Eustachian valve endocarditis: Rare case reports and review of literature

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ABSTRACT

Eustachian valve endocarditis (EVE) is a distinctly rare and underdiagnosed entity. We report 2 new cases caused by vancomycin resistant *Staphylococcus aureus* and *Staphylococcus hominis* diagnosed on transesophageal echocardiography (TEE). Although, 63% of Eustachian valve endocarditis is caused by *Staphylococcus aureus*, we report the first case of vancomycin resistant *Staphylococcus aureus* and first case related to implantable venous access systems. EVE is now seen more commonly in elderly population with diverse microbial cultures and antibiotic sensitivities. TTE is the first modality for investigation of EVE, however a negative TTE does not preclude the diagnosis, as only 88% of cases were diagnosed on TEE.

Key words: Echocardiography, endocarditis, Eustachian valve, transesophageal, vancomycin

INTRODUCTION

Right sided valvular involvement in infective endocarditis has been well-described, but lesions affecting Eustachian valve are distinctly rare. In 1986, Edwards *et al.*[1] first described the entity of Eustachian valve endocarditis (EVE) in an autopsy study of a patient with overwhelming streptococcal sepsis. In the literature only few cases have been reported so far. Herein, we review the literature and describe 2 new cases in which the causative organisms were determined to be vancomycin resistant *Staphylococcus aureus* (VRSA), which is reported for the first time, and *Staphylococcus hominis*.

CASE REPORTS

Case 1

A 33 year old woman with a history of intravenous drug use was admitted with a history of fever, chills and cough for the last 4 days. On presentation, she was noted to have a temperature of 102.4 F (reference range 96.8-100.4 F), an elevated pulse rate (110 bpm) (reference range 55-90 bpm), respiration rate of 26 breaths/min (reference range 12-20 breaths/min) and low blood pressure (90/50 mm Hg) (reference range 100-139/55-84 mm Hg). Blood investigation showed an elevated white blood cell count (22,600 cells/μL) (reference range 4000-11000 cells/μL). Chest radiography demonstrated bilateral multiple cavitatory nodules consistent with septic emboli. Suboptimal images by transthoracic echocardiography (TTE) showed mobile vegetation on the tricuspid valve, along with severe tricuspid regurgitation. Eustachian valve was not visualized. Transesophageal echocardiography (TEE) revealed a 6-cm, echogenic mass attached to the eustachian valve, [Figures 1 and 2] along with pulmonary valve and right ventricular wall. During the next 2 days, the patient's condition was complicated by hemoptysis, renal failure and hypoxic respiratory failure requiring mechanical ventilation.

She received empiric antibiotic therapy with vancomycin, gentamicin and rifampin. Three blood cultures grew

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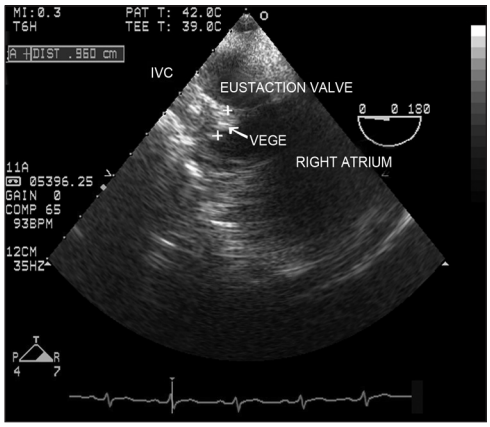

Figure 1: Transesophageal echocardiography image demonstrating a large vegetation (6 cm) attached to the Eustachian valve (Case 1)
VEGE**:** Vegetation, IVC: Inferior vena cava.

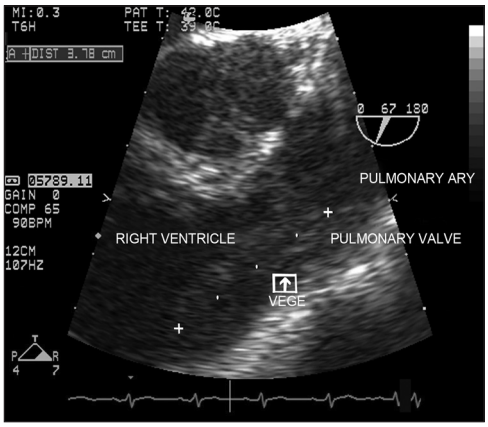

Figure 2: Transesophageal echocardiographic image of vegetation attached to the right ventricular wall (Case 1)
Pulmonary ARY**:** Pulmonary artery.

vancomycin resistant *Staphylococcus aureus* (VRSA). She was treated with intravenous daptomycin initially with 6 mg/kg every 24 hours, and was discharged after 4 weeks of antibiotics.

