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Case Report

An unusual case of *Corynebacterium striatum* endocarditis in a patient with congenital lymphedema and rheumatic heart disease

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ARTICLE INFO

Article history:
Received 6 May 2015
Accepted 14 July 2015
Available online 18 January 2016

Keywords:
Corynebacterium striatum
Lymphedema
Rheumatic heart disease
Endocarditis

ABSTRACT

Corynebacterium striatum (*C. striatum*) is a ubiquitous saprophyte with a potential to cause bacteremia. We report the first case of *C. striatum* endocarditis in a patient with congenital lymphedema and rheumatic heart disease.

1. Introduction

Corynebacterium species are aerobic, non-sporulating, gram positive bacilli that are often considered as non-pathogenic components of normal skin flora and mucosal membranes. Although frequently isolated in cultures, they are commonly considered as contaminants. Nevertheless, the ability of these bacteria to cause life-threatening disease is well established, and over the last decade, there have been increasing reports of

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http://dx.doi.org/10.1016/j.ihj.2015.07.026

their pathogenic potential in numerous clinical scenarios, including bacteremia and endocarditis. *Corynebacterium* species are now thought to cause 9% of early and 4% of late prosthetic valve endocarditis, but only 0.2–0.4% of cases of native valve endocarditis. Recognized risk factors for *Corynebacterium* endocarditis include preexisting cardiac disease, a prior history of bacterial endocarditis, and the presence of prosthetic devices, including intravascular access and auriculo-ventricular cerebrospinal fluid shunts.

Corynebacterium striatum (*C. striatum*) has been increasingly reported as the etiologic agent of various infections.[1] Blood cultures positive for *Corynebacterium* species and *C. striatum* in particular should never be overlooked.[2]

2. Case report

We report a first case of rheumatic heart disease complicated with infective endocarditis due to *C. striatum*. A 27-year-old male, who was a known case of rheumatic heart disease, presented with history of fever of 15 days duration. Also, he gave history of partial seizures involving left upper limb and face of 7 days duration. He had past history of swelling of both lower limbs up to knee since birth. On clinical examination, the patient had temperature of 99 °F or 37.2 °C, normal vital signs, no evidence of pallor, clubbing, or lymphadenopathy. He had bilateral lower limb induration and edema up to knees. Cardiovascular examination revealed a pansystolic murmur at the apex radiating to axilla. Lab findings revealed Hb 12.6 g/dl, WBC 14,500 cells/mm^3, 83.5% neutrophils, 11.7% lymphocytes, and Erythrocyte Sedimentation Rate 36 mm/h. The other biochemical parameters were within normal limits. 2D echocardiography revealed mitral valve prolapse with moderate mitral regurgitation and evidence of vegetation on the anterior mitral valve leaflet (Fig. 1). CT brain was done to find the cause of seizure, which was normal. The patient was empirically started with IV crystalline penicillin 20 lakh units 4th hourly and gentamicin 80 mg bd; later, the antibiotic was changed to IV cefuroxime 2 g iv bd based on the culture sensitivity report. The patient responded clinically to antibiotics and was asymptomatic after one week of antibiotics. Repeat echocardiography showed decrease in the size of vegetation. The patient was discharged and advised a regular follow-up.

3. Microbiological identification

Blood cultures were performed using BacT/Alert system. Two of the three samples gave positive signal for growth. Gram staining revealed Gram-positive short bacilli. Subcultures were performed on sheep blood agar and MacConkeys agar and incubated at 37 °C.

Blood agar was incubated in CO_2 incubator. Moist white colored colonies were observed in 48 h, Gram staining revealed short Gram-positive bacilli. Identification of the isolate as *C. striatum* was based on colony morphology, Gram staining, motility, the catalase, and oxidase reactions, and the results obtained was confirmed by use of the VITEK 2 Compact 2 system (BioMèrieux, l'Etoile, France) as *C. striatum*.

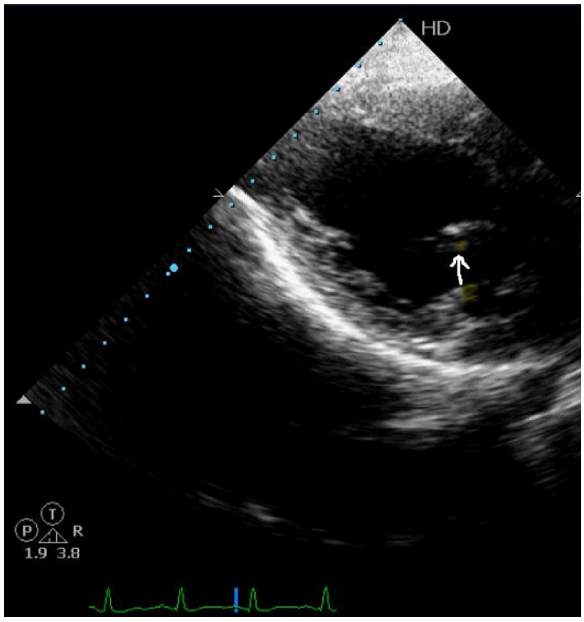

Fig. 1 – 2D ECHO revealing mitral valve prolapse with moderate mitral regurgitation with evidence of vegetation on the anterior mitral valve leaflet.

