Internal Medicine
The Japanese Society of Internal Medicine
doi: 10.2169/internalmedicine.9374-22
Intern Med 61: 3373-3376, 2022
http://internmed.jp

【 CASE REPORT 】

Right-sided Infective Endocarditis with Ventricular Free Wall Vegetation Caused by *Abiotrophia defectiva* in a Patient with Unrepaired Ventricular Septal Defect

Norihisa Miyawaki[1], Taiji Okada[1], Tadaaki Koyama[2] and Yutaka Furukawa[1]

Abstract:

To our knowledge, there have been no reports of right-sided infective endocarditis (RSIE) with ventricular free wall vegetation caused by *Abiotrophia defectiva*. We herein report a case of RSIE caused by *A. defectiva* with ventricular free wall vegetation in a 27-year-old man with ventricular septal defect (VSD). Computed tomography showed multiple bilateral pulmonary nodular shadows. Transesophageal echocardiography (TEE) demonstrated right ventricular free wall vegetation at the jet stream. Blood culture revealed *A. defectiva*. These findings are consistent with a diagnosis of infective endocarditis and septic pulmonary embolism. Treatment with ceftriaxone and gentamicin and subsequent surgical VSD closure improved the patient's condition without recurrence.

Key words: *Abiotrophia defectiva*, septic embolism, ventricular septal defect, transesophageal echocardiography, right-sided infective endocarditis

(Intern Med 61: 3373-3376, 2022)
(DOI: 10.2169/internalmedicine.9374-22)

Introduction

In Japan, right-sided infective endocarditis (RSIE) is typically seen in patients with congenital cardiac malformations (1), most commonly in patients with ventricular septal defect (VSD) (2). Without prior antimicrobial therapy, blood cultures are positive in >90% of cases (3). However, blood cultures may be negative in infective endocarditis caused by HACEK (*Haemophilus* spp., *Aggregatibacter* spp., *Cardiobacetrium* spp., *Eikenella corrodens*, and *Kingella* spp.), nutritionally variant *Streptococci* such as *Abiotrophia defectiva*, or fungal infections.

A. defectiva, part of the oral normal flora, requires vitamin B6 and L-cysteine to grow, which makes it difficult to culture and detect on regular blood agar (4). Furthermore, *A. defectiva* is a rare etiology of infectious endocarditis (5), most commonly presenting with heart failure and valvular pathologies (6, 7). RSIE caused by *A. defectiva* has been reported (8-10), but to the best our knowledge, there have been no reports of vegetation in the right ventricular free wall at the location impacted by VSD jet.

Echocardiography, especially transesophageal echocardiography (TEE), plays an important role in the diagnosis of infective endocarditis. Although TEE is more sensitive for the detection of valvular vegetation (11), few reports have investigated the role of TEE in RSIE.

We herein report a case of RSIE with ventricular free wall vegetation caused by *A. defectiva* in a patient with VSD, which was successfully treated with surgery and antibiotics.

Case Report

A 27-year-old man with VSD presented to another hospital with episodic back pain and fever for one year, which had occurred six months after undergoing dental treatment. He was referred to our hospital after chest computed tomography (CT) revealed multiple bilateral pulmonary nodular shadows (Fig. 1A-C).

[1]Department of Cardiovascular Medicine, Kobe City Medical Center General Hospital, Japan and [2]Department of Cardiothoracic Surgery, Kobe City Medical Center General Hospital, Japan
Received: January 11, 2022; Accepted: March 3, 2022; Advance Publication by J-STAGE: April 16, 2022
Correspondence to Dr. Taiji Okada, taiji_okada@kcho.jp