Case 2

A 86-year-old nursing home female with a history of dementia, severe aortic stenosis, congestive heart failure, chronic anemia secondary to gastrointestinal bleeding requiring implantable venous catheter insertion, presented with high grade temperature of 103^{0}F (reference range 96.8-100.4 F) rectally, pulse rate of 124bpm (reference range 55-90 bpm), respiration rate of 23 breaths/min (reference range 12-20 breaths/min) and blood pressure of 128/69 mm Hg (reference range 100-139/55-84 mm Hg). Laboratory data showed an elevated white blood count 30,000 cells/μL (86% polymorphs) (reference range 4000-11000 cells/μL), low hemoglobin of 7.1 g/dl (reference range 14.0-18.0 g/dl) and normal platelets (323,000/μl) (reference range 150,000-450,000/μl) Physical examination revealed a thin, cachectic female with a late peaking systolic murmur at the left sternal border, a soft S2 and few scattered rhonchi all over the lungs. Blood cultures grew *Staphylococcus hominis* resistant to penicillin G and sensitive to vancomycin.

TTE revealed moderate concentric left ventricular hypertrophy with ejection fraction of 70%, a heavily calcified aortic valve with severe aortic stenosis and moderate pulmonary hypertension; however, no vegetations were identified. The TEE revealed a 2 cm mass attached to the Eustachian valve. The patient received 4 weeks of vancomycin, 30 mg/kg in divided doses every 24 hours and became afebrile with second set of blood culture turning negative.

DISCUSSION

Eustachian valve is an embryological remnant of the sinus venosus, directing oxygenated fetal blood from inferior vena cava across foramen ovale, and into the left atrium. In adults, it is non-functional and is considered a benign rudimentary structure.

We present 2 new cases of EVE with a review of literature [Table 1]. In the previously reported cases, the age of patients ranged from 22 years to 76 years with a median age of 44.2 years, with a male to female ratio of 5:3. There seems to be an increasing trend in elderly population. In the present study, the age of the patients was 33 years and 86 years. The incidence of EVE is not well documented; however, a retrospective review by San Roman *et al.*[2] reported an incidence of 3.3% in patients with right sided endocarditis.

A predisposing factor was present in all but three cases,[3-5] with intravenous drug use (40% of the cases) being the most common. Other predisposing factors were presence of indwelling catheters, insertion of pacemaker wires, a history of rheumatic heart disease, and immunologic compromise (chronic alcoholism, human immunodeficiency virus – [HIV]) status. The increasing prevalence of indwelling catheters/devices may be the reason for increasing incidence of EVE in the elderly population. In the present study, a history of intravenous drug use was obtained in one case, and insertion of implantable venous catheter for repeated blood transfusions in the other case.

The blood culture revealed *Staphylococcus aureus* to be the most common pathogenic organism in 53% of the cases, which is consistent with previous reviews Sawhney *et al.*[6] The association of *Staphylococcus aureus* positivity on blood cultures with intravenous drug abuse (*IVDA)*

Table 1: Summary of reported cases of Eustachian value endocarditis, their location, microbial agents involved, and prognosis

Study	Age	Sex	Echocardiographic findings		Predisposing factor	Microbial agent	Vegetations diagnosed by		Prognosis
			Vegetati-ons on valves other than Eustachian	Other findings			TTE	TEE	
Case 1	33	F	Tricuspid	Trivial MR	IVDA	*VRSA*	0	+	D/C
Case 2	86	F	Mitral	Severe MR, mild TR	Implantable venous catheter	*Staphylococcus hominis*	0	+	D/C
Edwards *et al.*[1]	44	M	Mitral	-	Pneumonia	*Staphylococcus aureus*	-	-	-
Georgeson *et al.*[14]	33	M	0	Trivial TR	IVDA	*Staphylococcus aureus*	0	+	Pleural effusion
Navarro *et al.*[7]	33	M	0	-	HIV	*Staphylococcus hominis*	+	-	D/C
Paladoketti *et al.*[12]	36	M	0	0	IVDA		0	+	D/C
James *et al.*[8]	75	F	0	0	Pacemaker	*Enterobacter cloacae*	0	+	D/C
Punzo *et al.*[13]	73	F	-	Mild MS, Moderate MR, Moderate TR, Dilated LA, RA	H/o rheumatic fever	No growth	+	+	D/C
Sawhney *et al.*[6]	32	F	0	0	Indwelling central Venous Catheter	*Escherichia coli*	0	+	D/C
	68	M	0	Severe MR, Thickened Mitral Valve	Indwelling central Venous catheter	*Proteus vulgaris*	0	+	D/C
	39	F	0	0	IVDA	*Staphylococcus aureus*	0	+	D/C
	40	M	0	Mitral Valve thickened	IVDA	*Staphylococcus aureus*	0	+	D/C
San Roman *et al.*[2]	22	M	Tricuspid, Mitral	-	IVDA, HIV	*Staphylococcus aureus*	0	+	D/C
	25	M	Tricuspid	-	IVDA	*Staphylococcus aureus*	0	+	D/C
	30	M	Tricuspid, Pulmonary, Mitral	-	Hodgkin's lymphoma, central venous line, splenectomy	*Staphylococcus aureus*	0	+	D/C
	34	F	Tricuspid	-	Hodgkin's lymphoma, central venous line	*Staphylococcus aureus*	0	+	D/C
	23	M	0	-	IVDA	*Staphylococcus aureus*	0	+	D/C
Bowers *et al.*[15]	36	F	Tricuspid	Severe TR	IVDA	*Staphylococcus aureus*	0	+	Surgical excision and replacement of tricuspid valve
Schmidt *et al.*[4]	76	M	0	0	0	*Streptococcus viridians*	-	-	D/C
Gill *et al.*[3]	74	F	0	0	Absent	*Staphylococcus aureus*	0	+	Digital infarcts
Wong *et al.*[9]	70	F	0	Moderate MR	Chronic AF, Open Chole-cystectomy	*Multi-resistant Klebsiella pneumonia*	+	+	D/C
Veiga *et al.*[10]	45	M	0	0	Central venous catheter, CABG	*Klebsiella pneumonia*	0	+	D/C
Kennedy *et al.*[11]	29	F	0	Severe TR	IVDA	*Actinomyces israeli*	+	+	Surgical excision

