

## ФЛЕГМОН НА МЕКИТЕ ТЪКАНИ И ВТОРИЧЕН ЕНДОКАРДИТ 10 ГОДИНИ СЛЕД КОНГЕНИТАЛНА ХИРУРГИЯ И ИМПЛАНТАЦИЯ НА AICD

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## SOFT-TISSUE PHLEGMON AND SECONDARY ENDOCARDITIS 10 YEARS AFTER CONGENITAL CARDIAC SURGERY AND AICD-IMPLANTATION

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**Резюме.** Инфекциозният ендокардит на автоматичен имплантируем кардиовертер дефибрилатор (AICD) е рядко и потенциално фатално заболяване със значителна заболяемост и смъртност. Докладваме за случай на 23-годишен пациент от мъжки пол с ендокардит, свързан с устройство, след многократна имплантация на (AICD), свързан с камерни тахиаритмии. Поради инфекциозен ендокардит е извършена хирургична екстракция на AICD и протезиране на трикуспидалната клапа. На възраст между 4 и 10 години пациентът е имал предишни сърдечни операции поради перимембранозен междуклапален септален дефект (VSD) и обструкция на изходния тракт на дясната камера. Девайс-свързаният ендокардит е тежко усложнение след имплантиране на AICD. За потвърждаване на диагнозата ние рутинно използвахме ехокардиография и 18F-FDG PET/CT за допълнителна визуализация на находката.

**Ключови думи:** ендокардит, трикуспидална клапа, камерен септален дефект, ехокардиография, ядрено изображение, белодробна емболия, доклад за случай

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**Abstract.** Background: Infective endocarditis (IE) of Automated Implantable Cardioverter Defibrillator (AICD) is a rare and potentially fatal disease with a significant morbidity and mortality. Case Summary: We report on a case of 23-years-old male patient with device - related endocarditis after multiple implantation of (AICD) associated with ventricular tachyarrhythmias. Due to infective endocarditis we performed surgical extraction of AICD and tricuspid valve replacement (TVR). In age of 4 and 10 years the patient had previous cardiac surgeries for perimembranous ventricular septal defect (VSD) and right-ventricular outflow tract obstruction, respectively. Discussion: Device-related endocarditis is a severe complication after AICD implantation. For diagnosis verification we routinely use the echocardiography and 18F-FDG PET/CT scan as nuclear imaging. Tricuspid valve replacement is always recommended if the native valve is completely destructed.

**Key words:** endocarditis, tricuspid valve, ventricular septal defect, echocardiography, nuclear imaging, pulmonary embolism, case report

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## INTRODUCTION

Cardiac electronic device infection is a severe disease with significant morbidity and mortality. It is absolutely important to distinguish local device infection limited to the pocket of the device generator and device-related-infective endocarditis (IE) extending to the electrode leads,

valve leaflets and endocardial surface. The infection frequency of implantable electronic device varies widely between 0.13-19.9%, while device – related endocarditis is between 0.5-7% [1, 2, 3]. Patients who have diabetes mellitus, heart failure, renal failure, oral anticoagulant use and long-term corticosteroid use are at much higher risk of developing device-related infection.

## CASE PRESENTATION

The patient was a 23-year-old man with device-related endocarditis. He had surgically corrected VSD when he was 4 years old. In 2009, in age of 10 years he had a re-operation for enlargement of the right ventricular outflow tract (RVOT) and AICD implantation due to ventricular tachyarrhythmia. Due to device infection the AICD was twice explanted and reimplanted in 2009 and 2015, respectively.