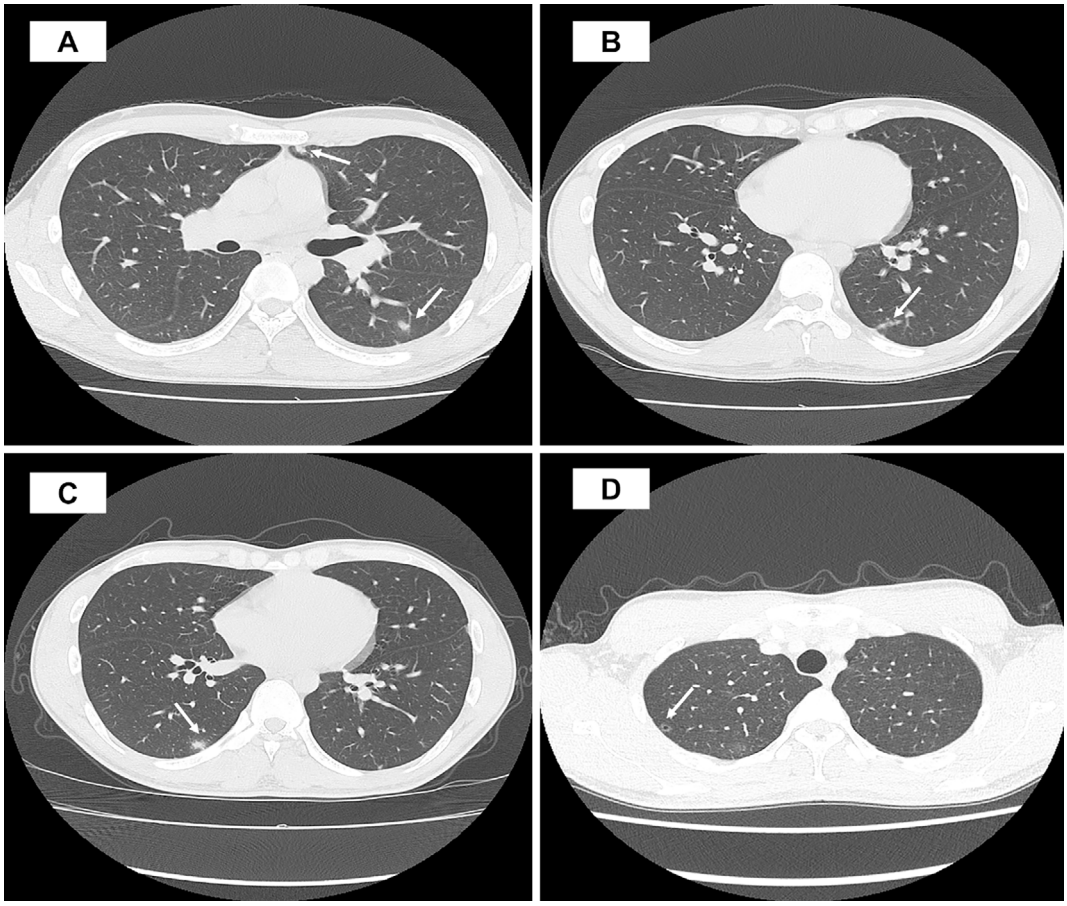

Figure 1. **Chest computer tomography showing progressive multiple nodular shadows in both lungs (arrow) at 7 months before admission (A), 6 months before admission (B), and 1 month before admission (C). Contrast-enhanced computed tomography on admission revealing resolution of the bilateral pulmonary nodular shadows (D).**

On admission, the patient denied any symptoms. Vital signs revealed slightly elevated blood pressure (126/84 mmHg) with a normal body temperature (37.0°C), pulse rate (85 beats/min), respiratory rate (16 breaths/min), and oxygen saturation (99% on room air). Chest auscultation showed a grade 4/6 pansystolic murmur, which was best heard along the left sternal border with clear breath sounds without rales. An abdominal examination revealed splenomegaly without hepatomegaly. A neurological examination was unremarkable. A peripheral examination showed no signs of endocarditis.

Blood tests in our hospital revealed an elevated C-reactive protein level (1.69 mg/dL; normal range, 0.0-0.5 mg/dL) with a normal white cell count (7.2×10^3/μL; normal range, 3.9×10^3-9.8×10^3/μL), D-dimer level (0.23 μg/L; normal range, 0-0.99 μg/L), and NT-proBNP level (111 pg/mL; normal range, 0-125 pg/mL).

Electrocardiography revealed no abnormalities. Transthoracic echocardiography (TTE) demonstrated a perimembranous VSD with no obvious valvular disease, vegetation, or pulmonary hypertension. Due to the absence of symptoms and only mildly elevated C-reactive protein level, the patient was managed at home. Blood cultures revealed Gram-positive cocci in two replicates, prompting emergent admission to the hospital and the initiation of ceftriaxone treatment (2 g, daily).