Antibiotic susceptibility was tested by the disk diffusion method in Mueller-Hinton agar supplemented with 5% blood. The isolate was susceptible to cefazolin (30 mcg), cefuroxime (30 mcg), linezolid (30 mcg), vancomycin (30 mcg), teicoplanin (30 mcg), cefotaxime (30 mcg) and ceftriaxone (30 mcg). The isolate was resistant to penicillin G 10 units, erythromycin (15 mcg), azithromycin (15 mcg), levofloxacin (5 mcg), and ciprofloxacin (5 mcg).

There is neither a recognized standard method nor specific guidelines published by Clinical Laboratory Standards Institute (CLSI) for antimicrobial testing of coryneform bacteria.[2] Hence, the susceptibility criteria of the CLSI for *Staphylococcus* spp. were used for all antibiotics tested except penicillin and ampicillin, for which thresholds for *Listeria* spp. were used.

4. Discussion

C. striatum is a part of the normal flora of the human skin and respiratory tract. Although rare, non-diphtheriae corynebacteria have known to cause endocarditis in individuals with predisposing risk factors such as prior cardiac disease, drug injection, indwelling prosthetic devices, or chronic hemodialysis, and only few cases involving *C. striatum* associated endocarditis, including native valve prosthetic valve and pacemaker-related endocarditis, are well being documented.[1–5] For many years, *Corynebacterium* species was long believed to have limited potential as a pathogen, and hence was usually considered a contaminant when isolated from a patient specimen. Evidence to support the role of *C. striatum* as a pathogen in immunocompromised and immunocompetent hosts is growing. Edward et al. have reported a case of *C. striatum* cutaneous infection in a patient with AIDS.[3] The role

of *C. striatum* as a nosocomial pathogen is also evolving.[3] David et al. have reported a case of spontaneous joint infection with *C. striatum*.[4] Olivia et al. have reported cardiac device related infection by *C. striatum*.[8]

Our patient was diagnosed with congenital lymphedema. Literature search on congenital lymphedema implies that in long-standing cases of lymphedema, the skin becomes scaly and cracked, and is prone for secondary bacterial or fungal infections (ref New York Health Care – lymphedema). In our case, the most likely route of entry/access of *C. striatum* would have been through minute abrasions on the skin. Gandham et al. have reported a case of necrotizing fasciitis of lower limb by *C. striatum* in a HBsAG positive patient, indicating deep seated infection secondary to trauma, which could have provided the portal of entry for *C. striatum*.[7] Superti et al. have reported a case of *C. striatum* skin and soft tissue infection of a malignant skin lesion.[7] Martin et al. isolated identical strains of *C. striatum* from a leg ulcer and from the blood stream in a patient with peripheral vascular disease, confirming the entry of the bacterium through the skin to the circulation.[2]

C. striatum, a normal skin commensal, is rarely isolated in native valve endocarditis and most often requires surgical management with appropriate antibiotics[1]; however, our patient recovered with medical management with injection of cefuroxime for six weeks. Blood cultures at the end of antibiotic treatment remained negative. The patient was asymptomatic at follow-up after one and three months.

The antibiotic susceptibility pattern of *C. striatum* is variable. Among strains of *C. striatum*, multidrug resistance may be spreading. A decade ago, isolates showed sensitivity to beta lactams (particularly penicillin G), fluoroquinolones, carbapenems, linezolid, and vancomycin.[4] In the last 5 years, pathogenic isolates showing multidrug resistance have emerged.[4] Significant number of authors have used vancomycin as initial therapy for suspected infection.[2–6]

5. Conclusions

C. striatum is a potentially pathogenic microorganism with the ability to produce infection like infective endocarditis in a native valve, which should never be ignored as a contaminant. It is a potential cause for infection in patients with established risk factors like rheumatic heart disease. *C. striatum* infections should be treated according to the results of the antibiogram, as it is an emerging multidrug-resistant organism. Universal hygiene measures should be observed, as this potentially infectious organism is able to cause nosocomial infections and colonization.

Conflicts of interest

The authors have none to declare.

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