Table 1: Contd...

Study	Age	Sex	Echocardiographic findings		Predisposing factor	Microbial agent	Vegetations diagnosed by		Prognosis
			Vegetati-Ons on valves other than Eustachian	Other findings			TTE	TTE	
Lopes *et al.*[16]	30	M	0	0	IVDA	*Staphylococcus aureus*	0	+	D/C
Maddury *et al.*[5]	52	M	Pulmona-Ry artery	None	None	*Not reported*	0	+	Surgical excision
Venkataraman *et al.*[17]	42	M	0	0	AICD CABG	*Staphylococcus aureus*	-	+	D/C
Chu *et al.*[18]	23 wks gestation	-	-	-	Intraumbilical, percutaneous lines, Chorio-Amnioitis	*Staphylococcus hemolyticus*	+	-	Death of the neonate

*AICD: Automatic implantable cardioverter defibrillator, AF: Atrial fibrillation, CABG: Coronary artery bypass grafting, *D/C- Discharged without complication, HIV: Human immunodeficiency virus, IVDA: Intravenous drug abuse, LA: Left atrium, MR: Mitral regurgitation, RA: Right atrium, TR: Tricuspid regurgitation, VRSA: Vancomycin resistant staphylococcus aureus*

and indwelling catheters was found to be in 100% of the cases. Other organisms that have been reported so far include *Staphylococcus hominis*,[7] *Enterococcus cloacae*,[8] *Escherichia coli*,[6] *Proteus vulgaris*,[6] *Streptococcus viridans,*[4] *Klebsiella pneumonia*[9,10] *and Actinomyces israeli.*[11] In our present report, the microorganisms isolated on blood cultures were VRSA and *Staphylococcus hominis*. This is the first case of VRSA EVE reported in the literature. In the second case, *Staphylococcus hominis*, a harmless commensal on human skin was the causative organism, likely related to the implantable venous access catheter insertion for chemotherapy (which is reported for the first time also). Like many other coagulase-negative *Staphylococci, Staphylococcus hominis* may occasionally cause infection in patients with compromised immune system.

Both cases in our series were detected on multi-planar TEE. The vegetations were not seen on TTE. The finding was in concordance with previous studies in which the vegetations were seen more frequently on TEE than TTE.[6] As the eustachian valve is situated posteriorly, the superiority of TEE over TTE is seen frequently.[12] Only in reports by Punzo *et al.*[13] and Navarro *et al.*[7] were TTE conclusive for EVE. In 2 other cases reported by Georgeson *et al.*,[14] TTE was frankly misleading, suggesting a ruptured chordae tendinae in one patient, and Chiari's network in the other. TEE allows not only better visualization of the Eustachian valve and Chiari network, but also easily differentiates pathological masses from these normal structures. Nevertheless, TTE remains the first imaging investigation of choice; and, TEE should be performed if clinical picture strongly suggests endocarditis and no lesions are identified on TTE.

Though EVE was first described in a post-mortem autopsy case, EVE seems to follow a benign clinical course presenting as right sided endocarditis, resolving with a 4-6 week course of culture sensitive antibiotics, as seen in 78% of patients described till date. Few patients had a transient worsening of symptoms requiring intubation, open heart surgery with removal of Eustachian valve.

CONCLUSION

Endocarditis of Eustachian valve is a rare and under diagnosed entity. It should be strongly considered when a patient has the clinical syndrome of right-sided endocarditis, but no vegetations are identified on TTE. We found the incidence of EVE to be highest in young intra-venous drug users, and *Staphylococcus aureus* to be the most common microbial agent. However, over the past 6 years EVE has been seen more frequently in the elderly population with culture positivity for diverse microbial agents. Multiplanar TEE remains the diagnostic tool of choice for detecting vegetations of Eustachian valve. Although EVE is rare, it may be unwise to rule out this diagnosis based solely on TTE, especially in the setting of persistent bacteremia or pulmonary emboli.[18]

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