On the 1st hospital day, contrast-enhanced CT showed no obvious embolism, splenomegaly, or nodular shadows in either lung field (Fig. 1D). On the 2nd hospital day, TEE revealed right ventricular vegetation on the free wall where the jet of the VSD was impacting (Fig. 2). These findings were consistent with a diagnosis of infective endocarditis with septic pulmonary embolism. A microbiologic examination confirmed that the etiology of the RSIE was *A. defectiva*, prompting continuation of ceftriaxone for six more weeks and the addition of gentamicin (3 mg/kg daily) for 2 weeks. Cranial magnetic resonance imaging showed no cerebral infarction.

On the 7th hospital day, blood cultures turned negative with no signs of heart failure. On the 16th hospital day, he was transferred to another hospital for continued antibiotic therapy. After completing six weeks of antibiotic therapy, the patient underwent surgical closure of the VSD. Intraoperatively, there were no infectious lesions near the VSD or vegetation on the right ventricular free wall. The patient's postoperative course was uneventful, with no recurrence of infection. He was discharged 10 days after surgery.

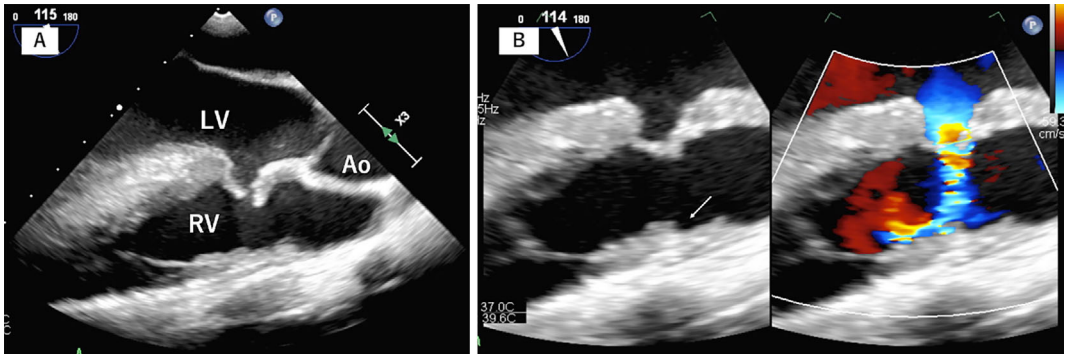

Figure 2. **Transesophageal echocardiography with Doppler demonstrating perimembranous VSD (A) with vegetation (arrow) in the free wall of the right ventricle where the jet from the VSD impacts (B). Ao: aorta, LV: left ventricle, RV: right ventricle**

Discussion

We described a case of RSIE with right ventricular free wall vegetation caused by *A. defectiva* in a patient with unrepaired VSD. To the best of our knowledge, this is the first reported case of RSIE caused by *A. defectiva*. In this report, we demonstrated the utility of TEE in the diagnosis of RSIE in a patient with VSD.

Infective endocarditis continues to pose a serious risk in patients with congenital heart disease. Despite improved antibiotic therapies and prophylactic measures, infective endocarditis is still associated with considerable mortality (12). Therefore, the early detection and management of infective endocarditis are important.

Echocardiography plays an important role in the diagnosis of infective endocarditis. Among various echocardiographic techniques, TTE allows an easy and accurate diagnosis of tricuspid vegetation in RSIE (13, 14). However, it has been reported that the consistency of TTE with subsequent intraoperative findings of vegetation was markedly lower in patients with RSIE in comparison to that in patients with left-sided infective endocarditis (2).

In this case, the clinical diagnosis of VSD and the echocardiographic diagnosis of right ventricular vegetation were confirmed. Aside from the right ventricular free wall vegetation, no other endocardial lesions were detected on physical examination or TTE. Our findings are consistent with the theory that a jet stream can cause endocarditis at the point of impact. VSD closure was performed due to the risk of recurrent endocarditis at the jet lesion. Therefore, TEE should still be considered in patients with RSIE, if no vegetation is observed on TTE.

Among the few reported cases of infective endocarditis due to *A. defectiva*, the majority underwent valvular replacement despite adequate antibiotic therapy (15-17). *A. defectiva* strains that are highly resistant to Penicillin G or multidrug-resistant have also been reported (18), and patients with penicillin-resistant *A. defectiva* IE may experience worse outcomes with a higher rate of recurrence in comparison to patients with IE caused by other viridans group streptococci. Blood cultures do not always predict the appropriate antibiotic treatment for this pathogen as it is difficult to culture on standard blood agar media without supplementation, which may lead to false-negative results. Therefore, it is prudent to suspect *A. defectiva* in patients with culture-negative endocarditis, and to use supplemented media when repeat blood culture is performed.

