et chirurgicaux classiques. Le patient ne présentait aucun facteur le prédisposant à cette infection inhabituelle. Ce cas pourrait permettre une meilleure compréhension du processus morbide, de la transmission et de la pathogénicité de *Bordetella hinzii*.

*Bordetella* species are gram-negative rod-shaped organisms that are known to cause respiratory tract infections. *Bordetella hinzii*, closely related to *Bordetella avium*, has been isolated from the respiratory tracts of poultry and, along with wild rodents, have been reported as possible reservoirs for human transmission.<sup>1,2</sup> The source of transmission and pathogenicity remains unclear. Limited reports have shown this rare transmission of infection in humans in the absence of contact with poultry to cause bacteremia, pulmonary, and digestive infection.<sup>1,3</sup> This transmission has only occurred in immunocompromised individuals reported to have acquired immunodeficiency syndrome, bone marrow and liver transplantation, myelodysplastic syndrome, Epstein–Barr virus, and cystic fibrosis.<sup>1</sup> Although respiratory infections in immunocompetent individuals have also been reported, its presence as a colonizer remains in question.<sup>1</sup> Only 1 previous case of *B. hinzii* endocarditis was reported in a setting of a bioprosthetic aortic valve in a patient who had chronic liver disease with portal hypertension and a history of adenocarcinoma of the colon.<sup>3</sup> We present a second case of infective endocarditis secondary to *B. hinzii* and a first case of this pathogen in a native aortic valve.

### Case Report

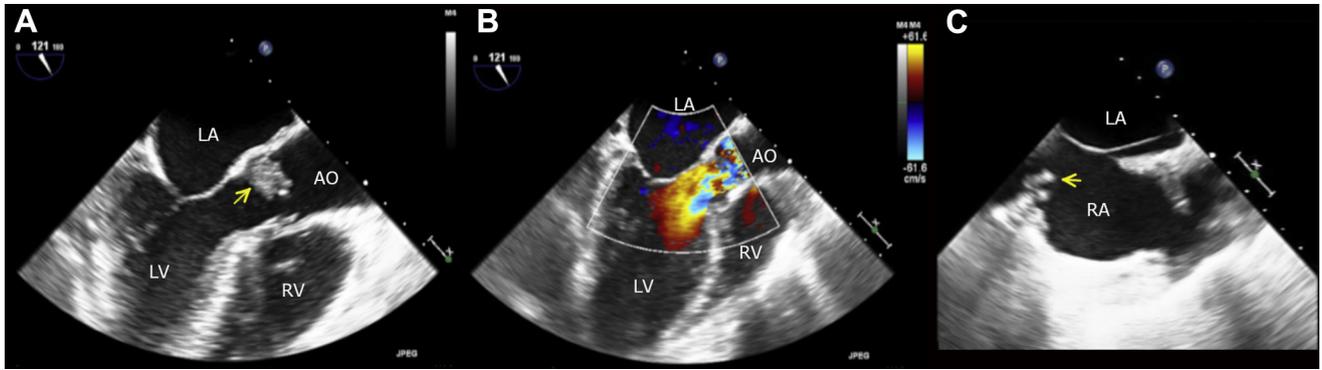
A 53-year-old white woman with a medical history of ulcerative colitis and no prior heart disease presented with fatigue, fever, shortness of breath, and cough for 3 weeks. Initial laboratory data showed a hemoglobin of 74 g/L, platelets at  $64 \times 10^9$  /L, and normal renal function. Within 6 days of admission, the patient developed acute renal failure, transaminitis, and lactic acidosis. Two sets of blood cultures grew *B. hinzii*. On the transesophageal echocardiogram, a large vegetation was noted on the noncoronary cusp, causing severe aortic insufficiency (Fig. 1A and B). A smaller pedunculated mass was noted at the junction of the right atrium and the inferior vena cava with features similar to a eustachian valve but with a high suspicion of vegetation (Fig. 1C). No evidence of septic emboli or valvular abscesses was observed. Urgent aortic valve replacement with an Intuity 21-mm Edwards valve (Edwards Lifesciences, Irvine, CA) was performed. Intravenous ceftazidime (2g three times a day) was initiated before surgery and continued for 2 weeks to treat bacteremia with a pan-sensitive antimicrobial profile. Repeat cultures, drawn 2 days later, were negative. The intraoperatively obtained aortic valve and right atrium pedunculated specimens were sent for culture and gram staining (Fig. 2A and B). No clear evidence for right atrium vegetation was appreciated upon review of the pathological specimen. Meanwhile, cultures of specimens did not grow an organism. Postoperatively, renal function slowly recovered with 2 weeks of monitoring in the Cardiac Unit. At 1-month follow-up, the dialysis catheter was removed.

