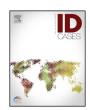


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A rare complication of a hemodialysis tunneled catheter: Case report of a superior vena cava and right atrium candida endocarditis



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ARTICLE INFO

Article history: Received 16 March 2020 Accepted 7 April 2020

Keywords: Candida albicans Endocarditis Tunneled cuffed catheters Hemodialysis

ABSTRACT

Infections remain an important cause of death among hemodialysis patients. This population have a higher risk of candidemia. Candida endocarditis its a rare but frequently fatal complication of candidemia.

A 64 year-old female presented with a purulent discharge at the insertion site of a hemodialysis tunneled cuff catheter. A catheter related bloodstream infection was suspected, cultures were obtained and wide-spectrum antibiotic therapy was administered. A multi sensitive *Candida albicans* was isolated. Transesophageal echocardiography showed a large vegetation located in the superior vena cava, in probable relation with a previous catheter. The first approach was antifungal treatment. Due to non-response, she did a surgical removal of the vegetation. Culture of the vegetation showed the same as the blood cultures. After one year she has no signs of relapse.

To improve the prognosis of this high mortality condition a high index of suspicion is necessary for early diagnosis and timely intervention.

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Introduction

Fungal endocarditis is a rare and serious disease [1,2]. It represents 1–10 % [1,3] of all infective endocarditis, with *Candida albicans* being isolated in 24 % of the cases [1].

Candida endocarditis (CE) is one of the most serious complications of candidemia with high morbidity and mortality [4] reaching 36 % of index hospitalization and 59 % at one year [5]. Larger vegetation and more arterial embolization than bacterial endocarditis may explain worse outcomes [6,7].

Presence of central venous catheters is a well-known risk factor for candidemia and CE. A biofilm is formed around the catheter walls that hinders the penetration of the antifungal.

Recent studies revealed high rate of candidemia in hemodialysis patients (0.04–0.32 cases per 1000 hemodialysis sessions per year) [8] and the presence of a catheter (vs. native graft or fistula) was an independent risk factor (Odds Ratio 3.24) [9].

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CE is very challenging and difficult to manage. Early diagnosis requires a high index of suspicion. An aggressive treatment strategy with a multidisciplinary approach that comprises both antifungal agents and surgical management is mandatory.

We present a case of endocarditis with a large vegetation by *Candida albicans* in the superior vena cava and right atrium associated to a tunneled catheter of hemodialysis.

Case

A 64 year-old white female presented with hypotension, tachycardia, purulent discharge and tenderness at the insertion site of a hemodialysis tunneled cuff catheter (TCC).

Past medical history included a 20-year history of type 2 diabetes mellitus (DM), with micro and macro-vascular complications such as diabetic retinopathy and nephropathy with chronic kidney disease. Although informed about the risks, the patient refused timely arteriovenous fistula placement for hemodialysis.

It was started hemodialysis trough a TCC inserted in the right internal jugular vein with real time ultrasound guidance. At the same time it was diagnosed a Drug Reaction with Eosinophilia and Systemic Symptoms (DRESS) Syndrome related to allopurinol so it was started oral prednisolone.

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Other medical problems included arterial hypertension, obesity, ischemic heart disease and congestive heart failure.

During a hemodialysis session she presented severe hypotension (Blood pressure $80/44\,\mathrm{mmHg}$) with pulse of 100 beats per minute and a body temperature of $37\,^\circ\mathrm{C}$. The physical examination showed local inflammatory signs with purulent discharge at the catheter insertion site. The remainder of the physical examination was normal.

Laboratory-test results revealed normocytic normochromic anemia of 7.9 g/dL (reference range (RR) 12.0–16.0 g/dL), white-cell count 18×10^3 μ L (RR $4.5-11 \times 10^3$ μ L) with neutrophil predominance (77.7 %; RR 40–70 %), high C reactive protein of 19 mg/dL (RR 0–0.5), procalcitonin 28 ng/mL (< 0.5 ng/mL). Other laboratory parameters were unremarkable.

Sepsis from a catheter related bloodstream infection was suspected, cultures of the blood and of the exudate were obtained and wide-spectrum antibiotic therapy with vancomycin (1 g after dialysis) and gentamicin (2 mg/Kg after dialysis) was administered.

A multi sensitive *Candida albicans* was isolated on blood and exudate cultures. The TCC was removed and the therapy was changed to 100 mg of micafungin intravenous (IV) once a day, as recommended for candidemia in nonneutropenic patients, in the Infectious Diseases Society of America (IDSA) guidelines [4]. After five days of therapy with micafungin, repeated blood cultures were negative. At that time, a non-tunneled temporary hemodialysis catheter was placed on the right internal jugular vein.

