Hellenic Journal of Cardiology (2016) 57, 134–137

Available online at www.sciencedirect.com

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journal homepage: http://www.journals.elsevier.com/hellenic-journal-of-cardiology/

CASE REPORT

Different types of tricuspid flail: Case reports and review of the literature

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Received 3 May 2014; accepted 20 June 2015
Available online 6 April 2016

KEYWORDS
Tricuspid regurgitation;
Leaflet flail;
Valvular disease

Abstract Tricuspid regurgitation (TR) is a common Doppler echocardiographic finding resulting from either intrinsic valve abnormalities or functional malcoaptation of structurally normal valves. TR caused by flail leaflets is most often post-traumatic, is caused by endocarditis or is a consequence of a myxomatously degenerated valve. The clinical presentation is severe and is characterized by excess mortality and high morbidity. Flail leaflets are reliably diagnosed using 2-dimensional and 3-dimensional echocardiography.

1. Introduction

Tricuspid regurgitation (TR) is a common Doppler echocardiographic finding resulting either from intrinsic valve abnormalities or functional malcoaptation of structurally normal valves. TR caused by flail leaflets is most often post-traumatic, is caused by endocarditis or is a consequence of a myxomatously degenerated valve. The clinical presentation is frequently severe and is characterized by excess mortality and high morbidity.[1] Flail leaflets are reliably diagnosed using 2-dimensional echocardiography[2] and 3-dimensional echocardiography.[3]

We hereby report three cases of patients with tricuspid flail of different aetiologies, diagnosed by an echocardiogram according to the guidelines of the European Society of Cardiology.[4]

2. Case 1

A 55-year-old man was admitted to our department due to worsening dyspnoea for two days. He was affected by

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Peer review under responsibility of Hellenic Cardiological Society.

http://dx.doi.org/10.1016/j.hjc.2016.03.007

arterial hypertension and did not have other history of systemic illnesses. However, he had suffered from blunt chest trauma one week before. A bobsled hit his thorax and abdomen while he was walking through a ski run. Clinical evaluation after the accident did not show any thorax or abdomen abnormality. At admission, his pulse rate was 80/min, his blood pressure was 100/70 mmHg, his body temperature was 36.8°C and his respiration rate was 16/min. Physical examination revealed a grade III/IV pansystolic murmur that was best heard at the fifth intercostal space along the left sternal border, hepatomegaly, and jugular distention. The electrocardiogram (ECG) showed normal sinus rhythm with right bundle branch block and right axis deviation. Chest X-ray showed a slightly increased cardiothoracic ratio. Routine laboratory tests were normal. A transthoracic echocardiogram (TTE) was performed. Left ventricular (LV) wall thicknesses, dimensions and ejection fraction (LVEF) were normal. There were no abnormalities in the valvular structures in the left side of the heart, but there was severe tricuspid regurgitation with dilated right atrium and right ventricle with normal wall motion. Systolic pulmonary arterial pressure was increased (58 mmHg). Severe tricuspid regurgitation was due to a flail leaflet of the tricuspid valve (Fig. 1, top). It was concluded that the tricuspid valve was injured after the blunt chest trauma. The patient underwent surgical repair with chordate reconstruction and annuloplasty. No complications occurred in the postoperative period.

3. Case 2

A 47-year-old white man was admitted to our hospital with dyspnoea at rest. He had a previous history of cardiac transplant for acute myocarditis and a recent myocardial biopsy for routine follow up to search for signs of rejection. At the current presentation, the physical examination revealed a grade IV/V pansystolic murmur at the fifth intercostal space along the left sternal border, hepatomegaly, peripheral oedema, and bilateral lower-lung crackles. Electrocardiography revealed sinus rhythm with spread and unspecified ventricular depolarizations in antero-inferior leads. Laboratory tests were normal. Upon his admission to the hospital, TTE revealed a LVEF of 60% with septal wall hypokinesis and increased left LV wall thickness (the results had been present in an echocardiogram 1 month earlier). Moreover, there was severe tricuspid regurgitation with dilated right atrium and right ventricle with normal wall motion. Systolic pulmonary arterial pressure was increased (43 mmHg). Severe tricuspid regurgitation was due to a flail leaflet of the tricuspid valve, most likely caused by biotome (Fig. 1, middle). The patient's condition was initially stabilized with intravenous diuretics, and then he underwent surgical repair with chordae reconstruction and annuloplasty.