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See page 1604.e18 for disclosure information.



**Figure 1.** (A) Midesophageal long-axis view of large aortic valve vegetation, (B) severe aortic valve insufficiency, and (C) bicaval view of pedunculated mass at the right atrium and inferior vena cava junction.

## Discussion

This case is unique because *B. hinzii* is not well known for its endocarditis pathogenicity. The one previously reported case of *B. hinzii* endocarditis was similar in presentation with no contact with poultry.<sup>3</sup> Other reported cases of human transmission have mostly occurred in immunocompromised individuals. Our workup included a bone marrow biopsy, serum flow cytometry, serum electrophoresis with immunofixation, and free light chain analysis. Laboratory and polymerase chain reaction analyses for pneumocystis, aspergillosis, tuberculosis, nocardia, Epstein–Barr virus, cytomegalovirus, herpes simplex virus, acquired immunodeficiency syndrome, and hepatitis were performed. Conditions such as celiac disease, systemic lupus erythematosus, and rheumatoid arthritis were also ruled out.

Inflammatory bowel disease has been reported as a predisposing factor in causing infective endocarditis. Bacteremia during the disease exacerbation is proposed to result from facilitation of invasion through increased transmucosal permeability.<sup>4</sup> However, in the case presented, a computed

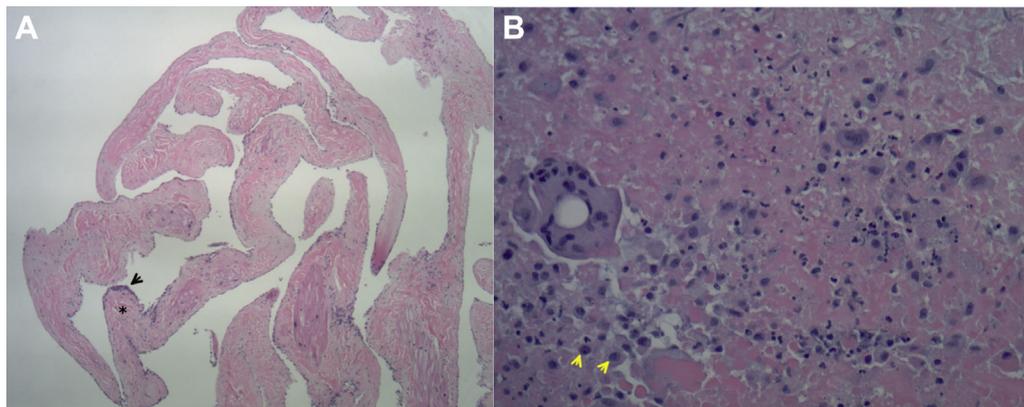
tomography of the abdomen did not show any acute infectious processes. With no previous use of corticosteroids or immunomodulators in our case, a stable chronic inflammatory bowel disease is less likely the primary contributor.

## Conclusions

Our case further illustrates the human transmission of *B. hinzii*, suggesting for other routes or vectors that may not be limited to poultry, rats, and rabbits. This bacterium can affect immunocompetent individuals and in a manner of hematogenous spread to cause endocarditis. This bacterium's pathogenicity for infective endocarditis with quick response to systemic and surgical treatment can be better understood with future reports of cases.

## Disclosures

The authors have no conflicts of interest to disclose.



**Figure 2.** (A) Hematoxylin–eosin stain; 10× magnification; mass at the right atrium and inferior vena cava junction. Fibroelastic stroma (**asterisk**) are present, and their outer surface is lined with endothelial cells (**arrows**). (B) Aortic valve vegetation, 40× magnification, with multinucleated giant cells, and neutrophilic infiltrates (**arrows**) in granulation tissue indicating ongoing infectious process.

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