Conclusion

We encountered a case of RSIE with ventricular free wall vegetation caused by *A. defectiva* in a patient with unrepaired VSD at the point impact from the jet stream. While *A. defectiva* is a rare etiology of infective endocarditis, it is often associated with severe outcomes, highlighting the importance of early detection and management. In addition, while we demonstrated the role TEE in the diagnosis of RSIE, TEE should be considered in patients with no detectable vegetation on TTE.

The authors state that they have no Conflict of Interest (COI).

References

1. Niwa K, Nakazawa M, Tateno S, Yoshinaga M, Terai M. Infective endocarditis in congenital heart disease: Japanese national collaboration study. Heart **91**: 795-800, 2005.
2. Xie J, Liu S, Yang J, Xu J, Zhu G. Inaccuracy of transthoracic echocardiography for the identification of right-sided vegetation in patients with no history of intravenous drug abuse or cardiac device insertion. J Int Med Res **42**: 837-848, 2014.
3. Nakatani S, Ohara T, Ashihara K, et al. JCS 2017 guideline on prevention and treatment of infective endocarditis. Circ J **83**: 1767-1809, 2019.
4. Bouvet A, van de Rijin I, McCarty M. Nutritionally variant streptococci from patients with endocarditis: growth parameters in semisynthetic medium and demonstration of a chromophore. J Bacteriol **146**: 1075-1082, 1981.
5. Roberts RB, Krieger AG, Schiller NL, Gross KC. Viridans streptococcal endocarditis: the role of various species, including pyridoxal-dependent streptococci. Rev Infect Dis **1**: 955-956, 1979.

6. Satoh T, Fujita K, Ikeda M, Morimoto K, Sakamoto H. Separation experience of nutritionally variant streptococci from blood cultivation. J Jpn Soc Clin Microbiol **6**: 51-55, 1996 (in Japanese).

7. Boutoille D, Reynaud AE, Leautez S, Ponge T, Raffi F. Abiotrophia-related endocarditis: contribution of molecular biology. Presse Med **28**: 2149-2151, 1999 (in French).

8. Takayama R, Motoyasu M, Seko T, et al. A case of isolated tricuspid valve infective endocarditis caused by *Abiotrophia defectiva*. Int J Cardiol **118**: e3-e5, 2007.

9. Bhattacharya P, Mohammed A, Mizrahi E. Aorto-right ventricular fistula: a rare complication of *Abiotrophia* endocarditis. Oxf Med Case Rep **2017**: omx035, 2017.

10. Planinc M, Kutlesa M, Barsic B, Rudez I. Quadruple-valve infective endocarditis caused by *Abiotrophia defectiva*. Interact Cardiovasc Thorac Surg **25**: 998-999, 2017.

11. Sordelli C, Fele N, Mocerino R, et al. Infective endocarditis: echocardiographic imaging and new imaging modalities. J Cardiovasc Echogr **29**: 149-155, 2019.

12. Di Filippo S. Clinical outcomes for congenital heart disease patients presenting with infective endocarditis. Expert Rev Cardiovascular Ther **18**: 331-342, 2020.

13. Evangelista A, Gonzalez-Alujas MT. Echocardiography in infective endocarditis. Heart **90**: 614-617, 2004.

14. Herrera CJ, Mehlman DJ, Hartz RS, Talano JV, McPherson DD. Comparison of transesophageal and trans-thoracic echocardiography for diagnosis of right-sided cardiac lesions. Am J Cardiol **70**: 964-966, 1992.

15. Christensen JJ, Facklam RR. *Granulicatella* and *Abiotrophia* species from human clinical specimens. J Clin Microbiol **39**: 3520-3523, 2001.

16. Ramos J, Dos Santos L, Vidal L, et al. A case report and literature overview: *Abiotrophia* defectiva aortic valve endocarditis in developing countries. Infection **42**: 579-584, 2014.

17. Hashimoto T, Jodo S, Furusaki A, et al. A woman with infectious endocarditis caused by Abiotrophia defectiva. Intern Med **43**: 1000-1004, 2004.

18. Cañas MA, Téllez A, García de la Mària C, et al. Development of high-level daptomycin resistance in *Abiotrophia* and *Granulicatella* species isolates from patients with infective endocarditis. Antimicrob Agents Chemother **65**: e0252220, 2021.