**Table 1. Timeline**

2004	Congenital surgery for perimembranous VSD (in age of 4-years)
2009	Re-operation for RV outflow tract stenosis and Implantation of AICD
2009	Extirpation and AICD re-implantation due to device infection
2015	Second AICD re-implantation due to recurrent infection
Nov. 2019	Soft-tissue phlegmon of generator pouch and extraction of three RV leads
Jan. 2020	Non-thrombotic (septic) pulmonary embolism
April 2020	Surgery for TV endocarditis

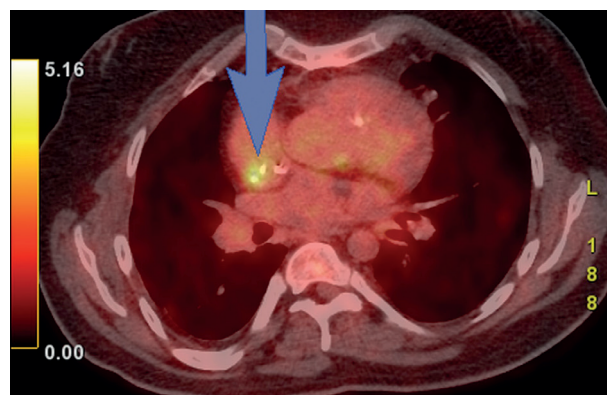
The patient was admitted to our institution in November 2019 with soft-tissue phlegmon of device pouch with isolated *Klebsiella oxytoca*, and no other inflammatory symptoms. We performed surgical device explantation and percutaneous extraction of 3 electrodes from right ventricle and right atrium through subclavian vein using a lasso technique from femoral vein. After the procedure there was a fragment of the electrode left in the right ventricle. Antimicrobial therapy was applied according to the microbial protocol with intravenous administration of meropenem 3 g daily, amikacin 1 g daily, and ciprofloxacin 400mg daily. After performing clinical and instrumental examination we confirmed that there were no signs of IE. Two sets of blood culture were negative.



**Fig. 1.** Bilateral septic pulmonary embolism

Two months later in January 2020 the patient was admitted to the cardiology department in our hospital. The performed contrast computed tomography (CT scan) revealed disseminated bilateral septic pulmonary embolism (Fig. 1). The left subclavian pouch of the infected AICD showed no signs of infection or inflammation. The patient experienced respiratory and heart failure. The physical examination showed poor general condition with fever of 39°C. The laboratory results were as follows: pH 7.60 (reference ranges 7.35-7.45); pCO<sub>2</sub> 23.0 mmHg (reference ranges 35-45 mm Hg); pO<sub>2</sub> 42.7 mmHg (reference ranges 80-100 mm Hg); HCO<sub>3</sub><sup>-</sup>: 26.9 mmol/l (reference ranges > 25 mmol/l); SaO<sub>2</sub> 87.7% (reference ranges 94-98%); BE: 3.0 mmol/l (reference ranges ± 2.5 mmol/l); D-dimer 3356.0 ng/ml (reference ranges < 600 ng/ml); CRP 215.7 mg/L (reference ranges 0.3-6 mg/L). Two sets of blood cultures grew *Staphylococcus epidermidis* and *Klebsiella oxytoca*. Trans-thoracic (TTE) and transesophageal echocardiography (TEE) was performed and there was no evidence of endocardial involvement. According to the actual microbiological protocol intravenous combination of Imipenem and Vancomycin were conducted. Two weeks later the patient was discharged in overall good condition with negative blood culture and without any inflammatory disorder. Home oral anticoagulation with Apixaban was prescribed.

In April 2020 the patient was admitted again to our institution with clinical and echocardiography features for infective endocarditis of the tricuspid valve and positive blood culture (*Staphylococcus epidermidis*). We performed positron emission tomography using white blood cells labeled with fluorine-18-fluorodeoxyglucose (18F-FDG-WBC PET). There was an increased FDG uptake around the part of right ventricular lead and in superior vena cava (SVC) (Fig. 2). Severe tricuspid valve regurgitation with vegetations and leaflets erosion was found. Large vegetations with measures 35-45 mm were also showing on the electrode in the right



**Fig. 2.** <sup>18</sup>F-FDG-WBC PET Scan

ventricle (Fig. 3). After isolation of *Klebsiella oxytoca* intravenous administration of meropenem treatment was started immediately for 22 days, according to recommendation of our department of microbiology. After multidisciplinary rounds and discussions third re-operation for surgical extraction of the lead fragments from the RV and tricuspid valve replacement (TVR) was considered indicated.