4. Case 3

A 62-year-old woman was admitted to our department with a diagnosis of suspected catheter endocarditis. A transoesophageal echocardiography (TEE) was performed a few days before at another centre for a persistent fever of

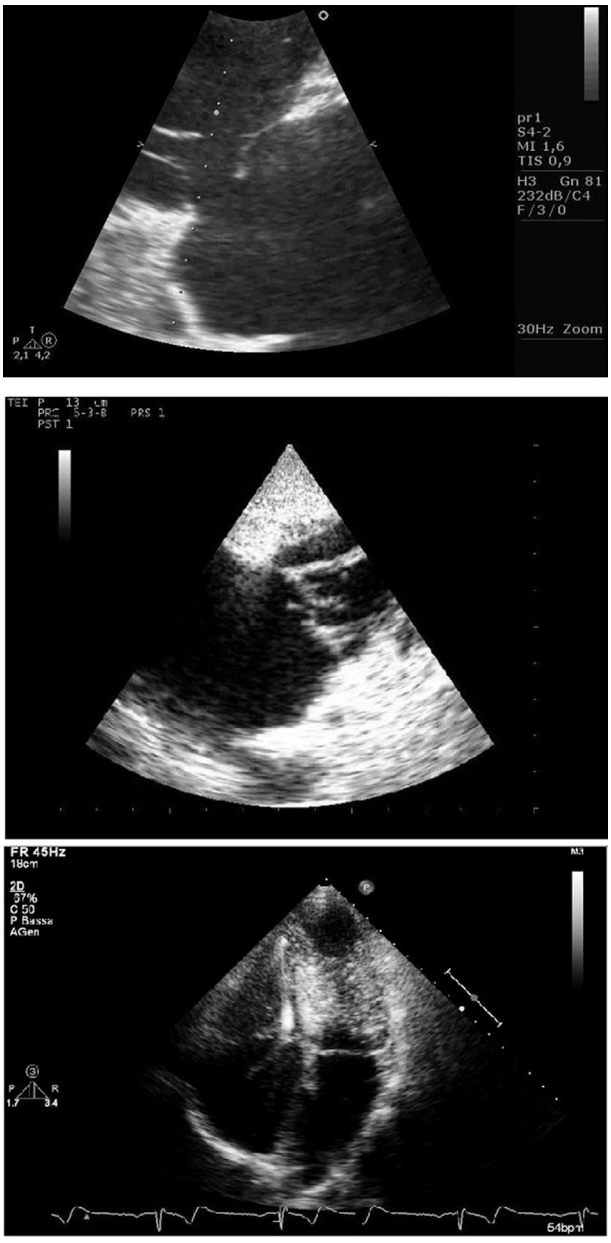

Figure 1 Top: Echocardiographic image of the tricuspid flail leaflet in case patient 1. Middle: Echocardiographic image of the tricuspid flail leaflet in case patient 2. Bottom: Echocardiographic image of the tricuspid flail leaflet in case patient 3.

unknown origin with leucocytosis and increasing C-reactive protein and erythrocyte sedimentation rate. It showed vegetation on the right catheter of the dual-chamber pacemaker (PM) implanted 3 years earlier for brady-tachy syndrome. During the hospitalization, the patient underwent catheter extraction and a repositioning procedure; both were well tolerated. After a few hours, the patient developed heart failure. She was evaluated by an echocardiogram that showed new onset mild tricuspid regurgitation with a flail leaflet of the same valve (Fig. 1, bottom) and an increased systolic pulmonary arterial pressure (40 mmHg). She was treated with intravenous diuretics with benefit. We concluded that the tricuspid valve was injured after the right catheter extraction.