Transesophageal echocardiography (TEE) showed a large mass (>5 \times 1.7 cm) located in the superior vena cava, occupying \sim 50 % of the lumen, adhering to the anterior wall with protrusion to the right atrium compatible with a vegetation in probable relation with a previous TCC. The therapy was adjusted to endocarditis (150 mg of micafungin/day). Other focalizations of this agent were excluded.

After multidisciplinary discussion with cardiology, cardiothoracic surgery and infectious diseases specialists it was decided to treat initially a 3 week course of antifungal due to the high surgical risk of this particular patient. The repeated TEE showed an increase of the vegetation that now filled the entire lumen of the superior vena cava, protruded into the right atrium cavity and reached the atrial face of the tricuspid valve (Fig. 1). The latter appeared to be preserved. Due to the non-response to the antifungal therapy it was decided to surgically remove of the vegetation. Before the surgery, a triple lumen non-tunneled temporary hemodialysis catheter was placed on right femoral vein. The right internal jugular catheter was removed. The procedure was done with an inflow-occlusion technique [10] and was successful with no complications (Fig. 2). Culture of the vegetation showed a multi-sensitive Candida albicans the same as the blood cultures. After surgery she underwent another eight weeks of therapy with micafungin (150 mg/daily). A PET scan (Positron Emission Tomography) with fluorodeoxyglucose was done and ruled out relapse or another focalization of the disease. After one year, the patient is asymptomatic with no signs of relapse.

Discussion

To our knowledge, this is the first case report of candida endocarditis with no valve involvement related to a TCC with a successful treatment.

The presence of central venous catheters, hemodialysis and glucocorticoid use constitute major risk factors for invasive candidiasis [11].

Recent studies, revealed high rate of candidemia in hemodialysis patients [8], reflecting a combination of elements including the disruption of anatomic barriers as well as co-morbid conditions, such as diabetes mellitus, extensive exposure to antibiotic agents,

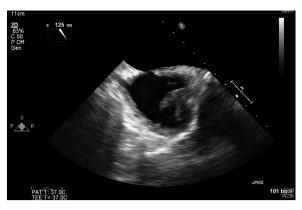


Fig. 1. Transesophageal echocardiography showing large vegetation filled the entire lumen of the visualized portion of the superior vena cava.

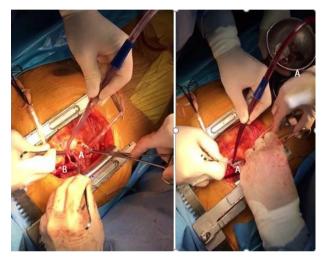


Fig. 2. Pictures of the heart surgery with inflow-occlusion technique. Inflow occlusion interrupts the cardiac filling making the field bloodless allowing the surgeon to do the required procedure. A- vegetation B- superior vena cava.

underlying renal dysfunction, and the dialysis process itself. In several studies, use of the catheter (vs. native graft or fistula) was an independent risk factor for candidemia [9].

This particular patient had additional risk because of glucocorticoid therapy. Sung et al. [10] showed that the higher prevalence of candidemia in dialysis patients is due a multiplicity of predisposing factors included corticosteroid.

The diagnosis in this case was not straightforward since there were few symptoms or signs that suggested endocarditis: no murmur, no vascular or immunologic phenomena as in the Modified Duke Infective Endocarditis Criteria.

The presentation of CE can be very nonspecific, so its important that we keep this diagnosis in mind and consider doing a TEE in patients with a CRBSI.

Furthermore, the role of micafungin in the treatment of CE is not unanimous. European Society of Clinical Microbiology and Infectious Diseases (ESCMID) 2012 guidelines [11] do not include echinocandins as therapeutic option. On the other hand 2016 IDSA guidelines [4] consider these antifungals a good choice. Their excellent safety profile with better tolerability and less toxicity even in high doses and prolonged treatment was the reason why we preferred it.

In some CE cases, isolated medical therapy is curative for patients who were not good candidates for surgery [5,12]. The size of the vegetation with a major risk of superior vena cava syndrome

or embolization and non-response to 3 weeks therapy were determinant to do decide on a surgical approach.

In summary, in hemodialysis patients, the risk of candidemia and the consequent CE is higher than in the general population. It is very important to do an effort to modify risk factors reducing catheters use (e.g., through patient education) and CRBSI as suggested by the Center for Disease Control and Prevention.

To improve the prognosis of this condition a high index of suspicion is necessary for early diagnosis and timely intervention.

Ethics approval and consent to participate

Not applicable.

Consent for publication

The patient consent for publication all the data expose including images.

Funding

Not applicable.

Declaration of Competing Interest

All authors declare that they have no competing interests.

Acknowledgements

Cardiology and Infection Disease Departments for all the support in these difficult case.

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