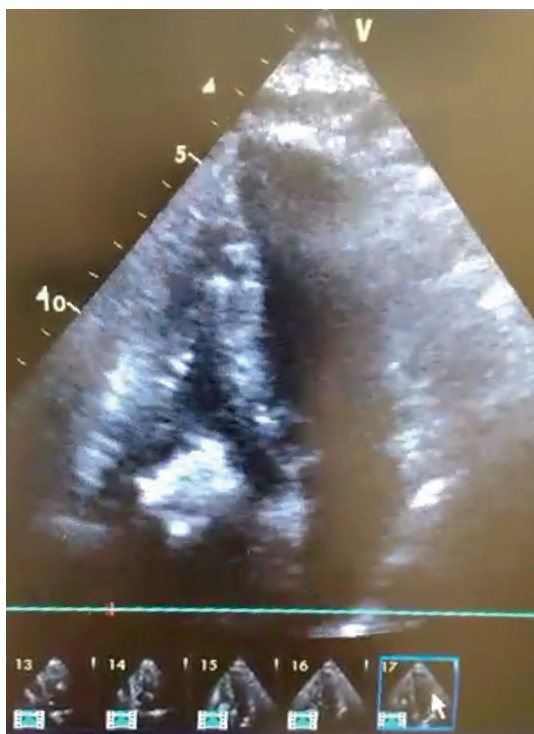


Fig. 3. IE of TV with huge valve vegetation

Redo cardiac surgery (3rd intervention) was performed in standard fashion. The tricuspid valve was completely destructed and needed to be replaced with mechanical valve (SJM 29 mm, Saint Jude Medical, MN, USA). In total three lead fragments and a lot of infected masses were sharply dissected and extracted from the RV septum and the SVC. Due to rupture of RV septum two teflon reinforced 2/0 GoreTex sutures were placed.

After 5 days ICU stay the patient was discharged on the 30th postoperative day in good general condition, negative blood culture and without any infective or inflammatory symptoms. Three years after the last hospitalization the young patient is doing very well without any restrictions and limitations.

## DISCUSSION

Device-related endocarditis is a severe complication after AICD implantation, which is very often hard to distinguish from the local device infection. The echocardiography is a gold standard for the assessment of concomitant endocarditis. In complex cases, when the

echocardiography does not rule out endocarditis, additional instrumental examination such as scintigraphy and F-FDG PET/CT scanning is necessary. Cardiac imaging plays an important role in the diagnosis, management and pre-operative evaluation of patients with device infection. The first report of cardiac infection detected by 18F-FDG PET was published in 2006 [6]. In 2015 Guidelines for the management of infective endocarditis, the European Society of Cardiology (ESC) addressed the use of nuclear medicine imaging for the diagnosis of IE [7].

Bensimhon et al. evaluated the diagnostic value of 18F-FDG PET/CT in 21 patients with presumed device infection, which were compared to 14 patients without infection [8]. 18F-FDG PET/CT had a sensitivity and specificity of 80% and 100%, respectively for diagnosis of infection. Johansen et al reported that the incidence of explantation of AICD due to infection was significantly higher after replacement procedures than after first implantation 2.06% versus 0.75% [5]. The classical Duke criteria for IE have lower sensitivity in such patients; therefore, modify criteria, including local signs of infection and pulmonary embolism, are considered as major criteria. Complete hardware removal (lead and device extraction) and prolonged antimicrobial treatment based on culture and susceptibility results must be applied [4].

TVR with mechanical prosthesis is a rare procedure. It is necessary in certain circumstances such as lack of possibility for repair and patients at higher risk of reoperation. In our case we used this approach because of exiting reoperation and the relatively young age of the patient. In one study with 265 TVRs with SJM valves in hospital death was 6.4%. Ten years survival rate was 86.6% [9].

In conclusion, the immediate extraction of infected AICD devices and its leads is absolutely mandatory, even in cases where second or third cardiac re-operation is required. The 18F-FDG-WBC PET is a very feasible approach to detect and localize the site of subtle infection with a high precision and sensitivity.

## LEARNING POINTS

The immediate extraction of infected AICD device and its leads is absolutely mandatory, even in cases where second or third cardiac re-operation is required.

The 18F-FDG-WBC PET is a very feasible approach to detect and localize the site of subtle infection with a high precision and sensitivity.

*No conflict of interest was declared*

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