5. Discussion

Flail tricuspid leaflet has been defined as a prolapsed leaflet of the tricuspid valve with excursion of the leaflet edge and/or free chords into the right atrium (RA) during systole.[5] TR caused by flail leaflets is permanent, organic, and mostly severe. Isolated organic TR is infrequent. However, there have been no comprehensive studies, and the experience regarding this valve disease is limited to case reports or short series. Accordingly, Messika-Zeitoun et al.[1] examined the causes, clinical presentations, and outcomes of patients with tricuspid flail leaflets. They reported a traumatic cause in 62% of patients, related to blunt chest trauma (mostly motor vehicle accident) in 50% and iatrogenic chordal severing (mostly right ventricular biopsy) in the other 50%. Non-traumatic causes were myxomatous (12%), infective endocarditis (8%), and congenital (3%). In 15% of patients with ruptured tricuspid chordae, no specific cause was defined, and these patients were considered idiopathic. TR caused by flail leaflets is a serious disease characterized over time by a cumulative high rate of events irrespective of its cause, suggesting that a surgical treatment should be considered early in its course. The clinical outcome after diagnosis exhibited excess mortality and high morbidity during follow-up. Even patients who were asymptomatic at presentation experienced high tricuspid-related event rates, predicted by severe enlargement of right-sided chambers on baseline echocardiography, independent of the cause. This poor outcome was also confirmed by the natural history beginning from the date of occurrence of the flail. Surgical repair of the tricuspid valve is generally feasible, with low operative mortality and marked symptomatic improvement. However, because surgical intervention is mostly performed at an advanced stage, atrial fibrillation (AF) is often refractory. Therefore, TR caused by flail leaflets is a serious disease characterized over time by a cumulative high rate of events independent of its cause, suggesting that an operation should be considered early in its course.[1]

In asymptomatic patients without severe right-side chamber enlargement, TR and right ventricular dysfunction might be progressive,[6] and close follow-up is warranted. Thus, the echocardiographic finding of tricuspid flail leaflets implies a high risk of long-term tricuspid-related complications and echoes data suggesting that severe TR, even of functional cause, might have dire outcome implications.[7,8]

In 1958, Parmley et al. were the first to describe tricuspid papillary muscle rupture from nonpenetrating chest trauma.[9] By that time, other authors had described a series of patients affected by tricuspid valve insufficiency following blunt chest trauma.[10–14]

Damage to the tricuspid valve with resultant tricuspid regurgitation induced by endomyocardial biopsy (EMB) following heart transplantation has been reported in several studies. EMB was initially noted to be associated with post-transplantation TR in the early 1990s. More reports related to iatrogenic damage of tricuspid apparatus induced by EMB were published later. The rate of flail tricuspid and TR prevalence as well as TR severity of these studies was 2.3 – 25% of flail tricuspid valve, 20.1-98% of TR and moderate or severe TR in 5.6-85% of patients.[2,5,15–18]

Williams et al.[5] examined TR in a population of patients after EMB, and they found that a moderate or severe TR was present in 32% of them. Of these patients, 14% had flail tricuspid leaflet, with 70% of these having severe TR. They had a higher right atrial pressure, a lower cardiac index, and greater right-sided cardiac dimensions.

Although implantation of a cardiac device can offer life-saving therapy, the procedure carries significant risks, including infection. The incidence of PM infections is an increasing problem. On the one hand, there is increasing use of implantable cardioverter defibrillator (ICD) and cardiac resynchronization therapy, and the other hand, our understanding of the presentation of cardiac device infection is steadily advancing. Moreover, the improved survival rates in patients bearing cardiac devices most likely lead to an increase in the number of "at risk" days for an infective complication.[19] The rates of septicaemia or endocarditis related to PM ranges from 0.5% to 2% in published studies.[20] Lead extraction was highly effective in treating PM or ICD related infection[21] and it is on class 1 indication in a North American Society for Pacing and Electrophysiology (NASPE) policy statement.[22] In large registries of lead extraction, major complications are reported in 1.9-2.5% and mortality in 0.6-0.8% of procedures.[23–25] In addition, a recent study suggested that TR caused by lead extraction is strongly associated with the use of additional tools beyond simple traction and with female sex (most likely because of more extensive fibrotic reaction and/or more fragile tissues than in men).[26]

To our knowledge, our clinical case n.3 is the first case of flail tricuspid leaflet caused by lead extraction.

In conclusion, there are several possible causes of tricuspid flail. We must pay attention to the lead extraction procedure, an efficacious treatment of PM infections, that on the whole could be an emerging cause of that pathology.

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